Creating the content for knowledge translation tools to prompt early referral for diagnostic assessment and rehabilitation services for children with suspected cerebral palsy.

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A thesis submitted to the McGill University in partial fulfillment of the requirements for the degree of Doctor of Philosophy (Rehabilitation Science)

December 5, 2019

DEDICATION

"I say, beware of all enterprises that require new clothes, and not rather a new wearer of clothes."
-Henry David Thoreau (*Walden; or, Life in the Woods*)

"The learning process is something you can incite, literally incite, like a riot."
-Audre Lorde (*Sister Outsider: Essays and Speeches*)

"The man who never alters his opinion is like standing water, and breeds reptiles of the mind."
-William Blake (*The Marriage of Heaven and Hell*)

"[E]njoy every sandwich."
-Warren Zevon (*Late Night with David Letterman, 30/10/2002*)

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ABSTRACT

Background: Early identification and early intervention are considered best practice for children with cerebral palsy. About half of children with CP are either born at term after uneventful pregnancies or are born preterm but >29 weeks gestational age; instead of receiving neonatal follow-up, these children are followed in their community by primary care practitioners (e.g. pediatricians, family physicians). These clinicians are uniquely positioned to identify and refer children suspected of having CP to medical specialists (e.g. child neurologists, developmental pediatricians) for diagnostic assessment and to rehabilitation specialists for intervention. However, primary care practitioners often do not have the advanced training in atypical child development that medical specialists receive and may not recognize the early features of CP during their developmental surveillance visits. Most research efforts to date have focused primarily on early detection of children considered to be at "high-risk" for CP, and consequently there are no CP-specific tools or resources available for a primary care practitioner to effectively apply in their context. A 'wait-and-see' approach to referral for diagnostic assessment has traditionally been favored, and parents have reported dissatisfaction with delays in the diagnostic process. With a critical window of neuroplasticity, and novel medical and rehabilitation interventions offering potential for neurorepair, it is essential that any unnecessary delays be mitigated. Educational knowledge translation tools for the primary care context are one strategy that may improve detection and referral of children suspected of having CP.

Aim: The overall aim of this doctoral thesis was to create the content (knowledge) needed for user-friendly knowledge translation tools for pediatric primary care practitioners. The objective being to increase their awareness of the early signs (features) of CP, in order to enhance early detection and subsequent simultaneous referral to medical specialists for diagnostic assessment and to other

health professionals for intervention for children suspected of having CP. The specific objectives were to: (i) Critically review the evidence on what is known about age at referral for diagnosis of CP, and identify factors potentially associated with delayed referral, (ii) Establish population-based evidence on current physician referral practices for diagnosis and rehabilitation services for children suspected of having CP, and identify factors associated with delayed referral, (iii) Determine the clinical features that manifest early in life and have a high association with CP or other neurodevelopmental disability, and (iv) Determine referral recommendations to occur simultaneously with referral for diagnostic assessment for children suspected of having CP.

Methodology: In order to achieve the overall aim, a sequential mixed-methods study was conducted using an integrated knowledge translation approach, with each component informing the next. The manuscripts contained in this thesis follow and document this progression and can be situated in two distinct phases. Phase One (Knowledge Synthesis): A scoping review of the literature on what is known about the age at which children are referred for diagnosis of CP, and factors associated with age at referral (**Manuscript 1**, "Age at referral for diagnosis and rehabilitation services for cerebral palsy: a scoping review", published in *Developmental Medicine & Child Neurology*); a single-site retrospective chart review documenting physician referral practices for diagnostic assessment and rehabilitation for children suspected of having CP, and factors associated with delayed referral (**Manuscript 2**, "Age at referral of children for initial diagnosis of cerebral palsy and rehabilitation: Current practices", published in *Journal of Child Neurology*); a national multi-site environmental scan documenting physician referral practices for diagnostic assessment and rehabilitation for children suspected of having CP, and factors associated with delayed referral (**Manuscript 3**, "Current referral practices for diagnosis and intervention for children with cerebral palsy: A national environmental scan", published in *Journal*

of Pediatrics). Phase Two (Knowledge Creation): Two nominal group processes (consensus methodology) were conducted with Canadian content-experts and knowledge-users to determine which features of CP should be used to prompt referral for diagnostic assessment, as well as which referral recommendations can be made concurrently (**Manuscript 4**, "Use of consensus methods to determine the early clinical signs of cerebral palsy", In Press in *Paediatrics & Child Health)*; and an online two-round Delphi survey was conducted with international experts in early identification and intervention for children with CP to assess the validity and generalizability of the results from the previous study (**Manuscript 5**, "International expert recommendations of clinical features to prompt referral for diagnostic assessment of cerebral palsy", Published in *Developmental Medicine & Child Neurology*).

Results: In Phase One (Knowledge Synthesis) it was found that: (i) The literature on age at referral for diagnosis of CP was sparse and based on older cohorts; some potential predictors of delayed referral were identified (**Manuscript 1**); (ii) An important subset of children with CP are experiencing pronounced delayed referrals for diagnostic assessment and for rehabilitation intervention, and factors collectively associated with delayed referral were identified (**Manuscripts 2 & 3**). In Phase Two (Knowledge Creation), the evidence generated in the first phase informed consensus groups and a Delphi survey to: (i) Identify six attributes that should be used to prompt referral for diagnostic assessment, two 'warning signs' agreed upon that should prompt closer monitoring and surveillance over time rather than immediate referral for diagnostic assessment, and formulate five referral recommendations to other health professionals (**Manuscript 4**); and (ii) Refine the results from **Manuscript 4** with input from international experts,and determine that they are indeed valid and generalizable, and can be used to enhance detection and prompt referral within the pediatric primary care context for children suspected of having CP.

Implications: A knowledge to practice gap was identified with respect to the identification and referral of children suspected of having CP. About half of children with CP either do not have a complicated birth history or do not meet criteria for close surveillance as part of neonatal follow-up programs and are thus followed by a primary care practitioner. It is therefore imperative that user-friendly knowledge translation tools are developed collaboratively to enhance early detection and referral strategies by primary care practitioners, and to optimize child and family functioning and adaptation. The research contained in this thesis has both demonstrated the need for such an educational knowledge translation strategy, and it has generated expert-informed content (knowledge) for this tool. To ensure that the final tools are end-user friendly, future research endeavors will involve the collaboration of relevant stakeholders in the primary care context (e.g. pediatricians, family physicians, parents of children with CP) to determine the optimal manner of delivering and disseminating this knowledge.

Keywords: cerebral palsy, delay, diagnosis, knowledge translation, primary care, referral

RÉSUMÉ

Contexte : Dans les cas de paralysie cérébrale chez l'enfant, le dépistage et l'intervention précoces sont considérées comme étant des pratiques exemplaires. Environ la moitié des enfants atteints de PC sont nés à terme après une grossesse sans incident ou nés prématurés à plus de 29 semaines d'âge gestationnel. Dès lors, au lieu de bénéficier d'un suivi néonatal, ces enfants sont suivis par des praticiens de première ligne dans les services de santé locaux (p. ex. pédiatres ou médecins de famille). Ces cliniciens sont particulièrement bien positionnés pour identifier et référer les enfants soupçonnés d'être atteints de PC à des médecins spécialistes (p. ex. neurologues pédiatriques ou pédiatres du développement) pour obtenir une évaluation diagnostique, ou à des spécialistes de la réadaptation pour une intervention. Toutefois, les praticiens de soins primaires ont rarement la formation avancée qu'ont les spécialistes en matière de développement atypique de l'enfant et ils pourraient ne pas reconnaître les caractéristiques initiales propres à la PC au cours des visites de surveillance du développement. Les études effectuées à ce jour se sont concentrées principalement sur l'identification précoce des enfants qui présentent un risque élevé de PC. Par conséquent, les praticiens de soins primaires n'ont pas d'outils ou de ressources spécifiques pour la PC qu'ils puissent efficacement appliquer dans le contexte qui est le leur. Traditionnellement, c'est l'approche attentiste qui a été favorisée en matière de référence pour une évaluation diagnostique, suscitant le mécontentement des parents concernés face au retard dans le processus diagnostique. Étant donné la fenêtre critique à l'intérieur de laquelle on peut introduire la neuroplasticité ou d'autres interventions médicales et de réadaptation novatrices offrant un potentiel de réparation neurale, il est essentiel que tout retard inutile soit évité. Les outils pédagogiques d'application des connaissances pour le contexte de soins primaires constituent une stratégie susceptible d'améliorer l'identification et la référence des enfants soupçonnés d'être atteints de PC.

Objectif: L'objectif global de la présente thèse de doctorat est de créer le contenu (connaissance) nécessaire pour élaborer des outils d'application des connaissances conviviaux pour les praticiens de soins primaires pédiatriques. Le but est de les sensibiliser davantage aux signes (caractéristiques) précoces de la PC afin d'améliorer son dépistage précoce chez les enfants et de faire en sorte que ces derniers soient référés en même temps à un spécialiste aux fins d'évaluation diagnostique et à d'autres professionnels de la santé aux fins d'intervention. Les objectifs spécifiques sont les suivants: (i) Faire une analyse critique des données existantes relatives à l'âge au moment de la référence pour un diagnostic de PC et identifier les facteurs potentiels menant à une référence tardive; (ii) Fournir des données probantes tirées d'études basées sur la population relativement aux pratiques actuelles en matière de référence pour un diagnostic et des services de réadaptation pour les enfants soupçonnés d'être atteints de PC, et identifier les facteurs liés à la référence tardive; (iii) Identifier les caractéristiques cliniques qui se manifestent tôt dans l'enfance et qui sont fortement associées à la PC ou à d'autres déficiences dans le développement neurologique; et (iv) Recommander la référence à des professionnels de la santé à faire en même temps que la référence pour une évaluation diagnostique dans le cas des enfants soupçonnés d'être atteints de PC.

Méthodologie: Afin d'atteindre l'objectif global, une étude séquentielle axée sur les méthodes mixtes a été réalisée au moyen d'une approche intégrée d'application des connaissances, où chaque composante éclaire la suivante. Les manuscrits contenus dans la présente thèse, correspondant à deux phases distinctes, retracent et documentent cet avancement. Première phase (synthèse des connaissances): revue exploratoire de la littérature relativement à l'âge auquel les enfants sont référés pour un diagnostic en PC, et aux facteurs liés à l'âge au moment de la référence (**Manuscr it 1**, « Âge au moment de la référence pour un diagnostic en PC : une revue exploratoire », publié

dans la revue *Developmental Medicine & Child Neurology*); examen rétrospectif monocentrique des dossiers, documentant les pratiques de références des médecins pour une évaluation diagnostique et une réadaptation pour les enfants soupçonnés d'être atteints de PC, et les facteurs liés à une référence tardive (**Manuscrit 2**, « Âge de l'enfant au moment de la référence pour un diagnostic initial de paralysie cérébrale et pour une réadaptation : pratiques actuelles », publié dans la revue *Journal of Child Neurology*); examen situationnel multicentrique d'envergure nationale documentant les pratiques médicales de référence pour une évaluation diagnostique et une réadaptation pour les enfants soupçonnés d'être atteints de PC, et les facteurs liés à une référence tardive (**Manuscrit 3,** « Pratiques actuelles en matière de référence pour un diagnostic et une intervention auprès d'enfants atteints de paralysie cérébrale : examen situationnel à l'échelle nationale », publié dans la revue *Journal of Pediatrics)*. Deuxième phase (création de connaissances) : la technique du groupe nominal (méthode de consensus) a été utilisée auprès d'un groupe d'experts du contenu canadiens et avec un groupe d'utilisateurs de connaissances canadiens en vue de déterminer lesquelles des caractéristiques de la PC devraient entraîner une réeference pour une évaluation diagnostique et quelles autres références peuvent être formulées concomitamment (**Manuscrit 4**, « Utilisation des méthodes de consensus pour identifier les signes cliniques précoces de la paralysie cérébrale », sous presse à la revue *Paediatrics & Child* Health); par ailleurs, un sondage Delphi en ligne en deux étapes a été administré à experts internationaux en identification et intervention précoces auprès d'enfants souffrant de PC afin d'évaluer la validité et la généralisabilité des résultats de l'étude précédente « Recommandations d'experts internationaux concernant les caractéristiques cliniques devant entraîner une référence pour une évaluation diagnostique de la paralysie cérébrale », publié dans la revue *Developmental Medicine & Child Neurology*).

Résultats : Dans la Première phase (synthèse des connaissances), on découvrait que : (i) Les écrits relatifs à l'âge au moment de la référence pour un diagnostic de PC sont peu nombreux et fondés sur des cohortes de patients plus âgés; quelques éléments de prédiction de référence tardive ont été identifiés (**Manuscrit 1**); et (ii) Un sous-ensemble important d'enfants souffrant de PC ont été référés pour une évaluation diagnostique et une intervention de réadaptation avec un retard significatif; des facteurs associés collectivement à la référence tardive ont également été identifiés (**Manuscrits 2 et 3**). Les données issues de la première phase ont servi aux groupes de consensus et à la préparation d'un sondage Delphi au cours de la Deuxième phrase (création de connaissances) afin d : (i) Identifier six éléments qui devraient inciter à la référence pour une évaluation diagnostique, convenir de deux « signaux précurseurs » qui, au lieu de référer immédiatement pour une évaluation diagnostique, devraient mener à effectuer une surveillance et un contrôle dans le temps plus serrés, et recommander cinq références à l'intention d'autres professionnels de la santé (**Manuscrit 4**); et (ii) Affiner les résultats découlant du Manuscrit 4 avec la contribution d'experts internationaux et confirmer leur validité, généralisabilité et utilité pour améliorer le dépistage précoce et la référence rapide des enfants soupçonnés d'être atteints de PC dans le contexte de soins pédiatriques de base.

Implications : Un décalage entre les connaissances et la pratique a été observé en matière d'identification et de référence des enfants soupçonnés d'être atteints de PC. Environ la moitié des enfants souffrant de PC se caractérisent par une naissance sans complications ou ne remplissent pas les critères justifiant une surveillance étroite dans le cadre des programmes de suivi néonatal, faisant en sorte qu'ils sont suivis par un praticien de soins primaires. Il est donc impératif que des outils conviviaux d'application des connaissances soient élaborés conjointement afin d'améliorer le dépistage précoce et les stratégies de référence par les praticiens de soins primaires, et

d’optimiser le fonctionnement et l’adaptation des enfants et des familles. La recherche réalisée dans le cadre de cette thèse a mis en évidence le besoin d’une telle stratégie éducative en matière d’application des connaissances, tout en apportant un contenu (connaissances) d’experts pouvant servir à la création d’un tel outil. Dans l’optique de rendre les outils conviviaux pour l’utilisateur final, les travaux de recherche à venir comprendront une collaboration entre les parties prenantes du secteur des soins primaires (pédiatres, médecins de famille, parents d’enfants atteints de PC, par exemple) afin d’identifier la manière optimale de présenter et de diffuser ces connaissances.

Mots-clés: paralysie cérébrale, retard, diagnostic, application des connaissances, soins primaires, référence

ACKNOWLEDGEMENTS

I wish to start by expressing the sincere gratitude I feel towards my supervisor, Dr. Annette Majnemer: Annette, thank you for being who you are, and for everything you have done for me. Considering that I initially did not apply to work with you because I figured there was slim-to-no chance of you taking me on, it goes without saying that I am quite content with how things unfolded! My PhD studies have been a time of great personal and professional growth. I attribute this to the expert mentorship you provided, the amazing learning and networking opportunities you fostered, and the respect for rigorous methodology and professionalism that you instilled in me. I found myself consistently inspired by the humility and humanity you demonstrate regardless of which of your many hats you happen to be wearing. Thank you for taking a chance on me, suffering six years of my penchant for excessive-hyphenation, and for the dignity and respect you showed me throughout our time together.

To the members of my supervisory committee, Dr. André Bussières and Dr. Laurie Snider: Thank you both for always being available when I needed you, for the attention to detail you always brought to your manuscript revisions, and for the style with which you provided feedback — your support throughout my studies has always been greatly appreciated.

To *The PROMPT Group*: Managing a team of 20 coinvestigators spread across the country could have been a nightmare, but it was an absolute pleasure working with you all. Thank you for your varied contributions to the research contained in this thesis. I wish to highlight the support I received from Dr. John Andersen, Dr. André Bussières, Dr. Darcy Fehlings, Dr. Adam Kirton, Dr. Patti Li, Dr. Maryam Oskoui, Dr. Charo Rodriquez, Dr. Michael Shevell, and Dr. Laurie Snider — I feel extremely fortunate to have had the opportunity to collaborate with and learn from such brilliant and inspirational clinician-scientists.

To everyone else who contributed to the research contained in this thesis, I wish to express my gratitude to: Jessica Goldschleger for your collaboration on the scoping review; Lara Hubermann for your various efforts with the pilot chart review; all the families who participated in the Canadian Cerebral Palsy Registry and provided the data for the national environmental scan; the local and national content-experts and knowledge-users who participated in the two consensus groups; and the international experts who participated in the Delphi survey.

To my fellow lab members (past and present), as we vagabonded from lab-to-lab-to-lab-to-lab-to-lab over the years, especially Anna Ehsan, Anna Radzioch Guérin, Deniz Keskinel, Nina Lantinga, Hana Macdougall, and Sossy Sahakian: Thank you for the various combinations of statistical/logistical/moral/social support you provided, it was always sincerely appreciated. Also, to those who came before me: Dr. Keiko Shikako-Thomas, Dr. Noémi Dahan-Oliel, and Dr. Marie Brossard-Racine; thank you for always letting me bend your ear, for the (sometimes unsolicited) Life advice, and for being there in times of need.

To the National Coordinators of the Canadian Cerebral Palsy Registry, Sasha Dyck (current) and Chris Saunders (former), and all the Site Coordinators (especially Mary McNeil and Kathleen O'Grady): It is thanks to your collective efforts and patience that we were able to successfully complete the national environmental scan – thank you all!

To my Graduate Program Directors, Dr. Isabelle Gélinas and Dr. Laurie Snider: Thank you for your support throughout my studies, for your 'good enough' advice, and for fostering a learning environment and opportunities that transcend the walls of the classroom and lab.

To all my SPOT Colleagues & students: I consider it a privilege to have been afforded the opportunity to teach as member of both the mental health and pediatric neurology teams

throughout my doctoral studies. I can only hope that you learned at least a fraction from me compared to what I learned from working with you over the years. Special thanks to Dr. Melissa Park, Assistant Professor Cynthia Perlman, and Dr. Hiba Zafran for the varied unwavering personal and professional support they provided me with throughout my PhD journey.

To my parents, Sheilagh and Michael: Thank you for always believing in and supporting me, not only during my doctoral studies, but throughout my life. I love and appreciate you both more than you will ever know.

To my friends: Thank you for your support and encouragement over the past six years, for making a point of checking-in during the stressful times, and for helping ensure that I was able to maintain a healthy occupational balance throughout this whole process.

Finally, I wish to acknowledge and express my sincere gratitude for the financial support that I was fortunate to receive throughout my PhD studies from the following sources: the Canadian Institutes of Health Research (Doctoral Student Stipend, MOP-133402); the Richard and Edith Strauss Canada Foundation (Musculoskeletal Doctoral Fellowship, renewed once); the Hoppenheim Fund & Montreal Children Hospital Foundation (Hoppenheim Scholarship); the McGill School of Physical and Occupational Therapy (Judith Kornbluth Gelfand Fellowship in Pediatric Rehabilitation; Graduate Research Enhancement and Travel Award; Travel Grant); the American Academy for Cerebral Palsy and Developmental Medicine (four Student Scholarships); NeuroDevNet (two Conference Scholarships); Kids Brain Health Network (Family Engagement in Research Trainee Scholarship; two Conference Travel Scholarships); and the EQUATOR CANADA Publication School & Ontario's Strategy for

Patient-Oriented Research Support Unit (EQUATOR Canada Publication School Trainee Scholarship).

PREFACE

i. Statement of originality

The elements of this thesis that are original scholarship and distinct contributions to the field of early identification and early intervention for children with cerebral palsy (CP) are fivefold:

(i) We demonstrated that the available scientific evidence on age at referral for diagnostic assessment and age at referral for rehabilitation services for children with suspected CP was sparse, and most importantly our synthesis found it was suggestive of high variation and that a subset of children with CP may be experiencing prolonged delays. Our findings highlighted that there was a need for population-based evidence that would provide a better understanding of what factors currently contribute to delays in referral;

(ii) We generated population-based evidence which described the current referral practices of physicians across Canada with respect to age at referral for diagnostic assessment and age at referral for rehabilitation services for children with CP, and also identified predictors associated with delayed referral;

(iii) Through two nominal group techniques (consensus methods) with Canadian content-experts and knowledge-users, we identified the following: Six clinical features, each of which should prompt a primary care practitioner to refer a child suspected of having CP to a medical specialist for diagnostic assessment; two clinical features, either of which should prompt closer monitoring rather than immediate referral for diagnostic assessment of CP; and five referral recommendations to co-occur with referral from the primary care context for diagnostic assessment of CP;

(iv) Through an online Delphi survey with international experts in early identification and early intervention for children with CP, we validated the results of the Canadian consensus groups and

found high concordance, suggesting that the features and referral recommendations are broadly generalizable within the primary care context;

(v) The collective evidence we generated will be used to inform the content of an educational knowledge translation tool designed for primary care practitioners. The aim will be to enhance detection of CP in the primary care context by increasing awareness of the features of CP that are observable early and which should prompt referral for diagnostic assessment and simultaneous referral to rehabilitation specialists.

ii. Contribution of authors

All five manuscripts included in this thesis are the work of the doctoral candidate, Zachary Boychuck (four as first author, one as second author), with extensive editing and feedback from his supervisor Dr. Annette Majnemer. In collaboration with the research team (see below), they conceptualized and designed the five studies, coordinated and supervised data collection, carried out the initial analyses, drafted the initial manuscripts, and reviewed and revised the manuscripts for publication.

This knowledge translation project had a team of 21 co-investigators (the PROMPT Group; Appendix A): Content-experts (e.g. child neurologists, developmental pediatricians, rehabilitation specialists, researchers), and knowledge-users (e.g. pediatricians, family physicians, parents of children with CP), representing varied stakeholders who were involved from study conceptualization through implementation of the various phases of this project depending on their role. As co-authors on the four manuscripts related to the CIHR-funded PROMPT project, all members of the PROMPT Group were offered the opportunity to review and provide feedback for each of the four relevant manuscripts in this thesis. All authors approved each final manuscript as submitted and agreed to be accountable for all aspects of the work. Substantial contributions to

each manuscript, resulting in inclusion as named authors on the publications (rather than being included in "the PROMPT Group") are highlighted below.

Manuscript 1: "Age at referral for diagnosis and rehabilitation services for cerebral palsy: A scoping review"

Dr. Bussières provided methodological guidance concerning the conduct of scoping reviews, and critically reviewed and revised the manuscript. Jessica Goldschleger was the second reviewer for the scoping review process.

Manuscript 2: "Age at referral of children for initial diagnosis of cerebral palsy and rehabilitation: Current practices"

Lara Hubermann participated in data collection and wrote the manuscript's first draft, both in direct collaboration with the doctoral candidate (Zachary Boychuck), and she has consented to this manuscript being included in this thesis (Appendix B). Dr. Shevell contributed to the recruitment of the sample, conceptualization and design of the study, and to the data analysis.

Manuscript 3: "Current referral practices for diagnosis and intervention for children with cerebral palsy: A national environmental scan"

Drs. Andersen, Fehlings, Kirton, Oskoui, Shevell and Snider contributed to the conceptualization and design of the study, participated in the data collection, and critically reviewed and revised the manuscript.

Manuscript 4: "Use of consensus methods to determine the early clinical signs of cerebral palsy"

Drs. Andersen, Fehlings, Kirton, Li, Oskoui, Shevell and Snider participated in data collection (national consensus groups; nominal group techniques), and critically reviewed and revised the manuscript. Drs. Bussières and Rodriguez provided methodological guidance

concerning the consensus process and nominal group technique, and critically reviewed and revised the manuscript.

Manuscript 5: "International expert recommendations of clinical features to prompt referral for diagnostic assessment of cerebral palsy."

Drs. Andersen, Fehlings, Kirton, Oskoui, Shevell and Snider participated in data collection, and critically reviewed and revised the manuscript. Drs. Bussières, Li and Rodriguez contributed to the conceptualization and design of the study, provided methodological guidance concerning the conduct of an online international Delphi survey, and critically reviewed and revised the manuscript.

iii. Thesis organization and overview

This thesis consists of five manuscripts. As per the guidelines of the Graduate and Postdoctoral Studies (GPS), this thesis also contains additional chapters that serve to connect the manuscripts and create a cohesive thesis that demonstrates the fullness of this research project. Consequently, some duplication in text was inevitable. What follows is a brief outline of this thesis.

Chapter 1 provides a literature review incorporating: an overview of CP, the importance of early detection and early intervention for neurodevelopmental disabilities, current practice with respect to early identification of CP, delays in referral for diagnosis and rehabilitation, knowledge translation and a critical synthesis.

Chapter 2 presents the rationale and objectives of this thesis.

Chapter 3 consists of **Manuscript 1** entitled "Age at referral for diagnosis and rehabilitation services for cerebral palsy: A scoping review". This study was a scoping review of the evidence on age at referral for diagnostic assessment of CP. It summarized the sparse existing evidence and suggested that subgroups of children with CP might potentially be experiencing

increased delays. **This manuscript was accepted for publication by *Developmental Medicine & Child Neurology* on July 31, 2018; Epub ahead of print on October 1, 2018; Published on July 3, 2019.**

Boychuck, Z., Bussières, A., Goldschleger, J., Majnemer, A., & the PROMPT group. (2019). Age at referral for diagnosis and rehabilitation services for cerebral palsy: A scoping review. *Developmental Medicine & Child Neurology,* 61(8), 908-914. DOI: 10.1111/dmcn.14034

Chapter 4 connects the first and second manuscripts.

Chapter 5 consists of **Manuscript 2** entitled "Age at referral of children for initial diagnosis of cerebral palsy and rehabilitation: Current practices". This was a feasibility study for the larger national environmental scan study. **This manuscript was accepted for publication by *Journal of Child Neurology* on March 14, 2015; Epub ahead of print on August 3, 2015; Published on March 1, 2016.**

Hubermann, L., **Boychuck, Z.**, Shevell, M., & Majnemer, A. (2016). Age at referral of children for initial diagnosis of cerebral palsy and rehabilitation: Current practices. *Journal of Child Neurology*, 31(3), 364-9. DOI:10.1177/0883073815596610

Chapter 6 connects the second and third manuscripts.

Chapter 7 consists of **Manuscript 3** entitled "Current referral practices for diagnosis and intervention for children with cerebral palsy: A national environmental scan". This study was a national environmental scan of Canadian physician referral practices for diagnostic assessment and rehabilitation services for children with CP. It described current referral practices and identified subgroups of children with CP who experience increased delays. **This manuscript was accepted for publication in *The Journal of Pediatrics* on September 13, 2019.**

Boychuck, Z., Andersen, J., Fehlings, D., Kirton, A., Oskoui, M., Shevell, M., Majnemer,

A., & the PROMPT group. (2020). Current referral practices for diagnosis and intervention for children with cerebral palsy: A national environmental scan. *The Journal of Pediatrics*, 216, 174-181.

Chapter 8 connects the third and fourth manuscripts.

Chapter 9 consists of **Manuscript 4** entitled "Use of consensus methods to determine the early clinical signs of cerebral palsy". This study used consensus methodology (nominal group techniques) to demonstrate agreement among Canadian content-experts and knowledge-users on 6 clinical attributes that should prompt referral to a medical specialist for diagnostic assessment of CP, 2 clinical attributes that should prompt close monitoring, and 5 referral recommendations to rehabilitation services/health professionals to occur simultaneously with referral for diagnostic assessment. **This manuscript was accepted for publication by *Paediatrics & Child Health* on February 4, 2019; Epub ahead of print on May 8, 2019; and is currently In Press.**

Boychuck, Z., Andersen, J., Bussières, A., Fehlings, D., Kirton, A., Li, P., Oskoui, M., Rodriguez, C., Shevell, M., Snider, L., Majnemer, A., & the PROMPT group. (in press). Use of consensus methods to determine the early clinical signs of cerebral palsy. *Paediatrics & Child Health.* DOI: 10.1093/pch/pxz061

Chapter 10 connects the fourth and fifth manuscripts.

Chapter 11 consists of **Manuscript 5** entitled "International validation of attributes that should PROMPT referral for diagnosis of cerebral palsy: A Delphi study". This study used consensus methodology (online Delphi survey) with international experts in the field of early detection and early intervention of CP to validate the clinical attributes and referral recommendations agreed upon by the Canadian consensus groups. The high concordance achieved

in this study suggests that the results are broadly generalizable, and can be used to inform the content of educational knowledge translation tools to improve the early detection of CP in the primary care context. **This manuscript was accepted for publication by *Developmental Medicine & Child Neurology* on March 13, 2019; Epub ahead of print on April 25, 2019; Published on December 2, 2019**.

Boychuck, Z., Andersen, J., Bussières, A., Fehlings, D., Kirton, A., Li, P., Oskoui, M., Rodriguez, C., Shevell, M., Snider, L., Majnemer, A., & the PROMPT group. (2020). International expert recommendations of clinical features to prompt referral for diagnostic assessment of cerebral palsy. *Developmental Medicine & Child Neurology*. 62(1), 89-96. DOI: 10.1111/dmcn.14252.

Chapter 12 is a summary of the findings of the five manuscripts, a discussion and critical synthesis of the results of the research contained in this thesis, and directions for future research.

The corresponding tables, figures and references are contained within each Manuscript. For Chapter 1, Chapter 2, Chapter 4, Chapter 6, Chapter 8, Chapter 10, Chapter 12 the corresponding tables, figures and references are presented at the end of this thesis.

Ethical approval is detailed within each manuscript.

CHAPTER 1:

INTRODUCTION

1.1 Cerebral palsy: An overview

Cerebral palsy (CP) is the fourth most common childhood disability (Statistics Canada, 2007), and the most common physical disability encountered in children (Hirtz et al., 2007) with an estimated prevalence of 2.11 per 1000 live births (Oskoui, Coutinho, Dykeman, Jetté, & Pringsheim, 2013). Rosenbaum et al. (2007), in their consensus definition, described CP as

> "a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to nonprogressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, perception, communication, cognition, and behavior, by epilepsy, and by secondary impairments (p. 9)."

CP is a lifelong condition and depending on the severity, it can affect an individual quite profoundly across the domains of the International Classification of Functioning, Disability and Health (ICF; World Health Organization, 2001). There is wide variability and heterogeneity of the clinical manifestations of CP. Severity of motor impairment and activity limitations is most often described using the five levels of the Gross Motor Function Classification System (GMFCS; Palisano et al., 1997). More than half of children with CP are classified as GMFCS Level I and II (mild motor impairment, independent ambulators), whereas approximately 30% are classified as GMFCS Level IV and V (more severe motor impairment, wheelchair users) (Novak, 2014; Shevell, Dagenais, & Oskoui, 2013) . In terms of topography, or type of CP, approximately 38% of children have hemiplegia, 36% have diplegia, and 26% have quadriplegia (McIntyre, Morgan, Walker, & Novak, 2011). A survey of international experts in CP was

conducted to inform the development of the ICF Core Sets for children with CP (Schiariti et al., 2013; Schiariti, Selb, Cieza & O'Donnell, 2015). The aim was to standardize the clinical assessment of CP across professionals and disciplines through the systematic use of the Core Sets, as there is currently no gold-standard for referral or assessment of children suspected of having CP. While the brain damage associated with CP usually occurs before birth, the early signs (features) often become increasingly apparent in infancy as motor skills develop. Therefore, the focus of clinical assessment evolves and relates to the child's developmental stage. Clinical manifestations, or features, gradually present themselves as the child matures and develops new milestones.

1.2 Early identification & early intervention for children with CP

For children suspected of having CP, early identification that prompts simultaneous referral to medical specialists for diagnosis and to rehabilitation professionals for intervention is widely accepted as best practice (McIntyre, Morgan, Walker, & Novak, 2011). High-level evidence supports the effectiveness of early interventions for children with CP at both the body structure and function level of the ICF (e.g. casting, hip surveillance, selective dorsal rhizotomy) and also at the activity and participation level of the ICF (e.g. bimanual training, constraint-induced movement therapy, home programs) (Novak & Honan, 2019; Novak et al., 2013).

American and Canadian pediatric professional societies widely support the early identification of developmental disabilities (Council on Children with Disabilities, 2006; Williams, Clinton, & Canadian Paediatric Society Early Years Task Force, 2011), and it is endorsed at a policy level (Government of Manitoba, n.d.; Dua, 2003). The benefits of early identification and early intervention are multifaceted. For the child, it enhances opportunities for neural repair and optimizes developmental outcomes, and for families it provides opportunities for

access to resources and psychosocial supports, and engages families as care partners from the onset.

Despite this, wide practice variations continue to be observed in the primary care setting. There is evidence that delayed referral can compromise the child's potential from a neurodevelopmental perspective (Shepherd, 2013). Delayed referral for diagnostic assessment can also be detrimental to the child's family from a psychosocial perspective; the diagnostic process in itself is a period of a crisis analogous to bereavement (Huang, Kellett & St-John, 2010; Schuengel et al., 2009). Furthermore, parental dissatisfaction with the disclosure process has been linked to later maternal depression, poor adaptive coping and increased parental stress (Baird, McConachie & Scrutton, 2000; Dagenais et al., 2006). Conversely, parental satisfaction with disclosure (early diagnosis, positive attributes of the physician, informative content) has been associated with better adaptation (Rentinck, Ketelaar, Jongmans & Gorter, 2006). Once a diagnosis is made, parents can begin the process of adaptation, and the possibilities for concrete action on behalf of their child is crystallized (Shevell & Shevell, 2013).

1.3 The importance of the primary care context

Primary care practitioners (e.g. pediatricians, family physicians) do not typically receive the advanced training in early childhood development that pediatric medical specialists (e.g. child neurologists, developmental pediatricians) do, and consequently wide variability currently exists in their related knowledge and practice behaviors (Liptak et al., 2006; Sices, Feudtner, McLaughlin, Drotar, & Williams, 2003). Pediatricians have expressed uncertainty about their ability to identify and manage motor delays, and that they are not confident regarding their knowledge about neuromotor assessment in general (Noritz & Murphy, 2013). While developmental screening tools exist for the primary care context, they tend to focus on milestone

acquisition (Frankenburk, Dodds, Archer, Shapiro, & Bresnick, 1992), and are not sensitive enough to capture the subtle early quality of movement features of CP. The primary care practitioner has only a limited amount of time during well baby care visits, and their comprehensive but brief surveillance must cover of all aspects of growth, development and general health. Pediatricians have expressed a need for more educational resources concerning the identification and care of children with motor delays (Noritz & Murphy, 2013).

Parents and families often rely on their pediatrician or family doctor for health-related information about their child; and represent a stakeholder group for whom information gathering and transmission are vitally important (Gentles, Lokker, & McKibbon, 2010). Hesse et al. (2005) found that while most adults rate physicians as their most highly trusted information source, almost half will seek health-related information online prior to consulting them. It can be challenging for families who are seeking information about concerns they may have about their young child's early motor development. Disparities between the readability and applicability of educational materials for parents/caretakers of pediatric patients and their literacy levels have been demonstrated (Davis et al., 1994). Although there has been widespread support for plain language usage in health-related literature, limited evidence from the parent's perspective exists to support general effectiveness (Otal et al., 2012). Complicating matters further is the accessibility of a rapidly evolving mode of accessing information through technology and social media, (i.e. Internet, apps) (MacDermid, Solomon, Law, Russell & Stratford, 2006; Miller, 2013; Novak, 2014), information that parents may not be able to critically appraise in terms of scientific merit, in order to make well-informed decisions about their children's health.

1.4 Early identification of CP: Current clinical reality

Historically there has been a tendency to avoid providing a diagnosis of CP before two two years of age, to be sure that the neuromotor impairments are not transient. Evidence on the age at which children are referred for diagnostic assessment or diagnosed with CP is sparse. The few available studies are older and report on birth cohorts from the mid-1950s through the 1980s, but suggest wide variations in age at diagnosis ranging from 10-21 months or later (Lock, Shapiro, Ross, & Capute, 1986; Palfrey, Singer, Walker, Butler, 1987; Stanley, 1979). Most of the current research on early identification has focused on children at high-risk of CP (e.g. children who are extremely premature, have encephalopathy, or who are neonatal intensive care unit graduates) (Morgan, Novak, Dale, Guzzetta, & Badawi, 2016; Morgan et al., 2016; Novak, 2014; Novak et al., 2017), a context in which one would expect earlier identification to occur. This is apparent with respect to what options are currently available to assist in early detection efforts. The Prechtl General Movement Assessment (GMA; Einspieler & Prechtl, 2005; Prechtl et al., 1997) is predictive of CP in high-risk infants and has a sensitivity of 98% (95% confidence interval, CI 74–100%) and a specificity of 91% (95% CI 83–93%) (Bosanquet, Copeland, Ware, & Boyd, 2013). Use of the GMA has been championed for use with children at high-risk for CP, particularly extreme preterm infants (Adde et al., 2010; Novak et al., 2017). The practicality of using the GMA in the primary care context is debatable though, as it requires considerable training and time to administer. This would not be feasible for the broad population of all infants followed in the primary care setting. A recent systematic review (Novak et al., 2017) demonstrated that a combination of standardized neurological examination, neuroimaging, and standardized motor assessments was most predictive of risk for CP. Specifically the guidelines suggest the following: before five months corrected age, application of magnetic resonance imaging (MRI) and the GMA, or the Hammersmith Infant

Neurological Examination (HINE; Haataja et al., 1999) and the Test of Infant Motor Performance (TIMP; Campbell, Osten, Kolobe, & Fisher, 1993); after five months corrected age, use of the HINE and either the Developmental Assessment of Young Children (DAYC; Voress, & Maddox, 1998, 2013) or Alberta Infant Motor Scale (AIMS; Piper, Darrah, Maguire, & Redfern, 1994), or Neuro Sensory Motor Development Assessment (NSM DA; Burns, Ensbey, & Norrie, 1989), or the combination of the HINE and either DAYC or Motor Assessment of Infants (MAI; Chandler, Andrews, & Swanson, 1980). The application of these recommendations is only feasible in the high-risk neonatal follow-up context where the appropriate specialists and access to imaging are readily available, but not by primary care practitioners during the limited time they have for surveillance visits. There is a lack of evidence on population-based samples, which makes it difficult (i) to determine which children with CP are experiencing delayed referral, and (ii) to identify the factors associated with delays in referral for diagnostic assessment and rehabilitation services. Both of these are crucial missing elements required to inform future early detection strategies.

In the primary care context, PCPs may lack awareness of the early motor signs of CP, and of the respective roles of rehabilitation professionals in early interventions for children with CP. Furthermore, there is typically a linear serial model of referral – where referral for diagnosis is carried out first and is then often a gateway to rehabilitation services. This collectively results in unnecessary delays in rehabilitation intervention (Noritz & Murphy, 2013; Novak, 2014). Thus, an important knowledge-practice gap (Graham et al., 2006) has been identified with respect to primary care practitioners, a gap that can be extended to parents who often rely on their pediatrician or family doctor for health-related information about their child, and for whom information gathering and transmission are vitally important (Gentles, Lokker, & McKibbon, 2010).

1.5 Knowledge translation

Researchers can use knowledge translation (KT) approaches to help reduce and narrow knowledge-to-practice gaps (Graham et al., 2006). The Canadian Institutes of Health Research (CIHR) defines KT as

> "a dynamic and iterative process that includes synthesis, dissemination, exchange and ethically-sound application of knowledge to improve… health…provide more effective health services and products and strengthen the health care system" (CIHR, 2016, http://www.cihr-irsc.gc.ca/e/29418.html#2).

There are two broad categories of KT. The first is *end-of-grant KT*, which refers to the development and implementation of a plan for raising awareness among knowledge-users of a research projects' results (Straus, Tetroe, & Graham, 2013), and which can range from the traditional way researchers have disseminated the results of their research (e.g. presenting at conferences, publishing in journals) to more intensive activities (e.g. interactive educational sessions) (Parry, Salsberg, & Macaulay, 2009; Straus, Tetroe, & Graham, 2013). The second is *integrated KT* (iKT), which is a participatory approach to conducting research collaboratively with stakeholders/knowledge-users in every phase of the project, whereby this collaboration shapes all aspects from the formulation of the research question to the dissemination of the results (CIHR, 2016). This approach is also known as 'collaborative research', 'action-oriented research', and 'co-production of knowledge' (Graham & Tetroe, 2007). Underlying this approach is the assumption that if stakeholders/knowledge-users are involved and help shape the research, the end result will likely be more relevant and useful to them than if they did not provide input (Straus, Tetroe, & Graham, 2013).

The field of KT research is an emerging field, with iKT and participatory action research

necessitating a paradigm shift for all stakeholders involved in healthcare research. This research approach encourages researchers to engage knowledge-users at each step of the research process, to include the Knowledge Creation phase of the knowledge-to-action cycle. The idea is that end-user involvement from the onset will result in a more acceptable, relevant and user-friendly protocol. This integrated patient-oriented research approach of multi-stakeholder engagement is more likely to foster "multilevel, system-wide and sustainable change very badly needed in our health care systems" (Kothari & Wathen, 2013, p. 190). For a research project that aims to increase awareness of the early motor features of CP amongst primary care practitioners and parents (i.e. knowledge users) through the creation of educational (knowledge translation) tools, the use of an iKT approach when possible will ensure that content (attributes of CP) is relevant, understandable and user-friendly.

1.6 Summary

There is a need for population-based research on more recent birth cohorts of children with CP, as it will provide a better understanding of the current referral practices of primary care physicians for young infants and children suspected of having CP. This will in turn help to identify potential subgroups of children with CP who are at a greater risk for experiencing delays in referral for diagnosis and for rehabilitation services. This information can then be used to help focus and inform the content of subsequent early identification efforts and strategies. An iKT approach involving all relevant stakeholders (e.g. primary care practitioners, medical specialists, parents of children recently diagnosed with CP, expert rehabilitation clinicians, and researchers), will ensure that the content for early detection strategies is relevant, readily applicable to primary care and likely to have impact on health care practices (Kothari & Wathen, 2013). Application of consensus methodologies can provide the knowledge needed to generate clinical features and referral

recommendations that may be used in the primary care context to enhance the early identification of CP. Together, these efforts should result in prompt referral to medical specialists for diagnostic assessment, and when appropriate simultaneous referral to other rehabilitation specialists for intervention, and improve health outcomes for children with CP.

CHAPTER 2:

RATIONALE, OBJECTIVES and DESIGN

2.1 Rationale

CP is the most common physical disability of childhood with lifelong consequences, impeding autonomy, health and participation (Statistics Canada, 2007). Early intervention and family education can optimize long-term outcomes for both the child and their family (McIntyre, Morgan, Walker, & Novak, 2011). Importantly, animal and human studies using novel neuroimaging technologies highlight the importance of initiating rehabilitation interventions as early as possible, to counteract maladaptive neural circuitry that evolves in the non-damaged regions of the brain with disuse of the affected regions. Early intensive therapeutic training can heighten brain reorganization in this critical period of plasticity, with associated significant improvements in the child's functional outcomes (Kirton, 2013). American and Canadian pediatric professional societies widely support the early identification of developmental disabilities (Council on Children with Disabilities, 2006; Williams, Clinton, & Canadian Paediatric Society Early Years Task Force, 2011), and it is endorsed at a policy level (Government of Manitoba, n.d.; Dua, 2003).

Existing screening tools focus on delayed milestone acquisition, but do not delineate attributes related to an abnormal quality of movement essential to the timely detection of CP. Indeed, additional delays in the detection of motor disorders in the primary care setting is a concern raised by the American Academy of Pediatrics (Noritz & Murphy, 2013). Parents of children diagnosed with CP have expressed their dissatisfaction associated with delays in the diagnostic process, with negative repercussions to their adaptive coping and health (Baird, McConachie & Scrutton, 2000; Dagenais et al., 2006).

Current research in the field of early identification of CP has focused on children at high-risk for CP (extreme prematurity, perinatal asphyxia, and other neonatal intensive care unit graduates), but approximately half of children eventually diagnosed with CP are born at term after an uneventful pregnancy, or are premature but above the gestational age cut-off (e.g. 30-36 weeks gestational age) to qualify for a neonatal follow-up program. Of note, when primary care physicians eventually refer the child to a medical specialist for diagnosis, they may not typically co-refer the child to rehabilitation specialists, thus further delaying the intervention process.

Although there is both a biologic imperative (better brain organization and development) and a psychosocial need (family coping and support) for earlier detection of CP, to prompt early diagnosis and intervention, existing tools to promote earlier identification are not feasible to implement or use in the primary care context. Access to health professionals with sufficient training to complete standardized neurodevelopmental assessments and the GMA and the cost of MRI testing (Novak et al., 2017) are more appropriately applied in the context of neonatal follow-up programs for very high-risk neonates. In the primary care context, in which primary care physicians do not have the time nor the skills to apply these early detection methods, another strategy was needed.

The aim of this thesis was to develop content for KT tools that will enable primary care practitioners to have easy access to the knowledge and capacity to detect attributes associated with CP earlier. This would prompt simultaneous timely referral to medical specialists for diagnosis and medical management, and to rehabilitation specialists to initiate early therapeutic interventions at a critical period of brain development, optimizing outcomes (e.g. reducing symptom impact). Finally, the added advantage of earlier referral for diagnosis and intervention is that parents

engaged in the process of detection will benefit from early access to resources and family supports to optimize physical, social, and emotional functioning in children with CP and their families.

The feasibility of this study was supported by our collaboration with the CP researchers of Kids Brain Health Network (a Canadian National Centres of Excellence), and the research infrastructure offered by the Canadian Cerebral Palsy Registry (CCPR) (http://neurodevnet.ca/cp-registry). The CCPR enabled national multi-site data collection of a population-based sample of children with CP as part of our environmental scan of current practices, and secure storage online via REDCap (https://neurodevnet.med.ualberta.ca/index.php). The feasibility of this study was also supported by active collaboration and engagement with all relevant stakeholders including both content-experts (pediatric neurologists, developmental pediatricians, rehabilitation professionals, researchers) and knowledge-users (pediatricians, parents of children recently-diagnosed with CP) through all phases of the study.

2.2 Objectives of this thesis

2.2.1 Overarching objective of this thesis

The focus of the research project for my doctoral thesis was on the early identification of CP by primary care providers (pediatricians, family practitioners, nurse practitioners). The ultimate goal was to identify the appropriate content to develop a KT tool that will prompt earlier and simultaneous referral of children suspected of having CP for diagnosis and rehabilitation services, thus decreasing the delays in referral currently experienced by children and families. Specifically, this project aimed to create the background (content) knowledge needed to inform the design a KT tool (e.g. pocketcard, poster/pamphlet, web-based application) to increase primary care providers and parents awareness of the early motor signs ('clinical attributes') of CP.

It is hypothesized that this increased knowledge will enable the earlier identification and thus prompt simultaneous timely referral to medical and rehabilitation specialists for children suspected of having CP. Ultimately, this should improve health outcomes for children with CP.

2.2.2 Specific objectives of this thesis

The specific objectives of this research endeavour were twofold:

1. To provide *evidence and a population-based* understanding of the current referral practices of physicians related to children suspected of having CP, including the identification of *potential predictors* of delayed referral.
2. To develop expert-informed content for educational knowledge translation tools by determining:
    a. What the *clinical signs (features)* of CP are that should prompt early referral for diagnostic assessment of children suspected of having CP; and
    b. To *which rehabilitation and other health professionals* (e.g. occupational therapy, physical therapy, speech-language pathology, ophthalmology) children suspected of having CP should simultaneously be referred to at the time they are referred to a medical specialist for diagnostic assessment.

2.3 Study design and methods

The research project described in this thesis applies a sequential mixed-methods design underpinned by an integrated knowledge translation approach, with each component informing the next. A combination of methodologies were employed during each phase.

2.3.1 Phase one (knowledge synthesis)

Specific Objective 1. A structured review of the literature was conducted using the *scoping review* methodology (**Manuscript 1**); a single-site *retrospective chart review* was conducted

(**Manuscript 2**), and a *national multi-site environmental scan* of physician referral practices was conducted (**Manuscript 3**).

2.3.2 Phase two (knowledge [content] creation)

Knowledge acquired in Phase One, from the literature and from current practices, was used to inform Phase Two.

Specific Objective 2a. Two nominal group processes (consensus methods) were conducted with national (Canadian) and local (Montreal, Quebec City) content-experts and knowledge-users (**Manuscript 4**); an online two-round Delphi Survey was conducted with international experts in the field of early childhood (**Manuscript 5**).

Specific Objective 2b. Two nominal group processes (consensus methods) were conducted with national (Canadian) and local (Montreal, Quebec City) content-experts and knowledge-users (**Manuscript 4**); an online two-round Delphi Survey was conducted with international experts in the field of early childhood (**Manuscript 5**).

CHAPTER 3:

MANUSCRIPT 1

Age at referral for diagnosis and rehabilitation services for cerebral palsy:

A scoping review

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This manuscript was published in *Developmental Medicine & Child Neurology.*

Boychuck, Z., Bussières, A., Goldschleger, J., Majnemer, A., & the PROMPT group. (2019). Age at referral for diagnosis and rehabilitation services for cerebral palsy: A scoping review. *Developmental Medicine & Child Neurology,* 61(8), 908-914. DOI: 10.1111/dmcn.14034

Accepted for publication: July 31, 2018.
Epub ahead of print: October 1, 2018.
Published: July 3, 2019.

Age at referral for diagnosis and rehabilitation services for cerebral palsy: a scoping review

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PUBLICATION DATA
Accepted for publication 31st July 2018.
Published online 1st October 2018.

ABBREVIATIONS

NICU	Neonatal intensive care unit
PCP	Primary care practitioner

AIM This study sought to: (1) determine what is known about age at referral for diagnosis and rehabilitation services for children suspected of having cerebral palsy (CP); and (2) identify factors associated with earlier referral.
METHOD A scoping review was conducted to summarize existing literature. We systematically searched Allied and Complementary Medicine, CINAHL, Cochrane Library, Embase, and PsycINFO for evidence published between 1979 and 2017 on age at referral for diagnosis or age at referral to rehabilitation services for children suspected of having CP. Quantitative and thematic analyses of the literature were performed.
RESULTS Our search yielded 777 articles, of which 15 met the inclusion criteria. Only one study focused on age at referral for diagnosis of CP (mean 16.6mo±19.2mo), with two on age at referral to rehabilitation services (means 13.9mo±15.8mo and 12.4mo). Potential predictors of earlier referral identified include referral source, type of CP, and a complicated birth history.
INTERPRETATION Evidence is sparse; however, available studies suggest high variation in the age at which children are being referred for diagnosis, typically ranging from 10 months to 21 months. Evidence indicates that subgroups of children with CP might be experiencing prolonged delays. Findings highlight the need to better understand what contributes to delays in referral for diagnosis and rehabilitation.

Cerebral palsy (CP) is the fourth most common childhood disability,[1] and the most common physical disability encountered in children,[2] with an estimated prevalence of 2.3 per 1000 live births.[3] A consensus definition of CP describes it as group of disorders of the development of movement and posture resulting from damage in the fetal or infant brain, often accompanied by secondary sensory impairments.[4] Current best practice for children suspected of having CP recommends early identification and referral both to medical specialists for diagnosis and to rehabilitation professionals for intervention.[5] Early identification of developmental disabilities is widely supported by the American and Canadian pediatric professional societies,[6,7] and is endorsed at a policy level.[8,9] There is evidence to suggest that delayed referral can limit a child's ability to reach their full potential in developmental skills.[10] Delayed referral for diagnosis can also be detrimental to the child's family from a psychosocial perspective. Diagnosis in and of itself is a period of crisis analogous to bereavement,[11,12] and parental dissatisfaction with the disclosure process has been linked to later maternal depression, poor adaptive coping, and increased parental stress.[13,14] Conversely, parental satisfaction with disclosure (e.g. early diagnosis, positive attributes of the physician, informative content) has been linked with better adaptation.[15,16] Failure to promptly identify and support parental psychosocial issues may contribute to mental health morbidity for the family of the child with CP.[17]

According to Canadian and Australian CP registries, approximately 40% to 45% of children with CP are born preterm;[18,19] however, only a subset, typically those no older than 29 weeks, are closely monitored by a neonatal follow-up program, with the children determined not to be at 'high-risk' of CP discharged to care in the community. In addition, many children with CP are born at term and may not have a high-risk history. Primary care practitioners (PCPs), such as pediatricians and family physicians, are thus uniquely positioned to serve a crucial role in identifying these children and referring them to medical specialists for timely diagnosis, and to rehabilitation professionals for intervention. Importantly, PCPs do not receive the same advanced training in early childhood development as child

 DOI: 10.1111/dmcn.14034

neurologists and developmental pediatricians. Not surprisingly perhaps, wide variability currently exists in their knowledge and practice with respect to identifying children with developmental disabilities.[20,21]

Apart from lacking an awareness of the early motor signs of CP, PCPs and parents may be unaware of the roles rehabilitation professionals have in early intervention. Unnecessary delays in intervention may result from the linear serial model of referral, where diagnosis is often the gateway for referral to rehabilitation services.[22,23] In summary, although early identification is widely endorsed, about half of children with CP do not benefit from close monitoring and PCPs may not be adequately informed to detect CP in this population. Thus, there is concern about delays in referral for diagnosis of CP.

The focus of this review is on age at referral rather than age at diagnosis as there may be strategies to optimize earlier detection by PCPs and decrease age at referral. Age at diagnosis may also be influenced by waiting lists, which is a systemic issue. Nevertheless, as part of this scoping review, we also documented age at diagnosis of CP and age at rehabilitation intervention as they may provide useful information related to potential predictors of earlier or delayed diagnosis. Therefore, the primary aim of this study was to determine from the existing literature what is known about age at referral for diagnosis and for rehabilitation services of children suspected of having CP. A second aim was to identify factors potentially predictive of earlier or delayed referral for diagnosis.

METHOD

A scoping review was conducted on the basis of the framework proposed by Arksey and O'Malley,[24] and informed by the enhancements proposed by Levac et al.[25]

Identifying the research question

The research question guiding this review was the following: what is known from the existing literature about the age at referral for diagnosis of children suspected of having CP, and what factors have been associated with earlier referral?

Identifying relevant studies

Given we anticipated that there would be a limited literature addressing this research question, we also included literature on age at diagnosis of CP, as well as age at referral to rehabilitation services. With the assistance of an information specialist, an electronic-database search strategy was developed in MEDLINE and adapted for Allied and Complementary Medicine, CINAHL, Cochrane Library, Embase, and PsycINFO to uncover studies reporting on age at referral for diagnosis of CP using a combination of keywords and Medical Subject Headings (MeSH) terms: cerebral palsy, spastic hemiplegia, spastic diplegia, spastic tetraplegia, spastic quadriplegia, dyskinesia, ataxia, age, delay, later, early, referral, consultation, primary healthcare, secondary care, tertiary healthcare, diagnosis, and detection. Articles were also identified through reference list screening/hand searching.

What this paper adds

- Evidence on age at referral for diagnosis of cerebral palsy is sparse.
- Potential predictors of delayed referral represent targets to minimize delays in diagnosis.
- A subset of children may be experiencing unnecessary delays in referral.

Study selection

Studies were limited to original published research and expert opinion that had undergone the rigor of peer review (grey literature, anecdotal, and expert-opinion sources were thus excluded), published in English or French, from 1979 to 2017 inclusively. The clinical population of interest was limited to infants and young people under 18 years of age with CP. Studies were limited to those with a primary focus on age at referral for diagnosis of CP and age at diagnosis of CP (e.g. studies focusing on risk factors for CP were excluded).

For this study, age at referral for diagnosis of CP was operationalized as the age in months at which a child was referred for diagnosis, usually from a PCP to a medical specialist (e.g. child neurologist, developmental pediatrician) for diagnosis of CP. Age at diagnosis of CP was operationalized as the age in months at which a child formally received a diagnosis of CP, usually from a medical specialist. For this study, ages at 'identification' or 'presentation' were included as age at diagnosis. Age at referral for rehabilitation services and age at rehabilitation intervention were similarly operationalized.

Three reviewers (ZB, JG, and AM) were involved in the study screening and selection process. Two levels of screening were performed: (1) title/abstract; and (2) full text. Initially all titles/abstracts were screened for eligibility by ZB and JG, with AM available to discuss and resolve potential discrepancies. Before this initial level of screening, a calibration exercise was conducted to ensure reliability in correctly selecting articles for inclusion: ZB and JG randomly selected 5% (n=30 out of 597) of the retrieved titles/abstracts, and interrater reliability exercises were performed using a predefined relevance criteria form, which demonstrated very good agreement (Cohen's κ=0.84). The two reviewers then divided and completed the remaining title/abstract screenings independently. The second level of screening involved reading the full text of each article retained from the title/abstract screening. ZB completed the full-text reviews, and AM was consulted as needed for further clarification of any ambiguities.

Charting the data

Using a descriptive–analytical method,[26] the authors applied an iterative approach in the development and integration of the data charting form. The following information was recorded for each study as available: author, year of publication, title, country of publication, research methodology/type of study, clinical population/sample

characteristics, information on age at referral for diagnosis, information on age at diagnosis, information on age at referral for rehabilitation, and information on age at initiation of rehabilitation services. Differences in extraction were resolved by discussion or with the involvement of a third reviewer (AM) if consensus could not be reached. Studies were excluded at the full-text review level if during data extraction some exclusion criteria were identified.

Collating, summarizing, and reporting the results

In accordance with Arksey and O'Malley,[24] no formal quality assessment of the included studies was made as the aim of this scoping review was to identify the breadth of the literature and the major areas of research activity with corresponding resulting themes. As suggested by Levac et al.,[25] we incorporated both a quantitative analysis (numerical summary) and qualitative analysis (identification of factors related to earlier or delayed referral). Also true to the scoping review methodology, the analytical description of studies is meant to identify directions for practice and gaps that should be addressed in future research.

RESULTS

Descriptive numerical analysis

As outlined in Figure S1 (online supporting information), a total of 777 articles were initially retrieved through the database searches, cross-referencing, and hand searching, with 15 eventually retained for the final analysis. Most studies were descriptive cohort studies with no testing of factors related to predictive factors of age at referral, and no specific recommendations for practice (Table I).

Age at referral for CP diagnosis

Only one study reported on age at referral for diagnosis. A retrospective chart review[27] found that children were referred for diagnosis at a mean age of 16.6 months, with high variability. Fifty-eight percent of children were referred before 1 year of age, 23% between 1 year and 2 years of age, 6% between 2 years and 3 years of age, and 13% were 3 years of age or older.

Age at CP diagnosis

Eight studies reported on age at diagnosis,[5,28–34] which ranged between 8 months and 24 months. Several other authors[23,35–37] suggested that CP can be diagnosed as early as 12 weeks, but may be more reliable as late as 36 months.

Age at referral to rehabilitation services

Only two studies reported on age at referral for rehabilitation services.[27,38] The mean age ranged between 12 months and 14 months, again with wide variability.

Age at initial rehabilitation intervention

Only one study focused on age at rehabilitation intervention. Colver[39] reported on the age at initiation of rehabilitation services for two cohorts of children with three types of CP. Children with quadriplegia received services earlier than the other subtypes. Age at intervention was earlier for children in the later birth cohort.

Qualitative analysis

Analysis of the included studies (n=10) revealed several potential predictors of earlier referral (Table SI, online supporting information).

Factors influencing age at referral for diagnosis

Three potential predictors of referral at a younger age for diagnosis were identified in one study.[27] First, referral source (operationally defined by the authors as a medical specialist [e.g. child neurologist] or PCP [e.g. pediatrician]) was identified. Medical specialists accounted for most of the referrals and flagged children for diagnosis significantly earlier than PCPs. Second, having had a complicated birth history (an initial admission to a neonatal intensive care unit [NICU]) was identified. Children initially admitted to a NICU were referred for diagnosis significantly earlier than children not initially admitted to a NICU. Third, type of CP (diplegia, hemiplegia, quadriplegia, mixed, 'other') was identified. Children with certain subtypes of CP (diplegia, hemiplegia) are experiencing significant delays.

Factors influencing age at diagnosis

Two potential predictors of a younger age at diagnosis were identified. Comparing type of CP, Lock et al.[31] found the mean ages of presentation for children with quadriplegia were earlier than for children with diplegia. Of note, the authors concluded that CP subtype is an insignificant predictor of earlier diagnosis, although it is important to note that their sample size (n=57) was small and did not include any children with hemiplegia.

The second potential predictor of a younger age at diagnosis was severity of motor impairment. One study[30] reported a median age at diagnosis of 11.1 months across CP types, although 6.6% were diagnosed at age 5 years or older, with an earlier diagnosis associated with a higher degree of motor disability. The Australian CP Registry indicated that the average age for a description of CP is 19 months, but the authors noted the wide variability in clinical practice with description as early as 1 week old for children with more severe impairment, to 5 years of age for children with mild to moderate severity.[5]

Factors influencing age at referral to rehabilitation services

Three potential predictors of referral at a younger age for rehabilitation services were identified. Hubermann et al.[27] suggested that a complicated birth history may be a factor. The subset of children initially hospitalized in the NICU was referred for treatment earlier than children who were discharged home and did not receive neonatal follow-up. Lindstrom and Bremberg[38] offered two potential predictors. The first was referral source, since most of the children referred at younger ages came from medical specialists with advanced clinical training in early infant

Table I: A summary of the characteristics of the included studies (*n*=15)

References	Country	Study design/ research methodology	Clinical population/sample characteristics	Information on age at referral for diagnosis; age at diagnosis; age at rehabilitation
Ashwal et al.[35]	USA	Practice parameter: literature review	All included studies had ≥20 patients	The diagnosis of CP is given before age 2y
Bennett[28]	USA	Commentary		Average age at diagnosis of CP was 18mo
Bosanquet et al.[36]	Australia	Systematic review	All included studies (*n*=19) assessed children at high-risk of CP only	Propose CP can be more reliably assessed and diagnosed at 36mo
Byrne et al.[29]	USA	Prospective cohort study	Infants at high-risk of CP: cohort A: 2014–2015 (*n*=70) cohort B: 2016–2017 (*n*=175)	Mean age at diagnosis: cohort A: 18mo±7mo, range 11mo–32mo cohort B: 13mo±4mo, range 4mo–29mo
Colver[39]	UK	Descriptive cohort study	All children of preschool age in Northumberland (3600 births each year)	Average age at which physiotherapy began for children with CP: birth years 1970–1977 quadriplegia (6.5mo) diplegia (23mo) hemiplegia (21mo) birth years 1978–1985 quadriplegia (6.5mo) diplegia (10mo) hemiplegia (12mo)
Granild-Jensen et al.[30]	Denmark	Descriptive retrospective cohort study	1291 children with CP born 1995–2003; registry, population-based	Median age at diagnosis reported was 11.1mo across CP types; 6.6% were diagnosed at age 5y or older
Hubermann et al.[27]	Canada	Retrospective chart review	103 children with CP, born 2002–2012	Mean age at referral for diagnosis (*n*=99) was 16.6mo±19.2mo (range 0.1mo–89.9mo) Mean age at referral to rehabilitation services (*n*=90) was 13.9mo±15.8mo (range 0.1mo–79.5mo)
Lindstrom and Bremberg[38]	Sweden	Retrospective cohort study	23 924 children born 1986–1990 (*n*=66 with CP)	Mean age of referral to a habilitation unit was 12.4mo (range 0.5mo–54mo)
Lock et al.[31]	USA	Retrospective chart review	738 consecutive children referred for developmental evaluation between 1982 and 1983 (*n*=57 with CP)	Mean age of presentation: diplegia: 15.9mo quadriplegia: 12.1mo
McIntyre et al.[5]	Australia	Review	N/A	Average age for a description of CP to be given is 19mo
Novak[23]	Australia	Review	N/A	CP is historically diagnosed around 12mo–24mo Diagnosis at 12wks possible for children at high-risk; early diagnosis for 'healthy term borns' requires further research
Novak et al.[37]	Australia	Systematic review	Children at high-risk for CP	Diagnosis is usually made between 12mo and 24mo, but now possible before 6mo corrected age
Palfrey et al.[32]	USA	Cross-sectional survey	All children in special education programs in five census districts (*n*=1726 children)	Mean age of identification of CP was 10.3mo
Stanley[33]	Australia	Retrospective cohort study	All children with the diagnosis of CP born in Western Australia between January 1st, 1956 and December 31st, 1975 (*n*=917 children with CP)	Mean age at diagnosis was 21mo; median age 11mo; 3% of children diagnosed later than 36mo
Tirosh et al.[34]	Israel	Population-based survey	All children part of developmental screening from January 1979 to December 1984; 29 108 children (*n*=66 children with CP)	At diagnosis of CP: 42.5% were 1mo–12mo 43.9% were 13mo–24mo 13.6% were 25mo–36mo

CP, cerebral palsy.

motor development. The second was severity of motor impairment, as the authors found that children with a 'mild' CP were referred much later than children with 'moderate' or 'severe' CP.

Factors influencing age at rehabilitation intervention

Type of CP was identified as a potential predictor of initiating rehabilitation services at younger age. Colver[39] reported that children with quadriplegia received the earliest intervention compared with children with other subtypes of CP.

DISCUSSION

To our knowledge, this is the first review of the literature on age at referral for diagnosis for children suspected of having CP. We performed a comprehensive search of

electronic databases, and had very good interrater reliability during the screening and abstraction phases.

The results of this scoping review demonstrate that available evidence is sparse about the current referral practices of PCPs for children with CP, especially with respect to the age at which children are currently being referred to medical specialists for diagnosis of CP. Most of the few available studies are older and report on birth cohorts from the mid-1950s to the 1980s, but they suggest variations in age at diagnosis ranging from 10 months to 21 months.[31–33] It is important to note that much of the literature found through this review, and used in clinical practice, is 'expert opinion' in nature. Current evidence lacks population-based data and strong methodological designs to support best-practices. In the single recent study that objectively explored age at referral for diagnosis, there was a high variability in the age at which children were referred (mean 16.6mo±19.2mo; range 0.1mo–89.9mo).[27] It is also important to note that most of the research so far in early identification and early intervention has focused on children considered to be at 'high-risk' of CP, potentially biasing the estimated age at referral to an earlier age.[5,23] Consequently, little is known and further research efforts are required for the almost half of children with CP born at term from uncomplicated pregnancies, or who are born preterm (i.e. 30–36wks gestational age) and subsequently may not have been in a neonatal follow-up program. In the two studies that reported age at referral to rehabilitation services, there was a similar high variability. Hubermann et al.[27] found wide variability in clinical practice, with some children experiencing delayed referral for diagnosis beyond 3 years of age. Similarly, Lindstrom and Bremberg[38] reported that some children experienced delays for referral as late as age 4 years 6 months.

Some factors have been identified as being potentially predictive of a younger age at referral both for diagnosis and for rehabilitation services. The first is *referral source*, with PCPs seeming to refer for diagnosis later than medical specialists. Since they often do not have the advanced training that medical specialists receive in early infant motor development, it is possible that PCPs may not recognize signs of early motor delay in their clinical practice for the subset of children who are discharged home without neonatal follow-up. It is important to acknowledge that medical specialists may be evaluating children with more severe impairment, or more high-risk infants through neonatal follow-up programs, which may in part explain the finding of earlier detection. The second potential predictor is having had *a complicated birth history*. The prenatal, perinatal, and postnatal risk factors for CP are well known, with some of the most important ones being low birthweight, known intrauterine infections, and multiple gestation.[40] Since these early risk factors are often associated with prompt neonatal follow-up that includes close developmental surveillance, children who fit this profile are more likely to be identified and referred earlier than those who are discharged home without neonatal follow-up. Not surprisingly, *severity of motor impairment* may be a third potential predictor of a younger age at referral, with children having more severe motor impairment being identified and referred earlier.

Synthesizing these results, it is possible that there are two catchments of children with CP: (1) children with a complicated birth history (initial NICU admission), who are referred early for diagnosis and rehabilitation services by medical specialists from neonatal follow-up programs; and (2) children who do not have a complicated birth history (no risk factors, no initial NICU admission), or are NICU graduates who do not meet criteria for entry to a neonatal follow-up program, and are discharged to the community and probably lost to follow-up until later age when other delays may present.

While early identification and early intervention are widely accepted as best practice for children suspected of having CP, the available evidence suggests that an important knowledge-to-practice gap may exist. If PCPs lack the knowledge of the early signs of CP, they will continue to experience challenges in detection in their practice, and children will continue to experience delayed referral for diagnosis and rehabilitation. Our findings have important implications for stakeholders, researchers, and decision-makers. Physicians (especially PCPs) and parents need to be better informed on the early motor signs of CP. Continuing monitoring and support to address potential concerns related to a child's motor development area are also needed.

This review, however, is not without limitations. We did not search the grey literature, and restricted the search to English and French publications published as of 1979, excluding abstracts and conference proceedings. These decisions possibly excluded some relevant studies. Also, as per scoping review methodology, the quality of the included studies was not assessed; thus there is a potential risk of bias inherent from the studies' low-quality methodological design. However, the purpose of this review was not to indicate what would be the best practice in the field, but rather to gather a scope of the existing evidence and current practices. Also, the lack of recent studies across different countries prevented us from making any meaningful comparisons across health care systems.

In a recently published strategic plan for CP research, the National Institute of Child Health and Human Development and the National Institute of Neurological Disorders and Stroke stress the importance of early intervention, linking successful outcomes with earlier diagnosis and intervention.[41] The potential benefits of earlier intervention continue to be supported in the literature. As an example, a recently published exploratory study found that the use of baby-constraint induced movement therapy with infants as young as 3 months to 8 months may improve functional outcomes for children with unilateral CP.[42] To effectively target this recommendation it is essential to better understand the current clinical picture for referral

practices. This study highlights the need for population-based data and high methodological designs on the current referral practices of PCPs and the factors that contribute to delays in referral and diagnosis. This would inform the development of evidenced-based knowledge translation tools to enhance early detection and prompt subsequent simultaneous referral for diagnosis, medical management, and rehabilitation services.

CONCLUSION

Literature is sparse on the current referral practices of PCPs related to children suspected of having CP. Although efforts have been made to promote developmental surveillance, PCPs may lack awareness of the early motor signs of CP, thus unnecessarily prolonging the delays currently experienced in diagnosis and intervention. This study highlights the need for population-based data on the current referral practices of PCPs and factors associated with delays in referral and diagnosis. This will enable the identification of the precise knowledge gaps that exist in terms of early recognition of CP, which will inform the targeted development of knowledge translation tools to enhance early identification and early intervention. Subsequently, continuing professional development initiatives addressed to PCPs should also be put in place to improve early detection and referral.

ACKNOWLEDGMENTS

The authors disclose receipt of the following financial support for the research, authorship, and publication of this article: ZB received salary support as a doctoral trainee from the Canadian Institutes of Health Research (MOP-133402), which supported this study in part. ZB also received a financial support from the Richard and Edith Strauss Canada Foundation as a musculoskeletal doctoral fellow, as well as from the Hoppenheim Fund/Montreal Children Hospital Foundation. We thank Pamela Harrison and Jill Boruff for their assistance with the database searches. We also express our gratitude to Deniz Keskinel, Sossy Sahakian, and Annahita Ehsan for their support with the database searches and in preparing the manuscript. No ethical review was required for this study. The authors have stated that they had no interests which might be perceived as posing a conflict or bias.

SUPPORTING INFORMATION

The following additional material may be found online:

Appendix S1: Members of the PROMPT Group.

Figure S1: Flow chart of study selection.

Table SI: Summary of the studies including information on factors potentially associated with a younger age at referral

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RESUMEN

EDAD DE DERIVACIÓN A LOS SERVICIOS DE DIAGNÓSTICO Y REHABILITACIÓN EN LA PARÁLISIS CEREBRAL: REVISIÓN DEL ALCANCE

OBJETIVO Este estudio buscó (1) determinar qué se conoce sobre la edad de derivación a los servicios de diagnóstico y rehabilitación de niños con sospecha de tener parálisis cerebral (PC) e (2) identificar factores asociados con una derivación más temprana.
MÉTODO Se llevó a cabo una revisión panorámica para resumir la literatura existente. Buscamos sistemáticamente en *Allied and Complementary Medicine, CINAHL, Cochrane Library, Embase,* and *PsycINFO* la evidencia publicada entre 1979 y 2017 sobre la edad de derivación para el diagnóstico o la edad de derivación a los servicios de rehabilitación de niños con sospecha de PC. Se realizaron análisis cuantitativos y temáticos de la literatura.
RESULTADOS Nuestra búsqueda arrojó 777 artículos, de los cuales 15 cumplieron los criterios de inclusión. Solo un estudio estaba enfocado en la edad de la derivación para el diagnóstico de PC (media 16.6m±19.2m), y dos sobre la edad de derivación a los servicios de rehabilitación (media 13.9m±15.8m and 12.4m). Los posibles predictores de la derivación más temprana incluyen la procedencia de la derivación, el tipo de PC, y una historia de nacimiento con complicaciones.
INTERPRETACIÓN La evidencia es escasa; sin embargo, los estudios disponibles sugieren que existe una alta variación de la edad en la que los niños son derivados para ser diagnosticados, típicamente oscilando entre los 10 meses y los 21 meses. La evidencia indica que subgrupos de niños con PC pueden estar experimentando retrasos prolongados. Los hallazgos resaltan la necesidad de comprender mejor lo que contribuye al retraso en la derivación para el diagnóstico y para la rehabilitación.

RESUMO

IDADE DE ENCAMINHAMENTO PARA DIAGNÓSTICO E SERVIÇOS DE REABILITAÇÃO PARA PARALISIA CEREBRAL: UMA REVISÃO ABRANGENTE

OBJETIVO Este estudo buscou (1) determinar o que se sabe sobre a idade de encaminhamento para diagnóstico e serviços de reabilitação para crianças com suspeita de paralisia cerebral (PC) e 2) identificar fatores associados com o encaminhamento precoce.
MÉTODO Uma revisão de escopo foi realizada para sintetizar a literatura existente. Nós sistematicamente buscamos a Allied and Complementary Medicine, CINAHL, Biblioteca Cochrane, Embase e PsycINFO por evidências publicadas entre 1979 e 2017 sobre idade no momento do encaminhmento para diagnóstico ou idade no momento do encaminhamento para serviços de reabilitação para crianças com suspeita de PC. Análise quantitativa e temática da literatura foram realizadas.
RESULTADOS Nossa busca resultou em 777 artigos, dos quais 15 atenderam aos critérios de inclusão. Apenas um estudo enfocou a idade de encaminhamento para diagnóstico de PC (média 16,6 m ± 19,2 m), com dois sobre a idade de encaminhamento para serviços de reabilitação (média 13,9 m ± 15,8 m e 12,4 m). Potenciais preditores do encaminhamento precoce incluíram a fonte do encaminhamento, o tipo de PC, e uma história de nascimento complicado.
INTERPRETAÇÃO A evidência é escassa; no entanto, os estudos disponíveis sugerem alta variação na idade em que crianças são encaminhadas para diagnóstico, tipicamente variando de 10 a 21 meses. A evidência indica que subgrupos de crianças com PC podem vivencar atrasos prolongados. Os achados enfatizam a necessidade de compreender melhor o que contribui para os atrasos no encaminhamento para diagnóstico e reabilitação.

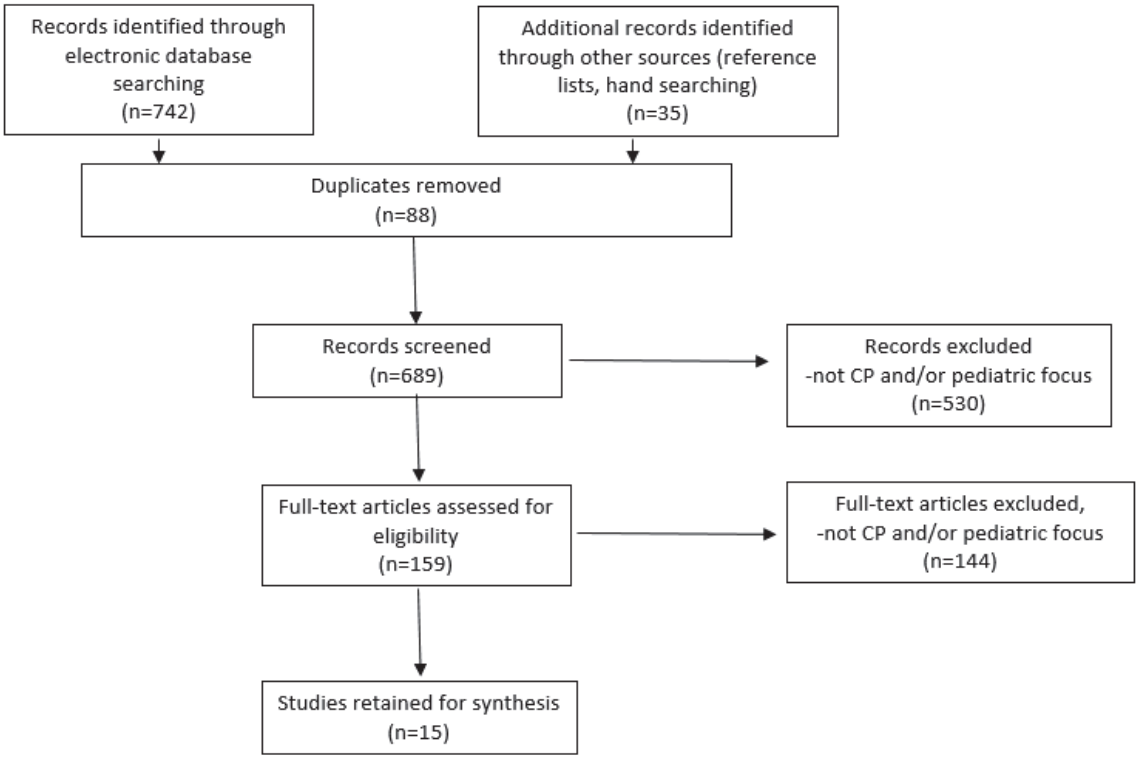

Figure S1. Flowchart of study selection.

Table SI: A summary of the studies including information on factors potentially associated with a younger age at referral

Reference	Information on factors associated with earlier referral
Bennett[28]	Severity of motor dysfunction
	Generally, the more severely affected child frequently can be diagnosed by 6mo of age, with milder cases taking longer
Colver[39]	Type of CP
	Both cohorts in this study (1970–1977, 1978–1985) reported that children with quadriplegia were receiving rehabilitation services considerably earlier than children with diplegia and hemiplegia
Granild-Jensen et al.[30]	Type of CP
	Median age at diagnosis was 11.1mo across CP types
	Children with a higher degree of motor disability were diagnosed earlier
Hubermann et al.[27]	Referral source
	81 out of 103 children were referred for diagnosis by a medical specialist (mean 13.6mo+15.7mo), whereas primary care providers referred much later (mean 28.8mo+27.1mo)
	Complicated birth history
	Children admitted to the neonatal intensive care unit (*n*=58) were referred for diagnosis at a mean age of 9.3mo+10.2mo, while children not initially admitted (*n*=36) were referred at a mean age of 28.1mo+24.9mo
	Type of CP
	Diplegia, *n*=21, mean age 31.6mo+24.9mo
	Hemiplegia, *n*=38, mean age 15.2mo+16.5mo
	Quadriplegia, *n*=23, mean age 10.9mo+16.9mo
	Mixed, *n*=1, mean age 2.6mo
	Other, *n*=16, mean age of 8.8mo+7.1mo
Lindstrom and Bremberg[38]	Severity of motor dysfunction
	Referral age for children with ‘mild’ CP (mean 12.4mo; range 0.5mo–54mo)
	Referral age for children with a ‘moderate’ or ‘severe’ CP (mean 7.1mo; range 0.5mo–23mo)
	Referral source
	Of the referrals at 2mo–3mo of age, five out of nine were from neonatologists/specialists
Lock et al.[31]	Severity of motor dysfunction

	Authors report severity of disability did not affect age of referral
McIntyre et al.[5]	Severity of motor dysfunction
	Description can be given anywhere between 1wk and 5y of age, depending on severity
Novak[23]	Complicated Birth History
	Diagnosis of CP as early as 12wk is now possible for high-risk populations (e.g. neonatal intensive care unit graduates)
Palfrey et al.[32]	Referral source
	Mean age at identification by physician 10.3mo
	Mean age at identification by non-non-physician (e.g. parents, teachers) 6.0mo
Tirosh et al.[34]	Referral source
	39% of the children with CP were not identified primarily as such by the referring pediatrician

CP, cerebral palsy.

Appendix S1
The PROMPT Group

Stakeholders: Content-Experts, Knowledge-Users

- Howard Bergman, MD – Department of Family Medicine, McGill University, Montreal, Quebec, Canada
- Zachary Boychuck, PhDc, OT – School of Physical and Occupational Therapy, McGill University, Montreal, Quebec, Canada
- Benjamin Burko, MD – Tiny Tots Medical Centre, Dollard-Des Ormeaux, Quebec, Canada
- Emmanuelle Dagenais, Parent of a child with CP – Montreal, Quebec, Canada
- Lynn Dagenais, PT – CHU Sainte-Justine, Montreal, Quebec, Canada
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CHAPTER 4:

INTEGRATION OF MANUSCRIPTS 1 AND 2

4.1 Research Questions of Manuscripts 1 and 2

Manuscript 1*:*

"What is known from the existing literature about the age at referral for diagnosis of children suspected of having CP, and what factors have been associated with earlier referral?"

Manuscript 2*:*

"To what extent do birth history, disability profile, sociodemographic variables, and type of referring physician influence the age at referral to a medical specialist for diagnosis and age at referral to rehabilitation specialists for intervention among children newly diagnosed with cerebral palsy?"

4.2 Integration of Manuscripts 1 and 2

The primary aim of Phase One of the research project contained in this thesis was to synthesize the available evidence on age at referral for diagnostic assessment and rehabilitation services for children suspected of having CP. The two main sources of information for this synthesis were (i) published scientific literature and (ii) current clinical practices. **Manuscript 1** was a study which used a scoping review methodology to explore the literature on age at referral for diagnostic assessment and rehabilitation services, which in turn provided the first half of the information necessary to conduct this synthesis. The second half of the required information came from a national environmental scan of Canadian physician referral practices (**Manuscript 3**). **Manuscript 2** was a feasibility study (local context, using a convenience sample) which provided preliminary data, and the methods used then informed the study contained in **Manuscript 3.**

CHAPTER 5:

MANUSCRIPT 2

Age at Referral of Children for Initial Diagnosis of Cerebral Palsy and Rehabilitation: Current Practices

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This manuscript was published in the *Journal of Child Neurology*.
Hubermann, L., **Boychuck, Z.**, Shevell, M., & Majnemer, A. (2016). Age at referral of children for initial diagnosis of cerebral palsy and rehabilitation: Current practices. *Journal of Child Neurology*, 31(3), 364-9. doi:10.1177/0883073815596610
Accepted for publication: March 14, 2015.
Epub ahead of print: August 3, 2015.
Published: March 1, 2016.

Journal of Child Neurology
2016, Vol. 31(3) 364-369

DOI: 10.1177/0883073815596610
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Age at Referral of Children for Initial Diagnosis of Cerebral Palsy and Rehabilitation: Current Practices

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Abstract

Objectives: This study describes current practices in the age at referral for diagnosis of cerebral palsy and factors that influence earlier referral. **Study Design:** Retrospective chart review (2002-2012). **Results:** Of 103 children referred for diagnosis, 81 were referred to a neurologist by other medical specialists at a mean of 13.6 $\pm$ 15.7 months, whereas primary care providers referred much later (mean = 28.8 $\pm$ 27.1 months). Children admitted to the neonatal intensive care unit were referred earlier (mean = 9.3 $\pm$ 10.2 months) than those not (28.1 $\pm$ 24.9 months). Referral to rehabilitation was similarly delayed. **Conclusions:** Primary care providers generated a minority of referrals, of concern given their role in developmental surveillance. Remarkably high variability suggests knowledge of cerebral palsy attributes varies widely among service providers. Half of children with cerebral palsy do not have a complicated birth history; subsequently, referrals for diagnosis and management are often delayed. New strategies are needed to optimize prompt referral by primary care providers.

Keywords

cerebral palsy, diagnosis, early identification, referral, rehabilitation

Received December 05, 2014. Received revised February 26, 2015. Accepted for publication March 14, 2015.

Cerebral palsy is the most common physical disability of childhood and manifests early in life, during which time the brain has the greatest potential for reorganization and plasticity. Therefore, early identification of developmental challenges is crucial for timely rehabilitation interventions that can greatly improve outcomes.[1,2] Earlier detection of cerebral palsy could be facilitated by providing primary care physicians with practical recommendations for timely referral to neurologists for diagnosis as well as by promoting the simultaneous referral to rehabilitation services. Practices of simultaneous referral currently exist for other developmental disabilities and have been proven successful.[3,4]

There is a general lack of awareness by primary care physicians of the early indicators of cerebral palsy and limited formal knowledge of the different roles of rehabilitation specialties in early intervention, which typically leads to referral to rehabilitation by a medical specialist rather than directly by the primary care practitioner (in a linear serial model of referral).[5,6] This contributes to unnecessary delays in intervention, which has been strongly criticized by parents who felt that their primary care providers dismissed their concerns and did not provide sufficient explanation of their child's developmental difficulties.[5] In a recent study on infants with perinatal stroke (hemiplegic cerebral palsy), the interquartile range of parental concern was 3.0 to 8.5 months, and for physicians was 4.5 to 17.5 months, with an interquartile range for subsequent diagnosis of 8.0 to 30.0 months.[7] Although screening tools exist to assist primary care providers (pediatricians, family physicians, nurse practitioners) in developmental surveillance, the uptake and consistent application of this

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information is often unsatisfactory.[8,9] Furthermore, screening tools have been designed for the early identification of developmental delays but not aberrant motor development that is more typical for young children with cerebral palsy. Tools currently available for developmental surveillance do not offer explicit referral guidelines or distinct, user-friendly clinical descriptors of atypical movements or postures seen in children with cerebral palsy.[10-12] Accordingly, these tools are lacking for identification of abnormal motor development and quality of movement, which characterize young children with cerebral palsy, as opposed to delays in development.[6]

Very little evidence has been published on the particular attributes that may be interpreted by primary care physicians as indicators of referral for early detection of cerebral palsy and there is minimal reported information about the process and timing of cerebral palsy diagnosis. One study by Sharkey et al[13] examined the relationship between age at referral for investigation of a physical disability (most eventually diagnosed as cerebral palsy) and developmental outcome at 18 months of age and found that those referred earlier (<9 months old) demonstrated significantly greater gains ($P < .001$, all areas of development) at follow-up developmental evaluation than those referred later, even though these 2 groups were developmentally similar as infants prior to intervention. Furthermore, there is a significant lack of evidence in the current literature concerning trends and patterns in age at referral to neurologists and rehabilitation specialists in relation to different variables (severity of motor impairment, complications during pregnancy, regional, and community factors). Among the limited existing findings, most date back to studies conducted more than 30 years ago and differ in their conclusions of timing of diagnosis. In Australia (birth cohorts 1956-1975), mean age of diagnosis was 21 months, whereas another study examining US birth cohorts of the 1970s and 1980s found mean age for diagnosis to be 10 to 12 months.[14-16] Moreover, existing evidence is often focused on subsets of high-risk children, such as those originally admitted to the neonatal intensive care unit after birth, rather than studying a more representative population-based sample with a wider range of types and severities of cerebral palsy. In a Swedish cohort of children diagnosed with cerebral palsy after formal developmental screening, mean age at diagnosis was 12.4 months. Children with severe cerebral palsy were diagnosed at 7.1 months. Indeed, when excluding those with a high-risk birth history, 24% were referred around 2 years of age and 16% were 44 to 54 months of age.[17]

Therefore, the objective of this study is to bridge the knowledge gap about the present state of cerebral palsy diagnosis and reveal the current timeline of age at referral to medical and rehabilitation specialists. The primary research question is to what extent do birth history, disability profile, sociodemographic variables, and type of referring physician influence the age at referral to a medical specialist for diagnosis and age at referral to rehabilitation specialists for intervention among children newly diagnosed with cerebral palsy.

There are thus 2 main objectives for this descriptive study:

1. Mapping the current referral practices of primary care practitioners concerning age at referral to medical specialists for diagnosis and age at referral to rehabilitation specialists for intervention.
2. Identifying subgroups of children with cerebral palsy who are more likely to experience delays in diagnosis and referral (type of cerebral palsy, uneventful birth history).

Methods

Sample Characteristics

Our study population was derived from the computerized database of a single pediatric neurologist (MS), containing patients seen at the following sites: the Montreal Children's Hospital Neurology Outpatient Clinic, the Montreal Children's Hospital Neonatal Follow-up Clinic, and the Children's Care Clinic (a suburban group practice). The medical records of the patients who met the following inclusion criteria were included: (1) diagnosis of cerebral palsy and (2) born between 2002 and 2012. Immigration to Canada after birth was the single exclusion criterion, as this may cause delay in diagnosis. The hospital's Director of Professional Services approved data collection and analysis for this study as is required for an anonymous chart review.

Operational Terms

Cerebral palsy was defined as a disorder of movement and posture that, according to the international consensus definition by Rosenbaum et al,[18] is often accompanied by secondary musculoskeletal problems and other comorbidities. For the purposes of this study, primary care provider/physician/practitioner was defined as a pediatrician, family physician, or nurse practitioner who works in the community and provides health surveillance and anticipatory guidance care that is focused on prevention and early detection of health conditions. As such, the role of the primary care provider would include the early identification of young children who are suspected of having cerebral palsy. The neurologist was considered to be the medical specialist that the child was referred to for possible diagnosis, investigation, and medical management of cerebral palsy. It was a pediatric neurologist in this study, but could also be a developmental pediatrician or any other medical specialist with expertise in developmental disability. Rehabilitation specialists in this study referred specifically to occupational therapists and physical therapists. In the context of this study, other medical specialists were considered to be any other type of subspecialist who could have referred the child to a neurologist for diagnosis (such as an orthopedic surgeon, neonatologist, cardiologist, or neurosurgeon). When considering potential predictor variables, birth history referred to admission to the neonatal intensive care unit and to history of prematurity. Each patient's disability profile was considered to be the specific cerebral palsy diagnosis received (subtype). Finally, sociodemographic variables included sex, parity (first born/first born with live twin/not first born), referral source (primary care, other specialist), and immigration to Canada.

Data Extraction Procedures

Three independent assessors reviewed the medical records of a random sample of patients that met the aforementioned inclusion criteria. The primary outcome of interest was: age at referral by a physician to a medical specialist (neurologist) for diagnosis of cerebral palsy, and age at referral to rehabilitation services (occupational therapy and physical therapy) for children suspected or first diagnosed with cerebral palsy. Age when first seen by the neurologist for diagnosis and age when first seen by rehabilitation services were documented. The following variables were noted as potential independent (predictor) variables: sex, parity (including information about adoption, twinship, and siblings when available), admission to a neonatal intensive care unit, type of physician (primary care physician, medical specialist) referring to the neurologist for diagnosis, and the type of cerebral palsy diagnosed (hemiplegia, diplegia, quadriplegia, mixed, and other). Immigration to Canada was noted for exclusion, as this could account for delays in referral.

In order to maximize consistency and uniformity of the medical chart reviews between raters, an inter rater agreement exercise was conducted. Two assessors independently reviewed 10 charts and subsequently compared findings for all variables. This revealed the areas most susceptible to discrepant interpretation and led to the creation of a clear and unified rating system understood and implemented by independent reviewers for the chart review process.

The most common and problematic discrepancies that arose were related to the date of referral to and date seen by a medical or rehabilitation specialist. Many charts showed that the patient was seen at the hospital during the first stay (within a few days or weeks following birth), but this usually did not involve a formal referral to the specialist or a scheduled appointment for the purposes of verification of a diagnosis of cerebral palsy. Therefore, it was agreed that this data would not be considered and that rather, the first referral by a primary care physician or medical specialist specifically for diagnosis of cerebral palsy and the first scheduled appointment with the medical specialist (neurologist) who diagnosed the child—typically found outside other medical visits or admissions to the hospital following birth—would be used. Furthermore, if the patient received an appointment and was seen by a rehabilitation specialist through a private institution before being seen at the hospital, this information was favored as it occurred at an earlier date. Between occupational and physical therapy, preference was given based on first date of occurrence of either. Concerning parity, birth order in relation to siblings was always registered, and any other relevant information was also noted when available (age and gender of siblings, twinship, adoption).

Statistical Analyses

Descriptive statistics were used to characterize the sample and describe age at referral patterns. We evaluated if particular exposure variables predicted the age first referred to a neurologist for diagnosis of cerebral palsy. Using the Statistical Package for the Social Sciences (SPSS), version 22.0, linear regressions, analysis of variance, and *t* tests were conducted. The assumptions of linear regression were verified.[19] Simple linear regressions were conducted with each of the 5 potential predictors (sex, parity, complicated birth history, referral source, type of cerebral palsy). The dependent variable was age (in months) at time of referral to a neurologist for diagnosis. A similar analysis was conducted with age (in months) at time of referral to a rehabilitation specialist as the dependent variable.

Results

Sample Characteristics

A total of 119 medical charts were reviewed. Cases (children diagnosed with cerebral palsy) where the patient immigrated to Canada (n = 16) in childhood were excluded, and analysis was conducted on the final retrospective sample comprising 103 patients. The sample consisted of 58 (56.3%) males and 45 females (47.3%). Information on parity was available in 95 cases: 50 (52.6%) were first born, 39 (41.1%) were not first born and had a living sibling, and 6 (6.3%) were first born with a living twin. Of the charts where information on perinatal/birth history was available (n = 98), 60 (61.2%) had been admitted to the neonatal intensive care unit compared to the 38 (38.8%) who had not. Of 103 patients, 81 (78.6%) had been referred to the neurologist for diagnosis by another medical specialist, compared to 22 (21.4%) who had been referred by a primary care physician. The cerebral palsy subtype was established for all 103 patients, and the majority had a spastic subtype of cerebral palsy: 39 (37.9%) hemiplegia, 24 (23.3%) diplegia, 23 (22.3%) quadriplegia, and 17 (16.5%) other.

Factors Influencing Age at Referral to a Medical Specialist

Of the charts where information on age at referral for diagnosis of cerebral palsy was available (n = 99/103), the range was highly variable (0.1-89.9 months, mean = 16.6 $\pm$ 19.2 months); 57 (58%) were referred at $\leq$1 year of age, 23 (23%) between >1 and <2 years of age, 6 (6%) between $\geq$2 and <3 years of age, and 13 (13%) were $\geq$3 years of age.

The subset of children hospitalized in the neonatal intensive care unit (n = 58) was referred to a neurologist for diagnosis earlier (mean: 9.3 $\pm$ 10.2 months) than the subset not originally in a neonatal intensive care unit (n = 36) (28.1 $\pm$ 24.9 months) ($\beta = -18.8$, $r^2 = 0.22$, $P < .001$). Furthermore, these results illustrate that an important subset of patients (10/32; 31.3%), who were not originally admitted to a neonatal intensive care unit, were over 2 years of age when they were referred to rehabilitation services, compared to patients in the neonatal intensive care unit, for which referral after 2 years of age was rare (3/53; 5.7%).

The majority of children (n = 81) were referred to a neurologist for diagnosis by other medical specialists (78.6%) at a mean age of 13.6 $\pm$ 15.7 months, whereas a primary care provider referred the remaining cases (21.4%) at a mean age of 28.8 $\pm$ 27.1 months ($\beta = -15.2$, $r^2 = 0.10$, $P = .002$). The majority of patients referred for diagnosis under 1 year of age (50/57; 87.7%) were referred by other specialists, not primary care practitioners. Primary care practitioners are unlikely to refer, and when they did, it was often at an older age: 7/19 (37%) were $\geq$2 years of age at referral to a neurologist for diagnosis. The distribution by age bracket of age at referral for diagnosis according to type of referring physician can be found in Table 1.

A significant difference was found between the various types of cerebral palsy and age at referral for diagnosis:

Table 1. Age at Referral to Neurologist and Rehabilitation Specialist by Referring Physician Type.

	Age at referral			
	≤1 y old	>1 and <2 y old	≥2 and <3 y old	≥3 y old
To neurologist for diagnosis				
By medical specialist (n = 80)	62% (n = 50)	23% (n = 18)	5% (n = 4)	10% (n = 8)
By primary care physician (n = 19)	37% (n = 7)	26% (n = 5)	11% (n = 2)	26% (n = 5)
To rehabilitation specialist for intervention				
By medical specialist (n = 72)	64% (n =46)	21% (n = 15)	4% (n = 3)	11% (n = 8)
By primary care physician (n = 18)	61% (n = 11)	22% (n = 4)	6% (n = 1)	11% (n = 2)

diplegia (n = 21), mean age of 31.6 ± 24.9 months; hemiplegia (n = 38), mean age of 15.2 ± 16.5 months; quadriplegia (n = 23), mean age of 10.9 ± 16.9 months; mixed (n = 1), age of 2.6 months; other (n = 16), mean age of 8.8 ± 7.1 months ($F = 5.357$, $P < .001$). Analysis of any association between sex and parity as potential predictor variables of referral age did not yield significant results. The degree to which the aforementioned potential predictor (independent) variables were associated with age at referral for diagnosis (dependent variable) can be found in Table 2.

Table 2. Results of Linear Regressions Conducted.

Potential predictor variables for earlier referral	Age at referral for diagnosis	Age at referral to rehabilitation services
Sex (male/female)	$\beta = -2.2$ $R^2 = .003$ $P = .571$	$\beta = -4.5$ $R^2 = .021$ $P = .178$
Parity (first born/first born with living twin/not first born)	$\beta = -1.6$ $R^2 = .006$ $P = .458$	$\beta = -2.2$ $R^2 = .017$ $P = .233$
Complicated birth history (admission to neonatal intensive care unit)	$\beta = 18.8$ $R^2 = .221$ **$P = .001$**	$\beta = 11.5$ $R^2 = .124$ **$P = .001$**
Referral source to neurologist for diagnosis (medical specialist/ primary care practitioner)	$\beta = -15.2$ $R^2 = .098$ **$P = .002$**	$\beta = -5.9$ $R^2 = .022$ $P = .159$
Type of cerebral palsy (hemiplegia/ diplegia, quadriplegia/other)	$\beta = -2.8$ $R^2 = .028$ $P = .099$	$\beta = -2.7$ $R^2 = .036$ $P = .072$

Factors Influencing Age at Referral to a Rehabilitation Specialist

For the charts in which there was information on age at referral to rehabilitation services (n = 90), the range was similarly variable (0.1-79.5 months, mean = 13.9 ± 15.8 months): 57 (63%) were referred at ≤1 year of age, 19 (21%) between >1 and <2 years of age, 4 (4%) between ≥2 and <3 years of age, and 10 (11%) were ≥3 years of age.

Once again, the subset of children originally hospitalized in the neonatal intensive care unit was referred significantly earlier (mean = 9.4 ± 10.8 months) than those not so admitted (mean = 20.9 ± 20.2 months) ($\beta = 11.5$, $r^2 = .12$, $P < .001$). The distribution by age bracket of age at referral to rehabilitation services according to original admission to a neonatal intensive care unit can be found in Table 3. Analysis of any association between sex and parity as potential predictor variables of referral age did not yield significant results. The distribution by age bracket of age at referral to rehabilitation services according to type of referring physician can be found in Table 1. The degree to which the aforementioned potential predictor (independent) variables were associated with age referred to rehabilitation services (dependent variables) can be found in Table 2.

Discussion

The data highlight a significant delay in referral for diagnosis, and remarkably, a paucity of referrals that were originated by primary care providers (especially family physicians). High variability (age range at referral: 0.1-90.0 months) in both physician groups suggests that knowledge of attributes of cerebral palsy differs widely. Overall, a delay in age at initial referral of children with cerebral palsy to medical specialists for diagnosis (42% of cases >1 year old) and to rehabilitation specialists (37% of cases >1 year old) was found. This delay is problematic because a formal diagnosis of cerebral palsy is a catalyst for (1) medical investigations to ascertain cause and effective management of comorbidities, (2) referral to rehabilitation specialists to provide therapeutic interventions to optimize function, and (3) educating families and providing the appropriate resources and supports to begin the process of family adaptation and coping.[20] Our findings of striking delay in the referral and diagnosis timeline therefore highlight the recognition of the need for early detection and intervention and the reality of delays in referral by primary care practitioners that could have deleterious impacts on child and family functioning and health.

This study also aimed to address the lack of evidence concerning trends and patterns in the referral timeline in relation to different variables. A minority (21.4%) of referrals came from primary care practitioners and these referrals occurred at a significantly later mean age compared to referrals provided by pediatric subspecialists. These results concerning primary care providers are of concern given the latter's vital role in early detection of developmental disabilities. This result underlines the lack of use and/or lack of specificity of existing

Table 3. Age at Referral to Neurologist and Rehabilitation Specialist by Original Admission to a Neonatal Intensive Care Unit.

	Age at referral			
	≤1 y	>1 and <2 y	≥2 and <3 y	≥3 y
To neurologist for diagnosis				
Originally admitted to a neonatal intensive care unit (n = 58)	71% (n = 41)	26% (n = 15)	0% (n = 0)	3% (n = 2)
Not originally admitted to a neonatal intensive care unit (n = 36)	36% (n = 13)	19% (n = 7)	17% (n = 6)	28% (n = 10)
To rehabilitation specialist for intervention				
Originally admitted to a neonatal intensive care unit (n = 53)	74% (n = 39)	21% (n = 11)	1% (n = 1)	4% (n = 2)
Not originally admitted to a neonatal intensive care unit (n = 32)	50% (n = 16)	19% (n = 6)	9% (n = 3)	22% (n = 7)

screening tools by primary care providers as well as their lack of application of knowledge and understanding of the early clinical indicators of cerebral palsy.[7,8] Our findings stress the need to develop user-friendly knowledge translation tools that will enable primary care providers to accurately and promptly detect attributes associated with cerebral palsy.

We also aimed to study a more representative sample consisting of a variety of types of cerebral palsy, as the research to date has tended to focus on subsets of high-risk children. The subset of children hospitalized in the neonatal intensive care unit was referred to a neurologist for diagnosis earlier than those not admitted. Often other specialists were providing services to these children for other reasons at the hospital, whereupon their suspicions about cerebral palsy prompted referral to neurology. Almost half of children with cerebral palsy are not born premature or do not have a complicated birth history and for these, referrals to specialists for diagnosis and management are postponed, often beyond 3 years of age. This suggests that children with an uneventful birth history are a particularly vulnerable subgroup as they are more likely to experience delays in eventual diagnosis and referral, and that future screening practices in the community (by primary care physicians) should be adjusted accordingly.

Age at referral to rehabilitation for assessment was similarly delayed, particularly for those not admitted to the neonatal intensive care unit. These results emphasize primary care physicians' poor understanding of the distinct roles of the various rehabilitation specialties, which typically leads to referral to rehabilitation by a medical specialist rather than the primary care practitioner (in a linear serial model of referral). In the recent published report by the American Academy of Pediatrics' Neuromotor Screening Expert Panel, the recommendation made for children with suspected neuromotor delay is that "concurrent referrals should be made to physical and/or occupational therapists while diagnostic investigations are proceeding," prior to formal medical diagnosis.[6 (p. 2024)] Furthermore, the urgency of simultaneous referral underscores the need for clinical decision tools that, if helpful, can minimize the well documented waiting times for treatment in this population and thereby promote better outcomes. Early interventions by rehabilitation therapists focus on enabling families to be effective caregivers in optimizing the functioning and health of their child with cerebral palsy. The anticipated impact is that parents will be more rapidly informed and better engaged in the process of detection, and will benefit from an earlier access to resources and family supports, thus potentially optimizing physical, social, and emotional functioning in children with cerebral palsy and their families.

An important limitation of this study is that it used a convenience sample; all charts reviewed belonged to patients of a single neurologist (MS) from 1 tertiary care hospital setting. In order to validate these findings, an environmental scan is essential, which is expected to provide a population-based understanding of current referral practices for initial diagnosis of cerebral palsy and rehabilitation services. Certain limitations are inherent in the chart review methodology. These include incomplete documentation, including missing charts, information that is unrecoverable or unrecorded, difficulty interpreting information found in the documents (such as jargon, acronyms, photocopies, and microfiches), and variance in the quality of information recorded by medical professionals.[21] We attempted to minimize most of these difficulties by conducting a reliability exercise and creating a standardized method of interpreting/collecting the data. Certain limitations such as unavailable information or the physicians' inconsistent recording of certain variables could not be controlled for. For example, in this feasibility study, the severity level according to the Gross Motor Function Classification System score could not be studied as part of the disability profile potential predictor variable as, too often, it was not recorded consistently in the medical chart.[22] A national study is now underway in 4 regions of Canada, which will enable the recruitment of a large population-based sample from the Cerebral Palsy Registry. Severity of cerebral palsy (Gross Motor Function Classification System, Manual Ability Classification System), perinatal factors, and etiologic determinants are collected in a standardized fashion in the registry and will be examined as potential determinants of early or late referral for diagnosis.

This scan of referral practices, conducted with a convenience sample drawn from the patient database of a single neurologist (MS) at a tertiary care pediatric hospital, reports objective contemporary evidence of delays in referral for diagnosis and to rehabilitation services for children with cerebral palsy. The results highlight that there are subgroups of children with cerebral palsy who are experiencing pronounced delays in this process (children who were not admitted to a neonatal intensive care unit; children with hemiplegia and especially diplegia). The considerable variability in the referral practices of both

primary care practitioners and medical specialists illustrates the knowledge gap that exists with respect to timely and simultaneous referral practices to medical and rehabilitation specialists for young children suspected of having cerebral palsy. A larger-scale, population-based environmental scan of referral practices would be crucial to better understand current referral practices and to inform the creation of knowledge translation decision tools designed to prompt earlier and simultaneous referral to appropriate specialists for diagnosis, medical management, and therapeutic intervention.

Acknowledgments

We are grateful to Joey Waknin and Christopher Saunders for their assistance with the data collection and chart review logistics. Anna Radzioch Guérin provided statistical training and support. Marie Belmonte provided the necessary administrative assistance to procure the charts for review in a timely manner. Zachary Boychuck was supported as a doctoral trainee by an operating grant from the Canadian Institutes of Health Research. In addition, we benefited from research infrastructure provided by the Montreal Children's Hospital Research Institute and the Centre de recherche interdisciplinaire en réadaptation du Montréal métropolitain; both funded by Fonds de recherche du Québec–Santé.

Author Contributions

AM, MS and ZB contributed to the study conceptualization and data analysis. LH wrote the manuscript's first draft.

Declaration of Conflicting Interests

The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

The authors disclosed receipt of the following financial support for the research, authorship, and/or publication of this article: Zachary Boychuck received salary support as a doctoral trainee from funding from the Canadian Institutes of Health Research (MOP-133402), which supported this study in part.

Ethical Approval

The Montreal Children's Hospital-McGill University Health Centre Associate Director for Professional Services provided approval for systematic chart review.

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CHAPTER 6:

INTEGRATION OF MANUSCRIPTS 2 AND 3

6.1 Research Questions of Manuscripts 2 and 3

Manuscript 2:

"To what extent do birth history, disability profile [type of CP], sociodemographic variables, and type of referring physician influence the age at referral to a medical specialist for diagnosis and age at referral to rehabilitation specialists for intervention among children newly diagnosed with cerebral palsy?"

Manuscript 3:

"To what extent do sociodemographic and child characteristics, birth history, CP profile [type of CP, severity/GMFCS level], and referral source for diagnosis influence the age at referral for diagnostic assessment and for rehabilitation services among Canadian children with CP?"

6.2 Integration of Manuscripts 2 and 3

In **Manuscript 2** we used a retrospective chart review methodology to describe current local referral practices with respect to age at referral for diagnostic assessment of CP, and identified factors that were potentially predictive of delayed referral. It also served as a feasibility study for the subsequent study. Informed by these results, in **Manuscript 3** we conducted a multi-site national environmental scan of Canadian physician referral practices for children suspected of having CP. It provided a population-based description of current practices with respect to age at referral from specialist and primary care contexts to pediatric medical specialists for diagnostic assessment of CP, and to rehabilitation specialists for intervention. Factors associated with delayed referral were also identified.

Chapter 7:

Manuscript 3

Current referral practices for diagnosis and intervention for children with cerebral palsy: A national environmental scan

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Keywords: Early Identification, Detection, Primary Care, Rehabilitation

Conflict of interest statement: The study sponsor had no role in the design; collection, analysis, or interpretation of data; writing of the report; or decision to submit the manuscript for publication. ZB and AM wrote the first draft of the manuscript. No honorarium, grant, or other form of payment was given to anyone to produce this manuscript. The authors declare no potential conflicts of interest with respect to the authorship and/or publication of this article.

Funding sources statement: AM received funding from Canadian Institutes of Health Research for the study "PROMPT Identification of Cerebral Palsy-Primary care Referral Of Motor-impaired children: Physician Tools" (MOP-133402); ZB received financial support from the Richard and Edith Strauss Canada Foundation as a Musculoskeletal Doctoral Fellow, as well as from the Hoppenheim Fund/Montreal Children Hospital Foundation.

Data Statement: The research data is confidential.

Abbreviations: CP **(**Cerebral palsy), CCPR (Canadian CP Registry), GMFCS (Gross Motor Function Classification System)

This manuscript was accepted for publication by *The Journal of Pediatrics* on

September 13, 2019; Published in Issue January 2020.

Boychuck, Z., Andersen, J., Fehlings, D., Kirton, A., Oskoui, M., Shevell, M., Majnemer, A., & the PROMPT group. (2020). Current referral practices for diagnosis and intervention for children with cerebral palsy: A national environmental scan. *The Journal of Pediatrics*, 216, 174-181.

Current Referral Practices for Diagnosis and Intervention for Children with Cerebral Palsy: A National Environmental Scan

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Objectives To describe current physician referral practices with respect to age at referral to medical specialists for initial diagnosis of cerebral palsy (CP) and rehabilitation specialists for intervention and to identify factors associated with delayed referral.

Study design National environmental scan of 455 children diagnosed with CP who were born in Canada between 2008 and 2011, selected from 4 sites within the Canadian CP Registry (Edmonton, Calgary, Toronto, and Montreal). Two sources of information were used—children's medical charts and the population-based registry, which provided corresponding data for each child. Primary outcomes extracted from the charts were age at referral for diagnostic assessment, age at diagnosis, age at referral for rehabilitation services, and age at initial rehabilitation intervention. Twelve variables were explored as potential predictors. Descriptive statistics, bivariate analyses, and multiple linear regressions were conducted.

Results Median age (in months) at referral for diagnostic assessment was 8 (mean: 12.7 ± 14.3), diagnosis 16 (mean: 18.9 ± 12.8), referral for rehabilitation services 10 (mean: 13.4 ± 13.5), and rehabilitation initiation 12 (mean: 15.9 ± 12.9). Lower maternal education, mild severity of motor dysfunction, type of CP, early discharge after birth, and region of residence explained between 20% and 32% of the variance in age at referral for assessment, diagnosis, referral for rehabilitation, and rehabilitation initiation.

Conclusions Findings suggest wide variability exists in the age at which young children with CP are referred to specialists for diagnosis and intervention. User-friendly tools are therefore needed to enhance early detection and referral strategies by primary care practitioners, to ensure early interventions to optimize developmental outcomes and enhance opportunities for neural repair at a younger age. *(J Pediatr 2019;■:1-8)*.

Cerebral palsy (CP) is a disorder of movement and posture resulting from injury to the developing brain, and it is the most common cause of physical disability in children.[1,2] Studies in animals have provided compelling evidence of the benefits of early intervention in optimizing brain development and function attainment.[3-6] Interventions using novel strategies (eg, intensive constraint and bimanual approaches) have the potential to modulate maladaptive circuitry of the damaged brain.[7-9] Early intensive training may facilitate brain organization during the critical period of plasticity, which combined with early family education and support, can enhance long-term outcomes for the child and family.[10-12]

North American pediatric societies widely endorse early identification of developmental disabilities by primary care practitioners to initiate interventions without delay.[13,14] Existing screening tools focus on delayed milestone acquisition but do not delineate attributes related to abnormal quality of movement essential to the timely detection of CP. The knowledge gap by primary care practitioners in the early detection of motor disorders has been a concern raised by the American Academy of Pediatrics.[15] Parents of children with CP have expressed their dissatisfaction associated with delays in the diagnostic process resulting in negative repercussions to adaptive coping and personal health.[16-18] Current research in the field of early identification of CP has focused primarily on children considered to be at high risk for CP who experienced neonatal adversity. However, approximately one-half of children eventually diagnosed with CP

CP	Cerebral palsy
CCPR	Canadian CP Registry
GMFCS	Gross Motor Function Classification System

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A.M. received funding from Canadian Institutes of Health Research for the study "PROMPT Identification of Cerebral Palsy-Primary Care Referral of Motor-impaired children: Physician Tools" (MOP-133402). Z.B. received salary support as a doctoral trainee from funding for the project from the Canadian Institutes of Health Research (MOP-133402); studentship support from the Richard and Edith Strauss Canada Foundation as a Musculoskeletal Doctoral Fellow; the Hoppenheim Fund/Montreal Children Hospital Foundation; and the Judith Kornbluth Gelfand Fellowship in Pediatric Rehabilitation. J.A. received support from Stollery Children's Hospital Foundation through the Women and Children's Health Research Institute. The other authors declare no conflicts of interest.

https://doi.org/10.1016/j.jpeds.2019.09.035

are born at term after an uneventful pregnancy or are born premature but above the gestational age cut-off for neonatal follow-up. Traditionally, physicians have assumed a "wait-and-see" approach for the diagnosis of CP.[10] In addition, if primary care practitioners do eventually refer the child for diagnostic assessment, they may not simultaneously refer the child to rehabilitation professionals, and thus the initiation of rehabilitation services may further be delayed.[19]

A recent scoping review found that published evidence described older studies of birth cohorts from the mid-1950s to the 1980s, and referral practices may have changed over time.[20] Most reported samples were not population-based, potentially biasing the data to favor documentation of earlier referral of more high-risk infants. Furthermore, the age of referral was found to be widely variable, with an important subset whose referral was delayed as late as 2-4 years of age. Results from a single-site study conducted by our group identified a subset of children with CP (hemiplegia and especially diplegia) who experienced delayed referral for diagnosis and rehabilitation and might be considered as potential priorities for efforts aimed at decreasing the age at referral.[19]

This study aimed to describe current practices within a population-based sample across multiple sites, with respect to the age at which children suspected of having CP are referred to medical and rehabilitation specialists and factors that are associated with earlier referral. More specifically, we aimed to determine to what extent do sociodemographic and child characteristics, birth history, CP profile, and referral source influence the age at referral for diagnostic assessment and for rehabilitation services among Canadian children with CP.

Methods

A historical cohort design was applied in this environmental scan of current physician referral practices for the diagnosis of CP and subsequent rehabilitation intervention. The cohort included children who were registered in a population-based registry, across four regions of Canada.

Participants were recruited via the CCPR (Canadian CP Registry; https://www.cpregistry.ca/), a voluntary, confidential national database of medical and sociodemographic information about children with CP and their families. Specifically, the CCPR serves to provide a national profile of children with CP, identify potential risk factors, collect epidemiologic data, and provide an infrastructure for researchers conducting population-based studies on CP. As part of the data-collection process, the CCPR gathers information via parental interviews and is also granted access to the medical charts of the mother and child. Participants were identified via the CCPR platform and had consented previously to having their information, including medical histories, potentially used in future research projects. Although no further contact with the children or parents was required, to access their medical charts, ethical approval was required and obtained from the following institutional research ethics boards: Conjoint Health Research Ethics Board, Calgary, Alberta (E-24218); Health Research Ethics Board–Health Panel, Edmonton, Alberta (Pro00015821); Holland Bloorview Research Ethics Board, Toronto, Ontario (10-179); McGill University Health Centre, Montreal, Quebec (09-234-PED); Centre hospitalier universitaire Sainte-Justine, Montreal, Quebec (3059); Comité d'éthique de la recherche des établissements du CRIR, Quebec (CRIR-517-0510); and the Shriner's Hospital for Children, Montreal, Quebec (A06-M64-11A).

Participants for this study included CCPR birth cohorts from 2008 to 2011 inclusive, allowing confirmation of diagnosis at 5 years of age. This restricted the sample to the 7 clinical regional centers that were enrolling participants between 2008 and 2011: Alberta Children's Hospital, Calgary, Alberta; Glenrose Rehabilitation Hospital, Edmonton, Alberta; Holland Bloorview Kids Rehabilitation Hospital, Toronto, Ontario; MAB-Mackay Rehabilitation Center/Montreal Children's Hospital/Hopital St-Justine/L'Institut de réadaptation en déficience physique de Québec/Shriners Hospital for Children, Quebec. Participants were excluded if (1) the child's family immigrated to Canada, as that could account for delays observed; or (2) the charts did not include information on at least one of the primary outcomes.

Data Sources and Extraction Procedures

At the onset of this study, the CCPR was not collecting data on age at referral for diagnostic assessment of CP, age at diagnosis of CP, age at referral for rehabilitation services, age at initiation of rehabilitation services, or referral sources. Working in collaboration with the co-directors of the CCPR and the National Coordinators, the authors created 5 additional variables that were subsequently added to the CCPR data collection manual: (1) date of referral for diagnosis; (2) referral source for diagnosis (medical specialist [eg, orthopedist, physiatrist, otolaryngologist] or primary care practitioner [eg, community-based pediatrician, family physician]); (3) date seen by a specialist for diagnosis; (4) date of initial referral for rehabilitation services; and (5) date of initiation of rehabilitation services.

Retrospective reviews of the medical charts of the child and mother were conducted across the 7 sites between November 24, 2014, and December 5, 2018, by 7 dedicated CCPR site coordinators or research assistants who were trained and familiar with both the CCPR and its REDCap online data platform. To ensure consistent data-collection strategies across sites and assistants, the data collection was standardized and incorporated into the CCPR Data Collection Manual. Support was provided as needed throughout the data collection process via telephone and e-mail. Using the child's date of birth and the information collected, we were able to determine the following primary outcomes of interest: age at referral by a physician to a medical specialist for diagnosis of CP, age at diagnosis of CP, age at referral to rehabilitation services, and age at initiation of rehabilitation services.

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CCPR. The following information was available for each participant via the CCPR and was extracted and explored as potential predictors of referral. Sociodemographic variables: sex; socioeconomic status (maternal and paternal education level); urban/rural (as per Statistics Canada mapping system: Postal Code^{OM} Conversion File, Reference Guide, 2016. Statistics Canada Catalogue no. 92-154-G). Birth history: gestational age (in weeks), extended hospitalization after birth (hospitalization ≥4 days); parity, prematurity (yes/no). CP profile: severity of motor dysfunction (Gross Motor Function Classification System [GMFCS] score; GMFCS Level I-III [ambulatory], GMFCS Level IV-V [non-ambulatory]); type of CP (hemiplegia, diplegia, triplegia/quadriplegia, or "other"). The final potential predictor considered was region (CCPR regional center; Edmonton, Calgary, Toronto, and Montreal).

Statistical Analyses

Using Statistical Analysis System, version 9.4 (SAS Institute, Cary, North Carolina), descriptive statistics were applied to characterize our population-based sample of children with a confirmed diagnosis of CP. Mean, SD, median, and range were determined for all 4 age outcomes (age at referral for diagnosis, age at diagnosis, age at referral for rehabilitation, age at initiation of rehabilitation services; in months, to one decimal place). Bivariate analyses were then conducted. Correlations were used to assess the association between the 4 age outcomes and the sole continuous potential predictor variable (gestational age); *t* tests/ANOVA were conducted with the 4 age outcomes and the 11 categorical potential predictor variables (sex; parity; urban/rural; maternal education; paternal education; extended hospitalization after birth; prematurity; severity of motor dysfunction; type of CP; referral source for diagnosis; and region). Informed by these results, multiple linear regressions were conducted with the potential predictors that demonstrated the most significance on the bivariate analyses as the independent variables, and the 4 age outcomes as the dependent variables.

Results

Of the 1850 children enrolled in the CCPR, 520 were born between the years 2008 and 2011 at the participating recruitment sites. Cases (children diagnosed with CP) for whom their family immigrated to Canada in early childhood were excluded (n = 60), as were cases for which the charts did not contain information on at least one of the primary age outcomes (n = 5). Thus, 455 participants with a confirmed diagnosis of CP at 5 years of age were retained as the sample for detailed analysis.

Table I provides an overview of child and sociodemographic characteristics, birth history, CP profile, and referral source. As expected, there was a preponderance of males. Almost one-half of children were first-born. The majority of families lived in urban settings, and parental education was primarily college education or greater. The median gestational age was 37.0 weeks, 43.9% of children were born premature (<37 weeks of gestation), and 69.3% of children required extended hospitalization after birth. The most common type of CP was hemiplegia, and most children were GMFCS Level I-III (ambulatory with or without assistance).

Outcome Variables

Table II provides detailed descriptive statistics for age at referral for diagnostic assessment, age at diagnosis, age at referral to rehabilitation specialists, and age at initiation of rehabilitation services. Most cases had documentation of age at diagnosis; however, a smaller subset (75.8%-83.3%) had documentation of age at referral for diagnostic assessment, age at referral to rehabilitation specialists, and age at rehabilitation initiation. All 4 outcomes demonstrated high variability across sites, and the data highlight that for each outcome there was a subset of children who were >1 year of age: age at referral for diagnostic assessment (>1 year: 37.6%), age at diagnosis (>1 year: 68.6%), age at referral for rehabilitation (>1 year: 40.1%), and age at initiation of rehabilitation (>1 year: 48.3%).

Bivariate Analyses

Table III presents the significance of all 12 predictor variables against all 4 age outcomes. All ages that follow are in months.

Factors Influencing Age at Referral for Diagnostic Assessment

Maternal Education. Children whose mothers completed a college/trade program were referred later (n = 73; mean: 15.9 ± 18.1; median: 12.0) than children whose mothers completed university (n = 96; mean: 10.4 ± 12.1; median: 6.0) and high school or less (n = 123; mean: 11.8 ± 11.3; median: 9.0; $F(2, 289) = 3.63$, $P = .0276$).

Extended Hospitalization After Birth. Children who went home within 3 days of birth (n = 100) were referred at a mean age of 16.7 ± 11.7 (median: 13.5), much later than those initially requiring intensive care (n = 220) who were referred at a mean age of 9.8 ± 13.1 (median: 4.0; $t[318] = 4.51$, $P < .0001$).

Severity of Motor Dysfunction. Children with GMFCS Level I-III (ambulatory, n = 245) were referred at a later age (mean: 14.4 ± 13.6; median: 12.0), compared with those with GMFCS Level IV-V (n = 95; mean: 8.3 ± 15.1; median: 3.9; $F[1, 338] = 13.09$, $P = .0003$).

Type of CP. Children with diplegia (n = 68; mean: 18.2 ± 16.7; median: 15.5), hemiplegia (n = 133; mean: 13.4 ± 12.7; median: 10.0), and "other" types of CP (n = 47; mean: 13.9 ± 19.0; median: 8.0) were referred significantly later than children with triplegia/quadriplegia (n = 97; mean: 7.2 ± 9.4; median: 3.9; $F[3, 341] = 8.90$, $P < .0001$).

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Table I. Sample patient characteristics

Potential predictor variables	Whole sample (n = 455)	Edmonton (n = 190)	Calgary (n = 89)	Toronto (n = 103)	Montreal (n = 73)
Sociodemographic variables					
Maternal education	(n = 395)	(n = 143)	(n = 86)	(n = 95)	(n = 71)
High school or less, n (%)	161 (40.8)	72 (50.3)	32 (37.2)	29 (30.5)	28 (39.4)
College/trade, n (%)	102 (25.8)	28 (19.6)	28 (32.6)	30 (31.6)	16 (22.5)
University, n (%)	132 (33.4)	43 (30.0)	26 (30.2)	36 (37.9)	27 (38.0)
Paternal education	(n = 363)	(n = 127)	(n = 83)	(n = 87)	(n = 66)
High school or less, n (%)	141 (38.8)	54 (42.5)	30 (36.1)	34 (39.1)	23 (34.8)
College/trade, n (%)	112 (30.9)	48 (37.8)	28 (33.7)	19 (21.8)	17 (25.8)
University, n (%)	110 (30.3)	25 (19.7)	25 (30.1)	34 (39.1)	26 (39.4)
Sex	(n = 455)	(n = 190)	(n = 89)	(n = 103)	(n = 73)
Male, n (%)	264 (58.0)	103 (54.2)	55 (61.8)	61 (59.2)	45 (61.6)
Female, n (%)	191 (42.0)	87 (45.8)	34 (38.2)	42 (40.8)	28 (38.4)
Urban/rural	(n = 405)	(n = 152)	(n = 79)	(n = 103)	(n = 71)
Rural, n (%)	16 (4.0)	9 (6.0)	3 (3.8)	0 (0.0)	4 (5.6)
Small population (1000-29 999), n (%)	56 (13.8)	37 (24.3)	19 (24.1)	0 (0.0)	0 (0.0)
Medium population (30 000-99 999), n (%)	31 (7.7)	16 (10.5)	15 (19.0)	0 (0.0)	0 (0.0)
Large urban population (100 000+), n (%)	302 (74.6)	90 (59.2)	42 (53.2)	103 (100.0)	67 (94.4)
Birth history					
Gestational age, wk	(n = 453)	(n = 189)	(n = 89)	(n = 102)	(n = 73)
Mean ± SD	35.1 ± 5.1	35.6 ± 4.8	34.8 ± 5.5	34.2 ± 5.4	35.4 ± 4.9
Median	37.0	38.0	37.0	36.0	37.0
Range	23.0-42.0	24.0-41.0	23.0-41.0	23.0-42.0	25.0-41.0
Extended hospitalization after birth	(n = 414)	(n = 177)	(n = 87)	(n = 83)	(n = 67)
No, n (%)	127 (30.7)	62 (35.0)	22 (25.3)	24 (28.9)	19 (28.4)
Yes, n (%)	287 (69.3)	115 (65.0)	65 (74.7)	59 (71.1)	48 (71.6)
Parity	(n = 448)	(n = 186)	(n = 88)	(n = 102)	(n = 72)
First child, n (%)	208 (46.4)	87 (46.8)	36 (40.9)	48 (47.1)	37 (51.4)
Second child, n (%)	133 (29.7)	52 (28.0)	31 (35.2)	31 (30.4)	19 (26.4)
≥Third child, n (%)	107 (23.9)	47 (25.3)	21 (23.9)	23 (22.5)	16 (22.2)
Prematurity	(n = 453)	(n = 189)	(n = 89)	(n = 102)	(n = 73)
No, n (%)	254 (56.1)	116 (61.4)	47 (52.8)	50 (49.0)	41 (56.2)
Yes, n (%)	199 (43.9)	73 (38.6)	42 (47.2)	52 (51.0)	32 (43.8)
CP profile					
Severity of motor dysfunction	(n = 445)	(n = 189)	(n = 89)	(n = 101)	(n = 66)
GMFCS Level I-III (ambulatory), n (%)	326 (73.3)	133 (70.4)	71 (79.8)	74 (73.3)	48 (72.7)
GMFCS Level IV-V (non-ambulatory), n (%)	119 (26.7)	56 (29.6)	18 (20.2)	27 (26.7)	18 (27.3)
Type of CP	(n = 453)	(n = 190)	(n = 89)	(n = 101)	(n = 73)
Hemiplegia, n (%)	179 (39.5)	83 (43.7)	37 (41.6)	33 (32.7)	26 (35.6)
Diplegia, n (%)	88 (19.4)	35 (18.4)	16 (18.0)	20 (19.8)	17 (23.3)
Tri/quadriplegia, n (%)	122 (26.9)	51 (26.8)	24 (27.0)	27 (26.7)	20 (27.4)
Other, n (%)	64 (14.1)	21 (11.1)	12 (13.5)	21 (20.8)	10 (13.7)
Referral source for diagnosis					
Referral source for diagnosis	(n = 371)	(n = 187)	(n = 79)	(n = 58)	(n = 47)
Medical specialist, n (%)	188 (50.7)	107 (57.2)	45 (57.0)	25 (43.1)	11 (23.4)
Primary care practitioner, n (%)	183 (49.3)	80 (42.8)	34 (43.0)	33 (56.9)	36 (76.6)

CP, cerebral palsy; *GMFCS*, Gross Motor Function Classification System; *n*, sample size; *SD*, standard deviation.

Referral Source. The majority of children (53.8%; n = 178) were referred for diagnosis by another medical specialist at a mean age of 9.1 ± 12.5 (median: 3.5), and children referred by a primary care provider (46.2%; n = 153) were significantly older (mean: 17.5 ± 15.3; median: 14.0; t[292.85] = 5.42, $P < .0001$).

Site. As detailed in Table II, the mean age of referral was younger for children in Edmonton and Calgary than those in Toronto and Montreal (F[3, 341] = 6.97, $P = .0001$). Analysis of any association between sex, parity, urban/rural, paternal education, gestational age, and prematurity as potential predictors of referral age did not yield significant results.

Factors Influencing Age at Diagnosis

Sex. Female children (n = 175) received a diagnosis at a mean age of 20.6 ± 14.1 (median: 18.0), which was delayed compared with male children (n = 245; mean: 17.6 ± 11.7; median: 15.0; t[331.33] = −2.27, $P = .0236$).

Maternal Education. Children whose mothers completed a college/trade program (n = 91; mean: 19.3 ± 16.8; median: 15.0) were diagnosed later than children whose mothers completed university (n = 122; mean: 15.5 ± 10.4; median: 13.0) and high school or less (n = 150; mean: 18.9 ± 9.9; median: 17.0; F[2, 360] = 3.42, $P = .0339$).

Severity of Motor Dysfunction. Children with GMFCS Level I-III were diagnosed later (n = 301; mean:

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Table II. Outcome variables

Age, mo	Whole sample (n = 455)	Edmonton (n = 190)	Calgary (n = 89)	Toronto (n = 103)	Montreal (n = 73)
Age at referral for diagnostic assessment	(n = 345)	(n = 177)	(n = 84)	(n = 38)	(n = 46)
Mean ± SD	12.7 ± 14.3	11.0 ± 13.1	11.0 ± 12.0	21.8 ± 15.8	14.5 ± 17.9
Median	8.0	6.0	6.0	18.0	9.5
Range	0.0-114.0	0.0-65.0	0.0-49.0	0.0-76.0	0.0-114.0
% 1-2 y old, n (%)	75 (21.7)	32 (18.1)	17 (20.2)	13 (34.2)	13 (28.3)
% >2 y old, n (%)	55 (15.9)	24 (13.6)	11 (13.1)	13 (34.2)	7 (15.2)
Age at diagnosis	(n = 420)	(n = 189)	(n = 83)	(n = 81)	(n = 67)
Mean ± SD	18.9 ± 12.8	20.1 ± 12.1	17.9 ± 12.2	19.6 ± 11.8	15.5 ± 16.0
Median	16.0	18.0	14.9	17.9	12.0
Range	0.0-122.0	0.0-67.0	1.0-51.0	3.0-56.0	0.0-122.0
% 1-2 y old, n (%)	189 (45.0)	98 (51.9)	30 (36.1)	36 (44.4)	25 (37.3)
% >2 y old, n (%)	99 (23.6)	46 (24.3)	21 (25.3)	22 (27.2)	10 (14.9)
Age at referral for rehabilitation services	(n = 379)	(n = 148)	(n = 74)	(n = 95)	(n = 62)
Mean ± SD	13.4 ± 13.5	10.6 ± 12.1	10.9 ± 10.7	21.4 ± 12.8	11.0 ± 15.8
Median	10.0	7.0	7.5	18.0	7.0
Range	0.0-117.0	0.0-61.0	0.0-47.1	2.0-76.0	0.0-117.0
% 1-2 y old, n (%)	90 (23.7)	21 (14.2)	14 (18.9)	40 (42.1)	15 (24.2)
% >2 y old, n (%)	62 (16.4)	19 (12.8)	7 (9.5)	32 (33.7)	4 (6.5)
Age at initiation of rehabilitation	(n = 370)	(n = 144)	(n = 70)	(n = 85)	(n = 71)
Mean ± SD	15.9 ± 12.9	13.7 ± 11.7	13.0 ± 11.0	23.8 ± 10.8	13.8 ± 15.4
Median	12.0	10.0	9.5	22.0	10.0
Range	1.0-122.0	1.0-64.0	1.0-48.0	5.9-62.0	2.0-122.0
% 1-2 y old, n (%)	103 (27.8)	29 (20.1)	16 (22.9)	39 (45.9)	19 (26.8)
% >2 y old, n (%)	76 (20.5)	19 (13.2)	10 (14.3)	38 (44.7)	9 (12.7)

SD, standard deviation; *n*, sample size.

20.5 ± 12.3; median: 18.0) compared with those with GMFCS Level IV-V (n = 110; mean: 14.7 ± 13.1; median: 13.0; F[1, 409] = 17.18, $P < .0001$).

Type of CP. Children with diplegia (n = 83; mean: 24.1 ± 13.0; median: 21.0), hemiplegia (n = 167; mean: 18.9 ± 12.4; median: 15.0), and "other" types of CP (n = 56; mean: 19.1 ± 17.6; median: 15.5) were diagnosed later than children with triplegia/quadriplegia (n = 113; mean: 14.7 ± 8.5; median: 13.0; F[3, 415] = 9.12, $P < .0001$). Analysis of any association between parity, urban/rural, paternal education, gestational age, extended hospitalization after birth, prematurity, referral source for diagnosis, and region as potential predictors of age of diagnosis did not yield significant results.

Factors Influencing Age at Referral for Rehabilitation Services

Maternal Education. Children whose mothers completed a college/trade program (n = 91; mean: 19.3 ± 16.8; median:

Table III. Bivariate analysis results

Potential predictor variables	Age at referral for diagnostic assessment	Age at diagnosis	Age at referral for rehabilitation services	Age at rehabilitation initiation
Sociodemographic variables				
Maternal education	*	*	*	*
Paternal education	-	-	-	-
Sex	-	*	-	-
Urban/rural	-	-	-	-
Birth history				
Gestational age	-	-	-	-
Extended hospitalization after birth	*	-	*	*
Parity	-	-	-	-
Prematurity	-	-	-	-
CP profile				
Severity of motor dysfunction	*	*	*	*
Type of CP	*	*	*	*
Referral source for diagnosis				
Referral source for diagnosis	*	-	*	*
Region				
Site	*	-	*	*

*Significant at the $P < .05$ level.
CP, cerebral palsy.

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15.0) were referred later than children whose mothers completed university (n = 122; mean: 15.5 ± 10.4; median: 13.0) and high school or less (n = 150; mean: 18.9 ± 9.9; median: 17.0; F[2, 331] = 3.08, P = .0474).

Extended Hospitalization After Birth. Children who did not require admission at birth (n = 8) were referred at a mean age of 16.6 ± 11.8 (median: 13.0), significantly delayed compared with children who were initially admitted (n = 249) who were referred at a mean age of 11.2 ± 12.4 (median: 7.1; t[345] = 3.71, P = .0002).

Severity of Motor Dysfunction. Children with GMFCS Level I-III were referred later (n = 270; mean:15.0 ± 12.7; median: 12.0) compared with children with GMFCS Level IV-V (n = 100; mean: 9.8 ± 15.1; median: 6.0; F[1, 368] = 11.12, P = .0009).

Type of CP. Children with hemiplegia (n = 149; mean: 14.4 ± 12.5; median: 11.0), "other" types of CP (n = 52; mean: 15.2 ± 18.7; median: 10.0) and especially diplegia (n = 70; mean: 18.2 ± 15.5; median: 14.0), were referred later than children with triplegia/quadriplegia (n = 107; mean: 8.0 ± 7.5; median: 6.0; F[3, 374] = 9.86, P < .0001).

Referral Source. Children who were referred for diagnosis by a primary care practitioner (n = 145) were referred for rehabilitation at a mean age of 16.6 ± 14.9 (median: 13.0), whereas those referred for diagnosis by a medical specialist (n = 159) were referred to rehabilitation at a mean age of 10.5.1 ± 12.7 (median: 6.0); t(302) = 3.87, P = .0001.

Site. As detailed in Table II, the mean age of referral was greater in Toronto compared with the other 3 sites (F[3, 375] = 16.83, P < .0001). Analysis of any association between sex, parity, urban/rural, paternal education, gestational age, and prematurity as potential predictors of rehabilitation referral age did not yield significant results.

Factors Influencing Age at Rehabilitation Initiation

Maternal Education. Children whose mothers completed a college/trade program (n = 83; mean:18.9 ± 17.4; median: 13.0) initiated rehabilitation later than children whose mothers completed university (n = 106; mean: 13.2 ± 8.9; median: 11.0) and high school or less (n = 135; mean: 16.3 ± 12.4; median: 12.0); (F[2, 321] = 4.51, P = .0117).

Extended Hospitalization After Birth. Children who were not admitted for care (n = 100) initiated rehabilitation at a mean age of 17.9 ± 12.2 (median: 13.0), significantly delayed compared with children who were initially admitted (n = 238) who began at a mean age of 14.0 ± 11.0 (median: 10.0; t[336] = 2.88, P = .0043).

Severity of Motor Dysfunction. Children with GMFCS Level I-III initiated rehabilitation later (n = 266; mean:17.2 ± 12.2; median: 13.0) compared with those with GMFCS Level IV-V (n = 95; mean: 12.9 ± 14.3; median: 9.0; F[1, 359] = 7.99, P = .0050).

Type of CP. Children with diplegia (n = 62; mean: 18.8 ± 11.1; median: 18.0), hemiplegia (n = 151; mean:16.7 ± 12.8; median: 12.0), and "other" types of CP (n = 56; mean: 18.6 ± 18.7; median: 14.0) received services later than children with triplegia/quadriplegia (n = 99; mean:11.2 ± 8.0; median: 9.0; F[3, 364] = 6.71, P = .0002).

Referral Source. Children who were referred for diagnosis by a primary care practitioner (n = 142) initiated rehabilitation at a mean age of 18.5 ± 15.2 (median: 13.0), and those referred by a medical specialist (n = 151) started at a mean age of 12.6 ± 10.1 (median: 9.0; t[242.89] = 3.85, P = .0001).

Site. As detailed in Table II, the mean age of initiation was greater in Toronto compared with the other 3 sites (F[3, 366] = 15.59, P < .0001). Analysis of any association between sex, parity, urban/rural, paternal education, gestational age, and prematurity as potential predictors of initiation age did not yield significant results.

Multivariate Analysis

Table IV presents the results for the multiple linear regression models. The following predictors explained between 20% and 32% of the variance for the four age outcomes of interest: maternal education, severity of motor dysfunction, type of CP, extended hospitalization after birth, and region.

Discussion

This environmental scan represents a population-based empirical study on current referral practices of Canadian physicians with respect to the diagnostic assessment and rehabilitation of children suspected of having CP. This study advances the results of a pilot study conducted by the authors,[19] which documented delays in referral for diagnosis and rehabilitation for a subset of children with CP, and identified potential predictors that may contribute to these delays. The results of the current study highlight that there is wide variability in the age at which young children with CP are referred to specialists for diagnosis and intervention. Children referred for diagnosis from primary care, children with mild motor dysfunction (GMFCS I-III), children with hemiplegia and especially diplegia, children not requiring an extended hospitalization after birth, and children whose mothers did not have a university-level education were observed to have been referred much later. Overall, greater maternal education (ie, university-level) was associated with earlier referral and earlier diagnosis. Referral and rehabilitation may be early in a subset of mothers with lower education (eg, high school or less); however, this finding may be confounded by a greater likelihood for high-risk (eg premature) delivery and need for closer follow-up. Although it is

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Table IV. Multiple linear regression results

Predictor variables	β	*P* value
Age (mo) at referral for diagnostic assessment		
Sex (female vs male)	2.1	.126
Maternal education		.028
High school	3.5	
College	4.4	
University	Reference	
Extended hospitalization after birth (yes vs no)	−4.3	.006
Severity of motor dysfunction (ambulatory vs non-ambulatory)	4.0	.038
Type of CP		.001
Diplegia	3.3	
Hemiplegia	−1.0	
Triplegia/quadriplegia	−5.5	
Others	Reference	
Referral source (specialist vs primary care)	−4.6	.001
Region		<.0001
Calgary	−1.3	
Edmonton	−2.8	
Toronto	9.2	
Quebec	Reference	
Age (mo) at diagnosis		
Sex (female vs male)	2.1	.113
Maternal education		.020
High school	4.2	
College	2.9	
University	Reference	
Extended hospitalization after birth (yes vs no)	0.5	.714
Severity of motor dysfunction (ambulatory vs non-ambulatory)	7.5	.0001
Type of CP		.009
Diplegia	−0.1	
Hemiplegia	−4.7	
Triplegia/quadriplegia	−5.0	
Others	Reference	
Referral source (specialist vs primary care)	−2.2	.104
Region		.025
Calgary	0.9	
Edmonton	2.2	
Toronto	7.0	
Quebec	Reference	
Age at referral for rehabilitation		
Sex (female vs male)	1.4	.335
Maternal education		.054
High school	4.0	
College	3.1	
University	Reference	
Extended hospitalization after birth (yes vs no)	−4.1	.016
Severity of motor dysfunction (ambulatory vs non-ambulatory)	3.9	.055
Type of CP		.001
Diplegia	5.1	
Hemiplegia	0.1	
Triplegia/quadriplegia	−4.4	
Others	Reference	
Referral source (specialist vs primary care)	−1.5	.352
Region		<.0001
Calgary	−0.1	
Edmonton	−0.7	
Toronto	11.9	
Quebec	Reference	
Age at initiation of rehabilitation		
Sex (female vs male)	0.5	.735
Maternal education		.009
High school	4.6	
College	4.7	
University	Reference	
Extended hospitalization after birth (yes vs no)	−3.2	.043
Severity of motor dysfunction (ambulatory vs non-ambulatory)	5.7	.004
Type of CP		.046
Diplegia	0.8	
Hemiplegia	−2.3	
Triplegia/quadriplegia	−4.9	
Others	Reference	
Referral source (specialist vs primary care)	−2.4	.101
Region		<.0001
Calgary	−0.8	
Edmonton	0.0	
Toronto	10.0	
Quebec	Reference	

CP, cerebral palsy.

not surprising that children with more severe motor dysfunction (GMFCS IV-V) and triplegia/quadriplegia are detected and referred earlier, these results highlight the need for improved awareness of the early motor signs and attributes of milder CP subtypes (ie, hemiplegia, diplegia).

Early identification and intervention for developmental disabilities is considered best practice, as such can capitalize on a critical period of brain plasticity that enhances the potential effects of intervention.[12] Thus, delays in identification or referral for diagnostic assessment provide additional and unnecessary prolongations to rehabilitation interventions, the consequences of which can be detrimental to both the child and family. One possible factor contributing to the delayed referrals from primary care providers is that knowledge of the early clinical features associated with increased risk of CP may differ. A recent scoping review synthesizing the evidence on the early clinical attributes of CP suggested that there is a lack of accessible and user-friendly signs that can be used with confidence by primary care practitioners in the early detection of CP.[21]

In an effort to address this knowledge gap, a recent consensus study used nominal group techniques with Canadian content experts and knowledge users to develop expert-informed content regarding early motor attributes consistent with CP that should prompt physician referral for diagnostic assessment, as well as concurrent referral recommendations for other interventions.[22] The results were then validated through an online Delphi survey of international experts in CP management, which resulted in the following: 6 clinical features that should prompt referral for diagnosis, 2 "warning sign" features that warrant monitoring, and 5 referral recommendations to other healthcare professionals to occur simultaneously with referral for diagnosis.[23]

This study is not without limitations. Selection and information bias are inherent in retrospective chart review methodology (eg, incomplete documentation, missing charts, and difficulty interpreting information found in the documents).[24] To mitigate this limitation, we provided training and a standardized manualized method of interpreting and collecting the data. The individual at each site who collected the data periodically reviewed any challenges with the first author, to ensure consistent data collection strategies across sites.

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Another limitation is related to recruitment bias where for some sites (ie, Toronto, Montreal) access to the children's community hospital charts was more restricted, resulting in missing data. Furthermore, both in Montreal and Toronto there were several staffing changes that limited data collection. Thus, the sample profiles and subsequent data from those regions may not be truly representative of the current practices across types and severities of CP. Finally, 62.4% of our sample came from Alberta, and thus the generalizability of the results to the rest of Canada would require further validation.

Approximately one-half of children with CP do not have a complicated birth history necessitating close surveillance by neonatal follow-up programs and are followed in their community by primary care practitioners. The results of this environmental scan can be used to inform the creation of user-friendly educational knowledge translation tools (eg, pocket card, Web-based resource, poster in waiting room) to enhance early detection and referral strategies by primary care physicians and parents. The ultimate aim is to decrease the delays identified in this study and to optimize child and family functioning and health. Efforts to disseminate this knowledge are now underway, and these results have been included in the most recent revision of The Rourke Baby Record,[25] an evidence-based health supervision guide for primary healthcare practitioners of children in the first 5 years of life, which has considerable reach in the context of Canadian primary care surveillance. ■

We thank Kids Brain Health Network for their support of the CCPR; Christopher Saunders and Sasha Dyck for their invaluable collaboration as CCPR National Coordinators; the CCPR site coordinators for their assistance with data collection: Mary McNeil (Calgary, Alberta), Kathleen O'Grady (Edmonton, Alberta), Danielle Guimont and Souad Rhalmi (Montreal, Quebec), Catherine Diamante (Quebec City, Quebec), Lauren Switzer and Hillary Carpenter (Toronto, Ontario); and Xun Zhang for support with statistical analyses. We also thank the research assistants Annahita Ehsan, Deniz Keskinel, Hana MacDougall, Sossy Sahakian, and Anna Radzioch Guerin for their support with data collection and in the preparation on this manuscript.

Submitted for publication Jun 28, 2019; last revision received Aug 20, 2019; accepted Sep 13, 2019.

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Data Statement

Data sharing statement available at www.jpeds.com.

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Appendix

Additional members of the PROMPT Group

Stakeholders: Content-Experts, Knowledge-Users

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CHAPTER 8:
INTEGRATION OF MANUSCRIPTS 3 AND 4

8.1 Research Questions of Manuscripts 3 and 4

Manuscript 3:

"To what extent do sociodemographic and child characteristics, birth history, CP profile, and referral source for diagnosis influence the age at referral for diagnostic assessment and for rehabilitation services among Canadian children with CP?"

***Manuscript 4*:**

(i) Participants in consensus Group One were asked to identify and come to consensus on: "What early clinical signs or attributes of CP should prompt referral to a medical specialist for diagnosis?"

(ii) Participants in consensus Group Two were asked to achieve consensus on the following: "At the time children are being referred to a medical specialist for diagnosis, to which health professionals, other than physicians, should children suspected of having CP also be referred?"

8.2 Integration of manuscripts 3 and 4

In **Manuscript 3** we conducted a national environmental scan to gather population-based evidence on Canadian physician referral practices in order to describe current practices with respect to age at referral to pediatric medical specialists for initial diagnostic assessment of CP and to rehabilitation and other specialists for intervention, and to identify factors that are associated with delayed referral. This evidence was synthesized with the results of **Manuscripts 1 and 2**, and was used to inform the first of the studies in Phase 2 of this research project. Participants in the studies described in **Manuscripts 4** and **5** were first informed of the findings of Phase 1, and considered these sources of evidence prior to applying consensus methods. In **Manuscript 4** we

used two consensus groups applying nominal group processes (consensus methodology) to determine if there was agreement among Canadian content-experts and knowledge-users concerning the clinical features that should prompt referral by a primary care practitioner to a medical specialist for diagnostic assessment of CP, and referral recommendations to other health professionals to occur simultaneously with referral for diagnostic assessment.

CHAPTER 9:

MANUSCRIPT 4

Use of consensus methods to determine the early clinical signs of cerebral palsy

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Financial Disclosure: AM received funding from Canadian Institutes of Health Research for the study "*PROMPT* Identification of Cerebral Palsy-*P*rimary care *R*eferral *O*f *M*otor-impaired children: *P*hysician *T*ools" (MOP-133402); ZB received financial support from the Richard and Edith Strauss Canada Foundation as a Musculoskeletal Doctoral Fellow, as well as from the Hoppenheim Fund/Montreal Children Hospital Foundation.

This manuscript was published in Paediatrics & Child Health

Boychuck, Z., Andersen, J., Bussières, A., Fehlings, D., Kirton, A., Li, P., Oskoui, M., Rodriguez, C., Shevell, M., Snider, L., Majnemer, A., & the PROMPT group. (2019). Use of consensus methods to determine the early clinical signs of cerebral palsy. *Paediatrics & Child Health*. DOI: 10.1093/pch/pxz061

Accepted for publication: February 4, 2019
Epub ahead of print: May 8, 2019
In Press

Paediatrics & Child Health, 2019, 1–8
doi: 10.1093/pch/pxz061
Original Article

Original Article

Use of consensus methods to determine the early clinical signs of cerebral palsy

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Abstract

Objectives: To develop expert-informed content regarding the early motor attributes of cerebral palsy (CP) that should prompt physician referral for diagnostic assessment of CP, as well as concurrent referral recommendations. This content will be used in the creation of knowledge translation (KT) tools for primary care practitioners and parents.

Methods: Two nominal group processes were conducted with relevant stakeholders, representing Canadian '*content experts*' and '*knowledge-users*', using an integrated KT approach.

Results: Six attributes were identified that should prompt referral for diagnosis. If the child demonstrates: Early handedness <12 months; stiffness or tightness in the legs between 6 and 12 months; persistent fisting of the hands >4 months; persistent head-lag >4 months; inability to sit without support >9 months; any asymmetry in posture or movement. Five referral recommendations were agreed upon: Motor intervention specialist (physical therapy and/or occupational therapy) for ALL; speech-language pathology IF there is a communication delay; audiology IF there is parental or healthcare professional concern regarding a communication delay; functional vision specialist (e.g., optometrist or occupational therapist) IF there is a vision concern (e.g., not fixating, following, or tracking); feeding specialist (e.g., occupational therapist, speech-language pathologist) IF there are feeding difficulties (e.g., poor sucking, poor swallowing, choking, and/or not gaining weight).

Conclusion: Rigorous consensus methods provided the initial evidence necessary to inform the content of tools to assist primary care providers in the early detection of CP. Results will be validated through a Delphi process with international experts, and user-friendly formats of this KT tool will be developed collaboratively with stakeholders.

Keywords: *Cerebral palsy; Child; Diagnosis; Primary health care; Referral and consultation; Rehabilitation.*

Cerebral palsy (CP) is the most common physical disability of childhood (1), with an estimated prevalence of 2.3/1,000 births (2), with lifelong consequences, affecting autonomy, health, and participation. For children suspected of having CP, prompt referral to medical specialists for diagnostic evaluation and to rehabilitation professionals for assessment and intervention is

Received: November 2, 2018; Accepted: February 4, 2019

widely accepted as best practice (3,4). Paediatric societies and governmental policies widely endorse early identification of developmental disabilities by primary care practitioners (PCPs), such as paediatricians and family medicine physicians (5,6). Early interventions incorporating child-initiated movement, parental education, and environment modification have the potential to optimize long-term motor outcomes for the child (7). Early psychosocial support for families is also important: Parents of children with CP have expressed dissatisfaction associated with delays in the diagnostic process, with negative repercussions to adaptive coping and health (8,9), while better long-term adaptation to having a child with a disability is associated with satisfaction with this diagnostic process (10,11).

Limited population-based evidence exists on the age at which children with CP are referred for diagnosis and intervention. Hubermann et al. (12) demonstrated that: PCPs refer later than medical specialists; subsets of children with CP experience delayed referral; and referral is delayed for rehabilitation. A Canada-wide environmental scan of referral practices is underway and preliminary results suggest similar patterns (13). One study on children with hemiplegia found that parental concern was typically at 4 to 6 months, whereas final diagnosis was only 12 to 18 months (14).

The early motor signs of CP are recognizable to medical and rehabilitation specialists who have advanced training in child development and neurologic status. PCPs, however, may lack the overt awareness of these early clinical signs, as well as the roles of rehabilitation professionals in early intervention. PCPs may use developmental screening tools that focus on the child meeting milestones (15). However, these tools are not sensitive enough to capture potentially subtle early attributes of CP such as aberrant quality of movement. For example, these tools can miss children with CP who demonstrate mild motor impairment (e.g., Gross Motor Function Classification System Level I and II [16]). These children represent more than half of the children with CP (17). A recent scoping review on the early clinical signs associated with CP found that evidence is sparse, with little discrimination between motor delay and CP (18). Furthermore, most studies focused exclusively on high-risk groups such as premature infants.

The primary aim of this study was to develop expert-informed content on early motor attributes of CP to be used in the creation of widely disseminated knowledge translation tools targeting PCPs and parents. For children suspected of having CP, prompt referral by PCPs to early-intervention services is recommended (19). A survey of American paediatricians revealed wide variability in knowledge and practices related to the identification of motor delays in children, including uncertainty with respect to how to diagnose and manage children with motor delays (20). Health professionals have traditionally taken a 'wait and see' approach to providing a diagnosis of CP, which often delays the child from receiving targeted early-intervention services during a critical period of brain plasticity, and can be potentially harmful to children and their families (3). Of concern, delays in age at referral to rehabilitation services have been documented for children with CP (12). Thus, a secondary aim of this study was to develop expert-informed content on referral recommendations for physicians to use simultaneously as they refer children suspected of having CP for diagnostic investigation.

It is considered best practice to use evidence-based research to inform clinical decisions (21), however, its absence necessitates drawing upon the expert opinion and experience of clinicians and other experts (22) via consensus methods where the steps in the process have been made explicit and can be repeated (23). They represent an improvement over informal consensus methods because the decision-making process is transparent, accountable and democratic (24). The nominal group technique (NGT) (25) has many advantages as a consensus method (26–28). This includes facilitated face-to-face meetings between participants and a structure that enables the active involvement of participants in each phase of the process of consensus-building.

RESEARCH QUESTIONS

i. Participants in consensus Group 1 were asked to identify and come to consensus on "*What early clinical signs or attributes of CP should prompt referral to a medical specialist for diagnosis*?"
ii. Participants in consensus Group 2 were asked to achieve consensus on the following: "*At the time children are being referred to a medical specialist for diagnosis, to which health professionals, other than physicians, should children suspected of having CP also be referred?*"

METHODS

Design

Two consensus groups were conducted using a NGT (25).

Participants

Twenty (n=20) national (Canadian) and local (Montreal, Quebec) stakeholders were invited to participate in the consensus process. These invitees represented '*content experts*' (child neurologists, developmental paediatricians, rehabilitation specialists) and '*knowledge-users*' (community paediatricians, family physicians, parents of children diagnosed with CP) in the primary care context of an early diagnosis of CP. Most of the invited participants were co-investigators on this project, though additional participants were identified through snowball sampling (personal communication with co-investigators).

Two invitees declined to participate citing unavailability. Consent for participation was obtained from all remaining 18 participants. This provided two purposive samples for the two consensus groups.

Consensus Group 1 consisted of 12 (n=12) participants: PCPs (paediatricians=2), medical specialists (paediatric neurologists=3; developmental paediatricians=2); parents of young children with CP (mother=1; father=1); researcher (paediatrician with expertise in primary care health services research=1); rehabilitation clinicians with expertise in early intervention (occupational therapist=1; physical therapist=1).

Consensus Group 2 consisted of thirteen (n=13) participants, with some overlap in participants from Group 1 (n=7): Medical specialists (paediatric neurologists=2; developmental paediatricians=2); parents of young children with CP (mothers=2); researchers (as above=1; occupational therapist with expertise in early diagnosis of CP=1); rehabilitation clinicians (occupational therapists=2; physical therapist=1; social worker=1; speech-language pathologist=1).

Data collection

Pre-NGT exercise

In advance of the face-to-face meeting for both groups, participants were asked to complete a brief online questionnaire, which primarily consisted of demographic information, and one question primed participants to begin thinking about the topic to be discussed. Participants in Group 1 were asked:

> *"In your experience, what are 1–2 early motor signs, postures and/or behaviours that are easily observable in the clinic or at home, that should be considered as early markers of CP which should prompt referral to a medical specialist for diagnosis?"*

Participants in Group 2 were asked:

> "*In your experience, to which medical specialists/health professionals, other than physicians, should children suspected of having CP be referred?"*

Procedures

Both group sessions were co-led in English by three facilitators familiar with the nominal group technique (AB, AM, ZB). Each group was provided with an overview of the most recent evidence in the field related to their group's question to help inform their decision making. Group 1 was provided with the preliminary results of two scoping reviews on early clinical attributes of CP (18) and age at referral for diagnosis of CP and to rehabilitation specialists for intervention (29). Participants also received the preliminary results of an environmental scan of referral practices of Canadian physicians (13) highlighting particular subgroups of children more likely to be referred later (i.e., children with hemiplegia or spastic diplegia; mild motor impairments), and a recent chart review on age at referral for diagnosis of CP and for rehabilitation services (12). Group 2 was similarly provided with the results of the scoping review on age at referral for diagnosis of CP (29), the preliminary results of the environmental scan (13), and relevant literature on rehabilitation interventions for children with CP (30).

Following this overview of existing evidence, the group facilitators ensured that the steps detailed in Table 1 were followed in the NGT process (31,32) in order to come to consensus. Items recorded during the *silent, private idea generation* were shared out loud with the group through a *round-robin approach*. In the *serial discussion of ideas* that followed regarding the merit of each item, clarifications and definitions were sought. Similar items were grouped together to facilitate item prioritization; optimal wording that represented each grouping was then determined collectively (e.g., 'asymmetry' as a clinical attribute to prompt referral), and items not specific enough for CP (e.g., 'paradoxical breathing patterns', 'clumsy child') were eliminated. This discussion resulted in the reduction and refinement of a final list of twelve items, which participants rated individually on paper. The results were tabulated by the facilitators and projected on a screen. A thematic analysis of the responses was then conducted. All participants in Group 1 and Group 2 completed anonymous feedback forms.

RESULTS

Identifying clinical attributes to prompt referral: Item generation

In total, 35 items were recorded during the *silent, private idea generation* were shared with the group (Supplementary Appendix A). In the *serial discussion of ideas*, it was suggested that an age or temporal qualifier should be attached to the attributes, as the timeframe in development at which they appear or persist was deemed relevant. Suggested age cut-offs were then proposed and added. Members of the group also felt that some attributes did not achieve the necessary high level of agreement but were still important to include. It was therefore proposed that although they should not necessarily be used to prompt referral for diagnosis immediately, they should be considered as a 'warning sign' to monitor the child. Through *discussion and consensus*, the final list of attributes were agreed upon, in which the observation of any single one should be used to prompt referral for diagnosis of CP (Table 2). The group was satisfied with the final list; thus, there was no need for a subsequent consensus process. The attributes agreed upon were: If the child demonstrates

- *Early handedness* before 12 months of age
- *Stiffness or tightness in the legs* between 6 and 12 months of age (e.g., unable to bring their toes to mouth during diapering)
- *Persistent fisting* of the hands beyond 4 months of age
- A *persistent head-lag* beyond 4 months of age

Table 1. Steps of the consensus process using nominal group technique

Silent, private idea generation: The facilitators guided the participants through a silent, individual idea generation phase. Participants were each provided with a paper with the research question ("*What are the early signs of cerebral palsy (CP) that should prompt referral for diagnosis?*"), and were asked to independently list every potential answer they were able to think of.

Round-robin recording of ideas: Participants took turns, proceeding in a clockwise manner around the table, providing one new response at a time from their individual list, which was recorded onto a master list, on a large easel pad flipchart visible to all participants. Duplicates were excluded, and the sharing continued until every unique response that had been written during the previous step was included on the master list.

Serial discussion of ideas: Led by the facilitators, the participants were able to ask for clarification of items on the master list, and a facilitated discussion enabled the group to cluster similar responses/items to shorten the master list for the participants. Some items were determined by the group not to be specific enough and were eliminated. When there was disagreement on an appropriate cut-off age, multiple versions were included to be considered.

Preliminary voting: Participants were provided with a listing of all potential items on a piece of paper and were asked to privately rank items as follows: (i) '*Exclude*', not sensitive enough for CP detection, (ii) '*Maybe*' - could be important, should be considered, or (iii) '*Include*' -high likelihood for positive diagnosis. The results were then submitted to the facilitators and tabulated during a short break.

Discussion and consensus: The results were shared with the participants on a screen. It was decided *a priori* that a high level of agreement would be used to determine which attributes and referral recommendations were retained for the final list. Considering the small sizes of the groups (Group 1, n=12; Group 2, n=13), consensus was defined as ≥70% agreement between participants for items in Group 1, and ≥85% agreement between participants for items in Group 2 (26,28). If consensus was achieved for particular items (exclude or include), no further discussion was required. If consensus was not achieved, the discussion and voting were to be repeated until consensus is achieved, or it was determined that a subsequent group is required.

Participant feedback: Participants in both groups were asked to submit written feedback on their experience in the consensus process. They were instructed to complete the form individually. The feedback forms were anonymous, with a colour-coded sticker indicating which stakeholder group they represented ('content expert', 'knowledge-user'). They were asked to respond to the following open-ended questions:

(1) How was your overall experience as a participant in this consensus group?
(2) To what extent do you feel you were engaged in the process of the group?
(3) To what extent do you feel your contributions were considered/your ideas were heard?
(4) What were the highlights for you of participating in this consensus group?
(5) What did you find challenging in participating in this consensus group?

- *An inability to sit without support* beyond 9 months of age
- *Any asymmetry* in posture or movement

The two '*warning signs*' agreed upon that should prompt closer monitoring and surveillance over time rather than immediate referral included: If the child demonstrates

- A *persistent startle (Moro) reflex* beyond 6 months of age.
- *Consistent toe walking or asymmetric-walking* beyond 12 months of age.

Recommendations for referral to other health professionals: Item generation

The second consensus group followed the same nominal group process as the first group. Following an initial *silent, private idea generation* phase, all items (n=18; Supplementary Appendix B) were shared out loud with the group in a *round-robin approach*. There was much discussion about *where* the child should be referred (e.g., type of institution or program). It was agreed that for purposes of generalizability, specifics regarding where to refer would not be appropriate as each community has a different locally specific service delivery model. Consensus was meant to focus on what specific type of nonphysician health professionals to simultaneously refer the child, in addition to a medical specialist for diagnosis. Similar items were clustered to facilitate item prioritization. This discussion resulted in the reduction and refinement of a final list of items (n=9) which participants then rated individually on paper in a *preliminary voting process* (Table 3). The group was satisfied with the final list; thus, there was no need for a subsequent consensus process. The referral recommendations agreed upon included the following:

- *Motor intervention specialist* (physical therapy and/or occupational therapy) **for ALL**
- *Speech-language pathology* **IF** there is a communication delay.
- *Audiology* **IF** there is parental concern and/or a communication delay.

Table 2. Preliminary ratings of clinical attributes for individual voting

Clinical attributes initially considered by consensus Group 1	Total rating (/36)* (N=12 raters)
1. Early handedness < 12 months	36
2. Early handedness < 15 months	28
3. Early handedness < 18 months	23
4. Stiffness or tightness in the legs: 6–12 months (unable to bring toes to mouth during diapering)	33
5. Persistent fisting > 2 months	25
6. Persistent fisting > 4 months	32
7. Persistent startle reflex > 4 months	20
8. Persistent startle reflex > 6 months	29
9. Persistent head lag > 4 months	33
10. Delayed sitting: refer for diagnosis if child is not sitting unsupported > 9 months	35
11. Toe walking: consistent toe walking or asymmetric > 12 months	26
12. Any asymmetry	32

*Scoring: A score of 36 indicates that all participants rated the attribute as 3, thus complete agreement by all to include the item.
1= 'Exclude', not sensitive enough, 2='Maybe', could be important, should be considered, 3= 'Include', high likelihood for positive diagnosis.

Table 3. Preliminary ratings for rehabilitation referral recommendations

Rehabilitation referral recommendations initially considered by consensus Group 2	Total rating (/39)* (N=13 raters)
1. *Motor intervention specialist* (physical therapy and/or occupational therapy) for ALL	34
2. Occupational therapy AND physical therapy ALL	27
3. Case manager/coordinator (e.g., social worker, nurse, infant developmental specialist)	29
4. Speech-language pathologist IF communications delay	37
5. *Audiology* IF no newborn screening	30
6. *Audiology* IF communications delay	35
7. *Audiology* IF parental concern	36
8. *Functional vision specialist* IF vision concern (e.g., not fixating, following, or tracking)	35
9. *Feeding specialist* (e.g., occupational therapist, speech-language pathologist, local expertise) IF there are feeding difficulties (e.g., poor sucking, poor swallowing, choking, and/or not gaining weight)	36

*Scoring: A score of 39 indicates that all participants rated the attribute as 3, thus complete agreement by all to include the item.
1= 'Exclude', not sensitive enough, 2='Maybe', could be important, should be considered, 3= 'Include', high likelihood for positive diagnosis.

- *Functional vision specialist* (e.g., optometrist or occupational therapist) **IF** there is a vision concern (e.g., not fixating, following, or tracking).
- *Feeding specialist* (e.g., occupational therapist, speech-language pathologist, local expertise) **IF** there are feeding difficulties (e.g., poor sucking, poor swallowing, choking, and/or not gaining weight).

Participant feedback

Participants in both groups uniformly identified their overall experience with the consensus process as being positive; reported feeling very engaged in the group consensus process; and expressed feeling that their ideas and contributions were carefully considered. Representative examples of comments appear in Supplementary Appendix C.

DISCUSSION

Delays in referral of children to medical specialists for the diagnosis of CP have been documented, with concomitant delayed referral to rehabilitation services (12,29). This ultimately leads to delayed intervention (13). We conducted two consensus groups to inform the content of a knowledge translation tool

(e.g., pocketcard, poster, website) being developed for PCPs and parents for wide dissemination. The consensus groups identified *six early clinical attributes* that should prompt referral for diagnostic evaluation of CP, and determined *five referral recommendations* to rehabilitation and other health professionals.

Much of the current research on early identification of CP has focused on children who are at 'high-risk' of developing CP. A recent systematic review reported that little has actually been published concerning clinical manifestations of CP if there are no discernable risks in the newborn period (4). Since more than half of the children with CP are born at term and many have uneventful birth histories (33), there is thus a substantial subgroup of children (i.e., 'lower-risk') who may not qualify for neonatal follow-up and are thus followed in the community by PCPs. Prematurity is an important risk factor for CP, with approximately 40 to 45% of children with CP born <37 weeks gestational age (17,34). However, neonatal follow-up programs typically only routinely follow those with extreme prematurity (e.g., ≤28 weeks), and therefore only a subset of these children are closely monitored in these programs. Indeed, 44.2% (408/923) of the children in the Canadian CP Registry who are born premature (<37 weeks) are ≤28 weeks, and 50.4% (465/923) are ≤29 weeks (S. Dyck, personal communication), so depending on neonatal follow-up inclusion criteria, about half or more are not followed. Recently published guidelines provide a detection pathway algorithm that includes recommendations to apply clinical neurological examinations (Hammersmith Infant Neurological Examination), magnetic resonance imaging, and motor tests (Prechtl Qualitative Assessment of General Movements, standardized motor assessments) in order to diagnose CP, and the evidence is strongest in terms of its predictive accuracy for 'high-risk' newborns (4). Indeed, this approach would not be cost-effective or feasible in the context of primary care. These guidelines were developed by an international consensus group, but to our knowledge, without the use of a formal consensus methodology (e.g., nominal group process, Delphi survey), and without testing in the community primary care context. Our consensus attributes focus specifically on infants who are not monitored closely as part of neonatal follow-up programs and do not necessarily have 'high-risk' profiles. Therefore, a detection strategy designed for the primary care context represents a critical need to enable early detection for a significant proportion of children eventually diagnosed with CP.

An important aspect for researchers to consider is involving key stakeholders (e.g., PCPs, parents of children with CP) as 'experts' with content knowledge and/or lived-experience (35). It is essential to select consensus methods which will provide the structure and opportunity for all panel members to participate equally, and to ensure that all voices are heard. Based on the feedback we received from both groups (Supplementary Appendix C), participants were able to express their opinions freely and the different perspectives were appreciated by all. This is in line with patient-oriented research strategies (www.pcori.org; cihr-irsc.gc.ca/e/41204.html; www.invo.org.uk).

There are strengths and limitations to use of consensus methodology. The main *strengths* of the nominal group technique include: (i) face-to-face meeting promotes interaction between experts; (ii) anonymous voting reduces outside bias and solicits true expert opinion; (iii) the design prevents any one member from dominating; (iv) ensures equal contribution by all; and (v) a time-efficient process. A major *limitation* is that face-to-face meetings may influence the contribution of participants who perceive that their perspectives are 'against-the-flow' and may not answer as candidly as if it were totally anonymous. It requires an experienced moderator to keep the process on-track and to prevent any one member from dominating. Additionally, these consensus groups included only national (Canadian) and local (Montreal, Quebec) content experts and knowledge-users, which may minimize the acceptability and generalizability of the results to other countries. A third potential limitation is that we did not involve stakeholders from all potential healthcare professions (e.g., audiologists, optometrists, nutritionists, educators, psychologists) in the consensus process.

Although the attributes were developed specifically for the detection of CP, we acknowledge that indeed these signs could mimic other neuromotor disorders. These would include for example, spinal muscular atrophy, congenital myopathy, Duchenne muscular dystrophy, and Charcot-Marie-Tooth disease. Nevertheless, for infants with these attributes, referral to a medical specialist is still warranted for prompt diagnosis.

Hutchings et al. (36) recommend a 'hybrid approach' to consensus method use in healthcare research. As an example, the pairing of nominal group technique and the Delphi method is complementary methodologically, since the former provides greater understanding of potential disagreement, while the latter provides greater reliability. Thus, the results of this study will be validated through a Delphi survey of international experts in CP, before finalizing the content for the knowledge translation tools for PCPs.

CONCLUSION

Using consensus methods, six attributes were identified that should prompt referral for diagnostic evaluation of CP, and five referral recommendations were agreed upon that should occur concurrently. Through this effort, it is anticipated that the present delays in referral of 'low-risk' but affected children for diagnosis and intervention will be shortened. The potential impacts are threefold. PCPs will have the knowledge and capability to detect attributes associated with CP early,

prompting simultaneous timely referral to medical and rehabilitation specialists. Rehabilitation specialists will be able to initiate therapeutic interventions much earlier at a critical period of brain development. Parents will be more rapidly informed and will benefit from early access to resources and family supports.

SUPPLEMENTARY DATA

Supplementary data are available at *Paediatrics & Child Health* Online.

ACKNOWLEDGEMENTS

The authors wish to express their sincere appreciation to the national and local content experts and knowledge-users who volunteered their time and contributed their invaluable expertise. The authors also wish to express their gratitude to our research assistants Annahita Ehsan, Anna Radzioch Guerin, and Sossy Sahakian for their support with the organization and administration of the consensus groups.

This consensus study was completed as part of a larger integrated knowledge translation study involved varied *content experts* and *knowledge-users*, and thus has 21 co-investigators (The PROMPT Group) who have been involved in the conceptualization and implementation of various phases of the project as appropriate.

The PROMPT Group

Stakeholders: Content experts, knowledge-users

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Funding Information: This study received funding from Canadian Institutes of Health Research for the study "*PROMPT* Identification of Cerebral Palsy-*P*rimary care *R*eferral *O*f *M*otor-impaired children: *P*hysician *T*ools" (MOP-133402); ZB received financial support from the Richard and Edith Strauss Canada Foundation as a Musculoskeletal Doctoral Fellow, as well as from the Hoppenheim Fund/Montreal Children Hospital Foundation and the 2017 Judith Kornbluth Gelfand Fellowship in Pediatric Rehabilitation from McGill School of Physical and Occupational Therapy Graduate Committee, during the conduct of the study.

Potential Conflicts of Interest: JA reports grants from Canadian Institutes of Health Research, Women and Children's Health Research Institute, and Glenrose Rehabilitation Hospital Foundation, during the conduct of the study. PL reports grants from the Canadian Institutes of Health Research and institutional support from Fonds de Recherche Santé - Québec during the conduct of the study. MO reports grants from the Canadian Institutes of Health Research during the conduct of the study as well as grants from the Cerebral Palsy Alliance Foundation, Kids Brain Health Network, and CIHR outside the submitted work. All authors have submitted the ICMJE Form for Disclosure of Potential Conflicts of Interest. Conflicts that the editors consider relevant to the content of the manuscript have been disclosed.

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Appendix A

All items suggested by participants in Group One as potential attributes

to prompt early referral for diagnosis of CP.

1. Stiffness in the limbs, noticed when handling the baby when bathing, dressing, baby's in supine, or held upright, when the upper and lower extremities move stiffly
2. Spastic patterns, fisting or maintaining arms/legs close to body
3. Poor suck
4. Weak or high pitched cry
5. Persistent primitive reflexes
6. Early handedness, asymmetry in terms of hand use or fine motor control; always rolling to one side
7. Cortical thumbs
8. Inability to reach and grasp
9. Abnormal or asymmetrical posture, stiffness
10. Motor milestone delay (rolling > 6 mo, sitting > 9 mo, crawling > 12mo, walking > 18mo)
11. "Early" attainment of milestones
12. Asymmetric pivoting, rolling to one side only, pull + stand from one side
13. Clonus ++
14. Decreased head control with difficulty keeping head midline
15. Low trunk tone (robustness or pliability of the chest)
16. Horizontal arm movements
17. Toe walking
18. Relates to age
19. Delayed secondary reflexes
20. Restricted repertoire of movement
21. In toeing (especially symmetrical)
22. Lack of reciprocal crawling
23. No babbling (at age)
24. Abnormal tendon reflexes
25. Focal hemi-body seizures (+/- fever)
26. Scissoring
27. Limb smaller or less muscular than other
28. Pull to stand with upper body, not lower (not push up lower extremity)
29. Dissociation of limbs, move in unison
30. Head lag, low tone
31. Jerky kicking rather than smooth kicking.
32. Paradoxical breathing patterns
33. Shallow cry
34. Trunk hyper extension
35. Clumsy child

Appendix B

All items suggested by participants in Group Two as potential referral recommendations to rehabilitation and other health professionals to co-occur with referral for diagnosis of CP.

Types of health professionals to refer to	*Conditions (any of the following)*
1. Physical Therapy	(a) for all children
	(b) if not imitating specific gestures like clapping, pointing, or if parent has concerns
	(c) and OT for all
2. Social Work	(a) if a diagnosis of CP is confirmed
3. Occupational Therapy	(a) for all children
	(b) and PT for all
	(c) for saliva management
4. Speech-Language Pathology	(a) for all children
	(b) if the child demonstrates communication difficulties
	(c) if the child demonstrates feeding and swallowing difficulties
	(d) for saliva management
5. Nutritionist	(a) if the child has difficulty gaining weight
6. Local motor program specialist (e.g. occupational therapist OR physical therapist)	(a) for all children
7. Audiologist	(a) for all children
	(b) if there are concerns about hearing or language development
8. Nursing	(a) for all children
9. Educators / early intervention specialists	(a) if the child has cognitive developmental abilities that are compromised
10. Optometry/vision worker	(a) if the child demonstrates fine motor, vision, communication and/or social skills difficulties
11. Language simulation program (early intervention specialists)	(a) if there is a waitlist for speech-language pathology
12. Psychology	(a) if the child is school age and/or demonstrates significant cognitive/behavioural difficulties

Appendix C

Participant feedback on the consensus group process

For Group One, which focused on the identifying the clinical attributes of CP, all participants described their overall experience as being that of a positive one.

> "*Very positive, all group members participated and the discussions resulted in consensus*." (content expert)
>
> "*Excellent. Great discussion & productive."* (knowledge-user)

The 'content experts' expressed appreciation for the unique perspective the parent participants, representing future 'knowledge-users', brought to the discussion.

> "*Great overall experience. Wonderful to hear all points of view. Parents input very valuable.*"

The 'knowledge user' parents expressed a similar sentiment.

> "*My overall experience was very good. I'm very happy I got to hear from all the experts and their point of view.*"

All participants reported feeling actively engaged in the group consensus process:

> "*Very much so. Parent involvement / opinions were welcomed*." (knowledge-user)
>
> "*Overall, the process was very engaging. Moderator was sure to engage everyone to the extent to which they were comfortable*." (content expert)

All participants reported feeling that their contributions were acknowledged and considered equally.

> *"I think that the group was very open to everyone's ideas and all input was acknowledged."(*content expert)
>
> *"Yes, very much. As a parent of a CP child, I think that getting together with other parents and doctors and experts should be done more often."* (knowledge-user)
>
> *"I felt that my contributions were well heard & led to further discussions."* *(*content expert)

With respect to the highlights identified by the participants, a recurring theme was a mutual appreciation of the participation of both professionals and parents as partners in the discussion.

> *"Hearing different perspectives, especially from primary care docs and parents."* (content expert)

> *"Being able to confer as an equal with professionals."* (knowledge-user)

Some of the challenges identified by the participants were primarily related to logistics and the environment in which the group was held.

> *"Location hard to get to by public transit. I took Uber."* (content expert)

> *"Honking of truck."* (knowledge-user)

For Group Two, which focused on referral to rehabilitation and other health professionals, all participants described their overall experience as being that of a positive one.

> *"Excellent."* (knowledge-user)

> *"Very interesting and positive.*" (content expert)

All participants reported feeling actively engaged in the group process.

> *"Everyone was included.*" (content expert)

> "*Very engaged – good opportunities for discussion."* (knowledge-user)

All participants indicated that they felt as though their participation and contributions were acknowledged and considered equally.

> *"I think that I was heard and taken into consideration."* (knowledge-user)

> *"I felt that everyone's opinions / ideas were heard & respected."* (content expert)

Of the highlights identified by the participants, a recurring theme was a mutual appreciation of the participation of both professionals and parents as partners in the discussion.

> "*Hearing different stakeholder opinions."* (content expert)

"Listening to all the different opinions and how they relate to my experience." (knowledge-user)

The challenges identified by the participants were primarily related to the environment in which the group was held.

*"Heat and hearing over fans." (*content expert)

"The acoustics due to the fans were not great." (knowledge-user)

CHAPTER 10:

INTEGRATION OF MANUSCRIPTS 4 AND 5

10.1 Research Questions of Manuscripts 4 and 5

Manuscript 4:

(i) Participants in consensus Group One were asked to identify and come to consensus on: "What early clinical signs or attributes of CP should prompt referral to a medical specialist for diagnosis?"

(ii) Participants in consensus Group Two were asked to achieve consensus on the following: "At the time children are being referred to a medical specialist for diagnosis, to which health professionals, other than physicians, should children suspected of having CP also be referred?"

Manuscript 5:

(i) To what extent is there international agreement regarding the early motor signs (clinical attributes) of CP that were identified that should prompt early referral for diagnostic assessment?, and

(ii) To what extent is there agreement concerning the referral recommendations to health professionals for children suspected of having CP, to be referred to at the same time as they are referred for a medical specialist for diagnostic assessment?

10.2 Integration of Manuscripts 4 and 5

In **Manuscript 4** we used two consensus groups applying nominal group techniques (consensus methodology) to demonstrate agreement among Canadian content-experts and knowledge-users on six clinical features that should prompt referral from a primary care practitioner to a medical specialist for diagnostic assessment of CP; two warning signs that should

prompt close monitoring rather than immediate referral to a medical specialist for diagnostic assessment, and five referral recommendations to health professionals to occur simultaneously with referral for diagnostic assessment. Since these results were obtained using only national (Canadian) and local (Montreal, Quebec) experts and knowledge-users, the goal of the **Manuscript 5** was to validate these results using a panel of international experts in early identification and early intervention for children with CP to ensure they are appropriate for the primary care setting and generalizable beyond the Canadian context.

CHAPTER 11:

MANUSCRIPT 5

International expert recommendations of clinical features to prompt referral for diagnostic assessment of cerebral palsy.

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Word Count: 3, 375

This manuscript was published in *Developmental Medicine & Child Neurology*.
Boychuck, Z., Andersen, J., Bussières, A., Fehlings, D., Kirton, A., Li, P., Oskoui, M., Rodriguez, C., Shevell, M., Snider, L., Majnemer, A., & the PROMPT group. (2019). International expert recommendations of clinical features to prompt referral for diagnostic assessment of cerebral palsy. *Developmental Medicine & Child Neurology*. DOI: 10.1111/dmcn.14252
Accepted for publication: March 13, 2019.
Epub ahead of print: April 25, 2019.
Published in Issue: December 2, 2019

International expert recommendations of clinical features to prompt referral for diagnostic assessment of cerebral palsy

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[†]**Members of the PROMPT group are listed in Appendix S1 (online supporting information).**
This article is commented on by Msall on page 13 of this issue.

PUBLICATION DATA
Accepted for publication 13th March 2019.
Published online 25th April 2019.

AIM To establish international expert recommendations on clinical features to prompt referral for diagnostic assessment of cerebral palsy (CP).
METHOD An online Delphi survey was conducted with international experts in early identification and intervention for children with CP, to validate the results obtained in two previous consensus groups with Canadian content experts and knowledge users. We sent two rounds of questionnaires by e-mail. Participants rated their agreement using a 4-point Likert scale, along with optional open-ended questions for additional feedback. Additionally, a panel of experts and knowledge-users reviewed the results of each round and determined the content of subsequent surveys.
RESULTS Overall, there was high-level of agreement on: (1) six clinical features that should prompt referral for diagnosis; (2) two 'warning sign' features that warrant monitoring rather than immediate referral for diagnosis; and (3) five referral recommendations to other healthcare professionals to occur simultaneously with referral for diagnosis.
INTERPRETATION There was high agreement among international experts, suggesting that the features and referral recommendations proposed for primary care physicians for early detection of CP were broadly generalizable. These results will inform the content of educational tools to improve the early detection of CP in the primary care context.

Cerebral palsy (CP), a childhood-onset physical disability, has an estimated prevalence of 2.3 out of 1000 live births.[1,2] Early detection and subsequent simultaneous referral for diagnosis and medical management and for rehabilitation intervention is widely accepted as best practice.[3,4] More than half of children with CP are born at term, and do not necessarily have high-risk perinatal histories. Therefore, instead of receiving close follow-up monitoring (e.g. neonatal follow-up clinics, neurologists, developmental pediatricians), they are followed by a primary care provider (e.g. pediatrician, family physician) in their community for well-child healthcare. Furthermore, those born preterm (<37wks gestational age) and in the neonatal intensive care unit may not meet criteria for neonatal follow-up (e.g. <29wks gestational age) with close surveillance. Thus, primary care physicians are well positioned to contribute to the early detection of CP, which consequently should lead to referral to a medical specialist for early diagnosis.

Early identification of CP can be challenging in the primary care context. Most primary care practitioners do not have the advanced knowledge in atypical child development that medical specialists have (e.g. child neurologists, developmental pediatricians), and while developmental screening tools exist for primary care providers, they are meant to identify significant delays in motor milestones but are not specific or sensitive enough to capture the early motor patterns characteristic of CP.[5] A recent review of clinical features associated with CP found that the available evidence is sparse and often based on expert opinion rather than empirical data and emphasizes risk factors rather than single objective clinical signs warranting referral.[6]

Furthermore, the focus of most of this research has been on high-risk groups (e.g. infants born very preterm, neonatal intensive care unit graduates with neonatal encephalopathy), and not community-based populations where most children with CP are initially seen.

In the absence of evidence-based research on early clinical features of CP that can be readily identified by primary care physicians (family physicians, pediatricians) in the context of short but comprehensive well-child care visits in the community, the opinions and experience of clinicians and other experts via consensus methods will be used to inform clinical decisions.[7] Our aim is to create content for educational knowledge translation tools to assist with the early detection of CP, designed specifically for the primary care context. These tools can be used to inform primary care practitioners and parents of the early signs of CP, with the aim of decreasing delays in detection (and subsequent delayed referral for diagnostic assessment and intervention) by increasing awareness of the early motor signs of CP. Our research team recently identified such features in a consensus investigation that involved regional and national clinical experts (developmental pediatricians, child neurologists, rehabilitation specialists) and knowledge users (primary care physicians, parents).[8] Using nominal group techniques, six distinct features were identified that should warrant primary care physicians to refer to medical specialists for diagnostic assessment of CP, when any single one of these is observed. Recommendations for simultaneous referral to other health professionals for assessment and intervention were put forth by consensus methods. To ensure clinical appropriateness and generalizability beyond the Canadian primary care context, these findings require additional validation.

The current study aimed to validate the results obtained in our two previous consensus groups with Canadian content experts and knowledge users.[8] The research questions guiding this study, which were posed to international experts in the field of early identification of CP, were the following: to what extent is there international agreement about (1) the early motor signs ('clinical features') of CP that were identified that should prompt early referral for diagnostic assessment and (2) the referral recommendations to other health professionals at the same time children suspected of CP are referred to medical specialists for diagnostic investigation?

What this paper adds

- International experts provide strong agreement on clinical features to detect cerebral palsy.
- Consensus on clinical 'warning signs' to monitor over time.
- Referral recommendations from primary care to specialized health services are identified.

METHOD

Research design

The Delphi technique, first developed by Dalkey and Helmer,[9] is a recognized formal consensus methodology that has been applied across varied healthcare settings and populations.[10–12] The Delphi method is structured and iterative, employing multiple survey rounds whereby the results of one informs the next survey.[13] It offers several advantages, including total anonymity (no face-to-face meetings) which enhances objective responses, and the online format enables the recruitment and participation of an international expert panel.[14–16] This Delphi study was integrated in a larger integrated knowledge translation project entitled 'PROMPT Identification of Cerebral Palsy: Primary-care Referral Of Motor-impaired children: Physician Tools' (the PROMPT study).

Participants

Respondent group

We identified a sample of experts, defined as clinicians from a variety of health professions with clinical and/or research expertise in the field of CP diagnosis and assessment and international professional reputation in this regard. This was not meant to be an all-inclusive list, but a representative sample with geographical variation. Accordingly, the research team identified a purposive sample of international experts by peer nomination and by scanning the Member Directory of the American Academy for Cerebral Palsy and Developmental Medicine. The final list generated included 51 experts from Asia, Australia, Canada, Europe, and the USA. Next, e-mails were sent to all 51 of the identified experts. The e-mail provided a brief overview of progress, informed them that they had been identified by our research team as an expert in the field of CP, and invited them to participate in a research project that aimed to promote early detection of CP by primary care providers (e.g. pediatricians, family physicians, nurse practitioners). Specifically, it was explained that their participation was requested to validate the findings of two previous consensus groups (using nominal group techniques with a sample of Canadian content experts and knowledge users). Consensus was achieved on: (1) specific clinical features that should prompt referral from primary care practitioners; and (2) other referral recommendations for children suspected of having CP to occur simultaneously with referral for diagnostic assessment. Interested participants were instructed to click on the link provided in the body of the e-mail, which led to a consent page containing the following: title and purpose of the research study, goals of the online survey, study procedures, possible risks and benefits, the individual's rights and option to withdraw from the study, confidentiality and sharing of results, and contact information. Provision of consent was required to proceed to the survey. Weekly reminders were sent to non-responders for 5 weeks for round one, and for 7 weeks for round two.

Procedure

Delphi survey instrument

Ethical approval for this study was obtained from the McGill Faculty of Medicine Research Ethics Office/Institutional Review Board (institutional review board study number

A00-E79-15A). Once consent was obtained, participants were provided access to a survey instrument via REDCap (https://www.project-redcap.org/), a secure online platform for administering surveys and collecting data, as this allowed anonymous and independent responses limiting potential social desirability bias. Before starting the survey, respondents were asked to complete a brief demographic questionnaire, consisting of three questions pertaining to their: (1) profession; (2) country of practice; and (3) number of years since obtaining their professional degree. Participants were then directed to the three-part survey for round one.

Round one

The first round of the Delphi survey consisted of a three-part questionnaire with a total of 13 items. Participants were asked to indicate their level of agreement with each item, which was rated using a 4-point Likert scale (strongly disagree; disagree; agree; strongly agree), in response to two questions: (1) whether the item should be included in our knowledge translation tool for primary care providers to assist with detection and referral practices; and (2) whether the wording of the item was clear and appropriate for the target audience. Participants were also given the opportunity to provide additional suggestions on other possible features or referral recommendations and on how to better articulate these features and recommendations. In part one, participants were asked to rate their level of agreement with each of the proposed separate features (n=6 items) that individually should prompt referral for diagnostic assessment of CP. In part two, participants were asked to consider the proposed 'warning signs' (n=2 items) to determine whether they agreed that either sign should prompt closer monitoring of the child's development at subsequent primary care follow-up visits. In part three of the survey, participants were asked to consider a series of referral recommendations (n=5 items) to other health professionals at the time the child was being referred for diagnostic assessment of CP. For the full list of items in each part of round one, refer to Appendix S2 (online supporting information).

Round two

Upon completion of round one, the results were reviewed by the advisory panel (see 'Data analysis' section) to determine whether there was high enough agreement to retain each item or not, and whether wording should be improved on the basis of suggestions provided. In addition, new features proposed by respondents were considered for the survey in round two. The second round of the Delphi survey focused specifically on optimizing the wording of the features and recommendations, since there was high agreement to retain all proposed features and referral recommendations. Round two consisted of a two-part survey with a total of six items. Participants were asked to choose from the revisions proposed by the advisory panel, which included several alternative descriptions of the feature based on recommendations from experts in round one so as to seek consensus, and respondents were again provided with the opportunity to give other suggestions to enhance the wording for each feature. In part one of the second round of the Delphi survey, participants were asked to consider the rewording of four features that should prompt referral for diagnostic assessment of CP. In part two, participants were asked to consider the proposed rephrasing of two referral recommendations. For the full list of items in each part of round two, refer to Appendix S3 (online supporting information).

Data analysis

Looking for agreement concerning item-inclusion and item-working, we a priori defined consensus as achieving at least 70% agreement by international experts for each item (i.e. feature, warning sign, or referral recommendation).[17] Agreement was considered achieved for an item if 'strongly agree' or 'agree' was indicated on the Likert scale. For each clinical feature/warning sign/referral recommendation, we were exploring agreement concerning (1) item-inclusion and (2) item-wording. A high cut-off for agreement (i.e. 70% a priori threshold) among experts was used specifically to assess agreement on item-inclusion, since the aim of this study was to validate the appropriateness and generalizability of each item beyond the Canadian context. Although we considered the level of agreement on item-wording to enhance the clarity of each item, further refinement of the item-wording will be performed in the next phase of this project. Specifically, we will be conducting focus groups with primary care practitioners and parents of children with CP, presenting them with the final lists of clinical features and referral recommendations, and collaborating with them on the optimal wording and formatting for the knowledge translation tools we are developing. As such, it was not as important that the agreement on item-wording reached this 70% threshold, since the final wording will be determined by the end-users.

A two-step analysis was conducted. Results were first reviewed by two primary authors (ZB, AM) and synthesized for presentation and discussion with the advisory panel. This panel included local and national content-experts and knowledge-users who had participated in the previous consensus groups and was composed of the following: pediatric neurologists (n=3), occupational therapists (n=3), developmental pediatricians (n=2), a community-based pediatrician (n=1), a family physician (n=1), and a mother of a child with CP (n=1). The role of the panel was to help determine what items from round one needed to be revisited in a second survey (round two), as part of the Delphi process. In addition, the panel reviewed all suggestions made by respondents of potential additional features or referral recommendations. These were carefully considered on the basis of the specificity of the suggested item to CP, the practicality of use in the primary care context, and the ability for primary care providers to understand and recognize the features. Upon consultation and discussion with this panel, only items from round one that

failed to meet at least 70% agreement, and/or that the panel determined from participant feedback required further clarification, were retained for round two.

RESULTS

Round one

Participants

The round one survey was completed by 76.5% (*n*=39 out of 51) of the invited participants. These included developmental pediatricians 28.2% (*n*=11), occupational therapists 20.5% (*n*=8), physical therapists 17.9% (*n*=7), child neurologists 12.8% (*n*=5), or 'other' 20.5% (*n*=8; neonatologists, pediatricians, pediatric physiatrist, researcher, orthopedic surgeon). These participants were from Australia (*n*=15), USA (*n*=8), Canada (*n*=9), Europe (*n*=5), Asia (*n*=1), and 'other' (*n*=1). More than half (66.7%; *n*=26 out of 39) of participants had obtained their professional degree between 21 and 50 years previously, while 25.6% (*n*=10) obtained theirs 11 to 20 years previously, 5.1% (*n*=2) indicated it was 5 to 10 years ago, and one (2.6%) responded that it had been 50 or more years since they obtained their professional degree.

Agreement

Agreement between participants on inclusion of each of the six clinical features that should prompt referral for diagnostic assessment of CP was very high (97.4–100.0%), while agreement on the wording of these features ranged from 65.8% to 84.2%. Agreement on the inclusion of the clinical features to be considered by primary care providers as 'warning signs' that should suggest monitoring rather than immediate referral for diagnostic assessment was similarly high (84.6–87.2%), while agreement on the wording of these warning signs ranged from 78.8% to 85.3%. Finally, referral recommendations to rehabilitation specialists and other health professionals by primary care practitioners to occur simultaneously with referral to a medical specialist for diagnostic assessment demonstrated similarly high agreement between participants (84.6–100.0%), with agreement on the wording of these recommendations ranging from 63.6% to 89.7%. For the full list of agreement on inclusion and wording, refer to Table I.

No new features were suggested with any consistency, and those suggested were determined by the advisory panel as not being specific to CP (e.g. 'delay in general activity level … low level of initiative of play and mobility'; 'feeding difficulties') or not feasible for primary care context (e.g. 'abnormal general movements assessment'; 'abnormal imaging'). The suggestions for additional 'warning signs' were felt to be similarly not CP-specific enough (e.g. 'irritability'; 'early signs of dysphagia'; 'not talking in sentences by 24 months'). Finally, of the additional referral recommendation suggestions, there was a recurrent theme of providing psychosocial support for families (e.g. 'families of a child diagnosed with CP or at high risk of CP should be offered early psychological support and preventative care for family mental health'; 'parental support with psychologist or social work'; 'a family support worker [e.g. psychologist, social worker] and information on where to find a parent-to-parent support program'). It was discussed and determined by the advisory panel that it would be important to include something in the tool related to acknowledging the potential need for, and availability of, psychosocial support for families of children recently diagnosed with CP.

Table I: Results of round one of the Delphi survey

Features (one or more of the following)	Agreement on inclusion (%)	Agreement on wording (%)
Part one: clinical features to prompt referral for diagnostic assessment		
1. The child demonstrates early handedness before 12mo of age	97.4	65.8
2. The child demonstrates stiffness or tightness in the legs between 6–12mo of age (e.g. unable to bring their toes to mouth during diaper/nappy change)	100.0	69.2
3. The child demonstrates persistent fisting of the hands beyond 4mo of age	100.0	89.7
4. The child demonstrates a persistent head lag beyond 4mo of age	97.4	84.2
5. The child is not able to sit without support beyond 9mo of age	100.0	76.9
6. The child demonstrates any asymmetry in posture or movement	84.6	63.6
Part two: 'warning sign' features to prompt monitoring rather than referral for diagnosis		
1. The child demonstrates a persistent startle (Moro) reflex beyond 6mo of age	87.2	85.3
2. The child demonstrates consistent toe-walking or asymmetric-walking beyond 12mo of age	84.6	78.8
Part three: referral recommendations to occur simultaneously with referral to a medical specialist for diagnosis		
1. All children should be referred to a motor intervention specialist (e.g. occupational therapist and/or physical therapist)	94.7	72.2
2. If the child manifests a delay in communication they should be referred to a speech-language pathologist	100.0	76.3
3. If the child manifests hearing concerns, a referral should be made to an audiologist	100.0	84.2
4. If the child manifests vision difficulties (e.g. not fixating, following, and/or tracking) a referral should be made to a functional vision specialist (e.g. optometrist or occupational therapist)	84.2	59.4
5. If the child manifests feeding difficulties (e.g. poor sucking, swallowing, choking, not gaining weight) a referral should be made to a feeding specialist (e.g. occupational therapist or speech-language pathologist)	73.7	73.0

Round two

Participants

All international experts from round one were invited to participate in round two, and 84.6% (*n*=33 out of 39) completed the second survey, along with three additional participants who did not participate in the initial round. These 36 participants included developmental pediatricians 25.0% (*n*=9), occupational therapists 22.2% (*n*=8), physical therapists 22.2% (*n*=8), child neurologists 13.9% (*n*=5), or 'other' 16.7% (*n*=6; neonatologists, pediatrician, pediatric physiatrist, researcher, child neuropsychiatrist). The countries/continents represented were Australia 36.1% (*n*=13), Canada 22.2% (*n*=8), USA 22.2% (*n*=8), Europe 16.7% (*n*=6), and Asia 2.8% (*n*=1). More than half (61.6%; *n*=22) of participants obtained their professional degrees between 21 and 50 years ago, while 30.6% (*n*=11) obtained theirs 11 to 20 years ago, 5.6% (*n*=2) indicated it was 5 to 10 years ago, while 2.8% (*n*=1) responded that it had been 50 or more years since they obtained their degree.

Agreement

In round one the agreement between international experts on item-inclusion was demonstrated to be very high for all features and for most referral recommendations; thus the focus of round two of the Delphi was on agreement related to item-wording. Four items related to features were noted to be less clearly articulated, and several suggestions were proposed by participants and refined by the advisory panel. Similarly, two items pertaining to referral recommendations were also deemed to require greater clarity, and revisions were drafted by the panel. For each item in round two, participants were presented with two to four of these proposed revisions (Appendix S3), and they were asked to indicate their preference from among the options. Agreement

Table II: Results of round two of the Delphi survey

Part one: clinical features to prompt referral for diagnosis

Features	Revisions proposed by the advisory panel	Agreement on wording (%)
Original item 1.1 The child demonstrates early handedness before 12mo of age	The child demonstrates a *hand preference* before 12mo of age	77.8 (*n*=28)
	The child demonstrates an *early hand preference*, before 12mo of age	22.2 (*n*=8)
Original item 1.2 The child demonstrates stiffness or tightness in the legs between 6–12mo of age (e.g. unable to bring their toes to mouth during diaper/nappy change)	The child demonstrates stiffness or tightness in the legs between 6–12mo of age (e.g. unable to bring their toes to mouth *when having their diaper/nappy changed*)	61.1 (*n*=22)
	The child demonstrates stiffness or tightness in the legs between 6–12mo of age (e.g. unable to bring their toes to mouth *during a diaper/nappy change*)	38.9 (*n*=14)
Original item 1.3 The child demonstrates persistent fisting of the hands beyond 4mo of age	The child keeps their hands *fisted (closed)* after the age of 4mo	13.9 (*n*=5)
	The child keeps their hands *fisted (closed/clenched)* after the age of 4mo	86.1 (*n*=31)
Original item 1.6 The child demonstrates any asymmetry in posture or movement	The child demonstrates *asymmetry of posture and movements* after the age of 4mo	22.2 (*n*=8)
	The child demonstrates *frequent asymmetry of posture and movements* after the age of 4mo	19.4 (*n*=7)
	The child demonstrates *consistent asymmetry of posture and movements* after the age of 4mo	36.1 (*n*=13)
	The child *habitually demonstrates asymmetry of posture nd movements* after the age of 4mo	22.2 (*n*=8)

Part two: referral recommendations to occur simultaneously with referral to a medical specialist for diagnosis

Referral recommendation	Revisions proposed by the advisory panel	Agreement on wording (%)
Original item 3.1 All children should be referred to a motor intervention specialist (e.g. occupational therapist and/or physical therapist)	All children should be referred to a motor intervention specialist (e.g. *pediatric occupational therapist and/or pediatric physical therapist*)	41.7 (*n*=15)
	All children should be referred to an occupational therapist and/or physical therapist *with expertise in child development*	25.0 (*n*=9)
	All children should be referred to an occupational therapist and/or physical therapist *with pediatric clinical experience*	33.3 (*n*=12)
Original item 3.4 If the child manifests vision difficulties (e.g. not fixating, following, and/or tracking) a referral should be made to a functional vision specialist (e.g. optometrist or occupational therapist)	If the child manifests vision difficulties (e.g. not fixating, following, and/or tracking) a referral should be made to an *optometrist or an ophthalmologist for assessment/evaluation, and to a functional vision specialist for intervention* (e.g. occupational therapist with expertise in pediatric vision; early childhood vision consultants)	38.9 (*n*=14)
	If the child manifests vision difficulties (e.g. not fixating, following, and/or tracking) a referral should be made to *an optometrist or an ophthalmologist, and to a functional vision specialist* (e.g. occupational therapist with expertise in pediatric vision; early childhood vision consultants)	61.1 (*n*=22)

between participants on the wording of the four clinical features was high overall, although it ranged from 36.1% to 86.1%. Agreement between experts on the wording of the two referral recommendations to occur simultaneously with referral to a medical specialist for diagnostic assessment ranged from 41.7% to 61.1%. The complete list of agreement on inclusion and wording for round two can be found in Table II.

There is no widely agreed upon stopping-point in the Delphi process.[18] The main focus of this study was on item-inclusion, since the final item-wording will be determined collaboratively with content-users (primary care practitioners, parents). As such, having achieved levels of agreement for item-inclusion that fall within an acceptable rate[17,19,20] and an adequate response rate,[17] it was determined that a third round of the Delphi was not required. Thus, the final list of features to prompt referral for diagnostic assessment, warning signs to monitor, and referral recommendations to occur simultaneously with referral for diagnosis can be found in Table III.

DISCUSSION

Results from this international Delphi survey further validate previous findings on features and recommendations for referral from two recent nominal groups with Canadian experts in early identification of CP.[8] Specifically, high-level agreement currently exists on: (1) the clinical features that should prompt referral for diagnosis; (2) 'warning signs' features that warrant monitoring rather than immediate referral for diagnosis; and (3) referral recommendations to other healthcare professionals to occur simultaneously with referral for diagnosis. The results of this international Delphi study complement the important recent work in the field of early identification and early intervention. Novak et al.[4] published an algorithm for early detection of CP or those at high-risk of CP, achieved by consensus. The algorithm, which focuses on both newborn and infant detectable risks, consists of a combination of a standardized clinical neurological examination (Hammersmith Infant Neurological Examination), brain imaging (magnetic resonance imaging [MRI]), and standardized motor evaluations, selected on the basis of the infant's age. At the age of less than 5 months, the Prechtl Qualitative Assessment of General Movements together with MRI results are recommended as early detection tools. In cases where MRI and/or the Prechtl Qualitative Assessment of General Movements are not available, a combination of the Hammersmith Infant Neurological Examination and the less sensitive Test of Infant Motor Performance is suggested. For infants older than 5 months of age, use of the Hammersmith Infant Neurological Examination together with results of the MRI and those of a standardized motor assessment is proposed. The motor tests available for infants younger than 5 months corrected age are the Developmental Assessment of Young Children, the Alberta Infant Motor Scale, and the Neuro-Sensory Motor Development Assessment, depending on previous examinations. While this combination may demonstrate diagnostic predictive accuracy for high-risk populations, which represents about half of children with CP, we note that the accuracy of these methods in infants with later discernable risks for CP (i.e. no high-risk perinatal history) is not yet known. Furthermore, these various standardized assessments require training, time to administer (e.g. ≥30min), and time to score and interpret the results. This may therefore not be feasible to integrate into the primary care context. Similarly, Noritz et al.[5] proposed an algorithm for surveillance and screening for pediatricians to begin the diagnostic and referral process earlier. Although an excellent synthesis, the neurological examination process is somewhat lengthy, and the focus is on detecting neuromotor impairment in general (i.e. to include degenerative disorders, and peripheral and central nervous system disorders), but is not CP specific. Our study offers a tailored and feasible method of identifying children with CP that can be readily integrated by primary care providers in clinical practice.

Table III: International consensus on clinical features, warning signs, and referral recommendations for cerebral palsy

Clinical features to prompt referral for diagnosis (one or more of the following)
1. The child demonstrates *a hand preference* before 12mo of age
2. The child demonstrates *stiffness or tightness in the legs* between 6–12mo of age (e.g. unable to bring their toes to mouth when having their diaper/nappy changed)
3. The child keeps their *hands fisted (closed/clenched)* after the age of 4mo
4. The child demonstrates a *persistent head lag* beyond 4mo of age
5. The child is *not able to sit without support* beyond 9mo of age
6. The child demonstrates *consistent asymmetry of posture and movements* after the age of 4mo
'Warning Sign' features to prompt monitoring rather than referral for diagnosis (either of the following)
1. The child demonstrates a *persistent startle (Moro) reflex* beyond 6mo of age
2. The child demonstrates *consistent toe-walking or asymmetric-walking* beyond 12mo of age
Referral recommendations to occur simultaneously with referral to a medical specialist for diagnosis
1. All children should be referred to a *motor intervention* specialist (e.g. pediatric occupational therapist and/or pediatric physical therapist)
2. If the child manifests a delay in communication they should be referred to a *speech-language pathologist*
3. If the child manifests hearing concerns a referral should be made to an *audiologist*
4. If the child manifests vision difficulties (e.g. not fixating, following, and/or tracking) a referral should be made to an *optometrist or an ophthalmologist*, *and* to a *functional vision specialist* (e.g. occupational therapist with expertise in pediatric vision; early childhood vision consultants)
5. If the child manifests feeding difficulties (e.g. poor sucking, swallowing, choking, not gaining weight) a referral should be made to a *feeding specialist* (e.g. occupational therapist or speech-language pathologist)

The availability of a tailored and feasible method for identifying children with CP that can be readily integrated in primary care settings has therefore become an imperative for family physicians and other primary care providers. Our previous work[8] demonstrated that there was a national agreement on: (1) the clinical features that should prompt early referral for a diagnostic work-up; (2) 'warning sign' features that warrant monitoring rather than immediate referral for diagnostic assessment; and (3) referral recommendations to other healthcare professionals to occur simultaneously with referral for diagnostic assessment. Results of this Delphi survey go beyond this and indeed confirm that there is international consensus on the proposed features and recommendations for referral.

This study is not without a few important limitations. First, the Delphi technique has its own inherent limitations, including the following: the feedback we provided participants from round one may have influenced their judgments in round two; and there is a potential for participant 'burnout' with the demands of each subsequent round.[16] However, we feel that this was a good fit considering the complementary nature between the nominal group technique and Delphi. Second, the choice of participants (or how 'experts in CP' were identified) was subject to the discretion of the authors, and many of those identified were the authors' colleagues, so this too may have influenced their participation. However, the criterion of being a recognized international expert in the diagnosis and treatment of CP was always respected (purposeful sampling), as well as the interest in maximizing geographical variation. Third, although there is no agreed definition of how to measure 'consensus', we did a priori establish the agreement cut-offs in the particular context of this investigation, as it has been largely recommended.[21] A final limitation is that we excluded lower middle-income countries, because their healthcare contexts and systems are quite different, and would require a different purposive sampling approach for their local contexts.

The very high agreement between international experts on both the features and referral recommendations in this study suggests that they are appropriate for use within the primary care context for detecting signs that warrant referral for diagnostic assessment and concurrent referrals for rehabilitation and other health professionals. This will inform the content of knowledge translation tools for the primary care context, the final wording and formatting of which will be determined through collaboration with relevant stakeholders (primary care practitioners, parents of children with CP). A qualitative descriptive study using focus groups will be conducted in the primary care context, to gather family physicians' and parents' views about the optimal manner of delivering the content that was validated through this Delphi survey.

ACKNOWLEDGEMENTS

The authors have stated that they had no interests that could be perceived as posing a bias or conflict. The authors disclose receipt of the following financial support for the research, authorship, and publication of this article: ZB received salary support as a doctoral trainee from funding from the Canadian Institutes of Health Research (MOP-133402), which supported this study; ZB also received financial support from the Richard and Edith Strauss Canada Foundation as a Musculoskeletal Doctoral Fellow, as well as from the Hoppenheim Fund/Montreal Children's Hospital Foundation as a trainee. We express our sincere appreciation to the international experts who volunteered their time and contributed their expertise. We also express our gratitude to research assistants Deniz Keskinel, Quynh Nguyen, and Anna Radzioch Guerin for their support with REDCap database management and in preparing this manuscript. This project was supported by infrastructure provided by the Centre for Interdisciplinary Research in Rehabilitation of Greater Montreal, the Research Institute of the McGill University Health Centre, and the CHILD-BRIGHT Network.

SUPPORTING INFORMATION

The following additional material may be found online:

Appendix S1: Members of the PROMPT group.

Appendix S2: Delphi survey instrument, round one.

Appendix S3: Delphi survey instrument, round two.

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RESUMEN

RECOMENDACIONES DE EXPERTOS INTERNACIONALES SOBRE CARACTERÍSTICAS CLÍNICAS PARA UNA DERIVACIÓN RÁPIDA PARA LA EVALUACIÓN DIAGNÓSTICA DE LA PARÁLISIS CEREBRAL

OBJETIVO Establecer recomendaciones de expertos internacionales sobre características clínicas para iniciar derivación para la evaluación diagnóstica de la parálisis cerebral (PC).
MÉTODO Se realizó una encuesta online tipo Delphi con expertos en identificación e intervención temprana de niños con PC a fin de validar los resultados obtenidos en dos grupos de consenso realizados previamente en Canadá con expertos en contenidos y usuarios. Se enviaron dos rondas de cuestionarios por correo electrónico. Los participantes calificaron su acuerdo con un puntaje de 4 puntos en una escala Likert y con preguntas opcionales de respuesta abierta para comentarios adicionales. Además, un panel de expertos y usuarios revisaron los resultados de cada ronda y determinaron el contenido de las encuestas subsiguientes.
RESULTADOS En general, hubo un alto nivel de acuerdo sobre: (1) seis características clínicas que requieren derivación rápida para el diagnóstico, (2) dos características de "señales de advertencia" que requieren monitoreo en lugar de referencia inmediata para el diagnóstico, y (3) cinco recomendaciones de referencia a otros profesionales de la salud que deben realizarse simultáneamente con la derivación para el diagnóstico.
INTERPRETACIÓN Hubo gran acuerdo entre los expertos internacionales, sugiriendo que las características y recomendaciones de referencia propuestas para los médicos de atención primaria para la detección de PC fue ampliamente generalizable. Estos resultados informarán el contenido de herramientas educativas para mejorar la detección precoz de PC en el contexto de atención primaria.

RESUMO

RECOMENDAÇÕES DE ESPECIALISTAS INTERNACIONAIS SOBRE ASPECTOS CLÍNICOS DISPARADORES DE ENCAMINHAMENTO PARA AVALIAÇÃO DIAGNÓSTICA EM PARALISIA CEREBRAL

OBJETIVO Estabelecer recomendações de especialistas internacionais sobre os aspectos clínicos disparadores de encaminhamento para avaliação diagnóstica em paralisia cerebral (PC).
MÉTODO Um levantamento online internacional do tipo Delphi foi realizado com especialistas em identificação e intervenção precoce para crianças com PC, para validar os resultados obtidos em dois consensos prévios com especialistas no conteúdo e usuários canadenses. Enviamos duas rodadas de questionários por email. Os participantes pontuaram sua concordância usando uma escala Likert de 4 pontos, junto com questões abertas opcionais para informações adicionais. Além disso, um painel de especialistas e usuários revisaram os resultados de cada rodada, e determinaram o conteúdo das pesquisas subsequentes.
RESULTADOS Em geral, houve alto nível de concordância em: 1) seis aspectos clínicos que devem disparar encaminhamento para diagnóstico. 2) dois 'sinais de alerta' que merecem monitoramento mas não encaminhamento imediato para diagnóstico, e 3) cinco recomendações de encaminhamento para outros profissionais da saúde simultaneamente ao encaminhamento para diagnóstico.
INTERPRETAÇÃO Houve alta concordância entre especialistas internacionais, sugerindo que os aspectos e recomendações para encaminhamento propostos para médicos na atenção básica para a identificação precoce da PC foram amplamente generalizáveis. Estes resultados informarão o conteúdo de ferramentas educacionais para melhorar a detecção precoce de PC no contexto da atenção básica.

Appendix B

Delphi Survey Instrument, Round Two

Round Two, Part One: 4 items
Attributes to PROMPT referral for diagnosis of cerebral palsy

1. Original item #1.1:
   The child demonstrates *early handedness* before 12 months of age.
   Proposed revisions:

a) The child demonstrates *a hand preference* before 12 months of age.
b) The child demonstrates *an early hand preference,* before 12 months of age.

2. Original item #1.2:
   The child demonstrates stiffness or tightness in the legs between 6-12 months of age (e.g. unable to bring their toes to mouth during *diapering*).
   ***Proposed revisions*:**

a) The child demonstrates stiffness or tightness in the legs between 6-12 months of age (e.g. unable to bring their toes to mouth *when having their diaper / nappy changed*).
b) The child demonstrates stiffness or tightness in the legs between 6-12 months of age (e.g. unable to bring their toes to mouth *during a diaper / nappy change*).

3. *Original item #1.3:*
   The child demonstrates *persistent fisting of the hands* beyond 4 months of age.
   ***Proposed revisions*:**

a) The child keeps their hands *fisted (closed)* after the age of 4 months.
b) The child keeps their hands *fisted (closed/clenched)* after the age of 4 months.

4. *Original item #1.6:*
   The child demonstrates *any asymmetry* in posture or movement.
   ***Proposed revisions*:**

a) The child demonstrates *asymmetry* of posture and movements after the age of 4 months.
b) The child demonstrates *frequent asymmetry* of posture and movements after the age of 4 months.
c) The child demonstrates *consistent asymmetry* of posture and movements after the age of 4 months.
d) The child *habitually demonstrates asymmetry* of posture and movements after the age of 4 months.

Round Two, Part Two: 2 items
Referral recommendations to occur simultaneously with referral for diagnosis of cerebral palsy

1. Original item #3.1:
   All children should be referred to a *motor intervention specialist* (e.g. occupational therapist and/or physical therapist).
   Proposed revisions:

a) All children should be referred to a motor intervention specialist (e.g. *pediatric occupational therapist and/or pediatric physical therapist*).
b) All children should be referred to an occupational therapist and /or physical therapist with *expertise in child development*.
c) All children should be referred to an occupational therapist and /or physical therapist with *pediatric clinical experience*.

2. Original item #3.4:
   If the child manifests vision difficulties (e.g. not fixating, following, and/or tracking) a referral should be made to a *functional vision specialist* (e.g. optometrist or occupational therapist).
   Proposed revisions:

a) If the child manifests vision difficulties (e.g. not fixating, following and /or tracking) a referral should be made to an *optometrist or an ophthalmologist for assessment/evaluation*, and to a *functional vision specialist for intervention* (e.g. occupational therapist with expertise in pediatric vision; early childhood vision consultants)
b) If the child manifests vision difficulties (e.g. not fixating, following and /or tracking) a referral should be made to an *optometrist or an ophthalmologist*, and to a *functional vision specialist* (e.g. occupational therapist with expertise in pediatric vision; early childhood vision consultants)

Appendix C

The PROMPT Group Members

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CHAPTER 12:
DISCUSSION

12.1 Summary of Findings

Early identification of children with suspected CP, followed by early referral to a medical specialist for diagnostic assessment and simultaneous referral to rehabilitation specialists for intervention is considered best clinical practice. Early identification in the primary care context is problematic. Specifically, primary care practitioners are often not aware of the early clinical features of CP. Furthermore, the profile of children with CP who may typically be identified in this context (e.g. term born, no apparent high-risk history, mild to moderate severity of motor impairment) might present an additional challenge to the primary care clinician. It was determined that educational KT tools designed for use in the primary care context could increase awareness of the early features of CP, and potentially address the gaps in knowledge that have been identified. The hope would be that in turn this would prompt early referral and decrease delays in diagnosis and rehabilitation trajectories for children with CP. This research project sought to create the knowledge essential to inform the content of these KT tools. Thus, the first phase of this research project involved a knowledge synthesis and the second phase focused on knowledge creation, for the eventual creation of knowledge translation tools.

Knowledge Synthesis

Manuscript 1 used a scoping review methodology to determine what was known about age at referral for diagnosis and rehabilitation services for children suspected of having CP, and to identify factors associated with earlier referral. A systematic review of literature between 1979 and 2017 on *age at referral for diagnosis* or *age at referral to rehabilitation services* for children suspected of having CP was conducted, and both quantitative and thematic analyses were performed. Of 777 studies, 15 were retained for final analysis. Overall, evidence was sparse and

was mostly based on older studies (birth cohorts from the mid-1950s to the 1980s). The available studies suggested there was high variation in the age at referral for diagnosis (range: 10-21 months), and indicated that subgroups of children with CP might be experiencing prolonged delays. Factors identified as being potentially predictive of age at referral for diagnosis and for rehabilitation included the following: referral source, type of CP and a complicated birth history. The results demonstrated that there was a need for population-based evidence on the current referral practices of primary care practitioners and on the factors that contribute to current delays in referral and diagnosis of children suspected of having CP (Specific Objective 1).

To address the needs identified above**, Manuscript 2** used a single-site retrospective chart review (using a convenience sample) to attempt to describe current referral practices with respect to age at referral for diagnosis of CP, and to identify factors that influence age at referral. Results indicated that 78.6% of children were referred for diagnosis by a medical specialist (mean: 13.6 + 15.7 months), whereas referral from primary care providers were significantly more delayed (mean: 28.8 + 27.1 months), and that children who were initially admitted to a neonatal intensive care unit were referred earlier (mean: 9.3+10.2 months) than those not (28.1+24.9 months). Referral to rehabilitation was similarly delayed. The results highlighted that referrals from primary care practitioners are considerably delayed, and that a subset of children with CP (e.g. children not initially admitted to a neonatal intensive care unit; children with hemiplegia and especially diplegia) were experiencing significantly delayed referral for diagnosis and rehabilitation. This served as a pilot for the following study (Specific Objective 1).

Informed by the results of the single-institution pilot study, the goal of **Manuscript 3** was to collect population-based evidence describing current physician referral practices with respect to age at referral for diagnosis of CP and to rehabilitation specialists for intervention, and to identify

factors associated with delayed referral. A national (multi-regional) environmental scan of 455 children (born in Canada between 2008-2011) who were diagnosed with CP was conducted, using both the medical charts of the children and the research infrastructure afforded by the Canadian Cerebral Palsy Registry. Age (in months) at referral for diagnostic assessment (mean: 12.7±14.3; median: 8.0), diagnosis (mean: 18.9±12.8; median: 16.0), referral for rehabilitation services (mean: 13.4±13.5; median: 10.0), and rehabilitation initiation (mean: 15.9±12.9; median: 12.0) were documented. Factors collectively associated with delayed referral were lower maternal education, milder severity of motor dysfunction, having hemiplegia or diplegia, early discharge from hospital after birth, and region of residence. These results confirmed what had previously been observed in the pilot study: highlighting that wide variation exists in the age at which referral for diagnostic assessment and rehabilitation interventions occur, that referrals from primary care practitioners tend to be significantly delayed, and that a subset of children with CP are experiencing prolonged delays for tend to be both assessment and rehabilitation referrals (Specific Objective 1).

The results of these three studies (**Manuscripts 1,2,3**) have contributed new knowledge to the literature on early identification of CP. These findings provide more contemporary estimates of age at referral, and highlight the wide variation and factors that are associated with prolonged delays in referring young children suspected of having CP for diagnostic assessment and simultaneous rehabilitation interventions. The wide variation in age suggests that some children are identified and referred early, highlighting the opportunity to enhance earlier identification and referral for others who are referred significantly later. A recent scoping review sought to identify the clinical features associated with the early detection of CP that can be used by the primary care provider, but the authors ultimately concluded that the evidence was sparse, inconsistent and often not specific to CP (Garfinkle, Li, Boychuck, Bussières, & Majnemer, in press). Primary care

practitioners often do not have the advanced training in atypical child development that medical specialists receive (e.g. developmental pediatricians, child neurologists), and may not recognize the early features of CP. It was thus determined that user-friendly educational knowledge translation tools, aimed at increasing awareness amongst primary care practitioners of the features of CP that should prompt referral for diagnostic assessment and for rehabilitation interventions, might be a strategy to decrease the delays currently experienced by a subset of children with CP. Thus, it was necessary to create the knowledge primary care practitioners need to enhance their early detection of CP. Indeed the educational tools may potentially be of benefit for parents to use as well.

Knowledge Creation

To address the knowledge gap identified in the studies contained in **Manuscripts 1-3**, the study in **Manuscript 4** sought to develop expert-informed content regarding the features of CP that are observable early on that should prompt referral for diagnostic assessment, as well as simultaneous referral recommendations for other investigations or interventions. This was achieved by conducting two nominal group processes (consensus methodology) with 18 national (Canadian) *content-experts* (e.g. medical specialists, rehabilitation clinicians and researchers with expertise in early intervention) and *knowledge-users* (e.g. primary care physicians, parents of children with CP). There were 6 possible attributes identified that should be used to prompt referral for diagnostic assessment; 2 'warning signs' agreed upon that should prompt closer monitoring and surveillance over time rather than immediate referral for diagnostic assessment; and 5 recommendations for simultaneous referral to other health professionals. This evidence was to be used to inform the content of educational knowledge translation tools for primary care practitioners, but a major limitation at this point was

that the results were obtained using only Canadian participants. Thus, further efforts were required in order to determine the validity and generalizability of these results (Specific Objectives 2a, 2b).

The study described in **Manuscript 5** sought to validate the results of the study in Manuscript **4** and to establish international expert recommendations on clinical features to prompt referral for diagnostic assessment of CP. An online Delphi survey of 42 international content-experts (peer-recognized experts in early identification and intervention for children with CP) was conducted. Consensus was achieved on the following: 6 clinical features to prompt referral for diagnosis; 2 'Warning Sign' features to prompt monitoring rather than referral for diagnosis; and 5 referral recommendations to occur simultaneously with referral to a medical specialist for diagnosis. The very high agreement observed between international experts suggests that the results presented in **Manuscript 4** were indeed valid and generalizable for use within a primary care context. Minor wording suggestions were provided and collectively endorsed to enhance the clarity of these recommendations. The refined results from the study in **Manuscript 5** will be used to inform the content of educational knowledge translation tools for primary care practitioners in an effort to prompt early referral for diagnostic assessment and rehabilitation interventions for children suspected of having CP (Specific Objectives 2a, 2b).

12.2 Original Contributions to the Literature and Implications for Practice

In the previous section the results from each study were summarized primarily in relation to each other, contextualized within the sequential knowledge translation approach that this research project employed. In this section the results are collectively situated and explored with respect to the current evidence in the field, and the implications of our findings for clinical practice.

The main clinical challenge that this research project addressed is that early identification of CP by primary care providers is problematic, and that many children with CP are experiencing

important delays in referral for diagnostic assessment and for rehabilitation services. These delays can be detrimental to the child's developmental outcomes and to the psychosocial functioning of the family. Almost half of children with CP are born at term, and/or receive no specialised neonatal follow-up (e.g. preterm survivors who do not meet the gestational age cut-off for the program) and receive well-baby care in their community context. Primary care providers thus have a pivotal role to play in the identification of these children, but aside from their limited training in atypical child development, there has as yet not been a user-friendly approach for primary care providers to adopt to facilitate early detection of CP. Novel strategies were needed to optimize prompt referral by primary care providers, with accessible and feasible tools to enhance early detection and referral strategies, which will ensure early diagnosis and interventions to optimize developmental outcomes and enhance opportunities for neural repair at a younger age.

In order to address this knowledge gap, we created expert-informed content for educational knowledge translation tools to prompt primary care practitioners to recognize the features of CP, and subsequently prompt them to refer children suspected of having CP to a medical specialist for diagnostic assessment, as well offering referral recommendations to co-occur if appropriate. As far as this author is aware, we produced the first scoping review on age at referral for diagnostic assessment of CP (**Manuscript 1**), the first environmental scan of Canadian physician referral practices related to referral for diagnosis and rehabilitation of children suspected of having CP (**Manuscript 3**), and the first expert-informed user-friendly CP-specific prompts for referral for diagnostic assessment and concurrent referral recommendations (**Manuscript 5**).

Early-detection solutions to date have primarily focused on children who are at high-risk for CP (e.g. children who are extremely premature, neonatal intensive care unit survivors with neonatal encephalopathy). These comprise about half of children with CP, and in most institutions,

they receive close neonatal follow-up. It's the other half of children with CP for which this research is primarily concerned with, as the early detection strategies to date are not optimally suited to capture them in the primary care context. For example, Novak et al. (2017) proposed an algorithm for early diagnosis in infants at high-risk for CP, which consists of a combination of neuroimaging studies, standardized general movements assessment, and standardized motor assessments. The high cost and time (i.e. several hours for all three by different testers) to administer what the authors propose is not feasible in the primary care context. For the approximately half of infants with CP who have high-risk indicators identifiable in the newborn period (Novak et al., 2017), these assessments are often completed by trained health professionals as part of the neonatal follow-up program. In primary care, the risk of CP is that of the population at large; about 2 per 1000 live births. Therefore, uniform comprehensive testing, as suggested in the guidelines for higher risk infants is not cost-effective or feasible. Additionally, the widespread adoption of the general movement assessment in the context of the general population is of concern, since its predictive validity in this context requires more rigorous psychometric testing (Darsaklis, Snider, Majnemer, & Mazer, 2011).

The anticipated clinical impacts of research project described in this thesis are three-fold. *Firstly,* primary care providers will have greater knowledge and capability to detect easily recognizable attributes associated with CP early, prompting simultaneous timely referral to medical and rehabilitation specialists. *Secondly,* rehabilitation specialists will be able to initiate therapeutic interventions much earlier at a critical period of brain development, optimizing developmental outcomes. *Finally*, parents and families will be more rapidly informed and better engaged in the process of detection and will benefit from early access to resources and family supports.

The relevance of this research project to the field of rehabilitative science is two-fold. Firstly, it emphasizes the importance of involving people with lived-experience (e.g. parents of children with CP) as collaborators and partners in research, validating their unique perspective as both 'content expert' and 'end-user'. Secondly, it highlights how adopting an integrated knowledge translation approach, where all relevant stakeholders are involved in the project from the start is a good-fit with rehabilitation research, one that warrants a more widespread application.

12.3 Directions for Future Research

The results obtained through the Delphi survey (**Manuscript 5**) will be used in a forthcoming qualitative descriptive study set within the primary care context. Focus groups will be conducted with primary care practitioners (e.g. pediatricians, family physicians) and parents of children with CP to determine how best to translate this knowledge into a user-friendly tool, possibly with multiple formats. A secondary objective could be to identify potential barriers and facilitators to using this new tool in clinical practice. Participants will be presented the 6 clinical features to prompt referral for diagnosis, 2 'warning sign' features to prompt monitoring rather than referral for diagnosis, and 5 referral recommendations to occur simultaneously with referral to a medical specialist for diagnosis. Their opinions will be sought concerning the optimal ways by which to deliver this information (e.g. what format[s] to use, what language and graphics to use, where/how to it make most accessible). The results of these focus groups will be used to inform the initial design and mock-ups of the knowledge translation tool, which will be shared with and discussed in an iterative consultation process with relevant stakeholders in the primary care context until a final version(s) is arrived at.

The Rourke Baby Record (RBR; Li et al., 2019) is an evidence-based health supervision guide for primary healthcare practitioners of children in the first five years of life. The authors of the RBR are currently working on a revision, and they have integrated the six clinical features

that should prompt referral for diagnosis of CP, the two ‘warning sign' features that warrant monitoring rather than immediate referral for diagnosis, and the five referral recommendations to other healthcare professionals that should occur simultaneously with referral for diagnostic assessment into the forthcoming version of the RBR. In Canada, the RBR is the criterion standard for health and developmental surveillance for children 0-5 years of age, and contains guidelines and information for comprehensive well baby/child care including: growth and nutrition monitoring, developmental surveillance, physical examination parameters, immunizations, and anticipatory guidance on safety, family, behaviour and health promotion issues. The RBR has been endorsed by the College of Family Physicians of Canada, the Canadian Paediatric Society, and the Dietitians of Canada. The RBR is available in English and French, and has been adapted to various locales and unique populations (e.g. Nunavut, Alberta First Nations, Ontario, Northwest Territories, and Nova Scotia) (Li et al., 2019). Thus, the considerable reach the RBR has in the context of Canadian primary care surveillance is another format in which to disseminate the knowledge we created through the research projects contained in this thesis. Another encouraging start to the dissemination of the results of this work which is currently ‘in press’ is that we were also recently consulted by a group of clinicians in British Columbia who are developing a diagnostic pathway for CP for their province, and who intend to incorporate our findings into their work.

Additionally, future research endeavors should focus on testing the KT tools to see if they do what they are designed to do (diagnostic accuracy), and on developing and evaluating a KT intervention to implement the new tools in the primary care setting. The latter, for example could include a piloted RCT using feasibility outcome measures (e.g. recruitment, adherence, retention) prior to embarking in a full-scale implementation study such as a cluster RCT.

12.4 Concluding Statement

CP is the fourth leading cause of childhood disability and is the most common physical disability seen in children. Early identification and subsequent early referral to medical specialists for diagnostic assessment and simultaneous referral to rehabilitation specialists for intervention is critical to optimize child and family outcomes. Detection by primary care practitioners is problematic, a concern given that most children with CP are in the care of primary care physicians and would benefit from more stringent surveillance. This doctoral project adopted sequential mixed-methodologies and employed a knowledge translation approach to create the content for educational tools designed to enhance the early detection and referral of children with suspected CP in the primary care context. This body of work presented in this thesis accomplished the following: it identified a knowledge gap in the literature related to age at referral for CP; it documented population-based evidence on current physician referral practices and identified factors associated with delayed referral; it generated the knowledge deemed essential to share with primary care practitioners to enhance their detection efforts; and it informed and validated the generalizability and appropriateness of these results through consultation with a panel of international experts. This resulted in the establishment of the following: six clinical features that should prompt referral for diagnostic assessment; two warning signs that warrant monitoring rather than immediate referral for diagnosis; and five referral recommendations to other healthcare professionals to occur simultaneously with referral for diagnosis. Through collaboration with relevant stakeholders, this knowledge will be tailored to the primary care context and disseminated in whatever format(s) are determined to be optimal to increase awareness and enhance early detection in the primary care context of children suspected of having CP.

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APPENDIX A

The PROMPT Group:
Co-investigators on Canadian Institutes of Health Research Operating Grant MOP-133402
"**PROMPT** Identification of Cerebral Palsy:
Primary-care **R**eferral **O**f **M**otor-impaired children: **P**hysician **T**ools"

Stakeholders: Content-experts, Knowledge-users

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- **Emmanuelle Dagenais, Parent of a child with CP** – Montreal, Quebec, Canada
- **Lynn Dagenais, PT** – CHU Sainte-Justine, Montreal, Quebec, Canada
- **Vasiliki (Betty) Darsaklis, OT** – Shriners Hospital for Children, Montreal, Quebec, Canada
- **Denis Leduc, MD** – Melville Paediatric Clinic, Westmount, Quebec, Canada
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APPENDIX B

Hubermann et al. (2019) Consent

Reply all | Delete Junk | •••

Hubermann et al., (2016) J. Child Neurol.

LH Lara Hubermann <lara.hubermann@gmail.com> Reply all |
Today, 6:27 PM
Zachary Boychuck, Mr

Inbox

Dear Zachary,

With regard to the publication cited below, I am writing to confirm that I have not previously included this article in a manuscript-based thesis submission. I hereby grant permission to include the article in your manuscript-based PhD dissertation submission.

Hubermann, L., Boychuck, Z., Shevell, M., & Majnemer, A. (2016). Age at referral of children for initial diagnosis of cerebral palsy and rehabilitation: Current practices. Journal of Child Neurology, 31(3), 364-9. DOI:10.1177/0883073815596610

Best regards,

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