Protocol for a Scoping Review and Qualitative Descriptive Study to Develop a Framework for Youth- and Family-Specific Engagement in Research

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Abstract

Background: Youth and families play an indispensable role in health research, given their unique lived experiences and expertise. Aligning research with patients' needs, values, and preferences can significantly enhance its relevance and impact;

however, recent research has highlighted various challenges and risks associated with youth and family engagement in health research. These challenges encompass the perils of tokenism, power imbalances and dynamics, questioning the motives behind engagement, limited accessibility to patient-friendly training for patient partners, as well as inadequate training on patient engagement for researchers, and the absence of equitable engagement tools. To address these risks and challenges, different patient engagement models, theories, frameworks, and guiding principles have been developed and adopted; to date, however, their transferability to youth- and family-specific engagement in research has been limited.

Objective: The objectives of this project are to 1) to determine the extent of the literature on the application of patient engagement models, theories, frameworks, and guiding principles in the context of youth-specific research; and 2) to determine how meaningful the key components and constructs of these models, theories, frameworks, and guiding principles are to youth and their family members.

Methods: This project will utilize an integrated knowledge translation approach and will consist of 2 phases: 1) a scoping review to identify patient engagement models, theories, frameworks and guiding principles in the context of youth-specific research, and 2) a qualitative descriptive study using one-on-one semi-structured interviews with youth and family members to understand their conceptualization of meaningful engagement in health research.

Results: The project will be carried out with funding from the Canadian Institutes of Health Research (CIHR).

Conclusions: The findings from this project will be foundational to the development of a youth- and family-specific engagement in research framework called the UNITE framework, and a subsequent validated measure.

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ABSTRACT

Background: Youth and families play an indispensable role in health research, given their unique lived experiences and expertise. Aligning research with patients' needs, values, and preferences can significantly enhance its relevance and impact; however, recent research has highlighted various challenges and risks associated with youth and family engagement in health research. These challenges encompass the perils of tokenism, power imbalances and dynamics, questioning the motives behind engagement, limited accessibility to patient-friendly training for patient partners, as well as inadequate training on patient engagement for researchers, and the absence of equitable engagement tools. To address these risks and challenges, different patient engagement models, theories, frameworks, and guiding principles have been developed and adopted; to date, however, their transferability to youth- and family-specific engagement in research has been limited.

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Results: The project will be carried out with funding from the Canadian Institutes of Health Research (CIHR).

Conclusions: The findings from this project will be foundational to the development of a youth- and family-specific engagement in research framework called the UNITE framework, and a subsequent validated measure.

Keywords: Youth and family engagement, patient engagement, implementation science, scoping review, qualitative

INTRODUCTION

Patients play an essential role in health research, given their unique lived experiences and expertise, which can significantly enhance the quality, relevance, and impact of research by aligning it with patient needs, values, and preferences [1, 2]. The overarching objective of patient engagement in health research is to generate research that contributes to improved healthcare service delivery, clinical outcomes, and population health [3]. Recent research has highlighted the myriad benefits and positive outcomes associated with patient engagement for patient partners, investigators, and research endeavours [4-8]. For example, engaging patients has influenced initial research priorities, study designs, interpretation of findings, healthcare interventions, and knowledge mobilization strategies, resulting in outcomes that more closely align with patient perspectives [7, 9]. When patient engagement is characterized as meaningful and authentic, it provides a rewarding experience for patients and researchers alike [10].

Other recent research, however, has highlighted various challenges and risks associated with patient engagement in health research. These challenges encompass tokenism [11-14], power imbalances/dynamics [12, 15], limited accessibility to patient-friendly training for patient partners [16], and the absence of equitable engagement tools [17]. These challenges can have lasting effects on patient partners, including mental and physical exhaustion, deteriorating health, doubts about the value of engagement, and a sense of personal failure, both to the team and the broader patient community [18]. A recent article by patient partners led by Richards et al. (2023) [18] on how patient engagement can falter, highlighted key themes including: “patient partners as a checkmark”, “unconscious bias towards patient partners”, “lack of support to fully include patient partners”, and “lack of recognizing the vulnerability of patient partners”[18].

To address these challenges and risks, various patient engagement models, theories, frameworks, and guiding principles (e.g., Strategy for Patient-Oriented Research (SPOR), Patient Engagement in Research (PEIR), Ways Community Members can Participate in the Stages of Research from Ontario Brain Institute) have been developed and adopted within the research community [10, 19, 20]. For example, the Patient Engagement in Research (PEIR) framework [10] includes eight key components, which collectively contribute to meaningful patient engagement in research: 1) procedural requirements, 2) convenience, 3) contributions, 4) support, 5) team interaction, 6) research environment, 7) feeling valued, and 8) benefits. A subsequent measure of meaningful patient engagement in research from the patient perspective has been developed (PEIRS-22) [21, 22].

This measure is designed to be completed by adult patients and family caregivers who partner with researchers on projects.

It is important to acknowledge that existing frameworks often predominantly emphasize the benefits of patient partners to the research project and team, sometimes overlooking the reciprocal benefits that may occur between the research team and patient partners, especially youth partners [23]. This skewed approach fails to fully consider the potentially extractive nature of research collaboration, as aptly described by Metz [23] where the research process can unintentionally exploit the knowledge and contributions of youth without adequately reciprocating in terms of personal and professional development opportunities. This issue takes on particular significance when crafting an engagement framework tailored for youth, as research teams have the unique potential to foster skill development, positively impact their career trajectories, and contribute to their holistic growth [23].

Despite the significant contribution of the PEIR framework [10] and the PEIRS-22 [21, 22], it lacks the incorporation of a comprehensive review of the evidence on existing frameworks, and its development was based on participants with limited diversity in terms of gender, race, education, primary diagnosis (all had arthritis), and age[10, 21, 22]. These limitations impede the transferability of this framework and measure to youth- and family-specific engagement in health research.

Furthermore, there is a model of engagement specifically designed for use with youth called the McCain Model of Youth Engagement [24]. However, it was developed solely in the context of youth mental health systems research for youth and young adults 15-29 years of age [24], limiting transferability (i.e., across different contexts and family members), hence highlighting the need to develop a broader framework (e.g., to be used in healthy youth). Accordingly, our research seeks to directly address these specific gaps in existing engagement models, theories, and frameworks by developing a yo**u**th a**n**d fam**i**ly-specific engagemen**t** in r**e**search (UNITE) framework. Future work will involve the development of a validated measure.

Research Objectives: This proposal has two objectives: 1) to determine the extent of literature on the application of patient engagement models, theories, frameworks, and guiding principles in the context of youth- and family-specific research; and 2) to understand how meaningful the key components and constructs of these models, theories, frameworks, and guiding principles are to youth and their family members. Collectively, these findings will be foundational to the development of the UNITE framework and a subsequent validated measure.

METHODS

Study Design: This study will be conducted in two phases, with Phase 1 consisting of a scoping review and Phase 2 consisting of a qualitative descriptive study.

Integrated Knowledge Translation (iKT) Approach: Integrated knowledge translation (iKT) is defined as a collaborative relationship between researchers and relevant knowledge users as partners that facilitate mutually beneficial decision-making related to a study or research program [25]. Six youths and four family members will be recruited to be part of the iKT panels. Youth will be recruited from previous projects in youth engagement [26] and the Bloorview Research Institute (BRI). Family members will be recruited from the BRI. The youth representative will lead the iKT Panels. The lead youth representative and research team decided to hold separate iKT panels (i.e., having a youth panel and a family panel with the opportunity to mix panels when needed). This is to ensure both groups feel comfortable and can freely express their unique perspectives. Activities of the panels may include planning study activities, participant recruitment, data collection and analyses, and knowledge mobilization. Panel discussions will be conducted in a manner that respects diverse perspectives and experiences. The "SPOREA Reflective Equity, Diversity and Inclusion (EDI) Exercise" will be conducted with iKT panel members to encourage dialogue and understanding around EDI [27].

Phase 1: Scoping Review on Patient Engagement Models, Theories, Frameworks, Guidance in Youth Health Research

The methodology for the scoping review will follow the methodological frameworks of the Joanna Briggs Institute (JBI) [28] and Khalil *et al* [29]. The scoping review protocol will be registered on the Open Science Framework Register (https://osf.io/), and be guided by and reported according to the Preferred Reporting Items for Systematic Review and Meta-Analysis Protocols (PRISMA-P) [30]. The results of the scoping review will be reported using the Preferred Reporting Items for Systematic Reviews and Meta-Analysis extension for Scoping Reviews (PRISMA-ScR) checklist [31].

Stage 1: Developing a Search Strategy*:* The Population, Concept, and Context (PCC) framework will be employed to guide the search strategy (Population: World Health Organization definition of "young people", ages 10-24 years [32]; Concept: patient engagement in research; Context: models, theories, frameworks, guiding principles). With the assistance of a research librarian, literature search strategies using medical subject headings and text words related to patient engagement and models, theories, frameworks, and guidance/guiding principles will be developed. The final search strategy will combine structure database-specific subject headings (as available) and keywords/synonyms. An Information Specialist with expertise in conducting searches for systematic and scoping reviews has drafted an initial search strategy using OVID Medline (Appendix A- Medline Search Strategy) and will work with the research team and iKT Panel to refine and finalize the search. The final search strategy will also undergo peer review using the Peer Review of Electronic Search Strategies (PRESS) Statement checklist with another librarian/information specialist [33].

The following databases will be searched: Medline, CINAHL, EMBASE, PsycInfo and Cochrane Central Register of Controlled Trials. Searches will be limited to English. Literature from 2013 to present will be captured, consistent with when the term patient engagement became frequently used [34]. We will also search the grey literature in specialized databases like OpenGrey, Grey Literature Report, and GreyNet International, platforms like arXiv, bioRxiv, and SSRN and databases such as ProQuest Dissertations and Theses.

Stage 2: Evidence Screening and Selection**:** All primary studies using a patient engagement in research model, theory, framework, or guiding principles among young people will be eligible for inclusion. We define young people as ages 10-24 years, consistent with the WHO definition [32]. We define a model as the essential elements or variables of a phenomenon or a specific aspect of a phenomenon; a theory as “a set of analytical principles or statements designed to structure our observation, understanding and explanation of the world” (pg.2); and a framework as an explanation of a phenomenon by organizing it into a collection of descriptive categories and the relationships between them [35]. Systematic reviews, meta-analyses, editorials, commentaries and non-specific conference proceedings will be excluded to focus on including primary results and not preliminary findings or ongoing research; however, the reference lists of such articles will be hand-searched for relevant articles.

Removal of duplicates as well as level 1 and level 2 screening will be managed through Covidence. To increase reliability, the Level 1 screening form will be piloted on a random sample of approximately 50 articles. Eligibility criteria descriptions will be revised if deemed necessary by the team or if low agreement (i.e., < 70%)[36] is observed, to improve the consistent application of the selection criteria**.** Agreement will be measured using Cohen's Kappa [37]. A pilot test of the level 2 screening will also be performed on approximately 25% of the articles, similar to the process for level 1 screening. For studies that are excluded at level 2, the reason for exclusion will be recorded. All screening will occur in duplicate and independently. When necessary, another reviewer will be sought to resolve conflicts.

Stage 3: Data Extraction*:* A standardized data extraction form will be developed by the research team and iKT panels, based on the JBI manual data extraction recommendations [28] and those recommended for the extraction, analysis, and presentation of results in scoping reviews [28, 38]. Extracted data will include *study characteristics* (e.g., study design, year of publication, geographic location)*, youth and family participant characteristics* (e.g., age of youth), and *details of the engagement models, theories, frameworks, and guiding principles* (e.g., components or values/principles and how they were enabled/enacted in the study), as well as study results. Additional categories for data extraction identified through discussions with the research team and iKT Panels will be added to the final data extract template as applicable. The data extraction template will be piloted for two to three articles to ensure all relevant results are extracted. All data will then be extracted in duplicate by two independent reviewers. Discrepancies in the extracted data will be discussed and resolved by the two reviewers. Quality/risk of bias will not be assessed, as this is not required in scoping reviews [31].

Stage 4: Data Analysis*:* The extracted data will be quantitatively summarized using numerical counts and qualitatively using content analysis [39]. The data will be grouped by the main components of the model, theories, frameworks, guiding principles (and how they were enabled/enacted), study designs and associated methods (e.g., one-on-one interviews). We will also synthesize data on how youth and family members were engaged throughout the research process, the types of outcomes collected, and results. Depending on the available data, sub-group analyses may be conducted by health condition, sex, gender-related variables, as well as other PROGRESS-Plus characteristics (e.g., race/ethnicity/culture/language, education) [40]. If feasible, we will contact the study authors of the included studies to confirm that all the data collected were included in the published article (i.e., and not excluded due to word count limitations of a journal).

Phase 2: Qualitative Study to Understand Key Components/Constructs for Development of a Framework for Youth- and Family-Specific Engagement in Research

***Study Design**:* Phase 2 will adopt a qualitative descriptive approach [41, 42]. Findings from the scoping review will inform the development of the interview guide for Phase 2, such as identifying what aspects of the included adult frameworks resonate with youth and family members, and which do not.

***Recruitment**:* We will recruit Canadian youth (ages 10-24) and their family members who have had experience in youth engagement in health research. Participants will be recruited via our professional networks including those of the iKT panel members (e.g., Kids Brain Health Network, Bloorview Research Institute Family Engagement Office, SickKids Patient and Family Engagement Office), social media pages, and email lists. We will also enlist the help of a diverse group of youth advisors who have participated in our research team's previous research [26, 43, 44].

***Data Collection**:* Youth and family participants will take part in separate, one-on-one, semi-structured telephone or online interviews with a member of the research team with expertise in qualitative research methods [45, 46]. All interviews will be digitally recorded and transcribed verbatim for data analysis. The interview guides will consist of questions focused on youth and family members' conceptualization of meaningful engagement in health research. The interview guide will be pilot-tested with various members of the research team and iKT Panel. We will use probes or recursive questioning during interviews to explore issues in greater depth and verify the interviewer's understanding of the collected information [47, 48].

***Data Analysis**:* We will use inductive thematic analysis, as described by Braun and Clark [49, 50] and consistent with the pragmatic orientation of this research [51]. Codes and themes will be refined through discussion with the larger research team. The software program NVivo version 14 will be used during the analysis of the transcripts to help organize the codes. Multiple aspects of trustworthiness will be utilised [52]. For example, we will demonstrate credibility via peer debriefing with various members of the research team and the iKT panel. Transferability will be accomplished by describing the study samples. Independent analysts will review the data and contest the themes to ensure dependability. Finally, confirmability will be accomplished by providing decision trails between data and interpretation[52].

Ethical Considerations: Ethics approval for Phase 2 of this project will be obtained from the first author's primary institution (Bloorview Research Institute, Holland Bloorview Kids Rehabilitation Hospital).

RESULTS

This work is supported by the Canadian Institutes of Health Research (CIHR) Healthy Youth Catalyst Grant. This study will take place between March 2024 and March 2025.

DISCUSSION

Expected Findings

The findings from this study will allow us to identify what is currently known about the application of patient engagement models, theories, frameworks and guiding principles (that are often designed for adults) in the context of youth-specific research, and to understand the importance of the components that make up these models, theories and frameworks from the perspective of youth and their families. This project lays the foundational work for developing a patient engagement framework called the UNITE framework, which will include equity, diversity and inclusion considerations, and the subsequent development and validation of an associated measure of engagement. The UNITE framework and associated measure will contribute to meaningful and sustained engagement of youth and their families in health research.

Strengths and Limitations

A strength of this study is the inclusion of grey literature (e.g., reports, policy literature) for Phase 1 (scoping review). This will provide a more comprehensive understanding of the concept (patient engagement in research) and context (models, theories, frameworks and guiding principles) that we are interested in, as well as mitigate publication bias [53]. In addition, the search strategy for Phase 1 will also be reviewed by the iKT panel and undergo peer review using the PRESS Statement checklist [33], which further strengthens the relevance, comprehensiveness, and quality of the search strategy. One limitation of this project is the potential for selection bias (specifically for Phase 2), where individuals who had either a very positive or very negative experiences with patient engagement in research may be more likely to participate in the study, which may limit the applicability of the study's findings. However, the adoption of the iKT approach and having the iKT panel (consisting of youth and their families) to assist in the recruitment for Phase 2 of the study, should result in the recruitment of a variety of individuals, with diverse interests.

Future Directions

We will use a variety of passive and active end-of-grant knowledge translation/mobilization approaches to disseminate our findings, which will be co-developed with our iKT Panel. Traditional knowledge translation will include dissemination through meetings locally, nationally and internationally, (e.g., PxP, For Patients, By Patients) and publications in peer-reviewed journals. Members of the research team are affiliated with and situated within pediatric institutions where the UNITE framework will be disseminated and implemented. Members of the research team will also discuss and distribute the UNITE framework within its expansive training curriculum and a planned youth-focused engagement in research course. Future research will also involve the implementation of the UNITE measure in a learning health system context. We will ensure youth and family voices are heard to develop a flexible communication plan that will suit diverse needs. We will co-develop plain language summaries with youth and family partners with clear, simple, and individualized messages for patient/family and community service organizations to augment the accessibility of the information.

Conclusions

The current proposal will lead to the development of a youth- and family-specific engagement in research framework, UNITE, with future research focused on the development of an associated, validated measure. The UNITE framework and measure will lay the foundation for meaningful and sustained engagement in health research by youth and their families, ultimately contributing to enhanced healthcare service delivery, improved clinical outcomes, and increased overall well-being and quality of life for youth and their families [1-3].

Conflicts of Interest

SM is the Editor-in-Chief of *JMIR Rehabilitation and Assistive Technologies*

ABBREVIATIONS

CIHR	Canadian Institutes of Health Research
iKT	Integrated Knowledge Translation
PEIR	Patient Engagement in Research
PEIRS-22	Patient Engagement in Research Scale-22

PRESS	Peer Review of Electronic Search Strategies
PRISMA-P	Preferred Reporting Items for Systematic Review and Meta-Analysis Protocols
PRISMA- ScR	Preferred Reporting Items for Systematic Reviews and Meta-Analysis Extension for Scoping Reviews
UNITE	Yo**u**th a**n**d Fam**i**ly-Specific Engagemen**t** in R**e**search

REFERENCES

1. Hayes H, Buckland S, Tarpey M. Briefing notes for researchers: public involvement in NHS, public health and social care research. INVOLVE: Eastleigh 2012.
2. Frank L, Forsythe L, Ellis L, Schrandt S, Sheridan S, Gerson J, et al. Conceptual and practical foundations of patient engagement in research at the patient-centered outcomes research institute. Qual Life Res. 2015 May;24(5):1033-41. PMID: 25560774. doi: 10.1007/s11136-014-0893-3.
3. Manafo E, Petermann L, Mason-Lai P, Vandall-Walker V. Patient engagement in Canada: a scoping review of the 'how' and 'what' of patient engagement in health research. Health Res Policy Syst. 2018 Feb 7;16(1):5. PMID: 29415734. doi: 10.1186/s12961-018-0282-4.
4. Ashcroft J, Wykes T, Taylor J, Crowther A, Szmukler G. Impact on the individual: what do patients and carers gain, lose and expect from being involved in research? J Ment Health. 2016;25(1):28-35. PMID: 26733079. doi: 10.3109/09638237.2015.1101424.
5. Staley K, Abbey-Vital I, Nolan C. The impact of involvement on researchers: a learning experience. Res Involv Engagem. 2017;3:20. PMID: 29062545. doi: 10.1186/s40900-017-0071-1.
6. Geissler J, Ryll B, di Priolo SL, Uhlenhopp M. Improving Patient Involvement in Medicines Research and Development:: A Practical Roadmap. Ther Innov Regul Sci. 2017 Sep;51(5):612-9. PMID: 30231692. doi: 10.1177/2168479017706405.
7. Gordon L, Dickinson A, Offredy M, Smiddy J. A research note on the benefit of patient and public involvement in research: The experience of prostate cancer patients regarding information in radiotherapy. Radiography (Lond). 2017 May;23(2):167-70. PMID: 28390550. doi: 10.1016/j.radi.2017.02.004.
8. Vanderhout SM, Bhalla M, Van A, Fergusson DA, Potter BK, Karoly A, et al. The Impact of Patient and Family Engagement in Child Health Research: A Scoping Review. The Journal of Pediatrics. 2023;253:115-28. doi: 10.1016/j.jpeds.2022.09.030.
9. Archambault PM, McGavin C, Dainty KN, McLeod SL, Vaillancourt C, Lee JS, et al. Recommendations for patient engagement in patient-oriented emergency medicine research. Cjem. 2018 May;20(3):435-42. PMID: 29690943. doi: 10.1017/cem.2018.370.
10. Hamilton CB, Hoens AM, Backman CL, McKinnon AM, McQuitty S, English K, et al. An empirically based conceptual framework for fostering meaningful patient engagement in research. Health Expect. 2018 Feb;21(1):396-406. PMID: 28984405. doi: 10.1111/hex.12635.

11. Richards Dawn DEFTHLNGTC. Tokenism: Seeing it. Fixing it. Perspectives from IMHA Patient Partners | IMHA Blog. 2021; Available from: https://blogs.ubc.ca/imhablog/2021/10/13/tokenism-seeing-it-fixing-it-perspectives-from-patient-partners/.

12. Ethics Guidance for Developing Partnerships with Patients and Researchers - Canadian Institutes of Health Research. 2019 [updated 2020-04-17]; Available from: https://cihr-irsc.gc.ca/e/51910.html.

13. Hahn DL, Hoffmann AE, Felzien M, LeMaster JW, Xu J, Fagnan LJ. Tokenism in patient engagement. Fam Pract. 2017 Jun 1;34(3):290-5. PMID: 27660557. doi: 10.1093/fampra/cmw097.

14. Johannesen J. Two-way street? A critical look at patient and family engagement | by Jennifer Johannesen. 2017.

15. Jennifer J. "The trouble with patient and public involvement (PPI)" - keynote at Cochrane Colloquium 2018 | by Jennifer Johannesen. @jenjohannesen; 2018.

16. Forsythe LP, Ellis LE, Edmundson L, Sabharwal R, Rein A, Konopka K, et al. Patient and Stakeholder Engagement in the PCORI Pilot Projects: Description and Lessons Learned. J Gen Intern Med. 2016 Jan;31(1):13-21. PMID: 26160480. doi: 10.1007/s11606-015-3450-z.

17. Forsythe LP, Carman KL, Szydlowski V, Fayish L, Davidson L, Hickam DH, et al. Patient Engagement In Research: Early Findings From The Patient-Centered Outcomes Research Institute. Health Aff (Millwood). 2019 Mar;38(3):359-67. PMID: 30830822. doi: 10.1377/hlthaff.2018.05067.

18. Richards DP, Poirier S, Mohabir V, Proulx L, Robins S, Smith J. Reflections on patient engagement by patient partners: how it can go wrong. Res Involv Engagem. 2023 Jun 12;9(1):41. PMID: 37308922. doi: 10.1186/s40900-023-00454-1.

19. Research CIoH. Strategy for Patient-Oriented Research - Patient Engagement Framework - CIHR. 2014 [updated 2014-07-02]; Available from: https://cihr-irsc.gc.ca/e/48413.html.

20. Ontario Brain Institute. Ways community members can participate in the stages of research. 2024.

21. Hamilton CB, Hoens AM, McQuitty S, McKinnon AM, English K, Backman CL, et al. Development and pre-testing of the Patient Engagement In Research Scale (PEIRS) to assess the quality of engagement from a patient perspective. PLoS One. 2018;13(11):e0206588. PMID: 30383823. doi: 10.1371/journal.pone.0206588.

22. Hamilton CB, Hoens AM, McKinnon AM, McQuitty S, English K, Hawke LD, et al. Shortening and validation of the Patient Engagement In Research Scale (PEIRS) for measuring meaningful patient and family caregiver engagement. Health Expect. 2021 Jun;24(3):863-79. PMID: 33729634. doi: 10.1111/hex.13227.

23. Metz A, Damschroder L. "There is a deep disconnect" - Laura and Allison talk about practical implementation. Essential Implementation2023.

24. Heffernan OS, Herzog TM, Schiralli JE, Hawke LD, Chaim G, Henderson JL. Implementation of a youth-adult partnership model in youth mental health systems research: Challenges and successes. Health Expectations. 2017;20(6):1183-8. doi: https://doi.org/10.1111/hex.12554.

25. Kothari A, McCutcheon C, Graham ID. Defining Integrated Knowledge

Translation and Moving Forward: A Response to Recent Commentaries. Int J Health Policy Manag. 2017 May 1;6(5):299-300. PMID: 28812820. doi: 10.15171/ijhpm.2017.15.
26. Bailey K, Allemang B, Vandermorris A, Munce S, Cleverley K, Chisolm C, et al. Benefits, Barriers and Recommendations for Youth Engagement in Health Research: Combining Evidence-Based and Youth Perspectives (Accepted). Res Involv Engagem. 2024.
27. Alliance SE. Reflective exercise. 2021.
28. Peters MDJ, Marnie C, Tricco AC, Pollock D, Munn Z, Alexander L, et al. Updated methodological guidance for the conduct of scoping reviews. JBI Evidence Synthesis. 2020;18(10).
29. Khalil H, Peters MD, Tricco AC, Pollock D, Alexander L, McInerney P, et al. Conducting high quality scoping reviews-challenges and solutions. J Clin Epidemiol. 2021 Feb;130:156-60. PMID: 33122034. doi: 10.1016/j.jclinepi.2020.10.009.
30. Moher D, Shamseer L, Clarke M, Ghersi D, Liberati A, Petticrew M, et al. Preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P) 2015 statement. Syst Rev. 2015 Jan 1;4(1):1. PMID: 25554246. doi: 10.1186/2046-4053-4-1.
31. Tricco AC, Lillie E, Zarin W, O'Brien KK, Colquhoun H, Levac D, et al. PRISMA Extension for Scoping Reviews (PRISMA-ScR): Checklist and Explanation. Ann Intern Med. 2018 Oct 2;169(7):467-73. PMID: 30178033. doi: 10.7326/m18-0850.
32. (WHO) WHO. Adolescent Health. Available from: https://www.who.int/health-topics/adolescent-health#tab=tab_1.
33. McGowan J, Sampson M, Salzwedel DM, Cogo E, Foerster V, Lefebvre C. PRESS Peer Review of Electronic Search Strategies: 2015 Guideline Statement. J Clin Epidemiol. 2016 Jul;75:40-6. PMID: 27005575. doi: 10.1016/j.jclinepi.2016.01.021.
34. Higgins T, Larson E, Schnall R. Unraveling the meaning of patient engagement: A concept analysis. Patient Education and Counseling. 2017 2017/01/01/;100(1):30-6. doi: https://doi.org/10.1016/j.pec.2016.09.002.
35. Nilsen P, Birken SA. Overview of theories, models and frameworks in implementation science. Handbook on Implementation Science. Cheltenham, UK: Edward Elgar Publishing; 2020.
36. Park CU, Kim HJ. Measurement of Inter-Rater Reliability in Systematic Review. hmr. 2015 02;35(1):44-9. doi: 10.7599/hmr.2015.35.1.44.
37. Cohen J. A coefficient of agreement for nominal scales. Educational and Psychological Measurement. 1960;20:37-46. doi: 10.1177/001316446002000104.
38. MDJ P, C G, P M, Z M, AC T, Khalil H. Chapter 11: Scoping reviews (2020 version). In: E A, Z M, editors. JBI Manual for Evidence Synthesis2020.
39. Stemler S. An Overview of Content Analysis. Practical Assessment, Research, and Evaluation. 2001 06/07;7:http://pareonline.net/getvn.asp?v=7&n=17.
40. O'Neill J, Tabish H, Welch V, Petticrew M, Pottie K, Clarke M, et al. Applying an equity lens to interventions: using PROGRESS ensures consideration of socially stratifying factors to illuminate inequities in health. J Clin Epidemiol. 2014 Jan;67(1):56-64. PMID: 24189091. doi: 10.1016/j.jclinepi.2013.08.005.

41. Sandelowski M. Whatever happened to qualitative description? Res Nurs Health. 2000 Aug;23(4):334-40. PMID: 10940958. doi: 10.1002/1098-240x(200008)23:4<334::aid-nur9>3.0.co;2-g.
42. Sandelowski M. What's in a name? Qualitative description revisited. Res Nurs Health. 2010 Feb;33(1):77-84. PMID: 20014004. doi: 10.1002/nur.20362.
43. Bailey K, Avolio J, Lo L, Gajaria A, Mooney S, Greer K, et al. Social and Structural Drivers of Health and Transition to Adult Care. Pediatrics. 2024 Jan 1;153(1). PMID: 38084099. doi: 10.1542/peds.2023-062275.
44. Munce SEP, Wong E, Luong D, Rao J, Cunningham J, Bailey K, et al. Patient, caregiver and other knowledge user engagement in consensus-building healthcare initiatives: a scoping review protocol. BMJ Open. 2024;14(5):e080822. doi: 10.1136/bmjopen-2023-080822.
45. Archibald MM, Ambagtsheer RC, Casey MG, Lawless M. Using Zoom Videoconferencing for Qualitative Data Collection: Perceptions and Experiences of Researchers and Participants. International Journal of Qualitative Methods. 2019;18:1609406919874596. doi: 10.1177/1609406919874596.
46. M.Q P. Qualitative research and evaluation methods (3rd ed.). Thousand Oaks, CA: SAGE Publications; 2002.
47. Tashakkori A, Teddlie C. SAGE Handbook of Mixed Methods in Social & Behavioral Research. Thousand Oaks, California2010. Available from: https://methods.sagepub.com/book/sage-handbook-of-mixed-methods-social-behavioral-research-2e.
48. Clarke V, Braun V, Hayfield N. Thematic Analysis. In: Smith JA, editor. Qualitative Psychology: A Practical Guide to Research Methods. London: SAGE Publications; 2015.
49. Braun V, Clarke V. Reflecting on reflexive thematic analysis. Qualitative Research in Sport, Exercise and Health. 2019 2019/08/08;11(4):589-97. doi: 10.1080/2159676X.2019.1628806.
50. Guba EG, Lincoln YS. Effective evaluation: Improving the usefulness of evaluation results through responsive and naturalistic approaches. San Francisco, CA, US: Jossey-Bass; 1981. xxv, 423-xxv, p. ISBN: 0-87589-493-3 (Hardcover); 1-55542-442-2 (Paperback).
51. Allemang B, Sitter K, Dimitropoulos G. Pragmatism as a paradigm for patient-oriented research. Health Expectations. 2022 2022/02/01;25(1):38-47. doi: https://doi.org/10.1111/hex.13384.
52. Research CIoH. Guide to Knowledge Translation Planning at CIHR: Integrated and End-of-Grant Approaches - CIHR. 2012 [updated 2012-12-06]; Available from: https://cihr-irsc.gc.ca/e/45321.html#a3.
53. Paez A. Gray literature: An important resource in systematic reviews. Journal of Evidence-Based Medicine. 2017;10(3):233-40. doi: https://doi.org/10.1111/jebm.12266.

Supplementary Files

Multimedia Appendixes

Medline search strategy.
URL: http://asset.jmir.pub/assets/ebf565bc901a8e34bc8d2e44cd7b332e.pdf

Peer-review reports.
URL: http://asset.jmir.pub/assets/6f9117d4e870468581a421b2888785a8.pdf