HEALTHCARE

POLICY

Politiques de Santé

Health Services, Management and Policy Research
Services de santé, gestion et recherche de politique

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Data Matters • *Discussion and Debate* • *Research Papers*

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HEALTHCARE
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Politiques de Santé

Health Services, Management and Policy Research
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Healthcare Policy/Politiques de Santé seeks to bridge the worlds of research and decision-making by presenting research, analysis and information that speak to both audiences. Accordingly, our manuscript review and editorial processes include researchers and decision-makers.

We publish original scholarly and research papers that support health policy development and decision-making in spheres ranging from governance, organization and service delivery to financing, funding and resource allocation. The journal welcomes submissions from researchers across a broad spectrum of disciplines in health sciences, social sciences, management and the humanities and from interdisciplinary research teams. We encourage submissions from decision-makers or researcher–decision-maker collaborations that address knowledge application and exchange.

While *Healthcare Policy/Politiques de Santé* encourages submissions that are theoretically grounded and methodologically innovative, we emphasize applied research rather than theoretical work and methods development. The journal maintains a distinctly Canadian flavour by focusing on Canadian health services and policy issues. We also publish research and analysis involving international comparisons or set in other jurisdictions that are relevant to the Canadian context.

Politiques de Santé/Healthcare Policy cherche à rapprocher le monde de la recherche et celui des décideurs en présentant des travaux de recherche, des analyses et des renseignements qui s'adressent aux deux auditoires. Ainsi donc, nos processus rédactionnel et d'examen des manuscrits font intervenir à la fois des chercheurs et des décideurs.

Nous publions des articles savants et des rapports de recherche qui appuient l'élaboration de politiques et le processus décisionnel dans le domaine de la santé et qui abordent des aspects aussi variés que la gouvernance, l'organisation et la prestation des services, le financement et la répartition des ressources. La revue accueille favorablement les articles rédigés par des chercheurs provenant d'un large éventail de disciplines dans les sciences de la santé, les sciences sociales et la gestion, et par des équipes de recherche interdisciplinaires. Nous invitons également les décideurs ou les membres d'équipes formées de chercheurs et de décideurs à nous envoyer des articles qui traitent de l'échange et de l'application des connaissances.

Bien que *Politiques de Santé/Healthcare Policy* encourage l'envoi d'articles ayant un solide fondement théorique et innovateurs sur le plan méthodologique, nous privilégions la recherche appliquée plutôt que les travaux théoriques et l'élaboration de méthodes. La revue veut maintenir une saveur distinctement canadienne en mettant l'accent sur les questions liées aux services et aux politiques de santé au Canada. Nous publions aussi des travaux de recherche et des analyses présentant des comparaisons internationales qui sont pertinentes pour le contexte canadien.

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EDITORIAL

Kicking Off the 2020s with *Healthcare Policy*

Disparities in health and healthcare plague provinces' residents. This means that different subgroups of the population have differences in health and healthcare. Examples include socio-economic status, location of residence and disability status. Disparities generate searching questions: does where you live, or who you are, affect your health or the quality or accessibility of healthcare you receive?

It is important for provincial ministries of health and social care to address the issue of disparities. Disparities limit gains in health among the population, induce additional healthcare utilization or spending and lay bare inequitable distribution of public resources. Resolving disparities is often challenging as causal pathways may be complex, ingrained in communities or very expensive to address.

Research on disparities is an important element of improving provinces' population health. Quantitative and qualitative researchers play complementary roles of observers and reporters of often-in-plain-sight disparities. Policy analysis plays the adjunct role of exploring intersections of legislation, strategy and program objectives and identifying short- and long-term options for decision-makers.

In this issue of *Healthcare Policy*, several papers identify and examine disparities in access to healthcare services. Tobias and colleagues (2020) examine the effectiveness of cancer screening programs among First Nations peoples, Jones and colleagues (2020) report on gender differences in access to surgery, whereas Martin-Misener and colleagues (2020) explore facets of rural and remote healthcare delivery through the role of nursing practices.

This issue also features an interprovincial comparison of childhood cancer costs by McBride and colleagues (2020), which provides valuable insights into variations in care delivery. Zoratti and colleagues (2020) report on a three-province comparison of drug reimbursement recommendations, whereas Bell and colleagues (2020) explore equity in

accessing allogeneic stem cell transplant and Antonipillai and colleagues (2020) explore Canadian refugee health policy. This issue also presents a Discussion and Debate article by Khan and colleagues (2020) that draws on international experience to inform biosimilar implementation.

Just as opioids and patient-oriented research gained policy makers', researchers' and the public's eye in the last decade, it is yet unclear which topics in health services and policy research will gain prominence over the next decade. Although I expect that disparity-focused research will feature prominently in future issues, the journal will aim to continue to publish equally valuable health services and policy research whose aim is to improve the health and quality of life of Canadians.

This is my inaugural issue as editor-in-chief. I'm indebted to the editorial team for their support, noting the valuable addition of Dianne Foster-Kent as editorial director, whose efforts will support authors, the editors and publication. I also acknowledge the contributions of the dedicated editors, reviewers and past editor-in-chief, Dr. Jennifer Zelmer. Through her leadership over a decade, *Healthcare Policy* publications are renowned for their high quality and deep reach into all levels of policy making. We also thank Dr. Eric Latimer for his hard work and years of service supporting the scholarship appearing in the journal.

We are also welcoming two successful scholars as incoming editors. Dr. Fiona Clement joins us from the Department of Community Health Sciences and the O'Brien Institute for Public Health at the University of Calgary, and Dr. Sabrina Wong joins us from the Faculty of Nursing and the Centre for Health Services and Policy Research at the University of British Columbia. We look forward to their engagement with authors and reviewers.

Despite some new faces, there is reason to expect continuity in content and editorial style. As the focus of this journal is healthcare policy, I encourage quantitative and qualitative submissions that will carefully develop policy perspectives and the implications of the research. I will also aim to support an increased number of Discussion and Debate submissions as readers will appreciate spirited examination of topical health and healthcare policy issues. Finally, I invite all authors to continue to support *Healthcare Policy* through the submission of Canadian-focused health services and policy research.

JASON M. SUTHERLAND, PhD
Editor-in-Chief

References

Antonipillai, V., J. Abelson, O. Wahoush, A. Baumann and L. Schwartz. 2020. Policy Agenda-Setting and Causal Stories: Examining How Organized Interests Redefined the Problem of Refugee Health Policy in Canada. *Healthcare Policy* 15(3): 116–31. doi:10.12927/hcpol.2020.26126.

Bell, J.A.H., Z. Schmilovich, D.Z. Buchman, M. Escaf, J. Costello and H.A. Messner. 2020. First Ready, First to Go: Ethical Priority-Setting of Allogeneic Stem Cell Transplant at a Major Cancer Centre. *Healthcare Policy* 15(3): 102–15. doi:10.12927/hcpol.2020.26127.

Jones, A.M., M. Koehoorn and C.B. McLeod. 2020. Gender Differences in Surgery for Work-Related Musculoskeletal Injury: A Population-Based Cohort Study. *Healthcare Policy* 15(3): 47–62. doi:10.12927/hcpol.2020.26131.

Khan, D., T. Luig, D. Mosher and D. Campbell-Scherer. 2020. Lessons from International Experience with Biosimilar Implementation: An Application of the Diffusion of Innovations Model. *Healthcare Policy* 15(3): 16–27. doi:10.12927/hcpol.2020.26133.

Martin-Misener, R., M.L.P. Macleod, E.C. Wilson, J.G. Kosteniuk, K.L. Penz, N.J. Stewart et al. 2020. The Mosaic of Primary Care Nurses in Rural and Remote Canada: Results from a National Survey. *Healthcare Policy* 15(3): 63–75. doi:10.12927/hcpol.2020.26130.

McBride, M.L., C. De Oliveira, R. Duncan, K.E. Bremner, N. Liu, M.L. Greenberg et al. 2020. Comparing Childhood Cancer Care Costs in Two Canadian Provinces. *Healthcare Policy* 15(3): 76–89. doi:10.12927/hcpol.2020.26129.

Tobias, J.K., J. Tinmouth, L.C. Senese, N. Jumah, D. Llovet, A. Kewayosh et al. 2020. Health Policy as a Barrier to First Nations Peoples' Access to Cancer Screening. *Healthcare Policy* 15(3): 28–46. doi:10.12927/hcpol.2020.26132.

Zoratti M. J., F. Xie, K. Thorlund, N. Allen and M. Levine. 2020. An Exploratory Analysis of Predictors of Concordance between Canadian Common Drug Review Reimbursement Recommendations and the Subsequent Decisions by Ontario, British Columbia and Alberta. *Healthcare Policy* 15(3): 90–101. doi:10.12927/hcpol.2020.26128.

ÉDITORIAL

Coup d'envoi de *Politiques de Santé* pour les années 2020

Les inégalités en santé et dans les services de santé minent la vie des résidents des provinces. En fait, ces différences sont une réalité pour plusieurs sous-groupes de la population. Ces groupes se définissent, par exemple, par le statut socioéconomique, le lieu de résidence ou un handicap. Les inégalités sont à l'origine de questions de recherche : est-ce que le lieu de résidence, ou l'identité d'une personne, affecte sa santé ou l'accessibilité et la qualité des services reçus?

Il est important que les ministres provinciaux de la santé et des services sociaux se penchent sur le problème des inégalités. Ces dernières limitent les gains en matière de santé dans la population, accroissent l'utilisation des services, exacerbent les dépenses en santé et font voir la répartition inéquitable des ressources publiques. Trouver une solution aux inégalités pose problème, car les liens de causalité sont souvent complexes, fermement ancrés dans les communautés ou très onéreux à rectifier.

La recherche sur les inégalités est un élément important pour l'amélioration de la santé des populations dans les provinces. Ceux qui font des recherches quantitatives et qualitatives jouent un rôle complémentaire d'observateurs et de divulgateurs d'inégalités souvent manifestes. L'analyse des politiques joue un rôle adjoint, en explorant l'intersection entre la législation, les stratégies et les programmes, tout en identifiant les possibilités à court ou long terme pour les décideurs.

Dans ce numéro de *Politiques de Santé*, plusieurs articles portent sur les inégalités d'accès aux services de santé. Tobias et collègues (2020) examinent l'efficacité des programmes de dépistage du cancer chez les Premières Nations. Jones et collègues (2020) font état de différences entre les sexes pour l'accès aux chirurgies. Martin-Misener et collègues (2020) s'intéressent aux facettes de la prestation des services de santé par la main-d'œuvre infirmière dans les régions rurales et éloignées.

Le présent numéro présente également le travail de McBride et collègues (2020) dans une comparaison interprovinciale des coûts associés au cancer chez l'enfant. Cette étude offre d'intéressantes pistes pour comprendre les variations dans la prestation des soins. De leur côté, Zoratti et collègues (2020) proposent une comparaison entre trois provinces au sujet des recommandations en matière de remboursement des médicaments. Bell et collègues (2020) se penchent sur l'équité d'accès aux greffes de cellules souches allogéniques. Finalement, Antonipillai et collègues (2020) explorent les politiques de santé canadiennes à l'intention de réfugiés. Par ailleurs, la section Discussion et débat propose un article de Khan et collègues (2020) qui met à profit l'expérience internationale pour éclairer la mise en œuvre des biosimilaires.

Alors que la question des opioïdes ou la recherche axée sur le patient ont gagné l'attention des décideurs, des chercheurs et du public au cours des dix dernières années, on ne sait pas encore quels thèmes de recherche gagneront en importance au cours de la prochaine décennie. Même si je m'attends à ce que la recherche sur les inégalités occupe une place de choix dans les prochains numéros de la revue, nous continuerons certainement de publier des recherches qui visent l'amélioration de la santé et de la qualité de vie des Canadiens.

Ceci est mon premier numéro en tant que rédacteur en chef. Je suis reconnaissant à l'équipe éditoriale pour son soutien et je salue l'apport précieux de l'éditrice en chef Dianne Foster Kent, dont les efforts soutiendront les auteurs, les éditeurs et la publication. Je remercie également les éditeurs, réviseurs et l'ancienne rédactrice en chef, Jennifer Zelmer. Grâce à son leadership pendant dix ans, *Politiques de Santé* a acquis une excellente réputation pour la qualité et la portée de ses articles à tous les niveaux de l'élaboration des politiques. Nous remercions également Eric Latimer pour son travail acharné et ses années de service à l'appui de l'excellence de la qualité des recherches publiée dans la revue.

Nous accueillons également deux chercheuses de talent comme nouvelles éditrices : Fiona Clement du Département des sciences de la santé communautaire et de l'Institut de santé Publique O'Brien (Université de Calgary) et Sabrina Wong de la Faculté des sciences infirmières et du Centre de recherche sur les politiques et les services de santé (Université de la Colombie-Britannique). Nous sommes heureux de pouvoir compter sur leurs conseils pour les auteurs et les examinateurs.

Malgré quelques nouveaux venus, il y a lieu de s'attendre à une continuité dans le contenu et le style éditorial. Étant donné que cette revue se concentre sur les politiques de la santé, j'encourage la soumission d'articles au contenu quantitatif ou qualitatif qui exposeront soigneusement les perspectives politiques et les implications de la recherche. Je viserai également à soutenir un nombre accru de soumissions pour la section Discussion et débat; les lecteurs apprécieront certainement un examen attentif des questions d'actualité en matière de santé et de politiques de santé. Enfin, j'invite tous les auteurs à continuer d'appuyer *Politiques de Santé* en soumettant des recherches sur les politiques et services de santé au Canada.

JASON M. SUTHERLAND, PhD
Rédacteur en chef

Références

Antonipillai, V., J. Abelson, O. Wahoush, A. Baumann et L. Schwartz. 2020. Programme d'élaboration des politiques et anecdotes : comment des intérêts organisés redéfinissent le problème des politiques de santé canadiennes à l'intention des personnes réfugiées. *Politiques de Santé* 15(3): 116–31. doi:10.12927/hcpol.2020.26126.

Bell, J.A.H., Z. Schmilovich, D.Z. Buchman, M. Escaf, J. Costello et H.A. Messner. 2020. Premier prêt, premier parti : établissement des priorités en matière d'éthique dans le cas de greffe de cellules souches allogéniques dans un grand centre d'oncologie. *Politiques de Santé* 15(3): 102–15. doi:10.12927/hcpol.2020.26127.

Jones, A.M., M. Koehoorn et C.B. McLeod. 2020. Différences entre les sexes dans la chirurgie pour les blessures musculosquelettiques reliées au travail : une étude de cohortes axée sur la population. *Politiques de Santé* 15(3): 47–62. doi:10.12927/hcpol.2020.26131.

Khan, D., T. Luig, D. Mosher et D. Campbell-Scherer. 2020. Leçons à retenir de l'expérience internationale quant à la mise en œuvre des biosimilaires : application du modèle de diffusion des innovations. *Politiques de Santé* 15(3): 16–27. doi:10.12927/hcpol.2020.26133.

Martin-Misener, R., M.L.P. Macleod, E.C. Wilson, J.G. Kosteniuk, K.L. Penz, N.J. Stewart et coll. 2020. Mosaïque de la main-d'œuvre infirmière en soins primaires dans les régions canadiennes rurales et éloignées : résultats d'une enquête nationale. *Politiques de Santé* 15(3): 63–75. doi:10.12927/hcpol.2020.26130.

McBride, M.L., C. De Oliveira, R. Duncan, K.E. Bremner, N. Liu, M.L. Greenberg et coll. 2020. Comparaison des coûts associés aux soins oncologiques chez les enfants dans deux provinces canadiennes. *Politiques de Santé* 15(3): 76–89. doi:10.12927/hcpol.2020.26129.

Tobias, J.K., J. Tinmouth, L.C. Senese, N. Jumah, D. Llovet, A. Kewayosh et coll. 2020. Les politiques de santé comme obstacle à l'accès des Premières Nations au dépistage du cancer. *Politiques de Santé* 15(3): 28–46. doi:10.12927/hcpol.2020.26132.

Zoratti M.J., F. Xie, K. Thorlund, N. Allen et M. Levine. 2020. Analyse exploratoire des prédicteurs de la concordance entre les recommandations de remboursement du Programme commun d'évaluation des médicaments et les décisions subséquentes de l'Ontario, de la Colombie-Britannique et de l'Alberta. *Politiques de Santé* 15(3): 90–101. doi:10.12927/hcpol.2020.26128.

PUBLISHER'S NOTE

Thank you, Dr. Jennifer Zelmer

Change is inevitable as we turn an important page in *Healthcare Policy*. Dr. Zelmer was welcomed as the second editor-in-chief of *Healthcare Policy* more than a decade ago succeeding Dr. Brian Hutchison who was the journal's inaugural editor-in-chief. At that time, we invited her to continue building on the foundation laid by her predecessor – establishing *Healthcare Policy* as Canada's preeminent scholarly journal of health services and policy research. No doubt readers will agree that Dr. Zelmer and her team of editors have accomplished that and much more during her tenure as the editor-in-chief, and for that, Longwoods Publishing is grateful.

As editor-in-chief, Dr. Zelmer worked with a Canadian and international network of scholars, and built relationships within the healthcare and health research community. This support was invaluable in ensuring the journal's ability to respond to existing and emerging issues such as quality improvement and patient safety, primary care reform, pharmacare and new services delivery models, among others. With her team, Dr. Zelmer has addressed these issues and offered a venue for opinions, facts, debates, research and insight to be expressed through the journal.

"Jennifer did a wonderful job [as editor-in-chief of *Healthcare Policy*]. It was a pleasure to work with her," says Raisa Deber, Editor, *Healthcare Policy*, and Professor, Institute of Health Policy, Management and Evaluation, University of Toronto.

Thanks to Dr. Zelmer, the discussion within the pages of *Healthcare Policy* continues to generate innovative ideas and debates with the goal of influencing health policy and improving health services in Canada. Please see her final editorial "Reflections on a Decade of *Healthcare Policy/Politiques de Santé*" (Zelmer 2019).

Moving forward, Dr. Zelmer will focus on her role as president and chief executive officer for the Canadian Foundation for Healthcare Improvement, where her passion for healthcare policy, research and governance will ensure the improvement of health and healthcare in Canada.

With change comes new opportunities and Longwoods is pleased to welcome Dr. Jason Sutherland, Professor, Centre for Health Services and Policy Research, University of British Columbia as the new editor-in-chief for *Healthcare Policy*. Dr. Sutherland joins Longwoods with significant expertise in policy, analytics, statistics, system performance, surgical outcomes and funding.

"Jason is remarkably strong, but a practically minded scholar who always makes sure that rigorous work finds a home with policy makers," says Adalsteinn Brown, editor-in-chief of *Healthcare Papers*, and Dean, Dalla Lana School of Public Health, University of Toronto.

While building a renewed vision for the journal, we at Longwoods are excited to watch and learn with you – the reader – as Dr. Sutherland challenges and motivates us. Please see his first editorial "Kicking Off the 2020s with *Healthcare Policy*" (Sutherland 2020).

MATTHEW HART
Chief Executive Officer
Longwoods Publishing

References

Sutherland, J.M. 2020. Kicking Off the 2020s with *Healthcare Policy*. *Healthcare Policy* 15(3): 6–9. doi:10.12927/hcpol.2020.26135.

Zelmer, J. 2019. Reflections on a Decade of *Healthcare Policy/Politiques de Santé*. *Healthcare Policy* 15(2): 6–7. doi:10.12927/hcpol.2019.26076.

NOTE DE L'ÉDITEUR

Merci à Dr Jennifer Zelmer

Le changement est inévitable alors que nous tournons une page importante de l'histoire de *Politiques de Santé*. Il y a plus de dix ans, la revue accueillait Jennifer Zelmer comme deuxième rédactrice en chef, succédant à Brian Hutchison, le premier rédacteur en chef de *Politiques de Santé*. Nous l'invitions alors à continuer de bâtir sur la lancée de son prédécesseur : faire de *Politiques de Santé* la revue scientifique par excellence pour la recherche sur les politiques et les services de santé au Canada. Les lecteurs conviendront sans aucun doute que Jennifer Zelmer et son équipe ont accompli cela et bien plus encore pendant son mandat. Pour cela, Longwoods Publishing lui doit reconnaissance.

À titre de rédactrice en chef, Jennifer Zelmer a pu compter sur un réseau canadien et international de chercheurs et elle a créé des relations dans les milieux de la recherche et des services de santé. Ce soutien a été inestimable pour garantir la capacité de la revue à répondre à des problèmes actuels ou émergents tels que l'amélioration de la qualité et de la sécurité des patients, la réforme des soins primaires, l'assurance médicaments et les nouveaux modèles de prestation de services. Avec le soutien de son équipe, Jennifer Zelmer a abordé ces enjeux et fait de la revue un lieu pour présenter des opinions, des faits, des débats, des recherches et des idées.

« Jennifer a accompli un travail formidable en tant que rédactrice en chef de *Politiques de Santé*. Ce fut un plaisir de travailler avec elle » a déclaré Raisa Deber, éditrice de *Politiques de Santé* et professeure à l'Institut de l'évaluation, de la gestion et des politiques en santé, Université de Toronto.

Grâce à Jennifer Zelmer, les débats de *Politiques de Santé* continuent de générer des idées et des questionnements qui influencent les politiques de santé et améliorent les services de santé au Canada. À cet effet, je vous suggère de consulter son dernier éditorial « Réflexions sur une décennie de *Politiques de Santé/Healthcare Policy* » (Zelmer 2019).

À l'avenir, Jennifer Zelmer se concentrera sur son rôle de présidente-directrice générale de la Fondation canadienne pour l'amélioration des services de santé, où son intérêt pour

les politiques, la recherche et la gouvernance en matière de soins de santé contribuera à l'amélioration de la santé et des services de santé au Canada.

Le changement entraîne de nouvelles opportunités et Longwoods est heureux d'accueillir Jason Sutherland, professeur au Centre de recherche sur les politiques et les services de santé de l'Université de la Colombie-Britannique, en tant que nouveau rédacteur en chef de *Politiques de Santé*. Jason Sutherland apporte à Longwoods une expertise considérable en politique, analyse, statistiques, performance du système, résultats chirurgicaux et financement.

« Jason est remarquablement fort. C'est un chercheur à l'esprit pratique qui veille toujours à ce qu'un travail rigoureux trouve sa place auprès des décideurs », a déclaré Adalsteinn Brown, rédacteur en chef de *Healthcare Papers*, et doyen à l'École de santé publique Dalla Lana de l'Université de Toronto.

Tout en renouvelant la vision de la revue, nous sommes ravis, à Longwoods, d'acquérir de nouvelles connaissances avec vous, notre lectorat, tout en étant interpellés et motivés par Jason Sutherland. Je vous conseille de consulter son premier éditorial « Coup d'envoi de *Politiques de Santé* pour les années 2020 » (Sutherland 2020).

MATTHEW HART
Président et chef de la direction
Longwoods Publishing

Références

Sutherland, J.M. 2020. Coup d'envoi de *Politiques de Santé* pour les années 2020. *Politiques de Santé* 15(3): 9–11. doi:10.12927/hcpol.2020.26135.

Zelmer, J. 2019. Réflexions sur une décennie de *Politiques de Santé/Healthcare Policy*. *Politiques de Santé* 15(2): 8–9. doi:10.12927/hcpol.2019.26076.

Lessons from International Experience with Biosimilar Implementation: An Application of the Diffusion of Innovations Model

Leçons à retenir de l'expérience internationale quant à la mise en œuvre des biosimilaires : application du modèle de diffusion des innovations

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Abstract

Although biosimilars offer cost savings in Canadian healthcare, uptake is low. We discuss the literature on international experiences with biosimilar adoption in the context of the Diffusion of Innovations model. We highlight potential challenges with biosimilar implementation and gaps in research needed to inform implementation efforts. We observe a lack of systematic description of implementation design and evaluation and a paucity of in-depth and engaged research to understand stakeholders' pragmatic considerations and the knowledge, messages and meanings that shape clinician and patient decisions to choose biosimilars.

Résumé

Bien que les biosimilaires permettent d'épargner des coûts pour les services de santé au Canada, leur adoption demeure faible. Nous commentons la littérature sur l'expérience internationale en matière d'adoption des biosimilaires au moyen du modèle de diffusion des innovations. Nous dégageons les défis potentiels quant à leur mise en œuvre et nous faisons état des lacunes en matière de recherche nécessaire pour éclairer les efforts de mise en œuvre. Nous observons un manque de description systématique des modèles de mise en œuvre et d'évaluation ainsi qu'une rareté de recherche approfondie pour comprendre les considérations pragmatiques des intervenants ainsi que les connaissances et les messages qui éclairent les décisions prises par les cliniciens et les patients au sujet des biosimilaires.

Background

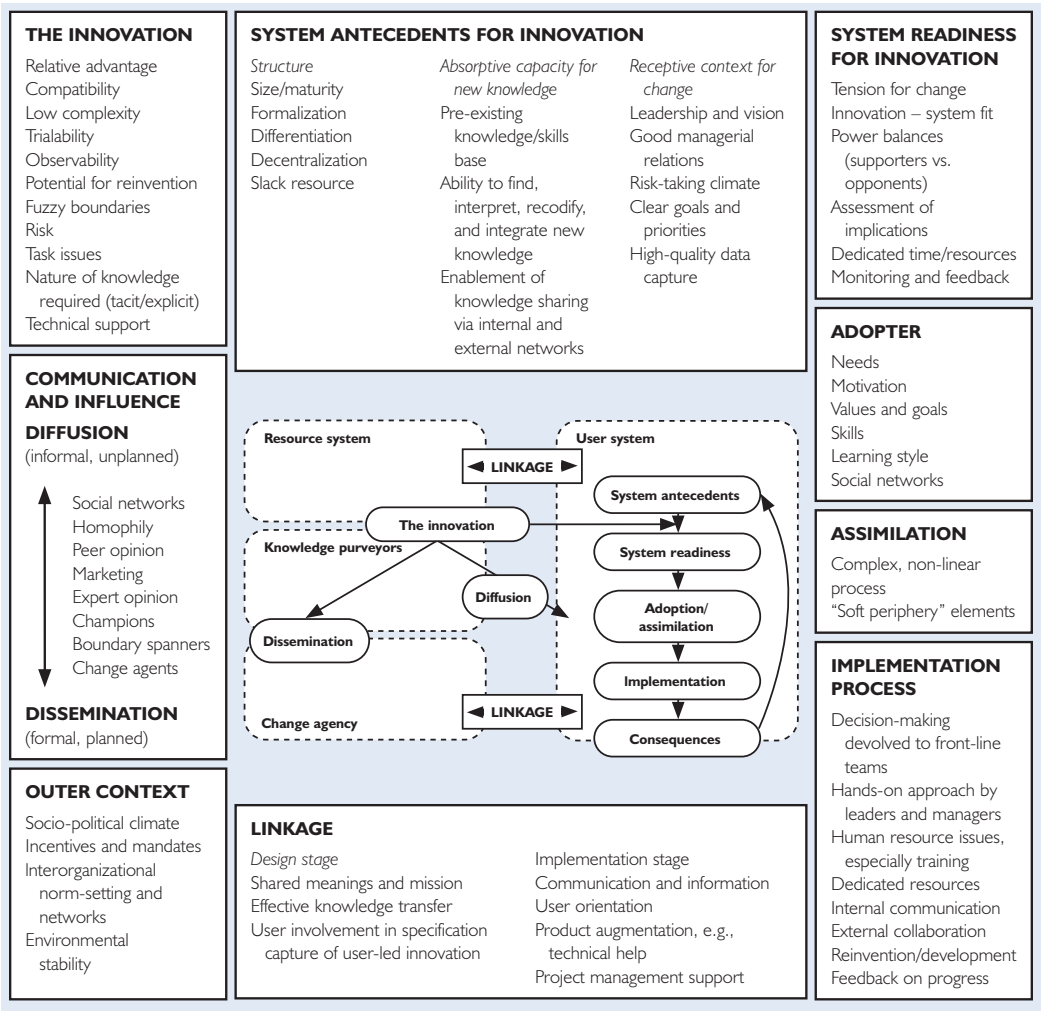
Biosimilars, an economical alternative to traditional biologic treatment, offer exciting opportunities to improve access to treatment (Elek et al. 2017) and cost savings as Canadian healthcare costs rise. However, despite these potential benefits, biosimilar uptake is lower than expected (Health Canada 2017).

To increase biosimilar adoption, several countries instituted biosimilar prescription policies to increase biosimilar uptake by patients new to biologics and biosimilar switching by patients already taking biologics. In Canada, the provincial formularies of Alberta and British Columbia cover the infliximab biosimilar Inflectra for treatment-naïve patients (CADTH 2018), but neither province had a switching policy until 2019–2020. The Patented Medicines Prices Review Board (2017) forecast a potential rapidly growing market for biosimilars, with 13 biologic drugs losing Canadian market exclusivity by 2022. Canadian healthcare needs appropriate steps to reap benefits.

What can Canada learn from international experiences with biosimilar policy and implementation? The literature reflects barriers to biosimilar uptake in translating knowledge, changing prescribing practices, and adjusting drug administration logistics and the benefit/risk perception of patients, providers and payers (Calvo et al. 2018; Cohen et al. 2016; Peyrin-Biroulet et al. 2017). However, the published literature seldom explores in

greater depth the key factors that implementation theory highlights. Implementation comprises active efforts to transition an innovation from adoption to routinization (Greenhalgh et al. 2004). Increasing biosimilar uptake and switching are complex implementation problems that benefit from implementation science. Despite the impact on access to biologics for patients and on cost-effectiveness in healthcare, surprisingly little is published on biosimilar implementation and uptake.

FIGURE 1. A conceptual model for Diffusion of Innovations in service organizations



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Greenhalgh et al. (2004) developed the Diffusion of Innovations model, a unifying conceptual model of factors and processes in spreading and sustaining innovations in health service organizations. The model is based on an extensive review of theoretical and empirical findings in the interdisciplinary literature (Figure 1). Here we discuss the literature on international experiences with biosimilar adoption in the context of Greenhalgh's model to highlight potential challenges with biosimilar implementation and gaps in research needed to inform implementation efforts.

Diffusion of Innovations: An Implementation Model

Implementing innovations in healthcare requires stakeholders to change how they think and practise in specific situations. People are not passive about innovations; they appraise them, challenge them, experiment with them, find purpose or not in them, develop feelings about them, work around them and adapt them to fit situations and goals (Greenhalgh et al. 2004). These processes happen individually, across social and professional networks and within structural constraints.

Greenhalgh et al.'s model details aspects of context, such as the socio-political climate and system antecedents, that shape conditions in which organizations and individuals operate and interact with innovation. The model conceptualizes influences on innovation adoption and implementation of organizational and individual actors, factors and processes and linkages between them (Table 1). For example, innovation qualities such as relative advantage, compatibility, trialability, low risk and soft periphery impact implementation success. Diffusion and dissemination are affected by knowledge transfer through social networks, peer opinions and champions. The model outlines conditions that promote system readiness to take up and implement innovations, such as tension for change, assessment of implications, power, monitoring systems and innovations–system fit.

Innovations require individuals to adopt new behaviour or technologies. Adoption decisions are influenced by the individual needs, motivations, values and meanings they (and others) ascribe to the innovation and decisions for their organization or professional network. Once an adoption decision is made, the model suggests actions that help routinize and integrate the innovation into regular practice: dedicating resources, communicating and collaborating, engaging stakeholders early and widely in decision-making and planning, giving management support, motivating and building practitioner capacity.

We conducted a narrative review on biosimilar implementation literature and policy from the years 2012 to 2018. The review focused on the experiences, attitudes and barriers to introducing biosimilars to physicians, specialists, patients and caregivers and professional and patient organizations, as well as on policy documents and position papers reflecting governmental implementation strategies and experiences. We searched the databases Medline, Scopus, Google Scholar, EMBASE and PubMed using the keywords "biosimilar*," and "subsequent entry biologic*," along with "survey*," "experience" and "perception." Manual searches of various governmental health service providers (such as Health Canada, the U.S. Food

and Drug Administration [FDA] and the National Health Service [NHS]) and professional groups that represent specialties in practice were conducted with an emphasis on biosimilar implementation policy and position papers with regard to this novel class of pharmaceuticals. Here we apply Greenhalgh et al.'s model to discuss components of the model where information is available and point out gaps in the literature that hinder a comprehensive assessment of implementation internationally.

TABLE 1. Diffusion of Innovations model definitions

Select model terminology	Explanation
Innovation	Novel technology, behaviours aimed at improving health
Diffusion	Innovation spread by passive, informal and unplanned means
Readiness	Capacity of an organization or system to implement, including tension for change, clear implications, established systems to monitor impact and availability of resources
Adoption	Individual process from first contact with innovation to decision to adopt it
Implementation	Planned efforts to routinize an innovation within an organization and workflow
Relative advantage	Clear, unambiguous advantage (effectiveness or cost)
Compatibility	Aligned with adopter's values and needs
Trialability	Potential for adopter to experiment with the innovation
Low risk	Innovation has a low level of perceived risk and low uncertainty in outcomes
Adaptiveness of the soft periphery	Organizational structures, systems and tools required for implementation can be adapted according to contexts within different systems/organizations
Tension for change	Adopters perceive current situation as intolerable
Power balance	Balance between supporters and opponents of an innovation
Monitoring systems	Strategies and skills to monitor and evaluate impact of an innovation

Innovation

Increasing biosimilar uptake has a clear, observable advantage: biosimilars are less expensive than biologics, with no significantly different clinical results (Health Canada 2017), although this is contested to varying degrees depending on the available published evidence. This advantage can lead to improved access to biologics treatment and cost savings for health-care systems or patients (Elek et al. 2017). Biosimilar introduction also has the potential for leveraging the adaptiveness of organizational structures, systems and tools required for implementation, which is an important innovation quality that supports implementation. For example, Scotland developed tools for education and adoption to fit the needs of individual adopters and user systems (Health Improvement Scotland 2018). These key features can support biosimilar implementation among adopters.

The advantages of biosimilars versus biologics are less clear when considering adopters' beliefs and values surrounding care. In Germany and Belgium, where biosimilars are accessible, physicians choose a biologic over a biosimilar for first-line therapy and prioritize treatment efficacy over cost-effectiveness (Sullivan et al. 2017; van Overbeeke et al. 2017). Controversies persist in specialist communities over biosimilar safety (Sullivan et al. 2017;

van Overbeeke et al. 2017). The debate is fuelled by efforts of originator manufacturers to retain their market share and to question the efficacy and safety of biosimilars (Cassels 2017; Milne et al. 2017).

The potential for adopters to experiment with biosimilars (trialability of the innovation) without risk is limited and may thus pose a challenge for adoption that implementation efforts should consider. Patients must be screened for eligibility and treatment and may need complete re-evaluation. Biologics patients express concerns about medication changes (Peyrin-Biroulet 2017; Waller et al. 2017). When the clinical stakes are high due to the debilitating nature of the illness being treated, there is more anxiety about change. This is particularly true if after a complex course of illness a person is finally enjoying improved function and quality of life. Thus, switching from an effective treatment is naturally more fraught than a new initiation in a person who is suffering. Thus, clinicians and patients may be reluctant to trial biosimilars.

Diffusion of Biosimilar Information

Multiple modes influence how clinicians incorporate new knowledge into clinical practices. These include clinical practice guidelines, primary clinical studies, trusted opinion leaders, clinical colleagues, continuing professional education events, media and industry marketing, clinical experience and patient/family values and preferences (Gabbay and le May 2016). Through these influences, clinicians develop clinical mindlines (Gabbay and le May 2016), which guide daily practice. In the case of complex interventions such as biosimilar initiation and switching, the mindlines of whole clinical teams, including, most importantly, patients, need to evolve as changes happen to care delivery.

When regulatory agencies grant market approval for new biosimilars, they aim to affirm biosimilar safety and efficacy, raise awareness of biosimilars and share knowledge tailored to prescribers. The FDA (2018) took tangible steps in promoting biosimilar use in its Biosimilar Action Plan, with network formation initiatives to facilitate data sharing between the FDA and foreign regulatory agencies to streamline drug application approval processes for European-approved biosimilars.

Professional organizations and patient advocacy groups have widespread influence for members, but few champion biosimilar use. The literature that explores the diffusion of information, for example, how these organizations develop their statements, how their funding relationship to pharmaceutical companies affects their position and how patients and physicians are impacted by their messaging, is not clear. The American College of Rheumatology (2018) position statement affirms the need for cost-effective treatments but raises concerns about the diligence of studies used for market approval.

The European League Against Rheumatism's (2018) position statement on biosimilars educates patients and emphasizes the need to strengthen patient input in policy formation. Some groups position themselves more strongly; Crohn's and Colitis Canada (n.d.) initiated a patient-driven letter-writing campaign to government about "forced switching." It is crucial

for implementation researchers and policy makers to reflect on how information and messaging reaches clinicians, teams and patients and how this diffusion of knowledge impacts adopters' attitudes to the introduction of innovations. These important influences on physician practice and patient decisions are little explored in the available literature.

Readiness

Greenhalgh et al.'s model (2004) defines the readiness of a clinical setting to implement innovations as arising from a perceived intolerable current situation (tension for change), clear implications of adopting an innovation to change the current situation, systems to monitor impact and resource availability to support innovation implementation.

The literature suggests a lack of tension to adopt biosimilars and little enthusiasm for biosimilars. A survey of Belgian rheumatology specialists and patients suggests significant adopter indifference to biosimilar approval (van Overbeeke et al. 2017). Rheumatologists perceived more risk with biosimilars because of a lack of clinical trials in specific indications. The literature repeatedly establishes the need to have more comprehensive data available to inform clinical decisions (Cohen et al. 2016; van Overbeeke et al. 2017) and to have monitoring systems in place to evaluate patient outcomes (Calvo et al. 2018; Health Improvement Scotland 2018).

Adopting biosimilars and facilitating switching require dedicated financial and human resources. These processes require increased clinic hours and extra staff and infrastructure to facilitate the switch and to collect data for monitoring patient outcomes. Determining organizational readiness to implement biosimilars requires a thorough evaluation of the clinical setting. Engaged management and front-line staff can develop locally suitable solutions to improve readiness. For example, Norway's initial policy to automatically switch patients from originator to biosimilar infliximab failed to attain the anticipated market share. This led to a concerted effort to consult stakeholders to develop a targeted educational campaign to promote biosimilar use and adjust pricing. This resulted in Norway's two approved biosimilars at the time reaching a majority market share (Institute of Health Economics 2016). The key to this more successful strategy to encourage the use of biosimilars was understanding organizational readiness and involving stakeholders in developing targeted strategies for biosimilar implementation.

Adoption by Individuals

Individual decisions to adopt and routinize an innovation are impacted by their needs, motivations, values, meanings ascribed to the innovation and decisions of their organization or patient/professional network. Relationship quality between adopters and organizations developing policy impacts the meaning attached to innovation (Greenhalgh et al. 2004).

Surveys and focus groups show patient concerns about clinical stability and treatment affordability with regard to biologics treatment and biosimilar switching. A focus group organized by five prominent Canadian patient organizations indicated that switching to a

different biologic or biosimilar is perceived as potentially disruptive (The Arthritis Society 2017). Surveys also indicate that patients lack knowledge about biosimilars and trust their physician's decision (Peyrin-Biroulet et al. 2017; van Overbeeke et al. 2017).

In 2015, the Government of the Netherlands framed biosimilar adoption as policies "for securing the affordability and accessibility of expensive medicines" (Government of the Netherlands 2015). This coincides with patient concerns about the affordability of biologic treatments. Where patients do not directly pay for drugs, policy efforts may tap into adopters' altruistic attitudes on collectively reducing health-associated costs. Messaging from government, professional and patient organizations, industry and media shapes meaning for adopters and fuels debates over cost savings, evidence quality, risks and safety (Cassels 2017; Rowland 2019). Divergent messaging poses challenges for adopters in making decisions about adopting biosimilars.

Current data also suggest that gaps of knowledge and uncertainty regarding biosimilars exist among physicians. Researchers who conducted a survey of US-based physicians and specialists concluded that there is a "significant need for evidence-based education" on biosimilars in the areas of understanding bioequivalence and differentiating between biologics and biosimilars (Cohen et al. 2016). Programs such as the FDA's Biosimilar Action Plan are designed to address these shortcomings by enhancing the resources available to prescribers regarding the approval of biosimilars (FDA 2018). In other surveys, European gastroenterologists and rheumatologists expressed doubts over the safety of switching patients to biosimilars (Sullivan et al. 2017; van Overbeeke et al. 2017). As a result of the uncertainty, in 2017, the European Medicines Agency and the European Commission published a guide for healthcare professionals on the benefits of biosimilars, unequivocally saying that there are "no differences" in the expected safety and efficacy between biologics and biosimilars.

Physicians and patients together negotiate knowledge, risk and implications. A recent survey of switched patients mentioned the lack of support from clinicians as a factor that negatively affected their switching experience (Attipoe et al. 2018). Health Improvement Scotland (2018) addressed this potential challenge by providing information letters to patients on biosimilars and treatment implications, preparing patients for physician encounters.

As an innovation, biosimilars cannot be assessed in isolation from their mode of administration and associated barriers and supports. In Canada, some biologics are linked to industry-sponsored patient support programs for drug access and delivery, such as Pfizer Inc.'s patient support program for their infliximab biosimilar, Inflectra (Pfizer Canada 2019). These programs are valued by patients and may drive an individual prescription decision (The Arthritis Society 2017).

Authoritative measures, such as Norway's initial adoption regulations, may boost initial implementation but not long-term routinization. Mandated (must-do's) adoption and switching may be the deciding factor for individual adoption. However, without stakeholder engagement and understanding of local context, mandated actions risk underestimating organizational capacity for the complex logistics of biosimilar implementation.

Implementation

Key to implementation are the features of the innovation, diffusion and dissemination, system and organization readiness and adoption.

In 2017, NHS England published the *Commissioning Framework for Biological Medicines (Including Biosimilar Medicines)*, which is, along with Scotland's (Health Improvement Scotland 2018), one of a few examples of biosimilar implementation found in the literature. The framework entrusted more than 150 Clinical Commissioning Groups (CCGs), bodies tasked with commissioning health services for regional NHS organizations, in England to engage stakeholders to gauge concerns and identify the cost/benefit of biosimilar uptake. CCGs and adopters then assess readiness and tailor implementation strategies for specific regions. The goal is 90% uptake by patients who have never before been treated with biologics within three months of framework enactment and 80% switching for existing patients within 12 months. Strategies include interprofessional clinical teams and tool kits to identify and educate patients eligible for switching. Financial incentives to providers fund educational programs and staff for extra clinic hours but are expected to cease over time as the numbers of patients eligible to switch fall.

The framework builds readiness by concerted support for innovation and adopter ability to monitor and evaluate patient outcomes, with CCG time and analytical resources dedicated to working with providers. NHS England recommends assessing the consequences of biosimilar implementation through pharmacovigilance and monitoring: reporting adverse drug reactions and observing financial benefits, clinical outcomes, patient perspectives and unexpected adoption challenges. Data are not yet published on the success of these measures.

Discussion

We reviewed the literature on the international experience with biosimilar implementation in the context of Greenhalgh et al.'s Diffusion of Innovations model. Using surveys, policy documents and position statements, we discussed biosimilar introduction as innovation, its diffusion process, challenges with clinical readiness and factors facilitating or hindering individual adoption.

Most information is from surveys on adopter perceptions and knowledge of biosimilars, which do not adequately explore the complex processes and circumstances that have an impact on patients, physicians and teams of allied healthcare providers (nurses, pharmacists) and healthcare organizations, as well as professional and patient associations' knowledge, meanings and practical challenges that shape clinician and patient decisions on biosimilars. There is little literature on the impact of relationships between patients, clinicians, organizations, industry and governing bodies on biosimilar implementation. Beyond examples from the UK and Scotland, the literature is scarce on implementation design and process evaluation.

Greenhalgh et al.'s model suggests that linkages between change agents, knowledge purveyors, resources systems and user systems are decisive for innovation diffusion and

implementation. Linkages build on networks, collaboration and relationships for understanding adopter readiness, creating shared meanings, transferring knowledge effectively and making decisions that align with adopter needs and values. Implementation can fail if relationships and bidirectional communication are not attended to. Crucial are effective stakeholder engagement and qualitative methods that better capture influences on patients' and on physicians' and teams' (biologics nurses, pharmacists) perceptions, resource constraints and decision-making early in policy and implementation design. Such engagement and research could help establish common ground and develop trust and shared language on the benefits and implications of introducing biosimilars, as well as uncover situations that could pose challenges for implementation.

The literature reveals that much of this sense-making and meaning-making around biosimilars is left to physician–patient encounters. Ideally, knowledge about biosimilar treatment and its implications for each patient are collaboratively negotiated during clinical conversations. This requires supporting providers and patients with consistent evidence-informed messaging, the time and resources for meaningful conversations and collaboratively created tools that satisfy their needs.

In the international experience, we observe a "disconnect" between organizations wishing to implement biosimilars into practice, adopters who must routinize the innovation and the research available to inform implementation. This disconnect goes beyond adopter values around biosimilars to understanding necessary resources that sustain biosimilar implementation in daily practice.

Evolving Context – An Addendum (January 1, 2020)

In the original article we reported that although Alberta and British Columbia are covering biosimilar medications as part of their provincial formularies, neither had instituted a switching policy. This is no longer the case.

Residents of British Columbia who are on an originator biologic being reimbursed by the provincial PharmaCare program are now required to switch to the biosimilar alternative as part of a two-phase initiative broken down by indication. The first phase (from May 27, 2019 to November 25, 2019) included patients who were taking the originator biologic for indications that included ankylosing spondylitis and rheumatoid arthritis. The second phase (from September 5, 2019 to March 5, 2020) was for patients who were taking the originator biologic for ulcerative colitis or Crohn's disease (Government of British Columbia 2019). Over the course of the two phases, the biologic would still be covered for the specified indication, however, the expectation is that patients and prescribers would work together to ensure the transition deadlines would be met (Government of British Columbia 2019).

In Alberta, however, all patients on one of the biologics with an approved biosimilar being covered by the provincial program are required to switch by June 30, 2020, regardless of indication (Alberta Blue Cross 2019).

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References

Alberta Blue Cross. 2019. "Government-Sponsored Biosimilar Initiative." Retrieved December 27, 2019. <https://www.ab.bluecross.ca/government-plan/biosimilar-initiative.php>.

American College of Rheumatology. 2018. American College of Rheumatology Position Statement: Biosimilars. Retrieved August 12, 2018. <https://www.rheumatology.org/Portals/0/Files/Biosimilars-Position-Statement.pdf>.

Attipoe, L., S. Patel, R. Brit, J. Crooks, K. Hunt and A. Grigoriou. 2018. What Factors Predict Good Patient Experiences of Switching from Reference Etanercept to an Etanercept Biosimilar in a South West London General Hospital? *Rheumatology* 57(3 Suppl): iii63. doi:10.1093/rheumatology/key075.285.

Calvo, B., J. Martinez-Gorostiaga and E. Echevarria. 2018. The Surge in Biosimilars: Considerations for Effective Pharmacovigilance and EU Regulation. *Therapeutic Advances in Drug Safety* 9(10): 601–08. doi:10.1177/2042098618790442.

Canadian Agency for Drugs and Technologies in Health (CADTH). 2018. *Biosimilars – Regulatory, Health Technology Assessment, Reimbursement Trends, and Market Outlook*. Ottawa, ON: CADTH. Retrieved August 2, 2018. <https://cadth.ca/sites/default/files/pdf/ES0317_biosimilars.pdf>.

Cassels, A. 2017, January 9. Why Biosimilars Should Be Interchangeable with Biologics. *Clinical Pharmacist*. Retrieved April 17, 2019. <https://www.pharmaceutical-journal.com/opinion/insight/why-biosimilars-should-be-interchangeable-with-biologics/20202121.article?firstPass=false>.

Cohen, H., D. Beydoun, D. Chein, T. Lessor, D. McCabe, M. Muenzberg et al. 2016. Awareness, Knowledge, and Perceptions of Biosimilars among Specialty Physicians. *Advances in Therapy* 33(12): 2160–72. doi:10.1007/s12325-016-0431-5.

Crohn's and Colitis Canada. (n.d.). The Issue: Forced Switching. Retrieved August 15, 2018. <http://action.crohnsandcolitis.ca/forced-switching>.

Elek, P., A. Harsányi, T. Zelei, K. Csetneki and Z. Kaló. 2017. Policy Objective of Generic Medicines from the Investment Perspective: The Case of Clopidogrel. *Health Policy* 121(5): 558–65.

European League Against Rheumatism. 2018, August. Biosimilars – Position Paper: Updating Position Statement from the European League Against Rheumatism (EULAR) Standing Committee of People with Arthritis/Rheumatism in Europe (PARE). Retrieved August 2, 2018. <https://www.eular.org/myUploadData/files/biosimilars_paper_updated_2018_09_14_dw.pdf>.

European Medicines Agency and the European Commission. 2017. *Biosimilars in the EU: Information Guide for Healthcare Professionals*. Retrieved August 4, 2018. <https://www.ema.europa.eu/en/documents/leaflet/biosimilars-eu-information-guide-healthcare-professionals_en.pdf>.

Gabbay, J. and A. le May. 2016. Mindlines: Making Sense of Evidence in Practice. *British Journal of General Practice* 66(649): 402–03. doi:10.3399/bjgp16X686221.

Government of British Columbia. 2019. "Biosimilars Initiative for Patients." Retrieved December 27, 2019. <https://www2.gov.bc.ca/gov/content/health/health-drug-coverage/pharmacare-for-bc-residents/what-we-cover/drug-coverage/biosimilars-initiative-patients>.

Government of the Netherlands. 2015, December. Appendix 2: Complete Set of Regulations for Securing the Affordability and Accessibility of Expensive Medicines. Retrieved August 12, 2018. <https://www.government.nl/binaries/government/documents/publications/2016/03/07/appendix-2-complete-set-of-regulations-for-securing-the-affordability-and-accessibility-of-expensive-medicines/appendix-2-set-of-regulations.pdf>.

Greenhalgh, T., G. Robert, F. Macfarlane, P. Bate and O. Kyriakidou. 2004. Diffusion of Innovations in Service Organizations: Systematic Review and Recommendations. *The Milbank Quarterly* 82(4): 581–629. doi:10.1111/j.0887-378X.2004.00325.x.

Health Canada. 2017. Health Canada's 2017 Biosimilars Workshop: Summary Report. Retrieved August 14, 2018. <https://www.canada.ca/en/health-canada/services/drugs-health-products/biologics-radiopharmaceuticals-genetic-therapies/applications-submissions/guidance-documents/biosimilars-workshop.html>.

Health Improvement Scotland. 2018, March. *Biosimilar Medicines: A National Prescribing Framework*. Retrieved August 2, 2018. <http://www.healthcareimprovementscotland.org/his/idoc.ashx?docid=93faeca2-1f4d-4ffc-a41f-7a17909ae236&version=-1>.

Institute of Health Economics. 2016. *Towards an Alberta Approach for Biosimilar Reimbursement – Summary Report of the IHE Biosimilars Forum October 6, 2016*. Retrieved July 20, 2018. <https://www.ihe.ca/download/towards_an_alberta_approach_for_biosimilar_reimbursement.pdf>.

Milne, V., J. Tepper and M. Taylor. 2017, November 13. Do Drug Funding Decisions Need PR? *Healthy Debate*. Retrieved April 17, 2019. <https://healthydebate.ca/2017/11/topic/drug-funding-advertisements>.

National Health Service (NHS) England. 2017. *Commissioning Framework for Biological Medicines (Including Biosimilar Medicines)*. Retrieved August 2, 2018. <https://www.england.nhs.uk/wp-content/uploads/2017/09/biosimilar-medicines-commissioning-framework.pdf>.

Patented Medicines Prices Review Board. 2017. Potential Savings from Biosimilars in Canada. Retrieved July 30, 2018. <https://www.pmprb-cepmb.gc.ca/view.asp?ccid=1304>.

Peyrin-Biroulet, L., S. Lönnfors, X. Roblin, S. Danese and L. Avedano. 2017. Patient Perspectives on Biosimilars: A Survey by the European Federation of Crohn's and Ulcerative Colitis Associations. *Journal of Crohn's and Colitis* 11(1): 128–33. doi:10.1093/ecco-jcc/jjw138.

Pfizer Canada. 2019. Inflectra. Retrieved June 1, 2019. <https://www.inflectra.ca/>.

Rowland, C. 2019, January 9. 'Marketers Are Having a Field Day': Patients Stuck in Corporate Fight against Generic Drugs. *The Washington Post*. Retrieved April 17, 2019. <https://www.washingtonpost.com/business/economy/drugmakers-alleged-scare-tactics-may-hold-back-competition/2019/01/09/612ac994-046d-11e9-9122-82e98f91ee6f_story.html?noredirect=on&utm_term=.5cc17a924c19>.

Sullivan, E., J. Piercy, J. Waller, C.M. Black and S. Kachroo. 2017. Assessing Gastroenterologist and Patient Acceptance of Biosimilars in Ulcerative Colitis and Crohn's Disease across Germany. *PLoS One* 12(4): e0175826. doi:10.1371/journal.pone.0175826.

The Arthritis Society. 2017, March. *Biosimilar Focus Group Project*. Retrieved August 6, 2018. <https://arthritis.ca/AS/media/pdf/About%20Arthritis/TAS_FocusGroupReport_Final_ENG_V5.pdf>.

U.S. Food and Drug Administration (FDA). 2018, July. *Biosimilars Action Plan: Balancing Innovation and Competition*. Retrieved August 15, 2018. <https://www.fda.gov/downloads/Drugs/DevelopmentApprovalProcess/HowDrugsareDevelopedandApproved/ApprovalApplications/TherapeuticBiologicApplications/Biosimilars/UCM613761.pdf>.

van Overbeeke, E., B. De Beleyr, J. de Hoon, R. Westhovens and I. Huys. 2017. Perception of Originator Biologics and Biosimilars: A Survey among Belgian Rheumatoid Arthritis Patients and Rheumatologists. *BioDrugs* 31(5): 447–59. doi:10.1007/s40259-017-0244-3.

Waller, J., E. Sullivan, J. Piercy, C.M. Black and S. Kachroo. 2017. Assessing Physician and Patient Acceptance of Infliximab Biosimilars in Rheumatoid Arthritis, Ankylosing Spondyloarthritis and Psoriatic Arthritis across Germany. *Patient Preference and Adherence* 11: 519–30. doi:10.2147/PPA.S129333.

RESEARCH PAPER

Health Policy as a Barrier to First Nations Peoples' Access to Cancer Screening

Les politiques de santé comme obstacle à l'accès des Premières Nations au dépistage du cancer

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Abstract

Background: First Nations peoples in Ontario are facing increasing rates of cancer and have been found to have poorer survival. Cancer screening is an important strategy to improve cancer outcomes; yet, Indigenous people in Canada are less likely to participate in screening. Ontario has established organized breast, cervical and colorectal cancer screening programs; this paper examines the health policy context that informs these programs for First Nations peoples in the province.

Method: This paper follows an embedded multiple-case study design, drawing upon a document review to outline the existing policy context and on key informant interviews to explore the aforementioned context from the perspective of stakeholders.

Results: Policies created by agencies operating across federal, regional and provincial levels impact First Nations peoples' access to screening. Interviews identified issues of jurisdictional ambiguity, appropriateness of program design for First Nations persons and lack of cultural competency as barriers to participation in screening.

Conclusion: Federal, provincial and regional policy makers must work in collaboration with First Nations peoples to overcome barriers to cancer screening created and sustained by existing policies.

Résumé

Contexte : Les membres des Premières Nations de l'Ontario sont confrontés à des taux de cancer croissants et leur survie est moins bonne. Le dépistage du cancer est une stratégie importante pour améliorer les résultats, pourtant, les Autochtones au Canada sont moins susceptibles de participer au dépistage. L'Ontario a établi des programmes organisés de dépistage du cancer du sein, du col utérin et colorectal. Cet article examine le contexte des politiques de santé qui sous-tendent ces programmes pour les membres des Premières Nations de la province.

Méthode : Cet article emploie une conception intégrée d'études de cas multiples, s'appuyant sur un examen de la documentation pour décrire le contexte politique en place et s'appuyant sur des entretiens avec des informateurs clés pour explorer le contexte susmentionné du point de vue des parties prenantes.

Résultats : Les politiques élaborées par les organismes fédéraux, régionaux et provinciaux ont une incidence sur l'accès des Premières Nations au dépistage. Les entrevues ont permis de révéler des problèmes liés à une ambiguïté territoriale, à la pertinence de la conception des programmes pour les Premières Nations et au manque de compétences culturelles comme obstacles à la participation au dépistage.

Conclusion : Les décideurs fédéraux, provinciaux et régionaux doivent travailler en collaboration avec les Premières Nations pour surmonter les obstacles au dépistage du cancer créés et maintenus par les politiques en vigueur.

Introduction: Cancer in Ontario's First Nations Population

Indigenous peoples throughout Canada (Table 1) continue to face pressing health inequities, including persistent disparities in health outcomes (Cunningham 2011; Health Council of Canada 2013; King et al. 2009; Marrett and Chaudhry 2003; Nishri et al. 2014; Tjepkema et al. 2009). In 2015, the Truth and Reconciliation Commission of Canada (TRC) called upon the federal government and all Canadians to take action to mitigate these health challenges (TRC 2015). This and other recent socio-political shifts have contributed to what Kingdon (2011) coined as a changing health policy system climate, which he defined as an increased capacity to develop policies targeting the system's capability to address long-standing issues. The increasing burden of several cancers among Indigenous peoples in Ontario is an example of a health challenge that needs to be addressed, as described in the calls to action put forward by the TRC. Our paper aims to explore the ways in which the current health policy shapes access to and participation in cancer screening among First Nations communities in Ontario and to highlight opportunities for policy change. Although cancers affect all Indigenous peoples throughout Canada (First Nations, Inuit and Métis), the scope of this paper is limited to exploring cancer policies as these apply to First Nations in Ontario.

In Canada, the provision of healthcare for First Nations and Inuit is a fiduciary responsibility of the federal government. This responsibility resulted from the 1867 *British North America Act*, which made First Nations and their lands an exclusive federal jurisdiction (Waldram et al. 2006). As such, several programs and services typically coordinated and delivered by provincial and municipal levels of government for non-Indigenous Canadians are federally provided on First Nations reserves. This programming entails the creation and oversight of health services in the community as well as the subsidizing of the cost of medical transportation to access provincial health services not available locally (Hurley 2000). This policy structure is highly contested and has often been criticized as being a patchwork approach, characterized by shifting responsibilities between federal and provincial governments as well as a lack of accountability and clarification around responsibilities (Dwyer et al. 2013; Lavoie 2013; National Collaborating Centre for Aboriginal Health 2011; Snyder et al. 2015). As policy and jurisdiction are often found to be discordant (Jordan's Principle Working Group 2015) for the delivery of healthcare to First Nations, it is critical to understand the impact that existing health policies have on participation in cancer screening if these services are to be improved.

Historically, First Nations in Canada have had much lower rates of cancer incidence and mortality than non-First Nations Canadians (Gillis et al. 1991; Morgan and Laing 1981; Young and Frank 1983). However, cancer has recently emerged as a significant health challenge. Although there is a paucity of detailed data on both the prevalence and incidence of cancer among all Indigenous populations in Ontario (Chiefs of Ontario and Cancer Care Ontario 2016), there are some data to suggest that the incidence of several cancers has risen quickly among First Nations in the province, and the survival rate is poorer than before (Marrett and Chaudry 2003; Nishri et al. 2014; Withrow et al. 2017).

TABLE 1. Key terms used

Key term	Definition
Aboriginal	Aboriginal Peoples of Canada is the term used within Section 35 of the 1982 *Constitution Act*. It includes Indian (First Nations), Inuit and Métis.
First Nations	First Nations are the largest Indigenous population in Canada, south of the Arctic. First Nations peoples throughout Canada are further classified as "status" and "non-status". Status entails formal recognition of First Nations identity by the federal government and subsequently guarantees access to federal treaty rights and programs (i.e., Non-Insured Health Benefits; see also: Hurley 2000).
First Nations reserves	First Nations reserves are tracts of land that have been set apart by the Crown for the use and benefit of a First Nations band and its status members.
Indigenous	Indigenous communities, peoples and nations have a historical continuity with pre-invasion and pre-colonial societies that developed on their territories and consider themselves distinct from other sectors of society that now prevail on those territories. In Canada, Indigenous populations comprise First Nations, Inuit and Métis peoples (see also: Indigenous and Northern Affairs Canada 2017; Kenrick and Lewis 2004; The United Nations 2007).
Inuit	The Inuit are the Indigenous peoples of Inuit Nunangat (Inuit Homeland), which spans the Arctic regions of Canada. There is a growing Inuit community in Ontario, with the majority living in Ottawa and Toronto.
Métis	The Métis are descendants of people born out of relations between Indian women and European men. They are a distinct aboriginal people with a unique history, culture, language and territory (Métis Nation of Ontario 2017).
Political territorial organization (PTO)	A PTO is a governing body representing the political aspirations of its First Nations communities to all levels of government. There are four PTOs operating in Ontario: The Association of Iroquois and Allied Indians (representing seven communities), Grand Council Treaty #3 (representing 28 communities in Ontario and Manitoba), Nishnawbe Aski Nation (representing 49 communities) and The Anishinabek Nation (representing 40 communities). There are also 13 independent First Nations communities who are not affiliated with any PTO.
Residential school system	The residential school system was a federal government assimilationist policy that operated from 1870 to 1996. Indigenous children were removed from their families and forced to attend government-funded institutions. The physical and mental impacts of residential school continue to manifest among survivors and their families (Kirmayer et al. 2003).
Truth and Reconciliation Commission of Canada (TRC)	The TRC was established in 2008 with a mandate to learn the truth about what happened in residential schools and inform Canadians about this history. Upon closing in 2015, the TRC released a document identifying 94 calls to action aimed at redressing the legacy of residential schools and advancing the process of reconciliation (TRC 2015).

Screening is a key strategy to reduce the burden of cancer. Through screening, persons without cancer symptoms are tested to identify risks; if needed, further diagnostic testing is offered. Screening cannot reduce the impact for all cancer types. Large randomized controlled trials have demonstrated the benefits of mammography to screen for breast cancer and fecal occult blood testing and flexible sigmoidoscopy for colorectal cancer (Canadian Task Force on Preventive Health Care 2011, 2016; Lauby-Secretan et al. 2015; Nelson et al. 2016; Tinmouth et al. 2016). For cervical cancer, well-designed ecological studies support the use of cervical cytology (Pap smears) for screening (Canadian Task Force on Preventive Health Care 2013; Cancer Care Ontario 2012b). In Ontario, Cancer Care Ontario (CCO) is the provincial agency responsible for advising the Ministry of Health and Long-Term Care (MOHLTC) on the cancer system, including access to cancer screening. CCO currently operates three organized screening programs: Ontario Breast Screening Program, Ontario Cervical Cancer Screening Program and ColonCancerCheck. Within each program,

screening guidelines have been developed based on expert reviews of available scientific evidence (Canadian Task Force on Preventive Health Care 2011; Murphy et al. 2011; Tinmouth et al. 2016; Warner et al. 2012). Table 2 provides an overview of the three organized screening programs currently operating within Ontario. Within CCO, the Indigenous Cancer Care Unit (ICCU) works to improve cancer care for Indigenous peoples throughout the province. ICCU is guided by the Joint CCO–Indigenous Cancer Committee (JOICC), which comprises members from each of the PTOs as well as other provincial Indigenous organizations.

TABLE 2. Ontario's organized cancer screening programs (adapted from: CCO 2016)

Screening program	Recommended screening test	Screening guidelines
Ontario Breast Screening Program (OBSP)	Digital mammography provided at an OBSP screening location every two years	Women in the age range 50–74 and have: no acute symptoms no personal history of breast cancer no current breast implants not had a mammogram within the past 11 months
High-risk OBSP	Digital mammography + magnetic resonance imaging every year	Women in the age range 30–69 and: having a physician's referral having no acute breast symptoms Fall into one of the following risk categories: known to be carriers of BRCA1 or BRCA2 gene mutation first-degree relative of a mutation carrier; has had genetic counselling and has declined genetic testing previously assessed by a genetic clinic as having >25% lifetime risk of breast cancer received radiation therapy to the chest before age 30 and at least eight years ago
Ontario Cervical Cancer Screening Program	Cytology (Pap) test performed at healthcare provider's office every three years	Women who are 21 years old and are or have been sexually active
ColonCancerCheck (CCC)	Guaiac fecal occult blood test (FOBT) completed at home every two years. Test obtained from family physicians/ nurse practitioners or by contacting Telehealth Ontario Note: Ontario will be switching from FOBT to the fecal immunochemical test for colorectal screening in 2019.	Men and women in the age range 50–74 and have: no first-degree relative who has been diagnosed with colorectal cancer no personal history of pre-cancerous colorectal polyps requiring surveillance or inflammatory bowel disease
CCC increased risk	Colonoscopy every 5–10 years	Men and women with a family history of colorectal cancer that includes one or more first-degree relatives who have been diagnosed with colorectal cancer, but do not meet the criteria for hereditary colorectal cancer syndromes

Unfortunately, Indigenous peoples throughout Canada are often less likely to be screened (Assembly of First Nations [AFN] 2009; Cancer Quality Council of Ontario 2019; Elias et al. 2011; Sheppard et al. 2010; Withrow et al. 2014). Several factors have been shown to limit First Nations' participation in cancer screening; these include difficulties accessing screening services, including coverage for costs of transportation to screening sites (First Nations Information Governance Centre 2012); inadequate health coverage for non-status First Nations individuals (Bent et al. 2007); negative experiences with the healthcare system, including racism (Allan and Smylie 2015; Brooks-Cleator et al. 2018; Loppie et al. 2014; Wylie and McConkey 2018); impact of intergenerational trauma, including experiences of residential schools, which leads to distrust of healthcare providers and resistance to engaging with the healthcare system (Browne and Fiske 2001; Kirmayer et al. 2003; Smith et al. 2005); low levels of awareness and community-based education emphasizing the importance of screening (Loppie and Wien 2005; O'Brien et al. 2009); and limited capacity and often lack of willingness of the healthcare system to engage Indigenous populations in disease prevention, acknowledge patient symptoms, and facilitate early-detection activities (Lavoie et al. 2016; Tjepkema 2002). The presence of other salient health issues such as food insecurity (Neufeld et al. 2017) and access to safe drinking water (Patrick 2011) also competes, understandably, with First Nations peoples' engagement with screening. Therefore, it is critical to address lower participation rates and later-stage cancer diagnosis among First Nations people to improve access to and use of cancer screening services.

Approach

We followed an embedded multiple-case study design (Yin 2014), drawing upon both a document review to describe the existing policy context and key informant interviews to explore this context from the perspective of multiple stakeholders. Research ethics board approval was obtained for this study through Sunnybrook Research Institute's Research Ethics Office, the University of Toronto Research Ethics Board and Health Canada and the Public Health Agency of Canada Research Ethics Board.

Selection of cases

Three regions were selected as cases based on Ontario's regional decision-making structure for cancer care (i.e., regional cancer programs [RCPs] are aligned with health regions, also called Local Health Integration Networks [LHINs]), with embedded units of analysis within each case including community service providers (e.g., primary care providers, hospitals, public health units) and First Nations communities. We also included overarching units of analysis such as governments and agencies at the federal (e.g., Health Canada, First Nations and Inuit Health Branch [FNIHB]) and provincial levels (e.g., MOHLTC, CCO). Table 3 provides an overview of our approach.

TABLE 3. Embedded multiple case study approach

Embedded and overarching levels of analysis	Case 1	Case 2	Case 3
Federal	Health Canada – First Nations and Inuit Health Branch; Canadian Partnership Against Cancer; Assembly of First Nations		
Provincial	Ministry of Health and Long-Term Care, Cancer Care Ontario, political territorial organizations		
Regional	Regional Cancer Program A	Regional Cancer Program B	Regional Cancer Program C
Sub-regional (First Nations community)	First Nations communities (urban, rural)	First Nations communities (rural, remote)	First Nations communities (remote)
Sub-regional (health service and support)		First Nations health service provider (urban)	First Nations health service provider (remote)

Our case selection criteria aimed for diversity of communities within the region in terms of variation in location of the First Nations communities (urban, rural and remote), First Nations community population sizes and political territorial organization (PTO) affiliation. Additional case selection criteria focused on diversity of regional characteristics, including total population of all First Nations communities within the regional boundaries and geographic location within the province (north/south).

Within the selected case regions, a number of First Nations communities were approached for inclusion as embedded units of analysis. Similar to the criteria used in selecting case regions, First Nations communities were approached based on location within the region (e.g., urban, rural and remote), population size and PTO affiliation. As relationships built upon trust are key to support successful research with First Nations communities (Castleden et al. 2012; Tobias et al. 2013), we also drew on pre-existing relationships between members of the research team and First Nations communities to finalize the selection. Further embedded units of analysis at the sub-regional level – such as local health service and support providers – were also included if identified as relevant to the study by a key informant.

Key organizations operating at federal and provincial levels were also included as overarching units of analysis for the case study. These included government ministries responsible for health and/or Indigenous populations; agencies/organizations responsible for organizing, financing, delivering and/or guiding cancer services; and organizations responsible for Indigenous populations.

Selection of policy documents

The objective of the document review was to develop an understanding of health policies impacting First Nations' access to cancer screening in Ontario. We searched publicly accessible databases (e.g., Medline, Google Scholar) and policy-related sources (e.g., Canadian Public Policy Collection) and we conducted general Web searches using a mix of key words and subject headings (e.g., MeSH) where relevant. We also conducted targeted searches of

key organizations' websites and asked key informants to identify relevant documents upon completion of their interview. Our search yielded a wide variety of documents, and we drew upon Pal (2014) to guide screening for inclusion, characterizing policy documents as those defining a course of action or inaction to be taken by an organization in addressing a given issue. Subsequently, we categorized the included policy documents under four headings: cancer screening, cancer services, general Indigenous health policy and Indigenous cancer screening policy.

Key informant interviews

Key informant interviews provided an interpretive lens on the policy context. A sampling frame was constructed based on available organizational information identified through the document review (e.g., organizational charts, Web profiles) and pre-existing contacts. Purposive sampling was used to guide recruitment of interview participants to ensure balanced representation for each case and level of analysis. Snowball sampling was also used as a secondary sampling method.

A semi-structured interview guide was developed and pilot-tested for the study. Interviews were conducted by the lead author, Joshua K. Tobias, either in person or by telephone between January 2015 and September 2016. Interviews were audio-recorded and transcribed verbatim. Thematic analysis guided exploration of the key informant interview data, drawing upon an iterative approach used to identify and refine themes. QSR International's NVivo 10 Software was used to both code and analyze data. Themes and coding framework were also reviewed and discussed through presentations given to the staff and leadership at CCO's ICCU and JOICC.

Results: Document Review

The document review identified 34 relevant documents, with nine relating to cancer screening, four to cancer services, 12 to general Indigenous health policy and nine to Indigenous cancer screening policy. Table 4 (available online at longwoods.com/content/26132) provides a summary of the policy documents included in the review.

Organizational landscape of cancer screening policy making for First Nations

The review of the documents listed in Table 4 clearly demonstrates that First Nations health policy in Canada is characterized by complex relationships between multiple federal, provincial/territorial, regional, municipal governments and Indigenous authorities. Each plays a unique role in a system of healthcare delivery rooted in 19th century legislation (e.g., the *British North America Act*, The *Indian Act*) and further supported by the 1979 Federal Indian Health Policy as well as Section 35 of the 1982 *Constitution Act*. As it pertains specifically to cancer screening, the FNIHB Non-Insured Health Benefits (NIHB) Program includes medical transportation benefits detailed within the NIHB Medical Transportation

Framework (AFN, 2014; Health Canada 2005). This includes services covered by provincial health plans, including cancer screening services recommended within Ontario's organized cancer screening programs. Cost for medical travel for screening and costs incurred while travelling to participate in screening are reimbursed through the program, which includes necessary health services that are unavailable on-reserve or in the patient's community of residence. This policy predominantly impacts access to mammography and colonoscopy.

The Canadian Partnership Against Cancer (CPAC), an independent organization funded by the federal government with a mandate to shape a national strategy for cancer control, contributes to the improvement of provincial screening programs by evaluating and analyzing emerging research as well as assessing evidence for screening guidelines. CPAC has also launched the First Nations, Inuit and Métis Action Plan on Cancer Control (CPAC 2011) aimed at improving the quality of Indigenous patients' cancer care. This initiative includes specific components addressing access to cancer-related programs and services in remote and rural communities.

Provincial-level policy context

In Ontario, MOHLTC has the overall responsibility of governance of the healthcare system, including its funding arrangements and service delivery throughout the province. This entails oversight of strategic policy direction and priorities, legislation, establishment of funding models and the monitoring of and reporting on the performance of the healthcare system. In 2006, the *Local Health System Integration Act* introduced a regional health service model to the province through the creation of 14 Local Health Integration Networks (LHINs). The Patients First: Action Plan for Healthcare (MOHLTC 2015b) provides guidelines for healthcare delivery in Ontario. The plan is centred upon improving access to services, coordinating and integrating care closer to home, providing education and information as well as sustaining the value and quality of the healthcare system. The MOHLTC-supported Northern Health Travel Grant provides financial support to all eligible residents of Northern Ontario requiring a medical specialist or services that are not available locally but rather only at a designated health facility that is at least 100 km away.

CCO is an agency of the Ontario Government and the principal advisor to MOHLTC on cancer services and the cancer system. CCO is governed by the provisions of both the 1990 *Cancer Act* and the 2009 Memorandum of Understanding with MOHLTC (CCO 2011). CCO's key commitments are detailed within formal cancer plan documents, with the most recent, Ontario Cancer Plan IV (CCO 2015c), including implementation of provincial cancer prevention and screening programs, managing the performance of the cancer system in collaboration with 14 regional cancer programs and LHINs and ensuring health equity across the cancer system. The mandate of ICCU is
currently set out in their Aboriginal Cancer Strategy III (CCO 2015b), which was collaboratively developed with First Nations, Inuit and Métis peoples throughout the province.

Regional-level policy context

The CSIC report (2001) addressed major concerns about Ontario's cancer system (although notably made no mention of cancer services for Indigenous peoples) and was the precursor to major restructuring of cancer services in 2004, ultimately resulting in the creation of RCPs (Sullivan et al. 2005). There are currently 14 RCPs; these are generally aligned with the LHIN structure, with the exceptions of the Toronto Central LHIN, which is supported by two RCPs, and the Mississauga-Halton and Central West LHINs, which are supported by the same RCP. The RCPs are based at regional cancer centres that are integrated with host hospitals, with the RCP Regional Vice President working under a dual-reporting relationship with the CEOs of the host hospital and CCO. Within this relationship, CCO and RCPs work together to promote high performance in the cancer system across the province, taking into account the unique needs of each region. Furthermore, the RCPs are responsible for ensuring that service providers meet the requirements and targets set out in their partnership agreements with CCO. CCO's ICCU works with RCPs, providing support to the Indigenous populations within their regions. ICCU and RCPs collaborate with Indigenous communities and organizations within the individual regions to develop Regional Indigenous Cancer Plans. These plans outline key cancer priorities and strategies for addressing those tailored to the unique needs of the Indigenous population within each region while aligning with both the time frame and strategic priorities outlined in the Aboriginal Cancer Strategies (CCO 2012a, 2015b).

Results: Key Informant Interviews

A total of 44 of the 68 invitees participated in the interview (participation rate: 65%). Of these, 13 were recruited from the overarching (federal, provincial) units of analysis, with the remaining 31 individuals located within the regional cases (case 1: $n = 9$; case 2: $n = 12$; case 3: $n = 10$).

Results of key informant interviews: perspectives on the policy context for cancer screening

Three key themes emerged from key informant interviews: jurisdictional ambiguity, appropriateness of cancer screening program design for First Nations and lack of cultural competence. Although cases were selected to achieve diversity across the range of case selection criteria identified, the cross-case comparison revealed consistent thematic similarities among the selected cases. As such, the results of key informant interviews across each level of analysis are presented thematically, with regional differences highlighted where relevant. Table 5 (available online at longwoods.com/content/26132) provides representative quotes for each of the three identified themes.

Jurisdictional ambiguity

Each interview participant was asked if they could identify the key organizations responsible for developing policies that influence how and when First Nations peoples throughout the

province receive cancer screening. Across all levels of analysis, very few participants could identify multiple organizations or describe the approach that organizations would use to develop relevant policies. This pointed to a lack of awareness of jurisdictional responsibilities as well as of how various organizations collaborate. Among interviewees who were able to identify specific organizations, most identified MOHLTC, FNIHB and/or CCO as primarily responsible for setting relevant policies.

Within the three cases selected, awareness of federal responsibilities for healthcare provision was high among federal-level informants as well as community (sub-regional) key informants across all cases. Notably, awareness of the federal responsibility for First Nations healthcare was low among regional and provincial key informants. All key informants were also largely unable to speak to the process of operationalizing responsibilities across federal and provincial jurisdictions. Within case 3, key informants described challenges in obtaining medical transportation to access mammography services. Although several community members have detailed their experiences of being denied medical transportation, key informants were unable to articulate which agency would be best positioned to collaborate with FNIHB to address the issue. Among individuals within the RCPs, there was a greater level of awareness pertaining to transportation barriers in regions with higher numbers of remote communities. Some key informants, especially those working in remote areas, were able to speak in detail about policies such as FNIHB's NIHB Medical Transportation Framework or MOHLTC's Northern Health Travel Grants.

When discussing policy development, key informants operating primarily within the overarching federal or provincial organizations could often identify multiple sources that they believed informed the creation of programs and policies specific to their own organization, such as clinical guidelines presented by the Canadian Task Force on Preventive Health Care. However, these individuals were often unsure how other organizations developed policies that impact on access to screening. For example, FNIHB participants were aware of CPAC as an organization but could not speak to CPAC's First Nations, Inuit and Métis Action Plan on cancer control recommendations regarding health administrative data or how these have supported the development of First Nation, Inuit and Métis (FNIM) identifiers within cancer databases.

Appropriateness of cancer screening program design for First Nations

Key informants in each of the three cases expressed that policies are developed largely to address the needs of the non-Indigenous population and that the unique needs of First Nations peoples are only considered at later stages. Typically, when facing difficulties in policy implementation, for example, current clinical guidelines for screening were often questioned in terms of appropriateness. Given the high rates of several cancer risk factors among First Nations, many community-level key informants suggested that screening program design may require modification.

In case 1, CCO policies and pathways for providing screening results to a First Nations individual were described as requiring adaptation to the reality of First Nations' daily life in

the region. It was noted that individuals in this region may have both an on-reserve address and an urban residence. As such, delays in the receipt of mailed correspondence such as invitations to screening, results or follow-up requests may occur. This issue was believed to be compounded by an absence of primary care practitioners and an increased reliance on walk-in clinics for healthcare.

At the organizational level, discussion regarding the appropriateness of a screening program design varied among key informants. Those working in First Nations organizations believed that processes had been developed without community voice. However, this often also included a recognition that the prevailing culture was slowly changing as a result of assertions for inclusion from First Nations leadership.

Many informants suggested that the key to improving cancer screening participation among First Nations is the Indigenous representation in both processes of policy development and evaluation. Although it was articulated that inclusion of Indigenous perspectives in policy evaluation remains rare, it was noted that opportunities for Indigenous voices to be heard in policy evaluation are increasing. In case 2, a committee of representatives from the First Nations communities in the region, CCO and RCP has been created. This group meets regularly to provide guidance on the development and implementation of the Regional Aboriginal Cancer Plan.

Lack of cultural competence

Those working closely with First Nations communities, typically at the regional and community levels, identified cultural competency issues as a barrier to participation in screening. Previous experience with racism when seeking healthcare was discussed as causing apprehension and anxiety about further engagement with the healthcare system, including hesitancy to participate in screening. Community-level informants from all cases spoke about having directly experienced overtly racist treatment in healthcare settings.

Some informants at federal and provincial levels who were responsible both for policy design as well as implementation recognized the need for increased cultural training for healthcare service providers. Importantly, however, a small number of participants at this level of analysis maintained that this type of training was not a priority and argued that staff should be focused on developing other skills.

Discussion and Conclusion

Given the increasing cancer incidence and poorer cancer-related survival rate among First Nations, our study sought to explore how existing policies shape access to and participation in cancer screening among First Nations communities in Ontario. We identified three key factors impacting the policy context for cancer screening: jurisdictional ambiguity, absence of an Indigenous-specific screening program design and lack of cultural competence. While the first reflects long-standing governance arrangements (i.e., federal responsibility for First Nations health) that are challenging to change but possible to clarify, the other two reflect processes that are more amenable to change and align well with the more general shift from

a predominantly medical model of healthcare to an increasingly patient-centred orientation (MOHLTC 2015b) toward health services and policy (e.g., increased First Nations voice in policy design).

Although there were differences in the First Nations population structure (e.g., population size, percentage urban/rural/remote) across each of the three cases, the overall descriptions of challenges inherent in cancer screening were quite similar in nature for all the three cases. For instance, difficulties pertaining to access to transportation for screening due to jurisdictional ambiguity or experiencing racism when seeking screening was. The inclusion of First Nations' representation in policy design and evaluation was discussed as being somewhat easier in case 3, where the majority of individuals live in First Nations communities located in rural areas.

Despite commitments made both federally and provincially (Jordan's Principle Working Group 2015), insights from key informants reveal that barriers to effective screening policies and programs are rooted in long-standing issues of jurisdictional ambiguity. This has resulted in both the creation of policies that do not necessarily reflect First Nations' lived realities and the increase in frustration with the cancer screening system for First Nations peoples and uncertainty regarding where best to address grievances. Our results echo existing critiques of the current Indigenous health policy structure (Dwyer et al. 2013; Lavoie et al. 2010; Lavoie 2013; National Collaborating Centre for Aboriginal Health 2011; Snyder et al. 2015) and corroborate the emerging discourse from within First Nations' health literature that supports the creation of a national Indigenous policy framework (Lavoie 2013; Richmond and Cook 2016).

Inadequate Indigenous-specific screening program design results from the relatively few Indigenous persons who are involved in the process of designing a health policy. Key policy stakeholders operating at all levels should seek to increase opportunities to include Indigenous communities and organizations throughout the province in developing and carrying out evaluations. Across Canada, First Nations communities have increasingly begun asserting self-determination within health service delivery – including policy design and evaluation – with measurable improvements in health outcomes (Lavoie et al. 2015; Richmond and Cook 2016). Within our study, several community-level informants shared their frustration with the NIHB program and its ambiguities in enabling access to screening. However – as discussed by one key informant involved in the process – it is encouraging to note that this program is currently under joint review within a collaborative process, including both Health Canada and Indigenous representatives from across the country.

The three cases produced consistent findings that suggest that developing effective and appropriate cancer screening policies requires increased attention to the specific realities and needs of Indigenous populations. Currently, screening guidelines in Ontario do not make individualized recommendations for the First Nations population. There is some limited evidence suggesting that developing guidelines specific to individual ethnic groups may improve participation in screening (Williams et al. 2016). A comprehensive review of this issue at the provincial or national level, inclusive of Indigenous voices, may help clarify how a screening

program design could be modified to better serve the needs of Indigenous communities.

CCO has entered into a number of formalized relationships with First Nations PTOs and independent communities through the signing of relationship protocols. These agreements provide public recognition of the relationship and enhance accountabilities, creating greater capacity for the inclusion of First Nations' within the development, implementation and evaluation of provincial cancer initiatives and policies.

Our results are in line with those of others who have emphasized the critical importance of enacting specific policies and procedures aimed at promoting increased cultural safety (Allan and Smylie 2015; Baba 2013; Brascoupé and Waters 2009). Culturally competent healthcare has the potential to improve health disparities by increasing awareness of and addressing root causes such as structural racism and discrimination (Brooks-Cleator et al. 2018; Churchill et al. 2017). Cultural competency is indisputably important for healthcare providers who interact with Indigenous peoples on a daily basis; it is equally important among those contributing to policy development and implementation (e.g., RCP/CCO Leadership). In February 2016, the provincial government declared that Indigenous cultural competency and anti-racism training would be mandatory for all public service employees (Ontario 2016). Cancer screening leadership should be proactive in identifying existing training opportunities and ensuring that their staff is educated and aware of Indigenous realities in Canada in the spirit of reconciliation. This endeavour may present resource capacity challenges specifically within the RCPs, where program leadership will need to find ways to accommodate staffing requirements (e.g., paid time for taking this training). The province should seek to support RCPs in facilitating staff participation with cultural competency training.

It is also important to note that key informants across all levels of the health system cited examples of successful initiatives and expressed optimism for the future of Indigenous cancer screening.

> We just recently had a cervical screening clinic in one of our [Indigenous] communities and it was really well attended. […] It worked. It was evidence of increasing momentum that comes when you keep investing the time to say this relationship and doing this with you is really important and I will come back to keep doing this with you even if the numbers aren't there. You never do the math on the cost per patient. (RCP Informant)

The political climate in Canada appears to have shifted to be more receptive to and aware of the unique needs of the Indigenous population. As such, cancer screening policy in Ontario should capitalize on this new landscape to develop and produce new initiatives. Sharing information on successful initiatives by leveraging knowledge translation and exchanging opportunities in the cancer system would facilitate the adaptation and adoption of any lessons learned.

The findings in this paper are focused on screening policies and perspectives, as these apply to First Nations populations in Ontario. However, Ontario is also home to growing Métis and Inuit populations, each of which faces unique challenges in accessing cancer screening. Recent research demonstrates that cancer is also an increasing burden for these two groups (Cancer Care Ontario 2015; Cancer Quality Council of Ontario 2016; Chiefs of Ontario and Cancer Care Ontario 2016; Métis Nation of Ontario and Tungasuvingat Inuit and Cancer Care Ontario 2017). It was outside the scope of this study to explore features of the policy context facing Métis and Inuit in the province that are unique to these populations or common to First Nations; future research, however, should build on the current work to explore these features.

In 2015, the TRC put forward 94 calls to action intended to redress the legacy of residential schools and advance the process of Canadian reconciliation. Among the 94 calls to action, seven explicitly focus on improving the health of Indigenous peoples. These include calls upon governments to recognize the current state of Aboriginal health and implement healthcare rights (call 18); establish measureable goals toward closing the gap in health outcomes (call 19); address jurisdictional disputes (call 20); and require medical and nursing students to take a course on Aboriginal health issues (call 24). Our study identifies key structural issues and processes that should be addressed by agencies shaping cancer screening policy to successfully implement the TRC calls to action focusing on health.

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References

Allan, B. and J. Smylie. 2015. *First Peoples, Second Class Treatment: The Role of Racism in the Health and Well-Being of Indigenous Peoples in Canada*. Toronto, ON: The Wellesley Institute.

Assembly of First Nations (AFN). 2009. *Access to Cancer Screening and First Nations*. Retrieved August 2016. <http://s3.amazonaws.com/zanran_storage/64.26.129.156/ContentPages/2467254705.pdf>.

Assembly of First Nations (AFN). 2014. *Action on Non-Insured Health Benefits*. Retrieved January 2016. <http://www.afn.ca/uploads/files/aga2014/non-insured_health_benefits_fe.pdf>.

Baba, L. 2013. *Cultural Safety in First Nations, Inuit and Métis Public Health: Environmental Scan of Cultural Competency and Safety in Education, Training, and Health Services*. Prince George, BC: National Collaborating Centre for Aboriginal Health.

Bent, K., J. Havelock and M. Haworth-Brockman. 2007. *Entitlements and Health Services for First Nations and Métis Women in Manitoba and Saskatchewan. (Project #150).* Winnipeg, MB: Prairie Women's Health Centre of Excellence.

Brascoupé, S. and C. Waters. 2009. Cultural Safety: Exploring the Applicability of the Concept of Cultural Safety to Aboriginal Health and Community Wellness. *Journal of Aboriginal Health* 5(2): 6–41. doi:10.3138/ijih.v5i2.28981.

Brooks-Cleator, L., B. Philipps and A. Giles. 2018. Culturally Safe Health Initiatives for Indigenous Peoples in Canada: A Scoping Review. *Canadian Journal of Nursing Research* 50(4): 202–13. doi:10.1177/0844562118770334.

Browne, A. and J. Fiske. 2001. First Nations Women's Encounters with Mainstream Health Care Services. *Western Journal of Nursing Research* 23(2): 126–47. doi:10.1177/019394590102300203.

Canadian Partnership Against Cancer (CPAC). 2006. *The Canadian Strategy for Cancer Control: A Cancer Plan for Canada*. Retrieved January 2015. <https://www.partnershipagainstcancer.ca/wp-content/uploads/2017/09/canadian-strategy-for-cancer-control-a-cancer-plan-for-canada.pdf>.

Canadian Partnership Against Cancer (CPAC). 2009. *Quality Determinants for Colorectal Screening in Canada*. Retrieved January 2015. <http://www.iccp-portal.org/sites/default/files/resources/QD_for_CRC_Screening_in_Canada_2009-10-05_v16.pdf>.

Canadian Partnership Against Cancer (CPAC). 2010. *Strategies to Maximize Participation in Cervical Cancer Screening in Canada*. Retrieved January 2015. <https://www.partnershipagainstcancer.ca/topics/cervical-cancer-screening-max-participation/#>.

Canadian Partnership Against Cancer (CPAC). 2011. *First Nations, Inuit, and Métis Action Plan on Cancer Control*. Retrieved January 2016. <https://content.cancerview.ca/download/cv/fnim/documents/fnimactionplannov11pdf?attachment=0>.

Canadian Task Force on Preventive Health Care (CTFPHC). 2011. Recommendations on Screening for Breast Cancer in Average-Risk Women Aged 40–74. *CMAJ* 183(17): 1991–2001. doi:10.1503/cmaj.110334.

Canadian Task Force on Preventive Health Care (CTFPHC). 2013. Recommendations on Screening for Cervical Cancer. *CMAJ* 185(1): 35–45. doi:10.1503/cmaj.121505.

Canadian Task Force on Preventive Health Care (CTFPHC). 2016. Recommendations on Screening for Colorectal Cancer in Primary Care. *CMAJ* 188(5): 340–48. doi:10.1503/cmaj.151125.

Cancer Care Ontario (CCO). 2011. *Ontario Cancer Plan III*. Retrieved January 2016. <http://ocp.cancercare.on.ca/cms/One.aspx?portalId=77515>.

Cancer Care Ontario (CCO). 2012a. *Aboriginal Cancer Strategy II*. Retrieved January 2016. <https://www.cancercareontario.ca/sites/ccocancercare/files/assets/CCOACS2.pdf>.

Cancer Care Ontario (CCO). 2012b. *Cervical Screening Program 2012 Report*. Toronto, ON: Cancer Care Ontario.

Cancer Care Ontario (CCO). 2015a. *Ontario Breast Screening Program (OBSP) Guidelines Summary*. Toronto, ON. Retrieved December 2015. <https://www.cancercareontario.ca/sites/ccocancercare/files/assets/OBSPGuidelinesSummary.pdf>.

Cancer Care Ontario (CCO). 2015b. *Aboriginal Cancer Strategy III*. Retrieved January 2016. <https://www.cancercareontario.ca/sites/ccocancercare/files/assets/CCOAboriginalStrategy3.pdf>.

Cancer Care Ontario (CCO). 2015c. *Ontario Cancer Plan IV*. Retrieved January 2016. <http://ocp.cancercare.on.ca/>.

Cancer Care Ontario (CCO). 2016. *Ontario Cancer Screening Performance Report 2016*. Toronto, ON: Cancer Care Ontario.

Cancer Quality Council of Ontario. 2016. *Prevention System Quality Index*. Retrieved February 2020. <https://www.cancercareontario.ca/sites/ccocancercare/files/assets/PSQI2016FullReport.pdf>.

Cancer Quality Council of Ontario. 2019. *Cancer System Quality Index (CSQI) 2019: Modifiable Risk Factors in Ontario First Nations, Inuit, Métis [Internet]*. Retrieved January 2016. <https://www.csqi.on.ca/2019/indigenous>.

Cancer Services Implementation Committee (CSIC). 2001. *Report of the Cancer Services Implementation Committee*. Retrieved August 2016. <http://www.longwoods.com/articles/images/hudson.pdf>.

Castleden, H., V. Sloan Morgan and C. Lamb. 2012. I Spent the First Year Drinking Tea: Exploring Canadian University Researchers' Perspectives on Community-Based Participatory Research Involving Indigenous Peoples. *The Canadian Geographer* 56(2): 160–79.

Chiarelli A., G. Doyle, M. Harrison, D. Howell, L. Kan, V. Mai et al. 2008. Guidelines on Performance Measurement for Organized Cancer Screening Programs. Toronto: Canadian Partnership Against Cancer.

Chiefs of Ontario and Cancer Care Ontario. 2016. *Cancer in First Nations in Ontario – Risk Factors and Screening*. Toronto. Retrieved September 12, 2017. <https://www.cancercare.on.ca/FirstNationsReport>.

Churchill, M., M. Parent-Bergeron, J. Smylie, C. Ward, A. Fridkin, D. Smylie et al. 2017. *Evidence Brief: Wise Practices for Indigenous-Specific Cultural Safety Training*. Toronto, ON: St. Michael's Hospital.

Cunningham, M. 2011. State of the World's Indigenous Peoples. United Nations, Department of Economic and Social Affairs, Chapter 5. Retrieved October 27, 2016. <www.un.org/esa/socdev/unpfii/documents/SOWIP_chapter5.pdf>.

Dwyer, J., A. Boulton, J. Lavoie, T. Tenbensel and J. Cumming. 2013. Indigenous Peoples' Healthcare: New Approaches to Contracting and Accountability at the Public Administration Frontier. *Public Management Review* 16(8): 1091–112. doi:10.1080/14719037.2013.868507.

Elias, B., E. Kliewer, M. Hall, A. Demers, D. Turner, P. Martens et al. 2011. The Burden of Cancer Risk in Canada's Indigenous Population: A Comparative Study of Known Risks in a Canadian Region. *International Journal of General Medicine* (4): 699–709. doi:10.2147/IJGM.S24292.

First Nations Information Governance Centre. 2012. *First Nations Regional Health Survey (RHS) 2008/10: National Report on Adults, Youth and Children Living in First Nations Communities*. Ottawa, ON: FNIGC.

Gillis, D.C., J. Irvine, L. Tan, S. Chiu, L. Liu and D. Robson. 1991. Cancer Incidence and Survival of Saskatchewan Northerners and Registered Indians, 1967–1986. *Arctic Medical Research* (Suppl): 447–51.

Health Canada – First Nations and Inuit Health Branch. 2005. *Medical Transportation Policy Framework: Non-Insured Health Benefits (NIHB) Program*. Retrieved January 2016. <http://publications.gc.ca/site/eng/367064/publication.html>.

Health Canada. 2011. *Ontario Health Services Integration Fund Integration Plan*. Retrieved August 2016. <https://www.canada.ca/en/health-canada/services/first-nations-inuit-health/health-care-services/improving-access-health-services/health-services-integration-fund.html>.

Health Canada. 2012. *The First Nations and Inuit Health Branch Strategic Plan: A Shared Path to Improved Health*. Retrieved January 2016. <https://www.canada.ca/en/health-canada/services/first-nations-inuit-health/reports-publications/first-nations-inuit-health-strategic-plan-shared-path-improved-health-health-canada-2012.html>.

Health Canada. 2016. *Medical Transportation for OHIP Funded Cancer Screening Appointments: Ontario Region – Non-Insured Health Benefits Program*. Retrieved August 2016. <https://www.wrh.on.ca/Site_Published/wrh_internet/Document.aspx?Body.Id=74052&LeftNav.QueryId.Categories=294>.

Health Council of Canada. 2013. *Progress Report 2013: Health Care Renewal in Canada*. Retrieved December 2016. <http://www.healthcouncilcanada.ca/rpt_det.php?id=481>.

Hurley, M. 2000. *The Crown's Fiduciary Relationship with Aboriginal Peoples*. Ottawa, ON: Library of Parliament.

Indigenous and Northern Affairs Canada. 2017. About Indigenous and Northern Affairs Canada. Retrieved August 2017. <https://www.aadnc-aandc.gc.ca/eng/1100100010023/1100100010027>.

Jordan's Principle Working Group. 2015. *Without Denial, Delay, or Disruption: Ensuring First Nations Children's Access to Equitable Services through Jordan's Principle*. Ottawa, ON: Assembly of First Nations.

Kenrick, J. and J. Lewis. 2004. Indigenous Peoples' Rights and the Politics of the Term 'Indigenous'. *Anthropology Today* 20(2): 4–9. doi:10.1111/j.0268-540X.2004.00256.x.

King, M., A. Smith and M. Gracey 2009. Indigenous Health Part 2: The Underlying Causes of the Health Gap. *Lancet* 374: 76–85. doi: 10.1016/S0140-6736(09)60827-8.

Kingdon, J. 2011. *Agendas, Alternatives, and Public Policies: Updated (2nd ed)*. Boston, MA: Longman.

Kirmayer, L., C. Simpson and M. Cargo. 2003. Healing Traditions: Culture, Community and Mental Health Promotion with Canadian Aboriginal Peoples. *Australasian Psychiatry* 11(Supp. 1): S15–S23. doi:10.1046/j.1038-5282.2003.02010.x.

Lauby-Secretan, B., C. Scoccianti, D. Loomis, L. Benbrahim-Tallaa, V. Bouvard, F. Bianchini et al. 2015. Breast-Cancer Screening – Viewpoint of the IARC Working Group. *NEJM* 372(24): 2353–58. doi:10.1056/NEJMsr1504363.

Lavoie, J. 2013. Policy Silences: Why Canada Needs a National First Nations, Inuit, and Métis Health Policy. *International Journal of Circumpolar Health* 72(1): 22690. doi:10.3402/ijch.v72i0.22690.

Lavoie, J., A. Browne, C. Varcoe, S. Wong, A. Fridkin, D. Littlejohn et al. 2015. Missing Pathways to Self-Governance: Aboriginal Health Policy in British Columbia. *International Indigenous Policy Journal* 6(1). DOI: 10.18584/iipj.2015.6.1.2. Retrieved February 2020. <https://ir.lib.uwo.ca/iipj/vol6/iss1/2>.

Lavoie, J., E. Forget and A. Browne. 2010. Caught at the Crossroads: First Nations Health Care, and the Legacy of the Indian Act. *Pimatisiwin: A Journal of Aboriginal and Indigenous Community Health* 8(1): 83–100.

Lavoie, J., J. Kaufert, A. Browne and J. O'Neil. 2016. Managing Metajoosh: Determinants of First Nations' Cancer Care Decisions. *BMC Health Services Research* 16: 402. doi:10.1186/s12913-016-1665-2.

Loppie, C. and F. Wien. 2005. *Our Journey: First Nations Experience of Navigating Cancer Care*. Halifax, NS: Cancer Care Nova Scotia.

Loppie, S., C. Reading and S. de Leeuw. 2014. *Aboriginal Experiences with Racism and Its Impacts*. Prince George, BC: National Collaborating Centre for Aboriginal Health.

Marrett, L. and M. Chaudry. 2003. Cancer Incidence and Mortality in Ontario First Nations, 1968–1991 (Canada). *Cancer Causes Control* 14(3): 259–68. doi:10.1023/A:1023632518568.

Métis Nation of Ontario. 2017. *Who are the Métis*. Retrieved September 2017. <http://www.metisnation.org/culture-heritage/who-are-the-m%c3%a9tis/>.

Métis Nation of Ontario and Cancer Care Ontario. 2015. *Cancer in the Métis People of Ontario: Risk Factors and Screening Behaviours*. Ottawa, ON. Retrieved January 2016. <http://www.metisnation.org/media/653628/mno-cco-report-screen.pdf>.

Ministry of Health and Long-Term Care (MOHLTC). 2010. *Aboriginal Health Strategy Overview*. Retrieved January 2016. <http://health.chiefs-of-ontario.org/sites/default/files/files/MOHLTC%20Update%20to%20COO%20Forum%20Feb%2024.pdf>.

Ministry of Health and Long-Term Care (MOHLTC). 2015a. *Northern Health Travel Grants*. Retrieved January 2016. <http://www.health.gov.on.ca/en/public/publications/ohip/docs/brochure_northern_en.pdf>.

Ministry of Health and Long-Term Care (MOHLTC). 2015b. *Patients First: Action Plan for Healthcare*. Retrieved January 2016. <http://www.health.gov.on.ca/en/ms/ecfa/healthy_change/docs/rep_patientsfirst.pdf>.

Morgan K. and L.M. Laing. 1981. The Incidence of Cancer in Registered Indians of Alberta, 1974–1978. *Chronic Diseases in Canada* 2: 33.

Murphy J., E. Kennedy and S. Dunn. 2011. *Cervical Screening: Evidence-Based Series*. Toronto, ON: Cancer Care Ontario.

National Collaborating Centre for Aboriginal Health. 2011. *The Aboriginal Health Legislation and Policy Framework in Canada*. Retrieved March 2016. <https://www.ccnsa-nccah.ca/495/The_Aboriginal_health_legislation_and_policy_framework_in_Canada.nccah?id=2>.

Nelson, H.D., R. Fu, A. Cantor, M. Pappas, M. Daeges and L. Humphrey. 2016. Effectiveness of Breast Cancer Screening: Systematic Review and Meta-Analysis to Update the 2009 U.S. Preventive Services Task Force Recommendation. *Annals of Internal Medicine* 164: 244–55. doi:10.7326/M15-0969.

Neufeld, H., C. Richmond and Southwest Ontario Aboriginal Health Access Centre. 2017. Impacts of Place and Social Spaces on Traditional Food Systems in Southwestern Ontario. *International Journal of Indigenous Health* 12(1): 93–115. doi:10.18357/ijih112201716903.

Nishri, D., A. Sheppard, D. Withrow and L. Marrett. 2014. Cancer Survival among First Nations People of Ontario, Canada (1968–2007). *International Journal of Cancer* 136(3): 639–45. doi:10.1002/ijc.29024.

O'Brien, B., J. Mill and T. Wilson. 2009. Cervical Screening in Canadian First Nation Cree Women. *Journal of Transcultural Nursing* 20(1): 83–92. doi:10.1177/1043659608322418.

Ontario Institute for Cancer Research (OICR). 2015. *Ontario Institute for Cancer Research Strategic Plan 2016–2021*. Retrieved January 2016. <https://oicr.on.ca/wp-content/uploads/2016/11/StrategicPlan2016-2021.pdf>.

Ontario Human Rights Commission. 2016. Join Us – Training on new creed policy. Retrieved February, 2020. <http://www.ohrc.on.ca/en/join-us-training-new-creed-policy>.

Ontario Local Health Integration Networks (Ont LHINs). 2016. Provincial Aboriginal LHIN Report 2014/15. Retrieved March 2016. <http://www.lhins.on.ca/Pan-LHIN%20Content/Provincial%20Aboriginal%20LHIN%20Network.aspx>.

Pal, L. 2014. *Beyond Policy Analysis: Public Issue Management in Turbulent Times, (5th ed.)*. Toronto, ON: Nelson Education.

Patrick, R. 2011. Uneven Access to Safe Drinking Water for First Nations in Canada: Connecting Health and Place through Source Water Protection. *Health & Place* 17(1): 386–89. doi:10.1016/j.healthplace.2010.10.005.

Richmond, C. and C. Cook. 2016. Creating Conditions for Canadian Aboriginal Health Equity: The Promise of Healthy Public Policy. *Public Health Reviews* 37(2): 1–16. doi:10.1186/s40985-016-0016-5.

Sheppard, A., A. Chiarelli, L. Marrett, L. Mirea, D. Nishri and M. Trudeau. 2010. Detection of Later Stage Breast Cancer in First Nations Women in Ontario, Canada. *Canadian Journal of Public Health* 101(1): 101–05.

Smith, D., C. Varcoe and N. Edwards. 2005. Turning Around the Intergenerational Impacts of Residential Schools on Aboriginal People: Implication for Health Policy. *Canadian Journal of Nursing Research* 37(4): 38–60.

Snyder, M., K. Wilson and J. Whitford. 2015. Examining the Urban Aboriginal Policy Gap: Impacts on Service Delivery for Mobile Aboriginal Peoples in Winnipeg, Canada. *Aboriginal Policy Studies* 5(1): 3–27. doi:10.5663/aps.v5i1.23259.

Sullivan, T., L. Thompson and H. Angus. 2005. Transforming Cancer Service in Ontario: A Work in Progress. *Healthcare Papers* 5: 43–51. doi:10.12927/hcpap.17385.

The United Nations. 2007. *Declaration on the Rights of Indigenous People*. Accessed January 2016. <https://www.un.org/development/desa/indigenouspeoples/wp-content/uploads/sites/19/2018/11/UNDRIP_E_web.pdf>.

Tinmouth, J., E. Vella, N. Baxter, C. Dubé, M. Gould, A. Hey et al. 2016. Colorectal Cancer Screening in Average Risk Populations: Evidence Summary. *Canadian Journal of Gastroenterology and Hepatology* 2016: 2878149. doi: 10.1155/2016/2878149.

Tjepkema, M. 2002. The Health of the Off-Reserve Aboriginal Population. Supplement to Health Reports Volume 13. Ottawa, ON: Statistics Canada, Catalogue 82-003.

Tjepkema, M., R. Wilkins, S. Senécal, E. Guimond and C. Penney. 2009. Mortality of Métis and Registered Indian Adults in Canada: An 11-Year Follow-Up Study. *Health Reports* 20(4): 31–51.

Tobias, J., C. Richmond and I. Luginaah. 2013. Community-Based Participatory Research (CBPR) with Indigenous Communities: Producing Respectful and Reciprocal Research. *Journal of Empirical Research on Human Research Ethics* 8(2): 129–40. doi:10.1525/jer.2013.8.2.129.

Truth and Reconciliation Commission of Canada (TRC). 2015. "Truth and Reconciliation Commission of Canada: Calls to Action. Manitoba, Canada. Retrieved March 2018. <http://trc.ca/assets/pdf/Calls_to_Action_English2.pdf>.

Tungasuvingat Inuit and Cancer Care Ontario. 2017. *Cancer Risk Factors and Screening among Inuit in Ontario and Other Canadian Regions*. Toronto, ON. Retrieved May 2018. <https://www.cancercareontario.ca/sites/ccocancercare/files/assets/InuitRiskFactorsReport.pdf>.

Waldram, J.B., A. Herring and T.K. Young. 2006. *Aboriginal Health in Canada: Historical, Cultural, and Epidemiological Perspectives*. Toronto, ON: University of Toronto Press.

Warner, E., R. Heisey and J. Carroll. 2012. Applying the 2011 Canadian Guidelines for Breast Cancer Screening in Practice. *CMAJ* 184(16): 1803–07. doi:10.1503/cmaj.120392.

Williams J. H. and S. M. Carter. 2016, October 6. An empirical study of the 'underscreened' in organised cervical screening: experts focus on increasing opportunity as a way of reducing differences in screening rates. *BMC Medical Ethics* 17(56). doi:10.1186/s12910-016-0143-z.

Withrow, D., A. Amartey and L. Marrett. 2014. Cancer Risk Factors and Screening in the Off-Reserve First Nations, Métis and Non-Aboriginal Populations of Ontario. *Chronic Disease and Injuries in Canada* 34(2–3): 103–12.

Withrow, D., J. Pole, D. Nishri, M. Tjepkema and L. Marrett. 2017. Cancer Survival Disparities between First Nation and Non-Aboriginal Adults in Canada: Follow-Up of the 1991 Census Mortality Cohort. *Cancer Epidemiology, Biomarkers & Prevention* 26(1): 145–51. doi:10.1158/1055-9965.EPI-16-0706.

Wylie, L. and S. McConkey. 2018. Insiders' Insight: Discrimination against Indigenous Peoples through the Eyes of Health Care Professionals. *Journal of Racial and Ethnic Health Disparities* 6(1): 37–45. doi:10.1007/s40615-018-0495-9.

Yin, R.K. 2014. *Case Study Research: Design and Methods (5th Edition).* Thousand Oaks, CA: Sage Publications Inc.

Young, T.K. and J.W. Frank. 1983. Cancer Surveillance in a Remote Indian Population in Northwestern Ontario. American Journal of Public Health 73(5): 515–20. doi:10.2105/ajph.73.5.515.

RESEARCH PAPER

Gender Differences in Surgery for Work-Related Musculoskeletal Injury: A Population-Based Cohort Study

Différences entre les sexes dans la chirurgie pour les blessures musculosquelettiques reliées au travail : une étude de cohortes axée sur la population

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Abstract

Objective: The objective of this study is to examine if women are less likely than men to receive surgery following work-related musculoskeletal injury in the Canadian province of British Columbia.

Methods: The study included 2,403 workers with work-related knee meniscal tear, thoracic/lumbar disc displacement or rotator cuff tear. Probability of surgery was compared by gender using Kaplan–Meier methods and Cox proportional hazards models.

Results: For each injury type, a smaller proportion of women received surgery compared to men (knee: 76% vs. 80%; shoulder: 13% vs. 36%; back: 13% vs. 19%). In adjusted models, compared to men, women were 0.87 (95% confidence interval [CI] [0.69, 1.09]), 0.35 (95% CI [0.25, 0.48]) and 0.54 (95% CI [0.31, 0.95]) times less likely to receive knee, shoulder or back surgery, respectively.

Conclusions: Probability of surgery following work-related musculoskeletal injury was lower for women than for men. Strategies to ensure gender equitable delivery of surgical services by workers' compensation systems may be warranted, although further research is necessary to investigate determinants of the gender difference and the impact of elective orthopaedic surgery on occupational outcomes.

Résumé

Objectif : L'objectif de cette étude est de voir si les femmes sont moins susceptibles que les hommes de subir une chirurgie suite à une blessure musculosquelettique reliée au travail dans la province canadienne de la Colombie-Britannique.

Méthode : L'étude porte sur 2 403 travailleurs qui ont eu des blessures au travail, soit une déchirure du ménisque du genou, un déplacement de disque dorsal/lombaire ou une rupture de la coiffe des rotateurs. La probabilité de subir une chirurgie a été comparée entre les sexes au moyen de la méthode de Kaplan–Meier et du modèle des hasards proportionnels de Cox.

Résultats : Une plus petite proportion de femmes ont subi une chirurgie comparativement aux hommes, et ce, pour chaque type d'intervention (genou : 76 % c. 80 %; épaule : 13 % c. 36 %; dos : 13 % c. 19 %). Selon les modèles ajustés, les femmes avaient, comparativement aux hommes, 0,87 (95 % intervalle de confiance [IC] [0,69–1,09]), 0,35 (95 % IC [0,23–0,48]) et 0,54 (95 % IC [0,31–0,95]) moins de chance de subir une chirurgie pour le genou, l'épaule ou le dos, respectivement.

Conclusion : La probabilité de subir une chirurgie suite à une blessure musculosquelettique reliée au travail était moins élevée chez les femmes que chez les hommes. Des stratégies sont nécessaires pour assurer une prestation équitable des services chirurgicaux dans le cadre des régimes d'indemnisation des accidents du travail, mais il faut approfondir la recherche afin d'étudier les déterminants des différences entre les sexes ainsi que l'impact des chirurgies orthopédiques non urgentes sur les résultats professionnels.

Introduction

"Identifying gender inequalities and addressing gender equity are ... central to good stewardship of health systems" (Payne 2009: Executive Summary). Gender differences in healthcare delivery have been documented for a number of conditions including congestive heart failure (Sheppard et al. 2005), colon cancer (Donovan and Syngal 1998), acute myocardial infarction (Arora et al. 2019; Jneid et al. 2008), critical illness (Fowler et al. 2007), renal disease (Bloembergen et al. 1997; Jindal et al. 2005) and hip and knee arthritis (Hawker et al. 2000; Travis et al. 2012). For each of these aforementioned findings, men were more likely to receive indicated health services than women. Although, such gender differences are not universal across conditions, populations or time because these are affected by culture, gender roles and other ecological factors.

Some of the strongest findings for gender differences in healthcare delivery have been for elective arthroplastic surgeries. Almost 20 years ago, an observational study (in 2000) found that in Canada, both men and women underused knee and hip arthroplasty, but the underuse was greater for women (Hawker et al. 2000). Interestingly, although women were as willing to undergo surgery as men, women were less likely to have discussed the possibility with a physician. Following this, a 2010 Canadian study used standardized patients to experimentally control for surgical indications and isolate the effects of patient gender on odds of recommendation for total knee arthroplasty. Results found that compared to women, men had twice the odds of recommendation from a family physician and 22 times the odds of recommendation from an orthopaedic surgeon (Borkhoff et al. 2008). Such findings are not limited to Canada. A 1996 American study found that men's odds of total hip arthroplasty were 1.26 times that of women's after controlling for confounders (Giacomini 1996). Likewise, an English study of gender differences in healthcare delivery leading up to total hip arthroplasty found that, after controlling for age, disease severity and willingness and fitness for surgery, women were less likely than men to consult their general practitioner, receive referral to specialist care, consult an orthopaedic surgeon and be on a surgical wait-list (Jüni et al. 2010). Differences in diagnostic indicators, care-seeking behaviours and willingness to undergo surgery have been unable to fully explain the differences in men's and women's use of arthroplastic surgery, and evidence indicates that physicians are more likely to recommend these surgeries to men than to women (Borkhoff et al. 2008, 2013; Hawker et al. 2000; Jüni et al. 2010). Susceptibility of physician's decision-making to subconscious gender-based stereotypes is a suggested explanation for this observed gender disparity in surgical decision-making (Borkhoff et al. 2008).

Little is known about gender differences in elective surgery for musculoskeletal conditions other than osteoarthritis. Surgical decision-making for other musculoskeletal conditions may also be susceptible to gender bias because of the elective nature of these surgeries and the cognitively complex process of interpreting multiple surgical indications and contraindications, some of which may intersect with stereotypical understandings of

gender. Although gender differences in surgery for musculoskeletal conditions are of interest to health systems that serve general populations, these are also of special interest to health systems that serve working populations, such as workers' compensation systems where musculoskeletal conditions are a leading cause of disability and cost spending (Baldwin 2004; WorkSafeBC 2016).

In Canada, workers' compensation systems insure wage-loss benefits and health services for work-related injury and illness, whereas provincial public health systems insure physician and hospital services for non-work-related injury and illness. Policy and practice surrounding surgical healthcare delivery varies notably between these two parallel health systems (Hurley et al. 2008), warranting individual examination of both systems. For example, in the British Columbia workers' compensation system, case managers can opt to expedite orthopaedic surgery for injured workers by sending them to a private surgical facility (for an added cost), in lieu of a public facility where wait-lists are longer. Such practices may introduce additional mechanisms for gender bias not existent in provincial public health systems. Organizational mandates also vary between these two types of health system. Timely return to work is a more central focus for workers' compensation systems than for provincial public health systems. Return-to-work goals may have a greater influence on surgical decision-making when the condition is work-related and insured by workers' compensation, and this may affect men and women differently.

The objective of the current study was to examine gender differences in surgery among workers in British Columbia with accepted workers' compensation claims for musculoskeletal injury. The study focused on three common types of work-related musculoskeletal injury where surgery is a potential treatment option: knee meniscus tear, rotator cuff tear and thoracic/lumbar intervertebral disc displacement. To our knowledge, gender differences in surgery for these musculoskeletal conditions have not yet been examined in either general or working populations, and gender differences in healthcare delivery for work-related injury or illness have received relatively little attention in the occupational health literature. Based on findings from knee and hip osteoarthritis patients in the general population, we hypothesized that for each type of work-related musculoskeletal injury investigated, women would have lower probability of surgical treatment than men with the same injury, after adjusting for age, physical job demands and clinical characteristics.

Methods

This retrospective population-based cohort study was designed in accordance with *The Strengthening the Reporting of Observational Studies in Epidemiology* statement for reporting observational studies (von Elm et al. 2008).

Data sources

Individual-level, linked administrative data sets from WorkSafeBC (Workers' Compensation Board of British Columbia) and Ministry of Health were used (British Columbia Ministry

of Health 2012a, b; WorkSafeBC 2012). During the study period, WorkSafeBC provided workers' compensation coverage for 94% of the provincial workforce (Association of Workers' Compensation Boards of Canada 2016), and all residents of British Columbia were required to be registered for public health insurance via the Ministry of Health.

Study sample

The study sample included workers with an accepted workers' compensation lost-time claim (at least one full day off work) for one of three common musculoskeletal injuries where surgical treatment could be a recommended treatment option: knee meniscus tear, rotator cuff tear and thoracic/lumbar intervertebral disc displacement with an injury date between May 1, 2009, and December 31, 2010. The study period was chosen based on the availability of the data. In the event that an individual had multiple claims meeting the eligibility criteria, only the first claim from the study period was retained for analysis.

Injury type was determined using four-digit *International Classification of Diseases, Ninth Revision (ICD-9)* codes recorded in the claims data set (836.2, 836.0 and 836.1 for knee meniscus tear; 840.4, 840.5 and 840.6 for rotator cuff tear; and 722.0 and 722.1 for thoracic/lumbar intervertebral disc displacement; Government of British Columbia 2017). The study sample was restricted to injured workers between the ages of 19 to 64 years who were registered for provincial healthcare during the year of injury, which was necessary to ensure 365 days of follow-up for surgical outcomes.

Study variables

Gender was the primary explanatory variable for all analyses and was ascertained from the self-reported "sex" field in the Ministry of Health data set. According to the Canadian Institutes of Health Research (2019), gender refers to socially constructed roles, behaviours, expressions and identities, whereas sex refers to biologically determined features including chromosomes, gene expression, hormone levels and function and reproductive/sexual anatomy. Because the "sex" field from the administrative data was self-reported, participants likely completed this field based on their gender identity (to the best of their ability with the options that were provided, i.e., male or female).

Although administrative records often lack measures of gender, disaggregation of the data based on sex can allow for analysis of gender issues (UNECE 2010), and this is the approach used here. Throughout this study, the term "gender" is used rather than "sex" because previous research indicates that differences in elective orthopaedic surgery between men and women are because of the effects of socially constructed gender roles on physician's surgical decision-making, rather than biological differences between men and women. Further, because sex is not a surgical indication for the conditions examined here, there is no reason to hypothesize that sex-based characteristics explain potential differences in the rates of surgery between men and women.

Musculoskeletal surgery (yes/no and time to surgery in days) within 365 calendar days of the workers' compensation injury date was the primary outcome variable. Surgeries were identified using the Ministry of Health and workers' compensation data sets, and all major surgeries involving arthroscopic or arthrotomic access to the joint of interest were included. Surgical wait time was calculated as the number of calendar days from injury date to surgery date, censored at 365 days. A time frame of 365 calendar days was used to increase the probability that surgical outcomes were directly related to the injury from the included compensation claim. All holidays, weekends and business days were included in the calendar days measure. Individuals who received surgery on the same day as their injury were excluded from the study sample because such injuries represent probable traumatic injuries that require little discretionary decision-making about surgical treatment that might be influenced by gender.

Potential confounders included injury type (knee meniscus tear, rotator cuff tear or thoracic/lumbar intervertebral disc displacement), age at the time of injury, prior workers' compensation claim, occupational strength requirements and the primary body position associated with the occupation. For the knee meniscus injury sample, an additional variable for co-morbidity was included for knee ligament strain at the time of injury or in the 12 months following injury. This variable was defined as no knee strain diagnoses or one or more diagnoses from a non-musculoskeletal-related physician or one or more diagnoses from a musculoskeletal-related physician. Orthopaedic surgeons, physical medicine and rehabilitation physicians and radiologists were considered musculoskeletal-related physicians. Individuals with diagnoses for knee ligament strain from a non-musculoskeletal-related physician and from a musculoskeletal-related physician were categorized as having a diagnosis from the latter. Similar variables were not included for the rotator cuff and intervertebral disc injury samples because the incidence of co-morbid musculoskeletal diagnoses for these injuries was low.

For the prior workers' compensation claim variable, individuals were classified based on their history in the past five years: (1) no prior accepted workers' compensation claim, (2) one or more prior accepted workers' compensation claims with no direct pathophysiological relation to the injury of interest and (3) one or more prior workers' compensation claims with at least one of these claims having a direct pathophysiological relation to the injury of interest. Prior claims with diagnoses indicating internal injury to the same joint as the joint of interest were considered to have a direct pathophysiological relation, but by definition, each claim represents an independent injury or incident.

Occupational strength requirements were classified as limited (handling loads up to 5 kg), light (handling loads of 5 kg but <10 kg), moderate (handling loads between 10 kg and 20 kg) or heavy (handling loads of more than 20 kg). The primary body position associated with the occupation was classified as sitting or other. Values for these two variables were obtained from the *2003 Career Handbook*, published by Human Resources Development

Canada (2003), which contains ratings for key occupational characteristics assigned by trained occupational analysts using a Delphi procedure. For the current study, ratings from the *Handbook* were assigned to workers based on the occupation code associated with the claim.

Analysis

All analyses were conducted using SAS 9.4 (SAS 2017). Ethical approval for the research project was obtained from the Behavioural Research Ethics Board at the University of British Columbia (H12-02239). All analyses were stratified by injury type.

The cumulative probability of surgery by wait time was compared for men and women using Kaplan–Meier methods and the log-rank test. The probability of women undergoing surgery relative to men on any day following injury was estimated using unadjusted and adjusted Cox regression models (Cox 1972). In addition to gender, adjusted models included the potential confounders of age, prior claim, occupational strength demands, primary body position associated with the occupation and, in the case of the knee sample, co-morbid diagnosis of knee ligament strain. The proportional hazards assumption was assessed graphically (Kaplan–Meier and log-minus-log plots) and by examining interactions with the log of the wait time variable. Only interaction terms for time-varying variables, significant at a 95% level of confidence, were retained in the final adjusted models.

Finally, sensitivity analysis was conducted to examine the proportion of surgeries in the study sample covered by workers' compensation insurance as well as the effects of restricting the outcome to only surgeries covered by workers' compensation insurance. For the latter, follow-up time was censored on the day of the first surgery if the surgery was covered by the public health system rather than workers' compensation insurance.

Results

Study sample

A total of 2,545 unique accepted lost-time claims for knee meniscal tears (n = 687), rotator cuff tears (n = 1,105) and thoracic/lumbar intervertebral disc displacements (n = 753) were identified in the workers' compensation data set, with injury dates occurring between May 1, 2009, and December 31, 2010. Of the 2,545 claims, 142 (5.6%) were removed because of one or more of the following: age of <19 or >64 years, not registered for provincial healthcare, missing data for gender or age, receipt of surgical treatment on the same day as the injury or the claim was not the first musculoskeletal injury for a worker during the follow-up period. The final study sample consisted of 2,403 included claims (knee meniscal tear = 640; rotator cuff tear = 1,042; intervertebral disc displacement = 721).

Knee meniscus injury sample

The meniscus injury sample was approximately one fifth of women (20.9%), with a similar mean age for women (47 years) and men (45 years). Approximately 13% of both women and men had a related prior claim, although men were more likely than women to have had an unrelated prior claim (49.4% vs. 39.6%). Men and women had different occupational strength demands (Table 1, available online at longwoods.com/content/26131). In particular, compared with women (10.5%), men were more likely to work in occupations with heavy strength demands (27.2%). Approximately 70% of both men and women had received a diagnosis of co-morbid knee ligament strain from either a non-surgical or a surgical specialist.

Rotator cuff injury sample

The rotator cuff injury sample consisted of approximately one third of women (34.4%), and the mean age was the same for women and men (46 years; Table 1). Women were more likely than men to have no prior claim (40.5% vs. 34.9%), whereas men were more likely than women to have an unrelated prior claim (55.3% vs. 46.9%). Men and women had different strength demands associated with their occupation at the time of injury, with a higher proportion of men having heavy occupational strength demands (25.6%) compared to women (5.3%).

Intervertebral disc displacement injury sample

The intervertebral disc displacement injury sample was just over one-quarter women (27.0%), with a similar mean age for women (43 years) and men (41 years; Table 1). Women were more likely than men to have no prior claim (49.7% vs. 35.4%), whereas men were more likely than women to have an unrelated prior claim (41.6% vs. 31.3%). Again, compared to women (7.6%), men were more likely to work in an occupation with heavy occupational strength demands (25.5%).

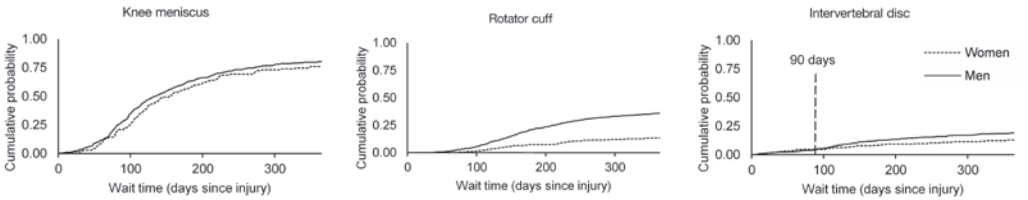
Overall, across the three injury samples, the distributions of the injured workers' age, prior claim status, occupational strength demands and occupational body position by gender were very similar.

Gender and surgery

Musculoskeletal surgery was most common in the meniscus sample with 509 workers (79.1%) with this injury receiving surgery within 12 months. A smaller proportion of workers from the rotator cuff (n = 294, 28.2%) and intervertebral disc (n = 125, 17.3%) samples went on to undergo surgery. Descriptively, by 365 days, the cumulative probability of surgery was higher for men than for women for all samples (Figure 1). Gender showed evidence of non-proportionality for the intervertebral disc sample. For this sample, gender had no effect on the probability of surgery within the first 90 days following injury (χ^2 = 0.00, p = 0.9697),

after which men were more likely to receive surgery than women. Relatively few surgeries were observed in the intervertebral disc sample prior to 91 days (4.6% of women [n = 9] and 4.6% of men [n = 24]). To account for this in the current study, analyses for the intervertebral disc sample were stratified by time and only results from the 91-to-365-day strata are reported for the Kaplan–Meier and Cox regression analyses. In the Kaplan–Meier analyses, the unadjusted effect of gender on wait time was significant for the rotator cuff (χ^2 = 57.05, p < 0.0001) and intervertebral disc (91 to 365 days; χ^2 = 5.02, p = 0.0250) samples but not the meniscus sample (χ^2 =1.86, p = 0.1729).

FIGURE 1. Cumulative probability of surgery by wait time and injury type for women and men with accepted workers' compensation claims



For the study confounders in unadjusted Cox regression models, age showed evidence of non-proportionality with surgery in the meniscus sample but not the intervertebral disc and rotator cuff samples. To account for this, an interaction term between age and the log of the wait time variable was included in the adjusted Cox regression model for the meniscus sample. Using men as the reference group, the unadjusted effect of gender in the Cox regression models was significant for the intervertebral disc (hazard ratio [HR] = 0.55, 95% confidence interval [CI] [0.32, 0.93]) and rotator cuff (HR = 0.32, 95% CI [0.24, 0.44]) samples but not the meniscus sample (HR = 0.86, 95% CI [0.69, 1.07]). Similar results were found for the full models adjusted for age, prior claim, strength demands, body position and knee ligament strain (knee sample only). The HRs for gender in the adjusted models for the intervertebral disc, rotator cuff and meniscus samples were 0.54 (95% CI [0.31, 0.95]), 0.35 (95% CI [0.25, 0.48]) and 0.87 (95% CI [0.69, 1.09]), respectively (Table 2).

In the sensitivity analysis, 95.3% of knee (485 of 509), 94.2% of shoulder (277 of 294) and 79.2% of back (99 of 125) surgeries were covered by workers' compensation insurance. When the surgical outcome was restricted to surgeries covered by workers' compensation insurance, the HRs and 95% CIs for the gender variable in the adjusted Cox regression models were similar to those found in the primary analysis (intervertebral disc – HR = 0.53, 95% CI = [0.28, 0.99]; rotator cuff – HR = 0.34, 95% CI [0.24, 0.47]; knee meniscus – HR = 0.83, 95% CI [0.66, 1.04]).

TABLE 2. Adjusted hazard ratios with 95% confidence intervals for surgery for workers with accepted workers' compensation claims stratified by injury type.

	Adjusted hazard ratio (95% confidence interval)		
	Knee meniscus tear	**Rotator cuff tear**	**Intervertebral disc[a]**
Gender			
Men	1	1	1
Women	0.87 [0.69, 1.09]	0.35 [0.25, 0.48]	0.54 [0.31, 0.95]
Age	0.85 [0.81, 0.91]	1.04 [1.03, 1.06]	0.98 [0.96, 1.00]
Age × log (wait time)	1.04 [1.02, 1.05]	Not significant[b]	Not significant[b]
Prior claim			
None	1	1	1
Other body part	0.91 [0.76, 1.09]	0.98 [0.78, 1.25]	0.85 [0.56, 1.29]
Same body part	1.46 [1.13, 1.89]	0.95 [0.64, 1.40]	0.86 [0.52, 1.43]
Strength demands			
Limited	1	1	1
Light	1.16 [0.85, 1.58]	0.83 [0.53, 1.27]	0.75 [0.36, 1.57]
Medium	1.09 [0.85, 1.40]	0.77 [0.55, 1.09]	0.92 [0.51, 1.66]
Heavy	1.13 [0.84, 1.51]	1.33 [0.89, 1.99]	0.82 [0.40, 1.68]
Body position			
Sitting	1	1	1
Other	0.77 [0.60, 0.99]	0.89 [0.65, 1.22]	1.01 [0.56, 1.84]
Knee strain			
None	1	Not applicable	Not applicable
Diag phys[c]	1.20 [0.96, 1.49]		
Diag msk phys[d]	1.33 [1.08, 1.64]		

a Time stratified analysis with only the 91-to-365-day strata reported here

b Interaction with the log of the wait time variable was examined for all study covariates. Only interaction terms significant at a 95% level of confidence were included in the adjusted analyses.

c Diagnosis for co-morbid knee ligament strain from a physician

d Diagnosis for co-morbid knee ligament strain from a musculoskeletal-related physician

Discussion

This study examined the relationship between gender and surgery among workers with accepted lost-time claims for musculoskeletal injury in the Canadian province of British Columbia. We found that after adjustment for confounders, women's probability of surgery for work-related knee meniscus tears, rotator cuff tears and intervertebral disc displacements was lower (0.87, 0.35 and 0.54 times, respectively) than men's. The findings are in agreement with those reported in literature from general populations, that is, women are less likely than men to receive hip or knee replacement surgery (Borkhoff et al. 2008; Giacomini 1996; Hawker et al. 2000; Jüni et al. 2010). The current study expands the knowledge base on gender differences in elective orthopaedic surgery to a workers' compensation population and to three types of common work-related musculoskeletal injury not previously reported on. Furthermore, after restricting the outcome to surgeries paid for by the workers' compensation system, women's probability of surgery was lower than men's probability. This demonstrates that the gender bias found in this study is not because of surgeries paid for solely by the public health system.

In our study, gender differences in surgery were more pronounced when surgery was not

the standard course of treatment, as was the case with work-related thoracic/lumbar disc displacements and rotator cuff tears. Respectively, less than 20% and 30% of workers with these injuries went on to undergo surgery. Conversely, nearly 80% of workers with knee meniscal tears went on to undergo surgery as a standard course of treatment. Women with knee meniscal tears were still less likely to undergo surgery than men, although the effect size was smaller and not statistically significant. Evidence shows that stereotypes have a larger impact on healthcare providers' clinical decision-making when there is an element of uncertainty or when the care provider experiences a high cognitive load (Burgess et al. 2004; Burgess 2010; Chinburapa et al. 1993; Hall 2002). Given this, we hypothesize that when surgery is a standard course of treatment, as was the case for knee meniscal tears in the study sample, there is less perceived uncertainty regarding indication for surgery, and clinical decision-making is less susceptible to bias by patient or worker gender.

The evidence base regarding the efficacy of the surgeries examined here is evolving. Recent systematic reviews have concluded unclear or no long-term benefits for arthroscopic knee meniscectomy and/or debridement, lumbar disc discectomy and open or arthroscopic subacromial decompression compared with non-surgical treatments (Coghlan et al. 2008; Gibson and Waddell 2007; Khan et al. 2014; Thorlund et al. 2015). As these findings are taken up into practice, surgeries that are currently the standard course of treatment, such as knee meniscectomy, may become less common. Should this be the case, physicians' tendency to refer men for surgery more often than women, especially in uncertain situations that involve higher degrees of discretionary decision-making, must be considered. A more conservative rate of elective surgery for musculoskeletal injury, as is currently the case for women compared to men, may be a preferred outcome.

Non-proportionality of the gender variable was observed in the intervertebral disc injury sample. No effect of gender was observed in the first 90 days following injury; however, from 91 to 365 days, men were more likely to receive surgery than women. Use of surgery during the first 90 days following intervertebral disc injury was low, and this likely reflects conservative use of surgery for this injury during this acute phase of injury when the prospect of return to work without surgical intervention is high (Frank et al. 1998). This finding is in line with clinical guidelines that recommend surgical discectomy only when pain is persistent and does not resolve with less invasive treatment (Chou et al. 2007).

Perception that return to work is of greater importance to men may lead physicians to refer men with work-related injury for surgery more often than women or to prioritize men's position on the wait-list. In Western cultures, men's status on the masculine hierarchy is strongly determined by their ability to produce income and/or the physicality of their work (Evans et al. 2011; Lee and Owens 2002). Inability to work due to health reasons, or inability to engage in physical labour, has a significant impact on masculine identities, more so than feminine ones (Evans et al. 2011; Lee and Owens 2002; Ostrander 2008). Further, based on recent Canadian estimates, men are over twice as likely as women to be the higher income recipient in an adult couple (Statistics Canada 2017). This may also contribute to the

perception that return to work is of greater importance for men, as in married or co-habiting heterosexual couples, as the household may be more reliant on men's income. For these reasons, physicians may be more likely to discuss surgery with men than with women or to refer men for surgery more often than women, because of disability–masculinity role conflict in the context of work disability and return-to-work outcomes and men's role as the higher income recipient within a household. These hypotheses are supported by the finding that in the general Canadian population, men have been shown to be more likely than women to have discussed the possibility of joint replacement surgery with a physician (Hawker et al. 2000). Such gendered perceptions could also affect decision-making in the workers' compensation system related to wait time for surgery where additional decisions are made about expediting surgery to improve return to work (Hurley et al. 2008). In the current study, men with musculoskeletal injuries were more likely to be in occupations with heavy physical demands than women. The perception that surgery is needed to be able to return to work more quickly for these types of occupations, particularly for men, may have added to the gender differences reported here.

There are some limitations to the current study. First, to control for injury severity, inclusion was limited to a small set of *International Classification of Diseases, Ninth Revision* diagnoses. Furthermore, the study sample was limited to lost-time claims, ensuring that all injuries were severe enough to warrant time off work, and injuries severe enough to result in same-day surgery were excluded. Despite this, the possibility of some remaining heterogeneity of injury severity cannot be entirely dismissed. This could cause differential bias if injury severity were associated with gender. However, there is little evidence to support such an association. In the meniscus injury sample, a measure of co-morbid knee ligament strain was developed to control for injury severity in the form of damage to multiple structures of the knee. This variable was related to surgery but not gender, demonstrating that in this instance, injury severity did not confound the relationship between gender and surgery. In addition to this, the models were adjusted for occupational strength demands to account for the possibility that workers with heavy occupational strength demands might have sustained more serious injuries, and the models were also adjusted for prior claims to control for potential differences in pathophysiological history of the injury as well as potential co-morbidities. Second, although the potential confounders of occupational strength demands (classified as limited, light, moderate or heavy based on the strength involved in the handling of objects) and the primary body position associated with the job (sitting vs. other) were included in the adjusted models, there was no available measure to capture the frequency of motion. Third, we were not able to control for workers' willingness to undergo surgery, and this may have contributed to the findings. However, a study on men's and women's use of elective hip or knee arthroplasty in the general Canadian population found that despite women's lower use of surgery, men and women were equally willing to undergo surgery (Hawker et al. 2000).

The authors concluded that women's lower rate of surgery was most likely due to the barriers that exist at the level of the doctor–patient interaction. Based on this, we hypothesize that a similar phenomenon is occurring here in that the results are not predominantly driven by a difference in men's and women's willingness to undergo surgery but rather a gender difference in the number and type of barriers encountered in the pathway of care. Fourth, the data did not allow us to examine surgery for non-work-related injuries, and thus, we are not able to isolate if the observed gender differences are unique to work-related injuries in a workers' compensation population or if these are a reflection of gender differences present in the general population. Finally, our findings may not be generalizable to worker populations where the policy and practice surrounding treatment of work-related musculoskeletal injury diverges from that used in British Columbia.

In conclusion, this study demonstrated a higher probability of surgery for men with work-related musculoskeletal injury compared to similar women with the same injury. Additional studies are needed to better understand if the results presented here represent a gender inequity in terms of underuse of surgery and/or extended surgical wait times for women versus overuse and/or expedited wait times for men, as well as the underlying mechanisms. As mentioned earlier, physicians' tendency to under-refer women for surgery may be an unintended beneficial outcome for women, especially in the context of newly emerging evidence to suggest non-efficacy. Further research is also needed to better understand how surgery following work-related musculoskeletal injury affects disability and occupational outcomes for men and women. With a better understanding of these issues, interventions to ensure gender-equitable treatment of work-related musculoskeletal injury can be developed. Potential interventions to address gender differences in surgery for work-related musculoskeletal injury that occur due to biased decision-making include training and awareness programs as well as the implementation and enforcement of standardized guidelines and protocols. Although we are not aware of existing interventions specific to gender differences in surgery for work-related musculoskeletal injury, or musculoskeletal injury in general, similar interventions have reduced disparities, improved patient outcomes and decreased physicians' likelihood of making a biased decision in other areas of healthcare delivery (Hannah and Carpenter-Song 2013; Herbert et al. 2010; Mosca et al. 2011; Stone and Moskowitz 2011; Teal et al. 2012; Wang et al. 2011). Ultimately, research of this kind will help workers' compensation systems determine the best use of surgical services to manage work-related musculoskeletal injury in a gender-equitable manner and reduce disability and improve return to work for men and women.

Disclaimer

All inferences, opinions and conclusions drawn in this article are those of the authors, and do not reflect the opinions or policies of the data stewards.

Conflict of interest

The authors have no conflict of interest to disclose.

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References

Arora, S., G.A. Stouffer, A.M. Kucharska-Newton, A. Qamar, M. Vaduganathan, A. Pandey et al. 2019. Twenty Year Trends and Sex Differences in Young Adults Hospitalized with Acute Myocardial Infarction: The ARIC Community Surveillance Study. *Circulation* 139(8): 1047–56. doi:10.1161/CIRCULATIONAHA.118.037137.

Association of Workers' Compensation Boards of Canada (AWCBC). 2016. *Detailed Key Statistical Measures Report*. Retrieved November 5, 2017. <http://awcbc.org/?page_id=9759>.

Baldwin, M.L. 2004. Reducing the Costs of Work-Related Musculoskeletal Disorders: Targeting Strategies to Chronic Disability Cases. *Journal Electromyography and Kinesiology* 14(1): 33–41. doi:10.1016/j.jelekin.2003.09.013.

Bloembergen, W.E., E.A. Mauger, R.A. Wolfe and F.K. Port. 1997. Association of Gender and Access to Cadaveric Renal Transplantation. *American Journal of Kidney Diseases* 30(6): 733–38. doi:10.1016/S0272-6386(97)90076-7.

Borkhoff, C.M., G.A. Hawker, H.J. Kreder, R.H. Glazier, N.N. Mahomed and J.G. Wright. 2008. The Effect of Patients' Sex on Physicians' Recommendations for Total Knee Arthroplasty. *CMAJ* 178(6): 681–87. doi:10.1503/cmaj.071168.

Borkhoff, C.M., G.A. Hawker, H.J. Kreder, R.H. Glazier, N.N. Mahomed and J.G. Wright. 2013. Influence of Patients' Gender on Informed Decision Making Regarding Total Knee Arthroplasty. *Arthritis Care and Research* 65(8): 1281–90. doi:10.1002/acr.21970.

British Columbia Ministry of Health. 2012a. *Medical Services Plan (MSP) Payment Information File*. Vancouver, BC: Population Data BC.

British Columbia Ministry of Health. 2012b. *Consolidation File (MSP Registration & Premium Billing*. Vancouver, BC: Population Data BC.

Burgess, D.J., S.S. Fu and M. van Ryn. 2004. Why Do Providers Contribute to Disparities and What Can Be Done About It? *Journal of General Internal Medicine* 19(11): 1154–59. doi:10.1111/j.1525-1497.2004.30227.x.

Burgess, D.J. 2010. Are Providers More Likely to Contribute to Healthcare Disparities Under High Levels of Cognitive Load? How Features of the Healthcare Setting May Lead to Biases in Medical Decision Making. *Medical Decision Making* 30(2): 246–57. doi:10.1177/0272989X09341751.

Canadian Institutes of Health Research (CIHR). 2019. *How to Integrate Sex and Gender into Research*. Retrieved April 29, 2019. <http://www.cihr-irsc.gc.ca/e/50836.html>.

Chinburapa, V., L.N. Larson, M. Brucks, M. Draugalis, J.L. Bootman and C.P. Puto. 1993. Physician Prescribing Decisions: The Effects of Situational Involvement and Task Complexity on Information Acquisition and Decision Making. *Social Science and Medicine* 36(11): 1473–82. doi:10.1016/0277-9536(93)90389-L.

Chou, R., A. Qaseem, V. Snow, D. Casey, J.T. Cross, P. Shekelle et al. 2007. Clinical Guidelines Diagnosis and Treatment of Low Back Pain: A Joint Clinical Practice Guideline from the American College of Physicians and the American Pain Society. *Annals of Internal Medicine* 147(7): 478–91. doi:10.7326/0003-4819-147-7-200710020-00006.

Coghlan, J.A., R. Buchbinder, S. Green, R.V. Johnston and S.N. Bell. 2008. Surgery for Rotator Cuff Disease. *Cochrane Database Systematic Review* (1): CDOO5619. doi:10.1002/14651858.CD005619.pub2.

Cox, D.R. 1972. Regression Models and Life-Tables. *Journal of the Royal Statistical Society Series B (Methodological)* 34(2): 187–220. Retrieved February 4, 2020.

Donovan, J.M. and S. Syngal. 1998. Colorectal Cancer in Women: An Underappreciated but Preventable Risk. *Journal of Women's Health* 7(1): 45–48. doi:10.1089/jwh.1998.7.45.

Evans, J., B. Frank, J.L. Oliffe and D. Gregory. 2011. Health, Illness, Men and Masculinities (HIMM): A Theoretical Framework for Understanding Men and Their Health. *Journal of Men's Health* 8(1): 7–15. doi:10.1016/j.jomh.2010.09.227.

Fowler, R.A., N. Sabur, P. Li, D.N. Juurlink, R. Pinto, M.A. Hladunewich et al. 2007. Sex- and Age-Based Differences in the Delivery and Outcomes of Critical Care. *CMAJ* 177(12): 1513–19. doi:10.1503/cmaj.071112.

Frank, J., S. Sinclair, S. Hogg-Johnson, H. Shannon, C. Bombardier, D. Beaton et al. 1998. Preventing Disability from Work-Related Low-Back Pain. New Evidence Gives New Hope – If We Can Just Get All the Players Onside. *CMAJ* 158(12): 1625–31.

Giacomini, M.K. 1996. Gender and Ethnic Differences in Hospital-Based Procedure Utilization in California. *Archives of Internal Medicine* 156(11): 1217–24. doi:10.1001/archinte.1996.00440100115013.

Gibson, J.N. and G. Waddell. 2007. Surgical Interventions for Lumbar Disc Prolapse: Updated Cochrane Review. *Spine* 32(16): 1735–47. doi:10.1097/BRS.0b013e3180bc2431.

Government of British Columbia. 2017. *Diagnostic Code Descriptions (ICD-9)*. Retrieved November 5, 2017. <http://www2.gov.bc.ca/gov/content/health/practitioner-professional-resources/msp/physicians/diagnostic-code-descriptions-icd-9>.

Hall, K.H. 2002. Reviewing Intuitive Decision-Making and Uncertainty: The Implications for Medical Education. *Medical Education* 36(3): 216–24. doi:10.1046/j.1365-2923.2002.01140.x.

Hannah, S.D. and E. Carpenter-Song. 2013. Patrolling Your Blind Spots: Introspection and Public Catharsis in a Medical School Faculty Development Course to Reduce Unconscious Bias in Medicine. *Culture, Medicine, and Psychiatry* 37(2): 314–39. doi:10.1007/s11013-013-9320-4.

Hawker, G.A., J.G. Wright, P.C. Coyte, I. Williams, B. Harvey, R. Glazier et al. 2000. Differences between Men and Women in the Rate of Use of Hip and Knee Arthroplasty. *New England Journal of Medicine* 342(14): 1016–22. doi:10.1056/NEJM200004063421405.

Herbert, K.H., B. Lopez, R. Horswell, L. Tamariz, A. Palacio, H. Li et al. 2010. The Impact of a Standardized Disease Management Program on Race/Ethnicity and Gender Disparities in Care and Mortality. *Journal of Health Care for the Poor and Underserved* 21(1): 264–76. doi:10.1353/hpu.0.0243.

Human Resources Development Canada. 2003. *Career Handbook (Revised) (2nd edn)*. Ottawa, ON: Government of Canada.

Hurley, J., D. Pasic, J.N. Lavis, A.J. Culyer, C. Mustard and W. Gnam. 2008. Parallel Payers and Preferred Access: How Canada's Workers' Compensation Boards Expedite Care for Injured and Ill Workers. *Healthcare Papers* 8(3): 6–14. doi:10.12927/hcpap.2008.19792.

Jindal, R.M., J.J. Ryan, I. Sajjad and M.H. Murthy. 2005. Kidney Transplantation and Gender Disparity. *American Journal of Nephrology* 25(5): 474–83. doi:10.1159/000087920.

Jneid, H., G.C. Fonarow, C.P. Cannon, A.F. Hernandez, I.F. Palacios, A.O. Maree et al. 2008. Sex Differences in Medical Care and Early Death after Acute Myocardial Infarction. *Circulation* 118: 2803–10. doi:10.1161/CIRCULATIONAHA.108.789800.

Jüni, P., N. Low, S. Reichenbach, P.M. Villiger, S. Williams and P.A. Dieppe. 2010. Gender Inequity in the Provision of Care for Hip Disease: Population-Based Cross-Sectional Study. *Osteoarthritis and Cartilage* 18(5): 640–45. doi:10.1016/j.joca.2009.12.010.

Khan, M., N. Evaniew, A. Bedi, O.R. Ayeni and M. Bhandari. 2014. Arthroscopic Surgery for Degenerative Tears of the Meniscus: A Systematic Review and Meta-Analysis. *CMAJ* 186(14): 1057–64. doi:10.1503/cmaj.140433.

Lee, C. and R.G. Owens. 2002. Issues for a Psychology of Men's Health. *Journal of Health Psychology* 7(3): 209–17. doi:10.1177/1359105302007003215.

Mosca, L., E.J. Benjamin, K. Berra, J.L. Bezanson, R.J. Dolor, D.M. Lloyd-Jones et al. 2011. Effectiveness-Based Guidelines for the Prevention of Cardiovascular Disease in Women–2011 Update: A Guideline from the American Heart Association. *Circulation* 123(11): 1243–62. doi:10.1161/CIR.0b013e31820faaf8.

Ostrander, R.N. 2008. When Identities Collide: Masculinity, Disability and Race. *Disability and Society* 23(6): 585–97. doi:10.1080/09687590802328451.

Payne, S. 2009. *Policy Brief 12. How Can Gender Equity be Addressed through Health Systems?* Copenhagen, Denmark: World Health Organization Regional Office for Europe. Retrieved February 4, 2020. <http://www.euro.who.int/__data/assets/pdf_file/0006/64941/E92846.pdf>.

SAS. 2017. Version 9.4. Cary, NC: SAS Institute.

Sheppard, R., H. Behlouli, H. Richard and L. Pilote. 2005. Effect of Gender on Treatment, Resource Utilization, and Outcomes in Congestive Heart Failure in Quebec, Canada. *The American Journal of Cardiology* 95(8): 955–59. doi:10.1016/j.amjcard.2004.12.033.

Statistics Canada. 2017. *Household Income in Canada: Key Results from the 2016 Census*. Retrieved May 6, 2019. <https://www150.statcan.gc.ca/n1/daily-quotidien/170913/dq170913a-eng.htm>.

Stone, J. and G.B. Moskowitz. 2011. Non-Conscious Bias in Medical Decision Making: What Can be Done to Reduce It? *Medical Education* 45(8): 768–76. doi:10.1111/j.1365-2923.2011.04026.x.

Teal, C.R., A.C. Gill, A.R. Green and S. Crandall. 2012. Helping Medical Learners Recognize and Manage Unconscious Bias toward Certain Patient Groups. *Medical Education* 46(1): 80–88. doi:10.1111/j.1365-2923.2011.04101.x.

Thorlund, J.B., C.B. Juhl, E.M. Roos and L.S. Lohmander. 2015. Arthroscopic Surgery for Degenerative Knee: Systematic Review and Meta-Analysis of Benefits and Harms. *BMJ* 350: h2747: 1–9. doi:10.1136/bmj.h2747.

Travis, C.B., D.M. Howerton and D.M. Szymanski. 2012. Risk, Uncertainty, and Gender Stereotypes in Healthcare Decisions. *Women and Therapy* 35(3–4): 207–20. doi:10.1080/02703149.2012.684589.

United Nations Economic Commission for Europe (UNECE). 2010. *Developing Gender Statistics: A Practical Tool: Reference Manual Prepared by the UNECE Task Force on Gender Statistics Training for Statisticians with Contributions from Various Experts*. Geneva, Switzerland: United Nations. Retrieved March 9, 2019. <http://www.unece.org/fileadmin/DAM/stats/publications/Developing_Gender_Statistics.pdf>.

von Elm E., D.G. Altman, M. Egger, S.J. Pocock, P.C. Gøtzsche, J.P. Vandenbroucke et al. 2008. The Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) Statement: Guidelines for Reporting Observational Studies. *Journal of Clinical Epidemiology* 61(4): 344–49. doi:10.1016/j.jclinepi.2007.11.008.

Wang, T.Y., D. Dai, A.F. Hernandex, D.L. Bhatt, P.A. Heidenreich, G.C. Fonarow et al. 2011. The Importance of Consistent, High Quality Acute Myocardial Infarction and Heart Failure Care: Results from the American Heart Association's Get with The Guidelines Program. *Journal of the American College of Cardiology* 58(6): 637–44. doi:10.1016/j.jacc.2011.05.012.

WorkSafeBC. 2012. *Claims and Firm Level Files*. Vancouver, BC: Population Data BC.

WorkSafeBC. 2016. *2015 Statistics*. Retrieved February 5, 2017. <https://www.worksafebc.com/en/resources/about-us/annual-report-statistics/2015-stats?lang=en>.

RESEARCH PAPER

The Mosaic of Primary Care Nurses in Rural and Remote Canada: Results from a National Survey

Mosaïque de la main-d'œuvre infirmière en soins primaires dans les régions canadiennes rurales et éloignées : résultats d'une enquête nationale

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Abstract

Background and objective: Nurses provide essential primary care (PC) in rural and remote Canada. We examined the practice context and responsibilities of this little-known understudied workforce.

Method: Data from Nursing Practice in Rural and Remote Canada II, a 2014 to 2015 pan-Canadian survey, were analyzed.

Results: Of 3,822 respondents, 192 identified that PC was their only practice focus (PC-Only), and for 111, it was one focus among others (PC-Plus). Proportionally more PC-Only than PC-Plus nurses had graduate education, were employed in larger communities and had experienced higher job resources and lower job demands. Proportionally fewer PC-Only than PC-Plus nurses followed protocols/decision support tools, dispensed medications and provided emergency services. Proportionally more PC-Only than PC-Plus nurses ordered advanced diagnostic tests/imaging, and fewer PC-Only than PC-Plus nurses performed and interpreted laboratory tests and diagnostic imaging on site.

Conclusion: Contributions of the rural and remote nursing workforce to PC are rendered invisible by contemporary characterizations of the PC workplace, limiting evaluation and improvement efforts.

Résumé

Contexte et objectif : Les infirmières fournissent des soins de santé primaires (SSP) essentiels dans les régions rurales et éloignées du Canada. Nous avons examiné le contexte et les responsabilités de la pratique de cette main-d'œuvre peu connue et mal étudiée.

Méthode : Nous avons analysé des données provenant d'une enquête pancanadienne sur la pratique infirmière dans les régions canadiennes rurales et éloignées, de 2014 à 2015 (*Nursing Practice in Rural and Remote Canada II*).

Résultats : Parmi 3 822 répondants, 192 ont indiqué que les SSP formaient l'essentiel de leur pratique (SSP-seul) et 111 ont indiqué que ces soins formaient une partie de leur pratique (SSP-plus). Toute proportion gardée, plus d'infirmières SSP-seul que SSP-plus ont reçu leur diplôme, ont été employées dans des grandes communautés, ont bénéficié de plus de ressources au travail et ont connu moins d'exigences au travail. Toute proportion gardée, moins d'infirmières SSP-seul que SSP-plus ont utilisé les outils d'aide aux protocoles ou à la décision, ont délivré des médicaments et ont fourni des services d'urgence. Toute proportion gardée, plus d'infirmières SSP-seul que SSP-plus ont prescrit une imagerie ou un test diagnostic poussé et moins d'infirmières SSP-seul que SSP-plus ont effectué et interprété des tests en laboratoire et des imageries diagnostiques sur les lieux.

Conclusion : La contribution de la main-d'œuvre infirmière dans les régions canadiennes rurales et éloignées demeure invisible en raison des caractérisations contemporaines du lieu de travail des SSP, ce qui limite les efforts d'évaluation et d'amélioration.

Introduction

Canada has experienced nearly two decades of incremental reforms in community-based primary healthcare (Hutchison 2013). Many of the changes have occurred in primary care (PC), which we understand is the first point of entry to and an ongoing point of contact with the overall healthcare system (Starfield 1992). Most Canadians receive care for the majority of their health concerns in PC, from birth to death, often with the same healthcare provider (Statistics Canada 2017).

Nurses now provide PC services in all provinces and territories (Martin-Misener et al. 2014). This is a dramatic change for nurses in southern regions of the country, but less so for nurses in rural and remote regions, where the vastness of Canada's land mass and low population density make PC health service delivery particularly challenging (Martin et al. 2018). Changes to the roles and responsibilities of physicians and nurses, sometimes called task-shifting, is a world wide strategy intended to improve accessibility to PC (Maier and Aiken 2016). Evidence from the US indicates that as PC physician recruitment for rural areas is declining, nurse practitioner (NP) recruitment is increasing (Barnes et al. 2018; Xue et al. 2019). A Canada-wide survey of nurses in rural and remote Canada found that twice as many registered nurses (RNs) working in PC versus RNs working in other care settings reported working beyond their scope of practice (MacLeod et al. 2019). Nurses have been the backbone of rural and remote PC for more than 50 years, yet very little is known about their deployment and practice.

The Canadian Institute for Health Information (CIHI) details the supply, distribution and practice characteristics of NPs, RNs, licensed practical nurses (LPNs) and registered psychiatric nurses (RPNs) in an annual regulated nurse report, the most recent of which is *Regulated Nurses 2018* (CIHI 2019a, b), and in the *Pan-Canadian Primary Health Care Indicator Update Report* (CIHI 2012). The CIHI-regulated nurse report uses registration data collected by provincial/territorial nurse regulatory organizations to identify trends in supply, distribution, employment and demographic characteristics. Place of work is grouped into the following four categories: *hospital* (hospital, mental health centre and rehabilitation/convalescent centre); *community health agency* (nursing station [outpost or clinic], home care agency, community health centre and public health department/unit); *nursing home/long-term care facility*; and *other place of work* (business/industry/occupational health office, private nursing agency/private duty, self-employed, physician's office/family practice unit, educational institution, nursing association/government and other). CIHI's indicator report defines workplace for a PC nurse as "a nursing station, community health centre, or a physician's office/family practice unit" (CIHI 2012, p. 109). Valuable as these definitions are, defining PC nurses using their self-reported practice location can be problematic in rural and remote settings that do not have a clear delineation of specialized places of work as seen in urban settings. Rural and remote healthcare contexts often require nurses to work in multiple practice areas, for example, home and acute care, PC and community care (NNPBC

2018). Therefore, equating workplace with type of health services offered may be inappropriate, particularly in smaller communities (Smith and Vandall-Walker 2017).

Health workforce solutions are needed to address the health needs of rural and remote populations that have high proportions of older adults and some of the highest rates of all-cause mortality in the country (Tam 2017). In Canada's rapidly changing healthcare environment, not knowing which nurses are providing PC and where they are employed is a serious information gap in planning and providing accessible high-quality rural and remote health services (British Columbia Ministry of Health 2015). The purpose of this study was to examine the practice context and responsibilities of PC nurses in rural and remote Canada.

Method

Design

Data for this analysis are from a pan-Canadian cross-sectional survey – *Nursing Practice in Rural and Remote Canada II*. MacLeod et al. (2017) provide a detailed description of the methods used. The definition of rural encompasses areas beyond the commuting zones of communities with 10,000 or more residents (du Plessis et al. 2001). Remote was not specifically defined. We used the phrase "rural and remote" because findings of previous research have shown more similarities than differences in how nurses define rural and remote workplaces (Kulig et al. 2008). The comprehensive questionnaire (27 pages in English/ 31 pages in French) was created based on a previous work (MacLeod et al. 2004; Stewart et al. 2005) and an integrated view of workforce planning (Tomblin Murphy et al. 2009). The questionnaire was developed iteratively by a 16-member research team with guidance from a 19-member advisory group composed of provincial and national nurse decision-makers. It was mailed in 2014 to 2015 to 10,072 regulated nurses practicing in rural and remote communities in each province and all regulated nurses in the territories.

Participants

A total of 3,822 out of the 9,622 eligible respondents completed the survey, resulting in a 40% response rate (N = 3,822). The questions that were used to identify the subsample of rural and remote PC nurses (staff nurses, NPs, clinical nurse specialists and managers) included those on primary place of employment and area of practice. Options for PC place of employment included those in CIHI's (2012) definition, such as nursing station, community health centre or physician's office/family practice unit, with additions of nursing stations/ outpost clinics/nurse clinics, family practice teams, NP-led clinics, multidisciplinary primary healthcare clinics. Only one response was permitted for this question. For area of current practice, respondents had the option of choosing all that applied from the following: acute care, primary care, community health, long-term care, home care, hospice/end-of-life/palliative care, mental health, and other. RPN respondents were excluded from the PC analysis owing to the small sample size (N = 29).

Measures

Survey content areas used in this analysis were as follows: individual (nurse type, gender, age, highest nursing education credential, region of residence and primary position), work community (population size and distance from advanced referral centre), workplace (required to be on call, job resources, job demands and primary health care engagement) and practice responsibilities (e.g., assessment [nine items], laboratory tests [five items], diagnostic tests [four items], diagnostic imaging [five items], diagnosis and referral [five items], therapeutic management [five items], emergency care and transportation [four items]). For brevity, we report on the subset of responsibilities that most distinguish the differences.

The 40-item Primary Health Care Engagement (PHCE) scale was composed of 10 subscales, each with three to five items (Kosteniuk et al. 2017). Items were rated on a five-point scale (1 = *strongly disagree* to 5 = *strongly agree*). The items in each subscale were summed and averaged to produce a subscale score indicating low agreement (1.0–3.0) or high agreement (>3.0). Higher scores represent perceptions of greater workplace engagement in PHC delivery. The 24-item Job Resources in Nursing (JRIN) scale and the 22-item Job Demands in Nursing (JDIN) scale were used to measure the overall job-related demands and resources across organizational, interpersonal and job position/task levels (Penz et al. 2018). The total summated scores ranged from 24 to 120 for the JRIN, interpreted as low (24–56), medium (57–88) or high (89–120) level of work-related resources. Similarly, the total summated scores on the JDIN ranged from 22 to 110, interpreted as low (22–51), medium (52–80) or high (81–110) level of work-related demands. Each of the six subscales within each scale was summated and then divided by the total number of items in each to produce a mean score, interpreted from a low level of agreement on that particular subscale (1.0–3.0) to a high level of agreement (>3.0).

Analysis

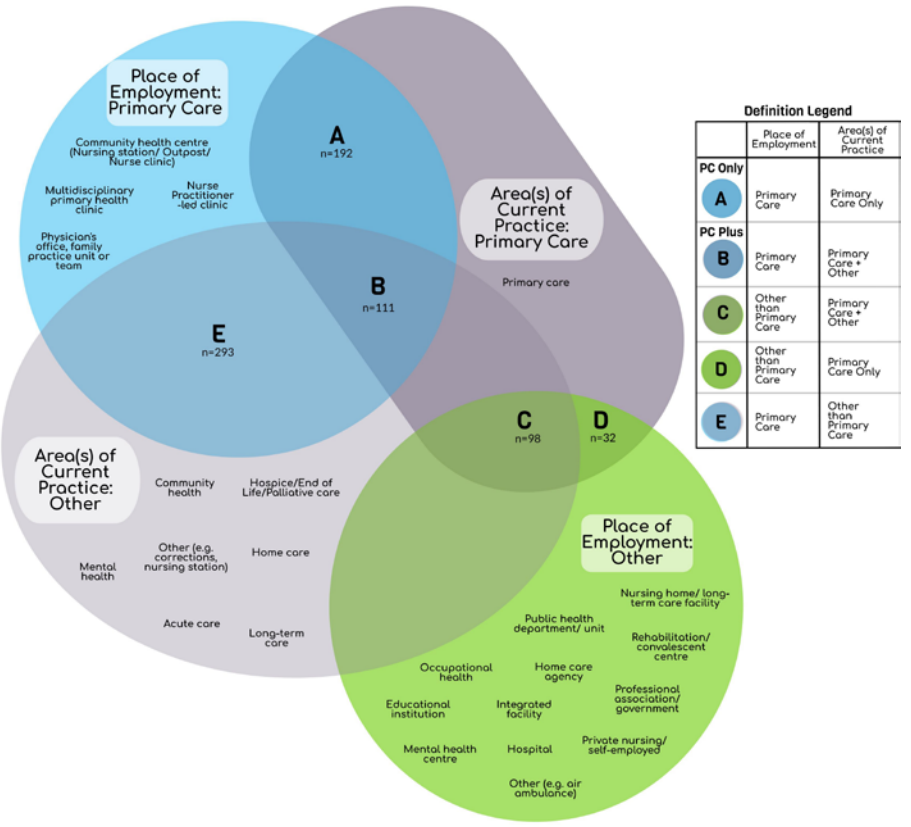
Data analyses were conducted using IBM SPSS V. 24.0 (2015). Descriptive statistics were used to analyze demographics of nurses employed in PC, characteristics of their places of employment and practice responsibilities. Chi-square analyses and *z*-test for proportions were used to determine between-group differences for categorical variables and to follow up significant results, respectively. Tables 1–3 and Appendix 1 present comparisons by group (PC-Only vs. PC-Plus), and Appendix 2 displays proportion testing for pairwise comparisons by nurse type (all these elements are available online at longwoods.com/content/26130). For between-group comparisons of continuous variables, multivariate analyses of variance using Wilk's criterion (λ) was the test statistic for the PHCE, JDIN and JDRN total summated scales and subscales. Significance was set at $p < 0.05$ for all tests.

Results

Responses to questions about place of employment and practice area helped divide the nurse respondents into five distinct groups (Figure 1). Group A, "PC-Only" (n = 192), identified

their place of employment as PC and their area of practice as PC only. Group B, "PC-Plus" (n = 111), identified their place of employment as PC and their area of practice as PC plus at least one other area, most commonly community health and or acute care. Demographic information about the nurses in Groups A and B is provided in Table 1. Group C (n = 98) identified their place of employment as other than PC and area of practice as PC plus at least one other area, most commonly acute care, long-term care and hospice/end-of-life/palliative care. This group included five NPs (5.1%), 42 RNs (42.9%) and 52 LPNs (52.0%). Group D identified their place of employment as other than PC, most commonly hospital, and their area of practice as PC-Only (n = 32). This group included six NPs (18.8%), 15 RNs (45.9%) and 11 LPNs (34.4%). Group E identified their place of employment as PC and their area of practice as other than PC (n = 293), most commonly community health (169; 57.7%) and home care (66; 22.5%). This group included 15 NPs (5.1%), 240 RNs (81.9%) and 38 LPNs (13%). In consultation with our advisory group, we prioritized comparative analysis of the groups that most aligned with CIHI's definition of PC practice, namely, Groups A and B (PC-Only and PC-Plus). Data are available for the other groups from the corresponding author.

FIGURE 1. Overlapping definitions of a primary care nurse based on area(s) of current practice and primary place of employment



As Table 1 shows (available online at longwoods.com/content/26130), Group A had significantly higher proportions of NPs and LPNs and fewer RNs than Group B. Group A also had a higher proportion of nurses with graduate education and a lower proportion of nurses in the 30–34 and >60 age categories. Proportionally, there were more Ontario and Atlantic nurses in Group A and more Territories nurses in Group B.

As shown in Table 2 (available online at longwoods.com/content/26130), proportionally fewer nurses in Group A than in Group B worked in communities with a population of <1,000 and at a distance of ≥1,000 km to an advanced referral centre. Fewer PC-Only than PC-Plus nurses were required to be on call. In both Tables 1 and 2, Group A signifies PC-Only nurses and Group B is PC-Plus.

Table 3 (available online at longwoods.com/content/26130) shows that in the analysis of the PHCE, JDIN and JRIN summated scales, the combined PHCE dependent variable had a significant main effect for group $F(8, 231) = 8.55$, $p < 0.001$, and partial $\eta^2 = 0.229$. Compared with nurses in Group A (PC-Only), nurses in Group B (PC-Plus) reported significantly higher scores on the summated PHCE scale and on the Accessibility/Availability and Population-Orientation subscales. Nurses in Group A (PC-Only) had significantly higher scores on the Quality Improvement subscale than nurses in Group B (PC-Plus).

The results from the full JRIN scale indicated there were statistically significant differences between groups $F(6, 261) = 4.09$, $p = 0.001$, and partial $\eta^2 = 0.086$. Nurses in Group A (PC-Only) reported significantly higher scores on the summated JRIN scale and on the subscales of Supervision, Recognition and Feedback; Staffing and Time; Technology; and Training, Professional Development, and Continuing Education than nurses in Group B (PC-Plus). The level of perceived job-related resources for both Groups A and B was in the medium range.

The results from the full JDIN scale indicated there were statistically significant differences between groups $F(7, 219) = 2.68$, $p = 0.011$, and partial $\eta^2 = 0.079$. Nurses in Group B (PC-Plus) reported significantly higher scores on the summated JDIN scale, the Equipment and Supplies subscale and the Safety subscale. The level of perceived demands related to their work for both groups was in the low-to-medium range.

Appendix 1 compares practice responsibilities of PC-Only and PC-Plus nurses, and is available at longwoods.com/content/26130. Proportionally fewer PC-Only nurses than PC-Plus nurses completed history-taking and physical examinations, independently made a nursing diagnosis, followed protocols/decision-support tools, dispensed medications and provided emergency care services and transportation. Although proportionally more PC-Only than PC-Plus nurses ordered advanced diagnostic tests and imaging, fewer performed and interpreted laboratory tests and diagnostic imaging on site. Box 1 outlines a fictional example of a PC-Plus nurse developed from survey results to illustrate this lesser-known practice group.

BOX 1. Fictional example of a PC-Plus nurse developed from survey results to illustrate this lesser-known practice group

Susan, a Registered Nurse, works in a health centre in a small town. It is a multidisciplinary clinic with PC provider offices on one "side" of the building, a separate entrance for patients seeking public health services, and an urgent care entrance that is staffed until 5 p.m. Susan works in the urgent care area. While other PC providers see booked patients most of the day and one is "on call" for the urgent care side, Susan triages all patients who enter the clinic seeking care. Some patients she can manage independently and others she consults with or refers to team members. There are limited lab and X-ray services in the clinic, which Susan often must perform, and she maintains her skills in emergency nursing. The health centre is just off a major highway, so in addition to myocardial infarctions or serious occupational/agricultural injuries coming through the door, Susan must be prepared to receive trauma victims from motor vehicle accidents or other similar events. Four mornings per month, she visits the assisted living complex across the road with another PC provider to help the staff with medication reconciliation and to complete assessments for residents that have new or chronic health concerns.

Recognizing that there are legislated scopes of practice, to explore what actually happens in practice, albeit through self-report data, we analyzed the sample by nurse type, (i.e., NP, RN and LPN). This full analysis is available in Appendix 2 (available at longwoods.com/content/26130). Proportionally more NPs than RNs completed history-taking and physical examinations and independently made a medical diagnosis; prescribed medications; ordered, performed, interpreted and followed up advanced diagnostic tests; ordered, interpreted and followed up diagnostic imaging; and made referrals to medical specialists. Proportionately fewer NPs than RNs dispensed medications, prescribed using protocols, performed laboratory tests and diagnostic imaging, and were involved in emergency care and transportation. Comparing RNs and LPNs, proportionately more RNs performed all practice responsibilities except ordering advanced diagnostic tests and advanced imaging, which few nurses of either type did.

Discussion

This study provides new information about nurses providing PC in rural and remote Canada, the context of their practice and their practice responsibilities. The distances involved and the small number of nurses offering PC to large geographically dispersed populations are staggering. Our results highlight the vital role nurses have in not only the delivery of a broad range of PC services but also their engagement and partnership with communities to improve health and health determinants. Our results illuminate the complex nature of PC in rural and remote Canada as well as the gaps in resources and services that should be enhanced to provide PC.

Nurses' responses to survey questions about their place of employment and practice focus reveal the diverse nature of their roles in PC and the important influence of context on practice (Hogg et al. 2008). Although nurses are regarded as the backbone of rural and remote healthcare, there is surprisingly little research about their practice in PC (Martin-Misener et al. 2014). This may explain the lack of focused attention to the nursing workforce in policy discussions about rural and remote PC. Filling this information gap is critical for decision-makers responsible for healthcare planning and delivery.

Our results reveal clear differences between nurses in PC workplaces who report PC as their only practice focus (PC-Only) and nurses who report PC as one of additional practice foci (PC-Plus). The features of the workplaces of PC-Only (Group A) nurses (i.e., more NPs, graduate education and communities with 5,000 plus people and within 500 km of a major referral centre) resemble interprofessional team-based models of PC also seen in urban areas (Hutchison et al. 2011). In contrast, PC-Plus nurses' (Group B's) workplace features (i.e., more RNs, baccalaureate education communities with <1,000 people and distance >1,000 km from major referral centres) are suggestive of health centres in more remote areas (Martin-Misener et al. 2008).

Nurses in both groups perceived engagement of their workplace in PHC delivery to be high; however, the PC-Plus nurses' scores were higher overall, and for Accessibility and Population-Orientation subscales (Kosteniuk et al. 2017). Population orientation refers to "the extent to which primary care providers assess and respond to the health needs of the population they serve" (Haggerty et al. 2007, p. 340). PC-Plus nurses were more likely than PC-Only nurses to report a good fit between their workplace and community needs. One interpretation is that because many PC-Plus nurses worked in remote communities in which multiple services are offered in one location (e.g., PC, public health and emergency care), they could perceive their workplace to be more oriented and responsive to population needs. PC-Only nurses had higher scores on the Quality Improvement subscale. This could be related to this group having a higher proportion of nurses with graduate education and/or having available staff, time and technology resources. Another possibility is that the more traditional workplaces of PC-Only nurses may have been influenced by the increasing focus on healthcare improvement in PC promoted by organizations such as the Canadian Foundation for Health Care Improvement and Accreditation Canada.

Nurses in Group B reported higher job-related demands and had lower perceived job-related resources than PC-Only nurses. Demands were particularly high with respect to safety (e.g., their personal safety being at risk) and equipment and supplies (e.g., necessary equipment or supplies being unavailable or in poor condition), issues that have been reported in previous studies (MacLeod et al. 2008; Martin-Misener et al. 2008). Interestingly, demands such as comfort with working conditions (which included items related to workload), preparedness/scope of practice and isolation were not significantly different between groups, even though higher proportions of PC-Plus nurses had on-call responsibilities and provided emergency care and transportation. It is possible that the differences in JRIN and JDIN results are related to the population size and geographic location of the communities in which nurses worked. Relative to each other, more PC-Plus than PC-Only nurses were employed in smaller-sized and more remote communities.

In our study, NPs' practice responsibilities aligned with their legislated scope of practice (CCRNR 2015). Not unexpectedly, some RNs had responsibilities extending beyond their legislated scope of practice. PC-Plus nurses, most of whom were baccalaureate-educated RNs, were employed in some of the smallest and remotest communities with high overall

job-related demands and fewer resources. That proportionally more PC-Plus than PC-Only nurses dispensed medications, performed and analyzed laboratory tests on-site, and provided emergency care and transportation is consistent with the role descriptions of nurses employed in communities without other on-site personnel to offer these services (Martin-Misener et al. 2008). It is possible that PC-Plus nurses who were the group more likely to be the recipients of task-shifting had different JRIN and JDIN scores because of this. The roles and responsibilities of these RNs encompass task shifting from not only physicians and NPs but also pharmacists, laboratory technicians and paramedics. Notwithstanding the small number of LPN participants, this study identifies previously undocumented task-shifting from RNs to LPNs in PC that requires monitoring.

Resourcing PC is a pressing equity concern given the overlap between rural and remote residence and Indigenous populations (Marchildon 2018). Task-shifting has long been a necessary strategy to improve access to rural and remote PC and may become even more common with movement toward team-based care in PC. In rural and remote communities, where team composition and size differ from urban areas, the work of nurses in filling gaps in access to other team members cannot remain invisible. Rather, the associated changes in nurses' roles and responsibilities must be accompanied by education and regulation to safeguard patient safety and quality of care (Freund et al. 2015; Moffit et al. 2018). Consideration of the skill mix of other team members is also needed to promote high-quality PC in rural and remote communities. Attending to these supports and increasing the number of NPs are approaches to prevent, or at least reduce, a double standard of care between rural/remote and urban communities (Tarlier and Browne 2011).

Lastly, our study has implications for optimizing measurement of PC performance in rural and remote Canada. The variability in how nurses responded to questions about primary place of employment and area(s) of practice turned what initially seemed like a straightforward approach to determine the subsample of interest into unanticipated complexity. Important first steps are to consider how to expand CIHI's 2012 definition of PC to be more inclusive of the workplaces where rural and remote PC nurses are employed and for nurse regulators to develop consistency in defining PC in registration databases. More information is needed about the other three groups of nurses identified in our study (Figure 1). The largest of these groups was nurses who identified that their place of employment was PC but that their area of practice was other than PC (n = 293). This could potentially represent nurses who are employed in PC settings, but have a different practice focus, for example, mental health or public health. Qualitative research to examine nurses' understanding of the nature of PC would be useful to inform definition changes.

This study was pan-Canadian in scope. A limitation is the small number of respondents from Quebec despite the survey being available in French. The inclusion of nurses in Whitehorse, Iqaluit and Yellowknife, despite the small number, may have diluted differences between Groups B and A. Because we did not define any area of practice, including PC, some nurses may have been unclear about what PC is. This may have influenced their responses to the survey, resulting in under- or overreporting of employment in PC.

Conclusion

The contributions of the rural and remote nursing workforce to PC include a mosaic rendered invisible by contemporary characterizations and interpretations. Current approaches to defining PC workplaces are insufficient to identify nurses providing PC in rural and remote Canada, limiting evaluation and improvement initiatives.

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References

Barnes, H., M.R. Richards, M.D. McHugh and G. Martsolf. 2018. Rural and Nonrural Primary Care Physician Practices Increasingly Rely on Nurse Practitioners. *Health Affairs (Millwood)* 37(6): 908–914. doi:10.1377/hlthaff.2017.1158.

British Columbia Ministry of Health. 2015. Rural Health Services in BC: A Policy Framework to Provide a System of Quality Care. Retrieved January 30, 2019. <https://www.health.gov.bc.ca/library/publications/year/2015/rural-health-policy-paper.pdf>.

Canadian Council of Registered Nurse Regulators (CCRNR). 2015. Nurse Practitioner Practice Analysis. Retrieved November 30 2018. <http://www.ccrnr.ca/nurse-practitioners.html>.

Canadian Institute for Health Information (CIHI). 2012. Pan-Canadian Primary Healthcare Indicator Report Update. Ottawa, ON. Retrieved January 30, 2019. <www.cihi.ca>.

Canadian Institute for Health Information (CIHI). 2019a. Regulated Nurses 2018 Canada and Jurisdictional Highlights. Retrieved October 30, 2019. <www.cihi.ca>.

Canadian Institute for Health Information (CIHI). 2019b. Regulated Nurses 2018 Methodology Guide. Retrieved October 30, 2019. <www.cihi.ca>.

Canadian Nurses Association (CNA). 2017. Nurse Practitioners. Retrieved January 30, 2019. <https://www.cna-aiic.ca/en/nursing-practice/the-practice-of-nursing/advanced-nursing-practice/nurse-practitioners>.

du Plessis, V., R. Beshiri, R.D. Bollman and H. Clemenson. 2001. Rural and Small Town Canada Analysis Bulletin, Definitions of Rural. Retrieved January 30, 2019. <https://www150.statcan.gc.ca/n1/en/pub/21-006-x/21-006-x2001003-eng.pdf?st=BNNRTf-P>.

Freund, T., C. Everett, P. Griffiths, C. Hudon, L. Naccarella and M. Laurant. 2015. Skill Mix, Roles and Remuneration in the Primary Care Workforce: Who Are the Healthcare Professionals in the Primary Care Teams across the World? *International Journal of Nursing Studies* 52(3): 727–43. doi:10.1016/j.ijnurstu.2014.11.014.

Haggerty, J., F. Burge, D. Gass, J.F. Lévesque, M.D. Beaulieu, R. Pineault et al. 2007. Operational Definitions of Attributes of Primary Health Care: Consensus among Canadian Experts. *Annals of Family Medicine* 5: 336–44. doi:10.1370/afm.682.

Hogg, W., M. Rowan, G. Russell, R. Geneau and L. Muldoon. 2008. Framework for Primary Care Organizations: The Importance of a Structural Domain. *International Journal for Quality in Health Care* 20(5): 308–13. doi:10.1093/intqhc/mzm054.

Hutchison, B., J.F. Levesque, E. Strumpf and N. Coyle. 2011. Primary Health Care in Canada: Systems in Motion. *Millbank Quarterly* 89(2): 256–88. doi:10.1111/j.1468-0009.2011.00628.x.

Hutchison, B. 2013. Reforming Canadian Primary Care – Don't Stop Half-Way. *Healthcare Policy* 9(1): 12–25. doi:10.12927/hcpol.2013.23481.

IBM Corp. Released 2016. IBM SPSS Statistics for Windows, Version 24.0. Armonk, NY: IBM Corp.

Kosteniuk, J.G., N.J. Stewart, C.P. Karunanayake, E.C. Wilson, K.L. Penz, J.C. Kulig et al. 2017. Exploratory Factor Analysis and Reliability of the Primary Health Care Engagement (PHCE) Scale in Rural and Remote Nurses: Findings from a National Survey. *Primary Health Care Research and Development* 18(6): 608–22. doi:10.1017/S146342361700038X.

Kulig, J.C., M.E. Andrews, N.L. Stewart, R. Pitblado, M.L.P. MacLeod and D. Bentham et al. 2008. How do Registered Nurses Define Rurality? *Australian Journal of Rural Health* 16: 28–32. doi: 10.1111/j.1440-1584.2007.00947.

MacLeod, M., J. Kulig, N. Stewart and R. Pitblado. 2004. Rural and Remote Nursing Practice: Final Report to Canadian Health Services Research Foundation. Retrieved February 7, 2020. <https://www.unbc.ca/sites/default/files/sections/rural-nursing/en/17-rrnfinalreport.pdf>.

MacLeod, M., R. Martin-Misener, K. Banks, M.A. Morton, C. Vogt and D. Bentham. 2008. I'm a Different Kind of Nurse: Advice from Nurses in Rural and Remote Canada. *Canadian Journal of Nursing Leadership* 21(3): 40–53. doi:10.12927/cjnl.2008.20060.

MacLeod, M., J. Kulig, N. Stewart, P. Anguish, M.E. Andrews, D. Banner et al. 2017. Who Are the Nurses that Work in Rural and Remote Communities in Canada and Why Do They Stay? Results of a National Survey. *Human Resources for Health* 15(1): 34. doi:10.1186/s12960-017-0209-0.

MacLeod, M.L.P., N.J. Stewart, J.G. Kosteniuk, K.L. Penz, J. Olynick, C.P. Karunanayake et al. 2019. Rural and Remote Registered Nurses' Perceptions of Working beyond Their Legislated Scope of Practice. *Nursing Leadership* 32(1): 8–19. doi:10.12927/cjnl.2019.25852.

Maier, C.B. and L.H. Aiken. 2016. Task Shifting from Physicians to Nurses in Primary Care in 39 Countries: A Cross-Country Comparative Study. *The European Journal of Public Health* 26(6): 927–34. doi:10.1093/eurpub/ckw098.

Marchildon, G.P. 2018. A Policy Research Agenda for Health Systems in Canada's North. *Healthcare Papers* 17(3): 35–40. doi:10.12927/hcpap.2018.25503.

Martin, D., A.P. Miller, A. Quesnel-Vallée, N.R. Caron, B. Vissandjée and G.P. Marchildon. 2018. Canada's Universal Health-Care System: Achieving its Potential. *The Lancet* 391(10131): 1718–35. doi:10.1016/S0140-6736(18)30181-8.

Martin-Misener, R., M. MacLeod, K. Banks, A. Michel Morton, C. Vogt and D. Bentham. 2008. There's Rural, and Then There's Rural: Advice from Nurses Providing Primary Healthcare in Northern Remote Communities. *Canadian Journal of Nursing Leadership* 21(3): 40–53. doi:10.12927/cjnl.2008.20062.

Martin-Misener, R., D. Bryant-Lukosius, C. Bullard, D. Campbell, N. Carter, F. Donald et al. 2014. *Optimizing the Role of Registered Nurses and Nurse Practitioners in Primary Care: Final Report*. Canadian Nurses Association. Retrieved January 30, 2019. <http://www.cna-aiic.ca/en/professional-development/canadian-network-of-nursing-specialties/whats-new-with-the-network/network-news-room/news-about-the-network/2014/optimizing-rn-roles>.

Moffit, P., R. Munday and J. Inman. 2018. Professionalism and Professional Conduct in the Northwest Territories and Nunavut Canada. In H. Exner-Pirot, B. Norbve and L. Butler (Ed.), *Northern and Indigenous Health and Healthcare* (pp. 249–59). Saskatoon, Saskatchewan: University of Saskatchewan.

Nurses and Nurse Practitioners of British Columbia (NNPBC). 2018. *Enhancing Rural and Remote Nursing Practice for a Healthier B.C.* Retrieved January 30, 2019. <https://www.nnpbc.com/pdfs/policy-and-advocacy/rural-and-remote/Enhancing-Rural-&-Remote-Nursing-Practice-for-a-Healthier-BC.pdf>.

Penz, K.L., J.G. Kosteniuk, N.L. Stewart, M.L.P. MacLeod, J.C. Kulig, C.P. Karunanayake et al. 2018. Development and Psychometric Evaluation of the Job Demands in Nursing Scale and Job Resources in Nursing Scale: Results from a National Study. *Nursing Open* 6(2): 348–66. doi:10.1002/nop2.215.

Smith, J.C. and V. Vandall-Walker. 2017. A Double Whammy! New Baccalaureate Registered Nurses' Transitions into Rural Acute Care. *Rural and Remote Health* 17(4): 4256. doi:10.22605/RRH4256.

Starfield, B. 1992. *Primary Care: Concept, Evaluation, and Policy*. New York, NY: Oxford University Press.

Statistics Canada. 2017. Primary Health Care Providers, 2016. Ottawa, ON. Retrieved January 30, 2019. <https://www150.statcan.gc.ca/n1/pub/82-625-x/2017001/article/54863-eng.htm>.

Stewart, N.J., C. D'Arcy, J.R. Pitblado, D.G. Morgan, D. Forbes, G. Remus et al. 2005. A Profile of Registered Nurses in Rural and Remote Canada. *Canadian Journal of Nursing Research* 37(1). Retrieved February 7, 2020. <https://cjnr.archive.mcgill.ca/article/view/1930>.

Tam, T. 2017. The Chief Public Health Officer's Report on the State of Public Health in Canada 2017 – Designing Healthy Living. Retrieved February, 2020. <https://www.canada.ca/en/public-health/services/publications/chief-public-health-officer-reports-state-public-health-canada/2017-designing-healthy-living.html>.

Tarlier, D. and A. Browne. 2011. Remote Nursing Certified Practice: Viewing Nursing and Nurse Practitioner Practice through a Social Justice Lens. *Canadian Journal of Nursing Research* 43(2): 38–61.

Tomblin Murphy, G., A. MacKenzie, R. Alder, S. Birch, G. Kephart and L. O'Brien-Pallas. 2009. An Applied Simulation Model for Estimating the Supply of and Requirements for Registered Nurses Based on Population Health Needs. *Policy Politics Nursing Practice* 10(4): 240–51. doi:10.1177/1527154409358777.

Xue, Y., J. A. Smith and J. Spetz. 2019. Primary Care Nurse Practitioners and Physicians in Low-Income and Rural Areas, 2010-2016. *JAMA 321* (1): 102–05. doi:10.1001/jama.2018.17944.

RESEARCH PAPER

Comparing Childhood Cancer Care Costs in Two Canadian Provinces

Comparaison des coûts associés aux soins oncologiques chez les enfants dans deux provinces canadiennes

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Abstract

Background: Cancer in children presents unique issues for diagnosis, treatment and survivorship care. Phase-specific comparative cost estimates are important for informing healthcare planning.

Objectives: The aim of this paper is to compare direct medical costs of childhood cancer by phase of care in British Columbia (BC) and Ontario (ON).

Methods: For cancer patients diagnosed at <15 years of age and propensity-score-matched non-cancer controls, we applied standard costing methodology using population-based healthcare administrative data to estimate and compare phase-based costs by province.

Results: Phase-specific cancer-attributable costs were 2%–39% higher for ON than for BC. Leukemia pre-diagnosis costs and annual lymphoma continuing care costs were >50% higher in ON. Phase-specific in-patient hospital costs (the major cost category) represented 63%–82% of ON costs, versus 43%–73% of BC costs. Phase-specific diagnostic tests and procedures accounted for 1.0%–3.4% of ON costs and 2.8%–13.0% of BC costs.

Conclusions: There are substantial cost differences between these two Canadian provinces, BC and ON, possibly identifying opportunities for healthcare planning improvement.

Résumé

Contexte : Le cancer chez l'enfant présente des problèmes uniques en matière de diagnostic, de traitement et de survie. Il importe d'effectuer une comparaison portant sur l'estimation des coûts, selon des étapes précises, pour renseigner la planification des services de santé.

Objectif : L'objectif de cet article est de comparer les coûts médicaux directs du cancer chez l'enfant, selon les étapes de soins, en Colombie-Britannique et en Ontario.

Méthode : Pour les patients qui ont reçu un diagnostic de cancer avant l'âge de 15 ans et pour le groupe témoin de personnes non cancéreuses au score de propension similaire, nous avons employé une méthodologie standard pour le calcul des coûts au moyen des données administratives de santé afin d'estimer et de comparer, étape par étape, les coûts dans les provinces.

Résultats : Les coûts attribuables au cancer pour les étapes à l'étude étaient de 2 à 39 % plus élevés en Ontario qu'en Colombie-Britannique. Les coûts pré-diagnostics associés à la leucémie et les coûts annuels pour un lymphome étaient >50 % plus élevés en Ontario. Les coûts des patients hospitalisés pour les étapes à l'étude (la principale catégorie de coûts) représentaient de 63 à 82 % des coûts en Ontario, contre 43 à 73 % en Colombie-Britannique. Les tests et procédures diagnostiques pour les étapes à l'étude comptaient pour 1,0 à 3,4 % des coûts en Ontario, contre 2,8 à 13,0 % en Colombie-Britannique.

Conclusion : Il y a d'importantes différences de coûts entre les deux provinces canadiennes, l'Ontario et la Colombie-Britannique, ce qui laisse possiblement place à une amélioration dans la planification des services de santé.

Introduction

The cost of treating cancer in Canada is increasing (de Oliveira et al. 2018). Healthcare funders must deliver high-quality and high-value care within sustainable healthcare systems. Assessment and comparison of performance of different healthcare systems, using recognized indicators such as patient-level utilization and costs over a disease course (Carinci et al. 2015), can identify opportunities for care and system improvement (Tarricone 2006). Differences between systems in policy/practice factors (e.g., locus of care, resource distribution and staff mix) and exogenous factors (e.g., labour costs, patient mix, facility size and remoteness) can point to opportunities to improve resource allocation, system performance, accessibility and sustainability (CIHI 2017; Cunningham 2000; Lipscomb et al. 2013; Robinson 1993; Tarricone 2006). Adult cancer-related resource distribution and costs were found to vary between Ontario (ON) and British Columbia (BC; de Oliveira et al. 2017a). Cancer costs for children likely differ from those for adults (CCS 2017; Ellison et al. 2009) because children have distinct cancer types often requiring lengthy complex therapy (Lanzkowsky et al. 2016; Nathan et al. 2019) at pediatric cancer centres, which provide more comprehensive care than adult facilities (Brand et al. 2016). Also, overall survival is higher (CCS 2017; Chan and Raney 2005; Ellison et al. 2009) among children, and most survivors experience late and/or long-term effects of cancer and/or its treatment (Lorenzi et al. 2011; Oeffinger et al. 2006).

Few studies have measured direct medical costs of childhood cancer care in Western countries (de Oliveira et al. 2017b, 2017c; Luo et al. 2002; Mueller et al. 2017; Nathan et al. 2019; Price et al. 2012). This observational study estimates cancer-attributable healthcare costs for children with cancer by phase of care in the provinces of ON and BC, using administrative data, to identify potentially policy-relevant interprovincial differences.

Methods

Ethics approvals and data management

The study was approved by institutional review boards at Sunnybrook Health Sciences Centre and the University of Toronto, Toronto, ON, and at BC Cancer/University of British Columbia, Vancouver, BC. Data set access was approved by data stewards (ministry individuals authorized to approve research access to government data). Identifying information was removed; individual consent was not required. ON and BC data were analyzed separately using parallel procedures, recognizing differences between provincial data sets.

Subjects

Eligible cases were residents of BC and ON with a first diagnosis of any cancer or tumour from January 1995 to June 2010 based on the International Classification of Childhood Cancer, Third Edition (Steliarova-Foucher et al. 2005), before the age of 15 years. These provinces represent 49.8% of the Canadian pediatric population and were the only Canadian

provinces with multiple years of detailed comparable data. ON cases were identified from the Paediatric Oncology Group of Ontario Network Information System, a registry and clinical database from the five ON pediatric cancer centres, which treat 96% of ON children diagnosed at <15 years (Greenberg et al. 2003). BC cases were identified from the BC Cancer Registry, which ascertains all cases diagnosed among BC residents using multiple sources, including the BC Children's Hospital (BCCH) and BC cancer treatment centres. Over 90% of children with cancer are referred to the BCCH. Cases with identical dates of diagnosis and death, missing or invalid codes for histology or sex or invalid provincial medical insurance plan numbers were excluded. We grouped patients into the three most common childhood cancers (leukemia, lymphoma and central nervous system [CNS] tumours) and a fourth group consisting of all other cancers.

Control subjects without cancer were selected from the provincial health insurance plan registries. Potential controls were randomly matched to cases on sex, birth year and month and assigned index dates corresponding to the diagnosis dates of the cases. A propensity score (probability of having cancer) was computed for each case and potential control, with sex, neighbourhood-level rurality of residence (Statistics Canada 2013) and co-morbidity as predictors. Co-morbidity was calculated for the year before diagnosis/index date, or from birth to diagnosis/index date if less than a year, using number of Aggregated Diagnosis Groups, a morbidity grouping generated from diagnostic codes in in-patient and outpatient administrative records, in a case-mix adjustment system validated for patients of all ages (Reid et al. 2001; Starfield and Kinder 2011). "Greedy matching" was used to select three controls with the closest propensity scores within 0.1 standard deviation of each patient's propensity score (Austin 2011; Austin and Mamdani 2006). Selected controls lived at least as long as the patients to whom they were matched.

Costs and data sources

Direct medical costs were tracked from 60 days before diagnosis or index date or from date of birth (whichever was later) until death or December 31, 2010, the latest date for which all data sets were available in both provinces. Costs from all years were converted to 2012 Canadian dollars using the Consumer Price Index (Statistics Canada 2012).

Data sets included information on in-patient hospitalizations, same-day surgery/procedures, chemotherapy, radiotherapy, physician services, diagnostic tests, home and community care, complex continuing care and outpatient prescription drugs. Supplementary Table 1 describes the data and costing methods and can be found at https://www.longwoods.com/content/26129.

Costs for hospital-based services were estimated by multiplying the resource intensity weight, a measure of resource utilization (CIHI 2016), of each visit by the cost per weighted case (CPWC), the cost of treating an average patient for a specific hospital (Wodchis et al. 2013), for the year of use (de Oliveira et al. 2017b, c). Because children have complex care needs, pediatric hospitals have higher CPWC values than general hospitals. Therefore, cost

estimates for in-patient hospitalizations, emergency department (ED) visits and same-day surgeries were generated using hospital-specific CPWC of pediatric hospitals (de Oliveira et al. 2017b, c) and the provincial average hospital CPWC for other hospitals.

Statistical analysis

Based on clinical relevance and joinpoint analysis (Baker et al. 1991; de Oliveira et al. 2017a; Kim et al. 2000; Yabroff et al. 2008), we defined (1) *pre-diagnosis phase*, including diagnostic testing, as the 60 days prior to diagnosis (or from birth to diagnosis in patients diagnosed before 60 days of age); (2) *initial phase*, encompassing primary treatment, from diagnosis date to 360 days after diagnosis; (3) *continuing phase* of variable length, involving surveillance and follow-up care; and (4) *terminal phase*, up to 360 days before death, for those who died. The continuing phase included the time between the end of the initial phase and the start of the terminal phase or the end of the study observation period (December 31, 2010). For terminal phase determination, we looked forward to December 31, 2011, to see if patients died within one year after the end of the observation period. Phase-specific mean costs per patient, including patients who did not use the service and had zero cost, and 95% confidence intervals were estimated overall, by cancer type and resource, for cases and controls. Costs for initial, continuing and terminal phases were standardized to annual costs, based on a 360-day year. Mean net (cancer-attributable) costs were estimated using a generalized estimating equation model (SAS Proc GENMOD), with case and control as binary clusters, to generate individual costs.

Results

Subjects

There were 1,503 cases (4,509 controls) in BC and 4,606 cases (13,818 controls) in ON available for analysis (Supplementary Table 2, available at https://www.longwoods.com/content/26129). Approximately 92% of cancer patients (1,390BC; 4,261ON) were part of the initial phase, 85% (1,293BC; 3,880ON) spent time in the continuing phase, and only 18%–19% (273BC; 893ON) entered the terminal phase. Sociodemographic characteristics and co-morbidity were similar between BC and ON cases.

TABLE 1. Phase-specific mean net costs by diagnosis* (Canadian $2012) for childhood cancer cases (1995–2010) in British Columbia and Ontario

Cancer diagnosis	*N* (%)	Pre-diagnosis	Initial	Continuing	Final
British Columbia					
Leukemia*	494 (32.9)	$2,879	$107,129	$14,436	$523,870
Lymphoma*	151 (10.0)	$5,264	$81,650	$4,865	$643,418
Central nervous system (CNS)*	345 (23.0)	$7,453	$84,372	$10,193	$231,412
Other§	513 (34.1)	$5,049	$105,455	$16,200	$208,000
Total	1,503 (100)	$4,909	$99,087	$13,133	$310,798
Ontario					
Leukemia*	1,636 (35.5)	$4,898	$156,225	$21,418	$432,010
Lymphoma*	459 (10.0)	$5,587	$96,380	$7,462	$373,492
CNS*	980 (21.3)	$7,910	$115,056	$10,878	$282,738
Other§	1,531 (33.2)	$7,993	$144,911	$14,970	$249,384
Total	4,606 (100)	$6,637	$138,161	$15,756	$316,303

* *International Classification of Childhood Cancer (ICCC), Third Edition (Steliarova-Foucher et al. 2005)*
§ *All other nine ICCC groups*

Costs

For all cancers combined, mean net costs in ON were higher than those in BC, across all phases of care (Table 1). The excess ranged from 1.8% (for the terminal phase) to 39.4% (for the initial phase). In both provinces, the highest mean net costs were in the terminal phase, followed by the initial phase. Pre-diagnosis (60-day) costs represented between 1.6%BC and 2.0%ON of terminal care (360-day) costs; continuing care costs totalled between 4.2%BC and 5.0%ON of terminal care costs.

TABLE 2. Phase-specific mean net costs by resource (Canadian $2012) for British Columbia and Ontario

Resource	Pre-diagnosis	Initial	Continuing	Final
British Columbia				
In-patient hospitalization	$2,146	$70,371	$7,016	$225,939
Physician services	$1,832	$8,610	$847	$46,826
Diagnostic tests and procedures	$626	$4,784	$864	$8,842
Chemotherapy	N/A	$7,476	$1,105	$8,630
Radiotherapy	N/A	$2,677	$285	$6,236
Total*	$4,909	$99,087	$13,133	$310,798
Ontario				
In-patient hospitalization	$4,735	$106,660	$9,943	$260,872
Physician services	$586	$7,201	$1,170	$20,463
Diagnostic tests and procedures	$273	$1,581	$536	$3,120
Chemotherapy	N/A	$13,240	$2,211	$10,519
Radiotherapy	N/A	$2,315	$132	$7,194
Total*	$6,177	$138,161	$1,556	$316,303

**Includes resources listed above plus emergency department visits, same-day surgery, outpatient prescription drugs, home/community care and complex continuing care.*

The ranking of the magnitude of diagnosis-specific costs across the phases was similar in both provinces (Table 1). Pre-diagnosis costs were highest for patients with CNS tumours and lowest for those with leukemia. The initial phase costs were highest for leukemia patients and lowest for patients with lymphomas. For the continuing phase, the highest costs were for patients with leukemia in ON and "other" cancers in BC, but in both provinces, patients with lymphoma had the lowest costs. For the terminal phase, the highest costs were for the leukemia patients in ON and the lymphoma patients in BC, and the lowest costs were for patients with "other" cancers in both provinces.

Higher diagnosis-specific costs were seen in ON than in BC for most phases of care. For leukemia patients, pre-diagnosis costs were 59% higher, initial treatment costs were 46% higher and annual continuing care costs were 48% higher. Terminal phase costs for leukemia patients were 18% lower in ON. For lymphoma patients, the provincial differences were not as large or consistent (ON 3% lower than BC in pre-diagnosis; 18% higher than BC for initial treatment; 53% higher than BC for continuing care; and 42% lower than BC for terminal care). Costs in ON were slightly higher than those in BC for CNS tumour patients; specifically, pre-diagnosis costs were 2% higher, initial treatment costs were 36% higher, continuing care costs were 7% higher and terminal phase costs were 22% higher. Among patients with "other" cancers, pre-diagnosis and initial treatment costs were 44% and 37% higher, respectively, in ON. Continuing care costs were 7% lower in ON, and terminal phase costs were 20% higher in ON.

The distribution of costs by resource varied between the provinces in all phases (Table 2). In-patient costs comprised a larger proportion of total costs in ON: pre-diagnosis (77%ON vs. 43%BC), initial (77%ON vs. 71%BC), continuing (63%ON vs. 52%BC) and terminal (82%ON vs. 73%BC). In contrast, physician services accounted for 7.4%–13.0% of ON costs and 6.4%–37.0% of BC costs, and diagnostic tests and procedures accounted for 1.0%–3.4% of ON costs and 2.8%–13.0% of BC costs. Notably, in the pre-diagnosis phase, 9.5% of ON costs, but 37% of BC costs, were for physician services, but in the continuing phase, 74% of ON costs were for physician services (vs. 6.4% in BC). The mean and proportion of day-surgery costs in continuing care (2.4%ON vs. 17.0%BC) were much lower in ON. The proportion of costs related to chemotherapy was higher in ON than in BC for the initial (9.6%ON vs. 7.5%BC), continuing (14.0%ON vs. 8.4%BC) and terminal (3.3%ON vs. 2.8%BC) phases, but the proportion attributed to radiotherapy was lower in ON than in BC for the initial (1.7%ON vs. 2.7%BC) and continuing (0.8%ON vs. 1.9%BC) phases. Net costs (total costs for cases minus costs for controls) were only slightly lower than total costs for all phases, in BC and ON, indicating that most costs for the cancer cases were cancer-related (Table 3).

TABLE 3. Phase-specific mean total and net costs (Canadian $2012), for childhood cancer cases in British Columbia and Ontario

Phase	British Columbia		Ontario	
	Mean Total Cost	Mean Net Cost	Mean Total Cost	Mean Net Cost
Pre-diagnosis	$4,997	$4,909	$6,637	$6,177
Initial	$99,648	$99,087	$139,453	$138,161
Continuing	$13,607	$13,133	$16,786	$15,756
Terminal	$311,398	$310,798	$319,883	$316,303

Net costs = Total costs for cases minus costs for controls

Discussion

This study compared cancer-attributable (net) childhood cancer medical costs in two Canadian provinces with publicly funded, comprehensive healthcare systems covering virtually all residents. We used the same criteria to select patients and used similar population-based registries and healthcare administrative data sets over the same period. Overall mean phase-specific costs were higher for ON than for BC, mainly because of higher in-patient hospital costs (the major cost category) in ON.

In both provinces, costs were highest in the terminal and initial care phases, reflecting cost patterns observed in adult cancer patients in Canada (de Oliveira et al. 2017a) and the US (Brown et al. 2002). However, costs were much higher for children because of the duration and intensity of childhood cancer treatments (Lanzkowsky et al. 2016). Childhood cancer-related diagnostic procedures, treatment protocols and follow-up care guidelines are highly standardized in North America but vary by diagnosis (Lanzkowsky et al. 2016). Consequently, we observed cost differences by diagnosis within provinces but similar diagnosis-specific costs between provinces.

The large proportion of hospital-based costs in our study is consistent with that reported in previous studies (Luo et al. 2002; Mueller et al. 2017; Rosenman et al. 2005). Most of the hospital visits in both provinces were at tertiary/quaternary centres for oncology care for individuals aged 18 years or younger. These centres would see patients with similar distributions of severity of disease, deliver similar care and have higher CPWC than other hospitals. ON's The Hospital for Sick Children has an estimated CPWC over twice as high as most other ON hospitals (KPMG 2012), and the estimated CPWC for BCCH is almost 60% higher than that for other BC hospitals. Furthermore, higher proportions of ON than BC patients were hospitalized in the initial (4.5% higher) and terminal (7.9% higher) phases, which would increase costs substantially. Despite similar proportions of users of physician services, diagnostic tests and procedures between provinces (data not shown), costs in BC were >50% lower for the initial and terminal phases, which could be related to fewer or less costly encounters per user. Differences in standard practice may partly explain these differences. There are more restrictive criteria for requesting clinical investigations, and more investigations undertaken as outpatients, in BC (personal communication with BC pediatric oncologist co-author [PCR]). There may also be inter-centre variations in patterns of care in

ON. These differences may contribute to lower hospital costs and higher physician services and diagnostic services costs in BC compared to in ON.

This study used geographically defined population-based administrative data sets documenting "medically necessary care" (Madore 2005). The systems in BC and ON are more similar to each other than to the system in Quebec, another large Canadian province, whereas the provinces with smaller populations have fewer well-developed data resources. Data sets included costs for most of the resources covered by provincial governments. This approach provides more accurate and comparable estimates of direct medical care costs than indirect data sources and non-representative populations. The "phase of care" approach is relevant to clinical care and patient experience.

Documentation of cost and utilization patterns of healthcare is fundamental to identifying opportunities to improve practice and policy. The next step is to explore the policy/practice-related and exogenous components of inter-provincial cost variations, including delivery models, resource issues, patient mix and socio-economic and geographic factors. A recent ON report on comparative hospital costs (CIHI 2017) concluded that most cost differences could be explained by exogenous factors or hospital management decisions, related more to system and delivery model factors than to institutional factors (CIHI 2017). More research has yet to be done to explore the results of the BC–ON adult study (de Oliveira et al. 2017a); therefore, no policy or practice changes have followed to date.

Some differences in costs may be owing to reporting artifacts. Some histology and diagnostic tests, counted in the ON claims data, were not recorded in the BC claims database because these are covered by institutional budgets. Some data sources were not comparable between provinces (e.g., ED visits, home care and outpatient prescription drugs). The proportion of patients with ED visits was similar between provinces in the pre-diagnosis and initial phases, whereas the proportion of users of home care varied dramatically between provinces, suggesting that home-care data sources might be measuring very different types of care or patient subgroups (data not shown). Although costs for in-patient hospitalization and physician services were estimated from identical or very similar databases in each province, some cost variation can be attributed to differences in the scope of services covered and recorded, as well as to difficulties in harmonizing data, missing data, changes in data capture over time and variable levels of data to estimate costs.

Time from data set availability to analysis completion resulted in an interval to reporting. However, five-year survival estimates for childhood cancer have been 82%–83% from 1999 onward (CCS 2008; CCS 2017), and the protocols generating these survival rates have not changed appreciably in recent years. We followed patients for up to 15 years after diagnosis, thus capturing costs of late effects of treatment within that time, but studies that tracked survivors of childhood cancer for up to 40 years following the diagnosis found that relative risks for late effects increased as patients aged (Geenen et al. 2007; Hudson et al. 2013; Lorenzi et al. 2011; Oeffinger et al. 2006). Therefore, net continuing care costs may change over time (Luo et al. 2002). Also, caregivers and survivors have many additional ongoing

costs, including out-of-pocket costs relating to loss of work days, treatment-related travel and family supports (Warner et al. 2015).

In conclusion, this large population-based study compared costs of childhood cancer care in two jurisdictions with similar healthcare systems and observed cost variation that was potentially modifiable, once determinants of variation are understood, in particular relating to resource allocation and system efficiencies. This type of analysis should provide useful guidance for other Canadian provinces, all of which operate under the *Canada Health Act*, and other within-country childhood cancer cost evaluations. The significant challenges in conducting cross-national comparative cost studies, in particular because of variation in data measurement, limit our current ability to produce valid comparisons among countries (Lipscomb et al. 2013). Although childhood cancer is rare (<1% all cancers; CCS 2008, 2017), survival in Canada and many Western nations is high (>80% overall) (CCS 2008; Ward et al. 2014) largely because of successful treatment, with many person years of life saved (CCS 2017; Chan and Raney 2005; Ellison et al. 2009). Therefore, strategies and interventions to improve care are likely to be cost effective; however, assessment of effect on patient survival and other outcomes is required to examine this question. Future work should assess costs for additional years in continuing care and update costs to account for newer treatments, to identify additional opportunities for improved system performance. Overall, comparative cost studies highlight differences in healthcare in different jurisdictions, and comparative results can support healthcare research and policy and care improvement.

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References

Austin, P.C. and M.M. Mamdani. 2006. A Comparison of Propensity Score Methods: A Case-Study Estimating the Effectiveness of Post-AMI Statin Use. *Statistics Medicine* 25(12): 2084–106. doi:10.1002/sim.2328.

Austin, P.C. 2011. An Introduction to Propensity Score Methods for Reducing the Effects of Confounding in Observational Studies. *Multivariate Behavioral Research* 46(3): 399–424. doi:10.1080/00273171.2011.568786.

Baker, M.S., L.G. Kessler, N. Urban and R.C. Smucker. 1991. Estimating the Treatment Costs of Breast and Lung Cancer. *Medical Care* 29(1): 40–49. doi:10.1097/00005650-199101000-00004.

Brand, S.R., L. Pickard, J.W. Mack and L.L. Berry. 2016. What Adult Cancer Care Can Learn from Pediatrics. *The Journal of Oncology Practice* 12(9): 765–67. doi:10.1200/JOP.2016.015057.

British Columbia Ministry of Health. 2011a: PharmaNet. V2. Population Data BC. Data Extract. MOH. (2011). Retrieved June 27, 2013. <http://www.popdata.bc.ca/data/health/PharmaNet>.

British Columbia Ministry of Health. 2011b: PharmaCare. V2. Population Data BC. Data Extract. MOH. (2011). Retrieved June 27, 2013. <http://www.popdata.bc.ca/data/health/Pharmacare>.

British Columbia Ministry of Health 2011c: Mental Health. V2. Population Data BC. Data Extract. MOH. (2011). Retrieved June 27, 2013. <http://www.popdata.bc.ca/data/health/mentalhealth>.

British Columbia Ministry of Health 2011d: Medical Services Plan (MSP) Payment Information File. V2. Population Data BC. Data Extract. MOH. (2011). Retrieved June 27, 2013. <http://www.popdata.bc.ca/data/health/msp>.

British Columbia Ministry of Health 2011e: Home & Community Care (Continuing Care). V2. Population Data BC. Data Extract. MOH. (2011). Retrieved June 27, 2013. <http://www.popdata.bc.ca/data/health/hcc>.

British Columbia Ministry of Health 2011f: Consolidation File (MSP Registration & Premium Billing). V2. Population Data BC. Data Extract. MOH. (2011). Retrieved June 27, 2013. <http://www.popdata.bc.ca/data/population/consolidationfile>.

Brown, M.L., G.F. Riley, N. Schussler and R. Etzioni. 2002. Estimating Health Care Costs Related to Cancer Treatment from SEER-Medicare Data. *Medical Care* 40(8 Suppl): IV-104–117. doi:10.1097/00005650-200208001-00014.

Canadian Cancer Society (CCS). 2008. *Canadian Cancer Statistics 2008*. Retrieved March 2, 2018. <https://www.cancer.ca/~/media/cancer.ca/CW/cancer%20information/cancer%20101/Canadian%20cancer%20statistics/Canadian-Cancer-Statistics-2008-EN.pdf?la=en.>

Canadian Cancer Society (CCS). 2017. *Canadian Cancer Statistics 2017.* Retrieved March 2, 2018. <http://www.cancer.ca/~/media/cancer.ca/CW/cancer%20information/cancer%20101/Canadian%20cancer%20statistics/Canadian-Cancer-Statistics-2017-EN.pdf>.

Canadian Institute for Health Information (CIHI). 2005. *Hospital Financial Performance Indicators 1999–2000 to 2002–2003*. Ottawa, ON: CIHI.

Canadian Institute for Health Information (CIHI). 2008. *The Cost of Acute Care Hospital Stays by Medical Condition in Canada, 2004–2005*. Ottawa, ON: CIHI.

Canadian Institute for Health Information (CIHI). 2016. *DAD Resource Intensity Weights and Expected Length of Stay (ELOS).* Ottawa, ON: CIHR. Retrieved March 2, 2018. <https://www.cihi.ca/en/access-data-and-reports>.

Canadian Institute for Health Information (CIHI). 2017. *Understanding Variability in the Cost of a Standard Hospital Stay.* Ottawa, ON: CIHI. Retrieved March 2, 2018. <https://www.cihi.ca/en/access-data-and-reports>.

Carinci F., K., Van Gool, J. Mainz, J. Veillard, E.C. Pichora, J. M. Manuel et al. 2015. Towards actionable international comparisons of health system performance: expert revision of the OECD framework and quality indicators. Retrieved July 4, 2019. *International Journal for Quality in Health Care* 27(2): 137–46. doi: 10.1093/intqhc/mzv004.

Chan, K.W. and R.B. Raney, Jr. 2005. *Pediatric Oncology*. New York, NY: Springer Science+Business Media, Inc.

Cunningham, S. J. 2000. Economic Evaluation of Healthcare – Is It Important to Us? *The British Dental Journal* 188(5): 250–54.

de Oliveira, C., R. Pataky, K.E. Bremner, J. Rangrej, K.K.W. Chan, W.Y. Cheung et al. 2017a. Estimating the Cost of Cancer Care in British Columbia and Ontario: A Canadian Inter-Provincial Comparison. *Healthcare Policy* 12(3): 95–108. doi:10.12927/hcpol.2017.25024.

de Oliveira, C., K.E. Bremner, N. Liu, M.L. Greenberg, P.C. Nathan, M.L. McBride et al. 2017b. Costs of Cancer Care in Children and Adolescents in Ontario, Canada. *Pediatric Blood & and Cancer* 64(11): e26628. doi: 10.1002/pbc.26628.

de Oliveira, C., K.E. Bremner, N. Liu, M.L. Greenberg, P.C. Nathan, M.L. McBride et al. 2017c. Costs for Childhood and Adolescent Cancer, 90 Days Prediagnosis and 1 Year Postdiagnosis: A Population-Based Study in Ontario, Canada. *Value in Health* 20(3): 345–56. doi:10.1016/j.jval.2016.10.010.

de Oliveira, C., S. Weir, J. Rangrej, M.D. Krahn, N. Mittmann, J.S. Hoch et al. 2018. The Economic Burden of Cancer Care in Canada: A Population-Based Cost Study. *CMAJ Open* 6(1): E1–10. doi:10.9778/cmajo.20170144.

Ellison L.F., P. De, L.S. Mery, P.E. Grundy and Canadian Cancer Society's Steering Committee for Canadian Cancer Statistics. 2009. "Canadian Cancer Statistics at a Glance: Cancer in Children". *CMAJ* 180(4): 422–24. doi:10.1503/cmaj.081155.

Geenen, M.M., M.C. Cardous-Ubbink, L.C.M. Kremer, C. van den Bos, H.J.H. van der Pal, R.C. Heinen et al. 2007. Medical Assessment of Adverse Health Outcomes in Long-Term Survivors of Childhood Cancer. *JAMA* 297(24): 2705–15. doi:10.1001/jama.297.24.2705.

Greenberg, M.L., R.D. Barr, B. DiMonte, E. McLaughlin and C. Greenberg. 2003. Childhood Cancer Registries in Ontario, Canada: Lessons Learned from a Comparison of Two Registries. *International Journal of Cancer* 105(1): 88–91. doi:10.1002/ijc.11004.

Hudson, M.M., K.K. Ness, J.G. Gurney, D.A. Mulrooney, W. Chemaitilly, K.R. Krull et al. 2013. Clinical Ascertainment of Health Outcomes among Adults Treated for Childhood Cancer. *JAMA* 309(22): 2371–81. doi:10.1001/jama.2013.6296.

Kim, H.J., M.P. Fay, E.J. Feuer and D.N. Midthune. 2000. Permutation Tests for Joinpoint Regression with Applications to Cancer Rates. *Statistics in Medicine* 19(3): 335–51. doi:10.1002/(SICI)1097-0258(20000215)19:3<335::AID-SIM336>3.0.CO;2-Z.

KPMG. 2012. *SickKids Socio-Economic Impact Study*. Retrieved May 27, 2018. <https://www.sickkids.ca/pdfs/About-SickKids/51294-Project%20Impact%20-%20Final%20Report%20-%20Sept%207%202012.pdf>.

Lanzkowsky, P., J.M. Lipton and J.D. Fish. 2016. *Lanzkowsky's Manual of Pediatric Haematology and Oncology (6th ed)*. San Diego, CA: Academic Press.

Lipscomb J., K.R. Yabroff, M.C. Hornbrook, A. Gigli, S. Francisci, M. Krahn et al. 2013. Comparing Cancer Care, Outcomes, and Costs across Health Systems: Charting the Course. *Journal of the National Cancer Institute Monographs* 2013(46): 124–30. doi:10.1093/jncimonographs/lgt011.

Lorenzi, M.F., L. Xie, P.C. Rogers, S. Pritchard, K. Goddard and M.L. McBride. 2011. Hospital-Related Morbidity among Childhood Cancer Survivors in British Columbia, Canada: Report of the Childhood, Adolescent, Young Adult Cancer Survivors (CAYACS) Program. *International Journal of Cancer* 128(7): 1624–31. doi:10.1002/ijc.25751.

Luo, W., R. Lane, K. Stobart, H. Morrison, D. Schanzer, R. Barr et al. 2002. The Medical Care Cost of Childhood and Adolescent Cancer in Manitoba, 1990–1995. *Chronic Diseases in Canada* 23(3): 83–90.

Madore, O. 2005. *The Canada Health Act: Overview and Options*. Retrieved August 13, 2018. <https://www.researchgate.net/publication/237773541_The_Canada_Health_Act_Overview_and_options>.

Mueller, E.L., M. Hall, J.G. Berry, A.E. Carroll and M.L. Macy. 2017. Healthcare Utilization and Spending by Children With Cancer on Medicaid. *Pediatric Blood and Cancer* 64(11): e26569. doi:10.1002/pbc.26569.

Nathan, P.C., K.E. Bremner, N. Liu, S. Gupta, M.L. Greenberg, M.L. McBride et al. 2019. Resource Utilization and Costs in Adolescents Treated for Cancer in Pediatric vs Adult Institutions. *Journal of the National Cancer Institute* 111(3): 322–30. doi:10.1093/jncl/djy119.

Oeffinger, K.C., A.C. Mertens, C.A. Sklar, T. Kawashima, M.M. Hudson, A.T. Meadows et al. 2006. Chronic Health Conditions in Adult Survivors of Childhood Cancer. *New England Journal of Medicine* 355(15): 1572–82. doi:10.1056/NEJMsa060185.

Price, R.A., E. Stranges and A. Elixhauser. 2012. Pediatric Cancer Hospitalizations, 2009: Healthcare Cost and Utilization Project Statistical Brief #132. Rockville, MD: *Agency for Healthcare Research and Quality.* Retrieved July 4, 2019. <https://www.ncbi.nlm.nih.gov/pubmed/22787680>.

Reid, R.J., L. MacWilliam, L. Verhulst, N. Roos and M. Atkinson. 2001. Performance of the ACG Case-Mix System in Two Canadian Provinces. *Medical Care* 39(1): 86–99. doi:10.1097/00005650-200101000-00010.

Robinson, R. 1993. Economic Evaluation and Healthcare. What Does It Mean? *BMJ (Clinical research ed)* 307(6905): 670–73. doi:10.1136/bmj.307.6905.670.

Rosenman, M.B., T. Vik, S.L. Hui and P.P. Breitfeld. 2005. Hospital Resource Utilization in Childhood Cancer. *Journal of Pediatric Hematology/Oncology* 27(6): 295–300. doi:10.1097/01.mph.0000168724.19025.a4.

Starfield, B. and K. Kinder. 2011. Multimorbidity and Its Measurement. *Health Policy* 103(1): 3–8. doi:10.1016/j.healthpol.2011.09.004.

Statistics Canada. 2013. "Postal CodeOM Conversion File (PCCF)." Retrieved May 27, 2018. <https://www150.statcan.gc.ca/n1/en/catalogue/92-154-X>.

Statistics Canada. 2017. "Distribution of Total Income, by Family Type, by Province and Territory (All Census Families)." Retrieved March 2, 2018. <https://www150.statcan.gc.ca/t1/tbl1/en/tv.action?pid=1110001201>.

Steliarova-Foucher, E., C. Stiller, B. Lacour and P. Kaatsch. 2005. International Classification of Childhood Cancer, Third Edition. *Cancer* 103(7): 1457–67. doi:10.1002/cncr.20910.

Tarricone, R. 2006. Cost-of-Illness Analysis: What Room in Health Economics? *Health Policy* 77: 51–63. doi:10.1016/j.healthpol.2005.07.016.

Ward, E., C. DeSantis, A. Robbins, B. Kohler and A. Jemal. 2014. Childhood and Adolescent Cancer Statistics, 2014. *Cancer Journal for Clinicians* 64(2): 83–103. doi:10.3322/caac.21219.

Warner, E.L., A.C. Kirchhoff, G.E. Nam and M. Fluchel. 2015. Financial Burden of Pediatric Cancer for Patients and Their Families. *Journal of Oncology Practice* 11(1): 12–18. doi:10.1200/JOP.2014.001495.

Wodchis, W.P., K. Bushmeneva, M. Nikitovic and I. McKillop. 2013. *Guidelines on Person-Level Costing Using Administrative Databases in Ontario*. Retrieved August 1, 2018. <https://tspace.library.utoronto.ca/handle/1807/87373>.

Yabroff, K.R., E.B. Lamont, A. Mariotto, J.L. Warren, M. Topor, A. Meekins et al. 2008. Cost of Care for Elderly Patients in the United States. *Journal of the National Cancer Institute* 100(9): 630–41. doi:10.1093/jnci/djn103.

RESEARCH PAPER

An Exploratory Analysis of Predictors of Concordance between Canadian Common Drug Review Reimbursement Recommendations and the Subsequent Decisions by Ontario, British Columbia and Alberta

Analyse exploratoire des prédicteurs de la concordance entre les recommandations de remboursement du Programme commun d'évaluation des médicaments et les décisions subséquentes de l'Ontario, de la Colombie-Britannique et de l'Alberta

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Abstract

Background: Concordance between Common Drug Review (CDR) recommendations and provincial plans has been studied previously. However, no study has, to the best of the authors' knowledge, examined the characteristics of CDR recommendations that may be associated with concordance.

Methods: Recommendation–decision pairs were collected from the CDR and the provincial plans of Ontario, British Columbia and Alberta. Concordance was evaluated by province. Characteristics of each CDR recommendation were collected, and associations with concordance were evaluated by logistic regression.

Results: Recommendation–listing concordance was high. Positive references to cost and clinical outcomes compared to placebo were statistically associated with concordance. Negative references to cost and to the consistency and certainty of economic evidence were statistically associated with discordance. However, these findings were inconsistent across the jurisdictions studied.

Conclusions: Although concordance was high, the ability of recommendation characteristics to explain the relationship between province and CDR listing decisions was limited. This exploratory study highlights the complexity of the reimbursement process and possible reasons for drug listing differences across jurisdictions.

Résumé

Contexte : La concordance entre les recommandations du Programme commun d'évaluation des médicaments (PCEM) et les régimes provinciaux a déjà fait l'objet d'études. Cependant, à la connaissance des auteurs, aucune étude n'a encore examiné les caractéristiques des recommandations du PCEM qui peuvent être associées à la concordance.

Méthode : Des paires recommandation–décision ont été recueillies auprès du PCEM et des régimes provinciaux de l'Ontario, de la Colombie-Britannique et de l'Alberta. La concordance a été évaluée selon les provinces. Les caractéristiques de chaque recommandation du PCEM ont été recueillies et les liens avec la concordance ont été évalués au moyen de la régression logistique.

Résultats : La concordance des listes de recommandations était élevée. Les références positives aux coûts et aux résultats cliniques par rapport au placebo étaient statistiquement associées à une concordance. Les références négatives aux coûts, à la cohérence et à la certitude des données économiques étaient statistiquement associées à une discordance. Cependant, ces résultats n'étaient pas uniformes parmi les provinces étudiées.

Conclusion : Bien que la concordance soit élevée, les caractéristiques des recommandations expliquent de façon limitée la relation entre une province et les décisions d'inscription au PCEM. Cette étude exploratoire met en évidence la complexité du processus de remboursement et les raisons possibles des différences entre les listes de médicaments d'une province à l'autre.

Introduction

The reimbursement process for publicly funded drug plans in Canada is complex, with multiple decision-makers acting across several jurisdictions. Reimbursement evaluations are conducted under the framework of health technology assessment, defined by the World Health Organization (2018) as the "systematic evaluation of properties, effects and/or impacts of health technologies and interventions". This multidisciplinary holistic approach broadens the evidence base to include medical, economic, social, organizational and ethical aspects of current and emerging technologies. Canada, like many European countries, has formally adopted health technology assessment practices as a means of informing decision-makers for drug reimbursements on public drug plans (Angelis et al. 2018; Mitton et al. 2006).

Until recently, the Common Drug Review (CDR) process, housed in the Canadian Agency for Drugs and Technologies in Health (CADTH), typically began once Health Canada approved a drug as safe for use. In June 2018, it was announced that Health Canada, CADTH and Quebec's Institut national d'excellence en santé et en services sociaux will align their respective drug review processes under Health Canada's Regulatory Review of Drugs and Devices initiative. Yet, the purpose of the review process remains to evaluate whether a drug qualifies for public payer reimbursement based on evidence of the drug's clinical and economic properties, alongside input from patient groups. Reimbursement decisions are issued and disseminated to the 18 participating drug plans delivered by federal, provincial and territorial ministries and agencies. However, the recommendation is non-binding and the final listing decision is at the discretion of the plan administrator, with funding allocated in the context of their unique jurisdictional mandates and priorities.

Given that CDR recommendations are non-binding, the final reimbursement decisions by participating drug plans may vary. Several studies have examined Canadian drug reimbursement decision concordance, both before and after the establishment of the CDR (Allen et al. 2016; Anis et al. 2001; Attaran et al. 2011; Gamble et al. 2011; MacDonald and Potvin 2004; Morgan et al. 2006; Morgan et al. 2009). However, the findings and conclusions from this body of evidence vary because of variations in study methodology, the jurisdictions represented and the period being studied.

Reimbursement submissions have also been studied to explore factors that may predict listing recommendation outcomes. For example, a study by Rocchi and colleagues examined factors associated with negative recommendations (Rocchi et al. 2012). Through univariate and multivariate analyses, the authors concluded that clinical uncertainty and price considerations, but not the findings from economic evaluations, were strongly predictive of listing recommendations. Similar research has been conducted in other reimbursement contexts, such as by Linley and Hughes, who examined the influence of policy and clinical and economic factors in Wales (Linley and Hughes 2012). Based on multivariate analysis, it was observed that the use of probabilistic sensitivity analyses to examine uncertainty positively influenced listing decisions. Interestingly, citing high-quality randomized controlled trials as

supporting clinical evidence was statistically significantly predictive of negative listing decisions. This finding raised questions about the perception of study quality in the context of decision-making, particularly with respect to the use of surrogate end points.

Although concordance and reasons for listing recommendations or decisions have been explored separately in the literature, no study has, to the best of the authors' knowledge, examined potential predictors of concordance between the CDR recommendations and Canadian provincial listing decisions. Thus, the first objective of our study is to describe the concordance between CDR recommendations and the listing decisions of the provinces of Ontario, British Columbia and Alberta. Beyond describing concordance between the CDR and provincial decisions, there is some value to investigators seeking formulary listings to make explicit the key elements that are associated with a decision outcome. Thus, the second objective is to propose predictors of concordance based on the published recommendation rationales issued by the CDR.

Methods

Study design

A database of CDR recommendations (2009–2017), including the listing recommendation, brand and generic drug name, indication, submission type and the recommendation date, was procured from the CADTH website (Canadian Agency for Drugs and Technologies in Health 2018). For the purpose of this study, only the initial drug submissions were considered (i.e., not re-submissions). The list of CDR recommendations was cross-referenced with the listing decisions by the provincial drug plan administrators of the Ontario Public Drug Programs (PDP) (Ontario Ministry of Health and Long-Term Care 2018), British Columbia PharmaCare (Government of British Columbia 2018) and the Alberta Drug Benefit List (DBL) (Alberta Health 2018) based on the intervention, indication and CDR recommendation date. The CDR recommendation date must have preceded the date of the provincial decision. We selected these provinces because, together, these represent approximately 80% of the Canadian population living in jurisdictions that participate in the CDR process. Recommendations were coded as "positive" irrespective of whether those were conditional (Allen et al. 2016). Similarly, provincial listing decisions were considered positive irrespective of limitations or special authorization requirements. Oncology drugs, which are evaluated through the separate pan-Canadian Oncology Drug Review process, were not considered.

A list of predictors reflecting various aspects of clinical evidence, economic evidence and evidence synthesis methods was developed by reviewing a sample of 25 CDR recommendation decisions purposefully selected to reflect a range of years, interventions and indications. Through a thematic analysis, novel reasons for recommendations were extracted from subsequent recommendation documents until no new themes emerged. Then, for each treatment–indication pair, the "Reasons for recommendation" section of published CDR recommendations was evaluated and predictors categorized as follows: "Present, in favour

of a positive recommendation"; "Present, in favour of a negative recommendation"; or "Not referenced".

All data were collected by a single reviewer (MZ) and maintained in Microsoft Excel workbooks. Analyses were conducted in R (v3.4.3, r-project.org).

Statistical analysis

For each province, concordance between CDR recommendations and provincial listing decisions was described first by the percentage of positive recommendations and decisions. The crude percentage of recommendations and decisions in concordance was then estimated and supplemented by Cohen's unweighted kappa coefficient to account for the possibility of agreement occurring by chance (Cohen 1960). Kappa coefficients were interpreted according to the guideline proposed by Landis and Koch (1977). Missing data (i.e., provincial listing decisions) were not imputed.

For each predictor, a binomial logistic regression model was fit to the data for each province–CDR paired set to estimate the odds ratio (OR) of concordance between the recommendation and listing decision when the predictor was present. The positive and negative predictor levels were evaluated separately against the reference category of the predictor being "Not referenced". Outcomes were interpreted as follows: Compared to when the predictor is not referenced, ORs greater than 1 and less than 1 are predictive of concordance and discordance, respectively, between the CDR and the province. For a predictor to be evaluated, we specified *a priori* that 10 observations, either in favour of a positive or negative recommendation, must be available in a given province–CDR paired set. Statistical significance was evaluated at the $\alpha = 0.05$ level.

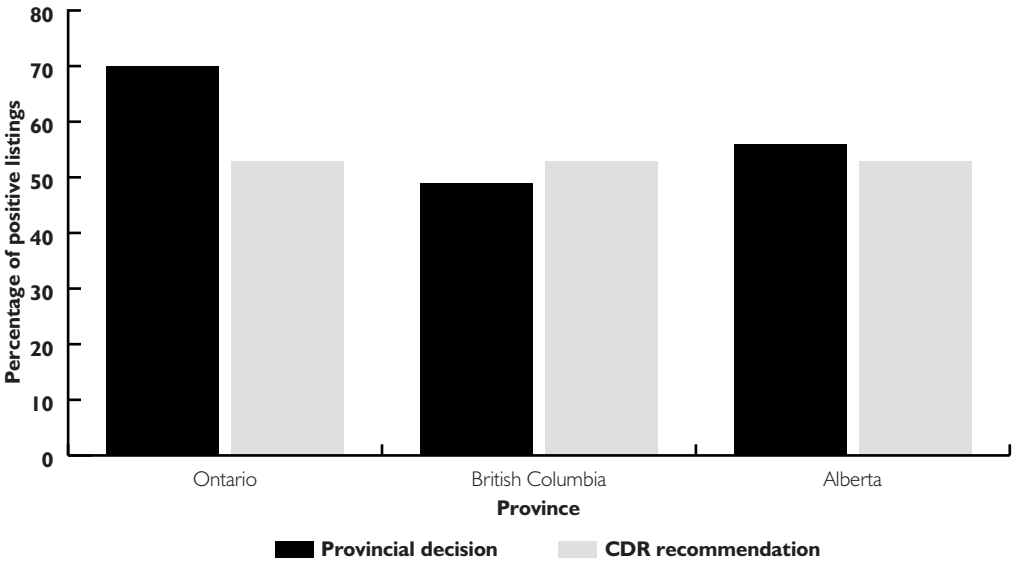
In cases where every observation has the same outcome, it is not possible to evaluate the province–CDR relationship by logistic regression. In these instances, a descriptive analysis was supplemented by Fisher's "exact test" to estimate the probability that this association was observed by chance (Fisher 1922).

Results

Common Drug Review recommendations

A database of 309 CDR recommendations, from January 28, 2009, to November 23, 2017, was available. Of these, 193 were identified as "New" submissions, with 125 (64.8%) positive listing recommendations (Figure 1). Over the time interval studied, the percentage of assessments with a conditional recommendation increased, from a low of 28.6% (2/7) of recommendations in 2009 to all recommendations in 2013 (8/8) through 2014 (17/17). Nearly all recommendations issued in 2017 included some condition (95.5%, 21/22). A complete list of recommendations and corresponding provincial decisions is presented in Appendix 1 (available online at longwoods.com/content/26128).

FIGURE 1. Percentage of positive Common Drug Review (CDR) listing recommendations and positive provincial listing decisions

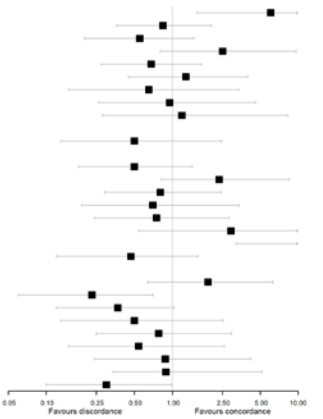


From the set of 25 CDR recommendations selected and used to generate predictors, 13 predictors were initially proposed. Data saturation achieved after 20 recommendations was reviewed. These predictors, with definitions and examples extracted from CDR recommendations, are presented in Appendix 2 (available online at longwoods.com/content/26128). Across the complete set of CDR recommendations, the median (interquartile range) number of predictors, positive or negative, was 2 (2, 3). From the 13 initial predictors, 5 were excluded from the analysis for having fewer than 10 references in either the positive or negative direction: clinical outcomes from non-comparative trials; replacement vs. currently available treatments; patient subgroup-specific evidence; evidence synthesis methods, only citing use of a network meta-analysis; and evidence synthesis methods, other than network meta-analysis. Many predictors were only evaluable at only the positive (clinical outcomes vs. placebo; safety or tolerability) or negative (consistency or certainty in clinical evidence; clinical methods; incremental cost-effectiveness ratio; consistency or certainty in economic evidence) level. The pattern of evaluability was similar across provinces. Appendix 3 (available online at longwoods.com/content/26128) presents the predictors as coded in CDR recommendations. A summary count of each predictor, arranged by province, is presented in Appendix 4 (available online at longwoods.com/content/26128).

A summary of the results of the logistic regressions, by predictor level and province, is presented in Figure 2.

FIGURE 2. Findings of logistic regressions to evaluate the ability of predictors to determine concordance between CDR recommendations and provincial listing decisions

	Predictor	Level	Province	Observations	Odds Ratio (95% CI)
Clinical factors	Clinical outcomes vs Placebo	Positive	ON	39	6.03 (1.57, 39.83)
			BC	53	0.84 (0.36, 2.05)
			AB	51	0.55 (0.20, 1.48)
	Clinical outcomes vs Active comparator	Positive	ON	40	2.51 (0.80, 9.60)
			BC	50	0.68 (0.27, 1.70)
			AB	48	1.28 (0.45, 3.98)
		Negative	ON	10	0.65 (0.15, 3.37)
			BC	18	0.95 (0.26, 4.56)
			AB	15	1.19 (0.28, 8.29)
	Safety or tolerability	Positive	ON	8	-
			BC	10	0.50 (0.13, 2.46)
			AB	9	-
	Consistency or certainty in clinical evidence	Negative	ON	31	0.50 (0.18, 1.44)
			BC	39	2.35 (0.82, 8.47)
			AB	37	0.80 (0.29, 2.44)
	Clinical methods	Negative	ON	13	0.70 (0.19, 3.39)
			BC	18	0.75 (0.24, 2.83)
			AB	17	2.91 (0.54, 54.11)
Economic factors	Cost	Positive	ON	38	17.76 (3.20, 333.79)
			BC	48	0.47 (0.12, 1.60)
			AB	45*	-
		Negative	ON	30	1.92 (0.64, 6.28)
			BC	48	0.23 (0.06, 0.70)
			AB	39	0.37 (0.12, 1.03)
	Incremental cost effectiveness ratio	Negative	ON	10	0.50 (0.13, 2.53)
			BC	18	0.78 (0.25, 2.94)
			AB	13	0.54 (0.15, 2.59)
	Consistency or certainty in economic evidence	Negative	ON	15	0.88 (0.24, 4.17)
			BC	18	0.89 (0.34, 5.15)
			AB	20	0.30 (0.10, 0.98)



Ontario

A review of the Ontario PDP database returned 105 listing decisions (70.5% positive) that corresponded to CDR recommendations. However, several drug list decisions (n = 34) were unavailable because of errors in the provincial database. Despite numerous attempts to contact provincial administrators, we were unable to obtain these data. Crude agreement between recommendations and listing decisions was estimated at 81.9%. Cohen's unweighted kappa was estimated at 0.625 ($p < 0.001$), indicating substantial agreement.

Positive references to cost (OR = 17.76) and to clinical outcomes compared to placebo (OR = 6.03) were statistically significantly associated with concordance. No negative references to predictors were statistically associated with concordance or discordance.

British Columbia

From the BC PharmaCare database, we identified 140 listing decisions (55.7% positive) corresponding to CDR recommendations. Crude agreement between recommendations and listing decisions was estimated at 81.4%. Cohen's unweighted kappa was estimated at 0.629 ($p < 0.001$), indicating substantial agreement.

Negative references to cost (OR = 0.23) were statistically significantly associated with discordance between CDR and BC PharmaCare. No other predictors were statistically significant.

Alberta

A review of the Alberta DBL returned 134 corresponding listing decisions (58.2% positive). Crude agreement between recommendations and listing decisions was estimated at 85.8%. Cohen's unweighted kappa was estimated at 0.71 ($p < 0.001$), indicating substantial agreement.

In all 45 observations of positive references to cost, we observed concordance between the CDR recommendation and the provincial listing decision. The probability of observing this by chance was evaluated by Fisher's "exact test" ($p < 0.05$). Negative references to consistency or certainty in the economic evidence (OR = 0.30) were statistically significantly associated with discordance between CDR recommendations and Alberta DBL listing decisions. No other predictors were statistically significant.

Interpretation

The purpose of this exploratory study was to examine concordance between CDR reimbursement recommendations and the subsequent decisions by three of the largest participating provincial plans as well as to explore possible predictors of concordance. Provincial concordance with recommendations was high across provinces. However, the ability of predictors to explain this relationship varied, both with respect to the type and direction of the predictor. Positive references to cost were statistically associated with concordance for Ontario and Alberta. Positive references to clinical outcomes compared to placebo were statistically associated with concordance for Ontario only. Negative references to cost were associated with discordance for British Columbia, whereas negative references to consistency or certainty in economic evidence were associated with discordance for Alberta. Interestingly, based on the evaluations by logistic regression, no positive reference to a predictor was statistically associated with discordance and no negative reference was statistically associated with concordance. This suggests that favourable evidence may be more likely to transcend jurisdictional boundaries, whereas negative evidence may need to be more closely examined in the reimbursement context. However, as many predictors were unevaluable because of the small number of observations and trends were largely inconsistent across jurisdictions, this inference is based on limited evidence.

The independent variables that met our *a priori* threshold for analysis were those that we anticipated as being particularly relevant to the decision-making process, namely, elements of clinical performance and cost. Generally, these considerations are the primary focus of much of the research leading to a decision to apply for reimbursement and, in some cases, are necessary to greenlight next steps in the market access pathway. For example, issues with safety, tolerability or unfavourable outcomes against a placebo may stall or terminate a drug's development because, under the current Canadian reimbursement framework, approval by Health Canada is required before seeking reimbursement. In addition, given that CDR recommendation rationales are short summaries, it would be unexpected to have reviewers comment positively on the consistency of clinical or economic evidence or on the methods of the supporting clinical research. Typically, we expected that reviewers would reserve comments on these factors for instances where evidence was unfavourable. Yet, given the relatively few statistical associations we observed, the summary rationale presented to support the CDR recommendation statement does not appear to sufficiently capture the decision-making

process, and the elements highlighted by the CDR may not be the primary driving factors at the provincial level. Future work may explore this by examining correlations between CDR recommendation justifications and the decisions provided in provincial listings.

Alignment of listing decisions in the Canadian context has been studied extensively, and conclusions have proven to be divisive (Allen et al. 2016; Anis et al. 2001; Attaran et al. 2011; Gamble et al. 2011; MacDonald and Potvin, 2004; Morgan et al. 2006; Morgan et al. 2009). Based on CDR recommendations from 2009 to the end of 2014, Allen and colleagues observed concordance rates with the CDR of 81.1%, 78.9% and 78.8% for British Columbia, Alberta and Ontario, respectively (Allen et al. 2016). With the exception of Alberta, we observed similar rates in the current study. However, whereas the current study was based only on new drug submissions, Allen and colleagues sourced the most recent recommendation listing. Gamble and colleagues examined agreement before and after CDR's implementation, collecting data from May 1999 to May 2009 (Gamble et al. 2011). Based on a set of 53 CDR recommendations, agreement varied from a low of 64.2% with Ontario to a high of 90.6% with New Brunswick and Nova Scotia, with concordance estimated at 69.8% and 83.2% for British Columbia and Alberta, respectively. Writing on the effectiveness of the CDR in Canada's national drug strategy, Attaran and colleagues criticized the considerable variability across the country based on a set of 369 pairwise observations (Attaran et al. 2011). Thus, there is a lack of consensus on the ability of the CDR process to standardize drug reimbursements across Canada's public payer drug plans.

Indeed, the establishment of the CDR in 2003 was heralded as a means of consolidating expertise and funding to support health technology assessments through a rigourous, evidence-based, centralized process. Yet, controversy on the usefulness of the process persists in the literature, given that individual drug plans ultimately have the decision to list or not list a drug irrespective of the CDR recommendations (Morgan et al. 2009). Arguably, the establishment of the CDR process particularly benefitted smaller provinces, such as those in Atlantic Canada, which may lack the resources and funding to fully execute health technology assessments on every new drug submission. Larger provinces, such as Ontario, may be more able to fund independent reviews specific to their jurisdictional context. Moreover, decision-makers representing larger patient populations may be afforded more flexibility in drug price negotiations. Ultimately, limitations on funding and patient needs decide whether recommendations are adhered to (Spitz 2013).

The reorganization of the Canadian reimbursement recommendation processes under Health Canada's Regulatory Review of Drugs and Devices initiative has the potential to further enhance the comprehensiveness and rigour by which submissions are assessed. However, the final reimbursement decision will remain with the individual public drug plans. Future research may examine differences in the rates of positive or negative recommendations and the reasons cited for these recommendations under this revised, integrated framework.

Strengths and limitations

The current study is characterized by several strengths. Observations of concordance were made based on a large, valid and directly sourced sample of CDR recommendations using robust methods, including both crude concordance and Cohen's kappa. Although missing data, specifically provincial listing decisions, were not imputed, the cause of missingness was assessed as either outside the jurisdiction of the provincial funding body being queried or that a decision had not yet been reached at the provincial level. The evaluation of predictors was conducted using robust statistical methods and was based on systematically collected evidence. However, there are several limitations to this work. This was an exploratory study, intended to be hypothesis generating rather than hypothesis testing, and thus, it is not appropriate to draw causal conclusions based on the data or analyses. In addition, given the number of analyses performed, it is reasonable to expect at least some relationships to generate spurious statistically significant results. Although the number of CDR recommendations identified was large, the number of predictor observations, either positive or negative, was comparatively small and fell below the *a priori* specified threshold in many cases. The predictors were restricted to the "Reasons for Recommendation" section of the CDR rationale, as the primary factors driving recommendations are highlighted here. Future studies may expand the scope to explore factors such as indication, first-in-class, year of submission and whether recommendations were conditional. Given that predictors were rarely referenced in isolation, interactions between predictors should be considered, as this may reveal important combinations of predictors that may better explain (dis)concordance. The jurisdictions studied, although representing the majority of patients in provinces participating in the CDR process, were restricted to three provinces. Evidence suggests differences in the level of concordance between large and small provinces, which may impact the evaluation of predictors of concordance. Nevertheless, the findings of the current study are valid with respect to the provinces studied, and we do not intend to extend these inferences beyond the jurisdictions specified here.

Conclusion

Previous studies have examined reasons for listing decisions, but this is, to our knowledge, the first study to examine factors for concordance across different decision-maker contexts. We observed substantial concordance between CDR recommendations and the provinces of Ontario, British Columbia and Alberta. In the predictor analysis, it was observed that positive references to cost (Ontario, Alberta) and clinical outcomes compared to placebo (Ontario) were statistically associated with concordance. Negative references to cost (British Columbia) and to the consistency and certainty of economic evidence (Alberta) were statistically associated with discordance between CDR recommendations and provincial listings. However, these findings were not consistent across the jurisdictions studied. Moreover, the exploratory nature of this study precludes the ascertainment of causal relationships between predictors and province–CDR concordance.

This study was motivated by the patchwork of drug coverage that persists in Canada, with inconsistent listing decisions across the country despite receiving a common CDR recommendation. Although the current study failed to reveal clear explanations for concordance, future work may describe trends, particularly with respect to the interplay of various predictors, that may help investigators preparing reimbursement submissions anticipate outcomes and prepare a better case when evidence to support important predictors is lacking. In addition, qualitative studies with provincial decision-makers may reveal motivations that are not sufficiently captured in the publicly available documentation.

Drug reimbursement decision-making is a complex and multidimensional process, and decision-makers must balance multiple forms of evidence, stakeholders and competing funding priorities. Thus, this study highlights the difficulty and limitations of presenting a simple explanation of a complex process.

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References

Alberta Health. 2018. *Interactive Drug Benefit List (iDBL)*. Retrieved March 7, 2018. <https://www.ab.bluecross.ca/dbl/publications.html>.

Allen, N., S.R. Walker, L. Liberti, C. Sehgal and M.S. Salek. 2016. Evaluating Alignment between Canadian Common Drug Review Reimbursement Recommendations and Provincial Drug Plan Listing Decisions: An Exploratory Study. *CMAJ Open* 4(4): E674–78. doi:10.9778/cmajo.20160006.

Angelis, A., A. Lange and P. Kanavos. 2018. Using Health Technology Assessment to Assess the Value of New Medicines: Results of a Systematic Review and Expert Consultation across Eight European Countries. *The European Journal of Health Economics* 19(1): 123–52. doi:10.1007/s10198-017-0871-0.

Anis, A.H., D. Guh and X. Wang. 2001. A Dog's Breakfast: Prescription Drug Coverage Varies Widely across Canada. *Medical Care* 39(4): 315–26. doi:10.1097/00005650-200104000-00003.

Attaran, A., R. Cartagena and A. Taylor. 2011. *The Effectiveness of the Common Drug Review in Canada's National Drug Strategy*. Halifax, NS: Atlantic Institute for Market Studies.

Canadian Agency for Drugs and Technologies in Health. 2018. *CADTH Common Drug Review (CDR) Reports*. Retrieved December 14, 2017. <https://cadth.ca/about-cadth/what-we-do/products-services/cdr/reports>.

Cohen, J. 1960. A Coefficient of Agreement for Nominal Scales. *Educational and Psychological Measurement* 20(1): 37–46. doi:http://dx.doi.org/10.1177/001316446002000104.

Fisher, R.A. 1922. On the Interpretation of χ^2 from Contingency Tables, and the Calculation of P. *Journal of the Royal Statistical Society* 85(1): 87–94. doi:10.2307/2340521.

Gamble, J.M., D.L. Weir, J.A. Johnson and D.T. Eurich. 2011. Analysis of Drug Coverage before and after the Implementation of Canada's Common Drug Review. *CMAJ* 183(17): E1259–66. doi:10.1503/cmaj.110670.

Government of British Columbia. 2018. PharmaCare Drug Review Results. Retrieved March 7, 2018. <https://fmdb.hlth.gov.bc.ca/>.

Landis, J.R. and G.G. Koch. 1977. The Measurement of Observer Agreement for Categorical Data. *Biometrics* 33(1): 159–74. doi:10.2307/2529310.

Linley, W.G. and D.A. Hughes. 2012. Reimbursement Decisions of the All Wales Medicines Strategy Group: Influence of Policy and Clinical and Economic Factors. *Pharmacoeconomics* 30(9): 779–94. doi:10.2165/11591530-000000000-00000.

MacDonald, K. and K. Potvin. 2004. Interprovincial Variation in Access to Publicly Funded Pharmaceuticals: A Review Based on the WHO Anatomical Therapeutic Chemical Classification System. *Canadian Pharmacists Journal* 137(7): 29–34. doi:10.1177/171516350413700703.

Mitton, C.R., M. McMahon, S. Morgan and J. Gibson. 2006. Centralized Drug Review Processes: Are They Fair? *Social Science and Medicine* 63(1): 200–11. doi:10.1016/j.socscimed.2005.11.049.

Morgan, S., G. Hanley, C. Raymond and R. Blais. 2009. Breadth, Depth and Agreement among Provincial Formularies in Canada. *Healthcare Policy* 4(4): e162–84. doi:10.12927/hcpol.2009.20685.

Morgan, S.G., M. McMahon, C. Mitton, E. Roughead, R. Kirk, P. Kanavos et al. 2006. Centralized Drug Review Processes in Australia, Canada, New Zealand, and the United Kingdom. *Health Affairs* 25(2): 337–47. doi:10.1377/hlthaff.25.2.337.

Ontario Ministry of Health and Long-Term Care. 2018. *Drug Submissions: Status for Single-Source Submissions*. Retrieved March 7, 2018. <http://www.health.gov.on.ca/en/pro/programs/drugs/status_single_source_subm.aspx>.

Rocchi, A., E. Miller, R.B. Hopkins and R. Goeree. 2012. Common Drug Review Recommendations: An Evidence Base for Expectations? *Pharmacoeconomics* 30(3): 229–46. doi:10.2165/11593030-000000000-00000.

Spitz, S. 2013. A Decade of the Common Drug Review. *CMAJ* 185(7): 554. doi:10.1503/cmaj.109-4443.

World Health Organization. 2018. *WHO Definition (EB 134/30)*. Retrieved March 12, 2018. <https://www.who.int/health-technology-assessment/about/Defining/en/>.

RESEARCH PAPER

First Ready, First to Go: Ethical Priority-Setting of Allogeneic Stem Cell Transplant at a Major Cancer Centre

Premier prêt, premier parti : établissement des priorités en matière d'éthique dans le cas de greffe de cellules souches allogéniques dans un grand centre d'oncologie

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Abstract

Medical advancements have now made it possible to provide allogeneic stem cell transplantation (allo-SCTs) to older patients and use stem cells from less well-matched donors. This has resulted in access to a life-saving modality for a greater number of patients with imminent life-threatening illnesses. However, resources have not always kept pace with innovation and expanded volumes. During the summer of 2015 in the province of Ontario, Canada, inadequate resources contributed to a capacity crisis, resulting in extended wait-lists for allo-SCT across the province. This situation presented unique ethical challenges, including the need for ongoing negotiations with health system partners and nimble process management to ensure timely delivery of care. This article reports on the process one organization used to determine how to equitably allocate scarce allo-SCT resources. With the ever-expanding landscape of new and emerging medical technologies, our experience has implications for the ethics of translating other increasingly expensive health technologies to clinical care.

Résumé

Les avancées médicales permettent aujourd'hui d'effectuer des greffes de cellules souches (allogreffe-CS) chez les patients plus âgés et d'utiliser moins de greffons de la part de donneurs qui, eux, peuvent être moins compatibles. Cela permet de sauver la vie d'un plus grand nombre de patients aux prises avec des maladies menaçant leur pronostic vital. Toutefois, les ressources ne suivent pas toujours le rythme des innovations ou l'expansion du nombre de cas. Au cours de l'été 2015, en Ontario, au Canada, des ressources inadéquates ont mené à une crise des capacités qui a donné lieu à une longue liste d'attente pour les allogreffes-CS dans la province. La situation présentait des défis d'ordre éthique particuliers, notamment le besoin de négociations continues entre les partenaires du système de santé et une gestion adroite des processus afin d'assurer une prestation de soins en temps opportun. Cet article fait état du processus mis en place dans une organisation afin de déterminer comment distribuer équitablement les rares ressources pour les allogreffes-SC. Avec l'abondance des nouvelles technologies émergentes, notre expérience a des répercussions sur l'éthique d'apporter, dans les soins cliniques, de nouvelles technologies de soins de santé toujours plus onéreuses.

Introduction

Among hematopoietic malignancies, patients with acute myeloid leukemia (AML) represent approximately half of the potential candidates for allogeneic stem cell transplantation (allo-SCT), in which the diseased bone marrow is replaced with the bone marrow from a healthy sibling or matched donor (Gratwohl et al. 2015). Recent medical and technological advancements in this area have allowed older populations to become candidates for allo-SCT and for the accommodation of less well-matched donors. This has led to an expanded volume of eligible patients. In Ontario, Canada, however, government funding for hospital

programs responsible for allo-SCT has not kept pace with these innovations and the increasing demand. This resource shortcoming contributed to a capacity crisis and resulted in a wait-list that made timely disease management difficult. The result was that patients on the wait-list became at risk of missing the opportunity for a potentially curative transplantation for their disease because of relapse or other medical co-morbidities thus exposing them to further medical complications, including death. While the province was developing a plan for the injection of resources, there arose the opportunity to send patients out of the country to receive allo-SCT. Each Ontario centre was confronted with the ethical challenge of managing wait-lists appropriately and deciding what criteria should be used to assign priority for transplantation at their centre.

Background

AML is a common form of leukemia among adults in Western countries. The median age of diagnosis is 65 years (Deschler and Lubbert 2006) and continues to increase (El Rassi and Arellano 2013). AML is a medically complex disease with multifaceted risk factors. Treatments for AML are labour- and resource-intensive, whether low-dose chemotherapy, best supportive care/symptom management or transplantation. Most patients require chemotherapy to reduce disease bulk, and they then typically undergo further treatments to encourage remission. Based on risk factors, allo-SCT may be offered as a post-remission strategy to obtain the best possible medical outcome. For patients with disease that has not responded to chemotherapy, transplantation remains the only curative option (Döhner et al. 2017). Hence, allo-SCT is an important, and sometimes the only viable, medical option for some patients with AML.

International standards indicate that the target time frame to receive allo-SCT is within 6 weeks of the patient being medically ready for transplantation and a donor being identified (Cancer Care Ontario 2017). The disease can be stabilized with inductions, but the longer a patient waits, the greater their risk of developing significant medical complications due to a compromised immune system and the greater their risk of death from uncontrolled disease or co-morbidities.

Allo-SCT includes the need to locate and secure an appropriate donor, a standardized process implemented at allo-SCT centres overseen by the Canadian Blood Services Stem Cell Registry. This process has also undergone recent technological innovation, necessitating further local facility resources and infrastructure. Donors and patients need to be human leukocyte antigen (HLA) compatible for donated stem cells or bone marrow to have the greatest chance of restoring a patient's immune system and eradicating AML (Hamilton and Copelan 2012). DNA-based testing for antibodies is enabling more precise donor typing to facilitate the determination of an HLA match. This requires the adoption and maintenance of costly new technology and the expansion of laboratory and pathology capacity, including

highly specialized staff. Also, matching donors to patients requires significant human resources and coordination to oversee the locating, typing and management of donor products. Finally, significant coordination at the local site is required to support the clinical needs of these highly complex patients from diagnosis to pre-transplantation and from transplantation to follow-up.

Resources available for allo-SCT in Ontario, however, have not kept pace with transplantation innovations and, therefore, the increased demand. Data compiled by the Worldwide Network for Blood and Marrow Transplantation indicate that the currently accepted target meant to apply across health systems is to perform 250 to 350 allo-SCTs per 10 million people (Gratwohl et al. 2015; Passweg et al. 2016), with transplantations performed within a target time frame of 6 to 8 weeks after disease remission. Available resources in Ontario, however, made possible 250 to 280 transplantations per 10 million people, within a 3- to 6-month post-remission time frame (Cancer Care Ontario 2017). This shortfall in resources led to a situation in which patients with very medically severe conditions were waiting for allo-SCT and potentially missing their window of opportunity for this treatment modality. An investigative report published in spring 2016 identified 255 AML patients as needing allo-SCT across three Ontario hospitals for the year ending on March 31, 2016. This number was contrasted with the 160 allo-SCTs performed across the three hospitals in 2010, prior to the expanded clinical indications for transplantation (Zlomislic 2016).

To ease clinical burden and promote timely care, the Ontario Ministry of Health and Long-Term Care (MOHLTC) developed a temporary process to refer patients to the US to receive allo-SCT and promised an increase in allo-SCT funding to Ontario hospitals over several years. The MOHLTC process required individual physicians to complete an application on behalf of their patient for government-insured medical services to be provided out of the country. It is the responsibility of each Ontario allo-SCT centre to notify patients of this option and forward individual applications, manage wait-lists appropriately and determine what criteria should be used to assign priorities for transplantation within their centre. Eligible patients were then presented with the choice of either staying in the province with the risk of expanded wait times or going out of the country, which involved weighing other benefits and downsides such as loss of wages for support persons (required to accompany patients out of the country) and out-of-pocket costs for travel, accommodation and meals for up to 100 days. Unlike an absolute scarcity of organs for transplantation, allo-SCT involves scarcity of capacity due to limited physical space, human and financial resources and timely donor access. Whereas organ transplantation is primarily about the availability and ethical allocation of a finite product, the former requires nimble and efficient process management of a volatile, dynamic and multidisciplinary system to ensure timely and quality care.

Hospital-based priority-setting decisions about fair allocation of limited treatment or therapies prioritize patients' medical need, expected health benefit or a cluster of

decision-making criteria (Martin et al. 2001; Mielke et al. 2003). At the Princess Margaret Cancer Centre in Toronto (hereafter the "Centre"), a hospital with the largest and most comprehensive malignant hematology program in Canada, priority-setting decisions were initially based on the principle of first-come, first-served, as physicians agreed that the majority of patients are very ill, so medical acuity could not be used as an indicator. However, senior leaders and physicians determined that an ethical framework to ensure fairness and transparency of the SCT decision-making process, as well as an overhaul of the process leading up to allo-SCT, was required. This article reports on the ethical priority-setting and systems improvement process that the Centre undertook to manage allo-SCT.

Developing Substantive Priority-Setting Criteria

In spring 2015, Centre leaders assembled an Allo-SCT Working Group (the "Working Group") comprising physicians, administrators and those involved in patient relations, bioethics and public affairs that met weekly. The purpose of the Working Group was to address the operational and clinical aspects of expanded indications for allo-SCT within limited capacity and resources. During these meetings, the need for an ethical decision-making process was identified by Working Group members out of concern for ensuring fair allocation of allo-SCTs.

The Working Group's ethics process built upon a preliminary ethical framework established by a group of Ontario bioethicists for allo-SCT priority-setting decisions (Wright et al. 2015), which was intended to be adapted to each local Ontario hospital where allo-SCT is performed. The framework was informed by accountability for reasonableness (A4R) and program budgeting and marginal analysis principles (Gibson et al. 2006) and included an iterative process for applying A4R principles (Table 1), as well as additional principles and an initial set of criteria to guide discussion (Table 2). A4R is an established ethical priority-setting process, which incorporates the principles of distributive and procedural justice, and includes five conditions to ensure a fair priority-setting outcome: relevance, publicity, revision, empowerment and enforcement (Table 2). Application of these principles and conditions throughout the priority-setting process helps to ensure that the resulting criteria are perceived as fair and reasonable.

Through six consensus-building meetings from September to December 2015, the Working Group applied the ethical framework and determined substantive priority-setting criteria for fair allo-SCT allocation (Table 1). This included identifying substantive criteria; ranking the relevance of each criterion from 0 to 5, 5 being the most relevant; and reaching consensus on the most relevant criterion to guide allo-SCT allocation decisions.

TABLE 1. Applying the accountability for reasonableness framework to allogeneic stem cell transplantation

Step	Elaboration
Step 1 – Determine the aim and scope of the priority-setting project	• Determine the reach/focus of the priority-setting project • Determine the scope of the priority-setting decision (local hospital, province, resources available, out of country)
Step 2 – Identify the priority-setting committee	• To support and participate in the development/implementation of the process • Identify stakeholders and chairs to lead the initiative
Step 3 – Clarify existing resource mix	• Determine the resources/funds allocated and the number of allo-SCTs available • Identify how many patients are being prioritized at one time
Step 4 – Develop decision criteria with stakeholder input	• Unanimously decide, define and objectively measure criteria to use for prioritization
Step 5 – Define the decision-making process	• Decide who makes the decisions and how these will be made • Consider how often the decision-making body will meet to review the wait-list and identify the data needed to make decisions
Step 6 – Communicate the decision and rationale	• Identify the means by which decisions and alternative treatments (if identified as not priority) and their rationale will be communicated to the patient's clinician
Step 7 – Provide a formal decision review process	• Determine if there will be an appeals process • Define the basis for and who can bring an appeal forward
Step 8 – Evaluate and improve the process	• Determine the means to evaluate the process, its impact on patient outcomes/experiences and clinician experience

TABLE 2. Accountability criteria and principles that guided stakeholder discussion

Principle/Criteria	Elaboration	
Distributive justice	A principle of justice that guarantees equality of opportunity and determines how to set fair limits to healthcare (Daniels 1981).	
Procedural justice	Deliberative democratic procedures that address issues of legitimacy (Daniels and Sabin 1997). This is used if there is a persistent disagreement about rationing.	
Accountability for reasonableness (A4R)	A priority-setting model known as A4R that guides decision makers toward unanimous criteria, relevant to the principles of distributive and procedural justice. This model describes a process by which limited resources can be allocated fairly and reasonably. Five conditions must be met:	
	Publicity condition	Priority-setting decisions and their rationales must be transparent and available to the public.
	Relevance condition	An objective condition that a fair-minded person can agree with even if their preferences and needs are contrary to the criterion. This condition aims to explain why more importance is placed on certain criteria than on others.
	Appeals condition	An opportunity to revise, amend and question priority-setting decisions when presented with further evidence and arguments.
	Empowerment condition	Power differences should be minimized to ensure effective stakeholder participation.
	Enforcement condition	There should be oversight to ensure that publicity, relevance, appeals and empowerment conditions are met.

Results

Aim and scope of priority-setting decisions

Applying the A4R framework (Table 1), the Working Group decided to focus on ethical decision-making about allo-SCT (instead of leukemia or malignant hematology, more broadly) and confined the scope to the Centre with the intent to collaborate with other Ontario hospitals and provincial partners throughout the process or in the near future. The Working Group also identified that the ethical issue was how to ensure fair allocation of limited transplantations due to a shortage of allo-SCT resources and capacity (staff, laboratory and beds), rather than a lack of donor stem cells.

Stakeholders

The Working Group emphasized the need for physician involvement in the ethical decision-making process to provide important medical input and foster collaboration with other physicians and program staff and to operationalize the process outcomes in a consistent fashion. A subgroup comprising allogeneic transplant and leukemia physicians, the medical director, senior leaders and the hospital bioethicist was formed to develop an initial set of substantive criteria to guide priority-setting decisions (Tables 1 and 3).

Developing substantive criteria

The Working Group subgroup identified nine priority-setting criteria as relevant for allo-SCT allocation within the Centre: time on wait-list, medical acuity, donor eligibility, type of transplantation, likelihood of benefit, efficiency, impact on other resources, patient willingness to go out of the country and donor availability (Table 3). The subgroup returned these criteria to the larger Working Group for a broader discussion.

Guided by the principles of distributive justice, in which resources are distributed fairly, and procedural justice, emphasizing a fair and democratic process, the Working Group considered the relevance of the identified criteria before ranking their importance in guiding allo-SCT decisions. Relevance relates to whether fair-minded individuals would consider a criterion's attributes important for guiding priority-setting decisions (Daniels 1981). Table 3 identifies the criterion, the decision as to whether the criterion is relevant to SCT allocation based on consensus from the Working Group and justification for its inclusion/exclusion in the Centre's allo-SCT priority setting. For further information on excluded criteria, please see Appendix 1 (available online at longwoods.com/content/26127).

TABLE 3. Substantive criteria for ethical decision-making about allogeneic stem cell transplantation

Criterion	Decision	Rationale
Time on wait-list	Primary criterion	This is considered the most ethically defensible criterion. Patients will be placed on the wait-list at the time of the transplant consult. Interpreted as "first come, first served," this criterion means that those at the top of the wait-list will be offered the next available transplant slot.
Medical acuity	Not relevant	All patients who require allogeneic stem cell transplant (allo-SCT) are urgent or acute, and it is difficult to determine who is more urgent. Applying this criterion may therefore depend on individual physician judgment and thus risks being applied inconsistently.
Donor eligibility	Not relevant	This is an eligibility criterion, not a priority-setting criterion. To be eligible to receive allo-SCT, patients must have consult eligibility, pre-transplant work-up availability, donor availability and informed consent. Those patients who meet eligibility requirements will be considered in order of time on the wait-list for allo-SCT.
Type of transplant	Not relevant	Some types of allo-SCTs are not currently offered (e.g., haploidentical). However, this is changing, and there will be more kinds of transplants performed in the long term, so allo-SCT will not be limited by this criterion.
Likelihood of benefit	Not relevant	No consensus was reached on how to determine "benefit." Physicians will differ in their reasoning and judgment, making this criterion subjective and therefore likely to be unfair.
Efficiency	Not relevant	The availability of related donors limits the current system. This raised "donor eligibility" as a possible criterion for decision-making; however, efficiency is a goal of the overall program, not a priority-setting criterion.
Impact on other resources	Not relevant	This criterion refers to the consequences of patients receiving or not receiving allo-SCT on the medical system (e.g., requiring further chemotherapy). By addressing the wait-list in a procedural fashion, the impact on other resources will be minimized.
Patient willingness to go out of the country	Not relevant	All patients who meet the international standards for transplant will be offered the opportunity to go out of the country.
Donor availability	Not a priority-setting criterion but is an eligibility consideration when working down the list	This criterion does not affect the patient's place on the allo-SCT wait-list. There is a possibility of cryopreserving unrelated or sibling donor products so they are available when the patient is ready for transplant. This criterion is a factor when assigning the transplant date.

Time on Wait-List as the Most Ethically Defensible Priority-Setting Criterion

The Working Group agreed that *time on wait-list* should be the only criterion to guide allo-SCT priority-setting decisions. This means a "first come, first served" system whereby patients are placed on the wait-list according to when they receive the initial allo-SCT physician consult. Time on wait-list was considered as the most ethically defensible criterion because it could be consistently and objectively applied. Patients at the top of the wait-list would be offered allo-SCT first. This criterion emphasized to the Working Group the need to streamline referral processes so that the timing of allogeneic consult does not unfairly disadvantage or advantage patients. Thus, as the priority-setting process evolved the perception

of what was at stake from an ethical standpoint also evolved. It became clear through multiple discussions with stakeholders that health systems improvement was also ethically salient because it impacted the availability and timeliness of transplantations.

Streamlining organizational processes to promote fairness and equity

The Working Group discussed operational ways to consistently identify who had "first come" so that they would be "first served." It was agreed that once patients were seen by the allo-SCT physician, and transplantation was medically recommended, their names would be added to a single wait-list dated with the time of their initial consult. An allo-SCT date would then be assigned by providing a date to the first eligible person on the list. To be eligible, the patient should have completed the pre-allo-SCT work up, have a donor available and have any co-morbidities under control. For patients to be fairly placed on the wait-list, workflow and organizational processes needed to be streamlined to ensure consult dates were fairly and efficiently allocated, and all consulting physicians consistently added their patients to the wait-list without delay.

Ethical values of equity and fairness also supported a change in the donor work-up to ensure timeliness of donor identification. Donor work-up went from being initiated *after* patients completed chemotherapy to being done *during* chemotherapy, before patients being medically ready for transplantation. At their initial clinical assessment, all AML patients had blood drawn for HLA typing and antibody testing and were asked for information regarding close relatives, so that transplantation search coordinators could begin to identify potential related donors and initiate a search for a matched unrelated donor.

A final consideration involved patients who experience co-morbidities while on the transplantation wait-list. The Working Group decided that these patients would receive the required treatment (e.g., antibiotics) without relinquishing their priority on the list. Patients who received allo-SCT but then later experienced a relapse would begin the wait-list process anew by participating in an initial transplantation consult.

Consideration and Rejection of Medical Acuity and Medical Benefit as Relevant Criteria

In contrast to the traditional principle of triage, our Working Group did not consider medical acuity as a relevant priority-setting criterion. To define medical acuity as a criterion, stakeholders aimed to develop an operational definition for sickness in which "sickness" included being at the highest risk of death, including co-morbidities. According to this criterion, the sickest patients would receive treatment first. However, the Working Group believed that these patients would be less likely to benefit medically from a transplantation. Furthermore, in the context of AML, the criterion of medical acuity would result in patients with the highest likelihood of leukemia remission/eradication never receiving prompt allo-SCT because their transplantation would be delayed continually in favour of someone else

who is sicker. The Working Group felt that this rationale was indefensible because as wait times were extended for patients with less medical acuity, they too would inevitably become sicker.

When discussing who would benefit medically the most from allo-SCT, the Working Group had difficulty reaching agreement. Some physicians argued that science was not yet advanced enough to make these predictions and to determine the best timing for a transplantation between two sick patients. Thus, determinations based on level of sickness or medical benefit would require very specific definitions and eligibility criteria that were subjective, were difficult to define and lacked a robust evidence base and standardized system to rank patients. Applying medical acuity or medical benefit would therefore require individual physician judgment and would carry the risk of being applied inconsistently, which would go against the ethical principle of fair and transparent decision-making upon which reasonable people would agree.

Finally, the Working Group distinguished eligibility criteria from priority-setting criteria and believed that medical acuity or medical benefit is related to the former but not the latter. Patients would first need to be eligible for a transplantation before priority-setting criteria could be applied. Medical acuity or medical benefit as well as other decision-making factors were perceived to inform whether a patient is eligible to receive a transplantation. The Working Group felt that a robust transplantation system would need to take into account disease differences, patient factors (e.g., medical co-morbidities) and the timely availability of a donor. Singling out medical acuity or medical benefit as a priority-setting criterion to determine which patient receives the next transplantation upsets the multifactorial parameters of clinical decision-making and the determination of transplantation eligibility.

Systems management and process improvement

The ultimate ethical goal as agreed upon by the Working Group was utilitarian: to get as many patients in need of transplantation access to transplantation as quickly as possible (i.e., greatest good for the greatest number). Therefore, the Working Group recognized that in addition to priority-setting criteria, a broader, systems-level approach to reducing impediments and enhancing efficiencies within the patient pathway to allo-SCT was ethically required, thus improving eligibility determination and the time frame to receive allo-SCT for patients.

A process to evaluate the system and improve workflow began at the Centre with a formal lean initiative in the fall of 2015 (Scoville and Little 2014). A range of mitigating circumstances were identified that should be taken into account to manage the wait-list, including co-morbidities, timely donor availability and bed capacity. As a result of the initiative, clinical managers and leaders completed current state process mapping, identified opportunities for improvement and delineated the characteristics of an ideal patient journey. The clinical team also identified and reported core metrics daily in huddles to ensure goals

were met. They created action plans, revised existing tools and streamlined the flow of information between coordinators and care teams. In addition, a weekly wait-list meeting was refined so that patient and donor status were reviewed consistently and systematically.

Appeals/revisions

A4R is an iterative process with a mechanism for appeals and revisions as needed, as situations change or at designated time intervals as agreed upon by stakeholders. Although the Working Group initially decided that the most ethical criterion to guide allo-SCT priority setting was "first-come, first-served," after implementing the criterion into Centre procedures, those involved in the allogeneic transplantation program found that this criterion alone did not provide appropriate flexibility because the first person on the wait-list may not be ready for allo-SCT. To allow greater transparency and flexibility within the wait-list so as to use all available allo-SCT slots and not disadvantage those patients who are ready for allo-SCT, the previously agreed-upon "first come, first served" criterion was refined to "first ready, first to go." Patients continued to be placed on the wait-list at their initial allo-SCT consult, but flexibility was permitted within the wait-list structure to allow for individuals' unique disease situation and mitigating circumstances (e.g., donor availability).

As discussions continued and the wait-list became more efficiently managed, it became apparent that wait-list patients could be grouped into four categories: patients ready for transplantation, patients who required some medical work-up, patients who required significant medical work-up and patients who required significant medical work-up and a donor had not yet been identified. Patients within these categories were reviewed at the weekly wait-list meetings to ensure consistency and timeliness of care.

Discussion

Since 2015, wait-lists at this Centre and other allo-SCT centres across Ontario have been significantly reduced as efficiencies, capacity-building efforts and coordination between transplantation centres in Ontario were realized (Cancer Care Ontario 2017). As the time frame to receive allo-SCT within Ontario decreased, there was less rationale for patients to accept going out of the country; however, each centre continued to offer this option, as it was, in part, the result of these patients going out of the country that allowed for a reduction in wait-lists in Ontario.

In 2017, there were 281 patients across Ontario who received an allogeneic transplantation (Cancer Care Ontario 2017). This was a 12% increase in all patients (not just AML) receiving SCT within Ontario from 2015/16 and a 70% increase in volume from 2009/10 (Cancer Care Ontario 2017). However, the number of patients eligible for SCT continues to grow and has not yet reached the expected volumes seen internationally. Therefore, conversation must continue among healthcare partners about the level of investment in health human resources and capital planning that is required to continue to meet current and future need.

Finally, although this article reports on an ethical priority-setting process with utilitarian goals, the individual patient experience should not be forgotten. Unlike other resource allocation systems in which there are limits to individual autonomy in favour of top-down decision-making to benefit public health (Devereaux et al. 2008), our Centre's process recognized patient choice as morally salient. Patients with life-threatening disease are faced with difficult care decisions. Although advances have been made and this is an evolving field, allo-SCT may be associated with significant morbidity and mortality and may not be consistent with a patient's values and preferences. Therefore, it is important that patients are well supported in their decision-making process to make informed decisions about their care, including the decision to not accept transplantation. This requires clinical teams communicating relevant information to support patient understanding but also addressing socio-economic barriers that might prevent some patients from selecting the out-of-country option. For example, by working together with healthcare partners in expanding financial coverage of associated costs for out-of-country transplantation.

Conclusion

By applying an ethical framework to allo-SCT resource allocation, stakeholders were able to set priorities within one Centre upon which it was believed fair and reasonable people would agree. Ethical priority-setting has been implemented in other healthcare contexts to determine the principles, criteria and processes that ought to support decision-making in resource and capacity scarcity (Christian et al. 2006; Frolic et al. 2009; Gibson et al. 2011; Silva et al. 2010). In all of these settings, it is crucial to focus on establishing a process to make timely and fair decisions about the allocation of limited goods and services.

Lessons learned from our experience include setting expectations early with key stakeholders regarding the time and human resource investment required to engage in comprehensive deliberative discussions. Referring back to shared goals when stakeholders grow weary or impatient helps to underscore the purpose and value of the process. A potential limitation of our process is that we did not involve patients directly in our priority-setting discussions. However, patients and families were engaged at the provincial level and provided important advocacy for system developments (including the out of the country program). We recognize that patients are perhaps the most important stakeholders because decisions directly impact their care. There is opportunity as part of the A4R framework for our Working Group to involve patients in revisiting and revising the process in the future.

Cancer care is seeing an increase in the number of those surviving or living with the disease due to major advances in the past decade in prevention, screening and high-quality treatment (Heymach et al. 2018). Adoptive cell immunotherapy (chimeric antigen receptor T-cell therapy) and precision medicine are major clinical cancer advances that use genetics and the body's own immune system to inform targeted treatment. These treatments have shown promising results for otherwise incurable malignancies (Hyman et al. 2017).

However, targeted therapies are increasingly expensive and raise complex access and equity issues related to the availability of potentially life-saving drugs. In this new era of precision medicine, policy makers are increasingly confronted with challenging ethical decisions that include deciding which drugs to fund and how to prioritize individuals for clinical trials of breakthrough therapies (Jecker et al. 2017). By sharing one Centre's process of hospital-based priority-setting, we hope to assist others in ethical priority-setting and policy making in our rapidly evolving and complex healthcare environments.

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References

Cancer Care Ontario. 2017. *Complex Malignant Hematology Services in Ontario: June 2017 – Year in Review*. Ontario, Canada: Complex Malignant Hematology Hematopoietic Cell Therapy Consultation Group.

Christian, M.D., L. Hawryluck, R.S. Wax, T. Cook, N.M. Lazar, M.S. Herridge et al. 2006. Development of a Triage Protocol for Critical Care during an Influenza Pandemic. *Canadian Medical Association Journal* 175(11): 1377–81. doi:10.1503/cmaj.060911.

Daniels, N. 1981. Health-Care Needs and Distributive Justice. *Philosophy & Public Affairs* 10(2): 146–79.

Daniels N. and J. Sabin. 1997. Limits to health care: fair procedures, democratic deliberation, and the legitimacy problem for insurers. *Philos Public Aff*. 1997 Fall;26(4): 303–50. doi: 10.1111/j.1088-4963.1997.tb00082.x.

Deschler, B. and M. Lubbert. 2006. Acute Myeloid Leukemia: Epidemiology and Etiology. *Cancer* 107(9): 2099–107. doi:10.1002/cncr.22233.

Devereaux, A.V., J.R. Dichter, M.D. Christian, N.N. Dubler, C.E. Sandrock, J.L. Hick et al. 2008. Definitive Care for the Critically Ill during a Disaster: A Framework for Allocation of Scarce Resources in Mass Critical Care: From a Task Force for Mass Critical Care Summit Meeting, January 26–27, 2007, Chicago, IL. *Chest* 133(5 Suppl): 51s–66s. doi:10.1378/chest.07-2693.

Döhner, H., E. Estey, D. Grimwade, S. Amadori, F.R. Appelbaum, T. Büchner et al. 2017. Diagnosis and Management of AML in Adults: 2017 Eln Recommendations from an International Expert Panel. *Blood* 129(4): 424–47. doi:10.1182/blood-2016-08-733196.

El Rassi, F. and M. Arellano. 2013. Update on Optimal Management of Acute Myeloid Leukemia. *Clinical Medicine Insights Oncology* 7: 181–97. doi:10.4137/cmo.s8528.

Frolic, A., A. Kata and P. Kraus. 2009. Development of a Critical Care Triage Protocol for Pandemic Influenza: Integrating Ethics, Evidence and Effectiveness. *Healthcare Quarterly* 12(4): 56–64. doi:10.12927/hcq.2009.21054.

Gibson, J., C. Mitton and G. Dubois-Wing. 2011. Priority Setting in Ontario's LHINs: Ethics and Economics in Action. *Healthcare Quarterly* 14: 35–46. doi:10.12927/hcq.2011.22649.

Gibson, J., C. Mitton, D. Martin, C. Donaldson and P. Singer. 2006. Ethics and Economics: Does Programme Budgeting and Marginal Analysis Contribute to Fair Priority Setting? *Journal of Health Services Research and Policy* 11(1): 32–37. doi:10.1258/135581906775094280.

Gratwohl, A., M.C. Pasquini, M. Aljurf, Y. Atsuta, H. Baldomero, L. Foeken et al. 2015. One Million Haemopoietic Stem-Cell Transplants: A Retrospective Observational Study. *Lancet Haematology* 2(3): e91–100. doi:10.1016/s2352-3026(15)00028-9.

Hamilton, B.K. and E.A. Copelan. 2012. Concise Review: The Role of Hematopoietic Stem Cell Transplantation in the Treatment of Acute Myeloid Leukemia. *Stem Cells* 30(8): 1581–86. doi:10.1002/stem.1140.

Heymach, J., L. Krilov, A. Alberg, N. Baxter, S.M. Chang, R. Corcoran et al. 2018. Clinical Cancer Advances 2018: Annual Report on Progress against Cancer from the American Society of Clinical Oncology. *Journal of Clinical Oncology* 36(10): 1020–44. doi:10.1200/JCO.2017.77.0446.

Hyman, D.M., T.W. Laetsch, S. Kummar, S.G. DuBois, A.F. Farago, A.S. Pappo et al. 2017. The Efficacy of Larotrectinib (Loxo-101), a Selective Tropomyosin Receptor Kinase (Trk) Inhibitor, in Adult and Pediatric Trk Fusion Cancers. *American Society of Clinical Oncology* 35. doi:10.1200/JCO.2017.35.18_suppl.LBA2501.

Jecker, N.S., A.G. Wightman, A.R. Rosenberg and D.S. Diekema. 2017. From Protection to Entitlement: Selecting Research Subjects for Early Phase Clinical Trials Involving Breakthrough Therapies. *Journal of Medical Ethics 43*(6): 391–400. doi:10.1136/medethics-2016-103868.

Martin, D.K., J.L. Pater and P.A. Singer. 2001. Priority-Setting Decisions for New Cancer Drugs: A Qualitative Case Study. *The Lancet* 358(9294): 1676–81. doi:10.1016/s0140-6736(01)06714-9.

Mielke, J., D.K. Martin and P.A. Singer. 2003. Priority Setting in a Hospital Critical Care Unit: Qualitative Case Study. *Critical Care Medicine* 31(12): 2764–68. doi:10.1097/01.ccm.0000098440.74735.de.

Passweg, J.R., H. Baldomero, P. Bader, C. Bonini, S. Cesaro, P. Dreger et al. 2016. Hematopoietic Stem Cell Transplantation in Europe 2014: More Than 40,000 Transplants Annually. *Bone Marrow Transplantation* 51: 786–92. doi:10.1038/bmt.2016.20.

Scoville, R. and K. Little. 2014. *Comparing Lean and Quality Improvement*. Cambridge, MA: IHI White Paper.

Silva, D.S., J.X. Nie, K. Rossiter, S. Sahni, R. Upshur and Canadian Program of Research on Ethics in a Pandemic. 2010. Contextualizing Ethics: Ventilators, H1N1 and Marginalized Populations. *Healthcare Quarterly* 13(1): 32–36. doi:10.12927/hcq.2013.21613.

Wright, L., A. Frolic, R. Sibbald, T. Foreman and J. Bell. 2015. Developing an Ethical Framework to Manage Access to Stem Cell Transplantation in Ontario. Ontario, Canada: Author.

Zlomislic, D. 2016, April 19. Ontario to Spend $100m Outsourcing Life-Saving Transplants to U.S. *The Star*. Retrieved February 5, 2020. <https://www.thestar.com/news/canada/2016/04/19/ontario-to-spend-100m-outsourcing-life-saving-transplants-to-us.html>.

RESEARCH PAPER

Policy Agenda-Setting and Causal Stories: Examining How Organized Interests Redefined the Problem of Refugee Health Policy in Canada

Programme d'élaboration des politiques et anecdotes : comment des intérêts organisés redéfinissent le problème des politiques de santé canadiennes à l'intention des personnes réfugiées

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Abstract

The development of refugee health policies is significant, given the increased volume of displaced persons seeking refuge in Canada and around the world. Changes to the Canadian refugee health policy, known as the Interim Federal Health Program (IFHP), limited healthcare access for refugees and refugee claimants from 2012 to 2016. In this article, we present a policy analysis using the case of the IFHP retrenchments to examine how political actors on opposing sides of the issue defined the problem using different causal story mechanisms. This analysis reveals that organized interests dramatically changed the problem definition of the IFHP reforms. Following their use of causal stories in redefining the problem, the courts declared that the reforms to refugee healthcare were a form of cruel and unusual treatment. Understanding policy strategies used by proponents of refugee healthcare coverage expansion is important for countries responding to the current, enduring refugee crisis.

Résumé

En raison du nombre de personnes déplacées qui cherchent refuge au Canada et ailleurs dans le monde, les politiques de santé à l'intention des réfugiés ont connu d'importants développements. Les changements apportés à la politique canadienne, connue sous le nom de Programme fédéral de santé intérimaire (PFSI), ont limité, entre 2012 et 2016, l'accès aux services de santé pour les réfugiés et les demandeurs d'asile. Dans cet article, nous présentons une analyse politique, en considérant les retranchements apportés à la PFSI, afin d'étudier comment les acteurs politiques ayant des vues opposées sur la question définissent le problème en utilisant différents mécanismes qui ont recours aux anecdotes. Cette analyse révèle que des intérêts organisés ont profondément transformé la définition du problème dans le cadre des réformes du PFSI. Suivant leur utilisation d'anecdotes dans la redéfinition du problème, les tribunaux ont déclaré que les réformes du système de santé en faveur des réfugiés constituaient une forme de cruauté et de traitement inhabituel. Il est important de comprendre les stratégies politiques employées par ceux qui proposent une expansion de la couverture des services de santé pour les réfugiés afin que les pays puissent répondre à la crise actuelle en matière de réfugiés.

Introduction

The United Nations High Commissioner for Refugees (UNHCR 2019) reports that there are 70.5 million forcibly displaced migrants worldwide, representing the highest level of forced migration since World War II. Following the protraction and persistence of refugee crises around the world, Canada has resettled more than 132,000 refugees and refugee claimants over the past four years, many of whom receive healthcare coverage under the Interim Federal Health Program (IFHP; Government of Canada 2019). The IFHP is a federally funded program established in 1957 that provides comprehensive healthcare insurance for

refugee populations seeking protection in Canada (CIC 2006; IRCC 2017). Before 2012, refugees and claimants received healthcare coverage for physician and hospital visits as well as supplementary care, including optical, dental and drug coverage. On April 25, 2012, the former Conservative government of Canada introduced cutbacks to health coverage provided under the IFHP. These retrenchments separated refugee recipients into categories that provided varying levels of coverage depending on their country of origin and immigration status, significantly limiting healthcare access for this vulnerable population (Campbell et al. 2012; CIC 2012; Table 1).

TABLE 1. 2012 Interim Federal Health Program reform information

Interim Federal Health Program group **Who is eligible?**	**Coverage** **What are they eligible for?**
Government-assisted refugees and other refugees who are receiving governmental resettlement assistance in the form of income support, including visa office-referred refugees and refugees coming to Canada through the Joint Assistance Sponsorship Program	**Expanded healthcare coverage** includes coverage of the following: • hospital services, • services of physicians, registered nurses and other healthcare professionals licensed in Canada, • laboratory, diagnostic and ambulance services, • supplemental services (audio care, home care occupational therapy, physiotherapy, dental care, optical care, etc.), • supplemental products (immunizations, medications) and • translation services for health purposes.
Privately sponsored refugees – Resettled refugees while under sponsorship who do not receive and have not received governmental resettlement assistance in the form of income support	**Healthcare coverage** includes coverage of hospital services, services of a doctor or registered nurse who is licensed in Canada and laboratory, diagnostic and ambulance services, with some limitations. Medications and vaccines only when needed to prevent or treat a disease posing a risk to public health or to treat a condition of public safety concern, such as HIV or tuberculosis (TB).
Refugee claimants who are from a designated country of origin – a country deemed safe by the Immigration Minster	**Public health or public safety healthcare coverage** includes coverage of hospital services, services of a doctor or registered nurse licensed in Canada, laboratory and diagnostic services and medication and vaccines, only if they are required to diagnose, prevent or treat a disease posing a risk to public health or to diagnose or treat a condition of public safety concern **Immigration medical examination**
Refugee claimants who are not from a designated country of origin	Healthcare coverage and immigration medical examination
People whose refugee claim has been suspended	Public health or public safety healthcare coverage and immigration medical examination
Rejected refugee claimants	Public health or public safety healthcare coverage and immigration medical examination
Persons for whom the Minister exercises discretion on his own initiative for humanitarian and compassionate considerations or for public policy considerations	Expanded healthcare coverage and immigration medical examinations

Within a month, professional organizations and advocacy groups collectively voiced concerns for refugees' restricted healthcare access. Following their organized protest and legal challenge, the Federal Court of Canada ruled that the IFHP cuts constituted "cruel and unusual" treatment, violating Section 12 of the *Charter of Rights and Freedoms* (CDRC v. AGC

2014). In response, the Federal Government of Canada announced "temporary measures" for the IFHP on November 4, 2014. This program revision was not a full reversal of the 2012 cuts, as ordered by the federal court, but it did restore some health services coverage for refugee women and children.

This policy analysis examines how political actors on opposing sides of the refugee healthcare cuts issue defined the problem to enact policy changes. It is based on the causal stories framework developed by Stone (1989, 2012), resting within the post-structural tradition of narrative policy research. Stone (2012) argued that the process of policy making entails a struggle over the meaning and significance of policy ideas and their influence on values embedded in community life. Disputes over collective community values drive policy debates articulated through relations of power and structures of governance. Subsequently, political discourse and language shape how policy ideas are communicated and translated into practice (Campbell 2002). An examination of the policy discourse will reveal the problem definitions and associated causal story constructions used by various government actors who initiated the IFHP cutbacks and by organized interest groups who called for its reversal. This analysis identifies stories or themes used to frame policy ideas of actors on both sides of the IFHP issue, contributing to our understanding of how political actors control interpretations, assign responsibility and influence policy decisions in refugee policy debates.

Analytical Framework and Methods

Stone's (1989, 2012) causal stories framework argues that causal ideas are at the core of understanding how difficult conditions or circumstances are transformed into political problems within the policy discourse. This process – referred to as "problem definition" – relies on the ability to attribute cause, blame and responsibility, while being amenable to human intervention (Stone 1989). Causal ideas are theories of causation that frame problems, strategically crafted using stories, symbols and numbers, applied by political actors on different sides of an issue to describe harms, assign responsibility and garner support to propose a policy solution. Throughout the problem definition process, political actors struggle for control over interpretations of the issue and compete to influence which causal idea becomes the main guide to policy (Stone 2012). These ideas are categorized into one of four causal theories that define problems based on the intentionality of the action and predictability of the consequences (Table 2). For instance, intentional causal theory suggests that the problem is derived from a deliberate action that produces expected consequences. Mechanistic causation refers to an unexpected action, such as a mechanical mishap, that leads to predictable outcomes, whereas accidental causal theory suggests that an unexpected action produces unpredictable consequences. Inadvertent causal theory indicates that the problem stems from an intended action resulting in unpredictable consequences.

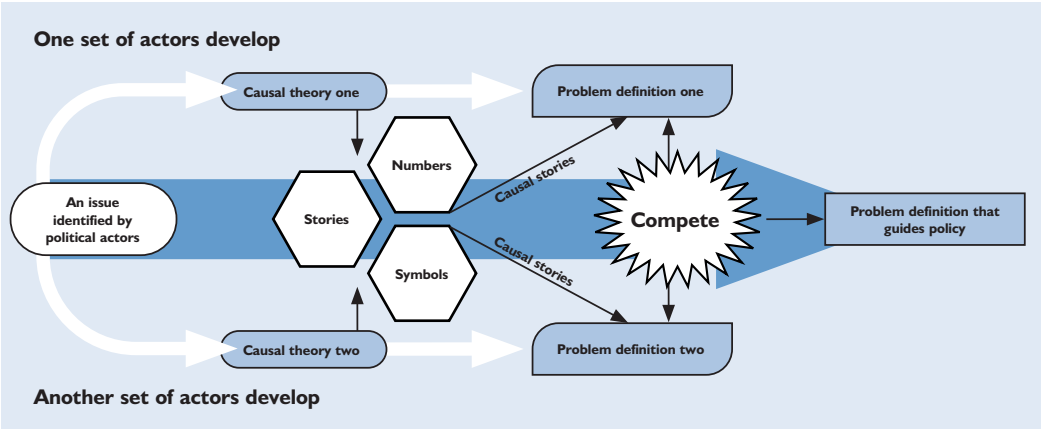
TABLE 2. Stone's (1989, 2012) theory of causal stories framework

	Consequences	
Actions	**Intended**	**Unintended**
Unguided	Mechanical cause[a] • Intervening agent • Machines	Accidental cause[b] • Fate • Natural disaster
Guided	Intentional cause[c] • Oppression • Blaming the victim	Inadvertent cause[d] • Unanticipated harmful side effects of policy

a Mechanistic causation – unguided action(s) resulting in predictable consequences.
b Accidental causation – unguided action(s) with unpredictable consequences (such as fate, the absence of human control).
c Intentional causation – guided action(s) leading to intended consequences (involving complete human control).
d Inadvertent causation – guided action(s) leading to unintended consequences.

Causal theories create the resulting problem definition using stories, symbols and numbers in the political discourse (Figure 1). According to Stone (1989, 2012), stories are composed with heroes, villains, problems and solutions and categorized into those involving either change or power struggles. *Stories of change* include *stories of decline*, which depict changes for the worse or stymied progress, whereas *stories of rising* depict successful transformations. *Stories of power* include those of *control* or *helplessness*, which represent the gain or loss of power, respectively. Symbols can include powerful literary devices, such as synecdoche and metaphor, as well as ambiguity, which synchronize motivations and values that fuel collective action. In this context, a synecdoche is defined as "a small part of the policy problem, used to represent the whole" (Stone 2012, p. 159). Numbers are descriptions of the world, derived from measuring and counting a problem, that support stories and symbols based on their interpretations (Stone 2012).

FIGURE 1. The problem definition process – transforming political issues into policy problems



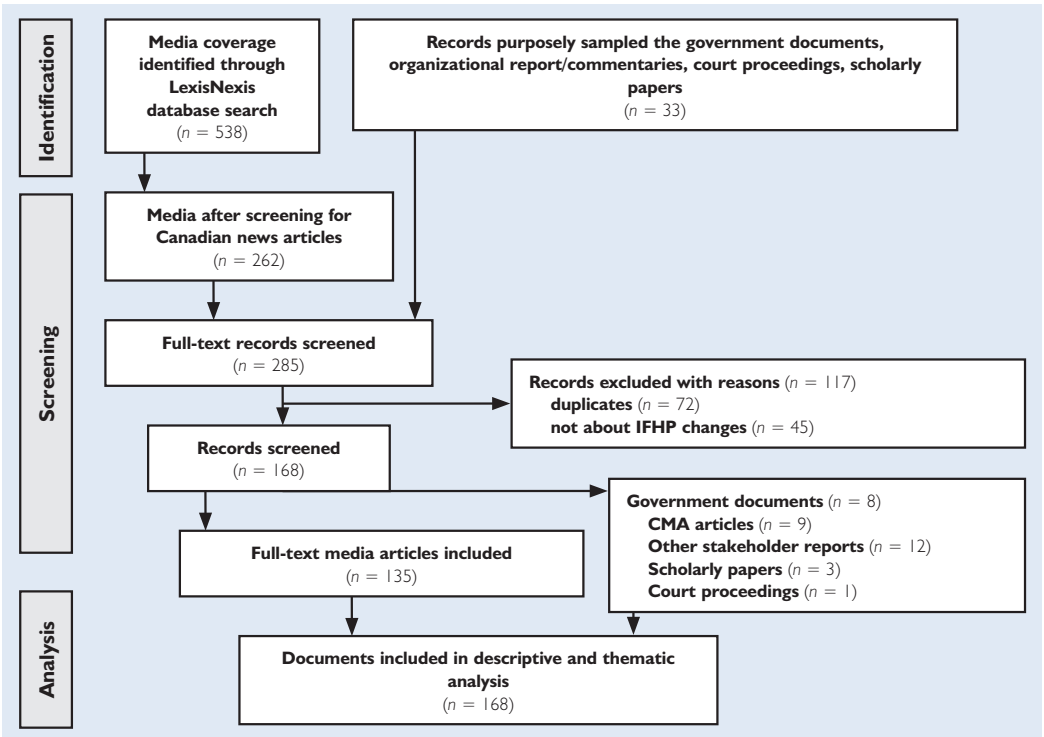
The problem (re)definition process occurs when at least two different groups of political actors develop causal theories, composed of mechanisms (stories, symbols and numbers) that frame divergent problem definitions, which compete against each other to gain precedence on the political agenda and guide policy-making processes.

We used the policy case of the IFHP retrenchments in 2012 and their partial reversal in 2014 to examine how each problem definition process was constructed and how different

causal mechanisms were used to define the problem by political actors on opposing sides of the issue (Yin 2009). An interpretive policy analysis of government documents, organizational reports, academic papers and a court proceeding was conducted. These documents were retrieved from ProQuest databases, PubMed, governmental and organizational websites and Google Scholar and examined to abstract different problem definitions of the refugee health policy reforms. In addition, news media articles on refugee health policy in Canada were searched using the LexisNexis database. Keywords included a combination of "Canada," "health policy," "refugee," "healthcare," "coverage" and "IFHP." The database search retrieved English-language newspaper articles only.

Canadian media reports (n = 262) were identified, and 135 articles were included in this study (Figure 2). News media coverage spanned nine provinces, of which 84% of articles were published in the top 10 newspaper sources (Appendix 1, available online at longwoods.com/content/26126). Documents (n = 33) published after the 2012 IFHP changes and before the introduction of the "temporary measures" to the IFHP in November 2014 were included. A content analysis was employed using a constant comparative approach to abstract themes of problem definition and causal stories, drawing on inductive discursive analysis techniques (Glynos et al. 2009). Triangulation of sources was conducted by assessing the consistency of data themes abstracted from the variety of documents analyzed in this study, strengthening the credibility and trustworthiness of the results (Patton 1999).

FIGURE 2. Media and document search strategy



Results

Causal theories were used by political actors on opposing sides of the issue to convey different representations of the problem of IFHP cutbacks: governmental actors used *intentional causation* and organized interests used *inadvertent causation*. This section examines each of these causal theories, deconstructing the causal stories used within each and revealing strategies used by political actors to gain support for their interpretation of the problem.

Causal Theory One: Intentional causation as a defense of the federal position

Federal government actors identified the issue of increasing refugee claims from European Union "democracies" that were rejected by the refugee determination system in Canada (Campion-Smith and Keung 2012; Appendix 2, available online at longwoods.com/content/26126). They transformed the issue into a political problem by framing the submission of failed asylum claims as the willful illegal action of fraudulent asylum seekers in Canada, which justified the implementation of the 2012 IFHP cutbacks. In this causal story, intentional causation is used to defend the federal government's position of withdrawing or limiting healthcare coverage to certain groups of refugees, whereby the problem's cause is assigned to an intended outcome resulting from guided, deliberate action. In turn, IFHP cutbacks were portrayed as a means to deter false refugee claims, contain costs and ensure fairness to Canadians.

Causal story strategies using intentional causation

DETERRING FALSE REFUGEE CLAIMS

The causal story portrayed by governmental actors is one of power and control, as the retrenchments are framed as a solution to "stop the abuse of Canada's generous and overburdened healthcare system by bogus refugees" (Keung 2012a). According to then Prime Minister Stephen Harper, healthcare benefits were removed "if we had clearly bogus refugees who have been refused and turned down" (Gulli 2015). In this case, a symbolic device, the *synecdoche*, is used to convey that "bogus refugees" define the entire problem and policy response. However, labelling all claimants who were refused refugee status as "bogus refugees" represents only part of the story. As a result, the IFHP cuts not only eliminated coverage for failed claimants, it also limited healthcare access for claimants awaiting a decision on their claims as well as for privately sponsored refugees (Table 1).

One year following the implementation of the IFHP reforms, the then Immigration Minister Chris Alexander stated that "Under the old, broken refugee system, abuse was commonplace. Thanks to our reforms, we've seen the number of asylum claims from safe countries fall by 87%" (Alexander 2014). The Minister presented a *story of rising* in which progress was made as a result of the policy response. Despite the use of numbers to justify this *story of change*, "numbers in policy debates cannot be understood without probing how people produced them," (Stone 2012, p. 159). Therefore, *ambiguity*, which is essentially the

capacity to have multiple meanings, underlies the origin of these numbers, urging the question of whether these figures were produced as a result of the IFHP cutbacks or whether they were generated as a result of the changes to the refugee determination system that same year (Bhuyan et al. 2014).

CONTAINING COSTS

Under the intentional causal problem definition, governmental actors present the causal story that increased intake of fraudulent refugees costs the healthcare system, and the IFHP cuts are a cost-containment measure. According to governmental actors,

> the cost of the IFHP continued to rise as a result of ... the increasing number of people eligible for IFHP coverage. For example, there were 105,326 people eligible for IFHP benefits in 2003, whereas, there were 128,586 people eligible for benefits in 2012. ... the IFHP cost Canadian taxpayers $50,600,000 in 2002/2003 and almost $91,000,000 in 2009/2010. As a consequence, cost containment was a driving principle underlying the decision to reform (CDRC v. AGC 2014).

This causal *story of decline* reveals that with increasing numbers of refugees, there were harms in the form of increased costs to taxpayers. Political actors strategically used numbers to assert that the rising cost phenomenon was occurring frequently, even though "overall expenditure on the IFHP is a tiny fraction (0.04%) of the percentage of total health expenditure in Canada" (Stall 2012).

Governmental actors used symbolic devices, such as the *container metaphor*, to convey that the IFHP costs were overflowing and needed a container to prevent spillover. Moreover, Stone (2012) indicated that *stories of decline* serve as the impetus for *stories of control*. As such, governmental actors used the empirical argument of containing costs to set the stage for the *story of control*, whereby they emphasize that the IFHP reforms would "ensure that tax dollars are spent wisely," saving taxpayers money (Keung 2012b).

ENSURING FAIRNESS TO CANADIANS

Governmental actors normatively argue that the cutbacks are a means to ensure fairness to Canadians. According to Alexis Pavlich, Immigration, Refugee and Citizenship, Canada spokeswoman,

> Canadians have been clear that they do not want illegal immigrants and bogus refugee claimants receiving gold-plated healthcare benefits that are better than those Canadian taxpayers receive (Keung 2013a; Komarnicki 2014).

Again, political actors use synecdochical labels such as "illegal immigrants" and "bogus refugees" to represent all who were affected by the reforms. Moreover, "gold-plated healthcare

benefits" is an *evocative metaphor* used to generate anger among Canadian citizens for having received fewer healthcare benefits than refugees. According to Stone, "the emotional impact of symbolic devices can make it harder for audiences to recognize and question the underlying factual assumptions" (Stone 2012, p. 177). In this case, the public overlooks the plight of refugees, who flee their homelands seeking refuge from endemic violence. The fact that refugees receive coverage "equivalent to Canadians on social assistance" (Payne 2014) was omitted by actors in this problem definition. Advocates describe the government's response to refugees as one that excludes refugees as "aliens who are treated with suspicion, not as guests needing help" (Stanbrook 2014). This label presents refugees as the other, an inhuman entity, undeserving of the social support and healthcare coverage that was previously provided to them.

Causal Theory Two: Inadvertent causation and the mobilization of organized interests

Organized interests transformed the issue of limited healthcare access for refugees into a political problem, in which the IFHP cuts were defined as "both inequitable and possibly inhumane in light of the extreme hardship and mistreatment many [refugees] have already experienced" (Arya et al. 2012, p. 1876). In contrast to the government's framing, interest groups used the theory of inadvertent causation to re-define the problem as one of guided action by the government with unintended consequences, "inadvertently introducing new system-level barriers to healthcare" (Arya et al. 2012, p. 1876). The IFHP reforms created suffering for refugees, generated ethical dilemmas for healthcare providers, threatened public health and downloaded costs to provinces, healthcare institutions and taxpayers. These organized interests included health professional associations, refugee-serving organizations, provincial governments and refugees who organized to instigate the legal challenge (Sanders 2012). See Appendix 3, available online at longwoods.com/content/26126.

Causal story strategies using inadvertent causation

PRODUCING PREVENTABLE SUFFERING FOR REFUGEES

The causal story depicted by organized interests is one of helplessness, in which tensions are portrayed explicitly on the assumption that situations were better in the past and have changed for the worse (Stone 2012). According to academic, media, legal and interest group reports (Barnes 2013; CCR 2013; CDRC v. AGC 2014; Raza et al. 2012; Seeking Solutions 2013; Sheikh et al. 2013), advocates conveyed that the situation before the reforms provided better access to healthcare for refugees, during which they received services equivalent to those received by Canadians on social assistance. In particular, organized interests conveyed how the IFHP reforms caused suffering for refugees on several accounts, for example, "Since the federal cuts, people with cancer cannot access chemotherapy, pregnant women are denied prenatal care, and diabetic children are not entitled to insulin medication." (Payne 2014). These *stories of helplessness* are *synecdoches*, representing parts of the whole problem.

Some of these stories are represented in the media as "horror stories" (Stone 2012). According to Dr. Buchman, who treated a 72-year-old failed refugee claimant,

> Her tumors were very large and disfiguring. Her chest wound was open and bleeding and infected. She was not eligible for cancer treatment ... we needed to find a place to accept her and allow her a peaceful, comfortable, dignified death (Keung 2014a).

Horror stories generate fear, as expressed by healthcare professionals: "Watching our patients become ill as a direct result of this policy has left us feeling desperate. We frankly fear for the lives of our patients" (Kraeker and O'Shea 2012). These *stories of helplessness* and horror are symbolic representations that allow people to identify with refugees, particularly the hardships and suffering endured as a result of the limited access to healthcare created by the IFHP reforms.

GENERATING ETHICAL DILEMMAS

A corresponding *story of helplessness* conveyed by organized interests in their problem redefinition is the loss of control by physicians and other healthcare workers in administering refugee care. According to Ontario's former Health Minister, Dr. Eric Hoskins,

> Cuts to the Interim Federal Health Program left refugee claimants unprotected and put our doctors in an untenable position, forcing them to choose who should be treated (Keung 2014b).

The ethical dilemma of placing healthcare providers in a position to deny providing care for a vulnerable group of people was a frequently discussed issue by the health provider community. According to advocates, "Healthcare workers should be deciding what care people need based on their illness – not their income or refugee status" (Hayes 2012).

THREATENING PUBLIC HEALTH

According to organized interests, the IFHP cutbacks harmed not only refugees but also the public. A causal *story of decline* is presented in which the reforms place the public at risk of developing communicable diseases. According to one interest group,

> Even though treatment for a select list of public health conditions remain covered [for refugees], the testing needed to diagnose these conditions often isn't, paradoxically. This results in a failure to protect either the public or the patient (Stanbrook 2014).

Again, powerful literary devices are used to portray the negative outcomes of retrenching diagnostic services, to generate fear and mobilize action. An example of the use of the *synecdoche* appears in the following statement from Dr. Gruner of the Canadian Doctors for Refugee Care:

> If they've got a cough, it could be tuberculosis but we're never going to know because they're not going to the doctor ... But they are going to the playgrounds, the schoolyards, the shopping centres, putting the rest of us at risk (Levitz 2013).

DOWNLOADING COSTS

Organized interests argue that the reforms generate harm through the IFHP cutbacks because they download the cost of refugee healthcare from the federal level to healthcare organizations, provinces and, simultaneously, taxpayers. A causal *story of decline* is portrayed, where taxpayers lose money if refugees are not cared for. According to advocates, because the reforms limit access to preventive and primary care,

> when a person with uncontrolled diabetes ends up in the emergency department ... Canadians will bear the burden of these policy changes through their taxpayer supported provincial health plans (Arya et al. 2012, p.1876).

The *story of decline* is further supported by empirical evidence in the form of numbers and facts, reported by hospitals that were absorbing the healthcare costs for refugees. According to media reports, "Sick Kids absorbed $131,615 in outstanding costs," and "the University Health Network ... expects to foot a bill of $800,000" as a result of the refugee healthcare cuts (Cauderella and Evans 2014; Evans et al. 2014; Keung 2013b).

One scientific study at the Hospital for Sick Children in Toronto, which examined emergency department (ED) admission rates six months before and after the IFHP cutbacks, demonstrated that the number of ED admissions among children doubled from 6.4% to 12.1%, with clinical significance (Evans et al. 2014). Empirical research facilitates the gain of political support when causal theories are successfully appealed to in scientific studies (Stone 1989). Thus, organized interests voiced the causal story supported by numbers and scientific evidence that the IFHP reforms limited refugees' access to preventive care, which subsequently increases ED visits, and costs to provinces and taxpayers.

Discussion

The IFHP policy case reveals important insights into the role and subsequent impact of causal stories in defining and re-defining policy problems. Our results demonstrate a dramatic change in the way that the IFHP reform was initially justified and then later represented using different causal theories and accompanying strategies to portray them. The causal stories, in turn, had considerable shaping effects on resulting policy decisions by (1) changing

interpretive social constructions of refugees, (2) garnering political support through both empirical and normative arguments, (3) assigning responsibility for the problem and (4) challenging or protecting the existing social order.

To control the interpretive frame, governmental actors defended the implementation of the reforms using intentional causal theory, depicting refugees as bogus and blaming them for deliberately submitting false claims to undermine the refugee determination system. The negative constructions generated by governmental actors fuelled a discourse of othering where providing refugees with healthcare generated the perception of "unfairness" or unequal healthcare opportunities among Canadian citizens, motivating public support. Governmental actors assigned blame by portraying the negative consequences of high healthcare costs on the supposed fraudulent actions of refugees. This analysis demonstrates that intentional causal theory was used as an instrument of social control to maintain existing global patterns of dominance over refugee reception, in which most Western host countries contain population movements to the global South, within regions of origin, and unevenly share the responsibility of refugee resettlement (Gottwald 2014).

To counter these stories, organized interests redefined the problem of the IFHP reforms using the theory of inadvertent causation, attributing the unintended effects of government action as the cause of suffering for refugees. Problem redefinition generated a normative shift, in which refugees were portrayed as doubly victimized, fleeing persecution only to endure intolerable suffering through restricted healthcare access. These social constructions humanized refugees through stories of relatable healthcare hardships in Canada, conveyed through select narratives of suffering or health decline following the reforms. Beatson (2016) argued that both governmental actors and advocates used simplistic framing strategies that other refugees as either bogus or the victim. The author recommends that future advocacy entail a human-rights-centred approach that shifts "the emphasis on access to healthcare from charity to obligation," legitimizing refugees as healthcare users while protecting them from fluctuating populist sentiment (Beatson 2016, p. 131). Our analysis reveals that in addition to changing the interpretive frame depicting refugees, causal stories are an important component of gaining political support through normative and empirical arguments. Following the IFHP cutbacks, normative arguments structured around core community values raised political awareness and public concern about the equity of refugee healthcare access, efficiency of the healthcare system and liberty of healthcare providers to appropriately practise. The assignment of blame on unintentional policy consequences placed accountability on the federal government to rectify not only refugee access to primary care but also the high burden of costs assumed by healthcare institutions and provincial governments. On the empirical level, costs for taxpayers were no longer solved by the reforms but were caused by them.

Moreover, shifting the location of responsibility from refugees to government action restructured alliances among refugee-serving groups, contributing to the growing mobilization of healthcare providers, advocacy groups, legal organizations, provincial governments

and even a few refugees themselves. The causal stories implicitly appealed for a redistribution of power, whereby organized interests explicitly requested the federal government to cease producing harm, or the "cruel and unusual treatment" of refugees, a dominant belief supported by the Federal Court in their decision that the IFHP reforms violated the *Charter of Rights and Freedoms* (CDRC v. AGC 2014). Holtzer et al.'s (2017) policy analysis recognizes the influence of external drivers such as the legal venue of the courts and the 2015 federal election that created opportunities for alternative causal stories to enter the political discourse. In addition to causal stories and external drivers, factors such as organized interest group interactions and institutional mechanisms that contributed to the full reversal of the IFHP retrenchments in 2016 require further investigation.

Limitations

There are several limitations to this study. First, a limitation of this study relates to the minimal exploration of interest group and institutional mechanisms for policy change. Second, although an examination of the way causal stories are used by political actors is valuable to understand the problem definition process and respond to the problematization of key issues, it is difficult to attribute select causal pathways to complex settings, such as politics. Finally, a limitation of this study involves the focus on English-language media sources only, which may have excluded important perspectives that were only covered in French-language news media.

Conclusion

Understanding causal story mechanisms used by advocates of refugee policy expansion is essential for those contesting restrictive measures implemented in response to enduring refugee crises around the globe. Restrictive refugee policy proponents construct migrants as the problem, portraying them as deviants eroding the regulated systems of host nations. The resulting political discourse situates moral responsibility and economic costs on refugees. By using Stone's causal stories framework, this analysis reveals strategies for organized interest groups to contest populist and anti-immigrant ideologies in the problem re-definition process. Ultimately, changing the policy in question involves transforming the interpretive framework by redefining the problem, composed of causal stories that generate empirical and normative strategies to dismantle opposing arguments, shift accountability and challenge the existing social order.

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Conflict of Interest

None identified.

References

Alexander, C. 2014. Abuse common in old refugee claim system: Calling Foul on Harper's Refugee Health Policy. *The Hamilton Spectator*. Retrieved February 6, 2020. <https://www.thespec.com/opinion-story/4620461-abuse-common-in-old-refugee-claim-system/>.

Arya, N., J. McMurray and M. Rashid. 2012. Enter at Your Own Risk: Government Changes to Comprehensive Care for Newly Arrived Canadian Refugees. *Canadian Medical Association Journal* 184(17): 1875–76. doi:10.1503/cmaj.120938.

Barnes, S. 2013. *The Real Cost of Cutting the Interim Federal Health Program*. Toronto, ON: Wellesley Institute. Pp. 1–19. Retrieved February 6, 2020. <https://www.wellesleyinstitute.com/wp-content/uploads/2013/10/Actual-Health-Impacts-of-IFHP.pdf>.

Beatson, J. 2016. The Stories We Tell about Refugee Claimants: Contested Frames of the Health-Care Access Question in Canada. *Refuge* 32(3): 125–34.

Bhuyan, R., B. Osborne, Z. Sajedeh and S. Tarshis. 2014. *Unprotected, Unrecognized: Canadian Immigration Policy and Violence against Women, 2008–2013*. Toronto, ON: Migrant Mothers Project, University of Toronto. Retrieved February 6, 2020. <http://www.migrantmothersproject.com/wp-content/uploads/2012/10/MMP-Policy-Report-Final-Nov-14-2014.pdf>.

Campbell, J. L. (2002). Ideas, Politics, and Public Policy. *Annual Review of Sociology* 28: 21–38. doi:10.1146/annurev.soc.28.110601.141111.

Campbell, M., A. Dalton and D. McKeown. 2012, May 17. *Health Impacts of Reduced Federal Health Services for Refugees*. City of Toronto, ON. Retrieved February 6, 2020. <http://www.toronto.ca/legdocs/mmis/2012/hl/bgrd/backgroundfile-47324.pdf>.

Campion-Smith, B. and N. Keung. 2012. Tories Say New Refugee Bill Will Make It Easier to Deal with Bogus Claims. *Toronto Star*. Retrieved February 6, 2020. <https://www.thestar.com/news/canada/2012/02/16/tories_say_new_refugee_bill_will_make_it_easier_to_deal_with_bogus_claims.html />.

Canadian Council for Refugees (CCR). 2013. *Refugee Health Care: Impacts of Recent Cuts*. Montreal, QC: Canadian Council for Refugees. Retrieved February 6, 2020. <http://ccrweb.ca/sites/ccrweb.ca/files/ifhreporten.pdf>.

Canadian Doctors for Refugee Care, v. Canada (Attorney General) (CDRC v. AGC). 2014. FC 651.

Cauderella, A. and A. Evans. 2014. No Such Thing as a 'Bogus Child'. *The National Post*. Retrieved February 6, 2020. <https://nationalpost.com/opinion/caudarella-evans-no-such-thing-as-a-bogus-child>.

Citizenship and Immigration Canada (CIC). 2012. *Information Sheet for Interim Federal Health Program Beneficiaries*. Retrieved February 6, 2020. <http://publications.gc.ca/collections/collection_2013/cic/Ci44-15-2012-eng.pdf>.

Citizenship and Immigration, Canada (CIC). 2006. *Interim Federal Health Program: Information Handbook for Health-Care-Providers*. Edmonton, Alberta: FAS Benefit Administration Ltd.

Evans, A., A. Caudarella, S. Ratnapalan and K. Chan. 2014. The Cost and Impact of the Interim Federal Health Program Cuts on Child Refugees in Canada. *PLos One* 9(5): e96902. <https://doi.org/10.1371/journal.pone.0096902>.

Glynos, J., D. Howarth, A. Norval and E. Speed. 2009. *Discourse Analysis: Varieties and Methods*. National Centre for Research Methods NCRM/014. University of Essex. Retrieved February 6, 2020. <http://repository.essex.ac.uk/4026/>.

Gottwald, M. 2014. Burden Sharing and Refugee Protection. In Fiddian-Qismeyeh, E., G. Loescher, K. Long and N. Sigona, eds., *The Oxford Handbook of Refugee and Forced Migration Studies* (pp. 525–40). Oxford, UK: Oxford University Press.

Government of Canada. 2019. *Open Government Portal*. Retrieved February 6, 2020. <https://open.canada.ca/en/open-data>.

Gulli, C. 2015, September 25. Harper Says Only Bogus Refugees are Denied Health Care. He's Wrong. *Maclean's*. Retrieved February 6, 2020. <https://www.macleans.ca/politics/harper-says-only-bogus-refugees-are-denied-health-care-hes-wrong/>.

Hayes, M. 2012. 'We Refuse to Co-operate'. *The Hamilton Spectator*. Retrieved February 6, 2020. <https://advance-lexis-com.libaccess.lib.mcmaster.ca/api/document?collection=news&id=urn:contentItem:56PS-K8B1-F197-51K5-00000-00&context=1516831>.

Holtzer, E., A. Moore-Dean, A. Srikanthan and K. Kuluski. 2017. Reforming Refugee Healthcare in Canada: Exploring the Use of Policy Tools. *Healthcare Policy* 12(4): 46–55. doi:10.12927/hcpol.2017.25099.

Immigration, Refugee and Citizenship, Canada (IRCC). 2017. *Canada – Admissions of Permanent Residents by Intended Province/Territory of Destination and Immigration Category, 2005–January 2016*. Retrieved February 6, 2020. <https://open.canada.ca/data/en/dataset/f7e5498e-0ad8-4417-85c9-9b8aff9b9eda>.

Keung, N. 2012a. Canadian Doctors, Nurses Join Protest against Cuts to Refugee Health Plan. *Toronto Star*. Retrieved February 6, 2020. <https://www.thestar.com/news/gta/2012/05/23/canadian_doctors_nurses_join_protest_against_cuts_to_refugee_health_plan.html>.

Keung, N. 2012b. Nurses and MDs Enlisted to Defy Refugee Health Cuts. *Toronto Star*. Retrieved February 6, 2020. <https://advance-lexis-com.libaccess.lib.mcmaster.ca/api/document?collection=news&id=urn:contentItem:5CX2-6T21-DY91-K306-00000-00&context=1516831>.

Keung, N. 2013a. Caught between Death and Debts; Federal Cutbacks to Refugee Health Program Mean Some No Longer Entitled to Free Treatment. *Toronto Star*. Retrieved February 6, 2020. <https://advance-lexis-com.libaccess.lib.mcmaster.ca/api/document?collection=news&id=urn:contentItem:5CXG-FCW1-DY91-K14G-00000-00&context=1516831>.

Keung, N. 2013b. 'I Did Not Choose to Have Cancer'; Refugee Claimant Told to Pay for Treatment as New Federal Rule Forces Hospitals to Absorb Refugee Health Costs – or Bill Patients. *Toronto Star*. Retrieved February 6, 2020. <https://advance-lexis-com.libaccess.lib.mcmaster.ca/api/document?collection=news&id=urn:contentItem:5CXG-K7P1-DY91-K324-00000-00&context=1516831>.

Keung, N. 2014a. Protesters Carry on Fight to Aid Refugee Health Care. *Toronto Star*. Retrieved February 6, 2020. <https://advance-lexis-com.libaccess.lib.mcmaster.ca/api/document?collection=news&id=urn:contentItem:5CSF-DY71-DY91-K0BX-00000-00&context=1516831>.

Keung, N. 2014b. Ottawa to Appeal Refugee Health-Care Decision; Advocacy Groups will Return to Court if Coverage Not Restored on Time, Lawyer Says. *Toronto Star*. Retrieved February 6, 2020. <https://advance-lexis-com.libaccess.lib.mcmaster.ca/api/document?collection=news&id=urn:contentItem:5DHH-NDS1-DY91-K22B-00000-00&context=1516831>.

Komarnicki, J. 2014. Alberta Looks to Fill Gaps in Refugee Care; Options to Be Examined for Health Funding. *Calgary Herald*. Retrieved February 6, 2020. <https://advance-lexis-com.libaccess.lib.mcmaster.ca/api/document?collection=news&id=urn:contentItem:5CFF-7HR1-DY2T-3161-00000-00&context=1516831>.

Kraeker, C. and T. O'Shea. 2012. Medical Professionals Compelled to Protest, Defend Their Patients' Rights; Refugee Health Cuts Are 'Mean-Spirited ... and Hurting Our Patients'. *The Hamilton Spectator*. Retrieved February 6, 2020. <https://advance-lexis-com.libaccess.lib.mcmaster.ca/api/document?collection=news&id=urn:contentItem:56PB-NC61-JDV5-F44P-00000-00&context=1516831>.

Levitz, S. 2013. Public Health, Purse at Risk: Doctors; Federal Cuts to Health Care for Refugees Short-Sighted: Advocates. *The Record (Kitchener-Waterloo)*. Retrieved February 6, 2020. <https://advance-lexis-com.libaccess.lib.mcmaster.ca/api/document?collection=news&id=urn:contentItem:58P7-16V1-F197-503N-00000-00&context=1516831>.

Patton, M.Q. 1999. Enhancing the Quality and Credibility of Qualitative Analysis. *Health Services Research* 34(5 Pt 2): 1189–1208.

Payne, E. 2014. Health Cuts Hurt Refugees, Doctor Says; Criticism Follows CMAJ Editorial on 'Irrational' Policy. Ottawa Citizen. Retrieved February 6, 2020. <https://advance.lexis.com/api/document?collection=news&id=urn:contentItem:5BD7-R8Y1-JBKR-J0WM-00000-00&context=1516831>.

Raza, D., M. Rashid, L. Redwood-Campbell, K. Rouleau and P. Berger. 2012. A Moral Duty: Why Canada's Cuts to Refugee Health Must be Reversed. *Canadian Family Physician* 58(7): 728–09, e365-7.

Sanders, C. 2012. Province Steps Up for Refugees. *Winnipeg Free Press*. Retrieved February 6, 2020. <http://www.winnipegfreepress.com/local/province-steps-up-for-refugees-169590316.html>.

Seeking Solutions Symposium. 2013. *Access to Health Care for the Uninsured in Canada*. Toronto, ON. Retrieved February 6, 2020. <http://www.womenscollegehospital.ca/assets/pdf/SEEKING%20SOLUTIONS%20REPORT.pdf>.

Sheikh, H., M. Rashid, P. Berger and J. Hulme. 2013. Refugee Health Providing the Best Possible Care in the Face of Crippling Cuts. *Canadian Family Physician* 59(6): 605–06.

Stall, N. 2012. Refugee Health Reforms Assailed. *Canadian Medical Association Journal* 184(10): E511–12. doi:10.1503/cmaj.109-4208.

Stanbrook, M.B. 2014. Canada Owes Refugees Adequate Health Coverage. *Canadian Medical Association Journal* 186(2): 91. doi:10.1503/cmaj.131861.

Stone, D. 1989. Causal Stories and the Formation of Policy Agendas. *Political Science Quarterly* 104(2): 281–300. <https://www.jstor.org/stable/2151585>.

Stone, D. 2012. *Policy Paradox: The Art of Political Decision Making, (3rd edition)*. New York, NY: WW Norton & Company.

United Nations High Commissioner for Refugees (UNHCR). 2019. *UNHCR Global Trends Forced Displacement in 2018*. Geneva. Retrieved February 6, 2020. <https://www.unhcr.org/globaltrends2018/>.

Yin, R. K. 2009. *Case Study Research: Design and Methods (4th ed.).* Thousand Oaks, CA: Sage Publishing.

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