Primary care performance for persons with dementia in Ontario: the impact of interdisciplinary primary care on health service use

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December 2019

A thesis submitted to McGill University, Faculty of Graduate Studies and Research in partial fulfilment of the requirements of the degree of Doctor of Philosophy (PhD)

DEDICATION

To my father, the original Dr. N. Sourial.

ABSTRACT

BACKGROUND: Dementia is a complex progressive disease with a high impact for patients, families and the health care system and requires integrated, person-centered and coordinated care. Interdisciplinary primary care may be beneficial to providing timely access to care and managing the wide range of needs of this vulnerable population. Ontario's introduction of Family Health Teams offers one of the most comprehensive examples of interdisciplinary primary care; however, its potential impact on health service use in the dementia population has been unexplored. Moreover, whether the introduction of interdisciplinary primary care practices and other recent primary care reforms have influenced trends in the management of dementia in primary care and health service use in both men and women is unknown. Finally, current evaluations of natural policy experiments such as interdisciplinary primary care have not fully accounted for the methodological complexities associated with determining an unbiased effect.

OBJECTIVES: This dissertation aimed to fill these substantive and methodological gaps through four objectives: 1) to develop a comprehensive framework of population-based, primary care performance and health service use indicators relevant to dementia and identify a subset of priority indicators; 2) to describe sex differences in primary care performance and health service use over time in persons with dementia in Ontario; 3) to provide guidance on the use of causal inference methods for appropriate confounder selection in the evaluation of natural policy experiments and 4) to apply causal inference methods to estimate the effect of interdisciplinary versus non-interdisciplinary primary care on health service use for persons with dementia in Ontario.

METHODS: For Objective 1, the framework was developed through the selection of an initial framework based on a literature review, identification of relevant indicators within the framework and enrichment based on existing dementia indicators and guidelines. Prioritization of indicators

was carried out through a stakeholder survey. For Objective 2, an observational repeated cohort of community-dwelling persons newly identified with dementia between 2002 and 2015 were extracted from linked health administrative data held at ICES in Ontario. Rates in indicators were age-standardized and stratified by sex. For Objective 3, the practice of testing baseline group differences for confounder selection in non-randomized studies and appropriateness of current reporting guidelines were assessed, and recommendations were proposed based on recent advances in causal inference. For Objective 4, using the same repeated cohort, these methods were then applied to compare emergency department and hospital use in persons with dementia within an interdisciplinary versus non-interdisciplinary primary care setting. A manuscript for the results of each objective has been either published, submitted or prepared for submission in a peer-reviewed journal.

RESULTS: In the first manuscript, a framework of 37 indicators across eight domains of performance (Accessibility, Integration, Effectiveness, Efficiency, Equity, Safety, Population Health and Patient-Centeredness) was developed. Continuity of care, early stage diagnosis and access to home care were consistently rated as priorities by stakeholders. In the second manuscript, 18 indicators from the framework were operationalized. Few differences were observed between sexes, although men had fewer diagnoses first recorded by the family doctor, more visits to specialists, less use of home care, more hospitalizations and readmissions, and longer discharge delays. Most indicators remained relatively stable over time for both men (median relative change: 13.7%; interquartile range (IQR): 4.5% to 29.7%) and women (median relative change: 15.7%; IQR: 5.9% to 31.5%). Notable improvements over time for both sexes included access to an interdisciplinary primary care team, use of home care and decreased use of long-term care. Areas of worsening included a higher occurrence of emergency department visits, lower continuity of

care and longer discharge delays. In the third manuscript, the misguided practice of using observed imbalances between study groups in non-randomized studies for confounder selection was explained; current reporting guidelines were found to be incomplete or inappropriate. A practical example was used to demonstrate how recent methods in causal inference can be used to better inform confounding. In the fourth manuscript, persons with dementia in an interdisciplinary primary care group were found to have a higher risk of having an emergency department visit (Relative risk (RR): 1.03; 95% CI:1.01-1.05) or non-urgent emergency department visit (RR:1.22; 95% CI:1.18-1.28) in the year following diagnosis compared to those in an non-interdisciplinary primary care group. Differences with respect to hospitalization outcomes were inconclusive.

CONCLUSION: The development and operationalization of a comprehensive framework of indicators sets a foundation for ongoing surveillance of trends and evaluation of health policies for persons with dementia using routinely available data at a population-level. Findings on sex differences in trends in indicators raise awareness on the similarities and differences in management and health system use for men and women newly diagnosed with dementia. These results underscore the importance of developing care plans and interventions adapted to their specific needs. Practical guidance on the use of causal inference methods may help to strengthen the evaluation of policies and interventions. That interdisciplinary primary care did not translate into a reduction in emergency department or hospital use suggests that more training and support in primary care teams may be needed to positively affect health service use in persons with dementia. This work will open avenues for future health services dementia research and increase the use of causal inference methods in health policy evaluation.

RÉSUMÉ

CONTEXTE : La démence est une maladie évolutive complexe qui a des répercussions importantes sur les patients, les familles et le système de santé, et qui nécessite des soins intégrés, coordonnés et axés sur la personne. L'interdisciplinarité en soins de santé primaires peut s'avérer bénéfique pour fournir en temps opportun les soins multiples et variés dont cette population vulnérable a besoin. La mise sur pied en Ontario des *Family Health Teams* offre l'un des exemples d'équipes interdisciplinaires en soins de santé primaires les plus complets et répandus. Cependant, son impact potentiel sur l'utilisation des services de santé des personnes vivant avec une démence n'a pas encore été étudié. De plus, depuis l'introduction de ces équipes interdisciplinaires et d'autres réformes en soins de santé primaires, on ne connait pas l'évolution de la gestion de la démence, ni de l'utilisation des services de santé par les hommes et les femmes. Enfin, l'évaluation des expériences naturelles, telle que l'introduction d'équipes interdisciplinaires, n'a pas pris en compte toutes les complexités méthodologiques associées à l'estimation non biaisée de leurs effets.

OBJECTIFS : Cette thèse visait donc à combler ces lacunes substantielles et méthodologiques et se divisait en quatre objectifs : 1) élaborer un cadre conceptuel d'indicateurs de performance en soins primaires et d'utilisation des services de santé adaptés à la démence et mesurables au niveau de la population, ainsi que définir un sous-ensemble d'indicateurs prioritaires; 2) décrire les tendances au fil du temps et les différences entre les hommes et les femmes vivant avec une démence en Ontario pour ces indicateurs ; 3) fournir des recommandations sur les avantages de l'utilisation des méthodes en inférence causale en comparaison avec les méthodes actuelles pour sélectionner les facteurs de confusion dans l'évaluation des politiques de santé et 4) utiliser ces méthodes pour estimer l'effet de l'interdisciplinarité sur le recours des services de santé en Ontario chez les personnes vivant avec une démence.

MÉTHODES : Pour l'objectif 1, l'élaboration du cadre conceptuel a été réalisée comme suit : d'abord par la sélection d'un cadre initial fondé sur une revue de la littérature, ensuite par l'identification d'indicateurs pertinents, et finalement par l'ajout d'indicateurs fondé sur les lignes directrices existantes en démence. La priorisation des indicateurs a été effectuée au moyen d'un sondage auprès des intervenants. Pour l'objectif 2, une cohorte observationnelle répétée de personnes nouvellement identifiées avec un diagnostic de démence entre 2002 et 2015 et vivant dans la communauté a été extraite des banques de données médico-administratives détenues à ICES en Ontario. Les taux des indicateurs ont été normalisés selon l'âge et stratifiés selon le sexe. Pour l'objectif 3, une pratique commune consistant à vérifier les différences entre groupes non-randomisés pour la sélection des facteurs de confusion a été examinée, et la pertinence des lignes directrices actuelles a été évaluée et des recommandations fondées sur les méthodes en inférence causale ont été proposées. Pour l'objectif 4, en utilisant la même cohorte répétée, ces méthodes ont ensuite été appliquées pour comparer l'utilisation des services d'urgence et hospitalisations chez les personnes vivant avec une démence ayant accès à une équipe interdisciplinaire ou non. Un manuscrit des résultats de chaque objectif a été publié, soumis ou préparé en vue de sa soumission dans une revue dotée de comité de pairs.

RÉSULTATS : Dans le premier manuscrit, un cadre conceptuel de 37 indicateurs répartis à travers huit domaines de performance (accessibilité, intégration, efficacité, efficience, équité, sécurité, santé de la population et soins axés sur le patient) a été élaboré. La continuité des soins, le diagnostic précoce et l'accès aux soins à domicile ont été considérés comme des priorités par les intervenants. Dans le deuxième manuscrit, 18 de ces indicateurs ont été opérationnalisés. Des différences minimales ont été observées entre les sexes, bien que les hommes aient eu moins de diagnostics enregistrés par le médecin de famille, plus de visites chez des spécialistes, moins de

recours aux soins à domicile, plus d'hospitalisations et de réadmissions, et des délais de congé plus longs. La plupart des indicateurs sont restés relativement stables au fil du temps tant chez les hommes (changement relatif médiane : 13,7 % ; écart interquartile (IQR) : 4,5 % à 29,7 %) que chez les femmes (changement relatif médiane : 15,7 % ; IQR : 5,9 % à 31,5 %). L'accès à une équipe interdisciplinaire de soins primaires, le recours aux soins à domicile et la diminution du recours aux soins de longue durée se sont améliorés au fil du temps chez les deux sexes. Parmi les domaines où la situation s'est détériorée, une fréquence plus élevée des visites à l'urgence, une plus faible continuité des soins et des délais de congé plus longs ont été observés. Dans le troisième manuscrit, l'utilisation des différences observées entre les groupes non-randomisés pour la sélection de facteurs de confusion a été discutée comme une mauvaise pratique. Les lignes directrices actuelles se sont avérées incomplètes ou inappropriées et une démonstration de l'utilisation des méthodes récentes en inférence causale pour mieux informer le choix et la gestion des facteurs de confusion a été faite. Dans le quatrième manuscrit, les personnes vivant avec une démence ayant accès à une équipe interdisciplinaire de soins primaires présentaient un risque plus élevé de visite à l'urgence (risque relatif (RR) : 1,03 ; IC à 95 % : 1,01-1,05) ou de visite non urgente (RR : 1,22 ; IC à 95 % : 1,18-1,28) durant l'année suivant le diagnostic comparativement au groupe n'ayant pas accès à une équipe interdisciplinaire. Les résultats relatifs à l'hospitalisation étaient peu concluants.

CONCLUSION : L'élaboration et l'opérationnalisation d'un cadre d'indicateurs mesurables au niveau de la population facilitera la surveillance continue des tendances et l'évaluation des politiques de santé pour les personnes vivant avec une démence. Les constatations sur les similitudes et des différences dans la prise en charge et l'utilisation des services chez les hommes et les femmes vivant avec une démence soulignent l'importance d'élaborer des plans de soins et

des interventions adaptés à leurs besoins spécifiques. Les recommandations pratiques sur l'utilisation des méthodes en inférence causale présentées serviront à renforcer l'évaluation des politiques et des interventions. Le fait que l'accès à des soins primaires interdisciplinaires n'a pas entraîné une réduction de l'utilisation des services d'urgence et hospitalisation suggère que plus de formation et de soutien aux équipes interdisciplinaires pourraient être nécessaires. Ces travaux ouvriront la voie à de futures recherches sur la démence dans les services de santé et augmenteront l'utilisation des méthodes d'inférence causale dans l'évaluation des politiques de santé.

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LIST OF ABBREVIATIONS

CIHR: Canadian Institutes for Health Research

CMAJ: Canadian Medical Association Journal

ED: Emergency Department

DAG: Directed Acyclic Graph

FHT: Family Health Team

FHO: Family Health Organization

FMG: Family Medicine Group

IPC: Interdisciplinary primary care

JAGS: Journal of the American Geriatrics Society

JAMA: Journal of the American Medical Association

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ACKNOWLEDGMENTS

Accomplishing a PhD and becoming a researcher has always been my ultimate dream. With what seemed like a distant goal several years ago was made a reality through the support and mentorship of many people. First, I would like to express my deepest thanks to my supervisor Dr. Isabelle Vedel for her endless guidance and mentorship during my doctoral degree. Thank you for always being available, for imparting me with your expertise in health services dementia research, coaching me in the strategic aspects of research, supporting me both financially and on a personal level and always having the right reassuring words and solutions to every problem. I would also like to sincerely thank my co-supervisor, Dr. Tibor Schuster, for opening my mind to modern causal inference, lending his expertise in navigating its complexities and nuances, teaching me the usefulness of framing methodological questions through DAGs and causal theory and supporting me to present my work at epidemiology and biostatistics conferences. To Dr. Howard Bergman, my career long academic mentor, thank you for convincing me to be the apple in the bowl of oranges and steering my career to primary care research. I will no doubt continue to need your words of wisdom as I embark on my role as researcher. I also would like to express my most sincere gratitude to Dr. Susan Bronskill, my committee member, for guiding my doctoral work and sharing her keen expertise in population-based health services research, dementia and sex and gender research as well as welcoming me as an ICES student and facilitating my remote access to the data. Thank you also to Dr. Jacob Etches, senior analyst at ICES, for extracting the cohort data and answering and re-answering my many questions. I would also like to thank Dr. Gillian Bartlett, who along with Dr. Vedel, gave me the confidence and push I needed to apply and successfully obtain a Vanier scholarship. To my team members, Geneviève, Claire, Mélanie, Marine, Mary and Martine, thank you for supporting me through endless iterations of results, tables and graphs, sharing successes

and failures together and all the laughter that made each day a joy to come in; you are, bar none, the best team I have ever had the pleasure to work with. A special thanks to Geneviève for the long friendship we have built and being my go-to life coach.

Finally, this dissertation would not have been possible without the unwavering support of my family. First and foremost, I owe much of this achievement to my husband Jimmy who took the parenting duties on countless evenings and weekends without ever complaining and for his emotional support through this long academic road. To my wonderful children, William and Matthew, thank you for always grounding me in the present and reminding me of my priorities. To my mother for always believing in me, babysitting on demand and her clinical wisdom on my research findings. To my sister for listening to the daily ups and downs and doing virtual homework together on weekends at the department. To my grandmother whom never quite understood what I did as a PhD student but whom I know would still be very proud. And to my cats, for keeping me company on those lonely days and late nights writing at home. It truly takes a village to raise a researcher and I am forever grateful to everyone who has been part of this incredible journey with me.

Acknowledgement for funding support

This work was funded through a Canadian Institutes for Health Research (CIHR) Vanier Canada Graduate Scholarship and a CIHR grant for the Canadian Consortium on Neurodegeneration in Aging (2014-2019) (ID# CNA-137794). This research was also supported by ICES, which is funded by an annual grant from the Ontario Ministry of Health and Long-Term Care. I also received several training awards from CIHR Institute of Health Services and Policy Research, the Department of Family Medicine (McGill University) and the Quebec Research Network on Aging to attend conferences within Canada, the United States and Europe.

PREFACE

Format of the thesis and contribution of authors

This dissertation follows the format of a manuscript-based thesis. It is comprised of four manuscripts, two of which have been published, one has been revised and resubmitted and one is in final preparation for submission.

As PhD candidate and first author on all manuscripts in this dissertation, I myself conceived of the idea for each study, elaborated the objectives and research design, conducted the data analysis and led the interpretation of findings as well as the writing of the manuscripts. The direction of this research work was guided by my supervisors Dr. Isabelle Vedel and Dr. Tibor Schuster and in consultation with my committee members, Dr. Susan Bronskill and Dr. Howard Bergman. Dr. Vedel and Dr. Schuster provided overall guidance on the protocol development, methods, analysis, interpretation of findings and presentation of results. Dr. Vedel's expertise in primary care dementia health services research ensured the relevance and originality of this work; Dr. Schuster's expertise in biostatistics and causal inference informed my methodological development, data analysis and interpretation of results. Dr. Bronskill provided expertise on dementia health services research using ICES health administrative data as well as contextual factors specific to the health system in Ontario. As an international expert in dementia research, Dr. Bergman provided input on the scope and pertinence of the research questions for stakeholders. While I obtained feedback and direction from supervisors and committee members, I take full responsibility for the integrity, quality, accuracy of this body of work and declare its content to be my original doctoral work. All co-authors of the papers approved their inclusion in this dissertation.

A list of all four manuscripts with specific author contributions is provided below:

Manuscript 1: Nadia Sourial PhD candidate, Claire Godard-Sebillotte MD PhD candidate, Susan E Bronskill PhD, Marine Hardouin BSc, Isabelle Vedel MD PhD. Framework and prioritization of dementia primary care performance indicators based on health administrative data. Submitted to the Canadian Journal of Aging.

NS made substantial contributions to the conception and design, analysis, interpretation of data, and the drafting, revisions and final approval of the manuscript. CGS contributed to the conception and design, interpretation of data, drafting, revision and final approval of the manuscript. SB contributed to the interpretation of data, revision and final approval of the manuscript. MH made substantial contributions to the design, analysis, interpretation of data, and the drafting, revision and final approval of the manuscript. IV contributed to the conception and design, interpretation of data, drafting, revision and final approval of the manuscript.

Manuscript 2: Nadia Sourial PhD candidate, Isabelle Vedel MD PhD, Claire Godard-Sebillotte MD PhD candidate, Jacob Etches PhD, Genevieve Arsenault-Lapierre PhD, Susan E Bronskill PhD. Sex differences in dementia primary care performance and health service use: A population-based study. Journal of the American Geriatrics Society (JAGS). 2020 Feb 5. doi: 10.1111/jgs.16347.

NS made substantial contributions to the conception and design, analysis, interpretation of data, and the drafting, revisions and final approval of the manuscript. IV contributed to the conception and design, interpretation of data, drafting, revision and final approval of the manuscript. CGS contributed to the conception and design, interpretation of data, drafting, revision and final approval of the manuscript. JE contributed to the acquisition of data, analysis, interpretation of

data, revision and final approval of the manuscript. GAL contributed to the conception and design, interpretation of data, drafting, revision and final approval of the manuscript. SE contributed to the conception and design, interpretation of data, drafting, revision and final approval of the manuscript.

Manuscript 3: Nadia Sourial PhD candidate, Isabelle Vedel MD PhD, Mélanie Le Berre MSc PT, Tibor Schuster PhD. (2019). Testing group differences for confounder selection in non-randomized studies: flawed practice. Canadian Medical Association Journal (CMAJ), October 28, 2019, 191(43): E1189-E1193; DOI: https://doi.org/10.1503/cmaj.190085 .

NS contributed to the study conception and design, analysis and interpretation of data, drafting and revision of the work and final approval the version to be published. MLB contributed to the study design, analysis and interpretation of data, drafting and revision of the work and final approval the version to be published. IV contributed to the study conception and design, interpretation of data, revision of the work and final approval the version to be published. TS contributed to the study conception and design, interpretation of data, drafting and revision of the work and final approval the version to be published.

Manuscript 4: Nadia Sourial PhD candidate; Tibor Schuster PhD; Susan Bronskill PhD; Claire-Godard-Sebillotte MD PhD candidate; Jacob Etches PhD; Isabelle Vedel MD PhD. Comparison of interdisciplinary versus non-interdisciplinary primary care on emergency department and hospital use in persons with dementia: A population-based study. To be submitted to the Journal of the American Medical Association (JAMA).

NS made substantial contributions to the conception and design, analysis, interpretation of data, and the drafting, revisions and final approval of the manuscript. TS contributed to the study design, interpretation of data, revision of the work and final approval the manuscript. SB contributed to the conception and design, interpretation of data, drafting, revision and final approval of the manuscript. CGS contributed to the conception and design, interpretation of data, drafting, revision and final approval of the manuscript. JE contributed to the acquisition of data, analysis, interpretation of data, revision and final approval of the manuscript. IV contributed to the conception and design, interpretation of data, drafting, revision and final approval of the manuscript.

Ethics approval

This dissertation was approved by the Research Ethics Board of McGill University and the Jewish General Hospital in Montreal, Canada. The use of ICES data in this project was authorized under section 45 of Ontario's Personal Health Information Protection Act, which does not require review by a Research Ethics Board. Please refer to Appendix A for ethics approvals.

STATEMENT OF ORIGINALITY

This dissertation generated new evidence that fills several important substantive and methodological gaps. A key contribution is the creation of a comprehensive framework of primary care performance and health service use indicators for the dementia population. This framework provides a foundation for the ongoing surveillance and evaluation of trends in dementia management at a population level using objective and relevant indicators. The framework I developed is currently being used in Ontario, Quebec and New-Brunswick by the Research on the Organization of Services for Alzheimer's (ROSA) team to describe and contrast trends within and across all three provinces. Based on this framework, this doctoral work produced the most comprehensive portrait to date on sex differences in primary care performance and health system use in persons with dementia, an area of dementia research which has, until now, been neglected.

This work also brings to the forefront how causal inference methods can be added to the toolbox of health services researchers to strengthen health policy evaluation and minimize bias. These methods were used to conduct a novel and robust evaluation of the impact of one key aspect of primary care reform in Canada: the introduction of a team-based, interdisciplinary approach to primary care. I measured the impact of interdisciplinary vs non-interdisciplinary primary care on health service use for persons with dementia in Ontario. Together, this evidence will help inform upcoming dementia strategies in Ontario, Canada and elsewhere based on comprehensive and sound evidence.

1 CHAPTER 1: INTRODUCTION

Dementia, including Alzheimer's disease, has been recognized by the World Health Organization (WHO) as a global public health crisis of the 21st century.[1,2] There are currently over 500,000 Canadians living with dementia. If current projections hold, the prevalence of dementia in Canada will double every 20 years due to population aging, affecting one in five baby boomers, with costs expected to surpass $800 billion over the next generation.[3-5] Many persons with dementia lack adequate management in terms of diagnosis, treatment and follow-up care. In addition, most suffer from multiple comorbidities and are more likely to have fragmented and poor coordination of care, resulting in the overuse of health services, including increased emergency department (ED) visits and hospital admissions.[1,2,4-8] Women, who are less likely to have informal caregivers or financial resources and who account for 2/3 of the dementia population, may be particularly vulnerable to suboptimal care.[9-11]

Interdisciplinary primary care (IPC) has been suggested as an ideal approach to managing the growing dementia population.[7,8,12-18] First, as the point of first contact into the health system, primary care is well suited to provide a person-centered, rather than a disease-centered approach to care. Moreover, given the scope of expertise required to deal with the complex range of cognitive, functional, social, and emotional problems associated with dementia as well as managing other chronic conditions and providing support to caregivers, an integrated multidisciplinary approach to diagnosing and managing dementia has been recommended.[1,5,12,17,18] At a system-level, in the last two decades, several Canadian provinces have introduced IPC models into their health systems with the aim of promoting better access and comprehensive care with a focus on prevention and management of chronic diseases.[7,19-24]

In Ontario, close to 200 Family Health Teams (FHTs), comprised of physicians, nurses, nurse practitioners, social workers, occupational therapists, pharmacists and other health professionals[25], were introduced across the province between 2005 and 2012. As one of the most wide-scale example of IPC models in North America,[26] FHTs may be particularly beneficial to meet the complex range of needs for persons with dementia compared to traditional physician-based practices. It is currently unknown, however, whether trends in health service utilization in the dementia population have changed since their introduction and whether any such changes were equitable for men and women with dementia.

Province-wide evaluation of IPC in terms of its potential effect on health service utilization has also been scarce and limited to the general population.[27-30] Moreover, comparisons in the use of health services, including ED and hospital use, between patients in an IPC setting versus other primary care models have shown mixed results. Inconsistencies have been partially attributed to limitations in isolating the "true" effect of each care model owing to the myriad of external and uncontrolled factors, such as voluntary enrolment and pre-existing differences in patient and practice characteristics across the different models.[27,28,30,31] Robust statistical methods that can meet these methodological complexities are essential to determine if and to what extent access to IPC can curb unnecessary health service utilization at the system level by improving access to and quality of care for dementia patients. New developments in the causal inference literature offer promising avenues to address these challenges. They have shown that, under certain specific conditions, the population-level causal effect of natural experiments, like the introduction of FHTs, can be reliably estimated.[32,33] A very limited number of studies have employed causal inference methods in other health policy evaluation settings but have not been explicit about how such methods can be used to strengthen the evaluation of such natural experiments.[34-37] It is thus

important to make these methods more accessible and provide guidance on how they can be used in practice.

This dissertation aimed to fill these important substantive and methodological gaps by describing changes over time in health service use in both men and women with dementia in Ontario and applying novel causal inference methods to estimate the effect of IPC versus non-IPC on health service, using Family Health Teams as a key example of IPC. This dissertation is organized as follows: First, a literature review will present an overview of dementia, the shift to IPC in the health system, the limited evidence and methodological challenges in evaluating the impact of IPC and the opportunities to apply advanced causal inference methods for this impact assessment. Following the literature review, the thesis rationale, knowledge gaps and specific objectives will be outlined. Each study will then be presented including the motivation, methods, results and interpretation. Finally, concluding remarks will summarize the body of work, its implications and future directions.

2 CHAPTER 2: LITERATURE REVIEW

2.1 Dementia: A health system priority

2.1.1 Overview of dementia

Dementia is a chronic disease and a degenerative neurological disorder that leads to memory loss, cognitive and functional impairment and eventually death. It is formally defined as "a clinical syndrome of cognitive decline that is sufficiently severe to interfere with social or occupational functioning". [6,38] Age is the strongest predictor of dementia. Low education, cardiovascular disease, hypertension, diabetes, smoking and head trauma are also considered to be strong risk factors.[2,4,6] Women account for 2/3 of dementia cases.[39] There are many subtypes of dementia with Alzheimer's disease accounting for 60 to 80 percent of dementia cases. Other types include vascular, mixed and Lewy body dementia.[40] Persons newly diagnosed with dementia have a life expectancy of between 7 to 10 years and often die of pneumonia due to their compromised immune system and susceptibility to infection.[5,6] Four drugs are available in Canada with modest efficacy in improving the symptoms of dementia: Exelon ®, Reminyl ® and Aricept ® (cholinesterase inhibitors) for mild to advanced dementia and Ebixa ® (memantine) for moderate to advanced dementia. [41] However, there are currently no pharmacological treatments that can cure or slow the disease.[41]

The prevalence of dementia among persons 65 years old and older in Canada has been estimated at 8%[42] and is projected to double over the next generation.[4,5] Some studies in the United States and other developed countries have shown dementia incidence may actually be decreasing. These trends have been mostly attributed to improvements in risk factors including low education and cardiovascular disease.[43-45] While this is promising and points to potential interventions for

preventable risk factors, dementia will continue to be an increasingly important challenge for our healthcare systems as populations continue to age.

2.1.2 Impact of dementia

2.1.2.1 Impact on patients and caregivers

Dementia is associated with multiple comorbidities, disability and death. On average, persons with dementia have two to three other chronic conditions such as diabetes, hypertension, cancer, depression, heart or lung disease.[46,47] They have been shown to have more comorbidities and a higher illness burden compared to matched cohorts without dementia.[48] These comorbidities can interact with dementia in complex ways, exacerbating dementia symptoms and complicating treatments for these conditions.[49] Dementia is also the leading cause of disability and institutionalization among older Canadians.[2,50,51] It contributes to 13% of years of life lived with disability - more than stroke (4%), heart disease (4%) and cancer (2%) - and accounts for almost half of long-term care users.[3,50] Dementia is reported as the 8th leading cause of death in Canada,[52] however, a recent study found that dementia is severely underreported at time of death and may in fact contribute to as many deaths as heart disease or cancer.[53]

Beyond the impact on patients, dementia also uniquely affects family caregivers. Twice as many dementia caregivers describe considerable financial, emotional and physical difficulties compared to caregivers of people without dementia, with 40% reported to suffer from depression.[6] Caregivers also contribute a large portion of informal hours of care affecting their ability to maintain stable employment resulting in lost income and productivity.[2,54] According to the Alzheimer's Society, these caregiver hours are expected to more than triple in the next 30 years.[5]

2.1.2.2 Impact on the health system

The dementia population represents a major cohort of high users within the health system.[50,55] Compared to older adults with similar health and demographic characteristics, persons with dementia have higher rates of emergency department (ED) visits, hospitalizations, physician visits, and home care services. [48,50] The incidence of hospital use, in particular, has been shown to be at least twice as high in the older population when dementia is present. [50,56] Dementia has also been associated with an increased risk of return visits to the ED within 30 days.[18] Overall, health service utilization costs have been calculated to be 5.5 times greater for those living with dementia than older persons without dementia.[57,58] In terms of costs to the overall health system in Canada, a 10-fold increase in total costs related to dementia is expected over the next generation, representing a cumulative economic burden of $872 billion.[5]

If current projections in dementia prevalence continue to hold, dementia will also create an increase in demand and shortfall of supply for long-term beds which will translate into a greater proportion of the dementia population being managed in the community, from 33% in 2008 to 43% by 2038.[5] This shift is projected to substantially increase the need for community care services as well as increased caregiver burden.[5]

2.1.3 Sex and gender differences in dementia

Sex and gender play an important role on the risk of dementia, its clinical presentation and progression. [9,39,59] Evidence is emerging that women have a higher lifetime risk of developing dementia not only due to longer longevity but also due to differences in biological and genetic factors related to brain aging as well as differences in disease and/or gender-related risk factors.[9,39,60] For example, older women are more likely to have a lower educational attainment

and engage in less exercise, both risk factors for dementia, due in part to historically greater parental roles in women than in men.[59] Women also have twice the risk of developing depression, a major risk factor for dementia, compared to men.[59,61] This risk difference has been attributed to hormonal differences throughout the life course as well as the increased role of women as caregivers.[39] Women, mainly spouses and daughters, account for 60% of informal caregivers for persons with dementia.[9] Caregivers of persons with dementia have been shown to be at increased risk of depression and even dementia compared to non-caregivers and caregivers of persons without dementia.[39,62]

There are also sex differences in the clinical presentation and progression of dementia. At similar early stages of cognitive impairment, women tend to perform better on cognitive tests than men due to improved reserves in verbal memory.[63] As such, women tend to be diagnosed later in the disease, leading to delayed management and more rapid decline after diagnosis than men.[39,63-65] On the other hand, men with dementia have been shown to have a higher prevalence of severe comorbidities such as arrhythmia, chronic obstructive pulmonary disease, and cancer than women.[50,66-68] They are also more likely to develop aggressive behavioral and psychological symptoms of dementia.[69,70]

Together, these sex-specific differences may have important consequences on the management of dementia and need for health services. However, despite emerging evidence on the role of sex and gender in the etiology of dementia, research on possible differences with respect to dementia management and health service use remains a neglected area of research.[39]

2.1.4 Inadequate dementia management

Lack of access to diagnosis, treatment and management throughout the course of the disease is a common challenge for men and women with dementia and reflect both an under and overutilization of services.[5] Even in high-income countries, only 50% of cases are diagnosed.[8] These suboptimal rates are mainly due to a scarcity of dementia specialists and adequately trained and supported primary care physicians resulting in delayed diagnoses.[5,8] This low diagnostic coverage in the early stages of disease often leads to an underutilisation of existing evidence-based interventions to improve functional status, support caregivers and preventive measures to avoid unnecessary emergency visits and hospitalizations.[2,8,71] The lack of professional support can also lead to increased patient anxiety and caregiver burden.[5]

At the same time, poor access and continuity of care in this vulnerable population often results in the overuse of hospital services, poor transitions of care, and increased overall health care costs.[72] This evidence of both under and overutilization of services points to a need for better management of dementia in the community.

2.1.5 Interdisciplinary primary care as the way forward to manage dementia

There is growing consensus that countries with a high-functioning primary care system provide more effective, efficient and equitable care to their population.[73-75] Within primary care, the use of IPC teams (including family physicians, nurse practitioners (NPs), nurses, social workers, occupational therapists and others) is considered by many to be an effective approach to delivering more timely, coordinated and comprehensive access to care, more efficient resource utilization and

better health outcomes for patients, compared to physician-only models of primary care, especially for the management of chronic diseases.[26,29,76-80]

IPC is also seen as a potentially advantageous approach to dementia care. [7,8,12-18] First, as the point of first contact into the health system, the primary care setting is well suited to provide a person-centered, rather than a disease-centered, approach to dealing with the complex range of cognitive, functional, social, and emotional problems associated with dementia, managing other chronic conditions and providing support to caregivers.[1,5,12,17,18] Unlike other chronic conditions, management of dementia relies less on medication and more on the integration of non-pharmacologic therapies from a wide range of healthcare providers.[17,81] Two studies on barriers and enablers to optimal primary dementia care found that multidisciplinary teams and case managers can promote better care.[16,82] Nurse practitioners, for example, can perform a wide variety of central tasks, such as conducting regular assessments of cognition and dementia symptoms, writing of referrals for tests, discussing possible treatment with patients and families, prescribing medication, following-up on imaging and test results, and in some cases making the initial diagnosis and care plan.[83] Nurse practitioners, nurses and social workers can also play an important role as "patient navigators" to help coordinate care across the health system and facilitate access to services.[29,84] Social workers can anticipate other needs such as respite care, financial services, counseling support groups, and crisis management.[17] Finally, occupational therapists can adapt the patient's home environment to deal with functional disabilities in tasks of daily living.[17]

2.2 Health systems' shift towards interdisciplinary primary care

2.2.1 Primary care reforms outside Canada

In the last two decades, many high-income countries, including the UK and United States, have implemented health reforms to re-organize their health systems around primary care. The creation of IPC teams has been a focus of these reforms. In the UK, most practices are now team-based, typically consisting of a group of family physicians, nurses and nurse practitioners (NPs) where NPs play a prominent role in decision making.[85] In the United States, the concept of a "patient-centered medical home" was developed by the Institute of Medicine in 2007 as a physician-directed interdisciplinary practice based on a whole-person perspective prioritizing enhanced access, coordinated and integrated care, quality and safety.[86,87] Patient-centered medical homes have been associated with improved patient care and outcomes, particularly for chronic disease management and prevention.[86]

2.2.2 Primary care reforms in Canada: A focus on Ontario's Family Health Teams, a prime example of interdisciplinary primary care

In 2000, Canada established an $800 million Primary Health Care Transition Fund to boost primary health care reform.[77] Acknowledging the growing body of evidence on the effectiveness of IPC teams in delivering high-quality, timely access to primary care, federal and provincial governments also set a goal that half of Canadians have access to team-based primary care by 2011.[88] Alberta, Ontario and Quebec have made the most system-wide changes with regards to the creation of interdisciplinary teams.[77]

Ontario's Family Health Teams (FHTs) are among the most comprehensive interdisciplinary primary care models in Canada and are one of the largest examples of a patient-

centered medical home in North America.[26] Between 2005 and 2012, 184 Family Health Teams were introduced over five waves of implementation.[25,89] They are considered the Ontario's government "flagship initiative in primary health care renewal".[90] The Government currently invests approximately $300 million annually in FHTs per year.[91] FHTs are formally defined as "an approach to primary health care that brings together different health care providers to co-ordinate the highest possible quality of care … each utilizing their experience and skills so that [patients] receive the very best care, when [they] need it, as close to home as possible." [89] The government's primary aim in implementing FHTs was to improve access to primary care.[29] Also considered key was the availability of health professionals to serve as "patient navigator to help guide patients through the health care system…and actively facilitate access to community-based services and secondary and tertiary care".[29] Other objectives included improved quality and continuity of care, increased patient and provider satisfaction and cost-effectiveness of primary care services.[29]

Several primary care enrolment models were rolled out over the last two decades in Ontario (Table 2.1). Of these models, only Family Health Organizations (FHOs) and Family Health Networks (FHNs) were eligible to apply to transition into FHTs and receive funding to recruit salaried health care professionals.[28] While FHTs retain the same elements as FHOs or FHNs, such as the remuneration method (blended capitation), use of electronic medical records, extended hours and access to 24/7 nurse teletriage services, the defining added-value of FHTs is their ability to offer "enhanced access to interprofessional, team-based care".[29] Teams are composed of three or more physicians, located at one or more locations, as well as nurses, nurse practitioners, social workers, dietitians, pharmacists, occupational therapists or other providers.[29] Interprofessional health providers can either be co-located or support multiple clinics within the FHT at different locations. FHTs vary in terms of team composition and programs depending on

local community needs, but all generally focus on chronic disease management, health promotion and disease prevention activities.[27] Compared to non-IPC models, FHTs have more formalized chronic disease management plans and a focus on patient-centered care.[92] The majority of FHTs today were previously FHOs.[93] As such, except for the added component of interdisciplinary primary care teams, FHTs share many of the same characteristics as FHOs including remuneration method, services and patient characteristics (Table 2.1). FHTs and FHOs also account for a large proportion of registered patients in Ontario (Table 2.1).

Table 2.1: Characteristics of primary care models in Ontario

Characteristic	Fee-for-service (FFS)	Community Health Centre	Comprehensive Care Model	Family Health Group (FHG)	Family Health Network (FHN)[a]	Family Health Organization (FHO) [a]	Family Health Team (FHT)
No. of patients	224,066	60,428	2,336,528		39,159	1,027,240	1,162,807
Year introduced	1966	1973	2005	2003	2001	2005	2005
Remuneration method	FFS	Salary	Enhanced FFS	Enhanced FFS	Blended capitation	Blended capitation	Same as FHO (FHN)[b]
Type of practice	Solo physician practice	Interdisciplinary primary care teams	Solo	Group physician practice	Group physician practice	Group physician practice	Same as FHO (FHN) + Interdisciplinary primary care teams
Patient enrolment	No	No	Yes	Yes	Yes	Yes	Yes
Extended hours	No	Yes	Yes	Yes	Yes	Yes	Yes

% of patients in major urban areas	79.5%	63.6%	79.5%	3.7%	66.5%	56.3%
% of patients in highest income quintile	-	13.5%	18.7%	26.0%	25.9%	21.0%
% recent registrants	8.1%	12.4%	9.4%	0.9%	2.9%	2.7%

2.3 Impact of interdisciplinary primary care on health service utilization in the general and older population

Evidence of the impact of IPC on health service use in the general population remains limited and has shown mixed results.[27-30,77,94,95] In the United States, while most studies have shown that IPC was associated with a decrease in ED, hospitalization, and avoidable hospitalizations, others have shown an increase or have been inconclusive.[30,95-98] One study in older adults comparing an IPC model to traditional practice found that patients in the IPC model had an 11% to 23% reduction in the incidence of ED visits, hospitalizations and avoidable hospitalizations compared to patients in traditional practice.[96] Inclusion of clinical pharmacists to a collaborative care–based IPC (i.e. patient-centered medical home) model was associated with a 23% reduction in hospitalizations.[95] Another study comparing pre- and post-utilization rates between team-based primary care and comparison practices found that team-based care was associated with a modest increase in ED and hospital use in the full patient sample, but with a 18% to 36% decrease among patients with 2 or more comorbidities.[97]

In Canada, studies in Quebec and Ontario have evaluated the impact of IPC reforms in the general or older population, with more positive evidence of impact in Quebec than Ontario. In Quebec, population-based studies have reported on the impact of Family Medicine Groups (*Groupe de médicine de famille*), the IPC model in Quebec, for older persons with chronic health conditions. Héroux et al. (2014) showed that enrolment into a Family Medicine Group caused a small reduction in ED visits for vulnerable older patients but no meaningful change in terms of hospitalizations.[34] A recent evaluation by Riverin et al. (2017) found similar results, with 4.2 fewer ED visits per 1000 hospital discharges among patients enrolled in a family medicine group compared to traditional primary care practices; findings in terms of hospital readmissions were

inconclusive.[35] Another study by these authors using the same older population found 25.1 fewer follow-up visits per 1000 discharges among patients in Family Medicine Groups versus traditional practice.[36]

In Ontario, the evaluation of IPC reforms has focused on the effect of FHTs in the general population. In 2014, the Conference Board of Canada (CBoC) released a report on a 5-year evaluation of FHTs commissioned by the Ontario Ministry of Health and Long-Term Care.[29] Using health administrative data provided by the Institute of Clinical Evaluative Sciences (ICES)[27], the CBoC report found that FHTs performed inconsistently compared to the other primary care models across different health service use indicators.[29] Table 2.2 is an extract from this report and summarizes these comparisons. FHTs had higher rates of ED use, 1-year readmission and visits to specialists than fee-for-service, higher ED use than FHOs but lower than FHNs. Differences in terms of avoidable hospitalizations were inconclusive in this report,[27,29] but another study found the risk of avoidable hospitalizations to be 6% higher in FHTs than in fee-for-service models.[30] Finally, a study comparing health care and primary care costs across Ontario primary care models found that patients in FHTs had higher total health care costs than other enrolment models.[94]

Table 2.2: Comparison of health service use across primary care models in Ontario (extract from the Conference Board of Canada report on the External Evaluation of the Family Health Team Initiative (2014))

Health Service Utilization: Comparison With FHTs, 2012

	CHC	EFFS	FFS	FHN	FHO
Emergency department visits per 100 population	+6.5*	-2.8	-6.5*	+5.8	-2.9*
Less urgent emergency department visits per 100 population	-2.3	-1.25	-1.6	+4.3	-1.6
Hospital admissions for four chronic conditions per 10,000 population	+45***	-5	-12	-1	-6
Hospital readmissions within 30 Days (%)	3.1%***	-0.2%	-0.5%	1.3%	0.0%
Hospital readmissions within one year (%)	4.9%***	-0.6%	-2.5%***	1.8%	-0.6%
Mean number of specialist visits per person	0.301***	0.003	-0.143**	-0.047	0.039

Comparator groups are: Community Health Centres (CHCs), Enhanced Fee-for-Service—Family Health Groups and Comprehensive Care Models (EFFS), Fee-for-Service (FFS), Family Health Networks that are not FHTs (FHNs), and Family Health Organizations (FHOs).
Note: Adjusted for rates at baseline, age, sex, income quintile, morbidity, comorbidity, rurality, and baseline rate. FHT is the comparison group, but FHT data are not included in the table because they were used as a reference point to report results for other models. Positive figures indicate higher amounts or percentages for the model in comparison to FHTs, while negative figures represent lower amounts or percentages for the model in comparison with FHTs.
*p<0.05, **p<0.01, ***p<0.005
Sources: ICES, 2012; FHT Patient Survey, 2012.

In terms of self-reported data on access and satisfaction with FHTs, patients and providers reported timelier and broader access to care in large part due to the availability of non-physician health professionals. More formalized chronic disease management plans and a focus on patient-centered care were also highlighted as strengths. A narrative synthesis of primarily qualitative studies of FHTs also revealed that patients and providers described better healthcare access, greater satisfaction, and enhanced quality of healthcare using a team-based approach; despite the degree of collaboration within FHTs considered to not have reached its full potential.[92] Overall, evidence to date on the effectiveness of IPC reforms on health service use in the general and older population has been mixed. FHTs, in particular, have been associated with higher health service use than other primary care models in Ontario.

2.4 Impact of interdisciplinary primary care on health service utilization in the dementia population

To our knowledge, no study has examined the impact of IPC reforms, including FHTs, on health service utilization in the dementia population. A limited number of intervention studies, mostly randomized controlled trials, have reported on the effect of enhanced IPC teams on health service use in persons with dementia.[99] These collaborative care interventions have consisted of IPC augmented with either a nurse navigator or case manager trained in dementia care, training for primary care providers, access to support by dementia medical specialists, and/or computer-based systems to facilitate assessments and referrals.[15,100-105] While these interventions have consistently shown improvements in clinical outcomes such as unmet needs, caregiver stress, patient quality of life and behavioral and psychological symptoms of dementia, evidence of their effect on health service use is scarce. In the United States, a trial of a telephone-based collaborative dementia care intervention consisting of a trained care team navigator, who provided education, support and care coordination with a team of dementia specialists (advanced practice nurse, social worker, and pharmacist) found the number needed to treat to prevent a single ED visit was 5; the effect on hospital use was inconclusive.[101] Three other studies with similar interventions failed to show improvements in ED or hospitalizations,[15,100,104] but two studies showed a delay in transition to long-term care.[100,105] In Ontario, Lee et al. (2010) demonstrated that implementation of collaborative memory clinics within the primary care practice can improve dementia care management.[103] A very recent unpublished provincial evaluation showed that patients enrolled within these interdisciplinary clinics were not visiting the ED as quickly and had a shorter length of stay in hospital than patients in traditional primary care. Patients receiving this enhanced IPC

approach also had a 30% higher increase in home care and a five-month longer delay, on average, in the time to long-term care admission.[106]

Overall, these findings suggest a positive effect of interdisciplinary primary care interventions in the care of persons living with dementia and possibly some aspects of health service use; however, population-wide evidence of the impact of system-wide IPC reforms, such as Family Health Teams, on health service utilization is still lacking.

2.5 Methodological challenges and opportunities in evaluating the impact of Family Health Teams

The limited and contradictory results of studies comparing FHTs to other primary care models may be partly due to inherent methodological complexities. In particular, the voluntary enrolment of physicians and patients into the various primary care models available create important challenges.

As physician and patient enrolment into FHTs was voluntary, the implementation of FHTs in the province represents a natural policy experiment. The lack of randomization, and therefore control over external factors, whether patient, practice or system-level factors, complicates the ability to reliably estimate the effect of this reform. Hutchison et al. (2011) cautioned that "the voluntary participation of patients and providers, and the confounding of primary care physicians' payment methods and organizational forms have made the evaluation of primary health care transformation challenging."[107] The results presented in Section 2.3 suggest that, while FHTs were designed to offer improved access, continuity and patient-centered care, patients enrolled in these models may in fact be consuming more health services than those in other primary care models. Authors of these studies have acknowledged that these findings may be partly artefactual and attributed to pre-existing differences in physician practices and patient case mix prior to

enrolment.[27,28,30,108] First, Glazier et al. (2012) suggested that given the voluntary enrolment of physicians into the various primary care models, payment incentives such as capitation and bonuses could have favored certain types of practices.[28] As capitation payments in Ontario are only adjusted for the age and sex of patients and not for comorbidity, physicians with more complex patients may have been less likely to switch into capitation-based models.[31,109] Second, physicians in capitation models are penalized when their patients seek primary care outside their practice, but not for ED use.[28] It may have, therefore, been more financially attractive for physicians in more rural areas, where there are fewer alternatives such as walk-in clinics or other primary care groups, to switch to a capitation-based model.[28] These reasons may partly explain why FHTs and other capitation-based models were found to have higher income patients, with fewer comorbidities, a greater rural population and higher ED rates than fee-for-service models.[28,31]

Lack of adequate control for these systematic differences threatens to bias the estimated effect of FHTs over other models and lead to misinformed policy decisions. More broadly, robust policy evaluation must be able to identify and control for variables that can contribute to confounding bias. Confounding occurs when variables that are common causes of both the exposure and outcome are not controlled for in the analysis, creating a spurious or biased association between the exposure and outcome. An understanding of the causal structure involved in the relationship between FHT affiliation and health system outcomes is critical to identify such sources of confounding and enable a robust evaluation. Statistical methods that are advanced enough to adapt to these complexities are needed to provide a valid assessment of the added-value of the FHT model over other models.

In order to attribute changes in health service use to the enrolment of dementia patients into a FHT, we ideally wish to know for every patient: *Would their health service use have been the same whether they were enrolled or not into the FHT?* While we can never observe both realities, advancements in causal inference have shown that, under certain specific conditions (for instance that all confounders be known and measured), the population-level causal effect of natural experiments or policies, can still be reliably estimated.[32,33,110] This causal inference framework formalizes the once vague concept of causality, using explicit mathematical notation to define and address causal concepts.[110] This development represents a paradigm shift in how we approach causal inference, opening up the possibility to attribute such claims to nonrandomized, observational data.

Methods developed under this causal framework are increasingly being used in other fields, including pharmacoepidemiology and health economics, but continue to be underutilized in primary care and health services research.[110] A primer which I developed on causal inference including practical applications was recently published as a methods brief within the Family Practice journal (Appendix B).[110] To our knowledge, only a few studies have employed causal inference methods to study primary care reform in Canada.[34-37] These studies showed that using these methods for primary care policy evaluation based on observational data is both feasible and robust. Guidance and better knowledge translation are needed to make these methods more accessible and practical for clinicians and researchers to use in mainstream primary care research.

Finally, health administrative data is one source of observational data that provides a unique opportunity to study effect of policies at a population-level using causal inference methods. This is especially true in the dementia population for whom national health survey data is limited and biased towards persons at early stages of the disease who can offer informed consent (e.g. the

Canadian Community Health Survey). While the use of administrative data is limited to what is routinely collected, the large number of linked databases and data sources increase the potential for confounding control. Moreover, as data is available on the near totality of the population of interest, sampling or selection bias is minimized.

The need for rigorous evidence-based knowledge on the impact of FHTs and other health reforms has been echoed in several reports.[8,27,28,77,111-113] There is now an opportunity to leverage large observational data and advances in causal inference to gain clarity about the population level effect of such reforms.

3 CHAPTER 3: KNOWLEDGE GAPS AND THESIS OBJECTIVES

3.1 Knowledge gaps

As stated in previous chapters, dementia is a devastating and complex disease with a high and growing burden for patients, families and the health care system and requires integrated, person-centered and coordinated care. Interdisciplinary primary care (IPC) models have been introduced in the last two decades in many health jurisdictions and have been proposed as a potentially beneficial way to care for this vulnerable population. However, evidence on population-level changes in dementia management within primary care since the onset of IPC is lacking. Specifically:

Gap #1: It is currently unknown if and how health service use in the dementia population has evolved over time.

Gap #2: It is currently unknown whether access to an interdisciplinary compared to non-interdisciplinary primary care model affects how persons with dementia use health services.

Ontario's Family Health Teams are one of the largest examples of IPC in North America and offer a unique opportunity in which to examine dementia management within a population-level, primary care context.

For Gap #1, I sought to describe trends in health service use in the Ontario dementia population over the last two decades using health administrative data and whether trends were shifting more towards management in primary care. To this end, it was necessary to construct a framework of indicators that would be suitable to measure primary care performance and health

service in the dementia population. Moreover, as previously mentioned, men and women with dementia may have differential health system experiences due to variable patterns of longevity, availability of informal caregivers and sex-specific manifestations of disease. I addressed this neglected component of health service research in dementia by conducting a comparative analysis of trends for women and men.

For Gap #2, current literature on the evidence of team-based care in the other populations revealed that methodological complexities associated with determining an unbiased effect of this natural policy experiment were not fully accounted for. My research aimed to apply the latest in methods in causal inference to better account for these complexities. In addition, as these methods remain underutilized in health service policy research, I also sought to provide accessible guidance to non-methodologists on how such methods can be used and how they can improve the evaluation of non-randomized interventions such as health reforms.

3.2 Thesis objectives

Overall, this thesis is comprised of four interrelated studies which build upon each other to answer the following objectives:

Objective 1: To develop a comprehensive framework of population-based, primary care performance and health service use indicators adapted to dementia and identify a subset of key priority indicators

Objective 2: To describe sex differences in primary care performance and health service use over time in persons with dementia in Ontario

Objective 3: To provide guidance on the use of causal inference methods for appropriate confounder selection in the evaluation of natural policy experiments

Objective 4: To apply causal inference methods to estimate the effect of interdisciplinary versus non-interdisciplinary primary care on health service use for persons with dementia in Ontario

To answer objective 1, I developed a framework of primary care performance indicators, adapted to the dementia population and measurable using population-level administrative data. The manuscript has been submitted to the Canadian Journal of Aging (**Manuscript 1: Framework and prioritization of dementia primary care performance indicators based on health administrative data**).

To answer objective 2, I described trends in these indicators for the dementia population over the last 15 years using health administrative data in Ontario. The manuscript has been published in the JAGS (**Manuscript 2: Sex differences in dementia primary care performance and health service use: A population-based study**)

To answer objective 3, I presented an overview of shortcomings in the selection of confounders for the estimation of interventions or policies based on non-randomized data and how causal inference methods can be used to strengthen the assessment of confounding. The manuscript has been published in the CMAJ (**Manuscript 3: Testing group differences for confounder selection in non-randomized studies: flawed practice**)

To answer objective 4, building on the findings of the previous three studies, I conducted an evaluation of the effect of FHT affiliation on health service use for persons with dementia in Ontario using causal inference methods. The manuscript is in preparation and will be submitted to the JAMA (**Manuscript 4: Comparison of interdisciplinary versus non-interdisciplinary primary care on emergency department and hospital use in persons with dementia: A population-based study)**

4 CHAPTER 4: FRAMEWORK AND PRIORITIZATION OF DEMENTIA PRIMARY CARE PERFORMANCE INDICATORS BASED ON HEALTH ADMINISTRATIVE DATA (MANUSCRIPT 1)

4.1 Preamble

Health systems in Canada and elsewhere continue to undergo significant reforms especially with regards to efforts to reinforce the role of primary care within the health system. Monitoring primary care and health system performance using readily-available indicators for persons with dementia who represent a growing cohort of vulnerable high users of the health system is important to ensure timely health policy decision-making.

In this first of four manuscripts, I developed a framework of primary care performance and health service use indicators adapted for persons with dementia and measurable using routinely available health administrative data. I also created and disseminated a survey to stakeholders involved in the provincial council for the ROSA research team (Appendix C) to identify a subset of priority indicators based on the framework. This framework served as the basis for my descriptive assessment of trends in primary care and health system performance for men and women with dementia in the second manuscript. This manuscript was submitted for publication within the Canadian Journal of Aging.

Title: Framework and prioritization of dementia primary care performance indicators based on health administrative data

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4.2 Abstract

OBJECTIVES: Given the growing number of persons with dementia managed in the community, indicators to measure the performance of primary care and health service use in this population using routinely collected data are needed in order to support dementia capacity planning. This study aimed to 1) develop a comprehensive framework of population-based, primary care performance and health service use indicators adapted to dementia and 2) identify a subset of stakeholder-driven priority indicators.

DESIGN: Framework development was carried out through the selection of an initial framework based on a rapid review and identification of relevant indicators and enrichment based on existing dementia indicators and guidelines. Prioritization of indicators was carried out through a stakeholder survey.

SETTING: Persons with dementia living in the community.

PARTICIPANTS: 109 stakeholders including clinicians, patients/caregivers, decision-makers/managers from three Canadian provinces (Ontario, Quebec, New Brunswick).

MEASUREMENTS: Primary care performance and health service use indicators.

RESULTS: Our framework comprised 37 indicators across eight domains of performance (Accessibility, Integration, Effectiveness, Efficiency, Equity, Safety, Population Health and Patient-Centeredness). Continuity of care, early stage diagnosis and access to home care were consistently rated as priorities. Equitable care was a specific priority among patients/caregivers; clinicians reported avoidable hospitalizations as among their priorities.

CONCLUSION: This comprehensive framework could set a foundation for the ongoing surveillance of trends and evaluation of health policies for persons with dementia at a population-level.

4.3 Introduction

Primary health care is seen by many as the way forward in managing the growing number of persons with dementia.[1-6] Moreover, with over 60% of persons with dementia cared for in community settings, a proportion expected to increase in the coming years, providing high-quality primary care for persons with dementia is increasingly being recognized as a public health priority.[7,8] In light of this changing landscape in dementia care, establishing appropriate indicators of the performance of the primary health care system and use of services in this population is essential to monitor and evaluate the impact of ongoing primary care reforms and dementia strategies. Rapid and objective measurement using routinely collected population-based data is also needed to equip health policy makers with timely evidence-based knowledge on areas of success and improvement and to facilitate comparisons and learning across jurisdictions. Indicators to measure the state of primary care for this population, however, remain scarce.

On the one hand, several general frameworks have been proposed to measure the performance of primary care.[9-11] These overarching frameworks aim to measure performance for the population at large and as such, it is currently unclear if these frameworks: 1) are applicable or appropriate for persons with dementia, 2) incorporate established indicators relevant to dementia and of importance to stakeholders and 3) can be measured at a population level using routinely collected data. On the other hand, a small number of studies have proposed or assessed quality indicators specific to the management of dementia within primary care,[12-14] but included a limited number of indicators, were based on local guidelines or targeted processes of care measurable through patient charts and therefore not readily measurable at a population level.

Given the growing role of primary care in the care of the dementia population, a formal elaboration and prioritization of indicators that can be used to measure and compare the quality of

primary care in persons with dementia is needed.[15] This study aimed 1) to develop a comprehensive framework of primary care performance and health service use indicators measurable using administrative data for persons with dementia and 2) to identify stakeholder-relevant priority indicators for ongoing surveillance and policy evaluation in this population.

4.4 **Methods**

4.4.1 Framework development

Our development of a primary care performance and health service use measurement framework for dementia was carried out in two steps: 1) the selection of an initial framework and 2) indicator identification and enrichment based on dementia guidelines.

4.4.1.1 Selection of an initial framework

A rapid review within published and grey literature of frameworks on health system or primary care performance was conducted.[16] This streamlined review strategy was selected to ensure timely knowledge transfer of our framework and indicators to stakeholders (decision makers including ministry representatives, clinicians, managers and patient/caregiver representatives) involved in the Research on Organization of Healthcare Services for Alzheimer's (ROSA), a pan-Canadian research team within the Canadian Consortium of Neurodegeneration in Aging.[17,18]

The literature search was performed using the MEDLINE (Ovid) and EMBASE databases with assistance from a specialized health librarian. The following concepts were used: "primary healthcare", "performance", "framework or indicators" (Supplementary Text S1). The search was limited to publications written in English between 2008 and 2018. The lower date limit was selected based on the publication of the Institute for Health Care Information (IHI)'s Triple Aim Initiative, which constituted a seminal work on the elaboration of frameworks in primary care.[10]

Grey literature was also searched by reviewing reports and recommendations from international, national and state/provincial institutes focused on health system performance and quality such as the IHI and Organisation for Economic Co-operation and Development (OECD).[19] The record selection was done by one reviewer (N.S.). A backward citation tracking approach and expert consultation was also used to ensure any other potentially relevant frameworks were also included in the review. The following exclusion criteria were applied to the identified records: 1=not a primary care framework (either a framework not including primary care or any set of indicators without an underlying framework), 2= framework entirely disease or population-specific aside from dementia (e.g. cancer, diabetes, maternal/child health), 3=framework not developed in the United States or Canada, 4=framework not operationalized or 5=framework indicators not potentially measurable using administrative data.

Of the eligible frameworks identified, an assessment of their relevance was done independently by two reviewers (N.S. and M.H.), disagreements were resolved with one verifier (I.V.).[20] Data on the following categories were extracted: comprehensiveness of the framework in terms of the number of performance domains/indicators, focus of the framework on patient-level indicators (rather than practice-level or system-level), pertinence of the framework indicators to the dementia population, and measurability using administrative data. All four categories were scored on a scale of 1 (low) to 4 (high). The framework with the highest overall score was selected.

4.4.1.2 Indicator identification and enrichment

Following the selection of a framework, an assessment of the appropriateness and feasibility of indicators within the framework was conducted by a panel of eight experts from our research group consisting of family physicians, geriatricians, primary care and health service researchers, and biostatisticians. Indicators were retained based on the following inclusion criteria: 1)

appropriate for an older population/dementia population; 2) not specific to a single disease or subgroup other than dementia (e.g. cancer, diabetes); 3) measurable at the patient-level (as opposed to practice-level or system-level); and 4) potentially measurable using routinely collected administrative data.

We also enriched the framework with indicators specific to dementia care based on current dementia care guidelines, quality indicators or reports from the US and Canada. Dementia care guidelines included those by the Alzheimer's Association,[21] American Geriatrics Society[22] and the Canadian consensus conferences on the diagnosis and treatment of dementia.[1] Dementia care quality indicators included indicators developed by the Dementia Measures Work Group (DWG),[23] Assessing Care of Vulnerable Elders (ACOVE),[24-27] and the Health Quality Ontario dementia care quality standards.[14] Three other sources were also considered: one on population-based quality of dementia care indicators,[12] and two systematic reviews.[28,29] We assessed the eligibility of the dementia-specific indicators based on the same inclusion criteria used for the general indicators.

4.4.2 Stakeholder prioritization

A survey including the complete set of indicators was distributed to key stakeholders within our stakeholder council group including clinicians, patient/caregiver representatives from the Alzheimer Society, managers and government representatives from three Canadian provinces (Ontario, Quebec and New-Brunswick). Stakeholders were asked to identify key priority indicators among the set presented. A snowball sampling technique[30] was used whereby participants also distributed the survey to colleagues within their own organizations. The survey was pre-tested with three stakeholders for clarity and face validity. Based on feedback obtained during the pre-test,

indicators measuring a similar concept were omitted from the final survey for clarity in identifying areas of priority.

Demographic information on province, type of stakeholder, age group and sex were collected. The relative frequency to which each indicator was selected as a priority was calculated. Indicators selected by at least 60% of stakeholders were considered as priority indicators. Cross-stakeholder comparisons in priority indicators were tabulated to determine common and stakeholder-specific priorities.

This study was approved by the Research Ethics Board of the Integrated Health and Social Services University Network for West-Central Montreal, Canada.

4.5 Results

4.5.1 Framework development

The literature review yielded a total of 358 peer-reviewed citations and 14 additional records obtained through backward citation searches, grey literature and expert consultation (Supplementary Figure S2). After duplicates were removed, 336 records were screened for eligibility. Of these, 274 were excluded because they either did not refer to a primary care framework (n=152), were entirely disease or population specific (e.g. diabetes) (n=89) or were not developed in the United States or Canada (n=33).

The full-text articles of the remaining 63 records were assessed. Of these, 16 references, referring to 12 distinct operationalized performance or quality indicator frameworks in primary care, were considered eligible (Supplementary Table S3).[11,14,15,19,31-42] Eligible frameworks included those developed by well-recognized organizations or institutions such as the Organisation for Economic Co-operation and Development (OECD), Agency for Health Research and Quality

(AHRQ), Institute for Health Care Information (IHI)'s Triple Aim Initiative, Canadian Institute for Health Information (CIHI), and Health Quality Ontario (HQO).

Among these eligible frameworks, the HQO Primary Care Performance Measurement Framework was rated highest across the categories assessed (Supplementary Table S3). It was comprehensive in terms of the number of performance domains/indicators (with nine domains of performance and 199 unique indicators), aligned with seminal theoretical frameworks including the IHI[31], focused mainly on patient-level indicators and included a large number of indicators pertinent to the dementia population and measurable using administrative data.

Eighteen (18) of the HQO indicators across eight domains of performance (Accessibility, Integration, Effectiveness, Efficiency, Equity, Safety, Population Health and Patient-Centeredness) were retained based on the inclusion criteria. Excluded indicators were mainly self-reported survey indicators for which there could be no equivalent health administrative measure (e.g. "*percentage of patients who report that their family physician/nurse practitioner involves them in as much as they want in decisions"*) and indicators referring to specific disease sub-cohorts other than dementia (e.g. diabetes, hypertension, cancer) or younger populations (children, perinatal health).

Nineteen (19) indicators from the dementia indicators and guidelines assessed met the inclusion criteria and were added to the framework (Figure 4.1).[1,12,14,21-29] These indicators included prescriptions for dementia medication, requests for blood tests, referrals to specialists and place of death. Indicators were matched to the most relevant domain within the framework.

Our final framework included 37 indicators (Figure 4.1).

Access	Integration	Effective Care	Efficient Care	Population Health	Safety	Patient-Centered Care
Access to a regular family doctor (or nurse practitioner)[a]	Continuity of care[a]	Diagnosis at an early stage of the disease[b]	Annual cost of health services[a]	Yearly flu shot (immunization for influenza)[a]	Having a high number of medications[b]	Access to counselling for patients[b]
After-hours access to the regular family doctor[a]	Phone calls between the regular family doctor and specialists[a]	Dementia diagnosed by the regular family doctor[b]	Duplicate medical tests (e.g. blood tests, brain CT / MRI scans)[b]	Pneumococcal immunization[a]	Potentially inappropriate prescriptions for medications (e.g. benzodiazepines, antipsychotics, anticholinergics, tricyclic antidepressants, trazodone) associated with serious side effects[a]	Access to counselling for caregivers[b]
Access to an interprofessional primary care team[a]	Hospitalizations[b]	Requests for blood tests originating from the regular family doctor[b]				Access to home care[b]
Visits to the regular family physician[a]	Length of time spent in hospital in the year following diagnosis of dementia[b]	Medications prescribed for dementia (e.g. Exelon®, Reminyl®, Aricept®, Ebixa®)[b]				Access to long-term care[b]
Physician specialty associated with the largest proportion of visits[b]	Discharge delay[b]	First medication for dementia prescribed by the regular family doctor[b]				Access to palliative end-of-life care[a]
Visits to the ED[b]	Avoidable hospitalizations[a]	Annual visit to the regular family doctor[a]				Number of days spent in hospital in last 3 months of life[b]
Non-urgent visits to the ED[a]	Visit to the regular family doctor within 7 days following a hospitalization[a]	Referrals to specialists in dementia (geriatrician, neurologist, psychiatrist) originating from the regular family doctor[b]				Dying at home[b]
	Readmission to the hospital within 30 days following a hospitalization[a]	Referrals to other specialists (e.g. cardiologist, oncologist) originating from the regular family doctor[b]				
Equity						
Equitable care across all patients (e.g. age, sex, income, region, immigrant status) [a]						

Figure 4.1:Framework of primary care performance and health service use indicators in dementia:

Footnote: [a] Indicator derived from the Health Quality Ontario (HQO) Primary Care Measurement framework; [b] Indicator derived from dementia-specific literature and expert opinion. ED: Emergency Department

4.5.2 Stakeholder prioritization

One-hundred and nine (109) stakeholders completed the survey. Participants were evenly distributed across the three provinces and were predominantly women, English-speaking and between 35 and 65 years of age (Table 4.1). Half of the participants were clinicians; nearly 20% were patient/caregiver representatives, 13% were managers and 8% were government representatives (Table 4.1).

Table 4.1: Characteristics of survey participants (N=109)

Characteristics	**N (%)**
Female	79 (74.5%)
English	71 (67%)
Age Group	
Less than 35	14 (13.2%)
35-44	31 (29.2%)
45-54	30 (28.3%)
55-64	24 (22.6%)
65 and over	7 (6.6%)
Missing	3 (2.8%)
Province	
New-Brunswick	39 (36.8%)
Quebec	38 (35.8%)
Ontario	31 (29.2%)
Saskatchewan	1 (0.9%)
Stakeholder group	
Clinician	54 (50.9%)
Patient / caregiver	20 (18.9%)
Manager	14 (13.2%)
Government representative	8 (7.5%)
Other	13 (12.3%)

The most frequently prioritized indicators among all participants were *access to a regular primary care provider* (81.7%), *continuity of care* (77.1%), *access to home care* (75.2%), *early stage diagnosis* (71.6%) and *avoidable hospitalizations* (63.3%) (Figure 4.2).

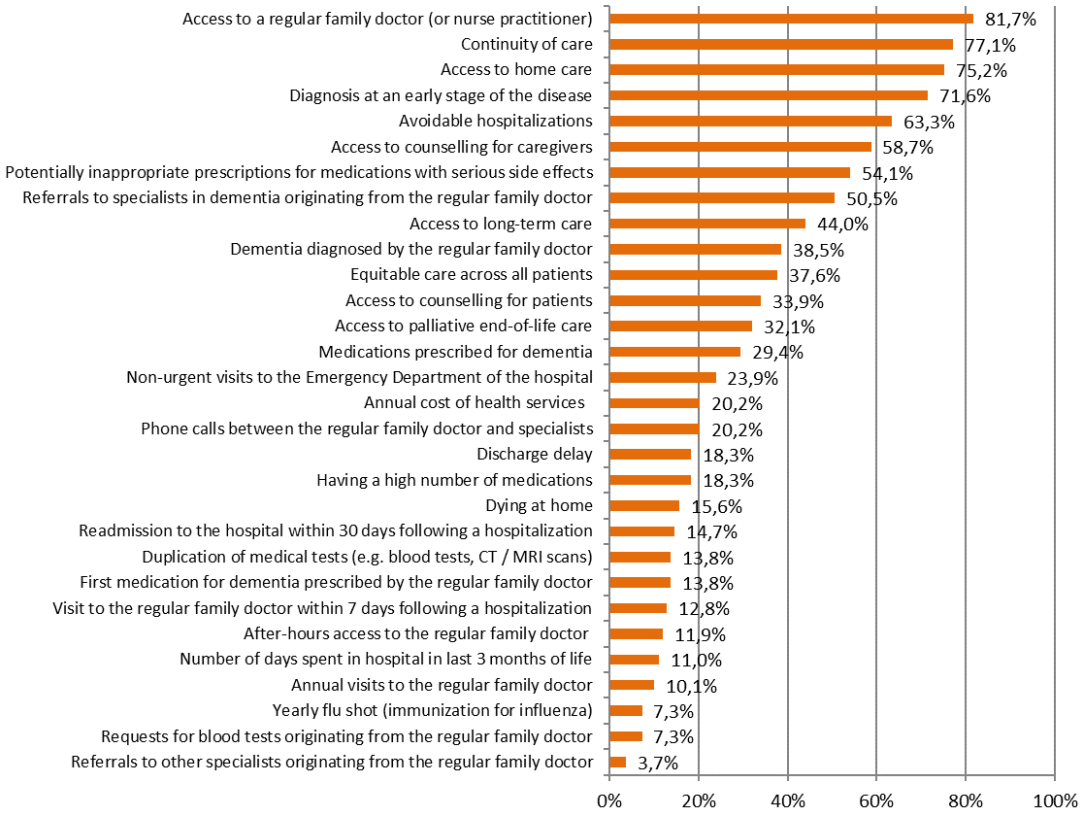

Figure 4.2: Proportion of stakeholders considering each indicator as a key indicator

Footnote: Based on pre-test feedback, seven indicators (access to an interprofessional primary care team, visits to the regular family physician, physician specialty associated with the largest proportion of visits, visits to the ED, hospitalizations, discharge delay and pneumococcal immunization) were considered similar in concept to other indicators and omitted from the stakeholder survey for clarity in identifying areas of priority)

Comparing priorities across stakeholder groups, we found that *continuity of care*, *early stage diagnosis, access to home care* were common priorities among all groups (Table 4.2). *Avoidable hospitalizations* were a greater concern among clinicians while *equitable care* was considered a higher target indicator by patient and caregiver representatives. Finally, government representatives rated referrals to specialists in dementia by the regular doctor among their priorities.

Table 4.2: Agreement across stakeholders

	Clinicians (n=54)	Government (n=8)	Managers (n=14)	Patient / caregiver representatives (n=20)	Others (n=13)	Overall (n=109)	Agreement
Continuity of care	x	x	x	x	x	x	5/5
Diagnosis at an early stage of the disease	x	x	x	x	x	x	5/5
Access to home care	x	x	x	x	x	x	5/5
Access to a regular family doctor (or nurse practitioner)	x		x	x	x	x	4/5
Access to counselling for caregivers		x	x	x	x		4/5
Potentially inappropriate prescriptions for medications	x		x	x			3/5
Avoidable hospitalizations	x				x	x	2/5
Referrals to specialists in dementia by regular doctor		x					1/5
Dementia diagnosed by the regular family doctor					x		1/5
Equitable care across all patients				x			1/5

4.6 **Discussion**

We developed a comprehensive framework of 37 population-based, primary care performance and health service use indicators across eight domains of performance adapted to the dementia population. By leveraging an existing validated primary care performance measurement framework as well as quality indicators in dementia care, we arrived at a set of indicators that would be both relevant to primary dementia care and measurable in routinely collected population-level data to support monitoring of dementia care performance over time and across care models and jurisdictions.

Our literature review confirmed the paucity of dementia indicators in current primary care performance frameworks. Current general primary care performance frameworks did not capture specificities around dementia care, limiting their use and scope. Among the operationalized primary care frameworks reviewed, while most contained indicators on the prevention or management of chronic diseases such as diabetes, cancer and hypertension, indicators relating to dementia were generally absent. Even within the HQO primary care framework that we assessed as most appropriate and relevant to use as a basis, only one of the original HQO indicators was directly focused on dementia care (proportion of patients with dementia receiving an annual follow-up).[37] This is consistent with reports citing the lack of consideration of dementia as one of the most important and common chronic diseases among older adults and its lack of representation from studies and discussions on chronic disease management.[43,44] Given that 1 in 5 baby boomers will live with dementia[45] and the high impact of this disease on patients, families and the health system, this substantive gap highlights the importance of bringing dementia management to the forefront of chronic disease management in primary care.

Within the dementia literature, proposed measures and guidelines such as those developed by ACOVE[24-27] and the Dementia Measures Work Group[23] primarily focused on process of care indicators assessed through chart review. These indicators provide a critical component of the evaluation of dementia care and allow for a detailed breakdown of the clinical management of dementia such as assessments of functional ability, driving, medication and caregiver status. Such an evaluation, however, requires primary data collection through chart abstraction and therefore cannot be easily scaled up a population-level. We sought to develop indicators that could be measured using routinely-collected health administrative data. These data are readily available and relatively inexpensive to access, compared with clinical data sources.[15] As dementia plans and initiatives continue to be implemented in many jurisdictions, the ability to analyze the effect of these policy changes using readily-available indicators is necessary to ensure efficient and timely decision-making.

Among the few population-based indicators proposed within the dementia literature, these frameworks either lacked a theoretical framework, were limited in scope or based on local guidelines.[12,14] We found only one primary dementia care framework, the HQO quality dementia standards. This framework, however, proposed only five indicators measurable using population-level data and related to only general health service use indicators such as emergency department visits and hospitalizations. Our framework provides an extension to this by considering a rich number of primary care specific and dementia-relevant indicators across several domains of performance.

While we strove to develop a comprehensive set of indicators, we also considered the value in prioritizing a subset of these indicators that would be feasible to be measured on an ongoing basis. Our stakeholder consultation allowed us to get a "pulse" on what end users wanted to know

and measure. The snowball technique for distribution of the survey also allowed us to engage with a broader network of key stakeholders and facilitated wider dissemination of our findings.[46] Among the priorities identified, continuity of care was unanimously reported as a key indicator of performance among all stakeholders. This finding is consistent with an international comparison of primary care performance indicators which found that family physicians perceived continuity of care as the most important dimension of quality of care.[41] Stakeholder-specific priorities were also brought to light. Perhaps not surprisingly, we found that clinicians rated avoidable hospitalizations as a priority, while patients and caregivers rated equitable care as one of their main concerns. A higher proportion of government representatives rated referrals to specialists in dementia by the regular doctor as a priority compared to other stakeholder groups. These differences point to underlying targets and goals specific to individual stakeholder groups. These identified contrasts also highlight the importance of involving all types of stakeholders that stand to benefit from the research within the research process and knowledge translation and exchange activities[47] to ensure that all priority lenses are included, not just those in common.

Several limitations should be acknowledged. First, although the rapid review provided a robust and methodologically sound approach to conducting the literature review, its streamlined approach may have caused some eligible frameworks to be missed. However, the use of a backward citation tracking approach and expert committee allowed us to mitigate this risk. The choice of indicators was also limited to variables that could be measured at a population-level using routinely-collected health administrative. As such, many self-reported or process-based indicators that were not adaptable into objective measures were not included in our framework. It would therefore seem advisable that any in-depth evaluation of primary dementia care performance be complimented with other data sources such as patient records in order to provide

these additional perspectives when needed.[15] In addition, other jurisdictions may have access to other routinely collected data, for example, nurse practitioner visits. In such cases, this framework could be expanded to include additional indicators as new data sources become available.

As dementia care continues to shift from secondary to primary care, routine monitoring of relevant and targeted indicators will become increasingly important. This is especially true given the development of national and sub-national dementia plans and strategies that are rooted in primary care.[48] A thorough description of care trajectories in this population, including gaps and inequities, will help orient the efforts of policy-makers in developing policies for the growing dementia population. In the companion joint article (reference to joint submission), we operationalized this framework using health administrative data and studied sex differences in trends in the performance indicators over a 12-year period. The identification of priority indicators will also help policy makers, program evaluators, and researchers narrow targets for evaluation. Overall, this framework will create a foundation for the surveillance of trends in the management of dementia in primary care at a population level and support the identification and scale up of policies and programs with the most potential to optimize care in this vulnerable population.

4.7 Acknowledgments

Conflicts of interest

The authors have no conflicts.

Author contributions

Nadia Sourial made substantial contributions to the conception and design, analysis, interpretation of data, and the drafting, revisions and final approval of the manuscript.

Claire Godard-Sebillotte contributed to the conception and design, interpretation of data, drafting, revision and final approval of the manuscript.

Susan E Bronskill contributed to the interpretation of data, revision and final approval of the manuscript.

Marine Hardouin made substantial contributions to the design, analysis, interpretation of data, and the drafting, revision and final approval of the manuscript.

Isabelle Vedel contributed to the conception and design, interpretation of data, drafting, revision and final approval of the manuscript.

We would also like to acknowledge the key input of experts, Dr. Howard Bergman and Dr. Tibor Schuster, in assisting in the selection of relevant and feasible indicators for our framework. We would also like to thank Dr. Vladimir Khanassov, Dr. Dan Murphy and Ms. Beatrice Débarges for their time and input in pre-testing the survey questionnaire. Finally, we express our sincere thanks to Ms. Genevieve Gore, research librarian, in helping to develop the search criteria for the rapid review. All persons who have contributed significantly to this work have been listed.

Sponsor's Role

This work was supported through Canadian Institutes for Health Research (CIHR) Vanier Canada Graduate Scholarships and a CIHR grant for the Canadian Consortium on Neurodegeneration in Aging, "Assessing care models implemented in primary health care for persons with Alzheimer's disease and related disorders" (2014-2019) (ID# CNA-137794). [17]

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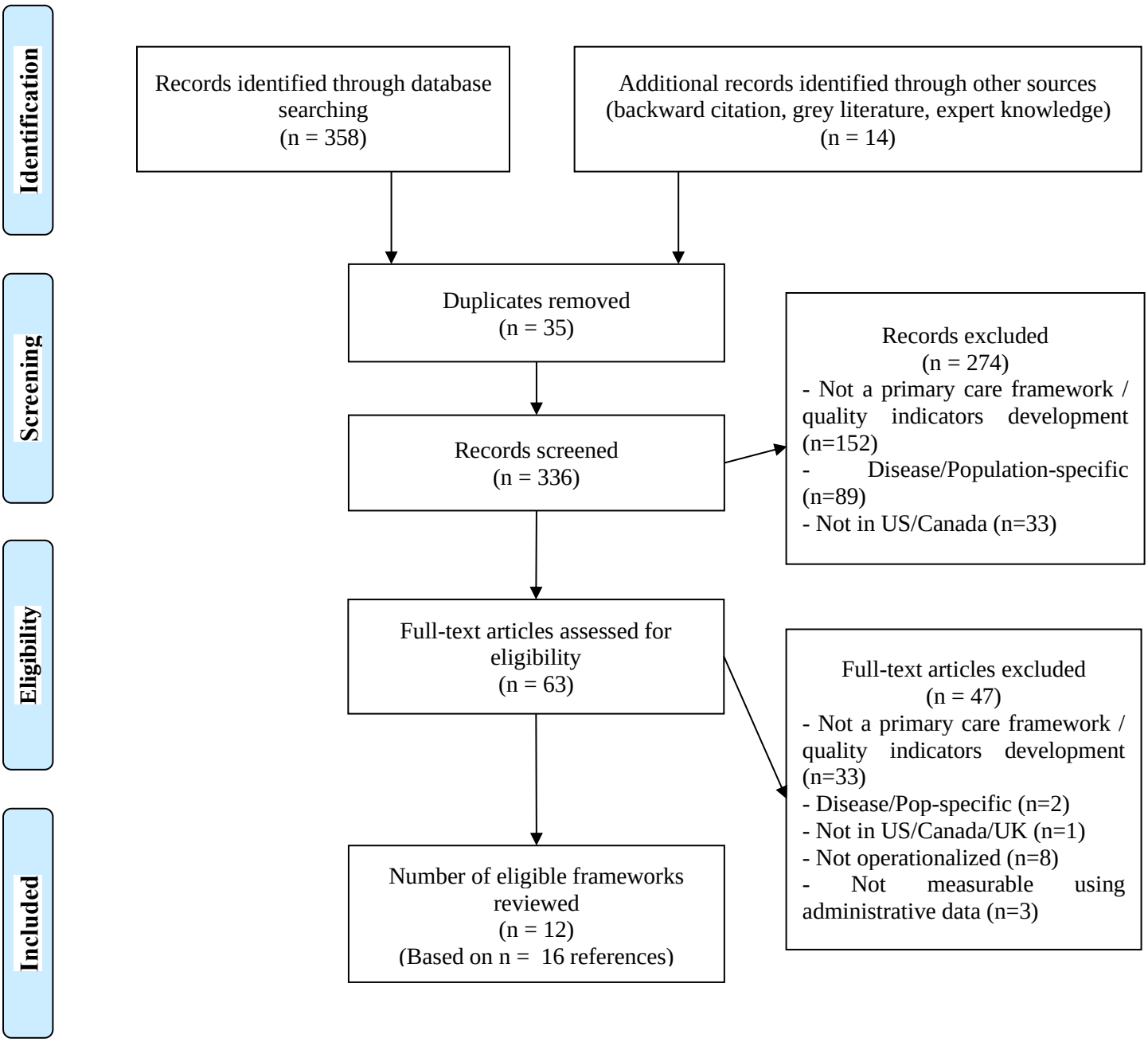
4.9 **Supplemental information**

4.9.1 Supplementary Text S1: Rapid review search strategy and results

Database: Embase <1996 to 2018 Week 09>
Search Strategy:
---
1 exp primary health care/ (125207)
2 (primary health care or primary healthcare or primary care).m_titl. (46293)
3 1 or 2 (130151)
4 performance measurement system/ (3858)
5 (primary care performance or health system performance or healthcare performance or health care performance or framework or indicators).mp. (334077)
6 (framework or indicators or performance).m_titl. (173590)
7 health care delivery/ (124000)
8 4 or 7 (127672)
9 3 and 5 and 6 and 8 (179)
10 limit 9 to (english and yr="2008 - 2018") (102)
****
Database: Ovid MEDLINE(R) Epub Ahead of Print, In-Process & Other Non-Indexed Citations, Ovid MEDLINE(R) Daily, Ovid MEDLINE and Versions(R) <1946 to February 21, 2018>
Search Strategy:
---
1 exp primary health care/ (134814)
2 (primary health care or primary healthcare or primary care).m_titl. (45109)
3 1 or 2 (148157)
4 Quality indicators, Health Care/ (13530)
5 (primary care performance or health system performance or healthcare performance or health care performance or framework or indicators).mp. (392530)
6 (framework or indicators or performance).m_titl. (204851)
7 "Delivery of Health Care"/ (78084)
8 4 or 7 (91018)
9 3 and 5 and 6 and 8 (423)
10 limit 9 to (yr="2008 - 2018" and english) (256)
****

4.9.2 Supplementary Figure S2: PRISMA flow diagram for selection of eligible framework



4.9.3 Supplementary Table S3: Scoring of eligible frameworks

Name of framework / Description of indicators	Comprehensiveness	Focus on patient-level	Pertinence for older population	Measurability in administrative data	Overall (out of 16)	Publication (First author; year)
	(1=low to 4= high)					
Primary care performance measurement framework (HQO)	4	3	3	3	**13**	HQO 2014[114], Haj-Ali 2017[115]
Toward a Primary Care Strategy for Canada (CFHI)	4	3	3	2	**12**	CFHI 2012[78]
Quality indicators of health system performance (IC/ES)	2	4	2	4	**12**	Stukel 2016[116]
Health System Performance Measurement Framework (CIHI)	4	2	2	3	**11**	CIHI 2012[117], 2016[118], Terner 2013[119]
Triple Aim Initiative (IHI)	3	3	2	2	**10**	Stiefel 2012[120], 2013[121]
Health Care Quality Indicators Project (OECD)	2	3	2	3	**10**	Carinci 2015[122]
Quality Standards for Dementia: Care for People Living in the Community (HQO)	2	3	3	2	**10**	HQO 2018[123]
Performance of primary healthcare Organizations (INSPQ)	3	2	2	3	**10**	Levesque 2010[124]
AHRQ Prevention Quality	2	3	1	3	**9**	AHRQ 2018[125]

Indicators (AHRQ)						
Primary health care performance measures (British Columbia U)	3	2	2	2	**9**	Broemeling 2009[126]
Quality Book of Tools (McMaster U)	3	1	2	1	**7**	Levitt 2014[127]
Process quality indicators in family Medicine (QUALICOPC study)	2	1	1	1	**5**	Pavlic 2015[128]

Abbreviations: IHI, Institute for Healthcare Improvement; OECD, Organization for Economic Cooperation and Development; AHRQ, Agency for Healthcare Research and Quality; CFHI, Canadian Foundation for Healthcare Improvement; CIHI, Canadian Institute for Health Information; HQO, Health Quality Ontario; INSPQ, Institut national de santé publique du Québec; QUALICOPC, Quality and Costs of Primary Care in Europe

5 CHAPTER 5: SEX DIFFERENCES IN DEMENTIA PRIMARY CARE PERFORMANCE AND HEALTH SERVICE USE: A POPULATION-BASED STUDY (MANUSCRIPT 2)

5.1 Preamble

Since the early 2000's, the province of Ontario introduced a number of changes to the primary care system including new physician payment models and funding to support physicians in transitioning from physician-based practices to interdisciplinary models as well as investments in home care. It is currently unknown, however, whether dementia management in primary care and health service use for persons with dementia has evolved over time since the introduction of these primary care reforms.

Based on the indicator framework developed in the first manuscript, the goal of this second manuscript was to operationalize indicators within the framework and describe population-level trends in primary care performance and health service use for persons with dementia in Ontario. Appendix D describes which subset of indicators within the framework were operationalized. At the same time, this manuscript addresses a recent call to action on the lack of attention and critical importance of examining sex differences in dementia.[39] The need for sex and gender based evidence of differences in the clinical detection, diagnosis, management, and treatment of AD was identified as a priority area. Sex is also among indicators within the cross-cutting domain of equity in the framework developed in Chapter 4. Given the emerging importance of sex in dementia research, I, therefore, sought to apply an equity lens to this analysis and shed light on possible sex differences in these trends. I used data from ICES to compare population-level trends in men and women with dementia in Ontario over a 12-year period covering the period of primary care reform in Ontario. Results from this descriptive study helped to inform the design and methods for the

impact evaluation study in Chapter 7 focusing on the effect of Ontario's Family Health Teams on health service use in the dementia population. This manuscript has been peer-reviewed and resubmitted for publication in the JAGS on December 6, 2019.

Title: Sex differences in dementia primary care performance and health service use: a population-based study

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5.2 Abstract

OBJECTIVES: Growing evidence points to underlying sex differences in the risk factors and clinical presentation of dementia. It is unclear, however, whether sex differences also exist in the management and health care utilization of persons with dementia. We compared primary care performance and health service use indicators for newly identified men and women with dementia in Ontario, Canada over a 12-year period.

DESIGN: Population-based, repeated cohort study between 2002 and 2014.

SETTING: Ontario, Canada.

PARTICIPANTS: 318,350 community-dwelling adults aged 65 years old and older newly identified with dementia, followed for up to one year.

MEASUREMENTS: Eighteen indicators of primary care performance and health service use were assessed.

RESULTS: Approximately 60% of the study population were women. Few differences in the indicators were observed between sexes, although men had fewer diagnoses first recorded by the family doctor, more visits to non-cognition specialists, less use of home care, more hospitalizations and readmissions, and longer discharge delays. Most indicators remained relatively stable over

time for both men (median relative change: 13.7%; interquartile range (IQR): 4.5% to 29.7%) and women (median relative change: 15.7%; IQR: 5.9% to 31.5%). Notable improvements over time for both sexes included access to an interprofessional primary care team, use of home care and decreased use of long-term care. Areas of worsening included a higher occurrence of emergency department visits, lower continuity of care and longer discharge delays.

CONCLUSION: These findings raise awareness on the similarities and differences in management and health system use for men and women newly diagnosed with dementia, particularly the imbalance in hospital and home care use. As health systems continue to adapt to meet the needs of the growing dementia population, policy makers and clinicians should be mindful to develop care plans and interventions that consider the influence of sex on the need for services.

5.3 Introduction

Ensuring equitable dementia care and access to services, notably for both men and women, is an essential dimension of high quality primary care and health system capacity planning.[1] Among older adults, sex differences in health service use have been previously reported.[2-8] Older women are more likely to live alone, have fewer financial resources and less likely to receive informal care at home; factors which could create barriers to care.[5] Older men tend to seek care less often than older women.[2,7,8] Sex differences in patterns of use in other health services such as visits to specialists, emergency department (ED) use, hospitalizations and home and long-term care have been inconsistent.[3-6]

In persons with dementia, research on sex differences in the epidemiology, risk factors and clinical presentation of dementia is emerging;[9-12] however, potential sex differences in the management of dementia in primary care settings and the use of other health services continue to be a neglected component of dementia research.[9,13] Assessing whether dementia care is equitable across sexes is important as men and women with dementia may not navigate the health system in the same way due to differences in the risk factors for and presentation of disease, socioeconomic factors, social support such as the availability of caregivers, and patterns of longevity.[13] More evidence regarding sex-based differences in dementia management and health service use is needed to support decision-makers in appropriately adapting our health systems and has been called upon as a key priority.[13,14]

In addition, over the last two decades, many health jurisdictions have implemented policy changes to strengthen their primary health care systems in an effort to improve care for the increasing number of persons with multiple chronic conditions.[15-20] These changes have included the introduction of patient-centered medical homes, pay for performance incentives, formal patient

rostering and enhanced home care programs and may have impacted persons with dementia, who represent a vulnerable cohort that particularly stands to benefit from better accessible and integrated care. As these recent changes were implemented without an explicit focus on sex- and gender-equity, it is of interest to explore whether the management and health service use of men and women with dementia may have varied differently over time.

This study aimed to describe sex differences in primary care performance and health service use over time in newly identified persons with dementia using population-level data from Ontario, Canada.

5.4 Methods

5.4.1 Setting and data sources

Ontario, Canada's largest province, is home to 2.3 million adults 65 years of age or older.[21] The provincial health insurance plan is centrally managed and covers the majority of costs of care including physician visits, hospital services, medical tests and prescription drugs for older persons. This setting thus offers a unique opportunity to study sex differences at a population-level.

We used population-based administrative data held at ICES in Ontario. ICES is an independent, non-profit research institute whose legal status under Ontario's health information privacy law allows it to collect and analyze health care and demographic data, without consent, for health system evaluation and improvement. Data from several administrative databases were linked using unique encoded identifiers and analyzed at ICES (see Supplemental Text S1). These data sources have been used extensively in health services research.[22]

This study was approved by the Research Ethics Board of McGill University in Montreal, Canada. The use of ICES data in this project was authorized under section 45 of Ontario's Personal Health Information Protection Act, which does not require review by a Research Ethics Board.

5.4.2 Design and population

A repeated cohort design was used to create and follow yearly cohorts of community-dwelling older adults, aged 65 years and older, with a new diagnosis of dementia between fiscal years 2002 (April 1st 2002 to March 31st 2003) and 2014 (April 1st 2014 and March 31st 2015). A previously validated algorithm was used to identify new cases of dementia in each year (see Supplemental Text S1).[23] The date of dementia diagnosis identified from this algorithm was used as the index date for each individual in the current study. We excluded individuals with missing age, sex, health identification number, those not considered residents of Ontario, and/or those who were in a long-term care facility on the index date within each year.

Individuals were followed for up to one year after their index date. Yearly cohorts of newly identified persons with dementia were selected for three reasons: 1) disease management needs and health care utilization in the year following diagnosis is frequently high (second only to the year prior to death);[24] 2) service requirements in the first year post-diagnosis tend to be more focused on community-based care and be fairly homogeneous; and 3) independent yearly cohorts with disjoint follow-up data were required to provide an accurate population-level portrait of change over time.

5.4.3 Indicators of primary care performance and health service use

Eighteen indicators of primary care performance and health service use were operationalized and assessed (Table 5.1). These indicators have been shown to be important

markers of high quality primary and/or dementia care.[25-28] Operationalization of the indicators was done through a rigorous and iterative process with a panel of experts including family physicians, geriatricians, primary care and health service researchers, epidemiologists, biostatisticians and senior analysts.

5.4.4 Analysis

All analyses were performed separately for each cohort year (2002-2003 to 2014-2015) and stratified by sex. The number of newly identified cases of dementia was calculated in each cohort year. Crude and adjusted rates with accompanying 95% confidence intervals (CI) for all study indicators were calculated. Adjusted rates were illustrated graphically over time. Indicators assessed at baseline (on the index date) were adjusted for age. Indicators assessed over the follow-up period were adjusted for age and person-time in order to adjust for differences in follow-up time. Direct standardization for age was based on the age distribution from the most recent Canadian national census for the following age groups: 65-69, 70-74, 75-79, 80-84, 85-89, 90 and older.[29] Person-time for each cohort was calculated as the time from the index date of dementia diagnosis to either death, institutionalization or end of the one-year follow-up, whichever occurred first. Follow-up was discontinued after institutionalization as the goal of the study was to measure primary care performance and health service use in persons with dementia cared for in the community.

Two types of comparisons between men and women were performed: 1) cross-sectional comparisons and 2) comparisons of trends. Cross-sectional comparisons were based on the graphical illustration and absolute differences in the indicator rates in each year for men and women. Comparisons of trends for men and women were based on the graphical illustration and relative change in the indicators from 2002 to 2014.

Given the near complete population of newly identified persons with dementia in Ontario included in this study, sex differences in study indicators were assessed using a descriptive approach. The process was carried out in two steps: first and principally, through discussion with our expert panel of experts and second, through feedback on the presentation of our interpretations to stakeholders involved in our research team.[30] The stakeholder consultation was carried out during a stakeholder council meeting including 38 patient and caregiver representatives, clinicians, researchers, managers and decision-makers from three Canadian provinces (Ontario, Quebec and New Brunswick).[30,31] Given the large number of indicators, the expert consultation process required numerous discussions over the course of approximately 18 months. We used both the illustrative graphs and the numerical rates in each cohort-year to arrive at a consensus on the direction of the difference (not meaningfully different, different or inconsistent) or change (improving, worsening or inconsistent) for each indicator. In assessing cross-sectional differences, we considered as meaningfully different, indicators which showed a consistent pattern in the direction of the difference (always higher in men or always higher in women). Similarly, for the interpretation of trends, indicators such as visits to the regular family doctor which clearly and consistently increased or decreased over time were categorized as either improving or worsening. Indicators which had more variation were subject to further discussion in order to reach consensus. Stakeholders provided additional insight to corroborate the results found and were in agreement with the interpretations established with the expert group.

As a negative or positive value in the relative change could represent either worsening or improving depending on the nature of the indicator (e.g. a positive change in rate of visits to the family doctor would represent improving but a positive change in the rate of avoidable

hospitalizations would represent worsening), relative changes were reported in absolute value and illustrated through a bi-directional, clustered bar chart, stratified by sex.

All analyses were performed using SAS© software, Version 9.4, SAS Institute Inc., Cary, NC, USA.

5.5 Results

Overall, 318,350 persons newly identified with dementia between 2002 to 2014 were included in the study. In each cohort year, women accounted for approximately 60% of the population and were nearly two years older than men (Table 5.1). In both men and women, the average age increased by roughly one year over the 12-year period.

Table 5.1 Description of cohorts of newly identified cases of dementia from 2002 to 2014 in Ontario, Canada

	Women					Men				
Fiscal Year	**Population 65+ in Ontario**	**Newly identified dementia in fiscal year among 65+**	**Rate of new cases**	**Age**		**Population 65+ in Ontario**	**Newly identified dementia in fiscal year among 65+**	**Rate of new cases**	**Age**	
	N	N	Rate per 100 person-years	Mean	SD	N	N	Rate per 100 person-years	Mean	SD
2002	820558	12096	1.5	81.1	± 6.8	642144	7773	1.2	79.2	± 6.7
2003	832814	12440	1.5	81.1	± 6.8	655132	7906	1.2	79.3	± 6.7
2004	845580	12512	1.5	81.1	± 6.8	668648	8128	1.2	79.4	± 6.7
2005	858303	12644	1.5	81.0	± 6.8	682769	8347	1.3	79.4	± 6.6
2006	873868	13157	1.5	81.2	± 6.8	699769	8871	1.3	79.7	± 6.6
2007	889912	13419	1.5	81.2	± 6.8	716901	8921	1.3	79.6	± 6.7
2008	908050	14493	1.6	81.4	± 6.9	736676	9634	1.3	79.7	± 6.8
2009	931362	14706	1.6	81.5	± 6.9	761194	9706	1.3	79.8	± 6.8
2010	954282	14885	1.6	81.7	± 7.0	785805	10085	1.3	80.0	± 6.9
2011	978761	14795	1.5	81.9	± 7.0	810897	10349	1.3	80.2	± 7.0
2012	1019127	15207	1.5	81.9	± 7.2	850879	10528	1.3	80.3	± 7.1
2013	1059418	15001	1.4	81.9	± 7.3	889697	10801	1.2	80.3	± 7.1
2014	1094988	14172	1.3	82.1	± 7.5	924719	10069	1.1	80.5	± 7.2
Absolute change (2002 to 2014)	**274430**	**2076**	**-0.2**	**1.0**		**282575**	**2296**	**-0.1**	**1.3**	
Relative change, %	**33.4%**	**17.2%**	**-13.3%**	**1.2%**		**44.0%**	**29.5%**	**-8.3%**	**1.6%**	

5.5.1 Cross-sectional comparisons

Six of the 18 indicators showed consistent cross-sectional sex differences across cohort years (Figure 5.1). We found that men tended to have a lower occurrence of dementia diagnosis first recorded by the regular family doctor (34.3 per 100 person-years in men vs 38.9 per 100 person-years in women in 2014), more visits to non-cognition specialists (2.6 visits per person-year in men vs 2.1 visits per person-year in women in 2014), more hospitalizations (for example, in 2014, 18.3 per 100 person-years in men vs 15.6 per 100 person-years in women), longer discharge delays (40.8 days per person-year in men vs 35.6 days per person-year in women in 2014), more frequent readmissions to the hospital within 30 days following a hospitalization (18.4 per 100 person-years in men vs 12.9 per 100 person-years in women in 2014) and lower use of home care (51.2 per 100 person-years in men vs 52.9 per 100 person-years in women in 2014) (Figure 5.1, Supplemental File S2).

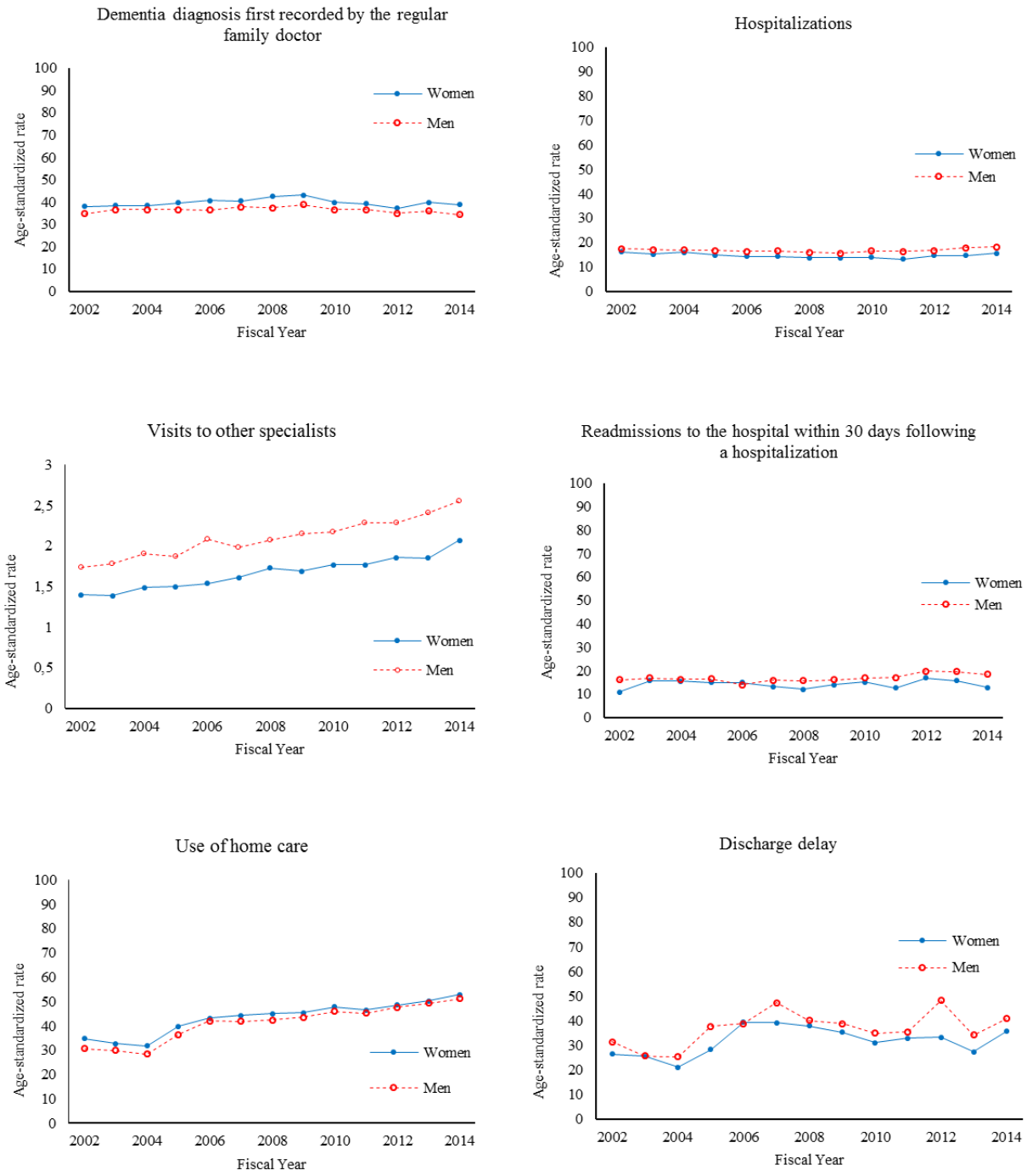

Figure 5.1 Age-standardized rates of primary dementia care and health service use indicators

Footnote: Operational definitions available in Supplementary Table S1.

No meaningful differences were observed by sex in half of the indicators examined (Supplemental Figure S1, Supplemental Table S2). Specifically, in each cohort year, there were no substantial differences in the rate of men and women with dementia in terms of having a regular family doctor, access to an interprofessional primary care team, medication dispensed for dementia, visits to the regular family doctor, visits to specialists in dementia care, continuity of care, visits to the emergency department and use of long-term care.

Sex differences in rates of avoidable hospitalizations, visits to the regular family doctor within 7 days following a hospitalization, and rate of persons dying at home were inconsistent across cohort-years (Supplemental Figure S1, Supplemental Table S2).

5.5.2 Comparison of trends

The relative change in the study indicators from 2002 to 2014 was comparable for men (median: 13.7%; IQR: 4.5% to 29.7%) and women (median: 15.7%; IQR: 5.9% to 31.5%) (Figure 5.2, Supplementary File S2).

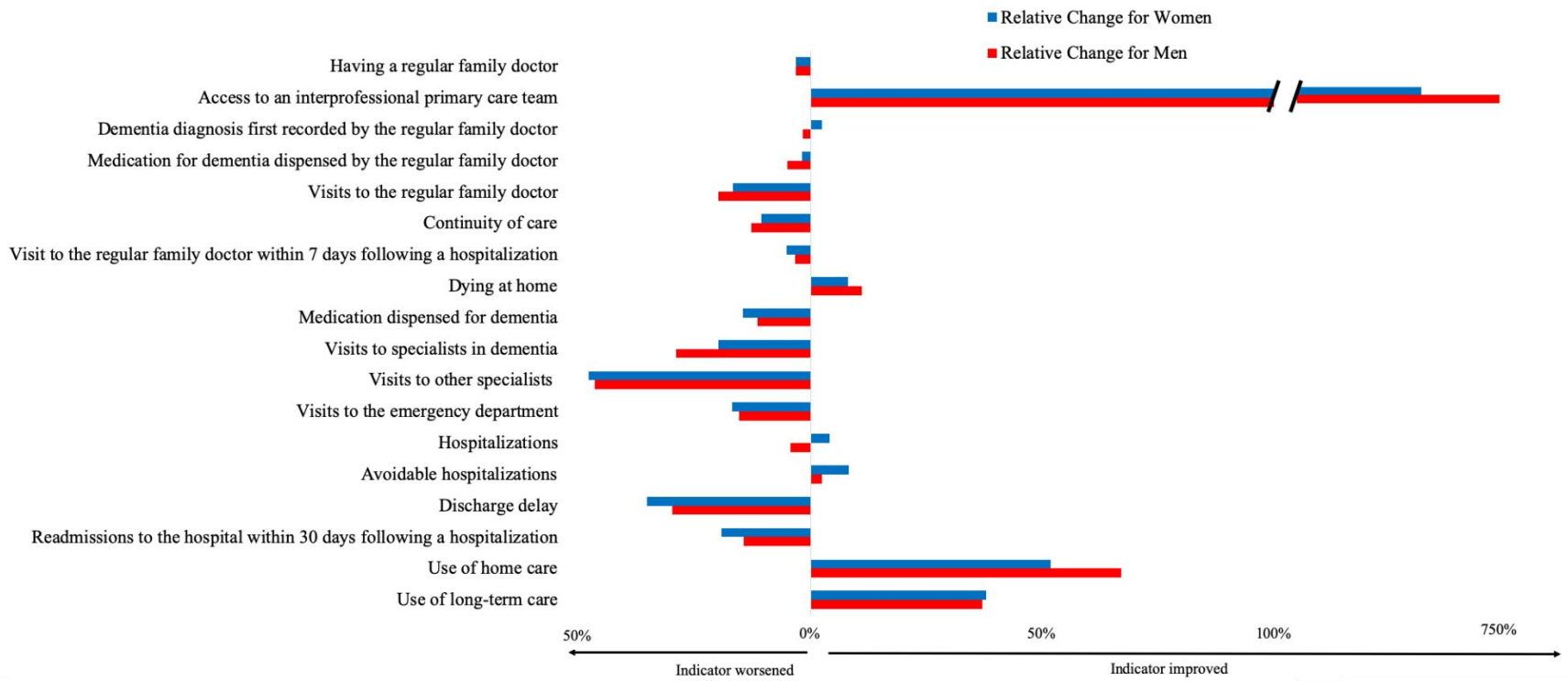

Figure 5.2 Relative change in primary care performance and health service use indicators from 2002 to 2014 for men and women with newly identified dementia in Ontario, Canada

Footnote: Operational definitions available in Supplementary File S1. As the sign of the relative change in the indicator could represent either worsening or improvement, relative changes are reported in absolute value.

Seven indicators showed no meaningful change over time in both men and women: having a regular family doctor, dementia diagnosis first recorded by the regular family doctor, visits to specialists in dementia care, overall and avoidable hospitalizations, visits to the regular family doctor within 7 days following a hospitalization and readmissions to the hospital within 30 days following a hospitalization. Six indicators worsened over time in both sexes: medication dispensed for dementia, visits to the regular family doctor, visits to non-cognition specialists, continuity of care, visits to the emergency department and discharge delay. Three indicators improved over time in both men and women: access to an interprofessional primary care team, use of home care and long-term care. Two (2) indicators had inconsistent trends in both men and women: medication for dementia dispensed by the regular family doctor and dying at home. (Figure 5.1, Supplementary File S1, Supplementary File S2)

5.6 Discussion

To our knowledge, this study is the first to present a comprehensive portrait of sex differences and trends in primary care performance and health system use in a population of persons with newly identified dementia. While there is a growing body of literature on sex-specific factors related to the risk and progression of dementia,[9-11] in our study, these underlying differences did not appear to translate into meaningful sex differences across many indicators. Moreover, despite system-wide primary care reforms over the last two decades more broadly, trends in primary care performance and health service use have remained mostly stable over time in both men and women with dementia.

Contrary to other reports in the general older population,[3,4,7,32,33] we did not find that women with dementia sought out ambulatory care more often than men in the year following diagnosis. There is little published evidence in the dementia literature with which to compare these

findings; however, one other study found similar results in a cohort of prevalent men and women with dementia.[34] The observed similarities in our study may reflect greater homogeneity in the disease management needs and health care utilization in the year following diagnosis than later in the disease process.[24] Availability of universal health care in Canada may have also facilitated equal opportunity in accessing care.[35]

Among the few indicators where sex differences were observed, we found that men more frequently used acute care hospitals in the year following dementia diagnosis while women used more home care services. These findings are in line with previous studies in the older and dementia population[3,36-38] and may reflect a number of underlying sex and gender-related factors including differences in the types and severity of comorbidities, functional ability and availability of family caregivers.[8,36,39-42] Men may require more adapted interventions or closer follow-up by a regular doctor to prevent acute exacerbation of their chronic conditions and potentially avoidable hospital use. In terms of home care, although women had consistently higher rates of home care use than men, it is still unclear whether the magnitude of the difference adequately portrays that demands for home care are adequately being met in both sexes, or whether there remain unmet needs. A study on gender differences in the availability of home care among persons with dementia found that perceived unmet needs for home care were twice as high among women than men.[34] A better understanding of home care needs in both sexes is needed to determine which services may still be needed to meet the demand and ensure equity.

In terms of trends over time, the observed increase in access to an inter-professional primary care team and use of home care in both men and women with dementia are consistent with system-wide shifts towards patient-centered medical homes and investments in home care, and also might partially explain the observed decrease in admissions to long-term care.[43-45] While the

interplay between these government initiatives and other competing forces make it difficult to claim a causal effect, it is possible that these system-wide efforts contributed to the trends observed.

Both men and women in our study experienced a decrease in continuity of care and increase in the rate of emergency department visits over time. Two studies exploring the association of continuity of care and health service use among persons with dementia found that higher levels of continuity were associated with a decreased risk of emergency department use.[50,51] Interventions targeting improved continuity of care in persons newly diagnosed with dementia may limit acute care hospital use in the year following diagnosis but the literature in this area remains scarce and requires further study.

5.6.1 Strengths and limitations

This study has important strengths. Our study explored a rich set of indicators providing a novel and comprehensive description of dementia management. Detailed operational definitions for the study indicators and hypotheses derived from these findings may help facilitate ongoing health services dementia research and identify areas of success, improvement and possible inequity that should be addressed in future programs and policy investments. Our use of repeated cross-sectional cohorts across multiple years allowed us to gage the consistency and reliability of our cross-sectional comparisons. The use of health administrative data in a universal health care system provided us with a unique opportunity to study potential sex differences for nearly 100% case ascertainment of the dementia population.

Some limitations should also be acknowledged. Population-level administrative data also did not allow for individual level assessment of appropriateness of care. For example, lack of dementia medication prescription or the decision to access long-term care may be appropriate in

some patients or based on shared decision making with the patient or family; visits to non-cognition specialists may also be appropriate in complex patients with serious comorbidities. Additional data sources such as patient surveys or records may be useful to compliment the objective indicators used to assess performance in this population. The observed decreases in primary care management may have also been partly linked to recent shifts in physician remuneration and task-sharing in team-based models which may have affected the reporting of services provided.[44,46] Finally, as this study was descriptive in nature, we could not separate how much of the observed changes in trends were attributed to secular versus dementia-specific changes or attribute causal interpretations to any observed sex differences.

5.7 Conclusion

Our results raise awareness on the similarities and differences in management and health system use for men and women newly diagnosed with dementia, particularly in the imbalance in hospital and home care use. That trends in many indicators of primary care performance and health service use have remained largely unchanged over time may point to the need for additional efforts to affect change in quality of care at a population level. As health systems continue to adapt to meet the complex needs of the growing dementia population, policy makers and clinicians should be mindful to develop care plans and interventions that consider the influence of sex and gender on the need for services.

5.8 Acknowledgements

5.8.1 Conflicts of interest

The authors have no conflicts of interest to report.

5.8.2 Author contributions

Nadia Sourial made substantial contributions to the conception and design, analysis, interpretation of data, and the drafting, revisions and final approval of the manuscript.

Isabelle Vedel contributed to the conception and design, interpretation of data, drafting, revision and final approval of the manuscript.

Claire Godard-Sebillotte contributed to the conception and design, interpretation of data, drafting, revision and final approval of the manuscript.

Jacob Etches contributed to the acquisition of data, analysis, interpretation of data, revision and final approval of the manuscript.

Genevieve Arsenault-Lapierre contributed to the conception and design, interpretation of data, drafting, revision and final approval of the manuscript.

Susan E Bronskill contributed to the conception and design, interpretation of data, drafting, revision and final approval of the manuscript.

We would like to acknowledge Dr. Tibor Schuster for his input on the design and analysis and Dr. Howard Bergman and Dr. Rick Glazier for their insight on the interpretation of the results. We also thank Ms. Mary Henein for creating and formatting all the tables and graphs in this manuscript. All persons who have contributed significantly to this work have been listed as authors.

5.8.3 Sponsor's role

This work was funded through Canadian Institutes for Health Research (CIHR) Vanier Canada Graduate Scholarships and a CIHR grant for the Canadian Consortium on Neurodegeneration in Aging (2014-2019) (ID# CNA-137794).

This study was supported by ICES, which is funded by an annual grant from the Ontario Ministry of Health and Long-Term Care (MOHLTC). Parts of this material are based on data and information compiled and provided by the Ontario MOHLTC, the Canadian Institute for Health Information, IMS Brogan Inc. and the Ontario Association of Community Care Access Centres. The analyses, conclusions, opinions and statements expressed herein are solely those of the authors and do not reflect those of the funding or data sources; no endorsement is intended or should be inferred. We thank IMS Brogan Inc. for use of their Drug Information Database.

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5.10 **Supplemental information**

5.10.1 Supplementary file S1: Operational definition for study indicators

Type of indicator	Indicator	Operational definition
Primary care	Having a regular family doctor	Rate of persons with dementia with access to a regular family doctor on the index date of diagnosis Numerator: Total number of persons with dementia with a regular family doctor on the index date of diagnosis, defined as: A history of at least three visits[b] to the same family doctor in the two years before the index date of diagnosis AND Scoring 50 or more on the Usual Provider of Care (UPC) index (maximum number of primary care visits[b] to the same family doctor / total number of primary care visits[b]) [129,130] Denominator: Total number of persons with dementia Subset (for UPC): Persons with dementia with at least 3 primary care visits[b] in the two years before the index date of diagnosis Rate adjustment: Age
	Access to an interprofessional primary care team	Rate of persons with dementia with access to an interprofessional primary care team (i.e. patient-centered medical home) on the index date of diagnosis

		Numerator: Total number of persons with dementia enrolled in an interprofessional primary care team on the index date of diagnosis defined as: Formal enrollment to a patient-centered medical home known as Family Health Teams in Ontario [25] Denominator: Total number of persons with dementia Rate adjustment: Age
	Dementia diagnosis first recorded by the regular family doctor [a]	Rate of persons with dementia with a first recording of dementia in the administrative data by the regular family doctor Numerator: Number of persons with dementia where index date of diagnosis is equal to a visit[b] to the regular family doctor Denominator: Total number of persons with dementia Subset: Persons with dementia with a regular family doctor Rate adjustment: Age
	Medication for dementia dispensed by the regular family doctor [a, c]	Rate of persons with dementia with at least 1 prescription for dementia medication dispensed in the year following the index date of diagnosis prescribed by the regular family doctor Numerator: Total number of persons with dementia with a prescription for dementia medication (cholinesterase inhibitors) dispensed in the year following the index date of diagnosis prescribed by the regular family doctor Denominator: Total person-time

		Subset: Persons with dementia with a regular family doctor Rate adjustment: Age and person-time
	Visits to the regular family doctor [a,b]	Rate of visits to the regular family doctor in the year following the index date of diagnosis Numerator: Total number of visits[b] to the regular family doctor in the year following the index date of diagnosis Denominator: Total person-time Subset: Persons with dementia with a regular family doctor on the index date of diagnosis Rate adjustment: Age and person-time
	Continuity of care	Continuity of care across physicians in the year following the index date of detection Numerator: Bice-Boxerman Continuity of Care (COC) Index [129] $\frac{(\sum_{i=1}^{p} n_i^2) - n}{n(n-1)}$ where n = all ambulatory visits to all types of physicians in the two years prior to the index date of diagnosis, p = total number of physicians. Referrals counted as visits were excluded. COC index varies from 0 to 1, with 0 indicating each patient visit was to different physicians and 1 indicating all visits were to the same physician [129] Denominator: Total person-time

		Subset (for Bice index): Persons with dementia with at least 3 visits to any physician in the two years prior to the index date of diagnosis Rate adjustment: Age and person-time
	Visits to the regular family doctor within 7 days following a hospitalization [a]	Rate of at least 1 visit[b] to the regular family doctor within 7 days of a hospital discharge in the year following the index date of diagnosis Numerator: Total number of persons with dementia with at least 1 visit[b] to the regular family doctor within 7 days of any hospital discharge occurring in the year following the index of date of dementia diagnosis Denominator: Total person-time Subset: Persons with dementia with a regular family doctor AND with at least 1 hospital discharge occurring in the year following the index of date of dementia diagnosis Rate adjustment: Age and person-time
	Dying at home	Rate of persons with dementia dying at home Numerator: Total number of persons with dementia who died in the community in the year following the index date of diagnosis, defined as: Deaths occurring in the community including home, rehabilitation or complex continuous care facilities, excludes deaths in the emergency department or hospital Denominator: Total number of persons with dementia who died in the year following the index date of diagnosis Subset: Persons with dementia deceased in year following the index date of diagnosis

		Rate adjustment: Age
Health service use	Medication dispensed for dementia [c]	Rate of persons with dementia with at least 1 prescription for dementia medication dispensed in the year following the index date of diagnosis Numerator: Total number of persons with dementia with at least 1 prescription for dementia medication (cholinesterase inhibitors) dispensed in the year following the index date of diagnosis Denominator: Total person-time Rate adjustment: Age and person-time
	Visits to specialists in dementia care [a]	Rate of visits to cognition specialists (geriatricians, neurologists, psychiatrists) in the year following the index date of diagnosis Numerator: Total number of visits to cognition specialists in the year following the index date of diagnosis Denominator: Total person-time Rate adjustment: Age and person-time
	Visits to other (non-cognition) specialists [a]	Rate of visits to non-cognition specialists in the year following the index date of diagnosis Numerator: Total number of visits to non-cognition specialists (all specialists other than geriatricians, neurologists, psychiatrists) in the year following the index date of diagnosis Denominator: Total person-time Rate adjustment: Age and person-time
	Visits to the emergency department	Rate of persons with dementia with at least 1 emergency department visit in the year following the index date of diagnosis Numerator: Total number of persons with dementia with at least 1 emergency department

		visit (unscheduled, not a transfer from another emergency department and with or without a subsequent hospital admission) in the year following the index date of diagnosis Denominator: Total person-time Rate adjustment: Age and person-time
	Hospitalizations	Rate of persons with dementia with at least 1 hospitalization (non-elective) in the year following the index date of diagnosis Numerator: Total number of persons with dementia with at least 1 hospital admission in the year following the index date of diagnosis Denominator: Total person-time Rate adjustment: Age and person-time
	Avoidable hospitalizations	Rate of persons with dementia with at least 1 avoidable hospitalization in the year following the index date of diagnosis Numerator: Total number of persons with dementia with at least 1 potentially avoidable hospitalization in the year following the index date of diagnosis, defined as: A hospital admission with a most responsible diagnosis of asthma, cardiac heart failure, chronic obstructive pulmonary disease, diabetes, hypertension, angina or seizures [30] Denominator: Total person-time Subset: Persons with dementia with at least 1 hospitalization Rate adjustment: Age and person-time
	Discharge delay	Rate of days delayed from hospital discharge

		Numerator: Total number of days of continued hospital stay after a patient is deemed medically fit to leave hospital (coded internally as *alternative level of care*) in the year following the index date of diagnosis Denominator: Total person-time Subset: Persons with dementia with at least 1 day in ALC Rate adjustment: Age and person-time
	Readmissions to the hospital within 30 days following a hospitalization	Rate of persons with dementia with at least 1 hospital readmission (non-elective) within 30 days of a previous hospital discharge in the year following the index date of diagnosis Numerator: Total number of persons with dementia with at least 1 hospital readmission within 30 days of a previous hospital discharge in the year following the index of date of diagnosis Denominator: Total person-time Subset: Persons with dementia with at least 1 hospital discharge in the year following the index date of diagnosis Rate adjustment: Age and person-time
	Use of home care	Rate of persons with dementia with at least 1 home care visit in the year following the index date of diagnosis Numerator: Total number of persons with dementia with at least 1 home care visit in the year following the index date of diagnosis Denominator: Total person-time Rate adjustment: Age and person-time

	Use of long-term care	Rate of persons with dementia admitted to long-term care in the year following the index date of diagnosis Numerator: Total number of persons with dementia admitted to long-term care in the year following the index date of diagnosis Denominator: Total person-time Rate adjustment: Age and person-time

Footnote:

[a] A regular family doctor is defined as per the first indicator "Having a regular family doctor"

[b] Primary care visits included office, home or phone visits by a family physician or general practitioner. Maximum of one visit per patient per physician per day.

[c] As memantine is not reimbursed or covered in the drug formulary in Ontario, only prescriptions for cholinesterase inhibitors were considered.

5.10.2 Supplementary file S2: Crude rates of primary care performance and health service use indicators for persons newly identified with dementia from 2002-2014 in Ontario, Canada

		Women			Men		
Performance indicator	Fiscal Year	Total population	Total events, n (%); mean (SD)	Age-standardized rate	Total population	Total events, n (%); mean (SD)	Age-standardized rate
PRIMARY CARE							
Having a regular family doctor, n (%)	2002	11051	10337 (93.5%)	93.7 (90.8-96.8)	7071	6686 (94.6%)	94.8 (91.7-97.9)
	2003	11360	10609 (93.4%)	93.1 (90.3-95.9)	7235	6797 (94.0%)	93.9 (90.9-96.9)
	2004	11503	10764 (93.6%)	93.5 (90.7-96.5)	7485	6988 (93.4%)	93.1 (90.2-96.1)
	2005	11718	10893 (93.0%)	92.4 (89.7-95.2)	7688	7172 (93.3%)	93.0 (90.1-96.0)
	2006	12316	11462 (93.1%)	92.7 (90.0-95.5)	8273	7724 (93.4%)	93.0 (90.1-95.9)
	2007	12581	11727 (93.2%)	92.9 (90.2-95.6)	8391	7830 (93.3%)	93.1 (90.3-95.9)
	2008	13611	12708 (93.4%)	93.3 (90.7-95.9)	9038	8407 (93.0%)	92.7 (90.0-95.5)
	2009	13849	12923 (93.3%)	92.9 (90.4-95.4)	9151	8538 (93.3%)	93.1 (90.4-95.9)
	2010	13905	12861 (92.5%)	92.0 (89.5-94.6)	9414	8761 (93.1%)	92.8 (90.2-95.5)
	2011	13749	12719 (92.5%)	91.7 (89.2-94.3)	9649	8991 (93.2%)	92.7 (90.0-95.3)
	2012	14052	12929 (92.0%)	91.9 (89.4-94.4)	9705	9007 (92.8%)	92.3 (89.7-94.9)
	2013	13738	12640 (92.0%)	91.3 (88.9-93.7)	9928	9203 (92.7%)	92.4 (89.8-95.0)
	2014	12884	11704 (90.8%)	90.8 (88.3-93.3)	9147	8424 (92.1%)	91.7 (89.1-94.4)
Absolute change				-3.0			-3.0
Relative change, %				3.2%			3.2%
Access to an interprofessional primary care team, n (%)	2002	12096	424 (3.5%)	3.6 (3.1-4.2)	7773	264 (3.4%)	3.5 (3.0-4.1)
	2003	12440	602 (4.8%)	4.8 (4.2-5.4)	7906	359 (4.5%)	4.4 (3.8-5.0)
	2004	12512	776 (6.2%)	6.0 (5.3-6.7)	8128	543 (6.7%)	6.8 (6.1-7.6)
	2005	12644	1172 (9.3%)	9.1 (8.3-9.9)	8347	750 (9.0%)	8.8 (8.0 -9.7)
	2006	13157	1628 (12.27%)	12.4 (11.5-13.4)	8871	1226 (13.8%)	13.9 (12.9-15.1)

	2007	13420	2401 (6.2%)	18.5 (17.3-19.7)	8921	1505 (16.9%)	17.26(16.1-18.5)
	2008	14493	3319 (22.9%)	22.9 (21.7-24.2)	9634	2179 (22.6%)	22.0 (20.7-23.2)
	2009	14706	3777 (25.7%)	25.2 (23.9-26.5)	9707	2347 (24.2%)	23.8 (22.5-25.2)
	2010	14885	4024 (27.0%)	27.2 (25.9-28.5)	10085	2785 (27.6%)	26.8 (25.5-28.2)
	2011	14796	4347 (29.4%)	28.4(27.1-29.8)	10349	3008 (29.1%)	28.9 (27.5-30.4)
	2012	15210	4521 (29.7%)	29.1(27.8-30.5)	10528	3151 (29.9%)	29.4 (28.0-30.8)
	2013	15001	4537 (30.2%)	30.9 (29.6-32.3)	10803	3167 (29.3%)	29.3 (28.0-30.7)
	2014	14172	4319 (30.5%)	30.1 (28.8-31.5)	10070	3113 (30.9%)	30.1 (28.7-31.6)
Absolute change				26.5			26.6
Relative change, %				732.3%			760.0%
Dementia diagnosis first recorded by the regular family doctor, n (%)	2002	10337	3996 (38.7%)	38.0 (36.1-39.9)	6686	2373 (35.5%)	34.9 (33.0-36.8)
	2003	10609	4189 (39.5%)	38.4 (36.6-40.2)	6797	2480 (36.5%)	36.5 (34.6-38.5)
	2004	10764	4301 (40%)	38.5 (36.7-40.4)	6988	2572 (36.8%)	36.6 (34.7-38.6)
	2005	10893	4389 (40.3%)	39.7 (37.9-41.6)	7172	2712 (37.8%)	36.5 (34.7-38.4)
	2006	11462	4659 (40.6%)	40.7 (38.8-42.6)	7724	2984 (38.6%)	36.4 (34.7-38.3)
	2007	11727	4791 (40.9%)	40.4 (38.6-42.3)	7830	3057 (39%)	37.8 (36.0-39.7)
	2008	12708	5437 (42.8%)	42.4 (40.6-44.2)	8407	3180 (37.8%)	37.4 (35.7-39.3)
	2009	12923	5534 (42.8%)	43.1 (41.3-44.9)	8538	3327 (39%)	38.9 (37.1-40.7)
	2010	12861	5206 (40.5%)	39.8 (38.1-41.5)	8761	3268 (37.3%)	36.6 (34.9-38.4)
	2011	12719	4979 (39.1%)	39.2 (37.5-41.0)	8991	3311 (36.8%)	36.6 (34.9-38.4)
	2012	12929	4937 (38.2%)	37.2 (35.6-38.8)	9007	3211 (35.7%)	35.0 (33.4-36.7)
	2013	12640	4936 (39.1%)	39.9 (38.3-41.7)	9203	3343 (36.3%)	36.1 (34.4-37.8)
	2014	11704	4544 (38.8%)	38.9 (37.2-40.6)	8424	2921 (34.7%)	34.3 (32.6-36.0)
Absolute change				0.9			-0.58
Relative change, %				2.4%			1.7%
Medication for dementia dispensed by the regular family doctor, n (%)	2002	10337	2392 (23.1%)	25.1 (23.5-26.7)	6686	1523 (22.8%)	24.6 (23.0-26.3)
	2003	10609	2708 (25.5%)	26.3 (24.8-27.8)	6797	1770 (26.0%)	27.6 (25.9-29.3)
	2004	10764	3053 (28.4%)	29.5 (27.9-31.2)	6988	1895 (27.1%)	28.0 (26.4-29.7)

	2005	10893	2995 (27.5%)	29.6 (28.0-31.3)	7172	1868 (26.1%)	27.5 (25.8-29.2)
	2006	11462	3312 (28.9%)	31.5 (29.8-33.2)	7724	2223 (28.8%)	28.9 (27.3-30.6)
	2007	11727	3502 (29.9%)	31.3 (29.7-32.9)	7830	2297 (29.3%)	29.5 (28.0-31.2)
	2008	12708	3880 (30.5%)	32.0 (30.4-33.6)	8407	2445 (29.1%)	29.5 (27.9-31.1)
	2009	12923	4161 (32.2%)	33.6 (32.1-35.2)	8538	2564 (30.0%)	30.7 (29.1-32.3)
	2010	12861	3751 (29.2%)	30.5 (29.0-32.0)	8761	2429 (27.7%)	29.2 (27.6-30.8)
	2011	12719	3421 (26.9%)	27.9 (26.5-29.4)	8991	2398 (26.7%)	26.9 (25.5-28.4)
	2012	12929	3356 (26%)	26.9 (25.5-28.3)	9007	2259 (25.1%)	25.2 (23.8-26.6)
	2013	12640	2939 (23.3%)	23.9 (22.6-25.2)	9203	2057 (22.4%)	22.9 (21.6-24.3)
	2014	11704	2756 (23.5%)	24.6 (23.3-26.0)	8424	1912 (22.7%)	23.4 (22.0-24.8)
Absolute change				**-0.5**			**-1.2**
Relative change, %				**2.0%**			**5%**
Visits to the regular family doctor, mean (SD)	2002	10337	3.5 (± 5.1)	3.9 (3.8-3.9)	6686	3.6 (± 4.5)	3.9 (3.8-4.0)
	2003	10609	3.5 (± 4.5)	3.8 (3.7-3.9)	6797	3.5 (± 4.4)	3.8 (3.7-3.9)
	2004	10764	3.6 (± 4.6)	3.9 (3.8-3.9)	6988	3.8 (± 4.7)	4.1 (4.0-4.2)
	2005	10893	3.5 (± 4.5)	3.9 (3.7-3.8)	7172	3.7 (4.8)	3.9 (3.8-3.9)
	2006	11462	3.5 (± 4.5)	3.8 (3.8-3.9)	7724	3.7 (± 4.5)	3.9 (3.8-3.9)
	2007	11727	3.4 (± 4.3)	3.6(3.5-3.7)	7830	3.6 (± 4.4)	3.8 (3.8-3.9)
	2008	12708	3.5 (± 4.3)	3.7 (3.6-3.7)	8407	3.6 (± 4.4)	3.8 (3.7-3.8)
	2009	12923	3.4 (4.1)	3.7 (3.6-3.7)	8538	3.5 (± 4.3)	3.6 (3.6-3.7)
	2010	12861	3.2 (± 4.0)	3.4 (3.3-3.5)	8761	3.4 (± 4.2)	3.5 (3.4-3.5)
	2011	12719	3.1 (± 4.0)	3.3 (3.3-3.4)	8991	3.2 (± 4.0)	3.3 (3.3-3.4)
	2012	12929	3.1 (± 3.8)	3.2 (3.1-3.2)	9007	3.1 (± 4.0)	3.3 (3.2-3.3)
	2013	12640	3.0 (± 3.8)	3.2 (3.1-3.2)	9203	3.1 (± 3.9)	3.3 (3.2-3.3)
	2014	11704	3.0 (± 4.0)	3.2 (3.2-3.3)	8424	3.0 (± 3.7)	3.1 (3.1-3.2)
Absolute change				**-0.7**			**-0.8**
Relative change, %				**16.8%**			**20.0%**
Continuity of care, mean (SD)	2002	7792	0.82 (± 0.29)	0.84 (0.81-0.88)	4887	0.83 (± 0.28)	0.86 (0.83-0.90)
	2003	7987	0.82 (± 0.29)	0.85 (0.82-0.88)	5056	0.82 (± 0.29)	0.84 (0.81-0.88)

	2004	8281	0.81 (± 0.30)	0.84 (0.8-0.87)	5325	0.81 (± 0.29)	0.84 (0.81-0.88)
	2005	8485	0.81 (± 0.30)	0.83 (0.8-0.86)	5497	0.81 (± 0.30)	0.84 (0.81-0.87)
	2006	8826	0.81 (± 0.30)	0.82 (0.79-0.85)	6011	0.80 (± 0.30)	0.83 (0.8-0.86)
	2007	9000	0.80 (± 0.31)	0.82 (0.79-0.85)	6011	0.81 (± 0.30)	0.83 (0.8-0.86)
	2008	9989	0.79 (± 0.31)	0.82 (0.79-0.85)	6544	0.78 (± 0.31)	0.81 (0.78-0.84)
	2009	10128	0.79 (± 0.31)	0.81 (0.78-0.84)	6579	0.79 (± 0.31)	0.81 (0.78-0.84)
	2010	10041	0.77 (± 0.32)	0.8 (0.77-0.83)	6774	0.77 (± 0.32)	0.8 (0.77-0.83)
	2011	9827	0.76 (± 0.33)	0.79 (0.76-0.81)	6976	0.76 (± 0.33)	0.78 (0.75-0.81)
	2012	10320	0.75 (± 0.33)	0.77 (0.74-0.8)	7041	0.76 (± 0.33)	0.78 (0.75-0.81)
	2013	10001	0.75 (± 0.34)	0.77 (0.74-0.79)	7306	0.75 (± 0.34)	0.78 (0.75-0.81)
	2014	9622	0.73 (± 0.34)	0.75 (0.73-0.78)	6806	0.72 (0.35)	0.75 (0.72-0.78)
Absolute change				**-0.09**			**-0.11**
Relative change, %				**10.7%**			**12.8%**
Visit to the regular family doctor within 7 days following a hospitalization, n (%)	2002	1672	288 (17.2%)	19.7 (16.0-24.0)	1140	219 (19.2%)	20.1 (16.7-24.1)
	2003	1642	297 (18.1%)	22.0 (17.7-26.95)	1155	222 (19.2%)	22.7 (18.6-27.5)
	2004	1702	311 (18.3%)	19.9 (16.3-24.0)	1224	262 (21.4%)	24.8 (20.6-29.6)
	2005	1707	306 (17.9%)	21.7 (17.9-26.0)	1186	226 (19.1%)	20.2 (16.7-24.1)
	2006	1712	280 (16.4%)	19.0 (15.2-23.6)	1298	234 (18%)	19.2 (15.9-23.1)
	2007	1833	293 (16%)	17.5 (13.9-21.8)	1279	241 (17.7%)	21.4 (17.8-25.5)
	2008	1914	331 (17.3%)	21.2 (17.3-25.6)	1364	232 (16.5%)	20.6 (17.0-24.9)
	2009	1941	343 (17.7%)	18.8 (15.5-22.5)	1404	238 (16.1%)	18.7 (15.5-22.5)
	2010	1985	327 (16.5%)	19.0 (15.6-23.1)	1482	255 (16.4%)	19.2 (15.9-22.9)
	2011	1926	282 (14.6%)	15.7 (12.4-19.5)	1553	269 (17.3%)	19.2 (16.0-22.8)
	2012	2130	361 (16.9%)	18.3 (14.8-22.4)	1552	279 (16.5%)	22.4 (18.7-26.7)
	2013	2132	330 (15.5%)	16.4 (13.2-20.1)	1696	295 (18.3%)	20.2 (16.8-24.0)
	2014	2120	326 (15.4%)	18.7 (15.1-22.8)	1615	257 (15.9%)	19.4 (15.8-23.6)
Absolute change				**-1.0**			**-0.7**
Relative change, %				**5.2%**			**3.4%**
Dying at home, n (%)	2002	745	190 (25.5%)	26.5 (18.3-37.2)	739	183 (24.8%)	26.9 (19.4-36.2)

	2003	681	170 (25.0%)	27.3 (20.0-36.3)	735	188 (25.6%)	21.9 (16.9-28.1)
	2004	672	211 (31.4%)	35.8 (25.7-48.5)	678	204 (30.1%)	29.2 (22.6-37.2)
	2005	634	200 (31.6%)	25.8 (17.1-37.4)	626	168 (26.8%)	28.3 (21.7-36.4)
	2006	686	200 (29.2%)	32.8 (23.7-44.4)	708	185 (26.1%)	29.0 (21.5-38.4)
	2007	746	202 (27.1%)	34.1 (24.2-46.7)	711	174 (24.5%)	21.^ (16.4-28.5)
	2008	802	206 (25.7%)	24.9 (18.2-33.3)	810	191 (23.6%)	24.0 (18.1-31.1)
	2009	801	224 (28.0%)	25.7 (19.0-33.8)	840	226 (26.9%)	27.1 (20.7-35.0)
	2010	848	231 (27.2%)	27.3 (20.2-36.1)	879	249 (28.3%)	26.6 (21.0-33.3)
	2011	812	241 (29.7%)	33.8 (23.5-47.1)	838	207 (24.7%)	30.5 (22.6-40.2)
	2012	905	255 (28.2%)	27.5 (21.1-35.2)	847	259 (30.6%)	26.1 (20.6-32.6)
	2013	879	257 (29.2%)	30.8 (22.9-40.5)	873	240 (27.5%)	25.8 (20.1-32.7)
	2014	885	278 (31.4%)	28.7 (21.3-37.8)	864	232 (26.9%)	29.9 (23.3-37.7)
Absolute change				**2.2**			**3.0**
Relative change, %				**8.1%**			**11.1%**
HEALTH SERVICE USE							
Medication dispensed for dementia, n (%)	2002	12,096	4982 (41.2%)	47.8 (45.7-50.0)	7,773	3126 (40.2%)	44.5 (42.4-46.5)
	2003	12,440	5636 (45.3%)	49.3 (47.3-51.3)	7,906	3643 (46.1%)	50.2 (48.2-52.3)
	2004	12,512	6089 (48.7%)	54.4 (52.3-56.6)	8,128	3840 (47.2%)	51.8 (49.7-54.0)
	2005	12,644	5867 (46.4%)	51.6 (49.6-53.7)	8,347	3825 (45.8%)	50.5 (48.4-52.7)
	2006	13,157	6408 (48.7%)	53.9 (51.8-56.0)	8,871	4181 (47.1%)	50.6 (48.5-52.7)
	2007	13,420	6565 (48.9%)	52.9 (50.9-54.9)	8,921	4351 (48.8%)	51.8 (49.8-53.9)
	2008	14,493	7122 (49.1%)	54.2 (52.3-56.2)	9,634	4599 (47.7%)	51.3 (49.4-53.3)
	2009	14,706	7276 (49.5%)	53.1 (51.2-55.0)	9,707	4658 (48%)	50.9 (48.9-52.9)
	2010	14,885	6826 (45.9%)	50.5 (48.6-52.3)	10,085	4539 (45%)	48.6 (46.7-50.5)
	2011	14,796	6414 (43.3%)	46.8 (45.1-48.6)	10,349	4437 (42.9%)	45.4 (43.6-47.2)
	2012	15,210	6234 (41%)	44.8 (43.2-46.6)	10,528	4251 (40.4%)	43.2 (41.5-45.0)
	2013	15,001	5479 (36.5%)	40.0 (38.5-41.6)	10,803	3925 (36.3%)	38.8 (37.2-40.4)
	2014	14,172	5146 (36.3%)	40.8 (39.2-42.5)	10,070	3644 (36.2%)	39.4 (37.7-41.1)
Absolute change				**-7.0**			**-5.1**
Relative change, %				**14.6%**			**11.4%**

Visits to specialists in dementia care, mean (SD)	2002	12,096	0.61 (±1.5)	0.90 (0.87-0.94)	7,773	0.67 (±1.5)	0.87 (0.84-0.90)
	2003	12,440	0.62 (±1.6)	0.87(0.84-0.9)	7,906	0.67 (±2.0)	0.86 (0.83-0.89)
	2004	12,512	0.68 (±1.9)	0.91 (0.88-0.94)	8,128	0.69 (±1.4)	0.88 (0.85-0.91)
	2005	12,644	0.69 (±1.6)	0.95 (0.92-0.98)	8,347	0.72 (±1.6)	0.9(0.87-0.93)
	2006	13,157	0.69 (±1.6)	0.89 (0.86-0.92)	8,871	0.74 (±1.5)	0.94 (0.91-0.97)
	2007	13,420	0.67 (±1.5)	0.90 (0.87-0.93)	8,921	0.74 (±1.9)	0.97 (0.94-1.0)
	2008	14,493	0.69 (±1.6)	0.93 (0.91-0.96)	9,634	0.71 (±1.5)	0.89 (0.86-0.91)
	2009	14,706	0.67 (±1.6)	0.92 (0.89-0.95)	9,707	0.71 (±1.4)	0.89 (0.86-0.91)
	2010	14,885	0.68 (±1.6)	0.90 (0.87-0.93)	10,085	0.75 (±1.6)	0.93 (0.90-0.96)
	2011	14,796	0.74 (±1.7)	0.98 (0.96-1.01)	10,349	0.77 (±1.5)	0.96 (0.94-0.99)
	2012	15,210	0.70 (±1.5)	0.94 (0.91-0.96)	10,528	0.77 (±1.7)	1.0 (0.97-1.0)
	2013	15,001	0.74 (±1.6)	1.0 (0.97-1.0)	10,803	0.80 (±1.6)	1.0 (1.0-1.1)
	2014	14,172	0.76 (±1.8)	1.1 (1.1-1.1)	10,070	0.84 (±1.6)	1.1 (1.1-1.1)
Absolute change				**0.18**			**0.25**
Relative change, %				**20.0%**			**29.1%**
Visits to other specialists, mean (SD)	2002	12,096	1.2 (±2.2)	1.4 (1.4-1.4)	7,773	1.5 (±2.6)	1.7 (1.7-1.8)
	2003	12,440	1.2 (±2.2)	1.4 (1.4-1.4)	7,906	1.6 (±2.7)	1.8 (1.7-1.8)
	2004	12,512	1.2 (±2.3)	1.5(1.4-1.5)	8,128	1.7 (±2.9)	1.9 (1.9-2.0)
	2005	12,644	1.3 (±2.5)	1.5 (1.5-1.5)	8,347	1.7 (±2.9)	1.9 (1.8-1.9)
	2006	13,157	1.4 (±2.4)	1.5 (1.5-1.5)	8,871	1.8 (±3.0)	2.1 (2.0-2.1)
	2007	13,420	1.4 (±2.5)	1.6 (1.6-1.7)	8,921	1.8 (±2.9)	2.0 (1.9-2.0)
	2008	14,493	1.4 (±2.6)	1.7 (1.7-1.8)	9,634	1.9 (±3.1)	2.1 (2.0-2.1)
	2009	14,706	1.4 (±2.4)	1.7 (1.7-1.7)	9,707	2.0 (±3.1)	2.2 (2.1-2.2)
	2010	14,885	1.6 (±2.7)	1.8(1.7-1.8)	10,085	2.0 (±3.1)	2.2 (2.1-2.2)
	2011	14,796	1.6 (±2.7)	1.8 (1.7-1.8)	10,349	2.1 (±3.2)	2.3 (2.3-2.3)
	2012	15,210	1.6 (±2.7)	1.9 (1.8-1.9)	10,528	2.1 (±3.2)	2.3 (2.3-2.3)
	2013	15,001	1.6 (±2.7)	1.9 (1.8-1.9)	10,803	2.2 (±3.4)	2.4 (2.4-2.5)
	2014	14,172	1.8 (±2.9)	2.1 (2.0-2.1)	10,070	2.3 (±3.5)	2.6 (2.5-2.6)
Absolute change				**0.67**			**0.81**

Relative change, %				**47.9%**			**46.6%**
Visit to the emergency department, n (%)	2002	12096	3446 (28.5%)	29.2 (27.7-30.8)	7773	2209 (28.4%)	29.9 (28.3-31.5)
	2003	12440	3491 (28.1%)	29.1 (27.7-30.7)	7906	2217 (28.0%)	30.3 (28.7-32.0)
	2004	12512	3639 (29.1%)	30.0 (28.5-31.6)	8128	2385 (29.3%)	30.5 (28.9-32.1)
	2005	12644	3633 (28.7%)	28.8 (27.3-30.2)	8347	2446 (29.3%)	30.1 (28.6-31.8)
	2006	13157	3905 (29.7%)	30.4 (28.9-32.0)	8871	2707 (30.5%)	31.7 (30.1-33.4)
	2007	13420	3941 (29.4%)	29.6 (28.2-31.1)	8921	2651 (29.7%)	30.8 (29.3-32.4)
	2008	14493	4356 (30.1%)	30.4 (28.9-31.8)	9634	2855 (29.6%)	29.6 (28.2-31.1)
	2009	14706	4444 (30.2%)	30.6 (29.2-32.0)	9707	2939 (30.3%)	30.5 (29.0-32.0)
	2010	14885	4653 (31.3%)	31.0 (29.6-32.4)	10085	3144 (31.2%)	32.0 (30.5-33.5)
	2011	14796	4634 (31.3%)	30.9 (29.5-32.4)	10349	3264 (31.5%)	31.4 (30.0-32.9)
	2012	15210	5003 (32.9%)	31.7 (30.4-33.1)	10528	3362 (31.9%)	32.1 (30.7-33.6)
	2013	15001	4956 (33.0%)	32.0 (30.6-33.3)	10803	3643 (33.7%)	33.3 (31.8-34.8)
	2014	14172	4959 (35.0%)	34.2 (32.8-35.7)	10070	3501 (34.8%)	34.5 (33.0-36.0)
Absolute change				**4.96**			**4.61**
Relative change, %				**17.0%**			**15.4%**
Hospitalizations, n (%)	2002	12,096	1943 (16.1%)	16.2 (15.1-17.4)	7,773	1320 (17.0%)	17.5 (16.4-18.8)
	2003	12,440	1928 (15.5%)	15.2 (14.1-16.3)	7,906	1323 (16.7%)	17.15(16.0-18.4)
	2004	12,512	1973 (15.8%)	15.9 (14.8-17.1)	8,128	1384 (17.0%)	16.8 (15.7-18.1)
	2005	12,644	1961 (15.5%)	14.8 (13.8-15.9)	8,347	1401 (16.8%)	16.8 (15.6-18.0)
	2006	13,157	1988 (15.1%)	14.4 (13.4-15.4)	8,871	1487 (16.8%)	16.4 (15.3-17.6)
	2007	13,420	2080 (15.5%)	14.4(13.5-15.4)	8,921	1455 (16.3%)	16.5 (15.4-17.6)
	2008	14,493	2149 (14.8%)	13.8 (12.9-14.8)	9,634	1578 (16.4%)	15.9 (14.9-17.0)
	2009	14,706	2215 (15.1%)	13.7 (12.9-14.7)	9,707	1581 (16.3%)	15.6 (14.6-16.7)
	2010	14,885	2295 (15.4%)	14.0 (13.1-14.9)	10,085	1687 (16.7%)	16.5 (15.5-17.6)
	2011	14,796	2258 (15.3%)	13.3 (12.4-14.2)	10,349	1787 (17.3%)	16.3 (15.3-17.4)
	2012	15,210	2519 (16.6%)	14.6 (13.7-15.5)	10,528	1832 (17.4%)	16.8 (15.7-17.8)
	2013	15,001	2521 (16.8%)	14.6 (13.8-15.5)	10,803	2028 (18.8%)	17.8 (16.8-18.8)
	2014	14,172	2539 (17.9%)	15.6 (14.7-16.5)	10,070	1967 (19.5%)	18.3 (17.2 -19.4)
Absolute change				**-0.65**			**0.76**

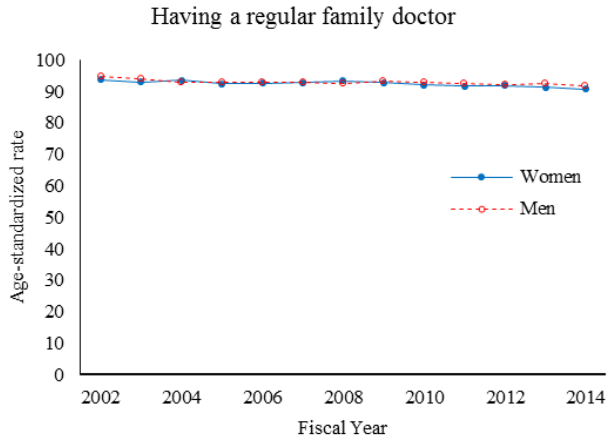
Relative change, %				**4.0%**			**4.3%**
Avoidable hospitalizations, n (%)	2002	1943	153 (7.8%)	9.8 (7.4-12.8)	1320	135 (10.23)	10.8 (8.5-13.5)
	2003	1928	168 (8.7%)	13.0 (10.0-16.7)	1323	133 (10.05)	10.9 (8.5-13.7)
	2004	1973	194 (9.8%)	13.5 (10.7-16.9)	1384	148 (10.69)	13.2 (10.3-16.7)
	2005	1961	139 (7.1%)	9.5 (7.1-12.5)	1401	125 (8.92%)	9.6 (7.5-12.7)
	2006	1988	148 (7.4%)	10.8 (8.0-14.2)	1487	135 (9.08%)	11.1 (8.6-14.2)
	2007	2080	159 (7.6%)	8.4 (6.5-10.7)	1455	141 (9.69%)	12.7 (9.9-15.9)
	2008	2149	164 (7.6%)	8.2 (6.1-10.9)	1578	143 (9.06%)	9.6 (7.6-1)
	2009	2215	147 (6.6%)	9.0 (6.6-12.2)	1581	126 (7.97%)	9.3 (7.1-12.1)
	2010	2295	159 (6.9%)	9.9 (7.4-12.7)	1687	148 (8.77%)	10.2(8.0-12.8)
	2011	2258	148 (6.6%)	9.4 (6.8-12.6)	1787	138 (7.72%)	9.3 (7.1-11.9)
	2012	2519	201 (8.0%)	13.3 (10.2-17.1)	1832	146 (7.97%)	8.6 (6.7-10.9)
	2013	2521	123 (4.9%)	6.1 (4.4-8.2)	2028	159 (7.84%)	9.7 (7.6-12.1)
	2014	2539	160 (6.3%)	9.0 (6.9-11.7)	1967	180 (9.15%)	10.5 (8.1-13.3)
Absolute change				**-0.8**			**-0.3**
Relative change, %				**8.2%**			**2.5%**
Discharge delay, mean (SD)	2002	679	24.3 (±32.1)	26.3 (25.5-27.1)	464	21.4 (±31.6)	31.4 (30.5-32.3)
	2003	687	19.0 (±23.4)	25.5 (24.6-26.4)	462	21.0 (±26.7)	25.7 (24.9-26.5)
	2004	735	19.0 (± 26.7)	21.0 (20.2-21.7)	462	19.4 (±28.8)	25.3 (24.5-26.2)
	2005	759	22.8 (±35.9)	28.1 (27.3-29.0)	523	25.4 (±45.9)	37.7 (36.7-38.7)
	2006	809	25.7 (±38.5)	39.3 (38.2-40.4)	593	31.4 (±46.6)	38.7 (37.8-39.6)
	2007	882	32.7 (±45.2)	39.2(38.4-40.0)	581	35.7 (±58.3)	47.2 (46.3-48.1)
	2008	922	31.8 (±48.9)	37.8 (36.9-38.7)	673	35.3 (±59.3)	40.1 (39.3-40.9)
	2009	983	31.4 (±52.7)	35.1 (34.3-36.0)	641	32.7 (±48.2)	38.7 (37.8-39.5)
	2010	1033	25.5 (±38.1)	31.0 (30.2-31.7)	703	28.8 (±42.0)	34.9 (34.2-35.6)
	2011	973	28.5 (±48.7)	32.8(32.0-33.6)	748	30.0 (±54.3)	35.4 (34.7-36.1)
	2012	1040	25.6 (±41.6)	33.2 (32.4-34.0)	726	37.8 (±79.2)	48.2 (47.2-49.1)
	2013	1086	22.9 (±43.4)	27.4 (26.7-28.0)	866	29.9 (±51.7)	34.1 (33.6-34.8)
	2014	1056	26.1 (±55.4)	35.6 (34.7-36.5)	801	31.6 (±56.7)	40.8 (40.0-41.6)

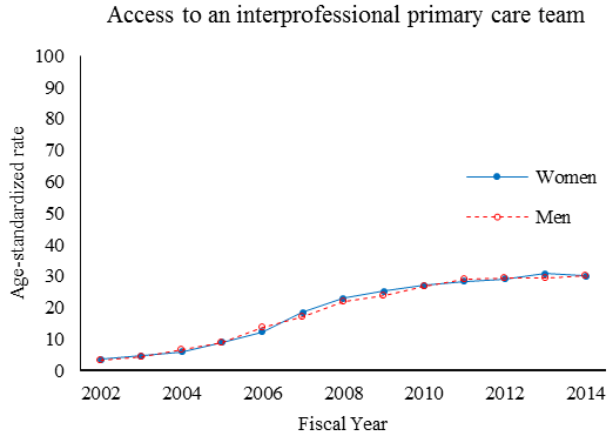
Absolute change Relative change, %				**9.28** **35.3%**			**9.38** **29.9%**
Readmissions to the hospital within 30 days following a hospitalization, n (%)	2002	1943	219 (11.3%)	10.8 (8.6-13.4)	1320	187 (14.2%)	16.1 (13.1-19.5)
	2003	1928	245 (12.7%)	15.6 (12.7-19.0)	1323	174 (13.2%)	16.9 (13.5-20.8)
	2004	1973	231 (11.7%)	15.5 (12.4-19.2)	1384	202 (14.6%)	16.3(13.1-20.0)
	2005	1961	250 (12.7%)	14.9 (12.0-18.2)	1401	199 (14.2%)	16.4 (13.2-20.1)
	2006	1988	232 (11.7%)	15.0 (11.8-18.8)	1487	182 (12.2%)	13.9 (11.1-17.3)
	2007	2080	229 (11%)	13.2 (10.4-16.5)	1455	186 (12.8%)	15.8 (12.8-19.4)
	2008	2149	245 (11.4%)	12.2 (9.5-15.1)	1578	208 (13.2%)	15.6 (12.7-19.1)
	2009	2215	253 (11.4%)	13.9 (10.9-17.5)	1581	207 (13.1%)	16.1 (13.0-19.5)
	2010	2295	294 (12.8%)	15.2(12.2-18.6)	1687	241 (14.3%)	16.8 (13.9-20.2)
	2011	2258	290 (12.8%)	12.5 (10.0-15.5)	1787	266 (14.9%)	17.0 (14.1-20.3)
	2012	2519	327 (13.0%)	16.9 (13.6-20.8)	1832	285 (15.6%)	19.7 (16.5-23.5)
	2013	2521	343 (13.6%)	15.6 (12.8-18.8)	2028	326 (16.1%)	19.5 (16.6-22.9)
	2014	2539	340 (13.4%)	12.9 (10.5-15.6)	1967	302 (15.4%)	18.4(15.4-21.8)
Absolute change Relative change, %				**2.1** **19.3%**			**2.3** **14.5%**
Use of home care, n (%)	2002	12,096	4493 (37.1%)	34.8 (33.2-36.5)	7,773	2381 (30.6%)	30.6 (29.1-32.3)
	2003	12,440	4411 (35.5%)	32.7 (31.3-34.3)	7,906	2395 (30.3%)	29.9.(28.4-31.5)
	2004	12,512	4318 (34.5%)	31.8 (30.4-33.4)	8,128	2368 (29.1%)	28.4 (26.9-29.9)
	2005	12,644	5494 (43.5%)	39.8 (38.2-41.5)	8,347	3130 (37.5%)	36.5 (34.8-38.2)
	2006	13,157	6432 (48.9%)	43.2 (41.5-44.9)	8,871	3811 (43%)	42.0 (40.2-43.9)
	2007	13,420	6675 (49.7%)	44.2 (42.6-45.9)	8,921	3865 (43.3%)	41.9 (40.1-43.7)
	2008	14,493	7384 (50.9%)	45.0 (43.4-46.7)	9,634	4369 (45.3%)	42.4 (40.8-44.1)
	2009	14,706	7715 (52.5%)	45.3 (43.8-46.9)	9,707	4444 (45.8%)	43.6 (41.9-45.3)
	2010	14,885	8139 (54.7%)	47.8 (46.2-49.5)	10,085	4917 (48.8%)	46.0 (44.3-47.8)
	2011	14,796	8049 (54.4%)	46.6 (45.0-48.2)	10,349	5090 (49.2%)	45.3 (43.6-47.0)
	2012	15,210	8631 (56.7%)	48.5 (46.9-50.2)	10,528	5386 (51.2%)	47.6 (45.9-49.4)

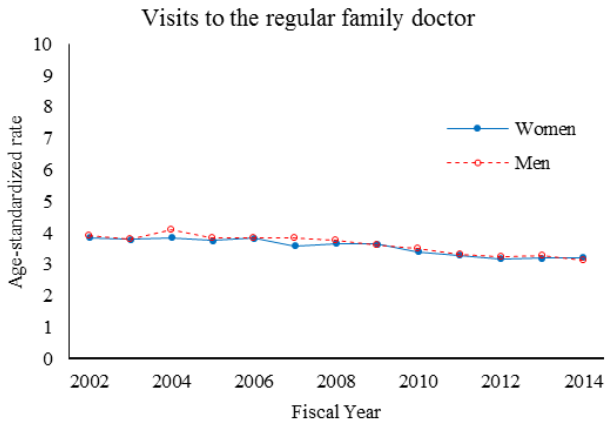
	2013	15,001	8820 (58.8%)	50.2 (48.6-51.8)	10,803	5790 (53.6%)	49.3 (47.6-51.1)
	2014	14,172	8618 (60.8%)	52.9(51.2-54.6)	10,070	5574 (55.4%)	51.2 (49.4-53.0)
Absolute change				**18.0**			**20.5**
Relative change, %				**52%**			**67.0%**
Use of long-term care, n (%)	2002	12,096	2113 (17.5%)	14.5 (13.6-15.6)	7,773	1125 (14.5%)	13.7 (12.7-14.7)
	2003	12,440	2204 (17.7%)	15.0 (14.0-16.0)	7,906	1111 (14.1%)	13.3 (12.3-14.4)
	2004	12,512	2142 (17.1%)	13.4 (12.6-14.3)	8,128	1154 (14.2%)	12.8 (11.9-13.8)
	2005	12,644	1961 (15.5%)	12.9 (12.0-13.8)	8,347	1156 (13.8%)	13.0 (12.0-14.0)
	2006	13,157	1942 (14.8%)	12.1 (11.2-13.0)	8,871	1083 (12.2%)	10.7 (9.9-11.6)
	2007	13,420	1785 (13.3%)	10.5 (9.8-11.3)	8,921	968 (10.9%)	10.1 (9.3-10.9)
	2008	14,493	1761 (12.2%)	10.0 (9.2-10.6)	9,634	1027 (10.7%)	9.4 (8.6-10.1)
	2009	14,706	1727 (11.7%)	9.0 (8.4-9.7)	9,707	904 (9.3%)	8.4 (7.7-9.2)
	2010	14,885	1767 (11.9%)	8.6 (8.1-9.4)	10,085	1015 (10.1%)	9.2 (8.4-10.0)
	2011	14,796	1613 (10.9%)	8.2 (7.6-8.9)	10,349	985 (9.5%)	8.4 (7.7-9.2)
	2012	15,210	1651 (10.9%)	8.6 (7.9-9.2)	10,528	931 (8.8%)	7.7 (7-8.4)
	2013	15,001	1672 (11.1%)	8.3 (7.7-8.9)	10,803	940 (8.7%)	7.6 (6.9-8.3)
	2014	14,172	1722 (12.2%)	9.0 (8.4-9.7)	10,070	1007 (10%)	8.6 (7.9-9.3)
Absolute change				**-5.5**			**-5.1**
Relative change, %				**37.9%**			**37.1%**

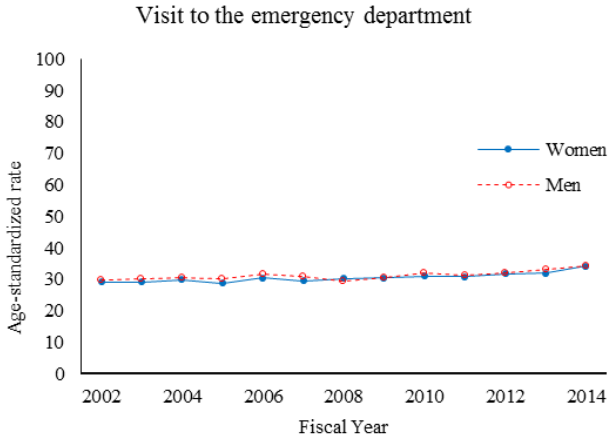
Footnote: Operational definitions available in Supplementary File S1. As the sign of the relative change in the indicator could represent either worsening or improvement, relative changes are reported in absolute value.

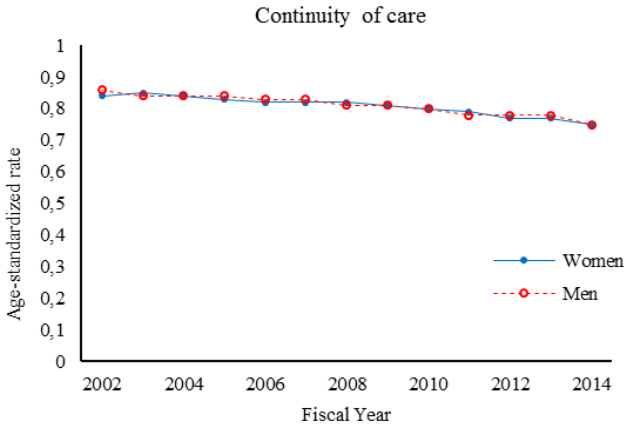
5.10.3 Supplementary file S3: Additional indicators of primary care performance and health service use

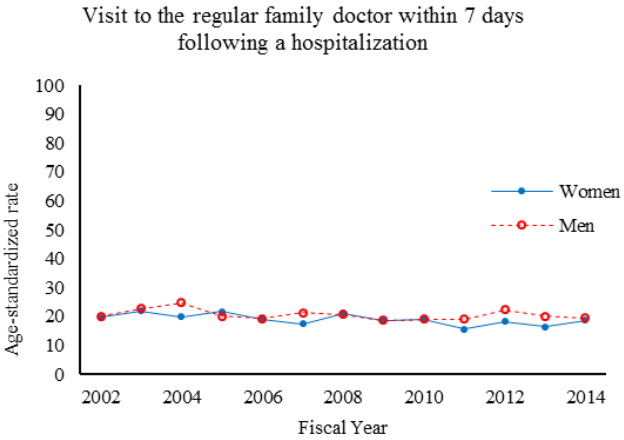




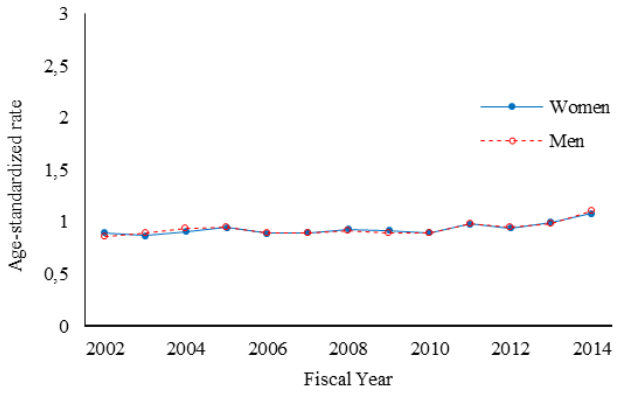




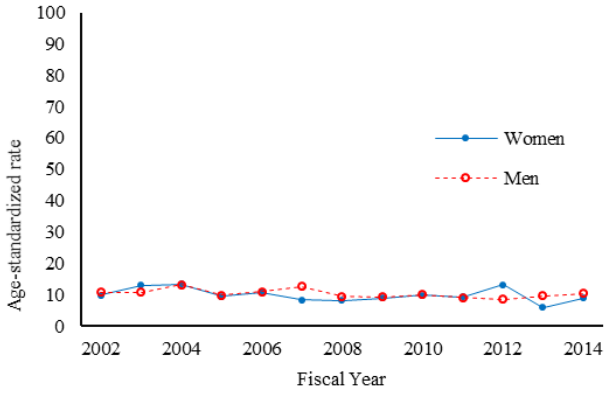




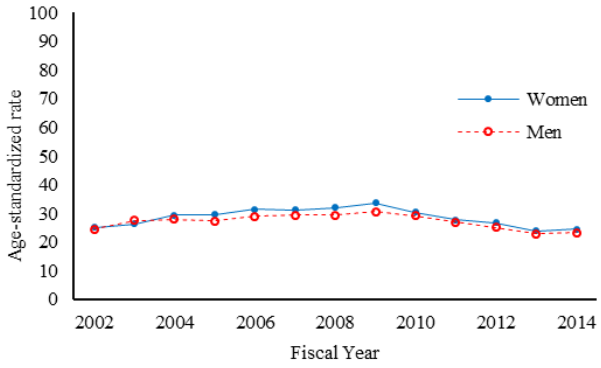
Visits to specialists in dementia care



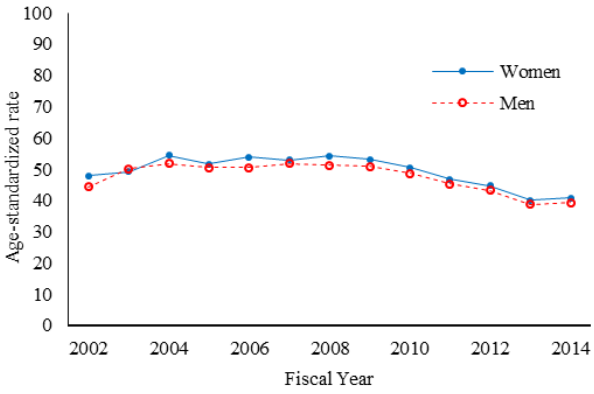
Avoidable hospitalizations



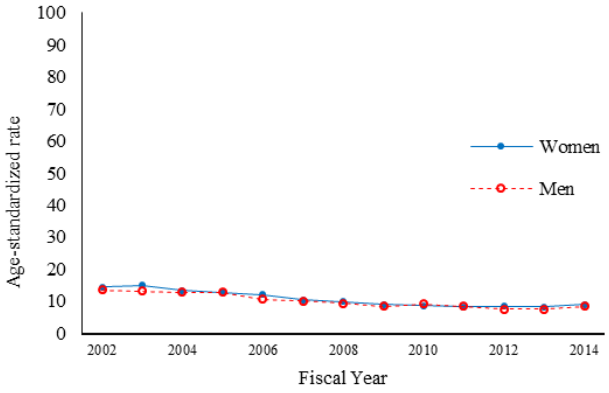
Medication for dementia dispensed by the regular family doctor



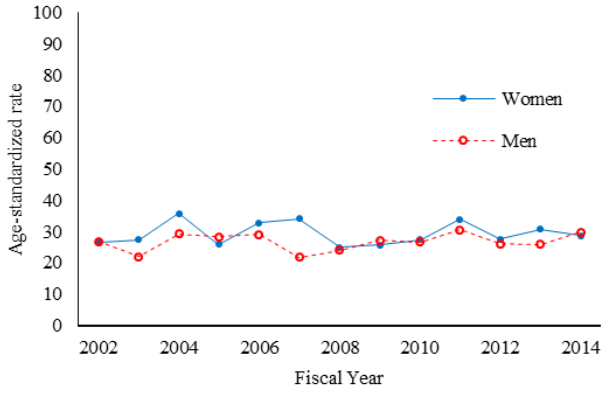
Medication dispensed for dementia



Access to long-term care



Dying at home



5.10.4 Supplementary file S4: Description of data sources and cohort creation

Description of data sources

Type of data	Database	Description
Demographics	Registered Persons Database (RPDB)	The RPDB provides demographic information about all individuals who are registered with the Ontario Health Insurance Plan, including their date of birth, sex, and home address.
Physician claims	Ontario Health Insurance Plan (OHIP)	The OHIP data are maintained by the Ministry of Health and Long-Term Care, records all claims for reimbursement by Ontario physicians for inpatient and ambulatory visits, consultations and procedures. The data also include claims from optometrists for publicly funded reimbursement and from laboratories for all diagnostic tests performed.
Primary care enrollment and group affiliation	Client Agency Program Enrolment (CAPE)	Managed by the Ministry of Health and Long-Term Care, the CAPE dataset identifies patients enrolled in different primary care models over time. A separate file provided by the Ministry of Health and Long-Term Care identifies the physicians that were part of a Family Health Team.
	Corporate Provider Database (CPDB)	The CPDB, maintained by the Ministry of Health and Long-Term Care, is a repository of provider and provider organization data. The CPDB receives regular updates of provider credentials from the College of Physicians and Surgeons of Ontario.
Acute care hospital use and ED visits	Discharge Abstract Database (DAD)	This DAD is compiled by the Canadian Institute for Health Information. It captures administrative, clinical and demographic information on hospital discharges, including deaths.
	National Ambulatory Care Reporting System (NACRS)	The NACRS is provided by the Canadian Institute for Health Information and contains demographic, clinical, and administrative data for all hospital- and community-based ambulatory care, such as day surgery and emergency department visits including chief complaint (reason for visit) in Ontario. NACRS data are available from 2002 onwards.

Dispensed prescription drugs	Ontario Drug Benefit (ODB)	The ODB database identifies the drug, dose and date for outpatient drug dispensations through publicly funded drug programs in Ontario. Eligible recipients are all Ontario residents aged ≥65 years and selected younger populations, including nursing home residents, patients receiving services under the Ontario Home Care program, those receiving social assistance, and residents eligible for specialized drug programs.
	Drug Identification Number (DIN)	The DIN data is supplied by IMS Brogan Inc and contains a near exhaustive list of DINs (Drug Identification Numbers) used in Canada from 1990 forward and includes information on the drug and product names and subclass information.
Home and long-term care services	Home Care Database (HCD)	The HCD is maintained by Health Shared Services Ontario and is a clinical, client-centered database that captures all home care services provided or coordinated by Local Health Integration Networks.
	Continuing Care Reporting System (CCRS)	The CCRS is a database that is maintained by the Canadian Institute of Health Information and contains demographic, clinical, functional and resource utilization information on individuals receiving continuing care services in hospitals or long-term care homes in Canada. Clinical data is obtained through the Resident Assessment Instrument, a patient assessment tool that assesses the care and needs of adult patients in hospital and community settings for in-home and placement services.
	Institution Information System	The Institution Information System is a series of related datasets containing information about Ontario health care institutions funded by the Ministry of Health and Long-Term Care.
Vital Statistics	Office of the Registrar General (ORGD) Vital Statistics Database	The ORGD is an annual dataset containing information on all deaths registered in Ontario starting on January 1 1990.

6 CHAPTER 6: TESTING GROUP DIFFERENCES FOR CONFOUNDER SELECTION IN NON-RANDOMIZED STUDIES: FLAWED PRACTICE (MANUSCRIPT 3)

6.1 Preamble

As was highlighted in the literature review **(**see **Section 2.5),** the non-randomized assignment of persons to a Family Health Team or other primary care model poses important challenges in estimating its true average causal effect, particularly to bias due to confounding given the underlying systematic differences in physician and patient characteristics across models. To determine the list of confounders for my impact evaluation (Chapter 7), I employed a causal inference technique, called Directed Acyclic Graphs, to visually represent the variables involved in the causal relationship between FHT affiliation and health service use.

As methods in causal inference remain underutilized in heath policy evaluation and health research in general, in this third manuscript, I sought to assess current practices and guidelines in confounder selection and provide guidance to clinicians and researchers on how new methods in causal inference, such as DAGs, can be used strengthen the estimation of non-randomized interventions, like FHTs. This manuscript has been published in the CMAJ:

Sourial N, Vedel I, Le Berre M, Schuster T. (2019). Testing group differences for confounder selection in non-randomized studies: flawed practice. CMAJ October 28, 2019 191 (43) E1189-E1193; DOI: https://doi.org/10.1503/cmaj.190085.

Title: Testing group differences for confounder selection in non-randomized studies: flawed practice

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6.2 Key messages

- Using observed imbalances between study groups (e.g. exposed and unexposed) to determine variables for confounding adjustment in non-randomized studies is an inappropriate practice
- This practice can misguide the selection of variables to control for in the analysis and bias study results
- Reporting guidelines on this practice are at times limited, vague and in some cases, erroneous
- Advances in causal inference offer new insights and solutions on handling confounding and should be incorporated in current reporting guidelines

6.3 Introduction

Non-randomized studies, including observational and quasi-experimental studies, are frequently used to determine the effect of a given exposure (e.g. a new practice, intervention or policy) on relevant outcomes in situations where random assignment to the exposed or unexposed group is not feasible or ethical.[1] A prevalent source of bias in causal inferences based on non-randomized studies is confounding. This type of bias occurs when characteristics of individuals that are causally linked to the outcome(s) of interest are imbalanced across the study groups. A known practice to identify these potential "confounders" is to statistically test for group imbalances based on the observed study data.[2-4] Characteristics found to be significantly imbalanced are then included in the statistical models in an attempt to reduce potential confounding bias.

While, on the surface, this practice seems reasonable, observed imbalances should not guide the selection of confounders and can in fact worsen the bias in estimating the exposure effect.

We review why testing group differences for confounder selection in non-randomized studies is inappropriate, assess shortcomings in established reporting guidelines and propose solutions based on recent advances in causal inference.

Why would researchers test group differences?

When imbalances are shown in variables that are conceptually consistent with confounders, this information can help corroborate knowledge on possible confounders. Researchers sometimes perform hypothesis tests to "confirm statistical significance" of observed imbalances and to inform the choice of variables for adjustment. For example, on reviewing articles published in the CMAJ in 2018 (Appendix 1), we found that, among the 34 non-randomized studies that compared two or more groups to assess the effect of an exposure, almost one quarter employed a form of statistical testing as a means of selecting confounders for model adjustment. While testing group imbalances can, at times, support the decision to include variables for adjustment, it can also create confusion in situations where results do not agree with preconceptions or knowledge. Testing becomes particularly problematic when used as the primary method to inform the choice of variables for confounding adjustment.

6.4 What are the pitfalls of testing group differences for confounder selection?

Limiting the search for confounders based on observed imbalances fails to consider possible unobserved variables that are relevant in the confounding mechanism.[5] Variables that are strong predictors of the outcome but only weakly associated with the exposure would be less likely to be selected for adjustment resulting in uncontrolled confounding and biased results. Earlier work by Sun et al. (1996) demonstrated that bivariate screening methods were insufficient to control confounding and could exclude important variables from the multivariable analysis.[2] Groenwold et al. (2011) showed that lack of adjustment for a baseline characteristic that is only marginally different between groups at baseline, but strongly associated with the outcome, can result in significantly overestimating the effect of the exposure.[4] At the other extreme, testing observed group differences may inadvertently identify as confounders, variables that are on the causal pathway between the exposure and outcome (so-called 'mediators') or variables that are a common effects of other variables for which at least one is linked to the exposure and one to the outcome

(so-called “colliders”) . There are many valid scenarios where identifying and measuring the effect of a mediator is of interest; for example, it may be more feasible or cost-effective to intervene at the level of the mediator rather than the exposure in order to affect the outcome.[6] However, when the purpose is to measure the total effect of an exposure, adjusting for mediators or colliders can, in fact, introduce rather than remove bias in the effect.[3,4,7] A more detailed review of mediation analysis is available elsewhere.[8]

Overall, relying on statistical testing for confounder selection can contribute to creating a paradox where true confounders may not be identified and, vice versa, where adjustment for non-confounders may create spurious associations.[3,4] Fortunately, more appropriate methods to assess confounding exist and will be discussed in a later section.

6.5 What guidance is provided in current reporting guidelines?

We reviewed relevant reporting guidelines and assessed the guidance, if any, provided for handling group imbalances in non-randomized studies or on confounder selection. Five guidelines were reviewed (Table 6.1).

Table 6.1: Relevant reporting guidelines related to testing group imbalances for confounder selection in non-randomized studies

Journal author/external reporting guidelines	Current guidance
ICMJE(9)	Does not include guidance on reporting of research methods including testing group imbalances for confounder selection in non-randomized studies Refers authors to STROBE
STROBE(11,12)	***Item 12 (a). Describe all statistical methods, including those used to control for confounding*** *"Investigators should think beforehand about potential confounding factors. This will inform the study design and allow proper data collection by identifying the confounders for which detailed information should be sought."* *"If groups being compared are not similar with regard to some characteristics, adjustment should be made for possible confounding variables by stratification or by multivariable regression"* ***Item #14: Descriptive data*** *"Inferential measures such as standard errors and confidence intervals should not be used to describe the variability of characteristics, and significance tests should be avoided in descriptive tables. Also, P values are not an appropriate criterion for selecting which confounders to adjust for in analysis; even small differences in a confounder that has a strong effect on the outcome can be important."*
TREND(14)	***Item #15 : Baseline equivalence - Data on study group equivalence at baseline and statistical methods used to control for baseline differences*** *"Example (baseline equivalence): the intervention and comparison groups did not statistically differ with respect to demographic data (gender, age, race/ethnicity; P > .05 for each), but the intervention group reported a significantly greater baseline frequency of injection drug use (P = .03); all regression analyses included baseline frequency of injection drug use as a covariate in the model"*
GRADE(13)	***5.2 Factors that can reduce the quality of the evidence*** *5.2.1 Study limitations (Risk of Bias)* *"Study limitations in observational studies:*

	• *Failure to develop and apply appropriate eligibility criteria (inclusion of control population)*
	  – *Under- or over-matching in case-control studies*
	  – *Selection of exposed and unexposed in cohort studies from different population*
	• *Failure to adequately control confounding*
	  – *Failure of accurate measurement of all known prognostic factors*
	  – *Failure to match for prognostic factors and/or adjustment in statistical analysis"*
	
	5.3. Factors that can increase the quality of the evidence
	
	<u>*5.3.3. Effect of plausible residual confounding*</u>
	"Rigorous observational studies will accurately measure prognostic factors associated with the outcome of interest and will conduct an adjusted analysis that accounts for differences in the distribution of these factors between intervention and control groups."

SAMPL(10)	Does not include specific guidance on reporting of research methods including testing group imbalances for confounder selection in non-randomized studies Refers authors to STROBE and TREND

<u>Abbreviations</u>:

ICMJE: International Committee of Medical Journal Editors; STROBE: STrengthening the Reporting of OBservational studies in Epidemiology; TREND: Transparent Reporting of Evaluations with Nonrandomized Designs; GRADE: Grading of Recommendations Assessment, Development and Evaluation; SAMPL: Statistical Analyses and Methods in the Published Literature

We found the level and appropriateness of the guidance varied (Table 6.1). Both the International Committee of Medical Journal Editors (ICMJE)[9] and the Statistical Analyses and Methods in the Published Literature (SAMPL)[10] offer no specific guidance and refer readers to Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) (Table 6.1).[11,12] STROBE provides appropriate guidance overall, recommending deciding on potential confounders at the study planning stage and discouraging the use of significance tests in confounder selection, but we found one statement that seemed somewhat misleading "If groups being compared are not similar with regard to some characteristics, adjustment should be made". Grading of Recommendations Assessment, Development and Evaluation (GRADE)[13] underlines

the importance of adequately selecting the variables for model adjustment as well as the risk of bias due to failure to control for confounding but offers no specific guidance on the methods that should or should not be used. Surprisingly, Transparent Reporting of Evaluations with Non-randomized Designs (TREND) explicitly promotes testing group differences as a tool to select variables for adjustment (Table 6.1).[14]

6.6 How should confounding variables be appropriately determined?

Confounding is a fundamental concept in causal inference, an area of research that has seen major developments in recent years.[15,16] The formal definition of confounders under this framework has also been the subject of recent debates.[17] Confounding is not something that can be determined or statistically tested using data alone.[3,18] Instead, selecting the set of confounders to adjust for should be considered at the design stage using a conceptual framework based on subject matter knowledge and published evidence.[3,11]

Causal diagrams known as Directed Acyclic Graphs (DAGs) are one among other conceptual design tools to aid in confounder selection and have been growing in popularity.[5,7,19] DAGs provide a visual conceptualization using arrows to represent the causal pathways involved in the exposure-outcome relationship (Figure 6.1). The graph is "directed" because arrows are unidirectional and "acyclic" because there is no path connecting a variable back onto itself. A key strength of DAGs is that, using graph theory, they can differentiate between confounders and other types of variables like mediators and colliders, to determine the set of confounders that must be taken into account when estimating the effect of interest (Figure 6.1). This is accomplished by examining the location of variables in the causal pathway and the causal links leading into and/or out of these variables.

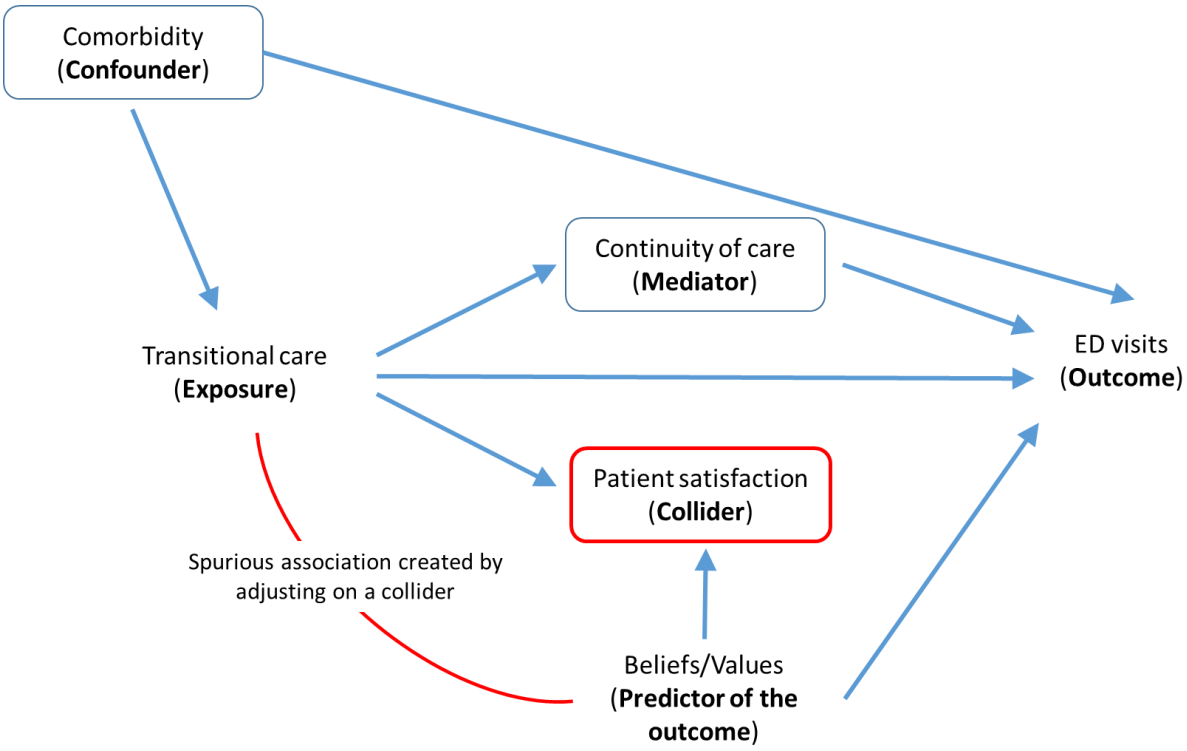

Figure 6.1: Hypothetical Directed Acyclic Graph and impact of adjusting for different types of variables in the causal pathway

Footnote: ED=Emergency Department. Boxes around a variable indicate adjustment. In this example, adjusting for comorbidity (confounder) would correctly block the spurious association between transitional care and ED visits due to the common cause of comorbidity. Adjusting for continuity of care (mediator) would block part of the total effect between transitional care and ED visits. Finally, adjusting for patient satisfaction (collider) would create a spurious association between transitional care and ED visits through patient beliefs/values, as indicated by the red arc connecting the colliding variables "exposure" and "predictor of the outcome".

Consider a hypothetical example (Figure 6.1) on the effect of a transitional care (TC) program (from hospital discharge back to the community) compared to usual care on subsequent emergency department (ED) visits. Suppose patients were not randomly assigned to either the TC or usual care group. We may decide to consider as confounders, variables found to be imbalanced between the groups, for example, the number of patient comorbidities, patients' level of continuity of care and satisfaction. However, mapping out the causal pathways through a DAG, we would be

able to uncover which of these variables should in fact be adjusted for and which should not. From the DAG, we see that:

- Patient comorbidity satisfies the conditions of a confounder, as it is a common cause of both group membership and going to the ED. Conclusion: Patient comorbidity *should* be adjusted for.
- If TC leads to better continuity of care, which in turn leads to decreased ED visits – then continuity of care would be a mediator and adjusting for it would block part of the total effect of TC on ED visits.

  Conclusion: Continuity of care *should not* be adjusted for if we are interested in estimating the total effect of TC on ED.
- If we consider that TC affects patient satisfaction but that patient beliefs/values also affect patient satisfaction and ED visits, then patient satisfaction is a common effect of both TC and beliefs – i.e. a collider. Adjusting for patient satisfaction would create a spurious association between TC and beliefs and, as beliefs, in this example, also predict patients' decision to go to the ED, a spurious association would be created between TC and ED visits (through beliefs), biasing the effect of TC on ED visits.

  Conclusion: Patient satisfaction *should not* be adjusted for.

A tabular summary of these assessments is also provided (Appendix 2). Thus, using a DAG at the design stage can help decide a priori on the variables and data that need to be collected. In this hypothetical example, only adjustment on patient comorbidity would be needed to ensure that the estimated effect of TC on ED visits is free of confounding bias.

Real-world DAGs are often more complex with a large number of inter-connected variables involved in the exposure-outcome relationship. Published tutorials and online tools are available to assist in developing and interpreting DAGs to tease out the set of confounders that should be adjusted for in a specific study.[20-22] It should be noted, however, that the utility of DAGs depends upon the quality of the evidence on which they are based. They also require a degree of subjective judgement and therefore cannot guarantee that all true confounders will be correctly identified. Nevertheless, DAGs remain a useful tool to better understand and visualize the complex

pathways involved in the exposure-outcome relationship and can help more rigorously determine sources of confounding.

In addition to DAGs, another approach to confounder selection is to adjust for variables that are known (or believed) to be predictive of exposure status, or predictive of the outcome, or both. This method has been shown to be sufficient to provide adequate confounding control.[23] Finally, for characteristics with observed group imbalances but which were not considered confounders at the design stage, bias factors or confounding functions can provide useful sensitivity analyses.[24,25] These methods calculate the magnitude by which the estimated effect of the exposure is affected by a potential confounder that was not controlled for or measured. As they incorporate the imbalance of a characteristic across exposure groups as well as its association with the outcome, these methods better reflect the triangular nature of confounding than imbalances with respect to exposure status alone. In their simplest form, bias factors are calculated by multiplying the difference in the prevalence of the confounder between the intervention and control group by the effect of the confounder on the outcome. For example, VanderWeele and Arah (2011) showed that if the prevalence of an unmeasured confounder is 30% higher in the intervention group than the control group and is associated with a 52% higher risk of having the outcome, then the magnitude of bias would be 0.30 x 0.52 = 0.16.[24] In other words, by *not* adjusting for this confounder, the exposure effect would be overestimated by 16%.

Confounding functions expand on bias factors by examining a range of different confounding scenarios. Their effect on the estimated effect of exposure can then be represented graphically to visualize the relationship between the degree of bias and shift in the estimated effect.[25] Details of the methods are provided elsewhere.[24, 25]

What are the implications for reporting guidelines on non-randomized studies?

Given the development of new tools and methods for confounder selection within the field of modern causal inference, an update to current guidance, with more uniformity across guidelines, on confounder selection in non-randomized studies seems warranted. We suggest these guidelines should 1) emphasize the selection of confounders at the design stage through the use of DAGs or other conceptual tools to avoid inadvertently adjusting for mediators and colliders; 2) delineate the pitfalls of relying on observed data and the results of statistical tests such as p-values, confidence intervals or univariate tests for confounder selection; and 3) propose the use of sensitivity analyses,

such as bias factors or confounding functions, at the analysis stage to assess the impact of unmeasured confounders. Engagement with end-users and authors of the reporting guidelines is also needed to formally test and revise to the guidelines to ensure clarity. Endorsement by journal editors and reviewers continue to play an important role in further dispelling the practice of using observed data to inform confounding.

6.7 Conclusion

Non-randomized studies represent an important portion of the medical literature and supply important evidence for practice or policy decision-making.[1] If the evidence produced from non-randomized studies for the purpose of causal inference is to be considered reliable, careful attention needs be paid to the quality of the methods aimed at addressing the various potential sources of bias such as confounding arising from the lack of randomization.

In light of the evidence presented, how has the practice of confounder selection based on statistical testing of group differences continued to "fly under the radar"? When misused, these statistical tests can mislead rather than inform evidence on the effectiveness or safety of exposures or interventions. With advances in causal inference, we are now in a position to promote better research practice by explicitly discouraging this practice and calling for more appropriate methods for confounder selection.

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6.9 Supplemental information

6.9.1 Appendix 1: Illustrative example of the practice of testing group imbalances for confounder selection

We conducted a systematic search within the Canadian Medical Association Journal, a high-impact general medical journal, of published articles in 2018 that focused on non-randomized studies comparing two or more groups to assess the impact or effect of some exposure. Using the keywords "observational", "cohort", "quasi-experimental", "historical control" and "case control", we identified 34 eligible studies. Two independent raters (N.S and M.L.B.) assessed the articles for use of statistical testing for confounder selection and resolved any disagreements through consensus.

Among the 34 studies, 8 (24%) (95% CI: 11% to 41%) employed a form of statistical testing to examine group imbalances, commonly reported through p-values or confidence intervals in the patient characteristics table. Authors either directly or indirectly reported results from these tests to inform the choice of confounders to include in their adjusted models. For example, one article reported “We selected potential confounding variables based on the literature and on p-values (< 0.2) after univariate comparisons.” Based on this example, the practice of confounder selection based on observed group differences remains present in the published literature.

6.9.2 Appendix 2: Summary of confounder, mediator and collider paths based on the hypothetical Directed Acyclic Graph in Figure 1 on the effect of transitional care (exposure) on ED visits (outcome)

Path*	Type of association	Status of adjusted* variable	Effect of adjustment
"transitional care" → ["comorbidity"] ← "ED visits"	Non-causal	"Comorbidity" is a common cause of the exposure and outcome, i.e. a confounder	Confounding correctly blocked by adjusting for "comorbidity"
"transitional care" → ["continuity of care"] → "ED visits"	Causal	"Continuity of care" is on the causal pathway between exposure and outcome, i.e. a mediator	Part of the effect of exposure on outcome erroneously blocked by adjusting for "continuity of care"
"transitional care" → ["patient satisfaction"] ← "beliefs/values" → "ED visits"	Non-causal	"Patient satisfaction" is a common effect of exposure and a predictor of the outcome, i.e. a collider	Spurious association between exposure and outcome erroneously created by adjusting for "patient satisfaction"

Footnote: ED: Emergency Department.

*Brackets around a variable indicate adjustment. Arrows represent causal associations between variables.

7 CHAPTER 7: COMPARISON OF INTERDISCIPLINARY VERSUS NON-INTERDISCIPLINARY PRIMARY CARE ON EMERGENCY DEPARTMENT AND HOSPITAL USE IN PERSONS WITH DEMENTIA: A POPULATION-BASED STUDY (MANUSCRIPT 4)

7.1 Preamble

Evidence on the effect of FHTs on health service use is scarce and has shown mixed results in the general population. There is currently no evidence of their impact in the dementia population, a cohort that stands to particularly benefit from a comprehensive, team-based approach to primary care. Studies that have compared the performance of FHTs to other models in Ontario have acknowledged limitations in accounting for possible sources of confounding. This last of four manuscripts in this dissertation builds on the previous chapters to attempt to fill two important gaps: 1) to provide new knowledge on the effect of FHTs on health service in the dementia population and 2) to create a more robust estimation of effect by minimizing the risk of bias due to confounding both at the design stage and analysis stage. This final manuscript makes use of indicators established through the framework developed in the first manuscript (Chapter 5), the cohort design and operationalization of indicators carried out in the second manuscript (Chapter 6) and the use of causal methods for confounder selection elaborated in the third manuscript (Chapter 7) as well as in the published methods brief (Appendix B). Moreover, this manuscript also makes use of underutilized methods in causal inference, namely propensity-based inverse probability weighting and propensity-score calibration, which explicitly consider measured and unmeasured confounding between study groups. This manuscript will be submitted for publication to the JAMA.

Title: Comparison of interdisciplinary versus non-interdisciplinary primary care on emergency department and hospital use in persons with dementia: a population-based study

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7.2 Abstract

INTRODUCTION: Persons with dementia are high users of the health system and are twice as likely to use the emergency department (ED) or hospital than those without dementia. Most also suffer from multiple comorbidities and are more likely to have fragmented and poor coordination of care, resulting in the overuse of health services. An interdisciplinary primary care (IPC) setting, including family physicians, nurses, nurse practitioners and other healthcare professionals, may be well suited to provide a timely, person-centered approach to managing the wide range of needs of persons with dementia. Whether access to this model of care results in less use of the ED or hospital in this vulnerable population is unknown. The wide-scale introduction of IPC in Ontario, Canada, offers a unique opportunity with which to fill this important knowledge gap.

OBJECTIVE: To compare ED and hospital use between persons newly diagnosed with dementia in an interdisciplinary primary care (IPC) versus non-IPC setting.

METHODS: Population-based, repeated cohort design using health administrative data. Overall, 95,323 community-dwelling persons 65 years old and older, newly diagnosed with dementia between 2005 and 2015 living in Ontario, Canada were followed for up to one year. The

intervention (IPC) group consisted of the subset in an IPC practice. Those belonging to a physician-only group practice were considered as the comparison (non-IPC) group. Outcomes were overall and non-urgent ED visits, overall and avoidable hospitalizations and 30-day hospital readmission. The association between IPC and study outcomes was estimated using inverse-probability weighting; sensitivity analyses for unmeasured confounding were also performed.

RESULTS: Almost half of individuals in the study belonged to the IPC group, 60% were female. Persons with dementia in the IPC group had a higher risk of having an ED visit (relative risk (RR)= 1.03; 95% CI=[1.01,1.05]) or non-urgent ED visit **(**RR=1.22; 95% CI=[1.18, 1.28]**)** in the year following diagnosis. Differences in hospitalization outcomes were inconclusive.

CONCLUSION: Among persons with dementia, access to IPC did not translate into a reduction in ED or hospital use and was found to increase non-urgent ED visits compared to non-IPC.

FUNDING: This study was supported through a Canadian Institutes of Health Research (CIHR) Vanier Canada Graduate Scholarship and a CIHR-Canadian Consortium on Neurodegeneration in Aging grant. TS was supported through funding obtained from the Canada Research Chairs program.

7.3 Introduction

Alzheimer's and other forms of dementia are complex chronic diseases with a high impact on patients, families and the health system.[1,2] Persons with dementia are among the highest users of the health system, with rates of emergency department (ED) and hospitalizations reported to be twice as high as for persons without dementia.[3] They also have a wide range of health needs placing them at increased risk of care fragmentation within the health system.[1,2,4,5] In addition to the management of cognitive, functional, social, and emotional problems associated with dementia,

the care of persons with dementia also requires management of multiple comorbidities and providing caregiver support.

Dementia has traditionally been diagnosed and managed by specialists,[1] but there is growing recognition that primary care providers may be best positioned to provide a timely, integrated and coordinated approach to dementia care.[1,6-13] As the point of first contact, they can facilitate a timely diagnosis and offer a person-centered, rather than disease-centered, approach to dementia care. Moreover, the management of dementia relies less on medication and more on the integration of non-pharmacologic interventions from a wide range of healthcare providers.[12,14] Given the scope of expertise required, an interdisciplinary primary care (IPC) approach, where family physicians work collaboratively with nurses, nurse practitioners and other healthcare professionals, has been recommended as a potentially beneficial strategy to diagnosing and managing most cases of dementia.[1,2,7-13,15]

Intervention studies have shown that IPC can improve access to integrated and coordinated care for persons with dementia compared to usual practice.[10,16-19] In turn, this may prevent conditions that lead them to use the ED or hospital such as infections, falls, delirium and exacerbation of chronic conditions.[20-22] To date, however, only a few intervention studies have evaluated the impact of IPC on ED and other health services in the dementia population.[10,18,23-25] One study showed a modest decrease in the number of ED visits[18] while three other studies reported inconclusive findings with respect to ED use, overall and avoidable hospitalization or hospital readmission.[10,24,25] Aside from intervention studies, to our knowledge, no study has examined the impact of wide-scale IPC implemented at a system-level on health service use in the dementia population.

In Ontario, Canada, close to 200 IPC practices were recently introduced by the government to improve access to care and chronic disease management.[26,27] These practices currently serve one in five residents within the province and include a wide range of health professionals.[27] This natural experiment offers an ideal setting in which to assess the effect of IPC over traditional physician-based practices on health service use in persons with dementia at a population-level.

The aim of this study was to compare ED and hospital use in persons newly diagnosed with dementia with access to interdisciplinary versus non-interdisciplinary primary care in Ontario.

7.4 Methods

7.4.1 Setting and data sources

Over two million adults 65 years of age or older currently live in Ontario, Canada's most populous province.[28] The provincial universal health insurance plan pays for all medically necessary inpatient, emergency, and physician services. The cost of prescription drugs is also covered for persons 65 years and older.

Patient information and data on the use of health services were extracted from the linked population-based health administrative data held at ICES in Ontario. ICES is an independent, non-profit research institute whose legal status under Ontario's health information privacy law allows it to collect and analyze health care and demographic data, without consent, for health system evaluation and improvement. ICES data are employed regularly for health research.[29] Data from several administrative databases were linked using unique encoded identifiers and analyzed at ICES. Data on patient demographics were extracted from the Registered Persons Database. The Client Agency Program Enrolment and Corporate Provider Database was used to link patients to the family doctor and corresponding primary care enrollment model to they belong. Physician

claims were identified through the Ontario Health Insurance Plan (OHIP). Data on ED visits and hospital admissions were obtained through the Canadian Institute for Health Information Discharge Abstract Database and National Ambulatory Care Reporting System. The Ontario Drug Benefit (ODB) and drug identification number databases provided information on dispensed prescription drugs. The use of home and long-term care services was determined through the Home Care Database, Resident Assessment Instrument, Continuing Care Reporting System and Ontario health care institutions. Admissions to long-term care were also determined through the OHIP and ODB databases. A database on vital statistics provided information on death.

This study was approved by the Research Ethics Board of McGill University in Montreal, Canada (study # A12-M42-18B). The use of ICES data in this project was authorized under section 45 of Ontario's Personal Health Information Protection Act, which does not require review by a Research Ethics Board.

7.4.2 Design and target population

A repeated cohort design was used to extract yearly cohorts of community-dwelling older persons, aged 65 years and older, in Ontario, newly diagnosed with dementia between April 1st and March 31st 2005 to 2015. We opted to focus on the period following dementia diagnosis as disease management needs and health care utilization in the year following diagnosis is high, tend to be homogeneous and focused on community-based care.[30] The index date of dementia diagnosis was based on a previously validated ICES algorithm.[31]

We restricted our target population to persons, who on their index date of diagnosis, were part of a Family Health Team (IPC group) or Family Health Organization (non-IPC group). Family Health Organizations are group-based physician practices and are one of the most common primary care patient enrollment models available in Ontario. They are also the largest of only two

models where physicians could apply to transition into a Family Health Team and receive government funding to support an interdisciplinary, team-based environment.[27,32,33] The comparison of Family Health Organizations with Family Health Teams was chosen to maximize internal validity in terms of comparability in patient, provider and geographic characteristics between study groups as well as matching on the type of remuneration method. Pre-existing differences in these characteristics across other primary care models have been previously discussed as a major source of confounding.[32,34]

Individuals with missing age, sex, health identification number, non-residents of Ontario, and/or those living in a long-term care facility on the incidence date were excluded. Persons identified in each group for one year following the diagnosis date of dementia or until long-term care admission or death, whichever occurred first.

7.4.3 Outcomes

Occurrence of an Emergency Department (ED) visit in the year following dementia diagnosis was selected as the primary outcome. As many of the most frequent reasons for ED visits in the dementia population relate to reasons potentially preventable through better access to primary care or disease management,[20] we hypothesized that access to IPC would result in a reduction in the use of ED. Secondary health service use outcomes were the occurrence of a non-urgent ED visit, hospitalization, avoidable hospitalization or 30-day hospital readmission in the year following diagnosis. A non-urgent ED visit was defined as an ED visit classified as less urgent or non-urgent according to the Canadian Acuity Triage Scale.[35] An avoidable hospitalization was defined as any hospitalization with a most responsible diagnosis of asthma, cardiac heart failure, COPD, diabetes, hypertension, angina or seizures.[36]

7.4.4 Potential confounders and other covariates

A directed acyclic graph was produced to visualize the variables involved in the causal relationship between access to IPC and use of the ED in order to identify potential confounders and predictors of ED use (eFigure 1).[37] The directed acyclic graph was informed by published literature and consultations with multiple stakeholders including patients and caregivers living in Ontario, family physicians and managers with knowledge of the primary care reforms in Ontario (39 stakeholders in total).

Data on age, income, rurality, recent immigrant status, comorbidity and resource utilization, were obtained for all persons in each cohort year and assessed at the incidence date of diagnosis. Neighbourhood level income quintiles and rurality (urban vs. rural) were determined from the 2011 national census.[38] An 'urban area' was defined as having a population of at least 1,000 and a density of 400 or more people per square kilometer.[39] Recent immigrant status was based on first registration into the Ontario health system within the last 10 years.[38] The Johns Hopkins Adjusted Clinical Group system was used to measure comorbidity and degree of resource utilization: comorbidity was defined using the Adjusted Diagnosis Group (ADG) , an aggregated number of comorbid condition types: 1-4 (low comorbidity), 5-9 (medium), 10+ (higher comorbidity); resource utilization was measured by the number of prior physician visits and by the Resource Utilization Band (RUB), a measure of overall morbidity and cost based on quintiles of expected resource use: 0 (non-users), 1 (least expected use) to 5 (highest expected use) (The Johns Hopkins ACG® System, v10).[40] Comorbidity and resource utilization were calculated using data in the two years prior to cohort entry.

Data on other identified potential confounders, including marital and caregiver status, dementia disease severity, behavioral symptoms, functional status, self-reported health status,

receipt of antipsychotic medication, were extracted for the subset of the cohorts who had received long-term home care services and for whom a Resident Assessment Instrument for Home Care (RAI-HC) was completed within three months prior to dementia diagnosis.[41]

7.4.5 Analysis

We used propensity score-based inverse probability weighting to estimate the average causal effect of IPC compared to non-IPC on the health service use outcomes.[42,43] The propensity score (PS) was calculated as the estimated probability of belonging to the IPC group, given the measured baseline covariates available for the entire population (age, income, rurality, recent immigrant status, comorbidity and degree of resource utilization). Inverse probability weighting reweights the sample based on the propensity score to create a pseudo-population in which exposure is independent of the measured confounders. This causal inference method therefore yields a marginal effect in that the measured effect is not conditional on the values of the model covariates and provides a population-level contrast.[42] Weights were calculated as 1/PS for persons in the exposed group and 1/(1-PS) for persons in the non-exposed group.[43] As the presence of extreme weights can increase the variability of the estimated effect, we then stabilized the weights by multiplying them with the prevalence of group membership for each individual.[43]

Covariate balance was verified graphically by comparing the propensity score and weight distributions in both groups and based on the standardized mean difference between groups for each covariate.[44] The presence of extreme propensity scores and weights was assessed to verify the plausibility of the causal condition of positivity. This condition requires that all persons in the study population be potentially exposable to either exposure group.[42] Practically, this implies that there should be both exposed (IPC) and unexposed (non-IPC) individuals at every level of the confounders. Extreme propensity scores (close to 0 or 1) or weights would create unstable

estimates from the inverse-probability weighting and indicate possible violations to the positivity assumption.

Risk differences and relative risks for the study outcomes were derived through bootstrap aggregating (bagging) of the effect estimates from 1,000 bootstrapped samples from the weighted sample.[45] Corresponding 95% confidence intervals were calculated as the 2.5th and 97.5th percentile of the distribution of the effect estimates.[43] The number needed to treat was calculated as the inverse of the risk difference and represents the number of cases needed to be exposed to IPC to prevent or cause one additional event. All outcomes were analyzed using the full analytical sample except for 30-day hospital readmission which was assessed in the subset of persons who had at least one hospitalization during the year.

7.4.5.1 Sensitivity analyses

Three sensitivity analyses for the primary outcome of overall ED visits were performed. First, in the construction of the directed acyclic graph, rurality surfaced as a major source of confounding, being both highly associated with exposure status[46] and ED visits.[47,48] While we included rurality in the calculation of the propensity-score, we also conducted a sensitivity analysis stratifying by rurality to account for possible residual confounding. Second, we used a causal inference method called propensity-score calibration to incorporate data on additional potential confounders and predictors available only for a subset of the population.[49] Under certain assumptions, this method reduces bias due to unmeasured confounding by leveraging richer subset data to estimate the propensity score in the main dataset.[49,50] In our study, population-level health administrative data were available for only a partial set of confounders, creating was is referred to as an "error-prone" propensity-score (PS_{EP}). A secondary data source, the RAI-HC, included baseline information on several important dementia-specific predictors of health service use identified in the directed

acyclic graph, such as disease severity, caregiver status and use of antipsychotics. These data, however, were only available within the subset of those receiving home care. Using propensity-score calibration, this enriched set of confounders in the subset data was used to estimate the so-called "gold-standard" propensity-score (PS_{GS}) in the population-level data. This estimation was conducted using regression calibration based on the following linear measurement error model in the subset data:

$$E[PS_{GS} \mid A, PS_{EP}] = \delta_0 + \delta_1 A + \delta_2 PS_{EP},$$

where A represents the exposure of interest (belonging to an IPC practice).[50]

Third, given the likelihood of residual confounding in this natural experiment evaluation, we also assessed the robustness of the estimated average causal effect for the primary outcome of overall ED visits through calculation of the E-value.[51] This value represents the minimum strength of association that an unmeasured confounder would need to have with both the exposure and the outcome, given the measured covariates, to nullify the estimated effect or reduce it to a magnitude that is deemed clinically irrelevant. [51]

All analyses were performed using SAS© software, Version 9.4, SAS Institute Inc., Cary, NC, USA, as well as *R* software, Version 3.4.2.[52]

7.5 **Results**

Between 2005 and 2015, 257,495 persons newly-diagnosed with dementia in Ontario were identified (Figure 7.1). Of these, 95,668 belonged either to the IPC or non-IPC group. The final analytical sample comprised 95,323 persons after exclusion of those with missing data (0.3%).

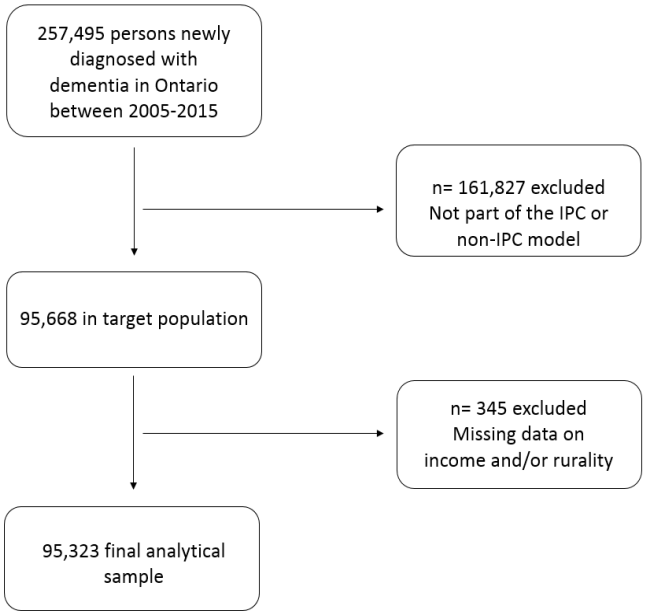

Figure 7.1: Flowchart for creation of the analytical sample

Table 7.1 presents the baseline characteristics in the original (unweighted) sample. On average, both groups were of similar age (mean (SD) ~ 81 (7) years old) and sex (~60% women). A larger proportion of those in the IPC group lived in a rural area than in the non-IPC group, with fewer physician visits in the year before diagnosis than in the non-IPC group (Table 7.1). Few variations were observed between groups in terms of recent immigration status, income, comorbidity and overall resource utilization. The distributions of the propensity score and stabilized weights were balanced between the two groups (eFigure 2). The lack of extreme values in the distributions supported the assumption of positivity. Absolute standardized mean differences for all baseline characteristics were negligible in the weighted sample (eFigure 3).

Table 7.1 Baseline characteristics of persons with newly-diagnosed dementia belonging to an interdisciplinary vs non-interdisciplinary primary care model in Ontario, Canada

Patient demographic	IPC group (N=46,830)	Non-IPC group (N=48,493)
Age, mean (SD)	81.2 (7.0)	81.4 (7.1)
Female, n (%)	27,809 (59.4%)	28,481 (58.7%)
Rural, n (%)	7,822 (16.7%)	5,093 (10.5%)
Recent immigrant, n (%)	633 (1.4%)	801 (1.7%)
Income quintile		
1 (low)	10,228 (21.8%)	9,006 (18.6%)
2	9,874 (21.1%)	9,962 (20.5%)
3	9,085 (19.4%)	9,222 (19.0%)
4	8,892 (19.0%)	9,785 (20.2%)
5 (high)	8,751 (18.7%)	10,518 (21.7%)
Adjusted diagnosis group, n (%)		
0-5 (low comorbidity)	14,788 (31.6%)	14,403 (29.7%)
6-10	21,339 (45.6%)	22,293 (46.0%)
> 10 (high comorbidity)	10,703 (22.9%)	11,797 (24.3%)
Physician visits in the year before diagnosis, mean (SD)	20.3 (14.6)	22.2 (15.8)
Resource utilization band, n (%)		
0 (low utilization)	547 (1.2%)	479 (1.0%)
1	337 (0.7%)	282 (0.6%)
2	1,969 (4.2%)	1,916 (4.0%)
3	20,662 (44.1%)	21,259 (43.8%)
4	12,338 (26.4%)	12,915 (26.6%)
5 (high utilization)	10,977 (23.4%)	11,642 (24.0%)

IPC: Interdisciplinary primary care

In the year following dementia diagnosis, persons newly diagnosed with dementia within the IPC group had a higher risk of overall ED visits (relative risk (RR)=1.03; 95 CI=[1.01,1.05]) and non-urgent ED visits (RR=1.22; 95% CI=[1.18, 1.28]). Differences in terms of overall hospitalization (RR=1.03; 95% CI=[1.00, 1.06]), avoidable hospitalization (RR=1.06, 95% CI=[0.95, 1.19]) and 30-day hospital readmission (RR=0.99; 95% CI=[0.92, 1.08]) were inconclusive.(Table 7.2)

Table 7.2 Comparison of interdisciplinary versus non-interdisciplinary primary care on emergency department (ED) and hospital use in persons newly-diagnosed with dementia in Ontario between 2005 and 2015

Outcomes	IPC group (N=46,829)	Non-IPC group (N=48,499)	Risk difference[¥] (95% CI)	Number needed to treat	Relative risk (95% CI)
Any ED visit, n (%)	15,398 (32.9%)	15,472 (31.9%)	1.0% (0.4%, 1.6%)	101	1.03 (1.01, 1.05)
Non-urgent ED visit, n (%)	4,201 (9.0%)	3,544 (7.3%)	1.7% (1.3%, 2.0%)	60	1.22 (1.18, 1.28)
Hospitalization, n (%)	7,930 (16.9%)	7,972 (16.4%)	0.5% (0.0%, 1.0%)	197	1.03 (1.00, 1.06)
Avoidable hospitalization, n (%)	624 (1.3%)	595 (1.2%)	0.1% (-0.0%, 0.3%)	927	1.09 (0.97, 1.22)
30-day readmission, n (%)	1,048 (13.2%)	1,060 (13.3%)	-0.1% (-1.1%, 1.0%)	993	0.99 (0.92, 1.08)

IPC: Interdisciplinary primary care; ED: Emergency Department
[¥] Difference in percentage points (risk in IPC group – risk in non-IPC group)

Pre-specified sensitivity analyses for the primary outcome (overall ED visits) showed that, within strata of rurality, the increased risk of ED visits was maintained among urban residents but not among rural residents (eTable 1). The effect of IPC on ED use was similar using the augmented set of confounders through propensity-score calibration (eMethods 1). In terms of sensitivity to unmeasured confounding, we found that, accounting for the other measured confounders, an

unmeasured confounder associated with both group membership and ED visits with a relative risk of 1.21 or higher could explain away the observed effect of 1.03 (eFigure 4).

7.6 Discussion

To our knowledge, this study is the first to compare interdisciplinary versus non-interdisciplinary primary care on emergency department and hospital use in persons with dementia at a population-level. We found that, in this population, access to an IPC practice did not decrease overall use of the ED and was estimated to have a 22% higher risk of non-urgent ED visits compared to non-IPC. The impact of IPC on overall and avoidable hospitalizations as well as hospital readmissions was inconclusive.

Our findings contribute to a mixed body of evidence on the impact of IPC over traditional physician-based practice on health service use. Within the general population, while some studies found that IPC was associated with a decrease in ED, hospitalization, and avoidable hospitalizations, others showed an increase in use or were inconclusive.[36,53-55] Studies within the older population or among persons with multiple comorbidities have pointed to a modest reduction in health service use, mainly ED use, for patients in team-based settings.[54,56-59]

Several factors may explain the observed lack of benefit of IPC in persons with dementia in our study. First, interdisciplinary primary care, without specific training or support for dementia care, may not be enough to impact emergency department and other health service use.[1,7] Where IPC has been shown effective in improving dementia care, these studies have been based on dementia-specific interventions including a nurse with training in geriatric or dementia care, a care navigator and/or support from cognition specialists.[10,16-19] While the IPC model in Ontario aimed to provide better prevention and management of chronic diseases, such as diabetes or hypertension, to our knowledge, dementia was never considered among these chronic conditions.[26] This is

consistent with reports citing the lack of consideration of dementia among chronic diseases in older adults,[60,61] and highlights the need to bring dementia management to the forefront of chronic disease management in primary care.[10,16,19] More targeted interventions such as case management within general IPC reforms and a better understanding of patient trajectories and decision-making leading to emergency department use in this population may be needed to help prevent avoidable health service use.[22]

Second, organizational characteristics within IPC teams may have also played a role in the findings observed. Team functioning, for example, has been shown to be an important predictor of reduced health service use including ED and hospital outcomes in early medical home implementation.[62] Clear scope of practice, collaboration, communication are cornerstones of high performing primary care teams.[63] However, as the shift to interdisciplinary care is still evolving, roles and responsibilities of team members may not yet be clear.[64] A qualitative study on the process of interprofessional collaboration in primary care team showed that interprofessional meetings could benefit from improvements in structure, patient-centredness and leadership.[65] As stated by Collier (2011), "building an effective team takes a lot more than corralling several health professionals under the same roof and hoping they get along".[66] Building trusting relationships, a positive team culture and stability is a process that can take several years.[66,67] A lack of clearly defined roles and protocols may render team members reluctant to take on the responsibility of treating patients, especially complex patients with dementia, for conditions which may require urgent care. Co-location in IPC is another important organizational feature. In Ontario, many IPC practices are not co-located, with interprofessional teams serving multiple clinics in an area. Co-location has been shown to be a key element in fostering effective team functioning.[68]

Third, persons with dementia are also particularly vulnerable to fragmented care due to their wide range of physical, cognitive and social needs.[1] Provider continuity, seeing the same professional each time, with the opportunity to establish a therapeutic, trusting relationship is therefore critical to patients and caregivers living with dementia.[1] Provider continuity has also been shown to result in improved preventive care than practice-level continuity.[69] A lack of consistency in the health care provider may inadvertently increase fragmentation of care and impede this therapeutic relationship, potentially leading patients or caregivers to seek emergency care.

Lastly, it is possible that the observed increase in ED use may be due to supply-induced demand where increased access to primary care through IPC may have fueled demand for other health services.[70] This phenomenon has also been observed in other evaluations of primary care reforms.[71]

Finally, results suggest that the effect of IPC on the risk of ED visits may be moderated by rurality, with findings pointing to a marginally beneficial effect in rural areas. Comprehensive primary care is especially important in rural and remote communities where alternative health services are limited. [72] As the ED is often more difficult to access in rural areas, IPCs embedded in underserved areas may have helped to fill unmet need and better access to care. Team building may also be facilitated in smaller communities than in urban centers.[73]

7.6.1 Strengths and limitations

This study has several important strengths. It employed novel causal inference methods at both the design and analysis stage to provide a robust assessment of IPC in dementia health service use. First, the elaboration of a causal diagram allowed for a thorough assessment of potential sources of confounding. Second, the use of inverse probability weighting to balance measured confounders

produced an estimate of the population-level average causal effect of IPC on study outcomes. Third, we conducted sensitivity analyses based on underutilized methods in causal inference, i.e. propensity-score calibration and E-values, to leverage the availability of subset data on additional confounders and ascertain the robustness of the estimated effect of IPC to unmeasured confounding.

The use of yearly cohorts of newly diagnosed persons with dementia focused our evaluation in a relatively homogeneous dementia population and disease period, minimizing additional sources of confounding due to heterogeneity in the stage of disease.[42,57] We also chose to compare two closely matched primary care models in Ontario (Family Health Teams and Family Health Organizations) to limit the number of potential unmeasured confounders, thereby increasing the internal validity of our assessment and allowing us to hone in on the "added value" of interdisciplinary teams.

Study limitations should also be considered in the interpretation of the findings. First, while we strove to maximize the internal validity by focusing the target population, this decision limited our ability to generalize the results to all newly diagnosed persons with dementia, regardless of their type of access to primary care. Heterogeneity in the exposure should also be acknowledged. For example, the time since transition into an IPC practice and organizational factors such as the team composition, functioning and co-location may have moderated the effect of IPC on the study outcomes. Information on the reasons for ED visits and hospitalizations in both groups would have also provided additional insight into the appropriateness or lack thereof in the use of hospital services. Finally, while every effort was made to limit bias due to confounding in our analysis, the possibility of bias due to unmeasured confounding remains.

7.7 Conclusion

In our study, persons with dementia in an interdisciplinary primary care setting did not use fewer health services compared to those in a non-interdisciplinary setting and may in fact access the emergency department more often for non-urgent reasons. While an interdisciplinary approach to dementia care may be ideal to manage the growing and complex dementia population, optimal team functioning and additional training and support from specialists may be needed to provide the extent of integrated and coordinated care required to reduce emergency department and hospital use in this vulnerable population.

7.8 References

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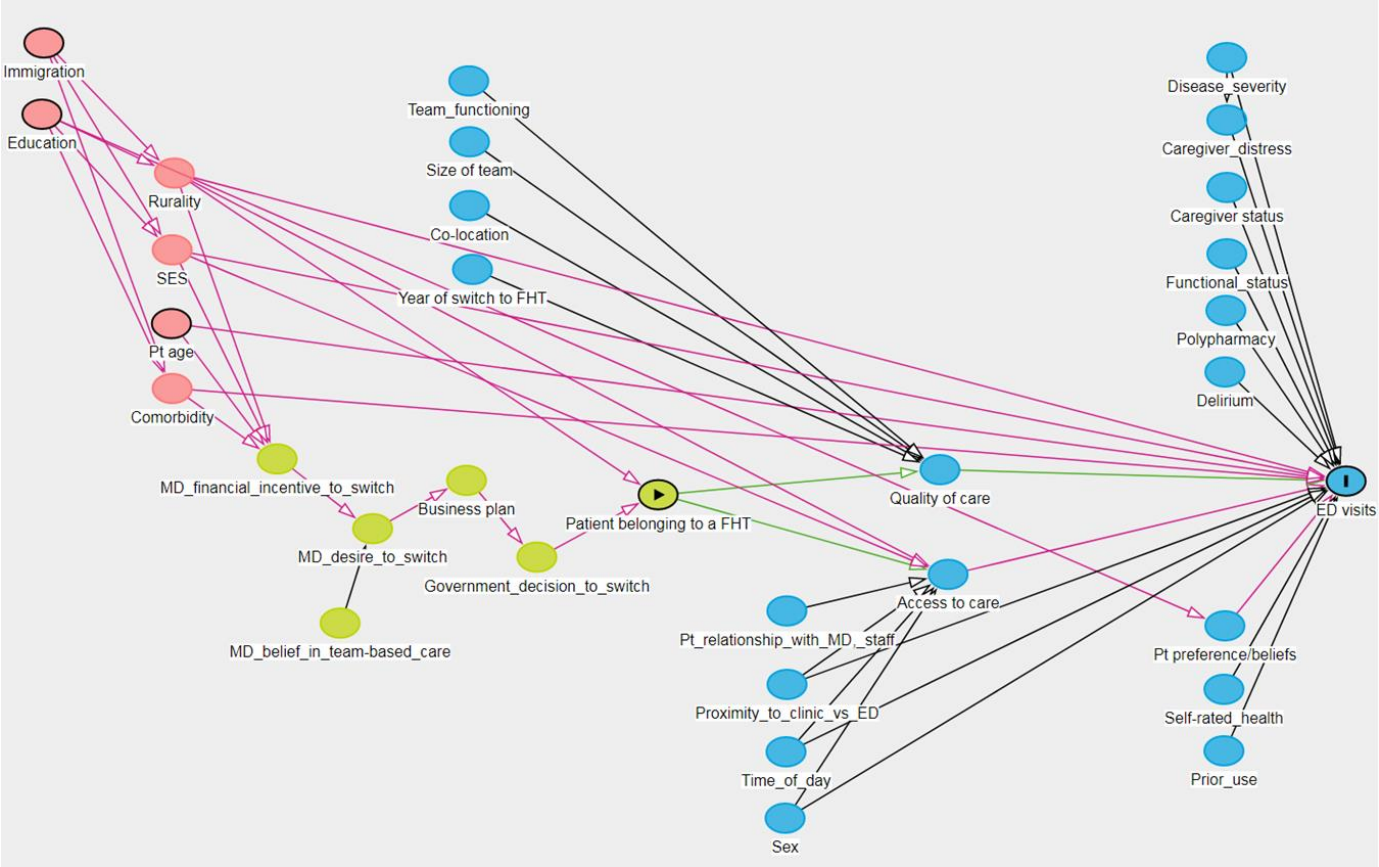
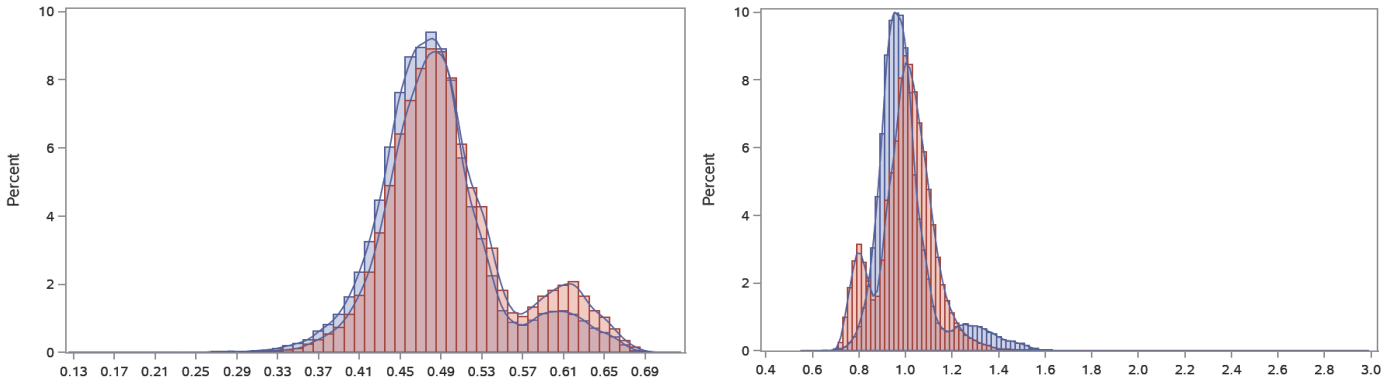
7.9 Supplemental information

7.9.1 eFigure 1: Directed Acyclic Graph of the relationship between affiliation to a Family Health Team (FHT) and the occurrence of an ED visit in the year follow diagnosis for persons with dementia in Ontario



SES: Socioeconomic status; Pt: Patient; MD: Medical doctor; FHT: Family Health Team; ED: Emergency department

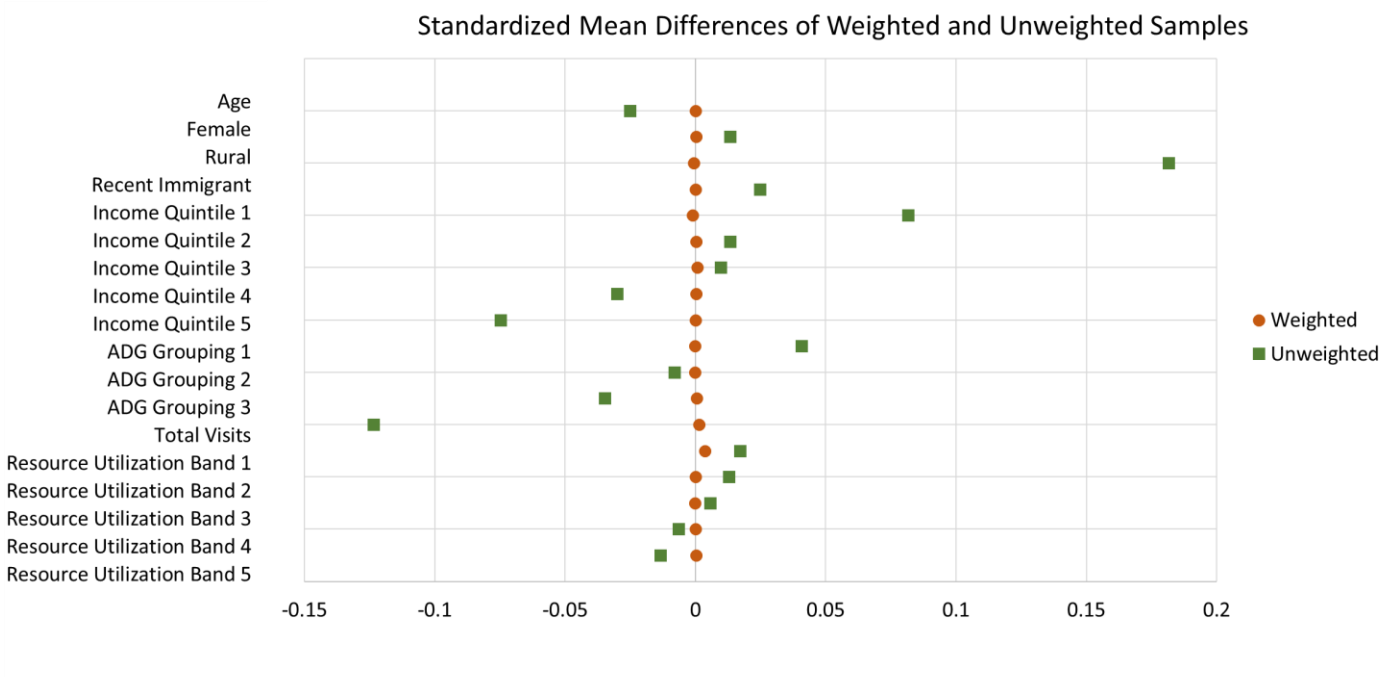
7.9.2 eFigure 2: Distribution of the propensity-score and stabilized weight in the exposed and unexposed group



Propensity-score

Stabilized weight

7.9.3 eFigure 3: Standardized mean differences in measured confounders in the unweighted and weighted sample



7.9.4 eTable 1: Effect of interdisciplinary primary care on emergency department visits in urban and rural persons newly identified with dementia in Ontario

Outcomes	IPC group (N=46,830)	Non-IPC group (N=48,493)	Risk difference[¥] (95% CI)	Relative risk (95% CI)
Among urban residents	*N=39,008*	*N=43,400*		
Any ED visit, n (%)	12,581 (32.3%)	13,568 (31.3%)	1.3% (0.7%, 2.0%)	1.04 (1.02, 1.07)
Among rural residents	*N=7,822*	*N=5,093*		
Any ED visit, n (%)	2,778 (35.5%)	1,898 (37.3%)	-1.5% (-3.2%, 0.3%)	0.96 (0.92, 1.01)

IPC: Interdisciplinary primary care; ED: Emergency Department

[¥] Difference in percentage points (risk in IPC group – risk in non-IPC group)

7.9.5 eMethods 1: Estimated effect of interdisciplinary primary care on overall emergency department visits using propensity-score calibration

The full sample consisted of all 95,343 persons newly identified with dementia in Ontario between 2005 and 2015 included in the analytical sample for which data on only a partial set of confounders (age, income, rurality, recent immigrant status, comorbidity and resource utilization) were available.

The subsample consistent of the subset of 11,246 persons who had received long-term home care services and for whom a Resident Assessment Instrument for Home Care (RAI-HC) was completed within three months prior to dementia diagnosis. In addition to the partial set of confounders already available, this subsample, through the RAI-HC, also had data on marital status, caregiver status, dementia disease severity, behavioral symptoms, functional status, self-reported health status, receipt of antipsychotic medication.

We defined:

A = Exposure (belong to an interdisciplinary primary care practice)

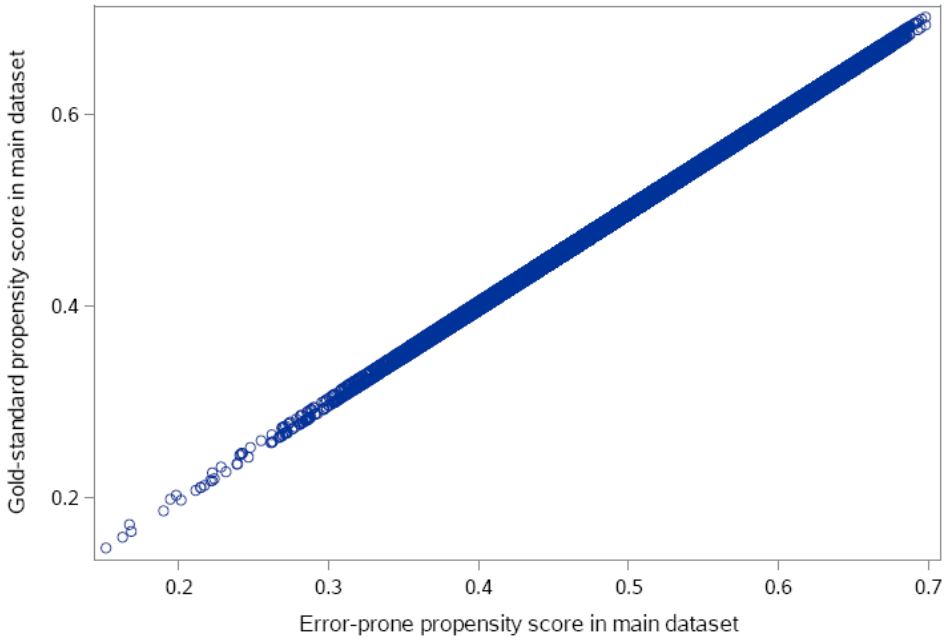
PS_{EP} = "Error-prone" propensity score based on the partial set of covariates

PS_{GS} = "Gold standard" propensity score based on the augmented set of covariates

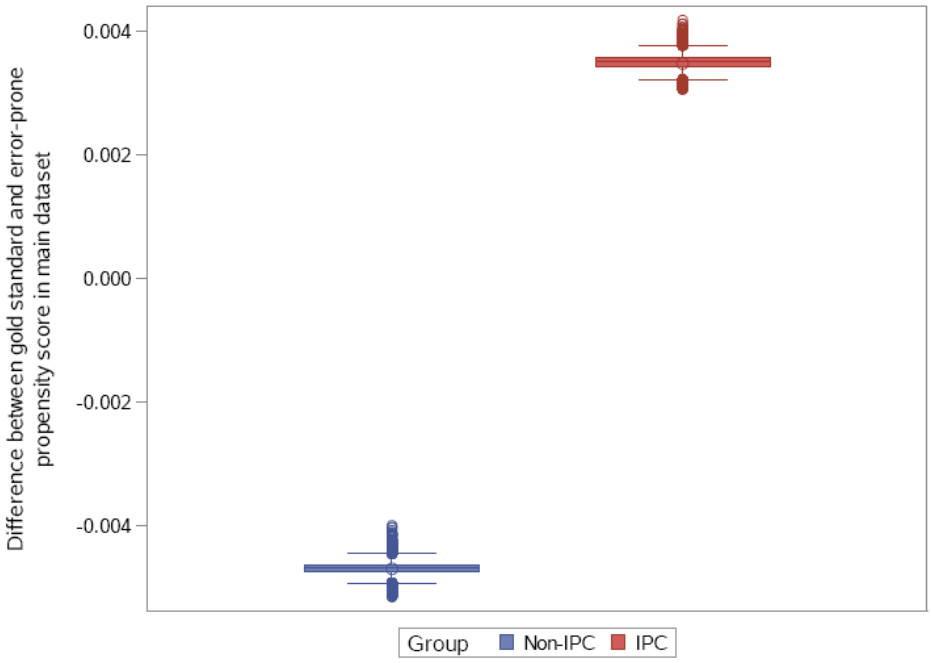
The linear measurement error model of the relationship between PS_{EP} and PS_{GS} in the subset data was calculated as:

$$E[PS_{GS} \mid A, PS_{EP}] = -0.00368 + 0.00821*A + 0.99789*PS_{EP},$$

The PS_{GS} in the full sample was estimated by applying these parameter estimates to the full sample. A comparison of the propensity-scores based on the PS_{EP} and estimated PS_{GS} in the full sample showed the scores to be highly correlated:

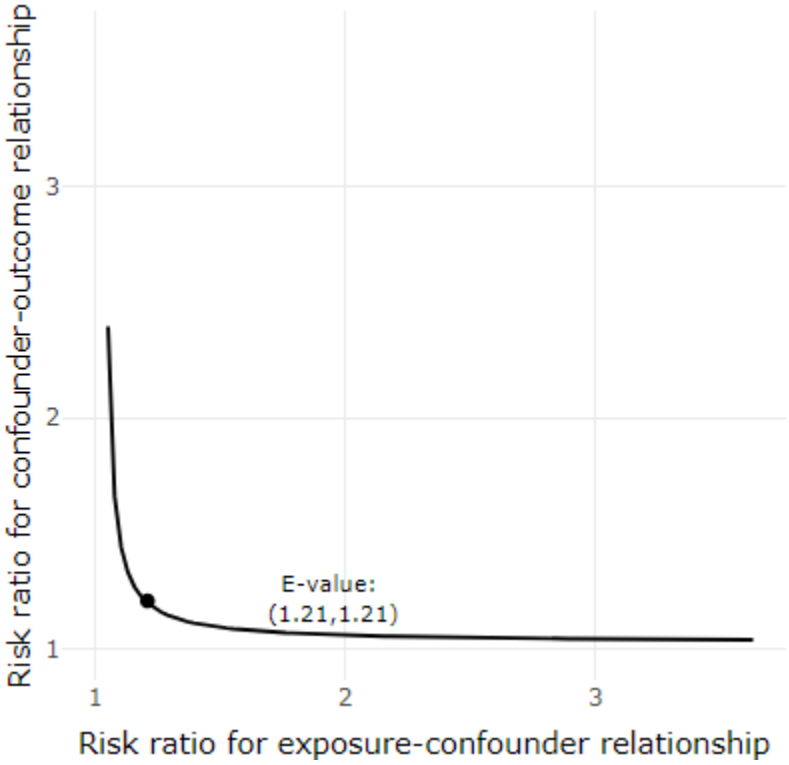


Comparing the distribution of the difference between the PS_{EP} and estimated, we found differences to be close to zero in each study group:



Finally, using the estimated PS_{GS}, the inverse-probability weighted relative risk of overall ED visits comparing IPC to non-IPC in the full sample was found to be equivalent to the original results based on the PS_{EP} (relative risk: 1.03; 95% CI: (1.01, 1.05)).

7.9.6 eFigure 4: Minimum strength of association between an unmeasured confounder and the exposure and outcome required to explain away estimated effect of IPC on the primary outcome, overall emergency department visits



8 CHAPTER 8: DISCUSSION

8.1 Summary of research

This dissertation aimed to shed light on the performance of the primary care system on health service utilization for persons with dementia, in particular, the impact of interdisciplinary primary care. To accomplish this aim, I conducted a series of four studies which built upon each other: (i) in the first manuscript, I developed a framework of dementia-relevant indicators to measure primary care performance and health service use at a population level (Chapter 4), (ii) in the second manuscript, I operationalized indicators within the framework to examine the evolution and equity in the performance of primary care and health service use for men and women with dementia since the onset of primary care reforms in Ontario (Chapter 5), (iii) in the third manuscript, I provided new guidance on the use of causal inference methods at the design and analysis stage to minimize confounding in evaluations of non-randomized interventions or policies (Chapter 6) and (iv) in the fourth manuscript, I used operationalized indicators and causal inference methods to estimate the effect of interdisciplinary versus non-interdisciplinary primary care on health service use in the dementia population (Chapter 7). Together, this research contributes new knowledge and methodological advances relevant to policy in the context of dementia and interdisciplinary primary care in Ontario and beyond.

The framework developed in the first manuscript grew out of the scarcity of and need for indicators of primary care performance adapted the dementia population that could be measured using health administrative data to monitor population-level trends and evaluate the impact of health system reforms. Existing primary care frameworks were not directly applicable to persons with dementia and current guidelines for dementia care focus mainly on clinical, process-based indicators that are not easily measurable at a population level. This framework combined a

validated primary care performance measurement framework with existing dementia quality indicators to assess dementia care and health service use across eight areas of primary care performance: accessibility, integration, effectiveness, efficiency, equity, safety, population health and patient-centeredness. Leveraging relevant indicators within both primary and dementia care constituted a novel approach to the creation of this framework. It is also the most comprehensive framework of its kind to date, offering 37 indicators that can be measured in routinely collected administrative data. The feedback of over 100 stakeholders including clinicians, researchers, managers, decision-makers and patient and caregiver representatives enabled the identification of a subset of indicators that can be used to target priorities for improvement or monitoring.

The second manuscript built upon the indicator framework and made use of health administrative data on nearly all newly diagnosed persons with dementia in Ontario between 2002 and 2015, over 300,000 persons in total. This manuscript operationalized 18 indicators from the framework to describe trends in primary care performance and health service use over time at a population-level and explored potential inequities between men and women. This study produced the most comprehensive portrait to date on the state of dementia management within primary care as well as an explicit and detailed exploration of sex differences in health service use in this population, addressing significant knowledge gaps in dementia research. Trends in most indicators were stable over time and similar in both and women; however, difference in some indicators did emerge. Increases in access to an interprofessional team and home care use and decreases in long-term care admissions were observed and likely reflected underlying system-wide reforms such as the introduction of Family Health Teams and investments in home care. Greater hospital and fewer home care use was seen in men than women.

In the fourth manuscript, the effect of interdisciplinary versus non-interdisciplinary primary care on emergency department and hospital use was ascertained using advanced causal inference methods. This final manuscript built upon the prior manuscripts in this dissertation by making use the operationalized framework of indicators and administrative data as well as prior knowledge on the feasibility and appropriateness of the study design and data for the study. Causal inference methods included the use of directed acyclic graphs, inverse probability-weighting based on the propensity-score for estimation of the population average causal effect, and propensity-score calibration and the E-value for sensitivity analyses. Directed acyclic graphs and bias factors like the E-value were proposed in the guidance published in the third manuscript to explicitly identify and handle measured and unmeasured confounding, a bias that is ubiquitous with non-randomized studies such as natural policy experiments.

Results from the impact evaluation in the fourth manuscript showed that, in the year following dementia diagnosis, persons with dementia belonging to an interdisciplinary primary care group (Family Health Teams) did not have fewer emergency department visits or hospital admissions than those belonging to a non-interdisciplinary primary care group (Family Health Organizations). Moreover, the risk of non-urgent emergency department visits was more than 20% higher in patients with access to interdisciplinary care. Sensitivity analyses on the effect of interdisciplinary primary care on overall use of the emergency department using an augmented set of confounders through propensity-score calibration showed similar results, but sensitivity analyses using the E-value showed that results may be sensitive to unmeasured confounding. Overall, this study was the first to estimate the effect of interdisciplinary primary care on health service use in persons with dementia. It is also the first evaluation of interdisciplinary primary care

teams to use a combination of novel causal inference methods to address methodological limitations from prior evaluations.

8.2 Implications for Practice and Policy

The evidence provided in this dissertation has several implications for practice and policy. First, while men and women with a recent dementia diagnosis appear to have mostly similar patterns of health service use and management within primary care, the assessment of equity in dementia care and health services planning remains essential. Moreover, sex differences seen with respect to hospital use and home care may reflect underlying sex and gender-related health and sociodemographic factors and deserve further exploration to identify to what extent these factors contribute to the trends observed. Consideration should be given to better adapting interventions to sex specific needs to avoid or postpone hospital use. Finally, a better understanding of unmet needs in both sexes is needed to determine whether the differences observed reflect that needs are equitably being met or not.

Second, given these mostly unchanged patterns of primary care performance over time and the negative findings on the impact of interdisciplinary primary care on emergency department and hospital use, this evidence suggests that recent primary care reforms in Ontario, including the introduction of close to 200 Family Health Teams, have not yet been able achieve a positive impact on curbing potentially avoidable health service use in persons with dementia. These findings have several implications. First, Family Health Teams alone, without specific programs, training or support for the management of dementia, may not be sufficient to affect change in health service use in this complex population. Evidence to date supporting the value of interdisciplinary primary care in improving dementia care has been related to dementia-specific interventions.[15,100-105] While Family Health Teams were implemented with a range of programs to better prevent and manage

chronic disease, such as diabetes or hypertension, to our knowledge, dementia was never considered among these chronic conditions. This is consistent with reports citing the lack of representation of dementia among key chronic diseases in older adults.[131,132] Second, Family Health Teams are relatively new and may not yet have reached their full potential. Evidence in the general population also shows a lack of effectiveness of Family Health Teams in reducing health service use.[27-30,77,94] Teams may need additional time and support to help define roles and responsibilities, build trust and foster communication and coordination of care in order to achieve optimal team functioning.[29,91,133] The importance of provider continuity and co-location should also be important considerations in future policy decisions to strengthen interdisciplinary primary care.[134-137] Overall, the recognition and inclusion of dementia in chronic disease management and support to optimize team functioning may be needed before any impact of interdisciplinary primary care on emergency department and hospital use can be seen in persons with dementia at a population-level.

8.3 Implications for Health Services Research

This research contributes to the advancement of the science and practice of health services research by making causal inference methods more accessible in two important ways: 1) by creating guidance for clinicians and researchers on the use of these methods in designing and analyzing non-randomized studies in program and policy evaluation and 2) by demonstrating how they can be used in practice to achieve a robust impact evaluation that leverages conceptual knowledge and minimizes bias. Future evaluations should be more explicit on sources of confounding, data sources needed to measure them and consider limiting the scope of their comparisons to a subset of population where conditions for causal inference are more likely to be met.

The framework developed and operationalized in the first and second manuscript may also provide decision-makers and researchers with dementia-relevant indicators that can be easily implemented to support decision-making through timely monitoring of dementia care performance, equity in the management of dementia and future evaluations of IPC and dementia initiatives such as the Ontario dementia strategy.[138]

8.4 Future Directions

This work highlights several future directions for health services dementia research. First, additional work is needed to complete the operationalization of the indicator framework developed in the framework. Operational definitions for indicators that rely on billing codes, such as visits for counselling, are complex as codes vary according to billing practices. An assessment of variations in billing practices across the province is needed in order to provide a thorough measurement of these indicators. Second, the descriptive assessment of sex differences in health service use within the dementia population could not conclude on how sex and gender may be influencing, or not, these patterns of use. A qualitative study with patients and caregivers living with dementia may help shed light on the results of this assessment and uncover potential inequities in the management of men and women with dementia. Third, this thesis demonstrated the feasibility and advantage of using causal inference methods to examine health policies. More guidance and knowledge translation is needed to bridge the gap between the fields of epidemiology and biostatistics and the fields of primary care and dementia research to demystify causal inference methods and promote their use within the armamentarium of health services dementia researchers. The use of propensity-score calibration, in particular, stands out as a highly promising avenue for leveraging additional data sources to improve the evaluation of natural policy experiments. However, assumptions underlying this method are still being developed and require further study.

Finally, further research is needed to confirm the reasons why interdisciplinary care increased non-urgent emergency department visits. Qualitative interviews with patients and caregivers may help to shed light on their reasons for choosing to visit the emergency department and whether these reasons may relate to sub-optimal organizational characteristics within team-based practices, increased care fragmentation or supply-induced demand for additional services. Studies linking organizational survey data on Family Health Teams with administrative data may be able to examine the moderating and mediating factors involved in the relationship between interdisciplinary primary care and health service use.

8.5 Conclusion

This doctoral work aimed to provide a better understanding of differences in dementia management and health service use for men and women with dementia and examine the role of interdisciplinary primary care in order to target and inform policies and practices for optimal primary care management and health service use within community-based persons with dementia. The four studies conducted in this dissertation demonstrated that it was feasible to develop, operationalize and assess a comprehensive set of population-based indicators to measure changes in the performance of primary care and in health service use and to apply innovative causal inference methods in estimating the effect of a natural policy experiment. Overall, findings suggest that more support within primary care teams may be needed to positively affect health service use in both men and women with dementia. This work will open avenues for future health services dementia research and increase the use of causal inference methods in health policy evaluation.

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10 APPENDIX A: ETHICS APPROVAL

McGill

Faculty of Medicine
3655 Promenade Sir William Osler #633
Montreal QC H3G 1Y6

Faculté de médecine
3655, Promenade Sir William Osler #633
Montréal, QC H3G 1Y6

Fax/Télécopieur: (514) 398-3870
Tel/Tél: (514) 398-3124

CERTIFICATION OF ETHICAL ACCEPTABILITY FOR RESEARCH INVOLVING HUMAN SUBJECTS

The Faculty of Medicine Institutional Review Board (IRB) is a registered University IRB working under the published guidelines of the Tri-Council Policy Statement, in compliance with the Plan d'action ministériel en éthique de la recherche et en intégrité scientifique (MSSS, 1998), and the Food and Drugs Act (17 June 2001); and acts in accordance with the U.S. Code of Federal Regulations that govern research on human subjects. The IRB working procedures are consistent with internationally accepted principles of Good Clinical Practices.

At a full Board meeting on 14 January 2019, the Faculty of Medicine Institutional Review Board, consisting of:

Alain Brunet, PhD	Kelly Davison, MD
Carolyn Ells, PhD	Catherine Lecompte
Athanasios Katsarkas, MD	Blossom Shaffer, MBA
Lucille Panet-Raymond, BA	Sylvia Villeneuve, PhD
Alexandra Pasca, LL.M.	

Examined the research project **A12-M42-18B** titled: *Impact of interdisciplinary primary care on the health service utilization of persons with dementia in Ontario*

As proposed by: Dr. Isabelle Vedel (Applicant) to _____ (Granting Agency, if any)

And consider the experimental procedures to be acceptable on ethical grounds for research involving human subjects.

14 January 2019 (Date) — Chair, IRB — Dean/Associate Dean

Institutional Review Board Assurance Number: FWA 00004545

McGill

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December 10, 2019

Dr. Isabelle Vedel
Department of Family Medicine
5858 Chemin Côte-des-Neiges, Suite 300
Montreal QC H3S 1Z1

RE: IRB Study Number A12-M42-18B
Impact of interdisciplinary primary care on the health service utilization of persons with dementia in Ontario

Dear Dr. Vedel,

Thank you for submitting an application for Continuing Ethics Review for the above-referenced study.

The study progress report was reviewed and Full Board re-approval was provided on December 9, 2019. The ethics certification renewal is valid until **December 18, 2020.**

The Investigator is reminded of the requirement to report all IRB approved protocol and consent form modifications to the Research Ethics Offices (REOs) for the participating hospital sites. Please contact the individual hospital REOs for instructions on how to proceed. Research funds may be withheld and / or the study's data may be revoked for failing to comply with this requirement.

Should any modification or unanticipated development occur prior to the next review, please notify the IRB promptly. Regulation does not permit the implementation of study modifications prior to IRB review and approval.

Regards,

Roberta M. Palmour

Roberta Palmour, PhD
Chair
Institutional Review Board

cc: Nadia Sourial
A12-M42-18B

2019-05-17

Dr. Isabelle Vedel
c/o: Geneviève Arsenault-Lapierre
email: genevieve.arsenault-lapierre@mail.mcgill.ca

Object: Project MP-05-2018-680, 17-021 - Continuing review ethics approval.
Assessing Care Models Implemented in Primary Health Care for Persons With Alzeimer's Disease and Related Disorders

Dear Dr. Vedel,

Thank you for the continuing review submission of the research project indicated above.

A delegated review of the research project was provided by member(s) of theFirst-Line Psychosocial of the Integrated Health and Social Services University Network for West-Central Montreal. The research project was found to continue to meet scientific and ethical standards for conduct at the Integrated Health and Social Services University Network for West-Central Montreal.

The following documents were approved or acknowledged by theFirst-Line Psychosocial of the Integrated Health and Social Services University Network for West-Central Montreal:

- Continuing Review Submission Form F9 - 8252
- Approved documents (Appendix L_FIC_conf attendees_ENG_V4fev2019.pdf)
- Approved documents (Appendix L_FIC_conf attendees_FR_V4fev2019.pdf)
- Approved documents (Appendix L_FIC_stakeholders_ENG-papier_V4fev2019.pdf)
- Approved documents (Appendix L_FIC_stakeholders_FR-papier_V4fev2019.pdf)
- Approved documents (CCNA_full protocol_V22jan2019.docx)
- Approved documents (Appendix L_FIC_stakeholders_ENG_V4fev2019 (1).docx)
- Approved documents (Appendix L_FIC_stakeholders_FR_V4fev2019 (1).docx)

This will be reported to the First-Line Psychosocial and will be entered accordingly into the minutes of the next meeting, to be held on June 7, 2019.

The approval of the research project is valid until 2020-05-17.

In addition to the WCMH - Jewish General Hospital site, it is expected that the ethical approval of this project granted by the MBM Committee will be applied to the following establishments for the following investigators:

- CIUSSS de la Capitale-Nationale
- CIUSSS de l'Estrie-CHUS
- CIUSSS du Centre-Sud-de-l'Île-de-Montréal
- CISSS de Laval
- CISSS de l'Abitibi-Témiscamingue
- CISSS de Chaudière-Appalaches
- CIUSSS de l'Ouest-de-l'Île-de Montréal

All research involving human subjects requires review at recurring intervals. To comply with the regulation for continuing review of at least once per year, it is the responsibility of the investigator to submit an Annual

Privacy Impact Assessment
ICES Project

PC Administrative Tracking

Event	Date
Receive from PI/FA	May 10, 2017
Forward to PL	May 12, 2017
Receive from PL	May 17, 2017

About this Form: *This ICES Project PIA Form is used to request and document, or amend, approval for the collection and use of ICES Data for an ICES Project. It is current and complete as of the date of Privacy approval below. The exception is amendments that do not involve changes to Project Objectives or requests to add further data to Schedule 1, which are effective upon submission to ICES' Privacy Office. Failure to provide all required information will delay approval. Do not use this form to request permission to disclose ICES Data; contact the Privacy Office.*

A. WHY ARE YOU COMPLETING THIS FORM?

To request permission to conduct a new ICES Project

For new ICES Projects, proceed to Section B

☐ Add an ICES Data Holding	☐ Add an ICES Scientist or ICES Collaborating Researcher
☐ Remove an ICES Data Holding Specify:	☐ Change estimated Project Completion
	☐ Change Project Objective
☐ Collect additional data for this ICES Project	Other (Specify):

B. PROJECT

Title (As appears on the PAW)	Evaluating the impact of primary care reforms in Ontario on the quality of care and health service use of persons with dementia					
TRIM	2018	0900	990	000		
Project Contact	Jacob Etches				**Email**	jacob.etches@ices.on.ca

C. PROJECT SUMMARY & OBJECTIVES

Summary (Attach proposal and describe in 50 words or fewer)

☑ Proposal attached

This project examines population-level trends in the quality of care and health service use for newly diagnosed persons with dementia, overall and contrasted by sex, in the last 15 years in Ontario.

Project Objectives (Indicate EPM for each objective that will Evaluate or enable Planning or Management of the health system or health services in Ontario)

1.	To describe variations in indicators of quality of care and health service use in the year following diagnosis of dementia among community-dwelling older adults in Ontario in the last 15 years	EPM
2.	To examine differences in these patterns of care and service use between men and women with dementia in Ontario	EPM

Project Activation Worksheet (PAW)

☐ Rapid Response Request (Available for Knowledge User requests that require approval within 1-2 business days)

Internal Office Use: Administrative Tracking

		DATE (MMM/DD/YYYY)
Date received from PI/FA		
Sent to PL/Designate for signoff (if applicable)	☐ N/A	
Signed by PL/designate (if applicable)	☐ N/A	
Sent to Finance		
TRIM # received (entered in Section E)		

Person submitting the PAW:	Jacob Etches	jacob.etches@ices.on.ca
Project Contact:	Jacob Etches	jacob.etches@ices.on.ca

A PAW is required for all ICES Projects and will result in a TRIM number, cost centre and ICES staff to be assigned. The PAW (including budget, 1 page protocol/DCP/approved grant proposal and grant award letter) should be submitted to the Research Program Coordinator (RPC) or Facility Administrator. Grant award letters are required for newly awarded grants only. For multi-project grants, funding confirmation need be submitted only once.

SECTION A: PROJECT INFORMATION (for MSS)

ICES Research Program:	(HSPE) Health System Planning and Evaluation
DAS Project Type (if applicable)	Select DAS Project Type (if applicable):
ICES Site:	ICES Central
Principal Investigator (PI):	Nadia Sourial
PI Type:	ICES Student/Fellow/Post-Doctoral Trainee/Visiting Scholar
PI Email:	nadia.sourial@mail.mcgill.ca
Responsible ICES Scientist:	Susan Bronskill
Anticipated Project Start Date:	May 1 2017 ☐ N/A

Do you require ICES Research and Analysis staff to assist with the completion of the ICES Project Privacy Impact Assessment (PIA) or Data Set Creation Plan (DCP) forms?

These staff hours will be billed (TRIM'ed) to your project.

Please specify from drop down list below who will complete the required ICES forms.

☑ YES ☐ NO — Select who will complete required ICES forms (PIA/DCP)

PROPOSAL TITLE (Limit to 147 characters)

Evaluating the impact of primary care reforms in Ontario on the quality of care and health service use of persons with dementia

PROPOSAL Short Title or Acronym (if proposal has a known acronym to be used) is required for project folder creation. Limit to 55 characters.

Impact of primary care reforms on persons with dementia

PROPOSAL DESCRIPTION

There is global consensus that primary care physicians are best positioned to provide timely access to patient-centered care, with support from specialists, for patients with multiple chronic diseases, including patients with dementia. In the last two decades, several Canadian provinces, in particular Ontario, have invested heavily in reforms to build up primary care.In 2001, Ontario began rolling out new primary care models (PCMs) with a range of remuneration methods, incentives and interdisciplinarity: Family Health Networks (2001, blended capitation, extended hours, incentives), Family Health Groups (2003, blended fee-for-service, extended hours, incentives), Family Health Organizations (2005, blended capitation, extended hours and services, incentives), and Family Health Teams (2006, blended capitation, extended hours, incentives, interdisciplinary teams). These four models were added to the traditional models of primary care including Community Health Centres (salaried interdisciplinary

SECTION B: ICES PROGRAM and SITE REVIEWS

Please complete this section and indicate which applies to this proposal. *Select only one option.*

☑	Stand-alone grant (e.g. operating grant) reviewed through the ICES Grant pre-submission process; ***scope and objectives have not changed since review***	Complete section D
☐	Stand-alone grant (e.g. operating grant) reviewed through the ICES Grant pre-submission process; ***scope and objectives have changed since review***	Complete section C & D
☐	Project funded by an umbrella Foundation Grant, Team Grant or similar	Complete section C & D
☐	Proposal reviewed by AHRQ Committee	Complete section D
☐	Proposal reviewed by a local site **and** does not require ICES Central resources	Complete section D
☐	A contract/research services agreement is in place for this proposal and has been reviewed by a Program/DAS	Complete section D
☐	PAW being submitted to amend existing information in MSS (e.g. to update or change billing information, project site transfer, etc.)	Complete section D
☐	None of the above apply	Complete section C & D

11 APPENDIX B: PAPER PUBLISHED ON INTRODUCTION TO CAUSAL INFERENCE FOR NON-RANDOMIZED STUDIES IN PRIMARY CARE

Sourial N, Longo C, Vedel I, Schuster T. (2018). Daring to draw causal claims from non-randomized studies of primary care interventions. Family Practice. 35(5): 639–643.

Daring to draw causal claims from non-randomized studies of primary care interventions

Nadia Sourial[a], Cristina Longo[a], Isabelle Vedel[a], Tibor Schuster[a].

a. Department of Family Medicine, McGill University, Montreal, Canada

Introduction

Primary care interventions, including new primary care policies or quality improvement programs, are often evaluated without the use of randomized controlled trials (RCTs), as randomizing who receives the intervention can be infeasible for many practical, ethical, and political reasons (1). In these cases, evidence on the effect of the intervention must stem from non-randomized studies (e.g. quasi-experimental studies, natural experiments or observational studies) which presents many complexities to isolating the causal effect from the many sources of bias and threats to validity including concurrent events, lack of comparability across groups, selection bias, etc. Faced with these barriers, researchers often conservatively accepted that determining causal effects in such non-randomized settings is unattainable and have become complacent with claims of "association" rather than "causation".

Recent methodological developments in the causal inference literature, however, have shown that, if specific conditions hold, the causal effect of non-randomized interventions can still be reliably estimated (2, 3). These advancements represent a paradigm shift in how we approach omnipresent causal questions, opening up the possibility of making causal claims even with non-randomized data. Methods developed under this causal framework are becoming increasingly used in many other fields including epidemiology (4), pharmacosurveillance (5, 6) and health

economics (7), but have yet to permeate into mainstream primary care research. Given that many primary care studies are conducted outside the randomized setting, causal inference methods offer enormous potential to this field including applications in practice-based research, health services research, pragmatic trials, and quality improvement initiatives.

This methods brief provides 1) an overview of the causal inference framework and its underlying conditions and 2) practical examples of how its analytical methods can be applied to reduce bias in the estimation of the effect of common primary care interventions. For more in-depth readings, seminal references are provided throughout the text.

What is the causal inference framework?

Let's consider an example where we want to evaluate the effect of a new primary care intervention, say the introduction of interdisciplinary primary care teams, on an outcome, such as chronic disease management. Suppose this intervention occurred naturally in several different practices and we wish to compare disease management for patients in these team-based practices (intervention group) to patients in solo practices (comparison group). In order to attribute changes in disease management to the intervention and claim a *causal effect*, we ideally wish to know for every patient: "Would their disease management be the same whether they received interdisciplinary care or not?". From Figure 1, this question equates to the theoretical situation on the bottom, where we would expose everyone to the intervention and then, in a counterfactual world, withhold the intervention from everyone, and compare their outcome on disease management. In reality, of course, we can only observe the situation at the top, where each patient either receives the intervention or not, and so, we can only observe the outcome for the exposure

actually received. The fact that we can only ever observe one of the two potential outcomes for each person is what is called the "fundamental problem of causal inference" (2).

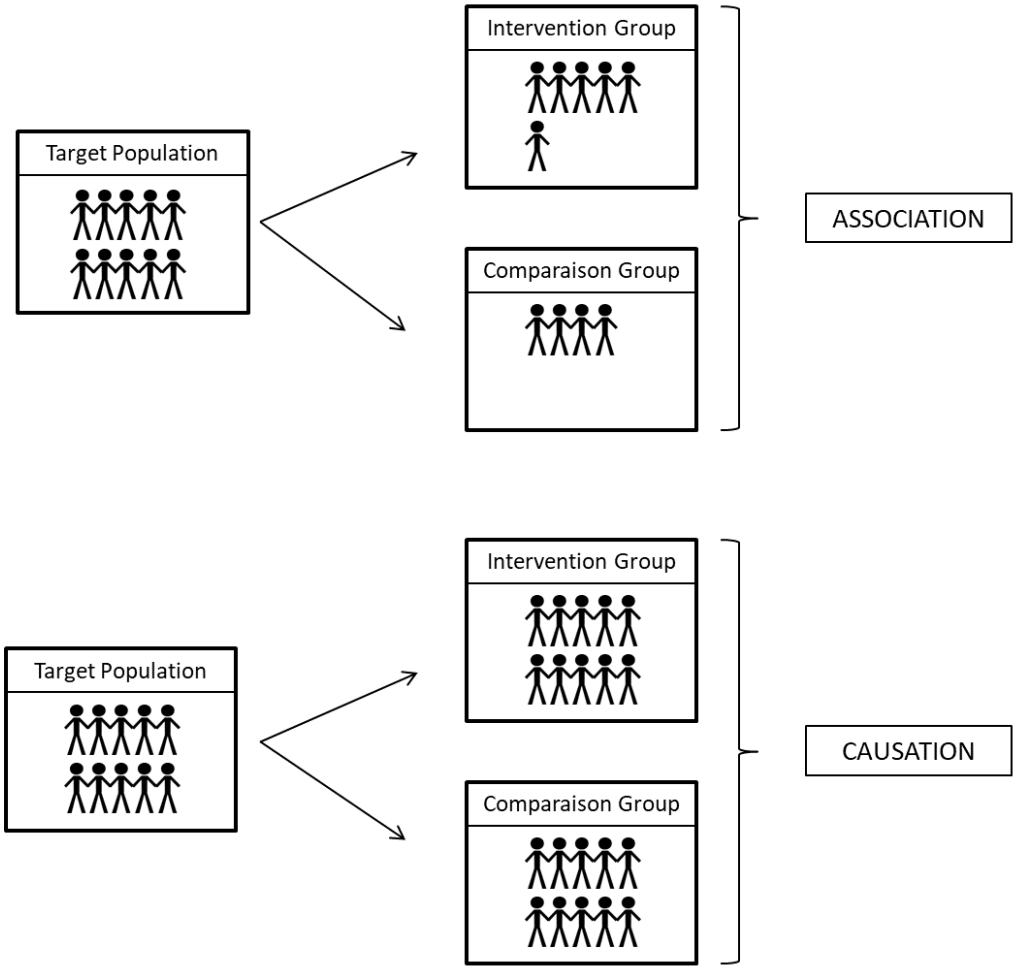

Figure 1: Illustration of causation vs association

The causal inference framework (also known as the potential outcomes framework) formalizes the once vague concept of causality, using explicit mathematical notation to define and address these causal concepts. It maps out the conditions needed to use the observed data at the top of Figure 1 to infer to the theoretical situation on the bottom of Figure 1, essentially allowing

us to extend from “association” to “causation”. In the case of non-randomized studies, this framework shows that the key is to consider these studies as if they were pseudo-randomized (2). While this may seem out-of-reach, this requirement simply relies on three conditions being met: 1) consistency, 2) positivity and 3) exchangeability (2).

1. *Consistency*

Consistency refers to the condition that the intervention to be evaluated be well-defined and specific enough to warrant an unambiguous and meaningful estimate of the causal effect. In other words, the intervention should be the same for all study subjects and be implemented in the same way. For example, the intervention on interdisciplinary primary care teams should specify the exact team composition (doctor, nurse, social worker, pharmacist, etc) and clinics applying the intervention should adhere to the same intervention guidelines on roles and responsibilities of the team members, schedule for team meetings, etc. Otherwise, variations on the intervention would make it difficult to attribute a single causal effect.

2. *Positivity*

Positivity requires that all persons in the study population be *potentially exposable* to the intervention and comparison group. In our example, this would imply that any patient from the target population could, in theory, have received the interdisciplinary care intervention. One scenario where this condition might be violated would be in the case of regional barriers, for example, where only patients with primary care providers in urban areas could access this new intervention.

3. Exchangeability

The exchangeability condition, also known as "no unmeasured confounding", refers to the interchangeability of patients between the intervention and comparison group. This means that if we swapped the patients in intervention group and those in the comparison group, the expected difference in the outcome would remain unchanged (2). While this is theoretically guaranteed under randomization, in the case of non-randomized allocation of interventions, this is often not justifiable as there are usually imbalances or systematic differences in the characteristics of the patients in each group. For example, patients receiving the interdisciplinary care intervention may be older, less educated, have more comorbidities, etc. When systematic imbalances of covariates across intervention groups are causally linked to the outcome of interest, we call them "confounders". A key mathematical result within the causal inference framework is that if we can control for all existing confounders, then receiving the intervention or not becomes independent of any variables that may cause the outcome, as is the case in an RCT, allowing for the estimation of a causal effect.

There are other scenarios when the exchangeability condition may be violated. Adjusting for variables that are on the causal pathway between the intervention and outcome (mediators) or variables that are affected by both the intervention and an unmeasured covariate of the outcome (colliders), can actually induce rather than reduce bias in the estimate of the effect (8). A diagram of the causal relationships between variables, known as a Directed Acyclic Graph (DAG) (Figure 1), can help to distinguish between these types of variables and determine which analytical approach is needed to address the different sources of bias. Practical examples of DAGs and these analytical approaches are presented in the next section.

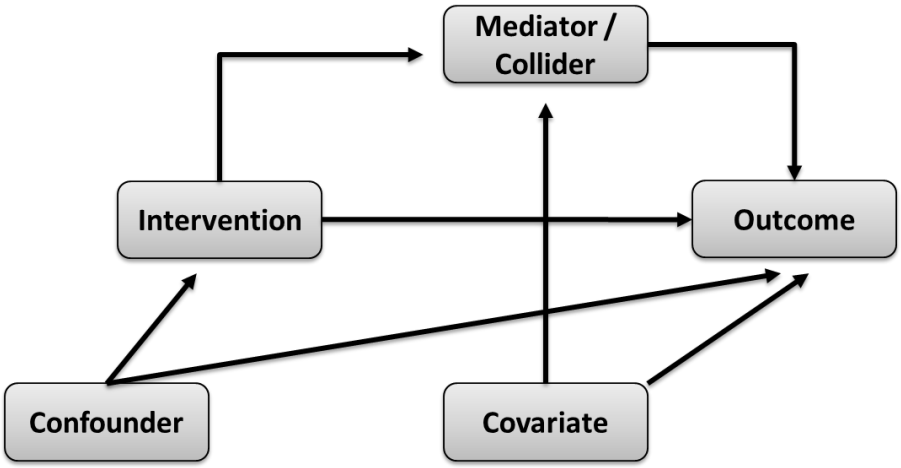

Figure 2: Causal relationships between variables in a Directed Acyclic Graph (DAG)

Applications of causal inference methods in primary care research

Now that we have reviewed the conditions (consistency, positivity and exchangeability) for the estimation of causal effects of non-randomized interventions, we now describe three causal inference methods that can be used to answer relevant primary care research questions that are implicitely or explicitely causal in nature. These methods address various threats to the exchangeability condition in ways that conventional regression techniques cannot.

1. *Marginal Structural Models*

Marginal structural models (MSMs) were primarily developed to overcome the limitations of conventional confounder adjustment methods with respect to biases arising from so-called time-dependent confounding (2, 3). A recent published article by Héroux et al. (2014) aimed to assess the impact of patient enrolment into an integrated primary care delivery model (Family Medicine

Groups or FMGs) on emergency department (ED) visits in Québec over a 3-year follow-up period (9). The presence or absence of chronic illnesses were believed to be confounders, affecting both patient enrolment into a FMG and the likelihood of ED visits. In addition, patient enrolment into a FMG was, in turn, also thought to influence chronic illness. When a confounder, like chronic illness, changes over time because it is influenced by prior exposure to an intervention (FMG), it also acts as a mediator in the causal pathway (Figure 3). When unmeasured covariates (underlying health status) are present (Figure 3), this creates a phenomenon known as 'time-dependent confounding' (10). Conventional regression adjustment for time-dependent confounders would induce a biased estimation of the intervention effect.

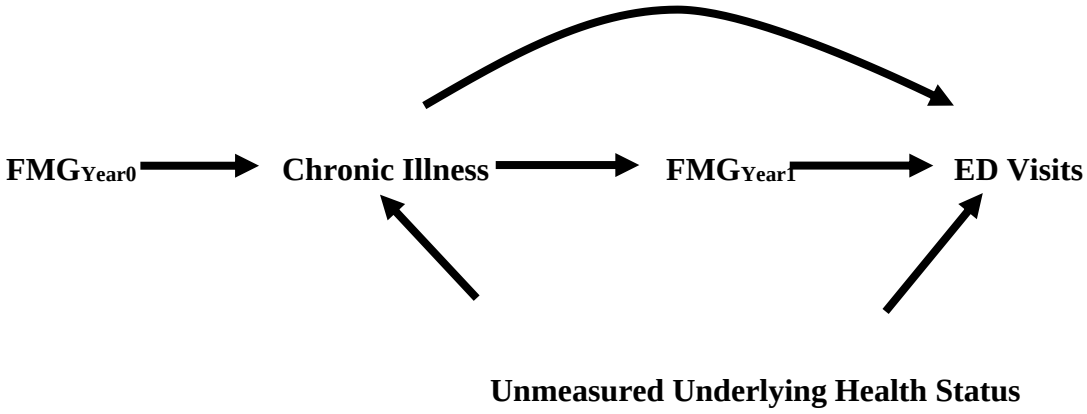

Figure 3: An example of time-dependent confounding when assessing the impact of Family Medicine Groups (FMGs) on emergency department (ED) visits

To address this issue, Heroux et al. (2014) analyzed their data with a MSM and compared their results to a conventional regression approach. The MSM uses a weighting approach to emulate the theoretical population shown on the bottom of Figure 1. This weighting, which is often derived from propensity scores, balances exposed and unexposed patients across all measured confounders, thus ensuring that the exchangeability condition holds (4). In the study, the

conventional regression model estimated a biased risk ratio of 0.979 (95% CI 0.963-0.995), while the MSM produced an unbiased risk ratio of 0.933 (95% CI 0.909-0.958). This example demonstrates the advantage of using MSMs in longitudinal studies where the exposure to the intervention and the confounders can vary over time.

2. Instrumental Variable Analysis

Instrumental variable (IV) analysis represents another important tool for causal inference in primary care research. Recall that the exchangeability condition requires that we know and measure all confounders of the relationship between an intervention and outcome. What happens when we know there are important confounders we cannot measure? IV analysis provides a "work-around" to estimate the causal effect of interventions, even in the presence of unmeasured confounding (11). It does this by finding an external variable, the IV, that satisfies the following assumptions: 1) it is strongly predictive of who receives the intervention; 2) it causes the outcome only through its relationship with the intervention; and 3) it cannot be influenced by other unmeasured predictors of the outcome (Figure 4) (12). Since the IV allows for the estimation of an intention to treat effect (blue arrow), it circumvents the bias introduced by unmeasured confounding.

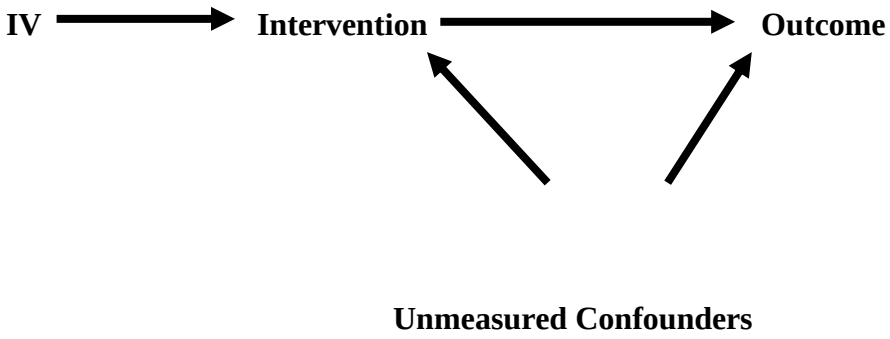

Figure 4: Causal diagram illustrating assumptions for instrumental variable analysis

A commonly used IV in pharmacoepidemiology is physician prescribing preference (13). In a database study assessing the short-term effects of COX-2 inhibitors vs other non-steroidal anti-inflammatory drugs (NSAIDs) on gastrointestinal toxicity, Brookhart et al. (2006) identified unmeasured confounding as a major threat to the validity of their findings (14). To address this concern, they analyzed data using IV analysis in addition to conventional regression and compared their results to published results from a previous RCT. Because prescribing different types of NSAIDs is thought to significantly vary between physicians, and the preference for NSAIDs is assumed not to be associated with any confounders, physician prescribing preference was selected as the IV. The IV analysis found a protective effect attributed to COX-2 inhibitors when compared to NSAIDS which was in agreement with the RCT. The conventional regression approach, on the other hand, found no statistically significant difference.

Good IVs can be hard to find and come with some additional assumptions but, when applicable, IV analysis provides an ingenious solution to dealing with unmeasured confounders, an all-too-common scenario in non-randomized studies.

3. Mediation Analysis: Decomposing Effects

Identifying mediators of a causal pathway between an intervention and health outcome is important for population health, as it allows for health policy experts to develop targeted solutions that can intervene at the level of the mediator. However, mediation analyses are susceptible to the same type of bias as time-dependent confounding and conventional adjustment methods fail to produce effects that satisfy the exchangeability condition for causal inference.

With the advent of causal inference methods such as MSMs and IV analyses, we now have the tools to decompose the total causal effect of an intervention into direct (blue arrow) and indirect or 'mediated' (red arrows) causal effects (Figure 5) (15). For example, in a recent French study investigating the mediators of lung cancer risk in men with varying degrees of education, the total causal effect of education level on lung cancer incidence was decomposed into direct and indirect effects mediated by smoking and other occupational exposures using MSMs (16). Menvielle et al. found that 31% of the total effect was mediated by cumulative lifelong smoking among men with a high-school degree (16). Based on their results, the authors recommended health policies targeting tobacco control to reduce socioeconomic disparities in lung cancer.

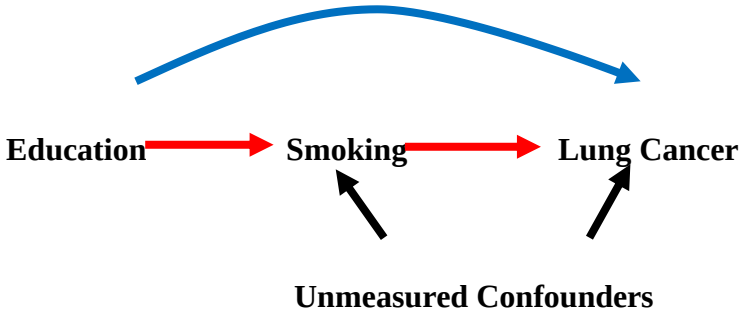

Figure 5: Direct and indirect causal effect of education level on lung cancer

Summary

In conclusion, the causal inference framework states that, when causal conditions hold (consistency, positivity, exchangeability), causal effects can still be estimated for non-randomized primary care interventions (Table 1). If one or more conditions are violated, the impact of these violations must be further investigated (for instance, through applying sensitivity analyses for unmeasured confounders).

Table 1: Assessing the conditions necessary to estimate the causal effect of interventions or policies

Causal Condition	Interpretation
Consistency	Is the intervention well-defined and implemented in the same way for all subjects?
Positivity	Does every subject in the target population have a chance to receive or not the intervention?
Exchangeability	Are all confounders known and measured?

Causal inference methods provide analytical tools to deal many sources of bias that cannot be dealt with using conventional regression methods: MSMs may be applied to overcome adjustment problems arising from time-dependent confounding, IV analyses can be used to address unmeasured confounding, and mediation analyses can elucidate causal pathways of an intervention effect (Table 2).

Table 2: Summary of advantages and application of causal inference methods

Research Aim	Conventional Regression Methods	Causal Inference Methods
Estimate a causal effect in the case where prior exposure to the intervention influences the confounder(s)	Induce rather than eliminate bias	MSMs overcome bias with a weighting approach
Estimate a causal effect in the case where some confounders are not measured	Cannot adjust for unmeasured confounding	IV analyses work around unmeasured confounding
Estimate direct and mediated causal effects of an intervention	Susceptible to bias due to adjustment of a mediator	MSM- and IV-based mediation analyses can decompose total causal effects into direct and mediated effects

New advances in causal inference offer promising ways to conduct our primary care studies, improve the quality of evidence that we produce and ensure that changes to our practices and health systems are based on sound, robust evidence of the causal effects of the interventions studied. Causal methods are the future and should be at the forefront of the quantitative armamentarium for primary care researchers.

Acknowledgements

We would like to thank Dr. Geneviève Arsenault-Lapierre for her input on the manuscript and Ms. Marine Hardouin for assistance with the figures. This methods brief was funded by the Canadian Institutes of Health Research - Vanier Canada Graduate Scholarship. The authors have no conflicts of interest to declare.

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12 APPENDIX C: SURVEY QUESTIONNAIRE

English Version

Choosing key indicators of care management in primary care for persons newly diagnosed with dementia

The goal of this survey is to get your input on which measures of care management for patients newly diagnosed with dementia in primary care are most important from your point of view.

As key stakeholders, your participation will be essential in deciding which measures should be prioritized and evaluated on a regular basis.

This survey should take around **15 minutes** to complete.

Your participation in this survey is completely voluntary. By completing this questionnaire, you consent to participate in this study. There are no known risks, side effects or disadvantages associated with this research study. We will ensure the confidentiality of the information collected. The information will, in no case, be transmitted to other persons not involved in this study. This survey has been approved by the research ethics committee of the Canadian Consortium of Neurodegeneration in Aging.

If you have questions about this survey or study, please contact Dr. Isabelle Vedel (isabelle.vedel@mcgill.ca) or Ms. Nadia Sourial (nadia.sourial@mail.mcgill.ca).

Thank you for your participation!

Demographic information for statistics purposes:

From what perspective are you answering this survey (check only 1)?

- ☐ As a patient / caregiver representative (e.g. Alzheimer Societies)
- ☐ As a clinician
- ☐ As a government representative
- ☐ As a manager
- ☐ Other

Please select your age group:

- ☐ Less than 35
- ☐ 35-44
- ☐ 45-54
- ☐ 55-64
- ☐ 65 and over

ROSA
Research on Organization of Healthcare Services for Alzheimers
Recherche en organisation des services sur l'Alzheimer

CIHR IRSC
Canadian Institutes of Health Research
Instituts de recherche en santé du Canada

Indicator of care management in primary care for persons newly diagnosed with dementia	**Check (✔) a maximum of 10** indicators that you feel are most important
1. Access to a regular family doctor (or nurse practitioner)	
2. Avoidable hospitalizations	
3. Non-urgent visits to the Emergency Department of the hospital	
4. Diagnosis at an early stage of the disease	
5. After-hours access to the regular family doctor	
6. Coordination between different health care providers	
7. Readmission to the hospital within 30 days following a hospitalization	
8. Visit to the regular family doctor within 7 days following a hospitalization	
9. Referrals to specialists in dementia (geriatrician, neurologist, psychiatrist) originating from the regular family doctor	
10. Referrals to other specialists (e.g. cardiologist, oncologist) originating from the regular family doctor	
11. Requests for blood tests originating from the regular family doctor	
12. Phone calls between the regular family doctor and specialists	
13. Dementia diagnosed by the regular family doctor	
14. Medications prescribed for dementia (e.g. Exelon®, Reminyl®, Aricept®, Ebixa®)	
15. First medication for dementia prescribed by the regular family doctor	

16. Annual visits to the regular family doctor	
17. Annual cost of health services	
18. Duplication of medical tests (e.g. blood tests, CT / MRI scans)	
19. Length of time spent in hospital in the year following diagnosis of dementia	
20. Equitable care across all patients (e.g. age, sex, income, region, immigrant status)	
21. Yearly flu shot (immunization for influenza)	
22. Potentially inappropriate prescriptions for medications (e.g. benzodiazepines, antipsychotics, anticholinergics, tricyclic antidepressants, trazodone) associated with serious side effects	
23. Having a high number of medications	
24. Access to counselling for patients	
25. Access to counselling for caregivers	
26. Access to home care	
27. Access to long-term care	
28. Access to palliative end-of-life care	
29. Number of days spent in hospital in last 3 months of life	
30. Dying at home	

COMMENTS:

French Version

Choisir les principaux indicateurs de gestion de soins en première ligne pour les personnes nouvellement diagnostiquées avec la démence

L'objectif de ce sondage est d'obtenir votre opinion sur l'identification des mesures de gestion de soins les plus importantes pour les patients atteints de démence en première ligne.

En tant que partie prenante, votre participation sera essentielle afin de déterminer les mesures qui devrait être priorisées et régulièrement évaluées.

Ce sondage devrait durer environ **15 minutes**.

Votre participation à ce sondage est entièrement sur une base volontaire. En complétant ce sondage, vous consentez à participer à cette étude. Il n'y a aucun risque, effet secondaire ou désavantage connu associé à cette recherche. Nous assurerons la confidentialité des informations collectées. Cette information ne sera en aucun cas transmise aux autres personnes impliquées dans cette étude. Ce sondage a été approuvé par le comité d'éthique de recherche du Consortium canadien en neurodégénérescence associée au vieillissement.

Si vous avez des questions sur ce sondage ou sur cette étude, veuillez contacter le Dre Isabelle Vedel (isabelle.vedel@mcgill.ca) ou Mme Nadia Sourial (nadia.sourial@mail.mcgill.ca).

Informations démographiques pour analyses statistiques

À quel titre remplissez-vous ce sondage (ne choisir qu'une réponse)?

- ☐ En tant que représentant patient/ proche aidant (ex. Sociétés Alzheimer)
- ☐ En tant que clinicien
- ☐ En tant que représentant du gouvernement
- ☐ En tant que gestionnaire
- ☐ Autre

Veuillez sélectionner votre groupe d'âge :

- ☐ Moins de 35 ans
- ☐ 35-44
- ☐ 45-54
- ☐ 55-64
- ☐ 65 ans et plus

ROSA
Research on Organization of Healthcare Services for Alzheimers
Recherche en organisation des services sur l'Alzheimer

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Indicateurs de gestion de soins en première ligne pour les personnes nouvellement diagnostiquées avec la démence	**Cochez (✔) un maximum de 10** indicateurs qui vous semblent les plus importants
1. Accès à un médecin de famille régulier (ou infirmière praticienne)	
2. Hospitalisations évitables	
3. Visites non-urgentes au département de l'urgence de l'hôpital	
4. Diagnostic à un stade précoce de la maladie	
5. Accès au médecin de famille régulier en dehors des heures ouvrables	
6. Coordination entre les différents professionnels de la santé	
7. Réadmission à l'hôpital dans les 30 jours suivant une hospitalisation	
8. Visite au médecin de famille régulier dans les 7 jours suivant une hospitalisation	
9. Visite à des spécialistes en démence (ex. : gériatre, neurologue, psychiatre) référés par le médecin de famille régulier	
10. Visites à d'autres types de spécialistes (ex. : cardiologue, oncologue) référés par le médecin de famille régulier	
11. Tests sanguins prescrits par le médecin de famille régulier	
12. Appels entre le médecin de famille régulier et les spécialistes	
13. Démence diagnostiquée par le médecin de famille régulier	
14. Prescriptions de médicaments pour la démence (ex. Exelon®, Reminyl®, Aricept®, Ebixa®)	
15. Première prescription de médicaments pour la démence prescrits par le médecin de famille régulier	

16. Visite annuelle au médecin de famille régulier	
17. Coût annuel des services de santé utilisés	
18. Duplication de tests médicaux (ex. tests de laboratoire, CT, IRM)	
19. Durée de temps passé à l'hôpital dans l'année suivant le diagnostic de démence	
20. Équité dans l'accès aux soins (ex. âge, sexe, revenu, région, statut immigrant)	
21. Vaccin annuel contre la grippe (immunisation contre l'influenza)	
22. Prescriptions potentiellement inappropriées de médicaments (ex. benzodiazépines, antipsychotiques, anticholinergiques, antidépresseurs tricycliques, trazodone) associés à des effets secondaires importants	
23. Avoir un nombre élevé de médicaments	
24. Accès au médecin de famille régulier pour du soutien psychologique pour les patients	
25. Accès au médecin de famille régulier pour du soutien psychologique pour les proches-aidants	
26. Accès aux soins à domicile	
27. Accès aux soins de longue durée	
28. Accès aux soins palliatifs en fin de vie	
29. Nombre de jours passés à l'hôpital durant les 3 derniers mois de vie	
30. Mourir chez soi	

COMMENTAIRES

13 APPENDIX D: LIST OF FRAMEWORK INDICATORS DEVELOPED IN MANUSCRIPT 1 AND JUSTIFICATION FOR SUBSET OPERATIONALIZED AND INCLUDED IN MANUSCRIPT 2

The table below presents the 37 indicators of primary care performance and health service use included in the framework developed in Manuscript 1 (Chapter 4). Of these indicators, 24 indicators were operationalized based on the available data within the linked health administrative data at ICES. Thirteen indicators were not operationalized within this doctoral work because they required a longer period than was feasible to accurately and fully consider how best to define them. For example, several indicators (e.g. after-hours access) were sensitive to billing practices and would necessitate a deeper exploration to capture all possible associated billing codes. Others required additional expertise outside the research team (e.g. annual cost of services) or a more thorough assessment of the appropriate medication codes for complex medication categories such as antidepressants. The finalization of the operationalization for these indicators has been cited in the overall discussion as future directions and is planned within the next phase of the larger research program within which this doctoral work fits.

Of the 23 operationalized indicators, 19 were included in Manuscript 2 (Chapter 5) on the examination of sex differences in trends of primary care performance and health service use. Given the very large number of indicators to present in a single manuscript, physician specialty associated with the largest proportion of visits, non-urgent ED visits, length of time spent in hospital in the year following diagnosis of dementia, diagnosis at an early stage of the disease were not included as per editors' request to minimize the number of figures and condense the manuscript.

Framework domain of performance	Framework indicator	Operationalized (if not, reason)
ACCESS		
	Access to a regular family doctor (or nurse practitioner)	Yes
	After-hours access to the regular family doctor	No (longer-term operationalization required, indicator sensitive to billing practices)
	Access to an interprofessional primary care team	Yes
	Visits to the regular family physician	Yes
	Physician specialty associated with the largest proportion of visits	Yes
	Visits to the ED	Yes
	Non-urgent ED visits	Yes
INTEGRATION		
	Continuity of care	Yes
	Phone calls between the regular family doctor and specialists	No (longer-term operationalization required, indicator sensitive to billing practices)
	Hospitalizations	Yes
	Length of time spent in hospital in the year following diagnosis of dementia	Yes
	Discharge delay	Yes
	Avoidable hospitalizations	Yes
	Visits to the regular family doctor within 7 days following a hospitalization	Yes
	Readmissions to the hospital within 30 days following a hospitalization	Yes
EFFECTIVE CARE		
	Diagnosis at an early stage of the disease	Yes

	Dementia diagnosis first recorded by the regular family doctor	Yes
	Requests for blood tests originating from the regular family doctor	No (longer-term operationalization required, indicator sensitive to billing practices)
	Medication prescribed for dementia (e.g. Exelon®, Reminyl®, Aricept®, Ebixa®)	Yes
	Medication for dementia prescribed by the regular family doctor	Yes
	Annual visit to the regular primary care physician	No (longer-term operationalization required, indicator sensitive to billing practices)
	Referrals to specialists in dementia (geriatrician, neurologist, psychiatrist) originating from the regular family doctor	Yes
	Referrals to other specialists (e.g. cardiologist, oncologist) originating from the regular family doctor	Yes
EFFICIENT CARE		
	Annual cost of health services	No (longer-term operationalization required, required expertise outside research team)
	Duplicate medical tests (e.g. blood tests, brain CT / MRI scans)	No (longer-term operationalization required; complexity of billing codes and could be performed in private)
POPULATION HEALTH		
	Yearly flu shot (immunization for influenza)	No (longer-term operationalization required, indicator sensitive to billing practices)
	Pneumoccocal immunization	No (longer-term operationalization required, indicator sensitive to billing practices)

SAFETY		
	Potentially inappropriate prescriptions for medications (e.g. benzodiazepines, antipsychotics, anticholinergics, tricyclic antidepressants, trazodone) associated with serious side effects	No (longer-term operationalization required, analysis of medication lists and codes)
	Polypharmacy	No (longer-term operationalization required, analysis of medication lists and codes)
PATIENT-CENTERED CARE		
	Access to counselling for patients	No (longer-term operationalization required, indicator sensitive to billing practices)
	Access to counselling for caregivers	No (longer-term operationalization required indicator sensitive to billing practices)
	Access to home care	Yes
	Access to long-term care	Yes
	Access to palliative end-of-life care	No (indicator sensitive to billing practices)
	Number of days spent in hospital in last 3 months of life	No (indicator sensitive to billing practices)
	Dying at home	Yes
EQUITY		
	Equitable care across all patients	Yes