EXTENT, DETERMINANTS, AND CONSEQUENCES OF COST-RELATED NON-ADHERENCE TO PRESCRIPTION MEDICATIONS AMONG PEOPLE WITH SPINAL CORD INJURIES IN CANADA

by

Shikha Gupta

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the degree of Doctor of Philosophy in Rehabilitation Science

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Abstract

The overall purpose of this doctoral thesis was to build an understanding of financial barriers faced by people with disabilities to pay for their prescription medications. The thesis was composed of four different studies that were designed and conducted logically to fulfill this objective. Study 1 was a scoping review that analyzed the type, extent, and quantity of research available on CRNA to prescription drugs in Canada. The study had three notable findings: first, between 4-36% of Canadians face CRNA to fulfill their medications; second, people between 18 and 64 years, with low income, poor health status, no insurance coverage, and high out-of-pocket cost on medications are at risk of facing CRNA; and third, CRNA to prescriptions may have an adverse effect on individuals’ health and may lead to other social consequences; however, the evidence for this is still underdeveloped. This research revealed an evidence gap that no studies are available to understand CRNA among people with disabilities. Study 2 was a theoretical paper that argued for the reconsideration of existing models on CRNA for application to people with disabilities. The study suggested an adapted model needed to examine CRNA within the context of disability. This adapted model served as a conceptual framework for study 3. Study 3 quantified the magnitude and risk factors of medication non-adherence among people with SCIs. The study revealed that the cost of prescribed medications spent by individuals with SCIs and the extent of CRNA is greater than that is reported for general Canadian households. Medication-related, disability-related, and socio-economic factors were the main influencers that determined the risk of facing CRNA among participants, parallel to what was proposed in the adapted model. Lastly, study 4 aimed to understand the processes underpinning medication cost burden through a qualitative inquiry. The study found five key strategies adopted by the participants to manage the burden of medication cost. From the overall findings of the thesis, I offer recommendations to improve financial access to medications among people with disabilities in general and people with SCIs in particular, within Canada.
Co-Authorship

The manuscripts presented in this thesis are the work of Shikha Gupta. For all manuscripts, Ms. Gupta was responsible for: conceptualization, developing the research questions and the study design; developing research tools; analyzing data; and writing the manuscripts. The co-authors of the manuscripts are Dr. Mary Ann McColl (Chapters 2-5), Dr. Karen Smith (Chapters 2-5), and Dr. Sara Guilcher (Chapter 2-5).

Chapter 2: Cost-related non-adherence to prescription medications in Canada: A Scoping Review. This manuscript has been published in Patient Preference and Adherence (Dove Medical Press). Dr. McColl provided inputs regarding idea conceptualization, developing the research question, analyzing data and guidance in writing the manuscript. Dr. Smith provided recommendations regarding conceptualization and development of the manuscript, interpretation of study results, and editorial feedback on the manuscript. Dr. Guilcher provided guidance in conceptualization and development of the manuscript, interpretation of study results, and provided editorial feedback on the manuscript.

Chapter 3: An adapted model of cost-related non-adherence to medications among people with disabilities. This manuscript has been published in Journal of Disability Policy Studies (The Hammel Institute). Dr. McColl provided inputs regarding development of the research question, guidance in analyzing data and writing the manuscript, and provided editorial feedback on the manuscript. Dr. Smith provided recommendations regarding conceptualization and development of the manuscript, interpretation of study results, and editorial feedback on the manuscript. Dr. Guilcher provided guidance in conceptualization and development of the manuscript, interpretation of study results, and provided editorial feedback on the manuscript.
Chapter 4: Prescription medication cost, insurance coverage, and cost-related non-adherence among people with spinal cord injury in Canada. This manuscript has been published in Spinal Cord (Springer Nature). Dr. McColl provided inputs regarding conceptualization of the research question, guidance in data analysis and writing the manuscript, and provided editorial feedback on the manuscript. Dr. Smith provided recommendations regarding conceptualization and development of the manuscript, interpretation of study results, and editorial feedback on the manuscript. Dr. Guilcher provided guidance in conceptualization and development of the manuscript, interpretation of study results, and provided editorial feedback on the manuscript.

Chapter 5: Managing medication cost burden: A qualitative study exploring experiences of people with disabilities in Canada. This manuscript has been published in the special issue on Disabilities, Health and Well-being of the International Journal of Environmental Research and Public Health (MDPI). Dr. McColl provided guidance in the conceptualization of research question, study design, and interview guide, provided inputs in analyzing data and writing the manuscript, and provided editorial feedback on the manuscript. Dr. Smith provided recommendations regarding conceptualization and development of the manuscript, interpretation of study results, and editorial feedback on the manuscript. Dr. Guilcher provided guidance in conceptualization and development of the manuscript, interpretation of study results, and provided editorial feedback on the manuscript.
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As it is commonly said: “It takes a whole village to raise a child!!” For someone like me, it actually took a whole village to raise my two children – Ananya and my thesis, at the same time. Therefore, I have a long list of people to thank and acknowledge, and I would like to start with:

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I am sincerely grateful to my participants, and my friends Andrea, Jason and Nicole from Spinal Cord Injury Ontario. This thesis is dedicated to you and those who live with a spinal cord injury. I am grateful to you for sharing your stories with me, providing me your valuable insights at every stage of the research process and inspiring me to pursue this work for the larger social change!

My parents and extended family: My parents made the person I am today. You were the most supportive in my difficult times, and never lost faith in my potential and my capabilities. I do not have enough words to thank you, so I will just say that I love you and miss you being here with me. I am equally blessed to have Atul’s parents as my in-laws as they have been a great support throughout my journey as a doctoral candidate and a new mother. I would also like to thank my extended family – Manoj, Nidhi, Sandeep and Dia for their affection to me all these years. Thank you so much.

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when I needed it the most. You looked after not only me but also Ananya and keep instilling me hope that “I can do this”. This thesis couldn’t have been completed in time without your love and support.

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In sum, I would like to thank all of you for helping me reach where I always wanted to be and making my dreams come true. I hope I make this world a little better for those this thesis is dedicated to.

Happy reading! ☺
Dedication

I would like to dedicate this thesis to William Kirby Rowe (March 24th, 1948 – August 25th, 2017), former executive director of the Canadian Paraplegic Association (now known as Spinal Cord Injury Canada), and a visionary leader and advocate for access and inclusion for people with disabilities in Canada. In his role as an executive director, he supported for spinal cord injury research and contributed to the development of new knowledge about spinal cord injury. Your life was an inspiration to many. I feel blessed to have met you.
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<th>Full Form</th>
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<tr>
<td>ATC</td>
<td>World Health Organization’s Anatomical Therapeutic Chemical (WHO-ATC) Classification system</td>
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<tr>
<td>BC</td>
<td>British Columbia</td>
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<tr>
<td>CAD</td>
<td>Canadian Dollars</td>
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<td>CIHI</td>
<td>Canadian Institute for Health Information</td>
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<td>COREQ</td>
<td>Consolidated Criteria for Reporting Qualitative Research</td>
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<td>CRNA</td>
<td>Cost-Related Non-Adherence</td>
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<td>CSD</td>
<td>Canadian Survey of Disability</td>
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<tr>
<td>HSREB</td>
<td>Health Sciences Research Ethics Board</td>
</tr>
<tr>
<td>ICF</td>
<td>International Classification of Functioning, Disability, and Health</td>
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<td>LMICs</td>
<td>Low and Middle-Income Countries</td>
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<td>NZ</td>
<td>New Zealand</td>
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<tr>
<td>OECD</td>
<td>Organization for Economic and Cooperative Development</td>
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<td>ON</td>
<td>Ontario</td>
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<tr>
<td>OOP</td>
<td>Out of pocket</td>
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<tr>
<td>PRISMA</td>
<td>Preferred Reporting Items for Systematic Reviews and Meta-analysis</td>
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<tr>
<td>QC</td>
<td>Quebec</td>
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<tr>
<td>SCI</td>
<td>Spinal Cord Injury</td>
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<td>SSK</td>
<td>Saskatchewan</td>
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<td>UK</td>
<td>United Kingdom</td>
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<td>USA</td>
<td>United States of America</td>
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<td>WHO</td>
<td>World Health Organization</td>
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Chapter 1

General introduction

1.1 Research statement

The overall purpose of this doctoral thesis was to build an understanding of the financial barriers faced by people with disabilities to pay for their prescription medications. In particular, the objectives were to explore the extent, determinants and consequences of cost-related non-adherence (CRNA) to prescription medications among people with spinal cord injuries (SCIs) in Canada. Prescription medications play an important role in the treatment and prevention of disease and disability, especially for people living with chronic health conditions such as stroke, diabetes, multiple sclerosis, and spinal cord injury (SCI) (Allin & Liporte, 2011; Asano & Finlayson, 2014; Campbell et al., 2014; Piette et al., 2014; Rouleau & Guertin, 2011). However, the evidence is emerging that many people in Canada face financial barriers to access necessary medications, leading to non-adherence and poor health outcomes (Anis et al., 2005; Campbell et al., 2014; Law et al., 2012; Tang, Ghali & Manns, 2013).

People with disabilities are one of the heavy users of medications and also one of the economically disadvantaged groups in Canada (Canadian Survey of Disability, CSD, 2017). They constitute around 22% of the total Canadian population (15 years and above), out of which nearly 14% live in poverty. In 2017, around 13% (836,690 persons) reported that they were unable to purchase one or more of the required medications in the past 12 months because of the cost (Stats Canada, 2017). Although Canada has a
universal public health insurance program, it excludes universal coverage of prescription drugs (Kratzer, Chang, Allin, & Law, 2015). The provincial governments provide public funding for prescription drugs, though the extent of coverage varies based on age, income, or type of disease (Daw & Morgan, 2012).

For people with disabilities, prescribed medications are often assumed to be covered under the provincial drug plans for social assistance recipients. But not all people with disabilities receive income assistance and many individuals with disabilities are increasingly engaging in paid work (Morris, Fawcett, Brisebois, & Hughes, 2018). However, there are many others who work part-time, work for small employers, or are self-employed. These individuals earn marginally but enough to remain ineligible for many income-based public drug programs (Dewa, Hoch, & Steele, 2005). Even those who do receive income-assistance, benefits are minimal and often are not sufficient to fulfil necessities (Reutter et al., 2009). Moreover, for people with disabilities, there are risk factors for forgoing medications, such as barriers to income and employment, higher susceptibility to health complications, complex medical regimens, additional health care costs, and the complexity of social assistance programs. Spinal cord injury (SCI) is one such example.

SCI is one of the most catastrophic conditions for individuals, their families and society because of the costly long-term health care needs (Krueger et al., 2013). Prescription drugs may pose a lifetime economic burden to manage complications and co-morbidities associated with a chronic SCI. The literature that exists on medication cost barriers in Canada has been mostly quantitative, generally examining prevalence of or risk factors associated with non-adherence to medications due to cost (Assayag et al.,
Studies examining economic burden of medications include older adults or individuals with chronic illnesses but is not specific to the experiences of people with disabilities. Within the SCI-specific literature, studies so far have explored medication use concerning individuals’ demographic and injury-related characteristics, or the intensity and prevalence of adverse drug reactions and polypharmacy within the SCI population (Kitzman, Cecil, & Kolpek, 2017). Therefore, in this thesis, I aimed to find out if people with SCI experience CRNA to prescription medications, and if yes, what are its determinants and consequences for them? In the subsequent sections, I will discuss each aspect of this research aim in detail.

1.2 Spinal cord injury and its impact

SCI is a traumatic or non-traumatic lesion of the spinal cord resulting in any degree of neurological impairment affecting motor, sensory, and autonomic function temporarily or permanently (American Spinal Injury Association, 2006). Depending on their type, cause, level and severity of injury, individuals with SCI fall into following sub-groups such as traumatic versus non-traumatic, paraplegic versus quadriplegic, complete versus incomplete, and acute versus chronic. The injury characteristics determine the neurological recovery, extent of secondary health conditions, and overall prognosis (Gupta, Jaiswal, Norman, & DePaul, 2019).

Available estimates suggest that there are more than 85,000 individuals living with an SCI in Canada (Farry & Baxter, 2010), though the numbers must have increased over the last 10 years. Individuals with SCI experience a large number of serious
secondary health conditions, many of which occur at a significantly higher rate in persons with SCI than in the general population (Jensen et al. 2013). The complications and co-morbidities associated with SCI impact many areas of one’s life. Impairments in bowel and bladder function, mobility, and autonomic functions, along with pressure ulcers and pain, are some of the consequences that directly impact one’s health with SCI (Adriaansen et al., 2013; McColl, Gupta, McColl, & Smith, 2018).

Associated disabilities and life changes render SCI as one of the catastrophic injuries because of the costly long-term health care needs. According to a national study, the estimated lifetime economic burden associated with a traumatic SCI in Canada ranges from $1.47 million for a person with incomplete paraplegia to $3.03 million for an individual with complete tetraplegia. Costs associated with hospitalizations ($0.17 billion or 6.5% of total costs), health care practitioner visits ($0.18 billion or 6.7%), equipment and home modifications ($0.31 billion or 11.6%) and attendant care ($0.87 billion or 32.7%) are the major direct costs (Krueger, Noonan, Trenaman, Joshi, & Rivers, 2013). Although prescription drugs did not constitute the largest portions of the health care costs for people with SCI in this study, the evidence is emerging that people with SCI are high users of medications (Guilcher et al., 2018; Kitzman, Cecil, & Kolpek, 2017).

People with SCI might need medicines to promote functional recovery or to treat various health conditions as they live with a SCI (Rabchevsky, Patel, & Springer, 2011). A study suggested that medication use increased 3.29 times in a population after acquiring a SCI (Jensen & Biering-Sørensen, 2014). Another study found that among a community-based cohort of people with a SCI in Quebec, more than 19 classes of drugs and more than 300 individual drugs were found to be administered (Rouleau & Guertin,
Other chronic conditions that individuals with SCI experience and may require long term use of medications include diabetes, cardiovascular diseases, respiratory problems etc. (Dryden et al., 2004; Hitzig et al., 2008; Rouleau & Guertin, 2011).

### 1.3 Drug coverage in Canada

The Canada Health Act, a framework to the Canadian Health System, covers medically necessary hospital, physician and limited long-term services but not prescription medications (Dewa, Hotch, & Steele, 2005). Countries similar to Canada in terms of the economy like Australia, Netherlands, New Zealand, Norway, Sweden, Switzerland and the United Kingdom have universal coverage of prescription medications while Canada does not provide such coverage (Squires, 2011). Prescription medications in Canada are paid for by the government drug plans, employer/ private insurance plans, and/or out-of-pocket as described here:

(a) **Federal drug benefit programs:** The federal government provides coverage to certain Canadians who are members of the eligible groups. Eligible groups include First Nations and Inuit, members of Canadian Armed forces, qualified veterans by Veteran Affairs Canada, and members of the Royal Canadian Mounted Police. These plans cover around one million individuals across Canada (Government of Canada, 2017).

(b) **Provincial drug benefit plans:** All provinces provide drug benefits to their eligible and uninsured residents for the eligible drugs. Overall, there are more than 100 public drug plans in Canada that fund around 42% of total pharmaceutical costs (Brandt, Shearer, & Morgan, 2018). To partially cover the cost for medications, all provinces use a combination of cost-sharing mechanisms such as premiums, copayments, co-insurance and deductibles. These terms are described in Table 1 (Demers et al., 2008).
The amount of out-of-pocket costs shared by individual Canadians vary by age, medication costs, and income level. It is to be noted that all provinces have different thresholds to define low-income and catastrophic drug costs, which results in different out-of-pocket costs paid by residents with similar age, income, and medication cost burden. Overall, deductibles are the most common form of cost-sharing mechanism across different provincial drug benefit plans and include annual amounts that patients pay for prescriptions before their provincial health insurance begins to pay for covered/eligible services. Individuals pay co-insurance and copayments after they have paid deductible each time a prescription is filled. Most of the provincial drug plans also involve a dispensing fee which is usually paid by the individual, and is calculated depending on various factors, such as type of pharmacy, distance to pharmacy, type of services a pharmacist is providing, provincial policies, type of drugs, complexity of regimen etc. It usually ranges between $6 to $18 (with some exceptions) and charged every time a prescription is filled (Patented Medicines Pricing Review Board of Canada, 2018). Table 2 describes various provincial drug benefit programs, their names, the eligible groups and the cost-sharing mechanisms.

**Table 1. Types of out-of-pocket costs**

<table>
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<tr>
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<th>A deductible is a fixed amount or a percentage of income that must be paid by the beneficiary before the insurer pays the premium.</th>
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<tr>
<td><strong>Copayment</strong></td>
<td>A copayment is a flat fee and constitutes a portion of the prescription cost that is not compensated by the insurer but must be borne by the beneficiary/insured person</td>
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<tr>
<td><strong>Co-insurance</strong></td>
<td>Co-insurance is the share (a percentage) of the prescription costs that beneficiaries pay directly at the point of service.</td>
</tr>
<tr>
<td><strong>Premium</strong></td>
<td>A premium is defined as a fixed amount that an insured person or a beneficiary must pay to be eligible for the insurance program</td>
</tr>
<tr>
<td><strong>Dispensing fee</strong></td>
<td>Dispensing fee is a professional fee a pharmacy charges every time a prescription is filled.</td>
</tr>
</tbody>
</table>
Table 2. Provincial drug programs in Canada

<table>
<thead>
<tr>
<th>Province</th>
<th>Name of the plan</th>
<th>Eligible groups</th>
<th>Out of pocket costs*</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Alberta</strong></td>
<td>1. Non-Group Coverage (Voluntary plan)</td>
<td>Albertans &lt;65 years of age and their dependents</td>
<td>Premiums: $44.45 (single) to $118 for family per month</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Co-pay: 30% to a maximum of $25 with some exceptions.</td>
</tr>
<tr>
<td></td>
<td>2. Coverage for Seniors</td>
<td>Albertans &gt;65 years of age and their dependents</td>
<td>Co-pay: 30% to a maximum of $25 with some exceptions.</td>
</tr>
<tr>
<td></td>
<td>3. Special programs (covers selected medications used in the direct treatment to patients)</td>
<td>Patients with Cancer, organ transplant, insulin therapy, communicable diseases etc.</td>
<td>Full Coverage</td>
</tr>
<tr>
<td><strong>British Columbia</strong></td>
<td>1. Fair Pharmacare (Voluntary plan)</td>
<td>All BC residents, regardless of age and income</td>
<td>Deductible: 1-3%, as per income.</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Co-insurance: 30% after deductibles</td>
</tr>
<tr>
<td></td>
<td>2. Special programs</td>
<td>Palliative care patients, first nations, cystic fibrosis patients, children at home, people on psychiatric medications, people living in residential care</td>
<td>Full coverage</td>
</tr>
<tr>
<td><strong>Manitoba</strong></td>
<td>1. Pharmacare</td>
<td>Uninsured Manitobans whose income is seriously affected by high prescription drug costs, regardless of age.</td>
<td>Deductible: ranges 3.17% -7.15 % depending on adjusted family income (minimum deductible is $100)</td>
</tr>
<tr>
<td><strong>New Brunswick</strong></td>
<td>1. Prescription Drug Program (NBPDP)</td>
<td>Uninsured residents of New Brunswick</td>
<td>Annual premiums: $200-$2000 as per income</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Co-pay: 30% per prescription</td>
</tr>
<tr>
<td></td>
<td>- Seniors</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>- Plan A for low income seniors</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>- Medavie Blue Cross Seniors medication program</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>- Other eligible populations: multiple sclerosis, cystic fibrosis, organ transplant, HIV-AIDS, special needs children.</td>
<td></td>
<td>a registration fee</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>a fixed copay: $9.05- $15 per prescription</td>
</tr>
</tbody>
</table>
| Newfoundland and Labrador | 1. Prescription Drug Program (voluntary coverage for eligible drugs) | - Seniors  
- Individuals having catastrophic drug costs (> 5-10% of annual income) | - Deductible- 5-10%, calculated depending on family income, low income seniors pay only a dispensing fee  
- Dispensing fee: $6 every time |
|---|---|---|---|
| | 2. Special programs | - Persons and families receiving income support benefits through the Department of Advanced Education and Skills  
- Individuals receiving services through the regional health authorities, including children in the care of Child, Youth and Family Services, and individuals in supervised care. | - Full coverage |
| Nova Scotia | 1. Family Pharmacare | - Seniors  
- Non-senior uninsured residents and their families with catastrophic drug costs | - Annual Premium: $0-$424 as per income  
- Co-insurance- 30% per prescription?  
- Deductible: 1-20% of net income up to 6-35% maximum  
- Co-insurance- 20% per prescription? |
| | 2. Special drug assistance programs (available for eligible drugs) | - Patients under palliative care, welfare children and people receiving disability or income support | - Co-pay: $5 per prescription? |
| Ontario | 1. Ontario Drug Benefit Program (ODB): covers the cost of certain drugs listed on provincial drug formulary | - Seniors (earning more than $20,000)  
- Low income seniors  
- Social assistance recipients  
- People receiving long term care  
- People less than 25 years and not covered by private insurance | - Deductible: $100 /year/ person  
- Copay: $6.11 per prescription  
- Co-pay: $2 per prescription  
- Co-pay: $2 per prescription  
- Co-pay: $2 per prescription |
| | 2. Trillium Drug Program (covers the cost of drugs listed on ODBP formulary) | - Ontario residents less than 65 years with high drug costs and no or limited private coverage | - Deductible: 4% of net income  
- Co-pay: $2 per prescription after deductibles are met |
<p>| | 3. Exceptional access Program (covers the cost of certain drugs not listed on ODBP) | - Individuals covered under one of the provincial drug programs | - Applicable as per age, income, medication costs |</p>
<table>
<thead>
<tr>
<th>Province</th>
<th>Program</th>
<th>Eligibility</th>
<th>Coverage/Co-pay Details</th>
</tr>
</thead>
<tbody>
<tr>
<td>Prince Edward Island</td>
<td>1. Drug Cost Assistance Program</td>
<td>Seniors &gt;65 years regardless of income or disease.</td>
<td>Co-pay: $8.25 plus $7.69 of the pharmacy professional fee for each prescription.</td>
</tr>
<tr>
<td></td>
<td>2. Generic Medication Program</td>
<td>For people less than 65 years and not having private insurance.</td>
<td>Maximum out-of-pocket costs of $19.95 for eligible generic prescription drugs</td>
</tr>
<tr>
<td></td>
<td>3. Special programs:</td>
<td>Social assistance recipients; People on long-acting injectable antipsychotic medication; People with cystic fibrosis; Chronic renal failure, HIV/AIDS, Diabetes, Ankylosing Spondylitis, Cancer, Crohn's Disease, Multiple Sclerosis, Plaque Psoriasis, Pulmonary Hypertension, Psoriatic Arthritis, Rheumatoid Arthritis, and Wet Age-Related Macular Degeneration.</td>
<td>Full or partial coverage depending on type of medication and illness</td>
</tr>
<tr>
<td>Quebec</td>
<td>1. Public Prescription Medication Insurance Plan (Mandatory drug plan)</td>
<td>Quebec residents not eligible for private insurance, regardless of their age and disease</td>
<td>Premium: $0 to $636/ person/ year as per income, Deductible: $21.75/ month, Co-pay: 37% per prescription</td>
</tr>
<tr>
<td></td>
<td>2. Special programs</td>
<td>Low-income seniors, persons with functional impairment, individuals who are living alone or with their parents and do not have access to a private plan.</td>
<td>Full coverage</td>
</tr>
<tr>
<td>Saskatchewan</td>
<td>1. Seniors drug plan</td>
<td>Seniors with a reported net income of $69,057 or less; People receiving social/income assistance, children, paraplegics, people with cystic fibrosis, and Chronic renal disease.</td>
<td>Co-pay: $25 per prescription</td>
</tr>
<tr>
<td></td>
<td>2. Special Support Program</td>
<td>Individuals with high drug costs and low family income</td>
<td>Deductible: 3-4% of total family income, Co-insurance: 35% per prescription after deductibles</td>
</tr>
</tbody>
</table>

*NOTE: Dispensing fees are not mentioned. Source: Government of Canada, 2017*
- All provinces have different plans for older adults (65 years or more) and those under 65 except Manitoba and British Columbia. Manitoba provides income-based coverage to individuals who are uninsured and bear substantial medication costs, regardless of age. British Columbia offers voluntary coverage to all British Columbia residents and poses income-based deductibles and co-insurance, regardless of age.

- Older adults with low-income receive full coverage in Quebec and have low or no cost-sharing regardless of medication cost burden except in Manitoba and British Columbia. They pay lower out-of-pocket costs for their prescription medications than seniors with high-income in Ontario, Saskatchewan, New Brunswick, and New Foundland and Labrador. There is no difference in cost-sharing between older adults with high and low-income in Alberta, Nova Scotia, and Prince Edward-Island.

- Older adults with high-income pay the lowest out-of-pocket costs in Prince-Edward-Island, Alberta, and Saskatchewan, and highest in Quebec, followed by Manitoba, British Columbia, Nova Scotia, New Brunswick, Newfoundland and Ontario.

- Individual with high-income, unless they have catastrophic drug costs, do not receive any provincial drug benefits except in British Columbia and Alberta, which provide voluntary drug coverage with cost-sharing based on income.

- Individuals with low-income can receive provincial drug benefits with income-based cost-sharing regardless of medication costs in five provinces - British Columbia, Quebec, Alberta, New Brunswick, Prince-Edward-Island. In other provinces- Ontario, Manitoba, Saskatchewan, Nova Scotia, Newfoundland, they are eligible for provincial drug benefits only if they have catastrophic drug costs.
- Social assistance recipients pay lower amounts for their prescription medications than older adults and non-recipient of social assistance. Ontario has the lowest out-of-pocket costs for social assistance recipients. Ontario is followed by New Brunswick, Nova Scotia, Newfoundland and Labrador, Prince Edward Island, Quebec and British Columbia. Saskatchewan, Manitoba and Alberta have higher cost-sharing for social assistance /welfare recipients.

- Certain groups of people receive full coverage, also called as first-dollar coverage, based on their disease status. Some common diseases for which full coverage is provided include cystic fibrosis, cancer, organ transplant, HIV-AIDS, special needs children, schizophrenia, thalassemia, chronic renal disease, psoriatic arthritis, rheumatoid arthritis, and age-related macular degeneration.

  (c) Private health insurance: The private plans are mostly provided by employers or purchased as supplementary or complementary insurance. Supplementary insurance includes coverage for additional health services that are not publicly covered. In contrast, the complementary health insurance complements coverage of government/social insured services by covering all or part of the extra costs not reimbursed otherwise (such as copayments) (Organization for Economic and Cooperative Development, OECD, 2019). Some of these private plans also provide family coverage. There are more than 100,000 private drug plans in Canada that cover approximately 60% of total Canadians and constitute around 36% of total spending on prescription drugs (Brandt et al., 2018).

  (c) Out-of-pocket: These include the direct costs paid by the individuals in the form of premiums, deductibles, copayments, co-insurance, pharmacy dispensing fees and
cash. National estimates suggest that 22% of total drug expenditures in Canada are out-of-pocket (Canadian Institute of Health Information, CIHI, 2018).

1.4 Rising prescription drug costs in Canada

Prescription medications are the topmost drivers of increasing healthcare costs in Canada and are the second topmost category of private spending (Law, Daw, Cheng, & Morgan, 2013). Canada has the second-highest spending on prescription drugs (C$926 per capita in 2017) among the OECD countries (CIHI, 2018; Brandt et al., 2018). The total spending on prescription drugs in Canada grew from $3.5 billion in 1980 to $27.2 billion in 2011 and $33.7 billion in 2018 (CIHI, 2018). These costs have grown rapidly with their share in total healthcare spending, nearly doubling from approximately 8.5% in the mid-1970s to an estimated 15.7% ($33.7 billion) in 2018. This trend is making prescription drugs the top cost drivers of the Canadian health care system.

Due to the rising expenditure on medicines, provincial governments are trying to reduce prescription drug costs from the healthcare budget. In several provinces, the drug coverage is limited to catastrophic coverage, which is the provision of a general level of coverage that protects individuals from drug expenses that threaten their financial security or cause undue financial hardship (Brandt et al., 2018). However, this type of coverage incorporates high deductibles for people who are already ill, leaving them to make difficult choices between the medications they need and other essentials. Additionally, most provincial and territorial governments have formulary restrictions or benefit caps to provide partial coverage to prescription medication costs. For example, most of these plans reimburse only the cost of generic substitutes of branded medicines, under a policy known as reference drug pricing (Tang et al., 2013). According to this
policy, if the brand-name drug is desired, then the patient must pay the difference. On the other side, where drug coverage is offered with low or no copayments for people receiving social assistance, there are wide variations in benefits (Tang et al., 2013). A recent government report measuring out of pocket costs borne by patients on medicines across Canada found that the patients who are less than 65 years of age and not receiving social assistance, out of pocket costs are highest due to the requirement to pay a premium (Clement et al., 2016). This scenario indicates that many people in Canada are underinsured, which might include low-income working Canadians with disabilities who are below the age of 65 years and earning a modest income and fall just above the threshold for public drug coverage programs (Lankin & Sheikh, 2012; Withers, 2016).

1.5 Impact of limited access to medicines on health

The financial burden of medications can lead to non-adherence, called cost-related non-adherence (CRNA), which may take four forms: unfilled prescriptions, delayed prescriptions, less frequent and/or smaller doses (Zheng, Poulse, Fulford, & Holbrook, 2012). A national study estimated that over 7.5 million Canadians do not have adequate drug coverage, and over 3 million Canadians are not able to fill their prescriptions due to cost (Law et al., 2018). The financial barriers to medication have consequences on access and utilization of other healthcare services (Lurk, DeJong, Woods, Knell, & Carroll, 2004). A few studies report that an increase in out-of-pocket expenditure on prescription drugs is related to a reduction in physician visits, filling prescriptions and an increase in hospital admissions (Anis et al., 2005; Campbell et al., 2014). Many insurance plans which now require copayments and deductibles reportedly are associated with adverse health outcomes (Tamblyn et al., 2001). For example, in
Quebec, the introduction of copayments was associated with an increased rate of emergency department visits among those receiving social assistance (Tang et al., 2013).

Apart from health outcomes, the financial burden of prescription drugs on patients have other social consequences. Researchers in the past have reported that patients make the necessary arrangements, such as cutting back on other basic expenses, to obtain their medications (Soumerai, Ross-Degnan, Avorn, & McLaughlin, 1991). Several contemporary studies have found that a rise in the annual health care costs on prescription medications place a household at an increased risk of food insecurity independent of other social determinants of health (Tarasuk et al., 2015; Law et al., 2018). Recently published national studies suggest that over 1 million Canadians cut back on food or have to borrow money to pay for their medications (Law et al., 2018; Kolhatkar et al., 2018).

Many national commission reports including Justice Hall Commission Report (1964), Romanow Commission Report (2002), Senate Kirby Report (2002), and House of Commons Standing Committee on Health Report (2018) have suggested to include pharmaceutical services in the basket of nationally insured services that are provided universally (Health Canada, 2019). Most recently, in 2018, the federal government commissioned an Advisory Council on the Implementation of National Pharmacare with a mandate of advising the government about a national drug program that would be affordable for the Canadians, the employers and the governments. However, the discussions and decisions around this issue stand unresolved due to the competing interests between insurance companies, pharmaceutical industry, patient groups and government (Brandt et al., 2018).
Overall, evidence from Canada shows that many people have to bear undue hardships due to out-of-pocket expenditure on prescribed medications, despite the provincial coverage. However, none of the studies have explored the implications of medication-related costs for people with disabilities such as those living with a SCI. Furthermore, over the past decade, many healthcare groups, advocacy associations and health policy researchers have proposed different Pharmacare models for Canada (Brandt et al., 2018; Dixon, 2013). Therefore, this study is crucial and timely to this context and contributes to the limited literature on access to medications for people with SCI and the role of health systems or health service providers in this phenomenon. The results might provide the policymakers with empirical evidence on the implications of costs of prescription medicines for people with long-term disabilities such as SCI for future policy directions and alternatives.

1.6 Research purpose and objectives

The overall purpose of this doctoral thesis is to build an understanding of financial cost barriers faced by people with disabilities to pay for their prescription medications. In particular, the objectives are to:

1. Synthesize the existing literature on CRNA to prescription medications in Canada and identify literature gaps in relation to people with disabilities.

2. Understand the phenomenon of CRNA to prescription medications from the lens of disability.

3. Describe the extent, determinants, and consequences of CRNA in people with SCIs.
1.7 Research design and worldview

The study uses a mixed-method approach and is conducted with an overarching pragmatist worldview. Worldviews are the philosophical assumptions and a basic set of beliefs of the researcher that guide the enquiry (Guba & Lincoln, 2005). The pragmatist worldview is typically associated with the mixed method research where the focus is on the consequences of research on the use of multiple methods of data collection to inform the problems under study. Thus, it is a pluralistic approach and oriented towards “what works” in practice (Creswell & Clark, 2011, p. 41). The pragmatists view reality in multiple ways, that is, as both singular (there may be a theory that operates to explain the phenomenon of study) as well as multiple (e.g., it is important to understand varied individual input into the nature of the phenomenon as well) (Creswell & Clark, 2011, p. 41).

Therefore, in this study, both deductive and inductive thinking are applied, and a mix of qualitative and quantitative methods are employed to achieve the research objectives. Quantitative methods are used within a post-positivist worldview in which the study is designed using a priori theory drawn from the previous literature, and pre-defined variables and outcomes are selected and measured. This is supplemented by a qualitative phase of the study adopting a constructivist worldview to build an in-depth understanding, and possibly explain the results obtained from quantitative approaches. Table 3 below highlights the research design and methods for specific studies conducted to fulfil each of the thesis objectives.
### Table 3. Research objectives and corresponding methods

<table>
<thead>
<tr>
<th>Objectives</th>
<th>Methods</th>
<th>Data Collection</th>
<th>Data Analysis</th>
<th>Results report</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Synthesize the existing literature on CRNA to prescription medications in Canada and identify literature gaps in relation to people with disabilities.</td>
<td>Scoping review, using Arskye O’Malley steps, Conceptual Framework: Piette Model</td>
<td>Scientific databases; keyword-based search; reference list searching</td>
<td>Descriptive and thematic analysis of the studies included; gaps in the literature</td>
<td>Manuscript 1 (Chapter 2 of the thesis)</td>
</tr>
<tr>
<td>2. Understand the phenomenon of CRNA to prescription medications from the lens of disability.</td>
<td>Critical analysis of theoretical models for CRNA, especially Piette Model</td>
<td>Review of the global literature on social determinants of disability and their potential impact on access to medications</td>
<td>Examination of CRNA model and suggested an adapted framework</td>
<td>Manuscript 2 (Chapter 3 of the thesis)</td>
</tr>
<tr>
<td>3. Describe the extent, determinants, and consequences of CRNA in people with SCIs.</td>
<td>Sequential explanatory mixed-method research: quantitative study → qualitative study</td>
<td>Online survey from 160 people with SCIs in Canada; personal interviews with 12 people who reported medication underuse due to cost</td>
<td>Quantitative analysis through SPSS; qualitative analysis through NVIVO</td>
<td>Manuscript 3 &amp; 4 (Chapters 4 and 5 of the thesis)</td>
</tr>
</tbody>
</table>

**1.8 Conceptual framework**

A conceptual framework given by Piette, Heisler, Horne, & Alexander (2006) is used for understanding the influence of patient, medication, clinician, and health system factors on individuals' adherence to medicines (Figure 1). According to this model, the cost-adherence relationship is modified by contextual factors, including patients' characteristics (e.g., age, ethnicity, and attitudes toward medications), the type of medications they are using (e.g., the complexity of dosing and the drug's clinical target), clinician factors (e.g., choice of medication and communication about medication costs), and health system factors (e.g., efforts to influence clinicians' prescribing and to help patients apply for financial assistance programs) (Piette et al., 2006). This framework was studied and adapted in examining the medication use, medication cost and consequences...
of prescription medication costs among people with SCI and finding the co-factors that influence these phenomena.

**Figure 1. Conceptual framework**

![Conceptual framework diagram](insert_image)

Notes: #: number; Rx: Drug; HRQL: health related quality of life; D/T: due to; Pt: patient

Source: Piette et al., 2006

### 1.9 Overview of the thesis

This is a manuscript style thesis that is divided into six chapters – introduction, four studies and a discussion and conclusion. Chapter 1 is the introductory chapter, where I introduce the research problem, its context and discuss the overall methodology. Chapter 2 is a scoping review that orients the readers to the breadth of the literature on CRNA to prescription drugs in Canada. Chapter 3 is a theoretical paper wherein I review the models available to understand medication non-adherence to find if they are representative of the issues faced by people with disabilities to access their prescription medications. Chapter 4 presents the findings of a cross-sectional survey conducted with
160 persons with SCI in Canada and quantify the extent and determinants of medication non-adherence due to cost among people with SCIs. Chapter 5 describes the strategies adopted by individuals to deal with medication cost burden and their downstream consequences through a qualitative inquiry. Finally, the last chapter, Chapter 6, presents the summary of the key findings integrated from the quantitative and qualitative studies, strengths and limitations of the work, its potential implications for policy, practice and research, and some of the efforts undertaken for knowledge translation.

1.10 References


Assayag, J., Forget, A., Kettani, F. Z., Beauchesne, M. F., Moisan, J., & Blais, L. (2013). The impact of the type of insurance plan on adherence and persistence with antidepressants: A


Government of Canada.


income profile of Canadians with disabilities aged 15 years and over, 2017. Statistics Canada.


Chapter 2

Cost-related non-adherence to prescription medications in Canada: A scoping review

2.1 The Fit of Manuscript 1 within the Dissertation

Manuscript 1, based on the scoping review study, contributes to the purpose of the dissertation by providing a synthesis of scientific evidence that exists on the topic of CRNA to prescription medications in Canada. This manuscript helps to understand the extent, determinants, and consequences of CRNA documented in the literature. This study also illuminates the knowledge gap in the field that exists in relation to people with disabilities in Canada. This knowledge synthesis built a strong foundation for further exploration of CRNA in the context of people with disabilities, specifically those with SCIs in the second, third and fourth manuscripts.
2.2 Abstract

The evidence is emerging that prescription medications are the topmost drivers of increasing healthcare costs in Canada. The financial burden of medications may lead individuals to adopt various rationing or restrictive behaviours, such as cost-related non-adherence (CRNA) to medications. Therefore, the purpose of this study is to provide an overview of the type, extent, and quantity of research available on CRNA to prescription drugs in Canada and evaluate existing gaps in the literature. The study was conducted using a scoping review methodology. Six databases were searched from inception till June 2017. Articles were considered for inclusion if they focused on extent, determinants, and consequences of CRNA to prescription medication use in the Canadian context. Variables extracted for data charting included author(s), year of publication, study design, the focus of the article, sample size, population characteristics, and key outcomes or results. This review found 37 studies that offered evidence on the extent, determinants, and consequences of CRNA to prescription medications in Canada. Depending on the population characteristics and province, the prevalence of CRNA varies between 4% to 36% in Canada. Canadians who are young (between 18-64 years), without drug insurance, have lower income or precarious or irregular employment and high out-of-pocket expenditure on drugs are most likely to face CRNA to their prescriptions. The evidence that CRNA has negative health and social outcomes for patients is insufficient. Literature regarding the influence of prescribing healthcare professionals on patients’ decisions to stop taking medications is limited. There is also a dearth of literature that explores patients’ decisions and strategies to manage their prescription cost burden.
2.3 Introduction

Prescription medications play an important role in the treatment and prevention of disease, especially for people living with chronic conditions (Allin & Laporte, 2011). However, costs associated with long-term use may pose a lifetime economic burden on people who are in need of those medications (Tang, Ghali, & Manns, 2014). Evidence is emerging that prescription medications are the topmost drivers of increasing healthcare costs in Canada (Law, Daw, Cheng, & Morgan, 2013). In 2015, spending on prescription medications in Canada increased by 9.2% compared with 2014 (Canadian Institute of Health Information, 2016). Public plans contributed 44%, private insurance paid for 35% and out-of-pocket payments made up the remaining 20% of the costs (Canadian Institute of Health Information, 2016).

Out-of-pocket costs for medications are the third topmost category of private spending (Morgan, Law, Daw, & Abraham, 2015). These potentially include direct out-of-pocket payments at the point of care, insurance premiums either paid directly or on payroll deduction, and user charges such as copayments or deductibles (Law et al., 2013). Households in the lowest income quintile pay an average of almost $500 per year for prescription drugs ($296), and health insurance premiums ($222) (Sanmartin, Hennessy, Lu, & Law, 2014).

In Canada, public health insurance is meant to cover all medically necessary hospital, physician and some long-term services but not prescription medications (Dewa, Hoch, & Steele, 2005). Furthermore, there is no national standard for drug coverage or drug purchasing in Canada (Morgan, Daw, & Law, 2013). People are either covered by the private insurance plans or provincial drug benefit plans that cover older adults, people
with disabilities, or people with catastrophic health needs (Kratzer, Chang, Allin, & Law, 2015) The extent of coverage, however, varies extensively among individuals as well as the provinces (Demers et al., 2008).

There is evidence that in the absence of insurance coverage for medications, patients are often in a position of having to make economic decisions about whether or not they will take their medications as prescribed. The decision to alter medication regimes for economic reasons is referred to as cost-related non-adherence (CRNA), such as stop filling prescriptions, delaying prescriptions, or taking less frequent and/or smaller doses to make them last longer (Zheng, Poulose, Fulford, & Holbrook, 2012). CRNA has been shown to have both direct or indirect effects on health and social outcomes of individuals, such as use of other medications and health services (doctor, specialist, and/or a hospital) and social consequences such as sacrificing other basic needs or taking loans to fulfil medication needs.

Piette and colleagues (2006) developed a conceptual model to understand the determinants of CRNA among patients with chronic illnesses (Piette, Heisler, Horne, & Alexander, 2006). According to this framework, the cost-adherence relationship is determined by the interplay of various factors in context, such as:

(a) characteristics related to patients themselves (e.g., age, income, employment status);

(b) medication usage and its type (e.g., importance of medications and complexity of dosing);

(c) clinician related factors (e.g., medication choice, support provided by the doctors, and communication about medication costs); and
(d) health system factors (e.g., mechanisms to help low-income patients get the financial assistance for filling necessary prescriptions) (Piette et al., 2006).

The framework developed by Piette and associates suggest that medication use and adherence is modified by various cost and non-cost factors, where some patients despite the costs use their medicines as per their prescription, while others report underuse or non-adherence despite having an apparent ability to afford their prescriptions (Piette et al., 2006). The framework was the first ever theoretically grounded conceptual model that laid the foundations to understand the construct of CRNA to medications in patients with chronic diseases. In the early 2000s, the national political debates about prescription cost pressures started emerging in the USA (Soumerai, 2004). However, at the time, the sound theoretical basis for understanding the cost-adherence relationships among chronically ill patients was lacking. Therefore, this work of authors proved timely and crucial, both academically and politically, and built the stage for research, policy, and practice considerations to address the issue of CRNA among patients. Since then, the model has been applied and adapted widely to understand CRNA in various populations such as older adults and patients with diabetes and other chronic illnesses (Piette, 2009; Piette, Beard, Rosland, & McHorney, 2011; Piette, Wagner, Potter, & Schillinger, 2004; Wagner, Heisler, & Piette, 2008; Zhang et al., 2014).

Due to the mounting attention to the increasing costs of prescription medications in Canada over the past decade, many healthcare groups, advocacy associations, and health policy researchers have proposed different Pharmacare models for Canada (Dixon, 2013; Picard, 2018). However, to date no homogeneous analysis has been done that can
inform Canadian policymakers and researchers regarding the extent, determinants, and consequences of CRNA to prescribed medications among Canadian people. Therefore, in this study, we aim to systematically map the literature on CRNA to prescription medications in Canada. We also report the type, extent, and quantity of research available on this topic and evaluate the existing gaps.

2.4 Methods

We conducted this study using a scoping review methodology developed by Arskey and O’Malley (2005) and supplemented by Colquhoun and other colleagues (except stakeholder consultation) (Armstrong, Hall, Doyle, & Waters, 2011; Colquhoun et al., 2014; Tricco et al., 2016). The Arksey & O’Malley (2005) framework for the scoping review process defines five main stages that include identifying the research question, identifying relevant studies, selecting studies, charting the data, and then collating, summarizing, and reporting the results. The search and review criteria were developed a-priori by the authors, in consultation with a health science librarian. Two independent reviewers screened the titles and abstracts (at the first stage of screening) and full-text articles (at the second stage for inclusion or exclusion of the articles) using a predefined charting form. However, the process was not linear. Our search strategy, criteria for article selection, and format for data charting was reviewed and revised several times in an iterative manner. Any disagreements in the final article selection were resolved with the guidance of senior authors on the paper.

Stage 1: Identifying the research question. The research question guiding this scoping review was “What does the existing literature inform about the extent,
determinants, and consequences of CRNA to prescription medications in Canada?”. We included studies that described:

(1) the prevalence, frequency, and types of CRNA;
(2) the determinants of CRNA;
(3) the evidence for health and social consequences of CRNA.

The conceptual framework developed by Piette and colleagues (referred above) was used to identify and include studies and subsequent data charting and coding for analyses.

**Stage 2: Identifying relevant studies.** Studies were located through a comprehensive search of major electronic bibliographic databases and search engines that included PubMed, CINAHL, ProQuest, Science Direct, Global Health, and Google Scholar. The search was done in June 2017 and updated on February 20, 2018. Reference list searching for some of the key articles was done to identify articles that might not have emerged in the initial database search. The search terms included a combination of subject headings and free text terms- prescription fees, drug costs, prescription drugs, prescription drug cost, drug insurance, pharmaceutical services, medication adherence, cost-related non-adherence to medicines, and Canada. This combination of keywords varied to some extent as per the different indexing schemes used in each of the databases. Also, there is no uniform terminology to refer to the concept of CRNA. Therefore, we used various combinations of common key terms that are used in the Canadian studies such as “cost-related barriers to prescription drugs” (Tang et al., 2014), “prescription drug cost-related non-adherence” (Law, Cheng, Dhall, Heard, & Morgan, 2012), “cost-related prescription non-adherence” (Hennessy et al., 2016; Kennedy & Morgan, 2009),
“effect of cost-sharing on use of medication” (Ungar, Kozyrskyj, Paterson, & Ahmad, 2008), “primary non-adherence with prescribed medication” (Tamblyn, Eguale, Huang, Winslade, & Doran, 2014), “medicine underuse due to cost” (Kemp, Roughead, Preen, Glover, & Semmens, 2010), “cost-related non-adherence to prescribed medicines” (Lee & Morgan, 2017), and “prescription non-adherence due to cost” (Kennedy & Morgan, 2006). The search strategy used to identify articles from PubMed is given in Box 1.

**Stage 3: Study selection.** The articles we selected after initial screening, based on the review of titles and abstract, were further screened based on the inclusion and exclusion criteria. Articles were considered for inclusion if they focused on extent, determinants and/or consequences of the financial burden of medications, in the Canadian context. We also limited selection to articles that were peer-reviewed, published in scholarly journals, and available in English. Studies were excluded if they did not focus on Canada or did not include Canadian population. The articles that were not available in English, did not have abstract or full text available were also excluded. We did not exclude any articles based on the study design, though the papers published as editorial letters, commentaries, news articles or case studies were excluded. Articles meeting criteria were reviewed by the first author, and consensus for inclusion was reached.
through subsequent discussions with the other authors. A flowchart representing this procedure is given in Figure 2.

Figure 2. PRISMA flowchart

Stage 4: Charting the data. The variables extracted for data charting from the selected studies included author(s), year of publication, study design, sample size, population characteristics, study purpose, the focus of the article, and key outcomes or results. The data charting was done in a spreadsheet based on which an analytical synthesis was prepared.

* Focused on generic versus branded pricing and prescribing, formulary of public drug programs, prescription auditing, polypharmacy.
Stage 5: Collating, summarizing, and reporting the results. Following the recommendations of Arksey and O’Malley’s (2005), results were reported using descriptive numerical summary and thematic analysis. A summary of descriptive findings was collated from the spreadsheet and presented below and in tables. Key themes used to extract data were developed based on the research question of the study and results are presented below.

2.5 Results

The initial database search retrieved a total of 740 articles out of which 37 articles were included in the final review. A summary of the selected 37 articles is presented in Table 4.

2.5.1 Characteristics of the records included in the study

Year published. Out of 37 studies, 21 studies were published in last five years, with a maximum number of studies published in 2014 (n=5) (Campbell et al., 2014; Després et al., 2014; Rotermann, Sanmartin, Hennessy, & Arthur, 2014; Sanmartin et al., 2014; Tamblyn et al., 2014). The remaining included (n=16) studies were published between 1999 to 2012.

Study design. A large number of the studies (n=19) used data from population-based surveys (self-reported; telephone or mail-based surveys) with sample size varying between 5,000 to 70,000 people. These surveys included Canadian Community Health Survey; Barriers to Care for People with Chronic Health Conditions; National Population Health Survey; International Health Policy Survey; Canadian Health Measure Survey; Ontario Health Survey; Family Expenditure Survey; and Survey of Household Spending. Nine studies (n=9) adopted retrospective or prospective cohort designs where population
cohorts were identified using an administrative database (Anis et al., 2005; Assayag et al., 2013; Després, Forget, Kettani, & Blais, 2016; Després et al., 2014; Hunter et al., 2015; Tamblyn et al., 2014; Thanassoulis et al., 2009; Ungar et al., 2008; Yao, Lix, Shevchuk, Teare, & Blackburn, 2018).

Studies that used administrative data utilized pharmacy databases, private insurance claims, public drug benefit insurance claims or electronic health records of the patients accessing primary care in public health institutions. Two studies (n=2) used data obtained from the National Prescription Drug Utilization Information System and the Canadian Pharmacists Association (Daw & Morgan, 2012; Demers et al., 2008). Three studies (n=3) were based on qualitative methods (Dhaliwal et al., 2017; Goldsmith et al., 2017; Guilcher et al., 2017). One study (n=1) adopted a natural experimental design (Wang, Lia, Sweetman, & Hurley, 2015), while one study (n=1) was based on interrupted time series analysis (Tamblyn et al., 2001). One of the included studies (n=1) was a systematic review (Lexchin & Grootendorst, 2004) while one study (n=1) was based on a small survey (Zheng, Poulouse, Fulford, & Holbrook, 2012).

**Participant characteristics.** The majority of the studies (n=17) included individuals or family households living in Canada. Twelve studies (n=12) involved patients having chronic illnesses/conditions such as cardiovascular diseases, depression, rheumatoid arthritis, heart failure, hypertension, diabetes, and asthma (Anis et al., 2005; Assayag et al., 2013; Campbell et al., 2014; Després et al., 2016; Després et al., 2014; Dhaliwal et al., 2017; Hennessy et al., 2016; Kratzer, Cheng, Allin, & Law, 2015; Millar, 2005; Thanassoulis et al., 2009; Ungar et al., 2008; Yao et al., 2018) Within these 12 studies, five involved both younger and older adult population with chronic conditions,
three included younger adults with chronic conditions, three studies included only older adults with chronic conditions and one study focused on children (<18 years) with chronic conditions. Three studies involved patients accessing primary care (n=3) (Goldsmith et al., 2017; Tamblyn et al., 2014; Zheng et al., 2012). One study (n=1) specifically focused on older adult population (>65 years) (Lee & Morgan, 2017), one included (n=1) older adults and social assistance recipients (Tamblyn et al., 2001), one review article (n=1) focused specifically on the vulnerable groups (defined as poor and frequent drug users) (Lexchin & Grootendorst, 2004) and one study (n=1) included only younger population (between 18-64 years) (Hanley, 2009). One study (n=1) included individuals who are homeless (Hunter et al., 2015) and one study (n=1) involved healthcare professionals and policymakers focusing on people with long-term neurological conditions (Guilcher et al., 2017).

**Geographical representation.** Out of 37 studies, 14 studies (n=14) were Pan-Canadian i.e. included participants/data from all Canadian provinces. Five studies (n=5) were conducted in Ontario (Hanley, 2009; Kratzer et al., 2015; Ungar et al., 2008; Zheng et al., 2012; Zhong, 2007), six (n=6) in Quebec (Assayag et al., 2013; Després et al., 2016; Després et al., 2014; Tamblyn et al., 2014, 2001; Thanassoulis et al., 2009), one in (n=1) Saskatchewan (Yao et al., 2018), one in (n=1) British Columbia (Anis et al., 2005) and one (n=1) in Alberta (Dhaliwal et al., 2017). Five studies (n=5) compared/studied prescription drug policy of selected provinces in Canada (Campbell et al., 2014; Goldsmith et al., 2017; Hennessy et al., 2016; Hunter et al., 2015; Thanassoulis et al., 2009). Four studies (n=4) included data from several countries that compared Canada with other countries such as United States (US), United Kingdom (UK), New Zealand.
(NZ), Australia, Netherlands, and Germany (Kemp et al., 2010; Kennedy & Morgan, 2006, 2009; Lexchin & Grootendorst, 2004).

**Focus of the included studies.** Of total 37 studies, twelve studies (n=12) focused on CRNA specifically (Assayag et al., 2013; Després et al., 2016; Després et al., 2014; Goldsmith et al., 2017; Hennessy et al., 2016; Kemp et al., 2010; Kennedy & Morgan, 2006, 2009; Law et al., 2012; Lee & Morgan, 2017; Yao et al., 2018; Zheng et al., 2012). Eight studies (n=8) focused on impact or consequences of prescription medication costs on health and/or social outcomes such as access or utilization of other healthcare services (Allin & Hurley, 2009; Anis et al., 2005; Dhaliwal et al., 2017; Hanley, 2009; Law et al., 2018; Lexchin & Grootendorst, 2004; Tamblyn et al., 2001; Wang et al., 2015). Six studies (n=6) analyzed the effects of having prescription drug insurance on prescription drug use (Demers et al., 2008; Dewa et al., 2005; Kapur & Basu, 2005; Kratzer et al., 2015; Millar, 2005; Thanassoulis et al., 2009) while four studies (n=4) focused on the impact of out-of-pocket costs and income on prescription drug use (Rotermann et al., 2014; Sanmartín et al., 2014; Ungar et al., 2008; Zhong, 2007) Three studies (n=3) analyzed and reviewed out-of-pocket costs of prescriptions including catastrophic drug costs across provinces or provincial drug benefit programs (Luffman, 2005; McLeod, Bereza, Shim, & Grootendorst, 2011; Sanmartín et al., 2014). Two studies (n=2) focused on general non-adherence with cost-related non-adherence to medications as one of the elements (Hunter et al., 2015; Tamblyn et al., 2014). Two studies (n=2) discussed general cost-related barriers to healthcare including financial burden of prescription medication costs (Campbell et al., 2014; Dhaliwal et al., 2017). One study (n=1) collected the
perspectives of various stakeholders on the affordability of necessary prescription medications for people with neurological conditions in Canada (Guilcher et al., 2017).

**Measurement/operationalization of CRNA.** The most common method used to measure CRNA was based on a survey question which asked participants: “During the past 12 months, have you ever taken less of your medication than prescribed because of cost such as skipping doses or not filling a prescription?” (n=6) (Hennessy et al., 2016; Kemp et al., 2010; Kennedy & Morgan, 2009; Law et al., 2012; Lee & Morgan, 2017; Zheng et al., 2012). Three studies (n=3) used ‘proportion or number of days covered’ method called as PDC/PPDC that measured adherence through the number of days covered by prescription refills over 1 year (Assayag et al., 2013; Després et al., 2016; Després et al., 2014). One study (n=1) defined CRNA as failure to obtain prescribed medication due to cost in the prior month (Kennedy & Morgan, 2006). One study (n=1) measured adherence over 365 days using medication possession ratio (MPR) (Yao et al., 2018). One qualitative study (n=1) focused on developing the typology of CRNA but did not mention any particular way of measuring it (Goldsmith et al., 2017)
Table 4. Descriptive summary of the studies included in the review

<table>
<thead>
<tr>
<th>Article</th>
<th>Province/Country</th>
<th>Population characteristic</th>
<th>Sample Size</th>
<th>Study design</th>
<th>Focus of the study</th>
<th>Outcome measurement</th>
</tr>
</thead>
<tbody>
<tr>
<td>Campbell et al, 2014</td>
<td>Manitoba, SSK, Alberta, BC</td>
<td>Adults with chronic conditions</td>
<td>N= 1849</td>
<td>Population based survey</td>
<td>Financial barriers to care including prescriptions</td>
<td>CRNA defined as stopped taking one or more medications for at least a week in last 12 months due to cost.</td>
</tr>
<tr>
<td>Hennessy et al, 2016</td>
<td>Manitoba, SSK, Alberta, BC</td>
<td>Adults with chronic conditions</td>
<td>N= 1849</td>
<td>Population based survey</td>
<td>OOP spending on prescriptions and CRNA</td>
<td>CRNA defined if patients did not get drugs needed due to cost in the past 12 months.</td>
</tr>
<tr>
<td>Tamblyn et al, 2014</td>
<td>Quebec</td>
<td>Patients accessing primary care</td>
<td>N= 15961</td>
<td>Retrospective cohort study</td>
<td>Medication non-adherence with CRNA as one of the elements</td>
<td>Non-adherence defined as not filling incident prescription within 9 months.</td>
</tr>
<tr>
<td>Hunter et al, 2015</td>
<td>ON and BC</td>
<td>Homeless and vulnerably housed adults</td>
<td>N= 716</td>
<td>Prospective cohort study</td>
<td>Medication non-adherence with CRNA as one of the elements</td>
<td>CRNA defined as not taking medication prescribed by a doctor because it is too expensive</td>
</tr>
<tr>
<td>Kratzer, Cheng, Allin, and Law, 2015</td>
<td>ON</td>
<td>Adults with chronic conditions</td>
<td>N= 2161311</td>
<td>Population based survey</td>
<td>Effects of private drug coverage on prescription use</td>
<td>Drug use was defined as having drugs in the past month and drug coverage was defined if private insurance covered all or part of the prescription medication cost.</td>
</tr>
<tr>
<td>Ungar et al, 2008</td>
<td>ON</td>
<td>Children with Asthma</td>
<td>N= 17046</td>
<td>Retrospective cohort study using administrative databases</td>
<td>Effect of cost sharing on prescription use</td>
<td>Cost-sharing levels were categorized as: zero cost sharing, less than 20% (low cost-sharing), and 20% or greater (high cost-sharing).</td>
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<tr>
<td>Article</td>
<td>Province/Country</td>
<td>Population characteristic</td>
<td>Sample Size</td>
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<td>Outcome measurement</td>
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<tr>
<td>McLeod et al, 2011</td>
<td>Pan-Canada</td>
<td>General Canadian population</td>
<td>N= 14430</td>
<td>Population based survey</td>
<td>Financial burden of prescription drug spending</td>
<td>Catastrophic OOP drug expenditure defined as households with drug budget share equal to or greater than 10%.</td>
</tr>
<tr>
<td>Després, Forget, Kettani, &amp; Blais, 2016</td>
<td>Quebec</td>
<td>Young adults with chronic conditions</td>
<td>N= 2872</td>
<td>Retrospective cohort study</td>
<td>Effects of OOP costs on adherence in privately insured patients</td>
<td>Adherence defined as proportion of days covered over 1 year (PDC)-the number of days supply of the medication during the follow-up period divided by the number of days of follow-up.</td>
</tr>
<tr>
<td>Després et al, 2014</td>
<td>Quebec</td>
<td>Young adults with chronic conditions</td>
<td>N= 1933</td>
<td>Retrospective cohort study</td>
<td>Effects of OOP costs on adherence in public and privately insured patients</td>
<td>Adherence defined as proportion of days covered over 1 year</td>
</tr>
<tr>
<td>Kennedy &amp; Morgan, 2009</td>
<td>Canada and USA</td>
<td>Adult American and Canadian population</td>
<td>N= 2980</td>
<td>Population based survey</td>
<td>Compare rates of CRNA for prescription drugs in USA and Canada</td>
<td>CRNA identified if participant responded yes to &quot;During the past 12 months, was there a time when you did not fill a prescription, or you skipped doses of your medicine, due to cost?”</td>
</tr>
<tr>
<td>Thanassoulis et al, 2009</td>
<td>Quebec, ON and BC</td>
<td>Older adults with chronic conditions</td>
<td>N= 67040</td>
<td>Cohort study using administrative data</td>
<td>Impact of type of drug coverage on medication use</td>
<td>Drug use was determined at 30 days of discharge, stratified by prescription plan in each province</td>
</tr>
</tbody>
</table>
- Direct expenditures defined as those not covered by insurance,      |
<table>
<thead>
<tr>
<th>Article</th>
<th>Province/Country</th>
<th>Population characteristic</th>
<th>Sample Size</th>
<th>Study design</th>
<th>Focus of the study</th>
<th>Outcome measurement</th>
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</thead>
<tbody>
<tr>
<td>Roterman, Sanmartin, Henessy, &amp; Arthur, 2014</td>
<td>Pan-Canada</td>
<td>General population</td>
<td>N= 11386</td>
<td>Population based survey</td>
<td>Determinants of prescription medication use including HH income</td>
<td>Drug use was determined if respondents had taken at least one prescription medication within two days of their household interview.</td>
</tr>
<tr>
<td>Allin &amp; Hurley, 2009</td>
<td>Pan-Canada</td>
<td>General population</td>
<td>N= 33161</td>
<td>Population based survey</td>
<td>Impact of drug coverage on physician utilization</td>
<td>Physician utilization measured by asking if person has seen a family doctor or specialist in last 12 months.</td>
</tr>
<tr>
<td>Kapur &amp; Basu, 2004</td>
<td>Pan-Canada</td>
<td>General population</td>
<td>N= n/a</td>
<td>Population based survey</td>
<td>Extent of drug coverage and financial burden of prescription drugs</td>
<td>Financial burden of prescription drugs calculated as out-of-pocket drug expenses of households as a proportion of household income.</td>
</tr>
<tr>
<td>Law, Cheng, Dhall, Heard, &amp; Morgan, 2012</td>
<td>Pan-Canada</td>
<td>General population</td>
<td>N= 5732</td>
<td>Population based survey</td>
<td>Extent and determinants of CRNA</td>
<td>CRNA defined as if costs led people who reported taking medication in past year to do anything to make their prescription last longer, not fill a new prescription or not renew a prescription.</td>
</tr>
<tr>
<td>Zhong, 2007</td>
<td>ON</td>
<td>General population</td>
<td>N= &gt;60,000</td>
<td>Population based survey</td>
<td>Inequality in drug use with respect to income</td>
<td>Drug utilization was determined by asking the participants: &quot;How many different numbers of prescription drugs have you taken in the last four weeks?&quot;</td>
</tr>
<tr>
<td>Article</td>
<td>Province/Country</td>
<td>Population characteristic</td>
<td>Sample Size</td>
<td>Study design</td>
<td>Focus of the study</td>
<td>Outcome measurement</td>
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<tr>
<td>Millar, 1999</td>
<td>Pan-Canada</td>
<td>Population diagnosed with a chronic disease</td>
<td>N= 70884</td>
<td>Population based survey</td>
<td>Availability of drug insurance and its effect on prescription drug use</td>
<td>Number of drugs taken in the past month used as an indicator of the influence of drug insurance coverage on medication use.</td>
</tr>
<tr>
<td>Luffman, 2005</td>
<td>Pan-Canada</td>
<td>General population</td>
<td>N= &gt; 20,000 HH</td>
<td>Population based survey</td>
<td>OOP prescription drug spending across various provinces</td>
<td>OOP drug spending referred to expenditures for medicines, drugs and pharmaceutical products prescribed by a doctor such as exclusions, deductibles and expenses over limits.</td>
</tr>
<tr>
<td>Lee &amp; Morgan, 2017</td>
<td>Pan-Canada</td>
<td>Older adult population</td>
<td>N= 5269</td>
<td>Population based survey</td>
<td>CRNA and its determinants</td>
<td>CRNA defined as not filling a prescription or skipping doses within the last 12 months because of OOP costs, among those who reported taking at-least 1 prescription.</td>
</tr>
<tr>
<td>Kemp et al, 2010</td>
<td>Australia, Canada, UK, USA, Netherlands, New Zealand, Germany</td>
<td>General population</td>
<td>N= 8898</td>
<td>Population based survey</td>
<td>CRNA and its determinants across countries</td>
<td>Cost related medication underuse assessed if there was a time in the last 12 months when respondent did not collect a prescription or skipped doses because of the cost? (yes/no).</td>
</tr>
<tr>
<td>Hanley, 2009</td>
<td>ON</td>
<td>Young adult population</td>
<td>N= 31630</td>
<td>Population based survey</td>
<td>Impact of prescription drug insurance on unmet healthcare needs</td>
<td>Unmet healthcare need was identified if participants decided not to seek care because he or she anticipated that a visit to a physician would result in a prescription.</td>
</tr>
<tr>
<td>Article</td>
<td>Province/Country</td>
<td>Population characteristic</td>
<td>Sample Size</td>
<td>Study design</td>
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<tr>
<td>Dhaliwal et al, 2017</td>
<td>Alberta</td>
<td>Individuals with heart disease</td>
<td>N= 13</td>
<td>Qualitative study</td>
<td>Experiences of patients who reported financial barriers to care including prescriptions</td>
<td>Cost-related non-adherence was identified if participants shared that they forgo their pills if they cannot afford it.</td>
</tr>
<tr>
<td>Dewa, Hoch, &amp; Steele, 2005</td>
<td>Pan-Canada</td>
<td>Community dwelling Canadians</td>
<td>N= 33,000</td>
<td>Population based survey</td>
<td>Characteristics of people covered or not covered for public prescription drug insurance</td>
<td>Having a drug insurance was identified if participant said yes to “Do you have insurance that covers all or part of the costs of your prescription medications?”</td>
</tr>
<tr>
<td>Demers et al, 2008</td>
<td>Pan-Canada</td>
<td>Individuals including social assistance recipients</td>
<td>N= 32* scenarios</td>
<td>Policy analysis</td>
<td>To examine the impact of variation in provincially funded public drug benefits on patients’ prescription drug costs having similar prescription needs</td>
<td>Cost-sharing strategies were examined in the form of premium, deductible, copayment and maximum annual contribution by the beneficiary; and pharmacists’ dispensing fees.</td>
</tr>
<tr>
<td>Kennedy &amp; Morgan, 2006</td>
<td>Canada and USA</td>
<td>Adult American and Canadian population</td>
<td>N= 8688</td>
<td>Population based survey</td>
<td>Extent and determinants of CRNA in two countries</td>
<td>CRNA was measured as failure to obtain prescribed medication due to cost in the prior month.</td>
</tr>
<tr>
<td>Guilcher et al, 2017</td>
<td>Pan-Canada</td>
<td>Health care professional, non-health professional, policymakers</td>
<td>N=180</td>
<td>Qualitative study</td>
<td>Perspectives of key stakeholders about the availability and use of prescription drugs for neurological conditions</td>
<td>n/a</td>
</tr>
<tr>
<td>Article</td>
<td>Province/Country</td>
<td>Population characteristic</td>
<td>Sample Size</td>
<td>Study design</td>
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<tr>
<td>Wang, Li, Sweetman, &amp; Hurley, 2015</td>
<td>Pan-Canada (Quebec versus rest of Canada)</td>
<td>Canadians between age of 12-64</td>
<td>N=10,653</td>
<td>Experimental study</td>
<td>Impact of universal prescription drug insurance on healthcare utilization and health outcomes</td>
<td>Drug insurance status was determined if a person reported having drug insurance. Measures of health care utilization include drug utilization, physician visits and hospitalization. Drug utilization measures the number of distinct medications taken in the previous month.</td>
</tr>
<tr>
<td>Zheng et al, 2012</td>
<td>ON</td>
<td>Patients visiting an outpatient clinic</td>
<td>N= 60</td>
<td>Small Survey</td>
<td>Predictors leading to CRNA</td>
<td>CRNA was assessed by asking patients to think for last 12 months and describe how frequently they left prescriptions unfilled, delayed filling prescriptions, took prescriptions with reduced frequency and lowered dosages because of the cost.</td>
</tr>
<tr>
<td>Goldsmith et al, 2017</td>
<td>ON and BC</td>
<td>Adults engaging in CRNA in past or currently</td>
<td>N=35</td>
<td>Qualitative study</td>
<td>Understand patients experiences of CRNA through typology development</td>
<td>CRNA was explored by asking participants’ most recent experience with stopping, reducing, or not filling a prescription medication due to out-of-pocket costs.</td>
</tr>
<tr>
<td>Yao et al, 2018</td>
<td>SSK</td>
<td>Older adults with chronic illnesses</td>
<td>N= 188109</td>
<td>Retrospective cohort study</td>
<td>Quantify the impact of drug benefit plan on medication adherence that capped out-of-pocket costs at $15 per prescriptions</td>
<td>Adherence was measured over 365 days using medication possession ratio (MPR).</td>
</tr>
<tr>
<td>Assayag et al, 2012</td>
<td>Quebec</td>
<td>Younger patients with</td>
<td>N= 2249</td>
<td>Matched cohort study</td>
<td>Adherence between privately and publicly</td>
<td>Adherence over 1 year was estimated using the proportion of</td>
</tr>
<tr>
<td>Article</td>
<td>Province/Country</td>
<td>Population characteristic</td>
<td>Sample Size</td>
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<tr>
<td>Daw &amp; Morgan, 2012</td>
<td>Pan-Canada</td>
<td>N/a</td>
<td>N= n/a</td>
<td>Analyses of administrative database</td>
<td>Review of provincial drug benefit programs to find factors leading to catastrophic drug expenditures</td>
<td>Assessed premiums and cost-sharing mechanisms in the form the expenses borne by the patient (or via supplementary private insurance): deductibles, copayments/co-insurance, and out-of-pocket limits to find catastrophic drug expenditures.</td>
</tr>
<tr>
<td>Anis et al, 2005</td>
<td>BC</td>
<td>Older adult patients with rheumatoid arthritis</td>
<td>N= 2968</td>
<td>Retrospective cohort study using administrative database</td>
<td>Effect of prescription drug cost-sharing on overall health care utilization</td>
<td>In people who reached the annual maximum copayment of $200 for any calendar year from 1997 to 2000, the outcomes assessed were number of hospital admissions, the number of physician visits and the total number of prescriptions filled.</td>
</tr>
<tr>
<td>Tamblyn et al, 2001</td>
<td>Quebec</td>
<td>Older adults and welfare recipients</td>
<td>N= 93950 elderly persons and 55333 adults on welfare</td>
<td>Interrupted time series analysis</td>
<td>Adverse effects of prescription drug cost-sharing on overall health care utilization</td>
<td>Mean number of drugs used per month, ED visits, and hospitalization, nursing home admission, and mortality before and after policy introduction.</td>
</tr>
<tr>
<td>Lexchin &amp; Grootendorst (2004)</td>
<td>Industrialized countries including Canada</td>
<td>Vulnerable population (poor and with chronic health conditions)</td>
<td>N=25 studies</td>
<td>Systematic review of literature</td>
<td>Effects of user fees for prescription drugs on drug use, other health services use, and overall health status</td>
<td>User fee included costs that patients pay out-of-their pockets for prescriptions such as in the form of copayment, deductibles, reimbursement limits etc.</td>
</tr>
<tr>
<td>Article</td>
<td>Province/Country</td>
<td>Population characteristic</td>
<td>Sample Size</td>
<td>Study design</td>
<td>Focus of the study</td>
<td>Outcome measurement</td>
</tr>
<tr>
<td>--------------</td>
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<td>---------------------------------------------------------</td>
<td>-------------------------------------------------------------------------------------</td>
</tr>
<tr>
<td>Law et al, 2018</td>
<td>Pan-Canada</td>
<td>General population</td>
<td>N=28091</td>
<td>Population based survey</td>
<td>Consequences for prescription drug costs</td>
<td>Health consequences that led to use of additional health care services such as a doctor visit or ED visit, and other consequences leading to trade-offs between prescriptions and other basic needs.</td>
</tr>
</tbody>
</table>

**Notes:** **Abbreviations:** BC, British Columbia; ED, Emergency Department; HH, household; OOP, out-of-pocket; ON, Ontario; SSK, Saskatchewan
2.5.2 Thematic Analysis

Based on the research question for the study, we used three themes related to extent, determinants, and consequences of CRNA to medications to prepare the data synthesis. These themes are represented and discussed here.

**Extent of cost-related non-adherence to prescription medications.** Out of total studies included, 9 studies measured the prevalence of CRNA within their participant population which varied between 4.1% to 35.6%, depending on the participant characteristics and provinces (Table 5) (Assayag et al., 2013; Campbell et al., 2014; Hennessy et al., 2016; Hunter et al., 2015; Law et al., 2012, 2018; Lee & Morgan, 2017; Tamblyn et al., 2014; Zheng et al., 2012).

Overall, the national prevalence of CRNA including general population was reported at 9.6% in 2012 (Law et al., 2012), which decreased slightly to 8.2% in 2018 as reported by the same group of authors in their recent study (Law et al., 2018). Further, in 2012, the extent of CRNA varied between 3.6% (95% CI 2.4-4.5) to 35.6% (95% CI 26.1%-44.9%) depending on income and availability of insurance (Law et al., 2012). This national study also found that rates of CRNA were lowest in Quebec (7.2%, 95% CI 4.5%-9.8%) and highest in British Columbia (17.0%, 95% CI 12.6%-21.4%) (Law et al., 2012).
Table 5. Extent of cost-related non-adherence to prescription medications

<table>
<thead>
<tr>
<th>Extent</th>
<th>Population</th>
<th>Province</th>
<th>Study</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>9.6% reported CRNA to medications ranging from 3.6% (95% CI 2.4-4.5) to 35.6% (95% CI 26.1%-44.9%) depending on income and availability of insurance.</td>
<td>General population</td>
<td>Pan-Canada</td>
</tr>
<tr>
<td>2</td>
<td>8.2% reported CRNA to medications who were prescribed at least 1 medication in last 12 months</td>
<td>General population</td>
<td>Pan-Canada</td>
</tr>
<tr>
<td>3</td>
<td>8.3% Canadians aged 55 years and older reported CRNA to medications</td>
<td>Senior population</td>
<td>Pan-Canada</td>
</tr>
<tr>
<td>4</td>
<td>15% reported CRNA to medications</td>
<td>Patients visiting out-patient clinic</td>
<td>Ontario</td>
</tr>
<tr>
<td>5</td>
<td>Prevalence of CRNA between privately and publicly insured individuals was 14% and 18% respectively</td>
<td>Non-senior patients with depression</td>
<td>Quebec</td>
</tr>
<tr>
<td>6</td>
<td>4.1% reported CRNA to medications</td>
<td>Adults with chronic conditions</td>
<td>Manitoba, SSK, Alberta, BC</td>
</tr>
<tr>
<td>7</td>
<td>13% reported stopped taking medications in last 12 months at least for a week due to cost</td>
<td>Adults with chronic conditions</td>
<td>Manitoba, SSK, Alberta, BC</td>
</tr>
<tr>
<td>8</td>
<td>31.3% of the incident prescriptions were not filled in the last 9 months</td>
<td>Patients accessing primary care</td>
<td>Quebec</td>
</tr>
<tr>
<td>9</td>
<td>26% reported CRNA to prescriptions</td>
<td>Homeless and vulnerably housed adults</td>
<td>Ontario and British Columbia</td>
</tr>
</tbody>
</table>

Focusing on older adult population, another recent national study reported that around 1 in 12 (8.3%) Canadians aged 55 years and older faced CRNA to prescription medications in 2014 (Lee & Morgan, 2017). Among people accessing primary care in Quebec, prevalence of CRNA was reported at 31.3% (Tamblyn et al., 2014). Among those who were homeless and precariously housed, CRNA was experienced by 26% of
the participants, residing in Ontario and British Columbia (Hunter et al., 2015). A study involving people with cardiovascular-related chronic conditions across four provinces (Manitoba, Saskatchewan, Alberta, British Columbia) reported that around 14% of the participants reported lack of drug insurance coverage out of which 4.1% faced financial barriers to accessing medications leading to non-adherence (Hennessy et al., 2016), while another study conducted with similar population in these four provinces reported that 13% of the respondents had stopped taking medications due to cost (Campbell et al., 2014).

**Determinants of cost-related non-adherence to prescription medications.**

Twenty-nine (n=29) studies identified the most common factors that predict someone’s risk of facing CRNA to medications. We describe these factors under four categories as per the Piette Model.

**Person/patient-related factors.** Within the person/patient-related factors, lower household or personal income (specifically below CAD 30,000 a year) and lack of regular employment are the primary predictors of CRNA reported by the highest number of studies (n=15). After individuals’ income and employment status; young age (i.e. <55 years) (n=12); poor health status (i.e. having a chronic illness, severe health condition or multiple co-morbidities) (n=10); and not having drug insurance (n=9) are the most common person-related factors that lead individuals to forgo their medications or skip or split doses due to cost. The national level studies suggest that Canadians who are younger, in worse health, have lower income, precarious or irregular employment and no drug insurance are most likely to face cost-related barriers to their prescriptions (Guilcher
et al., 2017; Kennedy & Morgan, 2006; Law et al., 2012; Lee & Morgan, 2017; Millar, 2005).

Having prescription drug insurance was also reported to be significantly associated with having access to prescription medication without financial barriers (Goldsmith et al., 2017; Kennedy & Morgan, 2006; Law et al., 2012, 2018; Lee & Morgan, 2017; Millar, 2005; Rotermann et al., 2014; Zheng et al., 2012; Zhong, 2007). A recent qualitative study exploring the typology of CRNA among adults who reported engaging in CRNA found that an array of factors such as individuals’ financial flexibility, the importance of the drug, burden of the drug costs, and having insurance interact with each other and influence CRNA in individuals (Goldsmith et al., 2017). A number of studies also reported that people with chronic conditions holding private drug insurance were more likely to use prescription drugs than those having public drug insurance (Després et al., 2014; Kemp et al., 2010; Kratzer et al., 2015; Lee & Morgan, 2017; Zheng et al., 2012). A study analysing risk of not having prescription drug insurance coverage reported that people residing in one of the Atlantic Provinces, Manitoba, or Saskatchewan; were young (less than age 25 or to a lesser extent age 25-34); had no post-secondary education; self-employed; working part-year or part-time; single persons living on their own; living in a rural area; and/or belong to households with lower middle income had a higher risk of not having prescription drug insurance coverage (Kapur & Basu, 2005).

Other factors that were reported in only a few studies included sex, education, relationship status, ethnicity, and place of residence. These studies reported that being female (Kennedy & Morgan, 2009; Law et al., 2018), having education less than high
school (Kapur & Basu, 2005; Zhong, 2007), living alone (Dewa et al., 2005; Kapur & Basu, 2005), being non-white, immigrant or aboriginal status (Dewa et al., 2005; Law et al., 2018; Zhong, 2007), and living in a rural area (Kapur & Basu, 2005) increased the risk of facing financial barriers to medications (Table 6).

Table 6. Factors associated with cost-related non-adherence

<table>
<thead>
<tr>
<th>Factors</th>
<th>Studies</th>
</tr>
</thead>
<tbody>
<tr>
<td>8 Race and Ethnicity: Aboriginal status, being non-white or immigrant (n=3)</td>
<td>Law et al, 2018; Dewa, Hotch, &amp; Steele, 2005; Zhong 2007</td>
</tr>
</tbody>
</table>
Factors | Studies
--- | ---
9 | Not having a primary physician (n=3) Hunter et al, 2014; Tamblyn et al, 2014; Kennedy & Morgan, 2006
10 | Women (Sex) (n=2) Kennedy & Morgan, 2009; Law et al, 2018
11 | Less Education (n=2) Kapur & Basu, 2004; Zhong 2007
12 | Living alone or not having spouse or partner (n=2) Kapur & Basu, 2004; Dewa, Hotch, & Steele, 2005
13 | Living in a rural area (n=1) Kapur & Basu, 2004

**Drug-related factors.** Within drug-related factors, high drug costs (i.e. >5% of annual household income or >$20 a month out-of-pocket) (n=10) were one of the major determinants of CRNA. In a number of studies, out-of-pocket spending between 5% to 20% on drugs predicted patients’ behaviour related to medication adherence (Campbell et al., 2014; Daw & Morgan, 2012; Demers et al., 2008; Després et al., 2016; Goldsmith et al., 2017; Hennessy et al., 2016; Kemp et al., 2010; Tamblyn et al., 2014; Ungar et al., 2008; Zheng et al., 2012). For example, three studies including people with cardiovascular conditions found that those spending more than 5% costs of medications out of their pocket were more likely to report CRNA than those spending less than 5% (Després et al., 2014; Després et al., 2016; Hennessy et al., 2016). Another study also reported that drugs in the upper quartile of cost were least likely to be filled (Tamblyn et al., 2014) in congruence with another smaller survey which found that spending more than $100 a month out-of-pocket increased the likelihood of CRNA among patients (Zheng et al., 2012). Quantifying the impact of senior drug benefit plan launched in 2007 that capped out-of-pocket costs at $15 per prescription for seniors on chronic medication reported that the impact of the program on adherence was consistently demonstrated in subgroups of patients receiving medications costing between $16 and $30 and those
costing ≥ $30 (Yao et al., 2018). A large retrospective study involving patients accessing primary care in Quebec also found that costly drugs and copayments for low-income groups (along with young age and more severe co-morbidities) were significantly associated with non-adherence (Tamblyn et al., 2014).

**Health-system related factors.** A large number of studies (n=11) focused on the effects of variations in public drug benefit programs on prescription drug costs (Campbell et al., 2014; Demers et al., 2008; Dewa et al., 2005; Hennessy et al., 2016; McLeod 2011; Thanassoulis 2009; Kapur & Basu, 2004; Law et al, 2012; Luffman, 2005; Daw & Morgan, 2012; Law et al, 2018). These variations in the provincial drug benefit programs were found to be having significant implications on the costs that patients pay for same medications and hence the extent of CRNA they face. For instance, McLeod et al (2011) found substantial interprovincial variation in the prevalence of catastrophic prescription drug costs paid by senior and social assistance households (McLeod et al., 2011). Demers and colleagues (2008) found that the eligibility criteria and cost-sharing mechanisms of public drug programs differed markedly across provinces, resulting in people with the same prescription needs bearing different financial costs (Demers et al., 2008). They found that older adults paid 35% or less of their prescription costs in 2 provinces, but elsewhere they may pay as much as 100%. With few exceptions, younger adults paid more than 35% of their prescription costs in every province, while most social assistance recipients paid 35% or less of their prescription costs in five provinces and pay no costs in the other five. In 2002, Ontario residents spent the least out-of-pocket cost for prescription drugs (less than CAD 300) while Saskatchewan spending the most (more than CAD 400). Families in Alberta, British Columbia, and Quebec spent less (between
CAD 300-350) than those in the Atlantic provinces (more than CAD 400) and Manitoba (between CAD 350-400); reflecting the differences in prescription drug coverage, employment status, health status and age structure of the provincial population (Luffman, 2005).

Three studies compared the reasons of self-reported medicine underuse due to cost across different countries having different health systems including Canada (Kemp et al., 2010; Kennedy & Morgan, 2006, 2009). An international study comparing Canada and six other countries found that approximately one-fifth of the respondents in Australia, Canada, NZ, the UK, and the US, with annual out-of-pocket costs over $500 reported under-using medicines due to cost (Kemp et al., 2010). This study found that cost-related underuse of medicines was least common in countries with lowest out-of-pocket costs and reduced copayments or cost ceilings for low income patients, the Netherlands and the UK. Analysis from two other international health policy surveys between Canada and the US found that Canadians were less likely than Americans to report CRNA (5.1% vs 9.9%; P < 0.001 in 2001 and 23.1% vs 8.0%; P < 0.001 in 2007); however, in both the countries, people without prescription-drug coverage were significantly more likely than those with insurance to report CRNA (Kennedy & Morgan, 2006, 2009).

One qualitative study collected data from policymakers and healthcare professionals and highlighted the effect of governance and structure of drug programs in Canada on access to drugs for individuals with neurological conditions. The study identified various factors such as shortage on drug formulary listings, lengthy processes for new drug approvals, the complexity of applying and confirming eligibility for coverage, piecemeal coverage across jurisdictions, and lack of collaboration among
public, private and industry sectors that affected access to prescription drugs for people with neurological conditions in Canada (Guilcher et al., 2017). The study reported that “participants identified frustrations with respect to the lack of standardization among Canadian jurisdictions as to which drugs are publicly covered under the provincial and territorial formularies” (p.393) and concluded that “these differences can impact choice of permanent residence, as participants described individuals relocating within Canada in order to obtain better drug coverage” (p.392) (Guilcher et al., 2017).

**Clinician-related factors.** Of 37 studies included in this review, three studies explored clinician/physician-related factors determining cost-adherence relationship for patients (Hunter et al., 2015; Kennedy & Morgan, 2006; Tamblyn et al., 2014). A retrospective study involving more than 15,000 patients accessing primary care reported that patients who had a greater proportion of physician visits with the prescribing physician had lower odds of non-adherence (Tamblyn et al., 2014). Similarly, among individuals who were homeless, a study found that lack of access to a physician was one among the most common reasons identified by participants for not adhering to their medications (Hunter et al., 2015). However, Kennedy and Morgan (2006) reported that the number of physician visits did not significantly predict non-adherence after controlling for other factors (Kennedy & Morgan, 2006). We did not find any other studies that explored the effect of support or propensity of prescribing healthcare professionals to take into account financial situations of patients on decisions that patients make to manage prescription cost burden.
Consequences of cost-related non-adherence to prescription medications. Of the 37 articles included, 9 studies discussed the potential impact that prescription drug costs can have on individual health or social outcomes (Table 7).

Health consequences. Evidence regarding the impact of CRNA on individual health outcomes such as disease exacerbation, poor self-reported health, increase in symptoms leading to increasing hospitalizations, emergency department visits, or mortality was limited and mixed. Of 7 studies that we found on this topic, three studies explored the effects of having drug insurance coverage on utilization of other healthcare services (Allin & Hurley, 2009; Hanley, 2009; Wang et al., 2015); three explored the effects of copayments or cost-sharing for drugs on the utilization of other healthcare services (Anis et al., 2005; Lexchin & Grootendorst, 2004; Tamblyn et al., 2001); and one explored the association between CRNA and health service utilization (Campbell et al., 2014).

A recent study by Wang and colleagues (2015) found that a mandatory universal drug insurance program substantially increased the physician visits among Canadians aged between 12-64 years. A study examining the impact of private drug financing on the utilization of physician services in Ontario revealed that people with prescription drug insurance make more physician visits than those without insurance (Allin & Hurley, 2009). A study by Hanley (2009) found that adults who did not have prescription drug insurance were 1.27 times more likely to report an unmet need for health care than those with insurance (Hanley, 2009).

Four studies reported that cost sharing for drugs in the form of copayments leads patients to forgoing essential medications and a decline in health care status, especially in
the vulnerable population (Lexchin & Grootendorst, 2004). A study conducted with elderly and social assistance recipients in Quebec in 2001 found that after copayments were introduced, prescription drug use reduced by 9% and 16% respectively which was further associated with increased rate of emergency department visits in this population (Tamblyn et al., 2001). Another study compared the prescription drug use, physician service utilization and hospital admissions among elderly patients with Rheumatoid Arthritis when they paid a co-payment for medications (cost-sharing period) versus when they did not (free period) (Anis et al., 2005) The study found that during the cost-sharing period, there were increased physician visits, fewer prescriptions filled and increased hospital admissions per month as compared to free period (Anis et al., 2005). Another study including people with cardiovascular conditions found that although self-reported financial barriers to drugs were not significantly associated with increased emergency department visits or hospitalizations, patients facing financial barriers to medications were 50% less likely to take statins (Campbell et al., 2014).

**Social Consequences.** Of total 9 studies we found on consequences of CRNA, only two (n=2) discussed the social outcomes and/or strategies adopted by the individuals to cope and manage the medication costs (Dhaliwal et al., 2017; Law et al., 2018). A recent national study highlighted the strategies or behaviours that patients adopt or engage in to manage the costs of their medications. Law and colleagues (2018) drawing from the Canadian Community Health Survey 2016 reported that around 1.2 million Canadians forwent basic needs such as food, heat and other health care expenses because of drug costs; and more than 100,000 reported an additional physician visit, emergency department visit, and hospital stay due to CRNA (Law et al., 2018). This was supported
by a qualitative study collecting data from individuals with heart diseases in Alberta which reported that individuals who faced financial barriers to medications prioritized essential medication over other non-essential medications and over healthy food and faced adverse clinical outcomes due to non-adherence to medications associated with cost (Dhaliwal et al., 2017).

<table>
<thead>
<tr>
<th>Impact/Consequences</th>
<th>Population</th>
<th>Province</th>
<th>Study</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>General population</td>
<td>Pan-Canada</td>
<td>Allin &amp; Hurley, 2009</td>
</tr>
<tr>
<td>2</td>
<td>Non-senior population</td>
<td>Pan-Canada</td>
<td>Hanley, 2009</td>
</tr>
<tr>
<td>3</td>
<td>Canadians between age of 12-64</td>
<td>Pan-Canada</td>
<td>Wang et al, 2015</td>
</tr>
<tr>
<td>4</td>
<td>Elderly patients with rheumatoid arthritis</td>
<td>British Columbia</td>
<td>Anis et al, 2005</td>
</tr>
<tr>
<td>5</td>
<td>Elderly and welfare recipients</td>
<td>Quebec</td>
<td>Tamblyn et al, 2001</td>
</tr>
</tbody>
</table>
Impact/Consequences | Population | Province | Study |
--- | --- | --- | --- |
6 | Cost sharing leads patients forgoing essential medications and to a decline in health care status. Copayments or a cap on the monthly number of subsidized prescriptions lower drug costs for the payer, but any savings are offset by increases in other health care areas. | Vulnerable population | OECD Countries including Canada | Lexchin & Grootendorst, 2004 |
7 | Many Canadians forwent basic needs such as food (about 730000 people), heat (about 238000) and other health care expenses (about 239000) because of drug costs. | General population | Pan-Canada | Law et al, 2018 |
8 | Some participants identified that their cost-related non-adherence led to adverse clinical outcomes. Some of them also ‘separated’ medications into essential and non-essential categories and prioritize medications over healthy food. | Individuals with heart disease | Alberta | Dhaliwal et al, 2017 |
9 | Self-reported financial barriers to drugs were not found significantly associated with increased number of emergency department visits or hospitalizations, though patients facing financial barriers to take medications were 50% less likely to take medications. | Adults with chronic conditions | Manitoba, SSK, Alberta, BC | Campbell et al, 2014 |

2.6 Discussion

The growing evidence on the barriers faced by Canadians to fulfil necessary medications has given a strong impetus to the national Pharmacare debate. Many negotiations and discussions are underway calling for a solution to this big drug-problem in Canada, and there has been a wide difference in the suggested propositions. Our study informs this conversation by answering three key questions that are at the core of this national discourse: first, how many Canadians face CRNA to fulfil their medications; second, who are at risk of facing CRNA; and third, what outcomes or consequences they face or might face due to CRNA. Answers to these questions will help policymakers and researchers in agenda-setting and future policy and programme development. In this
scoping study, we found 37 articles that offered some evidence on the extent, determinants and consequences of CRNA to prescription medications in Canada. Findings suggest that:

- Depending on the population characteristics and provinces, the prevalence of CRNA varies between 4% to 36%.

- The most common factors associated with CRNA include age (between 18-64 years), employment status or income, health status, lack of insurance coverage, and high out-of-pocket cost of medications. Though these factors may be confounded with each other.

- Evidence on the impact of CRNA to prescriptions on individual health outcomes such as disease exacerbation, poor self-reported health, increase in symptoms leading to an increase in hospitalizations or emergency department visits or mortality is limited and mixed.

- The literature regarding social outcomes and/or strategies adopted by the individuals to cope and manage the medication costs is absolutely insufficient.

Findings of our study resonate with the results from studies done in countries other than Canada, both having different and similar health care system arrangements for prescription medications. A national study from Israel reported that around 10% of chronically ill patients faced CRNA that was strongly associated with their lower income, unemployment, lack of physician explanation about the prescribed medication, and age (Simon-Tuval, Triki, Chodick, & Greenberg, 2014). A study from US showed strong correlation between copayments paid by the patients and medication underuse (Wagner et
al., 2008). Another study reported that age less than 65, lack of drug coverage, increased number of over-night hospitalizations, and greater functional limitations were associated with greater likelihood of CRNA among diabetic patients, while nursing home residence decreased risk (Zhang et al., 2014). CRNA was least common in countries with lowest out-of-pocket costs, and reduced copayments or cost ceilings for low-income patients (Kemp et al., 2010; Kennedy & Morgan, 2006, 2009). Overall, across all the countries, people without prescription-drug coverage were significantly more likely than those with insurance to report cost-associated non-adherence.

Although studies examining the consequences of CRNA on individual health outcomes are limited in the Canadian context, a number of studies from other countries report that CRNA to prescription drugs has a negative impact on the health outcomes of people who face these barriers (Piette, Wagner, Potter, & Schillinger, 2004; Rahimi, Spertus, Reid, Bernheim, & Krumholz, 2007; Sokol, McGuigan, Verbrugge, & Epstein, 2005). Internationally, evidence on social outcomes and/or strategies adopted by the individuals to cope and manage the medication costs is also emerging. A study exploring strategies patients use to reduce the cost burden of prescriptions across UK and Italy reported that commonly used strategies were not to get prescribed drugs at all, prioritizing by not getting all prescribed medicines or delay purchasing medicines until they got paid, or cost-consciously self-medicating with over-the-counter products for minor conditions (Atella, Schafheutle, Noyce, & Hassell, 2005). Another study from the US reported that patients coped with medication costs by obtaining free samples from physicians, splitting doses so medications last longer, buying drugs from other countries
and/or over the internet, or buying medications through the Veterans Administration (Musich et al., 2015).

**Methodological issues in the included studies.** It must be noted that available literature on CRNA within the context of Canada adopts no national or international standards to define, conceptualize and measure CRNA, which leads to lack of uniformity across the studies and hence the results drawn from these studies. Additionally, a majority of the studies are either survey based that used data from large population based national and international surveys, or cohort studies that used administrative claims databases. Both of these methods have some limitations that should be taken into consideration. First, the studies using claims data to assess adherence may not necessarily represent the actual consumption of prescribed medications or cannot account for medications that were not purchased due to cost (Després et al., 2016; McLeod et al., 2011). Second, most of the national surveys collected data over the phone (Campbell et al., 2014; Kemp et al., 2010; Kennedy & Morgan, 2006, 2009; Lee & Morgan, 2017). Though in national or international surveys, the sample was intended to represent the general population, telephone-based surveys may under-represent the most socially disadvantaged, individuals in remote areas, and individuals who do not own landline phones. It is also possible that some participants might not feel comfortable reporting under-using medicines because of cost, in which case the occurrence of cost-related underuse would be underestimated by these studies (Kennedy, 2018). Third, as most of the survey data was self-reported, it may have a recall bias or social desirability bias (Hennessy et al., 2016). Fourth, studies that analysed various provincial drug benefit programs or utilized claims data from provincial programs possibly included only those
drugs that were on the public formularies and hence could not have accounted for those drugs that did not fall onto the list or for which claims were rejected.

**Gaps in the current literature.** Within the Canadian context, there is a lack of literature that examines the effect of the propensity of prescribing healthcare professionals to discuss economic issues with their patients on determining cost-adherence relationships for them. It is important to explore and find how patients’ experiences can be improved through the support from clinicians. For example, prescribing less costly alternative or generic medication or having conversations about the medication costs may have a positive effect on decisions that patients make to manage their prescription cost burden (Law et al., 2018). Studies on strategies adopted by patients to cope with prescription cost burden, such as reducing the frequency, dose, or duration of medications; obtaining samples or generic substitutes; or substituting prescribed drugs with over the counter or herbal medications are also limited.

Furthermore, most of the available evidence in the Canadian context is drawn from the general population, senior population, and patients with chronic diseases, and nothing has been specifically studied for people with disabilities or people belonging to aboriginal communities. Evidence from other countries shows that people with disabilities and those belonging to aboriginal communities are at increased odds of facing CRNA. For example, indigenous patients belonging to aboriginal communities residing in Australia, Canada, and New Zealand were two to three times more likely to report CRNA compared with non-indigenous patients (Kemp et al., 2010). Similarly, studies show that severe disability, poor health status, low income, lack of insurance, and a high
use of prescriptions increase the likelihood of people with disabilities of engaging in CRNA (Kennedy & Erb, 2002; Naci et al., 2014).

**Recommendations for future research.** Future research should explore factors that influence patients’ decisions to alternative medicine regimens due to cost. Also, there is a need to explore the experiences of people while managing prescription drugs costs. Only two qualitative studies involved people facing these barriers directly and explored their experiences. These experiences are important to know, because access to necessary prescription medications might have implications that are beyond just health and healthcare, especially for socially vulnerable and disadvantaged (Kennedy, 2018). The role of health system governance in ensuring burden free access to prescription medications for all also needs to be investigated. Research is required that can analyse the structures (i.e. supports, institutions, resources), processes (i.e. access, roles, and functions) and outcomes (i.e. justice, equality and service) of provincial health and social policies, and their effect on cost-related access to medicines for people in Canada (Jongbloed, 2003).

**Study limitations.** Our study has certain limitations. One limitation is related to the generalizability of its findings. Given that the study specifically aimed to map the literature from Canada, this study might be helpful for Canadian policymakers to inform future policy directions and alternatives. Additionally, as scoping studies are intended to provide a wide spectrum i.e. quantity and breadth of literature, we did not assess the quality of included studies. Also, we only included articles published in English. Conducting the literature search in French would permit more confident claims regarding the comprehensiveness of the search strategy in this scoping review.
2.7 Conclusion

Due to the emerging attention on increasing costs of prescription medications in Canada and incidence of CRNA among Canadians over the past decade, many healthcare groups, advocacy associations, and health policy researchers have proposed different Pharmacare models for Canada. Findings of this scoping review suggest that what we know about the phenomenon of CRNA might be insufficient. Inquiry on CRNA is insufficient especially among the socially disadvantaged groups such as the indigenous population and people with disabilities; its impact on health outcomes, and access and utilization of other healthcare services. Future research should look at the effects of health-system factors and support from prescribing healthcare professionals on modifying the cost-adherence relationships for individuals. In summary, more evidence is required to inform whether national Pharmacare can ensure universal, timely, and burden free access to prescription medications for all or targeted policy efforts are required balancing the competing influences and demands.

2.7 References


national survey data. *Open Medicine, 5*(1), 1–9. Retrieved from http://www.scopus.com/inward/record.url?eid=2-s2.0-79251560043&partnerID=40&md5=972115c54d69d38b80e932e906b5e6e1


Chapter 3

An adapted model of cost-related non-adherence to medications among people with disabilities

3.1 The Fit of Manuscript 2 within the Dissertation

Manuscript 2 builds upon the findings of the first manuscript by examining the concept of CRNA within the context of people with disabilities. Since the inception of the idea to conduct this research, I was intrigued and bothered by the question: “Why do we need to study CRNA within people with disabilities, despite abundant studies already been done with the general population and people with chronic conditions?” I wanted to find if there are differences in the barriers faced by people with disabilities as compared to the general population to access their necessary medications; and what perpetuates the risk of facing such barriers between people with and without disabilities. This study began to answer those questions for me. In this study, I identified and studied the models available to understand medication adherence to find if they are representative of the issues faced by people with disabilities to obtain their prescription medications.
3.2 Abstract

Despite emerging evidence on cost-related non-adherence (CRNA) to prescription medications, there is little conceptualization and exploration of this phenomenon with respect to disability. Specifically, there is a gap in the literature that explores factors influencing medication cost-adherence relationship among individuals living with a disability. To advance research on and policy for CRNA to medications among people with disabilities, we need a framework that can contribute towards guiding solutions to this problem. We examined the applicability of Piette and colleagues’ existing model for CRNA to the context of people with disabilities and suggested an adapted model (CRNA-d) that can provide a more specific conceptualization of CRNA with respect to disability. The adapted CRNA-d model depicts that CRNA to prescription medications with respect to disability is a dynamic and multifaceted phenomenon, determined by various socio-economic, disability-related, medication-related, prescriber-related and system-related factors. We discuss how higher susceptibility to health complications, barriers to income and employment, additional healthcare costs, complexity of medical regimens, limited access to physician services and other policy-related factors increase the risk of persons with disabilities to face cost-related barriers to access their necessary medications.
3.3 Introduction

Prescription medications play an important role in the treatment and prevention of disease and disability, and promotion of health and well-being (Bigdeli et al., 2013; World Health Organization-WHO, 2003). However, with the worldwide increase in the cost of medication, cost-related medication underuse has now become a public health concern (Cutler, Fernandez-Llimos, Frommer, Benrimoj, & Garcia-Cardenas, 2018). Evidence is emerging that many low-income persons and those who have chronic health conditions forgo their medications due to cost, which is commonly referred to as cost-related non-adherence to medications (CRNA) (Soumerai et al., 2006). Despite emerging evidence on cost-related underuse of prescription medications, there is little conceptualization or exploration of this phenomenon with respect to disability (Gupta, McColl, Guilcher, & Smith, 2018).

People with disabilities, that constitute around 15% of the worlds’ population, are often economically disadvantaged and are much more likely to live below the poverty line as compared to their non-disabled counterparts (World Health Organization, 2011). Globally, people with disabilities often remain unemployed or underemployed and experience greater frequency of financial stress for basic necessities such as housing, healthy food, or essential drugs (Smith, 2013). Knowing that people with disabilities incur additional cost of living (Mitra, Palmer, Kim, Mont, & Groce, 2017), costs associated with long-term use of medications further pose a lifetime economic burden (Jensen & Biering-Sørensen, 2014). However, the majority of the global literature on cost of medications and its impact on medication use or adherence does not well represent people with disabilities. Studies that do exist measure the prevalence of cost-related
underuse or non-adherence to medications among people on social welfare or social assistance recipients at most (Kennedy & Erb, 2002; McLeod, Bereza, Shim, & Grootendorst, 2011; Naci et al., 2014). Thus, there is a gap in the literature that explores factors influencing medication cost-adherence relationship among individuals living with a disability.

To advance research and understand the phenomenon of CRNA to medications among people with disabilities, we need to have a framework, a system of concepts, assumptions, beliefs, or theories (Maxwell, 2011), that can contribute towards guiding solutions to this problem. An improved understanding of the cost-related barriers faced by those with disabilities will inform decision makers across the world while developing drug coverage policies that consider the needs of people with disabilities. Therefore, in this paper, our purpose is to examine how the existing model for CRNA apply or do not apply to the context of people with disabilities and suggest an adapted model that can depict more specific conceptualization of CRNA with respect to disability.

For the purpose of our study, we adopted the definition of disability provided by the International Classification of Functioning, Disability and Health (ICF) that defines disability as an umbrella term for impairments, activity limitations and participation restrictions and considers it as a complex phenomenon due to the interplay between persons’ nature of impairment and their socio-economic environment (WHO, 2002). Our intent behind using the general definition was to suggest a conceptual model that broadly addresses persons with disabilities. Overall, our discussions are supported by evidence from the global literature, and our previous and current experiences of working with people with disabilities.
3.4 Existing frameworks on cost-related non-adherence to medications

The most common frameworks that have been used in the literature to define, understand, or identify factors associated with CRNA to medications include the WHO’s framework of adherence to long-term therapies (2003) (De Vera, Mailman, & Galo, 2014; Després, Forget, Kettani, Blais, 2016); Andersen and Newman’s healthcare utilization model (2005) (Kennedy & Morgan, 2006, 2009); and a model on medication cost pressures and non-adherence developed by Piette and associates (2006) (Kemp, Roughead, Preen, Glover, & Semmens, 2010). Another recent notable work is done by Goldsmith and colleagues wherein authors have developed a typology of CRNA (Goldsmith et al., 2017). These models are discussed here briefly:

(a) WHO’s framework of adherence to long-term therapies: The WHO framework of adherence to long-term therapies was developed in 2003 that defines adherence as “the extent to which a person’s behaviour – taking medication, following a diet, and/or executing lifestyle changes, corresponds with agreed recommendations from a health care provider” (p. 3). The WHO framework suggests that adherence is a multidimensional phenomenon determined by the interplay of five sets of dimensions which include:

- patient-related dimensions (i.e. patients’ knowledge and beliefs about their illness, motivation to manage it, self-efficacy in their ability to engage in illness-management behaviours, and expectations regarding the outcome of treatment and the consequences of poor adherence.
• therapy-related dimensions (i.e. complexity of medical regimen, duration of treatment, previous treatment failures, frequent changes in treatment, immediacy of beneficial effects, side-effects, availability of support to deal with them)

• condition-related dimensions (i.e. severity of symptoms, level of disability (physical, psychological, social and vocational, rate of progression, severity of the disease, availability of effective treatments)

• healthcare team or system related dimensions (i.e. reimbursement by health insurance plans, medication distribution systems, knowledge and training for health care providers on managing chronic diseases, knowledge on adherence and of effective interventions for improving it, workload on health care providers, continuity of care by the doctor, communication style of the doctor)

• socio-economic dimensions (i.e. gender, marital status, age, educational level, health status, socioeconomic status, social support networks, living conditions, culture and lay beliefs about illness and treatment).

WHO framework serves as a broad framework that applies to several patient behaviours such as seeking medical attention, filling prescriptions, taking medication appropriately, obtaining immunizations, attending follow-up appointments, and executing behavioural modifications that address personal hygiene, self-management of asthma or diabetes, smoking, contraception, risky sexual behaviours, unhealthy diet and insufficient levels of physical activity, all of which are examples of therapeutic behaviours. While the WHO framework is very useful in understanding patient health seeking and adherence behaviours in general, it does not specifically focus on patient adherence behaviours under cost constraints.
(b) **Anderson and Newman’s healthcare utilization model:** Anderson and Newman’s model first developed in 1973, is a framework of societal determinants (technology and norms), healthcare system-level determinants (resources and organization), and individual determinants (predisposing, illness-level, and enabling factors) that explain the utilization and health seeking behaviour of individual patients for various health services such as physician, hospital, dental, medication, and nursing home. The authors of the model grouped healthcare utilization at four levels of primary, secondary, tertiary, and custodial healthcare services.

A major highlight of this model is its elaboration on individual characteristics: *predisposing factors* including demographic, social structural, and attitudinal-beliefs which exist prior to the onset of specific episodes of illness and suggest why certain individuals have higher or lower propensity to seek healthcare; *enabling components* which make health service resources available to the individual such as such as income, level of health insurance coverage, or other source of third-party payment, whether or not the individual has a regular source of care, the nature of that regular source of care, and the accessibility of the source; and *illness level components* that represent the most immediate cause of health service use such as symptoms, and clinical diagnosis. While this model serves as a useful framework to analyse factors affecting health seeking and utilization behaviours, it does not focus on patients’ adherence behaviours to therapies after seeking preliminary health service.

(c) **Piette model on medication cost pressures and non-adherence:** Piette and associates proposed the first conceptual model of CRNA to medications in patients who have chronic conditions to set the stage for research, policy, and practice considerations
to address the issue of CRNA among patients in 2006 (Piette, Heisler, Horne, & Caleb Alexander, 2006). According to this model, the cost-adherence relationship is determined by the interaction of a set of four factors: (a) patient characteristics (e.g., age, attitudes and beliefs toward medications), (b) drug or treatment characteristics (e.g., reason for taking medications and complexity of dosing), (c) clinician factors (e.g., medication choice, support provided by the doctors, their attitudes and communication about medication costs) and (d) health system factors (e.g., auditing clinicians' prescribing and mechanisms to help low-income patients to get the financial assistance for filling necessary prescriptions) (Piette et al., 2006) (Figure 1). Since the development on 2006, the model is used widely to understand CRNA in various populations such as older adults and patients with chronic illnesses (Piette, 2009; Piette, Beard, Rosland, & McHorney, 2011; Wagner, Heisler, & Piette, 2008; Zhang et al., 2014).

(d) Goldsmith model on CRNA typology: The typology of CRNA model is one of the most recent models developed by Goldsmith and colleagues based on a study conducted with adults who reported engaging in CRNA. The authors found that an array of factors such as individuals’ financial flexibility, the importance of the drug, burden of the drug costs, and having an insurance interact with each other and influence CRNA in individuals. Their model depicted the patterns of CRNA experience within individuals with varying financial and health situations: CRNA in persons with low financial flexibility, CRNA in persons with more financial flexibility, CRNA for low importance drugs and CRNA for high importance drugs. While this model focussed on CRNA specifically, it considered the interplay between four factors in determining CRNA experiences and did not focus on provider-related or system-related factors on CRNA.
3.5 Comparing existing models for their applicability to the context of people with disabilities

Out of the four models discussed above, the WHO model serves as the most comprehensive framework that provides a detailed and separate list of factors under following headings i.e. patient/demographic factors; patient socio-economic factors; therapy / medication related factors, condition / illness / disability related factors; clinician / prescriber related factors; and system / policy related factors to understand and determine patients’ adherence behaviours. The Anderson’s model focusses largely on patient-related factors in the form of predisposing, enabling and illness level factors and describe health service factors as different types and levels. The Piette Model provides a list of four group of factors affecting CRNA specifically but does not list the illness/disability-related factors. The Goldsmith model delves deeply but considers only a few factors within the patient socio-economic and medication related factors and explains the interplay among those.

Overall, among the four models discussed above, Piette model is the only model that focusses on CRNA to prescription medications specifically and comprehensively and aligns closely with the purpose of the present study which is to conceptualize CRNA with respect to people with disabilities. The Piette model will be therefore used, discussed and adapted further in this study. Please see table 8 that summarizes the advantages and disadvantages of the various models discussed above for their suitability to study CRNA within the context of present study.
Table 8: Advantages and disadvantages of various models

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<th>S.No</th>
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<th>Advantages</th>
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<td>3.</td>
<td>Piette model on medication cost pressures and non-adherence, 2006</td>
<td>The elements of the model provide a comprehensive list of factors associated specifically with CRNA behaviours in patients. It was the first conceptual model to study CRNA and set the stage for research, policy, and practice considerations to address the issue.</td>
<td>Since the development, the model is used widely to understand CRNA in various populations such as older adults and patients with chronic illnesses, however it has not been applied to the context of people with disabilities.</td>
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<td>4.</td>
<td>Goldsmith model on CRNA typology, 2018</td>
<td>This model focusses on CRNA specifically and depicts the patterns of CRNA experience within individuals with varying financial and health situations.</td>
<td>The model considers the interplay between four particular factors in determining CRNA and does not focusses on provider-related or system-related factors on CRNA.</td>
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**Piette model and its applicability to the context of people with disabilities:**

Although widely used in the United States, the Piette model has not been used to date with respect to people with disabilities, particularly in the developing countries. This is due to the fact that the model is based on American studies (Piette & Heisler, 2004), which do not necessarily represent people with disabilities living worldwide, especially in the countries where healthcare is not covered and paid out-of-pocket for people with disabilities. In order to examine this in depth, we will discuss the factors affecting CRNA...
as described by Piette at al. (2006) model one by one, to assess its importance or applicability to people with disabilities.

**Patient characteristics.** The first factor in Piette’s model that affects CRNA to medication is patient’s demographic and socio-economic characteristics. Piette and colleagues (2006) found that a person’s demographics, such as race, ethnicity, income, and age, have significant impact on CRNA. The authors also highlighted that individual’s medication related beliefs and perceptions lead to general medication non-adherence. While these factors may apply for people with disabilities, there are many more dimensions that may have a substantial effect on medication cost pressures and adherence for them. Some of these factors include- multi-dimensional nature of poverty resulting from disability; type, nature and severity of disability; health complications or co-morbidities leading to additional healthcare needs and costs; limited social support; and impact of age on disability.

**Poverty due to disability.** The World Report on Disability (2012) highlights that globally, people with disabilities are much more likely to be less educated, unemployed or underemployed and earn less than their non-disabled counterparts (WHO & The World Bank, 2011). In addition, they often face discrimination in employment, have limited access to transportation and to resources to promote self-employment and livelihood activities, which in turn put them at risk of poverty (Banks, Kuper, & Polack, 2017; Maart & Jelsma, 2014; Mitra, Posarac, & Vick, 2013; Sommers, 2006). A study by She & Livermore (2007) using a national survey data showed that at the similar income level, individuals with disabilities are much more likely to experience material hardship than their peers without disabilities. This means that incomes of those with disabilities would
need to be at least 3 times to reduce the prevalence of hardship to the level of others with incomes at the poverty level.

Evidence is robust that poverty and/or lack of regular employment is the primary predictor leading to CRNA to medications (Kapur & Basu, 2005; Kennedy & Morgan, 2006; Sanmartin, Hennessy, Lu, & Law, 2014), which is especially true for people with disabilities living in low and middle-income countries (WHO, 2004). A study looking at the relationship between disability and five dimensions of economic well-being (education, employment, medical expenditures, assets, and expenditures) across 15 developing countries found that persons with disabilities experience higher rates and severity of multiple deprivations than persons without disabilities (Mitra et al., 2013). Overall, global evidence suggests that persons with disabilities are significantly more likely to be multi-dimensionally poor (Agyemang & van den Born, 2018; Mitra et al., 2013, 2017; Pinilla-Roncancio, 2018), which in turn affects their affordability and access to healthcare including prescription medications.

**Type, nature, and severity of disability.** The nature of disability (i.e., physical, mental, sensory, neurological or intellectual) has an effect on the medication use, costs, and adherence. People with disabilities may need medicines during an acute stage, a relapse or at a chronic stage to treat various co-morbidities or health conditions that may or may not be directly related to their disability (Brichetto, Uccelli, Mancardi, Solaro, & Brichetto, 2003; Jensen & Biering-Sørensen, 2014). Furthermore, a person’s disability itself can be static, episodic, or progressive; short-term or long term; or painful or trivial (WHO & World Bank, 2011). Due to the heterogeneous nature of disability, CRNA issues can be very different for people with a minor physical disability, major cognitive and,
importantly, psychiatric or behavioral disabilities. For example, available research suggests that within people with disabilities, people with cognitive and developmental disabilities face more disparities in overall access to health care. Similarly, people with visual impairment as well as those with multiple limitations have been found at higher odds of forgone essential care (Horner-Johnson et al., 2014).

Additionally, the type of disability changes the relationship between financial resources and important quality of life outcomes. For example, in Canada, poverty and unemployment rates vary by type of disability. People who have a combination of physical-sensory, cognitive and mental disability are more commonly poor and jobless followed by those with a mental-cognitive disability and a physical-sensory disability (Wall, 2017). Similarly, another study found that even with insurance, people with multiple disabilities had more unmet healthcare needs as compared to people with a single disability (Reichard, Stransky, Phillips, McClain, & Drum, 2017). Therefore, the magnitude of the potential effects of disability on the relationship between medication cost and non-adherence will be greatly influenced by the disability status.

Susceptibility for health complications leading to additional healthcare costs. Susceptibility of persons with disabilities to develop additional complications, co-morbidities or chronic conditions is significantly higher as compared to the general population (Gudlavalleti et al., 2014; WHO & World Bank, 2011). Furthermore, disability can exacerbate comorbidity, and lead to development of frailty, all of which lead to additional need for medications (Fried, Ferrucci, Darer, Williamson, & Anderson, 2004). Due to unmet healthcare needs, individuals with disabilities use more health professionals’ and other health services than non-disabled individuals (McColl, 2005;
Apart from healthcare services, people with disabilities need extra services such as attendant care, assistive equipment, modifications to their home, special transportation, technological aids or rehabilitation services, all of which lead to added health care costs and burden (Krueger et al., 2013; Mitra, Findley, & Sambamoorthi, 2009; Maart & Jelsma, 2014). However, most of these costs are either not covered through the social welfare schemes or disability benefit programs, or are being capped for the cost, volume, or amount, leading to additional out-of-pocket cost for those living with a disability (Mitra et al., 2017).

**Lack of or inadequate family or social support.** With respect to social support, evidence suggests that being married or living with a partner and having family support significantly increase adherence, whereas homelessness or loneliness decreases adherence to necessary medications (DiMatteo, 2004; Kardas, Lewek, & Matyjaszczyk, 2013). However, a substantial proportion of people having disabilities live alone, are homeless, have small social networks or limited to no support from the family (Guilcher et al., 2012; Morris, 2001; Nishio et al., 2017). Their social support diminishes over time which often leads to depression, isolation, and poor health and well-being and makes them further vulnerable to CRNA (Beer et al., 2011; Mitra et al., 2017; Samuel, Alkire, Zavaleta, Mills, & Hammock, 2018).

**Impact of age on disability.** Several studies suggest that there are interrelationships between disability and frailty which creates vulnerability towards adverse health outcomes, dependency, falls, need for complex care, and mortality (Boeckxstaens et al., 2015; Curcio, Henao, & Gomez, 2014; Fried et al., 2004; Kojima, 2017; Wong et al., 2010). The process of ageing for people with disabilities begins earlier
than usual, with the effects of ageing getting superimposed on the effects of a disability (WHO, 2018). In addition, people with disabilities are often being prematurely removed or retired from the workforce while older persons in general population remain employed (Turcotte, 2014). A recent study reports that elderly people (>65 years of age) with disability experience higher out-of-pocket expenditures compared to other age groups (Mitra et al., 2017). Therefore, as the severity of disability or age of the person increases, the health, income and employment outcomes deteriorate (Hitzig, Campbell, McGillivray, Boschen, & Craven, 2010). This implies that while the need for prescription drugs may increase with time, the financial capacity of people with disabilities declines, thereby posing them at a greater risk for CRNA.

**Medication-related characteristics.** Another important determinant of CRNA highlighted in the Piette model is the type of medications and diseases for which medications are being taken. Authors indicated that patients value those medications that treat life threatening conditions or prevent future adverse events (essential medications). People are more likely to forgo symptomatic medications given for temporary relief or for less serious health problems (non-essential medications). Complexity of treatment regimen and propensity of side effects or adverse effects also affect people’s responses towards medication cost and adherence. While all these factors will apply to the context of people with disabilities, there are additional medication-related factors that may have a varied influence on CRNA and therefore, need to be considered differently.

**Complex and long-term medical regimens.** People with disabilities are often heavy users of prescription medications (Rouleau & Guertin, 2011). Specific to the nature
or type of disability, many people with disabilities have long-term medication needs (Patel, Milligan, & Lee, 2015). In addition, people with disabilities are often at a higher risk of being prescribed complex regimens of medications (e.g., analgesics, narcotics, anticonvulsant, antidepressant etc.), as well as multiple medications within each class e.g., multiple analgesic-narcotics (Kitzman, Cecil, & Kolpek, 2016). The long-term use of complex medical regimens can either directly affect cost and adherence or indirectly lead to side-effects which can limit one’s ability to return to work and gain financial flexibility to afford medications (Mohammed, Moles, & Chen, 2016; Sav et al., 2013; Wilson, Kataria, & McNeil, 2013).

**Need for over-the-counter substitutes and other health supplements.** People with disabilities may need to use a wide range of health products apart from the prescription medications such as alternative therapies (e.g., naturopathy, Ayurveda, homeopathy etc.), or other over-the-counter medications such as minerals, vitamins, amino acids, fish oil, or herbal products (Brichetto et al., 2003; Doan et al., 2013). These may have additional costs and therefore, affect their decisions or behaviors related to the use of prescription medication. Therefore, there exists an intricate relationship between various health and medication related characteristics, beyond being essential or non-essential, that may either decrease or increase CRNA in people with disabilities.

**Clinician-related factors.** Piette and colleagues highlighted various clinician-related factors that may have an influence on patients’ cost-related barriers to medications and hence their adherence. For example, very often physicians have limited time to discuss or have little knowledge about the cost of the medications that can lead to non-adherence. On the other hand, patients might themselves be hesitant to discuss their
economic insufficiency to access their prescriptions or are not able to build trust to have a
clear communication with their physicians. The authors suggested that if clinicians are
made aware, solutions could be developed to relieve patient’s cost pressures to
medications (Piette et al., 2006). While all these elements are significant to the context of
people with disabilities, there exists an additional barrier which people with disability
face, that is having access to a primary care provider. One of the common factors
highlighted by various studies associated with CRNA is having an access to a primary
care physician or a regular healthcare provider and relationship between doctor and
patients (Allin, Stabile, & Tuohy, 2010; Hunter et al., 2015; Kennedy & Morgan, 2006;
Tamblyn, Eguale, Huang, Winslade, & Doran, 2014; Wang, Lia, Sweetmanb, & Hurleyb,
2015).

Access to a health care provider. Literature suggests that people with disabilities
face various physical, attitudinal, and policy-related barriers to access services of their
primary healthcare providers (Donnelly et al., 2007; Manns & May, 2007; McColl, 2006;
Stillman, Frost, Smalley, & Williams, 2014). Doctors may not have accessible clinics due
to which people with disabilities are likely to miss their regular visits for health care or
may require assistance to attend their appointments (Popplewell, Rechel, & Abel, 2014).
Doctors often refuse to enroll patients with disability on their caseloads or roster as they
do not have any financial incentives for extra efforts in terms of time or service demands
(McColl et al., 2008). For example, with people having cognitive or auditory disabilities,
history-taking and other procedures might take longer. This may not leave any time to
discuss about prescription costs (McColl, 2006). Doctors may not be aware of the
financial situation of their patients and the disability itself may limit the discussion of
complex nuances. For a person having a disability, family physician acts as a channel to many disability-related benefits including that for prescription drug coverage. However, none of these administrative visits are covered by the government and must be paid for out-of-pocket (McColl, 2006). Therefore, all these barriers can influence whether persons with disabilities visit a doctor which in turn may affect their decisions regarding medication related cost-pressure and adherence.

Patient-provider relationship and communication. Multiple studies suggest that healthcare provider-patient relationship, concordance, and communication, such as doctor’s ability to provide appropriate information on drug administration, offering enough time to the patient, and doctor’s ability to demonstrate empathy and elicit or respect patient’s concerns have a great impact on patient’s decision to take or stop taking their medications (Kardas et al., 2013; Stavropoulou, 2011). However, evidence is emerging that these information and communication needs of people with disabilities remain particularly unmet (McColl et al., 2008; McColl, Aiken, McColl, Sakakibara, & Smith, 2012). People with disabilities often need to consult multiple healthcare providers and medication prescribers, and face poor coordination among providers, limited duration of visits with physicians and specialists which further limits the discussion on difficulties with insurance or finances; and results in inadequate information provision (De Vries McClintock et al., 2016). Primary care providers themselves acknowledge that they lack disability-specific knowledge which limit their ability to explain things to their clients with disabilities and propensity of developing therapeutic relationship with them (Kroll, Beatty, & Bingham, 2003; McColl et al., 2008). These elements leading to poor patient-
provider relationship or communication may put people with disabilities at an increased risk of engaging in medication non-adherence.

**Health system-related factors.** In the Piette model, health system characteristics have been identified as having a significant influence on patient’s extent of burden and responses to medications-related costs. The authors mentioned that determinants such as being insured or uninsured for prescription drug benefits or being treated in public or private systems could influence patient’s responses to medication cost burden. They also highlighted the long wait times or cumbersome application process to access assistance programs affecting medication adherence negatively. While all this is true for people with disabilities, the barriers related to healthcare programs or policies faced by people with disabilities are much more complex, dynamic and heterogeneous (Guiilcher et al., 2017).

**Complexity of health and social care.** In addition to policies related to healthcare, other policies or programs that are related to income security or housing also influence people with disabilities in accessing resources to afford therapies to maintain their health (Williamson et al., 2006). Moreover, these social assistance policies vary across countries or even in jurisdictions within countries or ministries (McColl, Jaiswal, & Roberts, 2017). For example, in Canada, depending on the eligibility criteria, the extent of prescription drug coverage varies extensively across provinces. Consequently, prescription drug costs vary for individuals having the same prescription needs (Demers et al., 2008; Kratzer et al., 2015). Even when people with disabilities are eligible to be covered by public insurance, they have to share the costs of medications in the form of copayments and deductibles that may lead to negative consequences on medication use
(Alan, Crossley, Grootendorst, & Veall, 2002). Also, sometimes the more expensive or new drugs or drugs required to treat uncommon conditions are not available on the public drug formulary despite the potential clinical benefits for people with disabilities (Guilcher et al., 2017). Consequently, the complexity and variability in policies need to be analyzed for their impact on medication related financial pressures for people with disabilities.

In summary, the model developed by Piette and colleagues (2006) has done the important groundwork by laying the foundation of the most common factors that could increase the risk of cost-related medication non-adherence in chronically ill patients. However, we would need to advance this knowledge and adapt the model with respect to the complex and heterogeneous barriers faced by people with disabilities while accessing their medications.

3.6 An adapted framework: CRNA-d

In order to present specific conceptualization of CRNA with respect to people with disabilities, we propose an adapted model and name it as CRNA-d (Cost-related non-adherence to medications for person with disability). This adapted model depicts that CRNA to prescription medications with respect to disability is a dynamic and multifaceted phenomenon, determined by various socio-economic (including demographic), disability-related, medication-related, prescriber-related and system-related factors (Figure 3). This proposed model is framed keeping the additional barriers faced by people with disabilities into consideration that pose them at greater risk of experiencing financial barriers to access their prescription medication needs. These factors are listed here:
(i) **Demographic and socio-economic factors:** Age; marital status or availability of social support; employment status (unemployed, underemployed, employed); income; access to non-employment related resources (pensions, loans, or disability benefits). All these factors can compound the impact of disability on cost-related medication underuse.

(ii) **Disability-related factors:** Nature or type of disability (temporary or long term; static or progressive; physical, mental, intellectual or sensory); severity of disability; presence of any co-morbidities (psychological issues or cognitive, physical impairment); additional healthcare costs (attendant care, home modifications, adaptive equipment etc.).

(iii) **Medication-related factors:** Type of medications (prescriptions, over the counter, natural health products, alternative therapies); conditions for which medications are taken; frequency of taking medications; out-of-pocket costs on medications; availability and type of drug insurance; cost-sharing mechanisms (premiums, copayments, deductibles).

(iv) **Prescriber-related factors:** Access to physician services; relationship with doctor and communication regarding medication cost; coordination of care; accessibility to clinics (accessible parking or transportation); prescriber knowledge of disability and disability-specific services.

(v) **System-related factors:** Availability and access to drug benefit programs; public drug formulary; availability and access to other social or healthcare services; and funding and resources for alternative therapies or equipment.
Figure 3. An adapted model: CRNA with respect to disability (CRNA-d)

In the pictorial representation of the adapted model, solid lines highlight predominant factors (socio-economic and medication-related factors) that affect CRNA among people with disabilities directly; dotted lines with arrows indicate the factors that may moderate the effect of predominant factors (demographic and disability-related factors) or indirectly affect CRNA among people with disabilities (prescriber-related and system-related factors); and dotted lines without arrows indicate the potential interaction among these factors. Also note that while some of the factors are concurrent with the previous model developed by Piette and colleagues, there are many other factors that are added to the model to depict more specific conceptualization of CRNA with respect to disability. We believe that a systematic investigation with further research would be required to find and quantify the potential impact of most relevant factors (especially disability-related factors) that determine cost-adherence relationship for people with disabilities.
3.7 Implications for policy

The growing evidence on the barriers faced by many people living in both developed and developing countries to access and afford necessary medications has given a strong push to the international debate (Agyemang & van den Born, 2018; Ahmadiani & Nikfar, 2016). Over the last few decades, many health care groups, advocacy associations and health policy researchers have called for national policies that can provide improved access to medicines for all (Gupta, 2016; Persaud & Ahmad, 2017). There have been efforts by the international organizations such as World Trade Organization as well as governments across the world to manage the issues related to drug shortages, manufacturing quality, marketing and price regulation, supply chain management, procurement processes, patenting, generic drug availability, and essential list of medications (Bigdeli, Peters, & Wagner, 2014; WHO and United Nations Development Programme, 2003). However, inquiry on extent, determinants, and consequences of cost-related barriers to medications especially among socially disadvantaged groups such as people with disabilities has been inadequate (Gupta et al., 2018).

Differences between the barriers faced by those with and without disabilities perpetuate the inequities in the availability of and access to prescription drugs (Guilcher et al., 2017). The proposed model (CRNA-d) is framed keeping additional barriers faced by people with disabilities into consideration that pose them at a greater risk of experiencing financial barriers to fulfill their prescription medication needs. Therefore, the present study will be crucial and timely to this context and contribute to the limited understanding on the medication cost pressures among people with disabilities. Further, it
may help to inform policy makers whether national drug coverage or targeted policy efforts are required to ensure burden-free access to medications for all. Though we believe that a systematic investigation with further research is warranted to find the most relevant variables that predict CRNA in people with disabilities and its relationship with specific types of disabilities.

3.8 Conclusion

Despite emerging evidence on CRNA to prescription medications among people with chronic conditions, there is little conceptualization or exploration of the phenomenon with respect to disability. In this paper, we tried to adapt the already existing framework developed by Piette and colleagues in 2006 that aimed to understand the factors associated with CRNA among chronically ill patients. While most of the factors highlighted in their model may apply to people with disabilities, there exists an array of additional barriers that people with disabilities face while accessing their necessary medications. Several of these include higher susceptibility to health complications or co-morbidities, barriers to income and employment, additional healthcare needs, other health care costs, barriers to access physician services and other environment or policy-related barriers. Therefore, the adapted CRNA-d model highlights the core elements and different dimensions that should be considered while examining the phenomenon of CRNA among people with disabilities. However, this model will need to be tested through research for the development of more specific and nuanced conceptualization of CRNA with respect to disability, and therefore, formulating strategies to reduce the extent and impact of CRNA among people with disabilities.
3.9 References


doi:10.1038/sj.sc.3101973


105
Statistics Canada, (75).


Chapter 4

Prescription medication cost, insurance coverage, and cost-related non-adherence among people with spinal cord injury in Canada

4.1 The Fit of Manuscript 3 within the Dissertation

Manuscript 3 builds upon the findings of the first two manuscripts by examining the magnitude and risk factors of CRNA among people with spinal cord injuries. The study is among the initial studies in Canada that quantified the phenomenon of CRNA within the context of people with disabilities. Thus, the manuscript complements the overall theme of the thesis by suggesting evidence on the extent and determinants of CRNA among people with SCIs.
4.2 Abstract

The purpose of this study was to describe the most common prescription medications used by people with spinal cord injury (SCI) and the extent of out-of-pocket cost, insurance coverage, and cost-related non-adherence (CRNA) for those medications. It was an observational study wherein data were collected through a cross-sectional online survey from individuals living with a SCI in Canada. We used descriptive statistics to describe the extent of drug cost, insurance coverage and CRNA among study sample, and analytical statistics to find association of CRNA with socio-demographic, injury-related and medication-related characteristics of the sample. Results suggested that individuals with SCI (n=160) used an average of 5 medications and spent a median of $49 (interquartile range: $234.75) per month on their medications. More than 90% of participants had some form of drug insurance, though 37% reported CRNA. The most common medications that were forgone due to cost included opioids, antidepressants, and drugs for genitourinary and muscular spasms. Individuals with paraplegia and non-traumatic SCI had higher drug costs, though injury-related characteristics did not influence CRNA. Female sex, higher drug expenditure and additional health care costs were significantly associated with CRNA. In conclusion, people with SCIs are at risk of experiencing CRNA to their prescription medications despite having insurance coverage. Decision-makers for the national Pharmacare in Canada should account for their concerns judiciously.
4.3 Introduction

The cost of living with a spinal cord injury (SCI) has been estimated between $1.47 to 3.03 million Canadian dollars in a lifetime (Krueger, Noonan, Trenaman, Joshi, & Rivers, 2013; McDaid et al., 2019). Secondary complications and health conditions make people with SCI high users of medications in comparison to the general population (Krause & Saunders, 2009; Rouleau & Guertin, 2010). Prescription medications now constitute one of the topmost categories of healthcare costs in Canada. In 2018, pharmaceutical drugs represented the second highest expenditure for health care (16%, after hospitals, 28%), and expenditure growth on drugs outpaced that for hospitals and physicians (Canadian Institute for Health Information, 2018).

In Canada, prescription drug coverage is not included in the universal health insurance scheme. Instead, there are five possible ways that people pay for prescription medication: private drug insurance, employer-sponsored insurance, provincial drug benefits (for adults >65 years, people on social assistance, or those with catastrophic drug costs), inclusion on a family member’s plan, or out of pocket. For all of the insurance options, the extent of coverage varies, and individuals may pay premiums, copayments or deductibles. Out-of-pocket drug costs for Canadians have been estimated between $0-$2500 a year, depending on the medication costs, insurance, age, employment and income (Campbell, Manns, Soril, Clement, & Campbell, 2017; Ontario Citizens’ Council, 2012).

In the absence of universal Pharmacare, many people choose to forgo their medications due to cost, a phenomenon called cost-related non-adherence (CRNA) (Law et al., 2018). CRNA has been observed among chronically ill populations, and those with
lower income, irregular employment, lack drug insurance and high drug costs (Campbell et al., 2014; Hennessy et al., 2016). No studies were found that explored this phenomenon among people with SCIs, who are high users of medication, in addition to other healthcare services due to their disability. Within the SCI-specific literature, studies so far have explored medication use with respect to individuals’ demographic and injury-related characteristics, or the intensity and prevalence of adverse drug reactions and polypharmacy within the SCI population (Guilcher et al., 2018; Kitzman, Cecil, & Kolpek, 2017).

This study aimed to answer the following questions:

(a) What are the most common medications used by people with SCIs in Canada?
(b) What are the costs of different types of medications and to what extent are they typically covered by insurance?
(c) To what extent is CRNA practiced by people with SCI in Canada, and who is more likely to engage in CRNA?

4.4 Methods

*Study design and data collection.* It was an observational cross-sectional study (Lavarakas, 2008). Data were collected through an online survey (with a choice of telephone survey). Generally, people with SCI demonstrate higher levels of health and computer literacy, and often use online methods to access health information (Goodman, Jette, Houlihan, Williams, 2008; Mayman, Perera, Meade, Jennie, Maslowski, 2016). Additionally, many researchers and national level organizations in Canada have used online surveys to collect health related information from people with SCI (Munce et al., 2014; Noreau, Noonan, Cobb, Leblond, Dumont, 2014). Therefore, expecting that these
features would favour higher levels of participation in the SCI population, we used online survey as a primary method for data collection. Ethical clearance for the study was obtained from the Health Sciences Research Ethics Board (HSREB#6023360) of Queen’s University (Appendix A).

A questionnaire was developed and validated through pilot testing, expert consultation (MAM, SJTG & KS), and testing its alignment with the CRNA component of the Canadian Community Health Survey of 2016. The questionnaire had 35 questions, including branching and adaptive questioning (Appendix B). It was pilot tested with four participants for the sensitivity of its items, language, content, appearance, and functionality. Participants were reminded to assemble their medications and other necessary medical records before starting to answer the survey. "Qualtrics™"- a web-based survey software that is compliant with the Canadian privacy and accessibility standards was used for data collection (Gupta, 2017).

**Sample and setting.** The sample for the study comprised individuals living in the community with a SCI in Canada. A SCI was defined as an injury that occurs when trauma (such as a fall, vehicle accident, act of violence) or disease (such as a motor neuron disease, tumour, infection, myelopathy, vascular, toxic, metabolic condition or a developmental disorder) damages the spinal cord, resulting in partial or complete paralysis (Farry & Baxter, 2010). The proposed sample size for the study was between 120-140 participants, which was calculated based on a formula suggested for survey-based studies (Charan & Biswas, 2013) (Appendix C). The inclusion criteria included individuals with a SCI, aged 18 years or more, living in Canada, and who were prescribed one or more medications at the time of the survey. Individuals living in a nursing home or
hospitalized at the time of the survey were excluded. Approximately 85,000 people in the community live with a SCI in Canada (Farry & Baxter, 2010). The sample of our study was reasonably congruent with the sample of a recent national study of Canadians with a SCI (Noreau et al., 2014), except that our sample included more females.

In total, 189 individuals responded to the online survey, out of which 160 were included in the final analyses, based on the completion of their responses (completion rate: 85%). The duplicate or fake entries ($n=12$) were excluded. Duplicate entries were identified through the internet protocol (IP) address provided in the log files and fake entries were identified by finding series or pattern in the responses, low mean time to finish the survey, and/or finding illogical answers. The log file gives details on the participants’ IP address; time of completion of the survey; survey completion rate and number of questions completed. If more than one entry was found from same IP address, or time of completion of the survey was less than 5 minutes (actual average time: 15 minutes); or survey completion rate was less than 50% (which means that half of the questions were unanswered), then those entries were removed from the analysis.

**Recruitment.** Participants were recruited over six months (May-October 2018) with the help of community-based organizations. We contacted five national organizations working with individuals with traumatic or non-traumatic SCIs in Canada. Most of these organizations have a client database and a network of local chapters to serve their clients in different locations or cities. These local chapters used their regular email listserv or a newsletter to share the study information with their clients. A few organizations posted the study information on their website or social media page too. The study protocol was reviewed and approved before it was disseminated among their
clients. To ensure our recruitment did not exclude people who do not use the services of these organizations, we posted study information at local out-patient clinics, hospitals and pharmacies with the help of a representative from the local chapters or members of our research team. The posted information had a QR code that participants could scan to go the survey link. Participants were offered a $5 gift card for their participation (see appendices D-G for participant information letter and recruitment materials).

**Study variables.** To find independent factors associated with CRNA, we divided the study variables into dependent and independent variables as follows:

**Dependent variable**
- *Cost-related non-adherence (CRNA):* CRNA was our primary outcome variable that was measured by asking participants if they have ever taken less of their reported medication than prescribed because of cost in the last 12 months (yes/no). This question was asked for each prescription they reported on the survey.

**Independent variables**
- *Medication use:* We asked our participants to list all medications prescribed to them in the last 12 months. A prescription medication was defined as a pharmaceutical drug that legally requires a medical prescription to be dispensed. The reported drugs were classified under 14 categories as per the World Health Organization’s Anatomical Therapeutic Chemical (WHO-ATC) Classification system (World Health Organization, 2013). To classify the drugs reported on our survey, the first author (SG) located the reported drug name on the ATC classification guide and categorized it as per its therapeutic subgroup (level 1 & 2). The drugs that belonged to different ATC categories (example: anti-inflammatory drugs) were categorized as per their
indicated use reported by the participants. Any drug names (e.g. new drugs) that were not on the WHO list were classified based on the information available through an online database (drugbank.ca, version 5.1.2, released 2018-12-20) (Wishart et al., 2018). The third author (KS), who is a medical physiatrist, confirmed the accuracy of the prescribed medication classification as per the standard of practice or recommended use.

- **Drug coverage**: Participants were asked the type of drug insurance they have for every medication they reported on the survey. The response options included provincial drug benefits, private drug insurance, employer-based insurance, family-based insurance, or no insurance.

- **Medication costs**: This involved information on monthly costs directly borne by the participants for their drugs (after insurance coverage) such as in the form of copay, deductibles, or pharmacist dispensing fee (open-ended). All amounts were collected in 2018 Canadian dollars. Participants listed these costs for every medication they reported on the survey.

- **Socio-demographic and injury-related variables** included age, sex, employment status, province, personal income, cause of injury, level of injury, the extent of the injury, and time since injury (open-ended).

- **Additional health care costs**: We asked participants to estimate the amount they spend every month on their other healthcare needs or supplies such as catheters, wheelchairs, special diet, and skin care supplies (open-ended).

**Analyses and reporting.** Descriptive statistics were used to present socio-demographic, injury-related information, and the type and number of medications.
reported by the participants. ANOVA, χ2 and t-tests were conducted to analyze drug costs, insurance coverage and CRNA with respect to participants' socio-demographic, injury-related, and medication-related characteristics. Binomial logistic regression was used to determine independent factors associated with CRNA among study sample. Statistical Package of Social Sciences (IBM SPSS V.24) was used for data analysis.

4.5 Results

Sample characteristics. The average age of our sample was 47 (±13) years; 56% were females; 62% lived in Ontario, and 40% were receiving disability benefits as their primary source of income. Of the total sample, 58% had SCI due to a traumatic cause; more than 60% had paraplegia and incomplete injuries. One-third (33%) of the sample had cervical injury, more than half (52%) had a thoracic level injury and rest (15%) had an injury at or below lumbosacral level. Mean time since injury was 18 ± 13 years.

4.5.1 Prescription medication use, cost, and insurance coverage

Medication use (Table 9). A total of 832 prescriptions were reported by our sample, representing 296 different medications. On average, participants used 5.47 (±3.03) medications and 48% of participants used >5 medications concurrently. The most common medications belonged to the category of the central nervous system (38%) that comprised of antidepressants, anticonvulsants, anxiolytics, antipsychotics, and opioids, generally prescribed for neuropathic pain, depression, anxiety, sleep, or mood-related disorders. Other most commonly prescribed drugs included antibiotics (11%) for bladder infections, drugs for the cardiovascular system (8%), skeletal muscle relaxants (9%), and anti-spasmodic or anticholinergic drugs for genitourinary spasms (6%).
Table 9: Medication profile of study sample of community-dwelling persons with spinal cord injury (n=160)

<table>
<thead>
<tr>
<th>Code</th>
<th>Drug therapeutic class</th>
<th>N= 832 (100%)</th>
<th>No. of different medications</th>
</tr>
</thead>
<tbody>
<tr>
<td>N</td>
<td>Nervous system</td>
<td>309 (37.5%)</td>
<td>98</td>
</tr>
<tr>
<td></td>
<td>• Anticonvulsants/anxiolytics/antipsychotics</td>
<td>115 (14.1%)</td>
<td>29</td>
</tr>
<tr>
<td></td>
<td>• Hypnotics/sedatives</td>
<td>12 (1.5%)</td>
<td>5</td>
</tr>
<tr>
<td></td>
<td>• Antidepressants</td>
<td>76 (9.2%)</td>
<td>23</td>
</tr>
<tr>
<td></td>
<td>• CNS Stimulants</td>
<td>10 (1.2%)</td>
<td>5</td>
</tr>
<tr>
<td></td>
<td>• Opioids</td>
<td>85 (10.3%)</td>
<td>27</td>
</tr>
<tr>
<td></td>
<td>• Migraine meds</td>
<td>6 (0.7%)</td>
<td>5</td>
</tr>
<tr>
<td></td>
<td>• Neuroprotective agent</td>
<td>5 (0.6%)</td>
<td>4</td>
</tr>
<tr>
<td>M</td>
<td>Musculoskeletal system</td>
<td>137 (16.5%)</td>
<td>26</td>
</tr>
<tr>
<td></td>
<td>• NSAIDs</td>
<td>31 (3.8%)</td>
<td>13</td>
</tr>
<tr>
<td></td>
<td>• Skeletal muscle relaxants</td>
<td>95 (11.5%)</td>
<td>10</td>
</tr>
<tr>
<td></td>
<td>• Biphosphonates</td>
<td>11 (1.3%)</td>
<td>3</td>
</tr>
<tr>
<td>J</td>
<td>Anti-infectives for systemic use</td>
<td>92 (11.1%)</td>
<td>26</td>
</tr>
<tr>
<td></td>
<td>• Antibiotics</td>
<td>89 (10.8%)</td>
<td>24</td>
</tr>
<tr>
<td></td>
<td>• Anti-viral</td>
<td>3 (0.4%)</td>
<td>2</td>
</tr>
<tr>
<td>A</td>
<td>Alimentary tract and metabolism</td>
<td>76 (9.1%)</td>
<td>32</td>
</tr>
<tr>
<td></td>
<td>• Proton pump inhibitors, bile acid sequestrants</td>
<td>29 (3.5%)</td>
<td>8</td>
</tr>
<tr>
<td></td>
<td>• Laxatives or stool softeners</td>
<td>18 (2.2%)</td>
<td>7</td>
</tr>
<tr>
<td></td>
<td>• Anti-hyperglycemic</td>
<td>15 (1.8%)</td>
<td>7</td>
</tr>
<tr>
<td></td>
<td>• Anti-inflammatory</td>
<td>10 (1.2%)</td>
<td>7</td>
</tr>
<tr>
<td></td>
<td>• Anti-allergic</td>
<td>4 (0.5%)</td>
<td>3</td>
</tr>
<tr>
<td>C</td>
<td>Cardiovascular system</td>
<td>70 (8.4%)</td>
<td>42</td>
</tr>
<tr>
<td></td>
<td>• Anti-hypertensives</td>
<td>13 (1.6%)</td>
<td>7</td>
</tr>
<tr>
<td></td>
<td>• Diuretics</td>
<td>6 (0.7%)</td>
<td>4</td>
</tr>
<tr>
<td></td>
<td>• Anti-coagulants/anti-lipemic</td>
<td>30 (3.7%)</td>
<td>15</td>
</tr>
<tr>
<td></td>
<td>• ACE Inhibitors/B-blockers</td>
<td>21 (2.5%)</td>
<td>16</td>
</tr>
<tr>
<td>G</td>
<td>Genito-urinary system and sex hormones</td>
<td>62 (7.5%)</td>
<td>28</td>
</tr>
<tr>
<td></td>
<td>• Sexual hormones and birth control</td>
<td>11 (1.3%)</td>
<td>9</td>
</tr>
<tr>
<td></td>
<td>• Anti-spasmodic or anti-cholinergic</td>
<td>51 (6.2%)</td>
<td>19</td>
</tr>
<tr>
<td>L</td>
<td>Antineoplastic and immunomodulating agents</td>
<td>26 (3.1%)</td>
<td>12</td>
</tr>
<tr>
<td></td>
<td>• Antineoplastic</td>
<td>3 (0.4%)</td>
<td>2</td>
</tr>
<tr>
<td></td>
<td>• Immunosuppressants/modulators</td>
<td>20 (2.4%)</td>
<td>9</td>
</tr>
<tr>
<td></td>
<td>• Beta-Interferon</td>
<td>3 (0.4%)</td>
<td>1</td>
</tr>
<tr>
<td>R</td>
<td>Respiratory system</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>• Bronchodilators</td>
<td>14 (1.7%)</td>
<td>8</td>
</tr>
</tbody>
</table>
Medication cost and insurance coverage (Table 10). To determine the specific drug classes with high out-of-pocket cost and lower coverage, we chose the top 8 therapeutic drug classes most commonly prescribed to our sample. These drugs constituted almost 80% of the total medications prescribed to our sample. Table 10 lists the costs of different types of medications and the extent they are typically covered by insurance. Within these eight drug classes, opioids had the highest cost, and lowest insurance coverage. Following this, anticonvulsants, skeletal muscle relaxants, and antidepressants had the highest cost for patients, whereas antibiotics, drugs for alimentary tract and metabolism, and genitourinary spasms had lowest insurance coverage.

Overall, individuals with SCI spent a median of $49 per month (interquartile range: $234.75). The majority of the participants (92%) had some form of drug insurance to help cover the cost of the medications (provincial drug benefit: 48%; employer-based insurance: 14%; private insurance: 50%; family-based insurance: 11%). Also note that 36 participants (22.5% of total sample) had more than one type of drug insurance. Within the
group of participants who had some form of drug insurance, 45% paid less than $5 a month and 12% paid more than $100 a month ($6-$20/month: 18%; $21-$50/month: 17%; $51-$100/month: 8%). In terms of burden of medications (expenditure on medications as a portion of their annual income), almost 30% of the participants (n=46) paid more than 10% of their annual income on the prescriptions (32.5% paid between 1-10% and 38.8% paid less than 1%). Though it should be noted that finding low estimates for medication costs for some individuals is not necessarily true as it may be due to CRNA among persons with relatively low income.

Although in our sample, females and individuals between 26 to 64 years of age had higher drug costs and lower insurance coverage, these were not statistically significant than males and individuals in other age groups. Similarly, those who were employed full-time; and earning >$60,000/year had higher drug costs and better drug coverage, these were statistically insignificant in comparison with other groups.

With respect to SCI related characteristics, individuals with non-traumatic SCI incurred significantly higher cost on their drugs ($: 2.26; $: 0.026), despite having higher insurance coverage, in comparison to those with traumatic SCI. Individuals with paraplegia had significantly higher drug costs ($: 2.12; $: 0.035), though their drug insurance did not differ significantly from those with tetraplegia. The completeness of injury and time since injury did not have any significant influence on prescription drug cost or coverage among the study sample.
Table 10: Out-of-pocket costs, drug insurance coverage and prevalence of CRNA for most common medications used among study sample of community-dwelling persons with spinal cord injury (n=160)

<table>
<thead>
<tr>
<th>Top 8 drug classes</th>
<th>N</th>
<th>Monthly cost Mean ± s.d. (range) ($CAD)</th>
<th>Insurance availability n (%)</th>
<th>Prevalence of CRNA n (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1 Anticonvulsants/anxiolytics/antipsychotics</td>
<td>115</td>
<td>38.90 ± 91.31 (585)</td>
<td>95 (82.6)</td>
<td>9 (7.8)</td>
</tr>
<tr>
<td>2 Skeletal muscle relaxants</td>
<td>95</td>
<td>24.78 ± 94.51 (900)</td>
<td>86 (90.5)</td>
<td>11 (11.6)</td>
</tr>
<tr>
<td>3 Antibiotics</td>
<td>89</td>
<td>13.02 ± 20.80 (100)</td>
<td>63 (70.8)</td>
<td>7 (7.9)</td>
</tr>
<tr>
<td>4 Opioids</td>
<td>85</td>
<td>54.67 ± 97.32 (550)</td>
<td>55 (64.7)</td>
<td>26 (30.6)</td>
</tr>
<tr>
<td>5 Antidepressants</td>
<td>76</td>
<td>23.11 ± 69.11 (585)</td>
<td>65 (85.5)</td>
<td>12 (15.8)</td>
</tr>
<tr>
<td>6 Drugs for alimentary tract and metabolism</td>
<td>76</td>
<td>11.38 ± 21.03 (125)</td>
<td>62 (81.6)</td>
<td>7 (9.2)</td>
</tr>
<tr>
<td>7 Cardiovascular drugs</td>
<td>70</td>
<td>10.25 ± 13.40 (59)</td>
<td>58 (82.9)</td>
<td>5 (7.1)</td>
</tr>
<tr>
<td>8 Drugs for genitourinary spasms</td>
<td>51</td>
<td>19.91 ± 28.75 (144)</td>
<td>41 (80.4)</td>
<td>9 (17.6)</td>
</tr>
</tbody>
</table>

4.5.2 The extent of CRNA among the study population and its association with various factors

Extent of CRNA (Table 10). Among the total sample, 59 (37%) individuals reported CRNA. Within these individuals, 28 (48%) individuals reported CRNA for one medication, while 31 (52%) reported CRNA for two or more medications. The most common drugs that were forgone due to high costs were opioids, antidepressants, and drugs for genitourinary and muscular spasms (Table 11). Table 11 presents the distribution and extent of CRNA among the study sample. Concerning participants' socio-demographic and SCI specific characteristics, we found that females ($\chi^2$: 8.47; $p$: 0.004), individuals between 26 to 64 years ($\chi^2$:10.72; $p$: 0.005), and those with non-traumatic SCI ($\chi^2$:7.58; $p$: 0.006) were significantly more likely to experience CRNA than other groups. Individuals who reported CRNA spent an average of $398 per month on their medications while those without CRNA spent $80 per month ($t$: 4.90; $p$<0.001).
Those who reported CRNA spent an average of $674 per month on their additional healthcare needs in comparison to $241 paid by those without CRNA (t: 3.91; p<0.001).

Drug insurance availability and the number of medications used did not differ significantly between CRNA and non-CRNA group.

### Table 11: Distribution and extent of CRNA among study sample of community-dwelling persons with spinal cord injury (n=160)

<table>
<thead>
<tr>
<th></th>
<th>Total sample n (%)</th>
<th>Extent of CRNA n (%)</th>
<th>Test statistic, significance level and effect size</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Total sample</strong></td>
<td>160 (100)</td>
<td>59 (37)</td>
<td></td>
</tr>
<tr>
<td><strong>CRNA w.r.t socio-demographic characteristics of study sample</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>Gender</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Males</td>
<td>70 (43.75)</td>
<td>17 (24.3)</td>
<td>$\chi^2$: 8.47, $p$: 0.004*</td>
</tr>
<tr>
<td>Females</td>
<td>90 (56.25)</td>
<td>42 (46.7)</td>
<td>$V$: 0.23</td>
</tr>
<tr>
<td><strong>Age (years)</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>&lt; 25</td>
<td>12 (7.5)</td>
<td>3 (25)</td>
<td>$\chi^2$: 10.72, $p$: 0.005*</td>
</tr>
<tr>
<td>26-64</td>
<td>129 (80.6)</td>
<td>55 (42.6)</td>
<td></td>
</tr>
<tr>
<td>&gt; 65</td>
<td>19 (11.9)</td>
<td>1 (5.3)</td>
<td></td>
</tr>
<tr>
<td><strong>Employment status</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Full-time</td>
<td>33 (20.6)</td>
<td>9 (27.3)</td>
<td>$\chi^2$: 2.22, $p$: 0.038</td>
</tr>
<tr>
<td>Part-time</td>
<td>108 (67.5)</td>
<td>44 (40.7)</td>
<td></td>
</tr>
<tr>
<td>Unemployed</td>
<td>19 (11.9)</td>
<td>6 (31.6)</td>
<td>$V$: 0.11</td>
</tr>
<tr>
<td><strong>Annual income (SCAD)</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>&lt;20,000</td>
<td>66 (41.3)</td>
<td>26 (39.4)</td>
<td>$\chi^2$: 4.17, $p$: 0.244</td>
</tr>
<tr>
<td>20,001-39,999</td>
<td>56 (35.0)</td>
<td>24 (42.9)</td>
<td></td>
</tr>
<tr>
<td>40,000-59,999</td>
<td>22 (13.8)</td>
<td>6 (27.3)</td>
<td></td>
</tr>
<tr>
<td>&gt;60,000</td>
<td>16 (10.0)</td>
<td>3 (18.8)</td>
<td>$V$: 0.16</td>
</tr>
<tr>
<td><strong>CRNA w.r.t injury-related characteristics of study sample</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>Cause of injury</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Traumatic</td>
<td>93 (58)</td>
<td>26 (28.0)</td>
<td>$\chi^2$: 7.58, $p$: 0.006*</td>
</tr>
<tr>
<td>Non-traumatic</td>
<td>67 (42)</td>
<td>33 (49.3)</td>
<td>$V$: 0.21</td>
</tr>
<tr>
<td><strong>Level of injury</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Tetraplegia</td>
<td>53 (33)</td>
<td>14 (26.4)</td>
<td>$\chi^2$: 3.72, $p$: 0.054</td>
</tr>
<tr>
<td>Paraplegia</td>
<td>107 (67)</td>
<td>45 (42.1)</td>
<td>$V$: 0.30</td>
</tr>
<tr>
<td><strong>Extent of injury</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Total sample n (%)</td>
<td>Extent of CRNA n (%)</td>
<td>Test statistic, significance level and effect size</td>
</tr>
<tr>
<td>----------------------</td>
<td>--------------------</td>
<td>----------------------</td>
<td>---------------------------------------------------</td>
</tr>
<tr>
<td></td>
<td>n (%)</td>
<td>n (%)</td>
<td></td>
</tr>
<tr>
<td>Complete</td>
<td>59 (37)</td>
<td>20 (33.9)</td>
<td>$\chi^2: 0.35$</td>
</tr>
<tr>
<td>Incomplete</td>
<td>101 (63)</td>
<td>39 (38.6)</td>
<td>$p: 0.551$</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>$V: 0.09$</td>
</tr>
<tr>
<td>Time since injury</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>&lt;5 years</td>
<td>35 (21.9)</td>
<td>11 (31.4)</td>
<td>$\chi^2: 0.82$</td>
</tr>
<tr>
<td>6-19 years</td>
<td>59 (36.9)</td>
<td>24 (40.7)</td>
<td>$p: 0.664$</td>
</tr>
<tr>
<td>&gt;20 years</td>
<td>66 (41.3)</td>
<td>24 (36.4)</td>
<td>$V: 0.07$</td>
</tr>
</tbody>
</table>

**CRNA w.r.t. medication-related characteristics of study sample**

|                      | 5.47 (±3.03)       |                      |                                                   |
| Number of medications used |                    |                      |                                                   |
| CRNA group            | 5.85 (±2.87)       | $p<0.30$             |                                                   |
| Non-CRNA group        | 5.26 (±3.09)       | $d: 0.19$            |                                                   |
| Monthly expenditure on drugs* | 49 (0.00, 234.75)  | $t: 4.90$            |                                                   |
| CRNA group            | 398.08 (±579.03)   | $p<0.001$            |                                                   |
| Non-CRNA group        | 80.25 (±229.11)    | $d: 0.72$            |                                                   |
| Drug insurance availability | 144 (92.2%)       | $X^2: 1.07$          |                                                   |
| CRNA group            | 55 (93.3%)         | $p: 0.29$            |                                                   |
| Non-CRNA group        | 89 (98.9%)         | $V: 0.08$            |                                                   |
| Additional healthcare costs | 127.50 (25.00, 378.75) | $t: 3.91$ |                                                   |
| CRNA group            | 378.75             | $p<0.001$            |                                                   |
| Non-CRNA group        | 241.46 (±392.71)   | $d: 0.57$            |                                                   |

Note: * This indicates median and interquartile ranges. $d$ indicates effect size for t-tests while $V$ indicates Cramer’s $v$ for chi-square tests; w.r.t: with respect to

**Independent factors associated with CRNA (Table 12).** All of the above variables that showed significant association with CRNA during bivariate analyses were tested for their independent effect in the regression model. The null hypothesis for logistic regression analyses was that there are no associations between CRNA and participant’s age, sex, cause of injury, out-of-pocket costs on medications, and additional
health care costs. The model was built through a backward elimination method. The final model revealed that sex, monthly drug expenditure and monthly additional health care costs were significantly associated with CRNA among study sample. The results indicated that:

- Females were three times more likely than males to face CRNA after controlling for other factors, i.e. when they had similar monthly drug costs and additional healthcare costs.

- With every $50 increase in the monthly expenditure on drugs, the risk of CRNA increased by 28% after other factors were controlled. In other words, monthly drug expenditure of $81 increased the risk of facing CRNA among individuals with SCIs by 50%.

- With every $50 increase in the additional healthcare costs, the risk of CRNA increased by 5% after other factors were controlled. In other words, a monthly expenditure of $405 on additional healthcare needs increased the risk of facing CRNA among individuals with SCIs by 50%.

Please note that these estimates were controlled for the province in order to account for the variability among provincial drug programs in Canada (Campbell et al., 2017). As the medication costs among our sample were rightly skewed, we tested for its non-linear effects (if any) on CNRA through stratification. This was done to ensure that the linearity assumption in regression modelling is not violated. We tested the final model for any confounding or potential interactions between the variables. The sex variable acted as a confounder as it was related to both our outcome variable (CRNA) and one of
the main variables (medication costs). We also ran residual and influential diagnostics to find out any potential outliers that could affect our estimates. When model accuracy was calculated before, and after removing influential observations, no significant improvement in the accuracy was found. Therefore, those observations were not removed from the analyses.

Table 12: Factors associated with CRNA in the study sample of community-dwelling persons with spinal cord injury (n=160)

<table>
<thead>
<tr>
<th>Independent factors</th>
<th>B</th>
<th>S.E.</th>
<th>Wald</th>
<th>Sig.</th>
<th>OR</th>
<th>95% C.I.</th>
</tr>
</thead>
<tbody>
<tr>
<td>Monthly expenditure on drugs</td>
<td>0.005</td>
<td>0.001</td>
<td>13.321</td>
<td>0.000</td>
<td>1.28*</td>
<td>1.26 - 1.28</td>
</tr>
<tr>
<td>$50</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Additional healthcare cost</td>
<td>0.001</td>
<td>0.001</td>
<td>4.621</td>
<td>0.032</td>
<td>1.05**</td>
<td>1.04 - 1.06</td>
</tr>
<tr>
<td>$50</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Females</td>
<td>1.132</td>
<td>.420</td>
<td>7.261</td>
<td>0.007</td>
<td>3.103</td>
<td>1.362 - 7.071</td>
</tr>
<tr>
<td>Constant</td>
<td>-2.988</td>
<td>.541</td>
<td>30.495</td>
<td>0.000</td>
<td>.050</td>
<td></td>
</tr>
</tbody>
</table>

OR: Odds ratio (adjusted); Correct prediction: 79.4%; df: 1; Hosmer and Lemeshow goodness of fit test $\chi^2$: 8.716, df: 8, p: 0.367; -2 Log likelihood: 156.332; Cox & Snell $R^2$: 0.288; Nagelkerke $R^2$: 0.393; Results are adjusted for province.

4.6 Discussion

This study is among the initial studies in Canada that explore the phenomenon of CRNA within the context on SCI. Findings suggest the following:

- Individuals with SCIs used 5-8 medications concurrently and spent a median of $49 per month (or $588/year) on prescribed medications.
- Although 90% of participants had some form of drug insurance, 37% reported CRNA.
• Opioids, antidepressants, and drugs for genitourinary and muscular spasms were most likely to be forgone due to costs.

• Being female, having higher drug expenditure and higher additional health care costs were significantly associated with CRNA.

International research has shown that annual household out-of-pocket spending over $500 a year perpetuates the risk of underusing prescription medications due to cost (Kemp, Roughead, Preen, Glover, & Semmens, 2010). This partially explains why the prevalence of CRNA among people with SCIs was found dramatically higher (37%) than that is reported for the general population (8-10%) and comparable to other vulnerable populations in Canada (36%) (Gupta, McColl, Guilcher, & Smith, 2018).

We also noted that out-of-pocket costs paid by the participants highly varied, suggesting that some people paid far more than the average while others paid far less. Two possible reasons can explain this. First, the variability in medication needs depending on one's age, and health status may have led individuals with SCI to differential use of medications. Second, the differences in the drug insurance coverage within people with SCIs may have caused them to bear different costs for similar medication needs. The sample of this study varied in terms of age, income, employment, and province they lived in. The differences in these characteristics might have affected their drug insurance arrangements and hence, their expenditure on drugs.

With respect to the type of medications, our study found that medications generally prescribed for pain, mental health conditions, muscular and urinary spasms were most likely to be forgone due to cost. A recent national study in Canada also found that medications for mental health conditions were most commonly reported for CRNA.
(Law et al., 2018). It must be noted that we could not identify specific medications that had a higher cost burden or prevalence of non-adherence for our participants. This was because medications that had restricted coverage and high costs for some people had no such implications for others, which affected patients' adherence decisions for those medications.

Another notable finding of this study was that people with SCIs faced CRNA despite having drug insurance and in some cases, more than one type of drug insurance. This finding can be explained by many possible reasons. First, several of these plans involve income-based deductibles (even for those with disabilities), which may be difficult to meet and have posed a financial burden on the individuals. Income-based deductibles have been demonstrated to lead to a reduction in overall drug use, especially for low-income and vulnerable patient populations (Law et al., 2019). A second possible explanation of this finding is the heterogeneity in the extent of drug coverage and cost-sharing arrangements provided under public versus private health insurance plans (Baicker & Goldman, 2011; McKillop et al., 2018). Previous research in Canada has shown that people who lack private drug insurance are more likely to face CRNA (Lee, Morgan & Lee, 2017). Of 70% of our sample who had public drug benefits only, they did not have additional drug coverage that a private health insurance offers. Another additional explanation for this finding can be that even a low deductible associated with public drug plans were difficult to manage for some people with SCIs. This may be especially true for people who are less than 65 years and have low-income with high additional healthcare costs. These costs may have posed an additional burden in such a way that they couldn’t afford to pay even a small deductible.
We also found that the burden of additional healthcare costs directly influenced the ability of individuals with SCIs to afford medications. This finding partly explains the differences in the barriers faced by people with disabilities as compared to the general population (Gupta, McColl, Guilcher, & Smith, 2019). It is important to note here that the additional health care costs that were considered for this study comprised costs for catheters, wheelchairs, special diet, or skincare supplies. These costs did not include cost for over-the-counter medications, home modifications, vehicular adaptations, attendant care, and cost of other rehabilitation services that people with SCIs may need. If these extra costs of living are accounted, the barriers to pay for medications may become even worse. Recent findings by Persaud et al. (2019) and Goldsmith et al. (2017) also indicate that there is a strong connection between financial burdens for medicines and other household needs, including other health expenditures.

It was also noteworthy that females were found to face a significantly higher risk for CRNA. This can be attributed to the fact that women are generally less likely than men to be employed, which affects their income and access to employer-based health insurance (Pelletier, Patterson, & Moyser, 2019). This finding aligns with the previous research that has shown that women with disabilities have the worst access to healthcare, including ability to afford medications from any other group such as men with and without disabilities and women without disabilities (Sakellariou & Rotarou, 2017).

4.7 Study limitations

As our study adopted a cross-sectional approach, it was not possible to capture how medication use, cost, or non-adherence changed among study participants over a period of time. The study is based on self-reported data that cannot be validated by
external data. Participants may under- or overestimate the medication costs and consequently lead to potential information bias. The recruitment process depended on voluntary participation, therefore, just like all surveys, our survey was also limited by its reliance on participants to provide accurate and honest responses. A 12-month retrospective time period to determine the occurrence of CRNA might be unequally estimated by participants compared to a more recent situation, which may introduce recall bias. Choice of an online survey was a limitation for those who are either not able to afford internet or do not use internet, though we provided the choice for a telephone survey. Lastly, due to the relatively small sample size, it was difficult to know whether our sample was reflective of the target population despite similarities in some key demographic characteristics.

4.8 Future directions

Future research can be directed towards understanding the impact of multiplicity and complexity of drug insurance arrangements on patients' adherence. Health services and policy researchers should also focus on the development and promotion of tools that inform cost-sensitive prescribing. These guides may help clinicians to choose between therapeutically similar medications with similar anticipated outcomes but often substantially higher prices. This study has implications for the proposed plans for a “universal” Pharmacare program in Canada by the federal advisory council. This study suggests that it may be an appropriate policy solution given evidence of patients falling through the cracks of the complex private/public system of Canada, even if 92% of them are apparently “covered” by a private or public plan of some kind.
4.9 References


Adherence, 12, 1699–1715. Retrieved from https://doaj.org/article/d8b4be3ec83246678566840f0e6a2650


Research, 46(D1), D1074-D1082.

Chapter 5

Managing medication cost burden: A qualitative study exploring experiences of people with disabilities in Canada

5.1 The Fit of Manuscript 4 within the Dissertation

Manuscript 4 builds upon the findings of the first three manuscripts by developing an understanding of the processes underpinning the medication cost burden through a qualitative inquiry. This particular study extended the findings obtained from the quantitative study and depicted the processes an individual with a disability such as a SCI might undergo or engage in after receiving a costly prescription and their tangible and non-tangible impacts at various levels.
5.2 Abstract

Despite the abundant literature on the burden of rising costs of prescription medications, there is limited research to explore how these costs affect people and the decisions they are forced to make within the context of disability. In this qualitative study we explored strategies adopted, factors influencing, and the impact of some of these strategies to manage the burden of medication cost among persons with disabilities. We interviewed 12 adults with spinal cord injuries living in Canada, using a general inductive approach to analyse data. We found that before cutting back on medications due to costs, participants generally tried and sought help from the government, employers, and/or their prescribers to improve their drug coverage. The key factors that participants considered while making decisions on the strategies included the cost and perceived importance of medications, their financial status, other competing needs, and their relationship with the prescribers. While some of their efforts were successful, many participants were still not able to obtain their medications as prescribed. In those cases, patients resorted to rationing strategies such as cutting back on medications, other essential needs, or selling assets. These strategies had serious implications on their health, healthcare utilization, and quality of life.
5.3 Introduction

Despite the abundant literature on the treatment burden of chronic illnesses (Demain et al., 2015; Rosbach & Andersen, 2017; Sav et al., 2013), there is limited research to understand the processes that underpin medication cost burden in general and especially among those with disabilities. The financial cost of living with a disability is substantially higher than that of living without a disability (Mitra, Palmer, Kim, Mont, & Groce, 2017). Our previous research has shown that the economic burden of medications faced by people with disabilities is higher than that experienced by their non-disabled counterparts (Gupta, McColl, Guilcher & Smith, 2020). For people with disabilities, barriers to income and employment, higher susceptibility to health complications, complex medical regimens, additional health care costs, and the complexity of drug and social assistance programs perpetuate the risk of forgoing medications (Gupta, McColl, Smith, & Guilcher, 2019).

Prescribed medications for people with disabilities are often assumed to be covered under the provincial drug plans but many are not. This leads to a situation where people stop taking their medications (generally referred to cost-related non-adherence, CRNA) or adopt other rationing behaviours due to cost (Piette, Heisler, Horne, & Alexander, 2006). However, we do not know what strategies are used to cope with the cost burden of medication, how people are making medication rationing decisions, and the impact of rationing strategies on individuals, their families, and the healthcare system.

The literature that does exist has been mostly quantitative, generally examining the prevalence or risk factors associated with CRNA. It includes older adults or individuals with chronic illnesses but is not specific to the experiences of people with
disabilities (Gupta, McColl, Smith, & Guilcher, 2018). Similarly, most commonly used theories that exist to define and understand medication non-adherence include the World Health Organization’s framework of general medication adherence (2003), Andersen and Newman’s healthcare utilization model (2005), and a model developed by Piette and associates (2006) (Anderson & Newman, 1973; Piette et al., 2006; Sabaté, 2003). However, all of these frameworks represent the factors that affect or increase someone’s risk of facing non-adherence to medications. None of these theories extend and depict the processes an individual with a disability might undergo or engage in after receiving a costly prescription.

To address this paucity of research, our research study explored the experiences of persons with disabilities managing their prescription drug costs. The specific objectives of this study were to find:

- The strategies that participants adopted to manage their medication cost burden.
- The factors that influenced an individual’s decisions to adopt those strategies.
- The impact of rationing strategies on individuals.

### 5.4 Methods

**Design.** This was a qualitative study wherein we adopted a general inductive approach to explore and describe experiences of the participants with medication cost pressures. The inductive approach helped to establish clear links between the study objectives with the raw data obtained from the interviews; and convey key themes or processes related to the underlying experiences of medication cost pressures among the
participants (Thomas, 2006). The ethical clearance for the study was given by the Health Sciences Research Ethics Board (HSREB#6023360) of Queen’s University.

**Context.** The rising cost of prescription drugs has been leading to the rising incidence of CRNA in Canada (Law et al., 2018). The prevalence of CRNA is two to five times higher in Canada in comparison to other countries with universal healthcare (The Commonwealth Fund, 2016). Although Canada has a universal public health insurance program, it excludes universal coverage for prescription drugs (The Commonwealth Fund, 2016). People are either covered by the private plans, mostly provided by their employers; or provincial drug benefit plans that cover seniors (>65 years), people on social assistance, or people with catastrophic health needs (Kratzer, Cheng, Allin, & Law, 2015). As a majority of the people with disabilities remain unemployed or underemployed, they either have to forgo their medications because of a lack of private insurance or have to rely on provincial drug benefits (if they qualify) to cover the costs of their medications.

Even when medications are covered by the public drug benefit programs, the extent of coverage varies extensively and individuals have to share the costs of medications in the form of premiums, copayments, and/or deductibles. Calculated according to the individual’s income, these deductibles may range from 3% to 13% of annual household income that comes around $2000 to $9000 for a middle-income family (Daw & Morgan, 2012; Hanley & Morgan, 2009). People who often work for small employers or are self-employed remain ineligible for the means-tested/income-based public drug programs (Dewa, Hoch, & Steele, 2005; Tang, Ghali, & Manns, 2014).
The growing evidence on the cost-related barriers faced by many Canadians, especially those with low-income, poor health status, and disability, has raised many equity concerns, challenging the core principles of the Canada Health Act (i.e., universality and comprehensiveness). According to the 2017 Canadian Survey on Disability, 13% of all people with disabilities \((n = 836,690)\) had unmet needs for prescription medications due to cost (Stats Canada, 2018). The prevalence of CRNA within people with disabilities is higher than that is found in general population in Canada (10%), suggesting that people with disabilities are more likely to face cost-related barriers to fulfil their medications (Stats Canada, 2018).

**Sample.** The sample for the study consisted of people with traumatic or non-traumatic spinal cord injuries (SCI) living in Canada. SCI affects around 85,000 Canadians living in the community (Farry & Baxter, 2010). The estimated lifetime economic burden associated with a traumatic SCI in Canada ranges from $1.47 million for a person with incomplete paraplegia to $3.03 million for one with complete tetraplegia (Krueger, Noonan, Trenaman, Joshi, & Rivers, 2013). People with SCI are high users of medications, taking between 5–14 different medications concurrently, often with complex regimens (Guilcher et al., 2018). Therefore, individuals with SCI represented an ideal population of high users of medications for this study due to the chronic nature of their condition, the presence of multiple consequences of injury and the level of disability associated with an SCI (Kitzman, Cecil, & Kolpek, 2017; Rick Hansen Institute, 2017).

The sample for this study was selected from a subset of participants who reported facing cost-related barriers or were self-identified non-adherent participants in our first
study (Gupta et al., 2020). In this first study, the overall sample was recruited with the help of community partners working with people with SCI including SCI Canada, Rick Hansen Institute, Canadian Spinal Research Organization, and other community-based organizations. An email invite was sent to their clients or the study information was posted on their e-newsletters or websites. To invite participants for this study, an email explaining the study details was sent to those participants who reported CRNA (n = 59) in the first study, of which 12 gave consent to participate for qualitative interviews. Written informed consent was obtained before interviews. Participants were offered $20 for their time and participation (see appendix H for participant information letter and consent form).

Data collection. A semi-structured interview guide (Table 13) was developed based on the objectives of the study and the key findings obtained from the quantitative phase of the study (Gupta et al., 2019). The interview guide was pilot tested with two participants and revised in light of their responses. The interviews were conducted either in-person or by telephone, depending on the availability and choice of the participants, over a period of six months starting from August 2018 to January 2019. Interviews were conducted by the first author, who was a PhD candidate and a trained occupational therapist with clinical experience of working with individuals with SCIs. The interviews lasted between 40–60 min. The interviews were recorded using two audio-recorders and transcribed verbatim. To maintain anonymity, all personal identifiers were removed before data analysis.
Table 13. Interview guide

You indicated in your survey that you sometimes have trouble paying for your medications, and that you even sometimes don’t take your medications as prescribed because of the cost. In this interview, I’d like to ask you more about that.

1. How often does this happen? When is the last time it happened?
2. Does it happen more often with some medications than with others?
3. How do you decide what to take and what not to take, or how to adjust your dosage to save money?
4. Do you tell your doctor when you are doing this? Do you and he/she discuss the cost of the medications prescribed?
5. What do you do to manage or cope with medication cost burden?
6. What about other supplies and non-prescription medications—do you ever economize on those?
7. What is your priority for spending on things for your or your family’s health?
8. How do you think your health has been affected by not taking your pills as prescribed, or by other compromises you have had to make because of costs?

Data analysis. The coding process in inductive thematic analysis started with the preparation of raw data files after data cleaning; close reading of the text to understand the content; the identification and development of general themes and categories; the re-reading to refine the categories and reduce overlap or redundancy among the categories; and creating a framework incorporating the most important categories (Guest, MacQueen, & Namey, 2012). The first two authors (SG, MAM) coded the four transcripts independently to ensure inter-coder consistency and peer examination of the codes developed by the first author. The coding scheme was confirmed and corrected by the senior authors for any imprecise code definitions or overlapping of meaning in the coding scheme. Eighty percent of the total codes were identified within the first eight interviews while no new subcategories emerged after 10 interviews were completed. Two
more interviews were conducted to confirm thematic saturation in data. These additional interviews verified that saturation is based on the widest possible range of data on the emerged subcategories. This process increased the comprehensibility of analysis and provided a sound interpretation of the data. The NVivo software was used to manage the data (QSR International, 2019).

**Research rigor.** Research rigor was ensured through an audit trail, member checking, and peer debriefing (Lincoln & Guba, 1985). For an audit trail, a logbook was maintained that contained the notes on the data collection process, the analysis process, and the final interpretations. To ensure the credibility of the findings, the transcribed interviews and result summary were made available to all the participants in order for the participants to assess and correct the accuracy of their accounts. Out of twelve, ten participants agreed with the result summary and the rest did not respond to our request. The research team met at regular intervals to provide critical inputs on the research methods and lead researcher’s interpretation of meanings and analysis. The peer-review process involved deliberations and debriefing of the emerging codes, categories, and their relationship with the data. We used the consolidated criteria for reporting qualitative research (COREQ) checklist (appendix I) to report the methods and findings of the study (Tong, Sainsbury, & Craig, 2007).

**5.5 Results**

The details of our sample are given in Table 14. Table 15 presents the coding scheme and definitions we adopted for the respective subthemes under each category. Figure 4 depicts the interplay of these subthemes which will be discussed further.
Table 14. Participant characteristics (n = 12)

<table>
<thead>
<tr>
<th>Participant Characteristics</th>
<th>n/Median (q1, q3) *</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (years)</td>
<td>57.5 (46, 58.25)</td>
</tr>
<tr>
<td>Females</td>
<td>8</td>
</tr>
<tr>
<td>Relationship status</td>
<td></td>
</tr>
<tr>
<td>• Married or in a relationship</td>
<td>5</td>
</tr>
<tr>
<td>• Single, divorced or never married</td>
<td>7</td>
</tr>
<tr>
<td>Education</td>
<td></td>
</tr>
<tr>
<td>• Up to high school</td>
<td>2</td>
</tr>
<tr>
<td>• College degree or certificate</td>
<td>7</td>
</tr>
<tr>
<td>• University degree and above</td>
<td>3</td>
</tr>
<tr>
<td>Work status</td>
<td></td>
</tr>
<tr>
<td>• Employed</td>
<td>2</td>
</tr>
<tr>
<td>• Retired</td>
<td>1</td>
</tr>
<tr>
<td>• On disability income</td>
<td>8</td>
</tr>
<tr>
<td>• Unpaid disability and unemployed</td>
<td>1</td>
</tr>
<tr>
<td>SCI-related characteristics</td>
<td></td>
</tr>
<tr>
<td>• Traumatic</td>
<td>6</td>
</tr>
<tr>
<td>• Paraplegia</td>
<td>9</td>
</tr>
<tr>
<td>• Incomplete</td>
<td>6</td>
</tr>
<tr>
<td>Time since injury (years)</td>
<td>20 (10, 28.5)</td>
</tr>
<tr>
<td>Median number of medications 1</td>
<td>9.5 (5, 13)</td>
</tr>
<tr>
<td>Monthly out of pocket cost of medications 1 (CAD)</td>
<td>316.5 (181.25, 398.75)</td>
</tr>
<tr>
<td>Type of insurance 2</td>
<td></td>
</tr>
<tr>
<td>• Public drug benefit program</td>
<td>8</td>
</tr>
<tr>
<td>• Employer based insurance</td>
<td>4</td>
</tr>
<tr>
<td>• Family based insurance</td>
<td>1</td>
</tr>
<tr>
<td>• Other</td>
<td>1</td>
</tr>
<tr>
<td>• No insurance</td>
<td>2</td>
</tr>
</tbody>
</table>

1 Include both prescribed and over-the-counter medications; 2 The numbers do not add up to 12 as a few participants had more than one type of drug insurance coverage. * q1 and q3 refer to the first (25th percentile) and third (75th percentile) quartiles respectively.
### Table 15. Coding scheme

<table>
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<tr>
<th>Strategies that participants adopted to cope with medication cost burden</th>
<th>Factors affecting decisions</th>
<th>Impact of rationing strategies</th>
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<td>Factors that influence participants’ decisions to adopt rationing strategies</td>
<td>Consequences of forgoing medications due to cost or financial stress caused due to medications</td>
</tr>
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<td>1. Trying to access public or employer-based drug benefits</td>
<td>1. Cost and perceived importance of medications</td>
<td>1. Decline in quality of life</td>
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<tr>
<td>statements when participants tried to go back to work, access public drug benefits, or other social assistance to manage medication cost burden</td>
<td>statements where participants decided to stop or continue taking a medication due to its cost and their perceived importance or severity of their health condition for which they were prescribed medication</td>
<td>statements when participants indicated that medication related financial burden reduced their general well-being or life satisfaction for them or their family members</td>
</tr>
<tr>
<td>2. Seeking help from prescribing doctors</td>
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</tr>
<tr>
<td>statements where participants shared that they requested their doctors or pharmacist for less expensive substitutes such as generics or over-the-counter medications</td>
<td>statements where participants stopped or rationed medications depending on their financial status or availability of financial resources at that particular time</td>
<td>statements when participants indicated that rationing medications due to cost has led to increase or worsening of their symptoms or experienced pain</td>
</tr>
<tr>
<td>3. Economizing on general or healthcare needs</td>
<td>3. Competing demands on resources for self and others</td>
<td>3. Psychological stress</td>
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<td>statements where participants shared that they cut back on other healthcare supplies or needs such as assistive devices, or general needs such as food, clothing, car, or leisure-related activities to manage medication cost burden</td>
<td>statements that reflected that participants made decisions to stop or ration on medications based on their priorities for their children, other significant health needs or basic life needs</td>
<td>statements when participants indicated that medication related financial burden caused mental or psychological stress</td>
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<tr>
<td>4. Rationing medications</td>
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</tr>
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<td>statements where participants shared that they decided to stop a medication temporarily or for a long duration, or taking smaller or less frequent doses, or postponed refill of a medication due to high cost</td>
<td>statements where participants stopped or rationed on medications in consultation with their doctors or lack thereof</td>
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<td></td>
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<tr>
<td>statements that reflected participants tried to sell their assets or borrowed money to manage financial burden due to medications and other healthcare costs</td>
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Overall, we found five key strategies adopted by our participants to manage the burden of medication cost. Out of these five, the first two were non-rationing strategies while the remaining three were rationing strategies. Before cutting back on prescriptions due to cost, participants adopted non-rationing strategies. They proactively tried and sought help from the government, employers, or their prescriber to improve their coverage or opt for more affordable or less expensive drug substitutes. While some of their efforts to manage costly prescriptions were successful, many participants were still not able to take their medications as prescribed. In those cases, patients resorted to rationing strategies such as cutting back on medications, other essential needs or selling assets. The key factors that participants considered while making decisions on these strategies included cost and perceived importance of drugs, their financial status, the basket of financial resources they had to meet competing needs, and consultation with the
prescribers. The rationing strategies caused financial stress and had serious implications on their health, healthcare utilization, and quality of life.

**Strategies to manage medication cost burden.** Five main strategies that were identified, in order of frequency, included:

1. **Trying to access public or employer-based drug benefits.** A common initial strategy that participants described was obtaining drug benefits through public drug insurance or employer-based insurance. Some participants who were on disability support tried to improve their coverage through more generous public drug benefit programs, such as exceptional drug coverage which allowed access to drugs that are not usually listed or covered through the provincial drug formulary. This is illustrated by a participant who shared:

   [Drug name 1] was not covered by the provincial Pharmacare, even though [drug name 2] was. I applied for it [Drug 1] under exceptional drug coverage, and they approved it on a yearly basis like I have to apply for it every year. [P/6]

   Participants described staying in current jobs to maintain employer-based drug insurance. Others who were self-employed at the time of the interview considered different employment to secure this type of drug coverage. A participant who had been self-employed in the past shared:

   I am considering going back to work. Part-time, but if needed, full-time too. Right now, I am concerned about money to survive until retirement. [P/5]
A few participants with a progressively worsening medical condition shared that they tried to maintain their jobs in order to remain eligible for employer-based drug benefits despite their worsening health situation. As a participant shared:

I had shift work, and it was very hard on me. In the last years, I tried to make it steady, but my body couldn’t take it. A lot of my disabled friends told me that I was practically killing myself to get those benefits. [P/9]

Participants who tried to access more generous public drug benefit programs also shared many challenges they faced while doing so. Examples of the challenges include: the complex paperwork, eligibility requirements, the heavy deductibles, and the delay or wait times to get accepted for the drug program. These challenges are highlighted in the quote below, as the participant describes trying to access a public drug benefit program, known as Trillium drug coverage:

Trillium, they have a huge [deductible] … you have to use up the deductible first, in order for it to be covered. My pills don’t work out to more than the deductibles, so it doesn’t apply to me. I tried to get on ODSP [Ontario Disability Support Program]. I have been turned down three times. We are (in) No Man’s Land. [P/3]

Participants without jobs or other regular incomes had to wait for the approval from the Trillium program. The wait time spanned from a few days to sometimes several weeks. This resulted in some participants managing without medications for that wait time.

It is frustrating as I have to submit [paperwork] first, to my insurance company, to my former employer, and then wait for them to get back to me, and then send all of this to Trillium. It’s such a delay. It is very frustrating because you may never know when you will get the money. [P/7]
It was evident from the participants’ accounts that while some of their efforts were successful, many participants were not able to improve their coverage.

2. Seeking help from prescribing doctors. The next common strategy that participants adopted to manage the burden of medication cost was to seek support from their doctors—either asking them to prescribe less expensive substitutes and alternatives or to help them enroll in a drug benefit program. Many times, doctors were able to help their patients access drug benefits or to prescribe less expensive and equally effective substitutes.

My doctors, all … my psychiatrist, my back specialist, and my family doctor they know I don’t have any benefits, so they always prescribe the cheaper alternative or over the counter alternative…or whatever, just for that reason. [P/2]

However, in some cases, this strategy was not successful, especially when generic or other effective substitutes were not available on the public drug formulary or not covered by the public drug benefit program. A few participants shared:

My doctor was concerned about me. She would give me the samples for some medication because I had to cut back and she didn’t want to see me pay out of pocket. But with [drug name], it was too costly, and she couldn’t help me there. She tried to find cheaper substitutes or the generic versions, but they were of the same price range. [P/4]

A participant suffering from a neurological condition shared how her doctor was cognizant of her financial situation as well as the time lag to get approval for the public drug benefits. As a result of this awareness, he applied for the benefits on her behalf well before her medical appointment.
My neurologist has to do that [application for drug benefit], and he is aware of the time-lapse. It has to be done every year. One year, he missed sending in the Section 8 form, and I was without drugs for a whole year. That would have been $4000–$5000 a month if I didn’t have coverage. [P/5]

Although a participant’s relationship with their doctors played a major role in managing the burden of their drug cost, the participant had to initiate the conversation about the barriers they faced to purchase their medications. This is illustrated by one participant here:

My family doctor does not [ask me about medication costs]. Most of them do not ask that question. I have to bring it up. The urinary urgency one … it was about $300. I couldn’t get it for several months. I went to my urologist and told him that I couldn’t get it because I couldn’t afford it. At that point, he prescribed one that was less expensive. [P/5]

3. **Economizing on general or other healthcare needs.** Participants who considered their medications essential shared that they either cut back on general needs such as food, clothing, car, leisure-related activities, or other healthcare supplies and needs—such as assistive devices. For example, a participant said:

I have stopped buying new clothes, and I shop at the thrift store. I have changed my spending habits, to make sure that I keep my health. Things like a gym membership, have also gone … [P/5].

Participants’ accounts showed that when they had to choose among the general necessities such as food, clothes, heat, or car; clothing and leisure-related activities were the first to be eliminated or reduced. Basic or more important needs such as food and heat were last to go.
I could not have cut back on food, as you can’t survive without eating. But I didn’t buy clothes for years. We didn’t go on any trips forever! I don’t remember the last time we went on a trip. We just cruised along from paycheck to paycheck. [P/4]

Most of these decisions were based on their health status, self-assessed health priority at the time, and their socio-economic status, as illustrated by this quote:

I have been needing new tires for my chair for two months (laughs). You have to make choices. Right now, my pressure sores are bad, and so that is my priority. I am not really going out much, stuck in bed, so the chair becomes my second priority. [P/9]

4. Rationing medications. Participants also described taking smaller or less frequent doses; either postponing a refill of medication or stopping their medications for a period of time. For example, a participant shared “[I] had to postpone an antibiotic by a week or till I got paid” [P/2]. Also, having access to benefits, or other financial resources had a significant effect on the frequency or timing of rationing medications. A participant shared:

When I know I am down to the last few, if I have ten pills left today and I know I am not getting paid for three weeks, I’ll take one every other day. I’ll stretch it out till I get money. [P/7]

A few participants also shared incidents when they were completely going without medications for a long period of time. To manage such situations, they chose between the two medications and sacrificed one to manage the other that was more important and/or costly to buy.

… Last month I could not afford [drug name] at the end of the month. So, I took my other pills, some of my three months pills I took a couple
of days, so I could afford the [drug name]. Now, right at this moment, I am short again until my husband has the money coming in. Right now, I am taking one of one pill today, and one of the other tomorrow to get me through till the weekend. [P/3]

Participants tried stretching-out medications slowly and gradually, depending on their perception regarding the effect of cutting back on medications and their financial situation.

… The recession hit, I was the last one hired, and first to go … For the next five years, I was doing temporary jobs. There were no benefits and I was paying out of pocket. That is when I started cutting back on the medication because I couldn’t afford to take it a hundred percent. It didn’t matter if I missed a needle. I thought I was going to die if I missed a needle. Turns out I was not, and I didn’t feel any difference. I thought if I missed two needles, then I would be affected. And like that, I missed three and four … I was taking medications for only 50 to 30 percent of the time. I was taking one needle a week, instead of every day. [P/4]

5. Selling assets or borrowing money. This was the last alternative that participants sought to manage the burden of their medication cost, especially when they had high additional disability-related expenses. They often tried to sell their house or car; use their available line of credit or borrow money from friends. Participants in our study who were living alone or did not have a significant other along with more complex healthcare needs had a greater tendency to consider selling their assets. When asked, “how do you manage this burden”, a participant living alone after a recent divorce from her husband shared:

I have considered selling my house and going to an apartment. I have considered selling my car and getting something cheaper. [P/5]
Participants who were living with their partners were generally more inclined to borrow money or use their line of credit, until the partner managed to find a job or other sources of financial support.

We got a huge line of credit on the house. We went into debt. We re-mortgaged the house and seriously considered selling the house. We just kept forking out this money and kept hoping we would win the lottery or something. [P/4]

A few participants who neither had any assets to sell nor had a line of credit were more reliant on family members for financial support. Relying on the family for personal needs, especially for adults aged 30 to 60 in our study, resulted in a feeling of guilt or emotional stress, as illustrated by a participant here:

I am the lucky one. I get support on top of ODSP because my family can give me some money. It is an unpeaceful relationship. I don’t want to keep that relationship, but I have to because it is the only way I can survive on income that I get from ODSP. [P/1]

Factors affecting decisions. This category involves factors that influenced participants’ decisions on the strategies adopted to manage the burden of their medication cost. Subthemes included:

1. Cost and perceived importance of medications. The first factor that influenced participants’ decisions regarding economizing on medications was the out-of-pocket cost of the medication. When participants had to choose between two medications for the same health condition, they often chose the ones that were more affordable to them. Sometimes, participants kept both medications, but they rationed the one that was costlier while taking the other one as prescribed.
I usually continue the three months one, because it’s cheap. The other one I cut off, because its, you know, sixty-seven dollars a month. I’ll stretch out the other one. [P/3]

In a few instances, participants chose between two medications that offer similar benefits, but have contrasting health risks and purchase price levels. For example, a participant who needed medication for sleep and pain shared that she chose the one that was less costly, though potentially harmful:

Given the choice of taking [drug name] and go to sleep, which is like a dollar, versus this other stuff which is $75 a bottle, I have to go with the highly dependent and habit-forming drug, unfortunately. [P/8]

In situations when participants were taking multiple medications for different health conditions, or multiple medications for a single health condition without much cost-difference, they often chose a medication based on its perceived importance, or severity of a negative health consequence should the medicine be eliminated or reduced, in a calculation that considered the individual’s budget and broad range of medical issues.

It [drug name] certainly cannot be used interchangeably for the heart or blood thinning either. But I make sure I cover that. I have to cover. That means if I have to do without something else ... I have to do without something else. [P/1]

In some incidents, decisions regarding choice of medications were often based on intuitive trial and error, especially when the cost of two medications did not differ much, or when participants were information-poor about adverse effects of choosing one medication over the other.

I am trying to decide what I can cut back. I am not really sure which one was good, because its trial-and-error. You try it, but you are not really sure if it is helping at all. [P/5]
2. Financial status and availability of resources. The second important factor that drove participants’ decisions on the strategies to manage the medication cost burden was the availability of insurance or other financial resources at a particular point in time. For example, participants who did not have regular employment or income, such as individuals who were self-employed or students, had to cut back and make choices between medications and other needs until their financial situation or insurance status changed.

Then I was not a student, and so I had no coverage. I was self-employed. My insurance was $100 a month. As a self-employed person, I could not afford that. It really was a double-edged sword. [P/8]

A participant who, at one point in her life, regularly took her medications as prescribed, had to eventually cut back because of her decreasing financial status.

There is a bunch of medicines I was taking over the years, which I could afford because the business was doing well. But because I have a fixed income and I am a single mom, I am going to have to cut back … [P/5]

Another participant who was injured in an automobile accident and had insurance decided to pay for all of his medications out of pocket after insurance coverage was exhausted.

I was covered by the Manitoba Automobile Insurance till it ran out. It was a $200,000 fund. I have used it up over the thirty years. It ran out about three or four years ago. Now I pay for all of that [medications and other disability-related expenses] out of pocket. [P/6]

3. Competing demands on resources for self and others. A third factor that determined participants’ decisions regarding managing the burden of medication cost was the need for acquiring certain personal and inter-personal benefits, or the need to avoid or
reduce certain personal and inter-personal threats. For example, some individuals, when their financial situation made them choose between medications and other basic needs such as housing, decided to manage without medications. For example, a participant shared:

I have to pay the bills first, otherwise, I don’t have a place to live! (laughs) For little things like medications, even though I need it, I workaround. [P/3]

Some of the participants described their serious financial hardships due to medication cost burden when they had to choose between having food or looking after their children, over paying for and optimally taking their medications.

I have shared custody over our son … I eat a lot of peanut butter when he is not around. I put my own needs aside so that I can look after my son. [P/7]

When they had more financial flexibility, they cut off things that were relatively less important or desirable versus the ones that were essential, i.e., medications. For example, a participant who had a chronic neurological condition decided to cut off additional help for their household chores, as well as other complementary medications before cutting back on necessary prescribed medications.

I have a cleaner and somebody to mow my lawn … somebody to help with my groceries… those things have fallen off. All the complementary medicines have all been cut off. [P/5]

In situations where participants had children or other dependents, they chose the needs of their children over the need for medications necessary to maintain their own health.
I would definitely choose his health over my health … I needed to cut back on some of my alternative medication so that I could buy him hockey equipment. [P/2]

4. Relationship and advice from prescribing doctors. Participants’ decisions regarding rationing or balancing the cost burden of medications was also influenced by their relationship with their doctors. Some participants shared that if they had a good relationship with their doctors, they were generally able to discuss their financial situation and the limited ability to afford their medications. In the majority of cases, in doing so, they were able to find options to afford their medication and avoid non-adherence. A participant who was on disability support income and had her doctor’s support found alternative ways to fund her medications, or at least make rationing decisions in consultation with her doctor.

They [my doctors] try to adapt and try to get something else … they try to get special coverage … I had doctors who would write several letters to make that special request … most times it is turned down by the government … in the end, I will do without … but doctors have been pretty good. [P/1]

A few participants did not have a highly supportive relationship with their doctor and felt that they had no opportunity to discuss the medication cost burden with them. Sometimes they felt that doctors did not have enough time for candid discussions within a short consultation period. In such cases, participants made purchase decisions on their own, depending on their affordability, and/or the perceived importance of the medication. For example, a participant said:

I don’t get the opportunity to discuss anything with him [neurologist], because he just does his neurological examination and then I am dismissed. There is no opportunity for discussion. [P/11]
Another participant shared that because she is served by a teaching hospital, this inhibits her ability to form a relationship with her doctor where she can discuss her finances and her inability to afford some of her medications.

I am at a teaching facility and every time I go, it is a different resident. I don’t find anybody who knows my file and I find educating them each time difficult. Sometimes that falls through the crack. [P/12]

**Impact.** This category highlights the consequences of rationing strategies adopted by participants to manage medication cost burden. Subthemes included:

1. **Decline in quality of life.** Not having enough money to pay for their basic medications and fulfil other basic needs had a negative impact on participants’ standard of living and overall quality of life. Financial hardships due to the burden of medication cost affected their security and freedom to live and enjoy life without worrying. They reported that they felt vulnerable, had a sense of scarcity, and were in constant stress managing the basic expenses within minimum financial resources. For example, a participant said:

   I haven’t bought new clothes in a few years. In terms of grocery shopping … I get by with anything in the freezer or in the cupboard. [P/7]

Sometimes their financial struggles were marginal while in other cases participants’ struggles were intense. Participants who were managing with a restricted income and who were navigating their chronic conditions alone frequently found themselves struggling with debt, having their credit seriously impaired, and using all of their savings:
I don’t do any recreational stuff. I don’t go out much either. I am still paying a mortgage, unfortunately, property taxes and all that great stuff. My whole cheque is eaten up… you could say I am living cheque to cheque. [P/9]

2. **Exacerbation of symptoms.** Several participants quoted that discontinuing medications had a negative impact on their health condition and led to the exacerbation of their symptoms. Some participants experienced the worsening of their symptoms much earlier and acutely while they were skipping doses. For example, a participant who was rationing between two medications for pain and depression shared:

> When I skim the [drug name 1], I notice the depression increases, and if I skim the [drug name 2], then my pain symptoms increase. They are progressing, and if I don’t take [drug name 2], then the symptoms would get much worse, which would then stress me out even more. [P/7]

Another participant who couldn’t afford her medications for a month experienced some of her symptoms worsening.

> I haven’t had it for four weeks, because it wasn’t in my budget this month. I thought I would be okay, but I am not. I can see that I have white patches under my eyes, and I am bruising easily. Just four weeks off it, and I can see I am pale. [P/9]

In other cases, participants noted the ill-effects of forgoing medications were evident much later when their health started deteriorating. For example, one of the participants who stopped one of her medications for a period of six months in the past was noticing a worsening of her symptoms and caught infections due to her compromised immunity:

> A while ago, I wasn’t able to get my [drug name], as it cost $90 a package, which was way beyond my coverage … I did stop for a long time, as I could not afford it. It’s an obscene cost. Now, I am able to identify the symptoms and know what is going on … I have a lot of flare-ups, to the point that it covers a lot
of my face. It’s very painful, and it brings on a Staph infection and that brings on a Strep infection, like flu. It can be very aggressive and painful. [P/8]

3. **Psychological stress.** Many of our participants shared that along with their physical health, their mental health was affected by not having enough money to pay for their medications.

The psychological stress of it all is the biggest burden. It’s so not measurable, and it’s there every day. What are you going to buy? Where are you going to cut back? What do you need to buy for the future? Like, the wheelchair, I think what do I need to cut back to afford that? That is a lot of pressure. [P/12]

The quote below is illustrative of a situation when participants were caught in a vicious cycle of stress which subsequently made their symptoms worse.

I didn’t have any [financial] support. I lost sleep, which causes immunodeficiency, which causes stress, which causes cold sores, which causes loss of sleep. It was just a cycle. [P/10]

In a few cases, participants’ accounts also showed that along with psychological stress, they faced emotional strain due to their restricted financial situation. For example, a participant who was on a disability support income had to ask her parents to buy some of her medications, which contributed to her feeling of humiliation and embarrassment.

I had to ask my parents for my meds, and it’s very, very humiliating and embarrassing. They are 90 and 91. Fortunately, they have been able to help me out but it causes a strain on them. I don’t like to do it, unless I absolutely have to. [P/7]

4. **Increased healthcare utilization.** The impact of the burden of the cost of medications was not just limited to patients’ physical and mental health but affected the public healthcare system too. Participants shared incidents when they had to visit the
hospital more often or stay at the hospital for long periods in order to access drugs needed
to manage their health condition. One of our participants was kept in the hospital for four
extra days because the medication he needed was not covered by any drug insurance plan
and was extremely costly to purchase.

When I was in the hospital, I was on an antibiotic for a bone infection … I was
allergic to the IV, so I had to take a pill … that pill was 1000 dollars a day and
only covered if you are in the hospital … They wanted to release me from the
hospital, but I had another four days to take the pill … so I had to stay at the
hospital for an extra four days … to get the 1000 dollar pill … [P/2]

Another participant who lived in a smaller city and had no access to an
interventional treatment involving a costly drug that she needed, was sent to the other
hospital in a city nearby every week for her treatment. She was being reimbursed for all
the costs involved.

I met a doctor in [city] who decides to send me to [name] hospital in [city]. This
hospital … what it does is it used a freezing element to freeze my nerves at the
back and my neck to relieve some of my pain. It did help to some degree. But
that meant every single week I am on the bus staying at a hotel for one night.
Incredibly costly to the healthcare system [P/1]

Overall, participants felt that cost savings to the healthcare system made through
restricting drug coverage would be negated. People who stop taking medications or
cutting back on food will ultimately end up using the healthcare system more frequently.

If a person is not able to access nutrition, they will use the healthcare system
more, and that is costly for the government … same for the provincial
government, if the ODSP is cut, then people will need ADP [assistive devices
program] more. If it doesn’t come out from one pocket, it will come out of
another. [P/10]
5.6 Discussion

To our knowledge, this is the first qualitative study in Canada that has explored patients’ experiences with managing the burden of the cost of their medications within the context of special vulnerable populations, such as those with a SCI. We found that before deciding to stop taking medications, participants tried a few non-rationing strategies to access their essential medications (Figure 4). Participants proactively tried and sought help from different potential sources including government, employers, or their doctors. They either tried to improve their coverage through public drug benefits or opted for less expensive drug substitutes. However, while some of these efforts were successful, many participants were still not able to fulfil their medication needs without facing financial hardships. In those cases, patients resorted to other strategies that had serious implications for them. For example, they economized on other healthcare or basic needs such as food, clothing, or leisure-related activities. Some considered selling their assets or borrowing money to manage their medications. In some instances, participants used those strategies in combination while in some only one of these strategies were used.

Even when individuals were covered through provincial drug benefits, they faced many systemic barriers including complex paperwork to meet eligibility requirements, heavy deductibles, restrictions on public drug formularies, and long wait times for getting approvals, all of which resulted in forgoing medications due to cost. These results support the findings from other studies that suggest that just having insurance does not safeguard individuals from facing financial hardships completely (Devoe et al., 2007; Goldsmith et al., 2017). This is specifically true for the people with disabilities who, despite receiving
subsidized care, face deprivation due to extra costs of living with a disability and may find even small copayments difficult to manage. Even among people with disabilities, we found that our participants were on a continuum of facing financial stress due to medications. Participants who were at the extreme end of financial stress often were living alone, had higher overall healthcare costs, poor financial resources, and poor social support.

The factors that participants considered while making rationing decisions included drug costs, their perceptions around the negative consequences of skipping medications, the basket of financial resources they had to fulfil their needs and that of their family, and the relationship with their doctors. Evidently, while most of these decisions were solely made by patients themselves, some decisions were guided or sanctioned by their doctors. Awareness by the medical professionals of the adherence barriers in patients has long been identified and proven as one of the effective strategies to protect against the non-adherence of medications (Nieuwlaat et al., 2014). To do this effectively, doctors need to be cognizant of the potential burdens for all patients and help those in need to navigate these barriers through care coordination. For example, there have been guidelines to help prescribers consider the generic and branded names of drugs, their market cost, and availability of insurance coverage when treating their patients with chronic medical conditions (Alberta College of Family Physicians, 2018).

The other main finding of the study was that financial barriers to medications not only affected patients’ physical health, but also impacted their overall quality of life, and highlights the costs and inefficiency impact to the healthcare system. Patients were trapped in a vicious cycle of stress and financial scarcity which led them to cut back on
basic needs or ask for money from friends or family. These stressors caused exacerbation in their symptoms which ultimately caused them to use more healthcare services, both related and unrelated to their medications. Studies involving other populations have also found that treatment burden operates in a cyclical manner and often has a catastrophic impact on patients, and their households which further trickles down to the healthcare system (Demain et al., 2015; Rosbach & Andersen, 2017; Sav et al., 2013). Studies from Canada, in particular, demonstrate that sometimes doctors have to keep their patients in the hospitals to allow access to drugs that are costly and not covered outside the hospitals (Guilcher et al., 2017; Wang, Lemmens, & Persaud, 2017).

5.7 Policy implications

This study has identified a complex array of choices made by the patients to access prescription medications. Yet they were not fully successful in improving the access. This lack of access adversely impacted their health and healthcare utilization. Thus, the issue needs to be addressed at the level of the healthcare provider, the healthcare system, community, and social services, employment, and insurance policies.

Recently, in response to the report from the Standing Committee on Health, the Canadian government announced the formation of a national advisory council on the implementation of an affordable Pharmacare in the 2018 budget (Government of Canada, 2018). The report of the advisory council that was recently released, recommended a universal single-payer public prescription drug system for Canada. This recommendation has given high hopes to Canadians, though how the implementation of these recommendations would benefit people with disabilities, in particular, remains elusive.
We propose that as Canada contemplates national Pharmacare, there are some short-term and long-term solutions that might be considered. In the short-term, prescribing healthcare professionals can play a key role by being cognizant of their patients who are at risk of out-of-reach medication costs, i.e., individuals without regular income or employment, with poor health status and disabilities, and those having high healthcare and prescription medication needs. Healthcare professionals can minimize this risk by choosing medications that have better coverage or lower costs for their patients. Healthcare professionals also need to let the patients define what out-of-reach financial costs are, as for some patients, even $50 per month can be out-of-reach. In the longer run, we propose that the government look into eliminating the systemic barriers of drug insurance arrangements (i.e., access and cost). The processes to get coverage through the provincial drug benefit programs should be simplified (e.g., yearly forms to be filled out by physicians is inefficient and burdensome to patients and physicians, especially for disabilities that are life-long). Public drug formularies should provide generous coverage to the drugs that are required by people with diverse disabilities. The heavy deductibles to meet eligibility requirements for exceptional drug coverage should be reconsidered and wait times to get approvals should be minimized.

Our results also showed that the minimum income afforded to individuals on disability requires that they ration their funds to access other healthcare needs or supplies. Government establish poverty cut-offs based on a minimum income required to access just enough food, housing and other basic necessities (Stats Canada, 2020). However, the extra costs of living incurred by people with disabilities do not count in those basic needs. In this case, many people with disabilities may appear to have minimum basic income on
paper to fall above poverty line, though in reality they may not have enough money to meet the minimum standards of decent living. Therefore, revisiting a minimum basic income for individuals with a disability would be important. Social assistance programs, including drug benefit programs need to consider the burden of extra costs of living with a disability. Employers may also ensure that disabled employees are appropriately accommodated, for example, with part-time work without restricting their access to group health benefits. These solutions may not only help Canadians with disabilities but also any Canadian who faces a financial barrier to access their necessary prescriptions.

5.8 Study limitations

This study has several limitations. Since our sample was small and comprised of people living with SCIs in Canada, our results may have limited generalizability to other populations or context. It would be interesting to compare our results with studies that explore the burden of medication cost for people with other types of disabilities. Because we used a qualitative approach at one point in time, we could not explore how these experiences changed over a period of time or with advancing age or disability. Although Canada is a multi-lingual country, the interviews were only conducted in English due to the feasibility for the research team. Conducting interviews in other languages might have allowed us to capture other diverse views. Some of the interviews were conducted telephonically which might have led to the absence of visual cues from participants' accounts. Though, telephone interviews are judged to be equally rich, detailed, and of high quality as the face-to-face interviews (Novick, 2008). Also, as the interviews were primarily conducted by the first author, the results might have been influenced by her own bias and experiences. However, we employed a number of methods to maximize
rigor, including member checking, peer review, rich description, and maintaining field notes.

5.9 Conclusions

The burden of medication costs among people with disabilities is overwhelming. Results of our study provide an in-depth evaluation of the processes underpinning as well as the after-effects of rationing or forgoing medications due to costs, for people living with complex disabilities such as those with a SCI. Despite the efforts made by the participants to improve coverage and fulfil their medication needs, many had to face serious hardships to manage their medication cost. These stressors exacerbated their symptoms which ultimately caused participants to use a greater range of healthcare services more frequently, both related and unrelated to their medications. These findings have important implications for healthcare policymakers to minimize systemic barriers within drug insurance programs, and prescribing healthcare professionals to be vigilant for patients who may encounter financial barriers fulfilling medications as well as future researchers to identify medication cost burden among people with other types of disabilities.

5.10 References


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Chapter 6

Discussion and Conclusion

This discussion and conclusion chapter is based on the integrated findings obtained from the quantitative and qualitative studies presented in chapters 2-5 and published separately. The integration of the findings was done through a narrative approach in a staged manner (Fetter, Curry, & Creswell, 2013). The staged approach to integration occurs “in multistage mixed methods studies when the results of each step are reported in stages as the data are analyzed and published separately” (Fetter et al., 2013, p. 2142), which is the case in this thesis.

Using the mixed-methods sequential explanatory design, the quantitative and qualitative data was connected at the intermediate stage when results obtained from the quantitative data analysis informed data collection of the qualitative study and guided the formation of the semi-structured interview guide. The quantitative and qualitative studies were also connected while selecting the participants for the qualitative study and conduct follow-up analysis based on the quantitative results. The development of the qualitative data collection tool was grounded in the results from the quantitative study to investigate those results in more depth through collecting and analyzing the qualitative data. Finally, an overall interpretation was framed, which is presented in this chapter, and implications of the integrated findings on future research, policy and practice are discussed. Figure 5 describes this process and depicts the various stages of integration, along with the specific aspects of thesis that were explored in subsequent phases or informed the subsequent phase.
Figure 5. Integration of quantitative and qualitative study findings

- Methods: Online survey of individuals living with a SCI across Canada (n=160)

- Prescription medication use, drug coverage availability and out-of-pocket costs; extent and determinants of medication nonadherence due to cost

- Purposefully selecting individuals who reported CRNA on the online survey.
- Developing interview questions to expand on: strategies to manage medication cost; role of prescribers and health system; consequences of medication cost barriers and non-adherence.

- Methods: Personal/telephone interviews with 12 individuals with SCI who reported underusing medications due to cost on the survey.

- Strategies adopted to manage medication cost burden.
- Factors that influenced an individual’s decisions to adopt those strategies.
- Impact of rationing strategies on individuals.

- Interpretation and discussion of implications and areas for future research.
• Study 1, which was a scoping review provided an analysis of the type, extent, and quantity of research available on cost-related non-adherence (CRNA) to prescription drugs in Canada. This study used the Piette model (2006) to systematically summarize the evidence for extent, determinants, and consequences of CRNA to prescription medications among Canadians. It also revealed an evidence gap for CRNA among people with disabilities and acted as a solid ground based on which other parts of the thesis were built.

• Study 2 was a theoretical paper that argued for the reconsideration of existing models on medication adherence. The study critically appraised the Piette model (2006) and suggested an adapted model to examine CRNA within the context of disability.

• This adapted model served as a conceptual framework for study 3 that quantified the magnitude, risk factors and the impact of medication non-adherence among people with SCI. Specifically, the study assessed prescription medication costs and coverage and CRNA among individuals with SCI. The survey instrument used in this study was developed based on the five groups of elements identified in the adapted model as having important implications on medication non-adherence within the context of disability (appendix J).

• Lastly, study 4 aimed to understand the processes underpinning medication cost burden through a qualitative inquiry. This study provided an in-depth understanding of certain aspects that were included in the online survey but remained unanswered, such as the role of prescriber- and system-related factors in managing medication cost burden (appendix J). The study offered a nuanced understanding of the complexities
involved in making decisions for medication rationing as well as its tangible and non-
tangible impact at various levels.

These four sets of studies that included a scoping review, a theoretical paper, a
quantitative study and a qualitative study, contribute to an emerging understanding of
factors affecting and implications of medication costs and non-adherence to medications
among vulnerable populations such as people with SCI in Canada. In this final chapter, I
summarize the key findings of the thesis that emerge from the four studies pooled
together, which are:

1. People with SCI spend more on prescription medications than the average Canadian
   and face a higher risk of facing financial barriers to access their medications.
2. Having drug insurance did not alleviate medication cost burden, and the type of
   insurance arrangement, i.e. public, private or a combination of both did not affect the
   risk of prescription underuse due to cost.
3. Other healthcare costs are one of the most important factors that affected cost-related
   medication non-adherence in people with SCI.
4. People with SCI used a hierarchical five-step strategy to deal with excess costs of
   prescription drugs.

From these findings, I offer recommendations to improve financial access to
medications among people with disabilities in general and people with SCI in particular,
within Canada. I also discuss the strengths and limitations of the thesis, and present
implications of the findings for future research, policy, and practice in the field of
healthcare and rehabilitation. In the end, I summarize some of the efforts I have made for
knowledge translation.
6.1 Summary of key findings

6.1.1 People with SCI spend more on prescription medications than the average Canadian and face a higher risk of facing financial barriers to access their medications.

The first notable finding of this thesis was that the average out-of-pocket cost spent by individuals with SCI on prescribed medications (around $197 a month) was five times higher than the average spending of $450 a year by a general Canadian household (Stats Canada, 2015). An apparent reason behind this finding is the heavy use of prescription medications by people with SCI. In our study, we found that 296 different medications were prescribed to the participants. Almost 50% of individuals were using more than five prescribed drugs concurrently, mostly for pain, spasticity, infections, and mental health conditions such as depression, anxiety and mood disorders. Recent studies report similar results and suggest that people with SCI use between 5–14 different medications, often with complex regimens (Cadel et al., 2019; Patel, Milligan, & Lee, 2015). The high out-of-pocket costs on medications in our sample was also related to the extent of non-adherence they reported. International research has shown that annual household out-of-pocket spending over $500 perpetuate the risk of underusing medicines due to cost (Kemp, Roughead, Preen, Glover, & Semmens, 2010). This explains why the prevalence of cost-related non-adherence among people with SCI was higher (37%) than that is reported for general (9.6%), or senior (8%) populations, adults with chronic conditions (4%-31%), or other vulnerable populations (26%) in Canada (Gupta, McColl, Smith, & Guilcher, 2018).
6.1.2 Having drug insurance did not alleviate medication cost burden and the type of insurance arrangement i.e. public, private or a combination of both did not affect the risk of prescription underuse due to cost. Total out-of-pocket costs paid in the form of copayments, deductibles or co-insurance determined CRNA.

Another notable finding of this thesis was that people with SCI faced financial barriers fulfilling their medication needs despite having drug insurance and, in some cases, more than one type of drug insurance. As found in the quantitative study (study 3), 92% of our participants had at least one of these insurance arrangements: provincial drug benefits; private drug insurance such as employer-based, family-based or other supplementary private drug insurance; or combination of both public and private insurance. We found that regardless of the type of insurance, individuals with SCI faced CRNA. This suggests that neither the public nor private drug insurance arrangements alleviate the financial burden of medications.

Many possible reasons can explain these findings. First, several of the provincial drug plans (British Columbia, Ontario, Manitoba, Quebec, and Saskatchewan) for seniors and those on social assistance involve income-based deductibles, copayment or co-insurance. Deductibles are the amounts that a patient pays out-of-pocket on the eligible drugs they buy before the coverage begins. Even for seniors, depending on the Canadian province, these deductibles can range from 1% to 5.5% of their net income or $100-$400 annually. For example, in Quebec, the provincial government, in 1996, introduced mandatory drug insurance for people not covered by employer-based or any other privately purchased drug plan. Under this plan, the previously insured beneficiaries (i.e. seniors and people on social welfare), who had first-dollar coverage for medications were
posed a set monthly deductible of $21.75 and 37% co-insurance for their medications.
Income-based deductibles have been demonstrated to lead to a reduction in overall drug
use for low-income and vulnerable populations (Law et al., 2019). These costs may have
been difficult to meet and pose a financial burden on the individuals in our sample.

For individuals less than 65 years of age and not receiving income support from
the government, provincial governments typically provide following three options: a
catastrophic drug coverage with deductibles ranging from 3% to 20% of annual income
plus a co-insurance of up to 35% (British Columbia, Ontario, Newfoundland and
Labrador, Nova Scotia, Prince Edward Island, Saskatchewan); a voluntary drug insurance
plan (Alberta, New Brunswick); or a mandatory drug insurance with mandatory yearly
premiums, and varied copayments or co-insurance (Quebec) (Brandt, Shearer, Morgan,
2018). It is important to note that the catastrophic drug coverage comes into play only
after a deductible limit is met. For example, the Trillium Drug Program in Ontario poses
a deductible at 4% of after-tax household income before the drug costs are covered by the
plan. Similarly, in Alberta, people must pay a premium of about $720 per year plus 30%
of prescription cost afterwards. In Newfoundland and Labrador, someone earning
$40,000 must spend around $3,000 in medication first each year to start being insured.
Therefore, it is possible that drugs that participants felt were non-urgent, or the drugs for
which the costs were less than a deductible threshold were forgone by our participants.

Analyses also showed that the cost-sharing (i.e. out of pocket drug costs paid by
the participants) were highest for those obtaining insurance either privately or both
publicly and privately, while the publicly insured individuals had lower levels of cost-
sharing. This means that with private health insurance too, high cost-sharing posed on
prescription drugs may have led to a reduction in medication usage among our sample. Additionally, under some drug insurance plans, coverage is provided through a reimbursement system where patients pay the total medication costs first and submit a claim for reimbursement. Therefore, it is a possibility that participants found it challenging to come up with such fees to be paid upfront and decided not to obtain a medication.

Other additional explanation for the finding that insurance does not mitigate CRNA completely can be that even a low deductible associated with public drug plans were difficult to manage for some people with SCI. This may be especially true for people who were less than 65 years and had low-income with high additional healthcare costs in our sample. The additional costs for over-the-counter medications, medical supplies such as catheters, wheelchair cushions or skincare supplies may have posed an additional burden in such a way that they could not afford to pay even a small deductible.

Additionally, the complexity involved in accessing public drug insurance benefits might have caused CRNA to the participants who were only eligible for or had access to publicly financed programs. In the qualitative study of the thesis, participants provided insights to challenges such as complicated eligibility requirements, hefty deductibles, and delay or wait times to get approvals, associated with public drug benefit programs. Besides, restrictions posed on the public drug formularies in terms of the medications that are covered and the maximum amount that is paid for different drugs might have contributed to the medication underuse. These barriers might have offset the drug benefits provided by the government long enough that caused people to skip their medications due to costs.
6.1.3 Additional healthcare costs are one of the most important factors that affected cost-related medication non-adherence in people with SCI.

The third notable finding of this thesis was the burden of out of pocket drug costs and additional healthcare costs directly influenced the ability of individuals with SCI to afford medications. The scoping study of the thesis listed the five most important factors that are associated with the risk of medication non-adherence due to cost in the general population: age, income, health status, insurance coverage, and out-of-pocket cost on medications. Age, income, insurance coverage and cause of disability were associated with CRNA in the SCI population as well when taken individually. But multivariate analysis rendered all of these factors insignificant when additional health care costs and medication costs were considered in the equation. As found in study 3, additional health care costs that comprised of other healthcare supplies such as bowel care supplies, bladder care supplies such as catheters, wheelchairs, special diet, and skincare supplies for people with SCI averaged around $400 per month, which is quite significant. Also, note that these costs did not include cost for over-the-counter medications, home modifications, vehicular adaptations, attendant care, and cost of other rehabilitation services that people with SCI need. If these extra costs of living are accounted for, the barriers to pay for medications may become even higher. These additional costs pose a burden that may have led individuals to ration their medications over other essential or healthcare needs. This finding explains the differences in the barriers faced by people with disabilities as compared to the general population (Mitra et al., 2017). It answers the question of what perpetuates the risk of facing CRNA between people with and without disabilities.
Further, this finding has important implications at the policy level. The poverty cut-offs established by the government are based on a minimum income required to access enough food, housing and other necessities. However, the extra costs of living incurred by people with disabilities do not count in those basic needs. In this case, many people with disabilities may appear to have minimum basic income on paper to fall above the poverty line. In reality, they may not have enough money to meet the minimum standards of decent living, as described in the poverty limits or cut-offs. Therefore, any public assistance scheme, including national Pharmacare, that does not consider the additional barriers faced by people with disabilities may not fulfil its intentions of financial risk protection in a true manner.

**6.1.4 People with SCI used a hierarchical five-step strategy to deal with excess costs of prescription drugs.**

The fourth notable finding of this thesis was that it found a cascade of participants' efforts before they begin to cut back on medications or adopt other rationing behaviours. The qualitative study of the thesis found five key strategies that participants took in a stepwise manner to manage the burden of medication cost. Out of these five, the first two were *non-rationing* strategies that included their efforts to improve their drug coverage and to seek help from the prescribers. When these were not successful, participants resorted to rationing strategies such as cutting back on medications, or other essential needs and selling assets.

Individuals’ strategies to tackle medication cost burden are essential to know because solutions to avoid medication rationing due to cost can be built upon these. Available research thus far suggests the adverse effects of medication underuse on
individuals’ health and other social outcomes. For example, one major pan-Canadian study reports that millions of Canadians forwent basic needs, such as food, heat, and other health care expenses because of drug costs (Law et al., 2018). In study 3, we also found that around 10% of the study participants spent less on food or heat, and 5% borrowed money to obtain necessary medications. However, the qualitative study, for the first time, reported on individuals' efforts before they resort to rationing strategies.

These participants’ efforts can be combined with system supports to avoid medication non-adherence due to costs. For example, patient benefits can be achieved if prescribers can have real-time information on drug benefits at the point of prescribing. A few US-based companies have made pharmacy benefit manager (PBM) tools and services (Lyles, 2017). These tools allow prescribers/pharmacists to compare two drugs for their patients, so they choose an alternative medicine that has better coverage or low-price for their patients. The drug insurance mechanisms can also be simplified. Many private health insurance plans have variable copays or co-insurance for various services. For instance, a plan that covers dental, prescription and eye care services might have a $50 copay for a visit to a dentist, 25% co-insurance for every prescription, but a $20 copay for every visit to an optometrist. Sometimes copayments also vary for the services provided at different facilities. Individuals may also have a different deductible if more than one family member is covered under the plan (Levy, 1992).

Similarly, administrative hassles associated with public drug plans can be reduced. In the qualitative interviews, participants alluded to the certain challenges that they face while accessing public drug plans. Available research also suggests that provincial differences within public drug plans lead patients with similar needs to bear
the different costs for medications (Demers et al., 2008). Therefore, healthcare managers can look into these strategies used by patients to deal with excess costs of prescription drugs and find out ways that support patients to avoid medication rationing.

In the next section, I discuss the strengths and limitations of the thesis, and present implications of the findings for future research, policy and practice.

6.2 Strengths of the doctoral thesis

The first strength of this thesis is its contribution to theory development using a systematic approach. This doctoral study resulted in two proposed theoretical frameworks. First is an adapted model for understanding the complex phenomenon of CRNA among people with disabilities (study 2). Second is a conceptual model on approaches to manage medication cost burden within the context of disability (study 4).

The thesis began with a scoping review. In the next step, I studied the related theories on the subject and suggested an adapted approach to understand the phenomenon within the target population better. The adapted model served as a tool for thinking about the aspect of CRNA among people with disabilities. The third study, using a quantitative approach, applied the adapted theoretical framework and identified the most critical predictors and confounders to the phenomenon in the population of interest. Some of the complex characteristics were further explored in the fourth study, using an inductive qualitative study. The final study resulted in a conceptual model that provides insights into decision-making involved while managing medication cost burden and impact of the same on individuals. These frameworks may inform future studies examining the phenomenon of medication underuse due to cost in other populations or contexts.
The other key strength of this thesis relates to its methodology. For the last few decades, quantitative approaches have dominated to inform the policy-oriented work. Though in recent years, the need for diverse approaches to guide solutions to complex health policy issues has been emphasized (Curry, Nembhard & Bradley, 2009; Fetter, Curry, & Creswell, 2013). To fulfil objective 3, this thesis adopted an explanatory sequential mixed-method approach. In this approach, quantitative data collection and analysis were followed by the qualitative data collection and analysis and two were integrated in a staged manner (Creswell & Clark, 2011; Fetter, Curry, & Creswell, 2013). The mixed-method approach provided a cohesive way to understand medication cost barriers among people with disabilities and a more meaningful interpretation of the data and the phenomenon being examined (Teddlie & Tashakkori, 2003). As illustrated in figure 6 above, while the quantitative phase found the most relevant factors that determine the risk of facing medication cost barriers among people with SCI. The qualitative study provided a rich characterization of these factors and extended our understanding of the impact of medication cost burden that emerged from participants’ accounts.

Another key aspect of the thesis was that it used an online survey (with a choice of telephone survey) to collect data from the participants. The choice of an online survey provided several benefits. First, as most of the people with SCI use wheelchairs, employing an internet-based method for data collection proved convenient and saved time and efforts that are often needed to get to a place to be interviewed. The survey was designed in a way that it could be completed at individuals’ own pace and saved to resume later. Internet-based administration of the survey would also have improved its
accessibility via a variety of devices such as laptops, phones, tablets, or desktop computers. It also provided absolute anonymity to the participants that might have been compromised in face-to-face interviews. The anonymity may have encouraged more honest answers to specific personal and sensitive questions that inquired about their financial barriers related to their health and disability. The online survey also helped to reach nationwide participants who would have been missed due to distance and other logistics challenges. This feature improved the scalability of the research. Eventually, the use of an online survey for data collection along with rigorous recruitment strategies resulted in a sample size larger than proposed initially.

The next strength relates to the ethical conduct of the thesis. Barring the large population-based surveys, there are only a few studies that have used internet-based methods to collect data from people with disabilities, partially due to the ethical concerns that may arise while using internet-based methods. To overcome this challenge, I studied the ethical conduct of online research in detail and addressed the anticipated ethical concerns using appropriate strategies adhering to the Personal Health Information Protection Act 2004 of Ontario. These details were published in the Journal of Research Practice in a paper titled “Ethical Issues in Designing Internet-Based Research: Recommendations for Good Practice” (Gupta, 2017).

The final strength of this thesis lies in its focus on one of the most debated and contemporary public health policy issues of National Pharmacare in Canada. The research related to high medication costs in Canada began in the early 2000s, whereas the debate for the best national Pharmacare strategy for Canada started to emerge post-2012 (Lemire, 2019). However, the current Pharmacare debate often misses the voices of
people with disabilities. Literature that is available on CRNA involves people with chronic illnesses/conditions such as cardiovascular diseases, depression, rheumatoid arthritis, heart failure, hypertension, diabetes, and asthma or general senior or non-senior population (Gupta et al., 2018). It is the first study in Canada that examines extent, determinants and the consequences of CRNA within the context of people with disabilities. Furthermore, though the evidence stands clear that a publicly financed Pharmacare is what Canada needs, the question of its implementation at its best to ensure medicines are accessible to all without financial hardships remains unanswered. The study provides preliminary insights on the multidimensional nature of financial barriers faced by people with disabilities to access their medications.

Study 3 provides a comprehensive list of the most common medications prescribed or used by people who have spinal cord related conditions. The list can help inform the work of the Canadian Drug Agency to build or renew a national formulary for prescribed drugs, as proposed in the budget of 2019 (budget.gc.ca). These features make this study timely and relevant to the current landscape of healthcare in Canada.

This thesis also indicates the need to integrate health and social services to address the social determinants of health for vulnerable populations. The study sets an example of research in the integrative rehabilitation sciences that draws on the public health approach. Integrative rehabilitation sciences involve rehabilitation services and intervention research and encompass public health domains such as health policy and law, health economics, or health administration and management (Stucki, Reinhardt & Grimby, 2007). In comparison to basic (i.e. human functioning) rehabilitation sciences, the amount of interdisciplinary research in the field of rehabilitation and disability policy
has been relatively less (Stucki et al., 2007). This research cuts across the areas of rehabilitation science, public health, health policy, economics, and contributes to the diversification of mainstream rehabilitation science research.

6.3 Limitations of the doctoral thesis

This thesis has certain limitations that relate to the generalizability of its findings. The scoping review conducted in the first part excluded the gray literature available in the form of published books, public reports, press releases, policy papers, government webpages and others, which might have added further details on the current state of this topic.

As noted in study 2, the heterogeneity within peoples’ disabilities may lead to different predictions and experiences of financial barriers to access medications. Therefore, the extent to which conclusions from studies 3 and 4 can be generalized to 85,000 persons with SCI in Canada is worth considering. It is unlikely that this thesis represented the diversity and heterogeneity of issues faced by people with SCI or other disabilities living in Canada. For instance, the insurance arrangements for those who sustain SCI in a motor vehicle accident versus non-traumatic causes can be very different. Therefore, with the increasing prevalence of non-traumatic SCI, further studies exploring the differences in insurance mechanisms and their impact on the two groups would become increasingly relevant.

Further, despite the dissemination of surveys across Canada, we had a limited representation of the provinces other than Ontario. Though it is also worth noting that Ontario is the most populated province with almost 40% of the Canadian population
(Stats Canada, 2018) and the five smallest provinces constitute not more than 10% of the Canadian population.

The other limitations of the thesis relate to the data collection. Although using an online survey provided several benefits in terms of ease and reachability, it might have been a limitation for those who are either not able to afford the internet or do not use the internet. The recruitment process did not allow for the selection of participants through a randomized process resulting in uneven participation across Canada. As the study was based on self-reported data that cannot be validated by external data, participants may under- or overestimate the medication costs and their burden. There is also a subjective component in the assessment of the consequences of the medication cost burden, which is influenced by personal beliefs. Further, a 12-month retrospective period to determine the occurrence of CRNA might be unequally estimated by participants compared to a more recent situation.

As the survey and interviews adopted a cross-sectional approach, it was not possible to capture how medication use, cost or extent of drug coverage change among study participants over a period of time. Although Canada is a multi-lingual country, the surveys and interviews were only conducted in English. Conducting surveys and interviews in other languages might have allowed us to capture other diverse views. For the qualitative study, some of the interviews were conducted through the telephone. Telephone interviews might have led to the absence of visual cues from participants' accounts. Also, as interviews were primarily conducted by myself, the results might have been influenced by my own biases and experiences, though I adopted strategies to ensure trustworthiness.
6.4 Implications of the doctoral thesis

Based on the findings from the thesis, I offer eight critical implications for future research, policy and practice below:

The first important area for future research is to explore the complexity of drug insurance arrangements and their impact on medication underuse. In this research, many of our participants had the uncoordinated mix of public and private health insurance arrangements, despite which they reported non-adherence. These findings about the insurance need to be further explored. Future research is needed to determine what additional features of a drug insurance program make it protective against CRNA.

Despite the abundant literature on general access to healthcare issues, empirical research on specific issues faced by people with disabilities to access their medications stands limited, both in developed and developing countries. Thus, it will be important to explore such challenges in poor resource settings, where healthcare and social policies are even more scarce. Results can then be compared for people with disabilities across different countries.

Another important future research will be to explore the issue of medication cost barriers among other disabilities using the two theoretical models proposed in this thesis. These issues can be different for someone with a minor physical disability and a major cognitive disability. The type of disability influences individuals’ financial status too. Thus, it will be important to explore the impact of the type of disability on financial access to medications and whether the five-stage hierarchy model can be applied to other populations.
Social programs related to income and housing influence access to healthcare among low-income individuals (Williamson et al., 2006). As found in studies 3 and 4 of the thesis, extra costs for services add to the medication cost burden and increase the risk of forgoing medications due to cost. These findings emphasize the need to consider social services along with healthcare services. This will help address the social determinants of health for vulnerable populations.

The next important implication for future research would be to take a longitudinal approach to examine the intersectionality between age, sex, disability and financial access to medications. As noted previously, the health, income and employment outcomes for people with disabilities deteriorate with advancing age or severity of disability (Hitzig, Campbell, McGillivray, Boschen, & Craven, 2010). Also, the process of ageing for people with disabilities may begin earlier than usual, thereby removing them from the workforce (Turcotte, 2014). This implies that while the need for medicines increases, the financial capacity of people with disabilities may decline with time, putting them at a higher risk for rationing essential medications. Therefore, longitudinal studies that explore how medication cost pressures change over a period of time for individuals with disabilities would make important contributions to future policy.

Thesis findings suggest that prescribers may have an impact on patients' decisions on rationing their medications. Though these findings emerged through the patients’ reports only. Studies are there, though very few, that suggest that primary care providers often face challenges such as compressed times or administrative hassles to provide the care that their patients need, especially to the ones that have a disability or other complex healthcare issues (McCull et al., 2008). More recent studies suggest that clinicians and
persons with SCI are not having important conversations about overall medication management (Cadel et al., 2020). Thus, exploring doctors’ perspectives on addressing the specific issue of medication cost burden among patients with disabilities can be a valuable research study. Understanding prescribers’ perspectives might illuminate important considerations for health services and systems strengthening.

Although this study has set the stage for research on financial barriers to medication access within individuals with SCI, a population-level analysis is needed to improve the generalizability and external validity of the findings. Researchers have found preliminary algorithms to identify people with traumatic and non-traumatic SCI in health administrative databases (Ho et al., 2017; Welk, Loh, Shariff, Liu, & Siddiqi, 2014). Studies are published that identify the prescription drug utilization claims made by patients with SCI above 66 years of age (Guilcher et al., 2018). Therefore, future studies may find prescription drug utilization for individuals with SCI who are under 65 years and receive social assistance or have catastrophic drug costs as their drugs are funded through the public drug program. The utilization data can then be combined with hospitalization and rehabilitation records to find the association of medication use with health and rehabilitation care utilization.

One limitation of the abovementioned use of health administrative databases is that it is not possible to find CRNA to prescription drugs through these databases. As noted in study 1, claims data do not necessarily represent the actual consumption of prescribed medications and cannot account for medications that were not purchased due to cost. Additionally, claims data from provincial drug benefit programs can only provide information for drugs that are on the public formularies and cannot account for drugs that
are not listed. Canadian Community Health Survey (CCHS) is the only nationally representative data source on this topic, which also contains a Health Utilities Index Mark 3 (HUI3) and a module on functioning in its 2018 version that can be used to identify people with disabilities (MacKenzie, 2013). Therefore, future research using population-level data can help researchers to move from generating evidence to generating evidence-informed policies that enhance provincial and national services for people with complex health care needs.

6.5 Knowledge translation

Given the increasing attention to improve the evidence use and uptake for impacting policy and practice, there has been a significant advancement in research on the science of knowledge translation (KT) (Gagliardi, Berta, Kothari, Boyko, & Urquhart, 2015; Grimshaw, Eccles, Lavis, Hill, & Squires, 2012). To ensure the findings of this study make an impact on future research, policy or practice, I employed various KT strategies. The important stakeholders identified for this purpose included policymakers, health care practitioners, patients/consumers, and researchers. I adopted knowledge translation channels that are best suited for the abovementioned stakeholders:

1. *Advocacy letter*: Recently, the Canadian government announced the formation of a National Advisory Council on the implementation of an affordable Pharmacare in their budget of 2018 (Grignon, Longo, Marchildon, & Officer, 2019). In January 2019, we wrote a letter to the council, sharing the key findings of our research and offered certain recommendations based on our findings. The letter is provided as appendix K of this thesis.
2. **Radio shows:** To reach the broader community within academia and outside, I participated in 3 radio interviews. Two interviews were organized by the Kelly and Company, which is a Toronto-based radio show with accessible broadcasting across Canada (www.ami.ca/category/kelly-and-company). The first interview focused on the general issue of financial access to medicines for people with disabilities and the relevance of this thesis in the Canadian context (dated March 26, 2018). As a follow up, another interview was organized by the same company in February 08, 2019 to discuss findings of the study and its implications within Canada and beyond. One more interview was held by the local radio station of Queen's University called CFRC 101.9 FM under their Grad Chat program dated January 28, 2019. This interview helped me to share the relevance and findings of this research with the academic community at Queen’s and other Canadian universities.

3. **Video Abstract:** Along with the written abstracts, many authors nowadays add a video abstract with their published articles to tell the research story in an accessible, succinct, and creative way (Czaran, Wolski, & Richardson, 2017; Zong, Xie, Tuo, Huang, & Yang, 2019). Because of their ability to overcome the constraints of written articles, video abstracts are becoming popular and are recommended by many mainstream publishing houses (Reupert, 2017; Spicer, 2014). To improve the visibility and impact of my work, I prepared a video abstract that was published on the website of Dove Medical press in May 2018 along with the first manuscript of this thesis (https://youtu.be/WyC7NEmp7wQ).

4. **Conference presentation:** To reach the scientific community and consumers, I presented the research findings at the 8th National Spinal Cord Injury conference
organized by the Canadian Spinal Cord Injury Rehabilitation Association in October 2019. This conference invited more than 300 delegates across North America, including primary care doctors, specialists, patients, consumer organizations, graduate students, researchers, and professional organizations working with individuals with SCI. The presentation won third place in the people's choice award.

5. *Newsletter communiqué*: The research proposal for the thesis was featured in a monthly newsletter of the Canadian Disability Policy Alliance, to invite citizen engagement and promote integrated knowledge translation. The Canadian Disability Policy Alliance is a national group of disability researchers, healthcare administrators, policy advocates, and policymakers that reaches up to 100 disability-related organizations across the nation. The newsletter featuring this study can be accessed through their website as well as this direct web link:


6. *Peer-reviewed publications*: Peer-reviewed publications have been the most common way to disseminate research findings within the scholarly community, including subject experts and reviewers. This thesis yielded a total of four manuscripts, out of which all are published in subject-specific reputed journals. Details on these journals are provided at the beginning of the respective chapters and in the appendices L-O.

7. *Twitter feeds*: Studies have emerged that suggest that the use of social media, especially Twitter, to promote one's research may improve its dissemination (Mollett, Moran, Dunleavy, 2011; Greer & Ferguson, 2011). Twitter provides an opportunity to tag a variety of stakeholders (such as patient groups, advocacy organizations,
researchers, policymakers and other like-minded groups) across the globe who may not be accessible quickly otherwise, and provides a metrics on the number of retweets, impressions, and engagements with the tweets. Therefore, I have been leveraging my Twitter account (@shikhaguptaot) to promote this research and reach out external audience since 2017. As per the metrics provided, the thesis related tweets have reached more than 5000+ people on Twitter.

8. **Infographic**: An infographic depicting a pictorial representation of the research findings was disseminated through Twitter. It will be further adapted to be used for dissemination to organizations that have helped me recruit my research participants, and to others who would benefit or have interest in this research. The infographic is attached as the appendix P.

9. **Article in Healthy Debate**: An article featuring this research has been submitted to the Healthy Debate. Healthy Debate is an online platform that publishes opinions and blogs about the healthcare system in Canada. It has been considered a powerful platform to create a constructive dialogue on a policy issue related to healthcare among the scholarly community.

10. **Future activities**: Other than these channels, I also plan to present my research findings at one of the annual meetings of the Spinal Cord Injury Ontario and to their local chapter meetings in Kingston. This exercise may help me to connect with people with SCI and gain their insights and feedback directly.

With these efforts, I hope that this research will contribute towards the global understanding of the extent, determinants and consequences of medication cost barriers.
and non-adherence among people with disabilities. This study may add to the evidence that may lead to improvements in policy, research and practice to ensure universal, timely, and burden-free access to prescription medications for all.

6.6 References


spinal cord injury/dysfunction and providers. Research in Social and Administrative Pharmacy. doi: 10.1016/j.sapharm.2020.01.014


Appendix A

Queen’s HSREB ethics approval

QUEEN'S UNIVERSITY HEALTH SCIENCES & AFFILIATED TEACHING HOSPITALS RESEARCH ETHICS BOARD (HSREB)

HSREB Initial Ethics Clearance

April 09, 2018

Ms. Shikha Gupta
Centre for Health Services and Policy Research
Queen’s University

ROMEO/TRAQ: #6023360
Department Code: EPID-616-18
Study Title: "Exploring the extent, determinants, and consequences of cost-related non-adherence to medications among people with spinal cord injuries". Lay Title: Paying for the pills: coverage or compromise?
Co-Investigators: Dr. M.A. McColl
Review Type: Delegated
Date Ethics Clearance Issued: April 09, 2018
Ethics Clearance Expiry Date: April 09, 2019

Dear Ms. Gupta,

The Queen's University Health Sciences & Affiliated Teaching Hospitals Research Ethics Board (HSREB) has reviewed the application and granted ethics clearance for the documents listed below. Ethics clearance is granted until the expiration date noted above.

- Thesis Proposal
- Recruitment Poster
- Recruitment Postcard
- Recruitment Message – Social Media Platform
- Interview Guide
- Survey
- Letter of Information/Consent Form – Interviews
- Letter of Information/Consent Form – Telephone Survey
- Letter of Information – Long Version

Documents Acknowledged:

- CORE Certificate – S. Gupta
- Department Letter of Approval – March 13, 2018

Amendments: No deviations from, or changes to the protocol should be initiated without prior written clearance of an appropriate amendment from the HSREB, except when necessary to eliminate immediate hazard(s) to study participants or when the change(s) involves only administrative or logistical aspects of the trial.

Renewals: Prior to the expiration of your ethics clearance you will be reminded to submit your renewal report through ROMEO. Any lapses in ethical clearance will be documented on the renewal form.
QUEEN’S UNIVERSITY HEALTH SCIENCES & AFFILIATED TEACHING HOSPITALS RESEARCH ETHICS BOARD (HSREB)

HSREB Renewal of Ethics Clearance

April 05, 2019

Ms. Shikha Gupta
Centre for Health Services and Policy Research
Queen’s University

TRAQ #: 6023360
Department Code: EPID-616-18
Study Title: “Exploring the extent, determinants, and consequences of cost-related non-adherence to medications among people with spinal cord injuries or related conditions”. Lay Title: Paying for the pills: coverage or compromise?
Review Type: Delegated
Date Ethics Clearance Effective: April 05, 2019
Ethics Clearance Expiry Date: April 05, 2020

Dear Ms. Gupta:

The Queen’s University Health Sciences & Affiliated Teaching Hospitals Research Ethics Board (HSREB) has reviewed the application. This study, including all currently approved documentation has been granted ethical clearance until the expiry date noted above.

Prior to the expiration of your ethics clearance, you will be reminded to submit your renewal report through TRAQ. Any lapses in ethical clearance will be documented below.

Lapses in Ethics Clearance: N/A

Yours sincerely,

[Signature]

Albert F. Clark, PhD
Chair, Queen’s University Health Sciences and Affiliated Teaching Hospitals Ethics Board

The HSREB operates in compliance with, and is constituted in accordance with, the requirements of the Tri-Council Policy Statement: Ethical Conduct for Research Involving Humans (TCPS 2); the International Conference on Harmonisation Good Clinical Practice Consolidated Guideline (ICH GCP); Part C, Division 5 of the Food and Drug Regulations; Part 4 of the Natural Health Product Regulations; Part 3 of the Medical Devices Regulations, and the provisions of the Ontario Personal Health Information Protection Act (PHIPA 2004) and its applicable regulations. The HSREB is qualified through the CTO REB Qualification Program and is registered with the U.S. Department of Health and Human Services (DHHS) Office for Human Research Protection (OHRP). Federalwide Assurance Number: FWA#: 00004184, IRB#: 00001173

HSREB members involved in the research project do not participate in the review, discussion or decision.
Appendix B

Survey questionnaire

Screening for eligibility

Thank you for your interest in our study. In order to confirm your eligibility, please provide following information. Please note that your data will be used for research purposes only. Data will be kept confidential. No third party will have access to data.

a. Do you have a spinal cord injury?

NOTE: A spinal cord injury or SCI, occurs when trauma (such as a fall, vehicle accident, act of violence) or disease (such as a motor neuron disease, tumor, infection, myelopathy, vascular, toxic, metabolic condition or a developmental disorder) damage the spinal cord, resulting in partial or complete paralysis.

Yes
No
May be

b. Are you 18 years or older?

Yes
No

c. Do you live in Canada?

Yes
No

d. Are you currently living in a hospital or a government funded long-term care home (e.g. Nursing home)?

Yes
No

e. In the last 12 months, have you been prescribed any medications to manage your health? Please think about both prescription and non-prescription medications.

NOTE: Prescription medications include medications that are prescribed by a regulated healthcare professional such as a physician, nurse, specialist, naturopath or a pharmacist. Non-prescription medications include alternative medicine that do not require a doctor’s prescription and you can buy them over the counter such as vitamin supplements or other herbal and natural products.

Yes
No

If “Do you have a spinal cord injury? = No” OR “Are you older than 18 years of age? = No” OR “Are you currently living in a hospital or a government funded long-term care institution = Yes” OR “Do you live in Ontario? = No” OR “In the last 12 months, have you been prescribed any medications to manage your health?”= No
Whoops! Sorry, we found that you are not eligible to participate in this study. If you would like to know about future research, you may provide following information. It will redirect participants to end of survey.

Otherwise, proceed:

Obtaining Consent

Thanks for your information. We have found that you are eligible to participate in this study!

Before you proceed, please read following information carefully.

INFORMATION: The purpose of our study is to identify whether people with spinal cord injuries face any financial barriers in accessing their necessary medications which leads to non-adherence or compromise on other basic needs. Approximately 120 participants will be recruited for this study over the course of 1 year. The survey will require approximately 10-15 minutes of your time for completion. Questions will be asked about the number and type of medications you use, the source of funding for your prescription medications and financial burden that you may face because of medication related costs. Most of these questions will have multiple choice answers and you will need to select the best answer that describes your situation. Your information will be kept completely confidential. No third party will be provided your data. You may decline answering any questions you do not wish to answer at any time without prejudice or penalty. For more information or for any queries and concerns, you can contact Shikha Gupta at shikha.gupta@queensu.ca or click here.

Do you provide your consent to participate in the study?
Yes
No

Skip To: End of Survey If “Do you provide your consent to participate in the study? = No”

Otherwise, proceed: Great! The next few questions will ask you about your medication use and its costs, and any barriers that may face because of the cost of medications.

NOTE: Please assemble all your medications and the calendar of your medical appointments before you begin the survey.

A. Your current medications

1. In the last 12 months, how many different medications have you received a prescription for, including any on-going prescriptions? (Please count number of medications that are prescribed by a healthcare professional and over-the-counter medications). ____

2. Please provide the following information with regard to your prescription medications (Medications that are prescribed by a healthcare professional such as a physician, nurse, specialist, naturopath or a pharmacist).

<table>
<thead>
<tr>
<th>Name of the medication</th>
<th>Health condition for which it is used? (Such as for bladder,</th>
<th>Do you have drug insurance to help cover the cost of this medication?</th>
<th>How much do ‘you’ have to pay monthly out-of-pocket for this medication?</th>
<th>In the past 12 months, have you ever taken this medication less due to cost?</th>
</tr>
</thead>
</table>
B. Your insurance coverage

4. What type of insurance do you have to help cover the cost of your medications?
   • Publicly funded- e.g. Ontario drug benefit program
   • Private insurance
   • Employer based insurance
   • Other type of insurance (such as family insurance)
   • No insurance

Display question 5 If “What type of insurance do you have to help cover the cost of your medications?” = Publicly funded

5. If you are covered by any of the two publicly funded programs, have you ever experienced any of these challenges? (Select all that apply)
   • Lengthy process for drugs to be approved
   • Required drug not on public formulary list
   • Complex paperwork to confirm eligibility
   • None
   • Not applicable

6. Do you share the cost of your medications in the form of co-pay, deductibles, insurance premiums, maximums or pharmacist dispensing fees?
   • Yes
   • No
   • I do not have insurance to cover the cost of medications

Display question 7 If “Do you share the cost of your medications in the form of co-pay, deductibles, insurance premiums… = Yes”

7. If yes, how much do you have to pay in the form of co-pay, deductibles, insurance premiums, maximums or pharmacist dispensing fees?
   • None
   • Less than $5 a month
   • Between $5 to $20 a month
   • Between $21 to $50 a month
   • Between $51 to $100 a month
   • More than $100 a month
C. Your health service use and strategies to manage costs

9. Do you have a regular healthcare provider that you regularly see or talk to when you need care or advice for your health?
   • Yes
   • No

Display question 10 If “Do you have a regular healthcare provider that you regularly see or talk to when you need care or... = Yes”

10. Is that regular healthcare provider a....?
   • Family Doctor or General Practitioner or Physician
   • Medical specialist such as physiatrist or a psychiatrist
   • Nurse practitioner
   • Therapist
   • Other (Please specify) ____________

11. How many outpatient/ambulatory care visits did you have in past 12 months? Please count total number of visits to all healthcare professionals (such as a general practitioner, specialist, surgeon, allergist, gynecologist, or psychiatrist). __

12. Do you discuss cost of your prescriptions with your prescribing healthcare professional or your pharmacist?
   • Yes
   • No
   • I do not face any cost related barriers to pay for my medications

13. Does your prescribing healthcare professional ask you if you can afford the medications they are prescribing to you?
   • Yes, always
   • Yes, sometimes
   • Never

14. Does your prescribing healthcare professional help you to manage the cost of your medications (such as prescribing other medications that are low-cost alternatives or generic substitutes, or referring you for drug benefits program?)
   • Yes
   • No
   • Sometimes

15. During the past 12 months, have you ever done any of these things to save money:

<table>
<thead>
<tr>
<th>Activity</th>
<th>Often</th>
<th>Sometimes</th>
<th>Rarely</th>
<th>Never</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Taken fewer pills or a smaller dose</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>2. Not filled a prescription at all</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>3. Put off or postponed getting a prescription filled</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>4. Used cheaper medication or substitutes such as herbal medication or vitamins</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>5. Taken your medication less frequently than recommended to “stretch out” the time before getting a refill</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
6. Worried about paying for your prescription medications
7. Borrowed money from someone to pay for medications
8. Spent less on food, heat, or other basic needs
9. Spent less on your other healthcare needs
10. Spent less on your family member's healthcare needs

16. In the past 12 months, were there any medications that were not prescribed to you because you couldn't afford it and you did not have insurance that could cover the cost?
   - Yes
   - No
   - Don’t Know

Display Question 17 If “In the past 12 months, were there any medications that were not prescribed to you because you... = Yes”

17. What health conditions were these prescriptions meant to treat? _______

Display Question 18 If “In the past 12 months, were there any medications that were not prescribed to you because you... = Yes”

18. Approximately, how much was this prescription going to cost you per month? Indicate amount in CAD. _____

19. During the past 12 months, have you ever taken one or more of your medications less than prescribed because of the following reasons, other than the cost?
   (a) Adverse reactions associated with drugs
   (b) Medications not necessary or less essential
   (c) Medication was not effective
   (d) No

Display question 20 If:
“During the past 12 months, have you ever engaged in the following due to medication-related costs: = Always” OR “During the past 12 months, have you engaged in the following due to medication-related costs: = Sometimes”

20. Do you think that being unable to afford your medications made your health worse?
   - Yes
   - No
   - May be

Display question 21 If “During the past 12 months, have you engaged in the following due to medication-related costs: = Always” OR “During the past 12 months, have you engaged in the following due to medication-related costs: = Sometimes”

21. Did being unable to afford your medications result in having to do any of the following:
   - Go to the emergency department
   - Be admitted to hospital
   - Go to the doctor which you would not have had to do otherwise
   - No
22. How many emergency department visits did you have in past 12 months? ____

23. How many over-night hospitalizations did you have in past 12 months? Please count number of nights that you have stayed in the hospital in last 12 months. ____

Thanks for the information you provided so far! The following section will ask you about your demographics and spinal cord injury related information.

D. Some general information about you…

24. What year were you born in? Please indicate the year. 19__

25. Your sex? Female _ Male _ I prefer not to answer _ I like to describe myself as____

26. What is your current marital status?
   • Married
   • Single/ Never married
   • Widowed
   • Separated/divorced
   • In a relationship

27. How do you define your ethnic or racial background? ______

28. What is the highest certificate, degree or diploma that you have completed?
   • Less than high school
   • High School diploma or certificate
   • College degree, diploma or certificate
   • University degree, diploma or certificate below the bachelor's level
   • Bachelor's degree or equivalent
   • Degree above Bachelor's level e.g. Master's or Ph.D degree

29. What is your current employment status?
   • Employed full time
   • Employed part time
   • Unemployed
   • Retired
   • Contractual work
   • On Disability
   • On unpaid disability
   • Student
   • Regular volunteer

30. What is your average net personal monthly income after tax and deductions? (Please provide your best estimate and indicate amount in CAD).________

31. What is your average net household monthly income after tax and deductions? (Please provide your best estimate and indicate amount in CAD). _______
32. Do you access any non-employment related sources of income such as pensions or disability benefits?
   • Yes
   • No

E. Information about your spinal cord injury

33. When did you acquire your spinal cord injury? Please mention the year. 19__

34. How did you acquire your injury?
   • Traumatic such as accident or fall
   • Non-traumatic such as tumor, cancer, or infection
   • Other (Please specify)

35. What is the level of your injury?
   • Cervical
   • Upper Thoracic (above T6)
   • Lower thoracic (below T6)
   • Lumbar
   • Sacral

36. What is the extent of your injury?
   • Complete
   • Incomplete

37. What amount of assistance do you require to perform your basic activities of daily living (such as dressing, bed mobility, transfers, eating etc.)?
   • Needs no assistance
   • Partial assistance
   • Total assistance

38. Except for medications, how much do you spend monthly on your health related needs? (Please indicate amount in CAD)
   • Special Diet ______
   • Bowel care supplies ______
   • Bladder care supplies______
   • Skin and wound care supplies ______
   • Pain treatments ______
   • Others (Please specify) __

Thanks, so much for providing the information!

Would you like to receive $5 Tim Hortons Gift Card as our thanks for completing the survey?
   • Yes
   • No

Are you willing to be contacted by the research team for an interview? You will be compensated for your time.
   • Yes
• No

If a participant response yes to these two questions, they will be redirected to a new survey which will collect their contact information, but this information would not be linked to their responses on previous survey.

Note that the information you provide below will not be linked to your responses on the previous survey.

First name (optional) ____
Last name (optional) _____
Contact number (optional) ______
Email Address (required) ______
City (optional) ______
Mailing address (optional) ______

End of Survey: Thank you for taking time to complete this survey. If you have any questions or concerns regarding this study, please feel free to contact Shikha Gupta at shikha.gupta@queensu.ca. You can also let us know if you would like to receive a summary of results at the end of this research project. To ensure that your session has been terminated, please clear your cookies and cache, then close your browser.
Appendix C

Formula for sample size calculation

Sample size calculation for this study

\[ N = \left( \frac{Z_{1-\alpha/2}}{d} \right)^2 \frac{p (1-p)}{d^2} \]

\[ N = \left( 1.96 \right)^2 \times 0.1 \times \left( 1 - 0.1 \right) \]
\[ \div \left( 0.05 \right)^2 \]

\[ = 138 \]

\[ Z_{1-\alpha/2} = \text{Standard error (type 1)} = 5\% \]
\[ p = \text{expected proportion of individuals who will report outcome of interest} \]
\[ d = \text{absolute error or precision} \]

(Charan & Biswas, 2013)
Appendix D

Participant Information Letter (Online Survey)

Extent, determinants, and consequences of cost-related non-adherence to medications among people with spinal cord injuries

You are invited to participate in a research study conducted by:

Principal Investigators:

Professor Mary Ann McColl
School of Rehabilitation Therapy
Queen’s University
mccollm@queensu.ca

Shikha Gupta, PhD Candidate
School of Rehabilitation Therapy
Queen’s University
shikha.gupta@queensu.ca

What is the purpose of this study?

The purpose of this study is to identify if people with spinal cord injury or related conditions face any financial barriers in accessing necessary medications, and if so, what are the consequences for them? Approximately 140 participants will be recruited for this study over the course of 1 year.

Please Note: We define spinal cord injury as “an injury that occurs when trauma (such as a fall, vehicle accident, act of violence) or disease (such as a motor neuron disease, tumor, infection, myelopathy, vascular, toxic, metabolic condition or a developmental disorder) damage the spinal cord, resulting in partial or complete paralysis.”

What will my responsibilities be if I participate in this study?

If you agree to participate, you can complete an online survey. You can also opt to answer the survey questions through phone. The survey contains 40 questions and will approximately require 15 minutes of your time for completion. Most of these questions will have multiple choice answers and you will need to select the best answer(s) that describe your situation. The questions will be about your demographic characteristics, spinal cord injury related characteristics, health service utilization, the number and type of medications you use, the source of funding for your prescription medications and any type of financial burden that you may face because of medications related costs.
What is the basis for selection and participation in this study?

The individuals who will be selected to participate in the study are of 18 years of age and above; living in the community and sustaining a spinal cord injury or related condition for more than 12 months and having one or more prescription medications at the time of the survey.

What are the possible benefits for me or society?

Prescription medications play an important role in the treatment and prevention of disease, especially for people with chronic conditions such as a spinal cord injury. However, in Canada, the coverage for prescription medications is not universal and varies across the provinces, depending on the individual health and socio-economic status. Evidence is emerging that people with poor health status face financial barriers to access necessary medications that may lead to poor health outcomes. Therefore, this study will try to find out whether people with spinal cord injury in Canada have access to necessary medications to ensure optimum health outcomes. The study will be beneficial to the people who face financial barriers to access their necessary prescription medications by bringing this issue to the knowledge of wider community including general public, researchers, policy makers, medical professionals and other stakeholders and contribute to driving change at the policy level.

What are the possible risks or discomforts?

There are no risks associated with your participation. Your participation in this study will not expose you to any risk or discomforts. Your information will be kept completely confidential and pseudonymous. No third party will be provided your data. You may decline answering any questions you do not wish to answer at any time without prejudice or penalty.

Will there be any payment or reimbursement if I participate in this study?

Your participation in this study will not provide you with any financial benefits but you will receive a $5 gift card after completing your survey as a token of appreciation for your time.

What information will be kept private?

Your data will not be shared with anyone, except with your consent. The information obtained by me will be kept in a locked cabinet. Your personal information will be kept separate from the survey data at a secure place. If the results are published, no names or identifying information will be released or published without your specific consent to the disclosure.

What if I change my mind about participating in the study?
Your participation in this study is absolutely voluntary. If you volunteer to be in this study, you may withdraw at any time, even after signing the consent form. In cases of withdrawal, your data will be destroyed if it is not aggregated and analysed. However, after data has been analysed and submitted for publication, it can’t be withdrawn.

**Will I find out about the study results?**

All participants will be given the opportunity to contact the student investigator (Shikha Gupta) at the end of the study to receive a summary of the study results.

**Can I get more information about participating as a study subject?**

This study has been reviewed for ethical compliance by the Queen's University Health Sciences and Affiliated Teaching Hospitals Research Ethics Board. If you have any concerns about your rights as a research participant please contact - Dr. Albert Clark, Chair - (toll-free number) - 1-844-535-2988 or clarkaf@queensu.ca. If you have questions or require more information about the study itself, please contact Shikha Gupta (shikha.gupta@queensu.ca; phone number: 6138761780). The information mentioned above will be discussed and all questions will be clarified prior to any involvement in the study.
Appendix E

Message for Social Media Recruitment

How do you pay for your medications? Through coverage or compromise? Please tell us!

Researchers at Queen’s University are looking for volunteers to complete a survey about medication costs and barriers that you might have faced to pay for your medications. The survey will take 15 minutes. You can complete it online or by telephone. In appreciation of your time, you will receive a $5 Tim Hortons Gift Card.

To start the survey, please click on this link:

Take the survey

For telephone survey, leave a text or voice message at:

613-876-1780

To know more, contact here:

shikha.gupta@queensu.ca
Appendix F

Recruitment Postcard

How are you paying for your pills? Through coverage or compromise? Please tell us!

WHO
We are looking for volunteers with a Spinal Cord Injury living in Ontario.

WHAT
We will ask you to complete a survey about your medication costs and any financial barriers that you might have faced.

HOW
The survey will take 15 minutes. You can complete it online or by telephone. You will receive a $5 Gift Card for your time.

To participate, contact Shikha Gupta @ shikha.gupta@queensu.ca
613-876-1780
Appendix G

Recruitment Poster

How are you paying for your medications? Through coverage or compromise?

Please tell us!

WHO?
We are looking for volunteers with Spinal Cord related conditions living in Ontario.

WHAT?
We will ask you to complete a survey about your medication costs and any cost-related barriers that you might have faced to pay for your medications.

HOW?
The survey will take 15-20 minutes. You can complete it online or by telephone.

In appreciation of your time, you will receive a $5 Tim Hortons Gift Card

If you are interested, contact at:
shikha.gupta@queensu.ca | 613-876-1780
Appendix H

Letter of information and consent form (qualitative interviews)

You are invited to participate in a research study!

Study Title: Exploring the extent, determinants, and consequences of cost-related non-adherence to medications among people with spinal cord injuries

Name of Researcher: Shikha Gupta, School of Rehabilitation Therapy, Queen’s University

Name of the supervisor: Mary Ann McColl, School of Rehabilitation Therapy, Queen’s University

I am Shikha Gupta, a PhD student in the School of Rehabilitation Therapy (SRT), working under the supervision of Dr. Mary Ann McColl. I am asking individuals with spinal cord injuries to take part in a research study exploring the extent, determinants, and consequences of cost-related non-adherence to medications among people with spinal cord injuries.

You indicated in your survey that you sometimes have trouble paying for your medications, and that you even sometimes don’t take your medications as prescribed because of the cost. In this interview, I’d like to ask you more about that. If you agree to take part, I will interview you for one hour at a public location of your choosing or through telephone. The interview will be audio-recorded and later transcribed. There are no known risks for taking part in this study. The study will be beneficial to the people who face financial barriers to access their necessary prescription medications by bringing this issue to the knowledge of wider community and contribute to driving change at the policy level.

There is no obligation for you to say yes to take part in this study. You don’t have to answer any questions you don’t want to. You can stop participating at any time without penalty. You may withdraw from the study up until December 31, 2018 by contacting me at shikha.gupta@queensu.ca.

I will keep your data securely for at least five years. Your confidentiality will be protected to the extent possible by replacing your name with a pseudonym for all data and in all publications. The code list linking real names with pseudonyms will be stored separately and securely from the data. Other than me, only a transcriber who has signed a Confidentiality Agreement will have access to any of the data.

I hope to publish the results of this study in my PhD thesis and academic journals and present them at conferences. I will include quotes from some of the interviews when presenting my findings. However, I will never include any real names with quotes, and I will do my best to make sure quotes do not include information that could indirectly
identify participants. During the interview, please let me know if you say anything you do not want me to quote.

For taking part in this study, you will receive a $20 in appreciation of your time.

If you have any ethics concerns please contact the Queen’s University Health Sciences and Affiliated Teaching Hospitals Research Ethics Board (HSREB) at 1-844-535-2988 (Toll free in North America) or email the HSREB Chair at clarkaf@queensu.ca.

If you have any questions about the research, please contact me, Shikha Gupta, at shikha.gupta@queensu.ca or my supervisor, Dr. Mary Ann McColl, at mccollm@queensu.ca or 613-533-6319.

This Letter of Information provides you with the details to help you make an informed choice. All your questions should be answered to your satisfaction before you decide whether or not to participate in this research study.

Keep one copy of the Letter of Information for your records and return one copy to the researcher, Shikha Gupta.

By signing below, I am verifying that: I have read the Letter of Information and all of my questions have been answered.

Name of Participant: ________________________________
Signature: _________________________________________
Date: ______________________________________________
## Appendix I

### COREQ (COnsolidated criteria for REporting Qualitative research) Checklist

A checklist of items that should be included in reports of qualitative research. You must report the page number in your manuscript where you consider each of the items listed in this checklist. If you have not included this information, either revise your manuscript accordingly before submitting or note N/A.

<table>
<thead>
<tr>
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<tr>
<td><strong>Domain 1: Research team and reflexivity</strong></td>
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<tr>
<td>Personal characteristics</td>
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<tr>
<td>Interviewer/facilitator</td>
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<td>Which author/s conducted the interview or focus group?</td>
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<td>Credentials</td>
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<td>What were the researcher’s credentials? E.g. PhD, MD</td>
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<td>Occupation</td>
<td>3</td>
<td>What was their occupation at the time of the study?</td>
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<tr>
<td>Gender</td>
<td>4</td>
<td>Was the researcher male or female?</td>
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<tr>
<td>Experience and training</td>
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<td>What experience or training did the researcher have?</td>
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<td><strong>Relationship with participants</strong></td>
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<tr>
<td>Relationship established</td>
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<td>Was a relationship established prior to study commencement?</td>
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</tr>
<tr>
<td>Participant knowledge of the interviewer</td>
<td>7</td>
<td>What did the participants know about the researcher? E.g. personal goals, reasons for doing the research</td>
<td>136-137</td>
</tr>
<tr>
<td>Interviewer characteristics</td>
<td>8</td>
<td>What characteristics were reported about the interviewer/facilitator? E.g. Bias, assumptions, reasons and interests in the research topic</td>
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<tr>
<td><strong>Domain 2: Study design</strong></td>
<td></td>
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<tr>
<td>Theoretical framework</td>
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<tr>
<td>Methodological orientation and Theory</td>
<td>9</td>
<td>What methodological orientation was stated to underpin the study? E.g. grounded theory, discourse analysis, ethnography, phenomenology, content analysis</td>
<td>134</td>
</tr>
<tr>
<td><strong>Participant selection</strong></td>
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<tr>
<td>Sampling</td>
<td>10</td>
<td>How were participants selected? E.g. purposive, convenience, consecutive, snowball</td>
<td>136-137</td>
</tr>
<tr>
<td>Method of approach</td>
<td>11</td>
<td>How were participants approached? E.g. face-to-face, telephone, mail, email</td>
<td>137</td>
</tr>
<tr>
<td>Sample size</td>
<td>12</td>
<td>How many participants were in the study?</td>
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</tr>
<tr>
<td>Non-participation</td>
<td>13</td>
<td>How many people refused to participate or dropped out? Reasons?</td>
<td>137</td>
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<tr>
<td><strong>Setting</strong></td>
<td></td>
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<tr>
<td>Setting of data collection</td>
<td>14</td>
<td>Where was the data collected? E.g. home, clinic, workplace</td>
<td>137</td>
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<tr>
<td>Presence of non-participants</td>
<td>15</td>
<td>Was anyone else present besides the participants and researchers?</td>
<td>NA</td>
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<tr>
<td>Description of sample</td>
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<td>What are the important characteristics of the sample? E.g. demographic data, date</td>
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</tr>
<tr>
<td><strong>Data collection</strong></td>
<td></td>
<td></td>
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</tr>
<tr>
<td>Interview guide</td>
<td>17</td>
<td>Were questions, prompts, guides provided by the authors? Was it pilot tested?</td>
<td>137-138</td>
</tr>
<tr>
<td>Repeat interviews</td>
<td>18</td>
<td>Were repeat inter views carried out? if yes, how many?</td>
<td>No</td>
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<tr>
<td>Audio/visual recording</td>
<td>19</td>
<td>Did the research use audio or visual recording to collect the data?</td>
<td>139</td>
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<tr>
<td>Field notes</td>
<td>20</td>
<td>Were field notes made during and/or after the interview or focus group?</td>
<td>139</td>
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<tr>
<td>Duration</td>
<td>21</td>
<td>What was the duration of the interviews or focus group?</td>
<td>137</td>
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<tr>
<td>Data saturation</td>
<td>22</td>
<td>Was data saturation discussed?</td>
<td>138-139</td>
</tr>
<tr>
<td>Transcripts returned</td>
<td>23</td>
<td>Were transcripts returned to participants for comment and/or</td>
<td>139</td>
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<tr>
<td>Domain 3: analysis and findings</td>
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<td>Data analysis</td>
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<tr>
<td>Number of data coders</td>
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<td>How many data coders coded the data?</td>
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<tr>
<td>Description of the coding tree</td>
<td>25</td>
<td>Did authors provide a description of the coding tree?</td>
<td>141</td>
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<tr>
<td>Derivation of themes</td>
<td>26</td>
<td>Were themes identified in advance or derived from the data?</td>
<td>138</td>
</tr>
<tr>
<td>Software</td>
<td>27</td>
<td>What software, if applicable, was used to manage the data?</td>
<td>139</td>
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<tr>
<td>Participant checking</td>
<td>28</td>
<td>Did participants provide feedback on the findings?</td>
<td>139</td>
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<tr>
<td>Reporting</td>
<td></td>
<td></td>
<td></td>
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<tr>
<td>Quotations presented</td>
<td>29</td>
<td>Were participant quotations presented to illustrate the themes/findings?</td>
<td>144-159</td>
</tr>
<tr>
<td>Data and findings consistent</td>
<td>30</td>
<td>Was there consistency between the data presented and the findings?</td>
<td>Yes</td>
</tr>
<tr>
<td>Clarity of major themes</td>
<td>31</td>
<td>Were major themes clearly presented in the findings?</td>
<td>Yes</td>
</tr>
<tr>
<td>Clarity of minor themes</td>
<td>32</td>
<td>Is there a description of diverse cases or discussion of minor themes?</td>
<td>Yes</td>
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</tbody>
</table>


Once you have completed this checklist, please save a copy and upload it as part of your submission. DO NOT include this checklist as part of the main manuscript document. It must be uploaded as a separate file.
Appendix J

Mapping of survey questionnaire on the adapted model

Demographic factors
- Age
- Sex
- Marital status
- Education
- Place of living
- Ethnicity

Socio-economic factors
- Employment status
- Additional healthcare costs
- Availability of social assistance
- Access to non-employment resources

Disability-related factors
- Nature of disability
- Type of disability
- Severity of disability
- Additional co-morbidities
- Need for alternative therapies and equipment

Medication-related factors
- Type of medications
- Out-of-pocket cost of medications
- Frequency of taking medications
- Availability of drug insurance
- Regimen complexity

System-related factors
- Access to public drug insurance
- Cost-sharing mechanisms
- Public drug formulary
- Availability and access to other social or healthcare services.
- Resources and funding for alternative therapies or equipment

Prescriber-related factors
- Access to a physician or prescriber
- Communication and relationship with prescribers
- Coordination of care
- Knowledge about disability-specific needs and services

Questions 1-2, 16-19

Questions 29-32

Cost-related non-adherence to medications with respect to disability

Questions 2, 15

Questions 9, 10, 12-14

Questions 4-7

Questions 24-28

* Underlined factors were explored in quantitative study while the italicized factors were explored in qualitative study.
Appendix K

Letter to the Advisory Council on Implementation of National Pharmacare

29th January, 2019

Dr. Eric Hoskins
Advisory Council on the Implementation of National Pharmacare Secretariat
Brooke Claxton Building
70 Colombine Driveway
Ottawa, ON K1A 0K9

Dear Dr. Hoskins,

We are writing to you today to contribute to the important work of the Advisory Council for the Implementation of National Pharmacare, and to share the results of our research:

Prescription cost barriers faced by people with disabilities in Canada (Gupta, McColl, Smith & Guilcher, 2019; full article linked).

Problem: There is growing evidence that even with comprehensive Pharmacare insurance, some members of the population face untenable costs for prescription medication. People with disabilities are one such group. The Canada Survey on Disability (2017) shows that despite being covered under the provincial social assistance plans, 13% of all persons with disabilities have unmet needs for prescription medication due to cost. People with disabilities constitute ~14% of the total Canadian population, and 23-28% live in poverty. In particular, those who experience barriers to employment, who have the health complications, and consequently the most complex medical regimens -- have the greatest risk of forgoing medications because of cost (Gupta, McColl, Guilcher & Smith, 2018; full article linked).

Evidence from our research: In our research with people with spinal cord injuries (n=160), we found that on average, participants used between 5-14 medications concurrently, and spent $197 per month on prescribed medications, with some over $2,000 per month. Although 92% of people with disabilities in our sample had some form of drug coverage, the prevalence of cost-related non-adherence to prescribed medications was far higher (37%) than in general population (9.6%), or even among seniors (8%) or other vulnerable populations (26%).

Medications for pain and mental health were most commonly forgone due to cost. Barriers included complex paper work to meet eligibility requirements, heavy deductibles, restrictions on public drug formularies, and long wait times for getting approvals. Our results showed that females, those between 26-64 years of age, and those low income or without regular employment were more likely to employ cost-related non-adherence to prescribed medications.
Financial barriers to medications have implications for healthcare and beyond. Those who rationed or skipped their prescribed medications had significantly more ambulatory and emergency visits in the past 12 months. They also cut back on basic needs such as food and other healthcare supplies. They sold assets such as a house or car or borrowed money from friends or family to manage the financial burden of their medications. Some faced serious financial hardships when having to choose between filling prescriptions and having food or looking after their children.

**Actions needed:** We respectfully submit the following recommendations to ensure that those in the greatest need are not left behind as Canada contemplates national Pharmacare. We recommend:

- Harmonization of drug insurance arrangements with other disability programs, to ensure full coverage of prescribed medication required by people with disabilities.
- Remove deductibles and minimize wait times for exceptional drug coverage for those with highest needs.
- Development of an information strategy for prescribing healthcare professionals, to assist them to be optimally aware of medication coverage and lowering costs for their patients.

These solutions will not only help Canadians with disabilities, but also any Canadian who faces a financial barrier to access their necessary prescriptions.

In summary, we hope that the results of our current work will provide meaningful guidance as you strategize how best to implement National Pharmacare that ensures equitable, burden-free, safe and timely access to prescription medications for all Canadians. Please do not hesitate to be in touch if you wish to know more about our work. Contact information is appended below.

Sincerely,

Shikha Gupta, MSc
Doctoral candidate
School of Rehabilitation Science
Queen’s University

Mary Ann McColl, PhD, MTS
Professor, Rehabilitation Science, Queen’s Unive
Associate Director, Centre for Health Services & Policy Research
Academic Lead, Canadian Disability Policy Alliance
Appendix L

Publication record of manuscript 1

Cost-related nonadherence to prescription medications in Canada: a scoping review

Shikha Gupta1
Mary Ann McColl1
Sara J Guilcher2
Karen Smith3

1School of Rehabilitation Therapy, Queen’s University, Kingston, ON, Canada; 2Leslie Dan Faculty of Pharmacy, University of Toronto, Toronto, ON, Canada; 3Department of Physical Medicine and Rehabilitation, School of Medicine, Queen’s University, Kingston, ON, Canada

Purpose: The evidence is emerging that prescription medications are the topmost drivers of increasing health care costs in Canada. The financial burden of medications may lead individuals to adopt various rationing or restrictive behaviors, such as cost-related nonadherence (CRNA) to medications. Therefore, the purpose of this study is to provide an overview of the type, extent, and quantity of research available on CRNA to prescription drugs in Canada, and evaluate existing gaps in the literature.

Methods: The study was conducted using a scoping review methodology. Six databases were searched from inception till June 2017. Articles were considered for inclusion if they focused on extent, determinants, and consequences of CRNA to prescription medications use in the Canadian context. Variables extracted for data charting included author(s), year of publication, study design, the focus of the article, sample size, population characteristics, and key outcomes or results.

Results: This review found 37 studies that offered evidence on the extent, determinants, and consequences of CRNA to prescription medications in Canada. Depending on the population characteristics and province, the prevalence of CRNA varies between 4% and 36% in Canada. Canadians who are young (between 18 and 64 years), without drug insurance, have lower income or precarious or irregular employment, and high out-of-pocket expenditure on drugs are most likely to face CRNA to their prescriptions. The evidence that CRNA has negative health and social outcomes for patients is insufficient. Literature regarding the influence of prescribing health care professionals on patients’ decisions to stop taking medications is limited. There is also a dearth of literature that explores patients’ decisions and strategies to manage their prescription cost burden.

Conclusion: More evidence is required to make a strong case for national Pharmacare which can ensure universal, timely, and burden-free access to prescription medications for all Canadians.

Keywords: Pharmacare, medication adherence, drug costs, drug insurance

Introduction
Prescription medications play an important role in the treatment and prevention of
Appendix M

Publication record of manuscript 2

An Adapted Model of Cost-Related Nonadherence to Medications Among People With Disabilities

Shishir Gupta, RPh, MD, Mary Ann McCoy, PhD, Sara J. T. Guilettie, PhD, Karen Smith, MD

First Published August 20, 2019 · Research Article · https://doi.org/10.1774/106425731866079

Abstract

Despite emerging evidence on cost-related nonadherence (CRNA) to prescription medications, there is little conceptualization and exploration of this phenomenon with respect to disability. Specifically, there is a gap in the literature that explores factors influencing medication cost-adherence relationship among individuals living with a disability. To advance research on and policy for CRNA to medications among people with disabilities, we need a framework that can contribute towards guiding solutions to this problem. We examined the applicability of Piette and colleagues’ existing model for CRNA to the context of people with disabilities and suggested an adapted model (CRNA to medications for persons with disability [CRNA-d]) that can provide a more specific conceptualization of CRNA with respect to disability. The adapted CRNA-d model depicts that CRNA to prescription medications with respect to disability is a dynamic and multifaceted phenomenon, determined by various socioeconomic, disability-related, medication-related, prescriber-related, and system-related factors. We discuss how higher susceptibility to health complications, barriers to income and employment, additional health care costs, the complexity of medical regimens, limited access to physician services, and other policy-related factors increase the risk of persons with disabilities developing CRNA to medications.
Appendix N

Publication record of manuscript 3

Prescription medication cost, insurance coverage, and cost-related nonadherence among people with spinal cord injury in Canada

Shikha Gupta1 · Mary Ann McColl2 · Karen Smith3 · Sara J. T. Golicher4

Revised: 4 September 2019 / Revised: 9 December 2019 / Accepted: 11 December 2019 © The Author(s), under exclusive licence to International Spinal Cord Society 2020

Abstract

Study design Observational cross-sectional study.

Objectives To describe the most common prescription medications used and the extent of out-of-pocket cost, insurance coverage, and cost-related nonadherence (CRNA) for those medications by people with spinal cord injury (SCI) in Canada.

Setting Community in Canada.

Methods It was an observational study wherein data were collected through a cross-sectional online survey from individuals living with an SCI in Canada. We used descriptive statistics to describe the extent of drug cost, insurance coverage and CRNA among study sample, and analytical statistics to find association of CRNA with sociodemographic, injury-related and medication-related characteristics of the sample.

Results Individuals with an SCI (n = 150) used an average of five medications and spent a median of $49 (interquartile range: $234.75) per month on their medications. More than 90% of participants had some form of drug insurance, though 37% reported CRNA. The most common medications that were forgone due to cost included opioids, antidepressants, and drugs for genitourinary and muscular spasms. Individuals with paraplegia and nontraumatic SCI had higher drug costs, though injury-related characteristics did not influence CRNA. Sex, monthly drug expenditure, and monthly additional healthcare costs were significantly associated with CRNA.

Conclusions People with SCIs are at risk of experiencing CRNA to their prescription medications despite having insurance coverage. Decision makers for the national pharmacare in Canada should account for their concerns judiciously.

Introduction

The cost of living with a spinal cord injury (SCI) has been estimated between 1.47 and 3.03 million Canadian dollars in a lifetime [1, 2]. Secondary complications and health conditions make people with SCI high users of medications in comparison to the general population [3, 4]. Prescription medications now constitute one of the topmost drivers of healthcare costs in Canada. In 2018, pharmaceutical drugs represented the second highest expenditure for healthcare (16%, after hospitals, 28%), and expenditure growth on drugs outpaced that for hospitals and physicians [5].

In Canada, prescription drug coverage is not included in the universal health insurance scheme. Instead, there are five possible ways that people pay for prescription medication: private drug insurance, employer-sponsored insurance, provincial drug benefits (for adults >65 years, people on social assistance, or those with catastrophic drug costs), inclusion on a family member’s plan, or out of pocket. For all of the insurance options, the extent of coverage varies, and individuals may pay premiums, co-payments, or deductibles. Out-of-pocket drug costs for Canadians have been estimated between $0 and $2000 a year, depending on the medication costs, insurance, age, employment, and income [6, 7].

In the absence of universal pharmacare, many people choose to forgo their medications due to cost, a phenomenon called cost-related nonadherence (CRNA) [8]. CRNA has been observed among chronically ill populations, and those with lower income, irregular employment, lack drug
Appendix O

Publication record of manuscript 4

Managing Medication Cost Burden: A Qualitative Study Exploring Experiences of People with Disabilities in Canada

Shikha Gupta 1,2*, Mary Ann McColl 1,3, Sara J.T. Guilcher 2,4 and Karen Smith 3

1 School of Rehabilitation Therapy, Queen's University, Kingston, ON K7L 3N6, Canada
2 Leslie Dan Faculty of Pharmacy, University of Toronto, Toronto, ON M5S 3M2, Canada
3 Department of Physical Medicine and Rehabilitation, School of Medicine, Queen’s University, Kingston, ON K7L 3N6, Canada
4 Correspondence: shikha.gupta@queensu.ca

Received: 14 July 2019; Accepted: 18 August 2019; Published: 23 August 2019

Abstract: Despite the abundant literature on the burden of rising costs of prescription medications, there is limited research to explore how these costs affect people and the decisions they are forced to make within the context of disability. In this qualitative study, we explored strategies adopted, factors influencing, and the impact of some of these strategies to manage the burden of medication cost among persons with disabilities. We interviewed 12 adults with spinal cord injuries living in Canada, using a general inductive approach to analyze the data. We found that before cutting back on medications due to costs, participants generally tried and sought help from the government, employers, and/or their prescribers to improve their drug coverage. The key factors that participants considered while making decisions on the strategies included the cost and perceived importance of medications, their financial status, other competing needs, and their relationship with the prescribers. While some of their efforts were successful, many participants were still not able to obtain their medications as prescribed. In those cases, patients resorted to rationing strategies such as cutting back on medications, other essential needs, or selling assets. These strategies had serious implications on their health, healthcare utilization, and quality of life.

Keywords: medication cost; spinal cord injury; treatment burden; disability; Canada

1. Introduction

Despite the abundant literature on the treatment burden of chronic illnesses, there is limited research to understand the processes that underpin medication cost burden in general and especially among those with disabilities. The financial cost of living with a disability is substantially higher than that of living without a disability [1]. Our previous research has shown that the economic burden of medications faced by people with disabilities is higher than that experienced by their non-disabled counterparts. For people with disabilities, barriers to income and employment, higher susceptibility to health complications, complex medical regimens, additional health care costs, and the complexity of drug and social assistance programs perpetuate the risk of forgoing medications [2].

Prescribed medications for people with disabilities are often assumed to be covered under the provincial drug plans but many are not. This leads to a situation where people stop taking their medications (generally referred to cost-related non-adherence, CRNA) or adopt other rationing behaviors due to cost [3]. However, we do not know what strategies are used to cope with the cost burden of medication, how people are making medication rationing decisions, and the impact of rationing strategies on individuals, their families, and the healthcare system.
Appendix P

Infographic

MEDICATION COSTS BURDEN
Evidence from people with spinal cord injuries in Canada

160 PEOPLE SURVEYED ACROSS CANADA

47.5% MEDICATION USE
- Used >5 medications concurrently
- 296 different medications prescribed
- Antidepressants, anticonvulsants, antipsychotics, and opioids most commonly prescribed

$588
- Median annual cost paid out-of-pocket
- 30% paid more than 10% of the annual income on prescriptions
- 92% had some form of drug insurance

37% COST–RELATED UNDERUSE
- Reported taking less prescription due to cost
- Females are at risk of cost-related underuse
- Out-of-pocket medication costs >$50 per month predictive of underuse

10% IMPACT OF UNDERUSE
- Spent less on food or heat to pay for prescriptions; 5% borrowed money
- Those who stopped medications had 4 more ambulatory visits and 3 more emergency room visits in last year

RESULTS CALL FOR ATTENTION FROM POLICY–MAKERS FOR NATIONAL PHARMACARE IN CANADA

Presented by:
Shikha Gupta, PhD Candidate, Queen’s University