A priority setting partnership to identify research priorities for expectant families and those with children to age 24 months: The Family Research Agenda Initiative Setting (FRAISE) project.

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Title: A priority setting partnership to identify research priorities for expectant families and those with children to age 24 months: The Family Research Agenda Initiative Setting (FRAISE) project.

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Abstract

Objective: The objective of this study was to identify the top 10 research priorities of expectant parents and caregivers of children up to age 24 months.

Design: An iterative, priority setting partnership using a modified James Lind Alliance approach was implemented in three phases. In Phase One, a steering committee of parents, clinicians, community agency representatives, and researchers was formed and established 12 categories of interest. In Phase Two, through in-person collaboration with steering committee members, an electronic survey to assess research priorities across 12 categories was developed. In Phase Three, the steering committee used several online and in-person consensus building workshops to establish the top 34 questions, which were circulated to the broader steering committee via online survey. Finally, the steering committee met in-person to determine and rank a top-11 list of parent-identified research priorities. Selecting 11 priorities as opposed to 10 priorities was the steering committee consensus.

Setting: This study was conducted in a community setting in a province in Western Canada.

Participants: Expectant parents and caregivers of children up to age 24 months. In total 596 participants consented with 480 completed surveys.

Results: Survey results providing 3232 responses, with 202 unique priorities. The top 10 (plus 1) research priorities included questions about: 1) developing healthy coping strategies and emotion regulation, 2) managing unexpected outcomes during pregnancy and labour/delivery, 3) prevention and treatment access for mental health concerns, 4) navigating health information, 5) creating multi-level supports for healthy relationships, 6) prevention and treatment of eczema and allergies, 7) managing developmentally appropriate risk taking, 8) culturally appropriate sleep strategies, 9) infant feeding, 10) supporting healthy child development, and 11) vaccine hesitancy.

Conclusions: The findings will direct future maternal-child research and health-promoting interventions, ensuring they are rooted in parent-identified priorities that represent contemporary needs.

Keywords: Parent, Priority Setting, Research Priorities, James Lind Alliance, Patient Engagement, Patient-Oriented Research, Participant Involvement.

Funding: This work was supported by The Alberta Children's Hospital Research Institute and Alberta Innovates.

Article Summary

Strengths and Limitations of This Study

This study had a large sample size (n = 480) for the online survey, comparable to other priority setting partnerships. Researchers also created space for broad representation of stakeholders including clinicians, community agency representatives, parents, and ethnically diverse representatives of the study location.

Limitations included the inherent and individual biases that self-selected, voluntary researchers and steering community members view the world with, which may reduce generalizability of the final priority list. Additionally, the research team was composed entirely of white heterosexual identifying women. Although attention was paid to ensure participation of fathers, individuals self-identifying as non-white, and those in non-heterosexual partnerships or single parents, these voices may not have been entirely represented in the final priorities.

- We engaged a core steering committee of 15 parents, four clinicians and two community agency representatives and a larger online group of steering committee members throughout the priority setting process.
- Together with the steering committee, we used a modified James Lind Alliance priority setting partnership approach.
- Four in-person priority setting workshops were hosted to build consensus around survey development, survey response theming, the top 30 list, and the final top 10 (plus one) list.
- We co-developed an online survey with the steering committee, using open-ended survey questions to elicit potential research questions from families of children up to 24 months of age.
- Each workshop was structured using nominal group technique to develop consensus around coding, theming, and development of the research priorities.

Introduction

Due to the critical and rapid development that occurs within the first three years of life, investing in early childhood has an estimated return of 800%.[1] Parents of young children have the greatest potential to optimize social and environmental conditions to foster optimal child health outcomes.[2] High parental expectations, intensive parenting, and increased access to information have drastically changed parenting experiences over the past 10-15 years.[3] The dynamic nature of the contemporary maternal-child and parenting landscape may be inadequately incorporated into existing research priorities and questions. Further, researchers and clinicians often encounter difficulties effectively translating and implementing research on child health and development.[4] As the knowledge base for supporting healthy developmental outcomes grows in breadth and scope, effectively prioritizing research investment is crucial to maximize impact and minimize research waste.[5]

Understanding health research priorities of knowledge users, such as parents, clinicians, and community agency representatives, is vital to conducting research that is more likely to contribute to meaningful changes in health outcomes.[6] Evidence generated without the consideration of the knowledge user will have minimal effect on clinical practice and reduce meaningful outcomes.[7] Further, there is growing awareness of the importance of adopting an intersectional lens when conducting health research to adequately address culturally and socio-politically appropriate parenting outcomes.[8] Employing an intersectional lens by centering the perspectives and meaning of parents, patients, or families will result in evidence and resultant policies that account for the diverse needs of patients and knowledge users.[9] Engaging knowledge users in research priority setting initiatives is recognized as an effective and ethical means of prioritizing the allocation of limited public research funds.[7] Not only does priority setting work promote researcher accountability, this integrative approach to knowledge translation may also reduce lag time between producing and implementing knowledge,[10] while contributing to the development of culturally and socially meaningful outcomes. Parental involvement in research may enhance engagement with, and uptake of, interventions and services that support early childhood health, resulting in more timely and effective care.

Priority setting partnerships

Patient-oriented research, specifically priority setting partnerships (PSPs), are increasingly identified as an effective method to decrease research waste and develop research outcomes meaningful to knowledge users, parents, and care providers.[7] PSPs bring together clinicians, researchers, and patients to identify and prioritize research uncertainties using a systematic and collaborative process.

Involving parents and caregivers in foundational development of research through PSPs is a feasible method to produce meaningful outcomes – by creating knowledge important to parents and providers, the likelihood of new evidence uptake and shifting healthcare practices is increased. To date, there have been no PSPs focused on identifying community-based research priorities for families of well children from conception to age 24 months. When purposefully employing a participatory action framework, PSPs can use intersectional principles by capturing perspectives and experiences of traditionally neglected populations.[8,9] Therefore, the objective of this study was to engage a diverse group of perspectives in identifying the top 10 research priorities of expectant parents and caregivers of children up to age 24 months.

Methods

The Family Research Agenda Initiative Setting (FRAISE) project used a modified James Lind Alliance (JLA)[11] approach to identify research priorities of parents, clinicians, and community agency representatives within a province in Western Canada.[12] Many PSPs use the JLA approach;[6] however, this methodology is resource intensive and may be impractical for lower resourced research studies and vulnerable populations. As such, a modified approach to the JLA method has emerged as a feasible alternative.[13] FRAISE applied a consensus-building and strengths-based approach, modelled after participatory action research and the JLA. James Lind Alliance PSPs bring together patients, caregivers, and clinicians, which requires sensitivity to varying participant capacities, ongoing effective communication, transparency in decision-making, and inclusivity of all views.[11] The modified JLA process utilized in the FRAISE project involved a series of iterative phases: (A) formation of a steering committee, including parents, clinicians and community agency representatives; (B) online survey development; (C) delivering the online survey to gather research uncertainties or questions from parents, clinicians and community agency representatives; (D) categorizing and/or grouping responses; and (E) determining the top 10 research priorities as directed by families, caregivers, and clinicians. In previous iterations of the JLA, determining priorities of vulnerable groups remained challenging in the priority setting process;[14] as such, the FRAISE project employed in-person techniques with key community organizations to ensure representativeness from a variety of socioeconomic backgrounds. Parent participants were given a $50 honorarium, childcare, and meals at each session. The Conjoint Health Research Ethics Board at the University of Calgary (REB17-0014) approved this research. We followed the REporting guideline for PRIority SEtting of health research (REPRISE)[15] for this study. All survery participants provided informed consent.

Steering committee

Between November 2017 and March 2020, we engaged a steering committee comprised of researchers, parents, clinicians, and community agency representatives. We aimed for 30 steering committee members who mirrored the regional population, including a minimum of 3% who were First Nations, Metis, or Inuit and 30% who identified as immigrants and/or visible minorities.[16] While membership was flexible to accommodate the realities of parenting young children, a core group of steering committee members emerged. The core group included 15 parents, four clinicians and two community agency representatives; a larger online group of steering committee members also provided feedback and consultation throughout the priority setting process.

Online survey development to identify research questions

To develop the online survey, our steering committee collaboratively identified 12 broad parenting topics of interest. Researchers then developed open-ended survey questions that could elicit potential research questions from families related to each of these 12 topics. The survey was refined by the steering committee using an iterative process of online and in-person engagement. The final survey was piloted with a small group parents who were unfamiliar with the FRAISE project. The survey was launched online using Qualtrics XM Survey Software © (Qualtrics, Provo, UT) in May 2018 and was promoted via Facebook and Twitter. In-person survey completion sessions were also held at various community and healthcare agencies to increase access to under-represented populations, particularly those without reliable internet access.

Priority setting workshops and consensus building

Four in-person priority setting workshops were hosted to build consensus around survey development, survey response theming, the top 30 list, and the top 10 list (Figure 1). Each session was structured using nominal group technique.[17] To analyze survey responses and identify top research priorities, core steering committee members were broken into six groups and provided with survey responses from four of the 12 topic areas. Each author acted as a group facilitator to guide the work, but not the content of the discussion. Each group member was provided with a question from the survey results and given time to silently generate ideas. Then, each group member shared their ideas; the facilitator clarified and recorded. Following group discussion to clarify ideas and priorities, group members voted and ranked each priority, with the top priorities moved forward to the steering committee for consideration. Consensus building concluded with discussion and agreement on which content to move forward from each session. After each in-person session, this process was repeated, using survey software with the broader online steering committee to develop consensus. In the spirit of true consensus building and PSP, researchers facilitated sessions allowing steering committee members to drive decisions and finalize results.[17] As such, modifications were made to the wording of the final top 10 list, resulting in an additional (11th) research priority.[13]

≪INSERT FIGURE 1: *Study flowchart* HERE≫

Role of the funding source

The funders of the study had no role in study design, data collection, data analysis, or report writing. All authors had access to study data. The corresponding author, EMK, had full access to all data and responsibility for the final decision to submit for publication.

Data analysis

Descriptive statistics were calculated from survey participants using Microsoft Excel. Reponses from the survey were captured verbatim in each of the 12 topic areas. All individual responses were printed and cut into individual items. In-person steering committee members worked together to group responses into themes, within each topic area. Reponses from each topic area were analysed by two separate groups to validate theming. Researchers then built the resultant themes (research priorities) into a code book. Research and practice experts provided feedback on the extent to which each potential research priority had been researched. These experts were asked to indicate if each potential research priority was: (A) well researched (systematic reviews and meta-analysis available), (B) somewhat researched (single studies, some inconsistent evidence on topic), (C) not researched (no studies), or (D) unsure. Experts were encouraged to provide comments or feedback on each of the potential priorities. This information was provided to the steering committee during a third consensus building workshop to develop the top 30 list. While the frequency that a particular priority was submitted in the online survey was considered important for subsequent prioritization, steering committee members did not rely solely on these counts to move items forward to the top 30 and top 10 lists.

The top 30 list was shared with the broader online steering committee and committee members were asked to select and rank their top 10 selections. These results were brought back to the fourth and final consensus building workshop to develop the top 10 priorities. The final top 10 (plus one) list was circulated online for final approval and steering committee members also completed an assessment of patient engagement.

Data Statement

The research priorities code book is available upon request from the corresponding author.

Patient and Public Involvement Statement

After the study and research objectives were conceptualized and funding was obtained, the public were engaged via membership in the steering committee. Steering committee members designed the survey and participated in data collection via disseminating the survey through their professional and social networks and provided advice on how to increase survey response rates for diverse groups. These members also provided feedback on the survey regarding participant burden and conducted data analysis in collaboration with the researcher team. In addition, steering committee members were sponsored to participate in local, regional, and national study dissemination via virtual and in-person child health rounds and conferences.

Results

In total, 596 participants consented to participate in the survey, of which 479 completed the survey. Of these, the majority were parents (76.3%; n = 130 tablet; n = 236 online), 16.0% were clinicians (n = 3 tablet; n = 74 online), 4.8% were early childhood educators (n = 10 tablet; n = 13 online), and 2.9% were community agency representatives (n = 0 tablet; n = 22 online). The majority of participants were recruited via social media and electronic networks (n = 337, 70.2%), with 29.8% (n = 143) recruited during targeted in-person sessions where participants were provided with a tablet to record responses. The mean age of all survey respondents was 34.1 years (SD = 9.1), with tablet-based respondents having a slightly younger mean age in years (32.1; SD = 5.9) than the mean age of online-based respondents (34.8; SD = 9.9). For parent participants who were not born in Canada (n = 65), the mean number of years in Canada were 10.7 (SD = 11.4), with tablet-based respondents having fewer mean years in Canada (M = 8.1; SD = 9.3) than online-based respondents (M = 13.6; SD = 13.0). Table 1 describes participant and household characteristics. The socioeconomic and ethnicity distribution of parent participants was comparable to population distributions.[16] Participation rates mirrored geographic population densities (Figure 2).

Table 1. FRAISE Survey participant characteristics.

	Total Completed (N = 480)		Online Survey (n = 337)		Tablet Survey (n = 143)	
All respondents	N	%	n	%	n	%
Employment status						
Working for pay, profit, or self-employed	233	48.5	182	54.0	51	35.7
Caregiving (including parental or maternity leave)	183	38.1	122	36.2	61	42.7
Not working, but looking	14	2.9	9	2.7	5	3.5
Going to school, retired, cannot work due to disability or illness or other	30	6.3	10	3.0	20	14.0
Completed post-secondary education (e.g., certificate or diploma program, undergraduate/graduate degree)	404	84.2	306	90.8	98	68.5
Ethnicity*						
Caucasian	333	69.4	256	76.0	77	53.8
Chinese	29	6.0	14	4.2	15	10.5
South Asian	14	2.9	10	3.0	4	2.8
Latin American	12	2.5	7	2.1	5	3.5
Indigenous	25	5.2	6	1.8	19	13.3
Caribbean	7	1.5	6	1.8	1	0.7
Filipino	6	1.3	4	1.2	2	1.4
Arab	5	1.0	3	0.9	2	1.4
Korean	4	0.8	3	0.9	1	0.7
Southeast Asian	7	1.5	3	0.9	4	2.8
Japanese	2	0.4	2	0.6	1	0.7
African	8	1.7	1	0.3	7	4.9
West Asian	2	0.4	0	0.0	2	1.4
Other	14	2.9	8	2.4	6	4.2
Prefer not to say	15	3.1	9	2.7	6	4.2
Clinicians	77	16.0	74	22.0	3	2.1
Nurse	47	9.8	47	13.9	0	··
Physician	7	1.5	7	2.1	0	··
Dietician	3	0.6	3	0.9	0	··
Occupational therapist	3	0.6	3	0.9	0	··
Other (midwife, social worker, etc.)	17	3.5	14	4.2	3	2.1
Community agency representative	14	2.9	14	4.2	0	··
Early childhood educator/care provider (e.g., nanny, daycare, preschool)	23	4.8	13	3.9	10	7.0
Parent	366	76.3	236	70.0	130	90.9
Mother	292	60.8	194	57.6	98	68.5
# pregnant	*136*	28.3	*85*	25.2	*51*	35.7
Father	71	14.8	40	11.9	31	21.7
Other (e.g., grandparent, aunt)	3	0.6	2	0.6	1	0.7
Parent respondents only (n = 366)						
Partnered	305	83.3	206	87.3	99	76.2
Number of households with children						
1 child in household	173	47.3	121	51.3	52	40.0
2 children in household	74	20.2	48	20.3	26	20.0
3 children in household	27	7.4	15	6.4	12	9.2
4^+ children in household	13	3.6	4	1.7	9	6.9
Born in Canada (Yes)	280	76.5	190	80.5	90	69.2
English as primary household language	318	86.9	206	87.3	112	86.2
Household income (yearly)						

Less than $40,000	66	18.0	17	7.2	49	37.7
$40,000 - $79,999	82	22.4	64	27.1	18	13.8
$80,000 - $119,999	69	18.9	54	22.9	15	11.5
$120,000 - $159,999	49	13.4	34	14.4	15	11.5
More than $160,000	53	14.5	35	14.8	18	13.8
I don't want to say	31	8.5	19	8.1	12	9.2

Note: Due to missing data, numbers in this table may not sum to the total number of completed responses; *indicates respondents could choose all that apply

≪INSERT Figure 2. Geographic distribution of responses HERE≫

In total, we received 3232 submissions of potential research priorities spread across all 12 topic areas (Figure 3). Survey participants ranked stress, emotional, and mental health; sleep; and infant feeding as the three most important priorities. The 3232 suggested priorities obtained via survey responses were themed and collapsed by the steering committee, resulting in 202 unique priorities (Figure 1) of which 34 were shortlisted for consideration in the top 10 list (Supplementary file). Steering committee members used this shortlist to build consensus on the final top 10 (plus one) list of research priorities (Table 2).

≪INSERT Figure 3. Number of raw submissions in each topic area from provincial survey HERE≫

Table 2. Final list and topic areas of the top 10 (plus one) research priorities from conception to two years of age for families in the community.

Rank	Question	Topic Area
1	How can families be supported to develop healthy coping strategies, emotional regulation, and resiliency in both parents and children?	Mental Health/Relationships
2	How can families access supportive care and information when things do not go as expected during pregnancy, labour, birth, or postpartum?	Access to Information/ Pregnancy, Labour & Birth
3	How can the healthcare system and providers ensure access to prevention and treatment of mental health concerns in a safe and trusting environment?	Mental Health
4	How do families navigate multiple sources of health information and access services tailored to their specific circumstances?	Access to Information

5	What supports and services can be developed at the individual, family, community, and government levels to build emotional/mental wellbeing, physical health, and healthy relationships?	Mental Health/Relationships
6	How can eczema, asthma, and allergies be more effectively prevented, assessed, and treated?	Immunity
7	In a constantly changing social and physical environment, how can families increase safety and manage developmentally appropriate risk-taking?	Child Development/Child Safety/Environmental Risk
8	How can sleep problems be prevented, assessed, and treated in a culturally appropriate way that is tailored to individual families?	Sleep
9	How can families be better supported to make informed, family-centred feeding decisions?	Feeding
10	How can families be better supported to promote healthy child development, recognize milestones, and access services for delay?	Child Development
11	With increasing vaccine hesitancy, how can individual, family and population health best be protected?	Immunity

Discussion

To our knowledge, this is the first attempt to identify community-based research priorities for families of well children from conception to age 24 months. Throughout all stages of the PSP, concerns for mental, social, and emotional well-being of parents and children were consistently expressed as priority areas. Comparing the top 10 (plus one) priorities with the initial 12 survey categories, we found that stress, emotional, and mental health were consistently a top concern for families and were identified in three of the top 10 (plus one) priorities. Additionally, parents repeatedly specified the need for structural health system changes at the individual, family, community, and government levels to focus on building emotional/mental well-being, physical health, and healthy relationships – taking the onus off of individual families to solve problems related to mental and physical wellness and prioritizing policy and systems shifts. As such, these research priorities highlight the need to situate individual- or family-focused research priorities within the context of complex health and social systems and involve multiple sectors such as government and health boards.

Across themes, the top 10 (plus one) research priorities reflect the importance of fostering access to information and developing information and services using a culturally sensitive framework. While identified as its own research priority (Table 2), concerns about access to information emerged in each topic area. The ubiquitous nature of access to information as a research priority raises a fundamental question about the accessibility of safe, reliable, and valid information for young families. Previous research from Canada indicates the majority of parents use the internet to access information about children's health, but many use unreliable sources of information.[18] In contrast, Australian women who are pregnant most often reported discussion with their midwife as an information source, while less than half reported using the internet to access information; group information sessions were the least preferred information sources.[19] This contrasts with a report from Devolin and colleagues[20] where Canadian parents rated the Internet (55.3%) and drop-in programs (42.9%) as preferred information sources. With such conflicting evidence, it is unsurprising that researchers and care providers have long struggled with providing safe and reliable information related to child and infant health. Healthcare providers and administrators should collaborate with researchers and parents to critically examine health information provision and consider transformative frameworks to create meaningful and innovative knowledge translation strategies to better communicate evidence.

Several research questions related to physical health emerged as important. For example, parents expressed concerns about how eczema, asthma, and allergies could be more effectively prevented, assessed, and treated; with this class of inflammatory disorders as one of the most common issues in early childhood, this is unsurprising. The fact that parents shared a strong desire for information about preventing these disorders, suggests this is a research area ripe for future development. Additionally, questions emerged about preventing injury while encouraging appropriate physical development and accessing information about culturally appropriate feeding and sleep. While some research exists in each of these areas, evidence is generally underdeveloped, and information needs to be communicated to parents in culturally appropriate and meaningful ways. Of particular importance given the emerging pandemic, parents were unwilling to leave the final steering committee meeting without including a research priority related to vaccine hesitancy. Diverse opinions emerged around this topic and it became clear that evidence-based information about vaccines and immunity was a valued priority.

Findings from this PSP compliment findings from other pediatric research PSPs that have focused on single issue priority setting, such as premature birth,[21] stillbirth,[22] chronic pain,[23] neurodisabilities,[24] or learning difficulties.[25] Despite the breadth in populations and that most existing PSPs were conducted in the UK, research priorities identified in these partnerships related to access to information, mental and emotional health, and healthy relationships, suggesting that these themes are consistently important to parents.

One notable exception to the general trend of conducting PSPs with specialized populations, is a Canadian PSP that developed research priorities for preventative care of children aged zero to five years.[26] Consistent with our top research priority of mental and emotional health in children and their parents, the top research question found by Lavigne and colleagues was "What are effective strategies for screening and prevention of mental health problems?" (p.750).[26] Our findings confirm and extend this priority of building mental and emotional wellness in children belonging younger age groups (and during conception) by broadening the focus to include a child's parents and protective factors of resilience. This suggests that parent, clinician, and researcher priorities are aligned with the emergence of infant mental health (ages zero to five) as a relational concept, whereby infant mental health is supported by optimizing the mental health and resiliency of children's caregivers (i.e. parents and family members).[1] Other similarities included: (A) supporting child development by improving identification and services for developmental delays; (B) developing effective interventions, supports, and services to improve mental health, physical health (obesity, physical activity), and healthy relationships (social skills); and (C) the impact and support of nutritional and feeding factors. Contrasts with our research priorities and those for children aged zero to five years included: (A) the impact of daycare attendance on child health, (B) behavior management in children, and (C) appropriate screen time for children. In addition, research priorities in our list that were not found in the list for children aged zero to five years included: (A) how families can access supportive care and information during unexpected experiences in the perinatal period; (B) access to information; (C) how eczema, asthma, and allergies can be more effectively prevented, assessed, and treated; (D) injury prevention and appropriate levels of risk; and (E) sleep problems in families. It should be noted that differences may represent prioritization related to different developmental stages and ages (i.e., screen time is not recommended for children under age two) and/or slight differences in prioritization method. For example, helping families identify appropriate childcare appeared in our top 34 list and aligns with a similar research priority related to impact of daycare attendance in the zero to five years list.

In a Delphi study of perceived research priorities of clinical staff at an Australian parenting centre, Hauck and colleagues[27] determined the top research priorities to be related to short- and long-term evaluation of the effectiveness of the centre's programs on children's behavior and stress levels, parental expectations, healthy family relationships, as well as parental use of sleep, settling, and feeding strategies. Determining the effectiveness of the centre's programme in the context of postpartum depression was also an identified priority.[27] Although these research priorities were not informed by parental participation and may have limited generalizability to other centres and/or countries, it is notable that issues related to sleep, infant feeding, healthy relationships, and parental mental health are captured by both ours and Hauck's[27] lists. This is not surprising as a study in the same geographical area suggested that breastfeeding, sleep issues, child development, helping children cope with emotions, and understanding child brain development were all in the top 10 (plus one) parenting topics ranked as somewhat or very important by parents.[20]

Clinical and research applications

To move forward with these research priorities, it is evident that multiple stakeholders must be engaged, and connections made across a variety of sectors and disciplines. Wording for the top 10 (plus one) research priorities requested consideration of "culturally appropriate" and "safe and trusting environments." Our findings highlight the need for increased emphasis around parenting education and culturally appropriate strategies to support mental health and well-being. Adopting an intersectional lens when conducting research, and providing care to pregnant and young families, will better address these considerations and provide meaningful evidence to inform health outcomes. Similar to how parents of diverse groups may have differing or intensified concerns relating to child health issues,[28] it is important to recognize that research priorities may differ based on community member's characteristics, specifically in terms of race, income, access to health services, and experiences, including racism. These top 10 (plus one) priorities were developed using an intersectional framework. Moving forward, research in these priority areas must continue to comprehensively and meaningfully include people with diverse sociopolitical backgrounds and experiences.[9]

The findings from this study should be considered in the context of its strengths and limitations. In terms of strengths, there was a large sample size for the online survey comparable to other PSPs; this created space for broad representation of stakeholders including clinicians, community agency representatives, parents, and ethnically diverse representatives of the study location. Limitations included the inherent and individual biases that self-selected, voluntary researchers and steering community members view the world with, which may reduce generalizability of the final priority list. Additionally, the research team was composed entirely of white heterosexual women and although attention was paid to ensure participation of fathers, individuals self-identifying as non-white, and those in non-heterosexual or single parent partnerships, these voices may not have been entirely represented in the final priorities.

Researching these family-driven priorities will reduce the knowledge to practice gap and result in higher quality health care services for families of young children; this will generate opportunities to improve services for families, including innovative delivery models and evidence-based treatment options to enable equitable access to services. Further, involving practitioners in the PSP will enhance the implementation of new evidence-based practice recommendations.[29] These outcomes, combined with decreased lag time from research to practice, will result in improved child and family outcomes. As a result of our strong collaborative partnerships and inclusion of policy makers in each stage of the PSP, the provincial health service has already utilized this information to streamline and enhance evidence-based information delivery to parents and care providers in the province. By sharing the FRAISE top 10 (plus one) research priorities, we invite other researchers to ensure their work aligns with patient-oriented research as a foundation for future initiatives targeted at improving outcomes in families with young children.

Author Contributions

EMK and MB drafted the manuscript, which all authors reviewed, provided feedback and approved the final version. EMK, MB, KSB, CG, and KMB designed the study. EMK, MB, KSB, CG, KMB and LTM developed the steering committee meeting content. EMK, MB, KSB, CG, KMB, LTM, SD, LC and the Steering Committee devised the data collection instruments. JW, PP, EMK, MB, KSB, and CG were responsible for data collection and for database design and management. EMK, MB, KSB, CG, KMB, LTM, JW, SD, LC and the Steering Committee completed the data analyses.

Declarations of interests

We declare no competing interests.

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References

1. Shonkoff JP. Capitalizing on advances in science to reduce the health consequences of early childhood adversity. *JAMA Pediatr* 2016; **170**: 1003–7.
2. Bronfenbrener, U. (ed.). Making human beings human: bioecological perspectives on human development. Thousand Oaks, CA: Sage, 2005.
3. Wall G. Mothers' experiences with intensive parenting and brain development discourse. *Womens Stud Int Forum* 2010; **33**: 253–63.
4. Albrecht L, Archibald M, Snelgrove-Clarke E, Scott SD. Systematic review of knowledge translation strategies to promote research uptake in child health settings. *J Pediatr Nurs* 2016; **31**: 235–54.
5. Chalmers I, Glasziou P. Avoidable waste in the production and reporting of research evidence. *Obstet Gyneco*l 2009; **114**: 1341–45.
6. Manafò E, Petermann L, Vandall-Walker V, Mason-Lai P. Patient and public engagement in priority setting: a systematic rapid review of the literature. *PLoS One* 2018; **13**: e0193579.
7. Chalmers I, Bracken MB, Djulbegovic B, et al. How to increase value and reduce waste when research priorities are set. *Lancet* 2014; **383**: 156–65.
8. Shimmin C, Wittmeier KDM, Lavoie JG, Wicklund ED, Sibley KM. Moving towards a more inclusive patient and public involvement in health research paradigm: the incorporation of a trauma-informed intersectional analysis. *BMC Health Serv Res* 2017; **17**: 539.
9. Bailey J, Steeves V, Burkell J, Shade LR, Ruparelia R, Regan P. Getting at equality: research methods informed by the lessons of intersectionality. *Int J Qual Methods* 2019; **18**: 1–13.
10. Mann BS, Manns BJ, Dart A, et al. An assessment of dialysis provider's attitudes towards timing of dialysis initiation in Canada. *Can J Kidney Health Dis* 2014; **1**: 3.
11. Cowan K, Oliver S. The James Lind Alliance guidebook. 2010. http://www.jlaguidebook.org/pdfguidebook/guidebook.pdf (accessed Nov 22, 2016).
12. Bright KS, Ginn C, Keys EM, et al. Study protocol: determining research priorities of young Albertan families (The Family Research Agenda Initiative Setting Project-FRAISE)-participatory action research. *Front Public Health* 2018; **6**: 228.
13. Breault LJ, Rittenbach K, Hartle K, et al. The top research questions asked by people with lived depression experience in Alberta: a survey. *CMAJ Open* 2018; **6**: E398–405.
14. Barnieh L, Jun M, Laupacis A, Manns B, Hemmelgarn B. Determining research priorities through partnership with patients: an overview. *Semin Dial* 2015; **28**: 141–46.
15. Tong A, Synnot A, Crowe S, et al. Reporting guideline for priority setting of health research (REPRISE). *BMC Med Res Methodol* 2019; **19**: 243.
16. Statistics Canada. 2011 National Household Survey: Statistics Canada. 2013. https://www12.statcan.gc.ca/nhs-enm/2011/dp-pd/dt-td/Index-eng.cfm (accessed Oct 7, 2020).
17. Carney O, McIntosh J, Worth A. The use of the nominal group technique in research with community nurses. *J Adv Nurs* 1996; **23**: 1024–29.
18. Pehora C, Gajaria N, Stoute M, Fracassa S, Serebale-O'Sullivan R, Matava CT. Are parents getting it right? A survey of parents' internet use for children's health care information. *Interact J Med Res* 2015; **4**: e12.
19. Grimes HA, Forster DA, Newton MS. Sources of information used by women during pregnancy to meet their information needs. *Midwifery* 2014; **30**: e26–33.

20. Devolin M, Phelps D, Duhaney T, et al. Information and support needs among parents of young children in a region of Canada: a cross-sectional survey. *Public Health Nurs* 2013; **30**: 193–201.
21. Duley L, Uhm S, Oliver S. Top 15 UK research priorities for preterm birth. *Lancet* 2014; **383**: 2041–42.
22. Heazell AEP, Whitworth MK, Whitcombe J, et al. Research priorities for stillbirth: process overview and results from UK Stillbirth Priority Setting Partnership. *Ultrasound Obstet Gynecol* 2015; **46**: 641–47.
23. Birnie KA, Dib K, Ouellette C, et al. Partnering for pain: a priority setting partnership to identify patient-oriented research priorities for pediatric chronic pain in Canada. *CMAJ Open* 2019; **7**: E654–64.
24. Morris C, Simkiss D, Busk M, et al. Setting research priorities to improve the health of children and young people with neurodisability: a British Academy of Childhood Disability-James Lind Alliance research priority setting partnership. *BMJ Open* 2015; **5**: e006233.
25. Lim AK, Rhodes S, Cowan K, O'Hare A. Joint production of research priorities to improve the lives of those with childhood onset conditions that impair learning: The James Lind Alliance priority setting partnership for 'learning difficulties'. *BMJ Open* 2019; **9**: e028780.
26. Lavigne M, Birken CS, Maguire JL, Straus S, Laupacis A. Priority setting in paediatric preventive care research. *Arch Dis Child* 2017; **102**: 748–53.
27. Hauck Y, Kelly RG, Fenwick J. Research priorities for parenting and child health: a Delphi study. *J Adv Nur* 2007; **59**: 129–39.
28. Garbutt JM, Leege E, Sterkel R, Gentry S, Wallendorf M, Strunk RC. What are parents worried about? Health problems and health concerns for children. *Clin Pediatr* 2012; **51**: 840–47.
29. Mathieson A, Grande G, Luker K. Strategies, facilitators and barriers to implementation of evidence-based practice in community nursing: a systematic mixed-studies review and qualitative synthesis. *Prim Health Care Res Dev* 2019; **20**: e6.

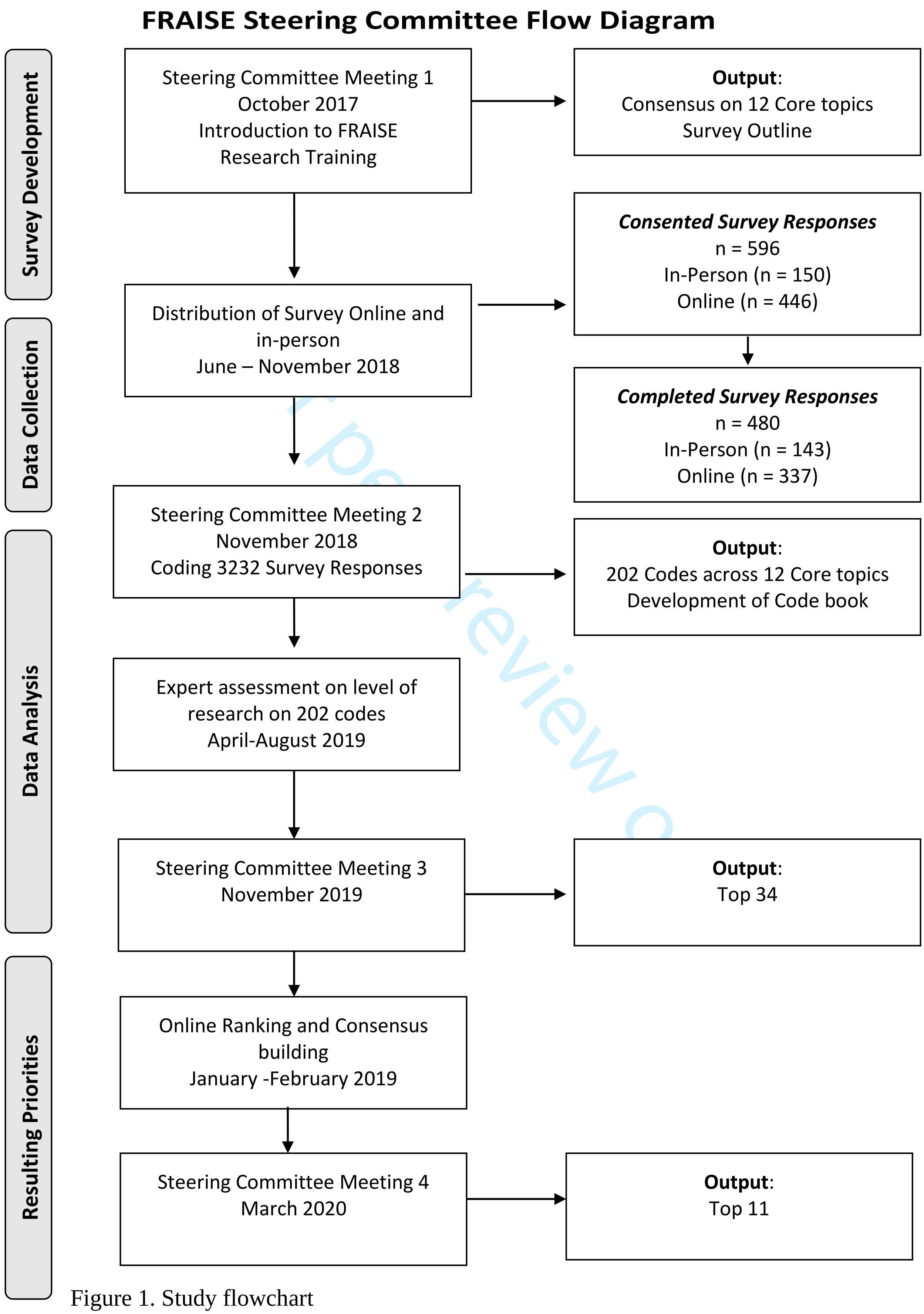

Figure 1. Study flowchart

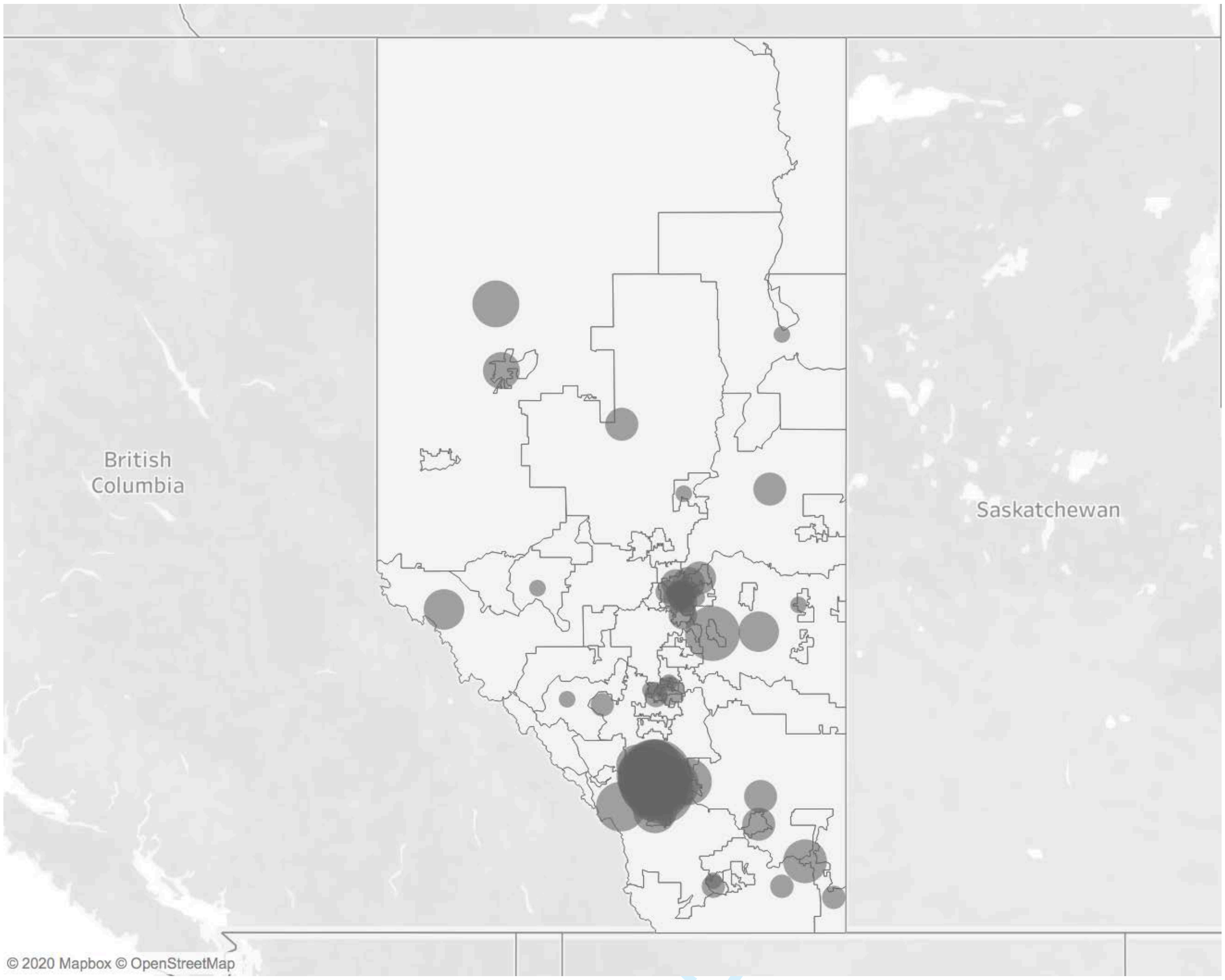

Figure 2. Geographic distribution of responses

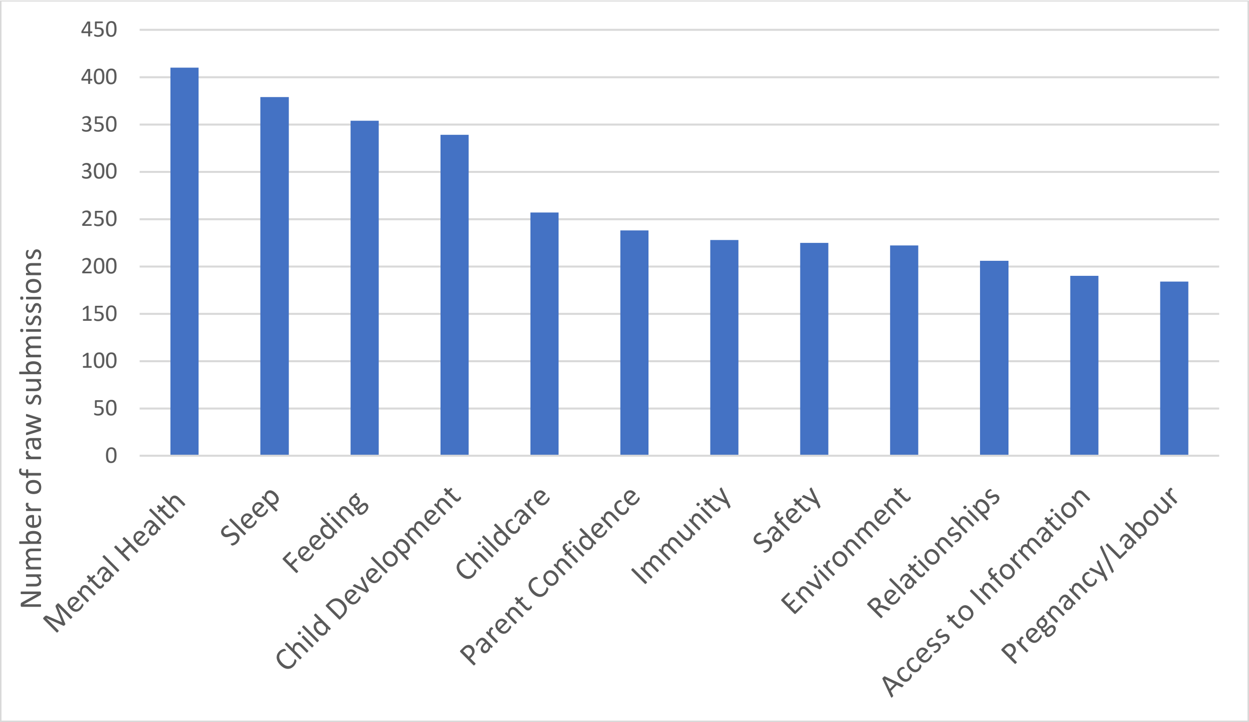

Figure 3. Number of raw submissions in each topic area from provincial survey

Supplemental Table. Top 34 Themes	
Question	**# of times in top 10**
How can families and caregivers of children 2 and under be supported to develop healthy coping strategies, emotional regulation, and resiliency in both themselves and their children?	15
How to access information/programs and navigate the healthcare system when you are from a minority/non-dominant group?	14
What is an age appropriate level of risk taking for children to promote their growth & development while maintaining adequate (good enough) safety?	12
How do parents manage multiple sources of information to make health decisions that are tailored to their specific circumstances and needs?	12
How can families (partners & extended family members), communities, and government encourage self-care and provide emotional and practical support for parents during the perinatal and early childhood period?	11
When things don't go as planned, how can parents get care, support, and information during pregnancy, labour, birth or the postpartum period?	11
What formats (i.e. in-person, online, group) do families prefer when accessing supports and services to develop and sustain healthy relationships and mental well-being?	10
How can health care professionals provide a safe and trusting environment that encourages women/families to disclose mental health concerns without fear of negative repercussions?	10
How to identify, prevent, and treat eczema, asthma, and allergies?	10
How do we create culturally appropriate, family specific information about sleep (co-sleeping, swaddling, sleep training, etc.) from conception to age 2?	10
What are the advantages and disadvantages of using a trauma-informed model of care to explore pregnancy, childbirth, and postpartum for all parents?	9
How do we prevent, assess, and treat sleep problems in parents and children?	9
What supports and services are required to support families to develop and sustain healthy relationships and mental wellbeing?	8
To minimize physician visits, what user centred information sources and formats are most acceptable and effective to provide parents and healthcare providers with reliable and up-to-date information?	8
How can we best support families with their infant feeding decisions?	7
With increasing vaccine hesitancy, how do we best protect individual and population health?	7
How can families during the perinatal and early childhood period access appropriate and evidence-based assessments, prevention, treatment, and support for mental health concerns?	7
How can we manage the effect of exposure to cannabis smoking to families with young children?	7
In children 2 and under, how can families and caregivers be supported around milestones, behaviours, and signs of delay?	7
In addition to vaccination, how can parents and children build their immunity?	6
What supports and services increase general parental confidence?	6
How can families during the perinatal and early childhood period manage emotional and physical wellbeing?	6
What are the necessary measures to increase safety for toddlers/children in an ever changing social and physical environment (during the early childhood period)?	6
How can communities and health care professionals provide culturally sensitive best practices for promoting healthy pregnancies & fetal/infant/child development?	6
How can we increase awareness and access to additional services and resources for families of young children?	6
What are expected and problematic sleep patterns in caregivers, infants, and toddlers?	6

What evidence-based community resources and services are available to help foster healthy relationships for expecting parents?	5
How can families be supported to identify the right fit of childcare, looking at quality, safety, and affordability while still incorporating their own beliefs and values?	5
How do we build on general confidence to generate transferable skills for parenting?	4
What are the consequences of sleep deprivation and problems in parents and children?	4
What are effective strategies for accessing/using my electronic health record and keeping track of my family's health information?	3
What should healthcare providers recommend for guidelines to support families to transition from milk feeding to solid foods?	2
How does infant feeding impact allergies?	2
How can parents during early childhood identify & prevent child safety risks?	2

Reporting Guideline for Priority Setting of Health Research (REPRISE)[1]

#	Item	Descriptor
		Context and Scope
1	Define geographical scope	Regional geographic area "within a province in Western Canada" (methods, pg. 7).
2	Define health area, field, focus	Focussed on a specific population of healthy young families and caregivers in the community setting. The population of interest is "families of well children from conception to age 24 months" (introduction, pg. 6).
3	Define the intended beneficiaries	Intended beneficiaries of these findings include researchers, care providers, community services representatives, and families themselves. "Develop research outcomes meaningful to knowledge users, parents, and care providers" (introduction, pg. 6).
4	Define the target audience of the priorities	The target audience who has the potential to fund future research or implement the identified priorities include care-providers, health services organizations, and researchers. "As a result of our strong collaborative partnerships and inclusion of policy makers in each stage of the PSP, the provincial health service has already utilized this information to streamline and enhance evidence-based information delivery to parents and care providers in the province. By sharing the FRAISE top 11 research priorities, we invite other researchers to ensure their work aligns with patient-oriented research as a foundation for future initiatives targeted at improving outcomes in families with young children." (discussion, pg. 18).
5	Identify the research area	The broad research area of the priority setting research encompasses health services and public health. "these family-driven priorities will reduce the knowledge to practice gap and result in higher quality health care services for families of young children" (discussion, pg. 17).
6	Identify the type of research questions	The types of research questions were broad, and ranged from etiology (i.e., asthma, eczema, allergy causes/diagnosis) to implementation (i.e., developing community supports). Questions also addressed psychosocial and behavioural research priorities. The researchers did not pre-define the type of research questions that were expected to be generated from the priority setting partnership. See Table 3 for top 11 research priority questions.

7	Define the time frame	The researchers outline the timeframe during which the research was conducted "between November 2017 and March 2020" (methods, pg. 8), however, the anticipated relevance of the research priorities is not addressed. Plans for to monitor or update the priorities with future prioritization activities are not discussed.
	Governance and Team	
8	Define the selection and structure of the leadership and management team	The priority setting was facilitated by the authors, however, the structure of the research team, relevance for inclusion, or technical expertise as a facilitator is not addressed. "Each author acted as a group facilitator to guide the work, but not the content of the discussion" (methods, pg. 9). A steering committee (smaller in-person group and larger online group) completed the prioritization process. The selection of the committee members and structure of the team is not outlined. Several groups are mentioned in the acknowledgements paper for contributing to the committee, but their role is not elaborated on in the manuscript. The authors simply state, "we engaged a steering committee comprised of researchers, parents, clinicians, and community agency representatives" and "core group included 15 parents, four clinicians and two community agency representatives; a larger online group of steering committee members also provided feedback and consultation throughout the priority setting process" (methods, pg. 8). The characteristics of, or any differences between, the in-person steering committee members and online members are not explicated. The need for participant diversity is noted, "We aimed for 30 steering committee members who mirrored the regional population, including a minimum of 3% who were First Nations, Metis, or Inuit and 30% who identified as immigrants and/or visible minorities", however, whether or not the goal for diversity was fulfilled is not described. The authors do note the limitation that the study may not be generalizable to all populations, "although attention was paid to ensure participation of fathers, individuals self-identifying as non-white, and those in non-heterosexual or single parent partnerships, these voices may not have been entirely represented in the final priorities" (discussion, pg. 17).
9	Describe the characteristics of the team	The demographics of the steering committee (on-line and in-person) are not presented. Furthermore, the affiliations or expertise of the researchers, clinicians, and community agency representatives are not presented.

		The authors do recognize that the research team was "composed entirely of white heterosexual women" (discussion, pg. 17), and also present the demographics of the broader sample who completed the survey to identify the initial parent questions that informed the final 11 themes, "Table 1 describes participant and household characteristics. The socioeconomic and ethnicity distribution of parent participants was comparable to population distributions. Participation rates mirrored geographic population densities." (results, pg. 11).
10	Describe any training or experience relevant to conducting priority setting	The presence of participant or researcher training, consultants, or past experience with priority setting research is not addressed in the body of the manuscript. However, the authors do acknowledge the "Alberta Strategy for Patient-Oriented Research Support Unit for their expertise, guidance and support in the developmental stages of this project" within the acknowledgement section.
Framework for Priority Setting		
11	State the framework used (if any)	The authors identify the use of "a consensus-building and strengths-based approach, modelled after participatory action research and the James Lind Alliance" (methods, pg. 7). The authors provide sound rationale for their use of a modified James Lind Alliance approach, recognizing that the conventional methodology is "resource intensive and may be impractical for lower resourced research studies and vulnerable populations" (methods, pg. 7).
Stakeholders and Participants		
12	Define the inclusion criteria for stakeholders involved in priority-setting	As mentioned above, the authors sought to include a diverse group of stakeholders who accurately represented the population of the research setting. Specific inclusion criteria or the process of identifying, approaching, and engaging stakeholders is not outlined. More broadly for the survey participants, efforts to engage diverse populations was achieved through in-person survey recruitment in conjunction with online surveys.
13	State the strategy or method for identifying and engaging stakeholders	The strategy for identifying stakeholders in the steering committee are not addressed in the body of the manuscript. Again, several groups and existing partnerships are noted in the acknowledgements for their contribution to the committee, however, strategies for stakeholder engagement are not outlined.
14	Indicate the number of participants and/or organizations involved	The authors briefly describe the number of stakeholders involved in priority setting, "we engaged a steering committee comprised of researchers, parents, clinicians, and community agency representatives" and "core group included 15 parents, four clinicians and two community agency representatives; a larger online group of steering committee members also provided

		feedback and consultation throughout the priority setting process" (methods, pg. 8). Although the number of in-person committee members is clear, there is no number provided for the larger online steering committee group. Furthermore, the organizational affiliation (or the nature of the organization, i.e., health, government etc.) of researchers, clinicians, or community representatives is not described.
15	Describe the characteristics of stakeholders	As noted above, apart from the number of steering committee members who were parents, clinicians, and community agency representatives in the in-person group, there is limited explanation of the steering committee characteristics.
16	State if reimbursement for participation was provided	The authors clearly state, "Parent participants were given a $50 honorarium, childcare, and meals at each session" (methods, pg. 7).
Identification and Collection of Research Priorities		
17	Describe methods for collecting initial priorities	The development of initial priorities appears to be two-fold. The authors and steering committee developed an initial list of 12 parenting topics, which then informed a survey where parents could ask questions about each of the 12 parenting topics. These parent questions relative to the 12 topics would then be analyzed to create the final top 11 priority list (methods, pg. 7-8). The authors do not elaborate how the initial 12 parenting topics were selected, apart from stating, "our steering committee collaboratively identified 12 broad parenting topics of interest" (methods, pg. 8). However, it is noted that the survey generated from the 12 topic areas was refined in collaboration with steering committee members, both online and in-person, and then pilot tested with "outside" parents. The subsequent data analysis of the themes and questions resulting from the provincial survey took place within in-person steering committee workshops "structured using nominal group technique" (methods, pg. 9), and through online consensus, "After each in-person session, this process was repeated, using survey software with the broader online steering committee to develop consensus" (methods, pg. 9).
18	Describe methods for collating and categorizing priorities	The initial broad priority suggestions and research questions were thematically grouped and analyzed through in-person steering committee workshops. "All individual responses were printed and cut into individual items. In-person steering committee members worked together to group responses into themes, within each topic area. Reponses from each topic area were analysed by two separate groups to

		validate theming. Researchers then built the resultant themes (research priorities) into a code book" (data analysis, pg. 10).
19	Describe methods and reasons for modifying (removing, adding, reframing) priorities	The authors did not play a role in determining the consensus and decisions to modify priorities. The decisions to move priorities forward to the final list was based on steering committee consensus both online and in-person. The authors appear to have provided members with tools to guide decisions (i.e., frequencies, evidence of the extent to which past research had been conducted), but this did not lead the steering committee decisions. "while frequency counts were considered as an important prioritization strategy, steering committee members did not rely solely on these counts to move items forward to the top 30 and top 10 lists" (data analysis, pg. 10). "In the spirit of true consensus building and PSP, researchers facilitated sessions allowing steering committee members to drive decisions and finalize results" (methods, pg. 9).
20	Describe methods for refining or translating priorities into research topics or questions	Apart from the aforementioned theming process, research questions were developed through consensus approach, committee members could initially generate ideas on their own, and then within their smaller group, and then eventually with the broader in-person steering committee members who collaboratively decided how to word the questions (methods, pg. 9).
21	Describe methods for checking whether research questions or topics have been answered	Once collapsed into themes by the steering committee, the authors disseminated the preliminary research priorities to experts in the subject matter. "Experts were asked to indicate if each potential research priority was: (A) well researched (systematic reviews and meta-analysis available), (B) somewhat researched (single studies, some inconsistent evidence on topic), (C) not researched (no studies), or (D) unsure. Experts were encouraged to provide comments or feedback on each of the potential research priorities" (data analysis, pg. 10).
22	Describe number of research questions or topics	An initial 12 broad parenting topics were identified to inform the provincial survey. The survey generated a total 3232 potential research priorities, which were analyzed and consolidated by the steering committee into 202 unique themes. The themes were further collapsed into a list of 34 research priorities, and then eventually into a top 11 list of research priorities. "Selecting 11 priorities as opposed to 10 priorities was the steering committee consensus" (abstract, pg. 2).
Prioritization of Research Topics/Questions		

23	Describe methods and criteria for prioritizing research topics or questions	The identification of priority themes and questions resulting from the provincial survey took place within in-person steering committee workshops “structured using nominal group technique” (methods, pg. 9). Decisions were based on group consensus between participants, with the authors facilitating the discussions but not leading or influencing the decisions. Although there is mention that steering committee members could score and rank the priorities, “The top 30 list was shared with the broader online steering committee and committee members were asked to select and rank their top 10 selections” (methods, pg. 10), the authors do not describe how group consensus was determined for the final list. There are no specific criteria that were used to guide the ranking or prioritization outlined in the manuscript. Steering committee members did have information regarding the level of evidence for each topic, however, this does not appear to have been the guiding factor for the ranking.
24	State the method or threshold for excluding research topics/questions	The authors do not describe a threshold or criteria for excluding priorities. The process for settling challenges to the prioritization process is not described in detail. Discussion, voting, and ranking were used to decide what priorities moved forward (methods, pg. 9), however, these processes are not described in detail.
		Output
25	State the approach to formulating the research priorities	The final priorities have been identified as 11 research questions (Table 3). How the questions were worded, and whether a specific format (i.e., PICO) was used is not explicated. Along with the top 11 research priorities, the researchers also identify the broad topic area (from the initial 12 survey domains) relevant to the priority.
		Evaluation and Feedback
26	Describe how the process of prioritization was evaluated	The usefulness, relevance, and reliability of the priorities are not evaluated. The authors do mention how more than one group themed the survey responses to enhance reliability in the early stages of prioritization, “Reponses from each topic area were analysed by two separate groups to validate theming” (data analysis, pg. 10). The findings have also been used by the provincial health organization already, which may speak to their relevance and utility. The authors also allude to evaluation of engagement from the steering committee member perspective, “steering committee members also completed an assessment of patient engagement.” (data analysis, pg. 10). However, this process, the findings, or implications for engagement evaluation are not described further.

27	Describe how priorities were fed back to stakeholders and/or the public, and how feedback (if received) was addressed and integrated	The authors state that the final priorities were sent to the committee for approval, "the final top 10 (plus one) list was circulated online for final approval" (data analysis, pg. 10). There was no revision or appeal process described if committee members did not agree with the final list/question wording.
	Implementation	
28	Outline the strategy or action plans for implementing priorities	The researchers describe how sharing the findings could inform future research to ensure it is aligned with relevant priorities for families by stating, "By sharing the FRAISE top 11 research priorities, we invite other researchers to ensure their work aligns with patient-oriented research as a foundation for future initiatives targeted at improving outcomes in families with young children" (discussion, pg. 18). However, no formal action plan for implementing the priorities is described.
29	Describe plans, strategies, or suggestions to evaluate impact	Although the authors mention how the provincial health service has already utilized the study findings to inform information delivery, formal plans to evaluate the impact of the priority setting findings are not outlined.
	Funding and Conflict of Interest	
30	State sources of funding	The funding came from a relevant funding agency for child health and patient-oriented research, "We are grateful for funding provided by the Alberta Children's Hospital Research Institute and by Alberta Innovates" (funding, pg. 19). The funder did not play a role in any portion of the research study, "The funders of the study had no role in study design, data collection, data analysis, or report writing" (methods, pg. 9).
31	Declare any conflicts or competing interests	The authors state no conflict of interest, "We declare no competing interests" (declaration of interests, pg. 19).

References:

1. Tong A, Synnot A, Crowe S, et al. Reporting guideline for priority setting of health research (REPRISE). *BMC Med Res Methodol* 2019; **19**(1): 243.

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Top 10 (plus 1) research priorities for expectant families and those with children to age 24 months in Alberta, Canada: Results from the Family Research Agenda Initiative Setting (FRAISE) priority setting partnership project.

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Abstract

Objective: The study objective was to identify the top 10 research priorities of expectant parents and caregivers of children up to age 24 months.

Design: A priority setting partnership using a modified James Lind Alliance approach was implemented. First, a core steering committee was formed, consisting of 17 parents, clinicians, and community agency representatives. Second, through in-person collaboration with steering committee members, we developed and distributed a survey to identify research priorities across 12 topics. In total, 596 participants consented and 480 completed the survey. Survey responses were grouped and themed into codes during a consensus-building workshop with steering committee members (n=18). Research and practice experts were consulted to provide feedback on which themes had already been researched. An in-person (n=21) workshop was used to establish the top 34 priorities, which were circulated to the broader steering committee (n=25) via an online survey. Finally, the core steering committee members (n=18) met to determine and rank a top 10 (plus one) list of research priorities.

Setting: This study was conducted in Alberta, Canada.

Participants: Expectant parents and caregivers of children up to age 24 months.

Results: Survey results provided 3232 responses, with 202 unique priorities. After expert feedback and steering committee consensus, a list of 34 priorities was moved forward for final consideration. The final top 10 (plus 1) research priorities included three priorities on Mental Health/Relationships, two priorities on each of Access to Information, Immunity, and Child Development, and one priority on each of Sleep, Pregnancy/Labour, and Feeding. Selecting 11 instead of 10 priorities was based on steering committee consensus.

Conclusions: The findings will direct future maternal-child research, ensuring it is rooted in parent-identified priorities that represent contemporary needs. To provide meaningful outcomes, research in these priority areas must consider diverse socioeconomic backgrounds and experiences.

Keywords: Parent, Priority Setting, Research Priorities, James Lind Alliance, Patient Engagement, Patient-Oriented Research, Participant Involvement.

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Article Summary

Strengths and Limitations of This Study

- The study had a large sample size (n = 480) for the online survey, comparable to other priority setting partnerships.
- Targeted recruitment strategies, including in-person recruitment at community and social service locations and father parenting classes, were used to successfully improve representation of diverse participants.
- Although attention was paid to promote genuine participation of diverse stakeholders and reach consensus on the research priorities, there is potential that these priorities were influenced by the steering committee members' and/or researchers' inherent and individual biases.
- While the partnership approach followed principles of participatory action research and was modelled after other published studies that used a modified James Lind Alliance methodology, this approach has not yet been validated.
- By creating a welcoming and family-friendly atmosphere and including alternate engagement strategies, parents were meaningfully engaged throughout the partnership process, resulting in the identification of parent-oriented research priorities, many of which differed from priorities previously established by researchers, funders, and industry.

Introduction

Due to the critical and rapid development that occurs within the first three years of life, investing in early childhood has an estimated return of 800%.[1] Parents of young children have the greatest potential to optimize social and environmental conditions to foster optimal child health outcomes.[2] High parental expectations, intensive parenting, and increased access to information have drastically changed parenting experiences over the past 10-15 years.[3] The dynamic nature of the contemporary maternal-child and parenting landscape may be inadequately incorporated into existing research priorities and questions. Further, researchers and clinicians often encounter difficulties effectively translating and implementing research on child health and development.[4] As the knowledge base for supporting healthy developmental outcomes grows in breadth and scope, effectively prioritizing research investment is crucial to maximize impact and minimize research waste.[5]

Understanding health research priorities of knowledge users, such as parents, clinicians, and community agency representatives, is vital to conducting research that is more likely to contribute to meaningful changes in health outcomes.[6] Evidence generated without the consideration of the knowledge user will have minimal effect on clinical practice and reduce meaningful outcomes.[7] Further, there is growing awareness of the importance of adopting an intersectional lens when conducting health research to adequately address culturally and socio-politically appropriate parenting outcomes.[8] Employing an intersectional lens by centering the perspectives and meaning of parents, patients, or families will result in evidence and resultant policies that account for the diverse needs of patients and knowledge users.[9] Engaging knowledge users in research priority setting initiatives is recognized as an effective and ethical means of prioritizing the allocation of limited public research funds.[7] Not only does priority setting work promote researcher accountability, but this integrative approach to knowledge translation may also reduce lag time between producing and implementing knowledge,[10] while contributing to the development of culturally and socially meaningful outcomes. Parental involvement in research may enhance engagement with, and uptake of, interventions and services that support early childhood health, resulting in more timely and effective care.

Priority setting partnerships

Patient-oriented research, specifically priority setting partnerships (PSPs), are increasingly identified as an effective method to decrease research waste and develop research outcomes meaningful to knowledge users, parents, and care providers.[7] PSPs bring together clinicians, researchers, and patients to identify and prioritize research uncertainties using a systematic and collaborative process.

Involving parents and caregivers in the foundational development of research through PSPs is a feasible method to produce meaningful outcomes – by creating knowledge important to parents and providers, the likelihood of new evidence uptake and shifting healthcare practices is increased. To date, there have been no PSPs focused on identifying community-based research priorities for families of well children from conception to age 24 months. When purposefully employing a participatory action framework, PSPs can use intersectional principles by capturing the perspectives and experiences of traditionally neglected populations.[8,9] Therefore, the objective of this study was to engage a diverse group of perspectives in identifying the top 10 research priorities of expectant parents and caregivers of children up to age 24 months.

Methods

The Family Research Agenda Initiative Setting (FRAISE) project used a modified James Lind Alliance (JLA)[11] approach to identify research priorities of parents, clinicians, and community agency representatives within Alberta, a province in Western Canada.[12] Many PSPs use the JLA approach;[6] however, this methodology is resource intensive and may be impractical for lower resourced research studies and vulnerable populations. As such, a modified approach to the JLA method has emerged as a feasible alternative.[13] These modifications included facilitation of the initial steering committee workshop by subject matter experts in patient engagement and priority setting partnerships from the Alberta Strategy for Patient-Oriented Research Support Unit (AbSPOR), rather than a JLA advisor. After this initial workshop, the lead authors, who are trained in group facilitation and community engagement, facilitated the remaining in-person workshops using consensus-building and shared-decision making strategies. During the analysis and prioritization of the potential research priorities, we used collective sensemaking to narrow down and rank the research priorities. Rather than conduct rapid literature reviews to determine the level of evidence available for the research uncertainties, we asked practice and research subject matter experts to rate the availability of evidence on research uncertainties. These rankings were then used by the steering committee members to help them prioritize the top 30 priorities. FRAISE applied a consensus-building and strengths-based approach, modelled after participatory action research and the JLA. James Lind Alliance PSPs bring together patients, caregivers, and clinicians, which requires sensitivity to varying participant capacities, ongoing effective communication, transparency in decision-making, and inclusivity of all views.[11] The modified JLA process utilized in the FRAISE project involved a series of iterative steps (Figure 1): (1) formation of a steering committee, including parents, clinicians and community agency representatives and online survey development; (2) delivering the survey to gather research uncertainties or questions from parents, clinicians and community agency representatives; (3) group and/or theme responses into codes; (4) consult with research and practice experts to determine which codes were already well researched, (5) consensus building to determine and rank the top priorities and (6) prioritize and rank top 10 research priorities as directed by families, caregivers, and clinicians.

In previous iterations of the JLA, determining priorities of vulnerable groups remained challenging in the priority setting process;[14] as such, the FRAISE project employed in-person techniques with key community organizations to ensure representativeness from a variety of socioeconomic backgrounds. These techniques included research assistants physically attending programs and services that provided targeted care to populations who were under-represented in the survey to recruit potential participants. To facilitate participation at these sites, research assistants carried electronic tablets that participants could use to complete the online survey before or after their programming or appointments. Parent steering committee members were given a $50 honorarium, childcare, and meals at each session. The Conjoint Health Research Ethics Board at the University of Calgary (REB17-0014) approved this research. We followed the REporting guideline for PRIority SEtting of health research (REPRISE)[15] for this study. All survey participants provided informed consent.

Steering committee

Between November 2017 and March 2020, we engaged a steering committee comprised of researchers, parents, clinicians, and community agency representatives. We aimed for 30 steering committee members who mirrored the regional population, including a minimum of 3% who were Indigenous, and 30% who identified as immigrants and/or visible minorities.[16] While membership was flexible to accommodate the realities of parenting young children, a core group of steering committee members emerged. The core group included 15 parents, four clinicians, and two community agency representatives. While we did not meet our target for Indigenous representation, we exceeded our aim for a diverse group, with five of the 15 parents identifying as a visible minority, two parents identifying as fathers, and two parents identifying as new to Canada. To accommodate the regionality and required flexibility of steering committee members, we provided opportunities for a larger online group of additional steering committee members. These members consisted of parents and providers who attended some, but not all workshops in person, as well as healthcare providers from other sites in Alberta, and directors and managerial staff who were unable to attend in-person workshops due to time constraints. The size of this group fluctuated throughout the study timeline, with a maximum of 10 members in addition to the core steering committee. The role of the larger steering committee was to provide online feedback and consultation throughout the priority setting process.

Online survey development to identify research questions

The first consensus building workshop focussed on introducing and training the core steering committee, orientating them to FRAISE, and developing the survey instrument. To develop the online survey, the core steering committee (n = 17) collaboratively identified 12 broad parenting topics of interest. Researchers then developed open-ended survey questions that could elicit potential research questions from families related to each of these 12 topics. The survey was refined by the core steering committee using an iterative process of online and in-person engagement. The final survey was piloted with a small group of parents who were unfamiliar with the FRAISE project. The survey was launched online using Qualtrics XM Survey Software © (Qualtrics, Provo, UT) in May 2018 and promoted via Facebook and Twitter. In-person survey completion sessions were also held at various inner-city community and healthcare agencies to increase the inclusion of under-represented populations, particularly those without reliable internet access.

Priority setting workshops and consensus building

Three additional in-person priority setting workshops were hosted with the steering committee to build consensus around survey development, survey response theming, the top 30 list, and the top 10 list (Figure 1). Each session was structured using a nominal group technique.[17] To analyze survey responses and identify top research priorities, core steering committee members were broken into six groups and provided with survey responses from four of the 12 topic areas. Each author acted as a group facilitator to guide the work and ensure that all voices were heard, but not to direct the content of the discussion. Each group member was provided with a question from the survey results and given time to silently generate ideas. Then, each group member shared their ideas; the facilitator clarified and recorded them. Following group discussion to clarify ideas and priorities, group members voted and ranked each priority, with the top priorities moved forward to the steering committee for consideration. Consensus building concluded with discussion and agreement on which content to move forward from each session. After each in-person session, this process was repeated, using survey software with the broader online steering committee to develop consensus. In the spirit of true consensus building and PSP, researchers facilitated sessions allowing steering committee members to drive decisions and finalize results.[17] As such, modifications were made to the wording of the final top 10 list, resulting in an additional (11th) research priority.[13]

≪INSERT Figure 1: FRAISE Study Flow Diagram HERE≫

Role of the funding source

The funders of the study had no role in study design, data collection, data analysis, or report writing. All authors had access to study data. The corresponding author, EMK, had full access to all data and responsibility for the final decision to submit for publication.

Data analysis

Characteristics of survey participants were calculated using descriptive statistics in Microsoft Excel. Responses from the survey were captured verbatim in each of the 12 topic areas. All individual responses were printed and cut into individual items. In-person steering committee members (n = 18) worked together to group responses into themes, within each topic area. Responses from each topic area were analysed by two separate groups to validate theming. Researchers then built the resultant themes (research priorities) into a codebook. Research and practice experts provided feedback on the extent to which each of the 202 codes had been researched. These experts were asked to indicate if each potential research priority was: (1) well researched (systematic reviews and meta-analysis available), (2) somewhat researched (single studies, some inconsistent evidence on topic), (3) not researched (no studies), or (4) unsure. Experts were encouraged to provide comments or feedback on each of the potential priorities. This information, in conjunction with the 202 codes, was provided to the steering committee during a third consensus building workshop (n = 21) to develop the top 30 list. Steering committee members were instructed to review and consider expert feedback when building consensus around which of the 30 priorities to move forward. While the frequency that a particular priority was submitted in the online survey was considered important for subsequent prioritization, steering committee members did not rely solely on these counts to move items forward to the top 30 and top 10 lists. While the original intent was to narrow down the priorities to a top 30 list, at the time of prioritization, the steering committee reached a consensus that the list should include 34 priorities.

The top 34 list was shared with the broader online steering committee and committee members (n = 25) were asked to select and rank their top 10 selections. These results were brought back to the fourth and final consensus building workshop (n = 18) to develop the top 10 priorities. The final top 10 (plus one) list was circulated online for final approval. Steering committee members were also asked to rank the top 10 (plus one) priorities on their perceived order of importance.

Data Statement

The research priorities codebook, containing the raw research priorities responses, is available upon reasonable request from the corresponding author.

Patient and Public Involvement Statement

The public was engaged via membership in the steering committee after the study and research objectives were conceptualized, and funding was obtained. Steering committee members designed the survey and participated in data collection via disseminating the survey through their professional and social networks and provided advice on how to increase survey response rates for diverse groups. These members also provided feedback on the survey regarding participant burden and conducted data analysis in collaboration with the research team. In addition, steering committee members were sponsored to participate in local, regional, and national study dissemination via virtual and in-person child health rounds and conferences, as well as manuscript authorship (LC, SD).

Results

In total, 596 participants consented to participate in the survey, of which 480 completed the survey. Of these, the majority were parents (76.3%; n = 130 tablet; n = 236 online), 16.0% were clinicians (n = 3 tablet; n = 74 online), 4.8% were early childhood educators (n = 10 tablet; n = 13 online), and 2.9% were community agency representatives (n = 0 tablet; n = 22 online). Most participants were recruited via social media and electronic networks (n = 337, 70.2%), with 29.8% (n = 143) recruited during targeted in-person sessions where participants were provided with a tablet to record responses. The mean age of all survey respondents was 34.1 years (SD = 9.1), with tablet-based respondents having a slightly younger mean age in years (32.1; SD = 5.9) than the mean age of online-based respondents (34.8; SD = 9.9). For parent participants who were not born in Canada (n = 65), the mean number of years in Canada was 10.7 (SD = 11.4), with tablet-based respondents having fewer mean years in Canada (M = 8.1; SD = 9.3) than online-based respondents (M = 13.6; SD = 13.0). Table 1 describes participant and household characteristics. The socioeconomic and ethnicity distribution of parent participants was comparable to population distributions.[16] Participation rates mirrored geographic population densities (Figure 2).

Table 1. FRAISE Survey participant characteristics.

	Total Completed (N = 480)		Online Survey (n = 337)		Tablet Survey (n = 143)	
All respondents	N	%	n	%	n	%
Employment status						
Working for pay, profit, or self-employed	233	48.5	182	54.0	51	35.7
Caregiving (including parental or maternity leave)	183	38.1	122	36.2	61	42.7
Not working, but looking	14	2.9	9	2.7	5	3.5
Going to school, retired, cannot work due to disability or illness or other	30	6.3	10	3.0	20	14.0
Completed post-secondary education (e.g., certificate or diploma program, undergraduate/graduate degree)	404	84.2	306	90.8	98	68.5
Ethnicity*						
Caucasian	333	69.4	256	76.0	77	53.8
Chinese	29	6.0	14	4.2	15	10.5
South Asian	14	2.9	10	3.0	4	2.8
Latin American	12	2.5	7	2.1	5	3.5
Indigenous	25	5.2	6	1.8	19	13.3
Caribbean	7	1.5	6	1.8	1	0.7
Filipino	6	1.3	4	1.2	2	1.4
Arab	5	1.0	3	0.9	2	1.4
Korean	4	0.8	3	0.9	1	0.7
Southeast Asian	7	1.5	3	0.9	4	2.8
Japanese	2	0.4	2	0.6	1	0.7

African	8	1.7	1	0.3	7	4.9
West Asian	2	0.4	0	0.0	2	1.4
Other	14	2.9	8	2.4	6	4.2
Prefer not to say	15	3.1	9	2.7	6	4.2
Clinicians	77	16.0	74	22.0	3	2.1
Nurse	47	9.8	47	13.9	0	··
Physician	7	1.5	7	2.1	0	··
Dietician	3	0.6	3	0.9	0	··
Occupational therapist	3	0.6	3	0.9	0	··
Other (midwife, social worker, etc.)	17	3.5	14	4.2	3	2.1
Community agency representative	14	2.9	14	4.2	0	··
Early childhood educator/care provider (e.g., nanny, daycare, preschool)	23	4.8	13	3.9	10	7.0
Parent	366	76.3	236	70.0	130	90.9
Mother	292	60.8	194	57.6	98	68.5
# pregnant	*136*	28.3	*85*	25.2	*51*	35.7
Father	71	14.8	40	11.9	31	21.7
Other (e.g., grandparent, aunt)	3	0.6	2	0.6	1	0.7
Parent respondents only (n = 366)						
Partnered	305	83.3	206	87.3	99	76.2
Number of households with children						
1 child in household	173	47.3	121	51.3	52	40.0
2 children in household	74	20.2	48	20.3	26	20.0
3 children in household	27	7.4	15	6.4	12	9.2
4^+ children in household	13	3.6	4	1.7	9	6.9
Born in Canada (Yes)	280	76.5	190	80.5	90	69.2
English as primary household language	318	86.9	206	87.3	112	86.2
Household income (yearly)						
Less than $40,000	66	18.0	17	7.2	49	37.7
$40,000 - $79,999	82	22.4	64	27.1	18	13.8
$80,000 - $119,999	69	18.9	54	22.9	15	11.5
$120,000 - $159,999	49	13.4	34	14.4	15	11.5
More than $160,000	53	14.5	35	14.8	18	13.8
I don't want to say	31	8.5	19	8.1	12	9.2
Note: Due to missing data, numbers in this table may not sum to the total number of completed responses; *indicates respondents could choose all that apply						

≪INSERT Figure 2. Geographic distribution of responses HERE≫

In total, we received 3232 submissions of potential research priorities spread across all 12 topic areas (Figure 3). When asked to rank the 12 broad parenting topics of interest, survey participants ranked stress, emotional, and mental health; sleep; and infant feeding as the three most important parenting topics. The 3232 suggested priorities obtained via survey responses were themed and collapsed by the steering committee, resulting in 202 unique priorities (Figure 1) of which 34 were shortlisted for consideration in the top 10 list (Supplementary Table).

Steering committee members used this shortlist to build consensus on the final top 10 (plus one) list of research priorities (Table 2).

≪INSERT Figure 3. Number of raw submissions in each topic area from provincial survey HERE≫

Table 2. Final list and topic areas of the top 10 (plus one) research priorities from conception to two years of age for families in the community.

Rank	Question	Topic Area
1	How can families be supported to develop healthy coping strategies, emotional regulation, and resiliency in both parents and children?	Mental Health/Relationships
2	How can families access supportive care and information when things do not go as expected during pregnancy, labour, birth, or postpartum?	Access to Information/ Pregnancy, Labour & Birth
3	How can the healthcare system and providers ensure access to prevention and treatment of mental health concerns in a safe and trusting environment?	Mental Health
4	How do families navigate multiple sources of health information and access services tailored to their specific circumstances?	Access to Information
5	What supports and services can be developed at the individual, family, community, and government levels to build emotional/mental wellbeing, physical health, and healthy relationships?	Mental Health/Relationships
6	How can eczema, asthma, and allergies be more effectively prevented, assessed, and treated?	Immunity
7	In a constantly changing social and physical environment, how can families increase safety and manage developmentally appropriate risk-taking?	Child Development/Child Safety/Environmental Risk
8	How can sleep problems be prevented, assessed, and treated in a culturally appropriate way that is tailored to individual families?	Sleep
9	How can families be better supported to make informed, family-centred feeding decisions?	Feeding
10	How can families be better supported to promote healthy child development, recognize milestones, and access services for delay?	Child Development
11	With increasing vaccine hesitancy, how can individual, family, and population health best be protected?	Immunity

Discussion

To our knowledge, this is the first attempt to identify community-based research priorities for families of well children from conception to age 24 months. Throughout all steps of the PSP, concerns for mental, social, and emotional well-being of parents and children were consistently expressed as priority areas. Comparing the top 10 (plus one) priorities with the initial 12 survey categories, we found that stress, emotional, and mental health were consistently a top concern for families and were identified in three of the top 10 (plus one) priorities. Additionally, parents repeatedly specified the need for structural health system changes at the individual, family, community, and government levels to focus on building emotional/mental well-being, physical health, and healthy relationships – taking the onus off individual families to solve problems related to mental and physical wellness and prioritizing policy and systems shifts. As such, these research priorities highlight the need to situate individual- or family-focused research priorities within the context of complex health and social systems and involve multiple sectors such as government and health boards.

Across themes, the top 10 (plus one) research priorities reflect the importance of fostering access to information and developing information and services using a culturally sensitive framework. While identified as its own research priority (Table 2), concerns about access to information emerged in each topic area. The ubiquitous nature of access to information as a research priority raises a fundamental question about the accessibility of safe, reliable, and valid information for young families. Previous research from Canada indicates the majority of parents use the internet to access information about children's health, but many use unreliable sources of information.[18] In contrast, Australian women who are pregnant most often reported discussion with their midwife as an information source, while less than half reported using the internet to access information; group information sessions were the least preferred information sources.[19] This contrasts with a report from Devolin and colleagues[20] where Albertan parents rated the Internet (55.3%) and drop-in programs (42.9%) as preferred information sources. With such conflicting evidence, it is unsurprising that researchers and care providers have long struggled with providing safe and reliable information related to child and infant health. Healthcare providers and administrators should collaborate with researchers and parents to critically examine health information provision and consider transformative frameworks to create meaningful and innovative knowledge translation strategies to better communicate evidence.

Several research questions related to physical health emerged as important. For example, parents expressed concerns about how eczema, asthma, and allergies could be more effectively prevented, assessed, and treated; with this class of inflammatory disorders as one of the most common issues in early childhood, this is unsurprising. The fact that parents shared a strong desire for information about preventing these disorders, suggests this is a research area ripe for future development. Additionally, questions emerged about preventing injury while encouraging appropriate physical development and accessing information about culturally appropriate feeding and sleep. While some research exists in each of these areas, evidence is generally underdeveloped, and information needs to be communicated to parents in culturally appropriate and meaningful ways. Of particular importance given the emerging pandemic, parents were unwilling to leave the final steering committee workshop without including a research priority related to vaccine hesitancy. Diverse opinions emerged around this topic, and it became clear that evidence-based information about vaccines and immunity was a valued priority.

Findings from this PSP complement findings from other pediatric research PSPs that have focused on single issue priority setting, such as premature birth,[21] stillbirth,[22] chronic pain,[23] neurodisabilities,[24] or learning difficulties.[25] Despite the breadth in populations and that most existing PSPs were conducted in the UK, research priorities identified in these partnerships related to access to information, mental and emotional health, and healthy relationships, suggesting that these themes are consistently important to parents.

One notable exception to the general trend of conducting PSPs with specialized populations is a Canadian PSP that developed research priorities for preventative care of children aged zero to five years.[26] Consistent with our top research priority of mental and emotional health in children and their parents, the top research question found by Lavigne and colleagues was "What are effective strategies for screening and prevention of mental health problems?" (p.750).[26] Our findings confirm and extend this priority of building mental and emotional wellness in children belonging to younger age groups (and during conception) by broadening the focus to include parents and protective factors for resilience. This suggests that parent, clinician, and researcher priorities are aligned with the emergence of infant mental health (ages zero to five) as a relational concept, whereby infant mental health is supported by optimizing the mental health and resiliency of children's caregivers (i.e. parents and family members).[1] Other similarities included: (1) supporting child development by improving identification and services for developmental delays; (2) developing effective interventions, supports, and services to improve mental health, physical health (obesity, physical activity), and healthy relationships (social skills); and (3) the impact and support of nutritional and feeding factors. Contrasts with our research priorities and those for children aged zero to five years included: (1) the impact of daycare attendance on child health, (2) behavior management in children, and (3) appropriate screen time for children. In addition, research priorities in our list that were not found in the list for children aged zero to five years included: (1) how families can access supportive care and information during unexpected experiences in the perinatal period; (2) access to information; (3) how eczema, asthma, and allergies can be more effectively prevented, assessed, and treated; (4) injury prevention and appropriate levels of risk; and (5) sleep problems in families. It should be noted that differences may represent prioritization related to different developmental stages and ages (i.e., screen time is not recommended for children under age two) and/or slight differences in the prioritization method. For example, helping families identify appropriate childcare appeared in our top 34 list and aligns with a similar research priority related to the impact of daycare attendance in the zero to five years list.

In a Delphi study of perceived research priorities of clinical staff at an Australian parenting centre, Hauck and colleagues[27] determined the top research priorities to be related to short- and long-term evaluation of the effectiveness of the centre's programs on children's behavior and stress levels, parental expectations, healthy family relationships, as well as parental use of sleep, settling, and feeding strategies. Determining the effectiveness of the centre's programme in the context of postpartum depression was also an identified priority.[27] Although these research priorities were not informed by parental participation and may have limited generalizability to other centres and/or countries, it is notable that issues related to sleep, infant feeding, healthy relationships, and parental mental health are captured by both ours and Hauck's[27] lists. This is not surprising as a study in the same geographical area suggested that breastfeeding, sleep issues, child development, helping children cope with emotions, and understanding child brain development were all in the top 10 (plus one) parenting topics ranked as somewhat or very important by parents.[20]

Clinical and research applications

To move forward with these research priorities, it is evident that multiple stakeholders must be engaged, and connections made across a variety of sectors and disciplines. Wording for the top 10 (plus one) research priorities requested consideration of “culturally appropriate” and “safe and trusting environments.” Our findings highlight the need for an increased emphasis on parenting education and culturally appropriate strategies to support mental health and well-being. Adopting an intersectional lens when conducting research, and providing care to pregnant and young families, will better address these considerations and provide meaningful evidence to inform health outcomes. Similar to how parents of diverse groups may have differing or intensified concerns relating to child health issues,[28] it is important to recognize that research priorities may differ based on community member’s characteristics, specifically in terms of race, income, access to health services, and experiences, including racism. These top 10 (plus one) priorities were developed using an intersectional framework. Moving forward, research in these priority areas must continue to comprehensively and meaningfully include individuals with diverse socioeconomic backgrounds and experiences.[9]

The findings from this study should be considered in the context of its strengths and limitations. In terms of strengths, there was a large sample size for the online survey comparable to other PSPs; this created space for a broad representation of stakeholders including clinicians, community agency representatives, parents, and ethnically diverse representatives of the study location. Limitations included the inherent and individual biases that self-selected, voluntary researchers and steering community members view the world with, which may reduce the generalizability of the final priority list. Given that the research team was composed entirely of white heterosexual women and although attention was paid to ensure participation of fathers, individuals self-identifying as non-white, and those in non-heterosexual or single parent partnerships, these voices may not have been entirely represented in the final priorities. Additionally, while this research PSP used a process based on a previously reported modified JLA approach,[13] these modifications have not been validated against the standardized JLA method. These modifications (e.g., using external group facilitators with expertise in patient engagement to establish the steering committee rather than a paid JLA advisor; consulting with experts on the level of evidence, rather than conducting rapid literature reviews on each potential research uncertainty) greatly reduced the cost of the project, thereby increasing feasibility. Additionally, the current approach incorporated principles of participatory action research (e.g., engaging with a community who have self-identified in reciprocal relationships, recognition of local knowledge, and incorporating processes of co-learning to take actions that will improve community member's wellbeing), which are widely recognized as critical in facilitating meaningful participant engagement and ensuring that participants' voices are represented throughout the research process.

Researching these family-driven priorities will reduce the knowledge-to-practice gap and generate opportunities to improve services for families, including innovative delivery models and evidence-based treatment options to enable equitable access to services. Further, involving practitioners in the PSP may enhance the implementation of new evidence-based practice recommendations.[29] These outcomes, combined with decreased lag time from research to practice, may result in improved child and family outcomes. As a result of our strong collaborative partnerships and inclusion of policymakers in each stage of the PSP, the provincial health service has already utilized this information to streamline and enhance evidence-based information delivery to parents and care providers in the province. By sharing the FRAISE top

10 (plus one) research priorities, we invite other researchers to ensure their work aligns with patient-oriented research as a foundation for future initiatives targeted at improving outcomes in families with young children.

Author Contributions

EMK and MB drafted the manuscript, which all authors reviewed, provided feedback, and approved the final version. EMK, MB, KSB, CG, and KMB designed the study. EMK, MB, KSB, CG, KMB, and LTM developed the steering committee workshop content. EMK, MB, KSB, CG, KMB, LTM, SD, LC and the Steering Committee devised the data collection instruments. JW, EMK, MB, KSB, and CG were responsible for data collection and database design and management. EMK, MB, KSB, CG, KMB, LTM, JW, SD, LC, and the Steering Committee completed the data analyses.

Declarations of interests

We declare no competing interests.

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Ethics Statement

Patient consent for publication

Not required

Ethics approval

The Conjoint Health Research Ethics Board at the University of Calgary (REB17-0014) approved this research.

References

1. Shonkoff JP. Capitalizing on advances in science to reduce the health consequences of early childhood adversity. *JAMA Pediatr* 2016; **170**: 1003–7.
2. Bronfenbrener, U. (ed.). Making human beings human: bioecological perspectives on human development. Thousand Oaks, CA: Sage, 2005.
3. Wall G. Mothers' experiences with intensive parenting and brain development discourse. *Womens Stud Int Forum* 2010; **33**: 253–63.
4. Albrecht L, Archibald M, Snelgrove-Clarke E, Scott SD. Systematic review of knowledge translation strategies to promote research uptake in child health settings. *J Pediatr Nurs* 2016; **31**: 235–54.
5. Chalmers I, Glasziou P. Avoidable waste in the production and reporting of research evidence. *Obstet Gyneco*l 2009; **114**: 1341–45.
6. Manafò E, Petermann L, Vandall-Walker V, Mason-Lai P. Patient and public engagement in priority setting: a systematic rapid review of the literature. *PLoS One* 2018; **13**: e0193579.
7. Chalmers I, Bracken MB, Djulbegovic B, et al. How to increase value and reduce waste when research priorities are set. *Lancet* 2014; **383**: 156–65.
8. Shimmin C, Wittmeier KDM, Lavoie JG, Wicklund ED, Sibley KM. Moving towards a more inclusive patient and public involvement in health research paradigm: the incorporation of a trauma-informed intersectional analysis. *BMC Health Serv Res* 2017; **17**: 539.
9. Bailey J, Steeves V, Burkell J, Shade LR, Ruparelia R, Regan P. Getting at equality: research methods informed by the lessons of intersectionality. *Int J Qual Methods* 2019; **18**: 1–13.
10. Mann BS, Manns BJ, Dart A, et al. An assessment of dialysis provider's attitudes towards timing of dialysis initiation in Canada. *Can J Kidney Health Dis* 2014; **1**: 3.
11. Cowan K, Oliver S. The James Lind Alliance guidebook. 2010. http://www.jlaguidebook.org/pdfguidebook/guidebook.pdf (accessed Nov 22, 2016).

12. Bright KS, Ginn C, Keys EM, et al. Study protocol: determining research priorities of young Albertan families (The Family Research Agenda Initiative Setting Project-FRAISE)-participatory action research. *Front Public Health* 2018; **6**: 228.
13. Breault LJ, Rittenbach K, Hartle K, et al. The top research questions asked by people with lived depression experience in Alberta: a survey. *CMAJ Open* 2018; **6**: E398–405.
14. Barnieh L, Jun M, Laupacis A, Manns B, Hemmelgarn B. Determining research priorities through partnership with patients: an overview. *Semin Dial* 2015; **28**: 141–46.
15. Tong A, Synnot A, Crowe S, et al. Reporting guideline for priority setting of health research (REPRISE). *BMC Med Res Methodol* 2019; **19**: 243.
16. Statistics Canada. 2011 National Household Survey: Statistics Canada. 2013. https://www12.statcan.gc.ca/nhs-enm/2011/dp-pd/dt-td/Index-eng.cfm (accessed Oct 7, 2020).
17. Carney O, McIntosh J, Worth A. The use of the nominal group technique in research with community nurses. *J Adv Nurs* 1996; **23**: 1024–29.
18. Pehora C, Gajaria N, Stoute M, Fracassa S, Serebale-O'Sullivan R, Matava CT. Are parents getting it right? A survey of parents' internet use for children's health care information. *Interact J Med Res* 2015; **4**: e12.
19. Grimes HA, Forster DA, Newton MS. Sources of information used by women during pregnancy to meet their information needs. *Midwifery* 2014; **30**: e26–33.
20. Devolin M, Phelps D, Duhaney T, et al. Information and support needs among parents of young children in a region of Canada: a cross-sectional survey. *Public Health Nurs* 2013; **30**: 193–201.
21. Duley L, Uhm S, Oliver S. Top 15 UK research priorities for preterm birth. *Lancet* 2014; **383**: 2041–42.
22. Heazell AEP, Whitworth MK, Whitcombe J, et al. Research priorities for stillbirth: process overview and results from UK Stillbirth Priority Setting Partnership. *Ultrasound Obstet Gynecol* 2015; **46**: 641–47.
23. Birnie KA, Dib K, Ouellette C, et al. Partnering for pain: a priority setting partnership to identify patient-oriented research priorities for pediatric chronic pain in Canada. *CMAJ Open* 2019; **7**: E654–64.
24. Morris C, Simkiss D, Busk M, et al. Setting research priorities to improve the health of children and young people with neurodisability: a British Academy of Childhood Disability-James Lind Alliance research priority setting partnership. *BMJ Open* 2015; **5**: e006233.
25. Lim AK, Rhodes S, Cowan K, O'Hare A. Joint production of research priorities to improve the lives of those with childhood onset conditions that impair learning: The James Lind Alliance priority setting partnership for 'learning difficulties'. *BMJ Open* 2019; **9**: e028780.
26. Lavigne M, Birken CS, Maguire JL, Straus S, Laupacis A. Priority setting in paediatric preventive care research. *Arch Dis Child* 2017; **102**: 748–53.
27. Hauck Y, Kelly RG, Fenwick J. Research priorities for parenting and child health: a Delphi study. *J Adv Nur* 2007; **59**: 129–39.
28. Garbutt JM, Leege E, Sterkel R, Gentry S, Wallendorf M, Strunk RC. What are parents worried about? Health problems and health concerns for children. *Clin Pediatr* 2012; **51**: 840–47.
29. Mathieson A, Grande G, Luker K. Strategies, facilitators and barriers to implementation of evidence-based practice in community nursing: a systematic mixed-studies review and qualitative synthesis. *Prim Health Care Res Dev* 2019; **20**: e6.

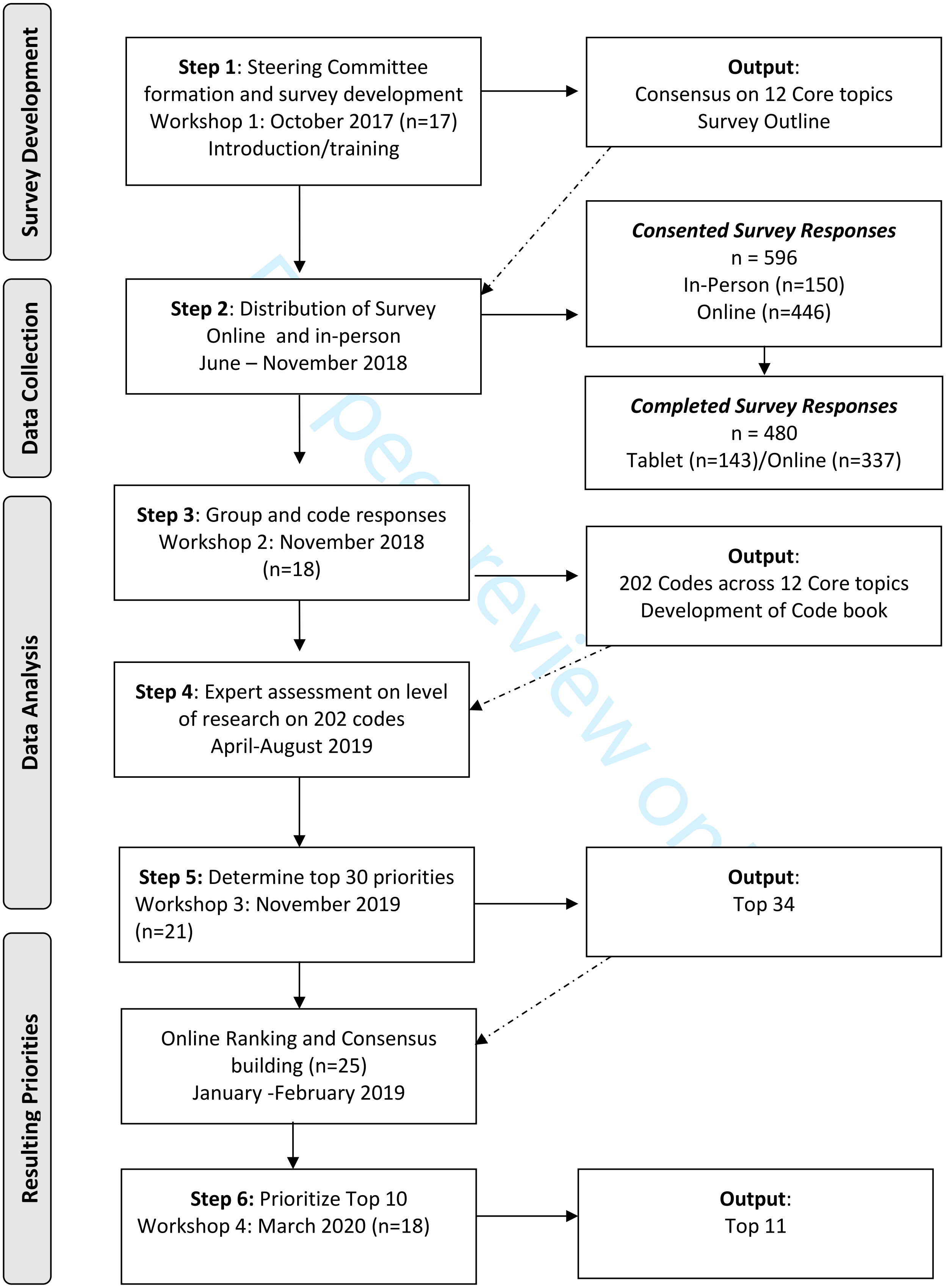
Figure Captions

Figure 1. FRAISE Study Flow Diagram

Figure 2. Geographic distribution of responses

Figure 3. Number of raw submissions in each topic area from provincial survey

FRAISE Study Flow Diagram



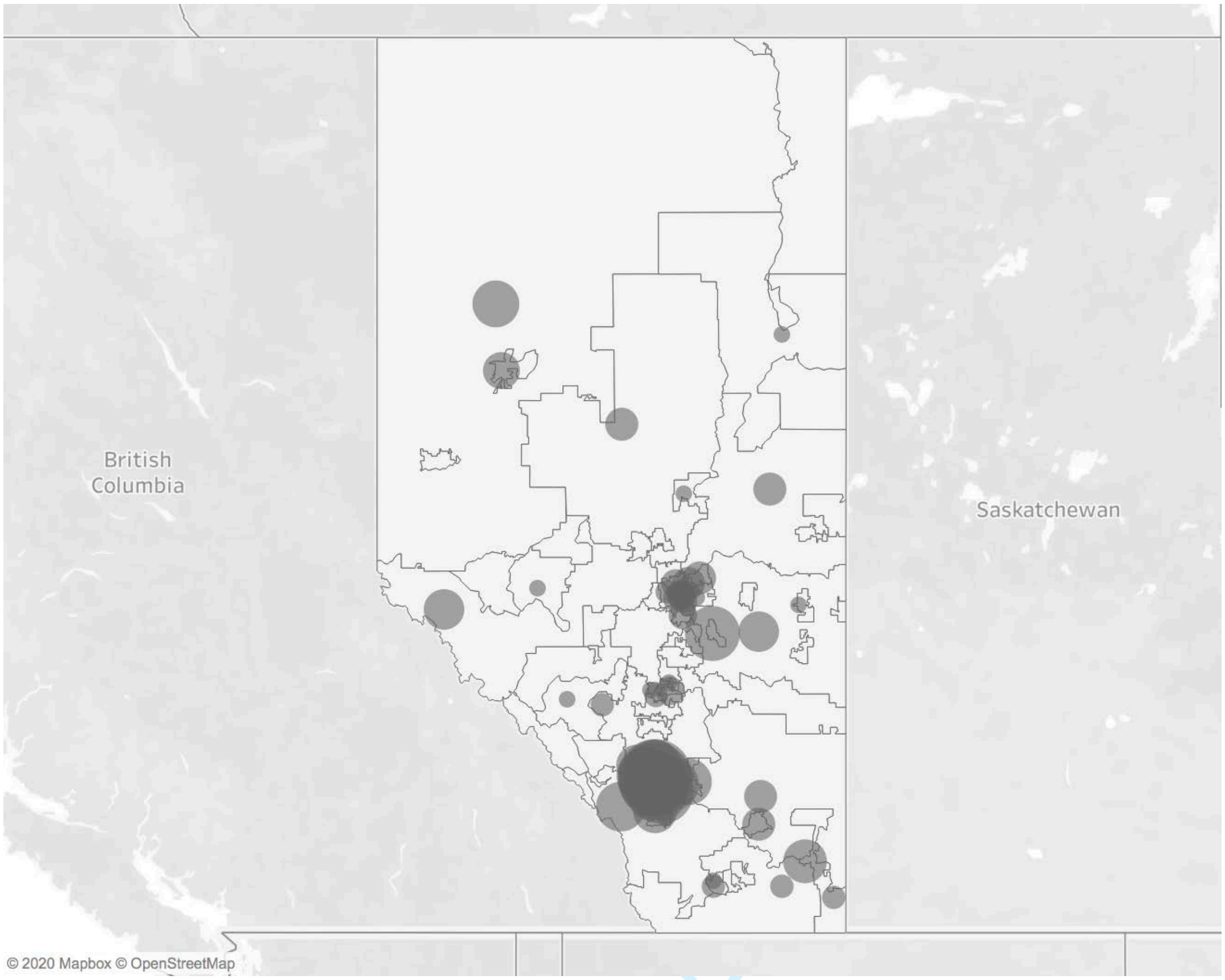


Figure 2. Geographic distribution of responses

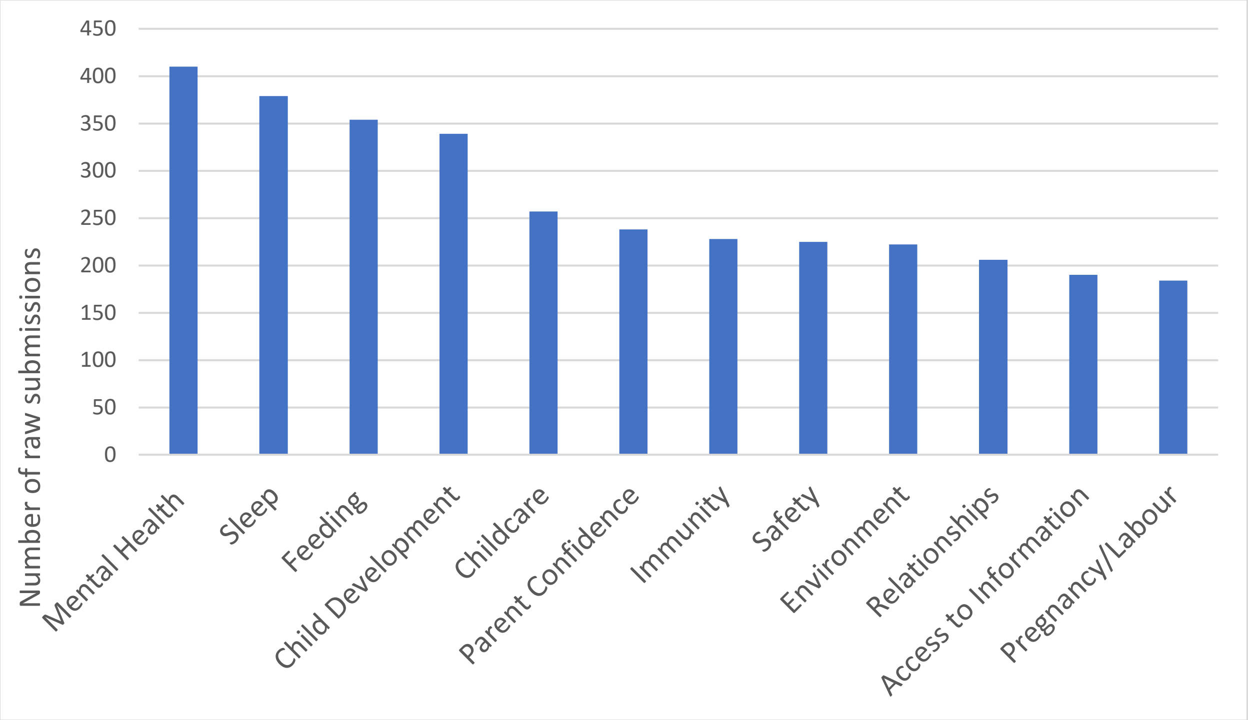


Figure 3. Number of raw submissions in each topic area from provincial survey

Supplemental Table. Top 34 Themes

Question	# of times in top 10
How can families and caregivers of children 2 and under be supported to develop healthy coping strategies, emotional regulation, and resiliency in both themselves and their children?	15
How to access information/programs and navigate the healthcare system when you are from a minority/non-dominant group?	14
What is an age appropriate level of risk taking for children to promote their growth & development while maintaining adequate (good enough) safety?	12
How do parents manage multiple sources of information to make health decisions that are tailored to their specific circumstances and needs?	12
How can families (partners & extended family members), communities, and government encourage self-care and provide emotional and practical support for parents during the perinatal and early childhood period?	11
When things don't go as planned, how can parents get care, support, and information during pregnancy, labour, birth or the postpartum period?	11
What formats (i.e. in-person, online, group) do families prefer when accessing supports and services to develop and sustain healthy relationships and mental well-being?	10
How can health care professionals provide a safe and trusting environment that encourages women/families to disclose mental health concerns without fear of negative repercussions?	10
How to identify, prevent, and treat eczema, asthma, and allergies?	10
How do we create culturally appropriate, family specific information about sleep (co-sleeping, swaddling, sleep training, etc.) from conception to age 2?	10
What are the advantages and disadvantages of using a trauma-informed model of care to explore pregnancy, childbirth, and postpartum for all parents?	9
How do we prevent, assess, and treat sleep problems in parents and children?	9
What supports and services are required to support families to develop and sustain healthy relationships and mental wellbeing?	8
To minimize physician visits, what user centred information sources and formats are most acceptable and effective to provide parents and healthcare providers with reliable and up-to-date information?	8
How can we best support families with their infant feeding decisions?	7
With increasing vaccine hesitancy, how do we best protect individual and population health?	7
How can families during the perinatal and early childhood period access appropriate and evidence-based assessments, prevention, treatment, and support for mental health concerns?	7
How can we manage the effect of exposure to cannabis smoking to families with young children?	7
In children 2 and under, how can families and caregivers be supported around milestones, behaviours, and signs of delay?	7
In addition to vaccination, how can parents and children build their immunity?	6
What supports and services increase general parental confidence?	6
How can families during the perinatal and early childhood period manage emotional and physical wellbeing?	6
What are the necessary measures to increase safety for toddlers/children in an ever changing social and physical environment (during the early childhood period)?	6
How can communities and health care professionals provide culturally sensitive best practices for promoting healthy pregnancies & fetal/infant/child development?	6
How can we increase awareness and access to additional services and resources for families of young children?	6
What are expected and problematic sleep patterns in caregivers, infants, and toddlers?	6

What evidence-based community resources and services are available to help foster healthy relationships for expecting parents?	5
How can families be supported to identify the right fit of childcare, looking at quality, safety, and affordability while still incorporating their own beliefs and values?	5
How do we build on general confidence to generate transferable skills for parenting?	4
What are the consequences of sleep deprivation and problems in parents and children?	4
What are effective strategies for accessing/using my electronic health record and keeping track of my family's health information?	3
What should healthcare providers recommend for guidelines to support families to transition from milk feeding to solid foods?	2
How does infant feeding impact allergies?	2
How can parents during early childhood identify & prevent child safety risks?	2

Reporting Guideline for Priority Setting of Health Research (REPRISE)[1]

#	Item	Descriptor
Context and Scope		
1	Define geographical scope	Regional geographic area "within a province in Western Canada" (methods, pg. 7).
2	Define health area, field, focus	Focussed on a specific population of healthy young families and caregivers in the community setting. The population of interest is "families of well children from conception to age 24 months" (introduction, pg. 6).
3	Define the intended beneficiaries	Intended beneficiaries of these findings include researchers, care providers, community services representatives, and families themselves. "Develop research outcomes meaningful to knowledge users, parents, and care providers" (introduction, pg. 6).
4	Define the target audience of the priorities	The target audience who has the potential to fund future research or implement the identified priorities include care-providers, health services organizations, and researchers. "As a result of our strong collaborative partnerships and inclusion of policy makers in each stage of the PSP, the provincial health service has already utilized this information to streamline and enhance evidence-based information delivery to parents and care providers in the province. By sharing the FRAISE top 11 research priorities, we invite other researchers to ensure their work aligns with patient-oriented research as a foundation for future initiatives targeted at improving outcomes in families with young children." (discussion, pg. 18).
5	Identify the research area	The broad research area of the priority setting research encompasses health services and public health. "these family-driven priorities will reduce the knowledge to practice gap and result in higher quality health care services for families of young children" (discussion, pg. 17).
6	Identify the type of research questions	The types of research questions were broad, and ranged from etiology (i.e., asthma, eczema, allergy causes/diagnosis) to implementation (i.e., developing community supports). Questions also addressed psychosocial and behavioural research priorities. The researchers did not pre-define the type of research questions that were expected to be generated from the priority setting partnership. See Table 3 for top 11 research priority questions.

7	Define the time frame	The researchers outline the timeframe during which the research was conducted “between November 2017 and March 2020” (methods, pg. 8), however, the anticipated relevance of the research priorities is not addressed. Plans for to monitor or update the priorities with future prioritization activities are not discussed.
	Governance and Team	
8	Define the selection and structure of the leadership and management team	The priority setting was facilitated by the authors, however, the structure of the research team, relevance for inclusion, or technical expertise as a facilitator is not addressed. “Each author acted as a group facilitator to guide the work, but not the content of the discussion” (methods, pg. 9). A steering committee (smaller in-person group and larger online group) completed the prioritization process. The selection of the committee members and structure of the team is not outlined. Several groups are mentioned in the acknowledgements paper for contributing to the committee, but their role is not elaborated on in the manuscript. The authors simply state, “we engaged a steering committee comprised of researchers, parents, clinicians, and community agency representatives” and “core group included 15 parents, four clinicians and two community agency representatives; a larger online group of steering committee members also provided feedback and consultation throughout the priority setting process” (methods, pg. 8). The characteristics of, or any differences between, the in-person steering committee members and online members are not explicated. The need for participant diversity is noted, “We aimed for 30 steering committee members who mirrored the regional population, including a minimum of 3% who were First Nations, Metis, or Inuit and 30% who identified as immigrants and/or visible minorities”, however, whether or not the goal for diversity was fulfilled is not described. The authors do note the limitation that the study may not be generalizable to all populations, “although attention was paid to ensure participation of fathers, individuals self-identifying as non-white, and those in non-heterosexual or single parent partnerships, these voices may not have been entirely represented in the final priorities” (discussion, pg. 17).
9	Describe the characteristics of the team	The demographics of the steering committee (on-line and in-person) are not presented. Furthermore, the affiliations or expertise of the researchers, clinicians, and community agency representatives are not presented.

		The authors do recognize that the research team was "composed entirely of white heterosexual women" (discussion, pg. 17), and also present the demographics of the broader sample who completed the survey to identify the initial parent questions that informed the final 11 themes, "Table 1 describes participant and household characteristics. The socioeconomic and ethnicity distribution of parent participants was comparable to population distributions. Participation rates mirrored geographic population densities." (results, pg. 11).
10	Describe any training or experience relevant to conducting priority setting	The presence of participant or researcher training, consultants, or past experience with priority setting research is not addressed in the body of the manuscript. However, the authors do acknowledge the "Alberta Strategy for Patient-Oriented Research Support Unit for their expertise, guidance and support in the developmental stages of this project" within the acknowledgement section.
Framework for Priority Setting		
11	State the framework used (if any)	The authors identify the use of "a consensus-building and strengths-based approach, modelled after participatory action research and the James Lind Alliance" (methods, pg. 7). The authors provide sound rationale for their use of a modified James Lind Alliance approach, recognizing that the conventional methodology is "resource intensive and may be impractical for lower resourced research studies and vulnerable populations" (methods, pg. 7).
Stakeholders and Participants		
12	Define the inclusion criteria for stakeholders involved in priority-setting	As mentioned above, the authors sought to include a diverse group of stakeholders who accurately represented the population of the research setting. Specific inclusion criteria or the process of identifying, approaching, and engaging stakeholders is not outlined. More broadly for the survey participants, efforts to engage diverse populations was achieved through in-person survey recruitment in conjunction with online surveys.
13	State the strategy or method for identifying and engaging stakeholders	The strategy for identifying stakeholders in the steering committee are not addressed in the body of the manuscript. Again, several groups and existing partnerships are noted in the acknowledgements for their contribution to the committee, however, strategies for stakeholder engagement are not outlined.
14	Indicate the number of participants and/or organizations involved	The authors briefly describe the number of stakeholders involved in priority setting, "we engaged a steering committee comprised of researchers, parents, clinicians, and community agency representatives" and "core group included 15 parents, four clinicians and two community agency representatives; a larger online group of steering committee members also provided

		feedback and consultation throughout the priority setting process" (methods, pg. 8). Although the number of in-person committee members is clear, there is no number provided for the larger online steering committee group. Furthermore, the organizational affiliation (or the nature of the organization, i.e., health, government etc.) of researchers, clinicians, or community representatives is not described.
15	Describe the characteristics of stakeholders	As noted above, apart from the number of steering committee members who were parents, clinicians, and community agency representatives in the in-person group, there is limited explanation of the steering committee characteristics.
16	State if reimbursement for participation was provided	The authors clearly state, "Parent participants were given a $50 honorarium, childcare, and meals at each session" (methods, pg. 7).
Identification and Collection of Research Priorities		
17	Describe methods for collecting initial priorities	The development of initial priorities appears to be two-fold. The authors and steering committee developed an initial list of 12 parenting topics, which then informed a survey where parents could ask questions about each of the 12 parenting topics. These parent questions relative to the 12 topics would then be analyzed to create the final top 11 priority list (methods, pg. 7-8). The authors do not elaborate how the initial 12 parenting topics were selected, apart from stating, "our steering committee collaboratively identified 12 broad parenting topics of interest" (methods, pg. 8). However, it is noted that the survey generated from the 12 topic areas was refined in collaboration with steering committee members, both online and in-person, and then pilot tested with "outside" parents. The subsequent data analysis of the themes and questions resulting from the provincial survey took place within in-person steering committee workshops "structured using nominal group technique" (methods, pg. 9), and through online consensus, "After each in-person session, this process was repeated, using survey software with the broader online steering committee to develop consensus" (methods, pg. 9).
18	Describe methods for collating and categorizing priorities	The initial broad priority suggestions and research questions were thematically grouped and analyzed through in-person steering committee workshops. "All individual responses were printed and cut into individual items. In-person steering committee members worked together to group responses into themes, within each topic area. Reponses from each topic area were analysed by two separate groups to

		validate theming. Researchers then built the resultant themes (research priorities) into a code book" (data analysis, pg. 10).
19	Describe methods and reasons for modifying (removing, adding, reframing) priorities	The authors did not play a role in determining the consensus and decisions to modify priorities. The decisions to move priorities forward to the final list was based on steering committee consensus both online and in-person. The authors appear to have provided members with tools to guide decisions (i.e., frequencies, evidence of the extent to which past research had been conducted), but this did not lead the steering committee decisions. "while frequency counts were considered as an important prioritization strategy, steering committee members did not rely solely on these counts to move items forward to the top 30 and top 10 lists" (data analysis, pg. 10). "In the spirit of true consensus building and PSP, researchers facilitated sessions allowing steering committee members to drive decisions and finalize results" (methods, pg. 9).
20	Describe methods for refining or translating priorities into research topics or questions	Apart from the aforementioned theming process, research questions were developed through consensus approach, committee members could initially generate ideas on their own, and then within their smaller group, and then eventually with the broader in-person steering committee members who collaboratively decided how to word the questions (methods, pg. 9).
21	Describe methods for checking whether research questions or topics have been answered	Once collapsed into themes by the steering committee, the authors disseminated the preliminary research priorities to experts in the subject matter. "Experts were asked to indicate if each potential research priority was: (A) well researched (systematic reviews and meta-analysis available), (B) somewhat researched (single studies, some inconsistent evidence on topic), (C) not researched (no studies), or (D) unsure. Experts were encouraged to provide comments or feedback on each of the potential research priorities" (data analysis, pg. 10).
22	Describe number of research questions or topics	An initial 12 broad parenting topics were identified to inform the provincial survey. The survey generated a total 3232 potential research priorities, which were analyzed and consolidated by the steering committee into 202 unique themes. The themes were further collapsed into a list of 34 research priorities, and then eventually into a top 11 list of research priorities. "Selecting 11 priorities as opposed to 10 priorities was the steering committee consensus" (abstract, pg. 2).
Prioritization of Research Topics/Questions		

23	Describe methods and criteria for prioritizing research topics or questions	The identification of priority themes and questions resulting from the provincial survey took place within in-person steering committee workshops "structured using nominal group technique" (methods, pg. 9). Decisions were based on group consensus between participants, with the authors facilitating the discussions but not leading or influencing the decisions. Although there is mention that steering committee members could score and rank the priorities, "The top 30 list was shared with the broader online steering committee and committee members were asked to select and rank their top 10 selections" (methods, pg. 10), the authors do not describe how group consensus was determined for the final list. There are no specific criteria that were used to guide the ranking or prioritization outlined in the manuscript. Steering committee members did have information regarding the level of evidence for each topic, however, this does not appear to have been the guiding factor for the ranking.
24	State the method or threshold for excluding research topics/questions	The authors do not describe a threshold or criteria for excluding priorities. The process for settling challenges to the prioritization process is not described in detail. Discussion, voting, and ranking were used to decide what priorities moved forward (methods, pg. 9), however, these processes are not described in detail.
Output		
25	State the approach to formulating the research priorities	The final priorities have been identified as 11 research questions (Table 3). How the questions were worded, and whether a specific format (i.e., PICO) was used is not explicated. Along with the top 11 research priorities, the researchers also identify the broad topic area (from the initial 12 survey domains) relevant to the priority.
Evaluation and Feedback		
26	Describe how the process of prioritization was evaluated	The usefulness, relevance, and reliability of the priorities are not evaluated. The authors do mention how more than one group themed the survey responses to enhance reliability in the early stages of prioritization, "Reponses from each topic area were analysed by two separate groups to validate theming" (data analysis, pg. 10). The findings have also been used by the provincial health organization already, which may speak to their relevance and utility. The authors also allude to evaluation of engagement from the steering committee member perspective, "steering committee members also completed an assessment of patient engagement." (data analysis, pg. 10). However, this process, the findings, or implications for engagement evaluation are not described further.

27	Describe how priorities were fed back to stakeholders and/or the public, and how feedback (if received) was addressed and integrated	The authors state that the final priorities were sent to the committee for approval, "the final top 10 (plus one) list was circulated online for final approval" (data analysis, pg. 10). There was no revision or appeal process described if committee members did not agree with the final list/question wording.
	Implementation	
28	Outline the strategy or action plans for implementing priorities	The researchers describe how sharing the findings could inform future research to ensure it is aligned with relevant priorities for families by stating, "By sharing the FRAISE top 11 research priorities, we invite other researchers to ensure their work aligns with patient-oriented research as a foundation for future initiatives targeted at improving outcomes in families with young children" (discussion, pg. 18). However, no formal action plan for implementing the priorities is described.
29	Describe plans, strategies, or suggestions to evaluate impact	Although the authors mention how the provincial health service has already utilized the study findings to inform information delivery, formal plans to evaluate the impact of the priority setting findings are not outlined.
	Funding and Conflict of Interest	
30	State sources of funding	The funding came from a relevant funding agency for child health and patient-oriented research, "We are grateful for funding provided by the Alberta Children's Hospital Research Institute and by Alberta Innovates" (funding, pg. 19). The funder did not play a role in any portion of the research study, "The funders of the study had no role in study design, data collection, data analysis, or report writing" (methods, pg. 9).
31	Declare any conflicts or competing interests	The authors state no conflict of interest, "We declare no competing interests" (declaration of interests, pg. 19).

References:

1. Tong A, Synnot A, Crowe S, et al. Reporting guideline for priority setting of health research (REPRISE). *BMC Med Res Methodol* 2019; **19**(1): 243.

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Top 10 (plus 1) research priorities for expectant families and those with children to age 24 months in Alberta, Canada: Results from the Family Research Agenda Initiative Setting (FRAISE) priority setting partnership project.

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Title: Top 10 (plus 1) research priorities for expectant families and those with children to age 24 months in Alberta, Canada: Results from the Family Research Agenda Initiative Setting (FRAISE) priority setting partnership project.

Authors: *Brockway, M., PhD [1,2]; *Keys, E.M., PhD [1,3], Bright, K. S., PhD [1,4], Ginn, C., PhD [1], Conlon, L. [5], Doane, S. [5], Wilson, J., BN [1], Tomfohr-Madsen, L., PhD [6,7], Benzies, K.M., PhD [1,7]

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Abstract

Objective: The study objective was to identify the top 10 research priorities for expectant parents and caregivers of children up to age 24 months.

Design: A priority setting partnership using a modified James Lind Alliance approach was implemented. First, a core steering committee was formed, consisting of 17 parents, clinicians, and community agency representatives. Second, through in-person collaboration with steering committee members, we developed and distributed a survey to identify research priorities across 12 topics. In total, 596 participants consented and 480 completed the survey. Survey responses were grouped and themed into codes during a consensus-building workshop with steering committee members (n = 18). Research and practice experts were consulted to provide feedback on which themes had already been researched. An in-person (n = 21) workshop was used to establish the top 34 priorities, which were circulated to the broader steering committee (n=25) via an online survey. Finally, the core steering committee members (n =18) met to determine and rank a top 10 (plus one) list of research priorities.

Setting: This study was conducted in Alberta, Canada.

Participants: Expectant parents and caregivers of children up to age 24 months.

Results: Survey results provided 3232 responses, with 202 unique priorities. After expert feedback and steering committee consensus, a list of 34 priorities was moved forward for final consideration. The final top 10 (plus 1) research priorities included three priorities on Mental Health/Relationships, two priorities on each of Access to Information, Immunity, and Child Development, and one priority on each of Sleep, Pregnancy/Labour, and Feeding. Selecting 11 instead of 10 priorities was based on steering committee consensus.

Conclusions: The findings will direct future maternal-child research, ensuring it is rooted in parent-identified priorities that represent contemporary needs. To provide meaningful outcomes, research in these priority areas must consider diverse socioeconomic backgrounds and experiences.

Keywords: Parent, Priority Setting, Research Priorities, James Lind Alliance, Patient Engagement, Patient-Oriented Research, Participant Involvement.

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Article Summary

Strengths and Limitations of This Study

- The study had a large sample size (n = 480) for the online survey, comparable to other priority setting partnerships.
- Targeted recruitment strategies, including in-person recruitment at community and social service locations and father parenting classes, were used to successfully improve representation of diverse participants.
- Although attention was paid to promote genuine participation of diverse stakeholders and reach consensus on the research priorities, there is potential that these priorities were influenced by the steering committee members' and/or researchers' inherent and individual biases.
- While the partnership approach followed principles of participatory action research and was modelled after other published studies that used a modified James Lind Alliance methodology, this approach has not yet been validated.
- By creating a welcoming and family-friendly atmosphere and including alternate engagement strategies, parents were meaningfully engaged throughout the partnership process, resulting in the identification of parent-oriented research priorities, many of which differed from priorities previously established by researchers, funders, and industry.

Introduction

Due to the critical and rapid development that occurs within the first three years of life, investing in early childhood has an estimated return of 800%.[1] Parents of young children have the greatest potential to optimize social and environmental conditions to foster optimal child health outcomes.[2] High parental expectations, intensive parenting, and increased access to information have drastically changed parenting experiences over the past 10-15 years.[3] The dynamic nature of the contemporary maternal-child and parenting landscape may be inadequately incorporated into existing research priorities and questions. Further, researchers and clinicians often encounter difficulties effectively translating and implementing research on child health and development.[4] As the knowledge base for supporting healthy developmental outcomes grows in breadth and scope, effectively prioritizing research investment is crucial to maximize impact and minimize research waste.[5]

Understanding health research priorities of knowledge users, such as parents, clinicians, and community agency representatives, is vital to conducting research that is more likely to contribute to meaningful changes in health outcomes.[6] Evidence generated without the consideration of the knowledge user will have minimal effect on clinical practice and reduce meaningful outcomes.[7] Further, there is growing awareness of the importance of adopting an intersectional lens when conducting health research to adequately address culturally and socio-politically appropriate parenting outcomes.[8] Employing an intersectional lens by centering the perspectives of knowledge users will result in evidence and resultant policies that better accounts for their diverse needs.[9] Engaging knowledge users in research priority setting initiatives is recognized as an effective and ethical means of prioritizing the allocation of limited public research funds.[7] Not only does priority setting work promote researcher accountability, but this integrative approach to knowledge translation may also reduce lag time between producing and implementing knowledge,[10] while contributing to the development of culturally and socially meaningful outcomes. Moreover, knowledge user involvement in research may enhance engagement with, and uptake of, interventions and services that support early childhood health, resulting in more timely and effective care.

Priority setting partnerships

Patient-oriented research, specifically priority setting partnerships (PSPs), is increasingly identified as an effective method to decrease research waste and develop research outcomes meaningful to knowledge users.[7] PSPs bring together knowledge users, including clinicians, researchers, and patients or other service users (i.e., parents of well-children), to identify and prioritize research uncertainties using a systematic and collaborative process.

Involving knowledge users such as parents and caregivers (i.e., those who provide direct care for children and who may or may not self-identify as parents, as well as professional caregivers who provide care to families with children) in the foundational development of research through PSPs is a feasible method to produce meaningful outcomes – by creating knowledge important to parents and caregivers, the likelihood of new evidence uptake and shifting healthcare practices is increased. To date, there have been no PSPs focused on identifying community-based research priorities for families of well children from conception to age 24 months. When purposefully employing a participatory action framework, PSPs can use intersectional principles by capturing the perspectives and experiences of traditionally neglected populations.[8,9] Therefore, the objective of this study was to engage a diverse group of perspectives in identifying the top 10 research priorities of expectant parents and caregivers of children up to age 24 months.

Methods

The Family Research Agenda Initiative Setting (FRAISE) project used a modified James Lind Alliance (JLA)[11] approach to identify research priorities of knowledge users (e.g., parents, clinicians, and community agency representatives) within Alberta, a province in Western Canada.[12] Many PSPs use the JLA approach;[6] however, employing an accredited JLA facilitator may be impractical for lower resourced research studies. As such, a modified approach to the JLA method has emerged as a feasible alternative.[13] These modifications included facilitation of the initial steering committee workshop by subject matter experts in patient and public engagement and priority setting partnerships (i.e., external facilitators from the Alberta Strategy for Patient-Oriented Research Support Unit (AbSPOR)), rather than a JLA advisor. After this initial workshop, the lead authors, who are trained in group facilitation and community engagement, facilitated the remaining in-person workshops using consensus-building and shared-decision making strategies. During the analysis and prioritization of the potential research priorities, we used collective sensemaking to narrow down and rank the research priorities. Rather than conduct rapid literature reviews to determine the level of evidence available for the research uncertainties, we asked practice and research subject matter experts to rate the availability of evidence on research uncertainties. These rankings were then used by the steering committee members to help them prioritize the top 30 priorities. FRAISE applied a consensus-building and strengths-based approach, modelled after participatory action research and the JLA. James Lind Alliance PSPs bring together patients or service users with lived experience and/or their carers (e.g., family members)and clinicians, which requires sensitivity to varying participant capacities, ongoing effective communication, transparency in decision-making, and inclusivity of all views.[11] The modified JLA process utilized in the FRAISE project involved a series of iterative steps (Figure 1): (1) formation of a steering committee, including parents, clinicians and community agency representatives and online survey development; (2) delivering the survey to gather research uncertainties or questions from parents, clinicians and community agency representatives; (3) grouping responses into codes; (4) consulting with research and practice experts to determine which codes were already well researched, (5) consensus building to determine and rank the top priorities and (6) prioritize and rank top 10 research priorities as directed by the steering committee.

In previous iterations of the JLA, determining priorities of those experiencing social vulnerability was a challenge;[14] as such, the FRAISE project employed in-person techniques with key community organizations to include responses from respondents with a variety of socioeconomic backgrounds. These techniques included research assistants physically attending programs and services that provided targeted care to populations who were under-represented in the survey to recruit potential participants. To facilitate participation at these sites, research assistants carried electronic tablets that participants could use to complete the online survey before or after their programming or appointments. Parent steering committee members were given a $50 honorarium, childcare, and meals at each session. The Conjoint Health Research Ethics Board at the University of Calgary (REB17-0014) approved this research. We followed the REporting guideline for PRIority SEtting of health research (REPRISE)[15] for this study. All survey participants provided informed consent.

Steering committee

Between November 2017 and March 2020, we engaged a steering committee comprised of researchers, parents, and caregivers (i.e., clinicians, community agency representatives). We aimed for 30 steering committee members who mirrored the regional population, including a minimum of 3% who were Indigenous, and 30% who identified as immigrants and/or visible minorities.[16] While membership was flexible to accommodate the realities of parenting young children, a core group of steering committee members emerged. The core group included 15 parents, four clinicians, and two community agency representatives. While we did not meet our target for Indigenous representation, we exceeded our aim for a diverse group, with five of the 15 parents identifying as a visible minority, two parents identifying as fathers, and two parents identifying as new to Canada. To accommodate the regionality and required flexibility of steering committee members, we provided opportunities for a larger online group of additional steering committee members. These members consisted of parents and caregivers who attended some, but not all workshops in person, as well as healthcare providers from other geographic sites in Alberta, and directors and managerial staff who were unable to attend in-person workshops due to time constraints. The size of this group fluctuated throughout the study timeline, with a maximum of 10 members in addition to the core steering committee. The role of the larger steering committee was to provide online feedback and consultation throughout the priority setting process.

Online survey development to identify research questions

The first consensus building workshop focused on introducing and training the core steering committee, orientating them to FRAISE, and developing the survey instrument. To develop the online survey, the core steering committee (n = 17) collaboratively identified 12 broad parenting topics of interest. Researchers then developed open-ended survey questions that could elicit potential research questions from families related to each of these 12 topics. The survey was refined by the core steering committee using an iterative process of online and in-person engagement. The final survey was piloted with a small group of parents who were unfamiliar with the FRAISE project. The survey was launched online using Qualtrics XM Survey Software

© (Qualtrics, Provo, UT) in May 2018 and promoted via Facebook and Twitter. In-person survey completion sessions were also held at various inner-city community and healthcare agencies to increase the inclusion of under-represented populations, particularly those without reliable internet access and who may be experiencing social vulnerability.

Priority setting workshops and consensus building

Three additional in-person priority setting workshops were hosted with the steering committee to build consensus around survey development, survey response theming, the top 30 list, and the top 10 list (Figure 1). Each session was structured using a nominal group technique.[17] To analyze survey responses and identify top research priorities, core steering committee members were broken into six groups and provided with survey responses from four of the 12 topic areas. Each author acted as a group facilitator to guide the work and ensure that all voices were heard, but not to direct the content of the discussion. Each group member was provided with research questions from the survey results and given time to silently generate ideas. Then, each group member shared their ideas; the facilitator clarified and recorded them. Following group discussion to clarify ideas and priorities, group members voted and ranked each priority, with the top priorities moved forward to the steering committee for consideration. Consensus building concluded with discussion and agreement on which content to move forward from each session. After each in-person session, this process was repeated, using survey software with the broader online steering committee to develop consensus. In the spirit of true consensus building and PSP, researchers facilitated sessions allowing steering committee members to drive decisions and finalize results.[17] As such, modifications were made to the wording of the final top 10 list, resulting in an additional (11th) research priority.[13]

≪INSERT FIGURE 1: FRAISE Study Flow Diagram HERE ≫

Role of the funding source

The funders of the study had no role in study design, data collection, data analysis, or report writing. All authors had access to study data. The corresponding author, EMK, had full access to all data and responsibility for the final decision to submit for publication.

Data analysis

Characteristics of survey participants were calculated using descriptive statistics in Microsoft Excel. Responses from the survey were captured verbatim in each of the 12 topic areas. All individual responses were printed and cut into individual items. In-person steering committee members (n = 18) worked together to group responses into themes, within each topic area. Responses from each topic area were analysed by two separate groups to validate theming. Researchers then built the resultant themes (research priorities) into a codebook. Research and practice experts provided feedback on the extent to which each of the 202 codes had been researched. These experts were asked to indicate if each potential research priority was: (1) well researched (systematic reviews and meta-analysis available), (2) somewhat researched (single studies, some inconsistent evidence on topic), (3) not researched (no studies), or (4) unsure. Experts were encouraged to provide comments or feedback on each of the potential priorities. This information, in conjunction with the 202 codes, was provided to the steering committee during a third consensus building workshop (n = 21) to develop the top 30 list. Steering committee members were instructed to review and consider expert feedback when building consensus around which of the 30 priorities to move forward. While the frequency that a particular priority was submitted in the online survey was considered important for subsequent prioritization, steering committee members did not rely solely on these counts to move items forward to the top 30 and top 10 lists. While the original intent was to narrow down the priorities to a top 30 list, at the time of prioritization, the steering committee reached a consensus that the list should include 34 priorities.

The top 34 list was shared with the broader online steering committee and committee members (n = 25) were asked to select and rank their top 10 selections. These results were brought back to the fourth and final consensus building workshop (n = 18) to develop the top 10 priorities. The final top 10 (plus one) list was circulated online for final approval. Steering committee members were also asked to rank the top 10 (plus one) priorities on their perceived order of importance.

Data Statement

The research priorities codebook, containing the raw research priorities responses, is available upon reasonable request from the corresponding author.

Patient and Public Involvement Statement

The public was engaged via membership in the steering committee after the study and research objectives were conceptualized, and funding was obtained,. Steering committee members designed the survey and participated in data collection via disseminating the survey through their professional and social networks and provided advice on how to increase survey response rates for diverse groups. These members also provided feedback on the survey regarding participant burden and conducted data analysis in collaboration with the researcher team. In addition, steering committee members were sponsored to participate in local, regional, and national study dissemination via virtual and in-person child health rounds and conferences as well as manuscript authorship (LC, SD).

Results

In total, 596 participants consented to participate in the survey, of which 480 completed the survey. Of these, the majority were parents (76.3%; n = 130 tablet; n = 236 online), 16.0% were clinicians (n = 3 tablet; n = 74 online), 4.8% were early childhood educators (n = 10 tablet; n = 13 online), and 2.9% were community agency representatives (n = 0 tablet; n = 22 online). Most participants were recruited via social media and electronic networks (n = 337, 70.2%), with 29.8% (n = 143) recruited during targeted in-person sessions where participants were provided with a tablet to record responses. The mean age of all survey respondents was 34.1 years (SD = 9.1), with tablet-based respondents having a slightly younger mean age in years (32.1; SD = 5.9) than the mean age of online-based respondents (34.8; SD = 9.9). For parent participants who were not born in Canada (n = 65), the mean number of years in Canada was 10.7 (SD = 11.4), with tablet-based respondents having fewer mean years in Canada (M = 8.1; SD = 9.3) than online-based respondents (M = 13.6; SD = 13.0). Table 1 describes participant and household characteristics. The socioeconomic and ethnicity distribution of parent participants was comparable to population distributions.[16] Participation rates mirrored geographic population densities (Figure 2).

Table 1. FRAISE Survey participant characteristics.

	Total Completed (N = 480)		Online Survey (n = 337)		Tablet Survey (n = 143)	
All respondents	N	%	n	%	n	%
Employment status						
Working for pay, profit, or self-employed	233	48.5	182	54.0	51	35.7
Caregiving (including parental or maternity leave)	183	38.1	122	36.2	61	42.7
Not working, but looking	14	2.9	9	2.7	5	3.5
Going to school, retired, cannot work due to disability or illness or other	30	6.3	10	3.0	20	14.0
Completed post-secondary education (e.g., certificate or diploma program, undergraduate/graduate degree)	404	84.2	306	90.8	98	68.5
Ethnicity*						
Caucasian	333	69.4	256	76.0	77	53.8
Chinese	29	6.0	14	4.2	15	10.5
South Asian	14	2.9	10	3.0	4	2.8
Latin American	12	2.5	7	2.1	5	3.5
Indigenous	25	5.2	6	1.8	19	13.3
Caribbean	7	1.5	6	1.8	1	0.7
Filipino	6	1.3	4	1.2	2	1.4
Arab	5	1.0	3	0.9	2	1.4
Korean	4	0.8	3	0.9	1	0.7
Southeast Asian	7	1.5	3	0.9	4	2.8
Japanese	2	0.4	2	0.6	1	0.7

African	8	1.7	1	0.3	7	4.9
West Asian	2	0.4	0	0.0	2	1.4
Other	14	2.9	8	2.4	6	4.2
Prefer not to say	15	3.1	9	2.7	6	4.2
Clinicians	77	16.0	74	22.0	3	2.1
Nurse	47	9.8	47	13.9	0	··
Physician	7	1.5	7	2.1	0	··
Dietician	3	0.6	3	0.9	0	··
Occupational therapist	3	0.6	3	0.9	0	··
Other (midwife, social worker, etc.)	17	3.5	14	4.2	3	2.1
Community agency representative	14	2.9	14	4.2	0	··
Early childhood educator/care provider (e.g., nanny, daycare, preschool)	23	4.8	13	3.9	10	7.0
Parent	366	76.3	236	70.0	130	90.9
Mother	292	60.8	194	57.6	98	68.5
# pregnant	*136*	28.3	*85*	25.2	*51*	35.7
Father	71	14.8	40	11.9	31	21.7
Other (e.g., grandparent, aunt)	3	0.6	2	0.6	1	0.7
Parent respondents only (n = 366)						
Partnered	305	83.3	206	87.3	99	76.2
Number of households with children						
1 child in household	173	47.3	121	51.3	52	40.0
2 children in household	74	20.2	48	20.3	26	20.0
3 children in household	27	7.4	15	6.4	12	9.2
4^+ children in household	13	3.6	4	1.7	9	6.9
Born in Canada (Yes)	280	76.5	190	80.5	90	69.2
English as primary household language	318	86.9	206	87.3	112	86.2
Household income (yearly)						
Less than $40,000	66	18.0	17	7.2	49	37.7
$40,000 - $79,999	82	22.4	64	27.1	18	13.8
$80,000 - $119,999	69	18.9	54	22.9	15	11.5
$120,000 - $159,999	49	13.4	34	14.4	15	11.5
More than $160,000	53	14.5	35	14.8	18	13.8
I don't want to say	31	8.5	19	8.1	12	9.2
Note: Due to missing data, numbers in this table may not sum to the total number of completed responses; *indicates respondents could choose all that apply						

≪INSERT Figure 2. Geographic distribution of responses HERE≫

In total, we received 3232 submissions of potential research priorities spread across all 12 topic areas (Figure 3). When asked to rank the 12 broad parenting topics of interest, survey participants ranked stress, emotional, and mental health; sleep; and infant feeding as the three most important parenting topics. The 3232 suggested priorities obtained via survey responses were coded, themed, and collapsed by the steering committee, resulting in 202 unique priorities (Figure 1). Of these 202 priorities, 34 were shortlisted for consideration in the top 10 list

(Supplementary Table). Steering committee members used this shortlist to build consensus on the final top 10 (plus one) list of research priorities (Table 2).

≪INSERT Figure 3. Number of raw submissions in each topic area from provincial survey HERE≫

Table 2. Final list and topic areas of the top 10 (plus one) research priorities from conception to two years of age for families in the community.

Rank	Question	Topic Area
1	How can families be supported to develop healthy coping strategies, emotional regulation, and resiliency in both parents and children?	Mental Health/Relationships
2	How can families access supportive care and information when things do not go as expected during pregnancy, labour, birth, or postpartum?	Access to Information/ Pregnancy, Labour & Birth
3	How can the healthcare system and providers ensure access to prevention and treatment of mental health concerns in a safe and trusting environment?	Mental Health
4	How do families navigate multiple sources of health information and access services tailored to their specific circumstances?	Access to Information
5	What supports and services can be developed at the individual, family, community, and government levels to build emotional/mental wellbeing, physical health, and healthy relationships?	Mental Health/Relationships
6	How can eczema, asthma, and allergies be more effectively prevented, assessed, and treated?	Immunity
7	In a constantly changing social and physical environment, how can families increase safety and manage developmentally appropriate risk-taking?	Child Development/Child Safety/Environmental Risk
8	How can sleep problems be prevented, assessed, and treated in a culturally appropriate way that is tailored to individual families?	Sleep
9	How can families be better supported to make informed, family-centred feeding decisions?	Feeding
10	How can families be better supported to promote healthy child development, recognize milestones, and access services for delay?	Child Development
11	With increasing vaccine hesitancy, how can individual, family, and population health best be protected?	Immunity

Discussion

To our knowledge, this is the first attempt to identify community-based research priorities for families of well children from conception to age 24 months. Throughout all steps of the PSP, concerns for mental, social, and emotional well-being of parents and children were consistently expressed as priority areas. Comparing the top 10 (plus one) priorities with the initial 12 survey categories, we found that stress, emotional, and mental health were consistently a top concern for families and were identified in three of the top 10 (plus one) priorities. Additionally, steering committee members repeatedly specified the need for structural health system changes at the individual, family, community, and government levels to focus on building emotional/mental well-being, physical health, and healthy relationships – taking the onus off individual families to solve problems related to mental and physical wellness and prioritizing policy and systems shifts. As such, these research priorities highlight the need to situate individual- or family-focused research priorities within the context of complex health and social systems and involve multiple sectors such as government and health boards.

Across themes, the top 10 (plus one) research priorities reflect the importance of fostering access to information and developing information and services using a culturally sensitive and humble framework. While identified as its own research priority (Table 2), concerns about access to information emerged in each topic area. The ubiquitous nature of access to information as a research priority raises a fundamental question about the accessibility of safe, reliable, and valid information for young families. Previous research from Canada indicates the majority of parents use the internet to access information about children's health, but many use unreliable sources of information.[18] In contrast, Australian women who are pregnant most often reported discussion with their midwife as an information source, while less than half reported using the internet to access information; group information sessions were the least preferred information sources.[19] This contrasts with a report from Devolin and colleagues[20] where Albertan parents rated the Internet (55.3%) and drop-in programs (42.9%) as preferred information sources. With such conflicting evidence, it is unsurprising that researchers and care providers have long struggled with providing safe and reliable information related to child and infant health. Healthcare providers and administrators should collaborate with researchers and parents to critically examine health information provision and consider transformative frameworks to create meaningful and innovative knowledge translation strategies to better communicate evidence. Several research questions related to physical health emerged as important. For example, parents expressed concerns about how eczema, asthma, and allergies could be more effectively prevented, assessed, and treated; with this class of inflammatory disorders as one of the most common issues in early childhood, this is unsurprising. The fact that parents shared a strong desire for information about preventing these disorders, suggests this is a research area ripe for future development. Additionally, questions emerged about preventing injury while encouraging appropriate physical development and accessing information about culturally appropriate feeding and sleep. While some research exists in each of these areas, evidence is generally underdeveloped, and information needs to be communicated to parents in culturally appropriate and meaningful ways. Of particular importance given the COVID-19 pandemic, parents were unwilling to leave the final steering committee workshop without including a research priority related to vaccine hesitancy. Diverse opinions emerged around this topic, and it became clear that evidence-based information about vaccines and immunity was a valued priority.

Findings from this PSP complement findings from other pediatric research PSPs that have focused on single issue priority setting, such as premature birth,[21] stillbirth,[22] chronic pain,[23] neurodisabilities,[24] or learning difficulties.[25] Despite the breadth in populations and that most existing PSPs were conducted in the UK, research priorities identified in these partnerships related to access to information, mental and emotional health, and healthy relationships, suggesting that these themes are consistently important to parents.

One notable exception to the general trend of conducting PSPs with specialized populations is a Canadian PSP that developed research priorities for preventative care of children aged zero to five years.[26] Consistent with our top research priority of mental and emotional health in children and their parents, the top research question found by Lavigne and colleagues was "What are effective strategies for screening and prevention of mental health problems?" (p.750).[26] Our findings confirm and extend this priority of building mental and emotional wellness in children belonging younger age groups (and during conception) by broadening the focus to include parents and protective factors for resilience. This suggests that parent, clinician, and researcher priorities are aligned with the emergence of infant mental health (ages zero to five) as a relational concept, whereby infant mental health is supported by optimizing the mental health and resiliency of children's caregivers (i.e. parents and family members).[1] Other similarities included: (1) supporting child development by improving identification and services for developmental delays; (2) developing effective interventions, supports, and services to improve mental health, physical health (obesity, physical activity), and healthy relationships (social skills); and (3) understanding the impact and support of nutritional and feeding factors. Contrasts with our research priorities and those for children aged zero to five years included: (1) the impact of daycare attendance on child health, (2) behavior management in children, and (3) appropriate screen time for children. In addition, research priorities in our list that were not found in the list for children aged zero to five years included: (1) how families can access supportive care and information during unexpected experiences in the perinatal period; (2) access to information; (3) how eczema, asthma, and allergies can be more effectively prevented, assessed, and treated; (4) injury prevention and appropriate levels of risk; and (5) sleep problems in families. It should be noted that differences may represent prioritization related to different developmental stages and ages (i.e., screen time is not recommended for children under age two) and/or slight differences in the prioritization method. For example, helping families identify appropriate childcare appeared in our top 34 list and aligns with a similar research priority related to the impact of daycare attendance in the zero to five years list.

In a Delphi study of perceived research priorities of clinical staff at an Australian parenting centre, Hauck and colleagues[27] determined the top research priorities to be related to short- and long-term evaluation of the effectiveness of the centre's programs on children's behavior and stress levels, parental expectations, healthy family relationships, as well as parental use of sleep, settling, and feeding strategies. Determining the effectiveness of the centre's programme in the context of postpartum depression was also an identified priority.[27] Although these research priorities were not informed by parental participation and may have limited generalizability to other centres and/or countries, it is notable that issues related to sleep, infant feeding, healthy relationships, and parental mental health are captured by both ours and Hauck's[27] lists. This is not surprising as a study in our same geographical area suggested that breastfeeding, sleep issues, child development, helping children cope with emotions, and understanding child brain development were all in the top 10 (plus one) parenting topics ranked as somewhat or very important by parents.[20]

Clinical and research applications

To move forward with these research priorities, it is evident that multiple stakeholders must be engaged, and connections made across a variety of sectors and disciplines. Wording for the top 10 (plus one) research priorities requested consideration of "culturally appropriate" and "safe and trusting environments." Our findings highlight the need for an increased emphasis on parenting education and culturally appropriate strategies to support mental health and well-being. Adopting an intersectional lens when conducting research, and providing care to pregnant and young families, will better address these considerations and provide meaningful evidence to inform health outcomes. Similar to how parents of diverse groups may have differing or intensified concerns relating to child health issues,[28] it is important to recognize that research priorities may differ based on community member's characteristics, specifically in terms of race, income, access to health services, and experiences, including racism. These top 10 (plus one) priorities were developed using an intersectional framework. Moving forward, research in these priority areas must continue to comprehensively and meaningfully include individuals with diverse socioeconomic backgrounds and experiences.[9]

The findings from this study should be considered in the context of its strengths and limitations. In terms of strengths, there was a large sample size for the online survey comparable to other PSPs; this created space for a broad representation of stakeholders including clinicians, community agency representatives, parents, and ethnically diverse representatives of the study location. Limitations included the inherent and individual biases that self-selected, voluntary researchers and steering community members view the world with, which may reduce the generalizability of the final priority list. Given that the research team was composed entirely of white heterosexual women and although attention was paid to ensure participation of fathers, individuals self-identifying as non-white, and those in non-heterosexual or single parent partnerships, these voices may not have been entirely represented in the final priorities. Additionally, while this research priority setting partnership used a process based on a previously reported modified JLA approach,[13] these modifications have not been validated against the standardized JLA method. Not employing an accredited JLA facilitator means that the study did not have the rigorous external supervision that other JLA-led studies benefit from. Our modifications (e.g., using external group facilitators with expertise in patient and public engagement to establish the steering committee rather than an accredited paid JLA advisor; consulting with experts on the level of evidence, rather than conducting rapid literature reviews on each potential research uncertainty) greatly reduced the cost of the project, thereby increasing feasibility. Additionally, the current approach incorporated principles of participatory action research (e.g., engaging with a community who have self-identified in reciprocal relationships, recognition of local knowledge, and incorporating processes of co-learning to take actions that will improve community member's wellbeing), which are widely recognized as critical in facilitating meaningful participant engagement and ensuring that participants' voices are represented throughout the research process.

Researching these family-driven priorities will reduce the knowledge-to-practice gap and generate opportunities to improve services for families, including innovative delivery models and evidence-based treatment options to enable equitable access to services. Further, involving practitioners in the PSP may enhance the implementation of new evidence-based practice recommendations.[29] These outcomes, combined with decreased lag time from research to practice, may result in improved child and family outcomes. As a result of our strong collaborative partnerships and inclusion of policymakers in each stage of the PSP, the provincial health service has already utilized this information to streamline and enhance evidence-based information delivery to parents and care providers in the province. By sharing the FRAISE top 10 (plus one) research priorities, we invite other researchers to ensure their work aligns with principles of patient-oriented research as a foundation for future initiatives targeted at improving outcomes in families with young children.

Author Contributions

EMK and MB drafted the manuscript, which all authors reviewed, provided feedback, and approved the final version. EMK, MB, KSB, CG, and KMB designed the study. EMK, MB, KSB, CG, KMB, and LTM developed the steering committee workshop content. EMK, MB, KSB, CG, KMB, LTM, SD, LC and the Steering Committee devised the data collection instruments. JW, EMK, MB, KSB, and CG were responsible for data collection and database design and management. EMK, MB, KSB, CG, KMB, LTM, JW, SD, LC, and the Steering Committee completed the data analyses.

Declarations of interests

We declare no competing interests.

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Ethics Statement

Patient consent for publication

Not required

Ethics approval

The Conjoint Health Research Ethics Board at the University of Calgary (REB17-0014) approved this research.

References

1. Shonkoff JP. Capitalizing on advances in science to reduce the health consequences of early childhood adversity. *JAMA Pediatr* 2016; **170**: 1003–7.
2. Bronfenbrener, U. (ed.). Making human beings human: bioecological perspectives on human development. Thousand Oaks, CA: Sage, 2005.
3. Wall G. Mothers' experiences with intensive parenting and brain development discourse. *Womens Stud Int Forum* 2010; **33**: 253–63.
4. Albrecht L, Archibald M, Snelgrove-Clarke E, Scott SD. Systematic review of knowledge translation strategies to promote research uptake in child health settings. *J Pediatr Nurs* 2016; **31**: 235–54.
5. Chalmers I, Glasziou P. Avoidable waste in the production and reporting of research evidence. *Obstet Gyneco*l 2009; **114**: 1341–45.
6. Manafò E, Petermann L, Vandall-Walker V, Mason-Lai P. Patient and public engagement in priority setting: a systematic rapid review of the literature. *PLoS One* 2018; **13**: e0193579.
7. Chalmers I, Bracken MB, Djulbegovic B, et al. How to increase value and reduce waste when research priorities are set. *Lancet* 2014; **383**: 156–65.
8. Shimmin C, Wittmeier KDM, Lavoie JG, Wicklund ED, Sibley KM. Moving towards a more inclusive patient and public involvement in health research paradigm: the incorporation of a trauma-informed intersectional analysis. *BMC Health Serv Res* 2017; **17**: 539.
9. Bailey J, Steeves V, Burkell J, Shade LR, Ruparelia R, Regan P. Getting at equality: research methods informed by the lessons of intersectionality. *Int J Qual Methods* 2019; **18**: 1–13.
10. Mann BS, Manns BJ, Dart A, et al. An assessment of dialysis provider's attitudes towards timing of dialysis initiation in Canada. *Can J Kidney Health Dis* 2014; **1**: 3.
11. Cowan K, Oliver S. The James Lind Alliance guidebook. 2010. http://www.jlaguidebook.org/pdfguidebook/guidebook.pdf (accessed Nov 22, 2016).

12. Bright KS, Ginn C, Keys EM, et al. Study protocol: determining research priorities of young Albertan families (The Family Research Agenda Initiative Setting Project-FRAISE)-participatory action research. *Front Public Health* 2018; **6**: 228.
13. Breault LJ, Rittenbach K, Hartle K, et al. The top research questions asked by people with lived depression experience in Alberta: a survey. *CMAJ Open* 2018; **6**: E398–405.
14. Barnieh L, Jun M, Laupacis A, Manns B, Hemmelgarn B. Determining research priorities through partnership with patients: an overview. *Semin Dial* 2015; **28**: 141–46.
15. Tong A, Synnot A, Crowe S, et al. Reporting guideline for priority setting of health research (REPRISE). *BMC Med Res Methodol* 2019; **19**: 243.
16. Statistics Canada. 2011 National Household Survey: Statistics Canada. 2013. https://www12.statcan.gc.ca/nhs-enm/2011/dp-pd/dt-td/Index-eng.cfm (accessed Oct 7, 2020).
17. Carney O, McIntosh J, Worth A. The use of the nominal group technique in research with community nurses. *J Adv Nurs* 1996; **23**: 1024–29.
18. Pehora C, Gajaria N, Stoute M, Fracassa S, Serebale-O'Sullivan R, Matava CT. Are parents getting it right? A survey of parents' internet use for children's health care information. *Interact J Med Res* 2015; **4**: e12.
19. Grimes HA, Forster DA, Newton MS. Sources of information used by women during pregnancy to meet their information needs. *Midwifery* 2014; **30**: e26–33.
20. Devolin M, Phelps D, Duhaney T, et al. Information and support needs among parents of young children in a region of Canada: a cross-sectional survey. *Public Health Nurs* 2013; **30**: 193–201.
21. Duley L, Uhm S, Oliver S. Top 15 UK research priorities for preterm birth. *Lancet* 2014; **383**: 2041–42.
22. Heazell AEP, Whitworth MK, Whitcombe J, et al. Research priorities for stillbirth: process overview and results from UK Stillbirth Priority Setting Partnership. *Ultrasound Obstet Gynecol* 2015; **46**: 641–47.
23. Birnie KA, Dib K, Ouellette C, et al. Partnering for pain: a priority setting partnership to identify patient-oriented research priorities for pediatric chronic pain in Canada. *CMAJ Open* 2019; **7**: E654–64.
24. Morris C, Simkiss D, Busk M, et al. Setting research priorities to improve the health of children and young people with neurodisability: a British Academy of Childhood Disability-James Lind Alliance research priority setting partnership. *BMJ Open* 2015; **5**: e006233.
25. Lim AK, Rhodes S, Cowan K, O'Hare A. Joint production of research priorities to improve the lives of those with childhood onset conditions that impair learning: The James Lind Alliance priority setting partnership for 'learning difficulties'. *BMJ Open* 2019; **9**: e028780.
26. Lavigne M, Birken CS, Maguire JL, Straus S, Laupacis A. Priority setting in paediatric preventive care research. *Arch Dis Child* 2017; **102**: 748–53.
27. Hauck Y, Kelly RG, Fenwick J. Research priorities for parenting and child health: a Delphi study. *J Adv Nur* 2007; **59**: 129–39.
28. Garbutt JM, Leege E, Sterkel R, Gentry S, Wallendorf M, Strunk RC. What are parents worried about? Health problems and health concerns for children. *Clin Pediatr* 2012; **51**: 840–47.
29. Mathieson A, Grande G, Luker K. Strategies, facilitators and barriers to implementation of evidence-based practice in community nursing: a systematic mixed-studies review and qualitative synthesis. *Prim Health Care Res Dev* 2019; **20**: e6.

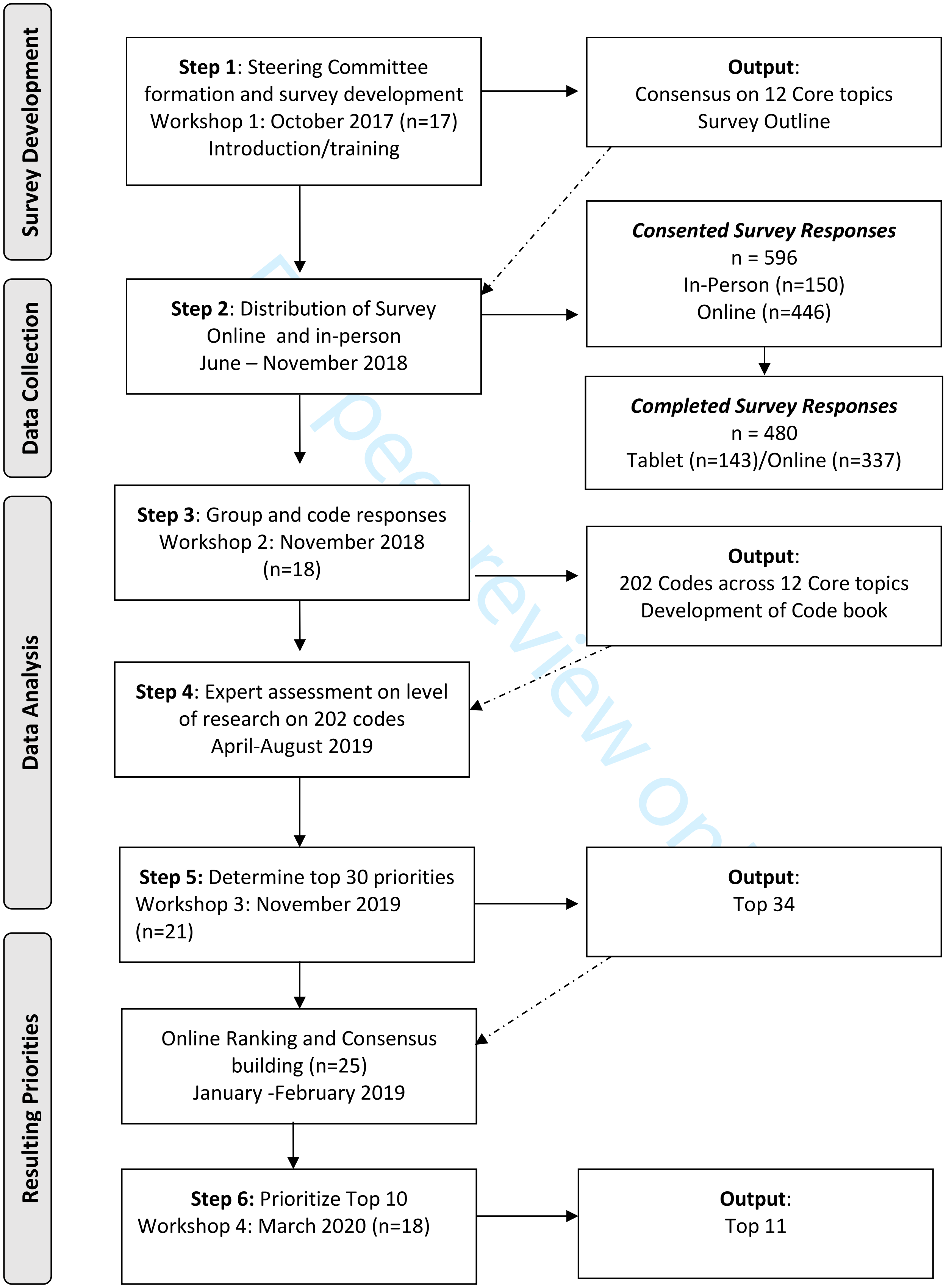
Figure Captions

Figure 1. FRAISE Study Flow Diagram

Figure 2. Geographic distribution of responses

Figure 3. Number of raw submissions in each topic area from provincial survey

FRAISE Study Flow Diagram



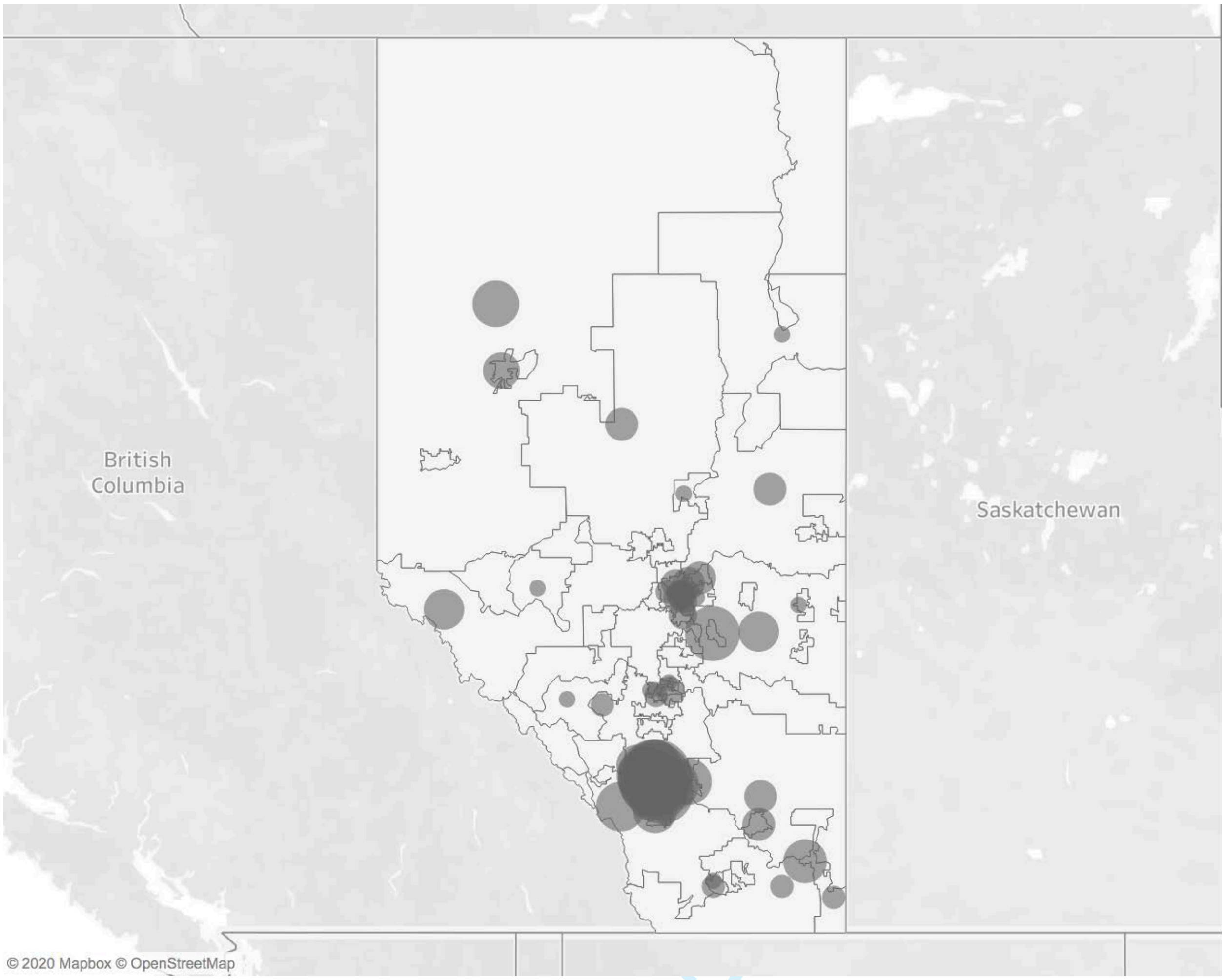


Figure 2. Geographic distribution of responses

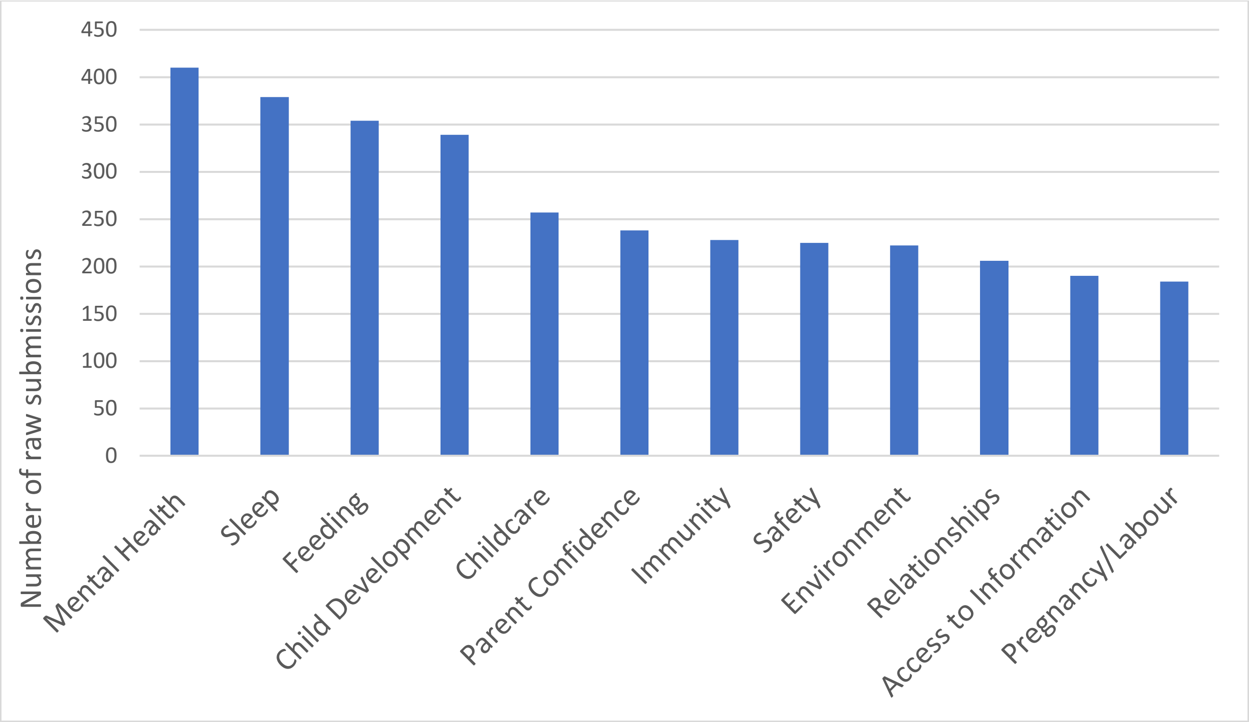


Figure 3. Number of raw submissions in each topic area from provincial survey

Supplemental Table. Top 34 Themes	
Question	**# of times in top 10**
How can families and caregivers of children 2 and under be supported to develop healthy coping strategies, emotional regulation, and resiliency in both themselves and their children?	15
How to access information/programs and navigate the healthcare system when you are from a minority/non-dominant group?	14
What is an age appropriate level of risk taking for children to promote their growth & development while maintaining adequate (good enough) safety?	12
How do parents manage multiple sources of information to make health decisions that are tailored to their specific circumstances and needs?	12
How can families (partners & extended family members), communities, and government encourage self-care and provide emotional and practical support for parents during the perinatal and early childhood period?	11
When things don't go as planned, how can parents get care, support, and information during pregnancy, labour, birth or the postpartum period?	11
What formats (i.e. in-person, online, group) do families prefer when accessing supports and services to develop and sustain healthy relationships and mental well-being?	10
How can health care professionals provide a safe and trusting environment that encourages women/families to disclose mental health concerns without fear of negative repercussions?	10
How to identify, prevent, and treat eczema, asthma, and allergies?	10
How do we create culturally appropriate, family specific information about sleep (co-sleeping, swaddling, sleep training, etc.) from conception to age 2?	10
What are the advantages and disadvantages of using a trauma-informed model of care to explore pregnancy, childbirth, and postpartum for all parents?	9
How do we prevent, assess, and treat sleep problems in parents and children?	9
What supports and services are required to support families to develop and sustain healthy relationships and mental wellbeing?	8
To minimize physician visits, what user centred information sources and formats are most acceptable and effective to provide parents and healthcare providers with reliable and up-to-date information?	8
How can we best support families with their infant feeding decisions?	7
With increasing vaccine hesitancy, how do we best protect individual and population health?	7
How can families during the perinatal and early childhood period access appropriate and evidence-based assessments, prevention, treatment, and support for mental health concerns?	7
How can we manage the effect of exposure to cannabis smoking to families with young children?	7
In children 2 and under, how can families and caregivers be supported around milestones, behaviours, and signs of delay?	7
In addition to vaccination, how can parents and children build their immunity?	6
What supports and services increase general parental confidence?	6
How can families during the perinatal and early childhood period manage emotional and physical wellbeing?	6
What are the necessary measures to increase safety for toddlers/children in an ever changing social and physical environment (during the early childhood period)?	6
How can communities and health care professionals provide culturally sensitive best practices for promoting healthy pregnancies & fetal/infant/child development?	6
How can we increase awareness and access to additional services and resources for families of young children?	6
What are expected and problematic sleep patterns in caregivers, infants, and toddlers?	6

What evidence-based community resources and services are available to help foster healthy relationships for expecting parents?	5
How can families be supported to identify the right fit of childcare, looking at quality, safety, and affordability while still incorporating their own beliefs and values?	5
How do we build on general confidence to generate transferable skills for parenting?	4
What are the consequences of sleep deprivation and problems in parents and children?	4
What are effective strategies for accessing/using my electronic health record and keeping track of my family's health information?	3
What should healthcare providers recommend for guidelines to support families to transition from milk feeding to solid foods?	2
How does infant feeding impact allergies?	2
How can parents during early childhood identify & prevent child safety risks?	2

Reporting Guideline for Priority Setting of Health Research (REPRISE)[1]

#	Item	Descriptor
		Context and Scope
1	Define geographical scope	Regional geographic area “within a province in Western Canada” (methods, pg. 7).
2	Define health area, field, focus	Focussed on a specific population of healthy young families and caregivers in the community setting. The population of interest is “families of well children from conception to age 24 months” (introduction, pg. 6).
3	Define the intended beneficiaries	Intended beneficiaries of these findings include researchers, care providers, community services representatives, and families themselves. “Develop research outcomes meaningful to knowledge users, parents, and care providers” (introduction, pg. 6).
4	Define the target audience of the priorities	The target audience who has the potential to fund future research or implement the identified priorities include care-providers, health services organizations, and researchers. “As a result of our strong collaborative partnerships and inclusion of policy makers in each stage of the PSP, the provincial health service has already utilized this information to streamline and enhance evidence-based information delivery to parents and care providers in the province. By sharing the FRAISE top 11 research priorities, we invite other researchers to ensure their work aligns with patient-oriented research as a foundation for future initiatives targeted at improving outcomes in families with young children.” (discussion, pg. 18).
5	Identify the research area	The broad research area of the priority setting research encompasses health services and public health. “these family-driven priorities will reduce the knowledge to practice gap and result in higher quality health care services for families of young children” (discussion, pg. 17).
6	Identify the type of research questions	The types of research questions were broad, and ranged from etiology (i.e., asthma, eczema, allergy causes/diagnosis) to implementation (i.e., developing community supports). Questions also addressed psychosocial and behavioural research priorities. The researchers did not pre-define the type of research questions that were expected to be generated from the priority setting partnership. See Table 3 for top 11 research priority questions.

7	Define the time frame	The researchers outline the timeframe during which the research was conducted "between November 2017 and March 2020" (methods, pg. 8), however, the anticipated relevance of the research priorities is not addressed. Plans for to monitor or update the priorities with future prioritization activities are not discussed.
Governance and Team		
8	Define the selection and structure of the leadership and management team	The priority setting was facilitated by the authors, however, the structure of the research team, relevance for inclusion, or technical expertise as a facilitator is not addressed. "Each author acted as a group facilitator to guide the work, but not the content of the discussion" (methods, pg. 9). A steering committee (smaller in-person group and larger online group) completed the prioritization process. The selection of the committee members and structure of the team is not outlined. Several groups are mentioned in the acknowledgements paper for contributing to the committee, but their role is not elaborated on in the manuscript. The authors simply state, "we engaged a steering committee comprised of researchers, parents, clinicians, and community agency representatives" and "core group included 15 parents, four clinicians and two community agency representatives; a larger online group of steering committee members also provided feedback and consultation throughout the priority setting process" (methods, pg. 8). The characteristics of, or any differences between, the in-person steering committee members and online members are not explicated. The need for participant diversity is noted, "We aimed for 30 steering committee members who mirrored the regional population, including a minimum of 3% who were First Nations, Metis, or Inuit and 30% who identified as immigrants and/or visible minorities", however, whether or not the goal for diversity was fulfilled is not described. The authors do note the limitation that the study may not be generalizable to all populations, "although attention was paid to ensure participation of fathers, individuals self-identifying as non-white, and those in non-heterosexual or single parent partnerships, these voices may not have been entirely represented in the final priorities" (discussion, pg. 17).
9	Describe the characteristics of the team	The demographics of the steering committee (on-line and in-person) are not presented. Furthermore, the affiliations or expertise of the researchers, clinicians, and community agency representatives are not presented.

		The authors do recognize that the research team was "composed entirely of white heterosexual women" (discussion, pg. 17), and also present the demographics of the broader sample who completed the survey to identify the initial parent questions that informed the final 11 themes, "Table 1 describes participant and household characteristics. The socioeconomic and ethnicity distribution of parent participants was comparable to population distributions. Participation rates mirrored geographic population densities." (results, pg. 11).
10	Describe any training or experience relevant to conducting priority setting	The presence of participant or researcher training, consultants, or past experience with priority setting research is not addressed in the body of the manuscript. However, the authors do acknowledge the "Alberta Strategy for Patient-Oriented Research Support Unit for their expertise, guidance and support in the developmental stages of this project" within the acknowledgement section.
Framework for Priority Setting		
11	State the framework used (if any)	The authors identify the use of "a consensus-building and strengths-based approach, modelled after participatory action research and the James Lind Alliance" (methods, pg. 7). The authors provide sound rationale for their use of a modified James Lind Alliance approach, recognizing that the conventional methodology is "resource intensive and may be impractical for lower resourced research studies and vulnerable populations" (methods, pg. 7).
Stakeholders and Participants		
12	Define the inclusion criteria for stakeholders involved in priority-setting	As mentioned above, the authors sought to include a diverse group of stakeholders who accurately represented the population of the research setting. Specific inclusion criteria or the process of identifying, approaching, and engaging stakeholders is not outlined. More broadly for the survey participants, efforts to engage diverse populations was achieved through in-person survey recruitment in conjunction with online surveys.
13	State the strategy or method for identifying and engaging stakeholders	The strategy for identifying stakeholders in the steering committee are not addressed in the body of the manuscript. Again, several groups and existing partnerships are noted in the acknowledgements for their contribution to the committee, however, strategies for stakeholder engagement are not outlined.
14	Indicate the number of participants and/or organizations involved	The authors briefly describe the number of stakeholders involved in priority setting, "we engaged a steering committee comprised of researchers, parents, clinicians, and community agency representatives" and "core group included 15 parents, four clinicians and two community agency representatives; a larger online group of steering committee members also provided

		feedback and consultation throughout the priority setting process" (methods, pg. 8). Although the number of in-person committee members is clear, there is no number provided for the larger online steering committee group. Furthermore, the organizational affiliation (or the nature of the organization, i.e., health, government etc.) of researchers, clinicians, or community representatives is not described.
15	Describe the characteristics of stakeholders	As noted above, apart from the number of steering committee members who were parents, clinicians, and community agency representatives in the in-person group, there is limited explanation of the steering committee characteristics.
16	State if reimbursement for participation was provided	The authors clearly state, "Parent participants were given a $50 honorarium, childcare, and meals at each session" (methods, pg. 7).
Identification and Collection of Research Priorities		
17	Describe methods for collecting initial priorities	The development of initial priorities appears to be two-fold. The authors and steering committee developed an initial list of 12 parenting topics, which then informed a survey where parents could ask questions about each of the 12 parenting topics. These parent questions relative to the 12 topics would then be analyzed to create the final top 11 priority list (methods, pg. 7-8). The authors do not elaborate how the initial 12 parenting topics were selected, apart from stating, "our steering committee collaboratively identified 12 broad parenting topics of interest" (methods, pg. 8). However, it is noted that the survey generated from the 12 topic areas was refined in collaboration with steering committee members, both online and in-person, and then pilot tested with "outside" parents. The subsequent data analysis of the themes and questions resulting from the provincial survey took place within in-person steering committee workshops "structured using nominal group technique" (methods, pg. 9), and through online consensus, "After each in-person session, this process was repeated, using survey software with the broader online steering committee to develop consensus" (methods, pg. 9).
18	Describe methods for collating and categorizing priorities	The initial broad priority suggestions and research questions were thematically grouped and analyzed through in-person steering committee workshops. "All individual responses were printed and cut into individual items. In-person steering committee members worked together to group responses into themes, within each topic area. Reponses from each topic area were analysed by two separate groups to

		validate theming. Researchers then built the resultant themes (research priorities) into a code book" (data analysis, pg. 10).
19	Describe methods and reasons for modifying (removing, adding, reframing) priorities	The authors did not play a role in determining the consensus and decisions to modify priorities. The decisions to move priorities forward to the final list was based on steering committee consensus both online and in-person. The authors appear to have provided members with tools to guide decisions (i.e., frequencies, evidence of the extent to which past research had been conducted), but this did not lead the steering committee decisions. "while frequency counts were considered as an important prioritization strategy, steering committee members did not rely solely on these counts to move items forward to the top 30 and top 10 lists" (data analysis, pg. 10). "In the spirit of true consensus building and PSP, researchers facilitated sessions allowing steering committee members to drive decisions and finalize results" (methods, pg. 9).
20	Describe methods for refining or translating priorities into research topics or questions	Apart from the aforementioned theming process, research questions were developed through consensus approach, committee members could initially generate ideas on their own, and then within their smaller group, and then eventually with the broader in-person steering committee members who collaboratively decided how to word the questions (methods, pg. 9).
21	Describe methods for checking whether research questions or topics have been answered	Once collapsed into themes by the steering committee, the authors disseminated the preliminary research priorities to experts in the subject matter. "Experts were asked to indicate if each potential research priority was: (A) well researched (systematic reviews and meta-analysis available), (B) somewhat researched (single studies, some inconsistent evidence on topic), (C) not researched (no studies), or (D) unsure. Experts were encouraged to provide comments or feedback on each of the potential research priorities" (data analysis, pg. 10).
22	Describe number of research questions or topics	An initial 12 broad parenting topics were identified to inform the provincial survey. The survey generated a total 3232 potential research priorities, which were analyzed and consolidated by the steering committee into 202 unique themes. The themes were further collapsed into a list of 34 research priorities, and then eventually into a top 11 list of research priorities. "Selecting 11 priorities as opposed to 10 priorities was the steering committee consensus" (abstract, pg. 2).
Prioritization of Research Topics/Questions		

23	Describe methods and criteria for prioritizing research topics or questions	The identification of priority themes and questions resulting from the provincial survey took place within in-person steering committee workshops “structured using nominal group technique” (methods, pg. 9). Decisions were based on group consensus between participants, with the authors facilitating the discussions but not leading or influencing the decisions. Although there is mention that steering committee members could score and rank the priorities, “The top 30 list was shared with the broader online steering committee and committee members were asked to select and rank their top 10 selections” (methods, pg. 10), the authors do not describe how group consensus was determined for the final list. There are no specific criteria that were used to guide the ranking or prioritization outlined in the manuscript. Steering committee members did have information regarding the level of evidence for each topic, however, this does not appear to have been the guiding factor for the ranking.
24	State the method or threshold for excluding research topics/questions	The authors do not describe a threshold or criteria for excluding priorities. The process for settling challenges to the prioritization process is not described in detail. Discussion, voting, and ranking were used to decide what priorities moved forward (methods, pg. 9), however, these processes are not described in detail.
	Output	
25	State the approach to formulating the research priorities	The final priorities have been identified as 11 research questions (Table 3). How the questions were worded, and whether a specific format (i.e., PICO) was used is not explicated. Along with the top 11 research priorities, the researchers also identify the broad topic area (from the initial 12 survey domains) relevant to the priority.
	Evaluation and Feedback	
26	Describe how the process of prioritization was evaluated	The usefulness, relevance, and reliability of the priorities are not evaluated. The authors do mention how more than one group themed the survey responses to enhance reliability in the early stages of prioritization, “Reponses from each topic area were analysed by two separate groups to validate theming” (data analysis, pg. 10). The findings have also been used by the provincial health organization already, which may speak to their relevance and utility. The authors also allude to evaluation of engagement from the steering committee member perspective, “steering committee members also completed an assessment of patient engagement.” (data analysis, pg. 10). However, this process, the findings, or implications for engagement evaluation are not described further.

27	Describe how priorities were fed back to stakeholders and/or the public, and how feedback (if received) was addressed and integrated	The authors state that the final priorities were sent to the committee for approval, "the final top 10 (plus one) list was circulated online for final approval" (data analysis, pg. 10). There was no revision or appeal process described if committee members did not agree with the final list/question wording.
	Implementation	
28	Outline the strategy or action plans for implementing priorities	The researchers describe how sharing the findings could inform future research to ensure it is aligned with relevant priorities for families by stating, "By sharing the FRAISE top 11 research priorities, we invite other researchers to ensure their work aligns with patient-oriented research as a foundation for future initiatives targeted at improving outcomes in families with young children" (discussion, pg. 18). However, no formal action plan for implementing the priorities is described.
29	Describe plans, strategies, or suggestions to evaluate impact	Although the authors mention how the provincial health service has already utilized the study findings to inform information delivery, formal plans to evaluate the impact of the priority setting findings are not outlined.
	Funding and Conflict of Interest	
30	State sources of funding	The funding came from a relevant funding agency for child health and patient-oriented research, "We are grateful for funding provided by the Alberta Children's Hospital Research Institute and by Alberta Innovates" (funding, pg. 19). The funder did not play a role in any portion of the research study, "The funders of the study had no role in study design, data collection, data analysis, or report writing" (methods, pg. 9).
31	Declare any conflicts or competing interests	The authors state no conflict of interest, "We declare no competing interests" (declaration of interests, pg. 19).

References:

1. Tong A, Synnot A, Crowe S, et al. Reporting guideline for priority setting of health research (REPRISE). *BMC Med Res Methodol* 2019; **19**(1): 243.