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Promoting health and addressing disparities amongst indigenous populations

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Promoting health and addressing disparities amongst indigenous populations

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Editorial: Promoting health and addressing disparities amongst Indigenous populations

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Editorial on the Research Topic
Promoting health and addressing disparities amongst Indigenous populations

Introduction

Indigenous populations worldwide endure persistent health inequities shaped by colonial legacies, systemic discrimination, and socio-economic marginalization. These disparities manifest in elevated rates of preventable diseases, limited access to healthcare, and poorer health outcomes compared to non-Indigenous populations. Discrimination can be a fundamental cause of the health inequalities that exist among Indigenous peoples. Additionally, it has a direct negative impact on health and wellbeing, as the mistreatment, stereotypes, and lack of quality care they experience discourage them from accessing health services. Therefore, generating multidimensional public policies that protect these groups is essential (1, 2). Despite global commitments to health equity, barriers such as cultural disconnects, geographic isolation, lack of resources, or geneal exclusion of Indigenous people, and inadequate policy implementation hinder meaningful progress. The studies in this editorial illuminate the multifaceted health challenges faced by Indigenous communities and underscore the urgent need for culturally grounded, community-driven solutions. By navigating through the breadth of research compiled here, we aim to highlight research studies that clearly advocate for Indigenous health equity and inspire action to bridge these gaps.

Exploring the research and key findings

This editorial presents 19 papers exploring diverse health challenges and innovative interventions for Indigenous populations. These studies exemplify how integrating cultural knowledge and community engagement can drive impactful changes.

One of the central themes is access to healthcare. In Australia, the uptake of health assessments among Indigenous populations remains limited due to logistical barriers, cultural disconnects, and systemic inefficiencies (Usher et al.). Indigenous clinical leadership emerges as a crucial enabler, highlighting the importance of empowering communities to lead health initiatives. Similarly, in Mexico, the COVID-19 pandemic exacerbated reproductive health inequities, halving contraceptive use among Indigenous women (Castro-Porras et al.). These findings emphasize the fragility of healthcare systems during crises and the need for resilient, inclusive solutions.

Mental health is another recurring focus. The adaptation of mental health interventions for Diné adolescents during the pandemic underscores how culturally tailored approaches can mitigate the psychological impacts of isolation and systemic inequities (Allison-Burbank et al.). In parallel, the validation of historical loss scales for Native Hawaiian adults highlights the intergenerational trauma affecting mental health, pointing to the necessity of tools that capture culturally specific experiences (Antonio et al.).

Addressing maternal and child health disparities is also vital. In Ecuador, community-driven sexual education programs emerged as a promising strategy to reduce teenage pregnancies in Indigenous populations (Tituaña et al.). Similarly, research in Aboriginal and Torres Strait Islander communities in Australia revealed that culturally safe maternal care can help reduce stillbirth disparities (Massi et al.). For Diné toddlers, an innovative language intervention combined traditional values with developmental care to address delays caused by systemic neglect (Billey et al.). These studies collectively advocate for interventions that respect Indigenous cultural values while addressing health inequities.

The impact of systemic discrimination on Indigenous health is undeniable. Research on American Indian and Alaska Native populations documented pervasive experiences of exclusion, stigmatization, and physical and verbal threats can contribute to poorer health outcomes if not addressed by the larger population (Begay et al.). Similarly, colonial legacies have entrenched disparities in brain health, with a review calling for culturally aligned care models that prioritize community connections and resilience (Henderson et al.).

Finally, environmental health emerges as a critical domain. Water contamination caused by mining in Diné communities exemplifies how systemic injustices extend to resource access (Tsosie). Innovative solutions, such as point-of-use water filters, reflect the resilience and ingenuity of Indigenous communities in addressing these challenges.

Summary of key contributions

- Preventative health assessments in indigenous populations of Australia (Usher et al.)

This study analyzed the uptake of health assessments by Indigenous Australians. Findings revealed barriers such as limited access to services, lengthy assessments, and cultural disconnects. Indigenous clinical leadership was identified as crucial to improving participation. The study emphasizes the need for culturally sensitive strategies to enhance health outcomes.

- Reduction in contraceptive use during COVID-19 in an Indigenous Mexican Community (Castro-Porras et al.)

This research documented a 50% decline in contraceptive use among Indigenous Mexican women during the pandemic. Challenges included supply disruptions and service inaccessibility, highlighting the need for resilient healthcare systems that address reproductive health needs during emergencies.

- Recommendations for Indigenous Substance Use Disorder (SUD) Treatment (Hirchak et al.)

A scoping review assessed effective interventions for Indigenous communities, emphasizing the integration of Indigenous knowledge and adapting Western frameworks. Community engagement and culturally centered approaches were highlighted as essential for improving treatment outcomes.

- Mapping the knowledge structure and trends in Australian Indigenous Health Research (Krahe et al.)

This scientometric analysis identified a shift from deficit-focused studies to applied, culturally safe approaches. The study calls for advancing equity through respectful collaborations with Indigenous communities and addressing structural barriers within healthcare systems.

- Colonial drivers and cultural protectors of brain health among Indigenous peoples (Henderson et al.)

This review explored how colonial legacies have shaped brain health disparities. It emphasized the need for culturally relevant care models and tools, as well as addressing systemic stressors like discrimination and unequal access to care.

- Small for gestational age and anthropometric body composition from early childhood to adulthood (Hansen et al.)

This cohort study examines changes in anthropometric measurements, including fat measures, across the life course comparing small for gestational age and non-small for gestational age individuals living in urban and remote communities.

- Honoring our teachings: children's storybooks as indigenous public health practice (Maudrie et al.)

This study described the development of a culturally grounded storybook promoting mental health and resilience among AIAN children. By integrating cultural teachings with public health guidance, the project highlighted storytelling as an effective community-based intervention.

- A community-embedded approach to increasing the health literacy of Aboriginal children in a regional area (Good et al.)

This study explores the implementation of a child-centered, co-designed and community-integrated program to improve health and wellbeing outcomes for Aboriginal children in the middle childhood years.

- A psychometric analysis of historical loss scales among native Hawaiian adults (Antonio et al.)

This study validated tools for measuring historical loss and its associated symptoms, such as depression and anxiety. It underscores the importance of addressing intergenerational trauma in developing culturally sensitive interventions for Native Hawaiian communities.

- **A** rural teledentistry care experience in a Mapuche Community in Chile (Beltrán et al.)

This study demonstrated the potential of teledentistry to address oral health disparities in rural Indigenous communities. The approach improved access to care and reduced health inequities for elders.

- Factors influencing survival and mortality in aboriginal Australians with bronchiectasis (Heraganahally et al.)

The study identified factors such as ICU visits and pseudomonas infections as increasing mortality risk, while higher BMI and better lung function were protective. These findings stress the need for targeted interventions to reduce mortality.

- Reducing teenage pregnancy in rural Ecuadorian Indigenous communities (Tituaña et al.)

The study emphasized the importance of culturally tailored sexual education programs to address teenage pregnancy. Community engagement and youth-focused approaches were deemed essential for reducing pregnancy rates among Indigenous adolescents.

- Adapting safety planning interventions for Diné communities (Allison-Burbank et al.)

This project culturally adapted mental health interventions for Diné adolescents during COVID-19. It highlighted the value of community-driven approaches in addressing mental health challenges.

- Development of "+language is medicine" for Navajo toddlers (Billey et al.)

The study created a culturally responsive intervention to address developmental delays in Diné toddlers. The program integrates Indigenous values and language, emphasizing culturally grounded approaches to early childhood health.

- Addressing cervical cancer disparities in Indigenous women in Latin America (Muslin)

This article highlighted structural and cultural barriers to HPV vaccination and cervical cancer screening. It advocates for community-driven, culturally tailored interventions to reduce disparities.

- Menopause, blood pressure, and osteoporosis in rural women (Jin et al.)

The study found associations between high blood pressure and osteoporosis in postmenopausal women, emphasizing the need for gender-sensitive healthcare strategies in resource-limited settings.

- Discrimination among American Indian and Alaska native populations (Begay et al.)

This study documented widespread experiences of discrimination and its detrimental health effects. It calls for public health strategies to mitigate these impacts on Indigenous communities.

- Mining legacies and water contamination in Navajo Communities (Tsosie)

This article explored water contamination challenges in Diné communities and proposed innovative, community-centered filtration solutions to improve water safety.

- Looking after Bubba for all our mob (Massi et al.)

This study addressed stillbirth disparities in Aboriginal communities, advocating for culturally safe, family-centered approaches and Indigenous birthing practices to improve maternal health outcomes.

Conclusion

The collective insights from these 19 studies underscore a compelling call to action: addressing health disparities among Indigenous populations is not only a moral imperative but a vital step toward achieving global health equity. Promoting Indigenous health demands more than addressing systemic inequities—it requires valuing cultural knowledge, empowering community leadership, and prioritizing co-designed, culturally sensitive solutions. The articles in this Research Topic provide valuable insights into the unique health challenges faced by Indigenous communities and offer pathways to bridge these gaps. However, much work remains to be done. Centering Indigenous voices in research, policy, and practice is essential for driving meaningful and sustainable progress. Only through sustained efforts and equity-driven policies can we hope to close the health gap and secure a healthier, more just future for all Indigenous populations.

Author contributions

EO-P: Conceptualization, Supervision, Validation, Writing – original draft, Writing – review & editing. RB: Conceptualization, Writing – review & editing. JV-G: Formal analysis, Writing – review & editing. JI-C: Investigation, Writing – review & editing.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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References

1. Ortiz-Prado E, Cevallos-Sierra G, Vasconez E, Lister A, Pichilingue Ramos E. Avoiding extinction: the importance of protecting isolated Indigenous tribes. *AlterNative.* (2021) 17:130–5. doi: 10.1177/1177180121995567

2. Wylie L, McConkey S. Insiders' insight: discrimination against Indigenous peoples through the eyes of health care professionals. *J Racial Ethn Health Disparities.* (2019) 6:37–45. doi: 10.1007/s40615-018-0495-9

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Preventative health assessments and indigenous people of Australia: a scoping review

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Given that Indigenous populations globally are impacted by similar colonial global legacies, their health and other disaprities are usually worse than non-indigenous people. Indigenous peoples of Australia have been seriously impacted by colonial legacies and as a result, their health has negatively been affected. If Indigenous health and wellbeing are to be promoted within the existing Australian health services, a clear understanding of what preventive health means for Indigenous peoples is needed. The aim of this scoping review was to explore the available literature on the uptake/engagement in health assessments or health checks by Indigenous Australian peoples and to determine the enablers and barriers and of health assessment/check uptake/engagement. Specifically, we aimed to: investigate the available evidence reporting the uptake/engagement of health checks/assessments for Australian Indigenous; assess the quality of the available evidence on indigenous health checks/assessments; and identify the enablers or barriers affecting Indigenous persons' engagement and access to health assessment/health checks. A systematic search of online databases (such as Cinhl, Scopus, ProQuest health and medicine, PubMed, informit, google scholar and google) identified 10 eligible publications on Indigenous preventive health assessments. Reflexive thematic analysis identified three major themes on preventive health assessments: (1) uptake/engagement; (2) benefits and limitations; and (3) enablers and barriers. Findings revealed that Indigenous peoples' uptake and/or engagement in health assessments/check is a holistic concept varied by cultural factors, gender identity, geographical locations (living in regional and remote areas), and Indigenous clinical leadership/staff's motivational capacity. Overall, the results indicate that there has been improving rates of uptake of health assessments by some sections of Indigenous communities. However, there is clearly room for improvement, both for aboriginal men and women and those living in regional and remote areas. In addition, barriers to uptake of health asessments were identified as length of time required for the assessment, intrusive or sensitive questions and shame, and lack of access to health services for some. Indigenous clinical leadership is needed to improve services and encourage Indigenous people to participate in routine health assessments.

KEYWORDS

indigenous, Australian, indigenous health assessments, preventive health, barriers, scoping review

Introduction

Given that Indigenous populations globally are impacted by similar colonial global legacies, their health and other disapritities are usually worse than non-indigenous people (1). This is similar to many other Indigenous groups across the globe (2, 3). In Australia, Indigenous populations also have higher reported morbidity and mortality rates (4, 5) increased susceptibility to chronic disease (1, 5, 6), and lower rates of engagement and access to preventative health care (4, 5, 7) due to the many factors including the lack of access to appropriate services and racism (6, 7). Indigenous people in Australia represent approximately 3.3 percent of the total population. However, this rate differs significantly between States and Territories and urban and rural/remote locations varying between 1.8 percent in major cities to 32 percent in remote and very remote locations (8). As preventative health care is known to have a positive impact on the management of chronic conditions (6), it is important to understand Indigenous peoples' access to these services and the enablers and barriers that affect access to available services.

Recent research suggests the rate of preventative health care being accessed has decreased during the recent pandemic (7), which is likely to have a negative impact on overall health of Indigenous people, especially those with chronic disease. Barriers to accessing preventative health care includes rurality, affordability, availability, lack of awareness of preventative health care services, and inappropriate services/resources (4, 5). Understanding the barriers and enablers of access to preventative health care by Indigenous people is important to help improve access to preventative health care services and to improve the overall health of communities. Hence, a scoping review is timely to explore the available literature related to Indigenous persons' health-seeking behaviours regarding preventative health care.

Primary, secondary and tertiary prevention is defined as the following: Primary prevention focuses prevention of disease using health promotion strategies and interventions to target at risk populations; Secondary prevention focuses on intervention such as early detection of disease through screening and interventions; Tertiary prevention focuses on reducing the impact of an existing disease (9). For the purpose of this scoping review we will focus on health assessment or health checks which are used as both a primary and secondary prevention tool (10).

Health checks/assessments were identified in the Indigenous chronic disease package as preventative measure for chronic disease (1, 5) and as a key performance indictor of health by the National Indigenous Reform Agreement (5). Health assessment was first introduced into Medical Benefits Scheme (MBS) for Indigenous and Torres Strait Islander people aged over 55 years in 1999 and for those aged between 15 and 54 in 2004 (11). The main item number for Indigenous health assessment in Australia is MBS 715, which usage rate, according to Australian Bureau of Statistics, increased from 11% in 2010–11 to 29% in 2016–17 (12). This demonstrates an increase in engagement in the health assessment/check for Indigenous people, but indicates that more needs to be done to increase engagement and access to have an impact on health outcomes and chronic disease for Indigenous Australians. However, it is important to recognise that access to preventive health varies across Australia with people living in regional, rural and remote locations having less access to these services (13). Given the importance of this information to the future development of the preventive health check strategy for Indigenous people in Australia, we aimed to summarize the exisiting evidence using a standardized scoping review methodology.

The aim of this scoping review was to explore the available literature on uptake/engagement in health assessment or health check, for Indigenous Australian peoples and to determine the barriers and enablers of health assessment/check uptake/engagement. Specifically, we aimed to map the (1) evidence reporting the uptake/engagement of health checks/assessments for Australian Indigenous; and, (2) the enablers or barriers affecting Indigenous persons' engagement and access to health assessment/health checks.

Methods

Design

A scoping review methodology was selected as it was deemed the most appropriate method to explore and examine the available evidence in this specified field, and to allow the research team to provide a scope of what is reported in the literature around a particular concept, to identify gaps in the literature, and highlights areas of future research (14, 15). The eligibility criteria for inclusion and exclusion of literature in this review were determined using the PCo (Population and Context).

Inclusion Criteria

1. Studies were included if indigenous perspectives were found for a study population with uptake/barriers to healthcare facilities.
2. Indigenous peoples worldwide
3. Published in English
4. Original research including qualitative, quantitative and mixed methods. Grey literature includes Google Scholar
5. Full text available

Exclusion Criteria

1. Literature reviews (relevant articles from these included), commentaries, editorials, book reviews, letters to the editor, or where the full text was not available.
2. Non-English publications

Search terms

Indigenous OR First Nation* OR Aborig* OR Torres Strait Islander AND Health Assessment OR Health Check

Search strategy

The search included a comprehensive strategy to identify the available literature pertaining to Indigenous health assessment/health checks uptake using the search terms. One reviewer performed and conduct the initial search of evidence to determine key terms and develop the search string. A health librarian was consulted to ensure the databases and search string would produce the desired results. The search was pilot tested in one database (selected by the research team) to ensure the search strategy was robust enough to capture the required evidence, before the search strategy was finalised. The search was then conducted by two researchers across the following databases Cinhl, Scopus, ProQuest health and medicine, PubMed, informit, google scholar and google. For each database the relevant papers were identified and the reference, title, abstract and keywords were exported as .Ris file into EndNote and duplicates removed. The remaining results were exported to Covidence, where two reviewers undertook title and abstract screening followed by full text screening. The reviewers meet to discuss any conflicting decisions, if the two reviewers were unable to make a final decision a third reviewer was consulted and final decision made. The PRISMA flow chart (16) was used to report results of the screening process. It is worth mentioning that given critical appraisal and risk of bias is not required for scoping reviews (17), this was not included in the manuscript.

Data extraction and analysis

One reviewer extracted data from the evidence included in the review using the standardized Joanna Briggs Institute data extraction and checked by a second reviewer. The data extraction tool gathers specific information on population, context, culture, geographical location, study methods, the phenomena of interest relevant to the review objectives, and source type. Disagreements between the two reviewers were resolved through discussion, or by a third reviewer. A reflexive thematic analysis was undertaken to extract findings under common themes. Following the six steps narrated by Braun and Clarke (18), which was commonly used in the previous health research (19, 20), reflexive thematic analysis was done. First, two authors familiarised with the data of the finally included studies, followed by deep immersion with the data by reading and re-reading. Second, initial coding was then generated. Third, the codes were used to form initial themes. Fourth, the collapsing and refining the codes and themes. Fifth, identifying the story in support of the themes. Finally, a review of the themes and subthemes was conducted to check whether they reflected the meaning of the coded extracts and data set appropriately. The findings were reviewed by the research team.The findings and illustration of findings are available in the Supplementary file.

Results

The search identified 195 sources of literature, 60 duplicates were removed, leaving 135 for title and abstract screening. Of these 24 were retrieved for full-text screening, of which 9 meet the inclusion criteria. Reference lists of these 9 papers were searched and a further 4 were identified for title and abstract screening of which only 1 meet the inclusion criteria ($n = 10$; see Figure 1).

A quality assessment/risk of bias was undertaken of the included studies using the Mixed Methods Assessment Tool (MMAT) (21). All 10 studies were rated as medium to high quality, Table 1 presents the full results of the quality appraisal.

The characteristics of the studies included in this review are presented in Table 2. Of the 10 studies, seven studies collected quantitative data ($n = 7$) and three studies collected qualitative data ($n = 3$). All studies discussed Indigenous engagement with in regard to preventative health care, however few studies presented the view of Indigenous community members.

Data analysis revealed three main themes which address the aims and objectives of the scoping review: (1) uptake/engagement; (2) benefits and limitations; and, (3) enablers and barriers. Table 3 presents a summary of themes reported by each study.

Theme 1: uptake/engagement

Six of the 10 studies (1, 22–26) reported on indigenous people's uptake of health assessments. The findings from Bailie et al. (1) and Panaretto et al. (25) indicate a general improvement/increase in the uptake of health assessments and attendance at clinics. The study conducted by Butler et al. (22) showed that approximately one-third (32%) of Aboriginal and Torres Strait Islander adults living in NSW received a Medicare-funded health check over a 2-year period. Besides the adults, another study observed an increase in indigenous child health checks recorded in medical records (24). One study (23) reported on engagement levels during 2011–2012, 1,169 health checks completed in Orange. However, there are also contradictory findings explored in a research conducted by Robertson et al. (26) which demonstrated statistically significant reductions in total First Nations people's health assessments during the early wave of COVID-19 (March, April, and May 2020).

There are several predictors which played an important role in the Indigenous peoples' uptake of health assessments. Research conducted by Butler et al. (22) found that women had more health check compared to men. The study also pointed out that health check varies depending on residency or locality (inner regional vs. outer regional). For example, the overall health check among the inner regional residents (33.3%) was higher than the outer regional residents (4.7%).

Theme 2: benefits and limitations

Four studies (5, 23, 27, 28) have emphasised the early identification of chronic diseases and health risk factors, which can be done by timely health checks. The early identification of diseases protects the patient from further health complexities (5). The Indigenous community-targeted health design or project (such as the 'Share and Care Check') was found culturally appropriate to attract the Indigenous peoples for health checks in their childhood (28). The best part of 'Share and Care Check' is its comprehensive nature of health check. More benefits of health assessments have been noted in the existing research. Dutton et al. (23) explored that the primary benefit was identifying the common health risk factors which include overweight (41%) and smoking (26%). The second benefit was related to receiving advice from the health professional, vaccination, and referral. The primary identification of health problems among the patients opens the door for further investigation when required. For example, Dutton et al.'s (23) study showed that overall 41% of cases received advice; 27% were prescribed new medication; 13% were vaccinated; 41% had at least one blood test ordered and 32% had further investigation; 70% were given at least one referral, most commonly to a dentist; and 42% were advised to

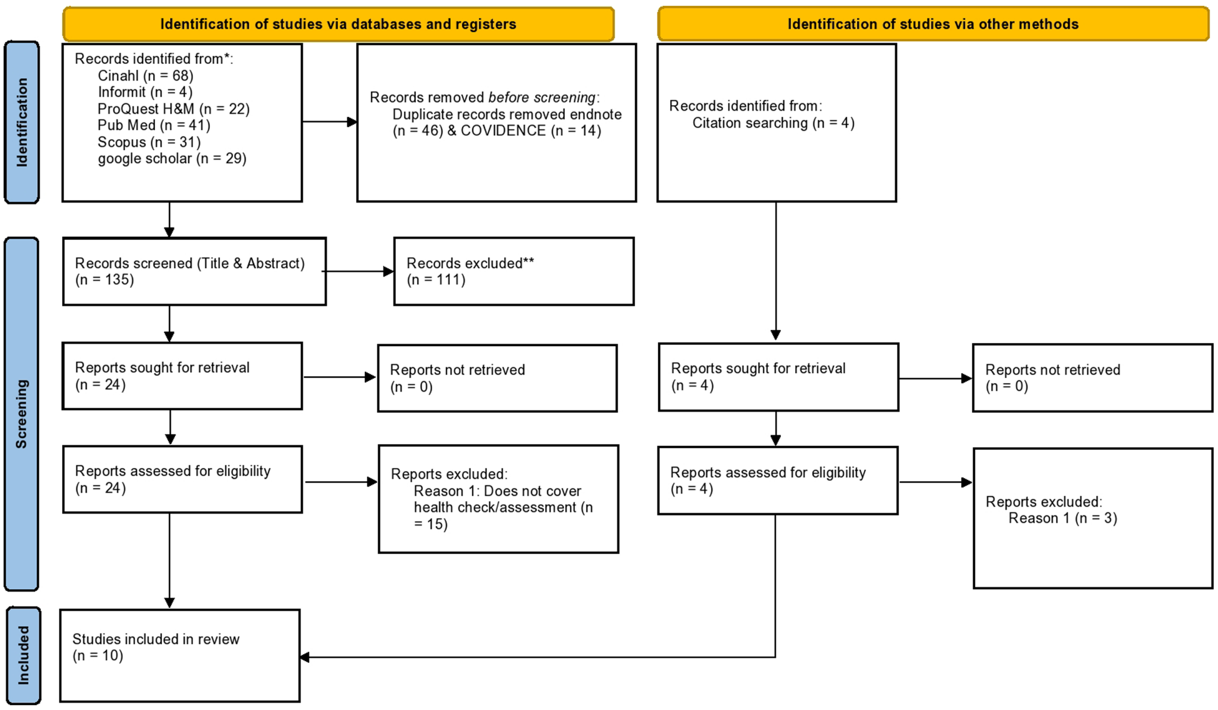

FIGURE 1
PRISMA flow chart.

return for a review. Despite the higher rate of referral, it is worth noting that there is tendency of not completing the recommended dental care treatment among the Aboriginal clients (29). It is quite unknown why they are reluctant to uptake health workers' referral for care. Time and costs associated with dental care could be an important reason of why Aboriginals are less keen to uptake medical care. Differences between medical care and dental care conditions that operate at a clinic or community level may affect uptake of dental care. This needs to be practically addressed. In addition to overweight and smoking, several new health problems (such as skin, ear, and dental problems) were also discovered during extended diagnosis. Other than the treatment-related benefits, another study [conducted by Jennings et al. (27)] focused on the financial benefits to clients, including subsidised medications and allied health consultations.

Spurling et al. (30) disclosed several limitations of health assessments. These limitations should be considered as obstacles for Indigenous peoples in accessing health services. One of the limitations is feeling shame to go to the doctor, which was evident from the following statements reported by Spurling et al. (30).

> *"I think the health checks are really important for Aboriginal and Torres Strait Islander people because some people ….feel shame to go to the doctor, and if they leave it too long there could be a problem building in their body [Liam]"* (p. 551).

Sometimes medical professionals do not go to the in-depth level or to the heart of peoples' health problems. This was considered a significant limitation in the study of Spurling et al. (30). The following statement would illustrate this clearly-

> *"I'm not sure whether it paints a really honest picture of exactly where my health's at. I think that [it] probably can go a bit more in depth [Edward]"* (p. 551).

Participants also mentioned that they felt identity crisis and/or their social world and health were not properly addressed by the existing health assessment procedures, which are mainly disease-focused. For example-

> *"I don't see how a doctor is going to solve an identity crisis. It's a social thing… [Bradley]"* (p. 551).

Theme 3: enablers and barriers

Three studies reported findings on this theme (5, 27, 28). Jennings et al. (27) proposed how to encourage Indigenous people attend regular health checks. Most felt that current community health promotion activities were inadequate, and the difficulties reaching an often-transient urban population were raised. Therefore, health promotion at the community level should be considered an important enabler to improve HC uptake. Indigenous community engagement alongside the direct promotion of the HC is necessary to make sure that more Indigenous people become interested in regular health checks. The study discovered that the existing community health activities were inadequate and hard to avail. The following statement of a participant mentioned in the study conducted by Jennings et al. (27) would make it clear-

TABLE 1 Quality appraisal MMAT.

Methodological quality of the included quantitative (descriptive) studies using MMAT (yes = 1, no = 0)									
Study	Clear research objectives/ questions	Data addressing research objectives/ questions	Relevancy of sampling strategy	Sample representativeness of target population	Appropriateness of measurement	Low risk of nonresponse bias	Appropriateness of analysis to answer research question	Total points	Ratings (6–7 = high quality, 4–5 = medium, >4 = low quality)
Reid et al. (20)	1	1	1	0	0	0	1	4	Medium
Methodological quality of the included qualitative studies using MMAT (yes = 1, no = 0)									
Study	Clear research objectives/ questions	Data addressing research objectives/ questions	Appropriate approach to answer research question	Adequate data collection methods to address research question	Findings adequately derived from data/ adequate data analysis	Interpretation of results sufficiently verified by data	Coherence between data sources, collection, analysis, and interpretation	Total points	Ratings (6–7 = high quality, 4–5 = medium, >4 = low quality)
Jennings et al. (19)	1	1	1	1	1	1	1	7	High quality
Schütze et al. (3)	1	1	1	1	1	1	1	7	High quality
Spurling et al. (2)	1	1	1	1	1	1	1	7	High quality
Bailie et al. (4)	1	1	0	1	1	0	1	5	Medium
Methodological quality of the included quantitative (non-randomized) studies using MMAT (yes = 1, no = 0)									
Study	Clear research objectives/ questions	Data addressing research objectives/ questions	Participants representativeness	Measurements appropriate	Complete outcome data	Confounders accounted in design and analysis	Conducting intended intervention	Total points	Ratings (6–7 = high quality, 4–5 = medium, >4 = low quality)
Butler et al. (14)	1	1	1	1	1	1	1	7	High quality
Dutton et al. (15)	1	1	1	1	1	1	1	7	High quality
McAullay et al. (16)	1	1	1	1	1	1	1	7	High quality
Panarett et al. (17)	1	1	1	1	1	1	1	7	High quality
Robertson et al. (18)	1	1	1	1	1	1	1	7	High quality

TABLE 2 Literature characteristics.

Citation	Aims/objectives	Study Design and methodology	Sampling	Analysis methods	Overall results	Country
Bailie et al. (4)	To describe patterns of uptake of Indigenous-specific health assessments and associated follow-up items, and examine the barriers and enablers to delivery and billing of follow-up over the first 3 years of implementation of the Indigenous Chronic Disease Package (ICDP)	- Quantitative data- The SSE was a formative evaluation covering 24 urban, regional and remote locations in all Australian states and territories. - Data were collected, analysed and reported in 6-monthly intervals over five evaluation cycles between 2010 and 2012 - Quantitative data from-Focus groups, in-depth interviews and discussions with key informants.	- Purposive - Of the 581 individual interviews done through the Sentinel Sites Evaluation (SSE), 63 contained specific information about the follow-up of health assessments - Of the 58 group interviews, 31 contained information relevant to this study, which included 103 participants - Of the 72 community focus groups, 69 provided data on access to services Qualitative data on barriers and enablers to delivery of and billing for follow-up were obtained from individual and group interviews with a range of key informants from Aboriginal Health Services (from SSE)	- Analysis of SSE data using a socioecological framework - Thematic analysis - Used an iterative approach to categorise these themes	- Aggregated data show a general improvement in uptake of health assessments and follow-up items after the baseline period - Barriers and enablers to delivery and billing of follow-up care using a socioecological framework were identified at five levels of influence: patient, interpersonal, health service, community and policy. - Negative past experiences affected patients' willingness to attend follow-up appointments. - Health service providers felt that short consultation times meant they had limited opportunity to explain reasons for referral for follow-up care to patients. This was related in part to shortage of service providers, including GPs, allied health professionals, Aboriginal Health Workers (AHWs) and practice nurses - Barriers related to Indigenous social and economic disadvantage included poor availability of transport to attend follow-up appointments and high or unpredictable cost of allied health services.	Australia
Butler et al. (14)	To quantify claims for the Aboriginal and Torres Strait Islander health check (MBS item 715) in a 2-year period among Aboriginal and Torres Strait Islander adults from the general population of New South Wales, Australia, in relation to sociodemographic and health characteristics, including prior CVD and CVD risk factors	- Quantitative - Survey questionnaire - Self-reported baseline questionnaire	- Random sampling - The study involved 1753 Aboriginal and Torres Strait Islander adults	- Frequencies and proportions were calculated for the sample according to participant characteristics, for the total sample and by claim for a health check - Logistic regression was used to estimate odds ratios (ORs) and 95% confidence intervals (95%CI) for receiving a health check in relation to participant Characteristics - The significance of the addition of an explanatory variable to the model was determined using the Wald joint test of significance. - Analyses were undertaken using Stata 14.1	- Approximately one-third of participants received a Medicare-funded health check over a 2-year period in this large population-based study of Aboriginal and Torres Strait Islander adults living in NSW, those who were disadvantaged, lived remotely, had CVD risk factors or established CVD, and had poorer self-rated health were the most likely to receive a health check. - Overall, 32% of participants had received at least one health check in the 2-year period from 1 January 2014 to 31 December 2015 - Use of GP services and poorer self-rated health remained strongly associated with receiving a health check - Most participants (91%) made at least three GP visits per year in the follow-up period, with 45% making more than 10 visits per year; 2% had no record of an MBS claim for a GP service.	Australia, NSW

(Continued)

TABLE 2 (Continued)

Citation	Aims/objectives	Study Design and methodology	Sampling	Analysis methods	Overall results	Country
Dutton et al. (15)	This study aimed to document (1) The number of each type (older person, adult, child) of AHA performed at the OAMS in 2011 and 2012; (2) The risk factors and new morbidities identified (3) The initial actions, management and 6-month follow up of any abnormalities detected	- Quantitative - Retrospective data extraction clinical records (2 year study)	- −1,169 AHAs were performed 41% child, 53% adult and 6% older person AHAs	- Descriptive statistical analysis was performed using SPSS software (version 21).	- 1,169 AHAs were performed: 52% (612) in 2011 and 48% (557) in 2012 (Table 1). Of these, 148 had two - AHAs, none had more. - Twenty-six per cent of the Orange Aboriginal population received an AHA in 2012 - The most common risk factors overall were being overweight and smoking - For all cases that were overweight and for all participants who currently smoked and intended to quit, only 37 and 60%, respectively, received an intervention. - Identification of skin and ear problems and poor dentition were similar. - The OAMS more commonly identified hypertension (18% adults) compared with the Inala adult AHAs (12%). - The OAMS changed information management systems in December 2011 and there may have been under-reporting of risk factors and intervention in the earlier period	Australia Orange
Jennings et al. (19)	The study sought to identify barriers and enablers to undertaking health checks in an urban Aboriginal Medical Service	- Qualitative - Semi-structured interviews	- Purposive - Of 30 clinical employees at the Aboriginal and Torres Strait Islander medical service (AMS), 25 staff (10 Aboriginal Health Workers (AHWs), 8 nurses and 7 doctors) participated in 20 interviews (five paired). - Eight AHWs and three nurses identified as Aboriginal, and two AHWs identified as Torres Strait Islander. Three participants were male: one AHW and two doctors.	- An inductive approach content analysis was used to identify patterns and themes in the Data - NVivo 9 software was used to assist data management and analysis. - All Aboriginal and Torres Strait Islander staff but one, explicitly and without prompting, identified community health promotion and outreach as important enablers to improve HC uptake	- Data analysis revealed that successful completion of HCs was contingent upon several interconnected components, including the client attending the AMS and consenting to the HC, and staff initiating and completing it. Barriers and potential enablers were identified at each of these stages, in addition to overarching systems within the clinics. - The AMS lacked a service-wide approach for conducting health checks (HCs), with different systems between clinics, and different systems recounted by staff within clinics - Maintaining client-centeredness was a concern for many staff who identified competing priorities in clients' sometimes stressful lives. - The study provided important insights into the barriers (e.g., inadequate practice systems and a lack of confidence for some staff in HC initiation and undertaking lifestyle brief intervention, socio-culturally sensitive health check content, and a lack of community engagement with HCs specifically, and preventative health care in general.) and enablers of increasing HC uptake.	Australia; Brisbane

(Continued)

TABLE 2 (Continued)

Citation	Aims/objectives	Study Design and methodology	Sampling	Analysis methods	Overall results	Country
McAullay et al. (16)	The primary objective of this study was to determine whether participation in the ABCD programme was associated with improved care and outcomes for Indigenous children. A secondary objective was to assess if quality differed by geographic location.	- Quantitative medical records audit - Data were collected from 59 Australian primary health-care centres providing services to Indigenous people and participating in the programme - 6-year study period (2008–2013) (February 2008 and December 2013)	- Random sample 30 records from each clinic - There were 2,360 individual file audits conducted in the 59 centres during the period February 2008 to December 2013. Only four were non-remote centres (323 individual file audits)	- Crude and adjusted logistic generalised estimating equation models were used to examine the effect of year of audit on the delivery of care. - Odds ratios (ORs) and 95% confidence intervals (95% CI) were calculated to compare the outcomes from 2008 to other subsequent years and to assess the time trend. - Data analyses were conducted using STATA 13.1	- Over the study period, the percentage of children included in recall systems significantly improved from 84% (n = 357) in 2011 to 95% (n = 415) in 2013 (OR 2.44, 95% CI 1.44–4.11) - Complete data were available for all items except checks on parent–child interaction, skin and oral health, which were available from 2011 to 2013 only. Weight checks remained consistently high (96–98%) and haemoglobin checks remained low (52–66%) from 2008 to 2013. All other child health check items showed statistically significant improvements over time (skin, oral, ears, hearing, development, interaction) (Table 2). - Hearing assessment improved the most, from 52% (n = 105) in 2008 to 89% (n = 378) in 2013 (OR 2.17, 95% CI 1.60–2.94). Skin checks improved from 73% (n = 309)	Australia; SSA, NT, WA
Panaretto et al. (17)	To examine improvements in the delivery of clinical care against key performance indicators.	- Quantitative - Longitudinal time point data (database- QAIHC) - Indigenous and non-indigenous pts. presenting to a Queensland Aboriginal and Islander Community Controlled Health Service - Data collection- June 2010 to February 2012	Convenience - The study data have been collated from data extracted by the QAIHC Core Indicator report in the Pen CAT tool. - The data collection thus represents a 'live' whole of the service patient snapshot.	- Descriptive statistics usin SPSS v19 - Proportions and 95% CIs or medians and IQRs using SPSS V.19.	Aboriginal and Torres strait islander Attendance at clinics increased from 273,692,010 to 55,441 in 2012 - The aggregated performance of participating services for health assessment increased over time. In October 2011, 8,697 (44.1%—43.4, 44.8) of the regular patients had a current health assessment.	Australia Queensland
Reid et al. (20)	To integrate cultural considerations and developmental screening into a First Nations child health check	Quantitative (Questionnaire, descriptive statistics) A short questionnaire survey via phone was conducted	Convenience - Participant's presented to GP clinical and were eligible were entered into REDCap database (developed) - A total of 118 children participated in the Share and Care Check between June 2019 and February 2020. Fifty-five caregivers consented for their child's data to be used for research purposes, and 28 caregivers consented and participated in a short feedback questionnaire.	- Descriptive statistics were reported as means and standard deviations for normally distributed continuous data, or medians and interquartile ranges for non-normally distributed data. - Normality was assessed using a Shapiro–Wilk test. Categorical variables were reported as frequencies and percentages.	The current study provides: (1) preliminary outcomes documenting cultural connections and developmental needs; and (2) feedback from caregivers regarding their experience of the Share and Care Check. theme 2 reports findings related to this lit review All caregivers reported the Share and Care Check was culturally appropriate, and the majority also reported that it was helpful (n = 23; 85.2%). A key positive feature noted by caregivers (n = 11; 40.7%) was the comprehensive nature of the health check. However, four caregivers (14%) reported that the health check took too long.	Australia, Queensland Gidgee

(Continued)

TABLE 2 (Continued)

Citation	Aims/objectives	Study Design and methodology	Sampling	Analysis methods	Overall results	Country
Robertson et al. (18)	The study aimed to assess the impact of the COVID-19 pandemic on First Nations people health assessments using an interrupted time series model.	- Quantitative (data extracted from Australian MBS database) - MBS item numbers included 715 (face-to-face health assessments), and 92,004 and 92,016 (temporary COVID-19 telehealth services).	- -Convenience	- Additive triple exponential smoothing (TES) is a forecasting method used to model and predict observations in a time series (health assessments) - Percentage differences between observed and predicted health assessments between January and June 2020 were calculated with 95% CI - Observed values falling outside the 95% CI of the model's prediction were considered statistically significant ($p < 0.05$).	- There was no significant difference between observed and predicted First Nations people health assessments in January, February, and June 2020. However, we found a statistically significant decrease in health assessments in March (16.5%), April (23.1%), and May 2020 (17.2%). The proportion of total health assessments delivered via telehealth was 0.5, 23.6, 17.6, and 10.0% for March, April, May, and June 2020, respectively - Telehealth health assessments did not entirely mitigate the reduction in face-to-face health assessments for First Nations people during the first wave of the COVID-19 pandemic	Australia
Schütze et al. (3)	This study explores some of the reasons why the uptake of Health Assessment for Aboriginal and Torres Strait Islander People remains low in some metropolitan general practices.	- Qualitative - Semi-structured interviews were conducted	- Purposive - In total, 31 out of a possible of 44 participants agreed to take part in the study (eight out of eight GPs, two of four nurses, one of one allied health professional, four of six practice managers, 16 of 25 receptionists).	- Interviews were transcribed verbatim - Thematic analysis was performed in Nvivo version 9.2. - Authors reviewed the coding of five interviews to identify differing or additional insights or meanings, which then informed the subsequent analysis.	- This study confirmed previously described barriers to MBS-715 uptake in general practice, including low rates of Indigenous status identification and a lack of awareness of MBS-715. - Additional barriers found in this study were avoidance of billing health assessments	Australia
Spurling et al. (2)	This research aimed to identify the priority health issues of the Inala Aboriginal and Torres Strait Islander community	- Qualitative - The authors situated this research in the transformative paradigm - Conducted face-to-face, semi-structured interviews	- Purposive - Twelve men and nine women took part in the interviews	- Transcribed interview data were uploaded to NVivo 9 - Thematic analysis was performed	- 3 central themes- (1) complex, interrelated, intergenerational nature of health involving social, cultural and environmental determinants of health (SCEDH); (2) ambivalence about HAs; and (3) community strength. Theme 2 reported findings related to this literature review. - Most key informants had had a health assessment (HA) with only two saying they had never had one. Key informants' experience of Aboriginal and Torres Strait Islander HAs were mixed, as four key informants gave unqualified support for the capacity of HAs to detect medical problems early. - Participants' responses suggested that their view of health and the social world was not adequately covered by HAs, which measured health in a compartmentalised, disease-focussed way.	Australia

TABLE 3 Summary themes reported.

	Theme		
Citation	Uptake/engagement	Benefits and limitations	Enablers and Barriers
Bailie et al. (4)	X		
Butler et al. (14)	X		
Dutton et al. (15)	X	X	
Jennings et al. (19)		X	X
McAullay et al. (16)	X		
Panaretto et al. (17)	X		
Reid et al. (20)		X	X
Robertson et al. (18)	X		
Schütze et al. (3)		X	X
Spurling et al. (21)		X	

> *"You know, we need to advertise it …. we need client[s] to approach us and …. request it …. both parties have to agree and … like want it from the heart. We need to educate our people more. Tell them about the health check, the importance of health check. We need posters and pamphlets or whatever we can to provide that information to our clients, 'cause I look at here … it's like 20 years back [than at home] … like in the health knowledge of the community, like in the conscious[ness].' (AHW H13)"* (p. 154).

There were several studies (5, 27, 28) who reported on the potential barriers to health checks. Firstly, Reid et al. (28) noted that the health check took too long. When health check procedures take a long time, Indigenous people may be less likely to seek this preventive opportunity. Additionally, Schütze et al. (5) found that GP services are much too time-consuming. In addition, due to the lack of strong local leadership and good communication, the development of clinic-specific systems is impossible to embed the HCs as a routine practice within busy workplace settings. By quoting a statement of a doctor, the study conducted by Jennings et al. (27) illustrated-

> *"…it needs like a practice manager who's there to make sure it's functioning and without that it's quite ad hoc …. and so doing something extra like a health check just becomes sort of an extra burden rather than a routine practice… (Dr H18)"* (p. 152).

The above statement can be seen as a reflection of the lack of services support for completion of health assessments, which is an obvious barrier to a routine health check. Medical professionals suggested important indicators (such as encouraging clinical leadership and positive attitudes with audit and feedback of health checks) to uplift motivation toward health checks which eventually elucidates the barriers to health checks (27).

The study by Jennings et al. (27) also noted disorganised management within the hospital/clinic settings. The following statement of a nurse clearly expressed the concerns about the difficulties of health check-

> *"'…. that no one's got together and we don't have a system' (RN H20)'"* (27, p. 152).

In addition, another potential barrier is related to the business of the clinics and the time needed to complete health checks.

> *"'…. it's like the size of War and Peace!' (AHW H16A)'"* (27, p. 152).

> *"'…. with the Indigenous people…. you don't keep them for a long time … otherwise they'll just get up and go out…. (RN H01-Aboriginal) '"* (27, pp. 152–153).

As a result of the above-mentioned barriers, it is important for health services to organise regular annual health assessment appointments for Indigenous people.

> *"…. in order to get a health check here you have to have an appointment, so …. that's one of the biggest barriers … you know 'oh, come back next week for a health check, yeah?' no, they're not going to come back … they got what they need now … we really need opportunistic health checks … that's what we need … (AHW H15)"* (27, p. 153).

Asking questions about lifestyle factors, particularly alcohol and smoking, but also the social history, including current home environment and overcrowding as a part of health check often deter Indigenous people from attending regular health assessment/checks services. This is not suoprising given the colonial history of child removal and other examples of colonoial interventions. Therefore, they consider health checks as difficult, sensitive, or invasive which can be illustrated by mentioning the statements used in the study conducted by Jennings et al. (27, pp. 153–154)-

> *"I felt like it was [Department of Communities] you know, the department, asking some of those questions, 'how many people living in your house?'…. that's not too bad, it's starting to get a little bit invasive but, 'does the mother drink, does the father drink?', 'how much do they drink?'…. What're we trying to achieve? We know*

> *we're gonna get social issues with a lot of these kids. So once you find out that Dad drinks every, whatever, or they're all smoking in the house, then what do you do? Again it comes back to capacity around implementing that information … (RN H07 Aboriginal)"*
>
> *"…. I think the hardest thing about the health check probably for both parties would be the lifestyle stuff, because that's the most personal …'* (Dr H18)."

Discussion

The review reveals strong uptake of health assessments with some sections of Indigenous communities. However, there is clearly room for improvement, particularly with men and those living in regional and remote areas. Butler et al.'s (22) work showed gender disparity with more women than men undergoing health assessment. This is likely because women often attend health services more than men (21), particularly between the ages of 15 and 44 years, partially because of reproductive health issues (31), and also because women are often responsible for taking children and others in their care to doctors appointments. There is a need to increase Indigenous men's engagement with regular health assessments, and in developing strategies for this to occur, it will be important for health services to engage in authentic and effective collaboration with Indigenous men to develop enhanced understandings of how services could be better configured to improve male participation. Strategies aimed at increasing uptake should consider cultural factors identified in this review such as potential for feelings of shame associated with the current questions included in health checks (30), and consider the need for an individualised approach to health assessment, with some evidence suggesting that assessments may be perceived by some Indigenous men as being superficial or lacking in adequate depth (30). There was also evidence that some Indigenous people felt the health assessment was too time consuming and this also affected uptake (28) as well as causing people to leave the clinic or hospital before the assessment is completed (27). Jennings et al. (27) also suggested a disorganised system in some disarray that was poorly prepared to meet the needs of community-dwelling Indigenous people. In light of these findings there is a need for closer collaboration with Indigenous communities and services, with a view to increasing efficiency of services, so that clients are not being left with a sense that their time is not being optimally respected and more likely to remain in the clinic until the assessment is completed.

There is clearly inequity in uptake of health assessments between inner and outer rural dwelling Indigenous people (22), and this inequity is mirrored in the broader population with outer rural dwelling people having less access to health services generally and associated poorer health outcomes (32). This disparity is often associated with distance and also with reduced health workforce availability. However, our review of the literature also suggests that urban-based services experience challenges associated with transient populations, and that Indigenous people may feel that community health services are inadequate to meet health needs, even in urban settings (27). Again, there is a need for enhanced and stronger collaboration between health services and Indigenous communities and specialist Indigenous-led services such as Aboriginal Community Controlled Organisations (ACCHOs) to work together to build stronger partnerships to enhance participation in health assessments. Furthermore, Jennings et al. (27) also highlighted the lack of clinical leadership in this important area, and that this lack of leadership resulted in services that lacked the motivation to undertake routine health assessments, or to work to streamline and improve service users experiences of health assessments. Therefore, we recommend that Indigenous clinical leadership is needed to improve services and to inspire and motivate Indigenous people and clinical staff to better participate in routine health assessments. In addition, we argue that strong Indigenous clinical leadership will provide an important clinical and cultural link between key stakeholders and this will also likely go some way towards improving community participation. It is therefore imperative that further research be conducted to better understand the reality of Indigenous peoples' preventive health attendance and access to services in Australia.

Of concern was the lack of literature that is from the perspective and voice of Indigenous people. The lack of Indigenous perspective means that the current evidence lacks an important position that must not be ignored.

Limitations

As with all reviews, there are some limitations to acknowledge in this review. Firstly, reports, grey literature, and books, which may be based on primary data, were not included in the review. There is thus a possibility that these sources may have yielded additional results. Secondly, the inclusion criteria focused on Indigenous peoples of Australia and hence the health seeking behaviours of other groups of Indigenous people may not be reflected in this review. Lastly, since the review only included literature published in English (which was a practical consideration due to authors' language limitations as well as to capture high-quality, peer-reviewed literature), the findings ignored the other language-based available evidence.

Conclusion

Regular preventive health checks are an important component of current health care services. In Australia, the implementation of annual health checks for all Indigenous people over the age of 55 was instigated in 1999 and for over 15 years since 2004 (mainly the MBS item #715). This scoping review was undertaken to investigate the uptake/engagement of health checks/assessments by Indigenous people of Australia and to identify the barriers or enablers affecting Indigenous persons' engagement and access to health assessment/health checks. The results indicate that there have been improving rates of uptake of health assessments by some sections of Indigenous communities. Our study found that Indigenous men and women living in rural areas need special attention in relation to regular health uptake. Aboriginal Australians are often found reluctant to uptake health assessments due to shame, recurring time needed for treatment purposes, culturally sensitive/intrusive questions associated with treatment procedures, and lack of health services. This study strongly suggests the need to ensure quality and culturally appropriate health services and Indigenous health leadership to improve the uptake of routine health assessments by Indigenous poeple.

It is also necessary for the Australian government to introduce new policies to support and encourage the regular uptake of health assessment by Indigenous People of Australia and provide the resources and services to ensure this occurs.

Data availability statement

The original contributions presented in the study are included in the article/Supplementary material, further inquiries can be directed to the corresponding author.

Author contributions

KU conceived the idea. KU and RJ developed the protocol. RJ and HK undertook the literature search and data extraction. KU, RJ, HK, and DJ conducted the data analysis. All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2023.1168568/full#supplementary-material

References

1. Bailie C, Matthews V, Bailie J, Burgess P, Copley K, Kennedy C, et al. Determinants and gaps in preventive care delivery for indigenous Australians: a cross-sectional analysis. *Front Public Health.* (2016) 4:34. doi: 10.3389/fpubh.2016.00034

2. Dwyer J, O'Donnell K, Willis E, Kelly J. Equitable care for indigenous people: every health service can do it. *Asia Pac J Health Manag.* (2016) 11:11–7. doi: 10.3316/ielapa.536721181318048

3. Ferdinand A, Lambert M, Trad L, Pedrana L, Paradies Y, Kelaher M. Indigenous engagement in health: lessons from Brazil, Chile, Australia and New Zealand. *Int J Equity Health.* (2020) 19:1–12. doi: 10.1186/s12939-020-1149-1

4. Peake RM, Jackson D, Lea J, Usher K. Meaningful engagement with aboriginal communities using participatory action research to develop culturally appropriate health resources. *J Transcult Nurs.* (2021) 32:129–36. doi: 10.1177/1043659619899999

5. Schutze H, Pulver LJ, Harris M. The uptake of aboriginal and Torres Strait islander health assessments fails to improve in some areas. *Aust Fam Phy.* (2016) 45:415–20. doi: 10.3316/informit.138534585530314

6. Bailie J, Schierhout GH, Kelaher MA, Laycock AF, Percival NA, O'Donoghue LR, et al. Follow-up of indigenous-specific health assessments-a socioecological analysis. *Med J Aust.* (2014) 200:653–7. doi: 10.5694/mja13.00256

7. Usher K, Bhullar N, Sibbritt D, Anubha Amarasena SS, Peng W, Durkin J, et al. Influence of COVID-19 on the preventive health behaviours of indigenous peoples of Australia residing in New South Wales: a mixed-method study protocol. *BMJ Open.* (2021) 11:e047404. doi: 10.1136/bmjopen-2020-047404

8. Australian Institute of Health and Welfare (2021). Profile of indigenous Australians. Available at: https://www.aihw.gov.au/reports/australias-welfare/profile-of-indigenous-australians [Accessed August 7, 2023].

9. NACCHO, RACGP (2018). National guide to a preventive health assessment for aboriginal and Torres Strait islander people: evidence base. Available at: https://www.racgp.org.au/FSDEDEV/media/documents/Clinical%20Resources/Resources/National-guide-3rd-ed-Sept-2018-web.pdf [Accessed December 15, 2022].

10. Commonwealth of Australia (2021). Annual healt assessment for aboriginal and Torres Strait islander people. Available at: https://www1.health.gov.au/internet/main/publishing.nsf/Content/mbsprimarycare_ATSI_MBSitem715 [Accessed November 3, 2022].

11. Kelaher M, Dunt D, Thomas D, Anderson I. Comparison of the uptake of health assessment items for aboriginal and Torres Strait islander people and other Australians: implications for policy. *Aust N Z Health Policy.* (2005) 2:21–1. doi: 10.1186/1743-8462-2-21

12. Australian Institute of Health Welfare (2017). Indigenous health check (MBS 715) data tool. Available at: https://www.aihw.gov.au/reports/indigenous-australians/indigenous-health-check-mbs-715-data-tool [Accessed November 17, 2022].

13. Australian Institute of Health and Welfare (2022). Rural and remote health. Available at: https://www.aihw.gov.au/reports/rural-remote-australians/rural-and-remote-health [Accessed August 7, 2023].

14. Bradbury-Jones C, Aveyard H. The incomplete scope of scoping reviews: a framework for improving the quality of reporting. *J Clin Nurs.* (2021) 30:e67–8. doi: 10.1111/jocn.15998

15. Harfield SG, Davy C, McArthur A, Munn Z, Brown A, Brown N. Characteristics of indigenous primary health care service delivery models: a systematic scoping review. *Glob Health.* (2018):14. doi: 10.1186/s12992-018-0332-2

16. Page MJ, McKenzie JE, Bossuyt PM, Boutron I, Hoffmann TC, Mulrow CD, et al. Statement: an updated guideline for reporting systematic reviews. *BMJ Open.* (2020) 2020:372. doi: 10.1136/bmj.n71

17. Pollock D, Ellen LD, Micah DJP, Andrea CT, Lyndsay A, Patricia M, et al. Undertaking a scoping review: a practical guide for nursing and midwifery students, clinicians, researchers, and academics. *J Adv Nurs.* (2021) 77:2102–13. doi: 10.1111/jan.14743

18. Braun V, Clarke V. Using thematic analysis in psychology. *Qual Res Psychol.* (2006) 3:77–101. doi: 10.1191/1478088706qp063oa

19. Kabir H, Maple M, Islam MS, Usher K. The paradoxical impacts of the minimum wage implementation on ready-made garment (RMG) workers: a qualitative study. *Indian J Labour Econ.* (2022) 65:545–69. doi: 10.1007/s41027-022-00375-9

20. Kabir H, Maple M, Islam MS, Usher K. A qualitative study of the working conditions in the readymade garment industry and the impact on workers' health and wellbeing. *Environ Occupl Health Pract.* (2022) 4:n/a–12. doi: 10.1539/eohp.2021-0020-OHW

21. Hong QN, Gonzalez-Reyes A, Pluye P. Improving the usefulness of a tool for appraising the quality of qualitative, quantitative and mixed methods studies, the mixed methods appraisal tool (MMAT). *J Eval Clin Pract.* (2018) 24:459–67. doi: 10.1111/jep.12884

22. Butler DC, Agostino J, Paige E, Korda RJ, Douglas KA, Wade V, et al. Aboriginal and Torres Strait islander health checks: sociodemographic characteristics and cardiovascular risk factors. *Public Health Res Pract.* (2022) 32:2103. doi: 10.17061/phrp31012103

23. Dutton T, Stevens W, Newman J. Health assessments for indigenous Australians at Orange aboriginal medical service: health problems identified and subsequent follow up. *Aust J Prim Health.* (2016) 22:233–8. doi: 10.1071/PY14120

24. McAullay D, McAuley K, Bailie R, Mathews V, Jacoby P, Gardner K, et al. Sustained participation in annual continuous quality improvement activities improves quality of

care for aboriginal and Torres Strait islander children. *J Paediatr Child Health*. (2018) 54:132–40. doi: 10.1111/jpc.13673

25. Panaretto KS, Gardner KL, Button S, Carson A, Schibasaki R, Wason G, et al. Prevention and management of chronic disease in aboriginal and islander community controlled health Services in Queensland: a quality improvement study assessing change in selected clinical performance indicators over time in a cohort of services. *BMJ Open*. (2013) 3:2083. doi: 10.1136/bmjopen-2012-002083

26. Robertson R, Mian M, Sreedharan S, Lau P. The impact of COVID-19 on first nations people health assessments in Australia. *Asia-Pac J Public Health*. (2021) 33:595–7. doi: 10.1177/10105395211011012

27. Jennings W, Spurling GK, Askew DA. Yarning about health checks: barriers and enablers in an urban aboriginal medical service. *Aust J Prim Health*. (2014) 20:151–7. doi: 10.1071/PY12138

28. Reid J, Anderson A, Cormack D, Reid P, Harwood M. The experience of gestational diabetes for indigenous Māori women living in rural New Zealand: qualitative research informing the development of decolonising interventions. *BMC Preg Childbirth*. (2018) 18:2103–8. doi: 10.1186/s12884-018-2103-8

29. Jones K, Keeler N, Morris C, Brennan D, Roberts-Thompson K, Jamieson L. Factors relating to access to dental care for indigenous south Australians. *J Health Care Poor Underserved*. (2016) 27:148–60. doi: 10.1353/hpu.2016.0042

30. Spurling GK, Bond CJ, Schluter PJ, Kirk CI, Askew DA. 'I'm not sure it paints an honest picture of where my health's at'–identifying community health and research priorities based on health assessments within an aboriginal and Torres Strait islander community: a qualitative study. *Aust J Prim Health*. (2018) 23:549–53. doi: 10.1071/PY16131

31. Bayram C, Pollack AJ, Britt H, Charles J. (2016). Why women see their GP more than men. Available at: https://theconversation.com/why-women-see-their-gp-more-than-men-49051?utm_source=twitter&utm_medium=bylinetwitterbutton [Accessed January 1, 2023].

32. Australian Institute of Health Welfare (2022). Rural and remote health. Available at: Rural and remote health - Australian Institute of Health and Welfare (aihw.gov.au) [Accessed December 22, 2022].

33. Hunt K, Adamson J, Hewitt C, Nazareth I. Do women consult more than men? A review of gender and consultation for back pain and headache. *J Health Serv Res Policy*. (2011) 16:108–17. doi: 10.1258/jhsrp.2010.009131

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Reduction in contraceptive use during the COVID-19 pandemic among women in an indigenous Mexican community: a retrospective crossover study

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Background: Being indigenous, being a woman, and living in poverty are social determinants that contribute to reduced access to healthcare, including reproductive health services. The COVID-19 pandemic might have exacerbated this lag.

Objective: This study explored how the COVID-19 pandemic affected the contraceptive use of a group of indigenous Mexican women and adolescents in their community.

Methods: Between June and December of 2021, 158 indigenous Mexican women who had experienced recurrent pregnancies were interviewed at two health centers in San Cristóbal de las Casas, Chiapas. Participants were either pregnant when they completed the questionnaire or had been pregnant during the COVID-19 pandemic. Women were asked about their contraceptive practices before and during the pandemic. The change in contraceptive practice was estimated using a logistic model.

Results: The COVID-19 pandemic reduced contraceptive use by 50%. Among women who wanted contraception, 58% did not receive it. During the pandemic, 77% of previous contraceptive users reported difficulty obtaining contraception, and only 23% sought family planning assistance.

Conclusion: During the COVID-19 pandemic, indigenous women in the studied community used fewer contraceptive methods and did not use intrauterine devices. Additionally, there was a decline in the percentage of women using contraceptives. These results highlight the impact on indigenous populations and the difficulties they could face in accessing reproductive health services during health emergencies.

KEYWORDS
contraceptive, indigenous, COVID-19, women, crossover study, Mexico

Introduction

Human rights gaps exist among certain populations in many lower-middle and low-income nations. One such group is impoverished indigenous women, whose living conditions, low educational access, and lack of employment opportunities determine their access to healthcare (1). The reproductive health of women from indigenous communities lags behind that of other women. Various factors, such as colonization's transgenerational negative effects, racism, low socioeconomic status, and lower education levels, have contributed to health disparities (2). Compared to other women, indigenous women have a greater gap in reproductive health (3, 4).

In Mexico, the 2020 Population and Housing Census identified 7,364,645 people aged 3 years or older who speak an indigenous language (6.1% of the total population), of whom 51.4% are women. According to the National Institute of Indigenous Peoples, there were 11,800,247 people in 2020 (9.4% of the country's total population). If they self-identify as indigenous, the number increases to 23.2 million people aged 3 years and older (19.4% of the total population in this age range) (5, 6).

It has been reported that Mexicans and Central Americans are poor, especially women and young women of reproductive age are the poorest (7). Similar inequalities occur in other countries; for example, in Australia was reported that there was lower access to general practitioners, allied health professionals, specialists, obstetrics service providers, and pathology and diagnostic tests among indigenous women who experienced stillbirths compared with non-indigenous women (8). There is another example of Canadian Indigenous women facing numerous health and societal challenges, including domestic violence (9).

Indigenous women in Mexico have lower rates of prenatal care, have a higher fertility rate, and become sexually active at a younger age (10). Among indigenous women, most decisions related to prenatal care are made by their partners (11, 12). Indigenous adolescent women have a higher fertility rate than their non-indigenous counterparts (13).

Unforeseen situations such as the COVID-19 pandemic could deepen the gap between the health services utilization by indigenous peoples in nations like Mexico; an example of this was found in the higher risk of death from this cause, especially outside the hospital (14). In this country, during the pandemic, emergency health authorities implemented containment measures that included converting healthcare facilities into "Covid care centers," limiting access to services that were not considered a priority during the emergency. For example, talks on the correct use of contraceptive methods or the availability of contraceptives (15).

The potential consequences on reproductive health services, particularly contraceptive methods, in indigenous communities, is one of the major concerns considering the vulnerability of indigenous populations. In this study, we employ an intersectional approach to examine the impact of the COVID-19 pandemic on the provision and use of contraceptive methods among indigenous Mexican women and adolescents. The intersectionality lens allows us to understand how various social factors, such as gender, ethnicity, and socioeconomic status, intersect and shape the experiences and vulnerabilities of this specific population. By considering the unique challenges impoverished indigenous women face, including their living conditions, limited educational access, and lack of employment opportunities, we can better comprehend the barriers they encounter in accessing reproductive healthcare. Moreover, the historical effects of colonization, systemic racism, and lower education levels contribute to health disparities in this population. By utilizing an intersectional framework, we can shed light on the complex dynamics that contribute to the reproductive health gap among indigenous women, particularly during times of crisis like the COVID-19 pandemic (16). This study aimed to explore how the COVID-19 pandemic affected the use of contraceptive methods in a group of indigenous Mexican women and adolescents.

Methods

Design and settings

We conducted a retrospective crossover study to investigate the impact of the COVID-19 pandemic on contraceptive use. The pandemic was regarded as a transient exposure, and the participants served as their own controls or matched pairs, transitioning between periods of varying risk, namely the pre-pandemic and pandemic periods. This design is typically used to study acute risks associated with transient exposures. The subjects were treated as matched pairs by crossing back and forth between periods of different risk (in this case: pre-pandemic and pandemic periods) (17, 18). To ensure the clarity of the survey questions used in this study, a pilot test was conducted with a sample of 27 participants. The pilot test served as an opportunity to refine the questionnaire and assess the participant's understanding of the items. The pilot test participants were included in the final sample of the study since no problem was found with the items or the responses. We collected data on use of contraceptive methods, sociodemographic characteristics, and beliefs about the COVID-19 epidemic through interviews following a standardized questionnaire. From June to December 2021, participants with a history of at least one previous pregnancy completed the questionnaire during their visit to the health center. As a result, they were pregnant when they filled out the questionnaire or were pregnant during the COVID-19 pandemic. This aimed to compare their pregnancy experiences related to contraceptive use during and before the pandemic. They were recruited as volunteer participants at two health centers in San Cristóbal de las Casas, México. Both health centers belonged to state services of the Mexican Ministry of Health; one belonged to the first level of care while the other one was from the second level of care. These were where the health center authorities provided us with facilities for conducting the study. In a designated private room trained personnel administered the questionnaire in Spanish. Additionally, a translator proficient in Spanish and Tzotzil (the most indigenous languages spoken in San Cristóbal de las Casas community) was present on-site throughout the data collection process.

Variables

The outcome variable was the percentage of participants who reported using contraceptives during and before the pandemic.

Ethical considerations

The Ethics and Research Committee of the School of Medicine of the National Autonomous University of Mexico approved the protocol of the present study under Research Protocol: FM/DI/115/2019. Participation in this study was voluntary. Before completion, participants were informed of their rights as outlined in the Helsinki Declaration (19). In addition, all participants were actively informed of the study's objective, their research rights, that there would be no consequences if they chose not to participate, and the confidential nature of their participation. This information was conveyed verbally and in writing. After that, all participants who were 18 years of age or older signed the informed consent form, indicating their agreement to participate. Participants under the age of 18, signed the informed assent form, which expressed their willingness to take part in the study, while their legal guardian or parent provided the informed consent. For illiterate women, consent or assent was obtained after ensuring their understanding of the form's content and confirming their proficiency in Spanish. Translator assistance was available throughout the study, although it was not necessary.

Statistical analysis

Categorical variables are expressed as frequencies and percentages (%), while continuous variables are described using means and standard deviations (SD). Age-adjusted logistic models were utilized to estimate changes in contraceptive use before and during the pandemic. The model included all factors with a value of $p < 0.05$. The Hosmer-Lemeshow test was used to evaluate the model's goodness of fit. The projected likelihood of using a contraceptive technique by age for each period was then plotted on a graph. The age intervals considered were from minimum to maximum, and between this period, we evaluated the probability every 2 years. The data were analyzed using the statistical software Stata version 15.0 (Stata Corp, College Station, Texas, United States).

Results

158 pregnant women who had previously given birth and lived in a predominantly indigenous community participated in this study. All participants understood the Spanish language.

The average age of the participants was 21.7 years old, and 13.3% were adolescents. 8.2% of participants were illiterate, nearly 95.0% spoke an indigenous language, and 4 out of 5 women did not attend school at the moment of the study. The main reason for not attending school was lack of money (32.0%), pregnancy (18.5%), and marriage (16.0%). One in two women reported home care was their main activity during the pandemic (Table 1).

Almost one-third of women used contraception during the pandemic (29.7%), and nearly half used contraception before the pandemic (47.0%). During the pandemic, 77.3% of contraceptive users reported difficulties finding a method. Among contraceptive users, 79% sought family planning counseling at a health center, and of these, 73% received it (Table 2). Before the pandemic, hormonal contraception was the most common form of contraception. Whereas, during the pandemic, the condom was. The implant was the contraceptive method that showed the greatest reduction since, during the pandemic, only 1 of the 10 previous users continued to use it (Figure 1).

TABLE 1 Sociodemographic characteristics of participants.

Characteristic	Percentage [*n*]
Age (years)[#]	21.7 [2.1]
Young women (<20 years)	13.3 [21]
School level	
Elementary or less	40.0 [58]
Secondary	52.4 [76]
High school or more	7.6 [11]
Illiteracy	8.2 [13]
School attendance	21.6 [33]
Economic problems	57.1 [64]
Pregnancy	34.9 [37]
Marriage	31.1 [32]
Own decision	27.8 [27]
Achieving the aspired level	10.2 [10]
Family decision	7.1 [7]
School far away	5.1 [5]
Personal problems	6.1 [6]
No school or space	5.1 [3]
Health problems	3.1 [3]
Religion	3.1 [3]
COVID-19	1.0 [1]
Marriage status	
Single	0.6 [1]
Marriage	84.2 [133]
Voluntary union	15.2 [24]
Main occupation during pregnancy	
Student	1.3 [2]
Housewife	47.1 [74]
Merchant	25.5 [40]
Work for several employers	24.8 [39]
Family-owned business	0.6 [1]
Indigenous language speaking	95.6 [151]
Location of residence	
San Cristóbal de las Casas	94.3 [147]
Other	5.7 [9]

San Cristóbal de las Casas, Chiapas, México, 2021. [#]Mean [Standard deviation]. Total sample = 158; Due to the non-response, some characteristics do not total to 100%.

There were more than double odds of not using any contraceptive method during the pandemic compared to the pre-pandemic period [odds ratio = 0.46 (95% CI 0.29, 0.73)] (Table 3). The plot for the probabilities resulting from the model shows that the probability of using any contraceptive method was less during the COVID-19 pandemic than in the previous period, independent of the participant's age (Figure 2).

TABLE 2 Contraceptive use and health service utilization before and during the pandemic.

Characteristic	Percentage [*n*]
Pre-pandemic contraceptive use (Rss = 158)	47.5 [75]
During the pandemic contraceptive use (Rss = 158)	29.7 [47]
Women who could not access any contraceptive method because of the pandemic (Rss = 158)	48.1 [76]
Change of contraceptive methods during the pandemic among pre-pandemic contraceptive users (Rss = 47)	25.5 [12]
Difficulty in obtaining a contraceptive method during the pandemic among pre-pandemic contraceptive users (Rss = 75)	77.3 [58]
Family planning counseling seeking at a health center among pre-pandemic contraceptive users (Rss = 75)	78.6 [59]
Get information about contraceptives when seeking counseling (Rss = 59)	72.9 [43]

San Cristóbal de las Casas, Chiapas, México, 2021.
Rss: reference sample size.

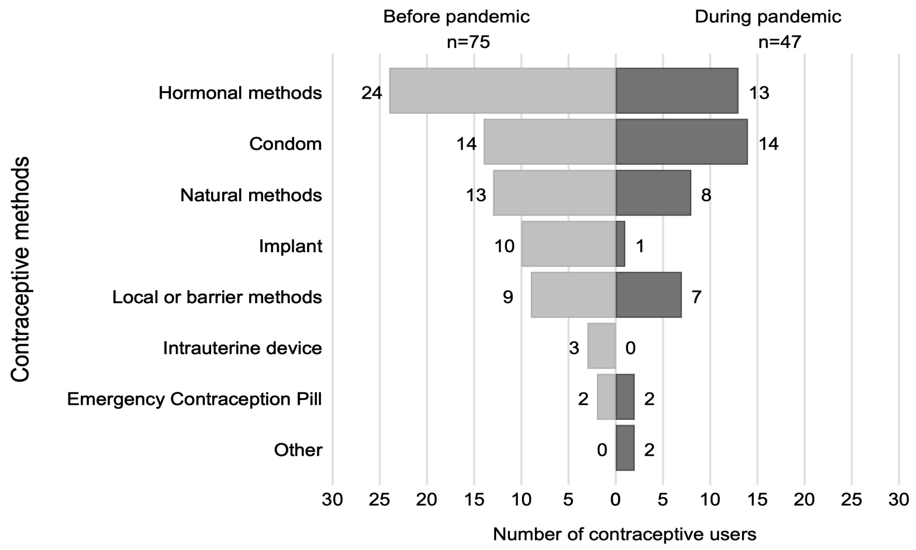

FIGURE 1
Comparison of contraceptive method use before and during the pandemic,

TABLE 3 Logistic model of using contraception*.

	OR [95% CI]	*p* value
Before pandemic	1	
During pandemic	0.46 [0.29, 0.73]	0.001

*Adjusted for age.

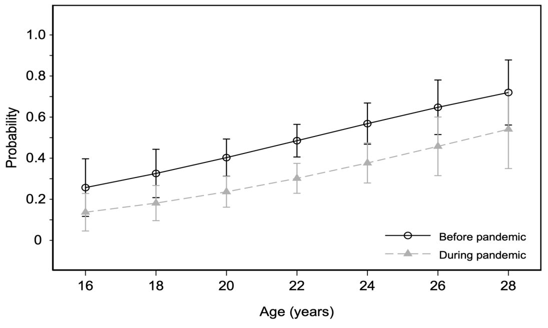

FIGURE 2
The estimated probability of contraceptive use before and during the pandemic by age.

Discussion

This study aimed to explore how the COVID-19 pandemic affected the use of contraceptive methods in a group of indigenous Mexican women and adolescents in their community. We found that less than half of the participants used any contraceptive method before the pandemic, and this percentage dropped to one-third during the pandemic.

The illiteracy rate in our study was 8.2% and only one in five women attended school. Lack of money (32%), pregnancy (18.5%) and marriage (16.0%) were the three main reasons why women did not attend school. In indigenous communities, marriage, community, and interpersonal relationships are institutionalized forms of gender oppression that increase indigenous women's vulnerability and influence their decisions regarding childbirth and health care, according to qualitative research methods (1) These conditions may partially explain why our study's participants did not attend school.

Education has a reducing effect on fertility and, above all, has an effect of delaying the timing of childbearing (20) and early pregnancy and motherhood have been strongly associated with school attendance, tertiary education, and marital or cohabitation status (21).

We found a lower percentage of participants (47%) using contraceptives before the pandemic. This result could be because the participants in our study were younger (under 28 years of age) and indigenous. Indigenous communities face significant barriers to accessing reproductive health services, which may explain the percentage of contraception use (22). Previous studies reported that lower contraceptive use in indigenous groups was associated with lower coverage ratio (23). In addition, Chiapas had the lowest percentage of sexually active women of childbearing age using contraceptive methods (63.0%); meanwhile, Mexico City reported 81.1% (24).

According to a report by the United Nations Population Fund in 2020, women with unmet contraception needs will experience a 19.6% increase in pregnancies and births resulting from the covid-19 pandemic. In this report, the analysis used economic data from Mexico and econometric models to estimate the increase (15). We found that women who could not have a method of contraception even though they wanted it was 58.3%. The small sample size could explain this result due to the small percentage of contraception users.

No significant differences were found related to being an adolescent for contraceptive use. Hubert et al. reported similar results. They analyzed the 2015 Mexican National Survey of Boys, Girls, and Women (ENIM) to explore the factors associated with pregnancies and births among young adults. According to their study, the indigenous background did not significantly influence the likelihood of an adolescent becoming pregnant. Among adult women, however, the indigenous group (87%) was significantly more likely than the non-indigenous group (64%) (25). However, our results do not agree with the reported in a previous study where the authors reported high rates of adolescent girls who did not use any form of family planning before becoming pregnant (26). Their study included only urban residents, which may explain the differences.

Concerning the type of contraceptive method used, a secondary analysis of data from the National Health and Nutrition Survey (Ensanut) for the periods 2012 and 2018–19 found, in the adolescent population, an increase in the use of long-acting reversible contraceptives (ARAP), among which are the intrauterine device and the contraceptive implant, in addition to condoms being the method most used by adolescents (21). In contrast, in the present study, among the methods least used by the participants are ARAP, with the intrauterine device being the least used and hormonal methods being the most used before and during the pandemic. This may be because the sample included people of different ages and most participants said it was hard to get a method during the pandemic. This is consistent with a study on the lack of ARAP availability in first-level units, provider-user relationships, and user information during the COVID-19 pandemic (27). Darney et al. analyzed data from three waves of a nationally representative, population-based survey to describe contraceptive use and education among rural adolescents (15–19) and young women (20–24) in Mexico. According to their study, adolescent and young adult women with high educational attainment, positive marital status, pregnancy experiences, and health insurance use modern contraceptives (28). According to recent research, indigenous women had a lower coverage ratio for modern contraception than non-indigenous women (CR: 0:73; CI 0:65–0:83) (3).

In addition, it was found that more than 70% of the sample reported difficulty in obtaining a contraceptive method. Based on previous epidemics, such as Ebola in 2014 (29) and Zika in 2015 (30), it has been documented that people stop attending health services even when needed due to fear of contracting the disease. In addition, health services may present limitations because they focus exclusively on care for patients infected with the virus. These data show the importance of identifying moderating variables for the use of contraceptive methods in healthcare situations, especially in indigenous populations whose demographic and social characteristics make them at-risk groups.

It has been found that 89.3% of indigenous language speakers know of some method of contraception, whereas 99.2% of non-indigenous language speakers do. The gap becomes wider when it comes to understanding how contraceptive methods work since only 75.3% of indigenous speakers know how they work, compared to 95.9% of non-indigenous speakers. It may explain, in part, the low prevalence of contraceptive methods (24).

Due to the diverse containment measures adopted in response to the pandemic, the consequences generated are also diverse. We found more than double the possibility of not using contraceptives during the pandemic compared to the pre-pandemic period. The results of our study are in line with the trend estimated using a model to analyze the consequence of the covid-19 pandemic on access to contraceptives in Latin America and the Caribbean. When developing the model, considerations included the need to purchase contraceptives privately, the shortage of contraceptives in public services, the discontinuity of sexual and reproductive health services, and the reluctance of users to visit clinics for fear of infection during the pandemic (15). There is a need to explore further the reasons for low contraceptive use in everyday situations, not just during emergencies. The consequences of some factors on contraceptive use, such as indigenous status, should be considered for future health policies.

Limitations

One notable advantage of this study is that it relied on primary data gathered through a questionnaire designed for this purpose. Moreover, the participants provided a comparison group by reporting their pregnancy experiences before the pandemic. While the authors recognize the potential for recall bias with a retrospective cross-over design, this is deemed less likely due to the non-traumatic nature of the questions asked. In Mexico, numerous indigenous groups speak distinct languages and reside in diverse communities with varying beliefs and perspectives, including reproductive health. Although our findings are based on a limited sample of participants from a specific community, they might be relevant for other communities sharing comparable characteristics.

Conclusion

During the COVID-19 pandemic, indigenous women in the studied community used fewer contraceptive methods and did not use intrauterine devices. Additionally, there was a decline in the percentage of women using contraceptives. The results highlight the impact on indigenous populations and the difficulties they could face in accessing reproductive health services during health emergencies.

Data availability statement

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding author.

Ethics statement

The studies involving human participants were reviewed and approved by The Ethics and Research Committee of the School of Medicine of the National Autonomous University of Mexico. Written informed consent to participate in this study was provided by the participant or the participants' legal guardian/next of kin if they were underaged.

Author contributions

All authors listed have made a substantial, direct, and intellectual contribution to the work and approved it for publication.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

References

1. Gamlin JB. You see, we women, we can't talk, we can't have an opinion\ldots. The coloniality of gender and childbirth practices in indigenous Wixárika families. *Soc Sci Med*. (2020) 252:112912. doi: 10.1016/j.socscimed.2020.112912

2. Valeggia C. The global and the local: health in Latin American indigenous women being indigenous in Latin America: from global to local biologies *Health Care Women Int*. (2017) 37:1–12. doi: 10.1080/07399332.2014.954704,

3. Armenta-Paulino N, Wehrmeister FC, Arroyave L, Barros AJD, Victora CG. Ethnic inequalities in health intervention coverage among Mexican women at the individual and municipality levels. *EClinicalMedicine*. (2022) 43:101228. doi: 10.1016/j.eclinm.2021.101228

4. Noreña-Herrera C, Leyva-Flores R, Palacio-Mejía LS, Duarte-Gómez MB. Disparidades étnicas no uso de serviços de saúde reprodutiva por mulheres colombianas indígenas e afrodescendentes. *Cad Saude Publica*. (2015) 31:2635–48. doi: 10.1590/0102-311X00016515

5. INEGI. *Estadísticas a propósito del día internacional de los pueblos indígenas*. México: INEGI (2022). Available at: https://www.inegi.org.mx/contenidos/saladeprensa/aproposito/2022/EAP_PueblosInd22.pdf

6. INEGI-INSP. *Censo de Población y Vivienda 2020, México*. (2021). Available at: https://www.inegi.org.mx/programas/ccpv/2020/ (Accessed June 23, 2022).

7. Emily D, Hernandez B, Mokdad AH. Antenatal care among poor and indigenous women in Central America and Mexico: a cross-country study of access, utilization, and barriers In: DA Schwartz, editor. *Maternal death and pregnancy-related morbidity among indigenous women of Mexico and Central America: an anthropological, epidemiological, and biomedical approach*. Cham: Springer International Publishing (2018). 133–44.

8. Callander E, Fox H, Mills K, Stuart-Butler D, Middleton P, Ellwood D, et al. Inequitable use of health services for indigenous mothers who experience stillbirth in Australia. *Birth*. (2022) 49:194–201. doi: 10.1111/birt.12593

9. Klingspohn DM. The importance of culture in addressing domestic violence for first Nation's women. *Front Psychol*. (2018) 9:872. doi: 10.3389/fpsyg.2018.00872

10. Hernández MF, Meneses E, Sánchez M. Mujeres indígenas y su acceso a los derechos sexuales y reproductivos, 2009 y 2014., *La situación demográfica de México 2016*. Ciudad de México: Consejo Nacional de Población (2017). p. 57–86. Available at: https://www.gob.mx/conapo/documentos/la-situacion-demografica-de-mexico-2016

11. Hughes J. *Gender, equity, and indigenous women's health in the Americas*. Washington, DC: Pan-American Health Organization (2004).

12. Wurtz H. Indigenous women of Latin America: unintended pregnancy, unsafe abortion, and reproductive health outcomes. *Pimatisiwin*. (2012) 10:271–382.

13. Vázquez-Sandrin G, Ortíz-ávila E. Family planning and indigenous population fertility in urban mexico. *Papeles de Poblacion*. (2020) 26:157–84. doi: 10.22185/24487147.2020.103.06

14. Ibarra-Nava I, Flores-Rodriguez KG, Ruiz-Herrera V, Ochoa-Bayona HC, Salinas-Zertuche A, Padilla-Orozco M, et al. Ethnic disparities in COVID-19 mortality in Mexico: a cross-sectional study based on national data. *PLoS One*. (2021) 16:1–10. doi: 10.1371/journal.pone.0239168

15. United Nations Fund for Population Activities México, Latin America and Caribbean Regional Office, & Gobierno de México. *Estimaciones del Impacto de la Pandemia de la COVID-19 en la Salud Sexual y Reproductiva en México* (p. 20). UNFPA México, UNFPA LACRO, SGCONAPO. (2021). Available at: https://lac.unfpa.org/es/resources/estimaciones-del-impacto-de-la-pandemia-de-la-covid-19-en-la-salud-sexual-y-reproductiva

16. Bowleg L. The problem with the phrase women and minorities: intersectionality-an important theoretical framework for public health. *Am J Public Health*. (2012) 102:1267–73. doi: 10.2105/AJPH.2012.300750

17. Maclure M. The case-crossover design: a method for studying transient effects on the risk of acute events. *Am J Epidemiol*. (1991) 133:144–53. doi: 10.1093/oxfordjournals.aje.a115853

18. Mittleman MA, Mostofsky E. Exchangeability in the case-crossover design. *Int J Epidemiol*. (2014) 43:1645–55. doi: 10.1093/ije/dyu081

19. Cook RJ, Dickens BM, Fathalla MF. World medical association declaration of Helsinki: ethical principles for medical research involving human subjects In: RJ Cook, BM Dickens and MF Fathalla, editors. *Reproductive health and human rights: integrating medicine, ethics, and law*. Oxford University Press (2003) 429–32.

20. Vignoli JR, Cavenaghi S. Adolescent and youth fertility and social inequality in Latin America and the caribbean: what role has education played? *Genus*. (2014) 70:1–25.

21. Villalobos A, Ávila-Burgos L, Hubert C, Suárez-López L, de la Vara-Salazar E, Hernández-Serrato MI, et al. Prevalencias y factores asociados con el uso de métodos anticonceptivos modernos en adolescentes, 2012 y 2018. *Salud Publica de Mexico*. (2020) 62:648–60. doi: 10.21149/11519

22. Ibáñez-Cuevas M, Heredia-Pi IB, Meneses-Navarro S, Pelcastre-Villafuerte B, González-Block MA. Labor and delivery service use: indigenous women's preference and the health sector response in the Chiapas highlands of Mexico. *Int J Equity Health*. (2015) 14:1–10. doi: 10.1186/s12939-015-0289-1

23. Armenta-Paulino N, Castelló A, Sandín Vázquez M, Bolúmar F. How the choice of ethnic indicator influences ethnicity-based inequities in maternal health care in four Latin American countries: who is indigenous? *Int J Equity Health*. (2020) 19:1–12. doi: 10.1186/s12939-020-1136-6

24. Fernández Moriano C. *Situación de la Salud Sexual y Reproductiva*. CONAPO, editor. México (2019). 4–32 p. Available at: https://botplusweb.portalfarma.com/documentos/2019/2/21/131476.pdf

25. Hubert C, Villalobos A, Abreu AB, Suárez-López L, de Castro F. Factors associated with pregnancy and motherhood among mexican women aged 15–24. *Cad Saude Publica*. (2019) 35:e00142318–2. doi: 10.1590/0102-311X00142318

26. Blackaller-Ayala J, Mejía-Mendoza ML, Ortiz-Villalobos RC, Gil-Hernández E, Benitez-Morales R, Laureno-Eugenio J. Condiciones socioculturales y experiencia del

embarazo en adolescentes de Jalisco, México: estudio cualitativo. *Rev Colomb Obstet Ginecol*. (2016) 66:242–52. doi: 10.18597/rcog.291

27. Gutiérrez-Galindo M, Santiago-Hernández K, Vargas-Campuzano E, Barquet-Bazán N, Moreno-Ortiz J. Artículo de revisión. Planificación familiar y covid-19: un servicio prioritario del primer nivel de atención. *Ocronos*. (2022) 5:15.

28. Darney BG, Weaver MR, Sosa-Rubi SG, Walker D, Servan-Mori E, Prager S, et al. The oportunidades conditional cash transfer program: effects on pregnancy and contraceptive use among young rural women in mexico. *Int Perspect Sex Reprod Health*. (2013) 39:205–14. doi: 10.1363/3920513

29. Bietsch K, Williamson J, Reeves M. Family planning during and after the west African Ebola crisis. *Stud Fam Plan*. (2020) 51:71–86. doi: 10.1111/sifp.12110

30. Marteleto LJ, Weitzman A, Coutinho RZ. Women's reproductive in Brazil during the Zika epidemic in Brazil. *Popul Dev Rev*. (2020) 43:199–227. doi: 10.1111/padr.12074

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Coming together for something good: recommendations from a scoping review for dissemination and implementation science to improve indigenous substance use disorder treatment

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Introduction: Dissemination and Implementation (D&I) science is growing among Indigenous communities. Indigenous communities are adapting and implementing evidence-based treatments for substance use disorders (SUD) to fit the needs of their communities. D&I science offers frameworks, models, and theories to increase implementation success, but research is needed to center Indigenous knowledge, enhancing D&I so that it is more applicable within Indigenous contexts. In this scoping review, we examined the current state of D&I science for SUD interventions among Indigenous communities and identified best-practice SUD implementation approaches.

Methods: PubMed and PsycINFO databases were queried for articles written in English, published in the United States, Canada, Australia, and New Zealand. We included key search terms for Indigenous populations and 35 content keywords. We categorized the data using the adapted and extended Reach, Effectiveness, Adoption, Implementation, and Maintenance (RE-AIM) framework that emphasizes equity and sustainability. RE-AIM has also been used as a primary model to consistently identify implementation outcomes.

Results: Twenty articles were identified from the original unduplicated count of over 24,000. Over half the articles discussed processes related to Reach, Adoption, and Implementation. Effectiveness was discussed by 50% of the studies ($n = 10$), with 25% of the articles discussing Maintenance/sustainability ($n = 4$). Findings also highlighted the importance of the application of each RE-AIM domain for meaningful, well-defined community-engaged approaches.

Conclusion: Finding indicated a need to prioritize Indigenous methods to culturally center, re-align and adapt Western treatments and frameworks to increase health equity and improve SUD treatment outcomes. Utility in the use of the modified RE-AIM and the continued modification for Indigenous communities was also noted.

KEYWORDS

dissemination and implementation science, indigenous research methods, community-based participatory research, American Indian and Alaska native adults, indigenous, scoping review, cultural centering

1. Introduction

Many Indigenous communities are interested in providing culturally responsive treatments for substance use disorders (SUD) to their communities (1). While alcohol and rates of other substance use varies greatly across Indigenous communities and reasons for these variation are complex (2–5), there is a need to understand how to better integrate culturally appropriate approaches specific to community and Tribal histories, culture, policy, and concepts of well-being and recovery to reduce the negative consequences of substance use more effectively (3–5). Over the last decade, research on evidence-based SUD treatments among Indigenous people has increased (e.g., motivational interviewing, community reinforcement approach, contingency management), which creates an opportunity to assess the strategies used to implement these treatments in community- and Tribal-based clinical settings (1, 6, 7).

In non-Indigenous focused research, dissemination and implementation (D&I) science models, frameworks, and strategies can guide and facilitate successful adoption, implementation, and sustainment of evidence-based practices to enhance participant outcomes. Indigenous D&I research is emerging and growing (8). Given the historical and ongoing harms created by extractive research practices, it is important that strategies related to implementation of SUD treatment use a community-engaged approach to facilitate equitable research partnerships (9) and work towards health equity.

Recent D&I and intervention research among Indigenous communities has commonly used a community-based participatory research (CBPR) framework to address chronic health conditions and health behaviors related to disease prevention [e.g., hypertension, cardiovascular disease, nutrition, substance misuse prevention and treatment (9) and wellness (10–12)]. CBPR has been widely adopted by Indigenous researchers and community research sites because the approach aligns with Indigenous values of centering the knowledge and expertise of the community, and the importance of Tribal sovereignty with applied outcomes that directly support culturally centered community wellbeing and capacity building. CBPR processes facilitate bi-directional learning and power-sharing between communities and researchers in every step of the process by addressing issues of equity, partnership voice and trust (11–14). While CBPR is thought of as an implementation approach, it has only more recently been conceptualized within the D&I context (15, 16).

Although there are numerous D&I frameworks, models, and theories to support program implementation (17), a commonly used framework to guide implementation outcomes is the Reach, Effectiveness, Adoption, Implementation and Maintenance (RE-AIM) Framework (18, 19) which has recently been expanded to include considerations around contextual factors, longer-term sustainability, and equity (20, 21). RE-AIM is comprised of five domains, each of which reference a particular area of evaluation: **R**each refers to the proportion and characteristics of people who are affected by and engaged in the intervention; **E**ffectiveness is how well the intervention works in a given setting; **A**doption is the proportion and representativeness of participating and non-participating providers and settings; **I**mplementation is the extent to which the intervention is delivered as intended (e.g., fidelity); and **M**aintenance (or Sustainability) is the extent to which the intervention becomes part of routine practice, as well as the long-term impact of the intervention.

While RE-AIM has evolved and been used within a range of contexts over the years (19), misconceptions about the framework persist. These assumptions primarily fall within four areas: that RE-AIM is simply for evaluation, the framework privileges quantitative over qualitative data, all dimensions must be weighted the same, and that the Maintenance phase encompasses only 6 months (22). Recently published studies have clarified that when appropriate (22–25), RE-AIM can incorporate qualitative and mixed methods designs, is not restricted to evaluation, and can be applied during dissemination, adoption, planning, and implementation (23). In addition, the framework has been expanded to include greater emphasis on sustainability (e.g., implementation for more than 1 year, integrating internal and external factors influencing implementation success) and health equity (21). For practitioners and researchers, applying RE-AIM during different phases of the project, while also maintaining a health equity and sustainability lens, better supports a multi-level approach that addresses the evolving needs related to capacity, as well as barriers and facilitators that many programs, organizations, and minoritized communities face. For Indigenous communities, the inclusion of a health equity and sustainability lens is particularly relevant for SUD treatment given health disparities in substance use related health outcomes, funding and capacity difficulties faced by Tribal health organizations.

The purpose of this scoping review is to inform future SUD evidence-based implementation research among Indigenous communities. A scoping review was identified by the study team as the appropriate approach because of the nascent nature of the state of the field. We sought to assess the existing evidence, clarify key concepts, and identify potential next steps to advance the science (26). While to our knowledge there are few completed published implementation trials for SUD interventions among Indigenous communities, we sought to inform future research and support practice change by examining a broad range of implementation strategies and processes among the more general Indigenous SUD treatment literature. Additionally, we reviewed whether Indigenous frameworks and worldviews have been centered in this work. Our guiding research question was what implementation processes or strategies are used among Indigenous communities for the uptake of evidence-based substance use treatment. We characterize the relevant research using the RE-AIM domains, contributing to previous work that has applied the framework with a focus on implementation (24, 25).

2. Methods

2.1. Sources and search protocol

Literature searches and eligibility assessment occurred in June and July of 2021 using Preferred Reporting Items for Systematic reviews and Meta-Analysis guidelines [PRISMA-Equity; search activities were completed by author MR (27)]. The search strategy was conducted within PsycINFO and PubMed/Medline databases. The second step in the search protocol involved all combinations of our population and content keywords. Population keywords included words used to describe and identify Indigenous populations. There were more than 30 content keywords including reference to dissemination and implementation frameworks and approaches. Combinations of each population term and all content keywords were administered via advanced search options with selections made for peer-reviewed articles written in English.

Search keywords were either combined with "AND" or in one plain search phrase (e.g., American Indian and Alaska Native; Māori "AND" dissemination and implementation science; implementation strategies; Explore, Preparation, Implementation and Sustainment; health equity implementation framework; Indigenous Implementation Framework). For a complete list, please refer to the Supplementary materials. The third step in the research strategy was to take the references identified by all combinations of search terms from each database extraction and save them as Research Information Systems (RIS) text files that were then uploaded into COVIDENCE, a systematic review management software (28). Duplicates were identified and removed within the software system.

2.2. Study selection

KH (descendent of the Eastern Shoshone Tribe/White, mixed European ancestry), MR (citizen of the Haliwa-Saponi Tribe), and KB (descendant of Filipino ancestry and immigrants) reviewed research articles for eligibility in COVIDENCE based on the title and the abstract. Study eligibility criteria was determined by the research team using knowledge of existing literature (e.g., anticipated state of the science), consideration of limitations of the research team (e.g., language capacity), and specific interest in SUD treatment implementation (29). Inclusion criteria were: (a) peer-reviewed, (b) written in English, (c) conducted with an Indigenous, adult (18 and older) population, (d) comprised of aims and outcomes related to substance misuse; (e) delivering an evidence-based intervention or practice based upon Western scientific criteria (e.g., efficacy or effectiveness randomized controlled trials), and (f) described one or more implementation processes or strategies. Exclusion criteria were: (a) studies completed entirely with non-Indigenous populations, (b) interventions intended for youth (younger than 18 years-old), (c) research that did not include outcomes related to substance use, and (d) research that did not discuss an evidence-based intervention (e.g., efficacy or effectiveness randomized controlled trials) for substance use. There was no exclusion based on publication date.

2.3. Data extraction

For articles which met initial eligibility criteria based on title and abstract, a full text review was conducted by KH and MR. Data extraction was completed by KH, MN (Turtle Mountain Band of Chippewa Indians), MR, KB, and MB (Spokane Tribe of Indians). Articles were initially reviewed independently and then underwent a second review by an alternate author to ensure validity and reliability of the extraction. None of the reviewers were individuals who had authored one of the articles. Consensus was determined and resolved by KH, MR, KB and MB. Final data review was carried out by KH and MR.

2.4. RE-AIM domain application

Once the final sample of articles was confirmed, the adapted RE-AIM framework was utilized for coding by two independent coders. While none of the studies were explicit D&I outcome studies, we coded the articles based upon what applied to the RE-AIM framework. This included articles that were clinical trials, methods papers, qualitative research, and case studies. Operational definitions based on the RE-AIM framework were identified and developed by the first author along with the review protocol (see Supplementary materials adapted from D'Lima et al. (30) and Shelton et al. (21)). The health equity and long-term sustainability extension codes were embedded within each existing domain. Codes were used to complete the data extraction within COVIDENCE, with final consensus completed in Excel.

For Reach, two questions were coded around the intended audience and who participated, as well as ways to better reach and engage the intended audience. Effectiveness was coded with three items related to whether the intervention was effective, if there had been any unintended consequences, and if intervention effectiveness was assessed over time. Adoption was assessed by three questions at the staff and setting level. This included application of the intervention and by whom, which staff were invited to participate versus excluded, and how staff were supported in delivering the intervention or could have been better supported. Implementation was assessed by six items ranging from efforts to culturally adapt the intervention, fidelity and delivery of the intervention, and costs. Maintenance was reviewed through the lens of sustainability of the intervention beyond 1 year and if multi-level contextual determinants were discussed that might impact sustainment.

3. Results

3.1. Description of articles

The initial search produced 44,301 articles. After the removal of 19,379 duplicates, an additional 24,689 were deemed ineligible through title and abstract screening and 234 articles underwent full-text review. After the full text-review, 20 articles were identified for inclusion (see Figure 1). Just over one-third were methods or protocol papers and about half of the studies included a randomization component. Of the studies identified, 65% included AI/AN adults, 20% Aboriginal Australian adults, 10% First Nations adults, and 5% Native Hawaiian adults. The evidence-based SUD programs being implemented (sometimes in combination) included: nicotine replacement therapy ($n = 8$), motivational interviewing ($n = 5$), community reinforcement approach ($n = 4$), medications for opioid use disorder ($n = 3$), contingency management ($n = 3$), community reinforcement approach and family training ($n = 2$), and recovery housing ($n = 1$). Primary substance use outcomes included smoking cessation ($n = 7$), alcohol use ($n = 6$), more than one substance or not specified ($n = 5$), and opioid use ($n = 3$). All the interventions focused on the individual, with a little over half also having a family, community, or organizational component ($n = 12$). Only two of the studies intentionally included implementation strategies and existing dissemination and implementation frameworks, but this was integrated retrospectively (Table 1).

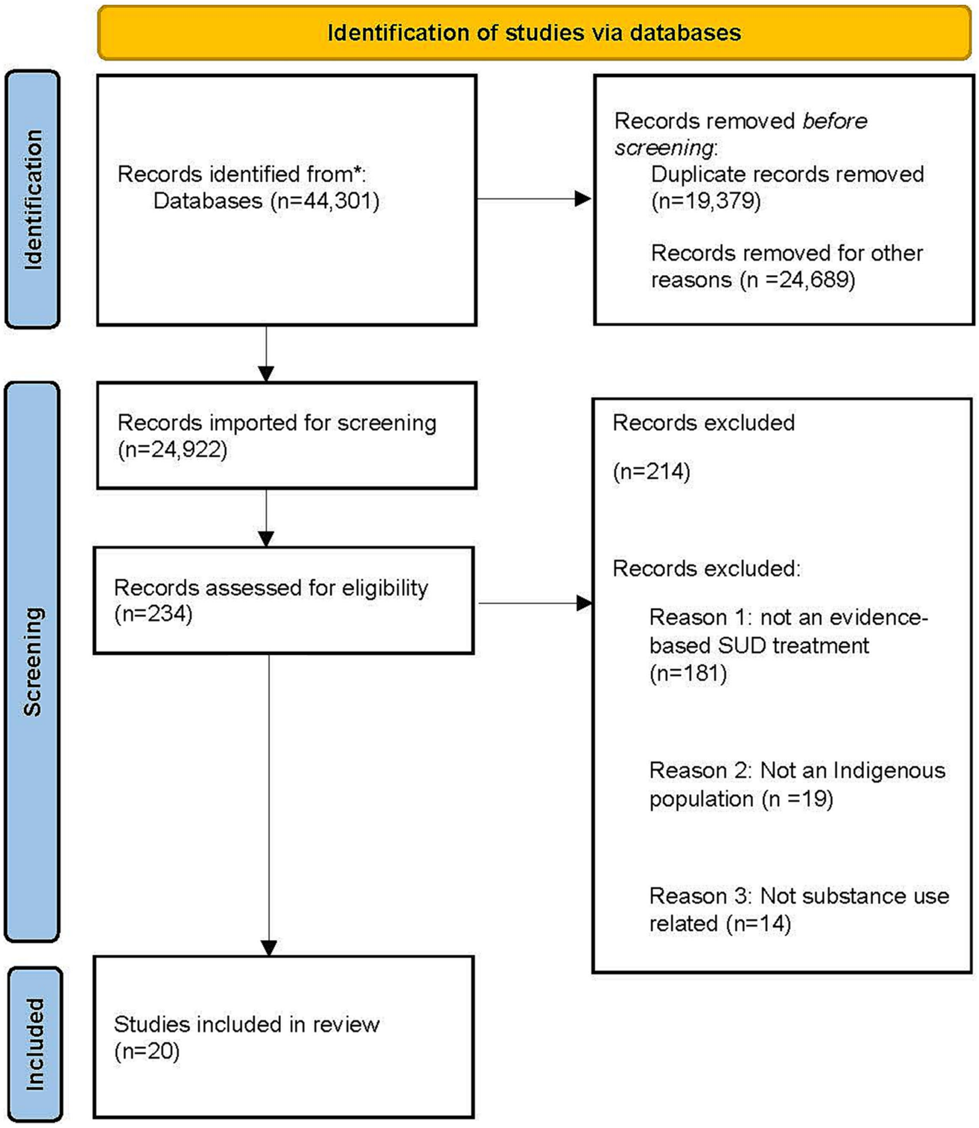

FIGURE 1
PRISMA flow diagram. Adapted from Page et al. (31).

3.2. Characterizing studies through the RE-AIM domains

Reach, Adoption, and Implementation were discussed by 60% of the studies. This was followed by Effectiveness (50% $n = 10$), with five studies discussing Maintenance/sustainability (25%, Table 2). Frequency and distribution of each of the domains coded in the literature are also presented (Figure 2). Below we provide descriptions of how the RE-AIM domain was characterized and include examples from the literature that highlight each domain. Definitions of domains and abbreviated summaries of examples can be found in Table 3.

3.3. Reach

Sixty percent of the studies reported Reach [$n = 12$; (33, 35–38, 40, 42, 45–48, 50)] with information mainly focused on inclusion and exclusion criteria for participants along with recruitment strategies (33, 35, 36, 40, 42, 45, 46, 48, 50). For example, Campbell and colleagues (35) noted that they recruited 58.8% ($N = 40$) out of the 68 clients that were eligible to participate in their study. In another study, the involvement of treatment providers from the community was also critical to Reach, with one study describing their recruitment efforts as including clinician referrals, radio newspaper, and digital (i.e., Facebook) ads, outreach at community events, and word-of-mouth (33). Reach was expanded in a pharmacist-led initiative through the Indian Health Service to increase access to medications for opioid use disorder [MOUD; (33)]. This was achieved by increasing the number of individuals initiated onto medications, poisoning awareness and procurement of naloxone kits that were distributed nationally. Other strategies around Reach included naloxone/drug take-back initiatives and engagement through culturally responsive educational tools (37).

Within the Reach domain, though not always referencing CBPR specifically, articles highlighted the importance of community buy-in and engaging collaborators on the project that were a part of the community and had knowledge of cultural protocols [e.g., how to appropriately greet an Elder; (50)]. Another common implementation strategy across studies was convening an advisory board (e.g., Community Advisory Board) or collaborative board to provide oversight on research activities, assist with recruitment and facilitate connections across organizations in the community. In one study, for example, the Community Advisory Board assisted in developing the culturally appropriate intervention title and how to brand the project in each community, identified and adapted measures, and guided focus groups to increase cultural acceptability of the intervention (40).

Trust was also emphasized in a few studies as another reason Reach was successful (36, 37). A trusting relationship between the participants and the research team was key. In these studies, it was argued that participants reported that they participated because they knew that the research team would manage their data with care, which speaks to data sovereignty and privacy. A unique aspect of Reach and trust was illustrated by a study completed in Australia (45). The authors described how "jealousy" played a significant role in recruitment. As reported by the authors, the spouses of participants were concerned that participation in research would provide opportunities for romantic infidelity. Research staff responded by developing strategies to make the intervention more welcoming and inclusive by providing opportunities to non-participating spouses. While specific to this community, this study demonstrates the importance of an engaged and flexible staff that can identify barriers and then develop solutions.

The need for flexibility in participant engagement strategies, and the ability to pivot based on community-generated solutions for health equity in substance use accessibility and capacity was, in fact, described across studies. For example, Orr and colleagues (47) initially intended to recruit AI/AN college students for their tobacco cessation program. However, recruitment efforts led to only 9 college students enrolling. The researchers then turned their recruitment strategy to Quitlines in states that had higher AI/AN rural populations (e.g., Alaska, New Mexico) and were able to complete their recruitment efforts. Another study identified recruitment challenges due to staff not having the time to recruit at the site, a short recruitment window, and eligibility criteria that were too stringent (38). One study team discussed needing to make changes to recruitment strategies when participants were screened eligible but declined to participate. Of note, the reasons for declining varied but were both practical and directly related to the research fit in the community. For example, participants reported not being comfortable with randomization, lack of interest in research, a preference for another type of treatment, and study location that was too far away (50).

3.4. Effectiveness

Out of the 20 studies reviewed, ten (50%) of them reported outcomes related to intervention effectiveness (1, 34, 36, 38, 40, 41, 45, 48–50). There were several studies where the primary outcome was tobacco cessation [$n = 7$; (32, 36, 38, 45, 47–49)]. In a tobacco cessation program for pregnant Aboriginal women, self-reported 12-week 7-day point-prevalence abstinence was 13.6% (38). In another smoking cessation study integrated across two geographically and culturally distinct Aboriginal primary care settings, no statistically significant difference was observed in quit rates, but the study was also statistically underpowered (45). Although the quit rate varied, up to 43% of patients were tobacco free for 3 months across a healthcare system with five locations serving Native Hawaiian people (49).

Interventions addressing alcohol and other substance use also resulted in favorable outcomes. In a study assessing the effectiveness of Motivational Interviewing and Community Reinforcement Approach among a Tribal community in the Southwest United States, at 8 months, percent days abstinent had increased for both alcohol and cannabis, as well as other substances [excluding tobacco; (47)]. In the primary outcome clinical trial of Motivational Interviewing and Community Reinforcement Approach, participants in both the treatment, and treatment as usual (TAU), improved in percent days abstinent and substance use severity [MICRA PDA =72.63%, TAU = 73.62%; (1)]. At the three-month follow up in another study, Aboriginal participants in the Community Reinforcement Approach intervention had significantly reduced their alcohol use. There were also reductions in frequency of days of alcohol consumed and number of drinks consumed per drinking day based on pre- and post- self-report (34).

3.5. Adoption

Sixty percent of the studies described processes and outcomes related to Adoption [$n = 12$; (1, 32, 36–38, 40–42, 44, 47–49)]. At the

TABLE 1 Frameworks and sample cultural adaptations.

Author	Indigenous community	Evidence based treatment culturally re-centered	Community engaged or CBPR	Western adaptation/ implementation frameworks	Indigenous frameworks	Sample cultural re-centering of evidence based treatment
Bar-Zeev (32)	Aboriginal Pregnant Women, Australia	No	No	No	No	• Stakeholder and Consumer Aboriginal Advisory Panel • Cultural liaison
Burduli (33)	AI Adults, United States	Yes	Yes	No	No	• Culturally appropriate rewards • Elders and community members led the intervention
Calabria (34)	Aboriginal and Torres Strait Islander Adults, Australia	~	~	No	No	~
Campbell (35)	Urban AI/AN Adults, United States	No	Yes	No	No	No
Daley (36)	Urban AI/AN Adults, United States	Yes	Yes	No	No	• Ceremonies, traditional and cultural activities related to tobacco, coping and stress • Culturally appropriate rewards
Duvivier (37)	American Indian Adults, United States	Yes	Yes	No	No	• Pharmacists developed culturally responsive materials • Efforts to reduced stigma and build individual/ community trust
Gould (38)	Aboriginal Pregnant Women, Australia	Yes	Yes	No	No	~
Hanson (39)	AI/AN Adult Women, United States	Yes	Yes	No	No	• Community members led the intervention • Community Advisory Board
Hirchak (40)	AI Adults, United States	Yes	Yes	• Interactive Systems Framework • ADAPT-ITT	No	• Focus groups • Speakers of the Native language led sessions
Hirchak (41)	AI/AN Adults, United States	Yes	Yes	• Quality Implementation Checklist	No	• Community Advisory Board • Community members led the intervention
Jason (42)	Snohomish Tribe, United States	Yes	Yes	No	No	• Residents tailored processes to meet their needs (e.g., talking circles, Indigenous artwork)
Kiepek (43)	First Nations, Canada	Yes	Yes	No	No	• Foods and medicines for a traditional diet provided • Elders-in-residence to lead cultural activities and ceremonies

(Continued)

TABLE 1 (Continued)

Author	Indigenous community	Evidence based treatment culturally re-centered	Community engaged or CBPR	Western adaptation/implementation frameworks	Indigenous frameworks	Sample cultural re-centering of evidence based treatment
Landry (44)	Elsipogtog First Nation (MiÍûåkmaq First Nations Band), Canada	Yes	Yes	No	No	~
Marley (45)	Miriuwung and other Aboriginal and Torres Strait Islander Adults, Australia	~	Yes (PAR[a])	No	No	• Community members led the intervention • Historical and present factors impacting communities specifically described
McDonell (46)	AI/AN Adults, United States	Yes	Yes	No	No	• Focus groups • Culturally appropriate rewards • Community members led the intervention
Orr (47)	Rural AI/AN Adults, United States	Yes	Yes	No	No	• Focus groups • Materials adapted (e.g., culturally relevant text messages)
Patten (48)	Yup'ik, Cup'ik or Athabascan, United States	Yes	Yes	• Cultural Variance Framework • Surface and Deep Structure Framework	No	• Counseling included an emphasis on positive cultural and community activities to cope with tobacco withdrawal • Community members led the intervention • Integrated traditional cultural practices, values and teachings (e.g., Yup'ik ways of being healthy)
Santos (49)	Native Hawaiian Adults, United States	Yes	Yes	No	No	• Materials were culturally responsive and included values of family, solidarity, and motivators for behavior change • Well respected community member and champions integrated for cultural relevance
Venner (50)	AI Adults, United States	Yes	Yes	No	No	~
Venner (1)	AI Adults, United States	Yes	Yes	No	No	• Speakers of the Native language led sessions • Traditional introductions • Culturally adapted and validated measures

~Unclear. [a]Participatory Action Research.

TABLE 2 Summary of RE-AIM domains included in each study.

Author	EBP	Study design	Reach	Effectiveness	Adoption	Implementation	Maintenance
Bar-Zeev (32)	NRT	Methods/Protocol Paper (SWRCT)[a]	No	No	Yes	No	No
Burduli (33)	CM	Methods/Protocol Paper (RCT)[b]	Yes	No	No	Yes	No
Calabria (34)	CRAFT	Quasi-Experimental	No	Yes	No	Yes	No
Campbell (35)	TES (CRA)	Mixed-Methods	Yes	No	No	Yes	No
Daley (36)	NRT/MI	Single Arm Clinical Trial	Yes	Yes	Yes	No	No
Duvivier (37)	MOUD	Descriptive	Yes	No	Yes	Yes	No
Gould (38)	NRT	SWRCT[a]	Yes	Yes	Yes	Yes	No
Hanson (39)	MI/CDC CHOICES	Methods/Protocol Paper (RCT)[b]	No	No	No	No	Yes
Hirchak (40)	MI/CRA/CRAFT	RCT (Case Study)[b]	Yes	Yes	Yes	Yes	Yes
Hirchak (41)	CM	RCT (Case Study)[b]	No	Yes	Yes	Yes	No
Jason (42)	Oxford House	Case Study	Yes	No	Yes	Yes	No
Kiepek (43)	MOUD	Case Study	No	No	No	Yes	No
Landry (44)	MOUD	Qualitative	No	No	Yes	No	Yes
Marley (45)	NRT	Qualitative	Yes	Yes	No	No	No
McDonell (46)	CM	Methods/Protocol Paper (RCT)[b]	Yes	No	No	Yes	No
Orr (47)	NRT/STOMP	Methods/Protocol Paper (RCT)[b]	Yes	No	Yes	Yes	No
Patten (48)	NRT	Methods/Protocol Paper (CRCT)[c]	Yes	Yes	Yes	No	No
Santos (49)	NRT	Case Study	No	Yes	Yes	Yes	Yes
Venner (50)	MI/CRA	Quasi-Experimental	Yes	Yes	No	No	No
Venner (1)	MI/CRA	RCT[b]	No	Yes	Yes	Yes	No

Adapted from D'Lima et al. (30). [a] Stepped wedge randomized controlled trial.

[b] Randomized controlled trial.

[c] Cluster randomized controlled trial.

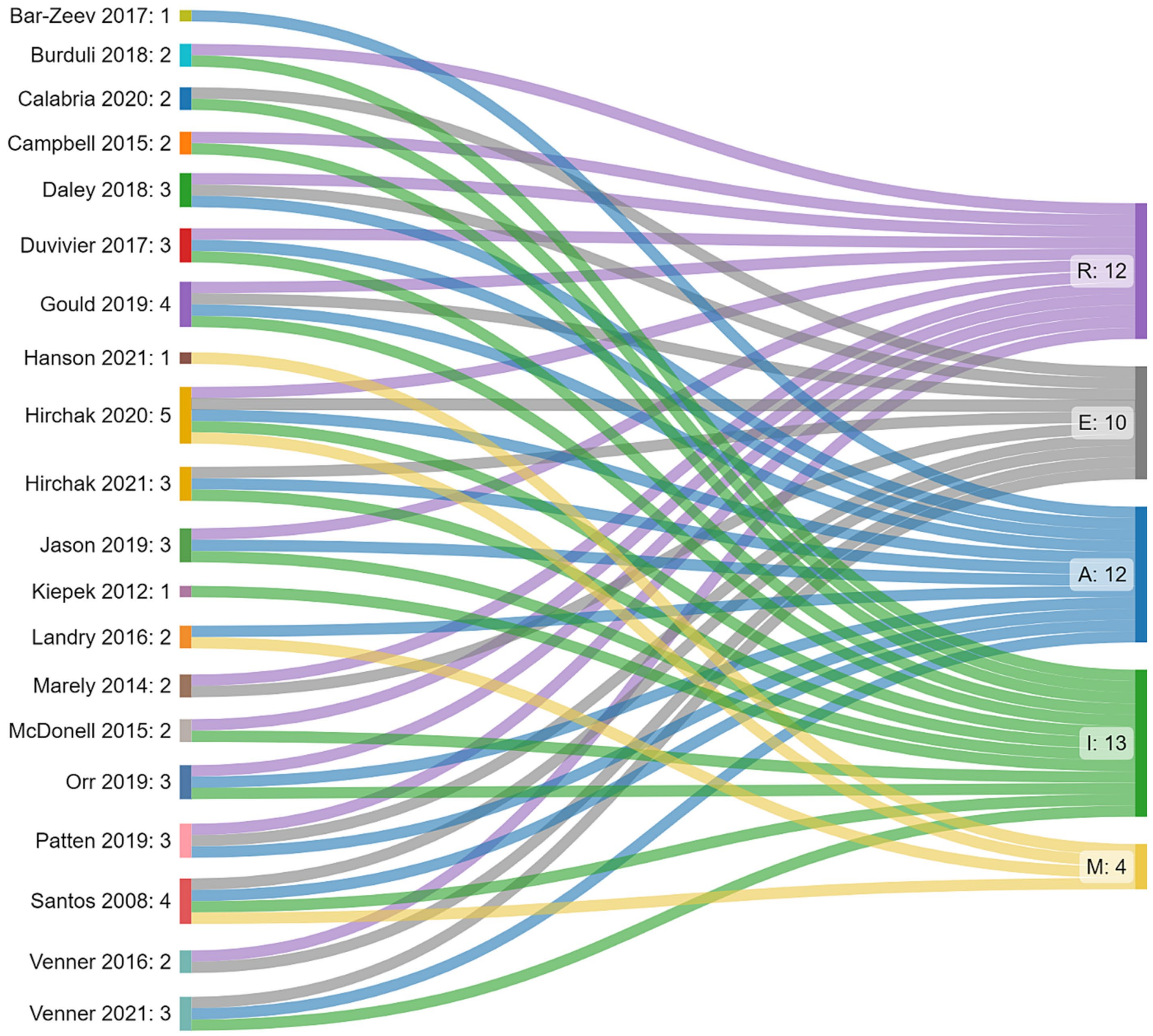

FIGURE 2
Distribution and frequency of RE-AIM domains assessed and discussed within each study.

staff level, authors highlighted strategies, for and barriers to, Adoption. Including enhancements to Adoption focused on the staff members themselves. While some studies had specific inclusion and exclusion criteria for staff (32, 38, 48), Indigenous staff and level of staff buy-in were highlighted as the key to increasing Adoption (37, 41, 44). The duration of the intervention also impacted Adoption. While shorter interventions are generally considered easier to Adopt, some of the studies highlighted that shorter project duration among Indigenous communities actually inhibited long-term Adoption and more time was needed to build relationships (36, 38, 40). Recommendations to increase Adoption included hiring qualified study staff who were trusted community members cross-trained to facilitate coverage (38). Also, the quality of the Tribal-organization and university partnership and the need for relationship-building at all levels was of noted importance, for managing potential distrust of research by the community, as well as to facilitate approval processes from multiple entities (1, 33, 41, 46).

At the organizational level, Adoption was discussed in the context of recruitment success, with higher recruitment translating to better adoption (36, 41, 45). One study focused on tobacco cessation located at a healthcare system with different locations across the Hawaiian Islands. Differences in adoption by site were explained by barriers such as lower level of administrative or clinical support, staff turnover, and lower organizational readiness to adopt a new protocol or service (49). Although the authors initially assumed locations would be similar because they were within the same health system, each of the five locations evidenced variation in community needs, representation on the governing board, use of their medical record system, or attitudes toward medications for tobacco cessation (49).

3.6. Implementation

Facilitators of Implementation were described at both the internal (i.e., organizational) and external levels, with 60% of studies touching upon this [*n* = 12; (1, 33–35, 37, 38, 40–43, 46, 47, 49)]. Organizations face an array of internal challenges (e.g., shifting priorities and

TABLE 3 RE-AIM domains and sample process.

Domain	Definition
Reach	% of individuals excluded, characteristics of those excluded, number or proportion of representativeness of individuals who participated, how to better engage the intended audience
Examples from the literature	
- 142 participants were screened for a alcohol and illicit drug contingency management intervention, but only 114 met inclusion criteria [i.e., self-reported AI adult 18+; seeking alcohol misuse or dependence and drug misuse or dependence treatment; a Diagnostic and Statistical Manual, fourth edition diagnosis of current alcohol dependence; current drug misuse defined as drug use in the last 30 days; ability to provide informed consent, read and speak English; Burduli et al. (33)] - The researchers described having a Community Advisory Board to guide the intervention and completed focus groups to culturally adapt and gain community feedback on study materials and processes to better engage the intended audience (46)	
Effectiveness	Was the intervention effective, for whom was it effective
Examples from the literature	
- In a tobacco cessation program specifically adapted for American Indian adults, after accounting for attrition, the overall quit rate at the end of the 3-month intervention period was about 41% and at 6 months post-baseline the final quit rate was approximately 22% [$p = 0.002$; Daley et al. (36)] -AI/AN participants who received a chance to draw for prizes when they submitted an alcohol-negative urine sample were 70% more likely to be alcohol abstinent compared to those in the control group (41)	
Adoption	By whom and where was the Evidence Based Treatment implemented, who was invited or excluded from implementation, intervention consistently applied (fidelity)
Examples from the literature	
- A tobacco cessation program among pregnant women in Alaska had the intention to hire a "Native Sister" from each participating village, but that this had not been feasible. One "Native Sister" was hired out of the Bethel, Alaska area to deliver the intervention at all the villages that did not already have one. Ultimately, the Bethel area had the best recruitment numbers because the prenatal visits were also located there (48) -MICRA had many supports related to counselor fidelity and intervention delivery. Experts in MI and CRA delivered the training and on-going technical assistance. Counselors were then also able to deliver the intervention in their Native language to further strengthen intervention adoption (1, 50)	
Implementation	Consistency of implementation strategies, costs of delivery of Evidence Based Treatment discussed, implementation strategies adapted/culturally adapted
Examples from the literature	
- In a self-paced educational module curriculum of TES, implementation strategies were consistent and included a multi-component approach of ongoing consultations from AI/AN clinical administrators, staff, and researchers on the development of the proposal, training, recruitment, and assessments. Collaborators made contributions to the interpretation of the data as well as the development of dissemination materials (35) - In a text messaging for tobacco cessation intervention, eight focus groups were conducted at four Tribal colleges in Montana, to support the cultural adaptation of the text messages. A total of 55 AI/AN Tribal college students who were currently smoking or were previous smokers participated across the 8 focus groups. The original messages designed for Māori young adults, and were reviewed by 2 AI research team members, all Māori specific messages and references were removed and those that were consistent to AI/AN cultures were retained for focus group review. This process yielded 104 culturally adapted text messages, of those 30 were newly developed by the focus groups (47)	
Maintenance	Implementation strategies discussed to sustain the program long-term or beyond 1 year after implementation
Examples from the literature	
- An intervention that addresses risk of alcohol-exposed pregnancies, discussed their plans for an economic evaluation that would be completed at the end of the 5-year study to assess the cost-savings related to the intervention and among participants in reducing alcohol related harms, and to assist future work in program sustainability (39)	

Adapted from Glasgow et al. (19) and Shelton et al. (21).

reduced funding) along with external factors that impact what types of interventions may be supported or can be reimbursed. Authors discussed strategies related to the need for capacity building within and outside of the organization, having champions at multiple levels within (e.g., direct service, supervisor levels) and outside of the organization [e.g., Tribal leadership; (1, 40, 41, 49, 50)] and holding information meetings about the intervention for service providers outside of the organization (i.e., community members and leaders). Within the organization, it was recommended that there be on-going face-to-face communication between research staff and site staff and sharing of information and lessons from Implementation including monthly ongoing technical assistance/facilitation (1, 35, 38, 40, 41, 45).

Modifications to the intervention to enhance acceptability, integration, and implementation included cultural adaptations tailored to each setting. Making cultural adaptations to evidence-based substance use disorder treatment was described as a more holistic approach that sits within the cultural framework of the partnering community. This can take the form of integrating appropriate representation (e.g., art, study staff from the community) knowledge, and Indigenous worldviews into the evidence-based treatment which may also increase engagement and adoption (33, 42, 43, 50) (Table 1). Seventy-five percent of the studies ($n = 15$) mentioned culturally adapting the intervention, but only 15.0% ($n = 3$) described a specific framework guiding the adaptations made. While 90% of the studies mentioned a process of community engagement, only one-third of the studies explicitly stated the use of CBPR. Of those, there was no description or definition of how CBPR was being interpreted. None of the studies explicitly identified an Indigenous framework used as a part of the program implementation or intervention adaptation.

Examples of the commonly described cultural adaptation processes and methods included: focus groups, having members of the community lead the intervention, and culturally adapting materials (e.g., including Indigenous languages or pictures that represent the community). One study was initiated when a Tribal elected official contacted the researchers following the development of a community sober living house to assist with evaluation of feasibility, acceptability, and effectiveness (42). The study was unique in that the residents culturally adapted the model and the implementation processes in real time. The communal nature of the sober living house aligned with Tribal culture, and participants were able to include cultural activities (e.g., sweat lodge, talking circles with the inclusion of sacred objects like feathers, and drumming circles). Further modifications included the house being run by the residents who decided all activities, house rules and procedures. The house was also inclusive of non-Tribal members, so that anyone would have a home if they needed one, reflecting the Tribe's values of providing social support and resources to those most in need (42).

Although funding is a core aspect of Implementation, it was not frequently discussed. Many of the studies did not directly specify the costs of delivering the evidence-based treatment, and, when funding was discussed, studies highlighted that it had enhanced Implementation efforts. For example, one study described how the total budget was dispersed across partnering communities during the duration of the research project and that this had been one of the major strengths of the collaboration [e.g., $1.8 million budget over 6 years for contingency management; (48)]. The authors of a multi-component intervention for smoking cessation among pregnant Aboriginal women did not list the entire cost of the intervention and delivery but noted that each site received $6,000 to support delivery of the intervention and staff contribution (38). In another study assessing recovery housing, Oxford Houses were argued to be self-sustaining and a lower cost option for post-treatment or recovery maintenance because each resident contributed to the cost of running the home. Professional staff were not employed (Oxford houses are operated by the residents) and residents' rent typically amounted to about $100 per week (42).

3.7. Maintenance/sustainability

Maintenance, or Sustainability, was the least discussed across studies with 20% of the articles including some discussion around this domain [$n = 4$; (39, 40, 44, 49)]. Multi-level factors related to sustainment suggested by a few authors included societal and policy forces (e.g., racism, historical trauma, poverty, and discrimination) but these components were not directly assessed within the studies (1, 41, 44, 45, 50). The studies that did highlight Sustainability also speculated on activities that may support or hinder the long-term delivery of the intervention. Authors also described future plans for program sustainment (39).

One qualitative study that examined a program delivering medications for opioid use disorder emphasized the need for community education and buy-in around harm reduction strategies to effectively sustain program activities. The need for education and training related to these medications was especially important to inform family members about how the medications worked to support treatment and how they were different from other types of opioids and illicit drugs (e.g., some family members were unsupportive of methadone maintenance treatment). Among those interviewed, there was a belief that the long-term impact of the medication for opioid use disorder program had been to increase cleanliness/safety (e.g., reduction in discarded needles) and reduce crime (e.g., burglary, vandalism) in the community. The program had already been on-going for 5 years by the time of publication, so institutionalization of the program was achieved, but the specifics of how this was accomplished were not discussed in detail (44).

4. Discussion

In this scoping review, we assessed implementation strategies and processes employed by researchers and Indigenous communities to enhance the uptake and delivery of evidence-based substance use disorder treatments, applying a widely used public health framework, RE-AIM: Reach, Effectiveness, Adoption, Implementation, and Maintenance. Across eligible studies, the most often discussed processes were Implementation, Reach, and Adoption followed by Effectiveness and Maintenance/sustainability. Of the 10 studies that described Effectiveness, all but one intervention led to statistically significant improved substance use outcomes. The present study highlights the need for additional implementation research with Indigenous communities to better understand implementation strategies and outcomes that will support the integration of evidence-based practices to address substance use disorders.

Many of the studies referenced the importance of a community engaged or CBPR approach but few defined or detailed what this meant or how the CBPR methods were applied. Several articles also mentioned having a Community Advisory Board or Research Review Board, which are key principles of CBPR, but how exactly this was developed and deployed remained unclear (for examples see 51, 52). Additionally, most of the studies did not outline the use of specific frameworks, or dissemination and implementation theories and models. Future research should be model-driven and systematically describe the methods used with Indigenous communities to support this emerging knowledge base. Strategies and approaches in D&I research must also be pursued thoughtfully so that an emphasis or privileging of Western worldviews does not hinder the use or development of Indigenous frameworks to braid collaborations [e.g., the Indigenist-Stress Coping Model (53), the Two-Eyed Seeing approach (54) or the He Pikinga Waiora Implementation Framework; (55)]. As noted by McCuistian and Colleagues (56), a need exists for a well-specified model and plan for community engagement and CBPR approaches with minoritized communities. This is likely to support reach and engagement within the community and ensure that the intervention is desired and integrated appropriately for and with the community. Prior evidence also suggests that this may lead to improved intervention effectiveness (57).

A primary challenge to sustainability from the studies assessed in this review was lack of or insufficient funding. Lack of program sustainment creates further issues around trust between researchers and communities (e.g., when programs or research projects are visible in the community and then suddenly disappear when the funding ends). Challenges in program sustainment also inhibit the ability to truly impact health equity (58). Additionally, we found that despite the large sample of articles identified through the initial search, there were few that described implementation research specifically for SUD treatments with Indigenous communities. More work is needed that is culturally congruent or grounded (and specifies the methods for conceptual grounding) to increase both the literature on implementation strategies as well as the literature on evidence-based treatments.

Another component of our analysis was related to health equity and the integration of Indigenous frameworks. Findings indicated that while some of the articles intended to positively impact substance use outcomes, many did not discuss the multi-level determinants necessary for interventions to work [e.g., the social determinants of health or the importance of overall improved quality of life; (57)]. A few authors suggested that societal and policy forces (e.g., racism, historical trauma, poverty, and discrimination) should be considered (1, 41, 44, 45, 50), but it was not explicit as to how the interventions might seek to address these factors. It is important that future SUD treatment research is placed within the larger colonial-settler context so that Indigenous people are not further stigmatized or blamed for the consequences of colonization and to ensure that meaningful systems change occurs.

There were notable strengths of this review and several limitations. Reviewers may not have extracted all relevant data for the specified domain due to lack of clarity or explicit description within the article. However, our analysis of these data included independent assessment from at least two reviewers and consensus building across reviewers to increase scientific rigor. Second, this research was focused on evidence-based treatments and used a Western framework to structure the review. This may have reduced our ability to examine community-centered approaches. While our final sample included 20 articles, the initial unduplicated count was close to 25,000. We believe this was due to the extensive population and content search terms (35+; see Supplementary materials). Future studies may want to include fewer keywords and more focused search terms to summarize any recent literature accordingly, as this research area continues to grow. Although we used the PRISMA-Equity Extension checklist, future research might consider the development of an *a priori* review protocol. Despite this, a strength of this review was the extensive assessment of implementation strategies and processes used by researchers and communities to effectively increase the health and well-being of Indigenous people.

5. Conclusion

Among Indigenous communities, the field of D&I is expanding. Research partnerships have led to the implementation of culturally adapted evidence-based treatments for substance use disorders that are more in alignment with culture and holistic conceptualizations of health and well-being. Given the continued growth in the field, we provide 6 broad recommendations from our findings to move the science forward. (1) The need for more SUD interventions among Indigenous communities that also include ways to address or reduce the disproportionate health inequities related to the social determinants of health that contribute to substance use. (2) While some studies addressed factors related to Implementation, Reach, Adoption and Effectiveness, very few addressed Maintenance/sustainability, which could reflect insufficient or lack of long-term funding for research as well as insufficient resources more broadly within Indigenous communities. More community generated and sustained funding is required. (3) Additional thoughtfulness and application of the extended RE-AIM to include a greater emphasis on equity and sustainability in D&I and treatment research among Indigenous communities was lacking. Future research could consider opportunities to expand RE-AIM to include adaptations to the domains that support the use of the framework specifically within Indigenous communities. (4) The field could benefit from a comprehensive discussion on applying Indigenous frameworks and worldviews within the context of Western D&I frameworks, models or theories. (5) CBPR plays an important role in D&I with Indigenous communities and our findings highlighted the need for more detailed descriptions of how this work is defined, employed, and assessed within partnerships. (6) Local research capacity building and front-loading engagement activities are needed to help with Reach, Adoption, and sustainment. Successful research partnerships require attention to trust and long-term relationship building. As models of sustainability evolve in the broader D&I community to reflect the challenges in the fit between interventions and local context, we hope that there will be more application of these methods and techniques to ensure interventions serve the needs of Indigenous communities.

Author contributions

KH: Conceptualization, Data curation, Formal analysis, Funding acquisition, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing, Investigation. OO: Conceptualization, Methodology, Writing – review & editing. MN: Conceptualization, Formal analysis, Methodology, Writing – review & editing. MR: Data curation, Formal analysis, Methodology, Software, Writing – original draft, Writing – review & editing. KB: Data curation, Formal analysis, Methodology, Software, Writing – review & editing. MB: Data curation, Formal analysis, Methodology, Writing – review & editing. JH: Conceptualization, Writing – review & editing. AH-V: Conceptualization, Writing – review & editing. AV: Conceptualization, Writing – review & editing. CP: Conceptualization, Writing – review & editing. KO: Conceptualization, Writing – review & editing. PB: Conceptualization, Writing – review & editing. KW: Writing – review & editing. LB: Writing – review & editing. MM: Writing – review & editing. KV: Conceptualization, Writing – review & editing, Data curation, Methodology. AC: Conceptualization, Writing – review & editing, Data curation, Methodology.

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Conflict of interest

KO was employed at KEAO Consulting LLC. KV currently has a conflict-of-interest management plan at the University of New Mexico.

The remaining authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2023.1265122/full#supplementary-material

References

1. Venner KL, Serier K, Sarafin R, Greenfield BL, Hirchak K, Smith JE, et al. Culturally tailored evidence-based substance use disorder treatments are efficacious with an American Indian southwest tribe: an open-label pilot-feasibility randomized controlled trial. *Addiction*. (2020) 116:949–60. doi: 10.1111/add.15191

2. Substance Abuse and Mental Health Services Administration. *Key substance use and mental health indicators in the United States: results from the 2020 national survey on drug use and health [internet]*. Rockville, MD: Center for Behavioral Health Statistics and Quality, Substance Abuse and Mental Health Services Administration (2021).

3. Greenfield BL, Venner KL. Review of substance use disorder treatment research in Indian country: future directions to strive toward health equity. *Am J Drug Alcohol Abuse*. (2012) 38:483–92. doi: 10.3109/00952990.2012.702170

4. Burlew AK, Copeland VC, Ahuama-Jonas C, Calsyn DA. Does cultural adaptation have a role in substance abuse treatment? *Soc Work Public Health*. (2013) 28:440–60. doi: 10.1080/19371918.2013.774811

5. Walter M, Carroll SR. Indigenous data sovereignty, governance and the link to indigenous policy In: M Walter, T Kukutai, SR Carroll and D Rodriguez-Lonebear, editors. *Indigenous data sovereignty and policy [internet]. 1st* ed. London: Routledge (2020)

6. McDonell MG, Skalisky J, Burduli E, Foote A, Granbois A, Smoker K, et al. The rewarding recovery study: a randomized controlled trial of incentives for alcohol and drug abstinence with a rural American Indian community. *Addiction*. (2021) 116:1569–79. doi: 10.1111/add.15349

7. McDonell MG, Hirchak KA, Herron J, Lyons AJ, Alcover KC, Shaw J, et al. Effect of incentives for alcohol abstinence in partnership with 3 American Indian and Alaska native communities: a randomized clinical trial. *JAMA Psychiatry*. (2021) 78:599–606. doi: 10.1001/jamapsychiatry.2020.4768

8. Blue Bird Jernigan V, D'Amico EJ, Keawe'aimoku Kaholokula J. Prevention research with indigenous communities to expedite dissemination and implementation efforts. *Prev Sci Off J Soc Prev Res*. (2020) 21:74–82. doi: 10.1007/s11121-018-0951-0

9. Holt CL, Chambers DA. Opportunities and challenges in conducting community-engaged dissemination/implementation research. *Transl Behav Med*. (2017) 7:389–92. doi: 10.1007/s13142-017-0520-2

10. Rae R, Belone L, Tafoya E, Yepa M, Cohoe-Belone B, Burbank I, et al. Family listening/circle program: the experience of community action projects to promote family and community wellness in three tribal communities in New Mexico. *Front Public Health*. (2023) 11:1091751. doi: 10.3389/fpubh.2023.1091751

11. Werito V, Belone L. Research from a Diné -centered perspective and the development of a community-based participatory research partnership. *Health Educ Behav*. (2021) 48:361–70. doi: 10.1177/10901981211011926

12. Walters KL, Johnson-Jennings M, Stroud S, Rasmus S, Charles B, John S, et al. Growing from our roots: strategies for developing culturally grounded health promotion interventions in American Indian, Alaska native, and native Hawaiian communities. *Prev Sci*. (2020) 21:54–64. doi: 10.1007/s11121-018-0952-z

13. Wallerstein N, Duran B. Community-based participatory research contributions to intervention research: the intersection of science and practice to improve health equity. *Am J Public Health*. (2010) 100:S40–6. doi: 10.2105/AJPH.2009.184036

14. Dickerson D, Baldwin JA, Belcourt A, Belone L, Gittelsohn J, Keawe'aimoku Kaholokula J, et al. Encompassing cultural contexts within scientific research methodologies in the development of health promotion interventions. *Prev Sci*. (2020) 21:33–42. doi: 10.1007/s11121-018-0926-1

15. Sacca L, Shegog R, Hernandez B, Peskin M, Rushing SC, Jessen C, et al. Barriers, frameworks, and mitigating strategies influencing the dissemination and implementation of health promotion interventions in indigenous communities: a scoping review. *Implement Sci*. (2022) 17:18. doi: 10.1186/s13012-022-01190-y

16. Belone L, Rae R, Hirchak KA, Cohoe-Belone B, Orosco A, Shendo K, et al. Dissemination of an American Indian culturally centered community-based participatory research family listening program: implications for global indigenous well-being. *Genealogy*. (2020) 4:99. doi: 10.3390/genealogy4040099

17. Nilsen P. Making sense of implementation theories, models and frameworks. *Implement Sci*. (2015) 10:53. doi: 10.1186/s13012-015-0242-0

18. Glasgow RE, Vogt TM, Boles SM. Evaluating the public health impact of health promotion interventions: the RE-AIM framework. *Am J Public Health*. (1999) 89:1322–7. doi: 10.2105/ajph.89.9.1322

19. Glasgow RE, Harden SM, Gaglio B, Rabin B, Smith ML, Porter GC, et al. RE-AIM planning and evaluation framework: adapting to new science and practice with a 20-year review. *Front Public Health*. (2019) 7:64. doi: 10.3389/fpubh.2019.00064

20. McCreight MS, Rabin BA, Glasgow RE, Ayele RA, Leonard CA, Gilmartin HM, et al. Using the practical, robust implementation and sustainability model (PRISM) to qualitatively assess multilevel contextual factors to help plan, implement, evaluate, and disseminate health services programs. *Transl Behav Med*. (2019) 9:1002–11. doi: 10.1093/tbm/ibz085

21. Shelton RC, Chambers DA, Glasgow RE. An extension of RE-AIM to enhance sustainability: addressing dynamic context and promoting health equity over time. *Front Public Health*. (2020) 8:134. doi: 10.3389/fpubh.2020.00134

22. RE-AIM [Internet]. Available at: https://re-aim.org/ (Accessed March 14, 2023).

23. Holtrop JS, Estabrooks PA, Gaglio B, Harden SM, Kessler RS, King DK, et al. Understanding and applying the RE-AIM framework: clarifications and resources. *J Clin Transl Sci*. (2021) 5:e126. doi: 10.1017/cts.2021.789

24. Glasgow RE, Battaglia C, McCreight M, Ayele RA, Rabin BA. Making implementation science more rapid: use of the RE-AIM framework for mid-course adaptations across five health services research projects in the veterans health administration. *Front Public Health*. (2020) 8:194. doi: 10.3389/fpubh.2020.00194

25. Heelan KA, Abbey BM, Bartee RT, Estabrooks PA, Malmkar A, Jacobs T, et al. Packaging of a pediatric weight management intervention and implementation blueprint for rural and Micropolitan communities: the Nebraska CORD 3.0 project. *Child Obes*. (2021) 17:S-62–9. doi: 10.1089/chi.2021.0171

26. Munn Z, Peters MDJ, Stern C, Tufanaru C, McArthur A, Aromataris E. Systematic review or scoping review? Guidance for authors when choosing between a systematic or scoping review approach. *BMC Med Res Methodol*. (2018) 18:143. doi: 10.1186/s12874-018-0611-x

27. Welch V, Petticrew M, Tugwell P, Moher D, O'Neill J, Waters E, et al. PRISMA-equity 2012 extension: reporting guidelines for systematic reviews with a focus on health equity. *PLoS Med*. (2012) 9:e1001333. doi: 10.1371/journal.pmed.1001333

28. Covidence [internet]. Melbourne, Australia: Veritas Health Innovation; (2023). Available at: www.covidence.org

29. Levac D, Colquhoun H, O'Brien KK. Scoping studies: advancing the methodology. *Implement Sci*. (2010) 5:69. doi: 10.1186/1748-5908-5-69

30. D'Lima D, Soukup T, Hull L. Evaluating the application of the RE-AIM planning and evaluation framework: an updated systematic review and exploration of pragmatic application. *Front Public Health*. (2022) 9:755738. doi: 10.3389/fpubh.2021.755738

31. Page MJ, McKenzie JE, Bossuyt PM, Boutron I, Hoffmann TC, Mulrow CD, et al. The PRISMA 2020 statement an update guideline for reporting systematic reviews. *BMJ*. (2021) 372:n71. doi: 10.1136/bmj.n71

32. Bar-Zeev Y, Bonevski B, Bovill M, Gruppetta M, Oldmeadow C, Palazzi K, et al. The indigenous counselling and nicotine (ICAN) QUIT in pregnancy pilot study protocol: a feasibility step-wedge cluster randomised trial to improve health providers' management of smoking during pregnancy. *BMJ Open*. (2017) 7:e016095. doi: 10.1136/bmjopen-2017-016095

33. Burduli E, Skalisky J, Hirchak K, Orr MF, Foote A, Granbois A, et al. Contingency management intervention targeting co-addiction of alcohol and drugs among American Indian adults: design, methodology, and baseline data. *Clin Trials*. (2018) 15:587–99. doi: 10.1177/1740774518796151

34. Calabria B, Shakeshaft AP, Clifford A, Stone C, Clare PJ, Allan J, et al. Reducing drug and alcohol use and improving well-being for indigenous and non-indigenous Australians using the community reinforcement approach: a feasibility and acceptability study. *Int J Psychol*. (2020) 55:88–95. doi: 10.1002/ijop.12603

35. Campbell ANC, Turrigiano E, Moore M, Miele GM, Rieckmann T, Hu MC, et al. Acceptability of a web-based community reinforcement approach for substance use disorders with treatment-seeking American Indians/Alaska natives. *Community Ment Health J*. (2015) 51:393–403. doi: 10.1007/s10597-014-9764-1

36. Daley CM, Daley SM, Pacheco CM, Smith TE, Talawyma M, McCloskey C, et al. Feasibility of implementing the all nations breath of life culturally tailored smoking cessation program for American Indians in multi-tribal urban communities. *Nicotine Tob Res*. (2018) 20:552–60. doi: 10.1093/ntr/ntx030

37. Duvivier H, Gustafson S, Greutman M, Jangchup T, Harden AK, Reinhard A, et al. Indian Health Service pharmacists engaged in opioid safety initiatives and expanding access to naloxone. *J Am Pharm Assoc*. (2017) 57:S135–40. doi: 10.1016/j.japh.2017.01.005

38. Gould GS, Bovill M, Pollock L, Bonevski B, Gruppetta M, Atkins L, et al. Feasibility and acceptability of indigenous counselling and nicotine (ICAN) QUIT in pregnancy multicomponent implementation intervention and study design for Australian indigenous pregnant women: a pilot cluster randomised step-wedge trial. *Addict Behav*. (2019) 90:176–90. doi: 10.1016/j.addbeh.2018.10.036

39. Hanson JD, Oziel K, Sarche M, MacLehose RF, Rosenman R, Buchwald D. A culturally tailored intervention to reduce risk of alcohol-exposed pregnancies in American Indian communities: rationale, design, and methods. *Contemp Clin Trials*. (2021) 104:106351. doi: 10.1016/j.cct.2021.106351

40. Hirchak KA, Hernandez-Vallant A, Herron J, Cloud V, Tonigan JS, McCrady B, et al. Aligning three substance use disorder interventions among a tribe in the Southwest United States: pilot feasibility for cultural re-centering, dissemination, and implementation. *J Ethn Subst Abus*. (2020) 21:1219–35. doi: 10.1080/15332640.2020.1836701

41. Hirchak KA, Jansen K, Lyons AJ, Herron J, Bergerson D, Shaw J, et al. Lessons shared: implementation of a randomized clinical trial for alcohol use disorders with five American Indian and Alaska native communities. *Collab J Community-Based Res Pract*. (2021) 4:5. doi: 10.33596/coll.76

42. Jason LA, Kassanits J, Reilly A, Bobak T, Guerrero M, Stevens E, et al. A promising recovery housing model for American Indian communities. *J Community Psychol*. (2019) 47:1926–36. doi: 10.1002/jcop.22237

43. Kiepek N, Hancock L, Toppozini D, Cromarty H, Morgan A, Kelly L. Facilitating medical withdrawal from opiates in rural Ontario. *Rural Remote Health*. (2012) 12:2193. doi: 10.22605/RRH2193

44. Landry M, Veilleux N, Arseneault JE, Abboud S, Barrieau A, Bélanger M. Impact of a methadone maintenance program on an aboriginal community: a qualitative study: Table 1. *CMAJ Open*. (2016) 4:E431–5. doi: 10.9778/cmajo.20150076

45. Marley JV, Kitaura T, Atkinson D, Metcalf S, Maguire GP, Gray D. Clinical trials in a remote aboriginal setting: lessons from the BOABS smoking cessation study. *BMC Public Health*. (2014) 14:579. doi: 10.1186/1471-2458-14-579

46. McDonell MG, Nepom JR, Leickly E, Suchy-Dicey A, Hirchak K, Echo-Hawk A, et al. A culturally-tailored behavioral intervention trial for alcohol use disorders in three American Indian communities: rationale, design, and methods. *Contemp Clin Trials*. (2015) 47:93–100. doi: 10.1016/j.cct.2015.12.010

47. Orr MF, Burduli E, Hirchak KA, Walsh Dotson JA, Young SL, Nelson L, et al. Culturally-tailored text-messaging intervention for smoking cessation in rural American Indian communities: rationale, design, and methods. *Contemp Clin Trials Commun*. (2019) 15:100363. doi: 10.1016/j.conctc.2019.100363

48. Patten CA, Lando HA, Desnoyers CA, Barrows Y, Klejka J, Decker PA, et al. The healthy pregnancies project: study protocol and baseline characteristics for a cluster-randomized controlled trial of a community intervention to reduce tobacco use among Alaska native pregnant women. *Contemp Clin Trials*. (2019) 78:116–25. doi: 10.1016/j.cct.2019.01.012

49. Santos LA, Braun KL, Kamuela Ae'a LS. Institutionalizing a comprehensive tobacco-cessation protocol in an indigenous health system: lessons learned. *Prog Community Health Partnersh*. (2008) 2:279–89. doi: 10.1353/cpr.0.0038

50. Venner KL, Greenfield BL, Hagler KJ, Simmons J, Lupee D, Homer E, et al. Pilot outcome results of culturally adapted evidence-based substance use disorder treatment with a southwest tribe. *Addict Behav Rep*. (2016) 3:21–7. doi: 10.1016/j.abrep.2015.11.002

51. Hirchak KA, Nadeau M, Vasquez A, Hernandez-Vallant A, Smith K, Pham C, et al. Centering culture in the treatment of opioid use disorder with American Indian and Alaska native communities: contributions from a National Collaborative Board. *Am J Community Psychol*. (2023) 71:174–83. doi: 10.1002/ajcp.12620

52. Yuan NP, Mayer BM, Joshweseoma L, Clichee D, Teufel-Shone NI. Development of guidelines to improve the effectiveness of community advisory boards in health research. *Prog Community Health Partnersh*. (2020) 14:259–69. doi: 10.1353/cpr.2020.0026

53. Walters KL, Simoni JM. Reconceptualizing native women's health: an "Indigenist" stress-coping model. *Am J Public Health*. (2002) 92:520–4. doi: 10.2105/AJPH.92.4.520

54. Venner KL, Donovan DM, Campbell ANC, Wendt DC, Rieckmann T, Radin SM, et al. Future directions for medication assisted treatment for opioid use disorder with American Indian/Alaska natives. *Addict Behav*. (2018) 86:111–7. doi: 10.1016/j.addbeh.2018.05.017

55. Oetzel J, Scott N, Hudson M, Masters-Awatere B, Rarere M, Foote J, et al. Implementation framework for chronic disease intervention effectiveness in Māori and other indigenous communities. *Glob Health*. (2017) 13:69. doi: 10.1186/s12992-017-0295-8

56. McCuistian C, Peteet B, Burlew K, Jacquez F. Sexual health interventions for racial/ethnic minorities using community-based participatory research: a systematic review. *Health Educ Behav*. (2023) 50:107–20. doi: 10.1177/10901981211008378

57. O'Mara-Eves A, Brunton G, Oliver S, Kavanagh J, Jamal F, Thomas J. The effectiveness of community engagement in public health interventions for disadvantaged groups: a meta-analysis. *BMC Public Health*. (2015) 15:129. doi: 10.1186/s12889-015-1352-y

58. Brown AF, Ma GX, Miranda J, Eng E, Castille D, Brockie T, et al. Structural interventions to reduce and eliminate health disparities. *Am J Public Health*. (2019) 109:S72–8. doi: 10.2105/AJPH.2018.304844

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Mapping the knowledge structure and trends in Australian Indigenous health and wellbeing research from 2003 to 2022: a scientometric analysis

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The health and wellbeing of Australian Indigenous peoples is a nationally sanctioned priority, but despite this, few studies have comprehensively analyzed the features and characteristics of the research in the field. In this regard, a comprehensive scientometric analysis and knowledge mapping to systematically summarize and discuss the current state of research, research trends, and emerging areas of research were conducted. Original articles and reviews published between 2003 and 2022 were obtained from the Web of Science Core Collection. CiteSpace and VOSviewer software were used to perform scientometric analysis and knowledge mapping. An examination of document and citation trends, authors, institutions, countries/regions, journals, and keywords was untaken, while co-citation, co-occurrence, and burst analysis provide insights and future development in this area. A total of 2,468 documents in this field were retrieved. A gradual increase in the number of documents over the past two decades is observed, with the number of documents doubling every ~7.5 years. Author Thompson SC and Charles Darwin University published the most documents, and 85.6% were affiliated with only Australian-based researchers. The *Australian and New Zealand Journal of Public Health* is the most prominent journal publishing in the field. The most commonly co-occurring keyword was “health,” and the keyword “risk” had the longest citation burst. Five keyword clusters were identified; “cultural safety” was the largest. This study articulates the knowledge structure of the research, revealing a shift from population-level and data-driven studies to more applied research that informs Indigenous peoples health and wellbeing. Based on this review, we anticipate emergent research areas to (1) reflect a more comprehensive understanding of the multidimensional factors that shape Indigenous health and wellbeing; (2) move beyond a deficit-based perspective; (3) respect cultural protocols and protect the rights and privacy of Indigenous participants; (4) address racism and discrimination within the healthcare system; (5) foster respectful, equitable, and collaborative research practices with Indigenous peoples; (6) provide culturally appropriate and effective interventions for prevention, early intervention, and treatment; and (7) ensure equitable change in systems to enhance access, quality, and outcomes in health and wellbeing.

KEYWORDS

Aboriginal and Torres Strait Islander, Indigenous, Australia, health, wellbeing, scientometric analyses, trends, knowledge

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1 Introduction

Aboriginal and Torres Strait Islander peoples (herein Indigenous[1]), represent the oldest continuing cultures in the world. Since the colonization of Australia, Indigenous peoples have experienced profound trauma and losses in social and emotional, health, and wellbeing through the devastation or fragmentation of traditional lands, languages, culture and community (Dudgeon and Walker, 2015; Paradies, 2016). Today, Indigenous peoples account for 3.8% (an estimated 984,400 people) of the total Australian population and are projected to reach 1 million people by 2028 (Australian Bureau of Statistics, 2022). Despite steady improvements in the life outcomes of Indigenous peoples over the past few decades, a notable gap compared to the wider community remains (Australian Institute of Health and Welfare, 2015). In an unprecedented shift in the way governments have previously worked, closing this gap is a national priority that embraces the strength and resilience of Indigenous culture and communities as a foundation for partnership and shared decision-making (Arabena et al., 2014; Australian Government, 2021; National Indigenous Australians Agency, 2022).

Indigenous peoples view health as a holistic concept that encompasses more than just the absence of disease or illness. It embraces elements like cultural identity and spiritual well-being, family and kinship, connection to the land and its care, traditional knowledge and beliefs, language preservation, and active participation in community life, along with access to ancestral lands for both individuals and communities (AIHW, 2022). Indigenous leadership plays a crucial role in enhancing research impact and ensuring its benefits for Indigenous communities. This is achieved by prioritizing activities that hold significance and align with the community's interests and cultural perspectives (Kiatkoski Kim et al., 2020). When research is led by Indigenous peoples, it has the potential to foster the creation of workforce development, strategies, policies, and procedures at regional, national, and global levels that genuinely support Indigenous peoples, all viewed through an Indigenous perspective.

Despite the previous reviews of Indigenous health research that have identified continued growth in outputs (Kinchin et al., 2017), lack of intervention research and research in urban settings (Jennings et al., 2021; McGuffog et al., 2023), and the need to hold to account health systems (Kennedy et al., 2022), there have been limited attempts to explore the evolution and current state of knowledge structure of the research. In 2006, Sanson-Fisher et al. (2006) examined the scientific literature related to the health of Indigenous peoples collectively from Australia, Canada, New Zealand and the United States at time points between 1987 and 2003. They conclude that the abundance of descriptive research is not considered an exemplar and encourage research organizations and researchers to consider this when developing research policies. In 2012, Derrick et al. (2012) published a bibliometric analysis of Indigenous health research in Australia (1972–2008). They conclude that while the volume of citations in selected health disciplines continues to grow, this still does not reflect the gravity of Indigenous health problems.

The National Health and Medical Research Council (NHMRC) has made significant commitments to Indigenous health and medical research in recent years (National Health and Medical Research Council, 2018). In 2018, they committed to allocate at least 5% of the Medical Research Endowment Account specifically to Indigenous health and medical research. The 2021 report demonstrates that this goal has been surpassed, evidenced by the funding of 206 active grants totaling over $58 million (National Health and Medical Research Council, 2021). However, despite more than 15 years since the introduction of the Australian Government's Closing the Gap strategy, the 2031 targets are not progressing as planned. The 2022 Lowitja Institute Close the Gap Campaign Report (Lowitja Institute, 2022) underscores the importance of sustained investment in research that informs policy and practice reform as a critical step towards empowering Indigenous communities and improving health and social outcomes.

One direction is to investigate the evolution of a research topic. In our attempt to detect trends in Indigenous health and wellbeing research, we combine modelling and visualization to establish a knowledge base that will have important value for academics, practitioners, and government departments to formulate public health strategies and provide support and guidance for future research.

In this study, we combine bibliometric and scientometric techniques to analyze the knowledge structure regarding research productivity, and collaboration across authors, institutions, and countries/regions, and to reveal trends and forecast emerging areas of research. Despite similar approaches in other fields (You et al., 2021), this study is the first to detail a scientometric analysis of the characteristics, knowledge structure and trends in Indigenous health and wellbeing research. The key objectives of this study include:

> *RQ*1. What are the trends and forecasted growth in documents?
>
> *RQ*2. Who are the most influential authors, institutions, countries/regions, and journals?
>
> *RQ*3. Which documents and keywords are the most impactful?
>
> *RQ*4. What are the dominant topics, trends, and emerging research areas?

Addressing these research questions will fill important gaps in the current body of knowledge, and advance our understanding of research related to the health and wellbeing of Australian Indigenous peoples.

2 Materials and methods

2.1 Research design

This study examines the published scientific literature related to Indigenous health and wellbeing using scientometric analysis and knowledge mapping. The detailed procedure is discussed in the following sections.

2.2 Scientometric analysis

Scientometric analysis is a quantitative research method that focuses on the analysis and mapping of scientific literature, to explore research themes and collaboration clusters, and to identify gaps and

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[1] To include all Aboriginal and Torres Strait Islander peoples, while recognizing that Aboriginal and Torres Strait Islander peoples have distinct cultures and identities.

trends (Mingers and Leydesdorff, 2015; López-Pernas et al., 2023). It involves the application of statistical and bibliometric techniques to evaluate and measure scientific activity to provide insights into the structure, growth, and impact of scientific knowledge (Noyons et al., 1999; Borgohain et al., 2021; Basumatary et al., 2022). Analyzing citation patterns, trends, and authorship networks, scientometric analysis can identify influential researchers, leading institutions, emerging research areas, and the overall development of scientific fields (Kastrin and Hristovski, 2021). In turn, this work can help researchers, policymakers, and institutions gain insights into the dynamics of scientific knowledge production, dissemination, and impact and it can inform decision-making related to resource allocation, funding strategies, identification of research trends, and evaluation of individual researchers, institutions, or research programs.

2.3 Search strategy and data collection

The selected source for the literature search and collection is the Web of Science Core Collection (WoSCC) database (Singh et al., 2021; Xia et al., 2021; Gusenbauer, 2022). A retrieval plan is detailed in Figure 1 for indexed documents relating to Indigenous health and wellbeing research, authored by researchers with an Australian affiliation, and published between 1 January 2003 and 31 December 2022. In order to mitigate the potential for bias resulting from ongoing database modifications, the retrieval and export of documents were executed on a single day (1 January 2023). We included papers that focused solely on Indigenous health and wellbeing, as well as those that incorporated data related to Indigenous health and wellbeing, such as the distribution of diseases or population-level risk factors. We adopted the Lowitja Institute search syntax for Aboriginal and Torres Strait Islander people (Lowitja Institute, 2022) and the final search string applied in the WoSCC: (TI = [Indigenous OR Aborigin* OR Torres Strait Islander* OR First AND (People* OR Nation*")] AND TS = [Australia AND (health OR wellbeing OR wellbeing OR well-being)] AND AD = [Australia] AND DOP = [01-Jan-2003 to 31-Dec-2022] AND Language = [English]).

Original articles and reviews (including early access articles) were included. A total of 2,800 documents were identified, of which, 332 records were excluded: meeting abstracts, editorial material, letters, and proceeding/data papers. Ethical approval was not applicable. For the present study, 2,468 documents (2,198 original articles and 279 reviews) were obtained as the final dataset for analysis. Data were obtained from WoSCC in .csv format for analysis, and in plain text format (full records and cited references) to visualize the data.

2.4 Measures

- Number of documents: To quantify the amount of research conducted over time, as distributed by authors, institutions, countries/regions, journals, and research categories.
- Number of citations: To examine trends in research over time, and to identify the most influential authors, institutions, countries/regions, and journals.
- Co-authorship: To evaluate collaboration networks of authors, institutions, and countries/regions, providing insight into the social connections across the domain.
- Co-citation: To analyze the frequency with which documents are cited together by other researchers, and to reveal clusters of interdisciplinary research trends.
- Co-occurrence: To identify related keywords and measure the strength of their links to visualize trending and emerging research themes.
- h-index: To quantify both productivity and impact.
- Journal Impact factor (JIF) and quartile in category (Q1-Q4) were obtained from the 2021 Journal Citation Report (Clarivate, 2022).

2.5 Data analysis and visualization

Based on the analysis approach, Microsoft Excel 2022 (Redmond, WA, United States) was used to analyze and graph document and citation metrics. The most influential documents were identified based on their citation count, and the top 10 authors, institutions, countries/regions, and journals were identified based on the number of documents. Journal research categories are presented as document counts and proportions.

VOSviewer (version 1.6.16) (Leiden University, Leiden, the Netherlands) was used to visualize the networks of authors and institutions (Fonseca Bde et al., 2016). Full-counting was applied and the threshold was set to ≥5 co-authored documents (Egghe et al., 2000). Based on these settings, the number of documents, citations, and total link strength (TLS) were determined. In this analysis, the nodes represent the author or institution, the size of the node represents the number of documents, and the lines between nodes represent co-authorship links. The thickness of the line depicts link strength. Clustering analysis was used to identify sub-clusters of collaboration from the overall structure of the literature (Rodriguez and Laio, 2014; Chen, 2017; Jiang et al., 2017; Singh et al., 2021).

CiteSpace (version 6.1.R6) (Wang and Lu, 2019) was used to explore the knowledge structure and research trends in the scientific literature. Parameters for this investigation were set to: time-slicing from 2003 to 2022 (4 years per slice), look back years = −1, link retaining factor = −1, top N% = 100%, top N = 50, and g-index = 25. Default settings for text processing and links were preserved and metrics such as citation burstiness, Sigma, Silhouette and betweenness centrality were reported.

To achieve this, the following scientometric techniques were employed:

- Co-citation analysis of documents quantifies how often documents are cited together, revealing influential publications with high citations (bursts) and related clusters.
- Keyword co-occurrence analysis identifies the most important keywords (extracted after searching the titles, abstract, and keywords) that represent the conceptual building blocks of the scientific literature.
- Burst detection of highly co-cited documents and keyword co-occurrences highlight significant literature and keywords (Su and Lee, 2010). The burst strength list is created using an algorithm mapping of hierarchical structure to capture increases in popularity within a specified period
- Cluster analysis divides the networks into clusters by extricating terms from the title, abstract, author keywords, and keyword plus

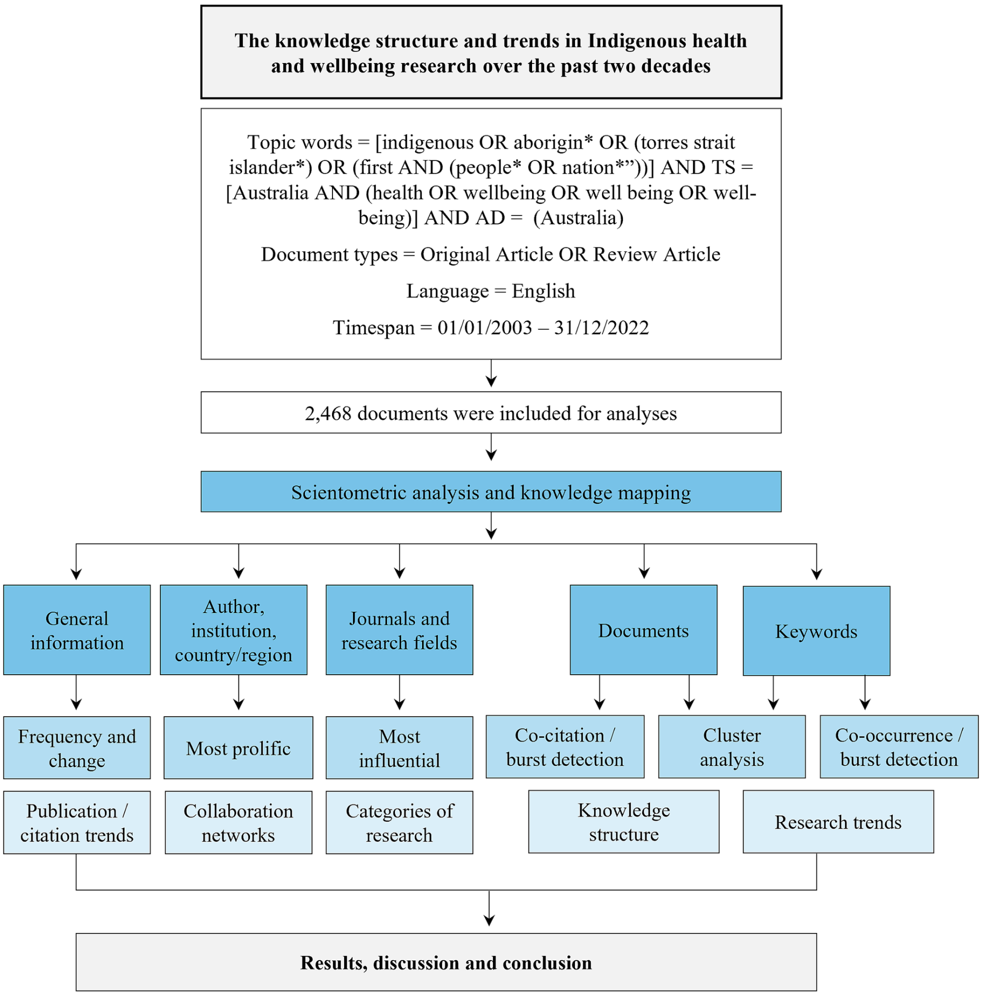


FIGURE 1
Methodological framework for the current study.

using the default algorithm log-likelihood ratio test. The top 50 keywords that appear in each time slice are presented and clusters are identified using distinct colors and labelled using CiteSpace.

- Timeline visualizations are generated from cluster analysis on a discrete horizontal axis. The term source includes title, abstract, author, keywords, and keyword plus of cited documents using the log-likelihood ratio algorithm. Clusters are arranged in a vertical manner descending in size, with the largest cluster at the top (#0). Each node represents a document, links between two nodes represent the co-citation/co-occurrence, and color corresponds to the year they most recently appeared. Documents with a citation burst and/or highly cited are denoted with a bright purple ring.

3 Results

3.1 Trends in publications and citations

Overall, publications have steadily increased from 21 documents in 2003, to 236 documents in 2022 (an average of 123 documents published/year). The average annual performance is 15.1% (95% CI 8.1 to 23.1%) and on average, the number of documents doubled every 7.5 years (Figure 2). In the first decade, the average annual performance of 23.2% (95% CI 13.7 to 36.9%) indicates an initial rapid growth period, which slowed to a stable 7.8% (95% CI 6.1 to 13.9%) in the second decade. A regression exponential trend line indicates consistent growth ($R^2 = 0.9792$) over time. A forecast estimate of future trends in Indigenous health and wellbeing research based on the equation of model fit indicates an additional 1,488 documents (1,136 original articles and 353 reviews) published in the next five years (2023–2027); representing an average annual performance of 6.20% (95% CI 4.7 to 10.9%).

3.2 Analysis of authors

The author network for Indigenous health and wellbeing research consists of 6,580 authors; 95% ($n = 2{,}344$) of documents are co-authored, and the median number of authors per paper is five (range: 1 to 65). The top 10 authors with the greatest number of documents are listed in Table 1 and collectively account for almost one quarter ($n = 608$; 24.6%) of all documents included in this study. These authors are affiliated with eight institutions, four of which are members of Australia's leading research-intensive universities, known as the Group of Eight (Go8) (The Group of Eight Ltd, 2022). Thompson, SC has the greatest number of documents (*n* = 87), total citations (*n* = 1,749), and *h*-index (Borgohain et al., 2021) related to the dataset in this study.

Figure 3A presents a visualization of the co-authorship network among 446 authors, grouped into 17 clusters with 3,197 links. In this analysis, Brown, A (cluster 2, 51 nodes) has the greatest number of co-authored documents (*n* = 57 and 79 links), Thompson, SC (cluster 7, 31 nodes) has the most citations (*n* = 931), and Ward, J (cluster 5, 36 nodes) has the highest TLS of 209. Other authors with notably high TLS include Brown, A (196), Garvey, G (cluster 8, 27 nodes) (179), and Thompson SC (157).

3.3 Analysis of institutions

A network of 1,616 institutions has contributed to the research; Table 2 lists the top 10 institutions. Of note, six of these institutions are members of Australia's Go8 (The Group of Eight Ltd, 2022) universities and one, the Menzies School of Health Research, is a young (38 years) medical health research institute dedicated to improving health outcomes for Indigenous Australians. Charles Darwin University ranked first with 508 articles followed by the University of Sydney (*n* = 476), and the Menzies School of Health Research (*n* = 417). A visualization of the institution network highlights four clusters. As can be seen in Figure 3B, cluster 1 (red, 56 nodes) is co-led by the University of Melbourne (links = 129, TLS = 799) and Charles Darwin University (links = 107, TLS = 775). Cluster 2 (green, 49 nodes) is led by the University of Sydney (TLS = 1,058), cluster 3 (blue, 33 nodes) is led by The University of Queensland (TLS = 720), and cluster 4 (yellow, 32 nodes) is led by the University of Western Australia (TLS = 641). Other institutions with high TLS include Flinders University (490), Menzies School of Health Research (481), and the University of Adelaide (473).

3.4 Analysis of countries/regions

A total of 54 countries/regions have contributed to the production of Indigenous health and wellbeing research over the past two decades; 85.6% (*n* = 2,113) are authored by Australian institutions only, and the remaining 355 publications are affiliated with authors predominantly from the United States (25.07%), Canada (24.22%), England (22.53%), and New Zealand (20.28%) (Table 3). While the number of documents co-authored with the People's Republic of China and Brazil is not high, the average citation rate is outstanding, suggesting that the quality and application of these publications are elevated.

3.5 Analysis of journals and research fields

Over the past two decades, a total of 632 journals have been the 2,468 documents. The top 10 most active journals are presented in Table 4 and account for 30% (n = 739) of all documents. The *Australian and New Zealand Journal of Public Health* has published the most documents (n = 113), followed by *the International Journal of Environmental Research and Public Health* (n = 96). The *Medical Journal of Australia* is the only Q1-ranked journal and has the highest impact factor at 12.766. Six of the 10 journals are classified as *Public, Environmental, and Occupational Health* based on the journal's research area (as categorized by WoSCC).

Figure 4 compares the top 10 journal categories for documents published in the first (2003–2012) and second (2013–2022) decades. Of note, six categories remain in the top 10, with "public, environment and occupational health," "healthcare sciences and services," and "general and internal medicine" as the top three categories. Documents published in journals categorized as "public, environmental and occupational health" had the greatest increase in the number of publications from 177 in decade 1 to 681 in decade 2, and as a proportion, publications categorized as "education and educational research" increased by 1,260%. Despite the categories of "substance abuse," and "biomedical social sciences," not being included in the

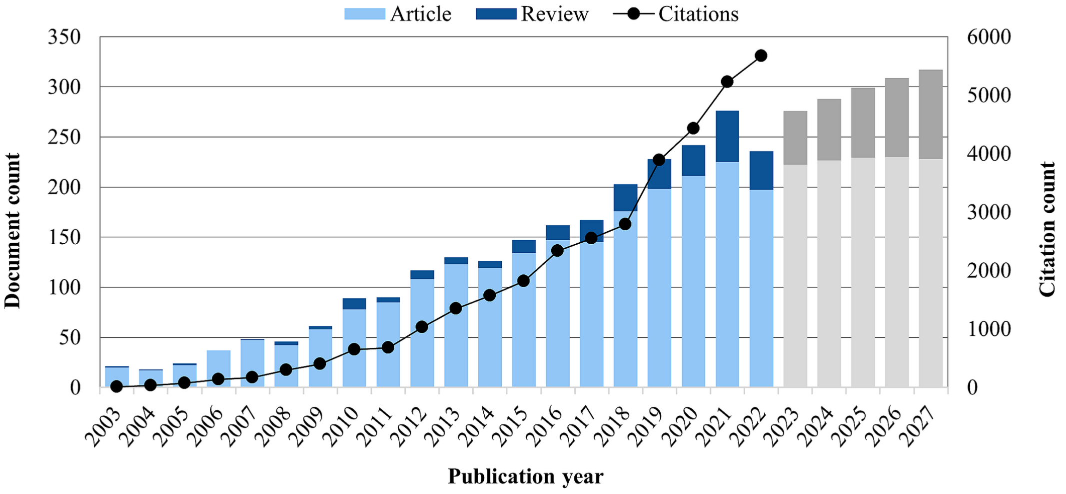


FIGURE 2
Document counts and citation growth trends of Indigenous health and wellbeing research (2003–2022), and document growth forecast for the next 5 years (2023–2027).

TABLE 1 Top 10 authors publishing Indigenous health and wellbeing research (2003–2022).

#	Author	No. of documents[a]	Total citations (average/document)[a]	*h*-index[a]
1	Thompson SC	87	1,749 (20.10)	24
2	Brown A	74	1,130 (15.27)	19
3	Jamieson LM	69	668 (9.68)	16
4	Eades S	65	940 (14.46)	18
5	Bailie R	61	1,203 (19.72)	19
6	Ward J	53	484 (9.13)	13
7	Garvey G	51	538 (10.55)	13
8	Clough A	40	671 (16.78)	16
9	Atkinson D	38	427 (10.95)	12
10	O'Dea K	36	761 (21.14)	16

[a] Metrics reported are calculated for the dataset included in this study only.

top 10 for decade 2, documents still increased by 2–2.5 times. "Ophthalmology" is the only category to decrease between decades.

3.6 Analysis of documents

Key steps to building a knowledge structure include identification of highly cited articles and key research areas. Figure 5A presents a visualization of the network of highly co-cited documents ($n = 241$, links = 2,987). To ensure a clearer picture, only nodes with ≥60 co-citations are labelled. The top three highly co-cited documents are the publications by Gracey and King (2009), Vos et al. (2009), and Bessarab and Ng'andu (2010). The publication by Carson et al. (2007), titled "social determinants of Indigenous health," examines the enduring health impacts of the Indigenous experience of dispossession, colonial rule, and racism. Published in 2007, it stands out as an influential document in the literature network (centrality score of 0.60). Published in earlier years, documents by King et al. (2009), and the NHMRC (National Health and Medical Research Council, 2018) continue to be consistently referenced and cited extensively in recent times.

Figure 5B shows the top 10 co-cited documents with the strongest citation bursts. Time is represented by the blue line, and the period when the document bust occurred, by the red line. The article with the longest citation burst lasting from 2007 to 2018 is titled "burden of disease and injury in Aboriginal and Torres Strait Islander peoples: the Indigenous health gap" (Vos et al., 2009). This study uses national population health datasets and Indigenous-specific epidemiological studies to measure the Indigenous health gap.

Based on the co-citation analysis network (Figure 5A), cluster analysis identified 10 research clusters (Trujillo and Long, 2018) which are visualized over time in Figure 5C (detailed in Supplementary File 1). The Q Score and S value (0.7965 and 0.9198, respectively) indicate that the network is reasonably divided, and the precision of clustering is high. The ranking of clusters is determined by the number of documents, where "disadvantage" (#0) is the largest cluster and "children" (#9) is the smallest. The clusters of "First Nations" (#1), "child health" (#2), and "social determinants" (#5) are trends in Indigenous health and wellbeing research in recent years.

3.7 Analysis of keywords

The keywords of a document reflect the research focus of publications. Here, we analyze the co-occurrence of keyword trends to explore research topics and frontiers in the field of Indigenous health and wellbeing. A network of 145 nodes (related keywords), and 862 links (connections), with a density of 0.0826 is shown in Figure 6A. The most commonly co-occurring keywords are "health" (363 times), "care" (187 times), "Australia" (184 times), "Indigenous health" (168 times), and "community" (166 times). Burst detection identified 42 keywords with a minimum duration of two years; the 25 keywords with the strongest citation bursts are shown in Figure 6B. The term "risk factors" had the longest burst period (2003–2015) and keyword bursts lasting until 2022 include: "Indigenous peoples," "cultural safety," "qualitative research," "social determinants," "smoking," "perspectives," "First Nations," "public health," "quality of life," and "colonization." These 10 keywords reflect the most recent research trends and belong to clusters labelled "cultural safety," "mental health" and "Indigenous health."

The keyword network was divided into five clusters which represent the main keyword categories of Indigenous health and wellbeing research (see Supplementary File 2). The evolution of keywords over time can be seen in Figure 6C. The largest cluster (#0) is "cultural safety" with 35 keywords related to cultural safety in healthcare, disparities, racism, barriers to participation, education, communication, quality of health services, women's health, and Indigenous populations. Cluster two is "mental health" (#1) with 29 keywords. The theme suggests an exploration of mental health policies, interventions, and research aimed at improving mental wellbeing and addressing the unique challenges faced by Indigenous peoples. The third cluster (#2) "Northern Territory" contains 29 topics, that together suggest an exploration of the patterns, diagnoses, and associations related to prominent health issues within Indigenous communities. The fourth cluster (#3) is "disease" with 22 topics that highlight various aspects related to disease, particularly in children, in the context of a place (Western Australia). The final cluster "Indigenous health" (#4) has 22 topics that cover primary healthcare, risk, prevalence, and determinants of health.

4 Discussion

This study provides an extended view of Indigenous health and wellbeing research published over the past two decades – offering insights into the achievements, knowledge structure, research trends and emerging areas. It is the first to apply scientometric techniques to represent the dynamic and structural features of the research visually.

Our analysis of 2,468 documents reveals prominent authors, institutions, countries/regions, and journals who have contributed to Indigenous health and wellbeing research over the past two decades - with an estimated 60% increase in the volume of documents by 2027. This growth likely reflects the growing national priorities for the health and wellbeing outcomes for Indigenous peoples and communities in Australia (National Health and Medical Research Council, 2018; Lowitja Institute, 2022), the availability of guidelines and frameworks related to the ethical conduct of research with Indigenous peoples and communities (National Health and Medical Research Council, 2018; Australian Institute of Aboriginal and Torres Strait Islander Studies, 2020), along with the significant commitment in targeted research funding (National Health and Medical Research Council, 2018). In

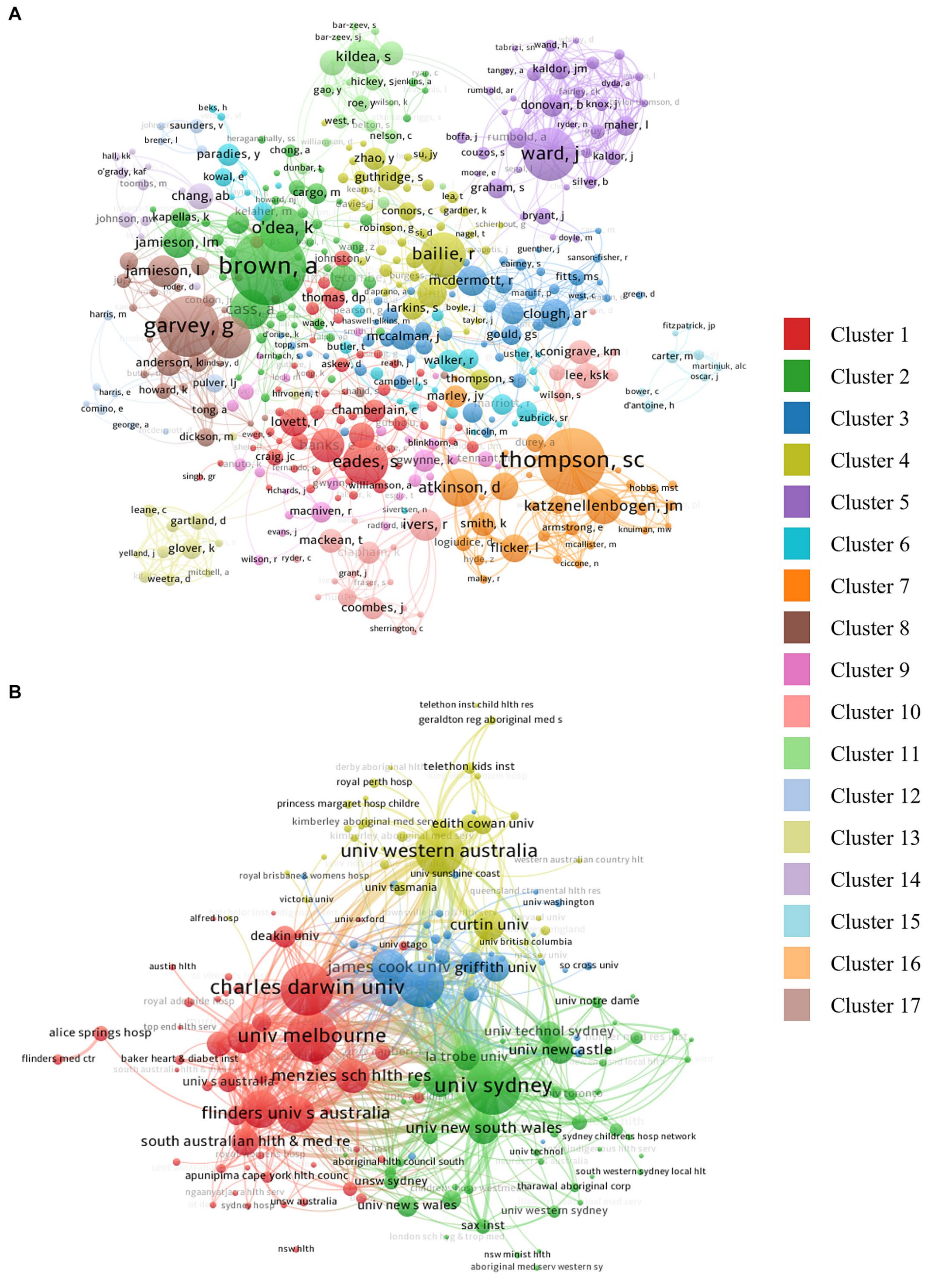


FIGURE 3
Visualization of the **(A)** authorship collaboration network, and **(B)** institutions co-authorship.

particular, the NHMRC has committed 5% of its total research funding budget to Indigenous health and medical research since 2008; in 2021 the NHMRC allocated 7.09% of funding or $58.1 million (National Health and Medical Research Council, 2018).

Each of the top 10 authors published at least 36 documents. Thompson SC had the greatest number of documents, number of total citations, and highest h-index. The focus of Professor Thompson's research is the prevention and management of chronic disease in remote communities and Aboriginal health. In a 2010 review of Australia's National bowel cancer screening program, barriers that exclude vulnerable minorities, including Indigenous groups, from participating in bowel cancer screening initiatives and the greater

TABLE 2 Top 10 institutions publishing Indigenous health and wellbeing research (2003–2022).

#	Institution	Location	Established	No. of documents[a]	Total citations (average/document)[a]
1	Charles Darwin University[b]	Northern Territory	2003	508	8,248 (16.11)
2	University of Sydney	New South Wales	1850	476	6,366 (13.32)
3	Menzies School of Health Research[c]	Northern Territory	1985	417	7,041 (16.72)
4	University of Melbourne	Victoria	1853	357	6,147 (17.17)
5	University of Western Australia	Western Australia	1911	354	5,441 (15.33)
6	University of Queensland	Queensland	1909	312	4,499 (14.42)
7	University of New South Wales	New South Wales	1949	302	3,338 (11.05)
8	Flinders University	South Australia	1966	237	2,697 (11.38)
9	University of Adelaide	South Australia	1874	215	2,593 (12.06)
10	James Cook University	Queensland	1970	206	3,341 (16.22)

[a] Metrics reported are calculated for the dataset included in this study only.
[b] Established after a merger between three institutions.
[c] An independent medical and research institute within Charles Darwin University.

TABLE 3 Top 10 countries/regions contributing to Indigenous health and wellbeing research (2003–2022).

#	Country/Region	No. of documents[a]	Total citations (average/document)[a]
1	USA	89	2,239 (24.88)
2	Canada	86	1,859 (21.62)
3	England	80	1,148 (14.17)
4	New Zealand	72	2,012 (27.56)
5	Scotland	12	199 (16.58)
6	People's Republic China	11	660 (55.00)
7	Brazil	10	626 (62.60)
8	South Africa	6	75 (12.50)
9	Germany	6	58 (9.67)
10	France	5	155 (31.00)

[a] Metrics reported are calculated for the dataset included in this study only.

incidence of late-stage cancer and mortality among Indigenous peoples are highlighted (Christou et al., 2010). Since then, studies to address barriers through appropriate health promotion and education have been rolled out. This includes the National Indigenous Bowel Screening pilot among 44 Indigenous primary healthcare centers (Menzies School of Health Research, 2020) in 2018 which is now available nationally.

According to our analysis, authors affiliated with Charles Darwin University had published the most documents ($n = 508$; 20.6%), and despite the University of Melbourne ranking fourth, it had the highest average citation rate of 17.7. The University of Sydney co-authored the greatest number of publications with international institutions ($n = 84$). It is worth noting that these lead institutions are renowned research and higher education organizations with prominent positions in Indigenous health research portfolios. Collectively, authors with affiliations to institutions based in the USA co-authored the most documents with Australia, albeit authors affiliated with the University of Toronto had the greatest single number of documents ($n = 31$).

The *Australian and New Zealand Journal of Public Health* published the largest number of documents ($n = 113$), and the *Medical Journal of Australia* had the highest impact factor (12.776) and citation count ($n = 3{,}251$). It is worth noting that most JCR partitions are categorized as Q2 and only one is a Q1 journal. These data will help researchers when they submit articles about Indigenous health and wellbeing in the future.

4.1 Knowledge structure

Central to the influence of relevant literature on the topic is the number of citations documents have received. In this study, 181 papers received only one citation, and 270 have not yet been cited. The average number of citations per document is 14.19 with a h-index of 68. Among the top 10 documents with the greatest citation rate (Supplementary File 3), nine of these papers are original articles and one is a review. Three are published in the *Medical Journal of Australia* (IF = 7.738) and two in the *Lancet* (IF = 79.321). The document that received the most citations ($n = 481$) accounts for 1.37% of the total citation count (Anderson et al., 2016) and is also recognized as a highly cited paper in the field of clinical medicine. Titled "Indigenous and tribal peoples' health (The Lancet-Lowitja Institute Global Collaboration): a population study," it is a large population study that reviews the health and social outcomes for Aboriginal and Torres Strait Islander peoples from across 23 countries. It is noteworthy that this article containing 65 authors and 43 affiliations, had one of the longest citation burst values lasting from 2019 to 2022. Reading the 10 most influential documents can assist researchers in gaining a foundational understanding of the knowledge structure pertaining to Indigenous health and wellbeing research.

The dynamic structure of research in this field is characteristic of documents with strong citation bursts and co-citation clusters. Statistics from CiteSpace identified that 44 articles broke out in recent years (2019–2022), of which 42 belonged to cluster #1 "First Nations" and two belonged to cluster #2 "child health" (Figure 5C). The "First Nations" cluster is concentrated between 2015 to 2022. Central to this cluster are papers that explore strengths-based approaches to Indigenous health. These include the paper by Askew et al. (2020) "Closing the gap between rhetoric and practice in strengths-based approaches to Indigenous public health: a qualitative study," Harfield et al. (2020) "Assessing the quality of health research from an

TABLE 4 Top 10 journals publishing Indigenous health and wellbeing research (2003–2022).

#	Journal	No. of documents[a]	Total citations (average/document)[a]	Category (Q1-Q4)[a]	JIF[a]
1	Australian and New Zealand Journal of Public Health	113	1,735 (15.65)	Public, environmental, and occupational health (Q2)	3.755
2	International Journal of Environmental Research and Public Health	96	522 (6.87)	Environmental sciences (Q2); Public, environmental, and occupational health (Q2)	4.614
3	BMC Public Health	89	1,285 (14.44)	Public, environmental, and occupational health (Q2)	4.135
4	Medical Journal of Australia	82	3,251 (35.40)	Medicine, general and internal (Q1)	12.776
5	BMC Health Services Research	78	1,405 (18.01)	Health care sciences and services (Q3)	2.908
6	BMJ Open	75	674 (10.21)	Medicine, general and internal (Q2)	3.006
7	Rural and Remote Health	56	570 (10.96)	Public, environmental, and occupational health (Q3)	2.733
8	Australian Journal of Primary Health	53	491 (9.44)	Health care sciences and services (Q4); Primary healthcare (Q4); Public, environmental, and occupational health (Q4)	1.72
9	Australian Health Review	49	621 (13.50)	Health care sciences and services (Q4)	1.837
10	Australian Journal of Rural Health	46	582 (14.20)	Nursing (Q3); Public, environmental, and occupational health (Q3)	2.606

[a]Metrics reported are calculated for the dataset included in this study only.
JIF, Journal Impact Factor according to Journal Citation Reports 2021; Q1–Q4 = quartile in category ranking by JIF.

Indigenous perspective: the Aboriginal and Torres Strait Islander quality appraisal tool," Paradies (2016) "Colonization, racism and indigenous health." Child health is another hotspot that presents in 2011 in the highly co-cited editorial titled "Social determinants and the health of Indigenous Australians" (Marmot, 2011). In this short paper, Professor Marmot discusses the large social inequalities and the 17-year age gap in life expectancy between Indigenous and non-Indigenous Australians. The influence of early childhood development is discussed in relation to access to education suggesting that the environment in early childhood is key to health status along the social gradient (Marmot et al., 2008). A central paper within this cluster is also the highly co-cited systematic review of interventions for Indigenous peoples with chronic diseases by Gibson et al. (2015).

4.2 Research trends

Next, we reflect on trends in the research based on the timeline view of keyword co-occurrence clusters (Figure 6C). In the early 2000s, Indigenous health and wellbeing research had a particular focus on the prevalence, impact, and risk factors associated with various chronic diseases, including mental health, infectious diseases, cardiovascular disease, obesity, renal disease, otitis media, and oral health. This early research sought to understand the social determinants influencing health outcomes, such as cultural safety, access to healthcare services, and the impact of disparities in healthcare provision. The research tended to explore patterns of disease prevalence and mortality rates among Indigenous populations, including children, women, adolescents, and infants, in efforts to develop preventive programs and effective management strategies for Indigenous peoples and communities, particularly in regions such as the Northern Territory and Western Australia. Ultimately, the research sought to articulate health disparities and outcomes from intervention studies in Indigenous communities.

During the mid-2000s, Indigenous health and wellbeing research shifted to understanding and improving the experiences and participation of Indigenous peoples in health services. It explored the role of communication and cultural knowledge in enhancing healthcare interactions and outcomes and it sought to identify the unique challenges faced by Indigenous women in accessing and receiving appropriate healthcare services. The impact of cultural factors on health-seeking behaviors, and the role of education and knowledge exchange in improving health literacy within Indigenous communities is also a focus. This research would inform strategies to improve healthcare delivery, promote culturally sensitive practices, and develop targeted interventions that address the specific needs and experiences of Indigenous peoples. Ultimately, the research sought to contribute by ensuring active participation and improved experiences in healthcare settings for Indigenous communities.

By the late 2000s, a surge in research to understand and mitigate the burden experienced by Indigenous peoples in Australia, particularly associated with the impact of racism, historical colonization, and socio-cultural determinants, can be observed. Although instances of racism in Aboriginal and Torres Strait Islander health have been documented since the introduction of Closing the Gap in 2020, it was the Black Lives Matter movement that garnered worldwide recognition (The Lancet, 2020). Studies addressing barriers that hinder Indigenous peoples' access to quality primary healthcare services and exploring strategies that promote cultural competence are trending. The research emphasizes the importance of health promotion and physical activity in improving the quality of life and endorses the perspectives and experiences of Indigenous peoples to gain a deeper understanding of their needs and challenges. Inevitably, this research is designed to inform health policies and initiatives aimed at reducing health disparities and addressing the impact of racism on health and wellbeing (Gatwiri et al., 2021). Unless the historical and contemporary determinants of Indigenous health and wellbeing are addressed, the development of a culturally appropriate and equitable healthcare system is ambitious.

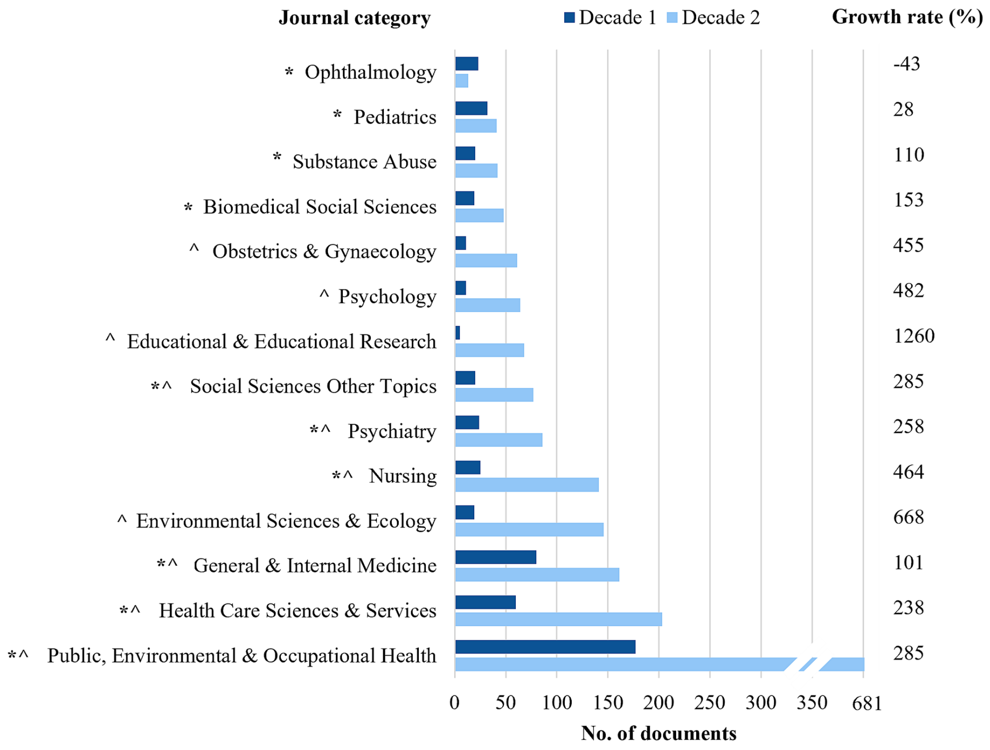


FIGURE 4
Top 10 journal research categories for documents published between 2003 to 2012 (decade 1) and 2013 to 2022 (decade 2). Growth rate (%) indicates the change in the number of publications between decade 1 and decade 2. *categories included in the top 10 for decade 1; ^categories included in the top 10 for decade 2.

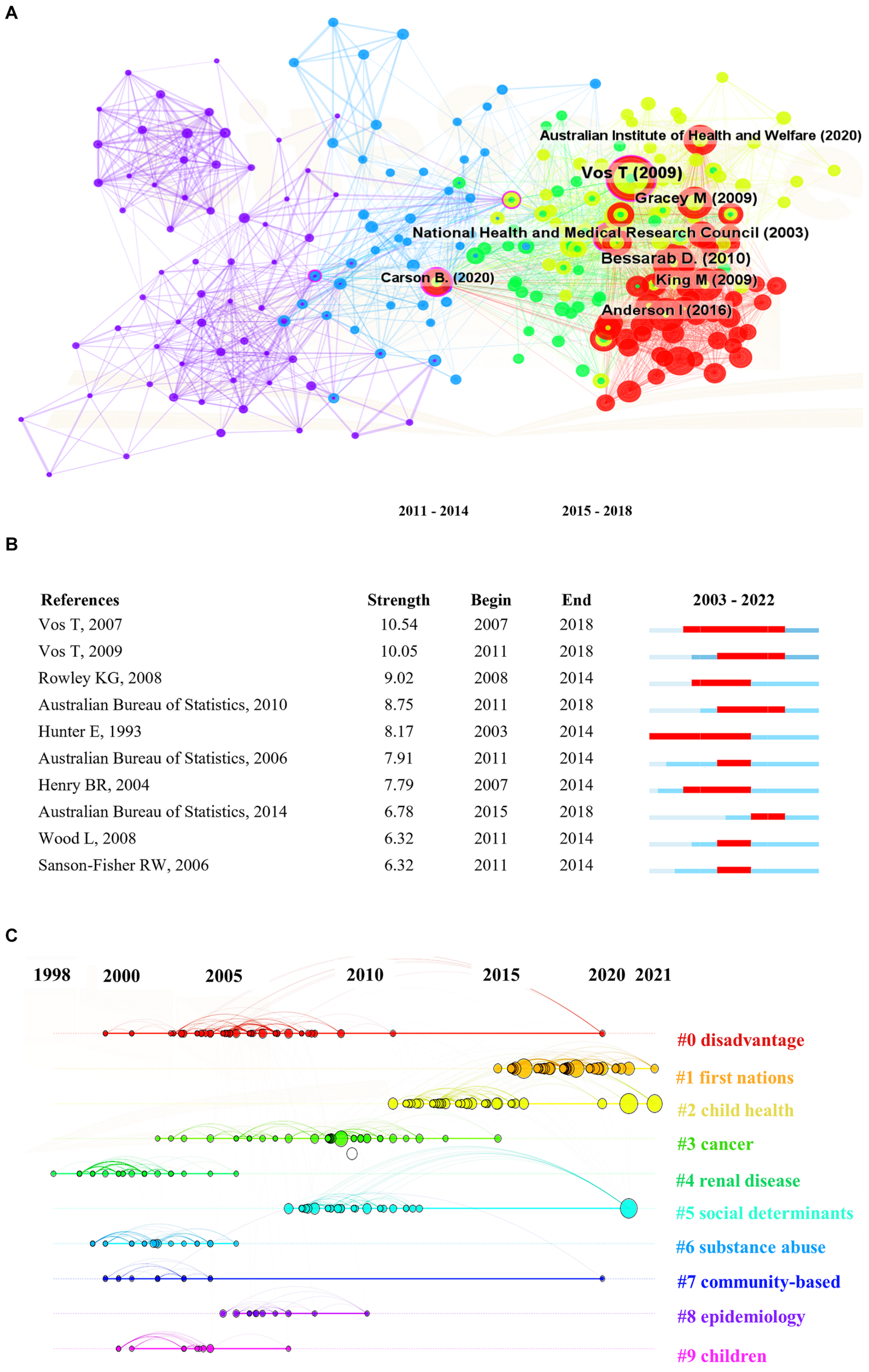
4.3 Emergent research areas

Despite the high level of investment, the gap in health and wellbeing outcomes between Indigenous and non-indigenous Australians remains alarmingly wide. Based on this study and through extensive reading of the literature, we consider the following areas to be emergent research approaches and practices (Figure 7).

Emergent approaches will contribute to a more holistic understanding of health and wellbeing, foster respect for Indigenous ways of knowing, and promote more equitable and inclusive research practices. An example is the concept of Two-Eyed Seeing introduced by Mi'kmaq Elders Albert and Murdena Marshall (Whiting et al., 2018). This approach seeks to integrate both Indigenous knowledge and Western scientific knowledge to address health disparities and promote wellbeing. It would involve combining traditional healing practices, cultural beliefs, and community wisdom with evidence-based medical and scientific approaches. Respecting the inherent strengths and knowledge present within Indigenous communities, while also valuing the advancements of Western medicine and research may include approaches such as:

- Intersectionality and holistic advances: Taking a more comprehensive understanding of the complex and multidimensional factors that shape Indigenous health, but in particular, wellbeing. This includes exploring the intersectionality of various factors, such as gender, age, socioeconomic status, highest educational attainment, and geographic location, to gain a more comprehensive and nuanced understanding of disparities, and lead to more effective interventions and policies that address the diverse needs and experiences of Indigenous peoples, recognizing their cultural strengths and promoting holistic health outcomes.
- Strengths-based and resilience-focused: Exploring the strengths and resilience within Indigenous communities, highlighting protective factors and successful health promotion strategies. This includes Indigenous leadership and governance in decision-making processes, policy development, and program implementation, or approaches that recognize the wisdom and guidance of Elders and traditional knowledge holders within communities. This approach aims to move beyond a deficit-based perspective, promote culturally appropriate and sustainable strategies, and empower Indigenous peoples to take an active role in their health and wellbeing.
- Indigenous data sovereignty and ethical research: A growing emphasis on Indigenous data sovereignty, which involves Indigenous control and ownership of data collected from the community. Researchers need to uphold ethical research practices that respect cultural protocols and protect the rights and privacy of Indigenous participants.

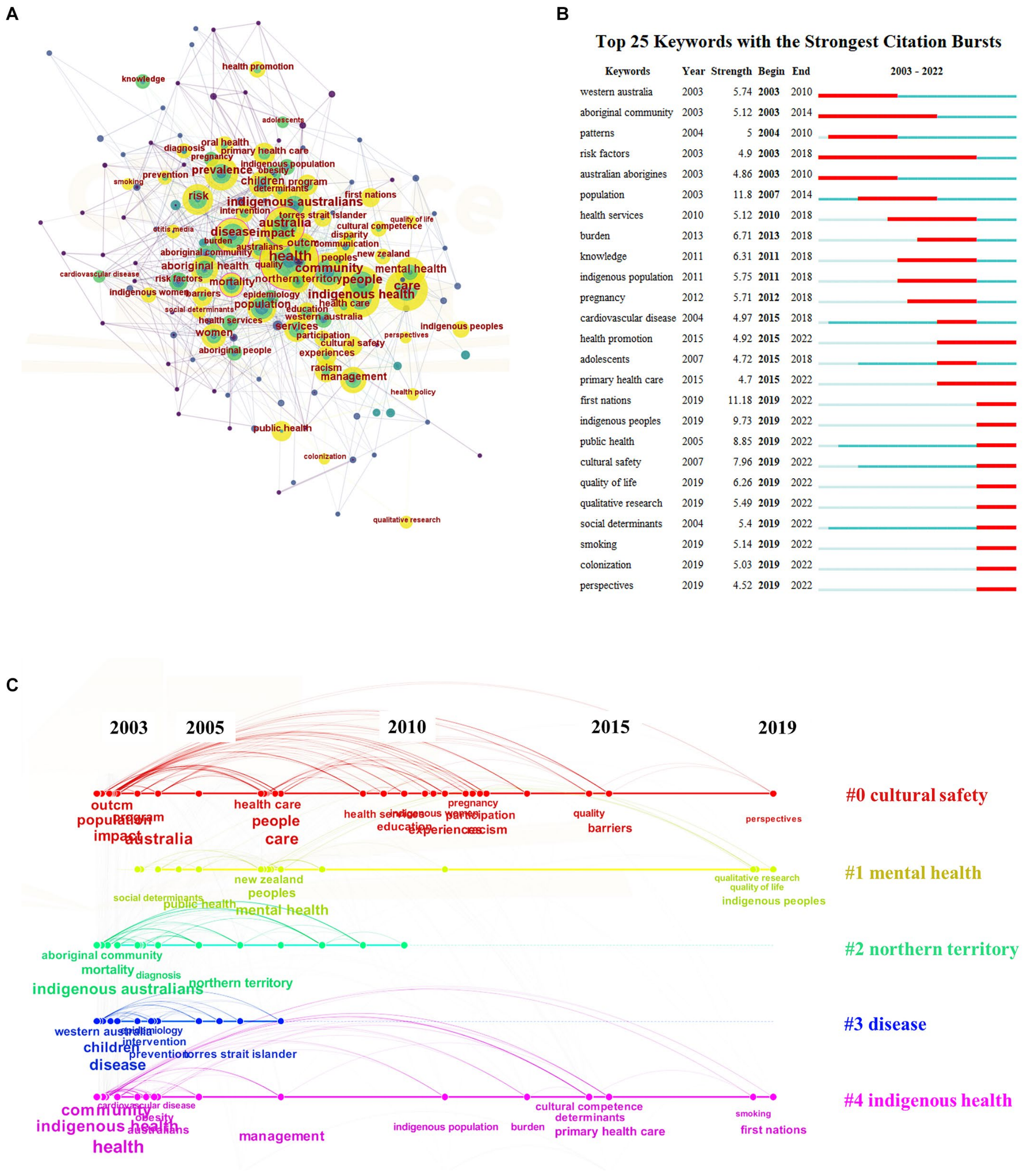
Emergent practices signify a shift toward more respectful, equitable, and effective research practices that honor Indigenous self-determination and prioritize the wellbeing of Indigenous peoples. An example is community-based participatory research that engages Indigenous communities as active partners in the research process,



References	Strength	Begin	End	2003 - 2022
Vos T, 2007	10.54	2007	2018	
Vos T, 2009	10.05	2011	2018	
Rowley KG, 2008	9.02	2008	2014	
Australian Bureau of Statistics, 2010	8.75	2011	2018	
Hunter E, 1993	8.17	2003	2014	
Australian Bureau of Statistics, 2006	7.91	2011	2014	
Henry BR, 2004	7.79	2007	2014	
Australian Bureau of Statistics, 2014	6.78	2015	2018	
Wood L, 2008	6.32	2011	2014	
Sanson-Fisher RW, 2006	6.32	2011	2014	

FIGURE 5
Visualization of the **(A)** document co-citation network, **(B)** top 10 documents with the strongest citation bursts, and **(C)** document co-citation clusters over time.

from defining research questions to interpreting results and implementing findings. This approach aims to address the historical power imbalances that have often characterized research involving Indigenous peoples and ensure that research is conducted in a way that aligns with the values, needs, and priorities of the community. Practices may include:



Top 25 Keywords with the Strongest Citation Bursts

Keywords	Year	Strength	Begin	End
western australia	2003	5.74	2003	2010
aboriginal community	2003	5.12	2003	2014
patterns	2004	5	2004	2010
risk factors	2003	4.9	2003	2018
australian aborigines	2003	4.86	2003	2010
population	2003	11.8	2007	2014
health services	2010	5.12	2010	2018
burden	2013	6.71	2013	2018
knowledge	2011	6.31	2011	2018
indigenous population	2011	5.75	2011	2018
pregnancy	2012	5.71	2012	2018
cardiovascular disease	2004	4.97	2015	2018
health promotion	2015	4.92	2015	2022
adolescents	2007	4.72	2015	2018
primary health care	2015	4.7	2015	2022
first nations	2019	11.18	2019	2022
indigenous peoples	2019	9.73	2019	2022
public health	2005	8.85	2019	2022
cultural safety	2007	7.96	2019	2022
quality of life	2019	6.26	2019	2022
qualitative research	2019	5.49	2019	2022
social determinants	2004	5.4	2019	2022
smoking	2019	5.14	2019	2022
colonization	2019	5.03	2019	2022
perspectives	2019	4.52	2019	2022

FIGURE 6
Visualization of the **(A)** keyword co-occurrence network, **(B)** keywords with the strongest citation bursts, and **(C)** keyword co-occurrence clusters over time. Year = is the earliest year of all publications being analyzed; strength = is an indicator related to the frequency of the keyword in a short time; begin and end = refers to the year of beginning and ending of the emergence of the keywords.

- Culturally responsive healthcare: Delving deeper into the concept of cultural safety in healthcare settings, examining strategies and interventions that promote culturally competent care delivery and addressing racism and discrimination within the healthcare system are warranted. This includes cultural awareness and competence training, communication collaborative decision-making, respect for cultural protocols and practices, workforce development, and addressing systemic issues and power imbalance.
- Indigenous-led research and community partnerships: There is increasing recognition of the importance of Indigenous-led research and community partnerships that foster respectful, equitable, and collaborative research practices and value Indigenous knowledge, perspectives, and self-determination. Research that involves close collaborations with Indigenous communities, ensuring their active participation in research design, implementation, and decision-making processes.

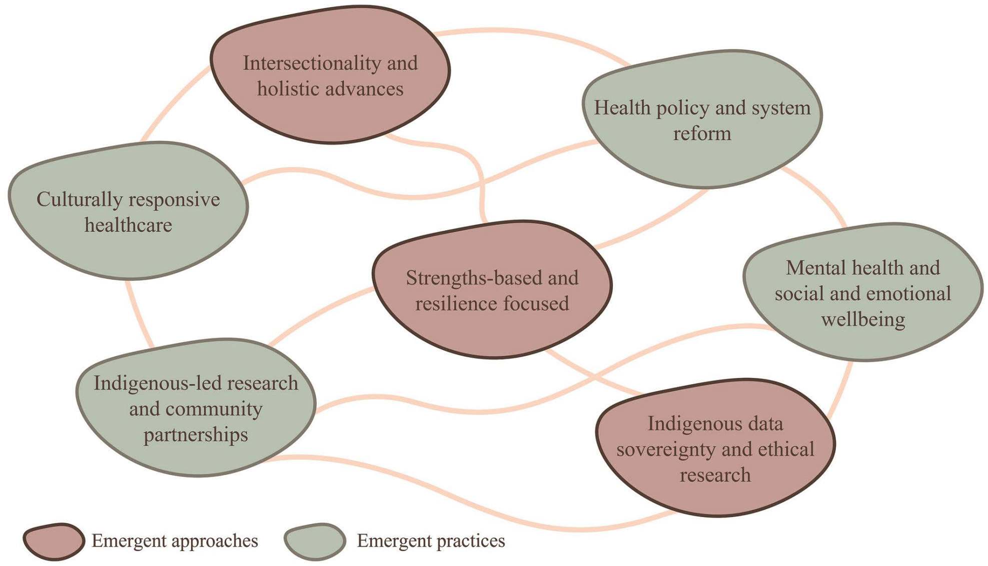


FIGURE 7
Emergent areas of research in Indigenous health and wellbeing research, as distributed by interconnected research approaches and research practices.

- Mental health and social and emotional wellbeing: While already a focus in earlier research, a deeper understanding of the specific mental health challenges faced by Indigenous peoples is culturally appropriate and culturally safe for prevention, early intervention, and treatment.
- Health policy and system reform: Continuing to inform health policies, and organizational and administrative procedures that advocate for system changes to reduce health disparities, improve access to quality healthcare services, and address the social determinants of health that contribute to Indigenous health inequities.

5 Strengths and limitations

To the best of our knowledge, this study is the first to systematically examine Indigenous health and wellbeing research using scientometric analysis and knowledge mapping - filling a significant gap in the existing literature. We review a substantial body of literature in a precise and objective manner, to provide insights for researchers engaged in the field. An exploration of the interrelationships among authors, institutions, countries/regions, journals, keywords, citations/co-citations, and references strengthens the robustness of our findings. Nonetheless, it is crucial to acknowledge the existence of limitations.

Firstly, we selected the WoSCC database as a comprehensive and respected platform for bibliometric analysis. Despite the standardization and consistency of publication records, there remains the potential that this approach is a non-exhaustive exploration of the literature (Kulkarni et al., 2009). Second, the influence of newly published articles might be undervalued due to their limited time for citation accrual. We acknowledge that document frequency is not a catalyst for change alone, so to mitigate this, a qualitative synthesis was applied in the analysis and interpretation of the results. Lastly, because software is used to conduct the analysis, there is a potential for errors or biases in our findings. For instance, journal names or research categorization might have changed over time, and there may be instances where two authors sharing the same name are repeatedly aggregated. All efforts to avoid these instances were applied before data analysis.

Despite not being within the scope of this study, we still acknowledge that distinguishing between research *on* Indigenous peoples and research *with* Indigenous peoples is critical. The latter necessitates a sense of relational accountability and research solely *on* Indigenous peoples often produces findings with diminished validity and reliability, and in the worst cases, it exacerbates the persistent overrepresentation of Indigenous populations facing significant challenges to their wellbeing.

6 Conclusion

This study represents a comprehensive scientometric analysis and knowledge mapping of Indigenous health and wellbeing research in Australia spanning 2003 to 2022. Our findings not only highlight a substantial and escalating focus of research within this field but also a change from population-level and data-driven studies towards community-based practices and applied research methodologies. Looking ahead in Indigenous health and wellbeing research, we can anticipate a growing emphasis on practices and methodologies that give precedence to forging robust partnerships *with* Indigenous communities. This shift away from conventional deficit mindsets, coupled with a heightened focus on recognizing cultural protocols and privacy considerations, will increasingly underpin the exploration of Indigenous individuals' experiences as they navigate their health and wellbeing. In an era of heightened awareness and significant investment in Indigenous health and wellbeing research in Australia, the imperative to articulate and prioritize outcomes for Indigenous peoples is more compelling than ever. By presenting this updated perspective based on two decades of published literature, this study not only provides an enhanced understanding of the knowledge in this field but also guides future research efforts.

Data availability statement

The original contributions presented in the study are included in the article/Supplementary material, further inquiries can be directed to the corresponding author.

Author contributions

MK: Conceptualization, data Curation, Formal Analysis, Investigation, Methodology. Visualisation, Writing – original draft, Writing – review & editing. KH: Conceptualization, Investigation, Writing – original draft, Writing – review & editing. PA: Conceptualization, Investigation, Writing – original draft, Writing – review & editing. CS: Investigation, Methodology, Resources, Writing – review & editing, Writing – original draft.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fsoc.2023.1290322/full#supplementary-material

References

AIHW. Indigenous health and wellbeing: Australian government (2022). Available at: https://www.aihw.gov.au/reports/australias-health/indigenous-health-and-wellbeing (accessed 07 June, 2023).

Anderson, I., Robson, B., Connolly, M., Al-Yaman, F., Bjertness, E., King, A., et al. (2016). Indigenous and tribal peoples' health (the lancet-Lowitja Institute global collaboration): a population study. *Lancet* 388, 131–157. doi: 10.1016/S0140-6736(16)00345-7

Arabena, K., Rowley, K., and MacLean, S. (2014). Building evidence about effective health promotion in Aboriginal and Torres Strait Islander communities. *Aust. J. Prim. Health* 20, 317–318. doi: 10.1071/PYv20n4_ED

Askew, D. A., Brady, K., Mukandi, B., Singh, D., Sinha, T., Brough, M., et al. (2020). Closing the gap between rhetoric and practice in strengths-based approaches to Indigenous public health: a qualitative study. *Aust. N. Z. J. Public Health* 44, 102–105. doi: 10.1111/1753-6405.12953

Australian Bureau of Statistics. *Estimates of Aboriginal and Torres Strait Islander Australians*. (2022) Canberra: Australian Bureau of Statistics.

Australian Government. National Aboriginal and Torres Strait Islander health plan 2021–2031. In: Department of health, Commonwealth of Australia (2021).

Australian Institute of Aboriginal and Torres Strait Islander Studies. *AIATSIS code of ethics for Aboriginal and Torres Strait Islander research*. Acton: AIATSIS (2020).

Australian Institute of Health and Welfare. *The health and welfare of Australia's Aboriginal and Torres Strait Islander peoples*. Canberra: AIHW (2015).

Basumatary, B., Verma, A. K., and Verma, M. K. (2022). Global research trends on aquaponics: a systematic review based on computational mapping. *Aquac. Int.* 31, 1115–1141. doi: 10.1007/s10499-022-01018-y

Bessarab, D., and Ng'andu, B. (2010). Yarning about yarning as a legitimate method in Indigenous research. *Int. J. Critic. Indig. Stud.* 3, 37–50. doi: 10.5204/ijcis.v3i1.57

Borgohain, D. J., Bhanage, D. A., Verma, M. K., and Pawar, A. V. (2021). Global research trends in augmented reality: scientometric mapping based on Scopus database. *Inform. Discov. Deliv.* 50, 387–403. doi: 10.1108/idd-08-2021-0081

Carson, B., Dunbar, T., Chenall, R. D., and Bailie, R. (2007) in *Social determinants of Indigenous health*. eds. B. Carson, T. Dunbar, R. D. Chenall and R. Bailie. *1st* ed (London: Routledge)

Chen, C. (2017). Science mapping: a systematic review of the literature. *J. Data Inform. Sci.* 2, 1–40. doi: 10.1515/jdis-2017-0006

Christou, A., Katzenellenbogen, J. M., and Thompson, S. C. (2010). Australia's national bowel cancer screening program: does it work for Indigenous Australians? *BMC Public Health* 10:373. doi: 10.1186/1471-2458-10-373

Clarivate. Journal citation reports (2022). Available at: https://jcr.clarivate.com/jcr/home (accessed 04 June, 2023).

Derrick, G. E., Hayen, A., Chapman, S., Haynes, A. S., Webster, B. M., and Anderson, I. (2012). A bibliometric analysis of research on Indigenous health in Australia, 1972-2008. *Aust. N. Z. J. Public Health* 36, 269–273. doi: 10.1111/j.1753-6405.2011.00806.x

Dudgeon, P., and Walker, R. (2015). Decolonising Australian psychology: discourses, strategies, and practice. *J. Soc. Polit. Psychol.* 3, 276–297. doi: 10.5964/jspp.v3i1.126

Egghe, L., Rousseau, R., and Van Hooydonk, G. (2000). Methods for accrediting publications to authors or countries: consequences for evaluation studies. *J. Am. Soc. Inform. Sci.* 51, 145–157. doi: 10.1002/(sici)1097-4571(2000)51:2<145::Aid-asi6>3.0.Co;2-9

Fonseca Bde, P., Sampaio, R. B., Fonseca, M. V., and Zicker, F. (2016). Co-authorship network analysis in health research: method and potential use. *Health Res. Policy Syst.* 14:34. doi: 10.1186/s12961-016-0104-5

Gatwiri, K., Rotumah, D., and Rix, E. A.-O. (2021). BlackLivesMatter in healthcare: racism and implications for health inequity among Aboriginal and Torres Strait Islander peoples in Australia. *Int. J. Environ. Res. Public Health* 18:4399. doi: 10.3390/ijerph18094399

Gibson, O., Lisy, K., Davy, C., Aromataris, E., Kite, E., Lockwood, C., et al. (2015). Enablers and barriers to the implementation of primary health care interventions for Indigenous people with chronic diseases: a systematic review. *Implement. Sci.* 10:71. doi: 10.1186/s13012-015-0261-x

Gracey, M., and King, M. (2009). Indigenous health part 1: determinants and disease patterns. *Lancet* 374, 65–75. doi: 10.1016/S0140-6736(09)60914-4

Gusenbauer, M. (2022). Search where you will find most: comparing the disciplinary coverage of 56 bibliographic databases. *Scientometrics* 127, 2683–2745. doi: 10.1007/s11192-022-04289-7

Harfield, S., Pearson, O., Morey, K., Kite, E., Canuto, K., Glover, K., et al. (2020). Assessing the quality of health research from an Indigenous perspective: the Aboriginal and Torres Strait Islander quality appraisal tool. *BMC Med. Res. Methodol.* 20:79. doi: 10.1186/s12874-020-00959-3

Jennings, W., Spurling, G., Shannon, B., Hayman, N., and Askew, D. (2021). Rapid review of five years of Aboriginal and Torres Strait Islander health research in Australia - persisting under-representation of urban populations. *Aust. N. Z. J. Public Health* 45, 53–58. doi: 10.1111/1753-6405.13072

Jiang, Y., Ritchie, B. W., and Benckendorff, P. (2017). Bibliometric visualisation: an application in tourism crisis and disaster management research. *Curr. Issue Tour.* 22, 1925–1957. doi: 10.1080/13683500.2017.1408574

Kastrin, A., and Hristovski, D. (2021). Scientometric analysis and knowledge mapping of literature-based discovery (1986–2020). *Scientometrics* 126, 1415–1451. doi: 10.1007/s11192-020-03811-z

Kennedy, M., Bennett, J., Maidment, S., Chamberlain, C., Booth, K., McGuffog, R., et al. (2022). Interrogating the intentions for Aboriginal and Torres Strait Islander health: a narrative review of research outputs since the introduction of closing the gap. *Med. J. Aust.* 217, 50–57. doi: 10.5694/mja2.51601

Kiatkoski Kim, M., Watkin Lui, F., Ah Mat, L., Cadet-James, Y., Bainbridge, R., and McCalman, J. (2020). Indigenous leadership in research in Australia. *J. High Educ. Policy M.* 43, 353–368. doi: 10.1080/1360080x.2020.1843220

Kinchin, I., McCalman, J., Bainbridge, R., Tsey, K., and Lui, F. W. (2017). Does Indigenous health research have impact? A systematic review of reviews. *Int. J. Equity Health* 16:52. doi: 10.1186/s12939-017-0548-4

King, M., Smith, A., and Gracey, M. (2009). Indigenous health part 2: the underlying causes of the health gap. *Lancet* 374, 76–85. doi: 10.1016/S0140-6736(09)60827-8

Kulkarni, A. V., Aziz, B., Shams, I., and Busse, J. W. (2009). Comparisons of citations in web of science, Scopus, and Google scholar for articles published in general medical journals. *JAMA* 302, 1092–1096. doi: 10.1001/jama.2009.1307

López-Pernas, S., Saqr, M., and Apiola, M. (2023). "Scientometrics: a concise introduction and a detailed methodology for mapping the scientific field of computing education research" in *Past, present and future of computing education research: A global perspective*. eds. M. Apiola, S. López-Pernas and M. Saqr (Cham: Springer International Publishing)

Lowitja Institute. *Transforming power: voices for generational change*. Canberra: Lowitja Institute (2022).

Lowitja Institute. Aboriginal and Torres Strait Islander health literature (2022). Available at: https://www.lowitja.org.au/page/research/lit-search (accessed 07 June, 2023).

Marmot, M. (2011). Social determinants and the health of Indigenous Australians. *Med. J. Aust.* 194, 512–513. doi: 10.5694/j.1326-5377.2011.tb03086.x

Marmot, M., Friel, S., Bell, R., Houweling, T., and Taylor, S. (2008). Closing the gap in a generation: health equity through action on the social determinants of health. *Public Health* 372, 1661–1669. doi: 10.1016/S0140-6736(08)61690-6

McGuffog, R., Bryant, J., Booth, K., Collis, F., Brown, A., Hughes, J. T., et al. (2023). Exploring the reported strengths and limitations of Aboriginal and Torres Strait Islander health research: a narrative review of intervention studies. *Int. J. Environ. Res. Public Health* 20:3993. doi: 10.3390/ijerph20053993

Menzies School of Health Research. *National Indigenous bowel screening pilot - final report* Darwin: Menzies School of Health Research (2020).

Mingers, J., and Leydesdorff, L. (2015). A review of theory and practice in scientometrics. *Eur. J. Oper. Res.* 246, 1–19. doi: 10.1016/j.ejor.2015.04.002

National Health and Medical Research Council. Road map 3 - a strategic framework for improving Aboriginal and Torres Strait Islander health through research. (2018). Available at: https://www.nhmrc.gov.au/about-us/publications/road-map-3-strategic-framework (accessed 21 June, 2023).

National Health and Medical Research Council. *Ethical conduct in research with Aboriginal and Torres Strait Islander peoples and communities: guidelines for researchers and stakeholders*. Canberra: Commonwealth of Australia (2018).

National Health and Medical Research Council. 2021 Aboriginal and Torres Strait Islander report card of achievements (2021). Available at: https://www.nhmrc.gov.au/health-advice/Aboriginal-and-torres-strait-Islander-health/Aboriginal-and-torres-strait-Islander-report-card-achievements-2021 (accessed 21 June, 2023).

National Indigenous Australians Agency. *Commonwealth closing the gap annual report 2022*. Canberra: National Indigenous Australians Agency (2022).

Noyons, E. C. M., Moed, H. F., and Van Raan, A. F. J. (1999). Integrating research performance analysis and science mapping. *Scientometrics* 46, 591–604. doi: 10.1007/bf02459614

Paradies, Y. (2016). Colonisation, racism and Indigenous health. *J. Popul. Res.* 33, 83–96. doi: 10.1007/s12546-016-9159-y

Rodriguez, A., and Laio, A. (2014). Machine learning. Clustering by fast search and find of density peaks. *Science* 344, 1492–1496. doi: 10.1126/science.1242072

Sanson-Fisher, R. W., Campbell, E. M., Perkins, J. J., Blunden, S. V., and Davis, B. B. (2006). Indigenous health research: a critical review of outputs over time. *Med. J. Aust.* 184, 502–505. doi: 10.5694/j.1326-5377.2006.tb00343.x

Singh, V. K., Singh, P., Karmakar, M., Leta, J., and Mayr, P. (2021). The journal coverage of Web of Science, Scopus and dimensions: a comparative analysis. *Scientometrics* 126, 5113–5142. doi: 10.1007/s11192-021-03948-5

Su, H.-N., and Lee, P.-C. (2010). Mapping knowledge structure by keyword co-occurrence: a first look at journal papers in technology foresight. *Scientometrics* 85, 65–79. doi: 10.1007/s11192-010-0259-8

The Group of Eight Ltd. Group of eight members (2022) Available at: https://go8.edu.au/ (accessed 07 June, 2023).

The Lancet (2020). Medicine and medical science: black lives must matter more. *Lancet* 395:1813. doi: 10.1016/S0140-6736(20)31353-2

Trujillo, C. M., and Long, T. M. (2018). Document co-citation analysis to enhance transdisciplinary research. *Sci. Adv.* 4:e1701130. doi: 10.1126/sciadv.1701130

Vos, T., Barker, B., Begg, S., Stanley, L., and Lopez, A. D. (2009). Burden of disease and injury in Aboriginal and Torres Strait Islander peoples: the Indigenous health gap. *Int. J. Epidemiol.* 38, 470–477. doi: 10.1093/ije/dyn240

Wang, W., and Lu, C. (2019). Visualization analysis of big data research based on Citespace. *Soft. Comput.* 24, 8173–8186. doi: 10.1007/s00500-019-04384-7

Whiting, C., Cavers, S., Bassendowski, S., and Petrucka, P. (2018). Using two-eyed seeing to explore interagency collaboration. *Can. J. Nurs. Res.* 50, 133–144. doi: 10.1177/0844562118766176

Xia, D., Yao, R., Wang, S., Chen, G., and Wang, Y. (2021). Mapping trends and hotspots regarding clinical research on COVID-19: a bibliometric analysis of global research. *Front. Public Health* 9:713487. doi: 10.3389/fpubh.2021.713487

You, Y., Wang, D., Wang, Y., Li, Z., and Ma, X. (2021). A bird's-eye view of exercise intervention in treating depression among teenagers in the last 20 years: a bibliometric study and visualization analysis. *Front. Psych.* 12:661108. doi: 10.3389/fpsyt.2021.661108

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Colonial drivers and cultural protectors of brain health among Indigenous peoples internationally

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Despite relatively higher rates of dementia among Indigenous populations internationally, research into drivers of disparities in brain health and cognitive function has tended to focus on modifiable risk factors over cultural understandings and contextual determinants. By seeking to characterize social and cultural factors that shape brain health and cognition in Indigenous populations, this mini scoping review expands prevailing schools of thought to include Indigenous knowledge systems. This reveals important gaps in culturally aligned care. It also reclaims horizons for research important to Indigenous Peoples that have garnered diminished attention in biomedical approaches. Twenty-three sources were included for data extraction. This synthesis of 23 sources includes health communication about dementia, health provider knowledge about Indigenous health, culturally relevant screening and assessment tools, and culturally grounded care models. Much of the focus is currently still on modifiable risk factors that reside at individual factors, whereas attention to wider social factors that impact populations is needed, as stressors through isolation, discrimination, and unequal care are widely reported. Going forward, identifying structural barriers to living well and recognizing the importance of connection to culture will benefit both Indigenous and non-Indigenous understandings of brain health.

KEYWORDS

Indigenous, brain health, cognition, dementia, Alzheimer's disease, social, culture, scoping review

1 Introduction

Colonization and ongoing colonial systems and structures continue to impact the health and wellness of Indigenous Peoples. Growing evidence indicates that Indigenous people experience higher risk of cognitive impairment, including dementia. Walker and colleagues (1) note how, in the international context, rates of Alzheimer's disease and related dementias (ADRDs) appear to be rising more rapidly in Indigenous compared to non-Indigenous populations. This disparity is concerning given that the proportion of people with an ADRD diagnosis across all populations is projected to more than double over the next 25 years (2, 3), meaning a greater proportion of

Indigenous Peoples of all ages may develop cognitive impairment. For instance, the prevalence of dementia in First Nations in the Canadian province of Alberta in 2009 was 7.5 per 1,000 compared to non-First Nations people at 5.6 per 1,000 (4). Moreover, evidence suggests that onset of dementia occurs at a younger age and that rates are rising more quickly for First Nations individuals, disproportionately affecting males (4).

While epidemiological research sheds light on prevalence and incidence rates of dementia among Indigenous populations ranging between 0.5 to 20% (5), explanatory frameworks for these are not fully developed. Little is known about social factors that may affect brain health and cognition that are specific to Indigenous communities with shared experiences of colonial oppression (6). Determinants of health known to play an important role in shaping Indigenous Peoples' experiences of health more generally include socioeconomic status; history of colonization; systemic racism; cultural perspectives and practices; healthcare access; and importantly, safety. Indigenous cultural perspectives on cognitive impairment and the aging process have garnered increasing attention (7–9), while other researchers focus on culturally grounded caregiving models (10) and cross-cultural communication and patient health education (11). While growing attention is on social and cultural factors that influence Indigenous Peoples' experiences of cognitive health, more research is needed. In one recent integrative review of literature on Indigenous perspectives on cognitive impairment and dementia, Racine and colleagues (12) concluded that a dearth of published information exists about Indigenous perspectives on aging or experiences of cognitive impairment and dementia. Racine et al. (12) urged researchers to more carefully examine the role of Indigenous knowledge, culture, and traditions in this domain, in order to decolonize dementia care.

We are a team of Indigenous and allied scholars and research associates located across what is now known as Canada, all affiliated with the Canadian Consortium on Neurodegeneration in Aging (CCNA). With several Indigenous co-authors, we address an erasure bias in Western scientific approaches that often minimize consideration of social and cultural factors to health and disease outcomes. The wider Indigenous cognitive health research team in the CCNA is organized around priority focus areas of advancing knowledge around the appropriate diagnosis of brain-related conditions in Indigenous populations, improved care for Indigenous Peoples with such conditions, and better understanding social and cultural realities that interface with brain health and cognitive function. Our work is generally community-based, and from this location this scoping review was an opportunity to bring together our collective learnings and knowledge. The objective of our scoping review is to characterize social and cultural factors that affect brain health and cognition with Indigenous populations, with particular interest in community understandings of these.

2 Methods

Scoping reviews are appropriately and commonly used to identify gaps in knowledge or explore the nature of published knowledge on a topic (13). Following Arksey & O'Malley's (14) six-part scoping review methodology and the PRISMA reporting standards for scoping reviews, we conducted a search of CINAHL Plus, Ovid Medline, EMBASE, SocINDEX, Scopus, and the Canadian Research Index from January 26 to February 1, 2022.

2.1 Search strategy

The search strategy was designed to retrieve social and cultural drivers of brain health and cognition among Indigenous Peoples. The team consulted with health librarians and experts in brain health and Indigenous health to supplement our own expertise in these areas. Recalling that the main shared experience of Indigenous Peoples internationally is colonization, our search focused on dual aspects of social worlds shaped by settler colonial systems: Indigenous cultures and their disruption by colonization. On one hand, this includes cultural practices, perspectives, and knowledge around the brain and its health, and on the other hand social environments characterized by oppression such as stigma, racism, marginalization, and resource disparities. Key search terms included those related to these social factors, brain and cognitive health, and Indigenous identities. No geographic restriction was imposed, nor a limit on the year of publication. Supplementary Table 1 displays key search terms.

2.2 Source selection

Sources needed to meet the following criteria for inclusion: (1) include an Indigenous population ('Indigenous' as defined by the United Nations Declaration on the Rights of Indigenous Peoples) (15); (2) include findings focused on brain health or cognition (including dementia or other neurodegenerative disorders); (3) discuss social or cultural factors (e.g., discrimination, socioeconomic disparities, social mobility, language) that may drive brain or cognitive health outcomes; and (4) be written in English. Two reviewers (SSC and JAF) independently conducted an initial screen of studies exported from the searches to the review management platform COVIDENCE, in the first stage reviewing only titles and abstracts for eligible inclusion. This was followed by a full-text scan of documents for eligibility conducted by the same reviewers. Sources were excluded if they were: (1) textbooks or handbooks (i.e., only providing a summary of existing work); (2) systematic and scoping review papers (i.e., not reporting original source material); (3) abstracts only; (4) animal research; (5) not Indigenous-focused (i.e., Indigenous Peoples appearing only as a comparator group among several); (6) no reference to social or cultural factors driving factors of brain health or cognition; or (7) outcomes were mental health disorders as defined by the Diagnostic and Statistical Manual of Mental Disorders (DSM-V-TR) due to our focus on brain health or cognition, not mental health. At this stage to identify further relevance, we also accessed titles and abstracts of sources that may have been missed in the original searches, reference lists from included studies (reviewed by SSC), with potentially relevant sources also assessed for title and abstract. A full-text review was then conducted by both reviewers (SSC and JAF). Individual results were compared and consensus of final inclusion of sources reached in discussion with the full project team.

2.3 Data extraction and analysis

Two reviewers (SSC and JAF) used a common data extraction form in Excel. Data fields included, when applicable: author(s), year of publication, article type, design, study date, sample, population demographics (age, sex/gender), details on Indigenous population included, geographic location, project collaborations (e.g., with community organizations), study objectives, social or cultural factors discussed, brain health or cognition outcome(s) examined, presentation of research rigor, findings, recommendations, and reported limitations. Special attention was paid to sex- or gender-reported data during extraction, reporting where relevant. Additionally, we searched each paper using the terms 'sex', 'gender', 'male', 'female', 'men', 'women', 'man', and 'woman' to ensure that no pertinent data was overlooked. This was undertaken to anticipate meaningful sex- or gender-based observations in or across included studies.

The wider author team then conducted a series of three consensus-building discussions based on a Nominal Group Technique (NGT), a structured small-group discussion method to reach consensus through brainstorming to identify and interpret emergent themes (16). Bringing together perspectives from our CCNA Indigenous cognitive health team, we generated an exhaustive list of observed categories in the data, then discussed what resonated and what was surprising in order to structure results presented here. Lead authors (RH and PR) then worked with transcripts from NGT discussions to discuss findings according to the Knowledge-Attitudes-Behavior (KAB) model (adapted from Knowledge-Attitudes-Practices [KAP] Model, where "practices" may be more narrowly medical than "behaviours"). Research suggests that the KAB model is often helpful for health behavior change theory (17). This offers a framework for organizing behavior change theory for health audiences, helping to orient the implications of findings for our colleagues in clinical practice, planning, and education. We take some liberty with the KAB model, as the inter-cultural nature of data presented here means that we treat "knowledge" within a constructivist lens (18), and "attitudes" may also reflect values or cultural principles.

3 Results

The search of the combined databases yielded 661 initial results. Sources were imported to COVIDENCE, where 131 were identified as duplicates and removed. From there, 530 underwent abstract and title screening, where 444 were excluded (Supplementary Figure 1). Eighty-six articles underwent full-text review, leading to an additional 72 being further excluded. Ultimately, 15 sources were identified as relevant from the initial database search. An additional 14 sources were identified as possibly relevant through hand-searching reference lists of eligible sources. These additional 14 underwent title, abstract and full-text screening, where eight were identified as eligible. A total of 23 articles were included for data extraction. Of these, the majority are in Australia ($n=8$), followed by United States ($n=5$), Canada ($n=4$), New Zealand ($n=3$), Guam ($n=2$), and Malayasia ($n=1$). The majority of articles included in the scoping review are quantitative in nature ($n=8$), followed by commentaries ($n=7$), qualitative methods ($n=5$), and mixed methods ($n=3$). Supplementary Figure 1 displays the PRISMA flow diagram.

Given the cultural and social diversity of Indigenous groups internationally, comparative differences are not suitable to link to explanatory frameworks here, though commonalities of primary interest. For the purpose of reporting findings, identified themes are broadly divided into categories of social versus cultural drivers impacting brain health and cognition. Key areas examined in this review include health communication about dementia, health provider knowledge about Indigenous health, culturally relevant dementia-screening and assessment tools, and culturally grounded care models. Important to note is a lack of sex and gender considerations found in the literature, suggesting more research is required (19).

3.1 Social drivers impacting brain health and cognition for Indigenous peoples

Fifteen articles identified social factors that affect brain health or cognition with Indigenous populations (10, 20–33). Studies acknowledged that population health research has historically focused on modifiable risk factors which include smoking, physical inactivity, and low educational attainment associated with dementia (24, 26, 30). Some note that the literature has begun to describe complexities in modifiable risk factors, such as education and occupation (21, 25, 27, 28). For instance, Radford et al. (28) compared skilled with unskilled employment (e.g., labor or entry-level jobs), identifying that many patients diagnosed with dementia had an occupational history of unskilled work. Three articles addressed low income as a risk factor for brain or cognitive health outcomes; however, the relationship was not explored as extensively as educational attainment (24, 26, 30). The inter-related connections between educational attainment, employment and other modifiable risk factors for dementia remains largely unexplored, especially within the context of Indigenous Peoples' health.

Several studies identified the impacts of colonization (10), institutional racism (31) and historical and ongoing trauma (9) as drivers of brain health or cognition. Radford et al. (28) measured cultural experiences and informal education using a scale entitled Retrospective Indigenous Childhood Enrichment (RICE). The RICE tool was developed to approximate cognitive stimulation throughout childhood outside of school in the Aboriginal Australian population. The paper also addresses childhood trauma as measured by the childhood trauma questionnaire (CTQ), a survey quantifying adverse childhood experiences ranging from separation from family to socioeconomic status. The CTQ survey indicated an association between childhood stress and late-life dementia diagnosis in Aboriginal Australian populations (28). Additional work to identify links between early childhood experiences and cognitive decline and dementia in adulthood are necessary to further understand the relationship between these outcomes.

Social impact and climate change were presented as a factor influencing Indigenous brain health in many papers. Data from Guam measured cycad exposure during traditional food gathering, preparation, and consumption of an Indigenous food called fadang, exploring this as a risk factor for neurodegenerative diseases (34). Although the association between fadang consumption was inconclusive, the study probes population-specific lifestyle factors such as exposure to eating fadang in young adulthood, highlighting potential sex differences to be further explored. Southern Inuit research participants from Eastern Canada identified dementia prevention as a small piece in the broader category of healthy aging (9). Participants in this study advocated for more access to foods associated with traditional diets in the region, such as wild game and locally-grown berries.

Disruption in the food chain due to environmental contamination and colonization has interrupted access to traditional foods, simultaneously interrupting opportunities to engage with the land – which was considered in the study community to be protective of mental wellness (9). A commentary by Farugia et al. (22) linked access to traditional food and Indigenous brain health to extreme heat. The authors explored the link between bushfires, access to care in rural environments, and the exacerbation of neurodegeneration among dementia patients exposed to extreme heat. Similarly, other work has highlighted that urbanization and climate change, including issues of urban migration, family structure, and community involvement, can have a lasting impact on brain health and dementia (10).

3.2 Cultural drivers impacting brain health and cognition for indigenous peoples

Eleven articles identified cultural factors that affect brain health or cognition in Indigenous populations (9, 10, 22, 25, 34–40). Some sources address physiological and functional symptoms resulting from the differences and tensions that lie between Indigenous perspectives and biomedical approaches (9, 37, 39). Pace (9) identified the increasing difficulty of being able to live in traditional ways on the land in one's own territory. This was seen to create inequity in who may receive needed services and thereby be able to age "in place," which the study authors note is often recommended as best practice. Additionally, findings indicate that place and culture foster identity maintenance, and are protective factors against cognitive decline (9). Similarly, Manly and Espino (39) outlined how culture shapes cognitive function and affects potential presentation of brain aging clinically, impacting efforts to diagnose and manage cognitive decline across groups. Both studies disrupt conventional western and biomedical parameters defining neurodegenerative conditions of the brain by highlighting protective aspects of cultural connection. This highlights the need for flexible and personalized diagnostic approaches inclusive of linguistic and epistemological differences across ethno-cultural groups (38).

Cultural understandings of dementia were noted to shape expectations of healthcare and influence preferences for informal caregiving in community networks over accessing formal caregiving services (10). Some sources outlined efforts to embed cultural norms within Western healthcare systems, while others emphasized that not all Indigenous Peoples may desire to access Westernized care as their conditions progress (22, 25, 35, 38). These studies note reasons anchored in cultural perceptions of the purpose of care itself for brain aging. One study illustrated this by contrasting Hawai'ian expectations that care focus on personalized support defined by an individual and their family or community, where a Western healthcare approach tends to emphasize mitigating all disease symptoms in the individual (35).

Most studies used sex and gender interchangeably and did not use these variables in a meaningful way. On study looked at differences in exposure to cycad and dementia risk by sex (34). Few papers (20, 28–30) looked at risk or predictive factors of dementia or cognitive decline based on sex or gender. This indicates a need for future research to intentionally include sex and gender analyses when undertaking primary research with Indigenous people living with dementia.

4 Discussion

Our review has highlighted the presence of structural drivers, both social and cultural, of Indigenous brain health and cognition. Much discourse in current evidence is shifting to focus on modifiable risk factors (i.e., childhood stressors). This emphasis nevertheless tends to neglect the influence of colonial structures on the autonomy of Indigenous Peoples to control these modifiable risk factors, such as income status or ability to remain in home territory or community. Instead, community-aligned perspectives reviewed here suggest critical attention be directed at systemic, structural, and social determinants of health, rather than individual ones. Large-scale policy and legislative changes to enshrine self-governance and self-determination in health and social care systems will be an important future change to create these solutions. Stites et al. (41) have established a framework for gathering structural and social determinants of health in ADRDs research which recognizes the importance of cultural values and perspectives. This may offer a crucial opportunity to identify structural barriers in order to decolonize and dismantle them.

The path to decolonizing brain health supports and care for Indigenous Peoples, therefore, includes emphasis on strengths of connection to culture. This includes cultural perspectives on brain aging encountered by many on our research team, who expected from our own community-based experiences to find in this review Indigenous community hesitation to defining brain aging simply in terms of disease and decline in favor of treating it as a natural component of the life course, where those exhibiting cognitive challenges may retain place and purpose in their social worlds. Embedding understanding of culture and social context will benefit all populations, Indigenous and non-Indigenous. For example, the Educating for Equity Care Framework developed for physicians providing care to Indigenous Peoples with diabetes, highlights colonization as a major health inequity and poses that while respecting diverse perspectives and experiences, culture "is a facilitator of the clinical relationship and patient capacity" (42, 43). Distinctions also need to be considered to avoid pan-Indigenizing populations. Understanding how cultural understandings of dementia and aging can differ among communities is imperative when developing appropriate responses. This approach is in alignment with the intention of Shkaabe Makwa (44), a team focused on driving culturally relevant system initiatives to achieve health equity and community wellness among First Nations, Métis, and Inuit populations. In addition, given that brain aging and cognition share many determinants with mental health outcomes, such as life stressors as risk factors and social connectedness as protective, the First Nations Mental Wellness Continuum Framework provides guidance and direction for how to appropriately embed cultural, social, and system factors (45).

In summary, listening to the experiences and asks of community members takes time, compassion, and meaningful relationship development. Sources reviewed here emphasize that it is imperative that dementia care be culturally safe and designed in collaboration with Indigenous Peoples themselves (19). Future research would do well to acknowledge that health and wellness extend beyond the biomedical model. This will aid in facilitating pathways that wholistically address the impact of bio-psycho-socio-cultural factors, though gaps in bridging evidence between these domains for improved care approaches clearly persist. This requires a willingness to learn and critically understand Indigenous values and principles broadly and locally, knowing that Indigenous-centered brain healthcare cannot be essentialized or reduced into universalized models of care. Studies discussed here highlight the importance of culture, family, community, respect, and trauma-informed care in ensuring that Indigenous Peoples' brain health and cognition are appropriately supported (46). Finally, the Truth and Reconciliation Commission of Canada: Calls to Action and the United Nations Declaration on the Rights of Indigenous Peoples (UNDRIP) are guides that draw attention to the inherent right for self-determination in healthcare, and as such are tools for affirming person, family, and community-centered.

Author contributions

RH: Writing – original draft, Writing – review & editing. JF: Writing – original draft, Writing – review & editing. SC: Writing – original draft, Writing – review & editing. AC-B: Writing – original draft, Writing – review & editing. AL: Writing – original draft, Writing – review & editing. JW: Writing – original draft, Writing – review & editing. LZ: Writing – original draft, Writing – review & editing PR: Writing – original draft, Writing – review & editing.

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Conflict of interest

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2024.1346753/full#supplementary-material

References

1. Walker J, Spiro G, Loewen K, Jacklin K. Alzheimer's disease and related dementia in indigenous populations: a systematic review of risk factors. *J Alzheimer's disease: JAD*. (2020) 78:1439–51. doi: 10.3233/JAD-200704

2. Alzheimer Society of Canada. Race and dementia. (2020). Available at: https://alzheimer.ca/en/take-action/change-minds/race-dementia [Accessed September 1, 2023].

3. Public Health Agency of Canada. A dementia strategy for Canada: together we aspire. (2019). Available at: https://www.canada.ca/en/public-health/services/publications/diseases-conditions/dementia-strategy.html [Accessed September 3, 2023].

4. Jacklin KM, Walker JD, Shawande M. The emergence of dementia as a health concern among first nations populations in Alberta. *Canada Canadian J Public Heal*. (2012) 104:e39–44. doi: 10.1007/BF03405652

5. Warren LA, Shi Q, Young K, Borenstein A, Martiniuk A. Prevalence and incidence of dementia among indigenous populations: a systematic review. *Int Psychogeriatr*. (2015) 27:1959–70. doi: 10.1017/S1041610215000861

6. King M, Smith A, Gracey M. Indigenous health part 2: the underlying causes of the health gap. *Lancet (British Edition)*. (2009) 374:76–85. doi: 10.1016/S0140-6736(09)60827-8

7. Hulko W, Camille E, Antifeau E, Arnouse M, Bachynski N, Taylor D. Views of first nation elders on memory loss and memory care in later life. *J Cross Cult Gerontol*. (2010) 25:317–42. doi: 10.1007/s10823-010-9123-9

8. Johnston K, Preston R, Strivens E, Qaloewai S, Larkins S. Understandings of dementia in low- and middle-income countries and amongst indigenous peoples: a systematic review and qualitative meta-synthesis. *Aging Ment Health*. (2020) 24:1183–95. doi: 10.1080/13607863.2019.1606891

9. Pace J. "Place-ing" dementia prevention and care in NunatuKavut. *Labrador Canadian J Aging*. (2020) 39:247–62. doi: 10.1017/S0714980819000576

10. Jacklin K, Pace JE, Warry W. Informal dementia caregiving among indigenous communities in Ontario. *Canada Care Manag J*. (2015) 16:106–20. doi: 10.1891/1521-0987.16.2.106

11. Webkamigad S, Rowe R, Peltier S, Froehlich CA, McGilton KS, et al. Identifying and understanding the health and social care needs of indigenous older adults with multiple chronic conditions and their caregivers: a scoping review. *BMC Geriatr*. (2020) 20:145–5. doi: 10.1186/s12877-020-01552-5

12. Racine L, Johnson L, Fowler-Kerry S. An integrative review of empirical literature on indigenous cognitive impairment and dementia. *J Adv Nurs*. (2021) 77:1155–71. doi: 10.1111/jan.14665

13. Munn Z, Peters MDJ, Stern C, Tuganaru C, McArthur A, Aromataris E. Systematic review or scoping review? Guidance for authors when choosing between a systematic or scoping review approach. *BMC Med Res Methodol*. (2018) 18:143. doi: 10.1186/s12874-018-0611-x

14. Arksey H, O'Malley L. Scoping studies: towards a methodological framework. *Int J Soc Res Method*. (2005) 8:19–32. doi: 10.1080/1364557032000119616

15. United Nations Declaration on the Rights of Indigenous Peoples. (2008). Available at: http://www.un.org/esa/socdev/unpfii/documents/DRIPS_en.pdf. [Accessed September 3, 2023].

16. Centers for Disease Control and Prevention. Gaining consensus among stakeholders through nominal group technique. (2018). Available at: https://www.cdc.gov/healthyyouth/evaluation/pdf/brief7.pdf. [Accessed May 15, 2023].

17. Schrader PG, Lawless KA. The knowledge, attitudes, & behaviors approach how to evaluate performance and learning in complex environments. *Perform Improvt (Int Society for Perform Improv)*. (2004) 43:8–15. doi: 10.1002/pfi.4140430905

18. Charmaz K. The power of constructivist grounded theory for critical inquiry. *Qual Inq*. (2017) 23:34–45. doi: 10.1177/1077800416657105

19. Chakanyuka C, Bacsu JDR, DesRoches A, Dame J, Carrier L, Symenuk P, et al. Indigenous-specific cultural safety within health and dementia care: a scoping review of reviews. *Soc Sci Med*. (2022) 293:114658. doi: 10.1016/j.socscimed.2021.114658

20. de Kok DS, Teh RO, Pillai A, Connolly MJ, Wilkinson TJ, Jacobs R, et al. What is the relationship between visual impairment and cognitive function in octogenarians? *N Z Med J*. (2017) 130:33–47.

21. Dyall L. Dementia: continuation of health and ethnic inequalities in New Zealand. *N Z Med J*. (2014) 127:68–80.

22. Farugia TL, Cuni-Lopez C, White AR. Potential impacts of extreme heat and bushfires on dementia. *J Alzheimer's disease: JAD*. (2021) 79:969–78. doi: 10.3233/JAD-201388

23. Garrett MD, Baldridge D, Benson W, Crowder J, Aldrich N. Mental health disorders among an invisible minority: depression and dementia among American Indian and Alaska native elders. *Gerontologist*. (2015) 55:227–36. doi: 10.1093/geront/gnu181

24. MacDonald JP, Barnes DE, Middleton LE. Implications of risk factors for Alzheimer's disease in Canada's indigenous population. *Canadian Geriatrics J*. (2015) 18:152–8. doi: 10.5770/cgj.18.159

25. Pollitt PA. The problem of dementia in Australian aboriginal and Torres Strait islander communities: an overview. *Int J Geriatr Psychiatry*. (1997) 12:155–63. doi: 10.1002/(SICI)1099-1166(199702)12:2<155::AID-GPS582>3.0.CO;2-M

26. Puun BI, Othman Z, Drahman I. Dementia among elderly Melanau: a community survey of an indigenous people in East Malaysia. *Intern Med J*. (2014) 21:468–71.

27. Radford K, Delbaere K, Draper B, Mack HA, Daylight G, Cumming R, et al. Childhood stress and adversity is associated with late-life dementia in aboriginal Australians. *Am J Geriatr Psychiatry*. (2017) 25:1097–106. doi: 10.1016/j.jagp.2017.05.008

28. Radford K, Lavrencic LM, Delbaere K, Draper B, Cumming R, Daylight G, et al. Factors associated with the high prevalence of dementia in older aboriginal Australians. *J Alzheimers Dis*. (2019) 70:S75–85. doi: 10.3233/JAD-180573

29. Reed DM, Brody JA. Amyotrophic lateral sclerosis and parkinsonism dementia on Guam, 1945-1972. 1. Descriptive epidemiology. *Am J Epidemiol*. (1975) 101:287–301. doi: 10.1093/oxfordjournals.aje.a112097

30. Smith K, Flicker L, Dwyer A, Atkinson D, Almeida OP, Lautenschlager NT, et al. Factors associated with dementia in aboriginal Australians. *Aust N Z J Psychiatry*. (2010) 44:888–93. doi: 10.3109/00048674.2010.491816

31. Shepherd SM, Ogloff JRP, Shea D, Pfeifer JE, Paradies Y. Aboriginal prisoners and cognitive impairment: the impact of dual disadvantage on social and emotional wellbeing. *J Intellect Disabil Res*. (2017) 61:385–97. doi: 10.1111/jir.12357

32. Glasson EJ, Sullivan SG, Hussain R, Bittles AH. An assessment of intellectual disability among aboriginal Australians. *J Intellect Disabil Res*. (2005) 49:626–34. doi: 10.1111/j.1365-2788.2005.00722.x

33. Lo Giudice D, Smith K, Fenner S, Hyde Z, Atkinson D, Skeaf L, et al. Incidence and predictors of cognitive impairment and dementia in aboriginal Australians: a follow-up study of 5 years. *Alzheimers Dement*. (2016) 12:252–61. doi: 10.1016/j.jalz.2015.01.009

34. Borenstein AR, Mortimer JA, Schofield E, Wu Y, Salmon DP, Gamst A, et al. Cycad exposure and risk of dementia, MCI, and PDC in the Chamorro population of Guam. *Neurology*. (2007) 68:1764–71. doi: 10.1212/01.wnl.0000262027.31623.b2

35. Braun KL, Browne CV. Perceptions of dementia, caregiving, and help seeking among Asian and Pacific islander American. *Health Soc Work*. (1998) 23:262–74. doi: 10.1093/hsw/23.4.262

36. Cullum S, Varghese C, Coomarasamy C, Whittington R, Hadfield L, Rajay A, et al. Predictors of mortality in Māori, Pacific Island, and European patients diagnosed with dementia at a New Zealand memory service. *Int J Geriatr Psychiatry*. (2020) 35:516–24. doi: 10.1002/gps.5266

37. Laditka JN, Laditka SB, Liu R, Price AE, Wu B, Friedman DB, et al. Older adults' concerns about cognitive health: commonalities and differences among six United States ethnic groups. *Ageing & Society*. (2011) 31:1202–28. doi: 10.1017/S0144686X10001273

38. Laditka SB, Laditka JN, Liu R, Price AE, Friedman DB, Wu B, et al. How do older people describe others with cognitive impairment? A multiethnic study in the United States. *Ageing & Society*. (2013) 33:369–92. doi: 10.1017/S0144686X11001255

39. Manly JJ, Espino DV. Cultural influences on dementia recognition and management. *Clin Geriatr Med*. (2004) 20:93–119. doi: 10.1016/j.cger.2003.10.004

40. Seto J, Davis J, Taira DA. Examining the association between different aspects of socioeconomic status, race, and disability in Hawaii. *J Racial Ethn Health Disparities*. (2018) 5:1247–53. doi: 10.1007/s40615-018-0471-4

41. Stites SD, Midgett S, Mechanic-Hamilton D, Zuelsdorff M, Glover CM, Marquez DX, et al. Establishing a framework for gathering structural and social determinants of health in Alzheimer's disease research centers. *Gerontologist*. (2022) 62:694–703. doi: 10.1093/geront/gnab182

42. Crowshoe LL, Henderson R, Jacklin K, Calam B, Walker L, Green ME. Educating for equity care framework: addressing social barriers of indigenous patients with type 2 diabetes. *Can Fam Physician*. (2019) 65:25–33.

43. World Alzheimer Report. Chapter 20. Sex, gender and cultural factors. (2021). Available at: https://www.alzint.org/u/World-Alzheimer-Report-2021-Chapter-20.pdf [Accessed November 6, 2023)].

44. CAMH. Shkaabe-makwa. (2023). Available at: https://www.camh.ca/en/driving-change/shkaabe-makwa. [Accessed September 25, 2023].

45. Thunderbird Partnership Foundation. First nations mental wellness continuum framework. (2022). Available at: https://thunderbirdpf.org/fnmwc/ [Accessed November 1, 2023].

46. LoGiudice D., Hughson J., Douglas H., Wenitong M., Belfrage M. *Culturally safe, trauma informed approach to cognitive impairment and dementia in older aboriginal and Torres Strait islander people [review of culturally safe, trauma informed approach to cognitive impairment and dementia in older aboriginal and Torres Strait islander people]*. (2022). 505–511. New York: New York Times Company.

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Small for gestational age and anthropometric body composition from early childhood to adulthood: the Aboriginal Birth Cohort study

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Background: In Australia the estimated rate of small for gestational age (SGA) births is 9% among non-Indigenous births compared to 14% among Aboriginal and Torres Strait Islanders. There is limited research investigating the effect of being born SGA on body composition later in life in Indigenous Australians.

Methods: Using data from the Aboriginal Birth Cohort longitudinal study, we compared the body composition of those born SGA to non-SGA by analysing anthropometric measures (height, weight, waist circumference, fat percentage [FAT%], body mass index [BMI], waist-to-height ratio, and A body shape index [ABSI]) collected at four follow-up periods (from childhood to adult). For cross-sectional analyses, linear regression models were employed to assess factors associated with anthropometric measures. For longitudinal analyses linear mixed models were employed to assess differences in anthropometric measures among SGA versus non-SGA individuals while adjusting for repeated measures.

Results: The analytic baseline cohort were those who participated in Wave 2 ($n = 570$). In cross-sectional analyses, across all waves those born SGA had smaller anthropometric z-scores compared to non-SGA individuals (β ranging from −0.50 to −0.25). Participants residing in urban environments were significantly larger in Waves 2 to 4 (β ranged 0.26 to 0.65). Those born SGA had higher ABSI scores in Waves 2 and 4 (β 0.26 and 0.37, respectively). In longitudinal analyses, those born SGA had smaller measures of body composition across the life course; these differences were larger in urban communities. In remote communities those born SGA had significantly higher ABSI scores during adolescence and young adulthood, and this difference was not observed in urban communities.

Conclusion: Indigenous Australians born SGA are smaller anthropometrically later in life compared to their non-SGA counterparts. In remote communities, those born SGA had higher levels of central adiposity compared to non-SGA.

KEYWORDS

small for gestational age, body composition, Indigenous Australian, anthropometrics, remote Australian communities

Introduction

Small for gestational age (SGA) is defined as a birth weight < 10th centile for gestational age, and those born SGA have an elevated risk of chronic diseases in adulthood, setting the stage for lifelong health disparities (1, 2). In Australia, the estimated rate of SGA births is approximately 9% in non-Indigenous and 14% among Aboriginal and Torres Strait Islander (hereafter, respectively, referred to as Indigenous Australians) births (3). Higher rates of SGA births are reported in remote Australian Indigenous communities (4) with comparable rates to low-to-middle income countries (5, 6).

The result of SGA followed by rapid weight gain during early postnatal life has been associated with increased long-term risks for central obesity, insulin resistance, impaired glucose tolerance, type 2 diabetes, hypertension, increased fat mass, and cardiovascular disease.

SGA is an adaptation to a less than ideal intrauterine environment. A period of catch-up growth occurs between 6 months and 2 years of age, followed by a trajectory of typical growth. However, SGA babies tend to be shorter and lighter than their appropriate for gestational age (AGA) cohorts (1, 2). The weight gain experienced by SGA babies typically has less fat-free body mass than AGA babies, hypothesized due to a consequence of the mismatch between the adaptations for survival *in-utero* and the abundant postnatal nutritional environment. This mismatch forms the basis of increased risk of central obesity, insulin resistance, compromised glucose tolerance, type 2 diabetes, hypertension, elevated fat mass, and cardiovascular disease seen in infants born SGA who undergo rapid weight gain during early postnatal life (2, 7–10). Obesity is a major risk factor for adult chronic diseases and the combination of SGA and later obesity amplifies this risk. Two studies analysing data from The Study of Longitudinal Indigenous Children (1,759 children born 2001–2008) reported that BMI is significantly lower among those categorised as being moderate-to-high prenatal risk (derived from gestational age, SGA, and birth weight) compared to those born full-term (11); and increased birth weight is associated with increased childhood BMI (12).

The Aboriginal Birth Cohort (ABC) is an Australian prospective longitudinal study investigating the long-term impact of early life factors on health and the burden of disease among Indigenous communities. The ABC study is the longest-running and largest Indigenous birth cohort in Australia with follow-up data collected over three decades among a cohort of 686 babies born to Indigenous Australian mothers. When first followed up at 11 years of age (Wave 2), and subsequently at 18 years of age (Wave 3), individuals born SGA remained significantly smaller anthropometrically compared to their non-SGA peers at both timepoints (13, 14), suggesting limited catch-up growth among those born SGA. The ABC cohort have since been followed up two more times, aged at 23–28 years (Wave 4) and 29–36 years (Wave 5). Recent research analysed anthropometric measures from Waves 2, 3, and 4 in association with socioeconomic status and remoteness, however SGA status was not part of the analyses (15).

The current study aims to extend the previous work of Sayers et al. (13, 14) by examining changes in anthropometric measures, including measures of fat, across the entire life-course comparing SGA and non-SGA individuals living in both urban and remote communities.

Methods

Study design

The ABC study is a prospective longitudinal cohort where data has been collected at birth and across four subsequent waves (birth to the age of mid-thirties). For the current study, we utilise both cross-sectional and longitudinal study designs. For the cross-sectional design we analyse data within each study wave separately, and for the longitudinal design we analyse data from all waves combined.

Setting and study participants

The data analysed in this study come from the ABC study and the recruitment methods have been reported in detail elsewhere (16). Briefly, 686 babies born to Aboriginal mothers at the Royal Darwin Hospital from 1987 to 1990 were recruited for the study. During that period the Royal Darwin Hospital served as the primary facility for Indigenous mothers, attracting over 90% of pregnant Indigenous mothers from a region spanning 400,000 km^2 in the “Top End” of the Northern Territory. Notably, 75% of the study cohort lived in remote communities, including the Arnhem, Victoria Daly, and Tiwi regions, while 25% resided in urban communities, including Darwin and its immediate surroundings.

Throughout their life course, the ABC cohort have been followed-up four times (Wave 2: at childhood aged 8–14 years; Wave 3: at adolescence aged 16–21 years; Wave 4: at young adult aged 23–28 years; and Wave 5: at adult aged 29–36 years). The ABC study recruited 686 Indigenous births (Wave 1) with 385 participating 36 years later (Wave 5), resulting in 301 participants being lost to follow-up during the study period, with 38 of these lost to death. As shown in Figure 1, study participants were sourced for follow-up at each wave regardless of participation in previous waves, resulting in non-continuous participation for some of the cohort. The anthropometric measures of interest were first collected at Wave 2, and therefore the baseline analytic cohort for the current study are those who participated in Wave 2 ($n = 570$ and excluding 1 participant in a wheelchair). The analyses include data from all subsequent waves, regardless of non-continuous participation.

Variables of interest

Demographic information

The demographic variables collected at each wave and analysed in this study were sex (male, female), age, location (remote, urban), and lifestyle factors smoking (yes, no), and alcohol use (yes, no) collected in Waves 3, 4, and 5.

Anthropometric measures (outcomes of interest)

The methods used to record anthropometric measures in the ABC study have been reported elsewhere (13, 14, 17). Briefly, in each phase of the ABC study, trained researchers assessed participants’ body size and shape while they wore lightweight clothing and were barefoot. Height measurements were recorded to the nearest millimetre using a portable stadiometer on a flat, hard surface. Weight and lean mass were measured to the nearest 0.1 kg using a digital electronic scale

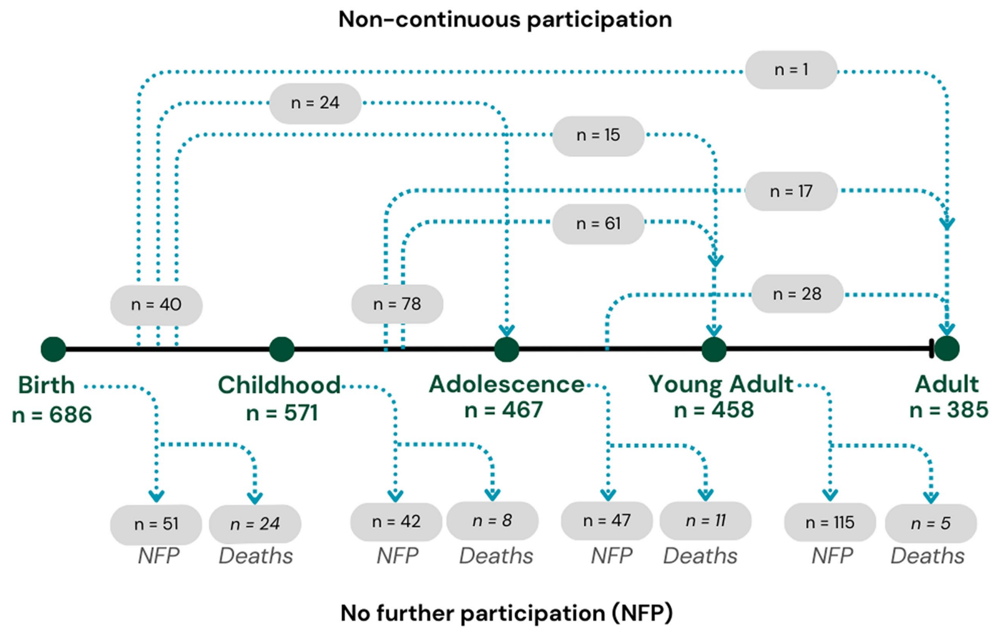

FIGURE 1
Life course participation numbers in the Aboriginal Birth Cohort study across waves 1 to 5.

(TBF-521, Tanita Corporation, Illinois, United States) and assessed through bioimpedance analysis. Waist circumference (WC) was measured in centimetres (cm) at a horizontal plane, midway between the lowest ribs and the iliac crests. For the current study, the following indices and ratios were computed: Body mass index (BMI) was calculated using the standard formula (weight[kg]/height[m]^2), fat percentage (FAT%) was derived by dividing fat mass by weight, waist-to-height ratio (WHtR) was calculated by dividing waist circumference by height, and the "A body shape index" (ABSI) was computed using the formula (WC/BMI^2/3 * Height^1/2). The ABSI, a relatively recent index developed by Krakauer and Krakauer (18), was employed as a measure of central obesity independent of BMI.

Small for gestational age (predictor of interest)

Measures of birth weight and length, and gestational age estimations taken at birth, have previously been described in detail (13). Small for gestational age (SGA) was defined as those with a birth weight < 10th centile for gestational age, and non-SGA as those with a birth weight ≥ 10th centile for gestational age.

Statistical analyses

Descriptive statistics, including frequencies, percentages, and means with standard deviations, are reported for demographic information and anthropometric measures. Analyses to compare anthropometric measures between non-SGA and SGA individuals were conducted both cross-sectionally and longitudinally. Cross-sectional analyses, within each wave, employed linear regression models to examine associations between anthropometric measures standardised as z-scores (calculated internally within each wave cohort), and potential predictors of sex (reference = male), age, SGA status (reference = non-SGA), and geographic location (reference = remote). All predictors were included in the models simultaneously. Smoking (yes/no) and alcohol (yes/no) were not collected at Wave 2 and therefore only included in the cross-sectional analyses for Waves, 3, 4, and 5. We additionally explored an interaction term involving SGA status and residential location to assess cross-differences in these groups.

Longitudinal analyses used linear mixed models to assess changes in anthropometric measures over time and differences between SGA and non-SGA and remote and urban residents. Random intercepts and slopes were included for each study participant to account for repeated measures (19). All anthropometric measures were standardised as z-scores (calculated internally across all waves combined) and were entered into models as outcomes regressed on sex (reference = male), age (with a polynomial cubed term), SGA status (reference = non-SGA), and geographic location (reference = remote). We present the results from these models two different ways: (1) plotting of the estimated marginal means for each age throughout the study period, categorised by SGA status and residential location and (2) post-hoc contrasts (e.g., differences in marginal means) between SGA and non-SGA estimated at ages 11, 18, 25, and 32 years (median age in each wave respectively), stratified by residential location.

All data preparation and analyses were performed using R version 4.2.2 (20). Linear mixed models were conducted using the lme4 package (21) and model contrasts and plotting of estimated marginal means were performed using the emmeans package (22).

Results

Descriptive characteristics

The baseline analytic cohort for this study are those who completed Wave 2 (*n* = 570), and of these, 443 (77.7%) completed

Wave 3, 426 (74.7%) completed Wave 4, and 363 (63.7%) completed Wave 5 (Table 1). Of the 570 in the baseline cohort, 444 (77.9%) were born in remote areas and 125 (21.9%) in urban areas. At Wave 2 there were 424 (74.4%) residing in remote areas, increasing to 85.3% residing in remote areas at Wave 5 (chi square 15.54, $p = 0.001$). Of the 570 in the baseline cohort, 115 (20.2%) were born SGA with the participation across waves remaining consistent (chi square 0.168, $p = 0.982$). The percentage of female participants increased slightly across the waves (Wave 2 = 46.6% to Wave 5 = 49.9%), however this was not statistically significant (chi square 2.39, $p = 0.496$). During Waves 3 to 5 there were 45 pregnancies belonging to 42 distinct females, which were excluded from analyses for the wave they were pregnant. At Wave 4, two of the pregnancies were multiparous and at Wave 5 one pregnancy was multiparous.

Cross-sectional analyses

Table 2 shows the mean and standard deviation (SD) for anthropometric measurements across the study waves. Compared to their non-SGA counterparts, individuals born SGA showed lower mean values for all anthropometric measures (except for WHtR) across all waves. In Figure 2, we present correlations between the anthropometric measures within each wave. Overall, BMI displayed the strongest positive correlations with other anthropometric measures (excluding height, $r = 0.75$ to 0.96). Additionally, WC exhibited consistently strong positive correlations with weight across all waves ($r = 0.87$ to 0.92). The positive correlation between height and weight was most pronounced during Wave 2 ($r = 0.83$), and then gradually diminished over the subsequent waves ($r = 0.51$, 0.40, 0.39 waves 3, 4, and 5 respectively). The ABSI exhibited a small negative correlation with BMI during Wave 2 ($r = -0.31$), whereas for subsequent waves BMI and ABSI were not significantly correlated.

Results from linear regression models examining factors associated with anthropometric z-scores can be found in Table 3. In summary, after controlling for sex, age, and residential location, individuals born SGA exhibited consistent reductions in anthropometric z-scores throughout the study waves compared to non-SGA individuals, with standardised coefficients ranging from −0.50 to −0.25. The only exception was WHtR, which showed no significant differences between SGA and non-SGA individuals in Waves 4 and 5. On the other hand, participants residing in urban environments were significantly larger for all anthropometric measurements within Waves 2 to 4 with coefficients ranging from 0.26 to 0.65. At Wave 5, this pattern only persisted for BMI and weight. The most substantial differences between genders were observed in FAT% with females consistently exhibiting higher values across all study waves (coefficients ranged from 1.06 to 1.19). Similarly, females also demonstrated higher WHtR values in Waves 3, 4, and 5 (coefficients ranged from 0.37 to 0.58) and weight (coefficients ranged from −0.42 to −0.33) in Waves 3, 4, and 5. Individuals who reported smoking consistently displayed reduced anthropometric measurements in Waves 3, 4, and 5, with the most prominent differences observed in Wave 5.

For ABSI scores, individuals born SGA displayed significantly higher scores in Waves 2 and 4 (coefficients = 0.26 and 0.37, respectively), whereas SGA was not associated with ABSI in other study waves. Females had significantly lower ABSI scores during Wave 2, however, this shifted over time with females exhibiting significantly higher ABSI scores in later waves. No other factors were found to be associated with ABSI scores, except for urban residents in Wave 2, who had significantly lower ABSI scores compared to their remote counterparts.

In all models presented in Table 3, we conducted further investigations to examine the influence of an interaction term involving SGA and residential location. Results showed statistically significant interaction terms only during Wave 4 (results not shown). In urban settings, non-SGA individuals exhibited significantly larger values across anthropometric measures compared to their SGA counterparts.

Longitudinal analyses

Based on the linear mixed models, the post-hoc contrasts between SGA and non-SGA are shown in Table 4. When estimating the marginal means for anthropometric measures (z-scores) at the ages of 11, 18, 25, and 32 years, contrasts showed that in the urban regions SGA individuals were consistently smaller than their non-SGA peers at 11, 18, and 25 years for BMI (differences = −0.35, −0.57, −0.60, respectively), FAT% (differences = −0.47, −0.62, −0.54, respectively), height (differences = −0.20, −0.43, −0.35, respectively), WC (differences = −0.37, −0.55, −0.66, respectively), weight (differences = −0.31, −0.67, −0.67, respectively), and WHtR (differences = −0.30, −0.44, −0.59, respectively). Whereas in remote communities, similar but smaller differences were observed between SGA and non-SGA at 11, 18, 25 and 32 years for BMI (differences = −0.21, −0.19, −0.33, −0.39, respectively), FAT% (differences = −0.26, −0.24, −0.25, −0.27, respectively), height (differences = −0.16, −0.21, −0.22, −0.24, respectively), WC (differences at 11, 25, and 32 years = −0.18, −0.21, −0.28, respectively), and weight (differences = −0.19, −0.25, −0.37, −0.42, respectively).

For the ABSI, in remote communities the SGA individuals had significantly higher scores at the ages of 18 years (difference = 0.26) and 25 years (difference = 0.34), whereas there was no difference in the ABSI between SGA and non-SGA in urban regions. For the WHtR, in urban regions SGA individuals were significantly smaller at the ages of 11 years (difference = −0.31), 18 years (difference = −0.44), and 25 years (difference = −0.59), but not at 32 years. Whereas, in remote communities WHtR was only smaller among SGA at 11 years (difference = −0.16).

Interpretation of Figure 3 shows that in remote communities the anthropometric trajectories followed a similar pattern for both SGA and non-SGA individuals, with significant differences for all measures except for WC and WHtR. In contrast, differences between SGA and non-SGA individuals in urban environments were most pronounced during the late-teens through mid-twenties, particularly for BMI, FAT%, WC, and weight. However, it's worth noting that these differences between SGA and non-SGA individuals in urban environments diminished towards 30 years of age. These results further confirm the interactions between SGA and residential location that were initially identified in Wave 4 cross-sectional analyses.

Further analyses were restricted to those residing in remote communities to assess the differences in anthropometric measures across genders and SGA status. Table 5 shows the post-hoc

TABLE 1 Descriptive characteristics of the study cohort from childhood to adult, stratified by small for gestational age (SGA) at birth.

	Wave 2 (Aged 8–14 yrs.)			Wave 3 (Aged 16–21 yrs.)			Wave 4 (Aged 23–28 yrs.)			Wave 5 (Aged 29–36 yrs.)		
	SGA	Non-SGA	All	SGA	Non-SGA	All	SGA	Non-SGA	All	SGA	Non-SGA	All
Total (*n*, row %)	115 (20.2)	455 (79.8)	570	89 (20.1)	354 (79.9)	443	82 (19.2)	344 (80.8)	426	71 (19.6)	292 (80.4)	363
Sex (*n*, column %)												
Male	60 (52.2)	243 (53.4)	303 (53.2)	40 (44.9)	182 (51.4)	222 (50.1)	36 (43.9)	170 (49.4)	206 (48.4)	38 (53.5)	145 (49.7)	183 (50.4)
Female	55 (47.8)	212 (46.6)	267 (46.8)	49 (55.1)	172 (48.6)	221 (49.9)	46 (56.1)	174 (50.6)	220 (51.6)	33 (46.5)	147 (50.3)	180 (49.6)
- Pregnant[a]	0	0	0	6 (6.7)	14 (4.0)	20 (4.5)	6 (13.0)	15 (8.6)	21 (9.0)	0	4 (2.7)	4 (1.1)
Birth location												
Remote	95 (82.6)	349 (76.7)	444 (77.9)	74 (83.1)	285 (80.5)	359 (81.0)	68 (82.9)	282 (82.0)	350 (82.2)	60 (84.5)	251 (86.0)	311 (85.7)
Urban	20 (17.4)	106 (23.3)	125 (21.9)	15 (16.9)	69 (19.5)	84 (19.0)	14 (17.1)	62 (18.0)	76 (17.8)	11 (15.5)	41 (14.0)	52 (14.3)
Wave location												
Remote	89 (77.4)	335 (73.6)	424 (74.4)	73 (82.0)	276 (78.0)	349 (78.8)	65 (79.3)	265 (77.0)	330 (77.5)	63 (88.7)	245 (83.9)	308 (84.8)
Urban	26 (22.6)	120 (26.4)	146 (25.6)	16 (18.0)	78 (22.0)	94 (21.2)	17 (20.7)	79 (23.0)	96 (22.5)	8 (11.3)	47 (16.1)	55 (15.2)
Smoking												
No	–	–	–	29 (32.6)	92 (26.0)	121 (27.3)	18 (22.0)	85 (24.7)	103 (24.2)	20 (28.2)	75 (25.7)	95 (26.2)
Yes	–	–	–	45 (50.6)	216 (61.0)	261 (58.9)	52 (63.4)	217 (63.1)	269 (63.1)	47 (66.2)	205 (70.2)	252 (69.4)
Missing	115 (100)	455 (100)	570 (100)	15 (16.9)	46 (13.0)	61 (13.8)	12 (14.6)	42 (12.2)	54 (12.7)	4 (5.6)	12 (4.1)	16 (4.4)
Alcohol												
No	–	–	–	48 (53.9)	188 (53.1)	236 (53.3)	31 (37.8)	145 (42.2)	176 (41.3)	37 (52.1)	128 (43.8)	165 (45.5)
Yes	–	–	–	32 (36.0)	144 (40.7)	176 (39.7)	41 (50.0)	161 (46.8)	202 (47.4)	30 (42.3)	152 (52.1)	182 (50.1)
Missing	115 (100)	455 (100)	570 (100)	9 (10.1)	22 (6.2)	31 (7.0)	10 (12.2)	38 (11.0)	48 (11.3)	4 (5.6)	12 (4.1)	16 (4.4)

[a] Percentage based on total females in that cell.

TABLE 2 Mean (SD) anthropometric measures from childhood to adult.

	Wave 2 (Aged 8–14 yrs.)		Wave 3 (Aged 16–21 yrs.)		Wave 4 (Aged 23–28 yrs.)		Wave 5 (Aged 29–36 yrs.)	
	n	Mean (SD)	*n*	Mean (SD)	*n*	Mean (SD)	*n*	Mean (SD)
Among all								
ABSI	537	0.082 (0.004)	415	0.080 (0.004)	394	0.082 (0.005)	355	0.083 (0.006)
BMI	569	17.0 (3.5)	422	21.6 (5.7)	404	23.7 (6.4)	359	25.3 (6.7)
Fat %	509	21.3 (9.4)	397	20.7 (12.1)	360	24.8 (12.1)	306	27.6 (10.0)
Height (cm)	570	143.8 (10.6)	423	167.8 (8.7)	405	167.9 (8.8)	359	167.5 (8.9)
Waist circ. (cm)	538	64.6 (9.6)	415	79.3 (14.8)	395	86.6 (15.7)	355	91.8 (17.1)
Weight (kg)	570	35.9 (11.8)	422	61.1 (19.2)	404	67.2 (20.1)	359	71.2 (20.4)
Waist/height ratio	538	0.4 (0.1)	415	0.5 (0.1)	395	0.5 (0.1)	355	0.5 (0.1)
Among SGA								
ABSI	107	0.083 (0.004)	80	0.080 (0.004)	74	0.083 (0.005)	71	0.084 (0.005)
BMI	114	15.9 (2.7)	83	20.0 (4.7)	75	21.8 (5.6)	71	23.5 (5.5)
Fat %	103	18.4 (8.2)	75	17.0 (10.3)	68	22.5 (11.4)	57	25.0 (10.1)
Height (cm)	115	142.8 (10.8)	83	164.5 (8.0)	76	164.7 (8.0)	71	164.4 (8.6)
Waist circ. (cm)	108	62.2 (7.8)	80	75.5 (12.7)	75	82.2 (13.1)	71	87.7 (13.7)
Weight (kg)	115	33.0 (9.8)	83	54.3 (13.7)	75	59.2 (15.3)	71	63.5 (15.0)
Waist/height ratio	108	0.4 (0.0)	80	0.5 (0.1)	75	0.5 (0.1)	71	0.5 (0.1)
Among non-SGA								
ABSI	430	0.082 (0.004)	335	0.080 (0.004)	320	0.082 (0.004)	282	0.083 (0.004)
BMI	455	17.2 (3.6)	339	21.9 (5.9)	329	24.2 (6.5)	288	25.8 (6.9)
Fat %	406	22.0 (9.5)	322	21.5 (12.3)	292	25.3 (12.2)	249	28.2 (9.9)
Height (cm)	455	144.1 (10.5)	340	168.6 (8.6)	329	168.7 (8.8)	288	168.2 (8.8)
Waist circ. (cm)	430	65.2 (9.9)	335	80.2 (15.2)	320	87.6 (16.1)	284	92.8 (17.7)
Weight (kg)	455	36.6 (12.2)	339	62.8 (19.9)	329	69.0 (20.7)	288	73.1 (21.1)
Waist/height ratio	430	0.5 (0.1)	335	0.5 (0.1)	320	0.5 (0.1)	284	0.6 (0.1)

contrasts between females and males (reference) estimated at the ages 11, 18, 25, and 32 years, stratified by SGA status. At age 18 years females had significantly higher ABSI scores than males, and the magnitude of these differences were similar in the SGA (gender difference = 0.50) and non-SGA groups (gender difference = 0.48). Then at ages 25 and 32 years the magnitude of the difference in ABSI score between females and males was almost two-fold in the SGA group (at 25 and 32 years: gender difference = 1.47 and 1.19 respectively) than in the non-SGA group (at 25 and 32 years: gender difference = 0.85 and 0.65). Females also had significantly higher FAT% and WHtR but the differences were relatively similar in the SGA and non-SGA groups. Figure 4 shows the estimated marginal means for males and females in remote communities, stratified by SGA status.

Sensitivity analyses

For the linear mixed models, we included tobacco smoking and alcohol use and only analysed data from Wave 3 onwards. The estimated marginal means at the ages of 18, 25, and 32 years can be found in Supplementary Table S1. Given that study participation was non-continuous, we limited the cross-sectional and longitudinal analyses to those who participated in all waves (*n* = 283) and results were similar to the those yielded in the main analyses, however SGA individuals were not significantly smaller across all anthropometric measures in Waves 4 and 5 (Supplementary Tables S2, S3). Within each wave, we checked for differences in anthropometric measures among those who continued to participate in the subsequent wave versus those who did not participate in the next wave. Results showed no differences in anthropometric measures for the transition of Wave 2 to Wave 3; for Wave 3 to Wave 4 those who did not continue to Wave 4 had significantly higher BMI, weight and WC; for Wave 4 to Wave 5 those who did not continue to Wave 5 had significantly higher WC and weight (Supplementary Table S4).

To compare the ABC study participant ABSI scores to those originally derived from the NHANES, we downloaded the data from the original authors (18) and plotted the ABSI scores for the ABC and NHANES cohorts across the age groups stratified by gender (Supplementary Figure S1). It was observed that the ABC study participants had significantly higher ABSI scores than the NHANES cohort, with more prominent differences among older females.

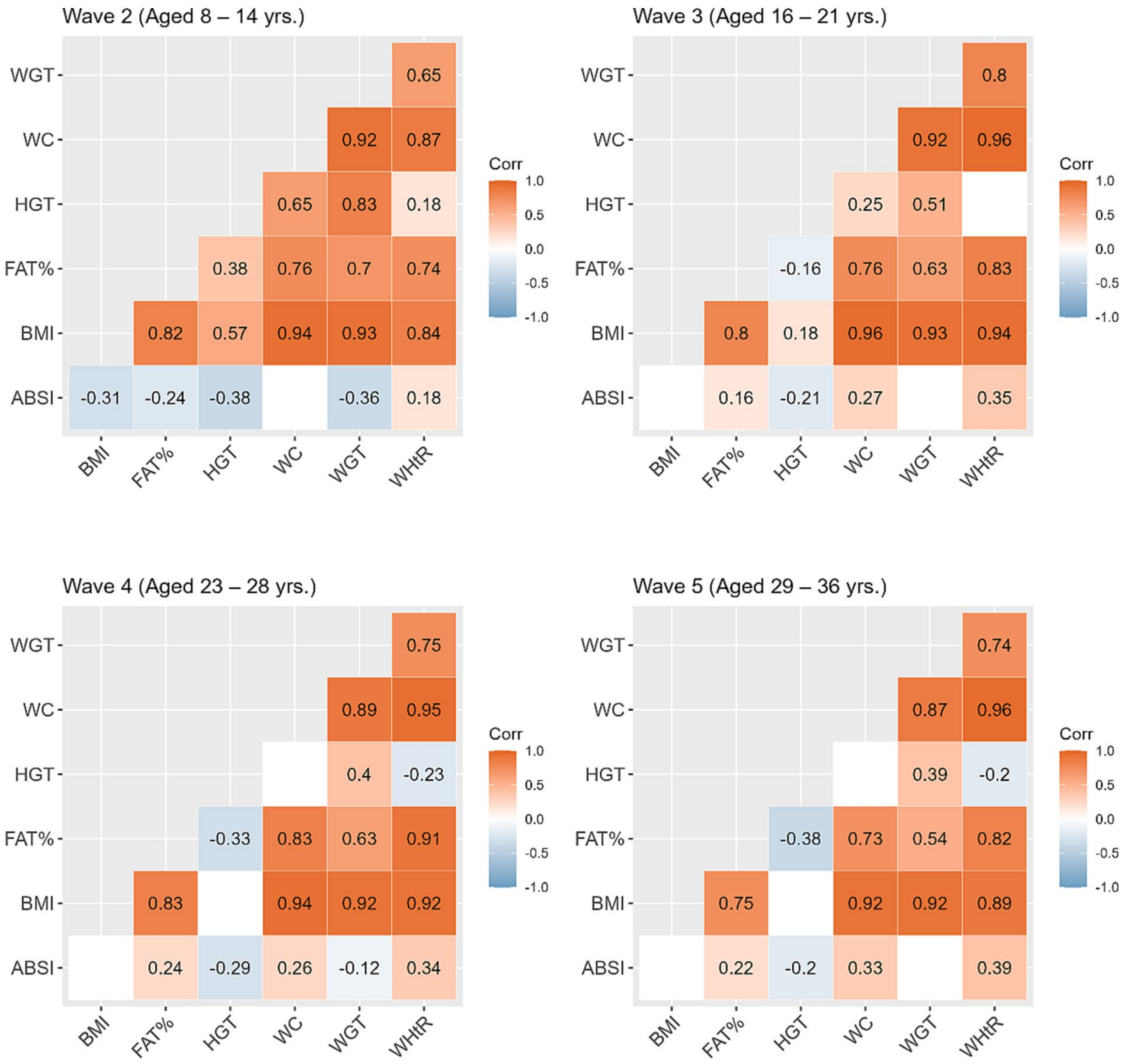

FIGURE 2
Correlations between the anthropometric measures within each study wave. Blank cells represent non-significant correlations. ABSI, a body shape index; BMI, body mass index; FATP, fat percentage; HGT, height; WC, waist circumference; WGT, weight; WHtR, waist/height ratio.

Discussion

We investigated the longitudinal changes in anthropometric parameters among Indigenous Australians who were born SGA compared to those born non-SGA. Our results consistently demonstrate lower anthropometric measurements in the SGA individuals from childhood through young adult, indicating that through their life course those born SGA are smaller than those born non-SGA. Although current literature on SGA and body composition later in life among Indigenous Australians is limited, our findings align with other studies (from Sweden, Netherlands, Japan, Greece, and a meta-analysis) reporting that individuals born SGA tend to be smaller during childhood (23–27).

Within each study wave most anthropometric measures were positively correlated (except for correlations with ABSI). Hence, the magnitude of the differences between SGA and non-SGA individuals were similar across different anthropometric measures, except for WHtR yielding no difference during the two latest waves. However, the disparity in anthropometric measures between SGA and non-SGA individuals was more evident in urban communities where non-SGA are larger than their SGA counterparts, in addition to being larger than both SGA and non-SGA in remote communities. Indigenous Australians nutritional intake has transitioned since colonisation from a traditional, varied and nutrient-dense diet, high in fibre and low in fat and refined carbohydrates, to an energy-dense westernised diet, high in fat and refined sugars (28). The geographic differential observed indicates the strong influence that residential environments have on growth and body composition, which could be driven by lifestyle factors such as dietary options available, food

TABLE 3 Cross-sectional analyses: associations between anthropometric z-scores and potential correlates within each study wave.

	ABSI	BMI	Fat %	Height	Waist circ.	Weight	Waist/height ratio
	Coefficient (95% CI)	Coefficient (95% CI)	Coefficient (95% CI)	Coefficient (95% CI)	Coefficient (95% CI)	Coefficient (95% CI)	Coefficient (95% CI)
Wave 2 (Aged 8–14 yrs.)							
SGA	0.26 (0.06, 0.46)	**−0.42 (−0.61, −0.23)**	**−0.42 (−0.60, −0.24)**	−0.25 (−0.40, −0.10)	**−0.36 (−0.55, −0.17)**	**−0.38 (−0.55, −0.21)**	**−0.32 (−0.52, −0.11)**
Female	**−0.30 (−0.46, −0.14)**	0.22 (0.07, 0.37)	**1.06 (0.91, 1.20)**	0.22 (0.10, 0.34)	*0.13 (−0.02, 0.29)*	0.24 (0.10, 0.38)	*0.04 (−0.13, 0.21)*
Urban	**−0.54 (−0.73, −0.35)**	**0.64 (0.46, 0.81)**	**0.36 (0.18, 0.53)**	**0.50 (0.36, 0.64)**	**0.56 (0.38, 0.74)**	**0.65 (0.49, 0.81)**	**0.39 (0.19, 0.59)**
Age	−0.20 (−0.27, −0.13)	0.22 (0.15, 0.28)	0.18 (0.12, 0.24)	**0.58 (0.53, 0.63)**	0.28 (0.21, 0.35)	**0.40 (0.34, 0.46)**	*−0.01 (−0.08, 0.07)*
Wave 3 (Aged 16–21 yrs.)							
SGA	*0.06 (−0.19, 0.31)*	**−0.37 (−0.62, −0.13)**	**−0.42 (−0.64, −0.20)**	**−0.37 (−0.56, −0.19)**	**−0.38 (−0.62, −0.14)**	**−0.45 (−0.68, −0.22)**	−0.28 (−0.53, −0.04)
Female	**0.53 (0.33, 0.73)**	*0.05 (−0.14, 0.24)*	**1.07 (0.90, 1.24)**	**−1.27 (−1.41, −1.12)**	*0.00 (−0.19, 0.20)*	**−0.38 (−0.56, −0.20)**	**0.37 (0.18, 0.57)**
Urban	*−0.14 (−0.40, 0.12)*	**0.56 (0.31, 0.81)**	**0.38 (0.16, 0.61)**	0.26 (0.06, 0.45)	**0.51 (0.26, 0.77)**	**0.61 (0.37, 0.85)**	**0.44 (0.19, 0.70)**
Age	*0.04 (−0.05, 0.13)*	*0.03 (−0.05, 0.12)*	*−0.01 (−0.09, 0.07)*	0.01 (−0.05, 0.08)	*0.05 (−0.04, 0.14)*	*0.04 (−0.05, 0.12)*	*0.05 (−0.04, 0.14)*
Smoking	*0.17 (−0.05, 0.38)*	−0.28 (−0.49, −0.08)	−0.20 (−0.38, −0.01)	0.00 (−0.16, 0.15)	−0.22 (−0.43, −0.01)	−0.26 (−0.45, −0.06)	−0.22 (−0.43, −0.01)
Alcohol	*0.05 (−0.18, 0.27)*	**0.34 (0.12, 0.55)**	0.26 (0.07, 0.45)	0.21 (0.04, 0.37)	**0.35 (0.13, 0.56)**	**0.36 (0.16, 0.56)**	**0.30 (0.08, 0.52)**
Wave 4 (Aged 23–28 yrs.)							
SGA	**0.37 (0.12, 0.62)**	**−0.41 (−0.67, −0.14)**	**−0.30 (−0.54, −0.06)**	**−0.38 (−0.57, −0.19)**	**−0.37 (−0.63, −0.11)**	**−0.50 (−0.75, −0.24)**	*−0.26 (−0.51, 0.00)*
Female	**0.69 (0.48, 0.90)**	*0.19 (−0.03, 0.40)*	**1.13 (0.93, 1.33)**	**−1.35 (−1.51, −1.20)**	*0.19 (−0.03, 0.40)*	**−0.33 (−0.54, −0.13)**	**0.58 (0.37, 0.80)**
Urban	*0.00 (−0.24, 0.25)*	**0.49 (0.23, 0.74)**	0.29 (0.05, 0.53)	0.28 (0.09, 0.46)	**0.49 (0.24, 0.75)**	**0.56 (0.31, 0.80)**	**0.40 (0.14, 0.65)**
Age	*−0.04 (−0.12, 0.05)*	*0.08 (−0.01, 0.17)*	*0.06 (−0.02, 0.15)*	*0.00 (−0.06, 0.07)*	*0.07 (−0.02, 0.16)*	*0.07 (−0.02, 0.16)*	*0.07 (−0.02, 0.16)*
Smoking	*0.03 (−0.19, 0.25)*	−0.29 (−0.52, −0.06)	−0.26 (−0.47, −0.05)	*−0.04 (−0.20, 0.13)*	−0.28 (−0.51, −0.05)	*−0.29 (−0.51, −0.07)*	−0.26 (−0.49, −0.04)
Alcohol	−0.27 (−0.50, −0.05)	*0.15 (−0.08, 0.38)*	*0.12 (−0.09, 0.33)*	*0.15 (−0.02, 0.32)*	*0.12 (−0.12, 0.35)*	*0.16 (−0.07, 0.38)*	*0.08 (−0.15, 0.31)*
Wave 5 (Aged 29–36 yrs.)							
SGA	*0.17 (−0.09, 0.43)*	**−0.33 (−0.58, −0.08)**	−0.29 (−0.53, −0.06)	**−0.48 (−0.66, −0.30)**	**−0.31 (−0.56, −0.05)**	**−0.49 (−0.73, −0.25)**	*−0.16 (−0.41, 0.10)*
Female	**0.52 (0.31, 0.74)**	*0.14 (−0.07, 0.34)*	**1.19 (1.01, 1.38)**	**−1.44 (−1.58, −1.29)**	*0.08 (−0.13, 0.30)*	**−0.42 (−0.62, −0.22)**	**0.48 (0.27, 0.69)**
Urban	*−0.19 (−0.49, 0.10)*	**0.34 (0.05, 0.63)**	*0.15 (−0.11, 0.41)*	0.22 (0.02, 0.43)	*0.27 (−0.03, 0.56)*	**0.41 (0.13, 0.69)**	*0.20 (−0.09, 0.49)*
Age	*0.01 (−0.06, 0.09)*	*0.03 (−0.04, 0.10)*	*0.01 (−0.05, 0.08)*	*0.00 (−0.05, 0.05)*	*0.04 (−0.03, 0.11)*	*0.03 (−0.03, 0.10)*	*0.04 (−0.03, 0.11)*
Smoking	*0.08 (−0.16, 0.32)*	**−0.64 (−0.87, −0.41)**	**−0.39 (−0.60, −0.18)**	*0.04 (−0.13, 0.20)*	**−0.56 (−0.80, −0.32)**	**−0.60 (−0.82, −0.37)**	**−0.55 (−0.78, −0.32)**
Alcohol	*−0.13 (−0.36, 0.10)*	0.25 (0.03, 0.47)	0.21 (0.01, 0.41)	*0.09 (−0.06, 0.25)*	*0.22 (−0.01, 0.44)*	0.27 (0.06, 0.48)	*0.18 (−0.04, 0.40)*

All potential correlates are entered into the models simultaneously. Wave 2 models included sex, age, SGA status, and residential location. Wave 3, 4, and 5 models included sex, age, SGA status, residential location, smoking, and alcohol use. To assist in observing stronger associations, figures in **bold** represent significant associations where the coefficient ≥ 0.30 (absolute). Figures italicised represent non-significant associations.

TABLE 4 Longitudinal analyses: differences in anthropometric z-score marginal means between SGA and non-SGA (reference) estimated at 11, 18, 25, and 32 years of age, stratified by residential location.

Stratified by	ABSI	BMI	Fat %	Height	Waist circ.	Weight	Waist/height ratio
	Difference (95%CI)	Difference (95%CI)	Difference (95%CI)	Difference (95%CI)	Difference (95%CI)	Difference (95%CI)	Difference (95%CI)
Age 11							
Remote	0.14 (−0.05, 0.33)	**−0.21 (−0.34, −0.07)**	**−0.26 (−0.44, −0.08)**	−0.16 (−0.29, −0.03)	−0.18 (−0.31, −0.04)	−0.19 (−0.30, −0.07)	−0.16 (−0.31, −0.01)
Urban	0.42 (0.00, 0.85)	**−0.35 (−0.57, −0.14)**	**−0.47 (−0.82, −0.13)**	**−0.20 (−0.42, 0.02)**	**−0.37 (−0.63, −0.11)**	**−0.31 (−0.50, −0.13)**	**−0.31 (−0.59, −0.02)**
Age 18							
Remote	**0.26 (0.04, 0.48)**	−0.19 (−0.36, −0.02)	**−0.24 (−0.43, −0.05)**	**−0.21 (−0.33, −0.10)**	*−0.12 (−0.28, 0.04)*	**−0.25 (−0.40, −0.10)**	*−0.05 (−0.23, 0.13)*
Urban	−0.06 (−0.50, 0.38)	**−0.57 (−0.83, −0.31)**	**−0.62 (−0.96, −0.27)**	**−0.43 (−0.62, −0.24)**	**−0.55 (−0.83, −0.28)**	**−0.67 (−0.90, −0.45)**	**−0.44 (−0.75, −0.12)**
Age 25							
Remote	**0.34 (0.10, 0.59)**	**−0.33 (−0.54, −0.12)**	**−0.25 (−0.45, −0.05)**	**−0.22 (−0.33, −0.11)**	**−0.21 (−0.41, −0.02)**	**−0.37 (−0.56, −0.18)**	*−0.14 (−0.37, 0.09)*
Urban	−0.04 (−0.54, 0.46)	**−0.60 (−0.94, −0.26)**	**−0.54 (−0.93, −0.14)**	**−0.35 (−0.56, −0.15)**	**−0.66 (−1.01, −0.32)**	**−0.67 (−0.98, −0.37)**	**−0.59 (−1.00, −0.19)**
Age 32							
Remote	0.28 (0.00, 0.56)	**−0.39 (−0.65, −0.13)**	**−0.27 (−0.50, −0.05)**	**−0.24 (−0.36, −0.11)**	**−0.28 (−0.52, −0.04)**	**−0.42 (−0.66, −0.19)**	*−0.20 (−0.48, 0.08)*
Urban	−0.18 (−0.91, 0.55)	*−0.31 (−0.80, 0.19)*	*−0.06 (−0.62, 0.49)*	*−0.28 (−0.59, 0.03)*	*−0.43 (−0.94, 0.08)*	*−0.38 (−0.82, 0.05)*	*−0.38 (−0.98, 0.22)*

These are post-hoc contrasts from the linear mixed models with interaction terms between SGA and residential location. Models included sex, age, SGA status, and residential location. To assist in observing stronger associations, figures in **bold** represent significant differences ≥0.20 (absolute). Figures italicised represent non-significant differences.

insecurity, and financial stress in the remote Indigenous communities (15, 17, 29), and/or people in urban areas having greater access to supermarkets and fast food outlets and a high proportion of energy-dense, nutrient-poor diet (28).

As expected, other publications analysing the anthropometric data from the ABC study have also reported this geographical divide with individuals in urban regions being larger compared to their counterparts in remote areas (15, 17, 30–32). This geographic differential has also been highlighted in other measures of health within the ABC study. For instance, Indigenous children residing in remote areas exhibit lower markers of lung function (33), haemoglobin levels, total cholesterol levels and systolic blood pressure (30), while Indigenous young adults residing in remote areas exhibit longer cognitive reaction times (34), lower maximal grip strength (32), lower iodine levels (35), and Indigenous females in remote areas exhibit adverse cardio-metabolic profiles (31).

BMI is a widely adopted metric for evaluating overall obesity by considering an individual's weight and height. It has limitations as it does not consider factors such as body shape or the distribution of muscle mass versus fat mass (36). In our study, BMI was observed to be lower among SGA individuals in both remote and urban regions with the difference in urban regions being far more pronounced. This is consistent with other studies reporting that those born SGA have a lower BMI later in life (26, 37). Two Australian studies analysing data from a remote Indigenous community in the Northern Territory found BMI to be inversely associated with all-cause mortality after follow-up at 9 years (38) and 18 years (39), however, increased WC was associated with increased risk of all-cause mortality (39). In the context of our study, these findings would suggest conflicting mortality risks for SGA individuals given that they showed significantly lower BMI and WC compared to those born non-SGA. It is important to note however, other large studies of non-Indigenous populations have reported a J-shape relationship between BMI and mortality showing that both lower and higher BMI is associated with increased risk of mortality (40, 41).

ABSI, on the other hand, incorporates WC as a key component in its calculation, and therefore intended to provide a better measure of central adiposity (18). In support of ABSI being independent of BMI our results showed no significant correlation between ABSI and BMI in Waves 3, 4, and 5. The ABSI and WHtR were the only two anthropometric measures that were inconsistent across the ages in terms of the contrasts between SGA and non-SGA individuals. In remote communities ABSI scores were significantly elevated among those born SGA compared to their non-SGA counterparts, and in urban regions there was no difference in ABSI scores between SGA and non-SGA despite the urban non-SGA individuals being the largest group across most anthropometric measures. This difference between SGA and non-SGA in ABSI scores in remote communities first appeared from adolescence, indicating an early-life predisposition to central adiposity among SGA individuals in remote communities. Earlier research among the ABC study participants at childhood showed that many had lower BMI with higher waist measurements (42).

Hence, although SGA individuals in remote communities are smaller in size, their higher ABSI scores indicate greater central adiposity. This interesting finding highlights the potential complex

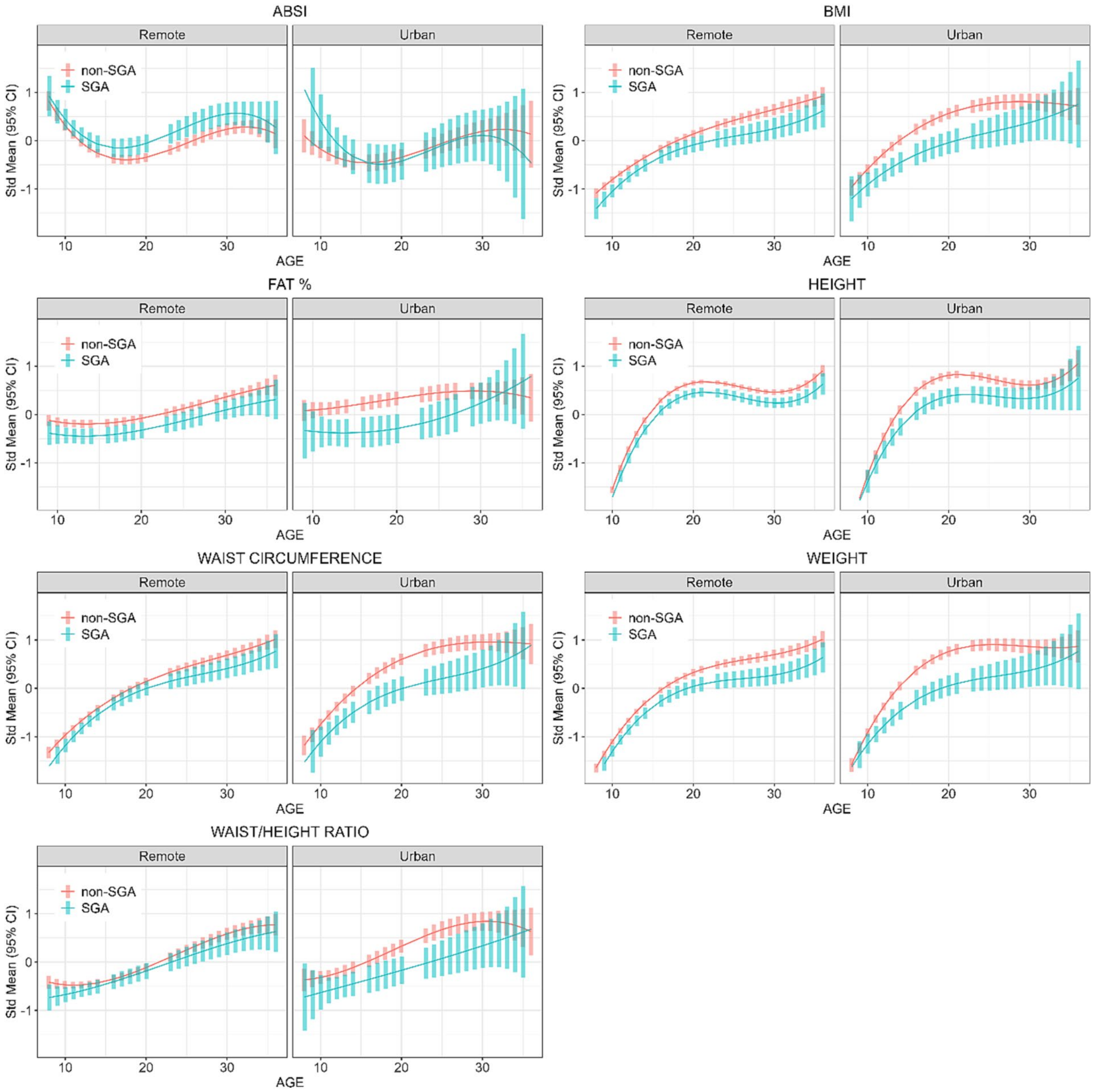

FIGURE 3
Longitudinal analyses: estimated anthropometric z-scores for ages across all waves, stratified by SGA status and Location. Models included sex, age, SGA status, and residential location.

interplay between possible early catch-up growth among those born SGA, lifestyle factors later in life, and the nutritional challenges prevalent in remote communities (29, 43). Rapid catch-up growth among SGA infants is a risk factor for being overweight/obese at 2–5 years of age (9) and is associated with increased fat mass at 9 years of age (37), and fat mass with central adiposity at 32 years of age (44). Lower birth weight combined with faster growth in the first 5 years has also been associated with increased hepatic fat in early childhood (45). Given that the first follow-up of the ABC study participants was at 8 years of age (Wave 2), we were unable to determine if any of our results, particularly higher ABSI scores (e.g., higher central adiposity) among those born SGA, are associated with catch-up growth during infancy. However, based on the assumption that Indigenous Australians in remote communities most likely experience similar lifestyle and nutritional challenges, it is therefore possible that individuals born SGA and residing in remote communities may exhibit a propensity for central adiposity, potentially mediated by the biological processes of catch-up growth and lifestyle factors. Consistent with this view, a study of 128 Australian Indigenous children found that children who experienced rapid weight gain in the first 12 months of life were 2.7 times more likely to be overweight at 9 years. Although the analyses were not stratified by gender, female children were 2.4 times more likely to be overweight at 9 years compared to male children (46).

Our cross-sectional findings (in models including SGA status) showed that, although females weighed less, they had higher FAT% with a larger WHtR and ABSI scores and this was more evident in remote communities. In analyses stratified by SGA status, the gender disparity in ABSI scores among SGA individuals was nearly twofold greater than observed in the non-SGA group. This observation implies

TABLE 5 Longitudinal analyses: within remote communities, differences in anthropometric z-score marginal means between female and male (reference) estimated at 11, 18, 25, and 32 years of age, stratified by SGA status.

Stratified by	ABSI	BMI	Fat %	Height	Waist circ.	Weight	Waist/height ratio
	Difference (95%CI)	Difference (95%CI)	Difference (95%CI)	Difference (95%CI)	Difference (95%CI)	Difference (95%CI)	Difference (95%CI)
Age 11							
Non-SGA	−0.18 (−0.35, −0.01)	*0.10 (−0.01, 0.21)*	**0.85 (0.69, 1.02)**	0.11 (0.01, 0.21)	*0.06 (−0.05, 0.17)*	*0.08 (−0.01, 0.18)*	*0.04 (−0.08, 0.17)*
SGA	**−0.36 (−0.69, −0.03)**	*0.09 (−0.12, 0.31)*	**0.83 (0.51, 1.16)**	**0.23 (0.04, 0.43)**	*0.05 (−0.16, 0.27)*	*0.13 (−0.06, 0.31)*	*−0.05 (−0.29, 0.19)*
Age 18							
Non-SGA	**0.48 (0.30, 0.66)**	*0.11 (−0.04, 0.25)*	**1.22 (1.05, 1.39)**	**−0.70 (−0.80, −0.61)**	*0.10 (−0.04, 0.24)*	**−0.24 (−0.36, −0.11)**	**0.42 (0.26, 0.58)**
SGA	**0.50 (0.13, 0.86)**	*−0.09 (−0.38, 0.19)*	**1.04 (0.70, 1.38)**	**−0.61 (−0.80, −0.43)**	*−0.08 (−0.36, 0.19)*	**−0.34 (−0.59, −0.10)**	*0.19 (−0.12, 0.50)*
Age 25							
Non-SGA	**0.85 (0.64, 1.06)**	*0.18 (−0.02, 0.37)*	**1.20 (1.01, 1.39)**	**−0.88 (−0.97, −0.79)**	*0.19 (0.00, 0.37)*	**−0.31 (−0.47, −0.14)**	**0.65 (0.44, 0.85)**
SGA	**1.47 (1.07, 1.87)**	*−0.09 (−0.46, 0.28)*	**0.93 (0.56, 1.30)**	**−0.85 (−1.03, −0.68)**	*0.12 (−0.23, 0.47)*	**−0.43 (−0.75, −0.12)**	**0.54 (0.14, 0.93)**
Age 32							
Non-SGA	**0.65 (0.42, 0.89)**	*0.18 (−0.07, 0.42)*	**1.01 (0.80, 1.22)**	**−0.86 (−0.96, −0.76)**	*0.13 (−0.10, 0.36)*	**−0.34 (−0.54, −0.13)**	**0.60 (0.34, 0.86)**
SGA	**1.19 (0.74, 1.63)**	*0.03 (−0.43, 0.50)*	**0.98 (0.57, 1.39)**	**−0.80 (−0.98, −0.61)**	*0.17 (−0.27, 0.60)*	*−0.36 (−0.75, 0.04)*	**0.60 (0.11, 1.10)**

These are post-hoc contrasts from the linear mixed models with interaction terms between SGA and sex. Models included sex, age, SGA status. To assist in observing stronger associations, figures in **bold** represent significant differences ≥0.20 (absolute). Figures italicised represent non-significant differences.

that in remote communities, Indigenous females born SGA tend to accrue a higher degree of central adiposity than SGA males by late adolescence. In support of this, other research conducted within the ABC study showed that Indigenous females residing in remote communities exhibit the highest predicted likelihood of having an adverse cardio-metabolic profile, a trend that was consistent across all levels of BMI. Furthermore, this pattern was not observed among their male counterparts (31).

To the best of our knowledge, there are no prior studies specifically investigating ABSI within Indigenous communities, making direct comparisons with existing research challenging. However, it is worth noting a recent study conducted on a sample of Australian adults ($n = 4{,}056$) reported a dose–response association with mortality across ABSI quartiles (47). In an Italian study of overweight and obese children aged 2–18 years, higher ABSI scores were associated with higher cardio-metabolic risk markers (48). Other studies have also reported that the ABSI performs well in predicting all-cause mortality (18, 49–51), hypertension (50), arterial stiffness in patients with type 2 diabetes (52), cardiovascular disease risk (53), and cancer (54).

Although research on SGA and body composition among Indigenous Australians is limited, studies among minority groups in low- and middle-income countries have reported positive associations between birth weight and body composition later in life with differences across genders. For example, in the Inuit population of Greenland (aged 18–61 years), birth weight was positively associated with BMI, waist circumference, fat mass index, fat-free mass index, and subcutaneous adipose tissue with weaker associations among females compared to males (55). In South Western Townships (Soweto), South Africa, a study of young adults (aged 22 years) reported that birth weight was positively associated with fat mass in males only, while relative weight gain in early life was associated with visceral adipose tissue in females (56). Another study in South Africa (infants aged 24 months) reported positive associations between weight gain and fat mass, fat-free mass, fat mass index, and fat-free mass index with no difference between genders (57). In Mexico, a study of Maya children (mean age 7.5 years) indicated that birth weight was positively associated with fat-free mass index and fat mass index among males only (58). A study of Brazilian adults (aged 35–74 years) found that low birth weight predicted higher levels of truncal fat in females but not in males (59, 60).

Limitations

Several limitations must be acknowledged in our study. Firstly, the children were first followed-up at 8 years of age, therefore early catch-up growth could not be analysed. Secondly, our study classified individuals into either remote or urban residential categories; however, we lacked information on the precise duration spent in each location, which complicates the ability to draw definitive conclusions regarding the association between residential location and growth patterns. Thirdly, although convenient, inexpensive and easy to use, Bioelectrical Impedance Analysis (BIA) underestimates body fat in the severely obese compared to DEXA (61). Lastly, a large portion of the study population resided in remote areas, limiting our ability to explore stratified analyses within the urban context.

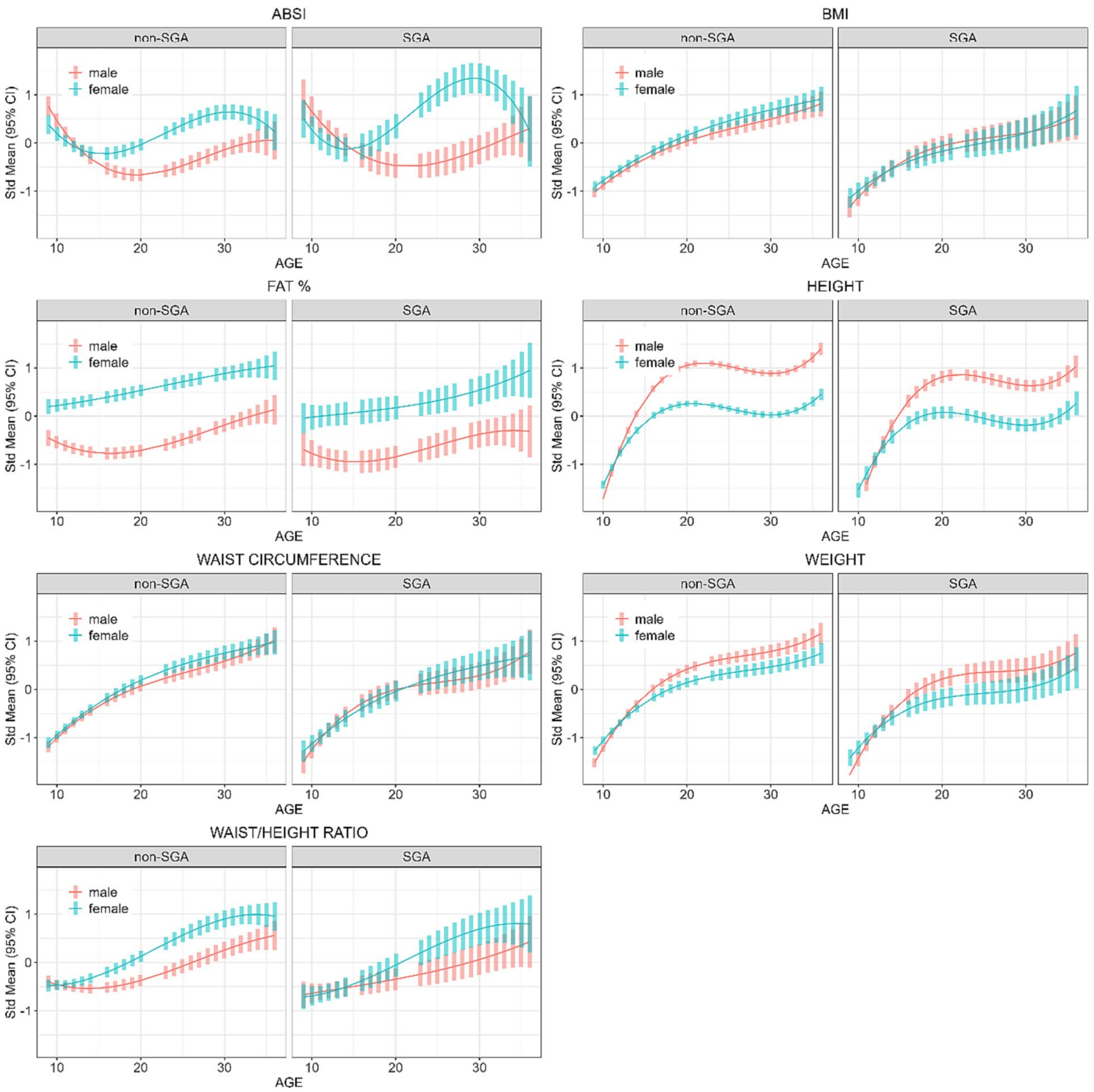

FIGURE 4
Longitudinal analyses within remote communities: estimated anthropometric z-scores for ages across all waves, stratified by sex and SGA status. Models included sex, age, SGA status.

Conclusion

The findings of our study showed that Indigenous individuals born SGA are smaller anthropometrically later in life compared to their non-SGA counterparts, and this disparity was more pronounced in urban regions. However, SGA individuals in remote communities had higher ABSI scores than non-SGA individuals, indicating that SGA individuals were more prone to accruing central adiposity. This result was driven by females in remote communities having higher ABSI and fat percentage than males, which was more evident among those born SGA. These findings highlight the complex relationships between early growth patterns, residential location, gender disparities, and central adiposity among Australian Indigenous born SGA.

Data availability statement

The original contributions presented in the study are included in the article/Supplementary materials, further inquiries can be directed to the corresponding author: belinda.davison@menzies.edu.au.

Ethics statement

The studies involving humans were approved by Human Research Ethics Committee of Northern Territory Health and Menzies School of Health Research (NT HREC). The studies were conducted in accordance with the local legislation and institutional requirements.

Written informed consent for participation in this study was provided by the participants' legal guardians/next of kin.

Author contributions

CH: Data curation, Formal analysis, Methodology, Writing – original draft. BD: Conceptualization, Data curation, Funding acquisition, Investigation, Methodology, Project administration, Resources, Writing – review & editing. GS: Conceptualization, Data curation, Funding acquisition, Investigation, Methodology, Project administration, Resources, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2024.1349040/full#supplementary-material

References

1. Nam HK, Lee KH. Small for gestational age and obesity: epidemiology and general risks. *Ann Pediatr Endocrinol Metab*. (2018) 23:9–13. doi: 10.6065/apem.2018.23.1.9

2. Cho WK, Suh BK. Catch-up growth and catch-up fat in children born small for gestational age. *Korean J Pediatr*. (2016) 59:1–7. doi: 10.3345/kjp.2016.59.1.1

3. Australian Institute of Health and Welfare. *Australia's mothers and babies 2017—in brief. Perinatal statistics series no. 35. Cat. no. PER 100*. Canberra: AIHW. (2019).

4. Sayers S, Powers J. Risk factors for aboriginal low birthweight, intrauterine growth retardation and preterm birth in the Darwin health region. *Aust N Z J Public Health*. (1997) 21:524–30. doi: 10.1111/j.1467-842X.1997.tb01746.x

5. Lee AC, Kozuki N, Cousens S, Stevens GA, Blencowe H, Silveira MF, et al. Estimates of burden and consequences of infants born small for gestational age in low and middle income countries with INTERGROWTH-21(st) standard: analysis of CHERG datasets. *BMJ*. (2017) 358:j3677. doi: 10.1136/bmj.j3677

6. Black RE. Global prevalence of small for gestational age births. *Nestle Nutr Inst Workshop Ser*. (2015) 81:1–7. doi: 10.1159/000365790

7. Hong YH, Chung S. Small for gestational age and obesity related comorbidities. *Ann Pediatr Endocrinol Metab*. (2018) 23:4–8. doi: 10.6065/apem.2018.23.1.4

8. Lu D, Yu Y, Ludvigsson JF, Oberg AS, Sorensen HT, Laszlo KD, et al. Birth weight, gestational age, and risk of cardiovascular disease in early adulthood: influence of familial factors. *Am J Epidemiol*. (2023) 192:866–77. doi: 10.1093/aje/kwac223

9. Li P, Lu Y, Qie D, Feng L, He G, Yang S, et al. Early-life weight gain patterns of term small-for-gestational-age infants and the predictive ability for later childhood overweight/obesity: a prospective cohort study. *Front Endocrinol (Lausanne)*. (2022) 13:1030216. doi: 10.3389/fendo.2022.1030216

10. Zheng M, Hesketh KD, Vuillermin P, Dodd J, Wen LM, Baur LA, et al. Understanding the pathways between prenatal and postnatal factors and overweight outcomes in early childhood: a pooled analysis of seven cohorts. *Int J Obes*. (2023) 47:574–82. doi: 10.1038/s41366-023-01301-9

11. Westrupp EM, D'Esposito F, Freemantle J, Mensah FK, Nicholson JM. Health outcomes for Australian aboriginal and Torres Strait islander children born preterm, low birthweight or small for gestational age: a nationwide cohort study. *PLoS One*. (2019) 14:e0212130. doi: 10.1371/journal.pone.0212130

12. Thurber KA, Dobbins T, Kirk M, Dance P, Banwell C. Early life predictors of increased body mass index among indigenous Australian children. *PLoS One*. (2015) 10:e0130039. doi: 10.1371/journal.pone.0130039

13. Sayers S, Mackerras D, Halpin S, Singh G. Growth outcomes for Australian aboriginal children aged 11 years who were born with intrauterine growth retardation at term gestation. *Paediatr Perinat Epidemiol*. (2007) 21:411–7. doi: 10.1111/j.1365-3016.2007.00852.x

14. Sayers S, Mott S, Singh G. Fetal growth restriction and 18-year growth and nutritional status: aboriginal birth cohort 1987–2007. *Am J Hum Biol*. (2011) 23:417–9. doi: 10.1002/ajhb.21133

15. Sjoholm P, Pahkala K, Davison B, Juonala M, Singh G. Socioeconomic status, remoteness and tracking of nutritional status from childhood to adulthood in an Australian aboriginal birth cohort: the ABC study. *BMJ Open*. (2020) 10:e033631. doi: 10.1136/bmjopen-2019-033631

16. Sayers SM, Mackerras D, Singh G, Bucens I, Flynn K, Reid A. An Australian aboriginal birth cohort: a unique resource for a life course study of an indigenous population. A study protocol. *BMC Int Health Hum Rights*. (2003) 3:1. doi: 10.1186/1472-698X-3-1

17. Davison B, Goodall J, Whalan S, Montgomery-Quin K, Howarth T, Singh G. Nutritional dual burden in indigenous young adults: the geographical differential. *Aust J Rural Health*. (2019) 27:14–21. doi: 10.1111/ajr.12439

18. Krakauer NY, Krakauer JC. A new body shape index predicts mortality hazard independently of body mass index. *PLoS One*. (2012) 7:e39504. doi: 10.1371/journal.pone.0039504

19. Murphy JI, Weaver NE, Hendricks AE. Accessible analysis of longitudinal data with linear mixed effects models. *Dis Model Mech*. (2022) 15:dmm048025. doi: 10.1242/dmm.048025

20. R Core Team. *A language and environment for statistical computing. R foundation for statistical computing*. Vienna, Austria. Available at: https://wwwR-projectorg/: (2022).

21. Bates D, Mächler M, Bolker B, Walker S. Fitting linear mixed-effects models Usinglme4. *J Stat Softw*. (2015) 67:1–48. doi: 10.18637/jss.v067.i01

22. Lenth R. emmeans: estimated marginal means, aka least-squares means. R package version 187. Available at: https://CRANR-projectorg/package=emmeans. (2023).

23. Lindstrom L, Ahlsson F, Lundgren M, Bergman E, Lampa E, Wikstrom AK. Growth patterns during early childhood in children born small for gestational age and moderate preterm. *Sci Rep*. (2019) 9:11578. doi: 10.1038/s41598-019-48055-x

24. Knops NB, Sneeuw KC, Brand R, Hille ET, den Ouden AL, Wit JM, et al. Catch-up growth up to ten years of age in children born very preterm or with very low birth weight. *BMC Pediatr*. (2005) 5:26. doi: 10.1186/1471-2431-5-26

25. Takayanagi T, Shichijo A, Egashira M, Egashira T, Mizukami T. Extrauterine growth restriction was associated with short stature and thinness in very low birthweight infants at around six years of age. *Acta Paediatr*. (2019) 108:112–7. doi: 10.1111/apa.14433

26. Elmrayed S, Pinto J, Tough SC, McDonald SW, Scime NV, Wollny K, et al. Small for gestational age preterm infants and later adiposity and height: a systematic review and meta-analysis. *Paediatr Perinat Epidemiol*. (2023) 37:652–68. doi: 10.1111/ppe.13002

27. Balomenou F, Rallis D, Evangelou F, Zisi A, Balomenou K, Tsekas N, et al. Is small for gestational age status independently correlated with body composition during childhood? *Eur J Pediatr*. (2023) 182:661–8. doi: 10.1007/s00431-022-04723-1

28. Whalan S, Farnbach S, Volk L, Gwynn J, Lock M, Trieu K, et al. What do we know about the diets of aboriginal and Torres Strait islander peoples in Australia? A systematic literature review. *Aust N Z J Public Health*. (2017) 41:579–84. doi: 10.1111/1753-6405.12721

29. Brimblecombe JK, Ferguson MM, Liberato SC, O'Dea K. Characteristics of the community-level diet of aboriginal people in remote northern Australia. *Med J Aust*. (2013) 198:380–4. doi: 10.5694/mja12.11407

30. Mackerras DE, Reid A, Sayers SM, Singh GR, Bucens IK, Flynn KA. Growth and morbidity in children in the aboriginal birth cohort study: the urban-remote differential. *Med J Aust*. (2003) 178:56–60. doi: 10.5694/j.1326-5377.2003.tb05063.x

31. Sevoyan A, Davison B, Rumbold A, Moore V, Singh G. Examining the relationship between body mass index and adverse cardio-metabolic profiles among Australian indigenous and non-indigenous young adults. *Sci Rep*. (2019) 9:3385. doi: 10.1038/s41598-019-40083-x

32. Howarth T, Davison B, Singh G. Grip strength among indigenous and non-indigenous Australian adults: a longitudinal study of the effects of birth size and current size. *BMJ Open*. (2019) 9:e024749. doi: 10.1136/bmjopen-2018-024749

33. Bucens IK, Reid A, Sayers SM. Risk factors for reduced lung function in Australian aboriginal children. *J Paediatr Child Health*. (2006) 42:452–7. doi: 10.1111/j.1440-1754.2006.00896.x

34. Pearce MS, Mann KD, Singh G, Sayers SM. Birth weight and cognitive function in early adulthood: the Australian aboriginal birth cohort study. *J Dev Orig Health Dis*. (2014) 5:240–7. doi: 10.1017/S2040174414000063

35. Singh GR, Davison B, Ma GY, Eastman CJ, Mackerras DE. Iodine status of indigenous and non-indigenous young adults in the top end, before and after mandatory fortification. *Med J Aust*. (2019) 210:121–5. doi: 10.5694/mja2.12031

36. Khanna D, Peltzer C, Kahar P, Parmar MS. Body mass index (BMI): a screening tool analysis. *Cureus*. (2022) 14:e22119. doi: 10.7759/cureus.22119

37. Balomenou F, Rallis D, Evangelou F, Zisi A, Balomenou K, Tsekas N, et al. Growth trajectories during infancy have a significant impact on body composition in childhood. *Nutr Res*. (2023) 116:37–47. doi: 10.1016/j.nutres.2023.05.007

38. Wang Z, Hoy WE. Body mass index and mortality in aboriginal Australians in the Northern Territory. *Aust N Z J Public Health*. (2002) 26:305–10. doi: 10.1111/j.1467-842X.2002.tb00176.x

39. Adegbija O, Hoy WE, Dong B, Wang Z. Body mass index and waist circumference as predictors of all-cause mortality in an aboriginal Australian community. *Obes Res Clin Pract*. (2017) 11:19–26. doi: 10.1016/j.orcp.2016.06.003

40. Bhaskaran K, Dos-Santos-Silva I, Leon DA, Douglas IJ, Smeeth L. Association of BMI with overall and cause-specific mortality: a population-based cohort study of 3.6 million adults in the UK. *Lancet Diabetes Endocrinol*. (2018) 6:944–53. doi: 10.1016/S2213-8587(18)30288-2

41. Klatsky AL, Zhang J, Udaltsova N, Li Y, Tran HN. Body mass index and mortality in a very large cohort: is it really healthier to be overweight? *Perm J*. (2017) 21:16–142. doi: 10.7812/TPP/16-142

42. Sellers EA, Singh GR, Sayers SM. Large waist but low body mass index: the metabolic syndrome in Australian aboriginal children. *J Pediatr*. (2008) 153:222–7. doi: 10.1016/j.jpeds.2008.02.006

43. Lee A, Rainow S, Tregenza J, Tregenza L, Balmer L, Bryce S, et al. Nutrition in remote aboriginal communities: lessons from Mai Wiru and the Anangu Pitjantjatjara Yankunytjatjara lands. *Aust N Z J Public Health*. (2016) 40:S81–8. doi: 10.1111/1753-6405.12419

44. Goedegebuure WJ, Van der Steen M, Smeets CCJ, Kerkhof GF, Hokken-Koelega ACS. SGA-born adults with postnatal catch-up have a persistently unfavourable metabolic health profile and increased adiposity at age 32 years. *Eur J Endocrinol*. (2022) 187:15–26. doi: 10.1530/EJE-21-1130

45. Cohen CC, Harrall KK, Gilley SP, Perng W, Sauder KA, Scherzinger A, et al. Body composition trajectories from birth to 5 years and hepatic fat in early childhood. *Am J Clin Nutr*. (2022) 116:1010–8. doi: 10.1093/ajcn/nqac168

46. Denney-Wilson E, Fatema K, Elcombe E, Ingram S, Harris M, Comino E. Associations between rapid weight gain in infancy and weight status among urban aboriginal children participating in the Gudaga study: nine-year results from a cohort study. *BMC Pediatr*. (2020) 20:224. doi: 10.1186/s12887-020-02121-w

47. Grant JF, Chittleborough CR, Shi Z, Taylor AW. The association between a body shape index and mortality: results from an Australian cohort. *PLoS One*. (2017) 12:e0181244. doi: 10.1371/journal.pone.0181244

48. Mameli C, Krakauer NY, Krakauer JC, Bosetti A, Ferrari CM, Moiana N, et al. The association between a body shape index and cardiovascular risk in overweight and obese children and adolescents. *PLoS One*. (2018) 13:e0190426. doi: 10.1371/journal.pone.0190426

49. Dhana K, Kavousi M, Ikram MA, Tiemeier HW, Hofman A, Franco OH. Body shape index in comparison with other anthropometric measures in prediction of total and cause-specific mortality. *J Epidemiol Community Health*. (2016) 70:90–6. doi: 10.1136/jech-2014-205257

50. Ji M, Zhang S, An R. Effectiveness of a body shape index (ABSI) in predicting chronic diseases and mortality: a systematic review and meta-analysis. *Obes Rev*. (2018) 19:737–59. doi: 10.1111/obr.12666

51. Lee H, Chung HS, Kim YJ, Choi MK, Roh YK, Chung W, et al. Association between body shape index and risk of mortality in the United States. *Sci Rep*. (2022) 12:11254. doi: 10.1038/s41598-022-15015-x

52. Bouchi R, Asakawa M, Ohara N, Nakano Y, Takeuchi T, Murakami M, et al. Indirect measure of visceral adiposity "A body shape Index" (ABSI) is associated with arterial stiffness in patients with type 2 diabetes. *BMJ Open Diabetes Res Care*. (2016) 4:e000188. doi: 10.1136/bmjdrc-2015-000188

53. Bertoli S, Leone A, Krakauer NY, Bedogni G, Vanzulli A, Redaelli VI, et al. Association of Body Shape Index (ABSI) with cardio-metabolic risk factors: a cross-sectional study of 6081 Caucasian adults. *PLoS One*. (2017) 12:e0185013. doi: 10.1371/journal.pone.0185013

54. Christakoudi S, Tsilidis KK, Evangelou E, Riboli E. A body shape index (ABSI), hip index, and risk of cancer in the UK biobank cohort. *Cancer Med*. (2021) 10:5614–28. doi: 10.1002/cam4.4097

55. Ronn PF, Smith LS, Andersen GS, Carstensen B, Bjerregaard P, Jorgensen ME. Birth weight and risk of adiposity among adult Inuit in Greenland. *PLoS One*. (2014) 9:e115976. doi: 10.1371/journal.pone.0115976

56. Prioreschi A, Munthali RJ, Kagura J, Said-Mohamed R, De Lucia RE, Micklesfield LK, et al. The associations between adult body composition and abdominal adiposity outcomes, and relative weight gain and linear growth from birth to age 22 in the birth to twenty plus cohort, South Africa. *PLoS One*. (2018) 13:e0190483. doi: 10.1371/journal.pone.0190483

57. Masiakwala E, Nyati LH, Norris SA. The association of intrauterine and postnatal growth patterns and nutritional status with toddler body composition. *BMC Pediatr*. (2023) 23:342. doi: 10.1186/s12887-023-04155-2

58. Azcorra H, Varela-Silva MI, Dickinson F. Birth weight and body composition in 6-to-8 years old Maya children. *Am J Hum Biol*. (2021) 33:e23542. doi: 10.1002/ajhb.23542

59. Resende CB, Luft VC, Duncan B, Griep RH, Molina M, Barreto SM, et al. Birth weight and body fat mass in adults assessed by bioimpedance in the ELSA-Brasil study. *Cad Saude Publica*. (2021) 37:e00061619. doi: 10.1590/0102-311x00061619

60. Rockenbach G, Luft VC, Mueller NT, Duncan BB, Stein MC, Vigo A, et al. Sex-specific associations of birth weight with measures of adiposity in mid-to-late adulthood: the Brazilian longitudinal study of adult health (ELSA-Brasil). *Int J Obes*. (2016) 40:1286–91. doi: 10.1038/ijo.2016.76

61. Catapano A, Trinchese G, Cimmino F, Petrella L, D'Angelo M, Di Maio G, et al. Impedance analysis to evaluate nutritional status in physiological and pathological conditions. *Nutrients*. (2023) 15:2264. doi: 10.3390/nu15102264

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Honoring our teachings: children's storybooks as indigenous public health practice

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Introduction: American Indian and Alaska Native (AIAN) communities continue to flourish and innovate in the face of the COVID-19 pandemic. Storytelling is an important tradition for AIAN communities that can function as an intervention modality. To support the needs of AIAN children and caregivers, we (a collaborative workgroup of Indigenous health researchers) developed a culturally grounded storybook that provides pandemic-related public health guidance and mental health coping strategies woven with Inter-Tribal values and teachings.

Methods: A collaborative workgroup, representing diverse tribal affiliations, met via four virtual meetings in early 2021 to discuss evolving COVID-19 pandemic public health guidance, community experiences and responses to emerging challenges, and how to ground the story in shared AIAN cultural strengths. We developed and distributed a brief survey for caregivers to evaluate the resulting book.

Results: The workgroup iteratively reviewed versions of the storyline until reaching a consensus on the final text. An AI artist from the workgroup created illustrations to accompany the text. The resulting book, titled Our Smallest Warriors, Our Strongest Medicine: Honoring Our Teachings during COVID-19 contains 46 pages of text and full-color illustrations. An online toolkit including coloring pages, traditional language activities, and caregiver resources accompanies the book. We printed and distributed 50,024 physical copies of the book and a free online version remains available. An online survey completed by $N = 34$ caregivers who read the book with their child(ren) showed strong satisfaction with the book and interest in future books.

Discussion: The development of this storybook provides insights for creative dissemination of future public health initiatives, especially those geared toward AIAN communities. The positive reception and widespread interest in the storybook illustrate how braiding AIAN cultural teachings with public health guidance can be an effective way to disseminate health information. This storybook highlights the importance of storytelling as an immersive learning experience through which caregivers and children connect to family, community, culture, and public health guidance. Culturally grounded public health interventions can be effective and powerful in uplifting AIAN cultural values and promoting health and well-being for present and future generations.

KEYWORDS

culturally grounded, American Indian/Alaska native, storytelling, COVID-19, indigenous research

Introduction

American Indian and Alaska Native (AIAN) peoples are the original stewards of Turtle Island, including the so-called United States (U.S.), and share a long history of resistance and self-determination in the face of ongoing colonization and structural racism. There are currently 574 federally recognized AIAN tribes and additional state-recognized tribes, all of which are ethnically, culturally, and linguistically diverse (1). Centuries of ongoing structural oppression have led to inadequate education and health care, poverty, and disproportionate burden of disease for many AIAN communities (2, 3). Limited access to health care is also a challenge for many Tribal communities located in rural areas, and urban AIANs who have limited health care funding (4–6). Some AIAN peoples may also be hesitant to seek health care services due to historical and contemporary mistreatment by government institutions and health care systems (7). Together these factors led to the devastating and disparate impact of the COVID-19 pandemic in many AIAN communities. Morbidity and mortality rates due to COVID-19 among AIAN peoples were some of the highest in the nation at various points during the pandemic, despite AIANs making up only 2.9% of the U.S. population (3, 8). In the early stages of the pandemic, physical distancing and lockdowns disrupted cultural practices and ceremonies that are vital to the spiritual and mental health of AIANs (9). Additionally, the loss of Elders and other loved ones had devastating impacts on AIAN mental health (10). Given the increased burden and associated impacts on family, community, and ability to practice cultural traditions, it is not surprising that increased rates of depression, anxiety, and stress challenged many AIAN communities during the pandemic (11).

The pandemic also presented significant disruptions to the lives of many AIAN children and their caregivers. Complex and rapidly evolving COVID-19 mitigation measures caused changes to education, daily routines, and social connection for children (12). Closures and shifts to virtual and hybrid school and community programming changed access to school-based services, peer support, and social interaction with others beyond their household (13). Children are particularly susceptible to the mental health impacts of events like the pandemic due to developmental stages and limited cognitive ability to understand complex situations, to independently develop and use coping strategies, and to adequately communicate their feelings (12). In addition to new and exacerbated stressors from the pandemic, caregivers navigated communicating complicated information about the rapidly evolving pandemic with their children (14).

Despite facing escalating challenges during the COVID-19 pandemic, AIAN communities continue to flourish and innovate to meet community needs. Strong kinship networks and community-driven values foster mutual support and care when navigating challenges and this was especially present during the pandemic (15). Prompted by increased media attention on AIAN communities during the pandemic, many Native-led organizations continue to advocate for structural and policy reforms and to raise public awareness of ongoing health inequities AIANs face (16). Tribal Nations demonstrated sovereignty and tailored and implemented local COVID-19 prevention responses to meet the urgent needs of their communities (17). For example, some communities established mutual aid networks to provide services such as patient transport, household food delivery, and distribution of personal protective equipment (17). Due to community-led and culturally grounded vaccination efforts, AIANs have consistently had the highest COVID-19 vaccination rates of any racial or ethnic groups in the United States (18, 19).

Intergenerational knowledge, such as the passing on of Tribal languages, traditional ecological knowledge, oral storytelling, and cultural teachings, plays an important role in cultural and holistic well-being for many AIAN communities (20, 21). Storytelling in many AIAN communities is a traditional and dynamic way of sharing knowledge, entertainment, and cultural teachings that can be harnessed as part of interventions to promote well-being (22). Multiple health promotion programs have incorporated culturally relevant storybooks to engage AIAN children through storytelling and promote positive health outcomes (23). Indigenous communities across the Earth also engage in storytelling to support well-being. For example, researchers in Canada created storybooks to help facilitate healing and reduce mental health distress stemming from intergenerational colonial trauma (24). In Australia, art and storytelling represent important communication and knowledge transmission traditions. The West Australian Indigenous Storybook project created storybooks in collaboration with Aboriginal Elders and a steering committee to share important cultural stories (25). This project began partly in response to negative media portrayals of Aboriginal Australians in media, recognizing the need to combat the detrimental health impacts of negative representation of Indigenous peoples in media (25, 26).

Accurate portrayal of contemporary AIAN peoples can do more than just engage an audience in health education and promotion, it can also serve to increase positive representations of Indigenous communities and peoples today, rather than the romanticized and harmful depictions of AIANs often seen in popular media (27). There is a woeful lack of accurate, respectful representation of AIAN peoples and communities in media, which contributes to severely limited views and understandings of AIAN identities and leads to harmful stereotypes (e.g., AI mascots) (27–29) and negative health, mental health, and academic outcomes for AIAN youth and adults (29–32). However, in both community and academic spaces, storytelling is a sacred responsibility that requires deep intentionality. As described by Mallory Whiteduck: “When we write, Native writers are responsible to our families, our communities, and the larger Native academic community. Our stories represent a fundamental love and respect for our homeland, and writing them ensures our children can return home regardless of their physical location. Through stories we can achieve decolonization by responding to past and ongoing oppression, while actively moving beyond it” (33). Storybooks hold promise as a multi-faceted resource to improve health and well-being through storytelling that expands modern representation of AIAN communities, peoples, and strengths. Further, the writing of storybooks requires dedication to community, and to the very act of storytelling. Stories cannot be separated from the storyteller, and therefore developing storybooks requires a deeply committed, intentional, and thoughtful writing team dedicated to not only responding to oppression but also to demonstrating active resistance and flourishing beyond oppression.

Considering that the inclusion of storytelling and other culturally appropriate materials and methods increase the effectiveness and sustainability of Indigenous public health projects (34), in March 2020, the Johns Hopkins Center for Indigenous Health (CIH) adapted *My Hero is You* (35). *My Hero is You* is a storybook aimed at helping children across the globe cope with the mental and social health impacts brought by the COVID-19 pandemic and developed by the Inter-Agency Standing Committee Reference Group on Mental Health and Psychosocial Support in Emergency Settings (36). The book's approach of delivering guidance and teachings through storytelling made it a compelling candidate to address the pandemic's impact on AIAN youth and communities (35). However, *My Hero is* You, lacked cultural specificity and important context unique to the experiences of AI/ANs during the COVID-19 pandemic. Therefore, the CIH convened a collaborative working group of AI/AN child health and development experts who represented diverse tribal affiliations, as well as professional and personal experience with AIAN children. The adaptation and dissemination of this book, *Our Smallest Warriors, Our Strongest Medicine: Overcoming COVID-19 (OSWOSM1),* is detailed in a publication by O'Keefe and colleagues (35).

After the release and distribution of *OSWOSM1*, the COVID-19 pandemic continued, and the public health response shifted from immediate crisis management to long-term, sustainable initiatives. To meet the ongoing and emergent needs of AIAN children and their caregivers in the changing pandemic landscape, the CIH created a culturally grounded sequel to *OSWOSM1*. This sequel, *Our Smallest Warriors, Our Strongest Medicine: Honoring Our Teachings during COVID-19 (OSWOSM2)*, was developed to provide continued support for mental and emotional health for AIAN children and their caregivers, promote updated public health guidance related to COVID-19, and to elevate and center Indigenous cultural strengths and teachings (37). To create the storybook, the CIH team (authors TM, FG, MC, AI, EH, and VO'K) re-convened an Indigenous collaborative workgroup (authors JA-B, JJ, LM, CA, and MR) from the first book, *OSWOSM1,* with the addition of an Indigenous child well-being researcher (author JS). The members of the collaborative workgroup (herein, workgroup) hold diverse Tribal affiliations and personal (i.e., many are parents themselves) and professional expertise in various aspects of child development and well-being. This manuscript describes the development and dissemination of *OSWOSM2*, a culturally grounded AIAN children's storybook.

Methods

Each member of our storybook development team brought their own unique cultural values, experiences, and expertise. Further each member had a unique experience with the COVID-19 pandemic and these experiences implicitly and explicitly shaped the creation of the storybook. In Table 1, authors who engaged in the development of the book reflect on their involvement with creating the storybook and the specific topics or experiences they hoped the book would represent. By communicating the hopes and intentions of our authorship team, we demonstrate that the authorship team engaged in storybook development with intentionality in the hopes that this book would not only respond to the oppression of the ongoing COVID-19 pandemic, but tell a story of survivance and thriving in the face of adversity.

Storybook creation process

The development of the storyline for *OSWOSM2* took place over 6 weeks from January to February 2021 via iterative virtual meetings (i.e., we did not determine how many meetings would be held but met until we had achieved our goals). During the development process, the workgroup discussed rapid changes in the COVID-19 pandemic public health response since the development of *OSWOSM1* and how these changes have affected AIAN children and their caregivers. Through these discussions the workgroup considered numerous AIAN community strengths demonstrated during the COVID-19 pandemic, such as innovative public health responses (e.g., utilizing Tribal sovereignty to implement stay-at-home orders to reduce COVID-19 spread) and cultural values that promote community health and wellness (e.g., caring for Elders, safely maintaining social and cultural connection). The workgroup agreed it was important for the storybook characters to convey diverse experiences, like living in an urban or reservation environment, and to represent mixed racial and Tribal identities.

The team came to consensus to use a similar format to *OSWOSM1*. This included keeping the main characters, twins Tara and Virgil, using the four directions as a foundation for illustrating distinct pandemic experiences, including COVID-19-related public health guidance at the time of development, and harnessing cultural teachings throughout the book. Workgroup members recommended specific content to be represented in the story to mirror their own family and community experiences (e.g., drive-through birthday celebrations, receiving COVID-19 vaccines, and sending care packages to loved ones). In addition, the workgroup agreed to the importance of addressing children's mental and emotional health in the storybook (e.g., by discussing coping strategies for grief and disconnection resulting from social distancing measures). Workgroup members provided specific examples of how cultural teachings offer frameworks and value systems that have supported the health of Indigenous communities since time immemorial. The workgroup agreed that honoring cultural teachings during the COVID-19 pandemic was an appropriate theme for the book as it reflected the way that many Indigenous communities across Turtle Island (i.e., North America) were responding to the pandemic, while grounding the story in traditional values and knowledge. For example, Indigenous communities in Wisconsin highlight four main themes that support health and wellness during (and before and after) the pandemic: "(1) helping my people, (2) honoring our elders, (3) self-determination, and (4) living in a good way" (38). Workgroup members discussed witnessing and learning about similar cultural teachings embedded in public health responses to the pandemic in their own and other tribal communities. The working group did not encounter any significant disagreements throughout the storybook development process. However, occasionally some suggestions for

TABLE 1 Reflections from members of the storybook development team.

Team member (Tribal affiliation if applicable)	Reflections
Tara Maudrie (Sault Ste. Marie Tribe of Chippewa Indians)	As an Anishinaabe woman, I have always found comfort and strength in my cultural teachings, especially during times of hardship. Through the storyline, I hoped to reinforce what our people have always known—that our strength lies in our connection to one another, land, and culture. As a child I had very little representation of Indigenous peoples in media and books and what representation of Indigenous peoples existed did not reflect my experience. My hope was that this book provided positive representation of Indigenous peoples and encouraged people to learn about and lean on their teachings during hard times.
Fiona Grubin	As a non-Indigenous person who strives to be an ally, I am grateful to support this important endeavor. I hoped to be a part of elevating and bringing the visions of the collaborative workgroup to life and, in doing so, contributing to more positive representation of Indigenous peoples in media. Along the way I appreciated the opportunity to learn from the group and the values and teachings that were discussed and included in the book.
Jessica Ullrich (Inupiaq, Nome Eskimo Community)	Children's books are a wonderful way to learn and for parents to engage in conversations with their children about important life lessons and teachings. Stories are our way of teaching, healing, growing, and feeling. My hope with this book was for children to feel supported and connected through story, even as they were experiencing the challenges of navigating a pandemic.
Joshuaa Allison-Burbank (Diné and Acoma Pueblo)	Young Indigenous children interpret the world in unique ways. The way they see and understand the world reflects a worldview that prioritizes connection and kinship. During the pandemic, critical connections to family, community, and the land were disrupted and this altered the worldview of these young children. This book provided the opportunity for loved ones to openly discuss what was happening in their community and offered strategies on how to talk about traumatic experiences linked to the pandemic. Most importantly, the use of shared book reading is a powerful way for caregivers to bond with their children and offer comfort during through positive attachment. During a time of isolation and social distancing, offering children's books and strategies for how to implement shared book reading was a special way to deliver much needed caregiver support.
Lisa Martin (Sault Ste. Marie Tribe of Chippewa Indians)	Teaching our young people how to deal with hard things, like grief, by honoring our teachings provides them with tools and options for coping. In this book I wanted to uplift approaches to dealing with hard things that were grounded in the beliefs and cultural practices of Indigenous communities.
Crystal Austin (Diné Nation)	In this book, I wanted to represent my experience as a parent and validate the experiences of other parents, it was difficult for many parents to navigate online and hybrid school. The safety checklist shared in the book is something I practice with my children, as it helped them to manage anxiety and to feel a sense of control, especially when so much of our lives felt out of control. I also wanted to highlight the good things even during hard times, like staying connected when we are physically apart. As Indigenous peoples, no one is ever truly alone. Our kinship within our communities gives us a sense of identity and reminds us that we are responsible for one another. This sense of connection enables our survival and our thriving as Indigenous peoples, even during hard times.
Joelle Joyner (Kauwets'a:ka)	I wanted to make sure there was representation of Indigenous peoples through diverse features, clothing, and environments in contemporary Indigenous communities through the illustrations I created for the book.
Marcy Ronyak (Confederated Tribes of the Colville Reservation)	Being a part of a unique opportunity to share public health messaging with Indigenous communities, ties closely to our storytelling practices. It was a highlight of my federal career, being able to work with an outstanding group of Indigenous allies who wanted to make a difference within a variety of cultural settings that supported a bond with caregivers, health education, and an opportunity to deliver messaging during a traumatic timeframe in our nation. The hope was that the book would provide an opportunity for connectedness during a time of isolation and social distancing.
Emily Haroz	It has been a true gift to work on these projects. I am not Indigenous, but I work every day to be an ally to Indigenous peoples and communities. Focusing on children provides me with hope for the future. Seeing contemporary Indigenous families and children in print is good for all kids to help them better understand the strength of Indigenous peoples and practices.
Victoria O'Keefe (Cherokee Nation/Seminole Nation)	As a Cherokee/Seminole woman, my community and cultural values shape how I live and everything I do, including collaborating on this storybook series. Through this book series, we have illuminated the inherent and continued Indigenous strengths, values, and traditions that wrap us in love, care, and protection, even during trying times like COVID-19. These strengths, values, and traditions have been passed down to us through our ancestors and will uplift our communities for generations to come.

elements of the story (e.g., an animal) had different cultural meaning across working group members. When this occurred working group members discussed thoroughly the meaning of the suggested element within their respective cultures, before reaching a solution that had the intended meaning across the represented cultural groups within the working group.

Based on a storyboard collaboratively developed from meetings with the workgroup, the CIH team (authors TM, VO'K, and FG) drafted the initial storyline text. We incorporated feedback from caregivers who read *OSWOSM1* with their children. Caregivers recommended less text per page to help children better engage with the book and providing guidance to consider reading the book in multiple sessions. Facilitated by the CIH team, versions of the storyline were reviewed iteratively with the workgroup until consensus was reached on the book's final text. We did this through regular virtual meetings where the draft storybook content was presented, reviewed, and discussed by workgroup members until everyone agreed that the draft was final. Next, AI artist and workgroup member (author JJ) created illustrations to accompany the final text over 3 months (March–May 2021). Illustrations were reviewed by the workgroup to ensure resonated with the diversity of AIAN experiences represented in the storyline. Following this process, the CIH team worked to compile the final text and illustrations together to create *OSWOSM2*.

Dissemination plans

The CIH team planned to facilitate all dissemination of *OSWOSM2* using similar strategies employed for distributing *OSWOSM1*, including online through the CIH webpage, CIH social media platforms (Facebook, Instagram, and Twitter), and emails to all Tribal, Inter-Tribal, Urban Indian Health Programs, school-based, clinic-based, and home-visiting organizations who received print copies of *OSWOSM1* (35). In keeping with dissemination efforts of *OSWOSM1*, we also made an online request form available for Native-serving organizations to request bulk orders of print copies of the book for wider distribution within their community. These requests were shipped out on a first come, first serve basis as funding allowed. Distributing print copies of the book was important to the CIH team, as at least 31% of people living on Tribal reservations report having no or unreliable internet connection access (39). Bulk shipments were sent to CIH offices in the Southwest and Midwest where the books were shared with communities through programs and community networks. The final *OSWOSM2* book and accompanying resources were made available for free download on the CIH website.[1] We implemented a series of social media posts to advertise availability of the book and resources and to celebrate the content of the book.

Caregiver survey

In conjunction with *OSWOSM2* dissemination, we promoted an optional survey intended for parents and caregivers to complete after reading the book with their children. The goal of this survey was to briefly assess the general impact and reach the book had on AIAN children and families. The caregiver survey was hosted using Qualtrics (40), an online data collection platform, and distributed via links, and QR codes embedded in emails and social media posts about the storybook, as well as on the back of all print copies of the storybooks. 13 survey questions assessed overall satisfaction with the book, whether new information and skills related to the COVID-19 pandemic or mental health coping were learned through reading the book, satisfaction with the book's cultural teachings and illustration of modern day AIAN people, and whether respondents would be interested in future books of this nature. Two open-ended questions asked about (1) what topics respondents might like to see in future books and (2) whether respondents had any additional feedback to share about the book. Respondents who completed the survey were entered for a chance to win one of five $20 visa gift cards. The Johns Hopkins Bloomberg School of Public Health Institutional Review Board determined this survey as not human subjects research and did not require IRB oversight (BSPH IRB #00016998). We did not seek specific Tribal approvals for this survey as it was intended, like the book, to reach a broad audience of any AIAN people. Additionally, participation in the survey was voluntary and the broad goal was to generally learn about how the book was received to help us improve future, similar programming and resources.

Results

Our Smallest Warriors, Our Strongest Medicine: Honoring Our Teachings during COVID-19

The final *OSWOSM2* storybook contains 57 pages, with 29 pages that include storyline text and 46 pages with full-page illustrations. Figures 1, 2 below illustrate examples COVID-19 messaging and illustrations from the final storybook. An online toolkit accompanied the book including: six coloring pages, six traditional language activities, a two page resource for reading and talking about the pandemic with children, a vocabulary page with relevant words from the story, and a worksheet to encourage reflection on the readers' cultural teachings. The storyline presented in *OSWOSM2* provides public health information and mental health education for children and parents that is grounded in the strengths and teachings of Indigenous communities.

The reader accompanies twins Tara and Virgil as they encounter emerging public health initiatives to mitigate the effects of the COVID-19 pandemic while maintaining social connections with friends and family (characters were introduced in *OSWOSM1*). Through their family the twins learn about vaccination efforts in their own community, and they learn from their friends located in the four directions about how they are staying connected and coping with the pandemic. Each of the four friends offers a different teaching accompanied by practical advice on how children can apply the teaching to promote well-being. For example, the Twins' friend Daniel shares through a letter how COVID-19 continues to impact his family's life and that when he feels scared or worried, he can talk about his feelings with family members. He says that his mom taught him that he carries the strength of his ancestors and that helps him feel strong. Later, the twins share with their family that Daniel taught them it is okay to feel all of their emotions, which reflects cultural values of holistically honoring all aspects of health, including one's emotions. The story concludes with the twins sharing with their family what they learned from each of their friends' experiences, cultural teachings, and their own message of hope for the future.

Dissemination

Our Smallest Warriors, Our Strongest Medicine: Honoring Our Teachings during COVID-19 books reached a wide variety of communities. In total, 50,024 print copies of books were distributed across all 12 of the Indian Health Service (IHS) regional areas in the United States, reaching 28 states, 105 tribes, and two First Nations communities in Canada (Figure 3). The books were shared with Indigenous families by 60 different urban and Inter-Tribal

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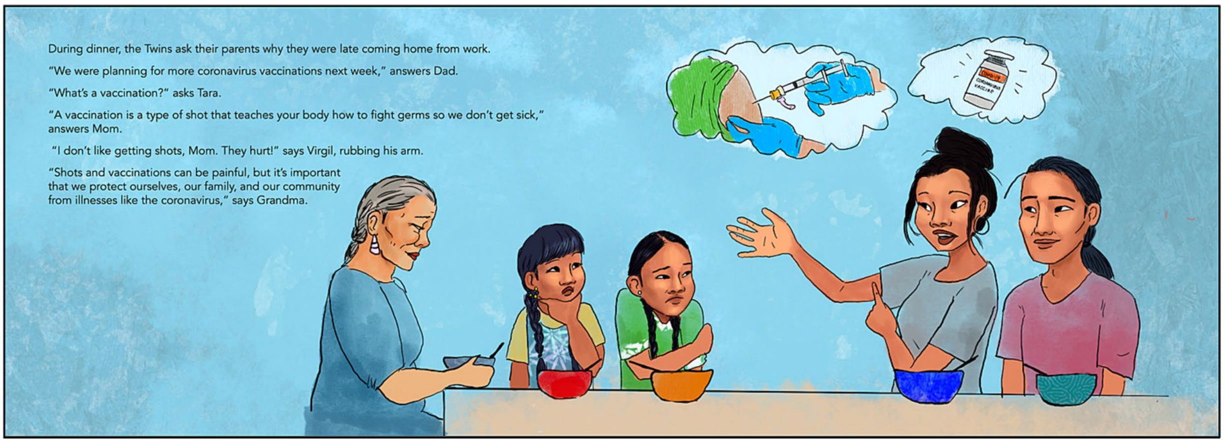
[1] bit.ly/cih-oswosm2



During dinner, the Twins ask their parents why they were late coming home from work.

"We were planning for more coronavirus vaccinations next week," answers Dad.

"What's a vaccination?" asks Tara.

"A vaccination is a type of shot that teaches your body how to fight germs so we don't get sick," answers Mom.

"I don't like getting shots, Mom. They hurt!" says Virgil, rubbing his arm.

"Shots and vaccinations can be painful, but it's important that we protect ourselves, our family, and our community from illnesses like the coronavirus," says Grandma.

FIGURE 1
OSWOSM2 storybook page about vaccines. Image credit: JJ.

"How are you, Sophie?" asks Tara.

Sophie tells the Twins that she has been feeling sad because her Grandmother died after getting sick from the coronavirus. "My Mom says that it's okay to be sad and miss my Grandma," says Sophie.

"When I miss her a lot, I try to think about all of the teachings I learned from my Grandma," Sophie tells the Twins. "That's why I go outside a lot because my Grandma taught me that we come from the land and to be grateful for the ways the land takes care of our body, heart, mind, and spirit."

FIGURE 2
OSWOSM2 storybook page about grief, healing, and land. Image credit: JJ.

organizations, 35 IHS clinics, 40 educational organizations, 20 Head Start programs (most of them Tribal Head Start programs), and 12 Family Spirit® Tribal home visiting affiliates (41). Between June 2021 and February 2023, CIH developed and published 14 social media posts promoting *OSWOSM2* on CIH accounts. These posts achieved a combined total reach of 12,284 impressions on Facebook, 12,274 impressions on Twitter, and 3,798 impressions on Instagram. The CIH was undergoing a re-branding and re-naming (formerly Center for American Indian Health) transition during the time books were being distributed which unfortunately meant we were unable to access relevant analytic information from the CIH website related to electronic downloads of the book.

Caregiver survey results

A total of 34 responses to the caregiver survey were determined to be valid for analysis. We implemented survey protections such as reCAPTCHA and responses with a high score for risk of being fraudulent were not analyzed. Of these 34 included responses, most respondents ($n = 30$) reported that a parent, caregiver, or other adult read the book with a child, and five of these people also reported the child had read the book on their own. When asked to rate their satisfaction with the book from 0 (not at all) to 5 (completely), the average response from $n = 23$ respondents was 4.8, indicating very high overall satisfaction. No responses below a four were received for this question. Satisfaction with the book's cultural teachings and representation of AIAN peoples through text and illustration was also high. 94% of respondents reported being extremely or somewhat satisfied with the cultural teachings presented in the books, 90% reported extremely or somewhat satisfied with the book's illustration and text depicting and describing modern AIAN peoples.

The data also show that most families who completed the survey learned new information about mental health coping and preventing COVID-19 from reading the book. 90% of respondents said that they had probably or definitely learned something about mental health coping, while 79% reported probably or definitely learning something about COVID-19 prevention. All but one respondent shared they

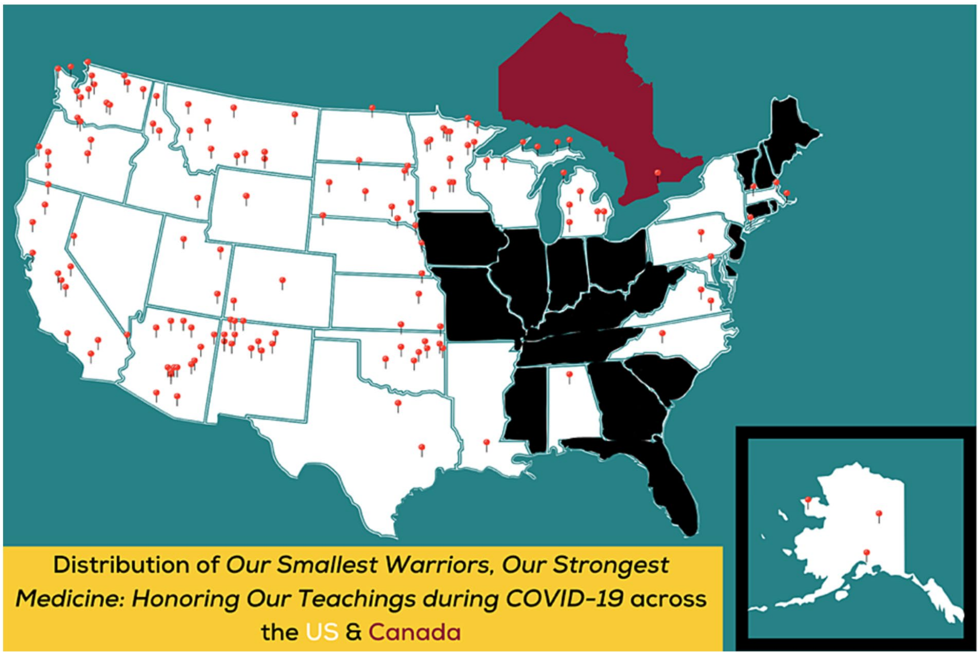


FIGURE 3
Distribution of OSWOSM2.

would be interested in future books on other health/wellness topics, and 16 people shared open-ended topic ideas for future books including mental health, language books, diabetes education, and wellness. Lastly, $n = 12$ respondents noted open-ended feedback about the book, all of which were positive and featured messages such as how much children enjoyed the books, appreciation for the Tribal traditions reflected in the book, and gratitude for the book. One respondent shared, “My kids love the illustrations of the book and said they were excited to see Natives like themselves. They mentioned that was them on the book or it was their cousins. They connected with the book and told their friends about it.” Another caregiver shared, “Thank you. The book made a lot of children very happy when they received it. It showed that we all can get COVID-19, and that it reflected tribal traditions and community.”

Discussion

The *OSWOSM* storybooks represent a resurgence and revitalization of Indigenous ways of knowing and being, which cultivate strength and innovation (42). Through developing *OSWOSM2* in collaboration with an Indigenous Workgroup, we used storytelling as a creative way to share public health information with children and their caregivers. As researchers who employ a Community-Based Participatory Research (CBPR) approach, developing and distributing the book to AIAN children and families was a form of creative dissemination and public service. Responding to the health needs of our communities is emblematic of our duty care for our communities in line with a CBPR approach. Our evaluation of the book showed that readers reported high satisfaction with the book and indicated that they learned new things about mental health coping and preventing COVID-19, further highlighting the power of creative dissemination and positive representation of Indigenous peoples in media.

Since time immemorial, Indigenous communities have told stories as a way to pass knowledge and teachings intergenerationally, to facilitate connectedness, and to share experiences across time (43). Engaging with storytelling is an important part of AIAN children’s development as it integrates dynamic learning with socialization and passes on cultural values and teachings in an engaging and developmentally appropriate way (44). Indigenous storytellers often tell the same stories multiple times, as with each telling there is something new to learn based on the listener’s growth, development, understanding, and life experience (45). Storytelling is both a practice and an Indigenous research method that provides an opportunity for children and their caregivers to actively engage in learning as they gather information from a story and return to that information iteratively as they navigate similar situations in their day to day life, encouraging them to continue to connect more deeply to different aspects of the story throughout time (43). Within a story are embedded values, emotions, behavioral actions, relational connections, and teachings that guide children on ways to live and be in the world in response to life’s joys, celebrations, and challenges.

Using a storytelling approach in *OSWOSM2* was not only responsive to the needs of children and their caregivers for easily understandable public health guidance, but reflective of the power of AIAN storytelling and Indigenous research methods to provide validation, and to facilitate connection, reflection, and discussion (43). An Ojibwe storyteller, writer, and scholar, Leanne Betasamosake Simpson, elaborates on the power of Indigenous storytelling: “For me, storytelling is a way of connecting to the land, and it is a way of connecting with the past, and it is also a way of connecting to the future…People are asked to see themselves in the story, to carry the story, to work with the story, and to find their own meaning within a story. Telling my children stories is like planting little seeds inside them. What I hope as a parent is that I have given my kids this garden of stories and then when life brings them challenges, they will have this body of knowledge that will provide them with comfort" (45). The format, content, and illustrations of *OSWOSM2* encourage children and their caregivers to see themselves in the story—these efforts were validated by caregiver responses to surveys which highlighted that children saw aspects of themselves and their experiences mirrored in the story. Integrating aspects of Indigenous storytelling with public health guidance encouraged children to process their emotions surrounding the pandemic and emphasized the power of cultural teachings to provide comfort and guidance as children navigate challenges. *OSWOSM2* builds on the rich tradition of AIAN storytelling through a storybook to provide children with a space to think, feel, learn, and gain wisdom, both from ancestral teachings and through their own family and community's response to the pandemic.

Efforts to mitigate the spread of COVID-19 often required communities to implement closures of schools and community programs. As public health understanding of the pandemic advanced, it became evident that a full return to in-person school would be delayed and that support was needed to help children navigate difficult learning and life routines (46). *OSWOSM2* responds to the needs of AIAN children and their caregivers by embedding child mental health practices in the story like spending time outside and practicing sharing feelings. The efforts to develop a culturally grounded storybook were aligned with our teams' values to honor community strengths and serve community needs and are one example of putting CBPR principles into practice (47, 48). The storybook is a creative, fast way to distribute much-needed mental health support to AIAN children that elevates protective AIAN values and beliefs. The storybook both represents and fosters creative expression, which contributes to positive connection with individual and community identities, provides a platform to share information about coping skills, and can augment the CBPR process (49). *OSWOSM2* responded to community feedback from *OSWOSM1*, integrating specific recommendations from our informal evaluation to meet the needs of AIAN children and their caregivers. Rapid dissemination of *OSWOSM* books during the COVID-19 pandemic aligns with CBPR values of being action-oriented with specific intent to benefit the community (47).

This project represents an example of an innovative approach to public health communication that is aligned with Indigenous methodologies and strengths. The second book in the *Our Smallest Warriors* series builds on the strengths of the previous book and met the demand for COVID-19 resources specifically for AIAN children that are reflective of evolving public health guidance. However, *OSWOSM2* is a culturally built and culturally grounded resource, where *OSWOSM1* is a cultural adaptation of an existing storybook. Culturally grounded resources offer significant advantages for Indigenous communities as they are created from community values and strengths, and therefore have stronger acceptability to community (50, 51). Despite its strengths, there are important limitations to consider when interpreting the results. While the creation of the book was done in a way to be inclusive and representative of many Indigenous Nations, there was no feasible way to incorporate all 574 federally recognized Tribal Nations, nor all state recognized tribes in a comprehensive way. Images and storylines may not generalize to all communities and peoples. The caregiver survey was voluntary and based on convenience sampling. This approach introduces potential sampling and selection bias, and we cannot be sure our results generalize to all people who interacted with the book and associated materials. However, as this was an example of public health practice, generalizability of results was not the intention. Rather, our goal with the survey and results is to be transparent and accountable to the greater community and to continuously improve our processes for future work related to storytelling as a public health modality. Further, our evaluation efforts did not specifically assess the acceptability or impact of messaging on children or their caregivers. Future evaluation efforts of children's storybooks may consider more robust evaluation efforts in order to make stronger conclusions about the impacts of storybook messaging.

In Indigenous communities storytelling is an intergenerational practice that builds on cultural traditions and teachings and encourages active learning. Our use of a storybook series brings this powerful tradition to the daily lives of AIAN children and families while serving as an Indigenous public health intervention. This storybook not only provided a health resource during a specific point in time (i.e., the COVID-19 pandemic), but created an opportunity for children, families, and communities to uplift and carry on cultural values and traditions now and in the future. We hope the *Our Smallest Warriors* series will inspire future culturally grounded efforts that braid Indigenous methodologies with public health dissemination.

Data availability statement

The datasets presented in this article are not readily available because data are not publicly available. Requests to access the datasets should be directed to TM, tmaudri1@jhu.edu.

Ethics statement

Ethical approval was not required for the study involving humans in accordance with the local legislation and institutional requirements. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent to participate in this study was not required from the participants in accordance with the national legislation and the institutional requirements.

Author contributions

TM: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Project administration, Resources, Writing – original draft, Writing – review & editing, Validation. FG: Conceptualization, Data curation, Formal analysis, Funding acquisition, Project administration, Resources, Writing – original draft, Writing – review & editing. MC: Data curation, Formal analysis, Project administration, Validation, Writing – original draft. JV: Data curation, Formal analysis, Writing – original draft. JS: Conceptualization, Writing – original draft. JA-B: Conceptualization, Writing – original draft. LM: Conceptualization, Writing – review & editing. CA: Conceptualization, Writing – review & editing. JJ: Conceptualization, Visualization, Writing – review & editing. MR: Conceptualization, Funding acquisition, Writing – review & editing.

KM: Writing – review & editing, Visualization. AI: Writing – review & editing, Project administration. EH: Conceptualization, Funding acquisition, Supervision, Writing – original draft, Writing – review & editing. VO'K: Conceptualization, Data curation, Funding acquisition, Investigation, Supervision, Writing – original draft, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

References

1. National Congress of American Indians (2020). Tribal nations and the United States: an introduction. Available at: https://www.ncai.org/tribalnations/introduction/Indian_Country_101_Updated_February_2019.pdf

2. Indian Health Service (2019). Indian health disparities. Available at: https://www.ihs.gov/sites/newsroom/themes/responsive2017/display_objects/documents/factsheets/Disparities.pdf

3. Aulandez KMW, Walls ML, Weiss NM, Sittner KJ, Gillson SL, Tennessen EN, et al. Cultural sources of strength and resilience: a case study of holistic wellness boxes for COVID-19 response in indigenous communities. *Front Sociol.* (2021) 6:612637. doi: 10.3389/fsoc.2021.612637

4. Rural Health Information Hub (2022). Rural tribal health. Available at: https://www.ruralhealthinfo.org/topics/rural-tribal-health

5. Warne D, Frizzell LB. American Indian health policy: historical trends and contemporary issues. *Am J Public Health.* (2014) 104:S263–7. doi: 10.2105/AJPH.2013.301682

6. Trahant MN. The story of Indian health is complicated by history, shortages & bouts of excellence. *Daedalus.* (2018) 147:116–23. doi: 10.1162/DAED_a_00495

7. Guadagnolo BA, Cina K, Helbig P, Molloy K, Reiner M, Cook EF, et al. Medical mistrust and less satisfaction with health care among native Americans presenting for cancer treatment. *J Health Care Poor Underserved.* (2009) 20:210–26. doi: 10.1353/hpu.0.0108

8. Indian Country Today (2021). Census: Native population increased by 86.5 percent. Available at: https://indiancountrytoday.com/news/2020-census-native-population-increased-by-86-5-percent#:%7E:text=The%20American%20Indian%20and%20Alaska%20Native%20population%2C%20alone%20and%20in,percent%20of%20the%20U.S.%20population

9. Koithan M, Farrell C. Indigenous native American healing traditions. *J Nurse Pract.* (2010) 6:477–8. doi: 10.1016/j.nurpra.2010.03.016

10. Solomon TGA, Starks RRB, Attakai A, Molina F, Cordova-Marks F, Kahn-John M, et al. The generational impact of racism on health: voices from American Indian communities. *Health Aff.* (2022) 41:281–8. doi: 10.1377/hlthaff.2021.01419

11. Tyra AT, Ginty AT, John-Henderson NA. Emotion regulation strategies predict PTSD during the COVID-19 pandemic in an American Indian population. *Int J Behav Med.* (2021) 28:808–12. doi: 10.1007/s12529-021-09964-2

12. Imran N, Zeshan M, Pervaiz Z. Mental health considerations for children & adolescents in COVID-19 pandemic. Pak. *J Med Sci.* (2020) 36:S67–72. doi: 10.12669/pjms.36.COVID19-S4.2759

13. de Figueiredo CS, Sandre PC, Portugal LCL, Mázala-de-Oliveira T, da Silva CL, Raony Í, et al. COVID-19 pandemic impact on children and adolescents' mental health: biological, environmental, and social factors. *Prog Neuro-Psychopharmacol Biol Psychiatry.* (2021) 106:110171. doi: 10.1016/j.pnpbp.2020.110171

14. Dalton L, Rapa E, Stein A. Protecting the psychological health of children through effective communication about COVID-19. *Lancet Child Adolesc Health.* (2020) 4:346–7. doi: 10.1016/S2352-4642(20)30097-3

15. Palimaru AI, Brown RA, Arvizu-Sanchez V, Mike L, Etz K, Johnson CL, et al. Risk and resilience among families in urban AI/AN communities: the role of young adults. *J Racial Ethn Health Disparities.* (2023) 10:509–20. doi: 10.1007/s40615-022-01240-7

16. Native Ways Federation (2023). Native nonprofit day 2023. Available at: https://www.nativeways.org/native-nonprofit-day/

17. Foxworth R, Redvers N, Moreno MA, Lopez-Carmen VA, Sanchez GR, Shultz JM. Covid-19 vaccination in American Indians and Alaska natives—lessons from effective community responses. *N Engl J Med.* (2021) 385:2403–6. doi: 10.1056/nejmp2113296

18. Ellenwood R, Boyd AD, Higheagle SZ. Communication and perspectives about COVID-19 and vaccinations among native Americans. *Sci Commun.* (2023) 45:172–94. doi: 10.1177/10755470221151184

19. Haroz EE, Kemp CG, O'Keefe VM, Pocock K, Wilson DR, Christensen L, et al. Nurturing innovation at the roots: the success of COVID-19 vaccination in American Indian and Alaska native communities. *Am J Public Health.* (2022) 112:383–7. doi: 10.2105/ajph.2021.306635

20. Substance Abuse and Mental Health Services Administration, Indian Health Service, & National Indian Health Board (2016). The national tribal behavioral health agenda. Available at: https://store.samhsa.gov/sites/default/files/d7/priv/pep16-ntbh-agenda.pdf

21. Ullrich JS. For the love of our children: an indigenous connectedness framework. *Alternatives.* (2019) 15:121–30. doi: 10.1177/1177180119828114

22. Hodge FS, Pasqua A, Marquez CA, Geishirt-Cantrell B. Utilizing traditional storytelling to promote wellness in American Indian communities. *J Transcult Nurs.* (2002) 13:6–11. doi: 10.1177/104365960201300102

23. Satterfield D, DeBruyn L, Lofton T, Francis CD, Zoumenou V, DeCora L, et al. "Make stories that will always be there": eagle Books' appeal, sustainability, and contributions to public health, 2006-2022. *Prev Chronic Dis.* (2023) 20:E26. doi: 10.5888/pcd20.220315

24. Cooper EJ, Driedger SM. Creative, strengths-based approaches to knowledge translation within indigenous health research. *Public Health.* (2018) 163:61–6. doi: 10.1016/j.puhe.2018.06.020

25. Stoneham M, Davies CR, Christophers R. The Western Australian indigenous storybook spins special yarns. 2021 mar 30 In: JH Corbin, M Sanmartino, EA Hennessy and HB Urke, editors. *Arts and Health Promotion: Tools and Bridges for Practice, Research, and Social Transformation.* Cham (CH): Springer (2021)

26. Balvin N, Kashima Y. Hidden obstacles to reconciliation in Australia: the persistence of stereotypes In: D Bretherton and N Balvin, editors. *Peace Psychology in Australia*. New York, NY: Springer Science + Business Media (2011). 197–219.

27. IllumiNative (2018). Reclaiming native truth research findings: compilation of all research. Available at: https://illuminative.org/wp-content/uploads/2022/06/FullFindingsReport-screen-spreads-1.pdf

28. Leavitt PA, Covarrubias R, Perez YA, Fryberg SA. "Frozen in time": the impact of native American media representations on identity and self-understanding. *J Soc Issues*. (2015) 71:39–53. doi: 10.1111/josi.12095

29. Fryberg SA, Markus HR, Oyserman D, Stone JM. Of warrior chiefs and Indian princesses: the psychological consequences of American Indian mascots. *Basic Appl Soc Psychol*. (2008) 30:208–18. doi: 10.1080/01973530802375003

30. O'Keefe VM, Wingate LR, Cole AB, Hollingsworth DW, Tucker RP. Seemingly harmless racial communications are not so harmless: racial microaggressions Lead to suicidal ideation by way of depression symptoms. *Suicide Life Threat Behav*. (2015) 45:567–76. doi: 10.1111/sltb.12150

31. Walls ML, Gonzalez J, Gladney T, Onello E. Unconscious biases: racial microaggressions in American Indian health care. *J Am Board Fam Med*. (2015) 28:231–9. doi: 10.3122/jabfm.2015.02.140194

32. Davis-Delano L, Gone JP, Fryberg SA. The psychosocial effects of native American mascots: a comprehensive review of empirical research findings. *Race Ethn Educ*. (2020) 23:613–33. doi: 10.1080/13613324.2020.1772221

33. Whiteduck M. "But it's our story. Read it.": stories my grandfather told me and writing for continuance. *Decoloniz Indig Educ Soc*. (2013) 2:72–92.

34. Mehl-Madrona L. What traditional indigenous elders say about cross-cultural mental health training. *Explore*. (2009) 5:20–9. doi: 10.1016/j.explore.2008.10.003

35. O'Keefe VM, Maudrie TL, Ingalls A, Kee C, Masten KL, Barlow A, et al. Development and dissemination of a strengths-based indigenous children's storybook: "our smallest warriors, our strongest medicine: overcoming COVID-19". *Front Sociol*. (2021) 6:611356. doi: 10.3389/fsoc.2021.611356

36. Inter-Agency Standing Committee (2020). My Hero is you, storybook for children on COVID-19. Available at: https://interagencystandingcommittee.org/iasc-reference-group-mental-health-and-psychosocial-support-emergency-settings/my-hero-you

37. Center for American Indian Health (n.d.). Our Smallest Warriors, Our Strongest Medicine: Honoring Our Teachings during COVID-19. Available at: https://caih.jhu.edu/programs/oswosm-honoring-our-teachings/

38. Dellinger M, Poupart AE. Lessons native American culture can teach us about resilience during pandemics and health care crises. *WMJ*. (2021) 120:S80–4.

39. American Indian Policy Institute (2020). Tribal digital divide policy brief and recommendations. Available at: https://aipi.asu.edu/sites/default/files/tribal_digital_divide_stimulus_bill_advocacy_04032020.pdf

40. Qualtrics, Provo, UT (2020). Version [version] of Qualtrics. Copyright © 2020 Qualtrics. Available at: https://www.qualtrics.com

41. Johns Hopkins Center for Indigenous Health (n.d). Family Spirit: promoting maternal and early child health. Available at: https://familyspiritprogram.org/

42. Watson A, Purkey E, Davison CM, Fu M, Nolan D, Mitchell D, et al. Indigenous strength: braiding culture, ceremony and community as a response to the COVID-19 pandemic. *Int J Indig Health*. (2022) 17:111–21. doi: 10.32799/ijih.v17i1.36716

43. Iseke A. Indigenous storytelling as research. *Int Rev Qual Res*. (2013) 6:559–77. doi: 10.1525/irqr.2013.6.4.559

44. Cajete GA. Children, myth, and storytelling: an indigenous perspective. *Glob Stud Child*. (2017) 7:113–30. doi: 10.1177/2043610617703832

45. Betasamosake Simpson L (2019). Reimagining the rich tradition of indigenous storytelling. Faith & Leadership. Available at: https://faithandleadership.com/leanne-betasamosake-simpson-reimagining-the-rich-tradition-indigenous-storytelling

46. Araújo LA, Veloso CF, Souza MC, Azevedo JMC, Tarro G. The potential impact of the COVID-19 pandemic on child growth and development: a systematic review. *J Pediatr*. (2021) 97:369–77. doi: 10.1016/j.jped.2020.08.008

47. Israel BA, Schulz AJ, Parker EA, Becker AB. Review of community-based research: assessing partnership approaches to improve public health. *Annu Rev Public Health*. (1998) 19:173–202. doi: 10.1146/annurev.publhealth.19.1.173

48. Burke JG, Hess S, Hoffmann K, Guizzetti L, Loy E, Gielen A, et al. Translating community-based participatory research principles into practice. *Prog Community Health Partnersh*. (2013) 7:115–22. doi: 10.1353/cpr.2013.0025

49. Gray N, Oré de Boehm C, Farnsworth A, Wolf D. Integration of creative expression into community-based participatory research and health promotion with native Americans. *Fam Commun Health*. (2010) 33:186–92. doi: 10.1097/FCH.0b013e3181e4bbc6

50. Okamoto SK, Kulis S, Marsiglia FF, Steiker LK, Dustman P. A continuum of approaches toward developing culturally focused prevention interventions: from adaptation to grounding. *J Prim Prev*. (2014) 35:103–12. doi: 10.1007/s10935-013-0334-z

51. Walters KL, Johnson-Jennings M, Stroud S, Rasmus S, Charles B, John S, et al. Growing from our roots: strategies for developing culturally grounded health promotion interventions in American Indian, Alaska native, and native Hawaiian communities. *Prev Sci*. (2020) 21:54–64. doi: 10.1007/s11121-018-0952-z

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A community-embedded approach to increasing the health literacy of Aboriginal children in a regional area: processes of co-design and local implementation

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Purpose: This research explores the implementation of a child-centred, co-designed, community-embedded program called 'Young Doctors for Life' (YDFL). YDFL is designed to improve health and wellbeing outcomes for Aboriginal children in the middle childhood years. Focus is given in this paper to the processes of program adaptation of the YDFL to ensure local cultural relevance, drawing on the experiences and perspectives of children, parents, schoolteachers, and the implementation team.

Method: Two focus groups with program stakeholders were convened. The first group consisted of three members from the local Aboriginal implementation team, and the second group comprised two members of the program design team. Children ($n = 22$) and schoolteachers ($n = 2$) participated in semi-structured interviews. Parent survey data ($n = 16$) were also collected and included. The data was analysed, guided by the five elements of implementation as outlined in the Hexagon Implementation framework (Capacity; Fit; Need; Usability; Support; and Evidence), which served as *a priori* themes.

Results: YDFL provides a promising example of how programs can be adapted with and for Aboriginal communities to support child health. Successful adaptation and implementation of this program required a co-design approach engaging program designers and the local implementation team. Community collaboration was also essential to identifying and addressing local community goals and aligning new programs with local service and cultural contexts.

Conclusion: Health programs to support positive child outcomes are more likely to be successful when they share their focus between the risks and challenges within a community, and the positive, protective factors that can be leveraged to support children to flourish. Stakeholder engagement and community leadership are necessary to achieve meaningful program adaptation and implementation in Aboriginal communities.

KEYWORDS

Aboriginal health, child-centred, co-design, community-embedded, implementation

1 Background and introduction

The 'Young Doctors for Life' (YDFL) program is run by the Malpa Project for Australian Aboriginal[1] children in their middle childhood years. It is a child-centred health intervention program designed to support improved child health outcomes in Aboriginal communities.

A particular focus in the program is health literacy and the role that children can play in advocating for positive health behaviours across their communities. The YDFL program is implemented only by invitation of community leaders and requires the formation of a local Aboriginal leadership and implementation team. Before the program commences with children, the local team assesses and adapts the program to embed local cultural elements and blend Western health practices with traditional knowledges. Adaptation of a program in response to local culture and needs is argued within the research literature to be critical to program sustainability and successful outcomes (1). In the Australian context, there is a strong call for Aboriginal people and communities to be at the centre of the service decision-making processes (2). This paper describes the local process of YDFL adaptation and implementation in one regional community, and how the program was received and experienced by local stakeholders.

Australian Aboriginal communities experience significant health inequities, similar to those of other Indigenous peoples in colonised nations (3, 4). These inequities are evident from childhood. For example, In Australia, Aboriginal peoples experience the highest documented burden of Rheumatic Heart Disease in the world (5), 90% of Aboriginal children living in remote areas have some form of Otitis Media (6) and there is a trachoma endemic within regional, rural, and remote Aboriginal communities, with Australia bewilderingly remaining the only high-income country reporting such widespread trachoma (7). The observation that, overall, health inequities for Aboriginal people continue despite significant government investment (2) leaves little doubt that there is a need to re-think the approaches and paradigms that underpin health service provision for Aboriginal children and their communities.

Reflective of a difficult socio-political and historical landscape, a discussion of the factors that underpin current health inequities experienced by Aboriginal children and their communities are beyond the scope of this paper. However, it is important to acknowledge that Australian Aboriginal communities experience high levels of intergenerational trauma as the result of strong state interventions, including: state-sanctioned child removal policies between 1909–1969 (8); the ongoing over-representation of Aboriginal children in the child protection system (9); high rates of incarceration (10, 11); and the ongoing struggles for the recognition of Aboriginal peoples as the custodians of their lands (12). An understanding of the complexity of this history and the impact of ongoing systemic racism is essential to redressing health inequities and honouring the resilience and self-determination of Aboriginal communities (13).

In addition to historical factors, research documents the complex array of social determinants associated with poor health outcomes in Aboriginal communities, including inadequate and overcrowded living conditions, and low levels of educational attainment, employment, and income (13). In contrast, the social determinants associated with positive health outcomes for Aboriginal communities include cultural identity, connection to kinship, knowledge and beliefs, language and participation in cultural activities, access to traditional lands and caring for country (14).

There is a growing number of researchers who argue that the investment in health programs has not achieved the anticipated improvements in health outcomes because of the lack of community co-design and consultation, which has rendered programs meaningless to communities and unsustainable (15, 16).

A major criticism of Aboriginal health programs is that they are frequently imposed on communities by service organisations who lack an understanding of the communities they intend to assist, who decide what problems exist and develop solutions without engaging in respectful communication or shared decision-making with the community (2, 17).

1.1 Culturally meaningful program implementation

This paper aims to explore the implementation of a child-centred health intervention program which aims to improve the health literacy of Indigenous children, and to encourage them to be health advocates within their community. An implementation science approach underpins this study, which employs a case study approach to examine the local adaptations that are necessary to ensure the program is locally meaningful, and the perceived impacts of the program. The voices of key stakeholders, including children, parents, teachers and the local implementation team are privileged in this study. The study seeks to contribute to the body of research on the importance of child-centred, co-designed, community embedded approaches to implementation to support meaningful outcomes and sustainability. When Aboriginal people are asked about their aspirations for health service improvement, focus is often given to the centrality of relationships rather than a focus on clinical disease. For example, Smith and colleagues interviewed 60 Aboriginal people of the Maningrida community in Arnhem Land (15). The participants spoke of the Maningrida construct of *urrutu*, which is the relationship between an individual, their kin, and their Country. They described wanting a public health care service built on trust and relationships, delivered by language-congruent Aboriginal health professionals guided by panels based on language groups. Bulloch and colleagues conducted case studies of three Aboriginal health and wellbeing services (18). All three services emphasised the importance of a community-driven approach that employed holistic health and person-centred practices for the delivery of effective services. For all three organisations, strengths-based approaches were inseparable from a community-driven, holistic service design (18).

Apart from being good and respectful practice, the leadership and engagement of Aboriginal people and communities in decision-making regarding the provision of health initiatives is reinforced by the United

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[1] The words Aboriginal and Indigenous are both used respectfully in this article. The term 'Aboriginal Australians' refers specifically to the First Nations people of mainland Australia. When the term 'Indigenous' is used in the Australian context, it respectfully refers to both Aboriginal people and the First Nations people of the Torres Strait. There are approximately 150 distinct language and cultural groups of Indigenous people in contemporary Australia, living both on and away from their traditional lands.

Nations Declaration on the Rights of Indigenous Peoples (19). Guided by the purposes and principles of its Charter, the United Nations upholds the right of Aboriginal families and communities to retain shared responsibility for the upbringing, training, education, and wellbeing of their children. Article 23 of the Charter includes the right of parents and the community to be actively involved in developing and determining health and social programs and to administer such programs through their own institutions. Social processes such as community co-design, deliberative decision-making, and participation can support public health improvements by engaging people in an outcome-oriented learning and capacity-building exchange (20). This process of evidence-informed decision-making is paramount when working in culturally diverse communities, as cultural assumptions that sit behind intervention programs must be challenged (21).

The international implementation science literature provides helpful frameworks for reflection on the appropriateness of intervention programs for diverse community contexts and the processes of place-based adaptation (22, 23). One useful example, the Hexagon analysis tool requires that decisions about program implementation in any community are guided by consideration of six essential indicators (24).

- Capacity to implement—are there staff who are appropriately qualified to implement the program or strategy? Is this program or strategy sustainable within this community?
- Fit with current initiatives—does the program align with community priorities? How does it fit within the existing service network and community values?
- Need—are the issues being addressed by the program seen as significant by all stakeholders and supported in data indicating need?
- Usability—is the program or initiative ready to be implemented? Does it need further adaptation to be meaningful and culturally appropriate in the local context?
- Support—is training and professional development related to this program or practice readily available? Is training culturally sensitive? Does it address issues of race equity, cultural responsiveness, or implicit bias? What is the source of training and professional development?
- Evidence—what is the strength of the existing evidence of effectiveness for this program or strategy?

The RE-AIM framework (Reach, Efficacy, Adoption, Implementation and Maintenance) is another widely used implementation science tool (25). RE-AIM guides program evaluations to consider factors beyond efficacy, and to incorporate measures and evaluation designs that broaden the range of factors beyond clinical measures only (26, 27). In recent years, RE-AIM has been used in evaluations of health promotion programs in Australian Aboriginal contexts (26). For example, it was employed in the evaluation of the Deadly River Mob (DLM) program, a peer-driven, incentivised health promotion program to reduce hepatitis C among Aboriginal and Torres Strait Islander peoples (26). The program was found to produce positive effects in a culturally safe way, with the employment of frontline staff seen as key to fostering community trust and engagement (28).

For the purposes of this study, the Hexagon analysis framework was employed because it gives stronger focus to decision making around implementation and adaptation which is aligned with the purpose of this research, where-as RE-AIM is more strongly focused on evaluation and measures of success (24, 29).

1.2 The Malpa project and the young doctors for life program

The Malpa Project is a not-for-profit organisation that is the home of the core YDFL program. YDFL offers the same information and core materials to all communities who invite them to deliver the program. However, a co-design process is undertaken with local Aboriginal Elders, to adapt the program, and shape the program content to be appropriate and responsive to community customs, knowledges, and needs. The passing of knowledge from Aboriginal Elders to young people is acknowledged as an important tradition in Aboriginal communities (30, 31). Respecting this sharing of knowledges, YDFL incorporates the sharing of the old and new ways with children to develop their role as health ambassadors and active agents of change within their own lives and communities. The program designers believe *'there is real wisdom in the old ways'* and see the program as an avenue through which to *respect* traditional cultures and healing practices while also sharing Western health knowledge. The language of the local people is incorporated into the program for each site wherever possible and is evident in the program names. For example, the program is called Tjitji Doctors in Alice Springs, and Bubup Doctors in Melbourne (32).

The Young Doctors for Life program commenced in 2012 with 12 students at Alice Springs in the Northern Territory of Australia. The program has grown so that 4,100 children across Australia are expected to have completed the program by the end of 2024.

Malpa has conducted their own internal evaluations. These evaluations, combined with a body of strong anecdotal evidence, point to many positive outcomes for the program, particularly as this relates to child and parent school engagement, improvements in health behaviours, and child sense of connectedness to culture.[2] There is a need to establish a formal evidence base for the effectiveness of the program.

1.3 The current project

The adaptation and implementation of the YDFL program in one regional community is reported in this paper. The perspectives and experiences of the local Aboriginal implementation team, the original YDFL design team, the participating children, their parents, and school staff are presented and discussed. The evaluation was guided by the following research questions:

1. What was the implementation process for the YDFL program.
2. Who was involved and what were the actions necessary to ensure that the program was relevant and culturally meaningful within the local community context?
3. What were the challenges associated with the adaptation for the cultural context, while maintaining core program components and the fidelity of the established YDFL program?

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[2] https://www.malpa.org.au/

4. What were the experiences and perspectives of children, parents, and school staff, regarding the implementation of the program and its relevance to the local context?

2 Methods

A qualitative case-study design using focus groups, interviews and surveys was employed to understand the processes and experiences of program adaptation and implementation in one regional site. Aboriginal children were recruited if they had completed YDFL training within 2 years of the study commencement date, with no child excluded. All parents and schoolteachers of the children taking part in the study were also asked to be involved in the research. No exclusion criteria were applied to adult participants.

A qualitative case study design was employed because we were interested in understanding the nuanced and context driven nature of program adaptation and implementation. The research literature argues that programs are most effective when they embed local knowledge, take into account environmental and place-based issues, and are culturally meaningful (18). While this argument is strong conceptually, there are very few descriptions within the literature of how this happens and when it has happened successfully. We hope to contribute to the literature by capturing this process in one community, privileging the experiences of the stakeholders directly involved. Future research will explore this process in other communities. We will look to identify the key support mechanisms of meaningful adaptation and the impact on child, family and community outcomes.

Questions used to guide the focus groups were based on the key implementation indicators of the Hexagon analysis tool (24), as described earlier. The larger program of research also examined program efficacy; however these findings will be reported in a subsequent paper. The focus of this paper is program adaption and implementation.

2.1 Aboriginal consultation

The research team was invited by the Malpa Project to undertake independent research and evaluation of their Young Doctors for Life (YDFL) program. The Malpa leadership team connected the researchers with the group of Aboriginal Elders who were leading the program delivery in the case study site. The researchers adhered to the Australian Aboriginal Health & Medical Research Council's five key criteria for conducting research with Aboriginal children and communities, including: demonstrating the overall positive benefit for Aboriginal health; ensuring Aboriginal community control of the research; establishing and sustaining cultural sensitivity; reimbursing participation costs; and enhancing Aboriginal skills and knowledge.

The first author, a doctoral student who conducted the field research, worked collaboratively and over a sustained time frame with the regional Aboriginal leaders of YDFL prior to commencing the research to support the development of trust and shared understanding of the method and appropriate community engagement strategies. The building of trust was supported by the fact that the field researcher was also a person with an Aboriginal family heritage. Following consultation with the local Aboriginal Medical Service, an Aboriginal reference group was formed and a formal invitation to commence the research was received from the regional YDFL project coordinator.

2.2 Ethics statement

This study was approved by the Aboriginal Health & Medical Research Council Human Research Ethics Committee (Reference number: 1561/19). This approval was recognised by the Western Sydney University Human Research Ethics Committee (WSU HREC). NSW State Education approval was obtained prior to interviewing children at their school. Privacy note: pseudonyms are used for children's names.

Ethical child researchers are reflexive in creating conditions where children have agency and are supported to share power as much as possible (33). Specifically, the researcher acknowledged and sought to address the implications of the child's perception of the adult researcher as unduly powerful as this may inhibit their free responses or make the child uncomfortable. The field researcher was always introduced to a child by an Aboriginal Elder. He was presented as a person with Aboriginal family heritage who was interested in what children thought about health issues, and specifically the YDFL program. He explained that he was not a teacher within the school system. The field researcher used child-friendly language and humour, dressed casually, and was relaxed to avoid replicating a teacher's role and to decrease power imbalances. He explained he was a researcher who saw children as experts on their lives and cared about what they think. The field researcher acknowledged the respect of children and adults following his invitation from the Aboriginal community to undertake the research.

2.3 Participants

This study engaged five groups of participants.

Group 1: *Members of the local implementation team*: the local implementation team comprised three participants who were all Aboriginal and responsible for the adaptation and implementation of YDFL in their region. The Aboriginal Elder in this group had primary school qualifications and all three participants had experience with face-to-face teaching and formal qualifications in community support.

Group 2: *Malpa program design team:* two senior members of the program design team participated in the project. Both participants had business and education qualifications.

Group 3: *Children:* the study involved two cohorts of children. Cohort 1, or the Retrospective Cohort, consisted of 14 children, aged 9–12 years (8 females and 6 males), who had completed the YDFL program within the last 2 years. Cohort 2 consisted of eight children, aged 7–11 years (5 females and 3 males), who were interviewed before and after program participation. No non-Aboriginal children were involved.

Group 4: *Parents*: 16 parents of children from Cohort 1 or 2 participated in the research.

Group 5: *School staff*: two schoolteachers of children from Cohort 2 participated in this research. One schoolteacher was Aboriginal, and the other was non-Aboriginal.

2.4 Recruitment and data collection

A face-to-face focus group was conducted with the local Aboriginal implementation team. Participants were provided with the focus group questions in advance of the session with assurances they could raise any issues that were most important to them without feeling restricted by the questions. Examples of questions included:

To what extent have you adapted the program specifically for the needs of the community?

Would you see the core program being easily adapted for other regional communities?

The program design team also participated in a one-off face-to-face focus group guided by questions informed by the Hexagon analysis tool.

Due to COVID lockdowns and the associated travel restrictions, as well as a series of natural disasters in the region including flooding, the original research plan to conduct a mixed methods pre and post design study with children participating in the program for a year was not feasible. Instead we interviewed children who had previously completed the program (Cohort 1), recruiting them with the assistance of local project champions. Nineteen interview questions were used to scaffold their reflections on program participation and any sustained learnings and impact. Later, when community access improved, we recruited another cohort of children (Cohort 2) to the study and proceeded with the original pre-post design, including gathering qualitative experiential data and measuring child outcomes. Only the findings that relate to program implementation are reported in this paper.

Parents of the participating children from both cohorts were invited to complete a hardcopy survey in their own time and return it to the field researcher. The survey was comprised of open-ended questions asking for their reflections on the program and its impact on their child and family.

School staff participated in individual, face-to-face semi-structured interviews to capture their views on the YDFL program and any observed program impacts on participating children. Dates of data collection for each participant group are shown in Table 1.

2.5 Analysis

All participants were given the opportunity to review the transcripts from their interviews and to add or subtract information. A thematic analysis was undertaken. The field researcher and one other research team member developed the initial coding framework using the six elements of the Hexagon analysis tool as *a priori* themes and identifying the unique data segments that captured how each participant constructed meaning based on personal experiences of the YDLF program implementation and adaption. Memo writing recorded researcher insights that afforded more depth and complexity to the coding process. Codes were refined and relationships with and between codes developed to identify themes and subthemes. The researchers have reported back to the local community and stakeholders, with plans to provide a final report after the completion of the study.

3 Results

The perspectives, experiences and health education approaches employed by the local implementation team in establishing the Malpa YDFL program in a regional community were explored. Data were analysed and organised according to *a priori* themes: Capacity; Fit; Need; Usability; Support; and Evidence. Each theme and their subthemes are discussed. Themes and subthemes are shown in Figure 1.

3.1 Theme 1 capacity

The research participants saw capacity as relating to both personal and available community resources. Their views were captured across three subthemes relating to the importance of local trainers with specific qualities, and adequate resourcing.

3.1.1 Subtheme 1A: YDFL trainers from the community

To have the YDFL trainers drawn from the local community was seen as imperative by all participants in the focus groups. A program designer said: "Another thing about the capacity to implement going beyond skill, one is that we require that one of the people is Indigenous, at least one preferably two."

When the implementation team was asked how important it was to be part of the community when the program was being implemented, they affirmed its importance, expressing their views based on their own experiences of living in the community.

> Yes, it gains trust, if people know – with the Indigenous community, if they do not know you and you were someone who flew in to do the same thing – they do not know you – they do not know what your motives are – they just assume you are another government worker or whatever. But if you are a member of the community and they have seen you out there. They are relaxed. (Implementation team member 1).

The importance of having Aboriginal trainers was also clear in the narratives of the children, who felt safer sharing their views when the adults were also Aboriginal.

TABLE 1 Participant data-gathering dates.

Participants	N	Age range (years)	Data collection: Pre—YDFL	Data collection: Post YDFL
Child cohort 1	14	9–12		June – December 2020
Child cohort 2	8	7–11	April 2021	November–December 2021 (disrupted by Covid lockdowns) May 2022
Parents	16		April 2021	November–December 2021 (disrupted by Covid Lockdowns) May 2022
Schoolteachers	2			May 2022

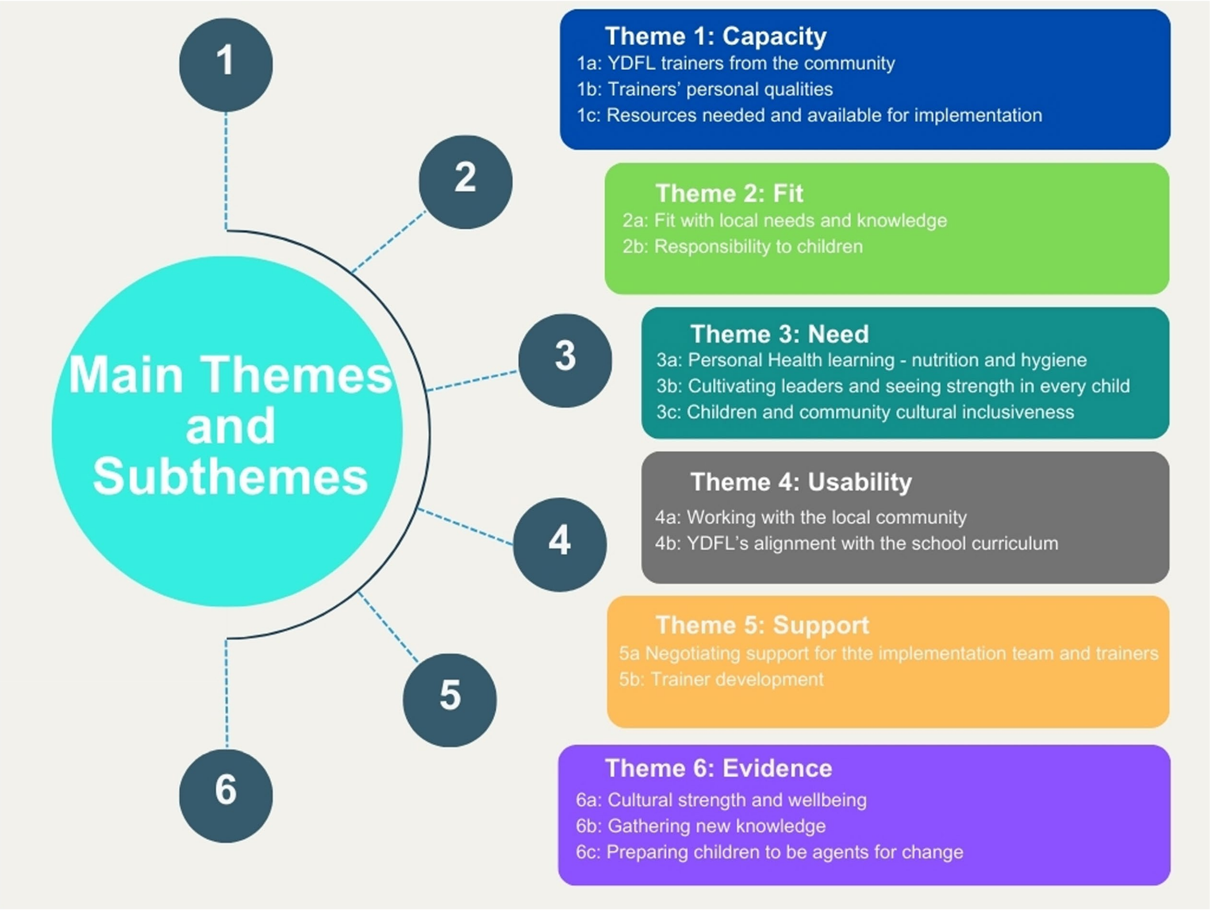

FIGURE 1
Themes and subthemes.

> It, sort of, made me a bit more confident to … made like everyone in our group to be, kind of, a bit more confident – and to, I guess, speak out because – because most of us or some of us, they feel like they cannot speak out because of, like, our colour and stuff. And even though that there's no racism at our school – that we know of, we still feel like that we cannot – do stuff, so that sort of helped us. (Irene, aged 11 years).

The children's schoolteachers also saw the leadership of local Aboriginal people as important to learning for the children, incorporating learning about their own heritage.

> The [local name] Doctors is one of the best things [for learning culture and heritage]. I learnt things that I did not, you know. And I always say to kids that—we are learning—we [teachers] are learning as well because we do not know everything. (Aboriginal schoolteacher).

3.1.2 Subtheme 1B: trainers' personal qualities

The personal approach of the individual trainers was seen as important to successful program implementation.

> … we want them [the trainers] to be imaginative and we want them to be able to put their own unique stamp on their project. So they need to have a capacity to try and to think differently in terms of how they engage the kids and how they connect with the school and their community. So they need to be good communicators. They need to have good interpersonal skills. Uh, they need that capacity like a good uh, a good sports coach to be able to energise the room, keep kids on focus and make them feel that somebody is genuinely interested in the kids themselves and what they are doing and have an idea about what they might become. (Program designer 1).

The children valued trainers that injected a lot of fun and supported them to find their voices. The routine yarning circles[3] and generally yarning with the children throughout the 15 week program was particularly valued. As Lucy (aged 11 years) commented "I like how we sing the song on the yarning circle mat. Yeah. Because we were all doing it, yeah. Yeah, having fun."

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[3] Yarning circles: A term used by some Aboriginal communities to refer to processes of bringing a group of community members together for the purpose of gathering specific information, in accordance with local community and cultural protocols.

A sense of fun, energy, and ability to empower children were seen as the most important qualities for the trainers, beyond their Aboriginality.

3.1.3 Subtheme 1C: resources needed and available for implementation

When asked if the YDFL was sustainable in their regional area, the implementation team Elder told the facilitator that the team receives funding from the Malpa Project for each program site implementation. It was a challenge to make the funding stretch, and the local team needed to make decisions and work to a budget to prioritise the needs of the children attending each YDFL program, as the Elder explained:

> I have cut back on the spending so much, but I have increased it on nutrition—I have increased the funds and how much we spend because I want them to taste good food not cheap food—more variety, so you have to spend more. (Implementation team member 1)

The Elder pointed out that the program is sustainable because of the small budget. However, in her view, if there were no funds available, the program would continue with the dedicated volunteers who were part of the program delivery: "You could deliver this with volunteers— you do not need a big budget—enough to buy the snacks and things like that."

3.2 Theme 2: fit

In the discussion of the core curriculum, program designers shared that they needed to be aware of the core curriculum, however there is a need to be adaptable to meet the needs at the community level, and in terms of the different health regulations across the states of Australia. Adaptability was critical.

3.2.1 Subtheme 2A: fit with local needs and knowledge

The adaptations do not happen at the level of head office, they happen in a co-design process with local implementation teams in response to the needs of communities: "We do not do the adaptations, no—the community creates their own, yeah— they are the ones adapting the program to make it relevant" (Program designer 1).

The implementation team stressed the importance of adapting the YDFL program to children's differing needs and knowledge depending on the context. One implementation team member, referring to one of their innovations, commented, 'We are the only people who do the song and the pledge - and I love it…it makes more ownership for the kids'.

Another implementation team member took pride in the program being locally driven and owned:

> The essence of this thing [program] is that that it is a home-grown … all the programs have commonalities but are really very, very different. [We] construct the ways of delivering according to what they [community] need and I think one of the real strengths of the program is this localisation… [by way of] it's structural flexibility… so community adaptation [is] right from the get-go and gives the local community ownership. (Implementation team member 1).

The children described being taught things that were responsive to their needs, evidence of the fit of the program as this relates to what was important to the children.

> I felt like they, uhm, have helped me a lot for the past years, because like I have got bullied at school and all that, but I think they [YDFL trainers and peers] help me with all of that and now, I do not really get bullied. And they helped me with being safe, and healthy, and not smoking and all that. (Mila, aged 10 years).

3.2.2 Subtheme 2B: responsivity to children

The implementation team explained that some children could not read and write or may have had learning disabilities. Nevertheless, they reported how they were responsive to children in an ongoing way across program delivery, so the challenges the children were facing did not stand in the way of their training as health ambassadors and becoming potential leaders for their peers and the wider community:

> You do [adapt] a lot and you find out what is happening to children…as adults we are adapting ourselves to the child, not having the child adapt to us, …one time I will do this and next week this—I'm [often] slightly trying to change and adapt the activity to the—sort of, need for the kids. (Implementation team member 3).

One of the children interviewed mentioned that he had a disability and found mainstream schooling difficult. When asked what he remembered most about attending the YDFL training he replied, "Probably how we, uh, sat in a circle on the mat and sang songs and talked, yeah" He went on to describe how he did not feel like he had a disability when he was with the group like this. In their responses, the parents said that their children's socialisation skills had improved following involvement in the YDFL program and indicated this was because of the trainer's responsiveness to their children. One parent spoke about how isolated her child had been, and how the YDFL implementation team had supported the friendship connections that were formed within the group:

> … it's helped with like my – with like people who have gone to [the Young] Doctors because we can talk about – like all this stuff. And they also talk – they also talk about like friendships and stuff and that helps us, uhm, I guess with feelings and stuff for friendship and – Yeah. (Irene aged 12).

Parents described witnessing their children's increased confidence to engage with others, including making new Aboriginal and non-Aboriginal friends. The program designers perceived that the trainers developed the ability to skilfully adapt the program in response to immediate need as they gained more and more hands-on experience. As one program designer said:

> It is much as possible, being experiential and hands-on ... they learn by implementing the program. They are discovering the boundaries that they may have thought constricted them, that

> only exist in their mind, and if they go off on the slightly strange tangent from our point of view, we want them to know that we are not looking over their shoulder. We are happy to support them and go wherever that might go, even if it sits slightly outside where we were thinking. (Program designer 2).

The program designers expressed belief in the values, skills, and expertise of each local program implementation team to deliver an adapted and tailored program to their community because they knew the children best. Programs were therefore centred on the children's needs.

3.3 Theme 3: need: empowering children as 'young doctors'

The heath challenges for Aboriginal children in the area were well known to all of the participants, and so there was a strong sense of shared need. The delivery of the program centred on key areas of need. The interview data described the growing ability of children to address those needs and become leaders within their own communities.

3.3.1 Subtheme 3A. personal health learning—nutrition and hygiene

Two priority issues for the case study community centred on nutrition and hygiene, and so the program was adapted to ensure emphasis on this form of personal health learning. The participants provided several anecdotes in which they could see the impact of their program in this respect. For example, children influenced their parents to purchase nutritious food, such as choosing grain bread instead of traditional white.

The implementation team described how they managed their budget to ensure they purchased wholesome food for their sessions with the children that focused on eating healthy foods. Following is an example of a local adaptation aligned with perceived local needs.

> I have increased how much we spend because I want them to taste good food, not cheap food … You are validating the kids, and you are validating that they are human beings. (Implementation team member 1).
>
> We do the food tasting—you try the different cheeses— 'ohh I've never had this before'— 'well, go get your mum to buy it'—and some would just stand there and keep eating— 'like can I take it on the bus?'—and they will take the packet home to show mum and dad. (Implementation team member 1).

The trainers also described discussing sensitive matters with the children relating to personal hygiene in an age-appropriate and non-offensive way. The implementation team, knowing the financial disparity in their regional area, and being aware that some families were struggling and not able to afford things like washing machines, worked to normalise the situation. For example, they created a game to help children take responsibility and look after their clothes:

> We did this one in relay teams where one washes [an item of clothing], drops it and the next one rinses it, one pegs it up and the next one puts it on an ironing board and the next one [folds it]. The children are very competitive, and they are saying, 'come on come on'—and the boys—the competitiveness about the whole thing—learning and they love the game. For two weeks in a row, we had to play the game because they just loved it. But they were learning that clothes do not get left in the basket ... They were learning that they could go into the bathroom and wash it if they do not have a washing machine. [It's about] learning how much water to wash your clothes [in] and taking responsibility to be respectful of your clothing and of your parents who have to buy them. (Implementation team member 3).

These extracts exemplify two important implementation components that support positive outcomes from the perspectives of the implementation team, namely: the value of games in facilitating behaviour change without shame or embarrassment; and the importance of cultivating a sense of personal responsibility. Having fun while learning new things and experiencing health education through games was a positive experience for the children. Extracts from children supported this perspective.

> [I liked] playing and learning about new stuff and playing things that you never played before. (Jade, aged 9 years)
>
> Then we'd do like some games and stuff—but they were also still educational which was really good. (Denis, aged 11 years)
>
> Making fun things every day…We made our own playdough… Yea, and we put it into—we made a stress ball. [What are the best things?] The activities. They made them more fun than they usually are. (Ivy, aged 11 years)

The schoolteachers agreed that the playful and fun nature of program implementation was impactful in supporting the learning of the children and facilitating their engagement.

> … You cannot have education boring. You are going to have education fun. It's going to be cultural, you know, it's a good way of learning, you know, in a fun, cultural, respected way, we believe anyway…I believe. (Aboriginal Schoolteacher)
>
> I think it's got a good balance because they do their talk time, and their workbook time, and then they also have their outside playtime, and then their food time, they love the food. So, no, I think it's a very good balance of everything, really. (Non-Aboriginal Schoolteacher.)

The implementation team was successful in marrying the need for information and health learning, with the need for connection and fun.

3.3.2 Subtheme 3B: cultivating leaders and seeing strength in every child

In the case study area, the implementation team felt there was a need for local leadership and saw their role as supporting children to become local health leaders.

If you give the kids the tools to lead, to be their own leaders and adults, they will step up to the plate. (Implementation team member 1).

It was particularly edifying for the implementation team when they saw child leadership demonstrated, and this encouraged them to continue prioritising supporting the children in the development of this skill. For example, when the school held a graduation ceremony for the kindergarten children, the YDFL implementation team were amazed to see the recently graduated 'young doctors' volunteer to lead the ceremony: 'They just stood up and said we can do this'. The implementation team strongly perceived that the involvement of the older children in the YDFL program gave them the confidence to support the younger children and be leaders and role models for them.

The implementation team described the selection process for children attending the YDFL program. In some instances, the school staff nominated the children; on other occasions, the selection of participants was made in consultation with the implementation team. The latter was considered to be ideal because this mitigated against the usual practices of school staff to select the children who they felt deserved a reward for their good behaviour.

> The teachers do not look at the kids as individuals before they pick them. He will be alright … he is naughty, so he will not be doing it. I do think it is good that the good kids do it because it is a reward for them, but the kids who muck up and stuff probably need it more. They are a bit lost and at some schools when we say [to the children] you are a leader, they say, 'no we are not, [name] is a leader—we could not be a leader.' We tell them that you were picked because you have something good to give—we build their self-esteem. (Implementation team member 3)

The implementation team advised that selecting participants who were potential leaders and health ambassadors required insight and understanding. As an example, an implementation team member related the following instance where leadership potential was not immediately obvious:

> We had two boys at [school name] who we had at a community play group ... We knew the mum and family, trouble kids who were always suspended. So, when we went to their school, we said that we want those boys. They [school] had not thought of them because they were looking at those who were trying to be good kids. But we had those boys, and we had a guest speaker who also worked with the boys elsewhere, and he said that he had never seen them sit quietly [before]. (Implementation team member 2).

The implementation team's intent was to empower children. The YDFL program is not taught in a didactic manner but is activity-based with a cultural focus. The implementation team felt that the children were often more comfortable in that space than in their mainstream classrooms, which was then reflected in their behaviour when engaged in YDFL activities.

There is scope to meet the individual needs of each child while maintaining fidelity to the core program, which is an important aspect of implementation. The mix of children and the ability to adapt to their needs was considered important by the program designers.

> I mean, most of our programs seem to choose a mix of people who are likely to be leaders, and sometimes there are kids who are currently leaders in a negative way, but they [trainers] want to pick up on their leadership ability and turn [them] around. We have some programs where more than 50% of kids are in out-of-home care. (Program designer 1).

The program designers spoke of the absence of a tiered reward system, with all Young Doctor graduates receiving the same certificate:

> We do not believe, in any type of class… we do not do any of that …you know competition is a good thing [and] rewarding excellence…we [just] want the kids to feel all proud and all in this together…and they do, and this is often the first certificate they have ever received… their parents feel proud… if you can empower one you can empower more than one. (Program designer 2).

Parents spoke positively of the social cohesion, equity, and confidence building within the YDFL program their children attended. For example, one parent said:

She gives things more of a go now than before, it has lifted her self-confidence. (Parent 3).

3.3.3 Subtheme 3C: children and community cultural inclusiveness

While the YDFL program has been designed for Aboriginal children, non-Aboriginal children are also welcome to attend in the spirit of inclusiveness. The implementation team and the program designers recognised the need to promote activities that may unite both Aboriginal and non-Aboriginal children and families from the regional area. "They [non-Aboriginal children] are coming from exactly the same socioeconomic group as the Aboriginal kids" (Program designer 1).

An Aboriginal child may invite a non-Aboriginal "buddy" to join them in the YDFL program. This was reported as a way of achieving inclusivity in the region.

> Then when they [non-Aboriginal] come to the [YDFL], they are so proud that they are part of the group … and we push it and say you are really special, and you have a role to play you have an important position and job [to tell people] … and they boast about it. They tell their non-Indigenous parents. (Implementation Team Member 1)

One implementation team member spoke of her wish to eliminate the 'black areas and 'white areas' of the regional township. The Elder of the implementation team (Team Member 1) spoke of her initiatives to improve a park in the town where people could visit and undertake activities or just rest there.

> So, to try and fill that gap, I went on the board of [name] park [with] the bush tucker garden and I am the chair. So, by helping the community I am helping the [YDFL program]. So, once the plants [re-grow], it has been run down for a while, (then) running school programs with the council with other groups for teenagers and parents [can occur]—the town is getting right back into weaving. I have been doing weaving at [name] Park, but I have to pass it on to the kids. And I hope they will come to a weaving group with us. (Implementation Team member 1).

These initiatives correspond to comments made regarding connecting to culture and understanding their environment. As one parent stated, "I think it is a great program for the kids, Aboriginal and non-Aboriginal, I think it helps the kids to connect to culture and understand culture while learning how to look after themselves and their environment." Such comments were reinforced by another parent who suggested changes that could help such as "more info about the program, [and to have] parents more involved, if they want to, and more Elder involvement."

3.4 Theme 4: useability

The YDFL program's usability relies on how successfully it is embedded within the target community. Being considered part of the local Aboriginal community was described by the implementation team as crucial to their efforts in the successful delivery of the YDFL program. The implementation team felt that their community membership and acceptance paved the way for program implementation by building trust and a spirit of collaboration. The local trainers' engagement with their own community was considered a distinct advantage by the Sydney-based program designers. This theme includes two subthemes that highlight usability through the adaptability of the YDFL program.

3.4.1 Subtheme 4A: working with the local community

The program designers rely on the local implementation team to embed the program within the community by working closely with the people from the community.

We do not know all those communities and there's no way we can ever know all those communities. So, we need those people on the ground to be able to do that for us…and they really do rise to the occasion. (Program designer 1)

Furthermore, the implementation team engages other local health service providers in the program to raise children's awareness of the local community health services available to them. This initiative included representatives from local services such as the dental service, a hearing service, a smoking prevention program, and health promotion educators who promote hygiene practices to prevent the spread of bacteria and viruses.

A frequent response from parents was that they observed the YDFL as a fun way for their children to learn about hygiene practices. They often commented on 'Mr Germ', a character in the program, and his related presentations, which reinforced good hygiene practices in a fun way. As one parent said:

> Mr. Germ and the black light, and teeth brushing [made my daughter] independent and clearer of her own environment [so she was able to understand], mask, cross contamination.

The relevance of the YDFL program became even more obvious to parents when the COVID-19 virus spread to regional and more remote areas of the state. The YDFL program was instrumental in supporting the children to understand the importance of personal care procedures.

> [Our child taught our family] Better hygiene, hand hygiene—showing family how to wash hands [during COVID]. (Parent 5)

> [My son] did not complain about washing hands and requirements of Covid 19. It prepared [him] for good hygiene practices with him regularly washing and sanitising his hands. (Parent 11).

3.4.2 Subtheme 4B: YDFL's alignment with the school curriculum

The program designers referred to YDFL's compatibility and enrichment of the NSW school core curriculum.

> I think part of the reason that is welcome is that it delivers on the core curriculum, and so, it's a different way of doing what they are required to do anyway. In a way, [it] is a lovely, sort of, an enrichment into the school but is not a bolt on the side. It's right in the middle. I think that's absolutely fundamental. If it's an add-on, it will not-will not be picked up. (Program designer 1).

The implementation team worked closely with the school to ensure that the program complemented the curriculum and the timing of lessons to align with what the school is working on.

> It's great because it fits straight in with our PDHPE syllabus [Personal Development, Health, and Physical Education], to teach about healthy relationships, healthy lifestyle, everything—and being good leaders and what good friends were about, so it all fits in perfect, yeah, and our-our kids love it. (Non-Aboriginal Schoolteacher).

Both schoolteachers observed the children's enthusiastic response to new learnings from the YDFL program, and the supportive relationship the YDFL trainers had developed with the children. They felt that participation in YDFL had a positive influence on the children's engagement with the wider school curriculum.

3.5 Theme 5: support

It is the role of the program designers and the regional program coordinator to ensure training support for the YDFL implementation team and trainers. This theme is discussed in two subthemes.

3.5.1 Subtheme 5A: negotiating support for the implementation team and trainers

The program designers, cognisant of the YDFL trainers' positive influence on the children, have developed support processes for their trainers during the delivery of the program. Nevertheless, focus group participants described early tensions between the program design team and trainers arising from the requirement for trainers to submit regular reports on the YDFL program. Trainers reported "finding the task overly complex and time consuming." To resolve these tensions, the participants in the focus groups described negotiation processes to balance the trainers' support needs and the accountability mechanisms required by the Malpa Project head office.

One of the difficulties the design team encounters is the geographical distance between themselves and the regional trainers. Regular contact enables the design team to become aware of the implementation team and trainers' concerns. As a result of negotiation between both parties, the process has become more streamlined, catering for both the educational reporting requirement, addressing the trainers' concerns, and ensuring that local Aboriginal voice is privileged.

The implementation team also established a mutual support system from within the broader YDFL community to share ideas and resources: "that's what we need…support from other groups… this resource works for us here and so on… what works for here may work for you." The program designers took notice and organised an electronic chat group to facilitate this networking. However, this was not taken up by other trainer groups. The implementation team continues to explore options to create communication networks between sites on suitable social media platforms.

3.5.2 Subtheme 5B: trainer development

Close liaison is required between the program designers and the local implementation coordinator to support ongoing implementation and adaption in line with local need. The program designers talked about how they kept in touch on a regular basis with the implementation team and responded promptly when advise was sought:

> They will frequently email me. Sometimes ring me or text me asking for advice about how to go about something…that is much more informal, but it is at least fortnightly contact with every program. The ongoing capacity development tends to happen through that [process]. (Program designer 1).

The program designers reported offering the YDFL trainers formal education courses (e.g., accreditation through TAFE), but to date there had not been strong interest in this offer. There was acknowledgement that the trainers were strongest when their local knowledge was honoured, and when they engaged in the process of experiential learning.

One implementation member described a process of 'research mindedness' in which the implementation team was constantly asking questions about what was needed, what was working, and how the program could respond.

> We do research, we do a lot of our own research. We look at things and bring it to the table, discuss it and say, 'do you like this or this.' We have regular meetings and talk about what we will do next week and what we will bring to the table. (Implementation team member 1).

3.6 Theme 6: evidence

In the Hexagon tool, the implementation element 'Evidence' is largely centred on previous research and practice evidence to support the decision to implement a particular practice or program in the first place. In our case study, 'evidence' was interpreted to mean ongoing observations to see whether children were responding to the program and adapting the program in response. The four subthemes under this heading, therefore, capture the implementation team's observations of positive change that fuelled their practice.

3.6.1 Subtheme 6A: cultural strength and wellbeing

In the delivery of the YDFL program, culture or cultural strength is not always spoken of directly. Culture is often taught through sharing information and stories, fun activities, and yarning circles. Below are examples of responses from children when they were asked if the YDFL program helped them feel strong in their culture.

> I reckon it did, yeah, even though that it wasn't about like culture, you still felt connected … being there with, like, basically with a few people, and like—being with kids, uhm, that, like, are Indigenous too and just learning about the same stuff. That's, like, the best part. (Irene, aged 12 years).
>
> Not power. Uhm, responsibility, uh, keeping people safe, uh, if you—if there's like a boulder that people still like get through, that you can move it. And uhm, if you have like a problem you have to like—uh, you have a problem like you like to move something, you can move it. (Len, aged 9 years).

YDFL provided an opportunity to strengthen friendships among Aboriginal children and facilitated the power that comes with joining together as a community to "keep people safe" (Len, aged 9 years).

Children's answers referred directly to cultural strength and the importance of their cultural identity for wellbeing.

> Uhm, like connecting to your culture like always—like when you are not—when you are weak with your culture, you are not learning about your culture. And when you are strong, you are always learning about it, and you have Elders talking to you. It's sort of like knowledge when you are strong. (Irene, aged 12 years).

Observations from the implementation team around the importance of strengthening children's connections with each other led to an expansion of their work into the broader community, such as encouraging children to become part of gardening and cultural groups.

3.6.2 Subtheme 6B: gathering new knowledge

The YDFL program is co-designed for local areas with traditional Aboriginal knowledge holders, Elders invited in by the YDFL trainers, in partnership with doctors, nurses, ambulance paramedics, dentists and nutritionists—whoever holds the knowledge that will empower the 'young doctors' to become health ambassadors. The children described being encouraged to open themselves to new knowledge and experiences. This willingness was evident in the narratives of the children.

> We had to learn like—when we like did not wanna try anything out—we had to [try] … Then I said to Auntie Jan, I said, 'Auntie [trainer], what is that?' and she said, 'It's passionfruit.' And then she said, 'Taste it,' and I said, 'No.' And then she opened it and then I said, 'Ewe, I do not wanna look at that.' And then I ate it and then it tasted good. (Margo, aged 9 years).
>
> ... learning new stuff…and learning like the Aboriginal, uhm, plants. And the berries that you can eat and not. And how to make the paint. I think—I think I remember once they put that on to protect their face from the sun. It like protects their face so they do not get burned when they hunt. Yeah. (Tess, aged 12 years).

The children demonstrated enhanced knowledge of nutrition.

> They've told us like the, I guess, good and bad food and that sort of changed how I see food. And they tell us like what you can and cannot eat and they—they also introduce us to new foods and salts and like jams and stuff like—uhm, like cultural sort of foods that sort of has changed my diet too. And like—yeah. I guess they are teaching us—what we used to eat, like, back in the day and that. (Irene, aged 12 years).

The children also gained new knowledge of hygiene that shaped their behaviours.

> So I would [now] always wash my [hands] before and after I eat dinner and after I go to the bathroom or outside, or I've touched the animal. Uhm, I've cleaned my room every Sunday, and I wiped down walls and clean my sheets and stuff, to make sure my room is all clean. And I always, uhm, get—I—if I've had water sitting in my drink bottle all day, I will refill it at night and put it in the fridge. And I would clean my dishes and fork, knives, and things like that after I've eaten, yeah. [Why is it good to wash your hands?] Uhm, so if you are touching things and then someone else goes to touch it, that they do not get your germs, if you have been—uhm, not picking your nose or playing with dirt or something, just so you can keep your germs to yourself and that. So, then people do not—because it's very important to keep yourself healthy and your hygiene. (Steph, aged 11 years).

The children also demonstrating that they were learning to access the health system.

> [If you were really sick] I would probably—I, think we'd go to the hospital straight up, they'd [parents] take me. [Who would help you there?] I think the nurses would see me first—and if something is badly wrong, they'd get a doctor. But if they can figure it out better, they'd give me the medicine I might need and tell my parents to what times and what days to take these. And make sure and come back maybe like two weeks tops to see if I'm doing okay. (Tess, aged 12 years).

The trainers advised that they wanted the program to be rich and stimulating first and foremost and prioritised this above cramming a large amount of information into the program. They chose to use an approach marked by gentle guidance and encouragement to help the children develop skills and gather new knowledge in the areas that were most relevant and appropriate for their community.

3.6.3 Subtheme 6C: preparing children to be agents for change

The schoolteachers witnessed a change in the children who participated in the program as this relates to helping their families and others. The felt that the program supported children to become agential.

> [The YDFL] is a really good program, I think. I think it's helped them. But it's helped them to help people who need help as well. Do you know what I mean? That sort of carries on....
>
> [the children say] I keep coming to school, learning all this good stuff, and then going home and saying, 'Mum, that's no good for you or dad, that's not—you cannot smoke that or drink that.' You know, the kids are seeing, you know, both sides of the good and the bad but, yeah. (Aboriginal school teacher).

The schoolteacher interviews in particular gave insight into how the YDFL program had effects beyond the classroom and into their families and communities. The implementation team saw this as evidence that their methods were having an impact and should be continued.

4 Discussion

This paper presents findings related to the adaptation, implementation, and participant experiences of the Young Doctors for Life (YDFL) program in a regional Australian Aboriginal community. The participants described a collaborative implementation process involving the national leaders of the YDFL program, the local Aboriginal YDFL program implementation team, the local Aboriginal community, children, and a local school where the YDFL program was delivered. It showcases the factors that are impactful in supporting the delivery of culturally appropriate health services in Aboriginal communities and the importance of locally meaningful program adaptation. Fixsen and Blase (34) propose that the successful adaptation and implementation of a program is a dynamic and ongoing process in response to the context and needs of local children, families, and communities. By way of their lively and energetic responses, the members of the implementation team conveyed their enthusiasm for community engagement in delivering the program. There are key learnings presented in this study that may assist the implementation of other programs intended for children in Aboriginal communities.

The processes employed by the YDFL program implementation team align with what is discussed in the research literature as best practice in culturally meaningful program adaptation, including the importance of: establishing local leadership, building relationships with the local community and program co-design; supporting the participation of children in child health education programs; and bringing together local and cultural knowledges with Western health knowledges (1, 18). Each of these elements is discussed.

4.1 Establishing local leadership, building relationships, and program co-design

Aboriginal Elders are vital leaders who enable community ownership and control of health initiatives that benefit their communities. Aboriginal cultural leadership is an expression of cultural values in action. The concept of cultural wellbeing was conveyed by Aboriginal Elders in the study by Cox and colleagues (35) as three interconnected themes: mentoring, cultural healing and balance between the community's cultural foundation and the delivery of health and service provider programs (35).

With widespread concern that health programs in Aboriginal communities are 'imposed' without respectful communication and shared decision-making (2) this study provides an example of one way in which a standing organisation might work effectively with

Aboriginal communities. In recent years we have seen a growing body of research that deliberately seeks to shift the power imbalance and ownership of programs, with examples including Australia's first National Aboriginal and Torres Strait Islander Cancer Framework (1); a drug and alcohol, and mental health comorbidity project in Adelaide, South Australia (36); and the development of a community rehabilitation and lifestyle service for a remote Aboriginal community in Northern Queensland (37). Use of co-design in each of these examples was found to be crucial to merging Western evidence and systems knowledge with local need and inclusion. This is fundamental in fostering ownership and knowledge translation for Aboriginal communities (1). In the current study, community acceptance of the program was evident and perceived to be strengthened by the implementation team who were all active members of the local Aboriginal community. Community collaboration was understood as essential to enable the implementation team to align the program with local community health initiatives and to partner with local stakeholders. This alignment with, and understanding of local supports, was found to build trust between program organisers, facilitators and the community in the development and implementation of the YDFL program. Ongoing health service engagement is supported by respectful community relations, community involvement, and co-design. Sustainability is reinforced when the program implementation team and trainers live in the community, enabling program uptake and ongoing assessment and improvements.

4.2 Supporting child participation

This study highlighted the importance of adapting programs to meet individual child needs. A study by Priest et al. (38) reported that Aboriginal children faced greater challenges than children in the general population in the achievement and maintenance of health and wellbeing due to several factors in their social, historical, and political environment. Therefore, essential to children's needs are programs that understand the local context and seek to ensure equity of access to high quality programs for all children. Child engagement and participation in positive experiences is crucial to counter-acting challenges in their lives and supporting positive outcomes (39).

A strong argument exists in the literature that improved child health outcomes will occur when health service providers listen to the voices of Indigenous children (40) and support them to be agents of positive change. The Secretariat of National Aboriginal and Islander Child Care called on researchers to hear the voices of Indigenous children as they explore domains of wellbeing, namely: safety, health, culture and connection, mental health and emotional wellbeing, home, and environment, learning and skills, empowerment, and economic wellbeing. One way this can be achieved is listening through yarning, the Australian Indigenous cultural form of conversation (41). There is a body of child participatory research that details the creative and arts-based qualitative methods that can be employed to support the engagement of children in research and the sharing of their perspectives (42). The findings from the current paper support the importance of participatory methods and seeing children as active stakeholders with a role to play in shaping program decision making.

4.3 Support for trainers

Trainer education was identified within the findings as key to successful program implementation. There were two educational aspects for trainers: firstly support from the program designers and, secondly, support within the implementation team. By providing training to local team members, new insights into the community context were gained and local knowledge was built into the program supporting its effective adaptation to the local context. The implementation team considered the implementation of program elements and offered suggestions to meet the local context before the program was delivered and throughout program implementation.

The importance of trainers' experiential learning as they engage in the education process has been recognised in previous educational research (43). Peer support and peer-mentoring programs have also gained popularity in higher education and other learning environments. (44). To enhance trainer's skills the design and structure of a mentoring program should be connected to the program goals (45). A mentoring program design is the key to peer mentoring program success. In the wider implementation of a health intervention, trainers' support and mentoring will require capacity for implementation costs and the required resources (24).

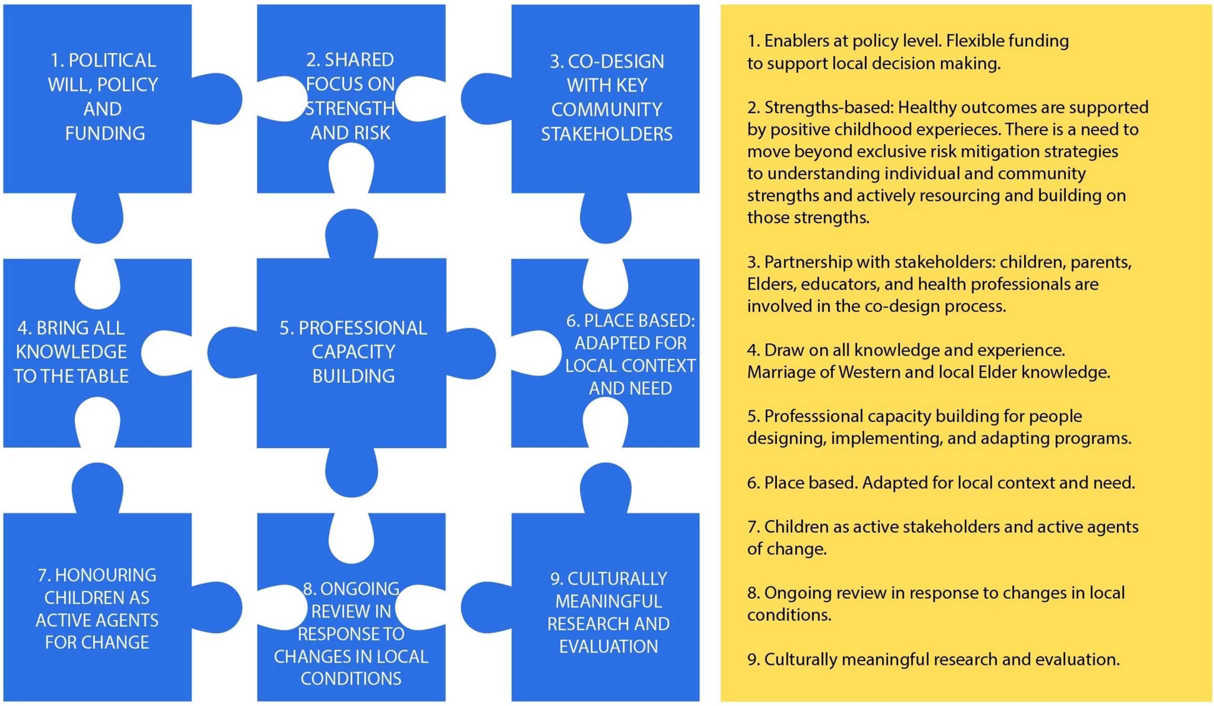
4.4 A framework for Aboriginal child health interventions

Translating research findings into practical outcomes and insights is complex and open to critique, notwithstanding the unpredictable results from theory to practice and the various levels of continuing assessment required (46). The following diagram depicts the complexity of developing health interventions for Aboriginal children and what needs to be considered when designing, disseminating evidence, and implementing the intervention. A successful intervention is not linear; however, it comprises a holistic sum of parts that require continuous evaluation in practice and research.

The information in Figure 2 uses the jigsaw analogy to indicate the non-linear conceptual framework of implementation science. Furthermore, each part of the process requires focus for all the parts to come together as an impactful whole-child health intervention. There is a need to review programs that have been implemented to refine and adjust where required over time. The analogy also applies to future research as researchers explore new ways to improve implementation, dissemination and adapting health interventions where Aboriginal children are at the centre of the process.

4.5 Future research

Jigsaw pieces 8 and 9 in Figure 2 refer to the full implementation phase of innovation and testing the application of the framework represented by the other pieces. In this phase, team members and educators support and expand the innovation using data for continuous improvement and improved outcomes. Future research will test the application of this framework, both in the expansion of research focused on the YDFL program and in relation to other Aboriginal child health programs. Future projects will include exploring the implementation of the YDFL



1. Enablers at policy level. Flexible funding to support local decision making.

2. Strengths-based: Healthy outcomes are supported by positive childhood experieces. There is a need to move beyond exclusive risk mitigation strategies to understanding individual and community strengths and actively resourcing and building on those strengths.

3. Partnership with stakeholders: children, parents, Elders, educators, and health professionals are involved in the co-design process.

4. Draw on all knowledge and experience. Marriage of Western and local Elder knowledge.

5. Professsional capacity building for people designing, implementing, and adapting programs.

6. Place based. Adapted for local context and need.

7. Children as active stakeholders and active agents of change.

8. Ongoing review in response to changes in local conditions.

9. Culturally meaningful research and evaluation.

FIGURE 2
Developing health interventions.

program in other diverse community settings, examining the long-term effects of YDFL on child and community outcomes, exploring how existing Aboriginal child health programs may be enhanced by engaging with children as active stakeholders and exploring the role of children as active agents of community change more broadly.

4.6 Limitations

This research was impacted by state-wide COVID-19 restrictions. For example, it was only possible to conduct the research in one regional community. Access to other regional or remote areas would have provided a more comprehensive study. COVID-19 travel restrictions also prevented the employment of a pre-post design for all of the participating children, and this compromised the rigour of the design to some extent.

There is potential for bias in the program evaluation as the children enjoyed the program activities. Their enjoyment could potentially mean that they overstated the impact of the program. Similarly, there may be biases from employees who believe in the importance of the program and may downplay challenges or drawbacks because they do not want to risk having the program discontinued. The research field officer has Aboriginal family heritage and belongs to an Aboriginal community healing and leadership group. While recognising his positionality provides a unique insight, to reduce any potential bias the analysis and interpretation of the data involved discussions and was conducted in collaboration with the research team.

4.7 Implications

The findings of this study have direct relevance to practice for those working to address the systematic differences in health status and outcomes for Aboriginal children. This paper emphasises the importance of participatory, community-driven models, and calls for consideration of the role of children as active agents for positive change. A child-centred approach to program delivery, in which programs are continuously adapted in line with the needs of local children, is vital. Health programs must incorporate aspects of local culture and healing practices as integral to the implementation of intervention programs if programs are to be meaningful, culturally appropriate, and respectful in local contexts. Leadership from local community members, with a foundation of local knowledge and culture, is a critical strength that should be prioritised. This study has shown that programs are strengthened when they build on existing health and community programs, and partner with a broad range of stakeholders across the health, education, social welfare, and community sectors.

5 Conclusion

The YDFL program was found to be collaborative in its implementation and delivery of culturally appropriate and child-centred health services in an Australian Aboriginal community and provides an example of locally meaningful program adaptation. Community co-design and collaboration was essential to identifying and addressing local community goals and aligning the program with local health initiatives. The community was an integral part of the program which addressed the needs of their children and complemented existing and accepted health initiatives. Importantly, adaptation and implementation of the program required the recognition of each child's individual needs and learning requirements. The inclusion of children in this process was fundamental to ensuring the program was fun, educational, appropriate, and relevant to the children's needs and lives. Community acceptance of the program was strengthened by having the implementation team and trainers as active members of the relevant community as it allowed trust to be built between both entities, which encouraged greater acceptance and buy-in of the program. Successful adaptation and implementation in part required a collaborative approach between the trainers and the children; an approach that focused on growing the healthy, positive, protective factors that already existed in their communities. For this program, as with all health interventions, it was important to consider all aspects of implementation science, and the successful adaptation and implementation of the program, to ensure sustainability and embed ongoing evaluation for continued improvement of the program over time.

Data availability statement

The original contributions presented in the study are included in the article/supplementary materials, further inquiries can be directed to the corresponding author.

Author contributions

PG: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Project administration, Writing – original draft, Writing – review & editing. RG: Conceptualization, Supervision, Writing – review & editing. CK: Conceptualization, Formal analysis, Supervision, Writing – review & editing. JC: Formal analysis, Supervision, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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References

1. Brands J, Garvey G, Anderson K, Cunningham J, Chynoweth J, Wallington I, et al. Development of a National Aboriginal and Torres Strait islander Cancer framework: a shared process to guide effective policy and practice. *Int J Environ Res Public Health*. (2018) 15:1–16. doi: 10.3390/ijerph15050942

2. Indigenous Allied Health Australia. Cultural responsiveness in action: An IAHA framework: IAHA; (2019). Available from: https://iaha.com.au/wp-content/uploads/2020/08/IAHA_Cultural-Responsiveness_2019_FINAL_V5.pdf (Accessed February 19, 2021).

3. Dossetor P, Freeman J, Thorburn K, Oscar J, Carter M, Jeffery H, et al. Health services for Aboriginal and Torres Strait islander children in remote Australia: a scoping review. *PLOS Glob Pub Heal*. (2019) 19:1–15. doi: 10.1186/s12913-019-4605-0

4. Schuch HS, Haag DG, Kapellas K, Arantes R, Peres MA, Thomson WM, et al. The magnitude of indigenous and non-indigenous oral health inequalities in Brazil, New Zealand and Australia. *Community Dent Oral Epidemiol*. (2017) 45:434–41. doi: 10.1111/cdoe.12307

5. Ralph AP, Noonan S, Wade V, Currie BJ. The 2020 Australian guideline for prevention, diagnosis and management of acute rheumatic fever and rheumatic heart disease. *Med J Aust*. (2021) 214:220–7. doi: 10.5694/mja2.50851

6. Kenyon G. Social otitis media: ear infection and disparity in Australia. *Lancet Infect Dis*. (2017) 17:375–6. doi: 10.1016/S1473-3099(17)30131-7

7. Shattock AJ, Gambhir M, Taylor HR, Cowling CS, Kaldor JM, Wilson DP. Control of trachoma in Australia: a model based evaluation of current interventions. *PLoS Negl Trop Dis*. (2015) 9:e0003474. doi: 10.1371/journal.pntd.0003474

8. Australian Human Rights Commision. Bringing them home: the historical context: AHRC; (1997). Available at: https://bth.humanrights.gov.au/significance/historical-context-the-stolen-generations (Accessed June 3, 2022).

9. Australian Institute of Health and Wellfare. Child protection: AIHW; (2021). Available at: https://www.aihw.gov.au/reports/child-protection/child-protection-system-in-australia (Accessed September 7, 2020).

10. Australian Institute of Health and Wellfare. Youth detention population in Australia 2020: AIHW; (2021). Available at: https://wwwaihwgovau/reports/youth-justice/youth-detention-population-in-australia-2020/contents/summary (Accessed May 4, 2022).

11. Australian Law Reform Commission. Pathways to Justice–Inquiry into the Incarceration Rate of Aboriginal and Torres Strait Islander Peoples (ALRC Report 133): ALRC; (2018). Available at: https://www.alrc.gov.au/publication/pathways-to-justice-inquiry-into-the-incarceration-rate-of-aboriginal-and-torres-strait-islander-peoples-alrc-report-133/ (Accessed October 4, 2020).

12. Burrows E. Chronicling the land rights movement: the democratic role of Australian indigenous land rights publications. *Media Int Australia*. (2016) 160:114–26. doi: 10.1177/1329878X16646218

13. Carson B, Dunbar T, Chenhall R, Bailie R. Social Determinants of Indigenous Health: ProQuest Ebook Central; (2020). Available at: https://ebookcentral.proquest.com/lib/uwsau/detail.action?docID=5145430 (Accessed February 4, 2022).

14. National Indigenous Australian Agency. Stretch reconciliation action plan 2022–2025: NIAA; (2022). Available at: https://www.niaa.gov.au/sites/default/files/publications/niaa-rap-2022-25.pdf (Accessed July 5, 2023).

15. Smith G, Kirkham R, Gunabarra C, Bokmakarray V, Burgess CP. 'We can work together, talk together': an Aboriginal health care home. *Aust Health Rev*. (2019) 43:486–91. doi: 10.1071/AH18107

16. Middleton P, Bubner T, Glover K, Rumbold A, Weetra D, Scheil W, et al. 'Partnerships are crucial': an evaluation of the Aboriginal family birthing program in South Australia. *Aust N Z J Public Health*. (2017) 41:21–6. doi: 10.1111/1753-6405.12599

17. Mokak R. Medicine & Society Oration: Lowitja; (2016). Available from: https://www.lowitja.org.au/page/news-and-events/lowitja-in-media (Accessed June 4, 2022).

18. Bulloch H FW, Bellchambers K. Aboriginal Health and Wellbeing Services: Putting community-driven, strengths-based approaches into practice: The Lowitja Institute; (2019). Available from: https://apo.org.au/sites/default/files/resource-files/2019-11/apo-nid269031.pdf (Acessed August 15, 2021).

19. United Nations. United Nations Declaration on the Rights of Indigenous Peoples (UNDRIP) Australian Human Rights Commission; (2007). [Available from: https://humanrights.gov.au/our-work/un-declaration-rights-indigenous-peoples-1 (Assessed August 10, 2022).

20. Rifkin SB. Examining the links between community participation and health outcomes: a review of the literature. *Health Policy Plan*. (2014) 29:ii98–ii106. doi: 10.1093/heapol/czu076

21. Farmer J, Currie M, Kenny A, Munoz S-A. An exploration of the longer-term impacts of community participation in rural health services design. *Soc Sci Med*. (2015) 141:64–71. doi: 10.1016/j.socscimed.2015.07.021

22. Eccles MP, Mittman BS. Welcome to Implementation Science. *Implement Sci: IS*. (2006) 1:1–3. doi: 10.1186/1748-5908-1-1

23. Estabrooks PA, Brownson RC, Pronk NP. Dissemination and implementation science for public health professionals: an overview and call to action. *Prev Chronic Dis*. (2018) 15:E162. doi: 10.5888/pcd15.180525

24. Metz A, Louison L. The hexagon: An Exploration Tool Hexagon Discussion & Analysis Tool Instructions: NIRN - University of North Carolina at Chapel Hill; (2018). Available from: https://implementation.fpg.unc.edu/wp-content/uploads/NIRN-Hexagon-Discussion-Analysis-Tool_September2020_1.pdf (Accessed June 4, 2022).

25. Dekker M, Jongerden IP, Caris MG, de Bruijne MC, Vandenbroucke-Grauls CMJE, van Mansfeld R. Evaluation of an infection control link nurse program: an analysis using the RE-AIM framework. *BMC Health Serv Res*. (2023) 23:140–12. doi: 10.1186/s12913-023-09111-5

26. Treloar C, Beadman K, Beadman M, Smith K-A, Christian J, Jackson AC, et al. Evaluating a complex health promotion program to reduce hepatitis C among Aboriginal and Torres Strait islander peoples in New South Wales, Australia: the deadly liver mob. *Harm Reduct J*. (2023) 20:1–8. doi: 10.1186/s12954-023-00885-9

27. Wagner B, Cross D, Adams E, Symons M, Mazzucchelli TG, Watkins R, et al. RE-AIM evaluation of a teacher-delivered programme to improve the self-regulation of children attending Australian Aboriginal community primary schools. *Emot Behav Diffic*. (2020) 25:42–58. doi: 10.1080/13632752.2019.1672991

28. Cama E, Beadman K, Beadman M, Walker M, Treloar C. Insights from the scale-up and implementation of the deadly liver mob program across nine sites in New South Wales, Australia, according to the RE-AIM framework. *Harm Reduct J*. (2023) 20:1–14. doi: 10.1186/s12954-023-00889-5

29. Holtrop JS, Estabrooks PA, Gaglio B, Harden SM, Kessler RS, King DK, et al. Understanding and applying the RE-AIM framework: clarifications and resources. *J Clin Transl Sci*. (2021) 5:e126. doi: 10.1017/cts.2021.789

30. Boulton J. *Aboriginal children, history and health: Beyond social determinants*. New York: Routledge (2016).

31. Harrison N, Sellwood J. *Learning and teaching in Aboriginal and Torres Strait islander education. 3rded.* Australia & New Zealand: Oxford University Press (2016).

32. Malpa. Young Doctors for Life (2021). Available from: https://www.malpa.org.au (Accessed February 4, 2022).

33. Stapf I, Biess C, Pfetsch J, Paschel F. Respecting children's rights in research ethics and research methods. *J Child Media*. (2023) 17:393–9. doi: 10.1080/17482798.2023.2235815

34. Fixsen D, Blase K. Implementation: The missing link between research and practice: National Implementation Research Network; (2009). Available from: https://files.eric.ed.gov/fulltext/ED507422.pdf (Accessed February 5, 2022).

35. Cox T, Hoang H, Mond J, Cross M. 'It all comes back to community!': A qualitative study of Aboriginal elders promoting cultural well-being. *Aust J Rural Health*. (2021) 29:909–17. doi: 10.1111/ajr.12791

36. Sharmil H, Kelly J, Bowden M, Galletly C, Cairney I, Wilson C, et al. Participatory action research-Dadirri-Ganma, using yarning: methodology co-design with Aboriginal community members. *Int J Equity Health*. (2021) 20:160–11. doi: 10.1186/s12939-021-01493-4

37. Cairns A, Geia L, Kris S, Armstrong E, O'Hara A, Rodda D, et al. Developing a community rehabilitation and lifestyle service for a remote indigenous community. *Disabil Rehabil*. (2022) 44:4266–74. doi: 10.1080/09638288.2021.1900416

38. Priest N, Mackean T, Davis E, Waters E, Briggs L. Strengths and challenges for koori kids: harder for koori kids, koori kids doing well - exploring Aboriginal perspectives on social determinants of Aboriginal child health and wellbeing. *Health Sociol Rev*. (2012) 21:165–79. doi: 10.5172/hesr.2012.21.2.165

39. HOPE. Healthy Outcomes From Positive Experiences: HOPE; (2020). Available from: https://positiveexperience.org/ (Accessed August 10, 2022).

40. Priest N, Thompson L, Mackean T, Baker A, Waters E. 'Yarning up with koori kids' - hearing the voices of Australian urban indigenous children about their health and well-being. *Ethn Health*. (2017) 22:631–47. doi: 10.1080/13557858.2016.1246418

41. Liew S, Gwynn J, Smith J, Johnson NA, Plotnikoff R, James EL, et al. The barriers and facilitators of sport and physical activity participation for Aboriginal children in rural New South Wales, Australia: a Photovoice project. *Int J Environ Res Public Health*. (2022) 19:1986. doi: 10.3390/ijerph19041986

42. Morton D, Bird-Naytowhow K, Pearl T, Hatala AR. "Just because they aren't human doesn't mean they aren't alive": the methodological potential of photovoice to examine human-nature relations as a source of resilience and health among urban indigenous youth. *Health Place*. (2020) 61:102268–10. doi: 10.1016/j.healthplace.2019.102268

43. Beardon C., Wilson J. (2013). Experiential learning: A handbook for education, Training and Coaching. Kogan Page, Limited, ProQuest Ebook Central. Available at: https://ebookcentral.proquest.com/lib/uwsau/detail.action?docID=1318826

44. Domínguez N. In: , P Collier , editor, Developing effective student peer mentoring programs: A Practitioner's guide to program design, delivery, evaluation, and training, Taylor & Francis Group, (2015). ProQuest Ebook Central. Available at: https://ebookcentral.proquest.com/lib/uwsau/detail.action?docID=4438627

45. Ortiz V, Virnoche M. In: , P Collier , editor, Developing effective student peer mentoring programs: A Practitioner's guide to program design, delivery, evaluation, and training, Taylor & Francis Group, (2015). ProQuest Ebook Central. Available at: https://ebookcentral.proquest.com/lib/uwsau/detail.action?docID=4438627

46. Rapport F, Clay-Williams R, Churruca K, Shih P, Hogden A, Braithwaite J. The struggle of translating science into action: foundational concepts of implementation science. *J Eval Clin Pract*. (2018) 24:117–26. doi: 10.1111/jep.12741

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Looking after bubba for all our mob: Aboriginal and Torres Strait Islander community experiences and perceptions of stillbirth

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The stillbirth rate among Aboriginal and Torres Strait Islander women and communities in Australia is around double that of non-Indigenous women. While the development of effective prevention strategies during pregnancy and improving care following stillbirth for women and families in communities has become a national priority, there has been limited progress in stillbirth disparities. With community permission, this study aimed to gain a better understanding of community experiences, perceptions, and priorities around stillbirth. We undertook an Indigenous researcher-led, qualitative study, with community consultations guided by a cultural protection protocol and within an unstructured research framework. A total of 18 communities were consulted face-to-face through yarning interviews, focus groups and workshops. This included 54 community member and 159 health professional participants across remote, regional, and urban areas of Queensland, Western Australia, Victoria, South Australia, and Northern Territory. Thematic analysis of consultation data identified common themes across five focus/priority areas to address stillbirth: Stillbirth or Sorry Business Baby care needs to be family-centered; using Indigenous "ways of knowing, being, and doing" to ensure cultural safety; application of Birthing on Country principles to maternal and perinatal care; and yarning approaches to improve communication and learning or education. The results underscore the critical need to co-design evidence-based, culturally appropriate, and community-acceptable resources to help reduce existing disparities in stillbirth rates.

KEYWORDS

stillbirth, Aboriginal and Torres Strait Islanders, Indigenous, grief and loss, perinatal loss, antenatal care, maternal and infant health, Sorry Business

Introduction

Stillbirth is a serious public health problem with far reaching psychosocial and financial burdens for families and communities (1). In Australia, there were 315,705 babies born in 2021, with ∼2,278, or 76% of perinatal deaths due to stillbirth (2). This equates to *six* babies being stillborn every day (birth of a baby without signs of life after 20+ weeks or weighing 400+ grams (3) making stillbirth the most common form of perinatal/infant death across the general Australian population (4, 5). There have been minimal changes in stillbirth rates in the last three decades and persistent disparities exist across high income country settings.

The 2021 Australian Census reported that 812,728 people had identified as being of Aboriginal and/or Torres Strait Islander origin, representing 3.2% of the total Australian population of 25,422,788 people (6). The stillbirth rates among Aboriginal and Torres Strait Islander (herein respectfully referred to as Indigenous) communities is typically about twice that of the general population (12 compared with 7.2 per 1,000 births, respectively) (2, 5) and there has been limited progress in reducing this disparity (2, 5, 7, 8).

Furthermore, when an Indigenous baby is stillborn, appropriate care models that consider women's cultural needs may be ill-understood, and often suboptimal, resulting in a greater burden of grief on Indigenous communities (9, 10). Data on causes of stillbirth in Indigenous communities indicate that many stillbirths are preventable (9). This indicates a greater need for improvements in maternity care and raising community awareness on prevention and care during pregnancy (1, 3, 9). However, limited resources are available for health professionals to provide both culturally responsive information and care and respectfully consider traditional practices around stillbirth prevention (7). Prevention of stillbirth and pregnancy care requires systemic change in the healthcare environment, ensuring co-led stillbirth risk reduction campaigns with Indigenous community-controlled organizations and service providers (7). The Stillbirth Center for Research Excellence (CRE) is working to improve prevention and care for Indigenous people when such losses occur (11), along with organizations such as the Stillbirth Foundation Australia (12), Red Nose (13), and Still Aware (14) focusing on awareness, education, research, and advocacy. Prior to the introduction of the Safer Baby Bundle, and with the exception of some Still Aware resources (14), there were very few robust resources available on the prevention of stillbirth (15).

Following the Senate Enquiry into stillbirth research and education (4) the National Stillbirth Action and Implementation Plan was launched (16). The Plan aims to reduce stillbirth rates by 20% or more from 2020 to 2025 in Australia and to improve bereavement care through various ways including calls for a concerted national public awareness campaign (17). Furthermore, the Plan recognizes the significant equity gaps in stillbirth prevention and care, including those that exist between mainstream populations and Indigenous peoples and members of some migrant and refugee communities (5). The Safer Baby Bundle is the national stillbirth prevention initiative of the Plan. The Safer Baby Bundle focuses on five elements of maternity care practices where evident-practice gaps exist: smoking cessation support; improving detection and management of fetal growth restriction; improving awareness and management of women with decreased fetal movements; provision of maternal safe sleeping advice; and improving decision-making around timing of birth for women with risk factors (18). However, these resources were developed for stillbirth prevention for the general Australian population and are unlikely to be effective in Indigenous populations (10, 11).

The recent "Still Six Lives" national media campaign used digital marketing and social media to increase awareness of stillbirth and educate on the three modifiable behaviors during pregnancy to reduce the risk of stillbirth; its evaluation showing some evidence of effectiveness in increasing the proportion of Australian women who were aware of these three evidence-based preventive actions that reduce the risk of stillbirth (19). Pollock et al. (20) reported in their study there were limited levels of knowledge, attitudes, and perceptions of stillbirth in the general population, which warranted the need for future public health campaigns. Furthermore, the study highlighted the need for future co-designed research that "addresses the culture, values, needs and wants of a stillbirth public health campaign" with Indigenous peoples (20). Therefore, it is critical to frame stillbirth risk messages to ensure they are relevant in an Indigenous cultural context, are tailored to a family-centered approach and also to the realities of women's and families' lives (7). Actions to address equity gaps such as the recommendation for culturally appropriate models of care during the perinatal period (16) can be achieved through consultation and partnership in stillbirth prevention and care with Indigenous Australian communities and health services, including resource co-design and implementation. Therefore, understanding Indigenous people's experiences, perceptions, and priorities around stillbirth is key to implementing effective culturally safe prevention strategies in Australian maternity health services.

This paper aimed to identify needs around maternity care to prevent stillbirth with and for Indigenous Australian peoples. The second aim of the paper was to identify appropriate language for the term stillbirth with Indigenous communities.

Methods

Project foundation and values

This collaborative consultation process was led by dedicated, experienced Indigenous researchers, who have deep cultural connections with the lands and communities they come from. Indigenous research team members followed a strengths-based approach, amplifying the voices, experiences and needs of Indigenous women, families, and community members, harnessing and highlighting the strengths and resilience of Indigenous people. Researchers used yarning methods to provide a comfortable, safe space for community members to share stories, ask questions and feel heard. Indigenous researchers ensured Indigenous "ways of knowing, being, and doing" were honored (21). Relationship building with certain Aboriginal and Torres Strait Islander communities and Aboriginal Community-Controlled Health Organizations (ACCHOs) in South Australia, Far North Queensland, Victoria, New South Wales, and Western Australia occurred from 2019 to 2022 and explored community experiences, perceptions, and priorities in relation to stillbirth prevention and maternity care.

This paper reports on consultations with Indigenous communities around Australia, which formed the qualitative component (Phase 1) of a wider program of work—the cultural adaptation of the Safer Baby Bundle for Indigenous communities. Community consultations informed resource co-design approaches for an Indigenous-led cultural adaptation of the Safer Baby Bundle resources for community, and health professional educational resources, which will be reported in a future publication.

Reflexivity of researchers

The Stillbirth CRE Indigenous Research Team is made up of Senior Advisor and Chair of the Stillbirth CRE Indigenous Advisory Group (IAG), Deanna Stuart-Butler (DSB), a descendant of the Arabana people of the "Pantu Parnda" (Lake Eyre) Region of South Australia. Deanna was leader of the Aboriginal Family Birthing Program in South Australia and conducted the consultations in Far North Queensland and South Australia. Carolyn Lewis (CL), a Yamatji Noongar woman and Aboriginal Research Fellow at Curtin University, led the consultations across Western Australia. Skye Stewart (SS), a Wergaia and Wemba Wemba midwife from Mallee Victoria, midwife and Research Coordinator led consultations in Victoria in discussion with Victorian Aboriginal Community Controlled Health Organization (VACCHO). Diana Jans (DJ), an Indigenous researcher with cultural connections to Far North Queensland undertook consultations in the Cape York region with DSB. Diana is a teacher, social worker, bereavement counselor and Narrative Therapist who has worked in human and child protective services and Apunipima Cape York Health Council in maternal, child and adolescent health since 2014. Senior research and clinical academics with extensive experience in midwifery, perinatal health (VF, PM, FB, SV) and Indigenous health (KMR, CS, SV) were integral in setting up and guiding the study. With expertise working in Indigenous health, SV worked closely with the team consulting in Queensland from the outset and played a major role getting the study underway, in consultations and qualitative analysis with the team. Early career researchers with experience in Indigenous health, perinatal health, and qualitative research (LM, RG, AB, LJ) were also part of the Stillbirth CRE Indigenous Research and authorship team. The Stillbirth CRE's National Indigenous Advisory Group, made up of Indigenous leaders in the maternity and infant health space, provided guidance and leadership from the outset of the community consultation work.

Study design

The study was guided by Participatory Action Research principles (PAR) (22–24), and Indigenous-specific collaborative frameworks (23, 25, 26). Participants were interviewed via open individual and/or group discussions, usually as part of yarning circles—a culturally appropriate method of data collection. Sharing stories through yarning circles has been a way of learning for Indigenous people throughout time (21, 27). These stories often reflect the participant's lived experience, affirm identity, and allow sharing of each other's life and culture (26). While yarning, Indigenous people weave stories together, which enables the listener to make family, community, and Country connections (21). Yarning is a process which occurs in various forms, and includes social, research, and therapeutic yarns and can often lead from one into the other (27). In yarning, Indigenous people begin with a social yarn, discussing social connections, and establishing trust (27). Then, in a consultative or research yarn, the researcher and participant/s talk freely on an agreed subject without being limited by directive questions. Stories are often featured, which may provide a layered illustration of multiple issues that can be explored from various angles (28). It is a way of showing respect, building trust and often can promote healing when discussing a sensitive issue like stillbirth (26). Yarns conducted were also at times therapeutic yarns (27), allowing the opportunity for women and family members to talk about their loss.

Consultations and yarning were facilitated by up to four Aboriginal researchers (DSB, DJ, CL, SS), and at times one or two non-Aboriginal researchers (SV, AB) with community members and Indigenous and non-Indigenous health care professionals caring for Indigenous women and families. Prior to consultations a group yarning/interview discussion guide was developed to help guide conversations. Sample questions from the yarning/discussion guide included: *Do you think stillbirth is discussed with Aboriginal and Torres Strait Islander women during pregnancy? What is the best way to share information about stillbirth and provide support for Aboriginal and Torres Strait Islander women during pregnancy? What education/training is needed for clinicians? Are there current gaps in educational resources for community and health professionals? Is stillbirth an appropriate name for us to use in our communities?* Although care after stillbirth was not the focus of the study, it is not possible to talk about stillbirth prevention without also discussing the devastating consequences of stillbirth. Therefore, a question around care after stillbirth and/or the community experience of stillbirth was also included. The discussion guide focused the consultations on stillbirth risks and prevention, and opportunities on how resources should be developed for community needs.

Context, collaborative consultation process and approach

The COVID−19 pandemic led to travel restrictions from 2020, which adversely impacted on the project timing and ability of the research team to visit many regional and remote communities. For this reason, a pragmatic approach was followed for the nationwide consultation. The CRE Indigenous Research Team decided to consult in targeted locations, that allowed for a national perspective ensuring meaningful consultations occurred at each site, while also prioritizing community safety during the pandemic. Face-to-face consultations with health professionals and community members were held in Far North Queensland (Cape York region), Western Australia, Victoria, New South Wales, and South Australia. The consultations started in 2019, and were finalized in 2022, including a delay of ∼10 months due to COVID-19 pandemic disruptions (throughout early 2020—late 2021). During this period travel restrictions included limited travel between some states as borders closed, particularly Queensland and Western Australia, as well as the restriction of visitors into many Indigenous communities to stop virus spread in remote areas. Online platforms were used at times; however, face-to-face discussions were preferred by community and therefore prioritized, resuming in late 2021/22.

The pragmatic research consultation meant that data collection encompassed several different approaches, including collecting data through forums, workshops, focus groups, face-to-face and at times online yarns. Consultations in some regions involved face-to-face information sessions on stillbirth and the Safer Baby Bundle resources (18) followed by informal discussions on how to adapt stillbirth prevention information with Indigenous communities. The regions consulted were urban, regional, rural, and remote areas, to ensure the inclusion of a wide breadth of experiences and perspectives.

Selection of participants

The choice of participant groups was based on existing relationships and networks between the Indigenous Research Team and ACCHOs and other health service organizations in South Australia, Victoria, Western Australia, and Queensland. The consultations focused on two participant groups: (1) Community members including Indigenous women of reproductive age (18 + years of age), men, and Elders; and (2) health care professionals, including maternity health professionals, and social and emotional wellbeing workers based in community health centers and clinics, program managers and policy makers, researchers working with Indigenous women in pregnancy and others who work with Indigenous communities. This included participants with lived experience of stillbirth. Staff information sessions at different locations were offered as both drop-in and open yarns, rather than structured interviews or focus groups. The two participant groups were interviewed/yarned separately, and some health care professionals who also identified as members of Indigenous communities provided feedback as both a community member and a health care professional, at the individual's discretion.

Data collection

Consultations included group and individual interviews/yarns held at health services, community centers and when requested at community members' homes. Most consultations were audio recorded, transcribed verbatim and all were de-identified. A small number of consultations were not recorded, at the discretion of Indigenous Researchers conducting the yarn. This may have been due to the sensitive nature of the topic of stillbirth and may also have been due to location of yarn and the comfort level of participants to speak freely with a recording device present. However, extensive field notes were taken on the discussion, topics covered, and stories shared.

Data analysis

Prior to coding, transcripts were checked for accuracy against the recording and de-identified. Analysis and interpretation were conducted collaboratively with the Stillbirth CRE's Indigenous Research Team using an iterative process. Indigenous Team members who collected the data, collaborated to refine the themes, and led the interpretation of the consultation data. Qualitative data from yarning circles and interviews were analyzed to identify themes using Framework Analysis (29). The Framework method offers a structured but adaptable approach to the thematic analysis of qualitative data (29). This method allows for the use of a pre-existing framework or set of key questions but is flexible enough to take account of new or differing concepts or issues that arise in the process of collecting data. To ensure conceptual consistency and interrater reliability at least two members of the research team conducted the analysis. The qualitative data analysis was conducted using NVivo 12 (QSR), and Word tables (Microsoft 365 Office). A summary of thematic findings and illustrative quotes and anecdotes are reported in Results.

Results

The analysis in this paper reports on the qualitative data collected from group and individual interviews and yarns conducted face to face with 135 participants in Far North Queensland (FNQ), South Australia (SA), Victoria (VIC), and Western Australia (WA); these were specifically aligned with the stated aims of this paper (see Table 1 for participant characteristics). These interviews form part of a broader set of consultations which were held in 18 communities, with 54 community members and 159 health care providers (many of whom also identified as Indigenous community members), across remote, regional, and urban areas of Australia (see Supplementary Table 1 for an outline of specific consultation characteristics).

Far North Queensland consultations

In total, 34 participants who ranged in age from 20 to 80 years took part in the consultations. Participants were predominantly female (85%); 55% identified as Aboriginal; 9% as Aboriginal and Torres Strait Islander or Torres Strait Islander; and 36% identified as non-Indigenous participants (from a variety of cultural backgrounds). Ten interviews were completed in this region, and all participants identified that even if they personally had not experienced stillbirth in their own life, they knew someone who had. Communities visited in the Cape York region included Weipa, Napranum, Mapoon, and Hopevale, with these sites chosen to provide a good representation of the community and the affiliation of the research team to Apunipima Cape York Health Council (the ACCHO for the Cape York region).

South Australia consultations

There were two consultations held in SA with the first consultation facilitated by the Aboriginal Communities and

TABLE 1 Participant characteristics—roles and Indigenous status.

Participants role	Female	Male	Total participants	Participants who identified as Indigenous
Far North Queensland				
Health professionals[a]	25	4	29	22
Community members, incl. Elders	4	1	5	5
Totals FNQ	**29**	**5**	**34**	**27**
South Australia				
Consultation 1				
Health professionals[a]	-	-	-	
Community members, incl. Elders	24	-	24	20
Total—Consultation 1	**24**		**24**	**20**
Consultation 2				
Health professionals[a]	15	-	15	13
Community members, incl. Elders	2	-	2	2
Total—Consultation 2	**17**	-	**17**	**15**
Totals SA	**41**		**41**	**35**
Western Australia				
Health professionals[a]	33	-	33	30
Community members, incl. Elders	16	1	17	17
Total WA	**49**	**1**	**50**	**47**
Victoria				
Health professionals[a]	2	-	2	2
Community members, incl. Elders	4	4	8	8
Total VIC	**6**	**4**	**10**	**10**
Overall totals	**125**	**10**	**135**	**92**

[a] Midwives, Aboriginal Health Workers, managers, policy makers, researchers, and other health professionals.

Cell counts with <5 were not tabulated to ensure confidentiality. Bold values indicate subtotals and total numbers of participants.

Families Health Research Alliance (ACRA) in Adelaide (Tarndanya), with 24 attendees including service providers, program managers, policymakers, planners, and researchers working with Indigenous families during pregnancy. The focus of the sessions was on stillbirth research priority setting for Indigenous families, review of current practice, cultural safety, and stillbirth within Indigenous birthing programs. The second consultation was held in Adelaide with 17 Aboriginal Maternal and Infant Care (AMIC) practitioners, health care providers and a community member. The session discussed how stillbirth prevention, care and bereavement care fit into the AMIC principles that guides their work; to ensure optimal care for Indigenous women and families in South Australia.

West Australia consultations

Four yarning circles were undertaken with community members and health services staff of rural, remote, and urban areas of Western Australia to elicit knowledge and experiences of stillbirth, risks, prevention, and care. Two of the four yarning circles held were two-way knowledge exchange workshops led by Elders and the Indigenous Researcher and conducted with Indigenous and non-Indigenous health professionals, community members, and families. Consultations in WA were held in the following locations: urban—Derbarl Yerrigan Health Service, East Perth (Booroolo); regional—South-West Aboriginal Medical Services (SWAMS), Bunbury; rural/regional—Geraldton Regional Aboriginal Medical Service (GRAMS), Geraldton and Bega Garnbirringu Health Service (BEGA), Kalgoorlie.

Victoria consultations

Five consultations with eight community members and two health services staff were held in rural, regional, and urban areas of Victoria to elicit their awareness, knowledge and experiences of stillbirth, risks, prevention, care during and after stillbirth. Consultations were held in various locations including urban—Melbourne (Naarm); rural/regional—Warnambool (Gunditjmara); Swan Hill (Wamba Wamba); Sea Lake (Wergaia); online consultation—with participants from bordering regional/rural area of NSW and Victoria.

Qualitative results

"Sorry Business Babies"—language for stillbirth

A key aim of the study included identifying a culturally appropriate term for stillbirth. "Sorry Business" is a term used by Indigenous Australians to encompass the time of traditional rites and customs related to death; it is a mourning period when a family or community member dies and describes all responsibilities that follow in accordance with traditional lore and custom, such as grief, loss and funerals (30, 31). Stillbirth is referred throughout this work as "Sorry Business Baby" or "Sorry Business Bubba." In other regions they have been referred to as "Star Babies." We started with the concept of Sorry Business and refined the use of this term after early consultations. "Sorry Business Bubba" was the term used by the Stillbirth CRE Indigenous Research team when referring to stillbirth, or when a baby passes away after 20 weeks of pregnancy or during birth.

> *When a woman and family are going through Sorry Business after having a stillborn baby, they suffer tremendous grief and loss. It is also a difficult time for their health carers. Stillbirth is a sensitive issue in Aboriginal and Torres Strait Islander communities—there is little open discussion.* (Deanna Stuart-Butler, Stillbirth CRE Indigenous Research Team Senior Advisor)

Based on consultation findings and early guidance from the Indigenous Advisory Group, the term was used by CRE Indigenous researchers to enable more open yarning around the taboo topic of stillbirth. The term was also used in the cultural adaptation of the Safer Baby Bundle project work.

> *Know that after stillbirth, Aboriginal families are in Sorry Business which is a cultural rite and responsibility. It means that we grieve collectively and there may be practices we do at this time to honor the bub who has passed. It might mean that we need more time when talking to us about next steps, like autopsy and other stillbirth investigations. It will be hard for family to make quick decisions. Sorry Business goes for days, weeks, or months and it doesn't matter if it is a bubba or an Elder, the Sorry Business and life of that person is equal.* (Aboriginal midwife and woman who experienced loss, VIC)

Awareness of the sacred nature of Sorry Business was identified as culturally appropriate practices that helped families after stillbirth, and were an important facilitator during consultations.

> *Culture is the protective factor for Aboriginal people. It is what keeps us well and safe. The death of anyone in Aboriginal community means there is a time of Sorry Business, which is a time of cultural rite and responsibility.* (Skye Stewart, Stillbirth CRE Indigenous Research Officer and Midwife)

Thematic findings—five priority areas

Overall, thematic analysis identified five key themes to address improvements in stillbirth prevention and bereavement care. These included: (a) Stillbirth or Sorry Business Baby care needs to be family-centered, (b) Using Indigenous "ways of knowing, being and doing" to ensure cultural safety, (c) Application of Birthing on Country principles to perinatal care, (d) Yarning approaches to improve communication, and (e) Learning and education through stories (see Figure 1). The five priority themes and illustrative quotes from consultations across the four regions are outlined below.

Stillbirth or Sorry Business Baby care needs to be family-centered

From these consultations it was clear that family-centered care was paramount during a time of grief and loss when a baby is stillborn. Participants stated that grief and loss services were needed across urban, rural, and remote areas, recognizing that a focus on more than just "mum and dad" was required. Health care workers were needed to empower the whole family, including parents and other family members involved in the decision-making process for the baby:

> *Lots of different things can happen and for [a] family to have the shock of their baby dying and then have to go through all these other things... "I've got all these doctors and people wanting me and yet I've got all my family, who are wanting me to tell them what happened and how come?"… Lots of pressures. Having that [health service] support, is a really good idea.* (Aboriginal health care provider, Cape York FNQ)
>
> *Definitely [there is] a lack of support service such a grief and loss services for the mother and other family members to access.* (Aboriginal midwife, WA)

Participants identified it was important that families feel enabled to make their own decisions for their stillborn baby, to avoid feeling scared and shamed, and to be able to ask for what they need—culturally, physically, and emotionally.

> *It feels like Aboriginal community have some shame job around... talking about it. At the same time, it feels like health providers are awkward or don't want to start the conversation in case there are questions they don't know how to answer. What is required for families is for health providers to be confident and kind in their conversations, as it needs to be talked about.* (Bereavement worker, VIC)

Family-centered care encompasses both the mother and father and generally the extended family as well. Culturally, even though birth is Women's Business,[1] participants outlined that there was a

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[1] Women's Business refers to the practices, ceremonies, and Lore that are specific and sacred to Aboriginal and Torres Strait Islander women. Women's Business is performed separately from Men's Business and can relate to health, upbringing, ceremonies, and maintenance of culturally significant

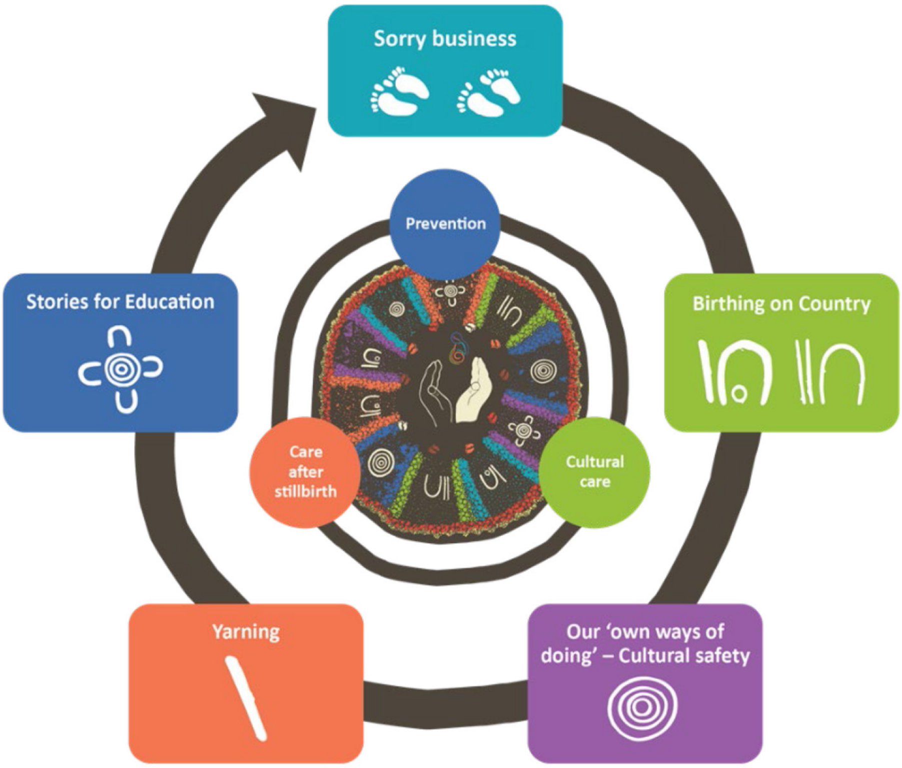

FIGURE 1
Five focus areas based on thematic analysis of consultations.

place for men in Sorry Business. This included how the health care team responded to the father or other male relatives. Participants indicated that men had responsibilities, not only in carrying their grief, but in supporting their partner. It is however important to note, that we recruited and only consulted with a small number of men, with the following reported by a male participant:

> *When things are tough, we have to stay strong and if that means there is a stillbirth, we need to support our women, as well as somehow keep carrying on, even in our own grief. Sometimes we have our own traumas, and we are treated badly by the hospital staff, it makes us shame job. We don't need to carry shame; we are doing our best and we need support too.* (Aboriginal father, VIC)

Using Indigenous "ways of knowing, being, and doing" to ensure cultural safety

Embedding Indigenous "ways of knowing, being, and doing" to ensure a culturally safe and responsive practice was identified as a key theme in all locations. Indigenous "ways of knowing, being, and doing" encompass the following principles as expressed by Martin and Mirraboopa (21):

> *"Recognition of our worldviews, our knowledges and our realities as distinctive and vital to our existence and survival; Honoring our social mores as essential processes through which we live, learn and situate ourselves as Aboriginal people in our own lands and when in the lands of other Aboriginal people; Emphasis of social, historical and political which shape our experiences, lives, positions and futures; Privileging the voices, experiences and lives of Aboriginal people and Aboriginal lands."*

This was highlighted in our study as being especially pertinent in relation to adverse situations during pregnancy and birthing healthcare experiences. Participants identified a lack of cultural safety that included: disempowerment; health professionals not always communicating around the time of stillbirth, which was essential to ensuring women and their families' understanding; lack of trust in people and the health system resulting in less confidence to engage. Racism was experienced by Indigenous people in general health care settings, which significantly impacted on cultural safety. Application of culturally safe practices in regional health centers was questioned, including health care staff's understanding of community customs; willingness to consult the family about the cultural and other arrangements appropriate for them.

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places and sacred sites; it is also a way of reconnecting with the ancient wisdom, tools, and culture to pass down the next generation of young women [Source: Cultural Ways—Learn—Australian Indigenous HealthInfoNet (ecu.edu.au)].

> *We don't shy away from hard yarns but if we feel unsafe by systems or people around us, then we shut down. It is a coping mechanism. We have been so hurt in the past and we protect ourselves from harm. If the health providers talk to the Aboriginal community in a way that shows cultural integrity and safety in their practice, then it gives that room for us to feel alright to open up. Aboriginal people want to receive health care. We just want that health care to be safe for us.* (Community member/parent, VIC)

Discussions related to this theme also identified that organizational and individual responsibility to deliver culturally safe care is of paramount importance; care needs to be considered, acknowledging of individual differences; professionals need to engage in cultural humility and reflective practice to ultimately deliver culturally responsive and responsible care that women, families, and communities deserve. Aboriginal maternal and infant health care staff and Aboriginal health worker participants identified several areas of need:

> *Aboriginal ways of knowing, being and doing surpass anything else and need to be at the center of the care and the decision-making process.* (Bereavement worker, VIC)
>
> *I know with our mob [health professionals should] ask because sometimes those women [sic bereaved mothers] are never given the privilege of being asked if they want to keep the placenta. And umbilical cord.... You've got something else of the baby... now we seem to see this resurgence of wanting to go home on country, wanting to know where their birth tree is or cave or you know? So, my thing is, ask.* (Aboriginal and Torres Strait Islander Health Worker, female Elder, Cape York, FNQ)

Embedding Indigenous "ways of knowing, being and doing" has been recognized as a key feature in culturally-safe maternal and infant health programs and services run through ACCHOs, as they foster cultural connections for women and families (32–35). Aboriginal community-controlled health organizations aim to deliver services in a way that engenders trust and are more acceptable to local communities.

> *As an organization [ACCHO], you know, we are about cultural safety and that honesty and an open approach. Otherwise, people will not trust us. If they don't trust us, then we may as well just stop doing what we do.* (Non-Indigenous doctor, male, Cape York, FNQ)

Birthing on Country principles for perinatal care

Birthing on Country is a metaphor for the best start in life for First Nations families (36). The term recognizes that when women give birth in Australia, they are doing so on the sovereign lands of the First Peoples of Australia who have never ceded ownership of their land, seas, and sky (36). Birthing on Country principles encompassed the delivery of holistic, continuity of care models for women and families, and where possible, for women to have a Cultural Primary Carer during pregnancy and beyond.

> *"Aboriginal led antenatal care [AHW/AMICs] at all appointments and culturally appropriate models of care, including continuity of care, which women trust and feel comfortable with are important."* [Aboriginal Maternal and Infant Care (AMIC) Worker, SA]

There is growing awareness and respect of the importance of Birthing on Country for Indigenous women, with a few initiatives now starting in maternity care in Australia (32, 35, 37). Birthing on Country not only refers to redressing the negative impact of colonization and returning childbirth services to Indigenous communities and control; it also encompasses services which are characterized as "community-based and governed; allow for incorporation of traditional practice; involve a connection with land and country; incorporate a holistic definition of health; value Indigenous and non-Indigenous ways of knowing and learning, risk assessment and service delivery; are culturally competent; and developed by, or with, Indigenous people" (36). As stated by a maternal and infant care worker:

> *"Supporting women to birth on Country or birth within community—Birthing programs are a positive step and should be rolled-out wider."* (AMIC worker, SA)

However, some participants stated that there were and continue to be challenges applying Birthing on Country principles reported, such as the management and loss of continuity of carer, with many women birthing at the hospital, which is a separate service and therefore without the ACCHO doctors and midwives, that provided their antenatal care at their local ACCHO. This is especially the case for women having to travel from remote and rural locations to birth in larger regional and urban maternity hospitals for delivery and/or manage later pregnancy complications. The practice of relocating women away from home for birthing presented additional challenges.

> *I think it's a combination of history and culture and a mistrust of organizations, and the fact that people have been let down possibly in the past... I think it's a loss of cultural norms for people who would have birthed on Country, and Elders in community, female Elders in community would have managed a lot of those processes. And then we've had the whole swathe of the western lifestyle negative influences coming in, you know, which have just complicated people's pregnancies and made it so much more difficult.* (Non-Indigenous doctor, male)

Yarning approaches to improve communication

"Yarning" methods are recognized as a culturally appropriate way of collecting information or data with Indigenous women and their families (27). Participants reported that when Indigenous families felt safe and cared for, they might be more likely to share. Therefore, health care practitioners were advised to provide space and opportunities for amplifying the voices of Indigenous peoples in their care. It was important that families knew that they were in control and able to make their own decisions. Using "strengths-based language or approaches" refers to those approaches that have their foundations in Indigenous ways of knowing, being and doing, and "view strengths not as the possessions of individuals, but as the structure and quality of the social relationships, collective practices and identities that are present in Indigenous communities (38). Strengths-based approaches should therefore be used in yarning with community about health. This was however identified by health professionals as an area in need of improvement."

> *Stillbirth is sad, how do you communicate effectively?* (non-Indigenous midwife, WA)
>
> *First thing [the non-Indigenous health care provider] said off the top of their head to them "you've got uncontrolled diabetes so you're at risk of stillbirth" and that woman is just sitting there, and you sort of sit there too and think 'Did [they] just really say that out loud?' and there was no care, it was just like [it's a] pre written record.* (Indigenous AMIC practitioner, SA)

Indigenous families highlighted that medical conversations, particularly around investigations after stillbirth, needed to happen however it was reinforced that it was important that the care from health professionals was culturally safe throughout the entire pregnancy. Aboriginal Health Workers (AHWs) were crucial for having a yarn with families and must be available at hospital at a time of Sorry Business. They can explain medical terminology, speak up and ask questions on behalf of the parents.

> *AHW/AMIC continuity of care to support women to understand and translate medical information [is needed].*
>
> *Education for non-Indigenous health workers to have conversations around Aboriginal culture and practices [is needed].* (AMIC worker, SA)

Stillbirth/Sorry Business Babies is a difficult topic and is generally understood to be a taboo subject to talk about in Indigenous communities. Therefore, a two-way exchange such as through yarning approaches needs to take place between health professionals and bereaved families. Open clear communication from the health care practitioner, in a way that is safe and respectful and holds cultural integrity is paramount:

> *Information that is new to you is hard to digest when you are talking about your precious baby that died. Talk calmly and don't rush the family if you are the health care practitioner caring for them. Just allow quiet space and don't expect too many words or for us to give the answers all at one time.* (Bereavement worker, female, VIC)
>
> *If the doc needs to refer… [and] there's this issue, I say "Can you change it to just you need someone to yarn to."* (Aboriginal and Torres Strait Islander Health Worker, female Elder, Cape York, FNQ)

Learning or education through stories

Indigenous cultural practices which have positive effects on children and communities include kinship relations, oral traditions that are often centered around stories, traditional knowledge systems, a collective community focus with respect for Elders' contributions, and spirituality (27, 39, 40). Ideas were highlighted such as the need for more Indigenous health professionals who have lived experience and knowledge of history and culture, who can share this through their stories for strengthening education. Additionally, stories could be used by non-Indigenous health professionals when explaining sensitive topics or medical information.

> *Education for clinicians—around complex trauma, how historical trauma, trauma associated with coming into a hospital, etc. may compound distress.*
>
> *Education for women/families on causes, and that often a stillbirth is unexplained, and no one's fault, is important to help women and families through the guilt after a stillbirth.* (AMIC worker, SA)

Discussion

We have reported on the processes undertaken by the research team that ensured a thorough and robust consultation around a sensitive topic to ensure representation from various Indigenous communities in most Australian States and Territories. Like other researchers and community leaders before them, this team led by Indigenous researchers, acknowledged that Indigenous women are not a homogenous group, rather they are characterized by differences in culture and traditions, languages, socio-economic circumstances, places of residence, educational levels, and employment status (40, 41). Despite the challenges imposed by the COVID-19 pandemic this team undertook a pragmatic approach and gained significant insights into this unmet need. The work highlighted the need for a culturally appropriate term for stillbirth and the yarning approach around the topic elicited communities' perspectives on a culturally appropriate term. Our findings have underscored that there are minimal culturally specific resources available, and limited understanding about responsive, culturally safe maternity and maternal and infant health services, particularly related to stillbirth for Indigenous people. The priority focus areas identified through this thematic analysis will consolidate approaches needed to address stillbirth in Indigenous communities. This which will include the next phases of development of appropriate stillbirth prevention resources based on the Safer Baby Bundle for Indigenous people.

Conducting respectful, consultative, and meaningful health research with Indigenous communities has been especially important when consulting around sensitive and less often spoken about topics such as stillbirth. Birthing on Country and continuity of midwifery care are key recommendations for community-based solutions to Closing the Gap in birthing and health inequalities that exist for Indigenous peoples (32, 35, 37). Reducing the stillbirth rate and improving the general health and wellbeing of Indigenous women is a key principle of Birthing on Country approaches (4, 32). Our work in this space, as well as the work of others in other health areas, highlights the importance of understanding these experiences from the perspective of the Indigenous communities to ensure culturally appropriate conversations about prevention strategies. Indigenous pregnant women who live in rural or remote areas may need to be transferred to the nearest regional or urban hospital for delivery, away from their family and community. This results in women being away from their connection to their cultural lands and Country, as well as being transported while delivering a Sorry Business baby into an unfamiliar health care team and system, that may not understand or recognize the importance of cultural practices during this time of grief and loss. For many Indigenous women and their families, delays, or prevention from undertaking these practices fundamentally escalates the stress of this event, and gentle supportive healthcare could help to alleviate this.

Our trustful, respectful, and thorough consultation process allowed for identification of contentious issues, gaps in awareness and education, and research priorities. Across all topics discussed during the national consultation undertaken, lack of continuity of care was identified as the main barrier in effective stillbirth prevention, investigation of stillbirth causes, and care for families following stillbirth. The importance of ensuring midwives and service providers have the education to provide continuous care for a family was noted as the main goal of caring for Indigenous families during the perinatal period. Recommendations focused on education and training for health care workers to support and empower pregnant women and their families. The yarning sessions undertaken also served to explore the feasibility and acceptability of education, training, and the development of culturally appropriate resources aimed to support Indigenous communities. Guidance for those undertaking future research based on our study findings includes the critical need to co-design evidence-based, culturally appropriate, and community-acceptable resources to help reduce existing disparities in stillbirth rates in Indigenous communities.

Lessons learned in conducting this research

Insights from this work can be translated to other settings include the need for: Indigenous leadership in the research process; flexibility and realistic time frames when working with Indigenous communities to support cultural protocols and priorities. During this project the COVID-19 pandemic period provided an extreme case where the team needed to manage several challenging conditions to support health and safety of Indigenous communities. Further learning's included understanding of diversity of Indigenous communities, understanding the distance and requirements of travel out to communities around the country. In our efforts to ensure face-to-face communications it became the project responsibility to undertake the travel to remote communities and gave the team additional insights into the challenges communities face to manage when there is a need for birthing elsewhere. A recommendation for researchers to attend more community yarning sessions to meet with women's groups, men's groups and visit more rural and remote communities was suggested. Inclusion of more men in the research process would have been of benefit, and having an Indigenous male to lead this, as well as having visible male representation in the development of future resources was also recommended.

Challenges

Although we have captured a series of consultations during this project, we did encounter a few challenges, the biggest being the COVID-19 pandemic and ensuing travel/access restrictions. Indigenous women, families, and communities already faced disadvantages in inequitable access to health care, and the COVID-19 pandemic exacerbated this in many instances. Although not relevant to our study, the pandemic further decreased access to health care in remote regions due to limitations on the ability to enter specific regions across the country and to visit communities during lockdown periods, including for researchers.

A particular challenge faced by the research team was the occasional pressures placed by the funder's deliverable timelines, and their limited understanding of culturally appropriate consultation processes. As is often the case, funding bodies have an expectation that research projects will deliver to a timeline of deliverables and apply significant pressure for this to occur. To enhance this understanding, a workshop was conducted by Stillbirth CRE Indigenous researchers and funding body representatives in September 2022. At times, travel restrictions from COVID-19 pandemic period changed extremely quickly and meant cancellations and not being able to reach more communities as planned. Unfortunately, there was little sympathy for this and continued to add to the burden of stress that this work entailed. The research team's commitment to follow Indigenous ways of knowing, being, and doing became a challenge in the circumstances, as it meant that significant education had to be provided to the funding body to ensure a culturally appropriate process continued.

During the pandemic period, there were occasional difficulties in securing health service staff to attend consultations due to prioritizing health care delivery for COVID-19 care. Similarly, it was at times challenging to have Indigenous community members participate due to the sensitive nature of the topics to be discussed. Despite these challenges, we were able to capture several viewpoints of women and family members with lived experience or awareness of others' experience of Sorry Business Babies, and health professionals who cared for women during pregnancy. To counteract the potential distress caused by the topic stillbirth, we included a follow up of women interviewed as needed and offered support by Social and Emotional Wellbeing Teams and/or by the Employee Assistance Program.

Opportunities

To overcome these challenges due to the COVID 19 restrictions we engaged with some community participants virtually. However, many Indigenous communities' encountered difficulties in access to equipment and internet connection. Further, there was some reluctance to use it, as for many people it was their first experience of these virtual meeting spaces. Virtual consultations were not as effective as face to face as it challenged the idea of a natural and supportive environment and restricted the flow of Indigenous ways of yarning especially when disclosing something as personal as a Sorry Business Baby.

From our consultations we recognized a gap in information available for Indigenous women who continue to have disproportionate burden of stillbirth in Australia and have needs requiring careful consideration in care after stillbirth. This project set out to explore experiences of stillbirth from the perspectives of Indigenous women and their communities, through discussions with community members and health professionals of Far North Queensland, South Australia, Victoria, and Western Australia. The Stillbirth CRE Indigenous Research Team worked with community level permission and direction, using yarning methods in data collection, and ensured data sovereignty through community ownership, and community benefit.

Conclusion

Our findings outlined the five key areas of focus to ensure stillbirth prevention and care of Indigenous peoples is culturally safe and responsive. Our study included a detailed consideration of Indigenous peoples' experiences of stillbirth and awareness of stillbirth risks, which to our knowledge, was the first time this has been undertaken with Australian Indigenous populations at scale. The findings provide a foundation of understanding for the development of culturally appropriate awareness and prevention resources and will support efforts to adapt or create existing resources. Consultation findings helped to inform and guide the co-design of culturally responsive resources for pregnant women to reduce stillbirth rates, and support families after the loss of their baby, and are an important step in addressing stillbirth prevention and care (42). Areas of future development include continued work to ensure Indigenous parents and community members have their voices, experiences and needs around Sorry Business babies heard at all levels within the policy-setting process through enabling conversations, and the development and dissemination of culturally appropriate resources, including in bereavement care. This research aims to contribute to the evidence-base in working toward optimal care of Indigenous families.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

This study was approved by the following Aboriginal-specific human research ethics committees in South Australia, and Western Australia, and included one with an Aboriginal review committee in Queensland: South Australian Health and Medical Research Institute, and South Australian Aboriginal Health Accord (***) (South Australia); Far North Queensland Human Research Ethics Committee (***); Western Australian Aboriginal Health Ethics Committee (HREC1073); Mater Misericordiae Ltd Human Research Ethics Committee (HREC/MML/****) and ratified by one university Human Research Ethics Committees (***). Due to the sensitive nature of this topic, and to minimize harm during interviews, a risk and safety strategy for community members and staff was put into place, guided by social and emotional wellbeing counselors of participating health services.

Author contributions

LM: Data curation, Formal analysis, Methodology, Project administration, Software, Writing—original draft, Writing—review & editing. CL: Data curation, Formal analysis, Investigation, Methodology, Validation, Writing—original draft, Writing—review & editing. SS: Data curation, Formal analysis, Investigation, Methodology, Validation, Writing—original draft, Writing—review & editing. DJ: Data curation, Formal analysis, Investigation, Validation, Writing—original draft, Writing—review & editing. RG: Data curation, Formal analysis, Methodology, Project administration, Software, Supervision, Writing—original draft, Writing—review & editing. LJ: Data curation, Formal analysis, Project administration, Software, Writing—original draft, Writing—review & editing. AB: Data curation, Formal analysis, Investigation, Software, Validation, Writing—original draft, Writing—review & editing. PM: Conceptualization, Data curation, Formal analysis, Methodology, Resources, Supervision, Validation, Visualization, Writing—original draft, Writing—review & editing. SV: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Supervision, Validation, Visualization, Writing—original draft, Writing—review & editing. FB: Conceptualization, Data curation, Formal analysis, Methodology, Validation, Visualization, Writing—original draft, Writing—review & editing. CS: Data curation, Formal analysis, Methodology, Resources, Supervision, Writing—original draft, Writing—review & editing. VF: Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Resources, Supervision, Validation, Visualization, Writing—original draft, Writing—review & editing. DS-B: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Supervision, Validation, Visualization, Writing—original draft, Writing—review & editing. KR: Data curation, Formal analysis, Methodology, Resources, Supervision, Visualization, Writing—original draft, Writing—review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2024.1385125/full#supplementary-material

References

1. Davey M-A, Wallace EM. Reducing stillbirth safely in Australia. *BJOG.* (2019) 127:581–9. doi: 10.1111/1471-0528.16038

2. Australian Institute of Health and Welfare. *Australia's Mothers and Babies.* Canberra: AIHW (2023).

3. Australian Institute for Health and Welfare. *Stillbirths and Neonatal Deaths in Australia 2015 and 2016: in Brief. Perinatal Statistic Series.* Canberra: AIHW (2019).

4. Commonwealth of Australia. Select committee on stillbirth research and education report. In: Senate DOT, editor. *Senate Printing Unit, Parliament House.* Canberra, ACT: Commonwealth of Australia (2018).

5. Flenady VJ, Middleton P, Wallace EM, Morris J, Gordon A, Boyle FM, et al. Stillbirth in Australia 1: the road to now: two decades of stillbirth research and advocacy in Australia. *Women Birth.* (2020) 33:506–13. doi: 10.1016/j.wombi.2020.09.005

6. Australian Bureau of Statistics A. *Estimates of Aboriginal and Torres Strait Islander Australians.* Canberra, ACT: Australian Bureau of Statistics A (2021).

7. Rumbold AR, Yelland J, Stuart-Butler D, Forbes M, Due C, Boyle FM, et al. Stillbirth in Australia 3: addressing stillbirth inequities in Australia: steps towards a better future. *Women Birth.* (2020) 33:520–5. doi: 10.1016/j.wombi.2020.08.012

8. Australian Institute of Health and Welfare. *Aboriginal and Torres Strait Islander Mothers and Babies.* Canberra: AIHW (2023).

9. Ibiebele I, Coory M, Boyle F, Humphrey M, Vlack S, Flenady V. Stillbirth rates among Indigenous and non-Indigenous women in Queensland, Australia: is the gap closing? *Int J Obstetr Gynaecol.* (2015) 122:1476–83. doi: 10.1111/1471-0528.13047

10. Callander E, Fox H, Mills K, Stuart-Butler D, Middleton P, Ellwood D, et al. Inequitable use of health services for Indigenous mothers who experience stillbirth in Australia. *Birth.* (2022) 49:194–201. doi: 10.1111/birt.12593

11. Homer CS. Addressing stillbirth in Australia: a national initiative. *Women Birth.* (2020) 33:505. doi: 10.1016/j.wombi.2020.09.011

12. Stillbirth Foundation Australia. *Stillbirth Foundation Australia.* Sydney, NSW (2024). Available online at: https://stillbirthfoundation.org.au/ (accessed January 30, 2024).

13. Red Nose. *Stillbirth Prevention.* Melbourne, VIC: Red Nose Australia (2024). Available online at: https://rednose.org.au/section/stillbirth-prevention (accessed 8 February 2024).

14. Still Aware. *Still Aware.* Adelaide, SA (2024). Available online at: https://stillaware.org/about-us/ (accessed January 30, 2024).

15. Andrews CJ, Ellwood DA, Gordon A, Middleton P, Homer CS, Wallace EM, et al. Stillbirth in Australia 2: working together to reduce stillbirth in Australia: the safer baby bundle initiative. *Women Birth.* (2020) 33:514–9. doi: 10.1016/j.wombi.2020.09.006

16. Commonwealth of Australia. *National Stillbirth Action and Implementation Plan.* Canberra, ACT: Health, D. O (2020).

17. Gordon A, Chan L, Andrews C, Ludski K, Mead J, Brezler L, et al. Stillbirth in Australia 4: breaking the silence: amplifying public awareness of stillbirth in Australia. *Women Birth.* (2020) 33:526–30. doi: 10.1016/j.wombi.2020.09.010

18. Andrews C, Ellwood D, Middleton P, Gordon A, Nicholl M, Homer C, et al. Implementation and evaluation of a quality improvement initiative to reduce late gestation stillbirths in Australia: Safer Baby Bundle study protocol. *BMC Pregn Childb.* (2020) 20:1–14. doi: 10.1186/s12884-020-03401-0

19. Chan L, Owen KB, Andrews CJ, Bauman A, Brezler L, Ludski K, et al. Evaluating the reach and impact of Still Six Lives: a national stillbirth public awareness campaign in Australia. *Women Birth.* (2023) 36:446–53. doi: 10.1016/j.wombi.2023.02.006

20. Pollock D, Shepherd CCJ, Adane AA, Foord C, Farrant BM. Knowing your audience: investigating stillbirth knowledge and perceptions in the general population to inform future public health campaigns. *Women Birth.* (2022) 35:e389–96. doi: 10.1016/j.wombi.2021.06.008

21. Martin K, Mirraboopa B. Ways of knowing, being and doing: a theoretical framework and methods for indigenous and indigenist re-search. *J Austral Stud.* (2003) 27:203–14. doi: 10.1080/14443050309387838

22. Baum F, Macdougall C, Smith D. Participatory action research. *J Epidemiol Community Health.* (2006) 60:854–7. doi: 10.1136/jech.2004.028662

23. Sharmil H, Kelly J, Bowden M, Galletly C, Cairney I, Wilson C, et al. Participatory Action Research-Dadirri-Ganma, using Yarning: methodology co-design with Aboriginal community members. *Int J Equity Health.* (2021) 20:160. doi: 10.1186/s12939-021-01493-4

24. Walker M, Fredericks B, Mills K, Anderson D. "Yarning" as a method for community-based health research with indigenous women: the indigenous women's wellness research program. *Health Care Women Int.* (2014) 35:1216–26. doi: 10.1080/07399332.2013.815754

25. Fredericks B, Adams K, Finlay S, Fletcher G, Andy S, Briggs L, et al. Engaging the practice of Indigenous yarning in action research. *Act Learn Act Res J.* (2011) 17:12–24. Available online at: https://search.informit.org/doi/10.3316/informit.463112109856313

26. Kennedy M, Maddox R, Booth K, Maidment S, Chamberlain C, Bessarab D. Decolonising qualitative research with respectful, reciprocal, and responsible research practice: a narrative review of the application of Yarning method in qualitative Aboriginal and Torres Strait Islander health research. *Int J Equity Health.* (2022) 21:134. doi: 10.1186/s12939-022-01738-w

27. Bessarab D, Ng'Andu B. Yarning about yarning as a legitimate method in Indigenous research. *Int J Crit Indig Stud.* (2010) 3:37–50. doi: 10.5204/ijcis.v3i1.57

28. Barlo S, Boyd WE, Pelizzon A, Wilson S. Yarning as protected space: principles and protocols. *AlterNative Int J Indig Peopl.* (2020) 16:90–8. doi: 10.1177/1177180120917480

29. Gale NK, Heath G, Cameron E, Rashid S, Redwood S. Using the framework method for the analysis of qualitative data in multi-disciplinary health research. *BMC Med Res Methodol.* (2013) 13:117. doi: 10.1186/1471-2288-13-117

30. Australian Indigenous Health Infonet. *Cultural Ways.* Perth, WA: Edith Cowan University (2023). Available online at: https://healthinfonet.ecu.edu.au/learn/cultural-ways/cultural-practices/ (accessed December 11, 2023).

31. Carlson B, Frazer R. "It's like going to a cemetery and lighting a candle": aboriginal Australians, sorry business and social media. *AlterNative Int J Indig Peopl.* (2015) 11:211–24. doi: 10.1177/117718011501100301

32. Marriott R, Reibel T, Coffin J, Gliddon J, Griffin D, Robinson M, et al. "Our culture, how it is to be us"—listening to Aboriginal women about on Country urban birthing. *Women Birth.* (2019) 32:391–403. doi: 10.1016/j.wombi.2019.06.017

33. Massi L, Hickey S, Maidment SJ, Roe Y, Kildea S, Kruske S. "This has changed me to be a better mum": a qualitative study exploring how the Australian Nurse-Family Partnership Program contributes to the development of First

Nations women's self-efficacy. *Women Birth*. (2023) 5:10. doi: 10.1016/j.wombi.2023.05.010

34. Kemp L, Grace R, Comino E, Jackson Pulver L, Mcmahon C, Harris E, et al. The effectiveness of a sustained nurse home visiting intervention for Aboriginal infants compared with non-Aboriginal infants and with Aboriginal infants receiving usual child health care: a quasi-experimental trial-the Bulundidi Gudaga study. *BMC Health Serv Res*. (2018) 18:1–13. doi: 10.1186/s12913-018-3394-1

35. Kildea S, Gao Y, Hickey S, Nelson C, Kruske S, Carson A, et al. Effect of a Birthing on Country service redesign on maternal and neonatal health outcomes for First Nations Australians: a prospective, non-randomised, interventional trial. *Lancet Glob Health*. (2021) 9:e651–9. doi: 10.1016/S2214-109X(21)00061-9

36. Molly Wardaguga Research Centre. *What is Birthing On Country?*. Darwin, NT: Charles Darwin University (2023). Available online at: https://www.birthingoncountry.com/ (accessed December 11, 2023).

37. Kildea S, Hickey S, Barclay L, Kruske S, Nelson C, Sherwood J, et al. Implementing birthing on country services for Aboriginal and Torres Strait Islander families: RISE framework. *Women Birth*. (2019) 32:466–75. doi: 10.1016/j.wombi.2019.06.013

38. Bryant J, Bolt R, Botfield JR, Martin K, Doyle M, Murphy D, et al. Beyond deficit: 'strengths-based approaches' in Indigenous health research. *Sociol Health Illn*. (2021) 43:1405–21. doi: 10.1111/1467-9566.13311

39. Bowes J, Grace R. Review of early childhood parenting, education and health intervention programs for Indigenous children and families in Australia. In: Studies AIOHAWAAIOF, editor. Canberra, ACT: Australian Institute of Health and Welfare (2014).

40. Lohoar S, Butera N, Kennedy E. *Strengths of Australian Aboriginal Cultural Practices in Family Life and Child Rearing*. Melbourne, VIC: Australian Institute of Family Studies (2014).

41. Hancock F, Epston D, Mckenzie W. Forging Treaty Hope: the application and relevance of narrative ideas and practices in developing Treaty-based policy and practice. *Community Dev J*. (2006) 41:453–66. doi: 10.1093/cdj/bsl027

42. Stilbirth Centre of Research Excellence. *Stronger Bubba Born—Looking After Bubba for All Our Mob*. Meanjin (Brisbane), QLD (2023). Available online at: https://strongerbubbaborn.org.au/ (accessed November 30, 2023).

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How to decrease teenage pregnancy: rural perspectives in Ecuador

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Introduction: This study aimed to understand the sociocultural context of teenage pregnancy in an Ecuadorian city with a large indigenous population, to gauge the acceptability of a multifaceted pregnancy prevention program for adolescents, and to elicit perspectives on the optimal program design from adolescents and adult key informants.

Methods: We ascertained qualitative data via an online, electronic survey administered from August to September 2020. Open- and closed-ended questions elicited perspectives relating to burden of adolescent pregnancies, acceptability of pregnancy prevention programs, and optimal design of future programs. Twenty-four adolescents (13–19 years of age) and 15 adult key informants working in the healthcare, business, and education sectors in Cotacachi completed the survey. Survey responses were analyzed using a structural and *in vivo* coding, and an inductive approach to consensus-building around key themes.

Results: Most adolescent survey respondents (75%) believed that teen pregnancy is "fairly common" in Cotacachi, and 41.7% believed differences in teen pregnancy rates are not associated with ethnicity. In comparison, 66.7% of adult survey respondents said teen pregnancy disproportionately occurs among indigenous teenagers. Additionally, 45.8% of adolescent and 80% of adult survey respondents believed that a comprehensive sexual education program would help reduce teenage pregnancy rates by imparting reliable sexual health knowledge. Adult respondents noted that the past programs were unsuccessful in preventing teenage pregnancy because of these programs' inability to fully engage teenagers' attention, very short time duration, or inappropriate consideration of cultural context.

Discussion: In Cotacachi, Ecuador, a sexual health education program is both desired and feasible according to adult and teenager key informants. A successful program must adapt to the cultural context and engage youth participation and attention.

KEYWORDS

teenage pregnancy, Ecuador, rural population, indigenous population, community-based participatory research, evidence-informed intervention

1 Introduction

Sexual and reproductive health "encompasses dimensions of physical, emotional, mental and social well-being in relation to sexuality" and it is not just the absence of disease (1). The lack of proper sexual and reproductive health services and education can contribute to the high incidence of teenage pregnancy in several countries (1–3). Adolescent mothers and their babies face very high risks for their health. In comparison to women 20–24 years old, adolescents have a higher risk of complications during pregnancy and childbirth such as eclampsia, postpartum endometritis, systemic infections (3) and ultimately, maternal mortality (4). Compared to neonates born to adult mothers, neonates born to adolescent mothers are more likely to experience low birth weight and other health complications derived from preterm delivery (3).

Latin America and the Caribbean (LAC) is one of the regions with the highest adolescent pregnancy rates in the world: there are 59 pregnancies for every 1,000 teenage women (15–19 years) (5). In comparison, Western Europe, Eastern Europe and Central Asia and North America have 8, 19 and 14 births per 1,000 adolescent girls and young women aged 15–19, respectively (6). In Ecuador, there are 64 pregnancies per every 1,000 adolescent women (5). Ecuador's indigenous population represent 7.7% of the total country's population (16′ 938.986) (7) and have historically been more vulnerable to adolescent pregnancy (8, 9). This vulnerability was exacerbated in 2014 when new restrictions to the Inter-Sectoral Strategy for Family Planning and Prevention of Adolescent Pregnancy program restricted reproductive health services for adolescent women, leading to increased birth rates by 8.5 births per 1,000 women in cantons with higher indigenous concentration (2).

Adolescent pregnancy also has severe socioeconomic effects. In Ecuador, the cost of caring for unintended pregnancy is over 5 times the cost of preventing it (10). A study among 6,487 young females (ages 15–19) in Ecuador estimated that school dropouts due to adolescent pregnancy represented a loss of $317 million in future income (10). Despite the health and economic impacts of unplanned adolescent pregnancies, funding for sexual and reproductive adolescent health is constantly threatened; for instance, the country's teenage pregnancy prevention funds diminished from $5 USD million in 2019 to $0 USD in 2020 (11).

Despite evidence-based recommendations to include comprehensive sexuality education (CSE) as part of education curriculums several years ago (12), Ecuador is still working to implement it into the education system. In fact, the Ministry of Education approved in October 2023, the "National Strategy for Comprehensive Sexuality Education," which will be implemented progressively in the education system until 2023 (13). The document is not publicly accessible; therefore, its content is still unknown. Previously to this announcement, educational authorities claimed that CSE it's part of the education system following a "transversal" strategy (14), whereby every subject (math, science, physical education, etc) at some point addresses a sexual education topic. However, Ecuadorian students from both private and public schools reported that their schools did not offer any continual sexual health education and that they lacked knowledgeable and reliable sources of accurate sexual health information (15).

Comprehensive sexual education works best to decrease adolescent pregnancy when combined with economic empowerment (16). Economic empowerment increases decision-making power in relationships, and access to contraceptives (16). For example, a multifaceted program in Uganda that offered vocational training alongside sexual health education found that, 4 years post-intervention, adolescents receiving the program were 4.9 percentage points more likely to engage in income generating activities, and with sharp declines showed decreases in adolescent pregnancy, marriage, cohabitation, and rape (17). Also, teenagers' beliefs regarding ideal ages at which to marry and bear children adjusted to an older age (17). A study of the Haiti Adolescent Girls' Initiative found that providing technical training with socio-emotional and soft-skills development increased empowerment on key socioeconomic dimensions, autonomy in personal mobility, and self-assertion in the intimate circle (18).

Despite these promising results, there are also contradictory reports in the literature. For instance, the 2009 Liberian Economic Empowerment of Adolescent Girls and Young Women (EPAG) program provided technical, cognitive, and emotional skills training, which included education on sexual and reproductive health. Six months after this intervention, researchers found that there were no changes in sexual behaviors among participants. Researchers claimed that sexual behavior was not an objective of the program, therefore these results are explained due to the lack of specificity and longer-term of the intervention (19). This suggests that the efficacy and optimal design of multifaceted adolescent pregnancy prevention programs merits further investigation.

To our knowledge, there has not been a multifaceted teenage pregnancy intervention with indigenous population. Due to the social complexity, interventions like the multifaceted programs in Uganda and Haiti have the potential to work in LAC countries if properly adapted. Public health interventions should never be imposed upon a population (20). Instead, they should be culturally adapted to the needs and desires of the specific population the intervention is meant to benefit (20). Without proper cultural adaptation, the intervention may also prove wholly ineffective when applied to the context of another community.

The objective of this study is to understand the sociocultural context of teen pregnancy in a small city in Ecuador (Cotacachi) which has a high indigenous population. This study also aims to gauge the acceptability of a multifaceted teenage pregnancy prevention program and how one may be best designed according to teen and adult key informants within the community.

2 Methods

2.1 Study setting

Participant enrollment and data collection occurred in Cotacachi, Ecuador. Cotacachi is a city of approximately 53,001 inhabitants located in the north of the country (7). The city's population is 80.1% rural and 41.7% of indigenous descent (7). In 2020, Cotacachi's public health system reported 210 pregnancies among adolescents between 10 and 19 years of age (21).

2.2 Study population

The study population included adult key informants working in Cotacachi and adolescent males and females living in Cotacachi.

Adults were recruited using a convenience sampling approach, selecting adults with experience working in the healthcare and education sectors who had experience working directly with adolescents or on topics of adolescent sexual health. We also recruited adults from the business sector with experience related to youth economic empowerment. We recruited a total of $n = 15$ adults to complete the survey, 5 from each of the three sectors. We additionally recruited $n = 24$ adolescents, ages 13–19, to complete the survey. The sample size was determined by data saturation. We consider data saturation when we identified redundancy in the data (22). Researchers were constantly monitoring the data collected and found data saturation with those participants.

Sexual health education is still taboo in Ecuador, especially in rural communities. Therefore, heterogeneous responses were appropriate to answer our research question, since the hypothetical program needs to be acceptable to youth, and also feasible and sustainable for adults to implement. This practice also agrees with data source triangulation, since it "involves the collection of data from different types of people, including individuals, groups, families, and communities, to gain multiple perspectives and validation of data." (23).

The local investigator who resides in Cotacachi and worked at the Cotacachi Public Health Center contacted participants (adults and teenagers) via phone call or text message from contacts provided by people in the community. Eligible adults employed within the healthcare sector were recruited in person. All individuals provided voluntary online informed consent/assent for study participation, which was approved by the IRB. The institution granted an exempt condition to this study because it was a "Research with data collection in an anonymized and digital manner, without human contact" All individuals were required to have a Level-1 reading ability (corresponding to completion of the seventh year of basic education in the Ecuadorian educational system).

2.3 Survey design

We decided to conduct this study through an online survey due to the pandemic conditions. Despite this method is underutilize, it has been described that "qualitative surveys may be more appropriate when: they are the best 'fit' for participants' needs (e.g., for very sensitive topics); a population is dispersed, hard to engage or access and/or diverse; a wide range of perspectives or positionings is sought […]" and that "*qualitative* survey datasets can provide richness and depth, when viewed in their entirety, even if individual responses might themselves be brief." (24). We developed two surveys based on successful multifaceted programs components and the target audience: adolescents and adult key informants. Surveys were designed by the investigative team and reviewed by native Spanish speakers from the community to ensure cultural accuracy. Both surveys ascertained demographic information followed by 18 multiple-choice and open-response questions that asked about views on adolescent pregnancy and previous adolescent pregnancy prevention programs, the acceptability of a theoretical multifaceted teenage pregnancy prevention program, and suggestions for program design components. The adult survey additionally asked whether the respondent had adolescent children, and their place of residence and employment sector. All surveys were developed in Spanish and programmed using Google Forms. Each survey took between 20 and 30 min to complete.

2.4 Data collection and analysis

Following the Handbook of mixed methods in social behavioral research, our design uses qualitative and quantitative approaches in the types of questions, data analysis procedures, and inferences (25), and thus is considered mixed-methods research.

Recruitment and data collection occurred from August 17th to September 18th, 2020. At enrollment and immediately after providing informed consent, participants received the questionnaire via email or text message. Interviewees completed the survey in Google Forms on their personal computer or Smartphone. All responses were recorded anonymously, and participants could skip any question they did not want to answer.

The data collected in the Google form platform was downloaded by the local investigator and stripped of identifiers for analyses. First, the data were classified into two groups: adult key informants and adolescents. Second, the frequency (%) was obtained for all socio-demographic characteristics, dichotomous (yes/no) questions, and multiple-choice questions. Last, the research team analyzed answers to open questions using structural coding and *In vivo* coding, which uses participants' own words to create an analytic coding scheme (26). Analysis was made in Microsoft Excel software (27). One of the investigators synthesized the major themes from each open question using an inductive approach. Meanings from these codes were used to produce clusters of themes, which were then discussed and agreed upon by the coding team over 5 iterations of reviews to create consensus (28). The research protocol was reviewed and approved by the bioethics committee of the Universidad San Francisco de Quito (Comité de Ética de Investigación en Seres Humanos, Study Code: 2020-036 M).

3 Results

3.1 Participants description

A total of $n = 24$ teenagers (13–19 years old) participated (Table 1). All of them lived in Cotacachi; three of them reported having a child at the time of the study. A total of $n = 15$ adult key informants (20–60 years old) participated in the study, five per each sector: education, healthcare, and commerce. Twelve of them lived in Cotacachi; nine of them reported having one or more children at the time of the study.

We wanted to understand the perceptions than teenagers and adults have regarding pregnancy during adolescence, and about prevention programs. Therefore, we divided the results in two main sections to helps us (1) understand the sociocultural context of teenage pregnancy in a city with high indigenous populations; and (2) gauge the acceptability of a multifaceted teenage pregnancy prevention program. By assessing this, we also have information about what are teenagers and adult key informants' expectations about an intervention that seeks to prevent pregnancies during adolescence, so we can

TABLE 1 Demographics of study participants.

Demographic categories	Teenagers *n* = 24		Key informants *n* = 15	
	N°	% of sample	N°	% of sample
Age (years)	13–15 y/o = 10 16–17 y/o = 8 18–19 y/o = 6	41.7% 33.3% 25%	20–40 y/o = 8 41–60 y/o = 7	53.3% 46.7%
Sex				
Female	11	45.8%	9	60%
Male	13	54.2%	6	40%
Ethnicity (Self-identified)				
Indigenous	7	29.2%	3	20%
Mestizo	16	66.7%	11	73.3%
White	1	4.2%	1	6.7%
Education				
Middle school	7	29.1%	0	0%
High school	17	70.8%	1	6.7%
Professional degree	0	0%	14	93.3%
Marital status				
Single	19	79.2%	5	20.8%
In a dating relationship	3	12.5%	0	0%
Married	0	0%	7	29.2%
Common law	2	8.3%	1	4.2%
Divorced	0	0%	2	8.3%
Internet and technology access				
Internet at home and access to a computer	18	75%	15	100%
Internet access with a smartphone only	3	12.5%		
Internet at home and owns a tablet	2	8.3%		
No internet at home, but owns a tablet	1	4.2%		

Common law is a status in Ecuador when a couple live together and have children but are not legally married.

understand how one may be best designed in the sociocultural context. We contrast adult key informants' interests with teenagers' points of view. A summary of the key points that emerged from this study to design a teenage pregnancy prevention program in an Ecuadorian indigenous and rural population is provided at the end of this section (Figure 1).

3.2 Sociocultural context of teenage pregnancy

We explored teenage and adult key informants' perceptions about teenage pregnancy. Qualitative results are described in Table 2. We present the most relevant findings in this section. Quantitative findings are described to provide as much detailed description about the study population and their preferences as possible.

3.2.1 Do you think adolescent pregnancy is common in Cotacachi? Why or why not?

Out of the 24 adolescents that participated, 20.8% choose the option "Yes, it's very common," 75% choose "Fairly common" and 4.2% choose "No, it's not common."

Table 2 summarizes the themes that emerged. *Lack of comprehensive sexual health education* was the most mentioned reason by teenagers that believe that adolescent pregnancy was very common or fairly common. One participant noted, for example:

> *"There is no accurate sexual health education, and we lack support and understanding from our parents when it comes to having a romantic partner."*
> Participant #19

Other reasons that the majority of adolescents claimed were *Little parental/family support* and *Lack of use of contraceptive methods.*

> *"Lack of information and communication with parents, not using contraceptive methods or misusing them."*
> Participant #17
>
> "Lack of trust with parents to talk about sexuality."
> Participant #7

Out of the 15 adult key informants, 66.7% choose the option "Yes, it's very common" and 33.3% choose "Fairly common." Similarly to adolescents, this group also identified lack of comprehensive sexual health education as the main cause of adolescent pregnancy in Cotacachi. They also agreed with teenagers regarding how, irresponsibility, family or sexual violence, and lack of access to

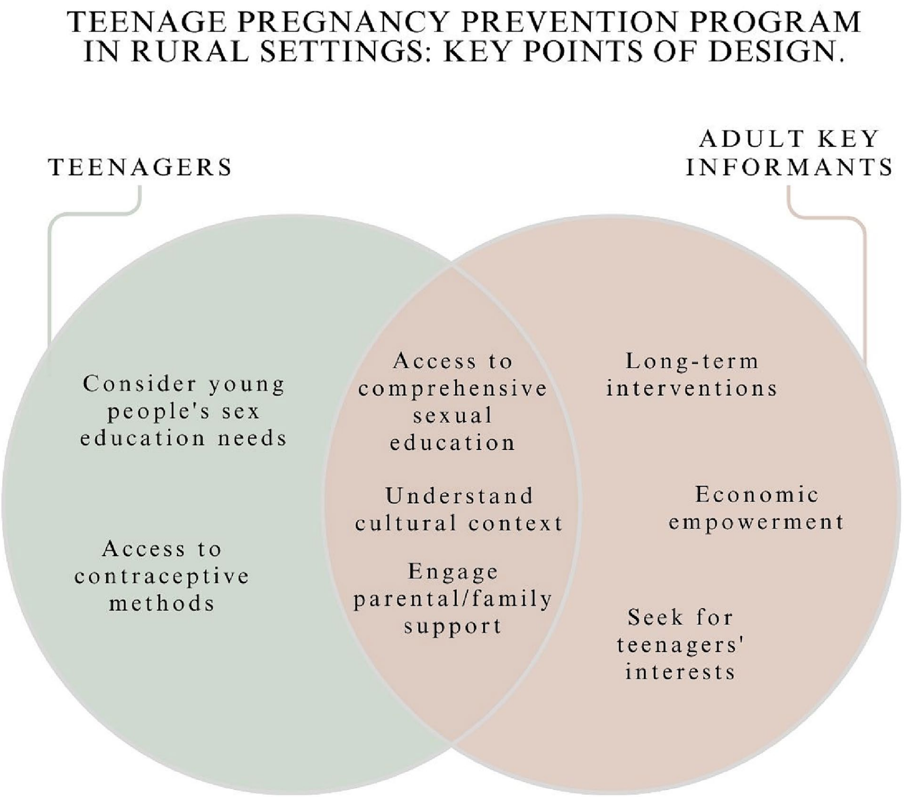

FIGURE 1
Key points to design a teenage pregnancy prevention program in a rural setting.

contraception perpetuates teenage pregnancy. However, the reasons more mentioned were little parental/family support, sexuality still treated as taboo and, culture and traditions.

Most adult respondents gave multiple answers. For example, one participant stated:

> *"[Teenage pregnancy is the result of] lacking access to preventative health information, sexual education by the educational system and in the family environment is deficient. Unfortunately, sexuality is still seen as a taboo; dogmas prevail in many cases."*
>
> Participant #4

3.2.2 Is Any racial/ethnic group more vulnerable to teenage pregnancy? If yes, which one and why?

A third of adolescents (33.3%) think that indigenous are more vulnerable to get pregnant during adolescence. Out of those who answered, "indigenous people," only three gave a reason, and stated that this happens because of their culture (Table 2).

> *"The ethnic group that I think [is more vulnerable] is indigenous because there are many women and men who fall in love young and do not mind have children."*
>
> Participant #32

In contrast, 41.7% of adolescents think that there is not an ethnic group more vulnerable to teenage pregnancy. Similarly, only three of them gave a reason.

> *"No, I think that everyone is responsible for what they do, and we should not necessarily be of an ethnic group to be more likely to have a young pregnancy."*
>
> Participant #26

Additionally, 4.2% of adolescents stated that both indigenous and mestizos[1] are vulnerable to adolescent pregnancy; and 20.8% skipped this question.

Among adult key informants, 66.7% believed that indigenous ethnicity made teens in Cotacachi to be more vulnerable to early pregnancy. Only 3 could explain their reasoning (Table 2).

> *"I think that the groups vulnerable to teenage pregnancy [are] mainly impoverished groups, where access to education is not essential, but rather survival. With these characteristics, indigenous groups may be more vulnerable."*
>
> Participant #13

Furthermore, 20% of adult key informants stated that there is not an ethnic group more vulnerable to teenage pregnancy, but they could not provide detailed explanations; and 13.3% skipped this question.

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[1] Mestizo in Ecuador in defined as "The result of crossing white with Indian." Arteaga, M. D. Los hijos y las hijas de África y sus descendientes en Cuenca de las Indias durante el gobierno de los Habsburgo 2014;(65):296.

TABLE 2 Themes about perceptions on adolescent pregnancy and an adolescent pregnancy prevention program.

Participant's answer	Respondent(s)	Emergent theme(s)
Question: do you think adolescent pregnancy is common in Cotacachi? Why or why not?		
Adolescent pregnancy is very common or fairly common in Cotacachi	Adolescents	• Lack of comprehensive sexual health education • Little parental / family support • Lack of use of contraceptive methods • Sexual violence • Irresponsibility • Early initiation of sexual activity
	Adult key informants	• Lack of comprehensive sexual health education • Little parental / family support • Culture and traditions • Sexuality still treated as taboo • Religious influence • Irresponsibility • Lack of use of contraceptive methods • Family or sexual violence • Greater risk in rural areas • Immaturity • Family or sexual violence • Gender inequality • Low economic level • Early marriage
Question: Is any racial/ethnic group more vulnerable to adolescent pregnancy? If yes, which one and why?		
Indigenous are more vulnerable to pregnancy during adolescence	Adolescents	• Their culture is different. • Young parenting is normalized. • Cultural ideology
	Adult key informants	• Cultural customs • Poverty • Lack of access to education • Machismo
There is not an ethnic group more vulnerable to adolescent pregnancy	Adolescents	• Responsibility lies on each individual, not on their culture. • An ethnicity does not conditionate an adolescent pregnancy. • The risk of teenage pregnancy is the same across every ethnicity in Cotacachi.
	Adult key informants	• Adolescent pregnancy occurs in every ethnicity. • Ethnicity is not the only problem.
Question: Do you think that a program that combines economic empowerment with sexual health education could decrease the rate of adolescent pregnancy in Cotacachi? Please explain your answer.		
Yes	Adolescents	• There is a need to receive sexual education. • More education decreases vulnerability. • Information can raise awareness about adolescent pregnancy consequences. • Information can prevent from early initiation of sexual activity.
	Adult key informants	• Economic empowerment can increase interest to educate in other fields. • Economic empowerment and sexual education can lead to have other vision of life and life goals. • Economic empowerment leads to women empowerment and women's economic independence. • Education improves decision-making capacity. • This program could increase motivation to be an entrepreneurship.
Maybe	Adolescents	• Not everyone could participate in the program. • It depends on every individual interest. • Some people do not take it seriously. • It depends more on the local culture.
	Adult key informants	• It depends on every individual interest. • In most cases adolescents ignore these workshops.
No and I do not know	Adolescents	• Irresponsible conduct would not change. • It would increase the interest to experiment [sexual activities] • Results depend on target audience and investigators effort.

3.3 Multifaceted teenage pregnancy prevention program acceptability and design

3.3.1 Previous teenage pregnancy prevention programs

We explored if there have been any teen pregnancy prevention programs in Cotacachi. Results are in Table 3. Teenagers could not specify any type of prior program and have contradictory opinions on their effectiveness. One claimed that prior programs had reduced teenage pregnancy, while another said that it was completely ineffective. The rest did not provide more information. This trend was also true for the adult key informants' group. Out of the adults with knowledge about previous interventions, two said that the programs were part of the Ministry of Health, while the rest did not provide more details about the programs. Those who could recall why past interventions did not work gave the following reasons: "inability to engage the teenagers' attention, being too short in duration, or being inappropriate to the cultural context."

3.3.2 Multifaceted teenage pregnancy prevention program acceptability

To evaluate this topic, we made the following question: "Do you think that a program that combines economic empowerment with sexual health education could decrease the rate of teenage pregnancy in Cotacachi? Please explain your answer." We organized findings quantitatively (Table 3) and qualitatively (Table 2).

Teenagers that answered "Yes" considered that this kind of intervention could work because it would increase awareness and knowledge regarding teen pregnancy. Many teenage respondents voiced that comprehensive sexual education would decrease their vulnerability.

> *"Education is the basis of any society, and it is necessary for young people to be informed and know the reality of sexual education."*
> Participant #19
>
> *"Yes, since this way they could help us raise better awareness about the problem that occurs when getting pregnant at an early age."*
> *Participant #27*

Key informants added that a teen pregnancy prevention program could improve decision-making, further encourage entrepreneurship, and empower women, including the economic field (thereby promoting their financial independence and security).

> *"Yes, because an economic empowerment program can offer economic independence to women and therefore leads to less dependence on their partner. Furthermore, if sexual education courses are given in the workshops, women can empower themselves, increase their self-esteem and believe in themselves."*
> *Participant #14*

Teens and adult key informants who answered "maybe" argue that the intervention's effectiveness would depend on people's interests. For example, one of the teenagers said:

TABLE 3 Perceptions on prevention programs.

Has there been any teen pregnancy prevention programs in Cotacachi?				
	Yes	No	I do not know	No response given
Teenagers	20.8%	29.2%	50%	0%
Adults key informants	33.3%	66.7%	0%	0%
Do you think that a program that combines economic empowerment with sexual health education could decrease the rate of teenage pregnancy in Cotacachi?				
	Yes	No	Maybe	I do not know
Teenagers	45.8%	8.3%	37.5%	8.3%
Adults key informants	80%	0%	20%	0%

> *"Maybe [the program would work], because some young people might listen to what they are taught but some may not."*
> Participant #31

Another adolescent claimed that a real change could not be achieved if the family and community does not change their ideology about this topic.

> *"Because it depends more on the culture, and information from the parents. In the [indigenous] communities from the age of 14 you are already an adult."*
> Participant #22
>
> *One participant, however, thought that the problem could be that "not all households would be able to attend [the economic empowerment workshops]."*
> Participant #7

Finally, only a few adolescents considered that this kind of program would not work or answered, "I do not know." One participant claimed that it is a shared responsibility. More themes can be found in Table 2.

> *"To be a good result, both researchers and the general population must take part."*
> Participant #20

3.3.3 Economic and personal empowerment interests

Participants reported their interest in three areas of economic empowerment: financial skills, life skills and vocational skills training (Table 4). On financial skills the question was "Economic empowerment is key within teen pregnancy prevention programs. The following are some relevant topics when starting a business and, in general, in financial education. Select the ones that you think teenagers between 13 and 19 years old will find useful and interesting (you can choose several options):"

Overall, we observed that teenagers do not have much interest in financial skills, for example 62.5% reported that they would like to learn

TABLE 4 Economic empowerment, life, and vocational skills topics of interest.

Topics	Teenagers ($n = 24$)	Key informants ($n = 15$)
	Financial	
Accounting skills	62.5%	60%
Ways to save money	62.5%	66.7%
Budget design	58.3%	80%
Investment and compound interest	50%	33.3%
Financial services in Ecuador	33.3%	40%
Debt management	33.3%	33.3%
Credit management	29.2%	46.7%
	Life skills	
Leadership	79.2%	73.3%
Conflicts resolution	66.7%	86.7%
Time management	62.5%	73.3%
Negotiation skills	58.3%	26.7%
	Vocational skills training	
Computing	83.3%	80%
Auto mechanics training	62.5%	60%
Farming	50%	73.3%
Crafts	50%	46.7%
Sewing	41.7%	53.3%
Carpentry	41.7%	40%
Hairdressing	37.5%	46.7%
Jewelry making	29.2%	33.3%

Teenagers and key informants' preferences in financial, life skills, and vocational topics.

about accounting and saving skills, and only 58.3% showed interest in learning budget design. Adult key informants selected the same topics, however, most of them choose budget design, in contrast with teenagers' preferences. We also ask for suggestions that were not included in our list. We identified that both adolescents and adults suggest that entrepreneurship should be included in the prevention program.

On life skills, the question was "What "life skills" tools would be important for the empowerment of an adolescent in Cotacachi? (you can choose several skills)." A list was provided. Leadership was the life skill most chosen by teenagers (79.2%), and conflicts resolution was most chosen by adult key informants (86.7%). Adolescents also suggested that the program should include training in communication skills, teamwork, responsibility, resilience and independence. In addition to this topics, adult key informants suggested: critical thinking, creativity/innovation, rights and obligations, self-esteem, life purposes, decision making, organization and perseverance.

Finally, on vocational skills training, computing was chosen as the skill in which young people from Cotacachi should be trained to generate income (83.3% of teenagers and 80% of adult key informants). The skills that adolescents suggested were gastronomy, art (music, painting, dance), driving, how to access to college, tourism, first aid, bakery, secretary, waiter, and marketing. Adult key informants suggested: clothing manufacturing, pottery, tourism, first aid/health, and leather crafting.

3.3.4 Sexual health education topics of interest

To explore their thoughts on this area, we made the following question: "Which themes do you think should be covered in sexual health education workshops for teenagers?"

Teenagers and adult key informants have different priorities in terms of sexual education for teenagers, those can be seen in Table 5. All adult key informants considered that life goals and family planning should be taught in sexual health education workshops. However, most teens (92%) selected the topic HIV/AIDS prevention, and only 50% of them chose life goals and family planning.

In addition to the topics proposed, adolescents suggested that the program should include training about how to properly take care of kids, relationships, harassment, sexual orientations, gender-based violence and masturbation. They also suggested that there should be workshops that include parents to "talk about sexuality with their children" Participant #7.

Adult key informants agreed with adolescents on the themes of childcare, and relationships, but they also suggested anatomy and physiology, how to act in a violence case, Human Rights, risks of teenage pregnancy, abortion, and mental health.

4 Discussion

We evaluated the perspectives of teenage and adult key informants from Cotacachi, a small city in northern Ecuador, about teenage pregnancy and their acceptability of a program that combines economic empowerment -through vocational and financial training- with sexual health education to prevent teenage pregnancy. Regarding the study subjects' perceptions of teen pregnancy incidence, most key informants (67%) pointed out that it is very common, meanwhile, most teenagers (75%) thought it was fairly common. These discrepancies (Table 2) in the perceived commonality of teen pregnancy between key informants and teenagers highlight the need for a nuanced understanding. Despite the discrepancies found, both groups concluded that it happens mainly due to lack of comprehensive sexual education and little parental or family support. This reaffirms the need to systematically implement comprehensive sexual education - an urgent need identified in similar teenage perspectives studies (29–31). From a race/ethnicity perspective, most teenagers did not associate higher pregnancy rates with a specific ethnic group. This contrasted with the adult key informants' view. Most of adult key informants explained that indigenous teenagers are more vulnerable to pregnancy due to poverty, lack of education, cultural customs, and machismo. Despite the teenagers' perception, teen pregnancy rates are higher among Ecuadorian indigenous youth. For instance, the frequency of teenage motherhood is 18.3% in indigenous teenagers versus 16.8% in non-indigenous teens between 15–19 years old (9). Additionally, indigenous teenagers in Bolivia, Guatemala, Ecuador, and Nicaragua have larger unmet needs for family planning than non-indigenous youth (8).

While exploring past programs' effectiveness, we found that nearly none of the adolescents were exposed to an intervention that seek to prevent teenage pregnancy (Table 3). The inadequacy of past programs, attributed to issues like short duration and cultural mismatch, highlights the necessity for programs to not only engage teenagers but also prioritize long-term strategies. We also found that there is an imperative need for

TABLE 5 Sexual education topics of interest.

Topics	Teenagers (*n* = 24)	Adult key informants *n* = (15)
HIV/AIDS prevention	91.7%	40%
Sexually transmitted infections	75%	93.3%
Consent and rape prevention	70.8%	66.7%
Use of contraceptive methods	66.7%	86.7%
How to obtain contraceptive methods?	62.5%	73.3%
Pregnancy	54.2%	80%
Life goals and family planning	50%	100%
Menstruation and related diseases	41.7%	46.7%
Child marriage and domestic violence	41.7%	60%

Family planning and contraceptive methods sometimes are used as interchangeable terms in Ecuador. But, family planning is the action to decide whether to have children or not; and when and how many children a person wants to have. In contrast, contraceptive methods are specific for a person who does not want to have children at that moment or never (depending on the method). We wanted to check if teenagers and adults key informants differentiate these terms.

culturally adapted interventions to tackle teen pregnancy effectively, particularly in highly indigenous settings.

By exploring the perspective of adult key informants and adolescents on a multifaceted program to prevent adolescent pregnancy, the study reveals a positive outlook from both teenagers and adult key informants regarding the potential success of a comprehensive teen pregnancy prevention program. Notably, teenagers emphasize the importance of reliable sexual health knowledge, while adult key informants stress the pivotal role of economic empowerment. Furthermore, it is interesting that adolescents would like their parents and family to be able to contribute to their training in sexual and reproductive health. This can be explained by the fact that the indigenous population is characterized by maintaining community life as one of the pillars of their cosmovision. The convergence of these perspectives emphasizes the holistic nature of successful interventions.

In terms of economic empowerment, we found that teenagers were less interested in financial education topics compared to adult key informants (Table 4). While little more than half of the teenagers chose accounting skills as the most important topic to learn in the financial area, more than three-quarters of adult key informants selected budget design as the most important topic. Future programs should take these differences into account when adapting an intervention directed to teenagers so that the programs would increase the chance of captivating teens' interests and involve them in the economic empowerment intervention.

Furthermore, we gathered key informants' and teenagers' interests in life and vocational skills topics. Both teens and adult key informants have the same top three life and vocational skill preferences: basic computer capabilities, auto mechanics training, and farming (Table 4). Their interests diverge from the courses on income-generating activities developed in other comprehensive teenage prevention programs (17). Understanding teenagers' preferences in this area is crucial to design highly accepted courses on income-generating activities.

Finally, we found major differences in sexual health education topics of interest between both groups. While most (92%) teenagers prioritized HIV/AIDS prevention to be part of reproductive health workshops, less than half (40%) of adult key informants selected this topic (Table 5). Meanwhile, all adult key informants agreed that life goals and family planning should be taught in sexual and reproductive health workshops aimed at teenagers, but just half of teenagers were interested in this topic. This finding reinforces the idea that sex education must consider young people's needs since they do indeed differ from those of adults (29).

The study's strengths lie in its inclusive representation of indigenous participants, offering unique insights into their perspectives on teen pregnancy prevention. The study's influence on the design of an ongoing intervention, involving teenagers from the outset, exemplifies a community-based participatory approach. However, limitations such as a small sample size and optional questions must be acknowledged, impacting generalizability. Therefore, results should be interpreted with caution. Furthermore, potential biases, including selective memory and social desirability, should be considered when interpreting results.

The study lays the groundwork for future research, emphasizing the need for larger, more diverse samples to enhance generalizability. The identified preferences and disparities among key informants and teenagers provide valuable guidance for tailoring future interventions. The ongoing digital intervention, with the involvement of a medical doctor from the indigenous population, exemplifies a promising approach to bridge the knowledge-action gap, ensuring cultural sensitivity in program implementation. Continued research is essential to ascertain the viability of teen pregnancy prevention programs in rural and indigenous settings, addressing the unique challenges and opportunities presented by these contexts.

Data availability statement

The original contributions presented in the study are included in the article/supplementary materials, further inquiries can be directed to the corresponding author.

Ethics statement

The studies involving humans were approved by Comité de Ética de Investigación en Seres Humanos (CEISH) de la Universidad San Francisco de Quito. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation in this study was provided by the participants' legal guardians/next of kin.

Author contributions

AT: Data curation, Formal analysis, Investigation, Methodology, Supervision, Writing – original draft, Writing – review & editing. KH: Conceptualization, Formal analysis, Investigation, Methodology, Writing – original draft, Writing – review & editing. OG: Conceptualization, Formal analysis, Investigation, Methodology, Supervision, Writing – review & editing. IP: Conceptualization, Formal analysis, Investigation, Methodology, Supervision, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

References

1. UNESCO, UNAIDS, UNFPA, UNICEF, UN Women, WHO. *International technical guidance on sexuality education*. Segunda: UNESCO (2018).

2. Galárraga O, Harris JE. Effect of an abrupt change in sexual and reproductive health policy on teen birth rates in Ecuador, 2008–2017. *Econom Hum Biol*. (2021) 41:100967. doi: 10.1016/j.ehb.2020.100967

3. World Health Organization (2023). Adolescent pregnancy. Available at: https://www.who.int/en/news-room/fact-sheets/detail/adolescent-pregnancy (accessed December 18, 2023)

4. Socolov DG, Iorga M, Carauleanu A, Ilea C, Blidaru I, Boiculese L, et al. Pregnancy during adolescence and associated risks: an 8-year hospital-based cohort study (2007–2014) in Romania, the country with the highest rate of teenage pregnancy in Europe. *Biomed Res Int*. (2017)

5. UNFPA. Estado de la población mundial 2022. Visibilizar lo invisible. (2022).

6. UNICEF UNICEF DATA. (2024). Early childbearing and teenage pregnancy rates by country. Available at: https://data.unicef.org/topic/child-health/adolescent-health/

7. Instituto Nacional de Estadísticas y Censos. Cifras clave del Censo Ecuador 2022. Instituto Nacional de Estadísticas y Censos; (2022). Available at: https://inec.censoecuador.gob.ec/public/Boletin_Segunda_Entrega_Trabajo.html

8. UNICEF. Health equity report 2016: analysis of reproductive, maternal, newborn, child and adolescent health inequities in Latin America and the Caribbean to inform policymaking. Panamá; (2016).

9. OPS, UNFPA, UNICEF. Acelerar el progreso hacia la reducción del embarazo en la adolescencia en América Latina y el Caribe. [Internet]. (2018).

10. Ministerio de Salud Pública del Ecuador, SENPLADES, UNFPA, SENDAS. *Estudio Costos de Omisión en Salud Sexual y Reproductiva en Ecuador*. Quito, Ecuador: Ministerio de Salud Pública del Ecuador, SENPLADES, UNFPA, SENDAS (2017).

11. Roa Chejín S. El comité para la prevención del embarazo adolescente, ¿otra oportunidad?. (2020).

12. UNESCO. Emerging evidence, lessons and practice in comprehensive sexuality education: a global review. (2015).

13. Ministerio de Educación. Unidos contra la violencia sexual en las aulas. (2024).

14. Arévalo GL Nadie puede defenderse de los monstruos que no conoce. GK (2019).

15. Reynolds C, Sutherland MA, Palacios I. Exploring the use of Technology for Sexual Health Risk-Reduction among Ecuadorean adolescents. *Ann Glob Health*. (2019) 85:57. doi: 10.5334/aogh.35

16. Nkhoma DE, Lin CP, Katengeza HL, Soko CJ, Estinfort W, Wang YC, et al. Girls' empowerment and adolescent pregnancy: a systematic review. *IJERPH*. (2020) 17:1664. doi: 10.3390/ijerph17051664

17. Bandiera O, Buehren N, Burgess R, Goldstein M, Gulesci S, Rasul I, et al. Women's empowerment in action: evidence from a randomized control trial in Africa. *Am Econ J Appl Econ*. (2020) 12:210–59. doi: 10.1257/app.20170416

18. World Bank Group. *Haiti adolescent girl initiative (AGI): Project report*. (2015)

19. Adoho F, Chakravarty S, Korkoyah DT, Lundberg M, Tasneem A. The Impact of an Adolescent Girls Employment Program: The EPAG Project in Liberia. (2014).

20. Movsisyan A, Arnold L, Evans R, Hallingberg B, Moore G, O'Cathain A, et al. Adapting evidence-informed complex population health interventions for new contexts: a systematic review of guidance. *Implement Sci*. (2019) 14:105. doi: 10.1186/s13012-019-0956-5

21. Torres C *Sala Situacional Distrito 10D03 Cotacachi: ENE-DIC 2020*. Ministerio de Salud Pública del Ecuador (2020).

22. Saunders B, Sim J, Kingstone T, Baker S, Waterfield J, Bartlam B, et al. Saturation in qualitative research: exploring its conceptualization and operationalization. *Qual Quant*. (2018) 52:1893–07. doi: 10.1007/s11135-017-0574-8

23. Carter N, Bryant-Lukosius D, DiCenso A, Blythe J, Neville AJ. The use of triangulation in qualitative research. *Oncol Nurs Forum*. (2014) 41:545–7. doi: 10.1188/14.ONF.545-547

24. Braun V, Clarke V, Boulton E, Davey L, McEvoy C. The online survey as a qualitative research tool. *Int J Soc Res Methodol*. (2021) 24:641–54. doi: 10.1080/13645579.2020.1805550

25. Tashakkori A, Johnson RB, Teddlie C. *Foundations of mixed methods research: Integrating quantitative and qualitative approaches in the social and behavioral sciences*. London: SAGE Publications (2020).

26. Saldaña J. *The coding manual for qualitative researches*. Great Britain: SAGE Publications Ltd (2009).

27. Meyer DZ, Avery LM. Excel as a qualitative data analysis tool. *Field Methods*. (2009) 21:91–12. doi: 10.1177/1525822X08323985

28. Tolley EE. *Qualitative methods in public health: A field guide for applied research*. Newark: John Wiley & Sons, Incorporated (2016).

29. Helmer J, Senior K, Davison B, Vodic A. Improving sexual health for young people: making sexuality education a priority. *Sex Educ*. (2015) 15:158–71. doi: 10.1080/14681811.2014.989201

30. Castillo Nuñez J, Derluyn I, Valcke M. Young people's expressed needs for comprehensive sexuality education in Ecuadorian schools. *Sex Educ*. (2018) 18:14–31. doi: 10.1080/14681811.2017.1374941

31. Fisher CM, Kerr L, Ezer P, Kneip Pelster AD, Coleman JD, Tibbits M. Adolescent perspectives on addressing teenage pregnancy and sexually transmitted infections in the classroom and beyond. *Sex Educ*. (2019) 20:90–00. doi: 10.1080/14681811.2019.1618257

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Menopause modified the association of blood pressure with osteoporosis among gender: a large-scale cross-sectional study

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Purpose: This study aimed to assess the potential association between blood pressure and osteoporosis in a rural population with limited resources. Existing evidence on this association is limited, particularly in such settings.

Methods: Data from 7,689 participants in the Henan Rural Cohort study were analyzed. Four blood pressure indicators [systolic blood pressure (SBP), diastolic blood pressure (DBP), mean arterial pressure (MAP), and pulse pressure (PP)] were measured. The logistic regression model and restricted cubic spline plots were used to assess the relationship between blood pressure indicators and osteoporosis prevalence.

Results: Positive trends were noted between blood pressure indicators and osteoporosis prevalence in the entire group and women ($P_{\text{trend}} < 0.05$ for SBP, MAP, and PP). Women with higher SBP and PP exhibited elevated odds of osteoporosis compared with those with the lowest SBP and PP (*OR*s ranging from 1.15 to 1.5 for SBP and 1.06 to 1.83 for PP). No such associations were found in men. These relationships were only evident in postmenopausal women. Dose–response analysis confirmed these findings. Excluding participants taking hypertension medication did not alter the results.

Conclusion: In resource-limited settings, higher SBP and PP are associated with the increased prevalence of osteoporosis in women, potentially influenced by menopause-related factors. This indicates that potential gender-based differences and social inequalities may affect bone health.

Clinical trial registration: The Henan Rural Cohort Study has been registered at the Chinese Clinical Trial Register (Registration number: ChiCTR-OOC-15006699) http://www.chictr.org.cn/showproj.aspx?proj=11375.

KEYWORDS

osteoporosis, blood pressure, rural health, gender-based differences, menopause

Introduction

Osteoporosis is a medical condition that causes a reduction in bone mass and deterioration of bone structure. This can lead to weakened bones that are more susceptible to fractures (1). In China, there has been a recent increase in the prevalence of osteoporosis, with rates reaching 13.5% in men and 29.0% in women (2). This increase is largely attributable to the aging population (3). It is estimated that between 60 and 120 million individuals will suffer from osteoporosis, resulting in ∼5.99 million fractures and an estimated cost of 25.43 billion dollars in 2050 (4, 5). This unexpected cost can place a significant economic burden on both society and families, particularly in rural areas where medical resources are limited (3).

Elevated blood pressure, a major risk factor for death and disability globally (6), accounts for 10.4 million deaths worldwide (7). Recent research has concentrated on the relationship between osteoporosis and blood pressure as they share common regulatory mechanisms (8). Several epidemiological studies have demonstrated that patients with hypertension are at an increased risk of bone loss and decreased bone turnover (9, 10). A cross-sectional study of 270 postmenopausal Turkish women indicated that hypertension was associated with low bone mineral density (BMD), while a meta-analysis of 1,43,0431 participants and 148,048 osteoporotic fracture cases revealed a higher risk of fractures among patients with hypertension (11). However, this association may be gender-based. A longitudinal study revealed that hypertension was associated with high femoral neck BMD in men and low BMD in women (12). In addition, the previous meta-analysis concluded a similar result, in which the association between hypertension and osteoporosis was more remarkable in women than in men (11).

Although numerous studies have yielded comprehensive results on the relationship between hypertension and osteoporosis, evidence in areas with limited resources remains scarce. Furthermore, the research on the association between specific blood pressure indicators, such as systolic blood pressure (SBP), diastolic blood pressure (DBP), mean arterial pressure (MAP), and pulse pressure (PP), and osteoporosis is also limited. As a result, this study aimed to indicate whether there would be gender-based variations in the association between blood pressure indicators and osteoporosis prevalence in areas with limited resources.

Methods

Study participants

This research used data from the Henan Rural Cohort study (ChiCTR-OOC-15006699), previously described in detail (13). Briefly, the cohort recruited 39,259 adults aged 18–79 years residing in five rural Henan counties (Suiping, Yuzhou, Xinxiang, Tongxu, and Yima) between 2015 and 2017. The study aimed to investigate the prevalence of chronic non-communicable diseases (NCDs) like hypertension, stroke, and osteoporosis in this rural population and to explore potential links between environmental exposures, genetics, and NCD risk. Eligibility criteria included the following: subjects were permanent residents, were healthier so as to answer our questionnaire, and did not move out in the following-up.

Among 39,259 individuals who participated in the Henan Rural Cohort study baseline, only 8,475 underwent bone mineral density (BMD) testing. To ensure data quality and minimize confounding factors, we excluded participants with missing BMD data ($n = 442$) and missing blood pressure measurements ($n = 304$). Additionally, to avoid potential disruptions in bone metabolism due to chronic health conditions, we excluded individuals with a history of stroke, cancer, or kidney failure ($n = 40$). This resulted in a final analysis sample of 7,689 participants. The study protocol was approved by the Zhengzhou University Life Science Ethics Committee [Code: (2015) MEC (S128)], and all participants provided informed consent.

Covariate measurement

Sociodemographic and lifestyle data**:** trained researchers collected data through a structured questionnaire on participants' age (categorized as 18–44, 45–64, and 65+), gender, education level (elementary or below, junior high school, and high school or above), marital status (married/cohabiting, unmarried/divorced/widowed), and average monthly income (<¥500, ¥500-¥1000, and >¥1000). Lifestyle habits: smoking and drinking habits were categorized as never, former, or current. Physical activity measured by metabolic equivalent (MET) hours per week was classified into low, moderate, and high. More vegetable and fruit intake was defined as one person who ate vegetables and fruits over 500 g/day on average while a high-fat diet was defined as consuming more than 75 g of fat/day on average. The body mass index (BMI) was calculated from the height and weight measured using standardized equipment. Medical information: menopause status was determined through the a questionnaire. Antihypertensive medication use was defined as taking antihypertensive drugs in the past 2 weeks.

Measurement of blood pressure and definition of hypertension

BP was measured using electronic sphygmomanometers (Omron HEM-7071A) on the right arm. After resting for at least 5 min in a seated position with their arm at heart level, participants had their BP measured three times (14, 15). The average of these three readings was used for analysis. The mean arterial pressure (MAP) and pulse pressure (PP) were calculated as follows:

MAP: 2/3 DBP + 1/3 SBP; PP: SBP – DBP

Hypertension was defined as

- Mean SBP $\geq$ 140 mmHg or DBP $\geq$ 90 mmHg
- Self-reported history of hypertension
- Taking antihypertensive medication in the past 2 weeks

Definition of osteoporosis

The BMD of the participants' heels was measured by trained staff blinded to the study, using a Sahara clinical bone densitometer.

Participants were positioned comfortably seated with specific leg angles for optimal measurement. Three measurements were taken on the left heel, or the other heel if a previous fracture was present (16). Osteoporosis was diagnosed using the WHO criteria based on T-scores calculated from the BMD measurements. The T score represents the number of standard deviations for which a person's BMD deviates from the average healthy adult population. Individuals with a T-score ≤ -2.5 were classified as having osteoporosis (17).

Statistical analyses

Considering that a potential gender-difference association between osteoporosis and blood pressure may exist, all analyses in this research were presented by gender. Continuous variables were expressed as mean ± SD, and categorical variables were expressed as counts with percentages. Gender-difference characteristics were detected by using one-way ANOVA or Pearson's chi-square test. Blood pressure indicators were divided into five groups, and the group with the lowest levels was set as the reference group. The prevalence of osteoporosis was presented by gender and blood pressure group, and unadjusted binary logistic regression was employed to detect the trend. With adjustment of age, gender, educational level, marital status, average monthly income, smoking, drinking status activity, high-fat diet, more vegetable and fruit intake, and BMI, the binary logistic regression model was used to explore the associations between blood pressure and osteoporosis, and to test the robustness of our findings, we further explored the relationship between blood pressure indicators and the risk of osteoporosis in the older adults by gender (≥50 years old). In addition, the restricted cubic spline interpolation plot was used to explore the trend. Considering menopause status is usually related to osteoporosis (18), a stratified analysis among women by menopause status was also conducted. To test the robustness of our findings, some sensitivity analyses excluding medications for hypertension were done. All analyses were conducted via R software version 4.0.3. All tests were two-tailed, and $P < 0.05$ was regarded as statistically significant.

Results

Baseline characteristics of participants

This research included 2,879 men (37.44%) and 4,810 women (62.56%). Osteoporosis was identified in 405 (14.07%) men and 1,089 (22.64%) women. Women were found to be older, have lower educational levels, higher average monthly income, and were less likely to smoke or drink, less physically active, with a lower rate of a high-fat diet, but had a higher intake of vegetables and fruits and a higher BMI compared to men (all P <0.05). Moreover, women had lower systolic blood pressure (121.72 ± 18.94 vs. 122.64 ± 17.58 mmHg), diastolic blood pressure (74.97 ± 10.86 vs. 76.52 ± 11.32 mmHg), and mean arterial pressure (90.56 ± 12.78 vs. 91.89 ± 12.73 mmHg), but higher pulse pressure (46.75 ± 12.53 vs. 46.12 ± 10.89 mmHg) than men (all P <0.05). See Table 1 for more details. In addition, the baseline characteristics of participants by osteoporosis and hypertension are shown in Supplementary Tables 1, 2.

Prevalence of osteoporosis in groups with different blood pressure indicators

The graph in Figure 1 shows the changes in osteoporosis prevalence in different blood pressure indicator groups. In the SBP subgroups, the prevalence of osteoporosis among women increased from 16.49% (95% CI: 14.54% and 18.44%) in the group with <110 mmHg to 30.17% (95% CI: 27.05% and 33.29%) in the 140-mmHg group (P_{trend} <0.001). However, this trend was not observed in men, who had prevalence rates ranging from 16.87% to 17.67% ($P_{\text{trend}} = 0.636$). In MAP subgroups, the prevalence of osteoporosis among women increased from 19.10% (95% CI: 16.66%—21.54%) in the group with <80 mmHg to 25.19% (95% CI: 20.88%—29.51%) in the 110-mmHg group (P_{trend} <0.001). Again, this trend was not observed in men, who had prevalence rates ranging from 18.92% to 22.12% ($P_{\text{trend}} = 0.132$). The prevalence of osteoporosis in women increased sharply with increasing PP, from 15.28% in the group with <40 mmHg to 42.08% in the 70-mmHg group (P_{trend} <0.001). However, this trend was not observed in men. The trends remained stable even when medications were excluded from the analysis (see Supplementary Figure 1).

Gender-difference association between blood pressure and osteoporosis

As depicted in Figure 2, among women, the odds ratios (OR) and their 95% confidence intervals (CI) for four categories were as follows: 1.15 (0.92, 1.43), 1.33 (1.06, 1.67), 1.23 (0.95, 1.59), and 1.50 (1.19, 1.90) for systolic blood pressure (SBP) and 1.06 (0.87, 1.29), 1.32 (1.06, 1.65), 1.27 (0.97, 1.66), and 1.83 (1.34, 2.52) for pulse pressure (PP) compared with the reference group. However, these positive associations between blood pressure indicators and osteoporosis were significantly observed only in diastolic blood pressure (DBP) among men. Nonetheless, the dose–response relationships were only found in women, as shown in Supplementary Figure 2. These results were consistent even after excluding the medication use, as demonstrated in Supplementary Figures 3, 4. Furthermore, we also explored the relationship between blood pressure indicators and the risk of osteoporosis in the older adults by gender (≥50 years old). The results are shown in Supplementary Tables 3, 4. The results were similar to those of the main analysis.

Sensitivity analyses

The results of sensitivity analyses are presented in Figure 3. The study found positive associations between SBP and PP, with osteoporosis only among postmenopausal women. The odds ratios

TABLE 1 Baseline characteristics of participants by gender.

Variables	Total	Men	Women	*P*
	N = 7,689	*N* = 2,879	*N* = 4,810	
Age, *n* (%)				<0.001
18-	1,137 (14.79)	365 (12.68)	772 (16.05)	
45-	4,889 (63.58)	1,766 (61.34)	3,123 (64.93)	
65-	1,663 (21.63)	748 (25.98)	915 (19.02)	
Educational level, *n* (%)				<0.001
Elementary school or below	3,377 (43.92)	949 (32.96)	2,428 (50.48)	
Junior high school	3,181 (41.37)	1,348 (46.82)	1,833 (38.11)	
High school or above	1,131 (14.71)	582 (20.22)	549 (11.41)	
Marital status, *n* (%)				0.138
Married/cohabitating	7,011 (91.18)	2,643 (91.80)	4,368 (90.81)	
Unmarried/divorced /widowed	678 (8.82)	236 (8.20)	442 (9.19)	
Average monthly income, *n* (%)				
<500 RMB	2,444 (31.79)	985 (34.21)	1,459 (30.33)	
500–1000 RMB	2,323 (30.21)	854 (29.66)	1,469 (30.54)	
≥1000 RMB	2,922 (38.00)	1,040 (36.12)	1,882 (39.13)	
Smoking status, *n* (%)				<0.001
Never	5,689 (74.90)	932 (32.99)	4,757 (99.73)	
Former	364 (4.79)	358 (12.67)	6 (0.13)	
Current	1,542 (20.30)	1,535 (54.34)	7 (0.15)	
Drinking status, *n* (%)				<0.001
Never	5,918 (77.90)	1,303 (46.12)	4,615 (96.71)	
Former	1,077 (14.18)	951 (33.66)	126 (2.64)	
Current	602 (7.92)	571 (20.21)	31 (0.65)	
Physical activity, *n* (%)				<0.001
Low	2,151 (27.98)	917 (31.85)	1,234 (25.65)	
Moderate	2,961 (38.51)	819 (28.45)	2,142 (44.53)	
High	2,577 (33.52)	1,143 (39.70)	1,434 (29.81)	
High-fat diet, *n* (%)	1,552 (20.18)	783 (27.20)	769 (15.99)	<0.001
More vegetable and fruit intake, *n* (%)	4,060 (52.80)	1,571 (54.57)	2,489 (51.75)	0.016
Hypertension, *n* (%)	1,932 (25.13)	694 (24.11)	1,238 (25.74)	0.110
Osteoporosis, *n* (%)	14,94 (19.43)	405(14.07)	1,089 (22.64)	<0.001
Postmenopausal women, *n* (%)	2,687 (34.95)	-	2,687 (55.86)	-
BMI (kg/m^2, mean ± SD)	24.65 ± 3.42	24.39 ± 3.37	24.80 ± 3.44	<0.001
SBP (mmHg, mean ± SD)	122.07 ± 18.44	122.64 ± 17.58	121.72 ± 18.94	0.034
DBP (mmHg, mean ± SD)	75.55 ± 11.06	76.52 ± 11.32	74.97 ± 10.86	<0.001
MAP (mmHg, mean ± SD)	91.06 ± 12.78	91.89 ± 12.73	90.56 ± 12.78	<0.001
PP (mmHg, mean ± SD)	46.51 ± 11.95	46.12 ± 10.89	46.75 ± 12.53	0.027

BMI, body mass index; SBP, systolic blood pressure; DBP, diastolic blood pressure; MAP, mean arterial pressure; PP, pulse pressure.

(95% confidence intervals) were 1.02 (0.78, 1.33), 1.18 (0.91, 1.53), 1.07 (0.80, 1.44), and 1.38 (1.06, 1.79) for SBP and 1.01 (0.79, 1.29), 1.20 (0.93, 1.55), 1.17 (0.87, 1.57), and 1.68 (1.19, 2.35) for PP. The findings were supported by the dose–response relationships (see Supplementary Figure 5) and by excluding subjects who were taking medication (see Supplementary Figures 6, 7).

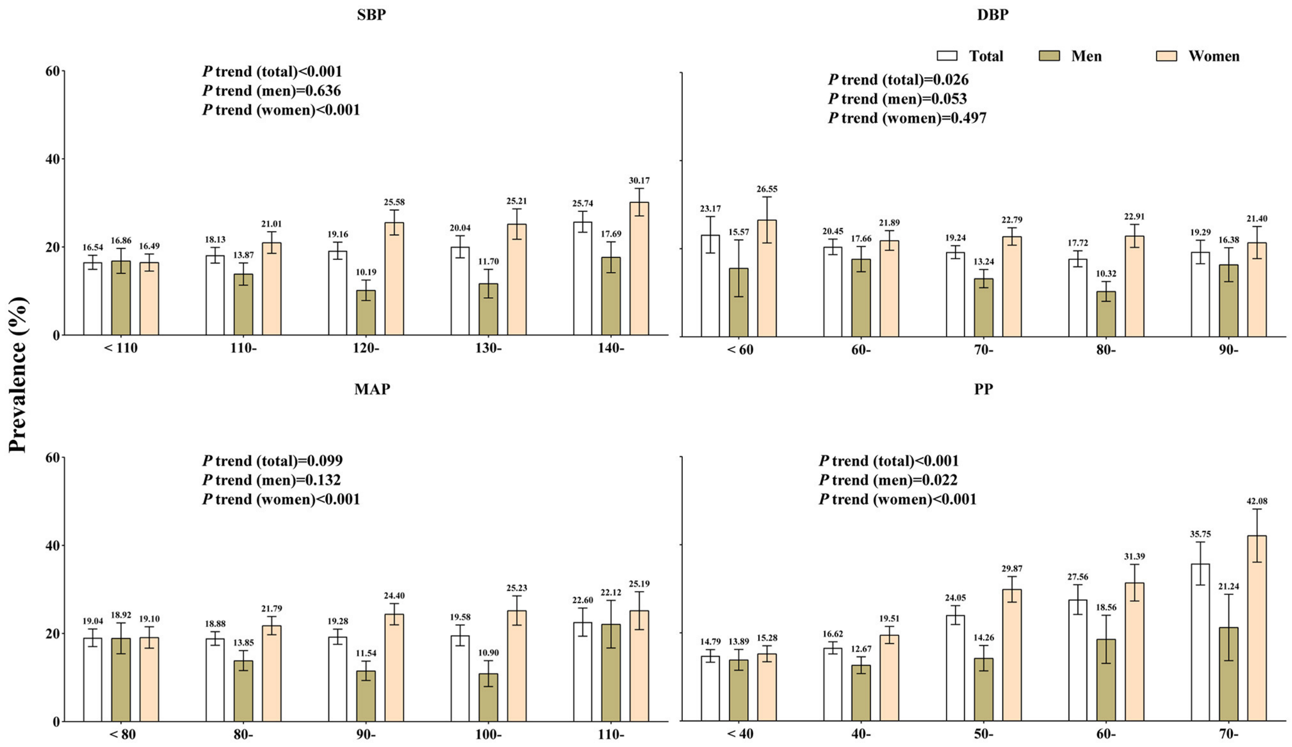

FIGURE 1
Prevalence of osteoporosis in the different blood pressure indicator groups by gender. SBP, systolic blood pressure; DBP, diastolic blood pressure; MAP, mean arterial pressure; PP, pulse pressure.

Discussion

This cross-sectional study focuses on people with limited resources and aims to explore the association between different blood pressure proxies and osteoporosis. It is the first study of its kind, and it seeks to detect potential gender-difference associations. The results show that with increase in SBP, MAP, and PP, the prevalence of osteoporosis increases among women. Positive associations between SBP and PP with the risk of osteoporosis were observed in women only. In sensitivity analysis, similar conclusions were observed in postmenopausal women merely. The results were still robust even after excluding subjects taking medications, which suggests that the gender–difference association between blood pressure indicators and osteoporosis might be due to menopause status.

The prevalence of osteoporosis in China varies among studies. However, women are consistently more susceptible to the condition (3). A recent study conducted in China found that the occurrence of osteoporosis is more prevalent among older women compared to older men. Specifically, in northern China, the prevalence of osteoporosis was 36.9% among older women and 19.8% among older men. In contrast, in northwestern China, the prevalence was 9.65% among older women and 8.08% among older men (19). According to a nationwide study, osteoporosis affected 6.46% of men and 29.13% of women (4). According to a recent study, 29.0% of women and 13.5% of men suffer from osteoporosis on a national level (2), whereas in this research, the prevalence of osteoporosis was 14.07% for men and 22.64% for women in Chinese rural areas. In rural areas of China, men were the primary workforce and engaged in agricultural work, while women typically stayed at home as housewives (20). This resulted in men having not only a higher socioeconomic status but also being exposed to sunlight for longer periods of time than women. Additionally, previous research has found that Asian women tend to avoid sunlight exposure and skin-tanning, resulting in shorter sunlight exposure and lower levels of serum 25 (OH) D compared to men (21). Therefore, the prevalence of osteoporosis was higher in women than in men.

Numerous research studies have repeatedly linked hypertension with prevalent osteoporosis (22). An early cohort study conducted in 3,676 older adults white women found that increasing SBP corresponded with an increasing rate of bone loss ($P_{\text{non-linear}} < 0.05$) (9), which was similar to our results. A recent cross-section study also indicated that both SBP and DBP were inversely related with the BMD of proximal femoral and lumbar vertebral and that the beta values were −0.382, −0.290, and −0.340 of SBP and −0.318, −0.340, and −0.304 of DBP (23). A similar association was also observed in Chinese population (24, 25). Conducted in Tibet, a retrospective cross-sectional study also found that SBP was inversely associated with BMD T score of the spine and femoral neck or hip among diabetic postmenopausal women (24). A recent case-control study also indicated that hypertension was positively associated with osteoporosis (26). In contrast to our results, Javed et al. (27) reported that hypertension was not correlated with low BMD at either lumbar spine or both femoral necks among African American women aged over 65 years. A retrospective analysis also pronounced that there was no

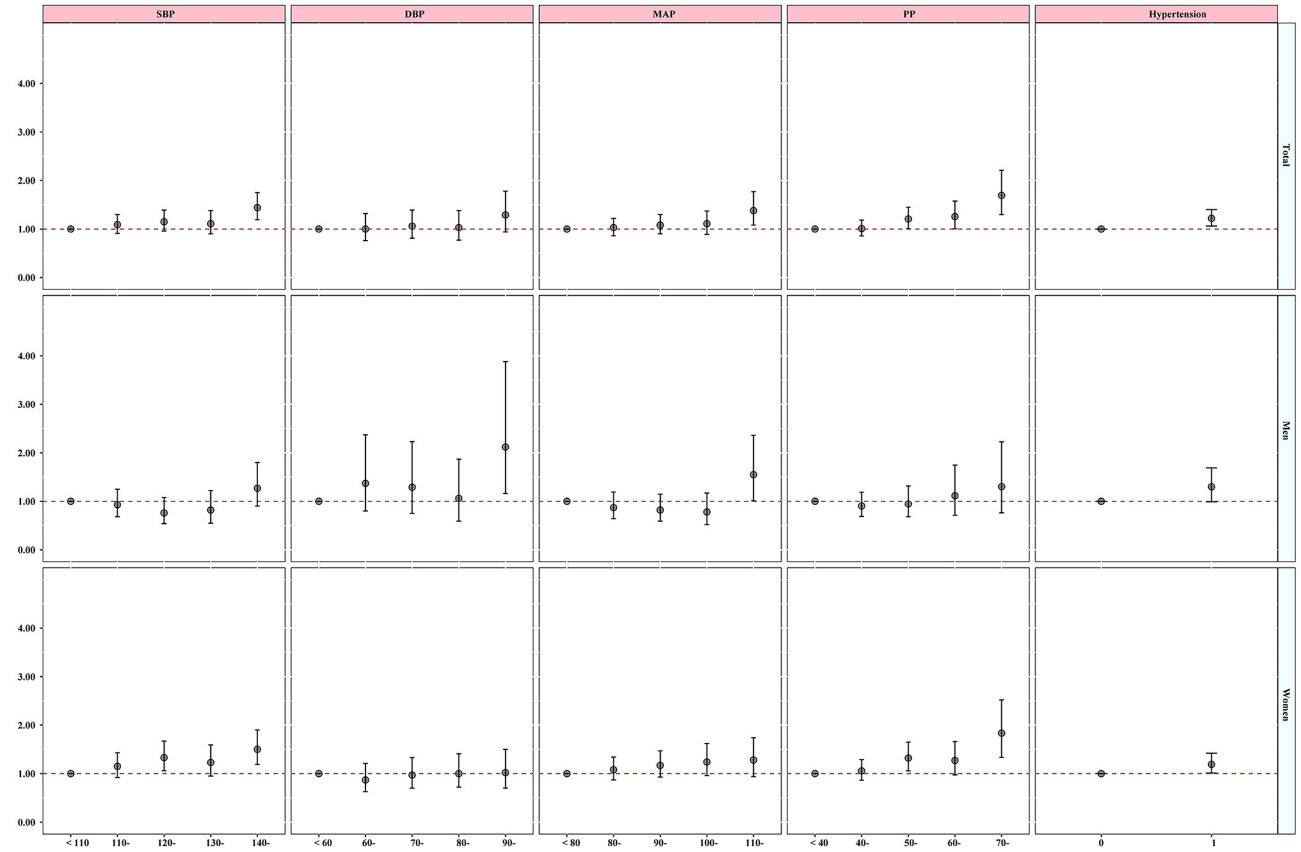

FIGURE 2
Gender-difference association on blood pressure indicators and hypertension with osteoporosis. The model was adjusted by age, gender (Total), educational level, marital status, average monthly income, smoking, drinking status, physical activity, high-fat diet, increased vegetable and fruit intake, and BMI.

significant difference between hypertension and non-hypertension participants in the BMD of the femur or the spine (28). Evidence from Korea National Health and Nutrition indicated that lumbar spine osteoporosis was not significantly associated with blood pressure (29). A population-based Mendelian randomization study conducted among European populations also revealed a potential positive association between the PP and forearm BMD (30). These differences may be the results of different regions, environmental exposures, lifestyles, races, and other underlying factors.

The gender–difference association of between blood pressure with osteoporosis was repeatedly observed in previous research. For instance, Loke et al. (25) observed that both SBP and DBP were negatively associated with BMD among women but not significantly associated among men. A recent longitudinal study also highlighted that the BMD of the femoral neck was lower in women with hypertension than those without hypertension (0.80 vs. 0.82), while in men, hypertension was positively associated with the BMD of the lumbar spine and femoral neck (12). Additionally, evidence from a meta-analysis pronounced that the association between fracture and hypertension was slightly stronger in women (pooled *OR* = 1.52, 95% CI 1.30–1.79) than in men (pooled *OR* = 1.35, 95% CI 1.26–1.44) (11). Given the positive association between blood pressure and osteoporosis disappeared in premenopausal women, the gender–difference association may be attributed to the menopause status. Additionally, a lower sample size in men may also contribute to this statistical insignificance.

Despite the potential mechanism of blood pressure and osteoporosis not yet being clarified, limited studies still provided various pieces of evidence. Calcium may be a primary bridge between blood pressure and osteoporosis (31). Previous research has reported that participants with hypertension had a higher calcium elimination and a lower intestinal absorption than non-hypertension participants, which contributed to a lower calcium concentration in the plasma (31, 32). To sustain a suitable blood calcium level, bones may break down and release calcium into the blood (33). Therefore, the bone may be porous and prone to fractures (34). In addition, recent research also found that hypertension corresponded with the low level of 25-hydroxy vitamin D and osteocalcin, which led to a low bone turnover (10). Recent studies have found that angiotensin receptor blockers, selective beta-adrenergic receptor blockers, and thiazide diuretics may improve bone trabecular number and bone density by stimulating osteoblast differentiation and reducing osteoclast generation (35, 36), which supported that the medications of hypertension might impact the association.

During menopause and postmenopause, the reduced estrogen level in women would contribute to an increased osteoclastic resorption activity without a suitable increase in osteoblastic

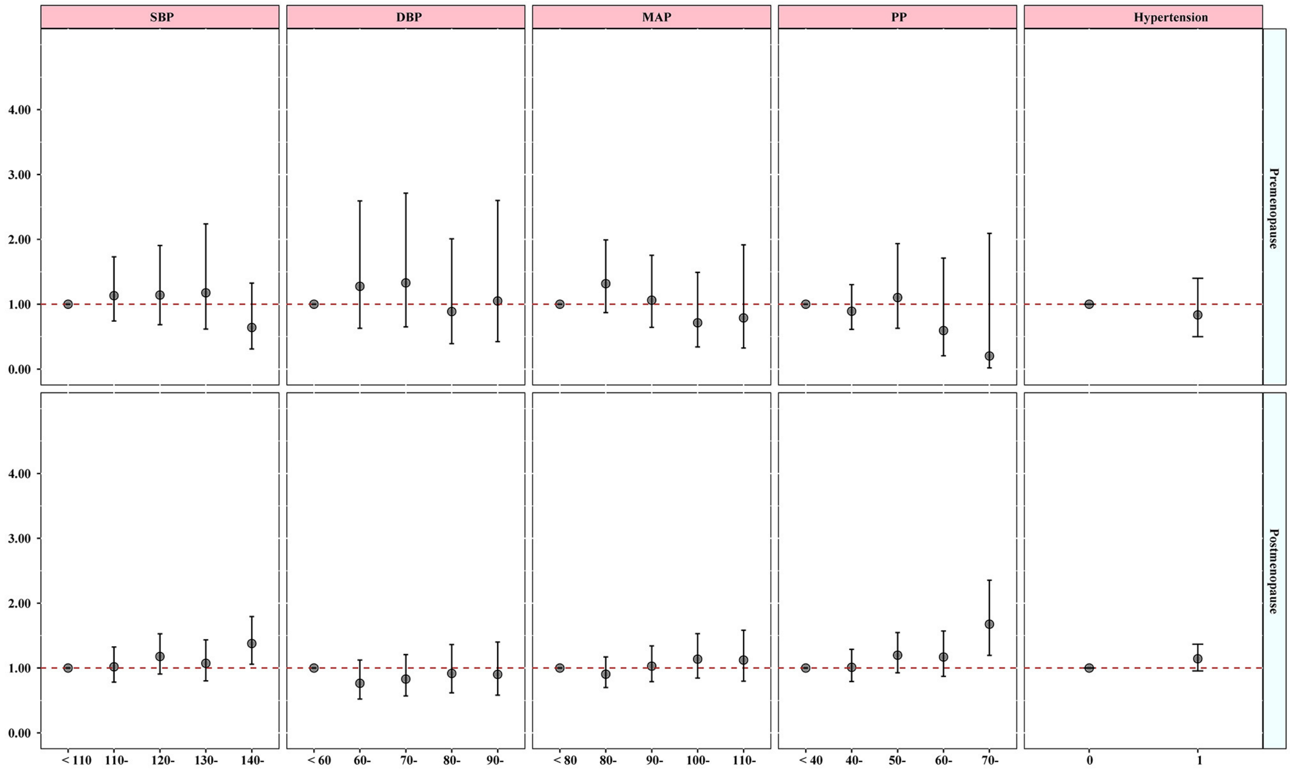

FIGURE 3
Association between blood pressure indicators and hypertension with osteoporosis considering menopause status. The model was adjusted by age, educational level, marital status, average monthly income, smoking, drinking status, physical activity, high-fat diet, increased vegetable and fruit intake, and BMI.

activity, which leads to a net loss of bone and a decreased bone strength (37–39). Thus, the low bone strength may explain that the significant association between blood pressure and osteoporosis was only observed among postmenopausal women but not among men or premenopausal women.

To the best of our knowledge, this is the first research to explore the gender–difference associations between divergent blood pressure proxies and osteoporosis among rural population. Despite that a large sample size and appropriate statistical methods could make this research more convincing, some limitations should be noted. First, only 7,689 participants from the Henan Rural Cohort study were included in this research and exclusion of participants with diseases and missing information may induce inevitable errors. Second, quantitative ultrasound (QUS) measures rather than X-ray absorptiometry (DXA) might underestimate osteoporosis prevalence; however, previous studies found it appeared capable of replacing dual X-ray considering its portability and low cost in the population-based study (40). Third, we did not examine the impact of vitamin D, calcium intake, and use of menopausal hormonal therapy and other medications on bone health, osteoprotegerin, and osteocalcin levels of the subjects, which may increase inevitable biases. Moreover, the unraveling reverse causality cannot be ruled out because of the survey based on a cross-sectional study. Finally, the results were based on the observations in rural populations, so caution is required when applying to other populations.

Conclusion

While this study found a positive association between higher blood pressure (systolic and pulse) and osteoporosis in women living in resource-limited areas, it is crucial to note that this association was only significant for postmenopausal women. This suggests menopause itself may be a key factor in the observed gender difference. Therefore, while these findings raise the possibility of using blood pressure as a screening tool for osteoporosis in postmenopausal women with limited resources, further research is necessary to determine the generalizability of this association to all women, especially those yet to experience menopause.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving humans were approved by the Zhengzhou University Life Science Ethics Committee. The studies were conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

Author contributions

HJ: Formal analysis, Investigation, Methodology, Writing – original draft. HZ: Formal analysis, Investigation, Writing – original draft. SJ: Formal analysis, Investigation, Writing – review & editing. XY: Investigation, Writing – review & editing. XL: Data curation, Investigation, Writing – review & editing. CW: Data curation, Funding acquisition, Project administration, Supervision, Writing – review & editing. GZ: Investigation, Validation, Writing – review & editing. JP: Methodology, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2024.1383349/full#supplementary-material

References

1. Hsu E, Pacifici R. From osteoimmunology to osteomicrobiology: how the microbiota and the immune system regulate bone. *Calcif Tissue Int.* (2018) 102:512–21. doi: 10.1007/s00223-017-0321-0

2. Cheng X, Zhao K, Zha X, Du X, Li Y, Chen S, et al. Opportunistic screening using low-dose ct and the prevalence of osteoporosis in china: a nationwide, multicenter study. *J Bone Miner Res.* (2021) 36:427–35. doi: 10.1002/jbmr.4187

3. Yu F, Xia W. The epidemiology of osteoporosis, associated fragility fractures, and management gap in China. *Arch Osteoporos.* (2019) 14:32. doi: 10.1007/s11657-018-0549-y

4. Zeng Q, Li N, Wang Q, Feng J, Sun D, Zhang Q, et al. The prevalence of osteoporosis in China, a nationwide, multicenter DXA survey. *J Bone Miner Res.* (2019) 34:1789–97. doi: 10.1002/jbmr.3757

5. Si L, Winzenberg TM, Jiang Q, Chen M, Palmer AJ. Projection of osteoporosis-related fractures and costs in China: 2010-2050. *Osteoporos Int.* (2015) 26:1929–37. doi: 10.1007/s00198-015-3093-2

6. Niu M, Zhang L, Wang Y, Tu R, Liu X, Wang C, et al. Lifestyle score and genetic factors with hypertension and blood pressure among adults in rural China. *Front Public Health.* (2021) 9:687174. doi: 10.3389/fpubh.2021.687174

7. Unger T, Borghi C, Charchar F, et al. 2020 international society of hypertension global hypertension practice guidelines. *Hypertension.* (2020) 75:1334–57. doi: 10.1161/HYPERTENSIONAHA.120.15026

8. McFarlane SI, Muniyappa R, Shin JJ, Bahtiyar G, Sowers JR. Osteoporosis and cardiovascular disease: brittle bones and boned arteries, is there a link? *Endocrine.* (2004) 23:1–10. doi: 10.1385/ENDO:23:1:01

9. Cappuccio FP, Meilahn E, Zmuda JM, Cauley JA. High blood pressure and bone-mineral loss in elderly white women: a prospective study. *Study Osteop Fract Res Group Lancet.* (1999) 354:971–5. doi: 10.1016/S0140-6736(99)01437-3

10. Hu Z, Yang K, Hu Z, et al. Determining the association between hypertension and bone metabolism markers in osteoporotic patients. *Medicine.* (2021) 100:e26276. doi: 10.1097/MD.0000000000026276

11. Li C, Zeng Y, Tao L, et al. Meta-analysis of hypertension and osteoporotic fracture risk in women and men. *Osteoporos Int.* (2017) 28:2309–18. doi: 10.1007/s00198-017-4050-z

12. Yang S, Nguyen ND, Center JR, Eisman JA, Nguyen TV. Association between hypertension and fragility fracture: a longitudinal study. *Osteoporos Int.* (2014) 25:97–103. doi: 10.1007/s00198-013-2457-8

13. Liu X, Mao Z, Li Y, Wu W, Zhang X, Huo W, et al. Cohort profile: the henan rural cohort: a prospective study of chronic non-communicable diseases. *Int J Epidemiol.* (2019) 48:1756-j. doi: 10.1093/ije/dyz039

14. Chobanian AV, Bakris GL, Black HR, et al. Seventh report of the joint national committee on prevention, detection, evaluation, and treatment of high blood pressure. *Hypertension.* (2003) 42:1206–52. doi: 10.1161/01.HYP.0000107251.49515.c2

15. Li R, Tian Z, Wang Y, Liu X, Tu R, Wang Y, et al. The association of body fat percentage with hypertension in a Chinese rural population: the henan rural cohort study. *Front Public Health.* (2020) 8:70. doi: 10.3389/fpubh.2020.00070

16. Lu YH, Geng JH, Wu DW, Chen SC, Hung CH, Kuo CH. Betel nut chewing decreased calcaneus ultrasound t-score in a large taiwanese population follow-up study. *Nutrients.* (2021) 13:655. doi: 10.3390/nu13103655

17. Chang CF, Lee JI, Huang SP, Geng JH, Chen SC. Regular exercise decreases the risk of osteoporosis in postmenopausal women. *Front Public Health.* (2022) 10:897363. doi: 10.3389/fpubh.2022.897363

18. Kanis JA, Cooper C, Rizzoli R, et al. Executive summary of the European guidance for the diagnosis and management of osteoporosis in postmenopausal women. *Calcif Tissue Int.* (2019) 104:235–8. doi: 10.1007/s00223-018-00512-x

19. Tian L, Yang R, Wei L, Liu J, Yang Y, Shao F, et al. Prevalence of osteoporosis and related lifestyle and metabolic factors of postmenopausal women and elderly men:

a cross-sectional study in Gansu province, Northwestern of China. *Medicine.* (2017) 96:e8294. doi: 10.1097/MD.0000000000008294

20. Pan X, Zhang Y, Xu L, Huang J, Zhao Q. An analysis of farmers' perception of the new cooperative medical system in Liaoning Province, China. *BMC Health Serv Res.* (2009) 9:230. doi: 10.1186/1472-6963-9-230

21. Wang LK, Hung KC, Lin YT, et al. Age, gender and season are good predictors of vitamin D status independent of body mass index in office workers in a subtropical region. *Nutrients.* (2020) 12:719. doi: 10.3390/nu12092719

22. Canoy D, Harvey NC, Prieto-Alhambra D, et al. Elevated blood pressure, antihypertensive medications and bone health in the population: revisiting old hypotheses and exploring future research directions. *Osteoporos Int.* (2022) 33:315–26. doi: 10.1007/s00198-021-06190-0

23. Cakmak HA, Cakmak BD, Yumru AE, Aslan S, Enhos A, Kalkan AK, et al. The relationships between blood pressure, blood glucose, and bone mineral density in postmenopausal Turkish women. *Ther Clin Risk Manag.* (2015) 11:1641–8. doi: 10.2147/TCRM.S95017

24. Zhou L, Song J, Yang S, Meng S, Lv X, Yue J, et al. Bone mass loss is associated with systolic blood pressure in postmenopausal women with type 2 diabetes in Tibet: a retrospective cross-sectional study. *Osteoporos Int.* (2017) 28:1693–8. doi: 10.1007/s00198-017-3930-6

25. Loke SS, Chang HW Li WC. Association between metabolic syndrome and bone mineral density in a Taiwanese elderly population. *J Bone Miner Metab.* (2018) 36:200–8. doi: 10.1007/s00774-017-0826-7

26. Chai H, Ge J, Li L, Li J, Ye Y. Hypertension is associated with osteoporosis: a case-control study in Chinese postmenopausal women. *BMC Musculoskelet Disord.* (2021) 22:253. doi: 10.1186/s12891-021-04124-9

27. Javed F, Khan SA, Ayers EW, Aziz EF, Akram MS, Nadkarni GN, et al. Association of hypertension and bone mineral density in an elderly African American female population. *J Natl Med Assoc.* (2012) 104:172–8. doi: 10.1016/S0027-9684(15)30140-1

28. Yazici S, Yazici M, Korkmaz U, Erkan ME, Baki AE, Erden I, et al. Relationship between blood pressure levels and bone mineral density in postmenopausal Turkish women. *Arch Med Sci.* (2011) 7:264–70. doi: 10.5114/aoms.2011.22077

29. Lee HT, Shin J, Min SY, Lim Y-H, Kim K-S, Kim SG, et al. The relationship between bone mineral density and blood pressure in the Korean elderly population: the Korea National Health and Nutrition Examination Survey, 2008-2011. *Clin Exp Hypertens.* (2015) 37:212–7. doi: 10.3109/10641963.2014.933971

30. He B, Yin L, Zhang M, Lyu Q, Quan Z, Ou Y. Causal effect of blood pressure on bone mineral density and fracture: a mendelian randomization study. *Front Endocrinol (Lausanne).* (2021) 12:716681. doi: 10.3389/fendo.2021.716681

31. Ilic K, Obradovic N, Vujasinovic-Stupar N. The relationship among hypertension, antihypertensive medications, and osteoporosis: a narrative review. *Calcif Tissue Int.* (2013) 92:217–27. doi: 10.1007/s00223-012-9671-9

32. Metz JA, Morris CD, Roberts LA, McClung MR, McCarron DA. Blood pressure and calcium intake are related to bone density in adult males. *Br J Nutr.* (1999) 81:383–8. doi: 10.1017/S0007114599000665

33. Hua P, Xiong Y, Yu Z, Liu B, Zhao L. Effect of chlorella pyrenoidosa protein hydrolysate-calcium chelate on calcium absorption metabolism and gut microbiota composition in low-calcium diet-fed rats. *Mar Drugs.* (2019) 17:348. doi: 10.3390/md17060348

34. Suzuki A, Minamide M, Iwaya C, Ogata K, Iwata J. Role of metabolism in bone development and homeostasis. *Int J Mol Sci.* (2020) 21:23. doi: 10.3390/ijms21238992

35. Zhang R, Yin H, Yang M, Lei X, Zhen D, Zhang Z. Advanced progress of the relationship between antihypertensive drugs and bone metabolism. *Hypertension.* (2023) 80:2255–64. doi: 10.1161/HYPERTENSIONAHA.123.21648

36. Ghosh M, Majumdar SR. Antihypertensive medications, bone mineral density, and fractures: a review of old cardiac drugs that provides new insights into osteoporosis. *Endocrine.* (2014) 46:397–405. doi: 10.1007/s12020-014-0167-4

37. Park JS, Choi SB, Rhee Y, Chung JW, Choi EY, Kim DW. Parathyroid hormone, calcium, and sodium bridging between osteoporosis and hypertension in postmenopausal Korean women. *Calcif Tissue Int.* (2015) 96:417–29. doi: 10.1007/s00223-015-9972-x

38. Li L, Qu Y, Jin X, Guo XQ, Wang Y, Qi L, et al. Protective effect of salidroside against bone loss via hypoxia-inducible factor-1alpha pathway-induced angiogenesis. *Sci Rep.* (2016) 6:32131. doi: 10.1038/srep32131

39. Kang IS, Agidigbi TS, Kwon YM, Kim D-G, Kim RI, In G, et al. Effect of co-administration of panax ginseng and brassica oleracea on postmenopausal osteoporosis in ovariectomized mice. *Nutrients.* (2020) 12:415. doi: 10.3390/nu12082415

40. Hoiberg MP, Rubin KH, Hermann AP, Brixen K, Abrahamsen B. Diagnostic devices for osteoporosis in the general population: a systematic review. *Bone.* (2016) 92:58–69. doi: 10.1016/j.bone.2016.08.011

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Factors influencing survival and mortality among adult Aboriginal Australians with bronchiectasis—A 10-year retrospective study

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Background: The prevalence of bronchiectasis among adult Aboriginal Australians is higher than that of non-Aboriginal Australians. However, despite evidence to suggest higher prevalence of bronchiectasis among Aboriginal people in Australia, there is sparce evidence in the literature assessing clinical parameters that may predict survival or mortality in this population.

Methods: Aboriginal Australians residing in the Top End Health Service region of the Northern Territory of Australia aged >18 years with chest computed tomography (CT) confirmed bronchiectasis between 2011 and 2020 were included. Demographics, body mass index (BMI), medical co-morbidities, lung function data, sputum microbiology, chest CT scan results, hospital admissions restricted to respiratory conditions and all-cause mortality were assessed.

Results: A total of 459 patients were included, of whom 146 were recorded deceased (median age at death 59 years). Among the deceased cohort, patients were older (median age 52 vs. 45 years, $p = 0.023$), had a higher prevalence of chronic obstructive pulmonary disease (91 vs. 79%, $p = 0.126$), lower lung function parameters (median percentage predicted forced expiratory volume in 1 s 29 vs. 40%, $p = 0.149$), a significantly greater proportion cultured non-*Aspergillus* fungi (65 vs. 46%, $p = 0.007$) and *pseudomonas* (46 vs. 28%, $p = 0.007$) on sputum microbiology and demonstrated bilateral involvement on radiology. In multivariate models advancing age, prior *pseudomonas* culture and Intensive care unit (ICU) visits were associated with increased odds of mortality. Higher BMI, better lung function on spirometry, prior positive sputum microbiology for *Haemophilus* and use of inhaled long-acting beta antagonist/muscarinic agents may have a favourable effect.

Conclusion: The results of this study may be of use to stratify high risk adult Aboriginal patients with bronchiectasis and to develop strategies to prevent future mortality.

KEYWORDS

First Nations, chest CT, morbidity, pulmonary, respiratory, Sputum culture, spirometry, severity

1 Introduction

Bronchiectasis is a chronic pulmonary condition that is clinically characterised by a vicious cycle of recurrent lower respiratory tract infections and airway inflammation (1, 2). Globally, there is emerging evidence to suggest that presence of bronchiectasis is associated with overall higher mortality rates (3–9). In the Australian context, approximately 3.3% of the population self-identify as of Aboriginal and/or Torres Strait Islander descent (from here on "Indigenous" is used to refer to global First nations populations, while "Aboriginal Australian/ population/patients/people" is used to specifically refer to Australians First Nations population) and the Northern Territory (NT) of Australia has the highest proportion of Australian Aboriginal people in comparison to all other Australian states and territories (10). Chronic respiratory disorders are reported to be highly prevalent among the adult Aboriginal Australian population (11, 12), and more so among those residing in the NT of Australia. In particular, prevalence of bronchiectasis is noted to be substantially higher among Aboriginal Australians compared to non-Aboriginal Australians (13, 14). Furthermore, hospital admission rates and overall mortality secondary to chronic respiratory disorders, including for bronchiectasis, are significantly higher among adult Aboriginal Australians in comparison to their non-Aboriginal counterparts (15–20).

In other diverse non-Indigenous ethnic populations, studies have demonstrated several factors which influence survival and mortality amongst patients with bronchiectasis such as, but not limited to lung function parameters, sputum microbiology, body mass index (BMI) and therapeutic interventions such as respiratory airway clearance (21–23). However, despite the high prevalence of bronchiectasis among Aboriginal Australians, there is scant evidence in the literature determining if these same factors are associated with survival or mortality in this population. The high prevalence of comorbidities, reduced lung function parameters (11–20, 24, 25), and unique environmental context of Aboriginal Australians which may predispose them to colonisation by other micro-organisms may indicate that other factors should be considered when predicting mortality risk. Hence, it is reasonable to explore those potential clinical parameters that may be influential for survival or mortality among adult Aboriginal Australians with bronchiectasis. This may lead on to identifying relevant clinical data for future interventions which would aid in reducing on-going adverse health consequences amongst adult Aboriginal patients suffering from bronchiectasis. Therefore, the aim of this study is to investigate and identify relevant clinical parameters that may indicate or influence survival and mortality in an adult Aboriginal Australian cohort diagnosed to have bronchiectasis over a 10-year study period (2011–2020) in the Top End Health Service (TEHS) region of the NT of Australia.

2 Materials and methods

2.1 Setting and study participants

This study was conducted at the respiratory and sleep division based at the Royal Darwin Hospital, a university affiliated tertiary care teaching hospital and Darwin Respiratory and Sleep Health, Darwin Private Hospital, within the TEHS, NT region of Australia. This study is a part of a larger project examining various aspects of bronchiectasis disease profiles among the adult Aboriginal population residing in the TEHS health district of the NT of Australia, which is inclusive of all adult Australian Aboriginal patients aged $\geq$18 years identified to have bronchiectasis via chest Computed tomography (CT) scan between 2011 and 2020.

2.2 Ethics

This study was approved by the Human Research Ethics governance/committee of the TEHS, NT and Menzies School of Health Research (Reference: HREC; 2019-3547). The authors acknowledge the rights of Australian Aboriginal people involved in this study, and as such conducted and reported according to strengthening and reporting of health research involving Aboriginal people (26), including consultations with institute Aboriginal representatives.

2.3 Clinical data assessed

Baseline demographics, age, sex, including smoking status and BMI when available were recorded. Patients usual place of residence as per community/suburb/postcode were collected, with further categorisation into the four health districts of the Top End (Darwin Urban, Darwin Rural, East Arnhem and Katherine), and by community when communities had >10 bronchiectasis cases were present (27). Presence of respiratory conditions alongside

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Abbreviations: BMI, body mass index; CAD, Coronary artery disease; COPD, chronic obstructive pulmonary disease; CI, confidence interval; CKD, chronic kidney disease; CT, computed tomography; FVC, forced vital capacity; FEV_1, forced expiratory volume in 1 s; HR, hazard ratios; ICS, inhaled corticosteroids; ICU, intensive care unit; IQR, interquartile range; LABA, long-acting beta antagonist; LAMA, long-acting muscarinic agent; NT, northern territory; ORs, odds ratios; TEHS, top end health service.

bronchiectasis and other medical comorbidities, including inhaled pharmacotherapy use [short-acting/long-acting beta antagonist (SABA/LABA), short-acting/long-acting muscarinic agents (SAMA/LAMA), inhaled corticosteroids (ICS)] were recorded. Details of chest CT scan findings, lung function test results [spirometry, forced vital capacity (FVC) and forced expiratory volume in 1 s (FEV_1)] and sputum microbiology results were also collected. Only respiratory related hospital admissions were extracted for assessment. All clinical parameters for this study were assessed via individual patients' electronic medical records. Mortality data was extracted through the hospital information system, death registry linkage was not utilised, and all-cause mortality was documented up until 31st December 2020. Further details on methods and study design are available form a recent report from our centre (27). The primary outcome in this study was all-cause mortality.

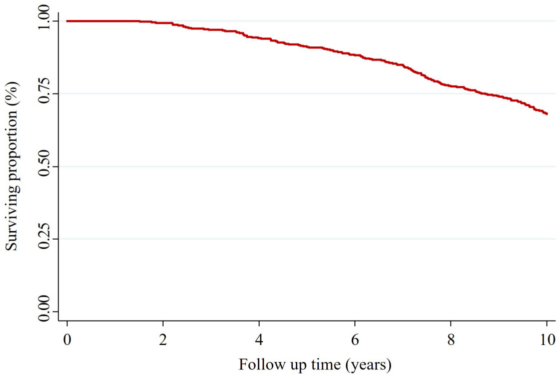

FIGURE 1
Kaplan-Meier survival graph across the study period (Jan 2011-December 2020).

2.4 Statistical analysis

Data were presented as median [interquartile range (IQR)] for continuous parameters, or number (%) for categorical parameters. Differences in demographic and clinical characteristics, radiological extent, hospitalisations and sputum cultures between surviving and deceased patients were tested via univariate logistic regression (categorical parameters) or quantile regression (continuous parameters). Stepwise Cox and quantile multivariate regressions were performed to identify factors associated with mortality and with age of death, respectively. The models included demographics (age, sex and residence location using Darwin area as reference), presence of comorbidities (restricted to those experienced by >10 patients in this cohort), radiological extent (bilateral involvement and ≥3 lobes effected), hospitalisation history (any hospitalisations, total time in hospital, any intensive care unit (ICU) visits), sputum results (presence of *haemophilus*, *streptococcus*, *moraxella*, *staphylococcus*, *aspergillus*, non-*Aspergillus* fungi, *klebsiella*, *mycobacterium* or *burkholderia*), and pharmacotherapy. A second multivariate Cox regression model was created (see **Supplementary material 2**) for mortality including BMI, current smoking and lung function, as only one-third of patients in our cohort had these data available. In the stepwise models, parameters with a p-value < 0.1 were considered for further analyses. Multicollinearity of included parameters was checked via Pearsons R^2, and pairs with an $R^2 > 0.5$ sequentially excluded until the models with the best fit were found. Results were presented as hazard ratios (HRs) (95% confidence intervals (CIs)) or beta (95% CI). All p-values were adjusted for multiple hypotheses testing via Romano-Wolf adjustment, utilising 750 bootstrap replications. Analyses were conducted in STATA IC 15 (College Station, Texas).

3 Results

3.1 Demographic and clinical details

In total 459 patients were retrospectively enrolled, of which 146 (31.8%) were recorded deceased by 31st of December 2020 (**Figure 1**). The median follow-up time for the surviving group was 10 years (IQR 10, 10), and for the deceased group was 7.1 years (IQR 4.6, 8.5). Patients who survived during this study period were younger upon enrolment [median 45 years (IQR 38.2, 53.5) vs. 52.2 years (IQR 43.4, 59.5), $p = 0.023$], had higher FEV_1 [40% (IQR 31, 35) vs. 29% predicted (IQR 24, 40)] and a lower prevalence of chronic obstructive pulmonary disease (COPD) (79 vs. 91%), lung cancer (3 vs. 7%), hypertension (58 vs. 73%), chronic kidney disease (CKD) (36 vs. 50%) and heart failure (7 vs. 12%) than patients who were recorded deceased (**Table 1**). However, following Romano-Wolf adjustment, aside from age, there were no statistically significant differences in relation to clinical, comorbidities nor inhaled pharmacotherapy prescriptions between the two groups. There were also no significant differences noted in mortality between health districts, nor at the community level (**Supplementary material 1**).

3.2 Chest CT findings and mortality

Chest CT scans demonstrated that the left lower lobe was the most commonly affected in both surviving (74%) and deceased (73%) cohorts, followed by the right lower lobe (59 and 69%, respectively) (**Table 2**). The left upper lobe appeared less commonly affected in the surviving cohort (19%) compared to the deceased cohort (27%), as was bilateral involvement (72 and 78%, respectively), however, the differences did not reach statistical significance.

3.3 Sputum cultures and mortality data

Sputum cultures were available for 425 patients [283 surviving (90.4%), 142 deceased (97.3%)]. *Haemophilus* was the most commonly cultured sputum overall, followed by non-*Aspergillus* fungi. Among those surviving, *haemophilus* (65 vs. 58%, $p = 0.593$) was the most commonly cultured organism, while among those deceased a greater proportion recorded non-*Aspergillus* fungi (65 vs. 46%, $p = 0.007$). *Pseudomonas* was also cultured less often in the

TABLE 1 Demographic and clinical characteristics of surviving and deceased patients.

Clinical parameters	Surviving ($n = 313$)	Deceased ($n = 146$)	RW-p
Age enrolment	45.02 (38.23, 53.51)	52.17 (43.41, 59.5)	0.007*
Age at death	–	59.17 (49.73, 67.42)	–
Sex (female)	173 (55.3%)	81 (55.5%)	0.987
BMI (kg/m^2)^	23.45 (19.57, 27.7)	21.67 (18.15, 25.12)	0.848
Darwin Urban	19 (6.1%)	15 (10.3%)	0.841
Darwin Rural	156 (49.8%)	70 (48.0%)	0.987
East Arnhem	78 (24.9%)	24 (16.4%)	0.649
Katherine	60 (19.2%)	37 (25.3%)	0.848
Never smoker^	15 (13.3%)	7 (18.9%)	0.960
Former smoker^	53 (46.9%)	11 (29.7%)	0.775
Current smoker^	45 (39.8%)	19 (51.4%)	0.861
FVC (% predicted)^	53 (42, 65)	44.5 (34, 53.5)	0.841
FEV$_1$ (% predicted)^	40 (31, 55)	29 (24, 40)	0.093
FEV$_1$/FVC^	0.66 (0.51, 0.76)	0.57 (0.43, 0.69)	0.828
COPD	247 (78.9%)	133 (91.1%)	0.086
Asthma	76 (24.3%)	41 (28.1%)	0.960
Lung cancer	8 (2.6%)	10 (6.8%)	0.576
HTN	182 (58.1%)	107 (73.3%)	0.093
T2DM	148 (47.3%)	76 (52.1%)	0.960
CKD	111 (35.5%)	73 (50%)	0.106
CAD	102 (32.6%)	58 (39.7%)	0.848
HF	21 (6.7%)	18 (12.3%)	0.676
SABA	191 (61%)	94 (64.4%)	0.987
SAMA	18 (5.8%)	15 (10.3%)	0.828
LABA	188 (60.1%)	91 (62.3%)	0.987
LAMA	147 (47%)	64 (43.8%)	0.987
ICS	170 (54.3%)	82 (56.2%)	0.987

*Indicates p-value < 0.05. ^BMI and lung function data was available for 169 patients ($n = 129$ and 40 surviving and deceased, respectively), and smoking data available for 150 patients ($n = 113$ and 37 surviving and deceased, respectively). BMI, body mass index; FVC, forced vital capacity; FEV$_1$, forced expiratory volume in 1 s; COPD, chronic obstructive pulmonary disease; HTN, hypertension; T2DM, type 2 diabetes mellitus; CKD, chronic kidney disease; CAD, coronary artery disease; HF, heart failure; SABA, short acting beta antagonist; SAMA, short acting muscarinic agent; LABA, long acting beta antagonist; LAMA, long acting muscarinic agent; ICS, Inhaled corticosteroids; RW-p, Romano-Wolf adjusted p-value.

TABLE 2 Radiological extent of bronchiectasis between surviving and deceased patients.

Location of bronchiectasis on chest CT scan	Surviving ($n = 313$)	Deceased ($n = 146$)	RW-p
RLL	185 (59.1%)	100 (68.5%)	0.305
RML	136 (43.5%)	60 (41.1%)	0.934
RUL	79 (25.2%)	47 (32.2%)	0.464
LLL	231 (73.8%)	106 (72.6%)	0.954
Lingula	89 (28.4%)	41 (28.1%)	0.954
LUL	60 (19.2%)	40 (27.4%)	0.305
Bilateral	225 (71.9%)	114 (78.1%)	0.490
Median number of lobes affected	2 (1, 3)	2 (2, 3)	1.000
$\geq$3 lobes effected	110 (35.1%)	58 (39.7%)	0.755

CT, computed tomography; RLL, right lower lobe; RML, right middle lobe; RUL, right upper lobe; LLL, left lower lobe; LUL, left upper lobe; RW-p, Romano-Wolff adjusted p-value.

TABLE 3 Sputum cultures for surviving and deceased patients.

Sputum microbiology	Surviving ($n = 283$)	Deceased ($n = 142$)	RW-p
Haemophilus	183 (64.7%)	82 (57.7%)	0.593
non-*Aspergillus fungi*	131 (46.3%)	92 (64.8%)	0.007*
Streptococcus	96 (33.9%)	54 (38%)	0.893
Pseudomonas	78 (27.6%)	65 (45.8%)	0.007*
Staphylococcus	36 (12.7%)	28 (19.7%)	0.286
Mycobacterium	35 (12.4%)	18 (12.7%)	0.971
Aspergillus	21 (7.4%)	16 (11.3%)	0.600
Klebsiella	18 (6.4%)	11 (7.7%)	0.936
Burkholderia	13 (4.6%)	7 (4.9%)	0.971
Other	280 (98.9%)	138 (97.2%)	0.600

*Indicates p-value < 0.05. RW-p, Romano-Wolff adjusted p-value. A total of 34 patients did not have sputum data available and were excluded from this table.

surviving cohort compared to the deceased (28 vs. 46%, $p = 0.007$) (Table 3).

3.4 Hospitalisations and mortality data

The majority of patients recorded at least one respiratory condition related hospitalisation during the study period, though a lesser proportion of those surviving did so (83%) compared to those deceased (95%) (Table 4). Patients who survived spent significantly less total time in hospital during the study window, with a median 12 days (IQR 3, 34) compared to those deceased with a median 34 days (IQR 12, 69) ($p = 0.027$). Half as many surviving patients recorded time in the ICU (22 vs. 42%, $p = 0.066$) or time on mechanical ventilation (6 vs. 12%, $p = 0.431$).

3.5 Regression analysis

In the Cox regression model (425 patients included), older age, presence of *Pseudomonas* and recorded ICU visits were associated with increased odds of mortality while cultured *Haemophilus* was associated with reduced odds of mortality (Table 5). In multivariate analysis use of LABA and LAMA were associated with reduced HR for morality, but this was attenuated with multiple hypothesis adjustment. In quantile regression (142 patients included), older age at the start of the study period and LABA prescription were associated with significantly older age at death, though the statistical significance associated with LABA prescription was attenuated following Romano-Wolf adjustment. Cox regression models for patients with lung function data, BMI and smoking data are displayed in **Supplementary material 2**. In the univariate cox regression model increasing FEV_1 [HR 0.96 (95% CI 0.94, 0.99), $p = 0.024$] and BMI were associated with reduced odds of mortality [HR 0.92 (95% CI 0.87, 0.98), $p = 0.051$], however, in the multivariate model ($n = 142$) only BMI showed a statistically significant association with mortality [HR 0.88 (95% CI 0.82, 0.954), $p = 0.017$].

TABLE 4 Hospitalisation information of surviving and deceased patients.

Hospitalisation data	Surviving ($n = 313$)	Deceased ($n = 146$)	RW-p
Any hospitalisations	260 (83.1%)	138 (94.5%)	0.119
Median number of hospitalisations	3 (1, 8)	5 (2, 12)	0.431
Total time in hospital (days)	12 (3, 34)	34 (12, 69)	0.027*
Any ICU	58 (22.3%)	58 (42%)	0.066
Total time in ICU (hours)	112.5 (64, 204)	143.5 (73, 302)	0.735
Any mechanical ventilation	16 (6.2%)	16 (11.6%)	0.431
Total time on ventilation (hours)	48.5 (30, 125)	58 (30.5, 243.5)	0.808

*Indicates p-value < 0.05. ICU, intensive care unit; RW-p, Romano-Wolff adjusted p-value.

4 Discussion

To the best of the authors knowledge, this is the first study to demonstrate potential clinical parameters that may influence survival or future mortality amongst an adult Aboriginal Australian population, particularly among those residing in the Top End, NT of Australia. This study has illustrated that increasing age, prior need for ICU admissions, and prior positive sputum microbiology demonstrating *Pseudomonas may* increase the risk of mortality. On the other hand, a higher BMI, better lung function on spirometry and prior positive sputum microbiology demonstrating *Haemophilus* may be associated with a favourable outcome.

Across various ethnically diverse non-Indigenous populations, hospitalisation and mortality data have been well documented in the literature (3–9, 21–23, 28–31). Although the available reports suggest that the burden of chronic respiratory disorders is much higher among global First Nations Indigenous people (including higher heath care utilisation and hospital admission rates), there is scant evidence pertaining to factors that may predict mortality or survival among First Nations Indigenous people (20, 32–38).

In comparison to international bronchiectasis cohorts, the current study cohort showed some unique clinical features. Most notably, the median age of death was 59.2 years, whereas in international data most deaths are among those aged >70 years (3). However, similar to as has been reported previously (3, 22, 39), we also noted a lower BMI in the deceased group at a median 22 kg/m^2. The reason for a lower BMI being associated

TABLE 5 Univariate and multivariate Cox regression models for mortality reporting HRs (95% CI) and quantile regression models for age of death reporting betas (95% CI) for parameters included from stepwise regression.

Univariate results	Cox regression for mortality	RW-p	Quantile regression for age of death	RW-p
Age	1.04 (1.03, 1.06)	0.001*	1.00 (0.95, 1.04)	0.001*
COPD	2.3 (1.3, 4.07)	0.016*	–	–
Haemophilus	0.8 (0.58, 1.12)	0.479	–	–
non-*Aspergillus fungi*	1.84 (1.31, 2.6)	0.001*	–	–
Pseudomonas	1.94 (1.4, 2.71)	0.001*	−0.49 (−6.89, 5.91)	0.912
Staphylococcus	1.56 (1.03, 2.36)	0.143	–	–
ICU	2.28 (1.64, 3.18)	0.001*	–	–
LABA	1.04 (0.75, 1.46)	0.802	2.91 (−3.35, 9.16)	0.581
LAMA	0.89 (0.64, 1.23)	0.688	–	–
Multivariate results	**Cox regression for mortality—($n = 425$)**	**RW-p**	**Quantile regression for age of death—($n = 142$) Pseudo R^2 0.824**	**RW-p**
Age	1.04 (1.02, 1.06)	0.001*	1.00 (0.96, 1.05)	0.001*
COPD	2.17 (1.13, 4.16)	0.107	–	–
Haemophilus	0.6 (0.41, 0.86)	0.037*	–	–
non-*Aspergillus fungi*	1.4 (0.96, 2.03)	−0.128	–	–
Pseudomonas	1.71 (1.2, 2.44)	0.027*	−1.00 (−2.05, 0.05)	0.202
Staphylococcus	1.64 (1.07, 2.53)	0.107	–	–
ICU	2.28 (1.58, 3.31)	0.001*	–	–
LABA	0.68 (0.47, 1)	0.128	1.72 (0.64, 2.8)	0.108
LAMA	0.67 (0.47, 0.96)	0.107	–	–

The Cox regression model included sputum cultures which were not available for 34 patients, therefore 425/459 patients were included for univariate modelling of these parameters, and in the multivariate model. *Indicates Romano-Wolf adjusted p-value < 0.05. COPD, Chronic obstructive pulmonary disease; ICU, intensive care unit; LABA, long acting beta antagonist; LAMA, long acting muscarinic agent; RW-p, Romano-Wolff adjusted p-value.

with mortality is unclear. It is reasonable to speculate that the resting energy expenditure and higher basal metabolic rate due to chronic respiratory tract infection, alongside chronic airway inflammation may be contributing to a lower BMI among patients with bronchiectasis as observed among patients with COPD (40, 41).

Among patients with bronchiectasis, multimorbidity, and especially respiratory comorbidities have been associated with significantly greater morbidity and mortality (4–6). Coexisting COPD in particular is shown to have a much worse prognosis (42). Among our cohort there was a significant prevalence of COPD, particularly among the deceased cohort at 91%, however, 79% of surviving patients were also noted to have COPD. This significant presence of disease amongst both surviving and deceased cohorts limits its ability to be used as a prognostic marker as it is in other populations.

In relation to laboratory parameters, lower lung function values, presence of *pseudomonas* on sputum microbiology and CT evidence of multi-lobar involvement are shown to be associated with poorer long-term prognosis among patients with bronchiectasis (3–9, 21). Among the deceased cohort in our study, lung function values were reduced (medians for FVC 45 vs. 53%, FEV_1 29 vs. 40%, FEV_1/FVC ratio 57 vs. 66 deceased vs. surviving, respectively) though this did not reach statistical significance. Although, this may not be statistically significant, it may have clinical relevance. Similar to what is observed in previous reports (6, 43), *Pseudomonas* was cultured more commonly among deceased patients than those surviving (45.8 vs. 27.6%). However, in our study in addition to *pseudomonas,* presence of non-*Aspergillus* fungi on sputum microbiology was significantly more common among deceased patients (64.8 vs. 46.3%). Further prospective studies may be useful to explore microbiology data in this population. The effects of therapeutic interventions, such as inhaled antibiotics (44) on survival and mortality has not been widely explored in the adult Aboriginal Australian patients with bronchiectasis. Further studies may be useful to investigate the beneficial effects of therapeutic modalities such as inhaled antibiotics, including N- Acetylcysteine in Indigenous patients (45). Use of ICS among patients with bronchiectasis is controversial (46, 47). Nonetheless, in our study more than half of the patients recorded an ICS prescription. ICS did not show any positive or negative association with mortality in our study. LABA and LAMA prescription however, was associated with reduced odds of mortality, and LABA prescription with an older age at death in multivariate regressions, though statistical significance of both was attenuated on multiple hypotheses adjustment. Nevertheless, in view of previous studies demonstrating positive impact on the use of LAMA among patients with bronchiectasis (48, 49), it is reasonable to assume that use of LAMA may have some favourable impact on survival in our study patients.

Among Aboriginal Australian patients, hospital admission rates secondary to respiratory conditions are reported to be higher than among non-Aboriginal Australian patients (50). In our study we noted that among the deceased cohort there was a significantly longer median time spent in hospital (34 compared to 12 days). Furthermore, a greater proportion among the deceased cohort required ICU care at some point (42 vs. 22%). ICU visits were one of the few parameters to retain a statistically significant association with increased mortality following multiple hypothesis correction, indicating that this factor should be considered as a marker for poor long-term prognosis among Aboriginal patients with bronchiectasis.

This study has demonstrated that several classic clinical parameters used for predicting survival, exacerbations/hospital admissions or mortality amongst adults with bronchiectasis may not be as applicable to Aboriginal Australians as in other non-Indigenous population (51–53). This is due to a significant background presence of comorbidities, high smoking rates, reduced lung function parameters, significantly younger age and generally lower BMI within this cohort. Given the disparity noted in some of the clinical parameters in comparison to other ethnic non-Indigenous patients as noted in this study and in general (54–69), moving forwards, it may be time to invest in establishing Indigenous specific bronchiectasis assessment and severity classification tool. This will be very valuable to identify high risk adult Aboriginal patients with bronchiectasis in order to guide clinical decision making and early interventions to reduce overall morbidity and mortality.

5 Limitation

This study's outcomes pertain to Aboriginal Australian people residing in the TEHS region of the NT of Australia and the results represented in this study cannot be generalised to the wider Aboriginal populations in Australia or for Indigenous people globally. Spirometry data and BMI values were not available for all patients, hence would have introduced a bias in the outcome observed. We also did not have data to represent therapeutic interventions other than inhaled pharmacotherapy use and moreover, we did not have dates of prescription of inhaled pharmacotherapy, nor to assess dosages or adherence to medication which would be significant confounders. Furthermore, with a high presence of COPD, CAD, and CKD in this population, which was observed among both deceased and surviving cohorts, it is uncertain if bronchiectasis is a primary or a secondary cause contributing to mortality. Nonetheless, this is the first study to assess factors predicting survival and mortality in an Aboriginal population, which could be useful to compare for any future similar studies in other Indigenous global populations.

6 Conclusion

Among adult Aboriginal Australian patients diagnosed to have bronchiectasis, advancing age, cultured *Pseudomonas* and prior ICU visits are strongly associated with mortality. Higher BMI, better lung function parameters, prior positive sputum microbiology for *Haemophilus* and use of LABA and LAMA may have a favourable effect. However, further studies are warranted in larger cohorts to determine if these findings are replicable. Furthermore, it is clear that efforts must be made to establish bronchiectasis severity assessment tools specific to Indigenous people to stratify high risk patients, so that interventions can be implemented to prevent mortality.

Data availability statement

The original contributions presented in the study are included in the article/**Supplementary material**, further inquiries can be directed to the corresponding author.

Ethics statement

The studies involving humans were approved by the Human Research Ethics Governance/Committee of the TEHS, NT and Menzies School of Health Research (Reference: HREC; 2019-3547). The studies were conducted in accordance with the local legislation and institutional requirements. The Ethics Committee/Institutional Review Board waived the requirement of written informed consent for participation from the participants or the participants' legal guardians/next of kin because Due to this research being a retrospective study–informed consent was waived by the Ethics Committee.

Author contributions

SH: Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – original draft, Writing – review and editing. CG: Conceptualization, Data curation, Investigation, Methodology, Resources, Software, Validation, Visualization, Writing – original draft, Writing – review and editing. SR: Conceptualization, Data curation, Investigation, Resources, Visualization, Writing – review and editing. DE: Conceptualization, Data curation, Investigation, Resources, Validation, Visualization, Writing – review and editing. AA: Conceptualization, Data curation, Investigation, Methodology, Resources, Software, Validation, Visualization, Writing – original draft, Writing – review and editing. TH: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – original draft, Writing – review and editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fmed.2024.1366037/full#supplementary-material

References

1. King PT. The pathophysiology of bronchiectasis. *Int J Chron Obstruct Pulmon Dis.* (2009) 4:411–9.

2. Smith MP. Diagnosis and management of bronchiectasis. *CMAJ.* (2017) 189:E828–35. doi: 10.1503/cmaj.160830

3. Mateus SP, Ribeiro-Alves M, Salles REB, Costa W, Costa CHD, Lopes AJ, et al. Mortality and comorbidities in patients with bronchiectasis over a 3-year follow-up. *Medicine (Baltimore).* (2022) 101:e32537. doi: 10.1097/MD.0000000000032537

4. Choi H, Yang B, Kim YJ, Sin S, Jo YS, Kim Y, et al. Increased mortality in patients with non-cystic fibrosis bronchiectasis with respiratory comorbidities. *Sci Rep.* (2021) 11:7126. doi: 10.1038/s41598-021-86407-8

5. Sin S, Yun SY, Kim JM, Park CM, Cho J, Choi SM, et al. Mortality risk and causes of death in patients with non-cystic fibrosis bronchiectasis. *Respir Res.* (2019) 20:271. doi: 10.1186/s12931-019-1243-3

6. Goeminne PC, Nawrot TS, Ruttens D, Seys S, Dupont LJ. Mortality in non-cystic fibrosis bronchiectasis: A prospective cohort analysis. *Respir Med.* (2014) 108:287–96.

7. Sciosciaa G, Alcaraz-Serranob V, Méndezc R, Gabarrúsd A, Fernández-Baratd L, Menéndezc R, et al. Factors associated with one year mortality in hospitalised patients with exacerbated bronchiectasis. *Arch Bronconeumol.* (2022) 58:773–7.

8. Biatobock DR, Paz M, Olmedo DWV, Barlem ELD, Ramos DF. Bronchiectasis: Morbidity and mortality in Brazil and its impact on hospitalization rates. *Rev Soc Cient Parag.* (2022) 27:61–73.

9. McDonnell MJ, Aliberti S, Goeminne PC, Restrepo MI, Finch S, Pesci A, et al. Comorbidities and the risk of mortality in patients with bronchiectasis: An international multicentre cohort study. *Lancet Respir Med.* (2016) 4:969–79. doi: 10.1016/S2213-2600(16)30320-4

10. Australian Bureau of Statistics. *Estimates of aboriginal and Torres Strait islander Australians*. Canberra, ACT: Australian Bureau of Statistics (2016).

11. McTaggart D. An overview of respiratory disease in indigenous communities: A comparison to the wider Australian population. *Med Stud J Aust.* (2010) 1:42–4.

12. Kruavit A, Fox M, Pearson R, Heraganahally S. Chronic respiratory disease in the regional and remote population of the northern territory top end: A perspective from the specialist respiratory outreach service. *Aust J Rural Health.* (2017) 25:275–84.

13. Howarth TP, Jersmann HPA, Majoni SW, Mo L, Ben Saad H, Ford LP, et al. The "ABC" of respiratory disorders among adult indigenous people – asthma, bronchiectasis and COPD among aboriginal Australians – a systematic review. *BMJ Open Resp Res.* (2023) 10:e001738. doi: 10.1136/bmjresp-2023-001738

14. Howarth T, Heraganahally SS, Heraganahally SS. Bronchiectasis among adult first Nations indigenous people – a scoping review. *Curr Respir Med Rev.* (2023) 19:36–51.

15. Heraganahally SS, Ghimire RH, Howarth T, Kankanamalage OM, Palmer D, Falhammar H. Comparison and outcomes of emergency department presentations with respiratory disorders among Australian indigenous and non-indigenous patients. *BMC Emerg Med.* (2022) 22:11. doi: 10.1186/s12873-022-00570-3

16. Pal A, Howarth TP, Rissel C, Messenger RL, Issac S, Connors C, et al. COPD disease knowledge, self-awareness and reasons for hospital presentations among a predominately Indigenous Australian cohort – a study to explore preventable hospitalization. *BMJ Open Res Res.* (2022) 9:e001295. doi: 10.1136/bmjresp-2022-001295

17. Heraganahally SS, Wasgewatta SL, McNamara K, Eisemberg CC, Budd RC, Mehra S, et al. Chronic obstructive pulmonary disease in aboriginal patients of the northern territory of Australia: A landscape perspective. *Int J Chron Obstruct Pulmon Dis.* (2019) 14:2205–17.

18. Mehra S, Chang AB, Lam CK, Campbell S, Mingi JJ, Thomas I, et al. Bronchiectasis among Australian aboriginal and non-aboriginal patients in the regional and remote population of the northern territory of Australia. *Rural Remote Health.* (2021) 21:6390. doi: 10.22605/RRH6390

19. Heraganahally SS, Wasgewatta SL, McNamara K, Mingi JJ, Mehra S, Eisemberg CC, et al. Chronic obstructive pulmonary disease with and without bronchiectasis in Aboriginal Australians – a comparative study. *Int Med J.* (2020) 50:1505–13. doi: 10.1111/imj.14718

20. Blackall SR, Hong JB, King P, Wong C, Einsiedel L, Rémond MGW, et al. Bronchiectasis in indigenous and non-indigenous residents of Australia and New Zealand. *Respirology.* (2018) 23:743–9. doi: 10.1111/resp.13280

21. Loebinger MR, Wells AU, Hansell DM, Chinyanganya N, Devaraj A, Meister M, et al. Mortality in bronchiectasis: A long-term study assessing the factors influencing survival. *Eur Respir J.* (2009) 34:843–9. doi: 10.1183/09031936.00003709

22. Lee JM, Lee SA, Han CH, Lee SM, Kim CJ, Lee SC, et al. Body mass index as a predictor of mortality in bronchiectasis: A nationwide population-based study. *Respir Med.* (2021) 180:106370. doi: 10.1016/j.rmed.2021.106370

23. Huang H-Y, Chung F-T, Lin C-Y, Lo C-Y, Huang Y-T, Huang Y-C, et al. Influence of comorbidities and airway clearance on mortality and outcomes of patients with severe bronchiectasis exacerbations in Taiwan. *Front Med.* (2022) 8:812775. doi: 10.3389/fmed.2021.812775

24. Howarth T, Gahreman D, Ben Saad H, Ng L, Heraganahally SS. Correlation of spirometry indices to chest radiology in the diagnosis of chronic airway disease among regional and rural Indigenous Australians. *Int Med J.* (2023) 53:1994–2006. doi: 10.1111/imj.16023

25. Heraganahally SS, Howarth T, Mo L, Sorger L, Ben Saad H. Critical analysis of spirometric patterns in correlation to chest computed tomography among adult Indigenous Australians with chronic airway diseases. *Expert Rev Respir Med.* (2021) 15:1229–38. doi: 10.1080/17476348.2021.1928496

26. National Health and Medical Research Council. *Ethical conduct in research with aboriginal and Torres Strait islander peoples and communities: Guidelines for researchers and stakeholders*. Canberra, ACT: Commonwealth of Australia (2018).

27. Gibbs C, Howarth T, Chen W, Ford PL, Abeyaratne A, et al. Bronchiectasis among indigenous adults in the top end of the northern territory, 2011-2020: A retrospective cohort study. *Med J Aust.* (2024) 220:188–95. doi: 10.5694/mja2.52204

28. Phua HP, Lim WY, Ganesan G, Yoong J, Tan KB, Abisheganaden JA, et al. Epidemiology and economic burden of bronchiectasis requiring hospitalisation in Singapore. *ERJ Open Res.* (2021) 7:00334–2021. doi: 10.1183/23120541.00334-2021

29. Sánchez-Muñoz G, López de Andrés A, Jiménez-García R, Carrasco-Garrido P, Hernández-Barrera V, Pedraza-Serrano F, et al. Time trends in hospital admissions for bronchiectasis: Analysis of the Spanish national hospital discharge data (2004 to 2013). *PLoS One.* (2016) 11:e0162282. doi: 10.1371/journal.pone.0162282

30. Dhar R, Singh S, Talwar D, Murali Mohan BV, Tripathi SK, Swarnakar R, et al. Clinical outcomes of bronchiectasis in India: Data from the EMBARC/respiratory research network of India registry. *Eur Respir J.* (2023) 61:2200611. doi: 10.1183/13993003.00611-2022

31. Ringshausen FC, de Roux A, Pletz MW, Hämäläinen N, Welte T, Rademacher J. Bronchiectasis-associated hospitalizations in Germany, 2005–2011: A population-based study of disease burden and trends. *PLoS One.* (2013) 8:e71109. doi: 10.1371/journal.pone.0071109

32. Roberts ME, Lowndes L, Milne DG, Wong CA. Socioeconomic deprivation, readmissions, mortality and acute exacerbations of bronchiectasis. *Intern Med J.* (2012) 42:e129–36.

33. Bibby S, Milne R, Beasley R. Hospital admissions for non-cystic fibrosis bronchiectasis in New Zealand. *N Z Med J.* (2015) 128:30–8.

34. de Boer S, Lewis CA, Fergusson W, Ellyett K, Wilsher ML. Ethnicity, socioeconomic status and the severity and course of non-cystic fibrosis bronchiectasis. *Intern Med J.* (2018) 48:845–50.

35. Einsiedel L, Pham H, Au V, Hatami S, Wilson K, Spelman T, et al. Predictors of non-cystic fibrosis bronchiectasis in Indigenous adult residents of central Australia: Results of a case–control study. *ERJ Open Res.* (2019) 5:00001–2019.

36. Steinfort DP, Brady S, Weisinger HS, Einsiedel L. Bronchiectasis in Central Australia: A young face to an old disease. *Respir Med.* (2008) 102:574–8.

37. Barton J, Scott L, Maguire G. Bronchiectasis in the Kimberley region of Western Australia. *Aust J Rural Health.* (2018) 26:238–44.

38. Vigneault LP, Diendere E, Sohier-Poirier C, Hanna MA, Poirier A, St-Onge M. Acute health care among indigenous patients in Canada: A scoping review. *Int J Circumpol Health.* (2021) 80:1. doi: 10.1080/22423982.2021.1946324

39. Onen ZP, Gulbay BE, Sen E, Yildiz OA, Saryal S, Acican T, et al. Analysis of the factors related to mortality in patients with bronchiectasis. *Respir Med.* (2007) 101:1390–7. doi: 10.1016/j.rmed.2007.02.002

40. Wilson DO, Donahoe M, Rogers RM, Pennock BE. Metabolic rate and weight loss in chronic obstructive lung disease. *J Parenter Enteral Nutr.* (1990) 14:7–11. doi: 10.1177/014860719001400107

41. Agha MA, El Wahsh RA. Basal metabolic rate in bronchial asthma and chronic obstructive pulmonary disease patients. *Egypt J Chest Dis Tuberc.* (2013) 62:39–44.

42. Mao B, Lu HW, Li MH, Fan LC, Yang JW, Miao XY, et al. The existence of bronchiectasis predicts worse prognosis in patients with COPD. *Sci Rep.* (2015) 5:10961. doi: 10.1038/srep10961

43. Venning V, Bartlett J, Jayaram L. Patients hospitalized with an infective exacerbation of bronchiectasis unrelated to cystic fibrosis: Clinical, physiological and sputum characteristics. *Respirology.* (2017) 22:922–7. doi: 10.1111/resp.13005

44. Karampitsakos T, Papaioannou O, Kaponi M, Kozanidou A, Hillas G, Stavropoulou E, et al. Low penetrance of antibiotics in the epithelial lining fluid. The role of inhaled antibiotics in patients with bronchiectasis. *Pulm Pharmacol Ther.* (2020) 60:101885. doi: 10.1016/j.pupt.2019.101885

45. Jayaram L, King PT, Hunt J, Lim M, Park C, Hu E, et al. Evaluation of high dose N– acetylcysteine on airway inflammation and quality of life outcomes in adults with bronchiectasis: A randomised placebo-controlled pilot study. *Pulm Pharmacol Ther.* (2023) 84:102283. doi: 10.1016/j.pupt.2023.102283

46. Martínez-García MÁ, Oscullo G, García-Ortega A, Matera MG, Rogliani P, Cazzola M. Inhaled corticosteroids in adults with non-cystic fibrosis bronchiectasis: From bench to bedside. A narrative review. *Drugs.* (2022) 82:1453–68. doi: 10.1007/s40265-022-01785-1

47. Håkansson KEJ, Fjaellegaard K, Browatzki A, Dönmez Sin M, Ulrik CS. Inhaled corticosteroid therapy in bronchiectasis is associated with all-cause mortality: A prospective cohort study. *Int J Chron Obstruct Pulmon Dis.* (2021) 16:2119–27. doi: 10.2147/COPD.S311236

48. Lee SY, Lee JS, Lee SW, Oh YM. Effects of treatment with long-acting muscarinic antagonists (LAMA) and long-acting beta-agonists (LABA) on lung function improvement in patients with bronchiectasis: An observational study. *J Thorac Dis.* (2021) 13:169–77. doi: 10.21037/jtd-20-1282

49. Jayaram L, Vandal AC, Chang CL, Lewis C, Tong C, Tuffery C, et al. Tiotropium treatment for bronchiectasis: A randomised, placebo-controlled, crossover trial. *Eur Respir J.* (2022) 59:2102184. doi: 10.1183/13993003.02184-2021

50. Williams P, Gracey M, Smith P. Hospitalization of aboriginal and non-aboriginal patients for respiratory tract diseases in Western Australia, 1988-1993. *Int J Epidemiol.* (1997) 26:797–805. doi: 10.1093/ije/26.4.797

51. Martínez-García MÁ, de Gracia J, Vendrell Relat M, Girón RM, Máiz Carro L, de la Rosa Carrillo D, et al. Multidimensional approach to non-cystic fibrosis bronchiectasis: The FACED score. *Eur Respir J.* (2014) 43:1357–67. doi: 10.1183/09031936.00026313

52. Chalmers JD, Goeminne P, Aliberti S, McDonnell MJ, Lonni S, Davidson J, et al. The bronchiectasis severity index. An international derivation and validation study. *Am J Respir Crit Care Med.* (2014) 189:576–85. doi: 10.1164/rccm.201309-1575OC

53. Nicolson CH, Holland AE, Lee AL. The bronchiectasis toolbox-A comprehensive website for the management of people with bronchiectasis. *Med Sci (Basel).* (2017) 5:13. doi: 10.3390/medsci5020013

54. Heraganahally SS, Timothy TP, Sorger L. Chest computed tomography findings among adult Indigenous Australians in the northern territory of Australia. *J Med Imaging Radiat Oncol.* (2022) 66:337–44. doi: 10.1111/1754-9485.13295

55. Heraganahally SS, Howarth TP, Lloyd A, White E, Veale A, Ben Saad H. The prevalence of bronchodilator responsiveness "asthma" among adult indigenous Australians referred for lung function testing in the top end northern territory of Australia. *J Asthma Allergy.* (2022) 15:1305–19.

56. Heraganahally SS, Silva SAMS, Howarth TP, Kangaharan N, Majoni SW. Comparison of clinical manifestation among Australian Indigenous and non–indigenous patients presenting with pleural effusion. *Int Med J.* (2022) 52:1232–41. doi: 10.1111/imj.15310

57. Seyedshahabedin MM, Howarth TP, Mo L, Biancardi E, Heraganahally SS. Flexible bronchoscopy indications and outcomes between indigenous and non-indigenous patients in the northern territory of Australia. *Int Med J.* (2022) 53:1634–41. doi: 10.1111/imj.15865

58. Sze DFL, Howarth TP, Lake CD, Ben Saad H, Heraganahally SS. Differences in the spirometry parameters between indigenous and non-indigenous patients with COPD: A matched control study. *Int J Chron Obstruct Pulmon Dis.* (2022) 17:869–81.

59. Howarth TP, Saad HB, Perez AJ, Atos CB, White E, Heraganahally SS. Comparison of diffusing capacity of carbon monoxide (DLCO) and total lung capacity (TLC) between Indigenous Australians and Australian Caucasian adults. *PLoS One.* (2021) 16:e0248900. doi: 10.1371/ journal.pone.0248900

60. Heraganahally SS, Kruavit A, Oguoma VM, Gokula C, Mehra S, Judge D, et al. Sleep apnoea among Australian aboriginal and non– aboriginal patients in the northern territory of Australia– a comparative study. *Sleep.* (2020) 43:zsz248. doi: 10.1093/sleep/zsz248

61. Doss AX, Howarth TP, Ng L, Doss SA, Heraganahally SS. Significance and prognostication of mediastinal lymph node enlargement on chest computed tomography among adult indigenous Australians. *J Med Imaging Radiat Oncol.* (2023) 67:726–33. doi: 10.1111/1754-9485.13569

62. Mishra K, Fazal R, Howarth T, Mutai J, Doss AX, Heraganahally SS. Cystic lung disease in adult Indigenous Australians in the northern territory of Australia. *J Med Imaging Radiat Oncol.* (2023) 68:67–73. doi: 10.1111/1754-9485.13593

63. Heraganahally S, Howarth TP, White E, Ben Saad H. Implications of using the GLI-2012, GOLD and Australian COPD-X recommendations in assessing the severity of airflow limitation on spirometry among an Indigenous population with COPD: An indigenous Australians perspective study. *BMJ Open Respir Res.* (2021) 8:e001135. doi: 10.1136/bmjresp-2021-001135

64. Heraganahally SS, Howarth T, Sorger L, Ben Saad H. Sex differences in pulmonary function parameters among Indigenous Australians with and without chronic airway disease. *PLoS One.* (2022) 17:e0263744. doi: 10.1371/journal.pone.0263744

65. Heraganahally SS, Howarth T, White E, Sorger L, Biancardi E, Ben Saad H. Lung function parameters among Australian aboriginal "apparently healthy" adults: An Australian Caucasian and global lung function initiative (GLI-2012) various ethnic norms comparative study. *Expert Rev Respir Med.* (2020) 23:1–11. doi: 10.1080/17476348.2021.1847649

66. Heraganahally SS, Monsi E, Gadil E, Maze D, Lynch S. Catastrophic effects of using cannabis via bucket bong in top end northern territory of Australia. *Am J Trop Med Hyg.* (2023) 55:1–6. doi: 10.4269/ajtmh.23-0393

67. Heraganahally S, Howarth TP, Issac S, Lloyd A, Ravichandran SJ, Abeyaratne A, et al. Exploring the appropriateness of prescribing practice of inhaled pharmacotherapy among aboriginal Australians in the top end northern territory of Australia: A retrospective cohort study. *BMJ Open Resp Res.* (2023) 10:e001508. doi: 10.1136/bmjresp-2022-001508

68. Heraganahally SS, Ponneri TR, Howarth TP, Saad HB. The effects of inhaled airway directed pharmacotherapy on decline in lung function parameters among indigenous Australian adults with and without underlying airway disease. *Int J Chron Obstruct Pulmon Dis.* (2021) 16:2707–20.

69. Heraganahally SS, Mortimer N, Howarth T, Messenger R, Issac S, Thomas I, et al. Utility and outcomes among indigenous and non-indigenous patients requiring domiciliary oxygen therapy in the regional and rural Australian population. *Aust J Rural Health.* (2021) 29:918–26. doi: 10.1111/ajr.12782

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Addressing the burden of cervical cancer for Indigenous women in Latin America and the Caribbean: a call for action

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Cervical cancer, primarily caused by human papillomavirus (HPV) infection, poses a significant global health challenge. Due to higher levels of poverty and health inequities, Indigenous women worldwide are more vulnerable to cervical cancer than their non-Indigenous counterparts. However, despite constituting nearly 10% of the population in Latin America and the Caribbean (LAC), the true extent of the burden of cervical cancer among Indigenous people in this region remains largely unknown. This article reviews the available information on cervical cancer incidence and mortality, as well as HPV infection prevalence, among Indigenous women in LAC. The limited existing data suggest that Indigenous women in this region face a heightened risk of cervical cancer incidence and mortality compared to non-Indigenous women. Nevertheless, a substantial knowledge gap persists that must be addressed to comprehensively assess the burden of cervical cancer among Indigenous populations, especially through enhancing cancer surveillance across LAC countries. Numerous structural, social and cultural barriers hindering Indigenous women's access to HPV vaccination and cervical cancer screening worldwide have been identified and are reviewed in this article. The discussion highlights the critical role of culturally sensitive education, community engagement, and empowerment strategies in overcoming those barriers. Drawing insights from the success of targeted strategies in certain high-income countries, the present article advocates for research, policies and healthcare interventions tailored to the unique context of LAC countries.

KEYWORDS

cervical cancer, HPV, indigenous, Latin America, Caribbean

1 Introduction

Cervical cancer is caused by human papillomavirus (HPV) infection, the most common sexually transmitted viral infection worldwide (1, 2). It has been estimated that, in the United States, more than 80% of sexually active women and men will acquire at least one HPV infection by the age of 45 years (3). In women, 90% of incident genital HPV infections clear within 2 years without any clinical impact (4). However, when persistence occurs, HPV becomes a risk factor for malignant transformation. Out of the more than 200 HPV types identified, 12 high-risk HPVs are responsible for virtually all cases of cervical cancers (5, 6).

Cervical cancer is one of the few cancers preventable through both vaccination and screening. Several prophylactic HPV vaccines are available in most countries and the World Health Organization (WHO) currently recommends one or two-dose vaccination of girls aged 9 to 14 years (7). As various high-risk HPV types are not covered by the vaccines, high-quality screening programs are also paramount to prevent cervical cancer. Screening can detect premalignant and early malignant lesions that can be treated with a very good prognosis (8). Three different methods can be used to identify women who have, or are at risk of, cervical precancerous lesions and early invasive cancer: (1) detection of genital high-risk HPV infection through a DNA-based test on cervical or vaginal samples; (2) microscopic examination of cervical exfoliated cells, known as the Papanicolaou (Pap) cytology test; or (3) visual inspection of the cervix with acetic acid, in low-resource settings (7). In high-income countries, the implementation of screening and treatment programs since the 1950s followed by the introduction of HPV vaccination since 2006 has led to a dramatic reduction in both the prevalence of HPV vaccine-types and closely related HPV types and the incidence and mortality of cervical cancer (9–14). Before the effectiveness of these prevention measures, the WHO launched in 2020 a strategy for the global elimination of cervical cancer, with three main objectives to be reached by 2030: 90% vaccination rates, 70% screening rates and treatment for 90% of the invasive cancers for women in all countries (15).

Unfortunately, challenges persist in low- and middle-income countries in implementing cervical cancer screening programs and ensuring access to HPV vaccines. As a result, these countries currently bear the largest burden of this preventable disease, with cervical cancer incidence and mortality worldwide strongly correlated with country income level, human development index and living standards (16, 17). At present, cervical cancer ranks fourth in cancer incidence and mortality among women globally (18). In 2020 alone, an estimated 604,000 new cases and 342,000 deaths were reported, with over 80% of these cases occurring in low- and-middle-income countries (18, 19). In Latin America and the Caribbean (LAC), most of the countries have implemented cervical screening and HPV vaccination programs, and trends in cervical cancer incidence and mortality have decreased over the past 20 years (20, 21). However, several barriers to screening access and vaccination uptake persist (22, 23), and the age-standardized incidence and mortality rates in LAC in 2018 were 14.6 and 7.1 per 100,000, respectively, ranking second after the African region and slightly higher than the global rates (13.1 and 6.9, respectively) (20). In 2020, cervical cancer remained the leading cause of cancer death in women in Belize, Honduras, Nicaragua, Bolivia and Paraguay (18).

An estimated 58 million Indigenous people lived in Latin America in 2018, accounting for 10% of the total population of the subregion (24). While no formal definition has been adopted in international law, a contemporary and inclusive understanding of "Indigenous peoples" has been developed and includes those who self-identify and are identified as indigenous, exhibit historical continuity with precolonial societies, possess distinct social, economic, or political systems, maintain unique languages, cultures, and beliefs, and constitute nondominant groups of society (25, 26). In most LAC countries, the determination of indigenous status in population censuses relies on self-identification by individuals (26). The distribution of Indigenous people varies widely across LAC, both in terms of absolute numbers and relative proportions (Table 1). Guatemala, Bolivia and Peru have the highest proportions of indigenous population, with 43.6, 41.5 and 26.0%, respectively. Meanwhile, Mexico is the country with the largest number of indigenous individuals, totaling around 25 million.

Despite the inherent cultural, geographic, and genetic diversity among the more than 800 distinct ethnic groups in LAC (57), Indigenous peoples share a common history of colonization and ongoing dispossession of traditional lands, resources and practices (58). Consequently, Indigenous peoples suffer the greatest structural inequalities in Latin America and are over-represented among the socially disadvantaged in almost every country (59). According to estimations from 2020, 45.5% of Indigenous people in Latin America live in poverty and 7.1% in extremely poverty, more than twice the rates for non-Indigenous people in the region (60). Furthermore, in most LAC countries, indigenous people continue to primarily live in rural areas associated with their ancestral territories, and when they live in cities, it is often in extreme poverty in marginal areas, with trouble accessing basic services and decent jobs (59). One consequence of this marginalization is that Indigenous people in LAC have disproportionally worse health than their nonindigenous counterparts (61). Specifically, Indigenous women are directly impacted by the strong association between poverty and cervical cancer.

The primary objective of this perspective article is to examine the existing data on the burden of cervical cancer among Indigenous women in LAC. Moreover, it aims to identify the barriers potentially hindering their access to HPV vaccination, cervical cancer screening and treatment services. Finally, the article explores proven strategies from high-income countries that have successfully addressed similar challenges, with a view toward their potential applicability within LAC nations.

The articles included in this paper were obtained through a systematic literature search conducted on the PubMed Medline database from its inception to February 2024, using the following terms: ("cervical cancer" OR "papillomavirus") AND "indigenous." All abstracts retrieved were reviewed and assessed to identify relevant studies, including peer-reviewed original articles, meta-analyses, reviews, and reports published in English, Spanish or Portuguese. Additional articles were gathered from the references of selected publications. Studies were excluded if they: (1) were unrelated to human papillomavirus infection or cervical cancer; (2) did not clearly define Indigenous people as the study population; (3) were not published in peer-reviewed journals. Furthermore, demographic and epidemiological data specific to each LAC country, including cancer registries and population censuses, were manually searched for on official governmental websites. As a result, findings were categorized and presented through a narrative synthesis in subsequent sections.

2 The burden of cervical cancer among Indigenous women in Latin America and the Caribbean

Because they frequently experience both ethnic and gender discrimination, Indigenous women often face the most profound structural inequalities, especially concerning poverty, education, and healthcare (59). Consequently, they are particularly vulnerable to cervical cancer. Over the last decades, numerous studies have

TABLE 1 Indigenous population, recording of indigenous status by cancer registries, and prevalence of cervical intraepithelial lesions and HPV infection among Indigenous women in Latin America and the Caribbean.

Country	Indigenous population[a] (% of general population)		Cancer registries recording indigenous status[d]	Prevalence of cervical intraepithelial lesions among Indigenous women, sample size n, study population, (study reference)[e]		Prevalence of cervical HPV infection among Indigenous women, sample size n, study population, (study reference)[f]	
South America							
Argentina	1,306,730	(2.9%)	No	4.3% (0.5% CC), n = 207, Guarani 7.9% (0.4% CC), n = 227, Pilaga	(27) (28)	64.3%, n = 207 Guarani 46.7%, n = 227 Pilaga	(27) (28)
Bolivia	4,910,670	(41.5%)	No	ND		ND	
Brazil	1,227,642	(0.6%)	Yes	4.0% (0.9% CC), n = 423 15.3%, n = 72, Parakana 10.7%, n = 84, Panará 5.1% (0.4% CC), n = 275 Yanomami 1.8%, n = 332, Macuxi, Wapishana 8.4%, n = 395, Quilombo 10.0%, n = 241 1.8%*, n = 3,231	(29) (30) (31) (32) (32) (33) (34) (35)	14.3%, n = 42, Parakana 42.9%, n = 49, Parakana 45.9%, n = 305, Yanomami 34.5%, n = 359, Macuxi, Wapishana 12.6%, n = 395, Quilombo	(30) (36) (32) (32) (33)
Chile	2,382,333	(12.4%)	No	ND		ND	
Colombia	2,255,697	(4.4%)	No	ND		63.8%*, n = 47 31.1%, n = 280	(37) (38)
Ecuador	1,301,887	(7.7%)	No	2.0%, n = 100, Kichwa	(39)	34.0%, n = 100, Kichwa 28.3%, n = 396, Kichwa, Shuar	(39) (40)
French Guiana	10000[b]	(3.5%)	No	ND		ND	
Guyana	78,492	(10.5%)	Yes	11.2% (0.8% CC), n = 2,250	(41)	19.3%, n = 1,423	(41)
Paraguay	140,206	(2.3%)	No	13.2%, n = 129, Ache, Ava Guaraní, Mbya Guaraní	(42)	23.2%, n = 181	(43)
Peru	8,673,449	(26.0%)	No	13.4%, n = 307, Shipibo-Konibo 2.1% ǂ, n = 48, Bora	(44) (45)	31.6%, n = 307 Shipibo-Konibo 35.4%(ns), n = 48, Bora	(44) (45)
Suriname	20,344	(3.8%)	No	ND		ND	
Uruguay	83,644	(2.4%)	Yes	ND		ND	
Venezuela	775,034	(2.7%)	No	1.8%, n = 57, Yekwana, Piaroa, Arawaco 14.3%(ns), n = 35, Eñepa 11.0%(ns), n = 82, Piaro	(46) (47) (48)	35.1%, n = 57 Yekwana, Piaroa, Arawaco 45.7%*, n = 35, Eñepa 72.0%(ns), n = 82, Piaro	(46) (47) (48)
North and Central America							
Belize	56,146	(17.4%)	No	ND		ND	
Costa Rica	123,337	(2.4%)	No	ND		ND	
El Salvador	13,037	(0.2%)	No	ND		ND	
Guatemala	7,956,939	(43.6%)	Yes	8.1% (0.5% CC), n = 222	(49)	ND	
Honduras	784,913	(7.8%)	No	ND		ND	
Mexico	25,280,302	(19.4%)	Yes	70.4%, n = 108, Nahuatl	(50)	70.4%, n = 108, Nahuatl	(50)
Nicaragua	422,250	(6.3%)	No	ND		ND	
Panama	698,114	(17.2%)	No	ND		ND	
Caribbean[c]							
British Virgin Islands	177	(0.6%)	Yes	ND		ND	
Dominica	2,576	(3.7%)	No	ND		ND	

(Continued)

TABLE 1 (Continued)

Country	Indigenous population[a] (% of general population)		Cancer registries recording indigenous status[d]	Prevalence of cervical intraepithelial lesions among Indigenous women, sample size n, study population, (study reference)[e]		Prevalence of cervical HPV infection among Indigenous women, sample size n, study population, (study reference)[f]	
St Vincent & the Grenadines	3.280	(3%)	No	ND		ND	

[a]Sources: Economic Commission for Latin America and the Caribbean (ECLAC), The sociodemographic impacts of the COVID-19 pandemic in Latin America and the Caribbean (LC/CRPD.4/3), Santiago, 2022. Argentina data: Instituto Nacional de Estadística y Censos, Censo Nacional de Población, Hogares y Viviendas 2022. Belize data: The Statistical Institute of Belize, Belize Population and Housing Census 2010. Brazil data: Instituto Brasileiro de Geografia e Estatística, Censo 2022. British Virgin Islands data: Government of the Virgin Islands, 2010 Population and Housing Census Report. Dominica data: Central Statistics Office of Dominica, 2011 Population and Housing Census. Ecuador data: Instituto Nacional de Estadística y Censos, Censo Ecuador 2022. Guyana data: Bureau of Statistics, 2012 Census. Panama data: Instituto Nacional de Estadística y Censo, XII Censo Nacional de Población y VIII de Vivienda 2023. Paraguay data: Instituto Nacional de Estadística, Resultados Preliminares del IV Censo Nacional de Población y Viviendas para Pueblos Indígenas 2022. Saint Vincent & the Grenadines data: Statistical Office, Government of Saint Vincent and the Grenadines, Population and Housing Census Report 2012. Suriname data: Algemeen Bureau voor de Statistiek, 2012 Census. [b]Due to the prohibition of ethnic statistics by the French Constitution, only estimates from unofficial studies collected by the Associations for the Defense of Indigenous Peoples of French Guiana are available. [c]Caribbean countries with distinct indigenous communities still in existence. [d]Sources: (26, 51–56). [e]Cervical intraepithelial lesions include: Atypical squamous cell of unknown significance (ASCUS), Low grade squamous cell intraepithelial lesion (LSIL); High grade squamous cell intraepithelial lesion (HSIL) and invasive cervical cancer (CC). Ethnics of the study population specified when available. [f]Prevalence of any-type HPV. *Rate significantly higher than in the non-Indigenous comparison group. (ns) Rate not significantly higher than in the non-Indigenous comparison group. ǂRate significantly lower than in the non-Indigenous comparison group. ND: No data.

extensively reported and characterized the disproportionately high burden of cervical cancer incidence and mortality among Indigenous women compared to non-Indigenous ones in high-income countries, namely Australia, New Zealand, Canada and the United States (62–68). A higher prevalence of HPV infection has also been observed among Indigenous women compared to the general population in these countries (69–72).

On the contrary, in the LAC region, the actual magnitude of the burden of cervical cancer among Indigenous women remains largely unknown and may be substantially under-estimated due to insufficient data collection. To the best of available information, indigenous status is currently recorded in the population-based cancer registries of only six out of the 24 LAC countries with Indigenous population (Table 1) (26, 51–56). However, none of these six countries publicly provide cancer rates for Indigenous people in their registry reports. Two recent studies have estimated cervical cancer mortality among Indigenous women in Brazil, reporting a mean age-standardized mortality rate of 6.7 per 100,000 between 2000 and 2020, the highest among all ethnic groups in the country and corresponding to a significant 80% increase in cervical cancer death risk compared to white women (73, 74). Regarding cervical cancer incidence, a study conducted on the Indigenous population in the State of Acre, located in the Brazilian Western Amazon, reported that, between 2000 and 2012, cervical cancer was the most frequent neoplasm among indigenous women, and cervical cancer incidence was significantly higher compared to the reference population (Standardized Incidence Ratio: 4.49) (75). Similarly, an analysis of the population-based cancer registry of Guyana from 2000 to 2009 highlighted that cervical cancer was significantly more common among Indigenous Amerindian women compared to other ethnic groups (53). Based on the available information, the assessment of cervical cancer risk among Indigenous women in the rest of LAC relies exclusively on studies investigating the prevalence of cervical HPV infection and cervical intraepithelial lesions within Indigenous populations. Table 1 summarizes the findings of these studies. Reported rates of any-type HPV infection among Indigenous women in LAC are notably high, ranging from 12.6 to 72.0%. Importantly, the majority of studies (15/19) identified higher cervical HPV prevalence rates compared to the meta-analysis estimates for the general populations of Brazil and Latin America, respectively 25.41 and 16.1% (76, 77). Furthermore, a wide range of cervical intraepithelial lesions prevalence rates have been reported among Indigenous women across LAC, varying from 1.8 to 15.3%, except for a study in Mexico which documented an exceptionally high prevalence of 70.4% among Nahuatl women (50). Notably, nine out of the 20 identified studies reported a prevalence of cervical lesions exceeding 10%, while the prevalence of cervical lesions in women worldwide generally remains under 10% (78).

In conclusion, the currently available data suggest that Indigenous women in LAC face a heightened risk of cervical cancer incidence and mortality, mirroring trends observed in Indigenous populations in high-income countries and reflecting the health inequities they experience. However, there remains a significant lack of data on cervical cancer trends among Indigenous women in most LAC countries, including countries like Guatemala, Bolivia, and Mexico, which have some of the highest proportions of Indigenous populations in the region. The wide variations observed in the prevalence of HPV infection and cervical abnormalities may reflect disparities in cervical cancer risk linked to the heterogeneity of Indigenous groups across LAC, including differences in geographical location, degree of isolation, and social and sexual behaviors (35). Consequently, further research is imperative to comprehensively assess the burden of cervical cancer among Indigenous women both regionally and within each LAC country.

3 Barriers to cervical cancer screening and HPV vaccination among Indigenous women

Indigenous women worldwide exhibit a higher prevalence of HPV infection as well as increased cervical cancer incidence and mortality rates compared to the general population. This disproportionate burden of cervical cancer in Indigenous women in both high- and low-and-middle-income countries could be explained by substantially lower rates of HPV vaccination (79–81), lower cervical screening coverage and longer time to clinical investigation (67, 82–85) in Indigenous women compared to non-Indigenous women. Numerous multifaceted barriers have been identified that hinder proper access for Indigenous women to HPV vaccination and cervical cancer screening and treatment (Table 2). While many of these obstacles are not exclusive to Indigenous individuals, the intricate interplay of structural, social and cultural barriers in cervical cancer prevention, diagnosis and treatment uniquely challenges Indigenous women, contributing to their disproportionate burden of this disease.

3.1 Structural barriers

Structural impediments play a pivotal role in limiting cervical cancer screening for Indigenous women. Most of them live in rural areas, where distance from health care centers and practitioners, coupled with the lack of personal resources, limit access to appropriate and timely health care, and pose challenges to screening, diagnosis and treatment (82, 88, 97, 103, 107, 110, 113). Furthermore, fragmentation of healthcare systems and economic constraints present additional challenges to the implementation of specific programs for prevention and control of cervical cancer among Indigenous women (23, 89, 90, 103, 106, 107, 110) (Table 2).

3.2 Social determinants

A complex interplay of social determinants, including limited health literacy, low education rates, low socioeconomic status, and isolation, exacerbates barriers to health services. Lack of awareness of cervical cancer and limited understanding of HPV role in its etiology has been identified as one of the main barrier contributing to low screening rates and HPV vaccination coverage among Indigenous women worldwide (see Table 2).

3.3 Cultural factors

Indigenous women often face discrimination and challenges in accessing culturally safe cervical screening and treatment services. Cultural differences and language barriers contribute to communication challenges with health care providers (82, 98, 99, 109, 110, 112). Furthermore, colonization disrupted Indigenous knowledge systems, de-valuing traditional practices and creating historical mistrust impacting their willingness to undergoing gynecological inspections and cervical cytology invasive procedure, as well as to participate in vaccination campaign (81, 114). Thus, mistrust in healthcare systems and HPV vaccines appears to be the first barrier to cervical cancer prevention and control among Indigenous women globally (see Table 2). Finally, community sensitivities regarding sexual health promotion and sexually transmitted diseases were also identified as one of the main limitation to both HPV vaccination and cervical cancer screening.

4 Strategies to improve cervical screening and HPV vaccination uptake for Indigenous women in Latin America and the Caribbean: recommendations for action

Cervical cancer poses a significant public health challenge for Indigenous women globally, demanding targeted strategies in response to the unique cultural and systemic factors hindering cervical screening and HPV vaccination uptake in Indigenous populations. Canada, Australia and New Zealand have implemented specific programs and initiatives of cancer screening and/or HPV vaccination that are showing promising results in reducing HPV-related diseases among Indigenous populations (97, 115–121). While taking example from these successful policies and strategies, it is imperative to devise solutions adapted to the socio-economic realities of LAC countries and the cultural diversity within Indigenous populations residing in the region.

4.1 Cultural sensitivity and education

Works in collaboration within Métis Nation communities in Canada have highlighted the importance of culturally-sensitive educational approaches to promote cervical cancer awareness and vaccination and to address misconceptions (115). Implementing culturally appropriate programs and ensuring that health care practitioners receive adequate support and resources to be able to incorporate cultural components into the delivery of information have been demonstrated to improve screening rates and HPV vaccination uptake among Indigenous women and adolescents in high-income countries (100, 102, 106, 114, 122, 123).

4.2 Community engagement and empowerment

Recognizing the influence of cultural factors on decision-making is crucial. Research from Canada and Australia has demonstrated that indigenous leadership plays a pivotal role in shaping research priorities, providing policy guidance, and developing strategies that are acceptable, appropriate and sustainable for Indigenous communities (64, 107, 116, 124).

Prioritizing collaboration with Indigenous leaders, strengthening intergenerational relationships and ensuring community involvement in program development and delivery are essential components for the success of HPV vaccination and cancer screening initiatives in high-income countries (97, 114). Community and peer support has been shown to positively contribute to overcoming negative attitudes toward cervical screening and vaccination cervical cancer screening barriers in Australia (120, 125), the United States (126) and Peru (127).

Finally, implementing mobile medical clinics to offer essential services has demonstrated its cost-effectiveness in overcoming access barriers for rural populations in low- and middle-income countries like South Africa (128) and Peru (129).

4.3 HPV self-testing implementation

Polymerase chain reaction (PCR)-based testing for high-risk HPVs on cervical or vaginal samples is now the gold standard for cervical cancer prevention (15). This screening method has shown higher sensitivity and negative predictive value compared to the Pap cytology test, thus allowing for larger intervals between screenings and providing greater protection against developing invasive cervical cancers (130–133). Another advantage of HPV

TABLE 2 Main barriers to HPV vaccination and cervical cancer screening in Indigenous population globally.

Country – Study population	Structural barriers			Social barriers		Cultural barriers					Ref.
Participant characteristics, sample size	Remoteness	Individual cost	Resource constraints in healthcare systems[a]	Limited knowledge about HPV, HPV vaccine and/or cervical cancer	Limited health literacy[b]	Sex and cultural taboos[c]	Mistrust in healthcare systems and/or in vaccine[d]	Language	Fear of pain	Culturally inappropriate health education delivery	
Barriers to HPV vaccination											
USA - American Indians (AI)											
M & F, parents/caregivers, 260					X		X				(86)
F, parents/caregivers, 50				X	X		X				(87)
M & F, healthcare providers, 35	X			X		X	X				(88)
M & F, healthcare providers, 31			X	X			X				(89)
M & F, healthcare providers, vaccine eligible individuals (all ages), parents/caregivers, 73			X			X	X				(90)
M & F, vaccine eligible individuals (>18 years), 57				X	X						(91)
F, community members, 194				X							(92)
USA - Alaska Natives (AN)											
F, vaccine eligible individuals (<18 years), 79				X			X		X		(93)
M & F, parents/caregivers, 79				X			X				(94)
USA - AI/AN											
M & F, healthcare providers, 319				X		X	X				(95)
USA - Native Hawaiians											
M & F, parents/caregivers, 189					X		X				(96)
Canada - First Nations											
M & F, community members, 24		X				X	X				(97)
Papua New Guinea - Papuans											
M & F, community members, 208					X	X	X			X	(98)
Peru - Quechuas											
F, parents/caregivers, 192						X	X	X			(99)
Subtotal	**1**	**1**	**2**	**8**	**5**	**6**	**12**	**1**	**1**	**1**	**14**
Barriers to cervical cancer screening											
Canada - Inuit											
F, community members, 27					X						(100)
F, community members, 175				X		X			X		(101)

(Continued)

TABLE 2 (Continued)

Country – Study population	Structural barriers			Social barriers		Cultural barriers					Ref.
Participant characteristics, sample size	Remoteness	Individual cost	Resource constraints in healthcare systems[a]	Limited knowledge about HPV, HPV vaccine and/or cervical cancer	Limited health literacy[b]	Sex and cultural taboos[c]	Mistrust in healthcare systems and/or in vaccine[d]	Language	Fear of pain	Culturally inappropriate health education delivery	
Canada - First Nations											
F, community members, 69 M & F, healthcare providers, 16						X	X				(102)
M & F, healthcare providers, 18	X		X		X						(103)
F, community members, 8						X	X				(104)
Australia - Aboriginal and Torres Strait Islander peoples											
F, community members, 29						X	X				(105)
M & F, healthcare providers, 13			X								(106)
M & F, community members, 368, healthcare providers, 179		X	X	X			X				(107)
New Zealand - Māori											
F, community members, 397						X			X		(108)
Papua New Guinea - Papuans											
M & F, community members, 208				X		X		X		X	(109)
Mexico - Mam, Nahua and Huichol											
F, community members, 122	X		X	X				X		X	(110)
Dominica - Kalinago											
M & F, community members, 42				X							(111)
Guatemala - Maya, Garifuna, Xinca											
F, community members, 5,728	X	X				X		X		X	(82)
Ecuador - Kichwa											
F, community members, 28							X	X			(112)
Peru - Shipibo, Quechua											
F, community members, 18	X	X									(113)
Subtotal	**4**	**3**	**4**	**5**	**2**	**7**	**5**	**4**	**2**	**3**	**15**
Total Barriers	**5**	**4**	**6**	**13**	**7**	**13**	**17**	**5**	**3**	**4**	**29**

F: female; M: male. Details on identified barriers: [a]Lengthy waiting time to get an appointment, Shortages of supplies, Shortages of female healthcare providers, Lack of funding, Competing clinical priorities, Workforce capacity limitations, Lack of recall and follow-up systems. [b]Participants reported that the healthcare provider did not mention the vaccine, Participants reported not knowing enough about the vaccine to be able to make a decision, not knowing where to seek information. [c]Concerns that HPV vaccine may encourage earlier or riskier sexual activity, Negative perceptions regarding sexual health promotion, Discomfort and vulnerability, Triggering distress and trauma, Stigma associated with sexually transmitted diseases, Lack of privacy and control, Women requiring permission from partner to go to the clinic, Fear of cancer. [d]Concerns about vaccine long-term safety and side-effects, Suspicion of Western medicine and preference for the use of traditional medicine, Complicated relationships with healthcare providers.

testing is that self-collected vaginal samples can be used, which can reduce cost, is noninvasive, and thus emerges as a promising solution to increase screening participation in settings with limited resources, access to health services, or where cultural barriers exist (134–139). In particular, self-collected HPV testing has been shown to be better accepted among Indigenous women in Australia (108, 140), New Zealand (141, 142) and Guatemala (143). Accordingly, in recent years, the Ministry of Health of Guatemala has worked to implement self-collection for HPV testing as an alternative screening method to increase screening coverage in vulnerable populations, especially rural and Indigenous women (144, 145). To the best of available information, this is the only governmental initiative dedicated to improve cervical cancer prevention and control among Indigenous populations that has been implemented in a LAC country.

5 Conclusion

Because they experience higher levels of poverty and health inequities, Indigenous women worldwide are more vulnerable to cervical cancer than non-Indigenous women. In high-income countries, the disproportionately high burden of cervical cancer among Indigenous populations has been extensively characterized. Studies have consistently revealed a higher prevalence of HPV infection and increased incidence and mortality of cervical cancer among Indigenous women compared to their non-Indigenous counterparts. In these countries, targeted programs and initiatives aimed at overcoming the multiple and complex structural, social, and cultural barriers hindering Indigenous women's access to cervical screening and HPV vaccination are yielding promising results.

On the contrary, in LAC, where Indigenous people constitute 10% of the population, the true extent of the burden of cervical cancer among Indigenous women remains largely unknown. Through an extensive review of published literature, this study presents the currently available data concerning the prevalence of cervical cancer and HPV infection among Indigenous women in LAC, highlighting the paucity of existing data and the critical gaps in research and healthcare interventions that need to be fill. While the findings discussed in this article offer valuable insights, one notable limitation is the absence of a meta-analysis, which could have facilitated a comparative assessment of HPV infection and cervical lesion prevalence rates between Indigenous women in LAC and the general population. Because indigenous status is not recorded in cancer registries of most LAC countries, accurately quantifying and understanding the burden of cervical cancer among Indigenous women in the region remains a challenge. Nevertheless, the limited available data suggest that Indigenous women in LAC face a heightened risk of cervical cancer incidence and mortality, as observed in Indigenous populations in high-income countries.

Strategies to improve cervical screening and HPV vaccination uptake for Indigenous women in LAC must be culturally sensitive, education-focused, and community-engaged. Detailed data on screening behaviors, vaccination uptake, and barriers specific to Indigenous populations in the region are essential. While taking example from successful policies and initiatives implemented in high-income countries, solutions adapted to the socio-economic realities of LAC countries and acknowledging the diversity among the more than 800 Indigenous groups in the region need to be found for bridging existing disparities and achieving the global goal of cervical cancer elimination.

Data availability statement

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding author.

Author contributions

CM: Conceptualization, Funding acquisition, Writing – original draft, Writing – review & editing.

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Conflict of interest

The author declares that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

References

1. de Sanjosé S, Diaz M, Castellsagué X, Clifford G, Bruni L, Muñoz N, et al. Worldwide prevalence and genotype distribution of cervical human papillomavirus DNA in women with normal cytology: a meta-analysis. *Lancet Infect Dis*. (2007) 7:453–9. doi: 10.1016/S1473-3099(07)70158-5

2. Forman D, de Martel C, Lacey CJ, Soerjomatarama I, Lortet-Tieulent J, Bruni L, et al. Global burden of human papillomavirus and related diseases. *Vaccine*. (2012) 30:F12–23. doi: 10.1016/j.vaccine.2012.07.055

3. Bruni L, Albero G, Rowley J, Alemany L, Arbyn M, Giuliano AR, et al. Global and regional estimates of genital human papillomavirus prevalence among men: a systematic review and meta-analysis. *Lancet Glob Health*. (2023) 11:e1345–62. doi: 10.1016/S2214-109X(23)00305-4

4. Rodríguez AC, Schiffman M, Herrero R, Wacholder S, Hildesheim A, Castle PE, et al. Rapid clearance of human papillomavirus and implications for clinical focus on persistent infections. *JNCI J Natl Cancer Inst*. (2008) 100:513–7. doi: 10.1093/jnci/djn044

5. Doorbar J, Quint W, Banks L, Bravo IG, Stoler M, Broker TR, et al. The biology and life-cycle of human papillomaviruses. *Vaccine*. (2012) 30:F55–70. doi: 10.1016/j.vaccine.2012.06.083

6. Chan CK, Aimagambetova G, Ukybassova T, Kongrtay K, Azizan A. Human papillomavirus infection and cervical Cancer: epidemiology, screening, and vaccination - review of current perspectives. *J Oncol*. (2019) 2019:1–11. doi: 10.1155/2019/3257939

7. Organization WH. Human papillomavirus vaccines: WHO position paper, December 2022. (2022) [cited 2023 Nov 4]. Available from: https://www.who.int/publications/i/item/who-wer9750-645-672

8. Cuzick J, Arbyn M, Sankaranarayanan R, Tsu V, Ronco G, Mayrand MH, et al. Overview of human papillomavirus-based and other novel options for cervical Cancer screening in developed and developing countries. *Vaccine*. (2008) 26:K29–41. doi: 10.1016/j.vaccine.2008.06.019

9. Checchi M, Mesher D, Panwar K, Anderson A, Beddows S, Soldan K. The impact of over ten years of HPV vaccination in England: surveillance of type-specific HPV in young sexually active females. *Vaccine*. (2023) 41:6734–44. doi: 10.1016/j.vaccine.2023.10.002

10. Palmer T, Wallace L, Pollock KG, Cuschieri K, Robertson C, Kavanagh K, et al. Prevalence of cervical disease at age 20 after immunisation with bivalent HPV vaccine at age 12-13 in Scotland: retrospective population study. *BMJ*. (2019) 365:l1161. doi: 10.1136/bmj.l1161

11. Chow EPF, Machalek DA, Tabrizi SN, Danielewski JA, Fehler G, Bradshaw CS, et al. Quadrivalent vaccine-targeted human papillomavirus genotypes in heterosexual men after the Australian female human papillomavirus vaccination programme: a retrospective observational study. *Lancet Infect Dis*. (2017) 17:68–77. doi: 10.1016/S1473-3099(16)30116-5

12. Patel C, Brotherton JML, Pillsbury A, Jayasinghe S, Donovan B, Macartney K, et al. The impact of 10 years of human papillomavirus (HPV) vaccination in Australia: what additional disease burden will a nonavalent vaccine prevent? *Eur Secur*. (2018) 23:30–40. doi: 10.2807/1560-7917.ES.2018.23.41.1700737

13. Sankaranarayanan R, Gaffikin L, Jacob M, Sellors J, Robles S. A critical assessment of screening methods for cervical neoplasia. *Int J Gynecol Obstet*. (2005) 89:S4–S12. doi: 10.1016/j.ijgo.2005.01.009

14. Ferlay J, Soerjomataram I, Dikshit R, Eser S, Mathers C, Rebelo M, et al. Cancer incidence and mortality worldwide: sources, methods and major patterns in GLOBOCAN 2012. *Int J Cancer*. (2015) 136:E359–86. doi: 10.1002/ijc.29210

15. World Health Organization. Global strategy to accelerate the elimination of cervical cancer as a public health problem. (2020) [cited 2023 Nov 4]. Available from: https://www.who.int/publications/i/item/9789240014107

16. Singh GK, Azuine RE, Siahpush M. Global inequalities in cervical Cancer incidence and mortality are linked to deprivation, low socioeconomic status, and human development. *Int J Matern Child Heal AIDS*. (2012) 1:17–30. doi: 10.21106/ijma.12

17. de Martel C, Georges D, Bray F, Ferlay J, Clifford GM. Global burden of cancer attributable to infections in 2018: a worldwide incidence analysis. *Lancet Glob Health*. (2020) 8:e180–90. doi: 10.1016/S2214-109X(19)30488-7

18. Sung H, Ferlay J, Siegel RL, Laversanne M, Soerjomataram I, Jemal A, et al. Global Cancer statistics 2020: GLOBOCAN estimates of incidence and mortality worldwide for 36 cancers in 185 countries. *CA Cancer J Clin*. (2021) 71:209–49. doi: 10.3322/caac.21660

19. Vaccarella S, Laversanne M, Ferlay J, Bray F. Cervical cancer in Africa, Latin America and the Caribbean and Asia: regional inequalities and changing trends. *Int J Cancer*. (2017) 141:1997–2001. doi: 10.1002/ijc.30901

20. Pilleron S, Cabasag CJ, Ferlay J, Bray F, Luciani S, Almonte M, et al. Cervical cancer burden in Latin America and the Caribbean: where are we? *Int J Cancer*. (2020) 147:1638–48. doi: 10.1002/ijc.32956

21. Torres-Roman JS, Ronceros-Cardenas L, Valcarcel B, Bazalar-Palacios J, Ybaseta-Medina J, Carioli G, et al. Cervical cancer mortality among young women in Latin America and the Caribbean: trend analysis from 1997 to 2030. *BMC Public Health*. (2022) 22:1–10. doi: 10.1186/s12889-021-12413-0

22. Nogueira-Rodrigues A, Flores MG, Macedo Neto AO, Braga LAC, Vieira CM, de Sousa-Lima RM, et al. HPV vaccination in Latin America: coverage status, implementation challenges and strategies to overcome it. *Front Oncol*. (2022) 12:984449. doi: 10.3389/fonc.2022.984449

23. Fernández-Deaza G, Serrano B, Roura E, Castillo JS, Caicedo-Martínez M, Bruni L, et al. Cervical cancer screening coverage in the Americas region: a synthetic analysis. *Lancet Reg Heal - Am*. (2024) 30:100689. doi: 10.1016/j.lana.2024.100689

24. Comisión Económica para América Latina y el Caribe (CEPAL)/Fondo para el Desarrollo de los Pueblos Indígenas de América Latina y el Caribe (FILAC), "Los pueblos indígenas de América Latina - Abya Yala y la Agenda 2030 para el Desarrollo Sostenible: tensiones y desafíos desde una perspectiva territorial", *Documentos de Proyectos* (LC/TS.2020/47), Santiago, 2020.

25. UNHCR. The United Nations Declaration on the Rights of Indigenous Peoples: A Manual for National Human Rights Institutions. (2013). 152 p. Available from: http://www.ohchr.org/Documents/Issues/IPeoples/UNDRIPManualForNHRIs.pdf

26. Moore SP, Forman D, Piñeros M, Fernández SM, de Oliveira SM, Bray F. Cancer in indigenous people in Latin America and the Caribbean: a review. *Cancer Med*. (2014) 3:70–80. doi: 10.1002/cam4.134

27. Tonon SA, Picconi MA, Zinovich JB, Nardari W, Mampaey M, Badano I, et al. Human papillomavirus cervical infection in Guarani Indians from the rainforest of Misiones. *Argentina Int J Infect Dis*. (2004) 8:13–9. doi: 10.1016/j.ijid.2003.03.001

28. Deluca GD, Basiletti J, González JV, Vásquez Díaz N, Lucero RH, Picconi MA. Human papilloma virus risk factors for infection and genotype distribution in aboriginal women from northern Argentina. *Med (Buenos Aires)*. (2012) 72:461–6.

29. Taborda WC, Ferreira SC, Rodrigues D, Stávale JN, Baruzzi RG. Cervical cancer screening among indigenous women in the Xingu Indian reservation, Central Brazil. *Rev Panam Salud Publica*. (2000) 7:92–6. doi: 10.1590/S1020-49892000000200004

30. Brito EB, Martins SJ, Menezes RC. Human papillomaviruses in Amerindian women from Brazilian Amazonia. *Epidemiol Infect*. (2002) 128:485–9. doi: 10.1017/S0950268802006908

31. Rodrigues DA, Ribeiro Pereira É, de Souza Oliveira LS, de Góis Speck NM, Gimeno SGA. Prevalence of cytological atypia and high-risk human papillomavirus infection in Panará indigenous women in Central Brazil. *Cad Saude Publica*. (2014) 30:2587–93. doi: 10.1590/0102-311x00152713

32. Fonseca AJ, Taeko D, Chaves TA, Da Costa Amorim LD, Murari RSW, Miranda AE, et al. HPV infection and cervical screening in socially isolated indigenous women inhabitants of the Amazonian rainforest. *PLoS One*. (2015) 10:e0133635. doi: 10.1371/journal.pone.0133635

33. Nascimento M d DSB, Vidal FCB, Silva MACN d, Batista JE, Lacerda Barbosa M d C, Muniz Filho WE, et al. Prevalence of human papillomavirus infection among women from Quilombo communities in northeastern Brazil. *BMC Womens Health*. (2018) 18:1. doi: 10.1186/s12905-017-0499-3

34. Barbosa MDS, Andrade de Souza IB, Schnaufer ECDS, Da SLF, Maymone Gonçalves CC, Simionatto S, et al. Prevalence and factors associated with trichomonas vaginalis infection in indigenous Brazilian women. *PLoS One*. (2020) 15:e0240323. doi: 10.1371/journal.pone.0240323

35. Novais IR, Coelho CO, Machado HC, Surita F, Zeferino LC, Vale DB. Cervical cancer screening in Brazilian Amazon indigenous women: towards the intensification of public policies for prevention. *PLoS One*. (2023) 18:e0294956. doi: 10.1371/journal.pone.0294956

36. Brito EB, Silva IDC, Stávale JN, Taromaru E, Menezess RC, Martins SJ. Amerindian women of the Brazilian Amazon and STD. *Eur J Gynaecol Oncol*. (2006) 27:279–81.

37. Camargo M, Soto-De Leon SC, Sanchez R, Perez-Prados A, Patarroyo ME, Patarroyo MA. Frequency of human papillomavirus infection, coinfection, and association with different risk factors in Colombia. *Ann Epidemiol*. (2011) 21:204–13. doi: 10.1016/j.annepidem.2010.11.003

38. Sarmiento-Medina MI, de Amaya MP, Villamizar-Gómez L, González-Coba AC, Guzmán-Barajas L. High-risk HPV prevalence and vaccination coverage among indigenous women in the Colombian Amazon: implications for cervical cancer prevention. Cross-sectional study. *PLoS One*. (2024) 19:e0297579. doi: 10.1371/journal.pone.0297579

39. Carrión Ordoñez JI, Soto Brito Y, Pupo Antúnez M, Loja CR. Infección por Virus del Papiloma Humano y citología cérvico-vaginal en mujeres indígenas del Cañar, Ecuador. *Bionatura*. (2019) 4:934–8. doi: 10.21931/RB/2019.04.03.10

40. Ortiz Segarra J, Vega Crespo B, Campoverde Cisneros A, Salazar Torres K, Delgado López D, Ortiz S. Human papillomavirus prevalence and associated factors in indigenous women in Ecuador: a cross-sectional analytical study. *Infect Dis Rep*. (2023) 15:267–78. doi: 10.3390/idr15030027

41. Kightlinger RS, Irvin WP, Archer KJ, Huang NW, Wilson RA, Doran JR, et al. Cervical cancer and human papillomavirus in indigenous Guyanese women. *Am J Obstet Gynecol*. (2010) 202:626.e1–7. doi: 10.1016/j.ajog.2010.03.015

42. Velázquez C, Kawabata A, Ríos-González CM. Prevalence of precursor lesions of cervical cancer and sexual/reproductive antecedents of natives of Caaguazú, Paraguay 2015-2017. *Rev salud pública Paraguay*. (2018) 8:15–P20. doi: 10.18004/rspp.2018.diciembre.15-20

43. Mendoza L, Mongelos P, Paez M, Castro A, Rodriguez-Riveros I, Gimenez G, et al. Human papillomavirus and other genital infections in indigenous women from Paraguay: a cross-sectional analytical study. *BMC Infect Dis*. (2013) 13:531. doi: 10.1186/1471-2334-13-531

44. Blas MM, Alva IE, Garcia PJ, Carcamo C, Montano SM, Muñante R, et al. Association between human papillomavirus and human T-Lymphotropic virus in indigenous women from the Peruvian Amazon. *PLoS One*. (2012) 7:e44240. doi: 10.1371/journal.pone.0044240

45. Martorell M, Garcia-Garcia JA, Gomez-Cabrero D, Del Aguila A. Comparison of the prevalence and distribution of human papillomavirus infection and cervical lesions between urban and native habitants of an Amazonian region of Peru. *Genet Mol Res*. (2012) 11:2099–106. doi: 10.4238/2012.August.6.14

46. Nicita G, Reigosa A, Torres J, Vázquez C, Fernández Y, Álvarez M, et al. Human papillomavirus (HPV) infection in an indigenous population of the Venezuelan Amazon. *Salus*. (2010) 14:51–9.

47. Fuenmayor A, Fernández C, Pérez V, Coronado J, Ávila M, Fernandes A, et al. Detection of precancerous lesions in the cervix and HPV infection in women in the region of Maniapure. *Bolivar State Ecancermedicalscience*. (2018) 12:884. doi: 10.3332/ecancer.2018.884

48. Vargas-Robles D, Magris M, Morales N, de Koning MNC, Rodríguez I, Nieves T, et al. High rate of infection by only oncogenic human papillomavirus in Amerindians. *mSphere*. (2018) 3:e00176-18. doi: 10.1128/mSphere.00176-18

49. Jeffries A, Beck-Sagué CM, Marroquin-Garcia AB, Dean M, McCoy V, Cordova-Toma DA, et al. Cervical visual inspection with acetic acid (VIA) and oncogenic human papillomavirus screening in rural indigenous Guatemalan women: time to rethink VIA. *Int J Environ Res Public Health*. (2021) 18:12406. doi: 10.3390/ijerph182312406

50. Torres-Rojas FI, Mendoza-Catalán MA, Alarcón-Romero LDC, Parra-Rojas I, Paredes-Solís S, Leyva-Vázquez MA, et al. HPV molecular detection from urine versus cervical samples: an alternative for HPV screening in indigenous populations. *PeerJ*. (2021) 9:e11564. doi: 10.7717/peerj.11564

51. Ministério da Saude - Instituto Nacional de Câncer José Alencar Gomes da Silva (INCA). Coordenação Geral de Prevenção e Vigilância (CGPV), Divisão de Vigilância e Análise de Situação. Manual de rotinas e procedimentos para registros de câncer de base populacional. *Instituto Nacional de Câncer José Alencar Gomes da Silva, Coordenação Geral de Prevenção e Vigilância, Divisão de Vigilância e Análise de Situação*. 2. ed. rev. atual. Rio de Janeiro: Inca. (2012). 240 p.

52. Comision contra el Cancer - Registro Nacional de Cancer. Registro Nacional de Cáncer, Uruguay - Material para Buscadores de Datos. *Registro Nacional de Cáncer - Área Vigilancia Epidemiológica, Área de Capacitación Técnico Profesional*. (2019). 26 p.

53. Best Plummer WS, Persaud P, Layne PJ. Ethnicity and cancer in Guyana, South America. *Infect Agent Cancer*. (2009) 4:S7. doi: 10.1186/1750-9378-4-S1-S7

54. Gobierno de la República de Guatemala, Ministerio de Salud Pública y Asistencia Social. Protoco de estudio: Evaluacion de alternativas para la deteccion temprana de cáncer cervicouterino en Guatemala. *Gobierno de la República de Guatemala, Ministerio de Salud Pública y Asistencia Social*. (2018). 37 p.

55. Brau-Figueroa H, Palafox-Parrilla EA, Mohar-Betancourt A, Brau-Figueroa H, Palafox-Parrilla EA, Mohar-Betancourt A. El Registro Nacional de Cáncer en México, una realidad. *Gac Mex Oncol*. (2020) 19:107–11. doi: 10.24875/j.gamo.20000030

56. Government of the United States Virgin Islands, Department of Health. Virgin Islands Central Cancer Registry - Physician's Cancer Report Form. *Government of the United States Virgin Islands, Department of Health*. (2016). 2 p.

57. Davis-Castro CY. Indigenous peoples in Latin America: Statistical Information. (Washington, D.C.: Congressional Research Service). (2023). 34 p.

58. Axelsson P, Kukutai T, Kippen R. The field of indigenous health and the role of colonisation and history. *J Popul Res*. (2016) 33:1–7. doi: 10.1007/s12546-016-9163-2

59. Pan American Health Organization. The Sociodemographic Situation of Indigenous Peoples in Latin America and the Caribbean. *Analysis in the Context of Aging and COVID-19*. Washington, D.C.: PAHO. (2023). Available from: https://doi.org/10.37774/9789275126479.

60. International Labour Organization. Implementing the ILO Indigenous and Tribal Peoples Convention No. 169: Towards an inclusive, sustainable and just future. (2019). 160 p. Available from: https://www.ilo.org/wcmsp5/groups/public/---dgreports/---dcomm/---publ/documents/publication/wcms_735607.pdf

61. Montenegro RA, Stephens C. Indigenous health in Latin America and the Caribbean. *Lancet (London, England)*. (2006) 367:1859–69. doi: 10.1016/S0140-6736(06)68808-9

62. Moore SP, Antoni S, Colquhoun A, Healy B, Ellison-Loschmann L, Potter JD, et al. Cancer incidence in indigenous people in Australia, New Zealand, Canada, and the USA: a comparative population-based study. *Lancet Oncol*. (2015) 16:1483–92. doi: 10.1016/S1470-2045(15)00232-6

63. Ministry of Health. Tatau Kahukura Māori health chart book 2015 (3rd edition). (2015). 68 p. Available from: https://www.health.govt.nz/publication/tatau-kahukura-maori-health-chart-book-2015-3rd-edition

64. Lawton B, Heffernan M, Wurtak G, Steben M, Lhaki P, Cram F, et al. IPVS policy statement addressing the burden of HPV disease for indigenous peoples. *Papillomavirus Res*. (2020) 9:100191. doi: 10.1016/j.pvr.2019.100191

65. Vasilevska M, Ross SA, Gesink D, Fisman DN. Relative risk of cervical cancer in indigenous women in Australia, Canada, New Zealand, and the United States: a systematic review and meta-analysis. *J Public Health Policy*. (2012) 33:148–64. doi: 10.1057/jphp.2012.8

66. Simkin J, Smith L, van Niekerk D, Caird H, Dearden T, van der Hoek K, et al. Sociodemographic characteristics of women with invasive cervical cancer in British Columbia, 2004-2013: a descriptive study. *C open*. (2021) 9:E424–32. doi: 10.9778/cmajo.20200139

67. Mazereeuw MV, Withrow DR, Nishri ED, Tjepkema M, Vides E, Marrett LD. Cancer incidence and survival among Métis adults in Canada: results from the Canadian census follow-up cohort (1992-2009). *Can Med Assoc J*. (2018) 190:E320–6. doi: 10.1503/cmaj.170272

68. Supramaniam R, Grindley H, Pulver LJ. Cancer mortality in aboriginal people in New South Wales, Australia, 1994-2002. *Aust N Z J Public Health*. (2006) 30:453–6. doi: 10.1111/j.1467-842X.2006.tb00463.x

69. Sethi S, Ali A, Ju X, Antonsson A, Logan R, Canfell K, et al. A systematic review and meta-analysis of the prevalence of human papillomavirus infection in indigenous populations – a global picture. *J Oral Pathol Med*. (2021) 50:843–54. doi: 10.1111/jop.13201

70. Jiang Y, Brassard P, Severini A, Goleski V, Santos M, Leamon A, et al. Type-specific prevalence of human papillomavirus infection among women in the Northwest Territories, Canada. *J Infect Public Health*. (2011) 4:219–27. doi: 10.1016/j.jiph.2011.09.006

71. Garland SM, Brotherton JML, Condon JR, McIntyre PB, Stevens MP, Smith DW, et al. Human papillomavirus prevalence among indigenous and non-indigenous Australian women prior to a national HPV vaccination program. *BMC Med*. (2011) 9:1–14. doi: 10.1186/1741-7015-9-104

72. Bennett R, Cerigo H, Coutlée F, Roger M, Franco EL, Brassard P. Incidence, persistence, and determinants of human papillomavirus infection in a population of Inuit women in northern Quebec. *Sex Transm Dis*. (2015) 42:272–8. doi: 10.1097/OLQ.0000000000000272

73. Góes EF, Guimarães JMN, da CC AM, Gabrielli L, Katikireddi SV, Campos AC, et al. The intersection of race/ethnicity and socioeconomic status: inequalities in breast and cervical cancer mortality in 20,665,005 adult women from the 100 million Brazilian cohort. *Ethn Health*. (2024) 29:46–61. doi: 10.1080/13557858.2023.2245183

74. de Melo AC, da Silva JL, dos Santos ALS, Thuler LCS. Population-based trends in cervical Cancer incidence and mortality in Brazil: focusing on Black and indigenous population disparities. *J Racial Ethn Health Disparities*. (2024) 11:255–63. doi: 10.1007/s40615-023-01516-6

75. de SO BMF, Koifman S, Koifman RJ, da Silva IF. Cancer incidence in indigenous populations of Western Amazon, Brazil. *Ethn Health*. (2022) 27:1465–81. doi: 10.1080/13557858.2021.1893663

76. Colpani V, Falcetta FS, Bidinotto AB, Kops NL, Falavigna M, Hammes LS, et al. Prevalence of human papillomavirus (HPV) in Brazil: a systematic review and meta-analysis. *PLoS One*. (2020) 15:e0229154. doi: 10.1371/journal.pone.0229154

77. Bruni L, Diaz M, Castellsagué X, Ferrer E, Bosch FX, De Sanjosé S. Cervical human papillomavirus prevalence in 5 continents: Meta-analysis of 1 million women with Normal cytological findings. *J Infect Dis*. (2010) 202:1789–99. doi: 10.1086/657321

78. Ting J, Rositch AF, Taylor SM, Rahangdale L, Soeters HM, Sun X, et al. Worldwide incidence of cervical lesions: a systematic review. *Epidemiol Infect*. (2015) 143:225. doi: 10.1017/S0950268814001356

79. Shafer LA, Jeffrey I, Elias B, Shearer B, Canfell K, Kliewer E. Quantifying the impact of dissimilar HPV vaccination uptake among Manitoban school girls by ethnicity using a transmission dynamic model. *Vaccine*. (2013) 31:4848–55. doi: 10.1016/j.vaccine.2013.07.073

80. Brotherton JML, Winch KL, Chappell G, Banks C, Meijer D, Ennis S, et al. HPV vaccination coverage and course completion rates for indigenous Australian adolescents, 2015. *Med J Aust*. (2019) 211:31–6. doi: 10.5694/mja2.50221

81. Poirier B, Sethi S, Garvey G, Hedges J, Canfell K, Smith M, et al. HPV vaccine: uptake and understanding among global indigenous communities – a qualitative systematic review. *BMC Public Health*. (2021) 21:2062. doi: 10.1186/s12889-021-12147-z

82. Gottschlich A, Ochoa P, Rivera-Andrade A, Alvarez CS, Mendoza Montano C, Camel C, et al. Barriers to cervical cancer screening in Guatemala: a quantitative analysis using data from the Guatemala demographic and health surveys. *Int J Public Health*. (2020) 65:217. doi: 10.1007/s00038-019-01319-9

83. Barcelos MRB, de Cássia Duarte Lima R, Tomasi E, Nunes BP, SMS D, Facchini LA. Quality of cervical cancer screening in Brazil: external assessment of the PMAQ. *Rev Saude Publica*. (2017) 51:67. doi: 10.1590/s1518-8787.2017051006802

84. Whop LJ, Baade PD, Brotherton JML, Canfell K, Cunningham J, Gertig D, et al. Time to clinical investigation for indigenous and non-indigenous Queensland women after a high grade abnormal pap smear, 2000-2009. *Med J Aust*. (2017) 206:73–7. doi: 10.5694/mja16.00255

85. Dasgupta P, Aitken JF, Condon J, Garvey G, Whop LJ, DeBats C, et al. Spatial and temporal variations in cervical cancer screening participation among indigenous and non-indigenous women, Queensland, Australia, 2008-2017. *Cancer Epidemiol*. (2020) 69:101849. doi: 10.1016/j.canep.2020.101849

86. Gopalani SV, Janitz AE, Burkhart M, Campbell JE, Chen S, Martinez SA, et al. HPV vaccination coverage and factors among American Indians in Cherokee nation. *Cancer Causes Control*. (2023) 34:267. doi: 10.1007/s10552-022-01662-y

87. Bowen DJ, Weiner D, Samos M, Canales MK. Exploration of New England native American Women's views on human papillomavirus (HPV), testing, and vaccination. *J Racial Ethn Health Disparities*. (2014) 1:45–51. doi: 10.1007/s40615-014-0009-3

88. Jacobs-Wingo JL, Jim CC, Groom AV. Human papillomavirus vaccine uptake: increase for American Indian adolescents, 2013–2015. *Am J Prev Med*. (2017) 53:162. doi: 10.1016/j.amepre.2017.01.024

89. Duvall J, Buchwald D. Human papillomavirus vaccine policies among American Indian tribes in Washington state. *J Pediatr Adolesc Gynecol*. (2012) 25:131. doi: 10.1016/j.jpag.2011.11.012

90. Schmidt-Grimminger D, Frerichs L, Black Bird AE, Workman K, Dobberpuhl M, Watanabe-Galloway S. HPV knowledge, attitudes, and beliefs among Northern Plains American Indian adolescents, parents, young adults, and health professionals. *J Cancer Educ*. (2013) 28:357–66. doi: 10.1007/s13187-013-0468-y

91. Hodge FS, Itty T, Cardoza B, Samuel-Nakamura C. HPV vaccine readiness among American Indian college students. *Ethn Dis*. (2011) 21:415–20.

92. Buchwald D, Muller C, Bell M, Schmidt-Grimminger D. Attitudes toward HPV vaccination among rural American Indian women and urban white women in the Northern Plains. *Health Educ Behav*. (2013) 40:704–11. doi: 10.1177/1090198113477111

93. Kemberling M, Hagan K, Leston J, Kitka S, Provost E, Hennessy T. Alaska native adolescent views on cervical cancer, the human papillomavirus (HPV), genital warts and the quadrivalent HPV vaccine. *Int J Circumpolar Health*. (2011) 70:245–53. doi: 10.3402/ijch.v70i3.17829

94. Toffolon-Weiss M, Hagan K, Leston J, Peterson L, Provost E, Hennessy T. Alaska native parental attitudes on cervical cancer, HPV and the HPV vaccine. *Int J Circumpolar Health*. (2008) 67:363–73. doi: 10.3402/ijch.v67i4.18347

95. Jim CC, Wai-Yin Lee J, Groom AV, Espey DK, Saraiya M, Holve S, et al. Human papillomavirus vaccination practices among providers in Indian Health Service, tribal and urban Indian healthcare facilities. *J Women's Health*. (2012) 21:372–8. doi: 10.1089/jwh.2011.3417

96. Dela Cruz MRI, Braun KL, Tsark JAU, Albright CL, Chen JJ. HPV vaccination prevalence, parental barriers and motivators to vaccinating children in Hawai'i. *Ethn Health*. (2020) 25:982. doi: 10.1080/13557858.2018.1473556

97. Henderson RI, Shea-Budgell M, Healy C, Letendre A, Bill L, Healy B, et al. First nations people's perspectives on barriers and supports for enhancing HPV vaccination: foundations for sustainable, community-driven strategies. *Gynecol Oncol*. (2018) 149:93–100. doi: 10.1016/j.ygyno.2017.12.024

98. Kelly-Hanku A, Newland J, Aggleton P, Ase S, Aeno H, Fiya V, et al. HPV vaccination in Papua New Guinea to prevent cervical cancer in women: gender, sexual morality, outsiders and the de-feminization of the HPV vaccine. *Papillomavirus Res*. (2019) 8:100171. doi: 10.1016/j.pvr.2019.100171

99. De La Cruz-Ramirez YM, Olaza-Maguiña AF. Barriers to HPV vaccine uptake in adolescents of the indigenous Andean community of Peru. *Int J Gynecol Obstet*. (2023) 162:187–9. doi: 10.1002/ijgo.14844

100. Tratt E, Sarmiento I, Gamelin R, Nayoumealuk J, Andersson N, Brassard P. Fuzzy cognitive mapping with Inuit women: what needs to change to improve cervical cancer screening in Nunavik, northern Quebec? *BMC Health Serv Res*. (2020) 20:529. doi: 10.1186/s12913-020-05399-9

101. Cerigo H, Macdonald ME, Franco EL, Brassard P. Inuit women's attitudes and experiences towards cervical cancer and prevention strategies in Nunavik, Quebec. *Int J Circumpolar Health*. (2012) 71:17996. doi: 10.3402/ijch.v71i0.17996

102. Wakewich P, Wood B, Davey C, Laframboise A, Zehbe I. Colonial legacy and the experience of first nations women in cervical cancer screening: a Canadian multi-community study. *Crit Public Health*. (2016) 26:368–80. doi: 10.1080/09581596.2015.1067671

103. Maar M, Burchell A, Little J, Ogilvie G, Severini A, Yang JM, et al. A qualitative study of provider perspectives of structural barriers to cervical Cancer screening among first nations women. *Womens Health Issues*. (2013) 23:e319–25. doi: 10.1016/j.whi.2013.06.005

104. O'Brien BA, Mill J, Wilson T. Cervical screening in Canadian first nation Cree women. *J Transcult Nurs*. (2009) 20:83–92. doi: 10.1177/1043659608322418

105. Butler TL, Lee N, Anderson K, Brotherton JML, Cunningham J, Condon JR, et al. Under-screened aboriginal and Torres Strait islander women's perspectives on cervical screening. *PLoS One*. (2022) 17:e0271658. doi: 10.1371/journal.pone.0271658

106. Jaenke R, Butler TL, Condon J, Garvey G, Brotherton JML, Cunningham J, et al. Health care provider perspectives on cervical screening for aboriginal and Torres Strait islander women: a qualitative study. *Aust N Z J Public Health*. (2021) 45:150–7. doi: 10.1111/1753-6405.13084

107. Manderson L, Hoban E. Cervical cancer services for indigenous women: advocacy, community-based research and policy change in Australia. *Women Health*. (2006) 43:69–88. doi: 10.1300/J013v43n04_05

108. Adcock A, Cram F, Lawton B, Geller S, Hibma M, Sykes P, et al. Acceptability of self-taken vaginal HPV sample for cervical screening among an under-screened indigenous population. *Aust New Zeal J Obstet Gynaecol*. (2019) 59:301–7. doi: 10.1111/ajo.12933

109. Kelly-Hanku A, Ase S, Fiya V, Toliman P, Aeno H, Mola GM, et al. Ambiguous bodies, uncertain diseases: knowledge of cervical cancer in Papua New Guinea. *Ethn Health*. (2018) 23:659–81. doi: 10.1080/13557858.2017.1283393

110. Allen-Leigh B, Uribe-Zúñiga P, León-Maldonado L, Brown BJ, Lörincz A, Salmeron J, et al. Barriers to HPV self-sampling and cytology among low-income indigenous women in rural areas of a middle-income setting: a qualitative study. *BMC Cancer*. (2017) 17:734. doi: 10.1186/s12885-017-3723-5

111. Warner ZC, Reid B, Auguste P, Joseph W, Kepka D, Warner EL. Awareness and knowledge of HPV, HPV vaccination, and cervical Cancer among an indigenous Caribbean community. *Int J Environ Res Public Health*. (2022) 19:5694. doi: 10.3390/ijerph19095694

112. Nugus P, Désalliers J, Morales J, Graves L, Evans A, Macaulay AC. Localizing global medicine: challenges and opportunities in cervical screening in an indigenous Community in Ecuador. *Qual Health Res*. (2018) 28:800–12. doi: 10.1177/1049732317742129

113. Nevin PE, Garcia PJ, Blas MM, Rao D, Molina Y. Inequities in cervical Cancer Care in Indigenous Peruvian Women. *Lancet Glob Health*. (2019) 7:e556–7. doi: 10.1016/S2214-109X(19)30044-0

114. MacDonald SE, Kenzie L, Letendre A, Bill L, Shea-Budgell M, Henderson R, et al. Barriers and supports for uptake of human papillomavirus vaccination in indigenous people globally: a systematic review. *PLOS Glob Public Heal*. (2023) 3:e0001406. doi: 10.1371/journal.pgph.0001406

115. Letendre A, Khan M, Bartel R, Chiang B, James A, Shewchuk B, et al. Creation of a Métis-specific instrument for Cancer screening: a scoping review of Cancer-screening programs and instruments. *Curr Oncol*. (2023) 30:9849–59. doi: 10.3390/curroncol30110715

116. Jumah NA, Kewayosh A, Downey B, Campbell Senese L, Tinmouth J. Developing a health equity impact assessment 'indigenous Lens tool' to address challenges in providing equitable cancer screening for indigenous peoples. *BMC Public Health*. (2023) 23:2250. doi: 10.1186/s12889-023-16919-7

117. Lawton B, MacDonald EJ, Storey F, Stanton JA, Adcock A, Gibson M, et al. A model for empowering rural solutions for cervical Cancer prevention (he Tapu Te Whare Tangata): protocol for a cluster randomized crossover trial. *JMIR Res Protoc*. (2023) 12:e51643. doi: 10.2196/51643

118. McGregor S, Saulo D, Brotherton J, Liu B, Phillips S, Skinner SR, et al. Decline in prevalence of human papillomavirus infection following vaccination among Australian indigenous women, a population at higher risk of cervical cancer: the VIP-I study. *Vaccine*. (2018) 36:4311–6. doi: 10.1016/j.vaccine.2018.05.104

119. Gibson-Helm M, Slater T, MacDonald EJ, Stevenson K, Adcock A, Geller S, et al. Te Ara Waiora–implementing human papillomavirus (HPV) primary testing to prevent cervical cancer in Aotearoa New Zealand: a protocol for a non-inferiority trial. *PLoS One*. (2023) 18:e0280643. doi: 10.1371/journal.pone.0280643

120. Moxham R, Moylan P, Duniec L, Fisher T, Furestad E, Manolas P, et al. Knowledge, attitudes, beliefs, intentions and behaviours of Australian indigenous women from NSW in response to the National Cervical Screening Program changes: a qualitative study. *Lancet Reg Heal West Pacific*. (2021) 13:100195. doi: 10.1016/j.lanwpc.2021.100195

121. Jackson J, Sonneveld N, Rashid H, Karpish L, Wallace S, Whop L, et al. Vaccine preventable diseases and vaccination coverage in aboriginal and Torres Strait islander people, Australia, 2016-2019. *Commun Dis Intell*. (2023) 47:127–59. doi: 10.33321/cdi.2023.47.32

122. Jamal S, Jones C, Walker J, Mazereeuw M, Sheppard AJ, Henry D, et al. Cancer in first nations people in Ontario, Canada: incidence and mortality, 1991 to 2010. *Heal reports*. (2021) 32:14–28. doi: 10.25318/82-003-x202100600002-eng

123. Maxwell AE, Young S, Rabelo Vega R, Cayetano RT, Crespi CM, Bastani R. Building capacity to address Women's health issues in the Mixtec and Zapotec community. *Womens Health Issues*. (2015) 25:403. doi: 10.1016/j.whi.2015.03.010

124. Maar M, Wakewich P, Wood B, Severini A, Little J, Burchell AN, et al. Strategies for increasing cervical Cancer screening amongst first nations communities in Northwest Ontario, Canada. *Health Care Women Int*. (2016) 37:478–95. doi: 10.1080/07399332.2014.959168

125. Butler TL, Anderson K, Condon JR, Garvey G, Brotherton JML, Cunningham J, et al. Indigenous Australian women's experiences of participation in cervical screening. *PLoS One*. (2020) 15:e0234536. doi: 10.1371/journal.pone.0234536

126. Suarez L, Lloyd L, Weiss N, Rainbolt T, Pulley L. Effect of social networks on Cancer-screening behavior of older Mexican-American women. *JNCI J Natl Cancer Inst*. (1994) 86:775–9. doi: 10.1093/jnci/86.10.775

127. Luque JS, Opoku S, Ferris DG, Guevara Condorhuaman WS. Social network characteristics and cervical cancer screening among Quechua women in Andean Peru. *BMC Public Health*. (2016) 16:181. doi: 10.1186/s12889-016-2878-3

128. Schnippel K, Lince-Deroche N, Van Den Handel T, Molefi S, Bruce S, Firnhaber C. Cost evaluation of reproductive and primary health care Mobile service delivery for women in two rural districts in South Africa. *PLoS One*. (2015) 10:e0119236. doi: 10.1371/journal.pone.0119236

129. Ferris DG, Shapiro J, Fowler C, Cutler C, Waller J, Condorhuaman WSG. The impact of accessible cervical Cancer screening in Peru-the Día del Mercado project. *J Low Genit Tract Dis*. (2015) 19:229–33. doi: 10.1097/LGT.0000000000000112

130. Kulasingam SL, Hughes JP, Kiviat NB, Mao C, Weiss NS, Kuypers JM, et al. Evaluation of human papillomavirus testing in primary screening for cervical abnormalities: comparison of sensitivity, specificity, and frequency of referral. *JAMA*. (2002) 288:1749–57. doi: 10.1001/jama.288.14.1749

131. Mayrand M-H, Duarte-Franco E, Rodrigues I, Walter SD, Hanley J, Ferenczy A, et al. Human papillomavirus DNA versus Papanicolaou screening tests for cervical Cancer. *N Engl J Med*. (2007) 357:1579–88. doi: 10.1056/NEJMoa071430

132. Rijkaart DC, Berkhof J, Rozendaal L, van Kemenade FJ, Bulkmans NWJ, Heideman DAM, et al. Human papillomavirus testing for the detection of high-grade cervical intraepithelial neoplasia and cancer: final results of the POBASCAM randomised controlled trial. *Lancet Oncol*. (2012) 13:78–88. doi: 10.1016/S1470-2045(11)70296-0

133. Ronco G, Dillner J, Elfström KM, Tunesi S, Snijders PJF, Arbyn M, et al. Efficacy of HPV-based screening for prevention of invasive cervical cancer: follow-up of four European randomised controlled trials. *Lancet*. (2014) 383:524–32. doi: 10.1016/S0140-6736(13)62218-7

134. Mezei AK, Armstrong HL, Pedersen HN, Campos NG, Mitchell SM, Sekikubo M, et al. Cost-effectiveness of cervical cancer screening methods in low- and middle-income countries: a systematic review. *Int J Cancer*. (2017) 141:437–46. doi: 10.1002/ijc.30695

135. Malone C, Barnabas RV, Buist DSM, Tiro JA, Winer RL. Cost-effectiveness studies of HPV self-sampling: a systematic review. *Prev Med (Baltim)*. (2020) 132:105953. doi: 10.1016/j.ypmed.2019.105953

136. Toliman P, Badman SG, Gabuzzi J, Silim S, Forereme L, Kumbia A, et al. Field evaluation of Xpert HPV point-of-care test for detection of human papillomavirus infection by use of self-collected vaginal and clinician-collected cervical specimens. *J Clin Microbiol*. (2016) 54:1734–7. doi: 10.1128/JCM.00529-16

137. Polman NJ, de Haan Y, Veldhuijzen NJ, Heideman DAM, de Vet HCW, Meijer CJLM, et al. Experience with HPV self-sampling and clinician-based sampling in women attending routine cervical screening in the Netherlands. *Prev Med (Baltim)*. (2019) 125:5–11. doi: 10.1016/j.ypmed.2019.04.025

138. Gravitt PE, Belinson JL, Salmeron J, Shah KV. Looking ahead: a case for human papillomavirus testing of self-sampled vaginal specimens as a cervical cancer screening strategy. *Int J Cancer*. (2011) 129:517–27. doi: 10.1002/ijc.25974

139. Scarinci IC, Litton AG, Garcés-Palacio IC, Partridge EE, Castle PE. Acceptability and usability of self-collected sampling for HPV testing among African-American women living in the Mississippi Delta. *Womens Health Issues*. (2013) 23:e123–30. doi: 10.1016/j.whi.2012.12.003

140. MacDonald EJ, Geller S, Sibanda N, Stevenson K, Denmead L, Adcock A, et al. Reaching under-screened/never-screened indigenous peoples with human papilloma virus self-testing: a community-based cluster randomised controlled trial. *Aust New Zeal J Obstet Gynaecol*. (2021) 61:135–41. doi: 10.1111/ajo.13285

141. Bromhead C, Wihongi H, Sherman SM, Crengle S, Grant J, Martin G, et al. Human papillomavirus (Hpv) self-sampling among never-and under-screened indigenous māori, pacific and asian women in aotearoa New Zealand: a feasibility study. *Int J Environ Res Public Health*. (2021) 18:10050. doi: 10.3390/ijerph181910050

142. Brewer N, Bartholomew K, Grant J, Maxwell A, McPherson G, Wihongi H, et al. Acceptability of human papillomavirus (HPV) self-sampling among never- and under-screened indigenous and other minority women: a randomised three-arm community trial in Aotearoa New Zealand. *Lancet Reg Heal - West Pacific*. (2021) 16:100265. doi: 10.1016/j.lanwpc.2021.100265

143. Gottschlich A, Rivera-Andrade A, Grajeda E, Alvarez C, Mendoza Montano C, Meza R. Acceptability of human papillomavirus self-sampling for cervical Cancer screening in an indigenous Community in Guatemala. *J Glob Oncol*. (2017) 3:444–54. doi: 10.1200/JGO.2016.005629

144. Gobierno de Guatemala, Ministerio de Salud Pública y Asistencia Social. Guía de atención integral para la prevención, detección y tratamiento de lesiones precursoras del cáncer cérvico uterino. *Gobierno de Guatemala, Ministerio de Salud Pública y Asistencia Social*. 2da edición. (2020). 76 p.

145. Gobierno de Guatemala, Ministerio de Salúd Publica y Asistencia Social. Plan estratégico para la prevención y control de cáncer cervicouterino, 2023-2030. *Gobierno de Guatemala, Ministerio de Salúd Publica y Asistencia Social*. (2023). 70 p.

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A rural teledentistry care experience: a geriatric approach to assessing oral health status and treatment needs in older adults from a Mapuche community in Chile

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Background: Limited attention has been given to oral health challenges faced by older Indigenous populations, especially in rural settings, where disparities exist. This study aims to assess oral health in a rural Mapuche community in southern Chile, utilizing geriatric technology support, and exploring the connection between geriatric health and oral well-being to fill a gap in this context.

Methods: A cross-sectional study was conducted involving 76 independent older adults from a rural Mapuche community who required dental care. Assessments were in a remote care setting gathering extensive data including comprehensive geriatric assessments, medical and dental conditions using a geriatric teledentistry platform (TEGO®). Statistical analysis involved descriptive analysis, logistic regression, and both multiple correspondence analysis and k-means cluster analysis.

Results: The sample comprised individuals with limited formal education and a high degree of vulnerability. Geriatric assessments unveiled cognitive deterioration, frailty, depression risk, and multimorbidity. A distribution of the DMFT index, number of remaining teeth, number of occluding pairs, number of teeth with restorative needs and other relevant clinical findings was conducted based on sociodemographic, and medical-geriatric-dental characteristics, and additionally, a Multinomial Logistic Regression Analysis of Dentition Variables in Relation to Geriatric Assessments was performed. The dental burden was substantial, with an average DMFT index of 25.96 (SD 4.38), high prevalence of non-functional dentition (89.3%), periodontal disease (83%), xerostomia (63.2%) and oral mucosal lesions (31.5%). Age, lower education, depression, daily medication number and sugary consumption frequency were associated with a decreased average number of teeth ($p < 0.05$). Multiple correspondence analysis and k-means cluster analysis identified 4 clusters, with the edentulous and functional dentition groups being the most distinct.

Conclusion: This study uncovers a substantial dental burden and intricate medical-geriatric conditions interlinked among Indigenous older adults in a rural Chilean Mapuche community. The implementation of a geriatric technological ecosystem in the community enabled the resolution of less complex oral health issues and facilitated remote consultations with specialists, reducing the necessity for travel to health centers. This underscores the need for innovative dental public health initiatives to address health disparities and improve the overall well-being of older Indigenous adults.

KEYWORDS

oral health, indigenous, oral disease burdens, teledentistry, aging, inequalities, geriatric assessment, older adults

1 Introduction

Numerous global references have focused on enhancing the oral health of older adults (1–5). However, limited attention has been directed toward the dual challenge faced by older Indigenous populations, stemming from restricted dental access and age-related concerns (6), which is further worsen by the disparities between Indigenous and non-Indigenous oral health outcomes (7–10). Moreover, in the present day, Indigenous communities are among the most disadvantaged globally (11, 12), facing an inequitable burden of health disparities, which differ across continents and are primarily linked to a mix of factors, such as socioeconomic disadvantages, communication barriers, transportation limitations, inadequate and inconsistent funding for community health services, limited access to culturally suitable healthcare, summed to unique social obstacles such as systemic racism, colonialism/neocolonialism, globalization, migration, transgenerational loss of language, land and culture, and even climate change (8, 13–16). Given potential disparities in funding for oral and general healthcare, it is imperative to thoughtfully consider the potential greater challenge of addressing oral health inequalities compared to general health inequalities (17).

Furthermore, rural communities face challenges associated with aging, marked by the departure of working-age individuals to urban areas and the influx of retirees from cities, intensifying the demand for healthcare (18). Coupled with these demographic shifts, rural health settings grapple with significative administrative, quality control, and logistical hurdles, including a shortage of trained personnel and limited access to specialists (19). Globally, the utilization of health information and communication technologies (e-health) has proven instrumental in overcoming geographical barriers, emerging as a viable option for providing oral care services in rural and remote areas (20). These technologies have positively impacted general health condition management, contributing to enhanced healthcare access, improved quality of life, and increased social support within Indigenous populations (21, 22). Despite these benefits, Indigenous e-health research remains underexplored and underfunded, predominantly focusing on younger Indigenous populations in developed countries (21–23).

Within the framework of human rights and sustainable development, initiatives have been undertaken to expedite endeavors aimed at enhancing Indigenous populations' access to healthcare as a means of promoting well-being, justice, and human dignity (24–26). Nonetheless, achieving this goal requires the development of scientific evidence regarding the epidemiological profiles of this population (24) and the formulation of community-tailored programs. Based on global experiences, it has been emphasized that strategies such as community health programs, student-led healthcare services, outreach initiatives, mobile clinics, and e-health could play a pivotal role in achieving this goal (27).

In that context, our group has made efforts in implementing an innovative teleodontology strategy for older adult residents of rural areas in Chile, with a special focus on the Mapuche community, which constitutes the most numerous Indigenous group in Chile, reaching around one million people in the central and southern regions of the country. 26% of the population in the La Araucanía Region, over 14 years old, identifies as such.

The absence of comprehensive epidemiological data among Latin American Indigenous groups represents a significant hurdle to advancing tailored oral health awareness and preventive initiatives (9). Moreover, a notable gap exists in initiatives addressing the distinctive requirements of older Indigenous individuals residing in rural settings, coupled with a scarcity of studies exploring the utilization of e-health, particularly e-oral healthcare, within this population, to the best of our knowledge. In this context, this study aims to assess the oral health status and treatment requirements of older adults within a rural Mapuche community in the La Araucania region of Chile, leveraging support from a geriatric technological ecosystem. Additionally, it seeks to explore the association between the sociomedical geriatric state and oral health, addressing an unaddressed area within the context of this Indigenous community.

2 Materials and methods

2.1 Ethical considerations

This cross-sectional study was developed as part of a Presidential Grant Project called Clinical Center for Innovation in Oral Health of the Older Adult in La Araucania region (Exempt Resolution No. 472). The main objective of this project is to

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Abbreviations: TEGO, Geriatric Dental Specialties Tele platform; OML, Oral Mucosal Lesion.

implement interventions aimed at preventing and treating oral diseases, along with initiatives to enhance oral health knowledge, behaviors, and oral health self-efficacy. Ethical approval for conducting this project was granted by the Universidad de La Frontera Ethics Committee, decision 109/22. All the participants agreed to participate by signing an informed consent and their confidentiality was granted by anonymization of personal information prior to data treatment.

2.2 Participants and setting

In Chile, 12.8% of the population belongs to Indigenous communities, Mapuche people are the most prominent group, making up 32.82% of the population in the La Araucania region (28). The traditional Mapuche territory spans from the Bio-Bio River to Chiloe Island, encompassing diverse territorial identities that diverge from official state boundaries; each holds its distinct socio-cultural, spiritual rationale, and medical-religious framework (29, 30). The Makewe-Pelale sector belongs to the Wenteche territory and is in the La Araucania region, specifically in the municipalities of Padre las Casas and Freire. It consists of approximately 80 Mapuche communities with an estimated population of 10.000 people; the residents of this sector are mainly small-scale farmers, with an average of only 1.5 hectares per capital (29). This rural sector does not have access to fluoridated water.

The first approach to the Makewe-Pelale community was through a social anthropologist (A.H) with many years of collaborating with the community. Then, a meeting was made with the Werkén, spokesperson for various Mapuche organizations (31), and the Trapilhue leaders to explain the project, learn about their perspectives and perceived needs. The community through its council designated an intercultural facilitator of the community (E.Q) who spoke Mapuzugun (the language spoken by the Mapuche community), to be in charge of recruiting and enrolling the participants. Due to the scarce statistical records in this geographic area and the poor connectivity, the recruitment was through a convenience sampling, in which the intercultural facilitator went house to house to recruit participants. As in Beltrán et al. (32), inclusion criteria considered older adults (60 years and older) requiring an emergency, priority, or dental check-ups. Requirements to provide dental care in a remote setting were specified as follows: patients with chronic diseases must be under pharmacological treatment according to medical indications; patients must be capable of receiving verbal instructions and have sufficient mobility to sit in a portable dental chair. If they met the selection criteria and agreed through informed consent, they were scheduled to receive dental care in an Experimental Campus of Universidad de La Frontera located in this area.

Patients were enrolled face-to-face by the facilitator trained for it. Data considered for the patient's registration included socio-demographic information such as their full name, national ID number, date of birth, contact phone, address, social register household (RSH) (33), ancestry, educational level, occupation, and social support. Barthel Index for Activities of Daily Living (34) was applied. All the data and instruments were loaded on a web platform named Geriatric Dental Specialties Tele platform (TEGO by its acronym in Spanish: "Tele Plataforma de Especialidades Geriátrico Odontológicas") (32). This platform constitutes a technological ecosystem for geriatric clinical care, incorporating an electronic dental assessment record, 3D models, and facilitating teleconsultations with specialists.

The setting was a classroom prepared in workstations for the integral assessment and treatment of the older adults, four fully portable dental equipment and a portable dental x-ray equipment were used.

The inclusion criteria considered the older adult population (over 60 years old) with a dental emergency or requiring some kind of priority dental care. The requirements for providing dental care in a mobile clinic were specified as follows: sufficient mobility to access a dental chair in a mobile clinic; patients with chronic diseases must be under pharmacological treatment according to medical indications; patients must be capable of receiving verbal instructions and must complete a triage before the provision of dental care. The exclusion criteria apply to patients with vital emergencies (e.g., anaphylactic crisis) that must be immediately assessed by a physician and require urgent care.

2.3 Data collection

The examinations were conducted by two dental students completing their final year of internship, who were trained and supervised by two dentists with extensive clinical experience in the field (C.A and F.M-S). Before the examinations, the examiners were trained on the study protocol and diagnostic criteria using a PowerPoint presentation. Inter- and intra-examiner agreement (kappa values) was calculated. For the inter-examiner agreement, the students examined a total of 445 dental surfaces across six patients, while for the intra-examiner agreement, they re-examined 245 dental surfaces 1 week later. The results showed substantial inter-examiner agreement (κ value = 0.66) and substantial to almost perfect intra-examiner agreement (κ values of 0.78 and 0.85, respectively).

Clinical data were recorded following the criteria and recommendations from well-established methods (35, 36). The TEGO platform integrates three anamnesis modules for integral asses of older adults, allowing the evaluation of:

2.3.1 Medico-geriatric data

a Cognitive state: The assessment was made through the Mini Mental state examination (MMSE) (37) or the Short Portable Mental Status Questionnaire (SPMSQ) (38), according to educational level. Cognitive deterioration was defined with <25 points in MMSE or >3 points in SPMSQ.
b Depression: The shortened Yesavage geriatric depression scale was evaluated (39).
c Frailty: Fried frailty phenotype (40) was applied.
d Nutritional status: Was calculated through Body Mass Index using the values for weight and height. A BMI ≥ 25 was considered overweight.
e Multimorbidity: Multimorbidity has been defined as the presence of two or more comorbidities (41), and was obtained from participants' reports.
f Number of daily medications: Medication consumption was obtained from the participants reports. Polypharmacy was defined as the consumption of five or more medication (42).

g History of falls in the past year, obtained from the participant report.

2.3.2 Dental-geriatric data

a Lifestyle-related characteristics: A first interview was conducted for the assessment of habits such as brushing frequency, use of dental floss, cariogenic diet consumption and unhealthy habits. The questions surveyed were: How many times per day do you brush your teeth: never/once a day/twice a day/ three times a day/more than three times a day?; Do you use dental floss?: yes/ no; concerning the consumption of a cariogenic diet, the questions were as follows: Do you consume sugary foods and/ or drinks (such as candies, cookies, cakes, sweet cakes, soda or sugary juices, coffee, tea, mate or milk with sugar) between meals: yes/no?. If the answer was yes, a question of the number of times in the day was added. Smoking habit was surveyed with the following question Do you smoke: yes/no?

b Last visit to dental care, obtained from the participant report.

c Swallowing disorders: The EAT-10 tool (43) was applied for the assessment of swallowing disorders. If the score was three or more, it was considered positive (44).

d Xerostomia assessment: It was surveyed the question Do you feel your mouth dry: yes/no?

e Dental Examination: Dental assessments were conducted employing dental mirrors and a periodontal probe, encompassing evaluations of teeth in both wet and air-dried states while considering both the coronal and root surfaces. Detailed explanations of the ICDAS-II scores for diverse levels of dental caries on all surfaces were previously provided (45). The computation of DMFT indices followed this approach: ICDAS scores 0 to 3 were considered as $D = 0$ (sound); scores 4 to 6 were interpreted as $D = 1$ (requiring restoration); missing teeth were designated as $M = 1$; and restored teeth without caries were attributed $F = 1$. Furthermore, the presence of non-carious tooth surface loss was also evaluated. The oral health status of the teeth was recorded in the platform's odontogram section.

f Periodontal assessment: was made with UNC-15 probe (Hu-Friedy, Chicago, Illinois, United States) performing a full-mouth recording on the SEPA periodontogram (46), which includes periodontal measurement at six sites per tooth. Gingival bleeding was evaluated after probing all teeth with an oral mirror. Periodontitis was defined when interdental $CAL \geq 2$ non-adjacent teeth, or Buccal or Oral $CAL \geq 3$ mm with $PPD > 3$ mm is detectable at ≥ 2 teeth (47); gingivitis was defined as $\geq 10\%$ bleeding sites with probing depths ≤ 3 mm; and gingival health was defined as $<10\%$ bleeding sites, with probing depths ≤ 3 mm (48).

g Missing teeth and number of occluded pairs: The number of natural teeth and tooth condition was assessed using the WHO criteria (35). The number of natural teeth was categorized into four groups: Total tooth loss (0 teeth), severe tooth loss (1–9 teeth), 10–20 teeth and functional dentition (21 or more teeth) (49, 50). The state of the teeth was uploaded in a novel 3D standardized model in TEGO (51). The analysis of the 3D model was used to ascertain the number of occluded pairs, where opposing pairs of maxillary and mandibular teeth were considered as a single occluded pair.

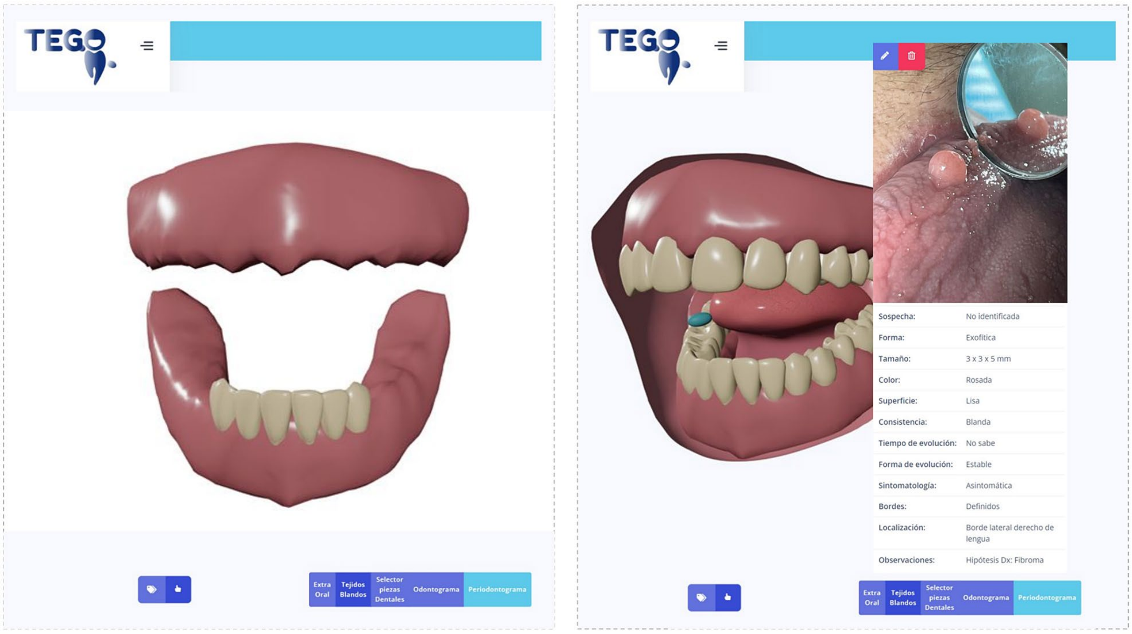
h Oral mucosal lesions (OML): Following WHO guidelines (35), a comprehensive examination of oral mucosa and soft tissues, involving both visual inspection of the perioral area and a systematic assessment of the oral mucosa, was conducted. Detected OML were documented on the platform with traditional descriptions (size, color. Limits, symptomatology, type of surface, consistency, localization, and evolution), 3D digital representations (utilizing an extra and intraoral 3D phantom), and clinical images (52) (see Figure 1). Specialists received timely information through auto-generated emails and provided responses directly through the platform. The assessment aimed to identify the presence or absence of OML, offering an initial clinical diagnosis and classification, including denture related lesion, infectious conditions, oral potentially malignant disorder, tumors and tumor-like lesions, pigmentations, immune-mediated conditions, physical and chemical injuries, and vascular malformations (53–56), while also noting the affected surfaces. Additionally, the oral pathology specialist provided treatment suggestions or follow-up recommendations.

i Prosthetic Evaluation: The assessment of dental prostheses involved evaluating rehabilitation requirements, the use of prostheses, and examining the state of appliances employed by individuals. During the clinical examination, a thorough analysis was conducted, encompassing defects, stability, retention, and the overall integrity of the appliance, all based on established criteria (57, 58).

Figure 2 depicts the TEGO comprehensive care pathway tailored for assessing and treating oral health within older adults of a rural Mapuche community. This pathway encompasses a holistic approach integrating teledentistry alongside conventional methods to address the unique oral health needs of this demographic. From initial screening to treatment plan implementation, each step was meticulously designed to consider cultural sensitivities, ensure patient-centered care, and leverage teledentistry for enhanced accessibility and outreach.

2.4 Statistical analysis

The descriptive analysis encompassed frequencies, means, standard deviations, and confidence intervals for the investigated parameters. Associations between outcomes and explanatory factors were investigated employing cross-tabulation alongside Pearson $\chi 2$ and Fisher's exact tests. Additionally, a Multinomial logistic regression analysis was conducted to examine the relationship between tooth loss variables and the medico-geriatric state of the participants. Multiple correspondence analysis and K-means cluster analysis was performed to gain insights into population profiles in concerning the number of present teeth. The data collection was recorded in a Microsoft Office Excel spreadsheet, a descriptive analysis of the data was carried out for which the mean and its respective standard deviation and frequency distribution table were determined, the Kolgomorov-Smirnov normality test was carried out., Pearson's Chi-Square test and Fisher's Exact test, Kappa index as a measure of agreement, multivariate analysis through: Multinomial Logistic Regression, Multiple



Sospecha:	No identificada
Forma:	Exofítica
Tamaño:	3 x 3 x 5 mm
Color:	Rosada
Superficie:	Lisa
Consistencia:	Blanda
Tiempo de evolución:	No sabe
Forma de evolución:	Estable
Sintomatología:	Asintomática
Bordes:	Definidos
Localización:	Borde lateral derecho de lengua
Observaciones:	Hipótesis Dx: Fibroma

FIGURE 1
Platform module interface, 3D standardized dental model, and 3D standardized intraoral and soft tissues examination. On the left side, the three-dimensional modeling depicts the most prevalent occlusion in the studied sample, using a standardized 3D phantom contained within the TEGO platform. On the right, there is the 3D phantom developed on the TEGO platform, which allows for generating a general representation of the intraoral soft tissues (mucous membranes and tongue), labeled in this case with a presumptive diagnosis of Fibroma; the interface enables remote evaluation and communication with an oral pathology specialist, through teleconsultation using the TEGO platform.

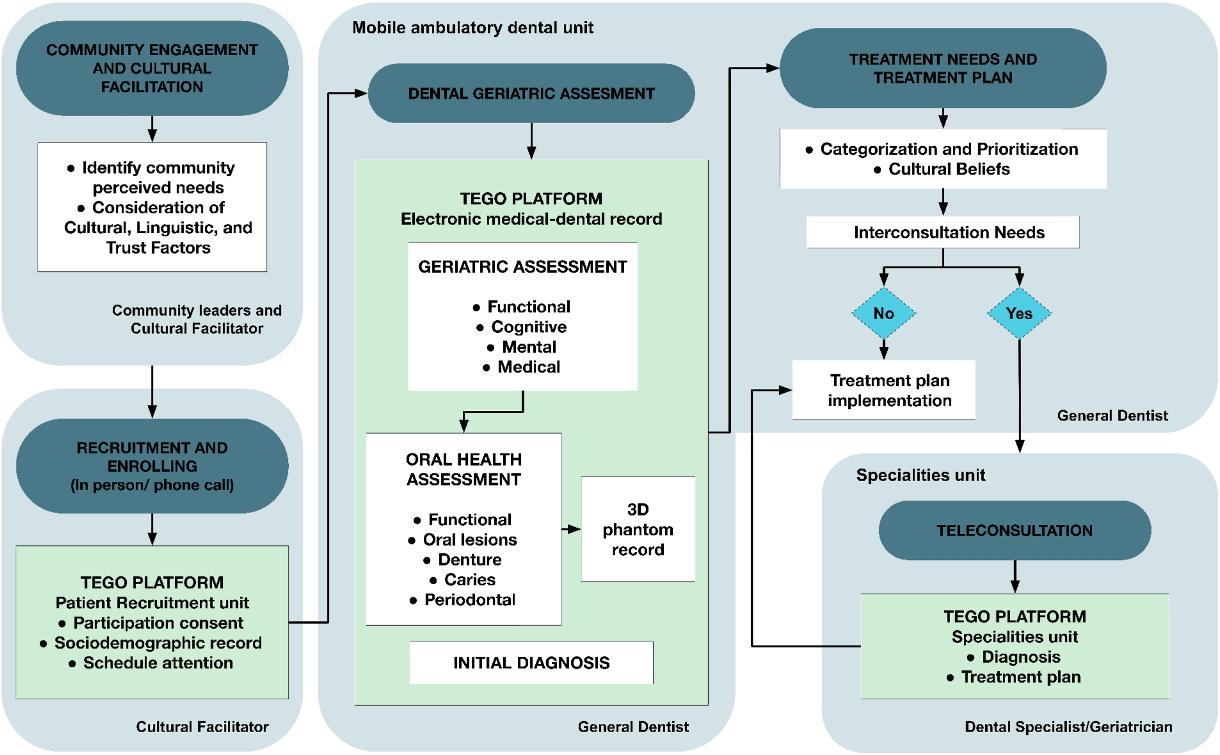


FIGURE 2
TEGO comprehensive care pathway. Operational flowchart of the attention and platform use workflow to support dental-medical care for older adults in a rural Mapuche community. Includes the Macroprocess description, the Units, and people involved in its development.

Correspondence Analysis and k-means Cluster Analysis. The SPSS statistical program (Statistics for Windows, version 23.0, IBM) was used to analyze the data.

Statistical significance was established at *p*-values <0.05. All statistical analyses were conducted employing SPSS software, version 23.

3 Results

At the conclusion of the survey, data were collected from a total of 76 participants. The participants had an average age of 70.55 years (SD 7.33), with 51.3% of them being women. Half of the older adults (51.4%) fell within the age range of 65 to 75 years. Most participants (78.9%) had 8 ears or less of formal education and were classified in the most vulnerable level of the RSH (98.4%). Almost half of the respondents considered themselves active by engaging in agriculture work (47.2%).

3.1 Geriatric assessment

In the mental and emotional evaluation, 20 participants (26.3%) had cognitive deterioration, and 32.9% were at risk of depression. The participants had a poor history of medical control, with 65.5% being overweight, 48.7% having uncontrolled blood pressure, and 18.4% having hyperglycemia. The average number of chronic diseases per participant was 1.58 (95% CI 1.27; 1.88), and 48.7% of them had multimorbidity. The daily consumption of medications varied from 0 to 10, with an average of 2.17 (95% CI 1.68; 2.66). As for functional status, almost all participants were entirely independent (93.4%). In terms of physical frailty, only one participant was classified as robust, while 51.3% exhibited frailty. Additionally, 39.5% of participants reported a history of falls in the past year.

3.2 Lifestyle characteristics

Regarding oral health behaviors, a considerable proportion (90.8%) of participants engaged in regular tooth brushing. The majority brushed their teeth two (37.8%) or three (32.4%) times a day and 26.2% flossed their tooth. The consumption of sugary items between meals was present in 28.9% of the participants, with an average of around two instances per day (mean 1.87; 95% CI 1.49; 2.23). Tobacco usage was observed in merely six individuals (7.9%). Additionally, 47.4% of participants had undergone a dental visit within the past year.

3.3 Dental caries experience

All the participants had a history of dental caries, as measured by the DMFT index, ranging from 13 to 32, with a mean of 25.96 (95% CI 24.88; 27.05) affected teeth. They had an average of 1.09 healthy teeth (95% CI 0.49; 1.69), 2.52 coronal caries (95% CI 1.89; 3.14), and 0.36 root caries (95% CI 0.16; 0.56) per teeth. Additionally, they had an average of 3.75 restorations (95% CI 2.74; 4.76) and six non-carious lesions (95% CI 5.12; 6.87).

3.4 Tooth loss

Concerning dentition status, the mean number of present teeth was 9.67 (95% CI 7.95; 11.39), 13 participants (17.1%) were fully edentulous, and only 10.7% of participants had functional dentition. Among those with partial dentition, the mean number of occluding pairs was 2.33 (95% CI 1.52; 3.15). The TEGO platform enabled the three-dimensional modeling of the most prevalent occlusal characteristics in the studied sample, with the most frequent findings being total maxillary edentulism and preservation of the lower anterior sextant (Figure 1).

3.5 Periodontal health

The prevalence of periodontal diseases was found to be substantial. Among the participants, 6.7% exhibited healthy periodontal conditions, whereas 10% presented with gingivitis. Notably, periodontitis was prevalent in 83.3% of participants.

3.6 Other oral manifestations

A total of 63.2% of participants self-reported experiencing symptoms of dry mouth, while nearly one in five (19.7%) reported having swallowing difficulties.

Both xerostomia and swallowing difficulty were found to exhibit significant correlations with each other ($p < 0.05$). Furthermore, they demonstrated correlations with multimorbidity ($p < 0.05$) and depression ($p < 0.01$). Additionally, swallowing disorders were also correlated with the number of medications consumed ($p < 0.01$).

30 oral mucosal lesions were found, which manifested in almost one-third of the participants ($n = 24$, 31.58%). Table 1 illustrates the distribution of all mucosal disorders observed in the sample. The most frequent anatomical location was Palate (33.3%), followed by Lower lip (26.7%). For 26.3% of the participants, a teleconsultation to oral medicine was conducted through the TEGO platform (Figure 1). The majority of cases were managed with conventional dental treatment, pharmacological management, or control and education. Biopsies were required for six cases.

3.7 Treatment needs

The average number of treatments performed was 3.79 [95% CI 3.57; 4.01]. Among the participants, 44.7% required fillings, with a mean of 2.14 [95% CI 1.52; 2.77] restorative treatments per tooth. This need for restorative treatment increased among the 75 years and older age group, as shown in Table 2. Extraction was indicated in 11.9% of participants. Almost all participants received preventive and promotional treatments, including oral health promotion (97.4%), scaling and dental prophylaxis (76.3%), and approximately one in five (19.7%) underwent fluoride varnish treatment.

Prosthetics were used by 76.3% of the participants, with 26.8% of individuals seeking consultations for issues related to prosthetics. Most dentures exhibited inadequacies, primarily attributed to cracks, fractures, or poor fit, needing repairs or replacements in a substantial proportion of the participants (85.5%). Relining and repair of the prosthetics were undertaken for 45% of the participants.

TABLE 1 Prevalence of oral mucosal lesions.

Classification	Oral mucosal lesion	n	%
Denture related lesion	Denture stomatitis	6	20.00
	Denture-associated inflammatory fibrous hyperplasia (Epulis Fissuratum)	3	10.00
Oral potentially malignant disorder	Actinic cheilitis	2	6.67
	Leucoplakia	2	6.67
	Erythroplakia	1	3.33
Vascular lesion	Vascular malformation	5	16.67
Tumors and tumor-like lesions	Irritation fibroma	4	13.33
Pigmentations	Solitary pigmented lesions	1	3.33
	Multiple-pigmented lesions	1	3.33
Immune-mediated conditions	Benign migratory glossitis	1	3.33
	Recurrent aphthous stomatitis	1	3.33
Physical and chemical injuries	Traumatic ulcerations	1	3.33
	Traumatic erythema	1	3.33

The need for dental prosthetics was significantly associated with overweight ($p < 0.05$) and sugar intake between meals ($p < 0.05$). The number of teeth requiring restoration showed a significant relationship with hyperglycemia ($p < 0.05$). Extractions were significantly correlated with multimorbidity ($p < 0.01$).

3.8 Sociomedical geriatric state and tooth loss

A multinomial logistic regression analysis was conducted to explore the relationship between geriatric health variables, categorized into four dimensions (social, functional, medical, and dental), and tooth loss-related variables, specifically the number of remaining teeth and the number of occluding pairs. The statistically significant models identified allow for predictions to be made about the studied variables by considering their geriatric characteristics. As illustrated in Table 3, the analysis of social factors reveals that individuals aged 65 and older with lower educational attainment (8 years or less) tend to exhibit a reduction in the number of teeth, with an estimated average decrease of 9.07 teeth. In the mental health assessment, the presence of depression and cognitive decline are associated with a decrease in the average number of remaining teeth and occluding pairs, respectively. Furthermore, a notable disparity emerges in the number of teeth among individuals who consume substantial amounts of sugars between meals, with an estimated difference of approximately 17 teeth.

A final multinomial regression analysis was conducted, incorporating variables that had been identified as statistically significant within each dimension. This culminated in the development of a conclusive model, capable of predicting the key variables pertaining to comprehensive geriatric assessment that warrant evaluation to ascertain the count of remaining teeth. These variables included age categorization, education level, frailty, cognitive decline, depression, the number of daily medications, and the frequency of sugar intake between meals

$$\left(Y = 16.00 + 0.96 * AgeCategorization - 3.71 * Education - 1.32 * Frailty - 0.59 * CognitiveDecline - 3.14 * Depression - 0.65 * Medications - 0.85 * FrequencySugarIntake;\ p = 0.013\right).$$

Additionally, age categorization, uncontrolled blood pressure, and the frequency of sugar intake between meals emerged as pertinent factors for determining the number of occluding pairs

$$\left(Y = 2.66 + 2.09 * Age\,Categorization - 1.19 * Uncontrolled\ Blood\ Pressure - 0.49 * Frequency\ Sugar\ Intake;\ p = 0.013\right).$$

In the multiple correspondence analysis (MCA), a two-dimensional model was obtained. Dimension 1 explains most of the model and includes variables such as remaining teeth, depression, cognitive decline, number of medications, frailty, age, and education level. Dimension 2 is composed of frequency of sugar intake and uncontrolled blood pressure. Figure 3A depicts clear and distinct patterns of association among variable categories in the MCA, revealing the formation of four clusters based on the number of remaining teeth. These clusters where further analyzed through k-means cluster analysis, identifying some common characteristics shared among the groups. However, two distinct groups stand out: the edentulous group (0 teeth) and the functional dentition group (21 and more teeth). The edentulous group comprises individuals with older age (65 and above), cognitive decline, depression, frailty, and polypharmacy. These characteristics, except for polypharmacy and cognitive decline, are also observed in the second group of patients with severe tooth loss (1–9 remaining teeth). Additionally, in alignment with the Multinomial logistic regression analysis, a noticeable association becomes evident in the fourth group, linking higher educational attainment, the absence of frailty, cognitive decline, depression and uncontrolled blood pressure, lower medication, and lower sugar consumption between meals, with the presence of functional dentition (Figures 3A,B).

4 Discussion

This study stands as one of the limited inquiries investigating the oral health condition, treatment requirements, and their interrelation with the socio-medical conditions of older Indigenous individuals in Chile. These objectives are distinctive because they concentrate on an often-overlooked group of older Indigenous individuals who should be assessed independently, rather than being grouped with other cohorts. This distinction is vital because the oral health status and disparities among Indigenous communities often go unnoticed in broader population metrics due to factors such as inadequate representation or constraints stemming from survey methodologies (59). Our findings reveal a significant oral health burden, marked by elevated rates of dental caries, periodontitis, dry mouth, and tooth loss. In addition, there is a substantial load of chronic diseases,

TABLE 2 Distribution of DMFT index, number of remaining teeth, number of occluding pairs, and number of teeth with restorative needs based on sociodemographic and medical-geriatric-dental characteristics.

	Variables	*n* = 76	DMFT Mean (SD)	Remaining teeth Mean (SD)	Occluding pairs Mean (SD)	Restorative needs Mean (SD)
Sex	Female	39	26.63(3.25)	11.20 (6.19)	2.20 (2.81)	1.63 (1.88)
	Male	37	25.65 (4.76)	12.72 (6.83)	2.34 (3.30)	2.76 (2.92)
Age groups	60–64 years	21	25.55 (4.11)	12.65 (6.51)	2.25 (2.65)	2.05 (2.35)
	65–74 years	39	26.47(4.34)	11.10 (6.68)	2.03 (3.33)	1.93 (2.42)
	75 years and more	16	26.44 (3.13)	13.22 (6.20)	3.11 (2.98)	3.33 (3.00)
Education level	8 or fewer years of education	57	26.54 (3.87)	11.05 (6.14)	1.80 (2.46)	1.83 (2.29)
	9 or more years	19	25.28 (4.46)	14.00 (7.03)	3.33 (3.93)	3.00 (2.81)
Currently employed	Yes	34	25.63 (3.38)	12.30 (5.57)	2.22 (2.62)	2.30 (2.43)
	No	42	26.59 (4.56)	11.66 (7.28)	2.31 (3.38)	2.09 (2.58)
Frailty phenotype	Pre-frail	36	26.43 (4.12)	12.70 (6.77)	2.93 (3.24)	2.67 (2.82)
	Frail	39	25.64 (3.95)	11.43 (6.20)	1.60 (2.74)	1.75 (2.05)
Cognitive decline	Yes	20	26.61 (3.55)	10.61 (4.52)	1.23 (2.01)	2.08 (2.46)
	No	56	26.02 (4.22)	12.33 (6.96)	2.57 (3.22)	2.22 (2.53)
Depression	Yes	25	27.21 (4.32)	9.43 (6.34)	1.57 (2.47)	1.70 (2.09)
	No	51	25.82 (3.97)	12.73 (6.42)	2.49 (3.18)	2.33 (2.61)
Multimorbidity	Yes	37	25.64 (4.49)	11.56 (6.77)	2.16 (3.05)	1.32 (2.06)
	No	39	26.53 (3.73)	12.23 (6.39)	2.35 (3.06)	2.82 (2.62)
Polypharmacy	Yes	27	27.518 (5.13)	8.15 (7.75)	1.48 (2.47)	1.39 (2.09)
	No	49	27.42 (4.04)	9.7 (7.14)	1.60 (2.86)	2.22 (2.33)
Dental care received during the past year	Yes	36	26.38 (4.07)	13.31 (6.45)	3.24 (3.16)	2.86 (2.56)
	No	40	25.93 (4.10)	10.63 (6.39)	1.33 (2.63)	1.53 (2.28)
Dry mouth	Yes	48	26.34 (4.23)	11.13 (6.67)	2.00 (3.10)	2.34 (2.61)
	No	28	25.81 (3.80)	13.43 (6.07)	2.76 (2.91)	1.90 (2.30)
Periodontitis	Yes	50	26.14 (4.03)	12.02 (6.62)	2.47 (3.14)	2.18 (2.54)
	No	10	26.20 (4.42)	11.69 (6.26)	1.30 (2.36)	2.20 (2.35)

encompassing cognitive deterioration, an increased risk of depression, the presence of multiple chronic conditions, and a prevalent state of frailty. This investigation sheds light on the significant disparities in both oral and overall health among Indigenous older adults, influenced by factors such as age, education, and lifestyle.

In examining caries experience, our findings reveal a substantial DMFT index of 26.47 within the 65 to 74 age group, indicating a noteworthy prevalence of dental caries in this demographic. While this index is lower than that reported in a Mapuche-Huilliche community in Chile (28.2) (60) and South American Indigenous groups (28.19) (9), it remains higher than figures from national studies (21.57–25.68) (57, 61–63), a trend that has been consistently observed in previous research (8, 9). Unlike many studies that focus primarily on investigating caries experience, this research provides a broader perspective by delving into the comprehensive oral health status of this community, revealing that health inequalities extend beyond caries to encompass tooth loss, periodontal disease, and xerostomia. Notably, when comparing the functional dentition of the 65–74 age group (5.3%) with figures from national surveys (30.2%) (64), a substantial disparity becomes evident. Likewise, significant discrepancies are observed in the prevalence of periodontitis when compared to Chilean non-Indigenous populations (43.3% vs. 28.8%) (65). While the challenges of periodontal epidemiological comparisons due to divergent case definitions are acknowledged, our study's outcomes align with those reported in a recent systematic review (7).

Consistent with these findings, the prevalence of dry mouth perception was notably higher (63.2%) than that reported in independent older populations (ranging from 13 to 40%) (66, 67). As for the prevalence of oral mucosal lesions, it is lower than previously reported in cross-sectional national studies (ranging from 34.2 to 53%) (54, 68, 69). Nevertheless, the most prevalent lesion in the participants is consistent with prior findings, as denture stomatitis emerges as the most common condition (54, 69, 70). It is noteworthy that oral potentially malignant disorders comprised 16.67% of all the OML, with a prevalence in our sample of 6.58%, which is higher than the reported in Chilean non-Indigenous populations (1.13–4.7%) (32, 54, 68, 70), and worldwide (4.47%) (71). One possible theory for this could be a greater accumulation of risk factors, such as increased sun exposure due to agricultural activities, local traumatic factors like ill-fitted prosthetics, poor oral health, and dietary elements like maté drinking (72, 73), underscoring the importance of oral health

TABLE 3 Multinomial logistic regression analysis of dentition variables in relation to geriatric assessments.

Social assessment				
Age categorization	8 or fewer years of education	Number of remaining teeth[a]	Number of occluding pairs[b]	*p*-value
60–64 years	Yes	11.16	-	0.006
	No	16.34	-	
≥65 years	Yes	7.27	-	
	No	12.44	-	
-	Yes	-	1.37	0.017
	No	-	3.16	
Functional assessment				
Frailty phenotype		Number of remaining teeth	Number of occluding pairs[c]	*p*-value
Yes		-	1.15	0.032
No		-	2.56	
Mental assessment				
Depression	Cognitive decline	Number of remaining teeth[d]	Number of occluding pairs[e]	*p*-value
Yes	-	6.00	-	0.002
No	-	11.50	-	
-	Yes	-	0.80	0.048
-	No	-	2.28	
Medical assessment				
Number of daily medications	Uncontrolled blood pressure	Number of remaining teeth[f]	Number of occluding pairs[g]	*p*-value
0	-	11.68	-	0.022
1	-	10.76	-	
2	-	9.85	-	
5	-	7.11	-	
7	-	5.28	-	
10	-	2.55		
-	Yes	-	1.05	0.019
	No	-	2.58	
Dental assessment				
Frequency of sugary intake between meals		Number of remaining teeth[h]	Number of occluding pairs	*p*-value
0		18.02	-	0.010
1		13.69	-	
2		9.36	-	
3		5.04	-	
4		0.71	-	

[a] Y = 12.44 + 3.89*Age categorization - 5.18*Educational level; [b] Y = 3.16–1.78*Educational level; [c] Y = 2.56–1.40*Frailty; [d] Y = 11.50–5.50*Depression; [e] Y = 2.28–1.48*Cognitive decline; [f] Y = 11.68–0.91*N° of medications; [g] Y= =2.58–1.52*Uncontrolled pressure; [h] Y = 18.02–4.33*N° sugary intakes.

promotion for the prevention of risk factors and oral self-examination in this population. In the context of oral potentially malignant disorders, age could be added as a risk factor, considering that this study focused on older adults (60 years and older), and, according to literature, Leucoplakias, as one of the main potentially malignant disorder are found in patients over 45 years old (74). In the same context, age also could predict probable malignization, considering that epithelial dysplasia is more frequently found in individuals over 41 years old (75). Predictive instruments like nomograms have been developed with the aim of predict malignant transformation of Leucoplakias, concluding that this transformation occurs mainly in patients over 50 years old (76). However, it is essential to approach

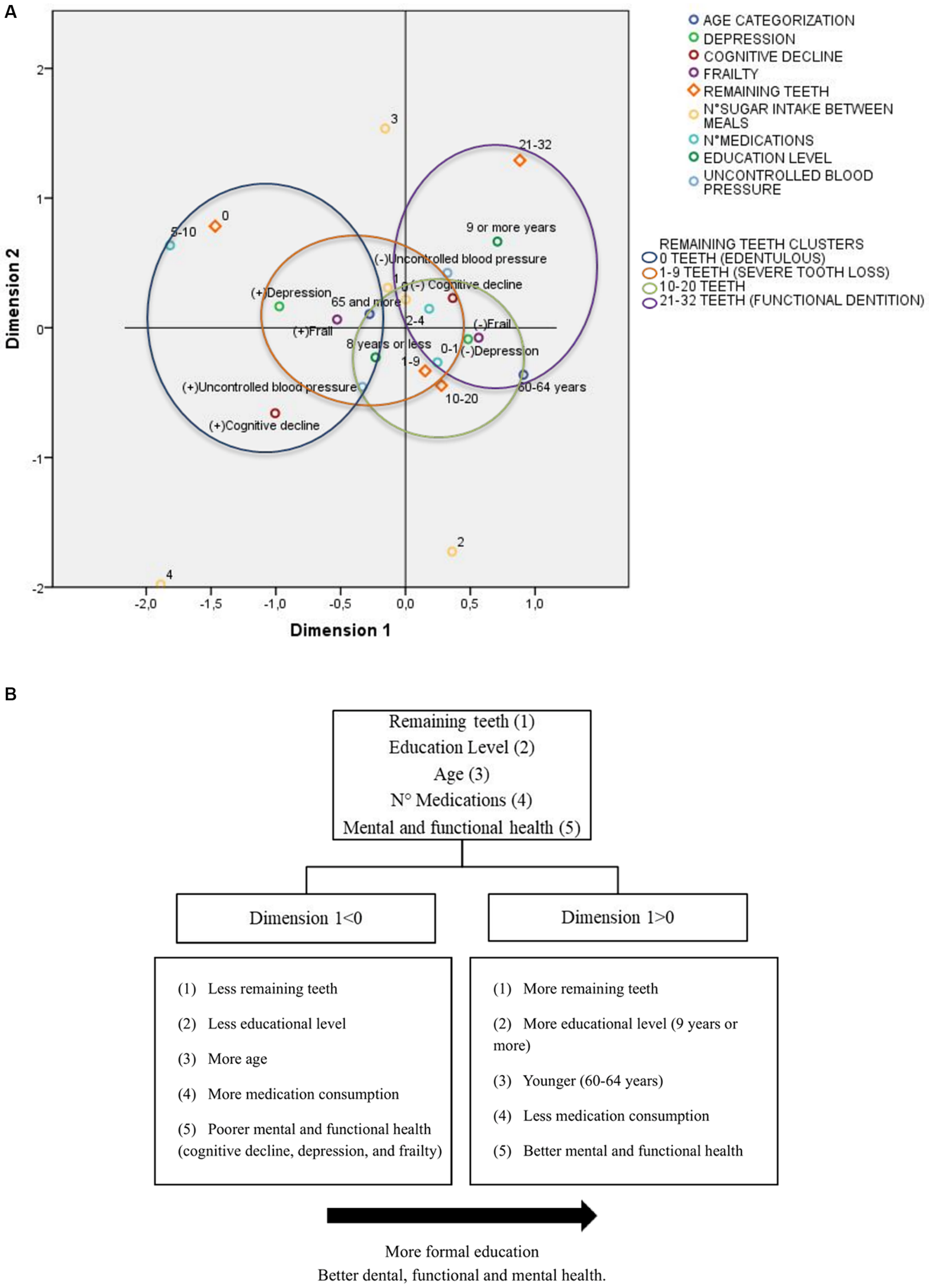


FIGURE 3
Profiles of remaining teeth using multiple correspondence analysis and k-means cluster analysis, grouped by geriatric assessment characteristics. **(A)** MCA dimensions discrimination measures grouped in clusters by the number of remaining teeth (0, 1–9, 10–20, and 21–32) by k- mean cluster analysis. **(B)** Positive and negative centroid coordinates for dimension 1.

these results with caution as they were based on clinical diagnosis, and histopathological confirmation is required. To the best of our knowledge, there is no existing evidence on the prevalence of swallowing disorders in Indigenous populations, as they are not routinely identified. The prevalence found in this study is in line with previous global studies, affecting approximately one in every five older individuals (77). Correlations were found with medication consumption, dry mouth, depression and multimorbidity. As the relationship between this geriatric syndrome and oral health conditions remains relatively unexplored (78), it is advisable for future studies to not only include screening but also to broaden their scope, considering its significant impact on health and quality of life. This expansion could encompass clinical evaluations, such as assessing temporomandibular joint disorders (79) and evaluating intra-oral compartment pressures (80).

The current study has identified a robust connection between the number of remaining teeth and several key factors, including age, education, mental and physical health, medication usage, and meal frequency. Individuals with fewer teeth tended to be older, have lower levels of education, experience more mental and physical health issues, consume a greater number of daily medications, and have a higher meal frequency. This observation is consistent with previous research findings in this age group (81–84).

Additionally, it has been reported that as certain Indigenous communities transition from traditional to more contemporary lifestyles, there is a concerning and rapid increase in lifestyle-related illnesses, such as obesity, cardiovascular diseases, and type 2 diabetes (85). Some of the issues highlighted by these findings underscore the importance of health promotion. This is because the primary drivers of complex noncommunicable diseases (NCDs), such as caries, periodontitis, oral cancer, diabetes, hypertension, and others, include an unhealthy diet, reduced physical activity, and tobacco usage (86, 87). Through our investigation, we have uncovered a compelling correlation between treatment requirements and both nutritional status and chronic conditions associated with dietary patterns. One plausible explanation for this phenomenon is the direct impact of the number of remaining teeth on dietary intake quality (88). For instance, a population requiring significant prosthodontic treatment due to tooth loss may have a reduced ability to chew and process certain foods, leading to dietary choices that rely on easily consumable carbohydrates and excessive sugar intake.

The use of technology to deliver care to remote settings has been successful in addressing accessibility barriers for Indigenous older adults (89–91). In this case, the integrated technological ecosystem in TEGO enabled teleconsultation and the provision of specialist guidance to patients. Additionally, it helped reduce the burden on the healthcare system by addressing a large number of low to moderate complexity treatment needs, while also decreasing the need for secondary care consultations, as only those who truly required specialist evaluation had to physically attend a healthcare facility. The integration of electronic records, as exemplified by the TEGO platform, played a pivotal role in enhancing the quality of care provided. This technology, empowered students to perform comprehensive assessments of rural older adults, guiding them in tailoring treatments based on the medical-geriatric status of the patients they evaluated. This community-based intervention also exposed them to the realities of rural practice and service conditions, preparing them to deal with situations they may not have encountered during their training, with remote support from dental specialist to address more complex queries. This approach aligns undergraduate curricula with the health needs of rural communities (92). Furthermore, our study draws parallels with the positive outcomes observed in rural Australia, where a collaborative endeavor between universities and the local community established a student-led clinic service improving access to timely and less invasive care for Indigenous populations (93). These outcomes underscore the potential for technology-driven solutions to bridge healthcare gaps and optimize the delivery of care to underserved communities, including Indigenous older adults in our study.

Considering that oral health disparities persist as a public health concern not only in Chile but also in various regions globally (94–96), this study underscores the necessity for a comprehensive analysis of factors contributing to health inequalities in the studied population. These factors include the historical socioeconomic determinants that have consistently shaped the life trajectories of this specific cohort (97), alongside with the rapid adoption of detrimental lifestyle habits. The study's identification of a high burden of oral disease, significant unmet oral healthcare needs, particularly in terms of restorative and prosthetic treatment; and the undeniable connection between oral health and overall well-being, highlights the importance of implementing a comprehensive and interdisciplinary healthcare approach, especially when working with Indigenous populations. This approach aligns with the Mapuche perspective on health, seamlessly integrating socio-spiritual and psycho-biological elements into their worldview and environmental interactions (13). However, integrating traditional health concepts faces challenges due to the prevailing specialization in the current healthcare system, which often hinders collaboration and regrettably overlooks the crucial principle of interdependence (98). To effectively address the substantial oral health challenges faced by Indigenous older populations dental public health programs must prioritize culturally appropriate strategies and embrace innovative, community-centered approaches (8).

While considering these observations, it is essential to recognize the constraints inherent in this study. Issues such as the scarcity of information regarding the entire population within this community, the wide dispersion of the population over a large geographical area, challenges in communication, and difficulties in transportation, led to the adoption of convenience sampling method. It has been acknowledged that studies of this kind often encounter challenges in engaging with Indigenous communities (99). Nevertheless, the involvement of an intercultural facilitator and the support from the local council facilitated the engagement of older adults' community members, especially those who exhibited hesitance in seeking medical and dental care. This approach also enabled the expression of specific concepts in Mapuzugun. The significance of community involvement and consideration of Indigenous perspectives has been previously emphasized (100, 101), and in this case, the program was approved by community leaders but designed and executed without active community participation, highlighting an area of priority for building trust and partnership in future initiatives. It is worth noting that the older individuals who participated were those with the means to travel to the remote care setting, potentially introducing a participation bias. Moreover, this subgroup might display varying levels of disease compared to more susceptible segments, like dependent older adults. Consequently, while this dataset provides valuable insights, it primarily offers preliminary information and should not be construed as indicative of the broader Mapuche older adult population. This is because the distinct cultural and social characteristics of Indigenous peoples hinder the generalizability to other communities. Additionally, this study is constrained by inherent limitations stemming from its cross-sectional design, impeding the establishment of causal relationships, and rendering it incapable of assessing the effectiveness of the employed program. This underscores the necessity for continued investigation in subsequent research efforts and partnership with local communities.

The study introduces an innovative approach to oral healthcare for Indigenous rural older adults, integrating a technological ecosystem and human capital development. This approach provides essential information to update epidemiological profiles of the Mapuche population. The intricate interplay among the presented variables presents a compelling opportunity for national and international healthcare policies to prioritize this group. Efforts should be directed at implementing a comprehensive and integrated horizontal approach of healthcare strategies, supported by technology, aimed at promoting oral health and endorsing the dignified aging of this population. While we believe that existing programs can benefit from the use of technology and home/community-based care, it is crucial to acknowledge the challenges related to technical capacity, structural limitations, and organizational support for an e-health venture in a rural setting, such as internet connectivity, relocation of healthcare personnel and equipment transport, among other factors. For the implementation of these strategies, it is necessary to encompass suitable and culturally competent interventions for oral health education and promotion, preventive, and treatment measures, as well as effective and sustainable financial resource and organizational planning characterized by a long-term view with the aim of preserving both the remaining dentition and supporting structures. These pilot initiatives hold significant potential for improving the oral health of Indigenous populations and, ultimately, for addressing disparities in oral healthcare. Furthermore, there is an urgent need for forthcoming studies to compile regional, national, and international statistics related to Indigenous oral health. Additionally, longitudinal studies are imperative for evaluating the long-term impact of these programs on changes in Indigenous oral health.

5 Conclusion

In conclusion, this study provides valuable insights into the oral health and general well-being of older Indigenous adults residing in a rural Mapuche community in Chile. The findings reveal a high burden of oral disease, including high rates of dental caries, periodontal disease, xerostomia, oral mucosal lesions, and tooth loss. Additionally, participants exhibited complex medical-geriatric conditions such as frailty, cognitive decline, depression, polypharmacy, and multiple chronic diseases, all of which were interrelated with tooth loss variables. The implementation of a geriatric technological ecosystem in the community facilitated the resolution of low to moderate complexity pathologies and enabled remote consultations, thereby reducing the necessity for travel to health centers. To address these significant oral health disparities among Indigenous populations, dental public health initiatives could incorporate innovative approaches like teledentistry, as demonstrated in this study. These programs should prioritize oral health education, prevention, and treatment, while fostering partnerships with the communities and considering the unique needs and perspectives of Indigenous populations. The ultimate goal is to promote dignified aging and enhance the overall health and well-being of these communities.

The research group has conducted previous studies regarding patients' acceptance of this tool. In this sense, the results showed high levels of patient satisfaction after receiving priority or urgent dental care, which reached above 75% in all dimensions of the questionnaire (Access to dental care, user treatment, platform, recommendation) (102).

Although economic costs may vary due to the types of specialty coverage, material costs, and the use of specialized dental equipment, considering the estimated fees for a specialist dentist, the system allows for the optimization of the specialist's time, enabling them to accommodate their schedule asynchronously within a timeframe that suits them. In this current project, we foresee the participation of a team of specialists in different areas of dentistry, with at least half a day of dedication each week. Within the context of the platform, the service of computer support is required, which in this case is supported by 2 professionals from the Institute of Educational Informatics of our university, plus the server costs to ensure a continuous service without interruption. However, in relation to the need for a stable internet network and the challenging geography of the territory in Chile, it becomes necessary to connect the use of these technologies in the future through satellite internet, in order to create a technological ecosystem that allows for comprehensive coverage of rural territories, which are the ones that unjustly have the least coverage of specialized health services. We believe that the combination of new technologies, like the ones we have fostered with the development of this technological platform, can be applied as teleodontological strategies to enhance coverage for older adults residing in remote geographical areas, respecting their cultural visions, such as the fair access to specialized dental health care in their own territory, also facilitating their travel to major urban centers and improving the interaction between dentists working directly in the communities.

Data availability statement

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to VB, victor.beltran@ufrontera.cl.

Ethics statement

The studies involving humans were approved by Universidad de La Frontera Ethics Committee. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation in this study was provided by the participants or their legal guardians/next of kin.

Author contributions

VB: Conceptualization, Funding acquisition, Project administration, Supervision, Writing – review & editing, Investigation, Methodology. FM-S: Conceptualization, Data curation, Investigation, Methodology, Writing – original draft, Writing – review & editing, Visualization. CA: Investigation, Project administration, Writing – review & editing, Data curation, Funding acquisition, Supervision. PN: Formal analysis, Writing – review & editing, Methodology. BV: Writing – review & editing, Investigation, Supervision, Visualization. CS: Investigation, Data curation, Writing – review & editing. PU: Investigation, Data curation, Writing – review & editing. WE: Writing – review & editing, Investigation, Supervision.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

References

1. Patel J, Wallace J, Doshi M, Gadanya M, Ben Yahya I, Roseman J, et al. Oral health for healthy ageing. *Lancet Healthy Longev*. (2021) 2:e521–7. doi: 10.1016/S2666-7568(21)00142-2

2. Petersen PE, Ogawa H. Promoting Oral health and quality of life of older people - the need for public health action. *Oral Health Prev Dent*. (2018) 16:113–24. doi: 10.3290/j.ohpd.a40309

3. World Health Organization (WHO). Decade of healthy ageing. 73rd World Health Assembly Decisions. (2020). Available from: https://www.who.int/news/item/07-08-2020-73rd-world-health-assembly-decisions [Accessed September 15, 2023].

4. FDI World Dental Federation. FDI policy statement on oral health for healthy ageing: adopted by the FDI general assembly: 24 September 2015, Bangkok, Thailand. *Int Dent J*. (2016) 66:7–8. doi: 10.1111/idj.12231

5. Petersen PE, Bourgeois D, Ogawa H, Estupinan-Day S, Ndiaye C. Policy and practice the global burden of oral diseases and risks to oral health. *Bull World Health Organ*. (2005) 83:686–93.

6. Slack-Smith L. Oral health in older indigenous peoples. *Gerodontology*. (2019) 36:91–1. doi: 10.1111/ger.12417

7. Nath S, Poirier B, Ju X, Kapellas K, Haag D, Jamieson L. Periodontal disease inequities among indigenous populations: a systematic review and meta-analysis. *J Periodontal Res*. (2022) 57:11–29. doi: 10.1111/jre.12942

8. Nath S, Poirier BF, Ju X, Kapellas K, Haag DG, Ribeiro Santiago PH, et al. Dental health inequalities among indigenous populations: a systematic review and meta-analysis. *Caries Res*. (2021) 55:268–87. doi: 10.1159/000516137

9. Soares GH, Pereira NF, Biazevic MGH, Braga MM, Michel-Crosato E. Dental caries in south American indigenous peoples: a systematic review. *Community Dent Oral Epidemiol*. (2019) 47:142–52. doi: 10.1111/cdoe.12436

10. Schuch HS, Haag DG, Kapellas K, Arantes R, Peres MA, Thomson WM, et al. The magnitude of indigenous and non-indigenous oral health inequalities in Brazil, New Zealand and Australia. *Community Dent Oral Epidemiol*. (2017) 45:434–41. doi: 10.1111/cdoe.12307

11. The World Bank. *Indigenous Latin America in the twenty-first century*. Washington, DC: World Bank Group (2015) Available at: http://documents.worldbank.org/curated/en/145891467991974540/Indigenous-Latin-America-in-the-twenty-first-century-the-first-decade.

12. Anderson I, Robson B, Connolly M, Al-Yaman F, Bjertness E, King A, et al. Indigenous and tribal peoples' health (the lancet–Lowitja Institute global collaboration): a population study. *Lancet*. (2016) 388:131–57. doi: 10.1016/S0140-6736(16)00345-7

13. Cuyul A. La política de salud chilena y el pueblo Mapuche. Entre el multiculturalismo y la autonomía mapuche en salud. (2013) [cited 2023. Available from: https://asuntosindigenas.gobiernosantiago.cl/wp-content/uploads/2017/05/La-politica-de-salud-chilena-y-el-pueblo-mapuche.pdf [Accessed Aug 28 2023].

14. Mejia GC, Parker EJ, Jamieson LM. An introduction to oral health inequalities among indigenous and non-indigenous populations. *Int Dent J*. (2010) 60:212–5. doi: 10.1922/IDJ_2565Jamieson04

15. King M, Smith A, Gracey M. Indigenous health part 2: the underlying causes of the health gap. *Lancet*. (2009) 374:76–85. doi: 10.1016/S0140-6736(09)60827-8

16. Deivanayagam TA, English S, Hickel J, Bonifacio J, Guinto RR, Hill KX, et al. Envisioning environmental equity: climate change, health, and racial justice. *Lancet*. (2023) 402:64–78. doi: 10.1016/S0140-6736(23)00919-4

17. Ravaghi V, Quiñonez C, Allison PJ. The magnitude of oral health inequalities in Canada: findings of the Canadian health measures survey. *Community Dent Oral Epidemiol*. (2013) 41:490–8. doi: 10.1111/cdoe.12043

18. Hage E, Roo JP, Van Offenbeek MAG, Boonstra A. Implementation factors and their effect on e-health service adoption in rural communities: a systematic literature review. *BMC Health Serv Res*. (2013) 13:1–16. doi: 10.1186/1472-6963-13-19

19. Hudson HE. Rural telemedicine: lessons from Alaska for developing regions. *Telemed e-Health*. (2005) 11:460–7. doi: 10.1089/tmj.2005.11.460

20. Emami E, Harnagea H, Shrivastava R, Ahmadi M, Giraudeau N. Patient satisfaction with e-oral health care in rural and remote settings: a systematic review. *Syst Rev*. (2022) 11:222–34. doi: 10.1186/s13643-022-02103-2

21. Maar MA, Seymour A, Sanderson B, Boesch L. Reaching agreement for an aboriginal e-health research agenda: the aboriginal telehealth knowledge circle consensus method. *Rural Remote Health*. (2010) 10:1299. doi: 10.22605/RRH1299

22. Reilly R, Stephens J, Micklem J, Tufanaru C, Harfield S, Fisher I, et al. Use and uptake of web-based therapeutic interventions amongst indigenous populations in Australia, New Zealand, the United States of America and Canada: a scoping review. *Syst Rev*. (2020) 9:1–17. doi: 10.1186/s13643-020-01374-x

23. Elliott G, Smith AC, Bensink ME, Brown C, Stewart C, Perry C, et al. The feasibility of a community-based Mobile telehealth screening Service for Aboriginal and Torres Strait Islander Children in Australia. *Telemed e-Health*. (2010) 16:950–6. doi: 10.1089/tmj.2010.0045

24. World Health Organization (WHO). The health of indigenous peoples. (2023) A76/A/CONF./1A76/A/CONF./1: 1–5. Available at: https://apps.who.int/gb/ebwha/pdf_files/WHA76/A76_ACONF1-en.pdf

25. United Nations. *United Nations declaration on the rights of indigenous peoples*. Geneva: UN Office of the High Commissioner for Human Rights (OHCHR). (2008).

26. Pan American Health Organization. *Strategy and plan of action on ethnicity and health 2019–2025*. Washington, D.C: PAHO (2019).

27. Gizaw Z, Astale T, Kassie GM. What improves access to primary healthcare services in rural communities? A systematic review. *BMC Primary Care*. (2022) 23:313. doi: 10.1186/s12875-022-01919-0

28. Instituto Nacional de Estadísticas de Chile (INE). Radiografía de género: Pueblos originarios en Chile 2017. (2018). Available from: https://www.ine.gob.cl/docs/default-source/genero/documentos-de-an%C3%A1lisis/documentos/radiografia-de-genero-pueblos-originarios-chile2017.pdf?sfvrsn=7cecf389_8 [Accessed Oct 16 2023]

29. Ibacache J, Chureo F, MCFall S, Quidel J. *Promoción de la Medicina y Terapias Indígenas en la Atención Primaria de Salud: El Caso de los Mapuche de Makewe-Pelale*. Washington, DC: WHO/PAHO (2001).

30. Plath O. *Folklore médico chileno: Antropología y salud*. *1st* ed. Santiago: Nascimento (1981).

31. El Poblete M. Pueblo Mapuche: Breve caracterización de su organización social. Santiago; (2019). Available from: https://obtienearchivo.bcn.cl/obtienearchivo?id=repositorio/10221/27459/1/BCN___Poblete___El_Pueblo_Mapuche._Breve_caraterizacion_de_su_organizacion_social_final.pdf. [Accessed Oct 15 2023].

32. Beltrán V, Von Marttens A, Acuña-Mardones P, Sanzana-Luengo C, Rueda-Velásquez SJ, Alvarado E, et al. Implementation of a Teledentistry platform for dental emergencies for the elderly in the context of the COVID-19 pandemic in Chile. *Biomed Res Int*. (2022) 2022:9285. doi: 10.1155/2022/6889285

33. Ministerio de Desarrollo Social Gobierno de Chile. Orientaciones al RSH N° 8 Cálculo de la Calificación Socioeconómica. (2019). Available from: http://www.registrosocial.gob.cl/docs/Orientaciones-complementarias-N8_calculo-CSE_VF.pdf [Accessed Oct 15 2023].

34. Bernaola-Sagardui I. Validación del índice de Barthel en la población española. *Enferm Clin*. (2018) 28:210–1. doi: 10.1016/j.enfcli.2017.12.001

35. World Health Organization. *Oral health surveys basic methods*. 5th ed. Geneva: World Health Organization. (2013).

36. Ismail AI, Pitts NB, Tellez M, Banerjee A, Deery C, Douglas G, et al. The international caries classification and management system (ICCMS™) an example of a caries management pathway. *BMC Oral Health*. (2015) 15:1–13. doi: 10.1186/1472-6831-15-S1-S9

37. Blesa R, Pujol M, Aguilar M, Santacruz P, Bertran-Serra I, Hernández G, et al. Clinical validity of the 'mini-mental state' for Spanish speaking communities. *Neuropsychologia*. (2001) 39:1150–7. doi: 10.1016/S0028-3932(01)00055-0

38. Martínez De La Iglesia J, Herrero RD, Vilches MCO, Taberné CA, Colomer CA, Luque RL. Cross-cultural adaptation and validation of Pfeiffer's test (short portable mental status questionnaire [SPMSQ]) to screen cognitive impairment in general population aged 65 or older. *Med Clin (Barc)*. (2001) 117:129–34. doi: 10.1016/s0025-7753(01)72040-4

39. Yesavage JA, Sheikh JI. Geriatric depression scale (GDS). *Clin Gerontol*. (1986) 5:165–73. doi: 10.1300/J018v05n01_09

40. Fried LP, Tangen CM, Walston J, Newman AB, Hirsch C, Gottdiener J, et al. Frailty in older adults: evidence for a phenotype. *J Gerontol*. (2001) 56:M146–57. doi: 10.1093/gerona/56.3.M146

41. Skou ST, Mair FS, Fortin M, Guthrie B, Nunes BP, Miranda JJ, et al. Multimorbidity. *Nat Rev Dis Primers*. (2022) 8:1–22. doi: 10.1038/s41572-022-00376-4

42. Masnoon N, Shakib S, Kalisch-Ellett L, Caughey GE. What is polypharmacy? A systematic review of definitions. *BMC Geriatr*. (2017) 17:1–10. doi: 10.1186/s12877-017-0621-2

43. Belafsky PC, Mouadeb DA, Rees CJ, Pryor JC, Postma GN, Allen J, et al. Validity and reliability of the eating assessment tool (EAT-10). *Ann Otol Rhinol Laryngol*. (2008) 117:919–24. doi: 10.1177/000348940811701210

44. Fernández-Rosati J, Lera L, Fuentes-López E, Albala C. Validez y confiabilidad del cuestionario Eating Assessment Tool 10 (EAT-10) para detectar disfagia en adultos mayores chilenos. *Rev Med Chile*. (2018) 146:1008–15. doi: 10.4067/s0034-98872018000901008

45. Ismail AI, Sohn W, Tellez M, Amaya A, Sen A, Hasson H, et al. The international caries detection and assessment system (ICDAS): an integrated system for measuring dental caries. *Community Dent Oral Epidemiol*. (2007) 35:170–8. doi: 10.1111/j.1600-0528.2007.00347.x

46. Sociedad Española de Periodoncia y Osteointegración (SEPA). Available at: https://sepa.es/periodontograma/. (2022). Periodontograma.

47. Botelho J, Machado V, Proença L, Mendes JJ. The 2018 periodontitis case definition improves accuracy performance of full-mouth partial diagnostic protocols. *Sci Rep*. (2020) 10:1–7. doi: 10.1038/s41598-020-63700-6

48. Chapple ILC, Mealey BL, Van Dyke TE, Bartold PM, Dommisch H, Eickholz P, et al. Periodontal health and gingival diseases and conditions on an intact and a reduced periodontium: consensus report of workgroup 1 of the 2017 world workshop on the classification of periodontal and Peri-implant diseases and conditions. *J Periodontol*. (2018) 89:S74–84. doi: 10.1111/jcpe.12940

49. Hobdell M, Petersen PE, Clarkson J, Johnson N. Global goals for oral health 2020. *Int Dent J*. (2003) 53:285–8. doi: 10.1111/j.1875-595X.2003.tb00761.x

50. Kassebaum NJ, Bernabé E, Dahiya M, Bhandari B, Murray CJL, Marcenes W. Global burden of severe tooth loss: a systematic review and meta-analysis. *J Dent Res*. (2014) 93:20S–8S. doi: 10.1177/0022034514537828

51. Beltrán V, Acuña-Mardones P, Fernández-Gil F, Acuña-Mardones D, Engelke W. Development of a teledentistry system for the elderly in the context of the COVID-19 pandemic: 3D diagnostic models integration in a web platform. *J Oral Res*. (2022) 2022:1–7. doi: 10.17126/joralres.2022.024

52. Venegas B, Rueda S, Flores M, Cerda A, Beltrán V. Prevention of Oral cancer through the implementation of a Teledentistry platform for the elderly. Journal of Oral. *Demogr Res*. (2022) 2022:1–8. doi: 10.17126/joralres.2022.027

53. Robledo-Sierra J, Mattsson U, Svedensten T, Jontell M. The morbidity of oral mucosal lesions in an adult Swedish population. *Med Oral Patol Oral Cir Bucal*. (2013) 18:e766–72. doi: 10.4317/medoral.19286

54. Espinoza I, Rojas R, Aranda W, Gamonal J. Prevalence of oral mucosal lesions in elderly people in Santiago, Chile. *J Oral Pathol Med*. (2003) 32:571–5. doi: 10.1034/j.1600-0714.2003.00031.x

55. Woo SB. *Oral pathology: A comprehensive atlas and text*. *1st* ed. Philadelphia: Elsevier Saunders (2012).

56. Neville B, Damm D, Allen C, Chi A. *Oral and maxillofacial pathology*. *4th* ed. Missouri: Elsevier (2016).

57. Mariño R, Giacaman RA. Factors related to unmet oral health needs in older adults living in Chile. *Arch Gerontol Geriatr*. (2014) 58:454–9. doi: 10.1016/j.archger.2014.01.003

58. Redford M, Drury TF, Kingman A, Brown LJ. Denture use and the technical quality of dental prostheses among persons 18–74 years of age: United States, 1988–1991. *J Dent Res*. (1996) 75:714–25. doi: 10.1177/002203459607502S11

59. Jamieson L, Hedges J, Peres MA, Guarnizo-Herreño CC, Bastos JL. Challenges in identifying indigenous peoples in population oral health surveys: a commentary. *BMC Oral Health*. (2021) 21:1–6. doi: 10.1186/s12903-021-01455-w

60. Angel P, Mc F. Cisternas, Moncada G. Prevalencia de caries, Pérdida de Dientes y Necesidad de Tratamiento en Población Adulta Mapuche-Huilliche de Isla Huapi prevalence of caries, teeth loss and treatment needs in adult Mapuche-Huilliche population in Isla Huapi Trabajo de Investigación. *Rev Clin Periodoncia Implantol Rehabil Oral*. (2010) 3:69–72. doi: 10.1016/S0718-5391(10)70044-6

61. Urzua I, Mendoza C, Arteaga O, Rodríguez G, Cabello R, Faleiros S, et al. Dental caries prevalence and tooth loss in chilean adult population: first national dental examination survey. *Int J Dent*. (2012) 2012:810170:1–6. doi: 10.1155/2012/810170

62. Beltrán V, Flores M, Sanzana C, Muñoz-Sepúlveda F, Alvarado E, Venegas B, et al. Tooth loss and caries experience of elderly Chileans in the context of the COVID-19 pandemic in five regions of Chile. *Int J Environ Res Public Health*. (2023) 20:1–11. doi: 10.3390/ijerph20043001

63. Quinteros ME, Cáceres DD, Soto A, Mariño RJ, Giacaman RA. Caries experience and use of dental services in rural and urban adults and older adults from Central Chile. *Int Dent J*. (2014) 64:260–8. doi: 10.1111/idj.12118

64. Ministerio de Salud (MINSAL). Informe Encuesta Nacional de Salud 2016–2017. Salud Bucal. (2019) Available from: https://goo.gl/oe2iVt. [Accessed Jul 5 2023].

65. Giacaman RA, Sandoval Salas D, Bustos Alvarez IP, Rojas Cáceres MA, Mariño RJ. Epidemiología del estado de salud periodontal en la VII Región del Maule, Chile. *Revista Clínica de Periodoncia, Implantología y Rehabilitación Oral*. (2016) 9:184–92. doi: 10.1016/j.piro.2016.07.002

66. Liu B, Dion MR, Jurasic MM, Gibson G, Jones JA. Xerostomia and salivary hypofunction in vulnerable elders: prevalence and etiology. *Oral Surg Oral Med Oral Pathol Oral Radiol*. (2012) 114:52–60. doi: 10.1016/j.oooo.2011.11.014

67. Ikebe K, Nokubi T, Sajima H, Kobayashi S, Hata K, Ono T, et al. Perception of dry mouth in a sample of community-dwelling older adults in Japan. *Spec Care Dentist*. (2001) 21:52–9. doi: 10.1111/j.1754-4505.2001.tb00225.x

68. Mariño RJ, Cueto A, Badenier O, Acevedo R, Moya R. Oral health status and inequalities among ambulant older adults living in Central Chile. *Community Dent Health*. (2011) 28:143–8. doi: 10.1922/CDH_2567Mariño06

69. Cueto A, Martínez R, Niklander S, Deichler J, Barraza A, Esguep A. Prevalence of oral mucosal lesions in an elderly population in the city of Valparaiso. *Chile Gerodontology*. (2013) 30:201–6. doi: 10.1111/j.1741-2358.2012.00663.x

70. Lozano C, Vergara C, Lee X. Prevalence of oral lesions and chronic non-communicable diseases in a sample of Chilean institutionalized versus non-institutionalized elderly. *J Oral Res*. (2018) 7:108–13. doi: 10.17126/joralres.2018.025

71. Mello FW, Miguel AFP, Dutra KL, Porporatti AL, Warnakulasuriya S, Guerra ENS, et al. Prevalence of oral potentially malignant disorders: a systematic review and meta-analysis. *J Oral Pathol Med*. (2018) 47:633–40. doi: 10.1111/jop.12726

72. Kumari P, Debta P, Dixit A. Oral potentially malignant disorders: Etiology, pathogenesis, and transformation into Oral cancer. *Front Pharmacol*. (2022) 13:825266. doi: 10.3389/fphar.2022.825266

73. Dasanayake AP, Silverman AJ, Warnakulasuriya S. Maté drinking and oral and oro-pharyngeal cancer: a systematic review and meta-analysis. *Oral Oncol*. (2010) 46:82–6. doi: 10.1016/j.oraloncology.2009.07.006

74. Shah JS, Dubey J. Prevalence and factors associated with oral potentially malignant disorders and oral squamous cell carcinoma: an institutional study. *J Cancer Res Ther*. (2023) 19:S536–44. doi: 10.4103/jcrt.jcrt_759_22

75. Pires FR, Barreto ME, Nunes JG, Carneiro NS, Azevedo AB, Dos Santos TC. Oral potentially malignant disorders: clinical-pathological study of 684 cases diagnosed in a Brazilian population. *Med Oral Patol Oral Cir Bucal*. (2020) 25:e84–8. doi: 10.4317/medoral.23197

76. Wang T, Wang L, Yang H, Lu H, Zhang J, Li N, et al. Development and validation of nomogram for prediction of malignant transformation in oral leukoplakia: a large-scale cohort study. *J Oral Pathol Med*. (2019) 48:491–8. doi: 10.1111/jop.12862

77. Rech RS, de Goulart BNG, dos Santos KW, Marcolino MAZ, Hilgert JB. Frequency and associated factors for swallowing impairment in community-dwelling older persons: a systematic review and meta-analysis. *Aging Clin Exp Res*. (2022) 34:2945–61. doi: 10.1007/s40520-022-02258-x

78. Drancourt N, El Osta N, Decerle N, Hennequin M. Relationship between Oral health status and oropharyngeal dysphagia in older people: a systematic review. *Int J Environ Res Public Health*. (2022) 19:3618. doi: 10.3390/ijerph192013618

79. Gilheaney Ó, Béchet S, Kerr P, Kenny C, Smith S, Kouider R, et al. The prevalence of oral stage dysphagia in adults presenting with temporomandibular disorders: a systematic review and meta-analysis. *Acta Odontol Scand*. (2018) 76:448–58. doi: 10.1080/00016357.2018.1424936

80. Engelke W, Jung K, Knösel M. Intra-oral compartment pressures: a biofunctional model and experimental measurements under different conditions of posture. *Clin Oral Investig*. (2011) 15:165–76. doi: 10.1007/s00784-009-0367-0

81. Cannon I, Robinson-Barella A, McLellan G, Ramsay SE. From drugs to dry mouth: a systematic review exploring Oral and psychological health conditions associated with dry mouth in older adults with polypharmacy. *Drugs Aging*. (2023) 40:307–16. doi: 10.1007/s40266-023-01017-5

82. Cademartori MG, Gastal MT, Nascimento GG, Demarco FF, Corrêa MB. Is depression associated with oral health outcomes in adults and elders? A systematic review and meta-analysis. *Clin Oral Investig*. (2018) 22:2685–702. doi: 10.1007/s00784-018-2611-y

83. Xu K, Yu W, Li Y, Li Y, Wan Q, Chen L, et al. Association between tooth loss and hypertension: a systematic review and meta-analysis. *J Dent*. (2022) 123:104178. doi: 10.1016/j.jdent.2022.104178

84. Gao C, Larvin H, Bishop DT, Bunce D, Pavitt S, Wu J, et al. Oral diseases are associated with cognitive function in adults over 60 years old. *Oral Dis*. (2023):1–9. doi: 10.1111/odi.14757

85. Gracey M, King M. Indigenous health part 1: determinants and disease patterns. *Lancet*. (2009) 374:65–75. doi: 10.1016/S0140-6736(09)60914-4

86. Chapple ILC, Bouchard P, Cagetti MG, Campus G, Carra MC, Cocco F, et al. Interaction of lifestyle, behaviour or systemic diseases with dental caries and periodontal diseases: consensus report of group 2 of the joint EFP/ORCA workshop on the boundaries between caries and periodontal diseases. *J Clin Periodontol*. (2017) 44:S39–51. doi: 10.1111/jcpe.12685

87. World Health Organization. Global oral health status report: towards universal health coverage for oral health by 2030. (2022). Available from: http://apps.who.int/bookorders. [Accessed Oct 15 2023].

88. Zhu Y, Hollis JH. Tooth loss and its association with dietary intake and diet quality in American adults. *J Dent*. (2014) 42:1428–35. doi: 10.1016/j.jdent.2014.08.012

89. Poprawski DM, Adams J, Bassal A. P0225 telemedicine: a novel approach of bringing oncology care closer to the patient. *Eur J Cancer*. (2014) 50:e72. doi: 10.1016/j.ejca.2014.03.269

90. Jin AJ, Martin D, Maberley D, Dawson KG, Seccombe DW, Beattie J. Evaluation of a mobile diabetes care telemedicine clinic serving aboriginal communities in northern British Columbia. *Canada Int J Circumpolar Health*. (2004) 63:124–8. doi: 10.3402/ijch.v63i0.17871

91. Choukou MA, Maddahi A, Polyvyana A, Monnin C. Digital health technology for indigenous older adults: a scoping review. *Int J Med Inform*. (2021) 148:104408. doi: 10.1016/j.ijmedinf.2021.104408

92. Strasser R, Kam SM, Regalado SM. Rural health care access and policy in developing countries. *Annu Rev Public Health*. (2016) 37:395–412. doi: 10.1146/annurev-publhealth-032315-021507

93. March S, Mangoyana C, Oakley P, Lalloo R, Walsh LJ. Positive impacts of oral health services provision by a student-led primary care clinic to an Australian rural indigenous community. *Aust Dent J*. (2023) 68:151–9. doi: 10.1111/adj.12960

94. Peres MA, Macpherson LMD, Weyant RJ, Daly B, Venturelli R, Mathur MR, et al. Oral diseases: a global public health challenge. *Lancet*. (2019) 394:249–60. doi: 10.1016/S0140-6736(19)31146-8

95. Elani HW, Harper S, Thomson WM, Espinoza IL, Mejia GC, Ju X, et al. Social inequalities in tooth loss: a multinational comparison. *Community Dent Oral Epidemiol*. (2017) 45:266–74. doi: 10.1111/cdoe.12285

96. Borgeat Meza M, Espinoza I, Carvajal P, Cuevas R. Changes in oral health inequalities in adults in Chile. *Community Dent Oral Epidemiol*. (2022) 50:506–12. doi: 10.1111/cdoe.12701

97. Jamieson LM, Elani H, Mejia GC, Ju X, Kawachi I, Harper S, et al. Inequalities in indigenous oral health: findings from Australia, New Zealand, and Canada. *J Dent Res*. (2016) 95:1375–80. doi: 10.1177/0022034516658233

98. Stange KC. The problem of fragmentation and the need for integrative solutions. *Ann Fam Med*. (2009) 7:100–3. doi: 10.1370/afm.971

99. Tsai C, Blinkhorn A, Irving M. Oral health programmes in indigenous communities worldwide—lessons learned from the field: a qualitative systematic review. *Community Dent Oral Epidemiol*. (2017) 45:389–97. doi: 10.1111/cdoe.12302

100. McIlduff CD, Acharibasam J, Starr V, Chapados M. Engaging indigenous older adults with technology use to respond to health and well-being concerns and needs. *Healthc Manage Forum*. (2022) 35:257–64. doi: 10.1177/08404704221103521

101. Moecke DP, Holyk T, Beckett M, Chopra S, Petlitsyna P, Girt M, et al. Scoping review of telehealth use by indigenous populations from Australia, Canada, New Zealand, and the United States. *J Telemed Telecare*. (2023):1–19. doi: 10.1177/1357633X231158835

102. Beltrán V, Acuña-Mardones P, Díaz J, Alvarado E, von Marttens A. TEGO: a new concept of teledentistry for the elderly through a web platform and mobile app in the context of the covid-19 pandemic. *J Oral Res*. (2022) S:1–8. doi: 10.17126/joralres.2022.023

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Development of a culturally enhanced caregiver-facilitated language nutrition intervention “+Language is Medicine” to address developmental delay in Diné (Navajo) toddlers

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Introduction: Developmental Delay (DD) is highly common in American Indian and Alaska Native (AI/AN; Indigenous) toddlers and leads to high numbers of AI/AN children who eventually need special education services. AI/AN children are 2.89 times more likely to receive special education compared to other children in the U.S., yet developmental disorders are more frequently under diagnosed and untreated in AI/AN infants and toddlers. DD, which can be identified as early as toddlerhood, can lead to negative impacts on developmental trajectories, school readiness, and long-term health. Signs of DD can be identified early with proper developmental screening and remediated with high quality early intervention that includes effective parent training. There are many evidence-based language facilitation interventions often used in Early Intervention programs. However, in communities in rural parts of the Navajo Nation where there are limited services and resources, infants and toddlers with early signs of DD are often missed and do not get the culturally responsive support and evidence-based intervention they deserve.

Methods: The community-based +Language is Medicine (+LiM) study team partnered with tribal home visitors, community members, and a Diné linguist/elder using a collaborative virtual workgroup approach in 2021 and 2022 to present the +LiM pilot study aims and to discuss strategies for enhancing a language intervention for toddlers experiencing DD in their tribal community. This paper will detail the stages of community engagement, intervention enhancement and preparation for field testing of the +LiM intervention to address elevated rates of DD in toddlers in the Northern Agency of the Navajo Nation.

Results: Two major outcomes from this collaborative workgroup included: (1) a team-initiated redefining of language nutrition to align with Indigenous values that center cultural connectedness and native language use and (2) a five-lesson caregiver-facilitated curriculum titled +Language is Medicine which includes caregiver lessons on language nutrition, language facilitation, shared book reading, pretend play, and incorporation of native language into home routines. These two workgroup outcomes were leveraged to develop a pilot pre–/post-intervention study to test the effectiveness of the +LiM intervention with caregiver-toddler dyads living on the Navajo Nation.

Discussion: Delivering tailored child interventions through tribal home visiting are cost-effective and innovative methods for reaching reservation-based families who benefit from culturally responsive parent coaching and instruction. The +LiM team has applied a precision tribal home visiting approach to enhance methods of early intervention for children with DD. Our enhancement process was grounded in Indigenous community-based participatory research that centered culture and language.

KEYWORDS

American Indian/Alaska native, indigenous, early childhood, developmental delay, infants, toddlers, tribal home visiting, indigenous language nutrition

1 Introduction

> "You must speak straight so that your words may go as sunlight into our hearts."
> ~ Cochise, Chiricahua

American Indian and Alaska Native (AI/AN; Indigenous) people make up highly language-rich communities that are full of stories, ceremonies, celebration, and humor. These communities continue to thrive through their oral traditions despite a long history of trauma and genocide brought on by outsiders who colonized their traditional homelands. Today, the effects of these traumas continue to impact parental responsiveness and attachment style which negatively impacts childrearing practices and communications styles with young children (1). It is through this context that we must explore child well-being and work toward trauma-informed and culturally responsive early childhood interventions that can steer early developmental trajectory in positive directions. Language-rich experiences are common in AI/AN households with strong oral traditions, Furthermore, it is established that these Indigenous families who maintain strong connections to tradition often engage in cultural routines that bring together family and community in which there is frequent language exchange. Therefore, this study team sought a family and community-level intervention approach that built on increasing positive parenting, attachment with young children, and increase of language nutrition. Language nutrition is a concept used to describe early language exposure that is rich in quality and quantity and aligns with a family's cultural and home routines (2). It is through the concept that this study and the cultural enhancements of early language facilitation strategies were planned to meet the rising incidence of developmental delay (DD) on the Navajo Nation.

1.1 Background

DD, which is defined as significant delays in two or more developmental domains of communication, problem solving, social emotional, motor, and self-help (3), has become highly common in AI/AN infants and toddlers. This has led to an overrepresentation in Individuals with Disabilities Education Act (IDEA) Part B and C programs with the higher incidence staying high following the COVID-19 pandemic. AI/AN children are 2.89 times more likely to receive special education services compared to other children in the United States (U.S.), yet neurodevelopmental disorders, including developmental delay, are more frequently underdiagnosed and untreated in AI/AN children (4, 5), DD, which can be identified as early as infancy and toddlerhood, can lead to negative impacts on developmental trajectories, school readiness, and long-term health (6, 7). Signs of DD can be identified early with proper screening and remediated with high quality early intervention that includes parent training (8). However, unequitable policies in states with many AI/AN children result in many referred children missing the opportunity for early intervention services. For example, in Arizona, children must have an established medical condition or a delay of at least two standard deviations (approximately 50 percent below the mean) in at least one developmental area, resulting in 66% of referred children not qualifying for services (9). For children who do qualify, their families often face long waiting times due to a lack of providers and system barriers that lead to delayed intervention and culturally responsive parental supports. Risk factors such as low socioeconomic status, low parental responsivity, and language-poor environments can limit early brain functioning and language development (10). However, protective factors include high parental responsivity, high print exposure, longer reciprocal caregiver-child interactions, and increased exposure to rich language and foster timely achievement of developmental milestones (11). Figure 1 (12), depicts how risk factors reduce long-term developmental trajectory and highlights the optimal timing for intervention to reduce risk factors for DD is shortly after birth to age 5 (13).

Addressing DD in early childhood can help prevent health inequities that manifest throughout adulthood. Risk factors for DD include decreased structured play with caregivers and reduced parent responsiveness. Children with DD encounter multiple academic and social challenges elevating their risk for chronic and behavioral health conditions later in life. The link between early developmental trajectories and health inequities is well established (14–16) to address this significant disparity, earlier identification of DD and expanded opportunities for early remediation are needed.

The Navajo Nation, the largest reservation in the United States, is a vast and culturally rich territory spanning portions of Arizona, New Mexico, and Utah. Home to the *Diné* people, the nation boasts a population that surpasses 300,000, making it one of the most populous Indigenous communities. Known for their strong emphasis on

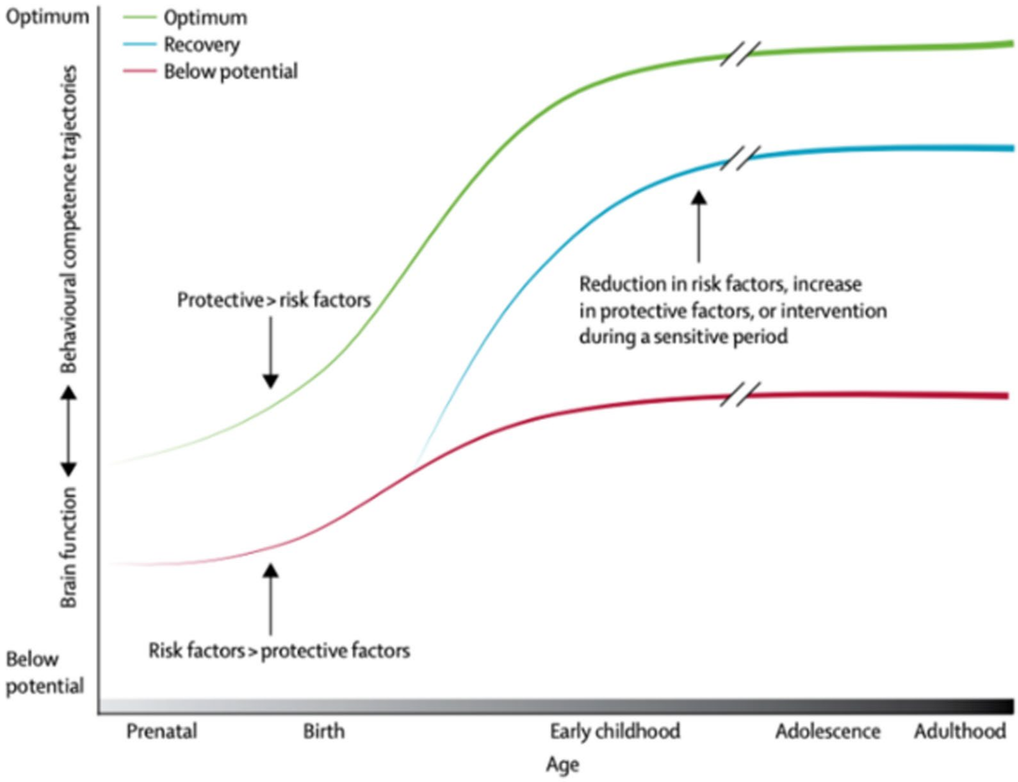

FIGURE 1
Brain function and behavioral competency trajectories across the lifespan.

traditional values of lifelong learning and reflection, the Diné people have a deep connection to the land, with a rich cultural heritage reflected in their art, language, and ceremonies. The Navajo Nation is renowned for its unique landscape, cornfields, valleys, and canyons. From early creation through present day, the Diné have always demonstrated resilience through initiatives focused on education, healthcare, language revitalization, and community-leveraged public health solutions. The community's strengths lie in its commitment to preserving its language, fostering cultural continuity, and implementing sustainable practices to improve the well-being of all Diné, especially in young children (17). The demographics of the Navajo Nation showcase a diverse population, with a blend of traditional and contemporary elements contributing to the unique fabric of this large sovereign nation. A core Diné philosophy of Hózhó can help to conceptualize Diné wellness and beliefs of living intentionally and in harmony with one's surroundings. This traditional teaching is used as a guide for how to organize the thoughts, actions, behaviors, and speech of the Diné people (18–21). When thinking about potential public health initiatives to improve child and family well-being, these characteristics, strengths, and ways of life must be considered when attempting to deliver evidence-based interventions (EBIs) in Diné households. Therefore, the overarching goal, when attempting to address early indicators of DD should be to provide evidence-based services that are culturally tailored and meet the whole child and their families (22). To accomplish this, there must be existing programmatic and workforce infrastructure in place in which the enhanced intervention can be delivered. Family support programs delivered by non-clinical, community-based providers, including Tribal Home Visiting (THV) programs, hold great promise for reaching families in underserved AI/AN communities in an effective way.

1.1.1 The power of tribal home visiting

Home Visiting has been recognized as a highly effective approach for promoting cognitive and social–emotional development in young children, with multiple program models showing positive outcomes with significant effect sizes, across diverse populations. THV programs specifically have shown that training community members (who are familiar with cultural beliefs, historical trauma, and language preferences of an Indigenous community) as home visitors, is a highly effective strategy for delivering health and developmental interventions (10, 11, 13, 23, 24). Further, THV programs, which are much more accessible than specialty early intervention clinical services, address social determinants of health in AI/AN communities, and are generally well-received – suggesting they could be an effective platform for dissemination of culturally-tailored parent coaching interventions, such as +LiM, to address early signs of DD. Family Spirit is a THV program developed by the Johns Hopkins Center for Indigenous Health (JHCIH) in collaboration with the Navajo, White Mountain Apache, and San Carlos Apache tribal communities. Family Spirit is currently the only THV model categorized as evidence-based for AI/AN communities and is delivered by community-based Health Educators to support caregivers during pregnancy and early childhood. +LiM is structured to mirror the core elements of the Family Spirit home visitation model which include relationship building, defined home visitation methods, utilization of strengths-based approaches, and defined structures for delivering the material. When considering the variety of systems and services that address early childhood needs, THV emerges as a valuable but largely untapped secondary prevention approach to mitigate early developmental risks within AI/AN children, harnessing the strength of culturally sensitive, family-centered interventions. By deploying knowledgeable professionals who understand the unique cultural contexts and challenges faced by AI/AN families, THV programs can effectively identify and address early developmental risks. Home visiting initiatives focus on empowering caregivers with the skills and resources needed to foster positive early childhood experiences. The home-based nature of these interventions ensures that they are tailored to the specific needs of each family, promoting a holistic approach that integrates cultural values and traditions. Through fostering strong caregiver-child relationships and offering support in the crucial early years, THV is not only poised to address developmental risks, but also represents a restoration of traditional community member roles and responsibilities related to caring for the next generation. By passing along skills, culture, and traditions to children that experience them with their caregivers, the intervention supports learning and healing so that children acquire and share these skills and behaviors with the children that they may care for when they become adults. In this way, enhancing THV programs guided by an implementation science framework, by equipping home visitors with early intervention training and resources can contribute to the preservation and revitalization of Indigenous cultures, reinforcing the resilience and well-being of AI/AN communities while also using proven research methods that lead to long-term sustainability of enhanced evidence-based therapies.

2 Methods

Due to the overburdening of IDEA Part B and C services on the Navajo Nation, a focus has been on identifying novel solutions to remediate DD early in childhood rather than to allow DD to progress and complicate growth and development through childhood. This section presents the initial enhancement process for two commonly used language facilitation approaches typically delivered by speech-language pathologists (SLP) including those working in IDEA Part C early intervention programs. This work on the Navajo Nation utilizes the Consolidated Framework for Implementation Research (CFIR) to operationalize the process of study concept development through the piloting phase of a culturally enhanced intervention described below (25). See Figure 2 for CFIR framework. The use of implementation strategies helps researchers to identify potentially successful determinants of new intervention development and to strategically plan for precision delivery to diverse communities such as the Navajo Nation. Therefore, the use of the CFIR stage of The THING, which included the selected evidence-based therapies common in early intervention (Innovation) was used to guide the adaptation process, hereinafter referred to as the enhancement process, of two evidence-based therapeutic models embedded within a caregiver-facilitated intervention being piloted on the Navajo Nation. In this initial implementation stage, evidence-based therapies are identified as suitable for the population of interest (toddlers with elevated DD risk) and the process of enhancement is established with community and regional early intervention expertise. This process of enhancement, which employs a strategic process, is the focus of this paper along with consideration of decolonization which can help Indigenous researchers to deconstruct historical and contemporary elements of colonialism (26).

2.1 Enhancement process

The research team utilized steps identified in a scoping study, see Table 1 (27) below, of adaptation frameworks for adapting public health evidence-based interventions (EBI) to organize how the +Language is Medicine (+LiM) intervention was developed and how language facilitation strategies, the Hanen "It Takes Two to Talk" and Enhanced Milieu Teaching (EMT), were enhanced to be delivered to toddlers in the Northern Agency of the Navajo Nation. The stages discussed in this paper are as follows: (1) assess the community, (2) understand the intervention(s), (3) select the intervention(s), (4) consult with experts, (5) consult with stakeholders, (6) decide what needs adaptation, (7) enhance the original program, (8) train staff, and (9) test the enhanced materials. The +LiM intervention is currently engaged in a pilot study and therefore, the remaining steps have been omitted for detailed evaluation after pilot study completion.

2.1.1 Assess the community needs

The Johns Hopkins Center for Indigenous Health (JHCIH) has continuously operated service-research sites on the Navajo Nation since the mid-1980s. Since 2013, JHCIH has operated a range of behavioral and mental health programs focusing on maternal and child health, promoting physical activity, and suicide prevention that have greatly improved the health and well-being of Diné families. JHCIH has delivered Family Spirit curriculum and developed and tested Family Spirit supplemental modules that target various health outcomes with children and their caregivers over the last 10 years in the Northern Agency of the Navajo Nation (24, 28, 29). The Northern Agency is located in the northern part of the Navajo Nation with 20 chapter houses or local governments to serve each community. Through JHCIH's established Family Spirit presence within this region and experience utilizing Diné community members as home visitors, the research team chose Shiprock, NM as the center location for the development and field testing of the +Language is Medicine intervention. Further, community partnerships in the Northern Agency, established through previous JHCIH Family Spirit implementation efforts, have indicated an increase referral rate of children to early intervention services. Additionally, the lead PI, a licensed Diné SLP has worked with the Indian Health Service and worked with the local Part C early intervention program. It was based on these firsthand experiences that the study team shifted their attention to secondary prevention efforts to respond to the high incidence of DD in this region of the Navajo Nation.

2.1.2 Understanding and selecting the EBIs

The Hanen "It Takes Two to Talk" approach (30), is an early language facilitation program designed to support caregivers in promoting language development of their young children (birth to 5 years old), particularly those with language delays or difficulties. Developed by The Hanen Centre, this evidence-based intervention focuses on empowering caregivers as the primary facilitators of their child's communication skills. "It Takes Two to Talk" employs interactive strategies to enhance language-rich interactions within everyday routines, emphasizing the importance of responsive and child-centered communication. The program provides a step-by-step

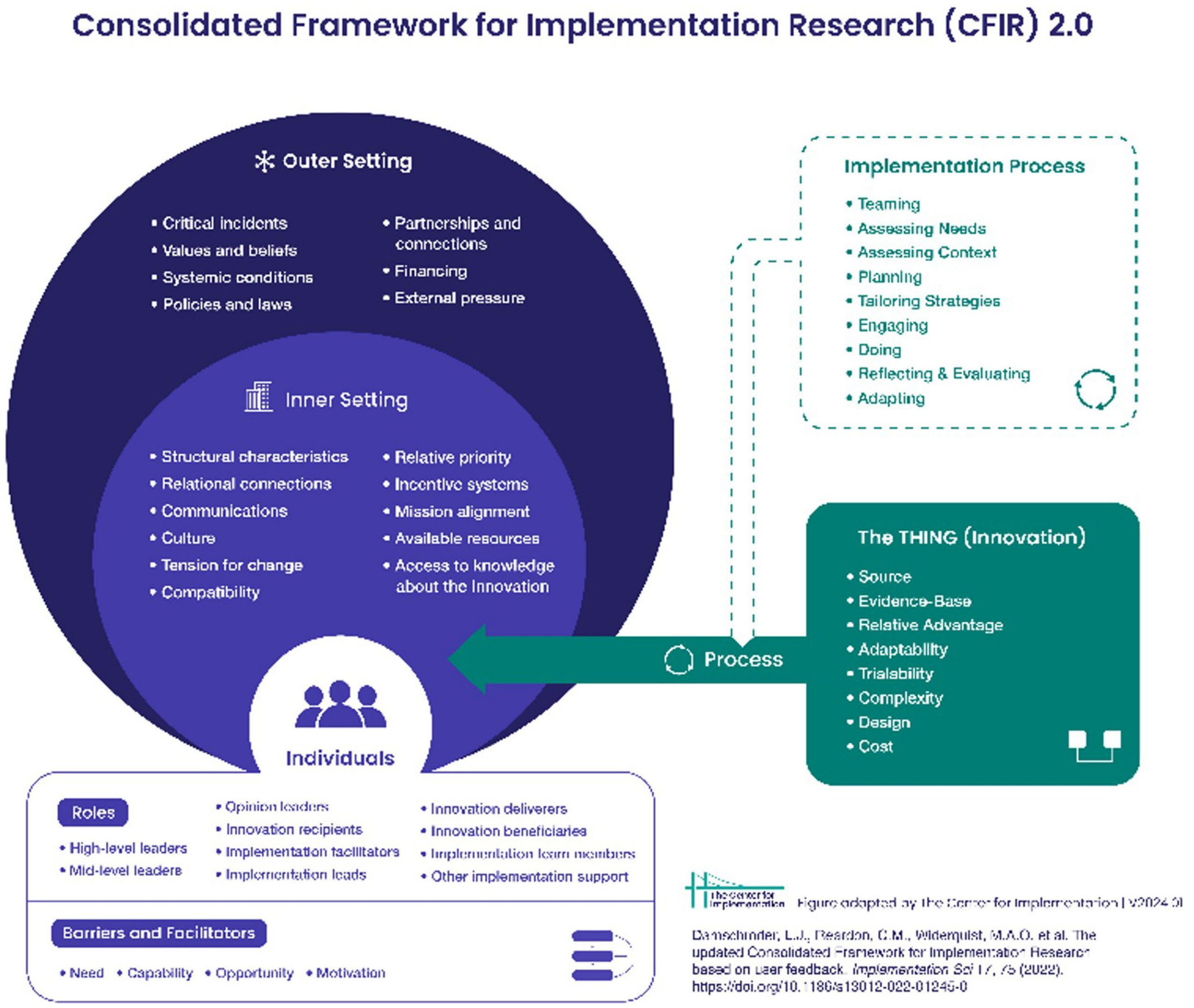

Figure adapted by the Center for Implementation | V2024.01

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FIGURE 2
CFIR promotes consistent use of constructs, systematic analysis, and organization of findings from implementation studies.

guide to educated caregivers as the primary facilitators through personalized coaching. Through this program, caregivers learn to recognize and respond effectively to their child's cues to create an enriched language-learning environment. By incorporating practical techniques into daily interactions, the program not only promotes language development but also strengthens the careiver-child bond. This intervention is recognized for its family-centered approach which acknowledges the crucial role caregivers play in supporting their children's communication skills during the foundational years of early childhood development.

Enhanced Milieu Teaching [EMT; (31)], is an evidence-based intervention that has proven effective in supporting toddlers with developmental delays in their language acquisition. EMT emphasizes creating an early language-rich environment within the child's natural context. This approach involves shared book reading and structuring play interactions based on the child's interest to encourage communication and language using naturalistic teaching strategies. With a focus on increasing the child's initiation of communication, EMT employs prompts, expansions, and modeling techniques to enhance language development. This method not only fosters the child's expressive and receptive language skills but also promotes generalization of these skills across various settings. EMT recognizes the importance of individualized, child-centered interventions and encourages active participation from caregivers and caregivers. EMT is a valuable tool for early intervention programs focused on supporting toddlers with developmental delays in achieving meaningful language milestones.

Incorporating shared book reading and pretend play instruction into home visiting programs is essential for fostering holistic child development and strengthening the parent/caregiver-child relationship within common caregiver-child routines. Shared book reading not only exposes children to language-rich environments but also promotes early literacy skills, vocabulary expansion, and cognitive development (32, 33). Playtime is an important opportunity for learning and brain development. Pretend play is when a child uses their imagination to do common everyday activities in their play by using an object, idea, or action to represent another object, idea, or action. (34, 35) Additionally, engaging in pretend play enhances a child's imaginative and social–emotional skills, allowing them to explore creativity, problem-solving, and interpersonal skills. Integrating these activities into home visiting programs not only supports children's cognitive and language development but also empowers caregivers with valuable tools to

TABLE 1 Key adaptation steps and descriptions.

Step name	Step descriptions
1. Assess community	• Identify behavioral determinants and risk behaviors of the new target population using focus group, interviews, needs assessment, and logic models. • Assess organizational capacity to implement the program
2. Understand the intervention	• Identify and review relevant EBPs and their program materials • Understand the theory behind the programs and their core elements
3. Select intervention	• Select the program that best matches the new population and context
4. Consult with experts	• Consult content experts, including original program developers, as needed • Incorporate expert advice into program
5. Consult with stakeholders	• Seek input from advisory boards and community planning groups where program implementation takes place • Identify stakeholder partners who can champion program adoption in new setting and ensure program fidelity
6. Decide what needs adaptation	• Decide whether to adapt implement original program • Theater test selected EBP using new target population and other stakeholders to generate adaptations • Determine how original and new target population/setting differ in terms of risk and protective factors • Identify areas where EBP needs to be adapted and include possible changes in program structure, content, provider, or delivery methods • Retain fidelity to core elements • Systematically reduce mismatches between the program and the new context
7. Adapt the original program	• Develop adaptation plan • Adapt the original program contents through collaborative efforts • Make cultural adaptations continuously through pilot testing • Core components responsible for change should not be modified
8. Train staff	• Select and train staff to ensure quality implementation
9. Test the adapted materials	• Pretest adapted materials with stakeholder groups • Conduct readability tests • Pilot test adapted EBP in new target population • Modify EBP further if necessary
10. Implement	• Develop implementation plan based on results generated in previous steps • Identify implementers, behaviors, and outcomes • Develop scope, sequence, and instructions • Execute adapted EBP
11. Evaluate	• Document the adaptation process and evaluate the process and outcomes of the adapted intervention as implemented • Write evaluation questions; choose indicators, measures, and the evaluation design; plan data collection, analysis, and reporting • Employ empowerment evaluation approach framework to improve program implementation

actively participate in their child's learning journey. Both shared book reading, and pretend play create opportunities for meaningful interactions, reinforcing positive relationships between caregivers and children. By emphasizing these activities as important routines in which caregiver coaching can be caregivers, home visiting programs can contribute significantly to the overall well-being and school readiness of young children. Equipping caregivers with practical strategies through coaching and modeling promotes families to continue to support their child's growth outside of structured intervention sessions, in a naturalistic environment.

2.1.3 Select the interventions

The previously mentioned EBIs, "It Takes Two to Talk" (30), and EMT (31), were chosen by the lead PI, a licensed Diné SLP. These two language facilitation approaches are commonly used in developmental early intervention (EI) programs that include home visits and parent coaching. The parent coaching model has been mostly adopted into Birth-to-Three EI programs because of its ability to have a positive impact on parent behavior rather than direct intervention with a child (36). Therefore, these two interventions were selected to be culturally enhanced for Diné families and adapted to be delivered by supervised tribal home visitors.

2.1.4 Consult with experts

Community advisory boards (CAB) play a crucial role in research with tribal communities by providing a culturally grounded and community-specific perspective (37–42). These boards help ensure that research respects tribal sovereignty, addresses community priorities, and incorporates local knowledge, fostering trust, collaboration, and the ethical conduct of research within the cultural context. A local CAB was convened and consisted of caregivers, community members, early childhood development professionals, school staff, and health professionals with various levels of Diné cultural knowledge to provide input to the +Language is Medicine curriculum modules. A Diné linguist/elder was a key CAB member who also provided translation work of +LiM intervention materials. 95% of the 23 CAB members identified as Diné. CAB members were partly identified through established community partnerships from previous and current JHCIH programs. Additional stakeholders, who were not early childhood professionals, teachers, or healthcare staff, were consulted to provide their parent and community insight. Early childhood professionals included individuals from early intervention programs (IDEA Part C) and other local THV programs servicing Diné families. CAB members were compensated for their time and participation in CAB meetings.

2.1.5 Consult with stakeholders

The research team virtually met with CAB members throughout the development of the +LiM curriculum. During CAB meetings, the Principal Investigator reviewed +LiM intervention content, including lessons and visuals, and CAB members provided live feedback. There were three CAB meetings over 4 months, giving time for the research team to develop the curriculum and incorporate feedback. The lead PI and tribal home visiting curriculum experts on the research team utilized the two EBIs previously described (Hanen "It Takes Two to Talk" and EMT) for concept development of five lessons promoting language nutrition. In the first CAB meeting, members provided feedback on the following proposed lesson topics: (1) Back and Forth with your Child (Reciprocal and Responsive Communication), (2) Reading Together (Shared Book Reading), (3) Following Your Child's Lead, (4) Encouraging Communication through Pretend Play, and (5) Incorporating Native Language into Daily Life. Open-ended questions to the CAB were utilized to facilitate discussion about the lesson concepts. Additionally, questions were asked to the CAB on engagement with young Diné families and how to best support Diné language use with infants and toddlers. In the second meeting, the team discussed a draft of the first lesson and asked questions to the CAB about the content, relatability, and flow of the lesson. Before the last meeting, lesson drafts were mailed to CAB members for review. In the last meeting, the +LiM team presented the remaining lessons for CAB feedback.

2.1.6 Decide what needs enhancement

Native language use and cultural relevance throughout the +LiM curriculum were key areas for enhancing the two previously mentioned EBIs. This included initial +LiM team discussions on how to promote language nutrition within home and cultural routines. CAB members provided additional recommendations to the curriculum after reviewing the draft lessons.

2.1.7 Enhance the original program

An enhancement plan was developed to incorporate CAB feedback into the full lesson drafts and training plan through the following steps: (1) compile, (2) apply, (3) design, (4) print, (5) train. CAB meeting feedback was compiled to highlight key edits and additions to lesson content and guided activities. The research team worked with the Family Spirit curriculum writer to apply the appropriate sections of each lesson with the cultural recommendations and additions. The evidence-based Family Spirit home visiting model was chosen due to its effectiveness in previous programs (11, 13, 24), and familiarity among the research team's community-based home visitors. In the design step, graphic design elements were finalized to improve the consistency in look and feel of the lessons. Complete draft lessons were reviewed and discussed in team meetings for a final round of curriculum revisions before being printed for training use. The community-based +LiM home visitors provided additional recommendations for curriculum revisions during the training period. After the training period, final curriculum versions were printed for pilot study use.

2.1.8 Train staff

The research team had prior experience in home visiting research practice and implementation. The community-based tribal home visitors were already trained and familiar with the delivery of the Family Spirit curriculum model. A precision THV approach was utilized to train field staff in the +LiM curriculum content. The PI trained the home visitors in administration of standardized child developmental assessments and curriculum intervention delivery. A combination of virtual and in-person meetings were utilized to train the tribal home visitors in data collection procedures and lesson delivery.

Pre-training in child developmental assessments was essential for understanding and recognizing developmental delays. Pre-training included review of developmental assessment materials which included the Ages & Stages Questionnaire – 3rd Edition [ASQ-3; (43)], and the Bayley Scales of Infant and Toddler Development – 4th edition (44), and a review of the procedure manuals. After an initial review of assessment materials, the PI utilized didactic teaching on administering and scoring these assessments. The home visitors shadowed virtual early intervention visits with the PI, followed by discussion and identification of language facilitation strategies from "It Takes Two to Talk" and Hanen. Following this observation of PI demonstration, a reliability training plan was developed for home visitors to practice administering assessments with Diné families. The PI and home visitors completed home visits together and completed assessment questionnaires individually, without coaching from the PI. Assessments were then scored and measured by the PI for reliability between the home visitors and the PI/SLP. The PI provided coaching and feedback after these practice home visits. The PI observed (live or video) home visitors in practice visits until they achieved 85% inter-rater reliability.

Final versions of the +LiM curriculum lessons were in development during the assessment training period prior to study launch. A series of training meetings were conducted with the home visitors on teaching the +LiM module. These meetings consisted of didactic teaching on key strategies of the +LiM intervention such as active listening, tailoring, Teach-Model-Coach, comprehension checks, and early intervention referrals. Home visitors also completed virtual role play sessions with the PI and team curriculum specialists which included live and retrospective coaching.

2.1.9 Test the enhanced materials

The enhanced +LiM curriculum is being implemented through a pilot study. Lesson fidelity checks are being completed by the PI for home visitor content competency. Sharing of pilot study results will be determined after pilot study completion and evaluation.

2.2 Timeline

This process of enhancement took place from December 2021 through December 2022. This included convening and meeting with the CAB, drafting lessons and incorporating CAB feedback, and training tribal home visitors in administration of child developmental assessments and lesson delivery.

3 Results

As a result of this collaboration with key community members, our research team produced two main products that drive the +Language is Medicine pilot study: (1) a culturally responsive interpretation of language nutrition that centered Diné knowledge and beliefs and (2) a five-lesson language nutrition curriculum based on EMT principles, shared book reading, increasing pretend play, and incorporating native language into family routines.

3.1 Indigenous language nutrition

Language nutrition, which describes early language exposure rich in quality and quantity and delivered in home routines, naturally became a center point of topic during meetings with CAB members. Language use, in the context of caregiver-child interactions, language use in ceremony, and exposure to Native languages were common references during conversations with CAB members and the study team. Language use that centers cultural routines and language revitalization directly correlates with the nourishment and sustenance of a community's linguistic heritage, cultural practices, and traditional ways of knowing (e.g., childrearing practices). In this context, language use was presented as a vital element of cultural identity, connecting individuals to their culture. The following themes were extracted from community-based conversations, in which the study team developed an approach for the enhancement of the two language facilitation strategies selected for the basis of the +LiM intervention (30, 31):

- Identifying naturalistic language-rich routines, such as using kinship terms, greetings, storytelling, and participating in ceremonies (e.g., Diné first laugh celebration)
- Aligning parent coaching topics with seasonal celebrations (e.g., planting and harvesting, winter games, birthday parties, and wood hauling)
- Alignment with Diné language revitalization efforts in early childhood programs based on local needs and family preferences
- Building on intergenerational learning and facilitating the transfer of language skills from elders to younger generations to foster linguistic knowledge and cultural understanding
- Embedding opportunities for discussion of traditional practices such as hunting, gathering, or craftsmanship which provides a context for language use that aligns with cultural values and caregiver practices
- Honoring Diné storytelling and oral traditions to preserve cultural narratives.

The process of establishing strong Indigenous language nutrition refers to nurturing the linguistic and cultural ecosystem in a holistic manner while acknowledging the interconnectedness of language, identity, and community well-being. Most critical, it is about sustaining and revitalizing a community's Indigenous language in a way that resonates with the cultural practices and values, fosters a sense of self-determination and intergenerational healing, and empowers caregivers to speak to their children in nurturing and respectful ways. This interpretation of language nutrition was developed internally by the study team because of engaging with our CAB members, spending time in the community, and applying methods of decolonization to the EBI enhancement process and establishing a basis for true community-leveraged enhancement strategies. It is with this redefinition that the study team moved into product development, which included a culturally tailored language nutrition curriculum that incorporated EMT and the Hanen Approach through parent coaching lessons.

3.2 The +language is medicine curriculum

A key component of the +LiM curriculum is an enhanced teach, model, coach approach to emphasize cultural routines and native language exposure in supporting multiple dimensions of child development. Key elements of +LiM's teach, model, coach are (1) use the Family Spirit home visiting model as a platform (2) engage caregivers as a child's first teacher (3) teach basic language nutrition concepts and language facilitation skills (4) demonstrate skills (live or video) (5) coach caregivers in practicing skills (6) exploring families' home, cultural routines and environment to identify opportunities to add concepts of Indigenous language nutrition, as previously described.

The +LiM curriculum consists of 5 lessons categorized into three content areas: introduction to +LiM and foundational knowledge (Lesson 1: Encouraging Conversations with Young Children), building strategies to support language acquisition (Lesson 2: Following Your Child's Lead), and identifying opportunities to apply skills and strategies in daily life (Lesson 3: Reading Together; Lesson 4: Pretend Play and Communication; Lesson 5: Native Language and Your Daily Routine).

A routines mapping activity was embedded in each lesson for families to identify home and cultural routines through which language facilitation strategies could be practiced and expanded upon. The activity utilizes a routines circle map consisting of four quadrants to represent household, outdoors, cultural, and community spaces. The goal of the routines mapping activity is for caregivers to identify in their daily routines when and where they can reinforce Indigenous language nutrition and practice language facilitation strategies with their child. This activity takes place at the beginning of the lesson and is referenced throughout the home visit. The routines map is revisited again at the conclusion of the lesson The CAB provided additional examples of daily routines such as narrating to a child the process of making dough for frybread (household routine), using rocks, sticks, or mud to pretend play (outdoors), singing a song or reciting a prayer in Diné Bizaad (Navajo language) with child (other cultural spaces), and greeting relatives in Diné Bizaad (community). These familiar routines are optimal places and moments for promoting language learning and practicing language facilitation strategies such as narration, imitation, labeled praise, and modeling through self-talk, parallel talk, and focused repeating.

4 Implementation supports

We also developed an implementation guide for use during the training of home visitors and serves as a reference point for how to further enhance the +LiM lessons. To help support the standardization of +LiM lesson delivery, a treatment fidelity checklist was developed by the PIdial to use by peers who will observe home visits.

5 Discussion

Tribal Home Visiting (THV), such as the Family Spirit home visiting curriculum, has shown strong promise in improving child and parental outcomes in tribal communities. Dedicated federal funding allows for the sustainability of these programs as long-term solutions to improving child developmental outcomes. Most importantly, the dedicated programming for AI/AN families allow for cultural enhancement and further tailoring of caregiver coaching interventions. The +LiM team aims to deliver their +LiM curriculum within this existing infrastructure and to further develop developmental surveillance and response efforts. Due to the shortage of special education personnel in reservation communities, a precision home visiting approach can be utilized to help meet the need. In this case, the +LiM team seeks to have a wider reach of skilled SLP services delivered at the tertiary prevention level (e.g., Birth-to-Three programs) and aim to deliver primarily through secondary prevention efforts (e.g., THV). With this model, the SLP can have a wider reach to families in underserved areas of tribal nations. In addition to this precision public health model, incorporating EBIs into home and cultural routines can take place. With this wider reach and ability to tailor interventions, the team can utilize community engagement to incorporate community values, practices, and traditions. This enhancement process demonstrates how EBIs used with young children with DD can be modified and adapted to incorporate cultural values and support native language use. Most importantly, this paper highlights the critical steps necessary to ensure that tribal community input is the driving factor of EBI cultural enhancement. Assessment of preliminary outcomes of enhancing EMT and Hanen for use with Diné toddlers with DD is the next major step for the +LiM research team and will be shared through future publications and community-based presentations.

Data availability statement

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding authors.

Author contributions

TB: Writing – original draft, Writing – review & editing. EK: Writing – original draft, Writing – review & editing. JM: Writing – original draft, Writing – review & editing. LM: Writing – original draft, Writing – review & editing. LJ: Writing – original draft, Writing – review & editing. MA-G: Conceptualization, Methodology, Writing – review & editing. AB: Writing – original draft, Writing – review & editing. JA-B: Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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References

1. Lange BCL, Callinan LS, Smith MV. Adverse childhood experiences and their relation to parenting stress and parenting practices. *Community Ment Health J*. (2019) 55:651–62. doi: 10.1007/s10597-018-0331-z

2. Head Zauche L, Darcy Mahoney AE, Thul TA, Zauche MS, Weldon AB, Stapel-Wax JL. The power of language nutrition for Children's brain development, health, and future academic achievement. *J Pediatr Health Care*. (2017) 31:493–503. doi: 10.1016/j.pedhc.2017.01.007

3. Shevell M, Ashwal S, Donley D, Flint J, Gingold M, Hirtz D, et al. Practice parameter: evaluation of the child with global developmental delay. *Neurology*. (2003) 60:367–80. doi: 10.1212/01.wnl.0000031431.81555.16

4. Collier C. *Seven steps to separating difference from disability*. California: Corwin Press (2011). 160 p.

5. Whitesell NR, Howley CT, Asdigian NL, Clifford C, Senehi N, Sarche M, et al. Community perspectives on developmental screening of American Indian and Alaska native children. *Infant Ment Health J*. (2022) 43:558–75. doi: 10.1002/imhj.21992

6. Snowling MJ, Adams JW, Bishop DV, Stothard SE. Educational attainments of school leavers with a preschool history of speech-language impairments. *Int J Lang Commun Disord*. (2001) 36:173–83. doi: 10.1080/13682820010019892

7. Merrell KW, Holland ML. Social-emotional behavior of preschool-age children with and without developmental delays. *Res Dev Disabil*. (1997) 18:393–405. doi: 10.1016/s0891-4222(97)00018-8

8. Choo YY, Agarwal P, How CH, Yeleswarapu SP. Developmental delay: identification and management at primary care level. *Singapore Med J*. (2019) 60:119–23. doi: 10.11622/smedj.2019025

9. Roberts MY, Curtis PR, Sone BJ, Hampton LH. Association of Parent Training with Child Language Development: a systematic review and Meta-analysis. *JAMA Pediatr*. (2019) 173:671–80. doi: 10.1001/jamapediatrics.2019.1197

10. Walkup JT, Barlow A, Mullany BC, Pan W, Goklish N, Hasting R, et al. Randomized controlled trial of a paraprofessional-delivered in-home intervention for young reservation-based American Indian mothers. *J Am Acad Child Adolesc Psychiatry*. (2009) 48:591–601. doi: 10.1097/CHI.0b013e3181a0ab86

11. Barlow A, Mullany B, Neault N, Compton S, Carter A, Hastings R, et al. Effect of a paraprofessional home-visiting intervention on American Indian teen mothers' and infants' behavioral risks: a randomized controlled trial. *Am J Psychiatry*. (2013) 170:83–93. doi: 10.1176/appi.ajp.2012.12010121

12. Walker SP, Wachs TD, Grantham-McGregor S, Black MM, Nelson CA, Huffman SL, et al. Inequality in early childhood: risk and protective factors for early child development. *Lancet*. (2011) 378:1325–38. doi: 10.1016/S0140-6736(11)60555-2

13. Barlow A, Mullany B, Neault N, Goklish N, Billy T, Hastings R, et al. Paraprofessional-delivered home-visiting intervention for American Indian teen mothers and children: 3-year outcomes from a randomized controlled trial. *Am J Psychiatry*. (2015) 172:154–62. doi: 10.1176/appi.ajp.2014.14030332

14. Bandini LG, Curtin C, Hamad C, Tybor DJ, Must A. Prevalence of overweight in children with developmental disorders in the continuous national health and nutrition examination survey (NHANES) 1999-2002. *J Pediatr*. (2005) 146:738–43. doi: 10.1016/j.jpeds.2005.01.049

15. Heslop P, Lauer E, Hoghton M. Mortality in people with intellectual disabilities. *J Appl Res Intellect Disabil*. (2015) 28:367–72. doi: 10.1111/jar.12196

16. Lauer E, McCallion P. Mortality of people with intellectual and developmental disabilities from select US state disability service systems and medical claims data. *J Appl Res Intellect Disabil*. (2015) 28:394–405. doi: 10.1111/jar.12191

17. Allison-Burbank JD, Conn A, Vandever D. Interpreting Diné epistemologies and decolonization to improve language and literacy instruction for Diné children. *Lang Speech Hear Serv Sch*. (2023) 54:707–15. doi: 10.1044/2023_LSHSS-22-00147

18. Mitchell F. *Navajo Blessingway singer: The autobiography of frank Mitchell, 1881–1967*. Albuquerque, NM: UNM Press (1978).

19. Jackson S, James IK, Attakai M, Attakai MN, Begay EF. *Amá Sani dóó Achei Baahané/the office of Diné culture, language, and community services*. Cortez, Colorado: Southwest Printing Co. (2004).

20. Kahn-John M. Concept analysis of dine Hozho: a dine wellness philosophy. *ANS Adv Nurs Sci*. (2010) 33:2. doi: 10.1097/ANS.0b013e3181dbc658

21. Kahn-John Diné M, Koithan M. Living in health, harmony, and beauty: the Diné (Navajo) Hózhó wellness philosophy. *Glob Adv Health Med*. (2015) 4:24–30. doi: 10.7453/gahmj.2015.044

22. Allison-Burbank JD, Reid T. Prioritizing connectedness and equity in speech-language Services for American Indian and Alaska Native Children. *Lang Speech Hear Serv Sch*. (2023) 54:368–74. doi: 10.1044/2022_LSHSS-22-00101

23. Sweet MA, Appelbaum MI. Is home visiting an effective strategy? A Meta-analytic review of home visiting programs for families with young children. *Child Dev Perspect*. (2004) 75:1435–56. doi: 10.1111/j.1467-8624.2004.00750.x

24. Rosenstock S, Ingalls A, Foy Cuddy R, Neault N, Littlepage S, Cohoe L, et al. Effect of a home-visiting intervention to reduce early childhood obesity among native American children: a randomized clinical trial. *JAMA Pediatr*. (2021) 175:133–42. doi: 10.1001/jamapediatrics.2020.3557

25. Damschroder LJ, Reardon CM, Widerquist MAO, Lowery J. The updated consolidated framework for implementation research based on user feedback. *Implementation Sci*. (2022) 17:75. doi: 10.1186/s13012-022-01245-0

26. Community-Based Global Learning Collaborative. What is decolonization, why is it important, and how can we practice it? (2021). Available at: https://www.cbglcollab.org/what-is-decolonization-why-is-it-important. Accessed March 03, 2024.

27. Escoffery C, Lebow-Skelley E, Udelson H, Böing EA, Wood R, Fernandez ME, et al. A scoping study of frameworks for adapting public health evidence-based interventions. *Transl Behav Med*. (2018) 9:1. doi: 10.1093/tbm/ibx067

28. Kenney A, Chambers RA, Rosenstock S, Neault N, Richards J, Reid R, et al. The impact of a home-based diabetes prevention and management program on high-risk American Indian youth. *Diabetes Educ*. (2016) 42:585–95. doi: 10.1177/0145721716658357

29. Barlow A, Varipatis-Baker E, Speakman K, Ginsburg G, Friberg I, Goklish N, et al. Home-visiting intervention to improve child care among American Indian adolescent mothers: a randomized trial. *Arch Pediatr Adolesc Med*. (2006) 160:1101–7. doi: 10.1001/archpedi.160.11.1101

30. The Hanen Centre. It Takes Two to Talk – Hanen Program for Parents of Children with Language delays. (2016). Available at: https://www.hanen.org/programs/for-parents/it-takes-two-to-talk.aspx Accessed January 25, 2024.

31. McCauley RJ, Fey ME, Gillan RB. *Treatment of language disorders in children*. Maryland: Paul H. Brookes Publishing Co, Inc. (2017). 87 p.

32. Sénéchal M, Pagan S, Lever R, Ouellette GP. Relations among the frequency of shared reading and 4-year-old children's vocabulary, morphological and syntax comprehension, and narrative skills. *Early Educ Dev*. (2008) 19:27–44. doi: 10.1080/10409280701838710

33. Dowdall N, Melendez-Torres GJ, Murray L, Gardner F, Hartford L, Cooper PJ. Shared picture book reading interventions for child language development: a systematic review and meta-analysis. *Child Dev*. (2020) 91:e383–99. doi: 10.1111/cdev.13225

34. Weitzman E, Greenberg J. *Learning language and loving it: A guide to promoting children's social, language, and literacy development in early childhood settings*. Toronto: The Hanen Centre (2002).

35. Hammer TR. *Pretend play*. Encyclopedia of Child Behavior and Development, Boston: Springer (2011).

36. Rush DD, Shelden ML. Evidence-based definition of coaching practices. (2005) Available at: https://fipp.ncdhhs.gov/wp-content/uploads/caseinpoint_vol1_no6.pdf Accessed January 25, 2024.

37. Yuan NP, Mayer BM, Joshweseoma L, Clichee D, Teufel-Shone NI. Development of guidelines to improve the effectiveness of community advisory boards in Health Research. *Prog Community Health Partnersh*. (2020) 14:259–69. doi: 10.1353/cpr.2020.0026

38. Balazs CL, Morello-Frosch R. The three R's: How community based participatory research strengthens the rigor, relevance and reach of science. *Environ Justice*. (2013) 6:1. doi: 10.1089/env.2012.0017

39. Cochran P, Marshall C, Garcia-Downing C, Kendall E, Cook D, McCubbin L, et al. Indigenous ways of knowing: implications for participatory research and community. *Am J Public Health*. (2008) 98:22–7. doi: 10.2105/AJPH.2006.093641

40. Christopher S. Recommendations for conducting successful research with native Americans. *J Cancer Educ*. (2005) 20:47–51. doi: 10.1207/s15430154jce2001s_11

41. Manson SM, Garroutte E, Goins RT, Henderson PN. Access, relevance, and control in the research process: lessons from Indian country. *JAH*. (2004) 16:58S–77S. doi: 10.1177/0898264304268149

42. Christopher S, Saha R, Lachapelle P, Jennings D, Colclough Y, Cooper C, et al. Applying indigenous community-based participatory research principles to partnership development in health disparities research. *Fam Community Health*. (2011) 34:246–55. doi: 10.1097/FCH.0b013e318219606f

43. Squires J, Bricer D. *Ages & stages questionnaires, third edition (ASQ-3): A parent-completed child monitoring system*. Maryland: Paul H. Brookes Publishing Co, Inc. (2009).

44. Bayley N, Aylward GP. *Bayley-4: Scales of infant and toddler development. 4th* ed. Texas: Pearson (2009).

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A psychometric analysis of the adapted historical loss scale and historical loss associated symptoms scale among native Hawaiian adults

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Objectives: The Historical Loss Scale (HLS) and Historical Loss Associated Symptoms Scale (HLASS) are standardized measures that have been accepted and previously validated among North American Indigenous communities and allow researchers to measure the impact of Historical Loss. Evidence of the psychometric properties of this instrument have not been assessed for Native Hawaiians, the Indigenous peoples of Hawai'i. The purpose of this study is to investigate the psychometric properties of the adapted HLS (aHLS) and HLASS for adults from multiple Hawaiian Homestead Communities throughout Hawai'i.

Methods: Data are based on cross-sectional surveys administered between 2014 and 2020. The final sample included 491 Native Hawaiian adults who were predominantly female (67.3%) and between the ages of 18–90 years, who were part of the larger study entitled the Hawaiian Homestead Health Survey. Factor analyses were conducted to determine the final model structures of each scale. Reliability and correlation matrices of items are also reported.

Results: The final factor structure of the aHLS model suggested 3 factors: (1) General loss of culture or cultural loss, (2) Intergenerational loss, and (3) Distrust and destruction of traditional foods. The final HLASS model also suggested 3 factors: (1) Depression and Anger, (2) Shame and Anxiety, and (3) Re-experiencing, fear, and avoidance.

Conclusion: These findings have implications for future research, practice, and education that explores the role of Historical Loss and associated symptoms in Native Hawaiians and Indigenous communities at large. In particular, measuring historical loss and associated symptoms in Hawaiian Homestead communities paves the way for quantitative assessments of historical trauma and healing in these communities.

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1 Introduction

Native Hawaiians, the Indigenous peoples of Hawaiʻi, were once described by foreign and western visitors and settlers as being robust and capable of great physical activity (1). Native Hawaiian lifestyles and worldviews center relationships where health is viewed as a sacred force maintained through balance and holistic approaches to health. The physical and environmental manifestation of balance was often reflected through complex, agricultural systems such as ahupuaʻa and ʻaha moku systems. Unbeknownst to foreigners, Native Hawaiians possessed an interdependent relationship with ʻĀina (land, nature, environment, that which feeds) that manifested in cultural practices and beliefs, which helped them maintain their physical, mental, spiritual, and emotional health (2–5). The large influx of westerners and foreigners in the Hawaiian Kingdom in the 1800s influenced many policies that interrupted the Native Hawaiian way of life, which shifted the sociopolitical power, contributing to land displacement and privatization of land, ultimately impacting Native Hawaiian health and wellbeing (6–10). After the illegal overthrow of the Hawaiian Kingdom in 1893, Hawaiʻi quickly became a major U.S. military site and tourist attraction. Since western contact, Native Hawaiians continue to face inequitable opportunities, including lower socioeconomic status and health inequities that negatively impact mental, physical, spiritual, and emotional health (11–13).

Today, the biomedical health status of Native Hawaiians remains one of the poorest in the state when compared to other major ethnic groups. Native Hawaiians make up 22.7% of Hawaiʻi's population (14) and are projected to grow to 47% by 2025 (15), yet they experience one of the lowest life expectancies in Hawaiʻi (16, 17). Chronic conditions, such as cardiovascular diseases, obesity, and diabetes are more prevalent in Native Hawaiians compared to other major ethnic groups (16, 18). Additionally, Native Hawaiians experience morbidity prevalence and mortality of the top five leading causes of death at greater rates than other major ethnic groups (16, 18).

Indigenous researchers theorize the health status of Indigenous populations as a direct result of historical trauma (19–22). Historical trauma refers to the enduring impact of systemic oppression, violence, dispossession, or other forms of harm on a community and collective group of people (19). Both current and past experiences contribute to collective trauma responses. The effects of these events and experiences can be transmitted intergenerationally, with long-lasting impacts that influence the biological, social, emotional, and mental health and wellbeing of the affected community (19). The Historical Trauma Conceptual Model conceptualizes the way mass trauma experiences, like loss of land and physical/psychological violence, impacts primary generations and secondary/subsequent generations through unresolved trauma responses (23).

In alignment with this model, the historical loss and cultural trauma experienced by Native Hawaiians directly impact generations subjected to mass death due to infectious disease, assimilative policies including language bans, and the illegal overthrow of the Hawaiian Kingdom (2, 9, 10, 24). Therefore, a culmination of traumatic events in Hawaiʻi, like colonization, cultural degradation, depopulation, and land ownership policies, are arguably a determinant of health for contemporary Native Hawaiians (12, 25). These historical losses continue to impact Native Hawaiians directly and indirectly today. We must validly and reliably measure Historical Loss to better understand and address their impacts. Whitbeck and colleagues developed two scales, the Historical Loss Scale (HLS) and the Historical Loss Associated Symptoms Scale (HLASS) to begin to enumerate historical trauma among North American Indigenous communities, who also faced mass trauma from colonialism (22). Thus, this study explores the psychometric properties of an adapted HLS and the HLASS among Native Hawaiian communities.

1.1 Historical trauma as a determinant of health

Scholars theorize the way historical trauma serves as an intergenerational determinant of health (19, 20, 22, 23). For instance, historical trauma theory builds on existing theoretical frameworks including psychosocial theory, political economic theory, and social ecological systems theory. These collective frameworks theorize the way disease is linked to physical and psychological stress that stem from the larger environment, political, economic, and structural determinants of health, and are multilevel, thereby impacting individuals intergenerationally and across the lifespan (23). Thus, historical trauma, or the deliberate and systematic trauma inflicted on a collective group or community results in disparities and inequities that persists across generations.

As with measurement and survey instrumentation as a whole, standardized and empirical measures of historical trauma are important to appropriately measure the health impacts of historical trauma, particularly as a risk factor and possible mediator and moderator of health. Valid and reliable measures may also advance health practice, research, and policy by exploring these impacts on health and wellbeing. For instance, empirical evidence that demonstrates the magnitude and severity of experiences related to displacement, violence, economic and political destruction and cultural dispossession that specifically result from colonialism, ongoing cultural and historical trauma, assimilative policies, and systemic oppression among Indigenous Peoples will help to inform health practice, research, and policy that fosters self-determination, and thus, health and wellbeing at large.

While this may be the case, limitations exist among measurements of historical trauma, primarily due to the difficulty in capturing the complex experiences of historical trauma that are unique to specific communities, with the potential for these impacts to vary between and within groups of people. In 2019, Gone et al. published a systematic literature review that explored literature related to Indigenous Historical Trauma (26). As noted by the authors, findings demonstrated impressive efforts and attempts to measure the impact of Indigenous Historical Trauma, yet the body of literature does not coherently demonstrate the empirical impacts of Indigenous Historical Trauma. Despite these limitations, and although the systematic literature review focused on Indigenous populations in the United States and Canada, the authors suggest refining existing measures of Indigenous Historical Trauma as solutions to remedy the urgent and pressing need to better understand the role of Indigenous Historical Trauma in health disparities and inequities experienced by Indigenous Peoples.

1.2 Historical loss scale (HLS)

One of the most widely used or adapted measures of Indigenous Historical Trauma is the Historical Loss Scale (HLS). The HLS was originally developed by Whitbeck and colleagues and validated with 143 American Indian adults who were parents of children between the ages of 10–12 years (22). According to the Exploratory Factor Analysis (EFA), the HLS was proposed as a unidimensional construct consisting of 12 items, which demonstrated high internal reliability (22). The original 12 items consisted of: (1) The loss of our land, (2) The loss of our language, (3) Losing our traditional spiritual ways, (4) The loss of our family ties because of boarding schools, (5) The loss of families from the reservation to government relocation, (6) The loss of self respect from poor treatment by government officials, (7) The loss of trust in White individuals from broken treaties, (8) Losing our culture, (9) The losses from the effects of alcoholism on our people, (10) Loss of respect by our children and grandchildren for elders, (11) Loss of our people through early death, (12) Loss of respect by our children for traditional ways.

Since the original publication of the HLS, researchers have proposed a multi-dimensional Historical Loss construct. For instance, in a later publication, Whitbeck and colleagues found that the original one-factor model demonstrated poor model fit (27). As a result, researchers proposed a 10-item 2-factor model. Factor one included items on government and institutional policies and practices including loss of land, loss of family ties because of boarding schools, loss due to government relocation, broken treaties, and poor treatment by government officials. Factor two focused on personal and cultural losses, including loss of language and spiritual ways, and loss of people to early death and via the effects of alcoholism (27). In another study, Armenta and colleagues proposed a higher-order construct of Historical Loss based on an adapted 10-item scale administered to 636 North American Indigenous adolescents (20). Based on the findings, researchers proposed a three-factor model: (1) General loss of culture, (2) Loss of people, and (3) Cultural mistreatment (20). Although the model with three subfactors and a larger Historical Loss factor demonstrated poor model fit, proposed models for second-order and third-order models indicated good model fit (20).

1.3 Historical loss associated symptoms scale (HLASS)

The HLASS was also originally developed by Whitbeck and colleagues based on the same sample of 143 American Indian adult parents and children (22). The 17-item HLASS was developed to empirically link perceptions of historical loss to various psychological symptoms found in qualitative literature. An exploratory factor analysis yielded two factors, anxiety/depression and anger/avoidance (22). Anxiety/depression includes five items reporting the experience of depression, anxiety, loss of concentration, isolation, and loss of sleep. Anger/avoidance includes seven items, which measures anger, discomfort around white people, shame, rage, fear and distrust, feeling like traumatic events are happening again, and avoiding places. A confirmatory factor analysis supported these results and a structural equation model yielded construct loading that were also consistent with the 2-factor model (22).

Despite evidence of a multidimensional, 2-factor model, other studies have used the HLASS as a single-factor. Wiechelt and colleagues examined the relationship between historical trauma symptoms and substance use and family cohesion among 120 adult American Indians (28). Results indicated that historical trauma symptoms were associated with past month alcohol use, lifetime use of non-marijuana illicit drugs, and lower family cohesion. The authors used the 12-item scale with single factor model and reported high internal consistency ($\alpha = 0.89$). Anastario and colleagues also examined the relationship between historical trauma symptoms and sexual risk behavior among 120 young American Indian men (29). As a single factor, the HLASS had high internal consistency ($\alpha = 0.88$). The 2-factor model also yielded good internal consistency, with a Cronbach's alpha of 0.73 for anxiety/depression and a Cronbach's alpha of 0.84 for anger/avoidance. Both factors were related to an increased likelihood of respondents' having sex with multiple concurrent partners.

1.4 Adapting HLS for native Hawaiians

Both HLS and HLASS were developed for and by North American Indigenous communities to assess Historical Loss and associated symptoms. In the Native Hawaiian context, the HLS was previously adapted to determine the relationship between Historical Loss and other measures of historical trauma (i.e., historical traumatic events), discrimination, and substance use (30). In the study by Pokhrel and Herzog, researchers omitted items from the original scale that were not relevant to the Native Hawaiian community, which resulted in an adapted 8-item survey administered to 128 Native Hawaiian community college students (*M*age = 27.5; SD = 9.5; 65% women). The 8-item survey administered by Pokhrel and Herzog consisted of the following items: (1) Loss of land, (2) Loss of language, (3) Loss of traditional spiritual ways, (4) Loss of self-respect from poor treatment by government officials, (5) Loss of culture, (6) Loss of respect by children for elders, (7) Loss of people through early death, and (8) Loss of respect by children for traditional ways.

Similar to Whitbeck et al.'s original scale, participants reported the frequency of thinking about historical loss as well as historical traumatic events. Historical loss and historical traumatic events both loaded on a factor of historical trauma. Interestingly, and according to structural equation modeling, the historical trauma factor had a weak and direct effect on substance use, which was mediated by perceived discrimination, resulting in a weak and indirect relationship with substance use. Despite the findings of this study, reliability and goodness of fit statistics were not reported for the HLS. Nonetheless, the adapted HLS was found to be positively and significantly correlated with historical traumatic events and discrimination, indicating convergent validity. For instance, the HLS implemented by Pokhrel and Herzog were moderately and positively associated with historical traumatic events, which was measured by eight items relevant to Native Hawaiian experiences and the perception of participants experiencing these historical traumatic events as well as the participant's parents, grandparents, great-grandparents, and great-great-grandparents.

1.5 Context of study

The Hawaiian Homes Commission Act was signed by U.S. Congress in 1921, with the goal of providing "rehabilitation" for the Native Hawaiian people. These government-sponsored lands span to include 203,500 acres throughout the islands of Hawai'i and are designated to Native Hawaiians who meet the government-assigned blood quantum qualifications, with legal implications for blood quantum as a government metric of Hawaiian identity and ability to reside on Hawaiian Homes. Despite experiences of adversity related to structural issues, Hawaiian Homestead communities serve as a kīpuka (a calm place in a high sea, deep place in a shoal, variation or change of form, or opening in a forest), or space where cultural practices and values continue to be perpetuated despite experiences related to ongoing cultural and historical trauma.

1.6 Purpose of study

Given the need to explore historical loss and associated symptoms in Native Hawaiian communities, the present study aimed to psychometrically test the newly adapted HLS (aHLS) and HLASS scales to support measurement of historical loss and associated symptoms among a Native Hawaiian population residing on Hawaiian Homestead communities.

2 Methods

2.1 Community-based participatory research approach

Community-based participatory research (CBPR) approaches promote social justice and health equity through research approaches that (re)distribute power to communities and ensures community and academic partners are engaged at every step of the research process (30). In this study, CBPR approaches were employed to develop a comprehensive health survey that was mailed to participants from select Hawaiian Homestead communities (See 2.2. Population/Sample Description for further description). Community and academic partnerships previously existed from other research projects and efforts to improve health outcomes for Native Hawaiian health, including a decade-long community-based research project that focused on diabetes and obesity-prevention. During the early phases of the research process, which started in 2013, community and academic partners engaged in various meetings to identify priority areas of interest for the comprehensive health survey. The aHLS and HLASS were of interest to better understand how measures of historical loss were associated with health and health-related outcomes for Native Hawaiian communities.

A comprehensive list of measures was compiled, which included the original HLS and HLASS. Community partners and leaders assisted with survey item adaptation of the HLS. When adapting items from the HLS, there was a strong interest to ensure items were appropriately measuring experiences of Native Hawaiians. Community partners and leaders dedicated time to review each HLS item and adapted the overall scale to better align with Native Hawaiian experiences and communities. Thus, there was a desire to omit items that did not appear relevant to Native Hawaiian experiences and re-word items to increase receptivity by individuals from Native Hawaiian communities. At the time of these initial meetings, there were no publications about the HLS and HLASS being assessed or measured in the Native Hawaiian community.

Given the nature of the survey funding mechanism, the comprehensive survey also included items that measured cancer prevalence, cancer-related health outcomes, and cancer-related behaviors. Based on community guidance and previous academic research, additional sections were included in the comprehensive health survey to assess for variables including demographics and socio-demographic factors; general health and health outcomes including family history of health conditions and health behaviors (including tobacco use, nutrition, physical activity); health-related factors including psychosocial factors, resilience-based factors, and social relations including discrimination, Hawaiian cultural affiliation, Historical Loss, and neighborhood connectedness. Community leaders and partners of this CBPR partnership were engaged in every step of the research process. In alignment with CBPR principles, community partners led survey recruitment, data entry, and data management. Community partners also assisted with the interpretation of data and provided annual community reports to disseminate a summary of the survey data to participants of the Hawaiian Homestead Health Survey. For instance, discussions around the findings and interpretation of data from this study was incorporated in annual meetings held by the Hawaiian Homestead Health Survey team. Furthermore, the final factor labels of study are based on the ongoing discussions and meetings that took place with the Hawaiian Homestead Health Survey team.

2.2 Population/sample description

The final sample of our study comprised 491 Native Hawaiian adult residents of the Hawaiian Homestead Lands from O'ahu and Hawai'i island. Lessees from select Hawaiian Homestead communities were invited to participate in the study based on a mailing list provided by the partner Hawaiian Homestead community. Participants were recruited using a convenience sampling method and based on established relationships with leaders and partners of the Hawaiian Homestead Health Survey team. Additionally, surveys were addressed to the specific "Lessee" of the property. To increase anonymity, the research team had no way to confirm who completed the survey. Instead, participant recruitment and compensation was provided by household.

The general response rate was around 30% of all households, with a total of 512 participants. Of the 512 respondents, 21 did not identify as Hawaiian and were excluded from the final sample of this study, for

TABLE 1 Participant characteristics ($N = 491$).

Characteristics	Mean (SD) or N (%)
Age (years)	55.5 (14.9)
Female (vs. male)	330/491 (67.28%)
Marital status	
Currently single (not married)	113 (23.2%)
Currently married	289 (42.6%)
Divorced/separated/widowed	85 (17.5%)
Educational attainment	
No high school diploma	20 (4.1%)
High school graduate/general education development	173 (35.5%)
Some college/technical school	154 (31.6%)
College graduate	141 (28.9%)
Income ($n = 476$)	
0-less than $30,000	88 (18.5%)
$30,000 to less than $50,000	92 (19.3%)
$50,000 to less than $75,000	76 (16.0%)
$75,000 or more	189 (39.7%)
Do not know or unsure	31 (6.5%)

a final sample size of 491. All participants in the final sample were Native Hawaiian adults, at least 18 years of age. Ages of participants ranged from 18 to more than 90 years, with a mean age of 55.5 (SD = 14.9). Survey respondents were primarily female ($N = 330$, 67.28%) and married ($N = 289$, 42.6%). The majority of the participants had a high school diploma (35.5%), followed by some college or technical school training (31.6%). Table 1 summarizes the characteristics of participants from this study.

2.3 Measures

The primary measures of this study included the aHLS and HLASS, which were included in the Hawaiian Homestead Health Survey. The HLS was originally a 12-item scale, which measured Historical Loss as a unidimensional factor. In alignment with CBPR approaches, the research team consulted community leaders regarding the face validity of the original 12-item scale. Based on continual community feedback, the final adapted scale consisted of eight adapted items. The newly adapted scale included the following items: (1) The taking of our land, (2) Fewer and fewer people using our traditional language, (3) Destruction of our culture and traditional spiritual ways, (4) Loss of respect for elders by our children and grandchildren, (5) Loss of respect by our children for traditional ways, (6) Distrust, resentment, or fear toward white people, (7) Destruction or damage of traditional foods, and (8) The destruction of natural resources and beauty due to pollution, mining, and other industries. This led to the adapted HLS (aHLS) (Table 2).

Similar to the original HLS scale, respondents who completed the aHLS rated the frequency of thinking about Historical Loss on a 6-point Likert scale, ranging from 1 (never) to 6 (several times a day). Thus, the following scores indicated the frequency in which participants thought of the historical loss item: 1 (never), 2 (yearly or only at special times), 3 (monthly), 4 (weekly), 5 (daily), and 6 (several times a day). In the original scale, items were reverse-coded, and thus, higher scores indicated more frequency of thoughts related to Historical Loss. In the aHLS, higher scores indicate greater frequency of thinking about Historical Loss.

The HLASS is a 12-item scale which measures the frequency of experiencing specific symptoms when thinking about the Historical Loss listed in the HLS. The specific symptoms include depression, anger, anxiety, being uncomfortable around White people, shame, loss of concentration, isolation, loss of sleep, rage, being fearful and distrusting of White people, feeling like it is happening again, and avoiding places that remind them of Historical Loss (Table 3). After much discussion and taking a CBPR approach, it was determined that the original HLASS scale would be appropriate to include in the comprehensive health survey without any adaptations. In other literature, these items have been found to load on to two factors, anxiety/depression (five items) and anger/avoidance (seven items) (22). Respondents rate the frequency of experiencing each feeling on a 5-point Likert scale, from 1 (never) to 5 (always). Thus, the following scores indicated the frequency in which participants experienced the associated symptoms when thinking about the historical loss: 1 (never), 2 (seldom), 3 (sometimes), 4 (often), and 5 (always). Similar to the HLS, higher scores of HLASS items indicate greater frequency of experiencing the associated symptoms (Table 4).

2.4 Procedures and statistical analyses

All procedures of this study were approved by community partners and the University of Hawaiʻi Institutional Review Board. Data from this study were based on the Hawaiian Homestead Health Survey administered between 2014 and 2020. The overall comprehensive survey was developed using CBPR approaches and measured demographic variables, general health measures, socio-cultural determinants of health, and psychosocial factors associated with health and health-related behaviors in adult Native Hawaiians residing on Hawaiian homestead lands.

Cognitive interviews were conducted in 2014 to pilot the comprehensive health survey. Cognitive interviewing is a method used to pre-test surveys and gather in-depth responses and insights about items to ensure constructs and surveys are measuring what they intend to measure, and thus, is a form of validity (31). In this case, the research team obtained verbal information about the drafted comprehensive health survey, which included items from the adapted Historical Loss Scale and Historical Loss Associated Symptoms Scale. Cognitive interviews were particularly helpful to reduce response errors and improve comprehension of the overall survey (31).

One survey packet was mailed to each household from select Hawaiian Homestead lands. Survey packets included a consent form, a personalized cover letter with information describing the purpose of the project, and the Hawaiian Homestead Health Survey. Those who consented to participate returned the completed survey in a pre-addressed envelope. Upon receipt of the completed survey, a $25 gift card was provided to the household as compensation for their time. An ID number was assigned to each survey to ensure confidentiality. Survey data were entered in REDCap, a secured, electronic database, then exported to SAS 9.4 to calculate participant

TABLE 2 List of historical loss scale items.

Items from Whitbeck et al.'s scale	Items from Pokhrel & Herzog's scale	Items from the aHLS from the Hawaiian homestead health survey
(1) The loss of our land	(1) Loss of land	(1) The taking of our land
(2) The loss of our language	(2) Loss of language	(2) Fewer and fewer people using our traditional language
(3) Losing our traditional spiritual ways	(3) Loss of traditional spiritual ways	(3) Destruction of our culture and traditional spiritual ways
(4) The loss of our family ties because of boarding schools		
(5) The loss of families from the reservation to government relocation		
(6) The loss of self respect from poor treatment by government officials	(4) Loss of self-respect from poor treatment by government officials	
(7) The loss of trust in White individuals from broken treaties		(6) Distrust, resentment, or fear toward white people
(8) Losing our culture	(5) Loss of culture	
(9) The losses from the effects of alcoholism on our people		
(10) Loss of respect by our children and grandchildren for elders	(6) Loss of respect by children for elders	(4) Loss of respect for elders by our children and grandchildren
(11) Loss of our people through early death	(7) Loss of people through early death	
(12) Loss of respect by our children for traditional ways	(8) Loss of respect by children for traditional ways	(5) Loss of respect by our children for traditional ways
		(7) Destruction or damage of traditional foods
		(8) The destruction of natural resources and beauty due to pollution, mining, and other industries

The items listed above are accompanied with the following prompt: Native Hawaiians have experienced many events, traumas, and changes since coming into contact with Europeans (White individuals). Below is a series of things like this that people have mentioned to us. Please indicate how often you think of these whether or not you are Native Hawaiian. Answer responses are scored as: (1) never, (2) yearly or only at special times, (3) monthly, (4) weekly, (5) daily, and (6) several times a day. Scoring follows the recommendation provided by Whitbeck et al. of having higher scores indicate higher levels of historical loss.

TABLE 3 List of historical loss associated symptoms scale items.

Items from Whitbeck et al.'s original scale
(1) Sadness or depression
(2) Anger
(3) Anxiety or nervousness
(4) Uncomfortable around white people (when you think of these losses)
(5) Shame (when you think of these losses)
(6) A loss of concentration
(7) Feel isolated or distant from other people (when you think of these losses)
(8) A loss of sleep
(9) Rage
(10) Fearful or distrust the intention of white people
(11) Feel like it is happening again
(12) Feel like avoiding places or people that remind you of these losses

The items listed above are accompanied with the following prompt: Now I would like to ask you about how you feel when you think about these losses. Answer responses are scored as: (1) never, (2) seldom, (3) sometimes, (4) often, and (5) always. Scoring follows the recommendation provided by Whitbeck et al. of having higher scores indicate higher levels of historical loss associated symptoms.

characteristics, conduct inter-correlation matrixes, and create Mplus files. Confirmatory Factor Analysis (CFA) and Exploratory Factor Analysis (EFA) were calculated using Mplus Version 8.5.

By default of the MPlus program, CFA consisted of geomin rotated solution, which is a type of oblique rotation, with correlations between factors provided in the Mplus output. Additional specifications were provided within the Mplus program including each item being treated as a categorical variable. For the purpose of this study, standardized factor loadings of the final CFA models are also provided in the figures. Model specifications of EFA analyses included labeling items as categorical variables. Similar to CFA models and by default of the Mplus program, Geomin rotation was utilized for final EFA models. The weighted least square mean and variance adjusted (WLSMV) estimator was utilized for all factor analysis models. This estimator is utilized when data is classified as categorical and uses pairwise deletion. Based on the missing pattern frequencies generated by MPlus, it was confirmed that 463 (out of 491, 94.3%) participants completed the full survey.

In cases where the research team conducted EFA, a cross-validation analysis was utilized. For this process, the final sample was randomly split into two halves, referred to as Sample 1 ($n = 245$) and Sample 2 ($n = 246$). There were no statistically significant differences between demographic variables such as age, gender, number of children, number of people living in household, number of years living on Hawaiian Homestead lands and representation from various homestead communities. During the cross-validation analysis process, Sample 1 was utilized for EFA models, which were then validated through a CFA of the selected EFA model using data from Sample 2. Items were considered to load on a factor if factor loadings were at least 0.4 or greater.

TABLE 4 Mean scores of the adapted historical loss scale (aHLS) and historical loss associated symptoms scale (HLASS).

aHLS or HLASS item or factor	Mean (SD)
Mean of adapted historical loss scale (aHLS) items*	
aHLS1 (the taking of land)	3.0 (1.4)
aHLS2 (fewer and fewer people using our traditional language)	3.0 (1.4)
aHLS3 (destruction of our culture and traditional spiritual ways)	3.1 (1.5)
aHLS4 (loss of respect for elders by our children and grandchildren)	3.6 (1.5)
aHLS5 (loss of respect by our children for traditional ways)	3.3 (1.5)
aHLS6 (distrust, resentment, or fear toward white people)	2.5 (1.5)
aHLS7 (destruction or damage of traditional foods)	2.7 (1.5)
aHLS8 (the destruction of natural resources and beauty due to pollution, mining, and other industries)	3.5 (1.6)
Mean of aHLS score based on Armenta's study*	
Mean of cultural loss factor	3.0 (1.3)
Mean of cultural mistreatment factor	3.5 (1.4)
Mean of aHLS score based on EFA*	
Mean of cultural loss factor	3.0 (1.3)
Mean of intergenerational loss factor	3.5 (1.4)
Mean of distrust and destruction of traditional foods factor	2.5 (1.4)
Mean of hierarchical factor	3.0 (1.2)
Historical loss associated symptoms scale (HLASS) items**	
HLASS1 (sadness or depression)	2.5 (1.1)
HLASS2 (anger)	2.6 (1.1)
HLASS3 (anxiety or nervousness)	2.0 (1.0)
HLASS4 (uncomfortable around white people)	1.8 (0.9)
HLASS5 (shame)	1.9 (1.0)
HLASS6 (a loss of concentration)	1.7 (0.9)
HLASS7 (feel isolated or distant from other people)	1.7 (0.9)
HLASS8 (a loss of sleep)	1.5 (0.8)
HLASS9 (rage)	1.6 (1.0)
HLASS10 (fearful or distrust the intention of white people)	2.1 (1.1)
HLASS11 (feel like it is happening again)	2.3 (1.1)
HLASS12 (feel like avoiding places or people that remind you of these losses)	1.8 (1.0)
Mean of historical loss associated symptoms scale (HLASS)**	
Mean of depression and anger	2.5 (0.9)
Mean of shame and anxiety	1.7 (0.7)
Mean of re-experiencing, fear, and avoidance	2.1 (0.9)
Mean of hierarchical factor	2.1 (0.7)

*In the table above, the total possible score for the mean of adapted historical loss scale (aHLS) score, cultural loss, intergenerational loss, and distrust and destruction of traditional foods ranged from 1 (never) to 6 (several times a day).

**In the table above, the total possible score for the mean of HLS score, cultural loss, intergenerational loss, and distrust and destruction of traditional foods ranged from 1 (never) to 5 (always).

Research over the decades suggest different approaches to reporting goodness-of-fit indices (32). While there are various goodness-of-fit indices that may be considered in structural equation modeling and factor analyses, there are two specific statistics that will be evaluated for the purpose of this study: (1) RMSEA, which considers the error or residual of a model by observing the discrepancy between observed values and predicted values when optimal parameters are chosen and (2) CFI, a goodness of fit statistic that determines the way the structural model fits the observed data adjusting for sample size. In particular, a model was considered to have good or acceptable fit based on goodness of fit statistics, including chi-square, degrees of freedom, Root Mean Square Error of Approximation (RMSEA) and Comparative Fit Index (CFI), with attention primarily given to RMSEA and CFI. For comparative purposes, values are also provided for the Standardized Root Mean Residual (SRMR) and Tucker–Lewis Index (TLI).

TABLE 5 **Summary of confirmatory factor analyses and exploratory factor analyses results and decision matrix for the adapted historical loss scale (aHLS).**

Model	Sample	Chi-square test of model fit	*df*	SRMR	RMSEA	CFI	TLI	Model fit decision
HLS CFA models								
Null	1&2 Combined	9519.83*	28	0.39	0.89	0.00	0.00	Poor fit
One-factor model	1&2 Combined	696.68*	20	0.05	0.29	0.93	0.90	Poor fit
Adapted 2-factor model (based on the suggested 3-factor model provided by Armenta et al. without the adapted items) **F1: items 1–3** **F2: items 4 and 6**	**1&2 Combined**	**8.345***	**10**	**0.01**	**0.05**	**1.00**	**1.00**	**Excellent fit; acceptable RMSEA, SRMR, and CFI**
Three factor model (based on the suggested model provided by Armenta et al. and the adapted items) F1: items 1–3 F2: items 4 and 6 F3: items 5, 7, and 8	1&2 Combined	388.71*	28	0.04	0.26	0.96	0.93	Acceptable SRMR and CFI, poor RMSEA, poor TLI
Three factor model (based on the suggested model provided by Armenta et al. and the adapted items) F1: items 1–2 F2: items 3, 4, and 6 F3: items 5, 7, and 8	1&2 Combined	578.29*	17	0.05	0.29	0.94	0.90	Acceptable SRMR, poor RMSEA, CFI, TLI

In the table above, the *indicates a chi-square test of model fit with a *p*-value that is less than 0.05 indicating statistical significance. The bolded row indicates the final model selected for this study.

RMSEA values range from 0 to 1, with lower values demonstrating better model fit (33, 34). The acceptable cutoff values for the RMSEA are rather debatable. Some argue that RMSEA values of 0.08 are acceptable, while others recommend an RMSEA cutoff value of 0.07 (33). Given the adapted HLS and implementation of new items, the cutoff selected for this study as "acceptable" is an RMSEA of 0.08 or less. SRMR uses similar goodness of fit criteria as RMSEA but differs due to its purpose of measuring differences between the observed correlation and the model implied correlation matrix. In this study, the RMSEA is more highly weighted due to the SRMR being positively biased, with greater bias for studies with smaller N sizes. CFI compares the proposed model to the null model, a model where there is no correlation between all of the observed variables. CFI works well with small samples and tends to be highly correlated with the TLI. CFI also ranges from 0 to 1, however, larger values indicate better model fit. Acceptable model fit is indicated by a CFI value of 0.95 or greater (33).

The minimal cutoff of Cronbach alpha for each scale and construct was set at 0.70. Final models were selected based on a combination of goodness of fit statistics and meaningfulness for the proposed models. Reliability was measured after the final models were selected using Cronbach's alpha and McDonald's Omega, which were calculated using JASP software. Different forms of validity include measures such as construct validity, including factorial validity, convergent validity, and discriminant validity (35). Construct validity, a form of validity that indicates that a scale is accurately measuring a construct of interest. Factorial validity exists when an existing hypothesized structure is confirmed through analyses such as CFA. Convergent validity exists when scales or tests overlap in measuring the same construct, while divergent validity exists when scales or tests do not correlate with one another due to the assumption that the scales are measuring two different constructs (35). With these definitions in mind, it is important to note that construct validity, specifically convergent validity, and discriminant validity, may not be achieved in this study due to the primary focus on the aHLS and HLASS. Instead, correlational matrices demonstrating patterns and associations between items and factors of the aHLS and HLASS are reported.

3 Results

3.1 HLS psychometric models

A summary of the CFA and EFA model results for the aHLS are presented in Tables 5, 6. A Confirmatory Factor Analysis (CFA) was conducted to determine model fit indices for a null model (with 0 correlations set for each item), one-factor model (22), and the suggested 3-factor model proposed by Armenta, Whitbeck, and Habecker (20) based on the items included in the aHLS. The 3-factor CFA model for the purpose of this study and based on the model suggested by Armenta and colleagues (20) comprised cultural loss, cultural mistreatment, and the newly developed items. While the two-factor model with original items (i.e., a two-factor model with the cultural loss and cultural mistreatment items) demonstrated good model fit with an RMSEA of 0.05 and CFI of 1.00, the added items with these two proposed factors demonstrated poor model fit (refer to Table 7).

TABLE 6 Summary of confirmatory factor analyses and exploratory factor analyses results and decision matrix for the adapted historical loss scale (aHLS).

Model	Sample	Chi-square test of model fit	*df*	SRMR	RMSEA	CFI	TLI	Model fit decision
HLS cross-validation models (based on 8-items)								
One-factor EFA model (items 1–8)	Sample 1	450.77*	20	0.09	0.30	0.93	0.90	Poor fit
Two-factor EFA model (items 1–8)	Sample 1	167.84*	13	0.04	0.23	0.98	0.95	Acceptable SRMR, CFI, and TLI, poor RMSEA
Three-factor EFA model (items 1–8)	Sample 1	87.51*	7	0.03	0.22	0.99	0.95	Acceptable SRMR, CFI, and TLI, poor RMSEA
Four-factor EFA model* **(items 1–8)** F1: items 1–3 F2: items 4–5 F3: items 6–7 F4: item 8	**Sample 1**	**2.46***	**2**	**0.01**	**0.03**	**1.00**	**1.00**	**Excellent fit; acceptable RMSEA, SRMR, and CFI**
HLS cross-validation methods (based on 7-items)								
One-factor EFA model (items 1–7)	Sample 1	368.11*	21	0.10	0.33	0.94	0.91	Poor fit
Two-factor EFA model (items 1–7)	Sample 1	107.04*	21	0.04	0.23	0.98	0.96	Acceptable SRMR, CFI, and TLI, poor RMSEA
Three-factor EFA model (items 1–7) F1: items 1–3 F2: items 4–5 F3: items 6–7	**Sample 1**	**5.65***	**21**	**0.01**	**0.06**	**1.00**	**1.00**	**Excellent fit; acceptable RMSEA, SRMR, and CFI**
CFA of 3-factor EFA model F1: items 1–3 F2: items 4–5 F3: items 6–7	Sample 2	31.67	21	0.01	0.09	1.00	0.99	Good fit
Hierarchical CFA of 3-factor EFA model F1: items 1–3 F2: items 4–5 F3: items 6–7 F4: F1 F2 F3	Sample 2	31.67	21	0.01	0.09	1.00	0.99	Good fit

In the table above, the *indicates a chi-square test of model fit with a *p*-value that is less than 0.05 indicating statistical significance. The bolded row indicates the final model selected for this study.

Due to poor model fit for the CFA models, an Exploratory Factor Analysis (EFA) was conducted using a cross-validation method. The EFA sample matrix correlation yielded the following eigenvalues based on data from Sample 1: 5.628, 0.734, 0.478, 0.462, 0.230, 0.188, 0.169, and 0.112. Thus, eigenvalues indicated that factor 1 represents the greatest magnitude of importance for the EFA models. However, based on a combination of model fit indices and meaningfulness of factors for sample 1, the 7 item, 3-factor model was selected as the best EFA model with an RMSEA of 0.06 and a CFI of 1.00. Of the eight adapted items, one item (Item 8) was omitted as it operated as a one-item factor when included in the EFA (refer to Table 8). A CFA of the selected 7-item, 3-factor model continued to demonstrate good fit based on data from Sample 2 with an RMSEA of 0.08 and CFI of 1.00. Figure 1 visually depicts the final selected psychometric model of aHLS as a hierarchical 3-factor model with standardized factor loadings.

3.2 HLASS psychometric models

A summary of the CFA and EFA model results for the HLASS are presented in Table 9. Goodness of fit statistics were assessed for the HLASS using CFA and EFA models. CFA models were conducted to determine goodness of fit statistics for a null model (with 0 correlations set for each item), one-factor model (27), and the suggested 2-factor model proposed by Whitbeck and colleagues (22). The suggested 2-factor model comprised anxiety/depression and anger/avoidance. Based on poor model fit for the HLASS CFA models, an EFA was conducted using a cross-validation method.

The EFA sample matrix correlation yielded the following eigenvalues based on data from Sample 1: 6.744, 1.386, 0.970, 0.621, 0.515, 0.461, 0.347, 0.295, 0.276, 0.173, 0.129, and 0.083. Thus, eigenvalues supported a multidimensional model for the HLASS, with

TABLE 7 Factor structure loadings for confirmatory factor analysis based on Armenta's model.

Item no.	Adapted historical loss scale (aHLS)	Factor 1: cultural loss	Factor 2: cultural mistreatment
Confirmatory factor analysis based on Armenta's model (Geomin model specification, standardized factor loadings, items 1–4 and 6, both samples, $n = 491$)			
1	The taking of our land	0.827	
2	Fewer and fewer people using our traditional language	0.882	
3	Destruction of our culture and traditional spiritual ways	0.925	
4	Loss of respect for elders by our children and grandchildren		0.718
6	Distrust, resentment, or fear toward white people		0.726

2 or 3 factors demonstrating magnitude of importance for the EFA models. Based on goodness of fit statistics for the EFA models using data from Sample 1, the three-factor model was selected. Goodness of fit statistics for the 3-factor EFA model with all items were acceptable with an RMSEA of 0.07 and a CFI of 0.99. However, of the 12 items included in the HLASS, three items were omitted. Two of the 12 items did not load on a factor including HLASS Item 4: Uncomfortable around white people (when you think of these losses) and HLASS Item 9: Rage, whereas Item 3: Anxiety or nervousness double-loaded on Factor 1 and 2 (refer to Table 10). Figure 2 visually depicts the final selected psychometric model of HLASS as a hierarchical 3-factor model with standardized factor loadings.

3.3 Reliability and inter-correlation matrix

Tables 11, 12 present the internal consistency coefficients (i.e., reliability) for the aHLS and HLASS scales and suggested subscales. Overall, the results demonstrated good reliability based on McDonald's Omega and the standardized Cronbach alpha, which ranged from 0.76 to 0.92. Inter-correlation matrices were generated to determine the bivariate relationships among the items and final factors included in the aHLS and HLASS. As shown in Tables 11–13, all the inter-factor and inter-scale correlations were statistically significant at the $p < 0.01$ level. The correlations demonstrated positive and significant relationships between the set of aHLS and HLASS items and factors, with correlations for the aHLS ranging from 0.39 to 0.93; correlations for the HLASS ranging from 0.25 to 0.90; and the correlation between aHLS and HLASS being 0.59. The inter-correlation matrices also demonstrated higher correlations for respective constructs.

4 Discussion

The overall purpose of this study was to estimate psychometric properties of the aHLS and HLASS in a Native Hawaiian sample. The findings of this study suggested both the aHLS and HLASS may be most acceptable as a three-factor model when measuring Historical Loss and associated symptoms in Native Hawaiian communities. The aHLS comprised factors that assessed for loss of culture, intergenerational loss, and distrust and destruction of traditional foods. The HLASS comprised factors that measured depression and anger, shame and anxiety, and re-experiencing, fear, and avoidance. The aHLS, which used seven of the eight adapted items proposed by community members and leaders, demonstrated good model fit for the Native Hawaiian sample. The proposed 3-factor model of the HLASS overlapped with the emotional responses identified through the original factor structure proposed by Whitbeck and colleagues (22). The results of this study also demonstrated good reliability based on McDonald's Omega and the standardized Cronbach alpha.

The HLS and HLASS were originally developed as standardized measures to assess the direct and indirect impacts of Historical Loss on other Indigenous populations, specifically North American Indigenous communities (22). Our findings support the use of the aHLS and HLASS in Native Hawaiian populations. Our findings further validate the importance of measuring Historical Loss and associated symptoms among Native Hawaiians as multidimensional constructs. This aligns with other research that supports the importance of addressing trauma, including cultural and historical trauma and community injustice, through a multidimensional lens (19, 36). For instance, the aHLS as a 3-factor model was most highly correlated with depression and anger followed by shame and anxiety. These associated symptoms of Historical Loss have substantial public health impacts and are negatively associated with multi-level social determinants of health, quality of life, as well as overall health including increased risk of chronic health conditions (37–39). By addressing Historical Loss as historical trauma, and thus, a multidimensional determinant of health, cultural strengths and values that address Historical Loss may directly and indirectly increase the resiliency of Native Hawaiians; thereby fostering wellness and wellbeing.

The development of the aHLS and pilot testing of the HLASS through community consultation helped to increase the face validity of these constructs. This is not surprising given the ongoing research that emphasizes the importance of taking a community-based and culturally sensitive approach to research with Native and Indigenous communities at large (1, 40, 41). In this case, engaging community in every step of the research process aided in the adaptation of culturally sensitive and appropriate measures of Historical Loss and associated symptoms of Historical Loss. The aHLS and the HLASS can be used to reliably assess the frequency with which Native Hawaiians think of Historical Loss and the negative emotions that are experienced when Native Hawaiians think of these Historical Loss.

Frequency distributions and mean scores indicated that on average, participants would think of the historical loss items on a monthly basis, with participants indicating greatest frequency of thinking about the loss of respect for elders by our children and grandchildren and the destruction of natural resources and beauty due to pollution, mining, and other industries. These findings correspond with Whitbeck et al.'s original published article on HLS, which indicated that the majority of participants would think of cultural loss or cultural mistreatment items (i.e., loss of our land, the loss of our family ties because of boarding schools, the loss of families from the reservation to government relocation, the loss of self-respect from poor treatment by government officials, and the loss of trust in White individuals from broken treaties) "never" or "yearly" during special times of the year. On the other hand, items from Whitbeck et al.'s original scale that assessed for loss of language, loss of

TABLE 8 Factor structure loadings of final exploratory and confirmatory factor analysis models for the adapted historical loss scale (aHLS).

Item no.	Adapted historical loss scale (aHLS)	Factor 1: loss of culture	Factor 2: inter-generational loss	Factor 3: distrust and destruction of traditional foods	Factor 4: destruction of natural resources
Exploratory factor analysis 4-factor model (Geomin rotated model results, all items, sample 1, $n = 245$)					
1	The taking of our land	**0.618***	−0.081*	0.054	0.324
2	Fewer and fewer people using our traditional language	**0.772***	0.036	0.128	−0.044
3	Destruction of our culture and traditional spiritual ways	**0.909***	0.089	−0.084	0.031
4	Loss of respect for elders by our children and grandchildren	−0.012	**0.971***	−0.014	0.022
5	Loss of respect by our children for traditional ways	0.064	**0.808***	0.094	0.006
6	Distrust, resentment, or fear toward white people	−0.011	0.041	**0.925***	0.005
7	Destruction or damage of traditional foods	0.058	−0.002	**0.608***	0.284*
8	The destruction of natural resources and beauty due to pollution, mining, and other industries	0.002	0.083	−0.001	**0.975***

Item no.	Adapted historical loss scale (aHLS)	Factor 1: loss of culture	Factor 2: inter-generational loss	Factor 3: distrust and destruction of traditional foods
Exploratory factor analysis 3-factor model (Geomin rotated model results, items 1–7, sample 1, $n = 245$)				
1	The taking of our land	**0.597***	−0.028	0.287*
2	Fewer and fewer people using our traditional language	**0.653***	0.014	0.236*
3	Destruction of our culture and traditional spiritual ways	**0.910***	0.054	−0.003
4	Loss of respect for elders by our children and grandchildren	0.188	**0.746***	−0.008
5	Loss of respect by our children for traditional ways	−0.003	**0.972***	0.059
6	Distrust, resentment, or fear toward white peoples	0.000	0.016	**0.877***
7	Destruction or damage of traditional foods	0.008	0.005	**0.860***

Item no.	Adapted historical loss scale (aHLS)	Factor 1: loss of culture	Factor 2: inter-generational loss	Factor 3: distrust and destruction of traditional foods
Final confirmatory factor analysis of 3-factor exploratory factor model (Geomin model specification, standardized factor loadings, items 1–7, sample 2, $n = 246$)				
1	The taking of our land	**0.831**		
2	Fewer and fewer people using our traditional language	**0.884**		
3	Destruction of our culture and traditional spiritual ways	**0.949**		
4	Loss of respect for elders by our children and grandchildren		**0.906**	
5	Loss of respect by our children for traditional ways		**0.931**	
6	Distrust, resentment, or fear toward white peoples			**0.759**
7	Destruction or damage of traditional foods			**0.936**

In the table above, the *indicates significance at 0.05 level. Loadings greater than 0.4 are bolded.

traditional spiritual ways, loss of culture, loss of people from the effects of alcoholism, loss of respect by our children and grandchildren for elders, loss of our people through early death, and loss of respect by children tended to have higher frequencies, with participants reporting thinking about these losses on a daily basis.

Additionally, reported associated symptoms were seldom experienced when thinking about the historical loss items. It is possible these lower mean scores are attributed to the final items that were ultimately adapted and included in these surveys. On the other hand, it is possible participants from this study do not necessarily view these items as "loss" or "historical loss" due to the ongoing resistance and resilience that continues to preserve and perpetuate Hawaiian knowledge, intergenerational strengths, and ways of knowing. This was showcased during our initial meetings, where community leaders and partners indicated the need to change items such as "loss of our land" to the "taking of our land." Re-framing this item demonstrates the way in which land is still present and nourishing us as people, but the current political and economic power structures limit the ability to be intimate with land due to the taking of land. This also sheds light on the ongoing fight for justice and self-determination, with implications for historical, present, and future

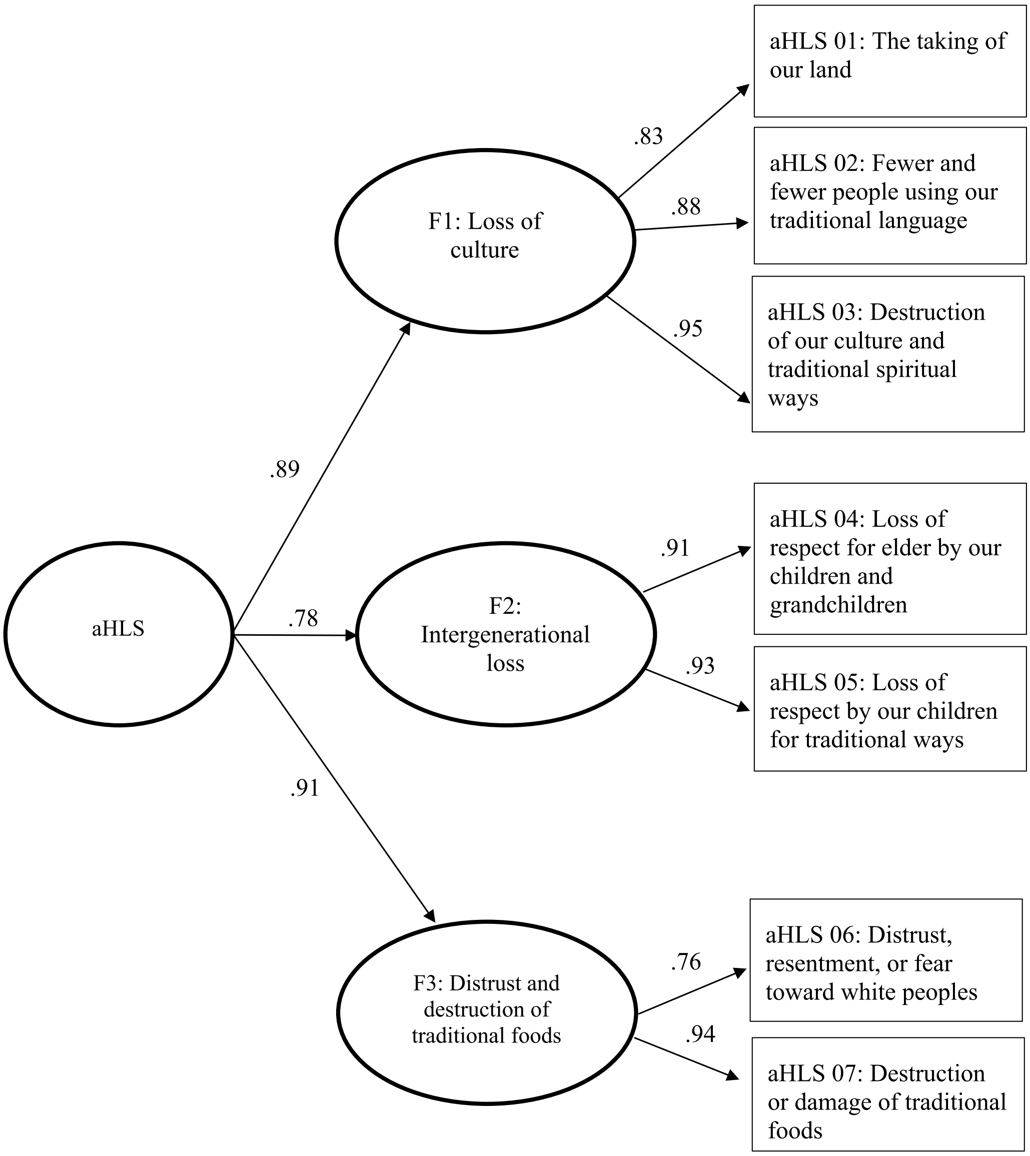

FIGURE 1
Psychometric model of aHLS as a hierarchical 3-factor model with standardized factor loadings.

loss. Thus, future studies that aims to explore historical trauma must also consider the impacts of larger and systemic barriers that also play a role in the health and wellbeing of Native Hawaiians, which extends further to Indigenous Peoples at large.

Overall, an exploration of historical trauma and symptoms associated with historical loss and historical trauma continue to be an urgent and important public health topic, especially given the social, cultural, economic, and political implications of present-day adversities. These adversities range to include the exacerbation of health factors through large public health issues including the COVID-19 pandemic (42), increased crises related to housing and affordable living, and devasting impacts of the natural environment, including the wildfires of Maui. Gaining a better understanding of Historical Loss and historical loss symptoms may assist with more upstream, systemic solutions that may better the health and wellbeing of Native Hawaiian through a social justice lens. For example, by associating intergenerational loss with current mental and emotional health conditions of Native Hawaiians, community, researchers, academicians, and practitioners will be able to illustrate the severity of addressing colonial atrocities like water rights issues in Hawaiʻi. To illustrate, historical and contemporary American capitalist ventures in Hawaiʻi, particularly plantations and tourism, resulted in the diversion of natural water ways, impacting the ancestral ways of life in which Native Hawaiians interact with land. Natural resources and agricultural systems that maintained Native Hawaiian health have been threatened by desecration, serving as a catalyst of health inequities for contemporary Native Hawaiians and the increased reliance on imported foods. Applying findings from aHLS and HLASS quantify the need to restore ancestral ways of life, including natural flowing waters, as means to address intergenerational loss.

4.1 Limitations

The findings from this study are based on cross-sectional survey data. As with any cross-sectional study, the findings from this study are limited

TABLE 9 Summary of confirmatory factor analyses and exploratory factor analyses results and decision matrix for the historical loss associated symptoms scale (HLASS).

Model	Sample	Chi-square test of model fit	*df*	SRMR	RMSEA	CFI	TLI	Model fit decision
HLASS CFA models								
Null	1&2 Combined	7283.10*	66	0.36	0.51	0.00	0.00	Poor fit
One-factor model	1&2 Combined	766.10*	66	0.07	0.17	0.91	0.90	Poor fit
Two-factor model F1: items 1, 3, 6, 7, and 8 F2: items 2, 4, 5, 9, 10, 11, 12	1&2 Combined	679.92*	53	0.07	0.16	0.92	0.91	Poor fit
HLASS EFA models (12 items)								
One-factor EFA model	Sample 1	496.55*	54	0.12	0.19	0.90	0.88	Poor fit
Two-factor EFA model	Sample 1	238.94*	43	0.07	0.14	0.96	0.93	Acceptable SRMR and CFI, poor RMSEA and TLI
Three-factor EFA model **items 1, 2** **items 5, 6, 7, 8,** **items 10, 11, 12**	**Sample 1**	**70.06***	**33**	**0.03**	**0.07**	**0.99**	**0.98**	**Good fit; acceptable RMSEA, SRMR, CFI, and TLI**
Four-factor EFA model	Sample 1	No Convergence	n/a	n/a	n/a	n/a	n/a	No Convergence
Five-factor EFA model	Sample 1	31.67*	16	0.02	0.06	1.00	0.99	**Excellent fit; acceptable RMSEA, SRMR, CFI, and TLI**
Six-factor EFA model	Sample 1	No convergence	n/a	n/a	n/a	n/a	n/a	No convergence
HLASS EFA models (9 items after dropping items 3, 4, and 9)								
One-factor EFA model	Sample 1	406.88*	27	0.15	0.24	0.89	0.86	Poor fit
Two-factor EFA model	Sample 1	175.57*	19	0.07	0.19	0.96	0.92	Acceptable SRMR and CFI, poor RMSEA and TLI
Three-factor EFA model **items 1, 2** **items 5, 6, 7, 8,** **items 10, 11, 12**	**Sample 1**	**28.44***	**12**	**0.02**	**0.07**	**1.00**	**0.99**	**Good fit; acceptable RMSEA, SRMR, CFI, and TLI**
HLASS CFA of EFA model								
CFA of the 3-factor EFA model F1: items 1, 2 F2: items 5, 6, 7, 8 F3: items 10, 11, 12	Sample 2	61.24*	24	0.03	0.08	0.99	0.98	**Acceptable fit; acceptable RMSEA, SRMR, CFI, and TLI**
Hierarchical CFA of the 3-factor EFA model F1: items 1, 2 F2: items 5, 6, 7, 8 F3: items 10, 11, 12 F4: F1 F2 F3	Sample 2	61.24*	24	0.03	0.08	0.99	0.98	**Acceptable fit; acceptable RMSEA, SRMR, CFI, and TLI**

In the table above, the *indicates a chi-square test of model fit with a *p*-value that is less than 0.05 indicating statistical significance. The bolded row indicates the final model selected for this study.

to data based on one point in time. As a result, causal statements may not be drawn. Furthermore, responses may have differed depending on the date in which the survey was administered. Survey administration took place between 2014 and 2020, with various significant and historical events taking place during this time, specifically in the context of Hawaiian history. For example, in 2015 and 2019, the threat of a Thirty Meter Telescope atop a sacred mountain, Mauna Kea, caused an uprising among many Native Hawaiians. Such an event illustrates the continual contention between Native Hawaiians and settlers who perpetuate Native Hawaiian cultural erasure, degradation, and trauma. These contextual factors are therefore important to consider for data interpretation. For instance, the forementioned present-day historical trauma may intersect with health consequences that stem from historical trauma of past, thereby resulting in negative health outcomes, including the inequities experienced by contemporary Native Hawaiians. Furthermore, although the data included participants from various Hawaiian Homestead communities, these findings may not be generalizable to other Hawaiians, including those who currently reside or do not reside on Hawaiian Homestead Lands as well as those who currently reside or do not reside in Hawaiʻi. Therefore, future research should consider further psychometric analysis of the proposed aHLS and HLASS among Hawaiian communities at large.

TABLE 10 Factor structure loadings of final exploratory and confirmatory factor analysis models for the historical loss associated symptoms scale (HLASS).

Item no.	Historical loss associated symptoms scale (HLASS)	Factor 1: depression and anger	Factor 2: shame and anxiety	Factor 3: re-experiencing, fear, and avoidance
Exploratory factor analysis 3-factor model (Geomin rotated model results, all items, sample 1, $n = 245$)				
1	Sadness or depression	**0.734***	0.045	0.089
2	Anger	**0.751***	−0.030	0.194
3	Anxiety or nervousness	**0.539***	**0.469***	−0.014
4	Uncomfortable around white people (when you think of these losses)	0.156	0.375*	0.388*
5	Shame (when you think of these losses)	0.216*	**0.556**	0.046
6	A loss of concentration	−0.009	**0.930**	0.036
7	Feel isolated or distant from other people (when you think of these losses)	−0.008	**0.756***	0.250*
8	A loss of sleep	0.137	**0.742***	−0.022
9	Rage	0.318*	0.223*	0.360*
10	Fearful or distrust the intention of white people	−0.053	0.013	**0.979***
11	Feel like it is happening again	0.081	−0.042	**0.874***
12	Feel like avoiding places or people that remind you of these losses	−0.002	0.225*	**0.624***

Item no.	Historical loss associated symptoms scale (HLASS)	Factor 1: depression and anger	Factor 2: shame and anxiety	Factor 3: re-experiencing, fear, and avoidance
Exploratory factor analysis 3-factor model (Geomin rotated model results, items 1–2, 5–8, 10–12, sample 1, $n = 245$)				
1	Sadness or depression	**0.731***	0.061	0.009
2	Anger	**0.907***	−0.007	−0.005
5	Shame (when you think of these losses)	0.225*	**0.561***	0.013
6	A loss of concentration	−0.026	**0.950***	0.008
7	Feel isolated or distant from other people (when you think of these losses)	−0.005	**0.781***	0.221*
8	A loss of sleep	0.094	**0.778***	−0.054
10	Fearful or distrust the intention of white people	−0.023	0.082	**0.895***
11	Feel like it is happening again	0.124	−0.012	**0.859***
12	Feel like avoiding places or people that remind you of these losses	0.018	0.258*	**0.595***

Item no.	Historical loss associated symptoms scale (HLASS)	Factor 1: depression and anger	Factor 2: shame and anxiety	Factor 3: re-experiencing, fear, and avoidance
Final confirmatory factor analysis of 3-factor exploratory factor model (Geomin model specification, standardized factor loadings, sample 2, $n = 246$)				
1	Sadness or depression	0.799		
2	Anger	0.850		
5	Shame (when you think of these losses)		0.702	
6	A loss of concentration		0.927	
7	Feel isolated or distant from other people (when you think of these losses)		0.955	
8	A loss of sleep		0.772	
10	Fearful or distrust the intention of white people			0.830
11	Feel like it is happening again			0.896
12	Feel like avoiding places or people that remind you of these losses			0.815

In the table above, the *indicates significance at 0.05 level. Loadings greater than 0.4 are bolded.

4.2 Conclusion

Measurement of the aHLS and HLASS in the Native Hawaiian homestead population paves the way for quantitative assessments of historical trauma in Native Hawaiian communities at large. A better understanding of how the three factors identified in the aHLS relate to the three factors identified in the HLASS is needed to better understand the relationship between historical loss and associated

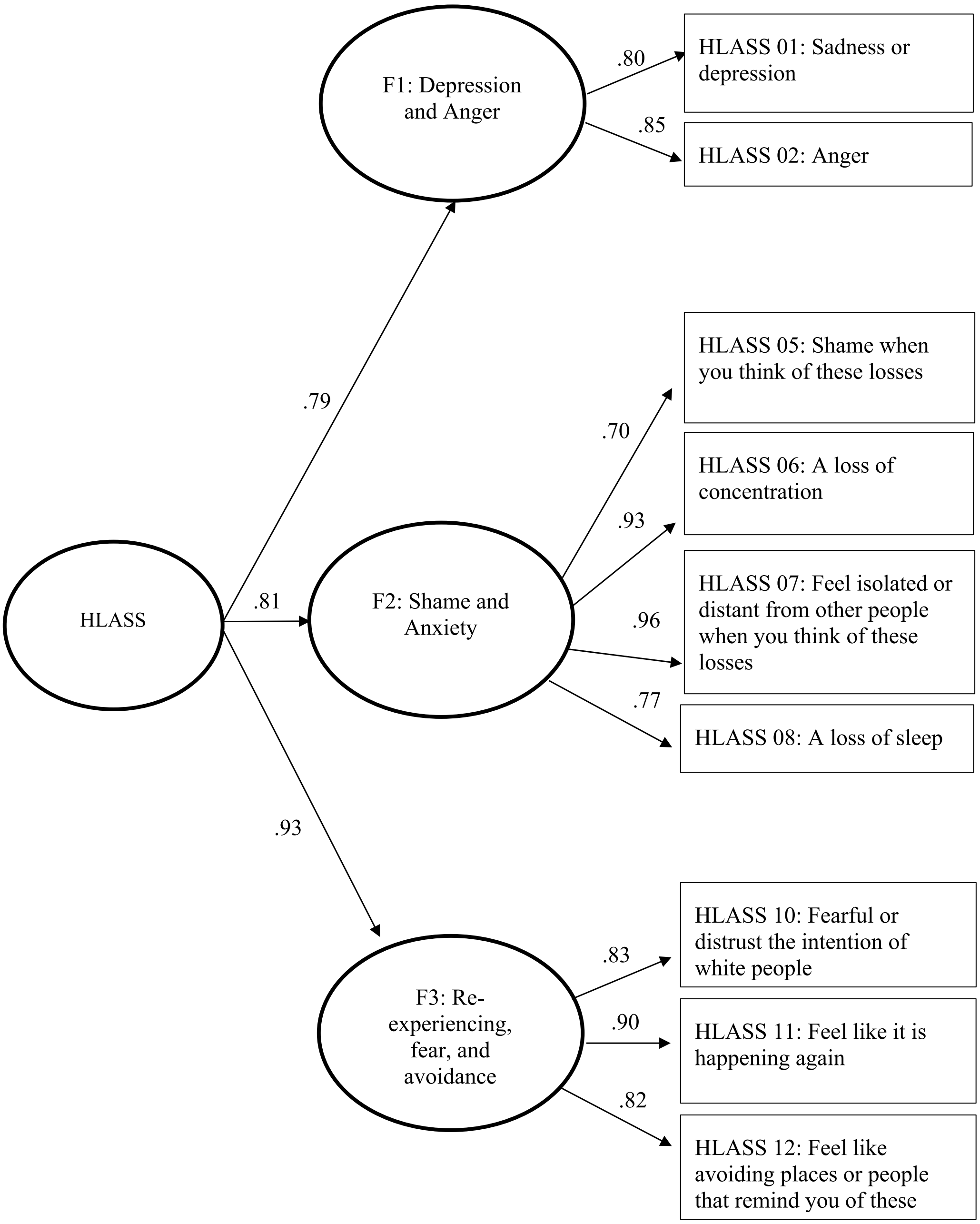

FIGURE 2
Psychometric model of HLASS as a hierarchical 3-factor model with standardized factor loadings.

symptoms. For instance, future studies could consider if thoughts related to the loss of culture, intergenerational loss, destruction of traditional foods, and distrust illicit the same reactions as measured by the HLASS. Additionally, future research should examine the strength of association between thoughts of certain losses and one HLASS factor compared to the others. This may also have implications for future research and practice that explores historical loss; symptoms associated with thoughts that influence mental, physical, and spiritual health in Native Hawaiian and Indigenous populations; and potential mediators and moderators of these relationships including impersonal or structural racism, cultural identity, cultural practice, or demographics such as age.

Most importantly, findings from this study may pave the way for efforts that aim to heal historical loss and associated symptoms among the Native Hawaiian community. An increased understanding of historical loss and its impact on health and wellbeing betters our understanding of resistance, resiliency, and the ability to overcome historical loss as a deep-seated determinant of health that stems from cultural and historical trauma and oppression. Initiatives that aim to heal the impact of listorical loss, including restoration of Native Hawaiian ways of knowing and cultural practices, including ʻĀina connectedness and land back, are critical in fostering mauli ola, optimal health and wellbeing, for the Native Hawaiian community and Indigenous Peoples at large.

TABLE 11 Reliability and inter-correlation matrix of the adapted historical loss scale (aHLS).

	Omega^a	Alpha^a	1	2	3	4	5	6	7	8	9	10	11	12	13
1. HLS item 1 (the taking of land)	–	–													
2. HLS item 2 (fewer and fewer people using our traditional language)	–	–	0.70												
3. HLS item 3 (destruction of our culture and traditional spiritual ways)	–	–	0.73	0.79											
4. HLS item 4 (loss of respect for elders by our children and grandchildren)	–	–	0.48	0.58	0.59										
5. HLS item 5 (loss of respect by our children for traditional ways)	–	–	0.50	0.58	0.65	0.83									
6. HLS item 6 (distrust, resentment, or fear toward white peoples)	–	–	0.55	0.56	0.58	0.47	0.52								
7. HLS item 7 (destruction or damage of traditional foods)	–	–	0.61	0.61	0.68	0.51	0.58	0.69							
8. EFA factor 01: cultural loss	0.90	0.89	0.89	0.91	0.93	0.61	0.64	0.62	0.70						
9. EFA factor 02: intergenerational loss	0.64	0.64	0.60	0.66	0.68	0.86	0.80	0.85	0.71	0.71					
10. EFA factor 01: cultural loss mean score	0.90	0.89	0.89	0.91	0.93	0.61	0.64	0.62	0.70	1.00	0.71				
11. EFA factor 02: intergenerational loss mean score	0.91	0.91	0.39	0.61	0.65	0.96	0.96	0.52	0.57	0.65	0.87	0.65			
12. EFA factor 03: distrust and destruction of traditional foods mean score	0.82	0.82	0.64	0.64	0.68	0.53	0.59	0.92	0.92	0.71	0.85	0.71	0.59		
13. HLS hierarchical mean score	0.92	0.91	0.80	0.84	0.88	0.78	0.82	0.78	0.83	0.89	0.93	0.89	0.88	0.88	

All inter-factor and inter-scale correlations were statistically significant at the $p < 0.01$ level.

[a] McDonald's omega and standardized Cronbach's alpha were based on the seven items of the HLS. Correlations were based on the mean of the three factors and the hierarchical factor of the HLS three-factor EFA model (items 1–7).

TABLE 12 Reliability and inter-correlation matrix of the historical loss associated symptoms scale (HLASS).

	Omega^a	Alpha^a	1	2	3	4	5	6	7	8	9	10	11	12
1. HLASS item 1 (sadness or depression)	–	–												
2. HLASS item 2 (anger)	–	–	0.61											
3. HLASS item 5 (shame when you think of these losses)	–	–	0.30	0.34										
4. HLASS item 6 (A loss of concentration)	–	–	0.36	0.35	0.52									
5. HLASS item 7 (feel isolated or distant from other people when you think of these losses)	–	–	0.34	0.39	0.53	0.76								
6. HLASS item 8 (A loss of sleep)	–	–	0.34	0.28	0.37	0.62	0.59							
7. HLASS item 10 (fearful or distrust the intention of white people)	–	–	0.35	0.45	0.29	0.43	0.47	0.30						
8. HLASS item 11 (feel like it is happening again)	–	–	0.45	0.50	0.38	0.43	0.47	0.25	0.75					
9. HLASS item 12 (feel like avoiding places or people that remind you of these losses)	–	–	0.34	0.38	0.36	0.41	0.50	0.38	0.57	0.58				
10. Factor 01: depression and anger mean score	0.75	0.75	0.90	0.90	0.35	0.39	0.41	0.35	0.45	0.53	0.40			
11. Factor 02: shame and anxiety mean score	0.84	0.83	0.41	0.42	0.76	0.88	0.88	0.77	0.46	0.48	0.51	0.46		
12. Factor 03: re-experiencing, fear, and avoidance mean score	0.85	0.84	0.44	0.52	0.40	0.49	0.56	0.35	0.89	0.90	0.81	0.53	0.55	
13. HLASS Hierarchical mean score	0.76	76	0.73	0.77	0.59	0.69	0.72	0.57	0.74	0.79	0.70	0.83	0.78	0.86

All inter-factor and inter-scale correlations were statistically significant at the $p < 0.01$ level.

[a] McDonald's omega and standardized Cronbach's alpha were based on the selected 10 items from the HLASS. Correlations were based on the mean of the three factors and the hierarchical factor of the HLASS.

TABLE 13 Reliability and inter-correlation matrix of the adapted historical loss scale (aHLS) and the historical loss associated symptoms scale (HLASS).

	1	2	3	4	5	6	7	8
1. HLS F1: cultural loss mean score								
2. HLS F2: intergenerational loss mean score	0.65							
3. HLS F3: distrust and destruction of traditional foods mean score	0.71	0.59						
4. HLS hierarchical mean score	0.89	0.86	0.88					
5. HLASS F1: depression and anger mean score	0.46	0.41	0.42	0.50				
6. HLASS F2: shame and anxiety mean score	0.35	0.30	0.40	0.40	0.46			
7. HLASS F3: re-experiencing, fear, and avoidance mean score	0.50	0.38	0.55	0.54	0.53	0.55		
8. HLASS hierarchical mean score	0.54	0.45	0.56	0.59	0.83	0.78	0.86	

All inter-factor and inter-scale correlations were statistically significant at the $p < 0.01$ level.

Data availability statement

The datasets presented in this article are not readily available because the data included in this project belongs to community partners. To increase data sovereignty, datasets are not provided publicly. Requests to access the datasets should be directed to MA, antoniom@hawaii.edu.

Ethics statement

The studies involving humans were approved by University of Hawaiʻi at Mānoa Institutional Review Board. The studies were conducted in accordance with the local legislation and institutional requirements. The ethics committee/institutional review board waived the requirement of written informed consent for participation from the participants or the participants' legal guardians/next of kin because of increased anonymity. Instead, survey packets were mailed to each household from select Hawaiian Homestead lands. Those who consented to participate returned the completed survey in a pre-addressed envelope. Completing the survey warranted consent.

Author contributions

MA: Conceptualization, Data Curation, Formal Analysis, Funding Acquisition, Methodology, Software, Validation, Visualization, Writing – original draft, Writing – review & editing. SK: Conceptualization, Formal Analysis, Methodology, Software, Validation, Writing – original draft, Writing – review & editing. CI: Conceptualization, Funding Acquisition, Methodology, Validation, Writing – original draft, Writing – review & editing. MW: Writing – original draft, Writing – review & editing. AD: Conceptualization, Funding Acquisition, Methodology, Project administration, Validation, Writing – original draft, Writing – review & editing. BK: Conceptualization, Funding Acquisition, Methodology, Project administration, Validation, Writing – original draft, Writing – review & editing. MKe: Conceptualization, Funding Acquisition, Methodology, Project administration, Validation, Writing – original draft, Writing – review & editing. SM: Writing – original draft, Writing – review & editing. KC: Conceptualization, Funding Acquisition, Methodology, Validation, Writing – original draft, Writing – review & editing. SA: Methodology, Validation, Writing – original draft, Writing – review & editing. MKa: Methodology, Validation, Writing – original draft, Writing – review & editing. JK: Conceptualization, Methodology, Funding Acquisition, Supervision, Validation, Writing – original draft, Writing – review & editing.

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Conflict of interest

SA and MKa were employed by Kapolei Community Development Corporation.

The remaining authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

References

1. Kaholokula JK, Ing CT, Look MA, Delafield R, Ki S. Culturally responsive approaches to health promotion for native Hawaiians and Pacific islanders. *Ann Hum Biol*. (2018) 45:249–63. doi: 10.1080/03014460.2018.1465593

2. Blaisdell K. Historical and cultural aspects of native Hawaiian health. *Soc Process Hawaii*. (1979) 1989:1–21.

3. Keli'iholokai L, Keaulana S, MCK A, Rogerson I, Deitschman K, Kamai JA, et al. Reclaiming ʻĀina health in Waimānalo. *Int J Environ Res Public Health*. (2020) 17:5066. doi: 10.3390/ijerph17145066

4. Paglinawan LK, Paglinawan RL, Kauahi D, Kanuha VK. Nānā i Ke Kumu = Look to the source. Helu ʻekolu = volume three. Honolulu: Liliʻuokalani Trust (2020).

5. Antonio MCK, Keaulana S, Keli'iholokai L, Felipe K, Vegas JK, Pono Research Hui W, et al. A report on the Ke Ola O Ka 'Āina: 'Āina connectedness scale. *Int J Environ Res Public Health*. (2023) 20:3302. doi: 10.3390/ijerph20043302

6. Kame'eleihiwa L. Native land and foreign desires = ko Hawaiʻi ʻāina a me nā koi puʻumake a ka poʻe haole: a history of land tenure change in Hawaiʻi from traditional times until the 1848 Māhele, including an analysis of Hawaiian aliʻi Nui and American Calvinists. Honolulu: Bishop Museum Press (1992).

7. Kame'eleihiwa L. Native land and foreign desires: Pehea lā e pono ai? How shall we live in harmony? Honolulu: Bishop Museum Press (2012).

8. Tengan TPK. Of colonization and Pono in Hawai'i. *Peace Rev*. (2004) 16:157–67. doi: 10.1080/1040265042000237699

9. Andrade T. E Ola Ka 'Ōlelo HAWAI'I: protecting the Hawaiian language and providing equality for Kānaka Maoli. *Indigenous Peoples' J Law Cult Resist*. (2020) 6:3–52. doi: 10.5070/P661051236

10. Andrade T. Hawaiʻi '78: collective memory and the untold legal history of reparative action for Kānaka Maoli. *Univ Penn J Law Soc Change*. (2021) 24:85–148.

11. Niheu K, Turbin LM, Yamada S. The impact of the military presence in Hawai'i on the health of Na Kānaka Maoli. *Pac Health Dialog*. (2007) 14:205–12.

12. Kaholokula JK. Mauli Ola: pathways to optimal Kanaka ʻŌiwi health. Honolulu, HI: University of Hawaii Press (2017).

13. Daniels S-AP, Kauahikaua L, Kaio C, Casson-Fisher JN, Ku T. Conceptualizing a new system of care in Hawai'i for native Hawaiians and substance use. *Hawai'i J Health Soc Welfare*. (2022) 81:43–51.

14. Wu Y, Braun K, Onaka AT, Horiuchi BY, Tottori CJ, Wilkens L. Life expectancies in Hawaiʻi: a multi-ethnic analysis of 2010 life tables. *Hawai'i J Med Public Health*. (2017) 76:9–14.

15. Look MA, Trask-Batti MK, Agres R, Mau ML, Kaholokula JK. Assessment and priorities for health & well-being in native Hawaiians & other Pacific peoples. Honolulu, Hawaiʻi: University of Hawaiʻi, JABSOM Department of Native Hawaiian Health (2013).

16. Look MA, Soong S, Kaholokula JK. Assessment and priorities for the health and well-being in native Hawaiians and Pacific islanders. Honolulu, Hawaiʻi: University of Hawaiʻi, John A. Burns School of Medicine, Department of Native Hawaiian Health (2020).

17. Wu Y, Uchima O, Browne C, Braun K. Healthy life expectancy in 2010 for native Hawaiian, white, Filipino, Japanese, and Chinese Americans living in Hawai'i. *Asia Pac J Public Health*. (2019) 31:659–70. doi: 10.1177/1010539519875614

18. Hawaii Health Data Warehouse (2022). Native Hawaiian race/ethnicity (DOH) community report. Available at: https://hhdw.org/report/community/indicators/ChronicDisease/RacEthDOH/2.html (Accessed December 1, 2023).

19. Brave Heart MYH, Chase J, Elkins J, Altschul DB. Historical trauma among indigenous peoples of the Americas: concepts, research, and clinical considerations. *J Psychoactive Drugs*. (2011) 43:282–90. doi: 10.1080/02791072.2011.628913

20. Armenta BE, Whitbeck LB, Habecker PN. The historical loss scale: longitudinal measurement equivalence and prospective links to anxiety among north American indigenous adolescents. *Cultur Divers Ethnic Minor Psychol*. (2016) 22:1–10. doi: 10.1037/cdp0000049

21. Walters KL, Beltran R, Huh D, Evans-Campbell T. Dis-placement and dis-ease: land, place, and health among American Indians and Alaska natives In: Social disparities in health and health care. New York, NY: Springer (2010). 163–99.

22. Whitbeck LB, Adams GW, Hoyt DR, Chen X. Conceptualizing and measuring historical trauma among American Indian people. *Am J Community Psychol*. (2004) 33:119–30. doi: 10.1023/B:AJCP.0000027000.77357.31

23. Sotero M. A conceptual model of historical trauma: implications for public health practice and research. *J Health Disparities Res Pract*. (2009) 1:93–108.

24. Osorio JK. Dismembering lāhui: A history of the Hawaiian nation to 1887. Honolulu: University of Hawaiʻi Press (2002).

25. Trask H-K. From a native daughter: colonialism and sovereignty in Hawaiʿi: Pbk. *Rev* ed. Honolulu, HI: University of Hawaiʿi Press (1999). Available at: https://ssrn.com/abstract=1350062

26. Gone JP, Hartmann WE, Pomerville A, Wendt DC, Klem SH, Burrage RL. The impact of historical trauma on health outcomes for indigenous populations in the USA and Canada: a systematic review. *Am Psychol*. (2019) 74:20–35. doi: 10.1037/amp0000338

27. Whitbeck LB, Walls ML, Johnson KD, Morrisseau AD, McDougall CM. Depressed affect and historical loss among north American indigenous adolescents. *Am Indian Alsk Native Ment Health Res*. (2009) 16:16–41. doi: 10.5820/aian.1603.2009.16

28. Wiechelt SA, Gryczynski J, Johnson JL, Caldwell D. Historical trauma among urban American Indians: impact on substance abuse and family cohesion. *J Loss Trauma*. (2012) 17:319–36. doi: 10.1080/15325024.2011.616837

29. Anastario MP, FourStar K, Rink E. Sexual risk behavior and symptoms of historical loss in American Indian men. *J Community Health*. (2013) 38:894–9. doi: 10.1007/s10900-013-9695-8

30. Pokhrel P, Herzog TA. Historical trauma and substance use among native Hawaiian college students. *Am J Health Behav*. (2014) 38:420–9. doi: 10.5993/AJHB.38.3.11

31. Minkler M, Wallerstein N. Community based participatory research for health. Hoboken, NJ, US: Jossey-Bass/Wiley (2003).

32. Willis GB, Artino AR Jr. What do our respondents think we're asking? Using cognitive interviewing to improve medical education surveys. *J Grad Med Educ*. (2013) 5:353–6. doi: 10.4300/JGME-D-13-00154.1

33. Jackson DL, Gillaspy JA Jr, Purc-Stephenson R. Reporting practices in confirmatory factor analysis: an overview and some recommendations. *Psychol Methods*. (2009) 14:6–23. doi: 10.1037/a0014694

34. Hooper D, Coughlan J, Mullen MR. Structural equation modelling: guidelines for determining model fit. *Electron J Bus Res Methods*. (2008) 6:53–60.

35. Hu L, Bentler P. Cutoff criteria for fit indexes in covariance structure analysis: conventional criteria versus new alternatives. *Struct Equ Model*. (1999) 6:1–55. doi: 10.1080/10705519909540118

36. Johnson B, Vogt WP. The SAGE dictionary of statistics & methodology: a nontechnical guide for the social sciences. *5th* ed. Los Angeles: SAGE (2016).

37. Evans-Campbell T. Historical trauma in American Indian/native Alaska communities: a multilevel framework for exploring impacts on individuals, families, and communities. *J Interpers Violence*. (2008) 23:316–38. doi: 10.1177/0886260507312290

38. Remes O, Mendes JF, Templeton P. Biological, psychological, and social determinants of depression: a review of recent literature. *Brain Sci*. (2021) 11:1633. doi: 10.3390/brainsci11121633

39. Hohls JK, König H-H, Quirke E, Hajek A. Anxiety, depression and quality of life—a systematic review of evidence from longitudinal observational studies. *Int J Environ Res Public Health*. (2021) 18:12022. doi: 10.3390/ijerph182212022

40. Staicu M-L, Cuţov M. Anger and health risk behaviors. *J Med Life*. (2010) 3:372–5.

41. Antonio MCK, Keaulana S, Keli'iholokai L, Ho-Lastimosa HI, Chung-Do JJ. Indigenous research methodologies with Kānaka 'Ōiwi to address health inequities: two case studies. *Hawai'i J Health Social Welfare*. (2023) 82:5–9.

42. McKinley CE, Figley CR, Woodward SM, Liddell JL, Billiot S, Comby N, et al. Community-engaged and culturally relevant research to develop behavioral health interventions with American Indians and Alaska natives. *Am Indian Alsk Native Ment Health Res*. (2019) 26:79–103. doi: 10.5820/aian.2603.2019.79

43. Kaholokula JK, Samoa RA, Miyamoto RES, Palafox N, Daniels S-A. COVID-19 special column: COVID-19 hits native Hawaiian and Pacific islander communities the hardest. *Hawai'i J Health Soc Welfare*. (2020) 79:144–6.

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Discrimination among American Indian and Alaska Native people: implications for public health communication

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Introduction: American Indian and Alaska Native People (AI/AN) have experienced discrimination stemming from sustained attempts to erase AI/AN People and their culture or livelihood. Research identifying the types of discrimination experienced by AI/AN People is needed to help individuals recognize discrimination in daily life. We examine experiences of discrimination among an urban AI/AN population using a mixed methods approach.

Methods: Self-identified AI/AN participants ($N = 303$, $n = 294$ with complete data; 63% women, mean age = 43 years) were recruited from the Denver-metro area in Colorado. Stress and coping models of discrimination guided our analysis. Exposure to discrimination was quantitatively assessed via the Brief Perceived Ethnic Questionnaire – Community Version (BPEDQ-CV), a self-report measure including four subscales assessing workplace discrimination, social exclusion, physical threat and harassment, and stigmatization. Participants responded to a laboratory recall task in which they described an episode of discrimination and their affective and coping responses. Content analysis was conducted on transcribed responses to illustrate discrimination exposure as reported in the BPEDQ-CV and in prior theoretical work on coping with discrimination.

Results: Repeated measures analyses revealed participants reported experiencing social exclusion more than other forms of discrimination, followed by reports of workplace discrimination, stigmatization, and physical threat. Consistent with these quantitative findings, participants were more likely to recall experiences of social threat (94%), including episodes of workplace discrimination, social exclusion, and stigmatization than physical threat and harassment. Almost half the participants (47%) reported confronting or directly addressing the discrimination, and 38% reported avoiding a direct approach. For 44% of participants, their predominant emotional response included internalizing emotions such as fear/sadness/embarrassment, and another 44% reported experiencing externalizing emotions, including anger.

Conclusion: Our descriptive findings present the experiences of urban AI/AN People who have experienced many forms of unjust and prejudicial treatment. These data can provide useful information to help the general public and AI/AN individuals more readily recognize and prevent discriminatory behavior, and consequently mitigate deleterious effects of discrimination on health.

KEYWORDS

Native American, American Indian and Alaska Native, Indigenous People, discrimination, microaggression, prejudice, resilience, mixed-method approach

1 Introduction

American Indian or Alaska Native (AI/AN) People have been persistently exposed to high levels of structural discrimination, including cultural and institutional threats (1). AI/AN People have faced systematic attempts to eradicate their identity and culture, including forced removal from their homes and placement in boarding schools whose purpose was to remove attachment to their culture (2, 3). At the individual level, AI/AN People have been exposed to a wide variety of interpersonal discrimination, defined as interpersonal maltreatment motivated by racial or ethnic bias (4–6).

Data on the prevalence of discrimination exposure among AI/AN People living on tribal lands or in urban communities is still limited. The findings suggest rates of discrimination against AI/AN populations are among the highest across ethnic/racial groups in the United States (U.S.) (2, 11), with more than half of AI/AN People reporting exposure to discrimination (2, 7–10). For example, findings on the prevalence of interpersonal discrimination targeted toward reservation-based AI/AN children and adolescents reveal that almost half of the participants reported facing discrimination at some point in their lives (8). Adult AI/AN participants were more likely than African American or White participants to report discrimination by healthcare professionals (11). Other studies report that a large proportion of participants experienced discrimination from employers (31%), police (32%), and healthcare institutions (23%) (12).

Racial and ethnic discrimination has been recognized as a public health threat by the American Medical Association (13). To address this threat among AI/AN People, there is a need for broader public awareness of the specific types of prejudice and discrimination AI/AN individuals face. This knowledge may guide the development of public health interventions to decrease discrimination by promoting increased awareness of common types of prejudicial communications and discriminatory behavior. Information presented in an accessible and relatable manner could heighten non-AI/AN individuals' awareness of words and actions that communicate prejudice, potentially reducing discriminatory behavior.

Knowledge about discriminatory acts may also protect AI/AN People from harms associated with discrimination. This information may help targeted Indigenous individuals more quickly recognize prejudicial and potentially discriminatory behavior, potentially reducing the stress evoked as individuals try to evaluate subtle forms of discrimination (14). Therefore, the present study aims to link findings from theoretical and empirical studies of discrimination with descriptions of lived experiences of discrimination and associated coping strategies reported by AI/AN People.

Key dimensions of interpersonal discrimination include a broad variety of maltreatment including social exclusion, unfair treatment at school or work, social and physical harassment, and stigmatization (15, 16). These discriminatory threats emerge from stereotypes about AI/AN People, including beliefs about their associations to substance abuse or criminal acts (17). Other discriminatory behavior reflects the traumatic history of AI/AN Peoples in the U.S. For example, AI/AN culture can be rendered invisible when members of other groups believe the AI/AN People only existed in the past and that none survived to the present day. When individuals are unaware of AI/AN communities, they may deny that an individual is Indigenous because they fail to recognize AI/AN People as a group whose members have unique languages, ways of knowing, educational pursuits, families, jobs, and certain cultural practices (e.g., wearing ceremonial apparel) (17, 18).

A limited body of quantitative research has examined the types of discrimination that AI/AN individuals face. Blair et al. (2) examined exposure to different types of interpersonal discrimination among a convenience sample of urban dwelling AI/AN People using the Brief Perceived Ethnic Discrimination -Community Version (Brief PEDQ-CV). The Brief-PEDQ-CV emerged from studies of ethnicity-related stress by Contrada et al. (16, 19) and Brondolo et al. (15). The Brief PEDQ-CV permits examination of different types of discrimination including racebased episodes of social threats (i.e., social exclusion, stigmatization and work or school-based discrimination) as well as physical threats. Findings on the types of discrimination reported by AI/AN People were compared to data from convenience samples of urban African American and Asian individuals recruited from communities in New York City (2). For AI/AN People, as is the case with other groups, social exclusion was the most prevalent form of discrimination, followed by workplace discrimination and stigmatization. Physical threat was the least common exposure.

These data are consistent with other studies examining variations in exposure to different types of discrimination. Among adolescent participants, studies have suggested that verbal harassment is the most common type of discriminatory behavior expressed by outgroups (7). Verbal harassment can include slurs, insults, verbal threats, criticism, and harmful exclusion. A recent report by D'Amico et al. (7) documented similar trends, reporting that a sizable percentage of participants reported being asked to prove the authenticity of their AI/AN status, consistent with the notion that invisibility is a salient form of discriminatory behavior.

Although participants report that race-based physical maltreatment is the least common type of maltreatment, physical threats remain a serious issue. A recent study reported that a quarter of adult participants in their sample indicated that they or their family members were subject to violence and threats (12). Studies including adolescents indicated that 14% reported exposure to physical threat (7).

Qualitative data are needed to facilitate the recognition of and understanding of these different types of discrimination. Qualitative data can provide insight into the lived experiences of discrimination in everyday life as they are drawn from personalistic accounts from individuals within the groups being studied (17, 20–22). As Robertson (17) noted, qualitative methodology allows researchers to, "[give] value and voice to their lived experiences in a historically contextualized way."

Existing qualitative studies have examined perceptions of the link between discrimination and specific issues of importance to the AI/

AN community, including substance use (21, 23), health trajectories (6, 24), academic achievement (25), and resiliency in the face of discrimination (26–29). For instance, some participants in the Skewes and Blume (21) paper identified discrimination as an underlying cause or perpetuator of their substance use. One participant even stated that, "Oppression is the overarching umbrella for all sickness with drugs and alcohol," (21). Solomon et al. (24) and Brondolo et al. (45) documented barriers associated with the Coronavirus Disease 2019 (COVID-19) pandemic that created health disparities in treatment and administration of vaccines. Other studies have provided insights into the ways in which a focus on tradition and interconnectedness among American Indian communities can build resilience against discrimination (28, 29).

However, there are gaps in knowledge. To date, existing studies of discrimination among AI/AN People have not explicitly tied descriptions of the lived experiences of discrimination to models of interpersonal discrimination derived from quantitative analyses, such as those of Contrada et al. (16, 19) or Krieger (4). These models describe distinctions among different types of discriminatory experiences. Analyses linking model-based quantitative data to qualitative findings could provide an accessible roadmap to understanding how to conceptualize discriminatory experiences and recognize them in different contexts. Combining quantitative and qualitative data can support Skewes and Blume's (21) call for educational interventions to reduce the discrimination AI/AN People have experienced and provide guidance for the development of public health interventions to help mitigate resulting health issues.

In the present study, we combined both quantitative and qualitative approaches to provide insight into discrimination exposure among urban AI/AN People. In contrast to qualitative studies of AI/AN People that incorporated inductive, grounded theory-based approaches that extrapolate themes (17, 18, 20, 21, 26), we employed another qualitative approach, content analysis, to provide illustrations of lived experiences of race-related discriminatory experiences, including social threat, stigmatization, work or school-based unfair treatment, and physical harassment. Content analysis was used to depict illustrations of specific aspects of extant theoretical frameworks of discrimination. Excerpts were analyzed to determine which types of discrimination and coping were identified.

In addition to illustrating experiences of discrimination, we expand the existing qualitative literature to include theoretically derived assessments of coping. Using frameworks developed by Krieger (27) and Krieger (4), coping responses have been categorized as either avoiding addressing the maltreatment or directly confronting the maltreatment. Approaches which focus on suppression or expression are also consistent with other research on the use of anger-coping strategies (i.e., anger-in vs. anger-out) in studies of coping with racism by Black and Hispanic Americans (46). We assess resilience as some researchers have reported that exposure to trauma (such as discrimination) can build resilience through the development of adaptive coping strategies, including seeking out social support (30).

2 Methods

2.1 Study population

The total sample population in this study consisted of 303 urban AI/AN People living in the Denver-metro area in Colorado. Nine participants chose to exit the study due to concerns raised during the screening tests (e.g., severely elevated blood pressure) or they did not attend study testing sessions, leaving 294 participants as the study sample.

In the U.S., AI/AN People make up roughly 2% of the population. They often identify either with one of the 575 federally- recognized or state-recognized tribes or with those that are unrecognized. Federally recognized tribes span 35 states with a large concentration located in the southwest region.

Roughly 46,000 AI/AN People live in the Denver-metro area alone (47). Denver has a growing population of urban AI/AN People because it sits between the Southwest and Midwest plains regions where many Tribal lands are located (47). Estimates suggest that 78% of AI/AN People live in urban areas; however, this number may fluctuate as many people are tied to their community, land, and relatives/family that are located on Tribal lands, but travel into urban areas for education, healthcare, or work (48). Over half (53%) of the participants in this study were affiliated with tribes from the Great Plains region of the U.S. while second largest group were affiliated with a tribe located in the Southwest (11%). Several participants noted more than one affiliation (20%). Forty participants did not respond with their tribal affiliation.

To participate in this study, we required participants to self-identify as an AI/AN individual, aged 18 or older, and residing in the Denver metro area. Participants were excluded if they had a pacemaker, were pregnant, were on dialysis, unable to have blood pressure measured, frequently used illicit drugs, were unable to provide consent, or who had severely elevated blood pressure. The study sample was comprised of 63.3% women ($n = 186$), and 36.7% men ($n = 108$), with a mean age of 43.4 (SD = 14.7, Range: 18–78 years). The majority ($n = 171$; 62.1%) were living at or below the poverty level, and 52 (11%) were living at three times or more than the poverty level; 47.6% ($n = 140$) had completed some college or more education and 52.4% ($n = 154$) had a high school diploma or less; and 38.2% ($n = 112$) were married.

2.2 Recruitment

Participants were recruited via word of mouth and via flyers that were sent electronically and posted physically inside the local Tribal community centers, health clinic, and public bulletin boards and at annual Tribal events. Participants could reach out to the study team by phone call or email to discuss their eligibility and to schedule a visit to the University clinic. Once contact was made, the participant was mailed a consent form detailing the benefits and limitations of the study before their first visit. Before their first visit, the participant was once again given details of the study and consent was obtained following a discussion to ensure voluntary and informed consent.

2.3 Data collection and mixed methods analyses

Urban AI/AN individuals participated in a field and laboratory study on stress and health that was funded by the American Heart Association. Informed consent was conducted in-person and privately at the research office. The research assistant reviewed all study procedures with each participant, using printed visual aids to depict the flow of the study session. Consent was provided in writing and participants were given a copy of the document. As a part of the first laboratory session, participants completed a quantitative measure of perceived ethnic discrimination and orally responded to a prompt to think about experiences of discrimination to describe, how they felt, and to detail the ways they coped with these experiences. Further details of the study have been previously published (31), and are publicly available at: http://tinyurl.com/5wbsc9ad. To gather information about the participants' experiences of discrimination, the interviewers provided the participants with a tape recorder in their room. The tape recorder was then turned on by the participant when a pre-recording prompt said, "Talk about a past experience in which you knew you were treated poorly or unfairly because you are American Indian, even if the other person did not mention your AI/AN cultural status. We will record what you say. We are not able to talk to you about this experience until your return visit the next day." Participants were also provided with the following six pilot tested written prompts to help them detail the act of discrimination that they planned to discuss: "What happened?," "Who were you with?," "Where were you?," "What did the other person do or say?," "How did you feel?," and "What did you do or say?." To answer these prompts, the participants were given 2 minutes to prepare, and then given the opportunity to discuss their discriminatory experience with no time limit. The response time per participant was 2.2 minutes on average. Participant responses to these prompts were recorded and transcribed and served as the data for the qualitative portion of the study.

The laboratory protocol in which participants provided descriptions of these experiences were administered and collected by research assistants in a clinic at the University of Colorado Boulder from 2016 to 2019.

2.4 Statement of positionality

The principal investigators of the study are an Asian American gender-queer woman (IB) and a White woman (EB). The Indigenous woman co-lead author (RB) served as a community advisory board member and is also a public health researcher who saw the value of the project. The research team asked her to lead this project and she was willing to help curate the message behind this paper. Her lived experiences as an Indigenous (Diné/Navajo), living in rural/urban settings provided a necessary and unique lens. The other co-lead author (MR), a White man, is a student who has spent several years working in the Collaborative Health Integration Research Program (CHIRP, directed by EB), a research training program focused on health disparities. He contributed to the analyses and organization of the data for the project which provided the structure needed to conclude the data. EB is a clinical psychologist who has devoted her research career to studying minority health disparities (45, 46). She met with the research assistants and coders each week to help them process the emotional impact of the stories that were conveyed. These discussions provided opportunities for her to learn about the types of discrimination the research participants reported and to understand the effect hearing these stories had on others. IB is a social psychologist who has dedicated her research career to understanding and addressing prejudice, racism, and health disparities. IB acknowledges her positionality as a descendant of people who came to this country as colonial/settlers and immigrants. The research assistants involved in the qualitative coding process included one White man and five men and women from underrepresented groups (e.g., Black, Latino/a, and Asian American). The research assistants received intensive training and supervision in explaining the study, administering the laboratory protocols, and training on the questions about discrimination. Weekly review sessions were held to support interviewers and ensure adherence to the testing protocol.

2.5 Measures

Quantitative assessment of exposure to racial discrimination was obtained with the Brief Perceived Ethnic Questionnaire – Community Version [BPEDQ-CV (15)]. The BPEDQ-CV is a short 17-item assessment of lifetime exposure to discrimination derived from the 34-item Perceived Ethnic Discrimination Questionnaire – Community Version [PEDQ-CV; (15)]. The BPEDQ-CV includes four scales that assess different dimensions of discrimination, including: exclusion/rejection, stigmatization/devaluation, discrimination at work/school, and physical threat/aggression. Items are scored on a Likert scale from 1 ("Never happened") to 5 ("Happened very often"). Some example items included: "Have you been treated unfairly by teachers, principals, or other staff at school," "Have others thought you could not do things or handle a job," and "Have others threatened to hurt you (ex: said they would hit you)?" A mean score was calculated such that higher scores indicated greater exposure to discrimination (time 1 $\alpha = 0.90$). Findings from our prior studies comparing new data from AI/AN individuals to data drawn from samples of urban African American and Asian individuals tested in similar circumstances in New York City, New York (2) which indicated the Brief PEDQ-CV demonstrated measurement invariance across all three urban groups. This suggests the Brief PEDQ-CV is a suitable tool for use with the AI/AN sample population. The Brief PEDQ-CV was administered on the first visit. A research assistant read the questions aloud to the participants and recorded their responses in RedCap.

2.6 Content analysis

Interviews were transcribed verbatim and were analyzed by six coders from St. John's University (three groups of two). The six coders included three men and three women who were all undergraduate students. The coders were trained by the research assistants at the University of Colorado Boulder, who developed the protocols, and by the principal investigators. Dedoose software was used to organize, manage the data, and apply the codes to the transcripts. Before beginning coding, Dedoose software calculated the Kappa statistic (inter-rater agreement) achieved when coding a pre-selected set of interview excerpts. Coding began once raters achieved Kappa levels above 0.60. Actual Kappa levels ranged from 0.68 to 0.87. Obtaining these kappa ranges involved an iterative process of refining codes and re-testing until sufficient agreement was reached among the coders.

Codes were generated *a priori* to reflect the types of discrimination and responses to discrimination based on prior work on racial and ethnic discrimination (15) and specific studies of discrimination facing AI/AN People (2, 31, 32). The codes and their explicit definitions were developed by researchers and research assistants (See Supplementary Tables S1–S7). A total of 41 codes listed under seven broader topics (See Supplementary Table S8) were created describing aspects of the participants' experiences of discrimination. In this paper we analyzed codes related to three topics (See Supplementary Tables S8, S9): (1) the act of discrimination (coding for different types of discrimination), (2) affective and behavioral responses to the act of discrimination (e.g., sad, frustrated, angry, confrontation, or avoidance etc.), (3) and evidence of resilience. Chi-Square analyses were performed to determine significant differences in frequency among related categories.

2.7 Ethics

This study was approved by St. John's University Institutional Review Board (IRB), University of Colorado Boulder IRB, and the Colorado Multiple Institutional Review Board (COMIRB). Feedback on the protocol, implementation, and interpretation of the results were provided by the community advisory board (CAB) composed of an all Indigenous (AI/AN) panel composed of six individuals living in the Denver-metro area. The CAB met with varying frequency over the course of several years, approximately 2–3 times a year. CAB members were initially recruited with the assistance of an AI/AN community liaison employed by the Centers for American Indian and Alaska Native Health (CAIANH). The goal was to include individuals who identify as AI/AN with a range of ages and gender, who are involved in the community and knowledgeable about AI/AN cultures and perspectives represented in the Denver-metro area. Board members were compensated for their time and expertise. All participants whose excerpts are cited in this paper provided written permission for their excerpts to be included and published.

3 Results

3.1 Quantitative analyses

Linear mixed models indicate that mean scores across the four scales significantly differed from each other (F(1,293) = 138.06, p <0 .001). Consistent with previous studies (7, 8), our findings suggest that social exclusion ($M = 1.89$; SE = 0.05) is the most prevalent type of discrimination, followed by workplace-based discrimination ($M = 1.66$; SE = 0.05), stigmatization ($M = 1.34$; SE = 0.05), and physical threats of violence ($M = 1.11$; SE = 0.06) (see Figure 1). Results of quantitative analyses of the qualitative data are presented in Figures 2–4.

3.2 Qualitative analyses

Responses were coded as acts of discrimination if the participant described specific acts of maltreatment related to their AI/AN status. We organized responses into the type of maltreatment including episodes of social exclusion, unfair workplace/school treatment, stigmatization (i.e., being called racial slurs or hearing references to negative stereotypes), and physical maltreatment. In addition to episodes of social exclusion that involved being ignored or left out, we included denial of the existence of AI/AN People or denial of an individual's Indigeneity as a specific form of social exclusion, as the individual was excluded both from the mainstream culture and their Indigenous culture to which they belonged. Figure 2 also details the frequency with which categories of "Act of Discrimination" codes were employed. To illustrate these categories, we included excerpts drawn from the participants' response to the laboratory discrimination-recall task which provide examples of specific types of discrimination

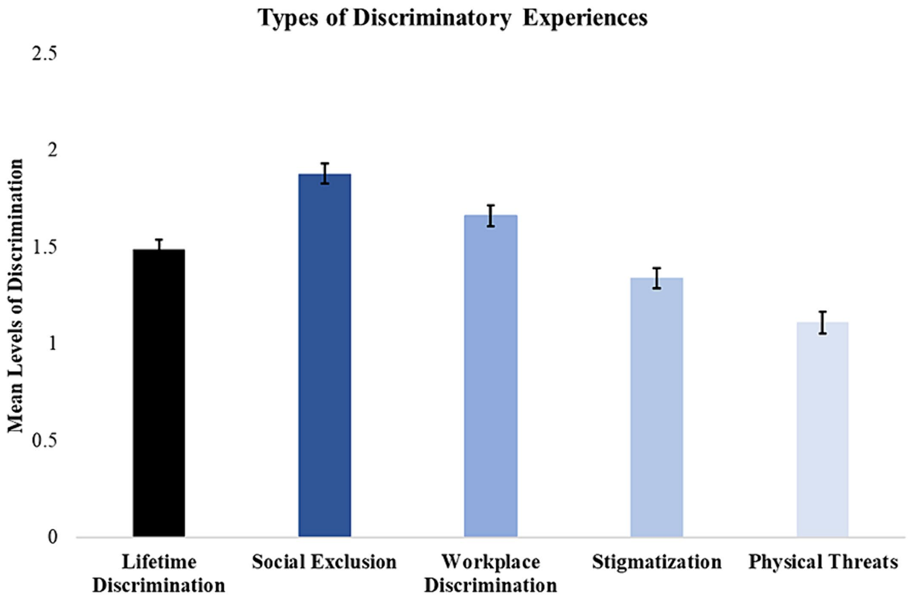

FIGURE 1
Mean rates of discrimination subtypes measured by the BPEDQ-CV faced by our sample ($n = 294$). Survey data suggest that social exclusion was the most common type of discriminatory experience ($M = 1.88$; SEM = 0.05), followed by workplace-based discrimination ($M = 1.66$; SEM = 0.05), stigmatization ($M = 1.34$; SEM = 0.05), and physical threats of violence ($M = 1.11$; SEM = 0.06).

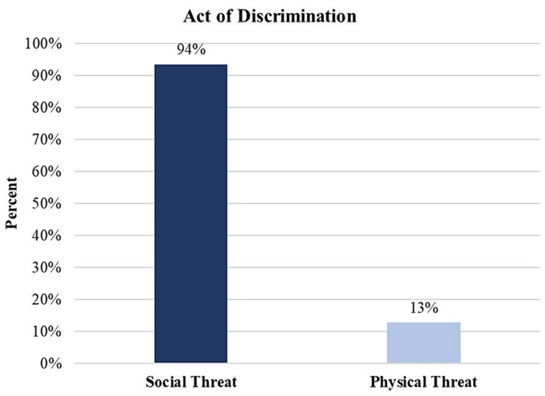

FIGURE 2
Act of discrimination experienced by AI/AN participants. Percentages represent the number of times the type of threat occurred across all participant excerpts divided by the total number of study participants ($n = 294$). As a consequence, they are not mutually exclusive codes. Some participants reported both social threats and physical threats, or not at all. Thus, the percentages presented in this figure exceed 100%. A chi-square goodness of fit test was performed to assess differences in the frequency of acts of discrimination, $X^2(1) = 179.45$, $p < 0.0001$. Results suggest that social threats are the most prevalent act of discrimination, more common than physical threat.

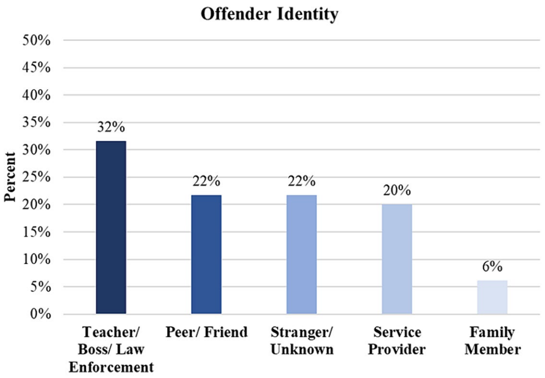

FIGURE 3
General identity of offender who committed a discriminatory act. Percentages represent the figures of authority of the offender identities divided by the total number of study participants ($n = 294$). As such, multiple types of offenders may have appeared in certain participants' excerpts. Thus, the percentages presented in this figure exceed 100%. A chi-square goodness of fit test revealed significant differences in observed frequencies among the offender subtypes, $X^2(4) = 48.41$, $p < 0.0001$. These findings suggest that authority figures, such as teachers, bosses, and law enforcement figures are more commonly perceived as perpetrators compared to other offender subtypes.

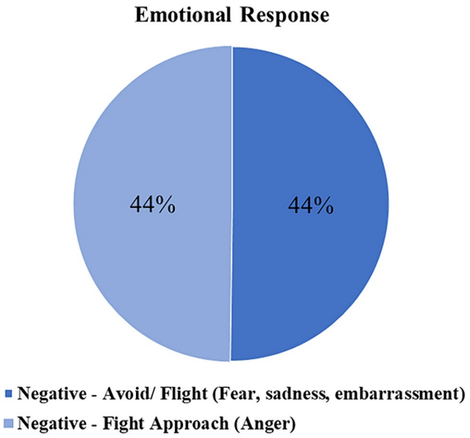

FIGURE 4
Emotional response by participants. Percentages represent the number of negative emotional flight approach (or fight approach) responses divided by the total number of study participants ($n = 294$). As a consequence, the types of emotional responses are not mutually exclusive, and some emotional responses may not have been recorded. Thus, the percentages presented in this figure do not sum up to 100%. A chi-square goodness of fit test was performed to assess differences in observed frequencies of emotional responses, revealing non-significant differences, $X^2(1) = 0.004$, $p > 0.05$. These findings indicate that anger and avoidance emotional responses are equally common responses to discriminatory behavior.

identified. It is important to note that some episodes reflect more than one type of discrimination. In such cases, these additional types are noted at the end of the excerpt. Detailed explanations of the characteristics associated with each code are provided in the Supplementary Tables S1–S7. The second topic included participant's "Responses to Discrimination." These responses were further organized into affective responses and then coping responses which could include episodes of avoidance and suppression, confrontation or directly addressing discrimination. The third topic was "Resilience" in the face of discrimination.

Lastly, we highlighted the groups who were the aggressors or offenders toward the AI/AN individuals. We found that individuals with authority (32%) (e.g., teachers, bosses, or law enforcement officials) were reported most frequently as committing an act of discrimination against the target individual, followed by peers and friends (22%), strangers (22%), service providers (20%) (e.g., store or restaurant employees, medical staff, etc.), and family members (6%) (see Figure 3).

3.3 A: Acts of discrimination

1: Descriptions of Social Exclusion

These lived experience descriptions of social exclusion include experiences of being rejected, ignored, and isolated (excerpt A.1.1) and ostracized (excerpt A.1.2). These episodes also include examples of being told AI/AN culture does not exist or that one is not AI/AN (excerpts A.1.3–A.1.7).

> Excerpt A.1.1 "As a child, I got information about my father from my grandma, and through the story she told me his ethnicity and everything. As a child, I did not really feel a part. I felt kind of isolated a lot, so it kind of gave, I guess, understanding to me why I felt so isolated and different, aside from my mom, brother, her husband. Like, they were like a family and it seemed like I was separate, and finding out that my dad was Native…So it gave me something to hold on to. Just going through the growing up stage, there was a lot of riding in a car with your head down between your legs so you do not get

> a chance to see -- you are not seen in the car as a part of the family riding in the car…Like, if she goes to visit -- or take my brother to visit his father, I would have to stay in the car. Like I wasn't accepted.” (Man, 30–40).
>
> A.1.2 “A few years ago, I went back home to visit some family members. Got a hotel room and invited my family to come over and swim, all the little relatives. And while we were swimming, a Caucasian woman came up to me and asked me how long we were planning on being in the pool. I asked her, well, why is that any of her business? She, then, told me that she did not want her kids in the same pool as us Native Americans. That we were swimming for more than two hours, and we had enough time to be in the pool, and we should leave because her kids needed to go swimming (Woman, 20–30). Note: This example could also be considered stigmatization if the perpetrator was implying that AI/AN children were unclean.
>
> A.1.3. “I was at…a nonprofit place and I had to fill out some paperwork…When I got to their forms where you fill out your ethnicity, race, all that good stuff, I actually wrote down Native and human, and the lady came back up to me and she said, “How are you Native American? You're too dark.”…it's not the first time that I mentioned being native and someone kind of just brushes it off like, no you are not, you do not fit the profile of what they ‘look like.’” (Woman, 20–30).
>
> A.1.4 “Well, I used to pass by a Native American cultural center on [Street Name], and one day I wanted to go in just to see what it was about… everybody was different from me. They had braids, long hair, and they looked at me, and they asked me, “What do you want? What are you doing here?” I said, “I want to learn about my people, my tribe.” He said, “But, you are not Native American.” (Man, 50–60).
>
> A.1.5. “When I was in high school for my senior year… there is Native American little classes and stuff where we can go and talk about us, what our ethnicity was and just share our stories…But this one boy and his sister… said that the brother would always get bullied because he had long hair and he would always come with a braid and everything in his hair and everybody thought he was a girl because he had long hair. That touched me because all my family has the long hair and everything. A lot of people just be like “Oh, he's a girl. He wants to have long hair and everything.” (Woman, 18–28).
>
> A.1.6 “When I was growing up, I was constantly teased and bullied over having long hair. They always made fun of me all the time. I was with my little brother, happened with him too, usually at school. The other people used to make homophobic comments and say things, implying that I was homosexual because I had long hair.” (Male, 30–40).
>
> A.1.7. “During the president's State of the Union address, he illustrated unemployment for the blacks, for the whites, for the Latinos, but never mentioned the first Americans. Many cases where the government has failed to address Indian issues, such as the rights of the pipelines up north. That was a heart-wrenching one for us natives. Any time we do something, it's always about not being recognized as an existing group of people…What about Native Americans? We're forgotten. Our rights do not count no more because we are first Americans.” (Man, 60–70).

2: Descriptions of Workplace or School Discrimination

These lived experience descriptions of workplace or school discrimination includes episodes in which individuals describe being assigned tasks (see excerpt A.2.1) or not assigned tasks (see excerpt A.2.2) as a function of biased assumptions about their capabilities. These biased assumptions may be driven by stereotypes about AI/AN People.

> A.2.1 “[The participant was at work and her manager assumed she was Latina and asked her to translate for a Spanish speaking customer] A coworker said “……. you could probably translate for us.’ I looked at her, and then I said, ‘What makes you think that?’ She said, ‘Well, your last name is [Participant's Last Name], is not it?’ I told her, ‘That does not have to do with my ethnicity. I'm not even Mexican or Hispanic at all.’ She just looked at me with a blank stare, and then she told me, “Well, what are you?” I told her, ‘I'm Native American, hundred percent…’ She was just like, ‘Oh, okay then. I just thought [Participant's Last Name] came from a Hispanic heritage.’ I just told her, ‘You're not the first. Everyone assumes that. You should not judge a book by its cover, though.’ (Woman, 20–30) Note: Example A.2.1 could also be considered a form of social exclusion involving the denial of one's ethnicity.
>
> A.2.2 “My experience occurred when I was at work. What happened was a patient's family member would not allow me to care for them because another co-worker had mentioned that I was Native American, and so I was asked to leave the room.” (Woman, 20–30).
>
> A.2.3 “When I told [my teacher] I wanted to be in the military and wanted to do these things, he told me I could not, saying that my writing skills and my speech and my way I carried myself, he said it was -- I would, it will never happen by the way I, by the way I was supposedly by the way my grades were. So, and I felt really bad about that; it made me feel, it made me feel like I could not do anything after that when he told me that.” (Man, 20–30).

3: Descriptions of Stigmatization

Some of these lived experience descriptions expressions of stigmatization include the invocation or assignment of negative stereotypes about the group (Excerpt A.3.1). Other expressions of stigmatization include verbal harassment, including slurs, of the targeted individuals (Excerpts A.3.2 – A.3.4).

> A.3.1 “I was at work. I was filling out my timesheet. Getting ready to go home. When this Caucasian gentleman he walked up to me, my supervisor at that, and he said to me, he asked me, he said, “What are you doing? Are you signing over the deed to the reservation?” (Man, 40–50). Note: Example could also be considered workplace discrimination.
>
> A.3.2 “I was at [Name] Middle School [State/City] with my friend, ***, who was also [Tribe Name]. We were outside after lunch. We were approached by two white girls who began to call us

> names. They called us "dirty Indians" and other derogatory names." (Woman, 40–50).
>
> A.3.3 "…just hanging out with everybody it would just get brought up from time to time that I was Native American and they would tease me about just by slapping their mouth and making, like, the hollering noise that's notorious for being done." (Man, 20–30).
>
> A.3.4 They would ask me if I smoke tobacco or if I'm alcoholic, or we'd occasionally talk about the Reservations and they would kind of tease me about that from time to time." (Man, 20–30).

4: Descriptions of Physical Threat

These lived experience descriptions of physical threat and harassment include implied or actual physical assault on the participant. These episodes are often accompanied by verbal maltreatment but are distinguished by menacing behavior, including the threat of physical violence (Excerpt A.4.1) or actual violence (A.4.2 and A.4.3). Quantitative data indicates that physical harassment scores on the Brief PEDQ-CV were lower than scores on measures involving social exclusion, stigmatization, and workplace discrimination. This pattern is consistent with the qualitative data in which a smaller proportion (13%) of individuals reported on an event in which they faced discrimination-related physical harassment. Examples of physical threats or harassment are illustrated below.

> A.4.1 "…we went for supplies to a nearby town to shop…one of my friends got spit in the face and told to go home. We were being followed by police. We were being followed by locals, who were -- wanted us to leave. They would tell us to go home, call us nasty names, whatever -- even the hotels, they would accost us." (Woman, 40–50).
>
> A.4.2 "I got attacked by a dog. I was with a white friend. I was at the park. The other person was a white guy. He had a dog and he let it loose and he was like "red meat." (Man, 40–50).
>
> A.4.3 "So this was a incident when I was about 13 years old. I got in a fight at school. It's a fist fight with a guy over a girl, and the police were called. And when they got there, I had a gun pointed in my face. I got hit in the back of the head with the gun, and this is just for getting into a fight. And the cop told me – I felt it was because it was my race and stuff – that I was – he was going to make sure I spent the rest of my life in jail." (Man, 40–50).

3.4 B: Responses to discrimination

These lived experience descriptions were used to classify emotional responses. Codes about emotions were applied if the participant identified the emotion they experienced following the discriminatory act. We categorized these emotions into two dimensions: anger-related emotions (e.g., including reports of feeling angry, mad, hostile, and related terms) and fear/sadness-related emotions (e.g., including reports of feeling sad, upset, afraid, anxious). This distinction between anger-related and fear/sadness related dimensions is common in the clinical literature (33). "First Party (Speaker) Response/ Intent" codes were applied to responses when participants were asked about their emotional responses to the situation and their coping responses to the situation and their own emotions. We found that 44% of the participants felt an anger-related emotion compared to 44% of participants who felt sadness or anxiety, including feeling fearful or upset in response to the act of discrimination. Figure 4 presents the frequency of "Emotional Response."

The next set of codes were used to classify coping responses. Two major categories were identified based on the literature (34): "Avoiding Confrontation/Emotional Suppression" and "Confront Directly/Addresses Discrimination." Responses were defined as "Confront/Directly Addresses Discrimination" confrontation if the participant addressed the act of discrimination by speaking up or doing something in response. Detailed definitions are located in Supplementary Tables S1–S7. Illustrative excerpts for these codes are highlighted in Descriptions 1 and 2 below. Figure 5 presents the frequency with which these different codes were applied. The graph shows that roughly 38% of the sample chose to "Avoid Confrontation/Avoid Escalation." In contrast, an estimated 47% of participants reported "Confront/Directly Addresses Discrimination."

1. Descriptions of Avoiding or Suppressing Emotion

For the "Avoiding Confrontation/Emotional Suppression" code, we combined "Avoiding Confrontation/Avoid Escalation" and "Inward Suppression of Emotion" because of conceptual and empirical overlap (35). Broadly, these lived experience descriptions were classified as "Avoidance or Suppression" if participants were aware of their distress and the injustice but chose to avoid confrontation and did not engage with the perpetrator (Excerpts B.1.1 and B.1.2) or if they suppressed the expression of their own emotions and responses following the act of discrimination (Excerpts B.1.3 and B.1.4).

> B.1.1. "I just had to ignore her and walk away because I did not want to go to jail." (Woman, 20–30).
>
> B.1.2. "It made me feel angry, but I did not portray that; of course upset, but I did not say much. Because it was at work, I tried to stay professional." (Woman, 40–50).
>
> B.1.3. "I shut down, and refused to talk to him, or to anyone." (Woman, 30–40).
>
> B.1.4. "So, and I felt really bad about that [The participant's teacher told him that he would never be able to join the military with his grades]; it made me feel, it made me feel like I could not do anything after that when he told me that. So I kind of just like, I did not say anything or do anything about it, I just kind of like kept it in as I grew up." (Man, 20–30).

2: Descriptions of Confronting or Directly Addressing Discrimination

These lived experience descriptions were classified as "Confronts/Directly Addresses Discrimination" if the participant directly identifies the maltreatment as discrimination (Excerpt B.2.1), asks for an apology (B.2.2), or persists in communicating with the perpetrators to complete their goals despite the discriminatory behavior (Excerpt B.2.3).

> B.2.1. "[The participant was filling out his timesheet at work] This Caucasian gentleman he walked up to me, my supervisor at that, and he said to me, he asked me, he said, "What are you doing? Are you signing over the deed to the reservation?" I was

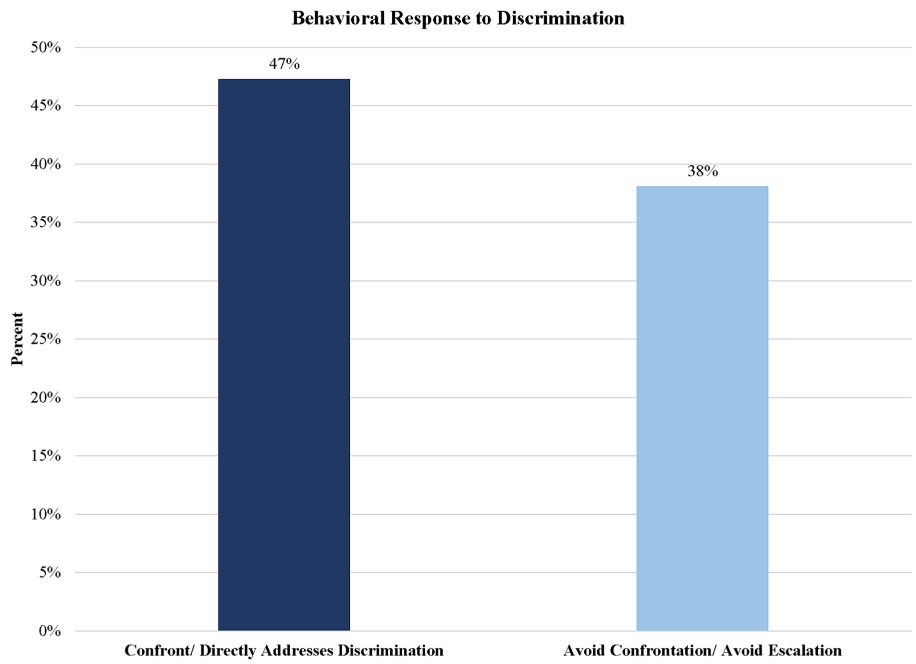

FIGURE 5
Individuals who confronted or avoided discrimination from others. Percentages represent the number of confrontations (or avoidance) divided by the total number of study participants ($n = 294$). As a consequence, some of the confrontation (or avoidance) responses were not mutually exclusive for some participants, or they may not have appeared at all for others. Thus, the percentages presented in this figure do not sum up to 100%. A chi-square goodness of fit test was performed to assess differences in observed frequencies of confrontational or avoidance behaviors, revealing non-significant differences, $X^2(1) = 2.90$, $p > 0.05$. These findings suggest that confrontational and avoidance behaviors are equally common responses to discriminatory behavior.

> shocked…[Participant discussed the issue with other supervisors, but they claimed it was a dead issue because he waited too long to report it]… I expressed to him that what he said is inappropriate and wrong, and he should not have said that. He claimed he wasn't trying to offend me, it was a joke. He said it in a joking manner. But how you could joke with someone you never even joked with before?" (Man, 40–50).
>
> B.2.2 In response to the incident described in A.2.1 (Following an episode in which the participant was asked to translate material as the manager assumed she was Hispanic). The participant went to the manager and asked for an apology, which she received.
>
> B.2.3 (Initial incident is described in excerpt A.1.4) I said, "But I am. I may look different, but I am." He said, "Well, what do you want to know?" I says, "Well, my tribe is [tribe name]," I said, "and who do I talk to about learning about my people?" (Man, 50–60).

3.5 C: Descriptions of resilience

This lived experience description was applied when participants reported ways in which they were resilient. Resilience was identified when individuals reported they were able to effectively manage discriminatory behavior or their emotional reaction to the discrimination. Often, they described reframing the experience or their reactions to the experience (Excerpt C.1-C.3); reported they experienced personal growth because of the way they handled it (Excerpt C.4), or valued their ability to lead others and themselves to change their perceptions (Excerpt C.5).

> C.1. "…I did not like it, but, you know, I was only one person. What could I do, you know. But I did not let it, you know, feed me that I was that person, you know, because I knew I wasn't you know. I knew I wasn't lazy. I knew I wasn't you know, dirty. I knew I wasn't uneducated, you know. So, you know, I did not let that affect me that much. It did affect me when, you know, it was happening, but like I said, I think as people get older and you, you know, observe, analyze and all that stuff, you know, you'll find yourself a better outcome than, you know, trying to be hateful back. You know, cause it do not solve nothing. And I wonder if, you know, those people in those surrounding towns ever understood that. You know, what is it going to resolve. You know, it just escalates and increases hatred, you know, treating people bad, you know." (Man, 40–50).
>
> C.2. "Now that I'm older I'm able to cope with it more and it does not really phase me… Like, just degrading stuff, like, it do not really get to me anymore because I know, like, how it is, and how my family is and stuff like that." (Man, 20–30).
>
> C.3. "There's not really much I can say or do. I just continue to pray for them, pray for peace in their heart to get their hate out for being judgmental against a person of a darker skin complexion when we all just pump the same blood. We all have one heartbeat, two eyes, one mouth, one nose. We're all human" (Man, 30–40).

> C.4. "You tend to ask why a lot because, you know, it (rejection) does not feel good. But when you find something that does make you feel good and something that you can identify with that other people cannot, then it takes away the bad feeling and replaces it with more of a good feeling, and that feeling is more like acceptance, you know. You guys might not accept me and that's because you do not understand, but I'm different and I have to accept me because you cannot understand because you are not me. So it just gave me an identity that I wasn't being given at home." (Man, 30–40).
>
> C.5 (continued from excerpt B.2.3) He escorted me over to a counselor. We sat down. We talked. But the feeling that I got was I wasn't perceived as Native American, because they seen the natural, my skin color, and just the overall appearance. But after a while, they got comfortable about being around me, and they started asking me questions. So, that ensued, I get to ask them questions. So, I start asking them about what tribe they are from and how they came to the community center, and it made me feel a little bit at ease. And by me talking to them and them talking -- and I'm talking back and forth, they got comfortable with me, and so they learned a little bit about me and my family history, and that's it. It just -- it was a different experience. It made me perceive people different, not because of the way they look or they dress, it's just by talking to them. And I feel, you know, I feel a little empowered." (Man, 50–60).

4 Discussion

To provide a framework for understanding the types of discriminatory experiences facing AI/AN People, we examined the quantitative data and provided qualitative accounts of these experiences in the participants' own words. These first-hand accounts can help build understanding and awareness for others to remember the types of maltreatment AI/AN People face in the U.S. Their lived experiences can provide an affective context, potentially motivating individuals to act to prevent and ultimately greatly reduce discrimination toward the First Peoples of North America.

The episodes of maltreatment presented by the participants were consistent with existing models of discrimination. Both social threats and physical threats were reported, with social threats more common (Figure 5). Example excerpts were provided of subtypes of social threat, including social exclusion, stigmatization, and discrimination. Many of the social threats experienced by AI/AN People conform to common stereotypes about underrepresented groups, consistent with the notion of stigmatization. These are all stereotypes that stigmatize AI/AN People and communicate that they are "other," less acceptable, and more dangerous than members of the majority group. These stereotypes include the ideas that AI/AN People are unclean, dangerous/savage, or alcoholic (17, 36). In the examples we included, participants were treated as if they were unclean (e.g., asked them to get out of the pool – Excerpt A.1.2.); dangerous (e.g., told them they would need to go to jail for the rest of their life – Excerpt A.4.3.); or savage (e.g., mimicking examples of Native calls as represented in movies – Excerpt A.3.3.). These stereotypes have been used to justify social and physical threat (17). As these examples demonstrate, negative stereotypes are translated into interpersonal maltreatment across a variety of contexts.

More specific to AI/AN groups are communications of invisibility both on an individual and a population level (18, 20, 37, 38). The communications of invisibility take several forms. In some cases, discriminatory treatment reflects expectations others have of AI/AN culture and AI/AN-specific phenotypes. For example, a perpetrator may view an AI/AN person as from another culture/ethnicity or declare their phenotype or name as inconsistent with their expectations about members of an Indigenous community. For two of the participants, a cultural attribute, such as long hair, is labeled as a problem with gender or sexual orientation rather than recognized as a marker of intelligence or strong Indigenous cultural identity (Excerpt A.1.5. and Excerpt A.1.6.). In other cases, invisibility is communicated through omission, when political leaders or news stories omit mention of AI/AN Peoples or their concerns (Excerpt A.1.7).

AI/AN People have also been stereotyped as less competent and lazy (17). Our participants provided examples of mistreatment consistent with these stereotypes, including experiences about others refusing to allow an AI/AN individual to treat a patient – Excerpt A.2.2. or asking if an AI/AN participant "will sign away the reservation" when completing forms at work– Excerpt A.3.1.

The stereotypes communicated can also be considered within the Stereotype Content Model proposed by Cuddy and Fiske (39). This model suggests that stereotypes can be organized along two primary dimensions: warmth and competence. The negative stereotypes, including those associated with dangerousness and invisibility are associated with low warmth. The acts of workplace discrimination and stigmatization are consistent with notions of low competence.

Perpetuation of these and other stereotypes can have significant consequences. These negative stereotypes and those associated with invisibility and denial of culture may have emerged from the need to justify stealing Indigenous lands (39–42). The narrative that AI/AN People were savages and had no civilized culture of their own further justified this theft of tribal lands (17). Stereotypes about criminality or irresponsibility may drive the high rates of arrest and incarceration to which AI/AN People are subject (43).

Consistent with other qualitative studies, the participants' experiences coping with discrimination by avoiding or confronting the perpetrators provide insight into the human toll of exposure to discrimination. Participants expressed concerns about the interpersonal or professional costs of communicating their anger (including, examples where people feared expressing their concerns because they may go to jail or would cause harm) (See Excerpts A.3.4, B.1.1). Participants also discussed the personal costs of the failure to express their distress, describing how upsetting it was to not communicate how they felt in the moment (See Excerpt B.1.4). Yet other participants provide models of ways to clarify and communicate, by asking for apologies and correcting assumptions (See Excerpts B.2.1, B.2.2, B.2.3, C.5). They frame their coping as a form of resilience, drawing pride from their culture and Tribal Nations. They show their ability to endure and address hardship and even have empathy for those that commit acts again them (See Excerpts C.1 – C.5).

Research has demonstrated the associations of discrimination and race related stress to poor health outcomes (44). The description of the participants' mental processes, as they describe their experiences and coping strategies, provides insights into the pathways through which discrimination can take a toll on physical and mental health. These findings and related data engender a call to action in the public health arena to educate the public about the types of discriminatory challenges facing the AI/AN community and support efforts to prevent discrimination and increase resilience.

There are several limitations to this study. Although we received qualitative data from a large number of AI/AN participants ($n = 294$), the data were collected from a group in the Denver-metro area; therefore, the results will not be similar to other Tribal groups living on reservation-based or rural areas and certainly does not reflect other urban Indigenous community's experiences. We used content analysis as a method to match to pre-existing structures, but did not probe or ask for clarification of the participants' experiences of discrimination. The data were collected in the context of a laboratory protocol asking for experiences of discrimination. The structured prompts for disclosure of discriminatory experiences may have limited the nature of the experiences communicated. However, the content of the laboratory prompts and the direction of the research were reviewed by the community advisory board from the time of study design to the end of data collection to ensure accuracy of experiences, understanding of the context, and to correct any assumptions researchers may have made in analyzing the data. In addition, the research assistants who coded participant excerpts were not AI/AN individuals, which may have resulted in some nuances of the participants' experiences being missed. However, five of the six coders were themselves members of other underrepresented groups (i.e., Black, Latino/a, or Asian). All coders had been participating in a research program investigating experiences of racial discrimination and their effects on health for at least 1 year.

5 Conclusion

Experiences of discrimination can take many forms and can be challenging to interpret. When these experiences are organized within standard models of discrimination, individuals may more readily recognize acts of discrimination and may be able to identify experiences of discrimination. This recognition can further facilitate the development of educational and public health interventions to prevent discrimination and mitigate its effects on the health and well-being of American Indian or Alaska Native People. In addition, those who are non-Indigenous may be able to become more effective allies and potentially reduce inadvertent communication of prejudice.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving humans were approved by St. John's University IRB, the University of Colorado Boulder IRB, and the Colorado Multiple Institutional Review Board (COMIRB). The studies were conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

Author contributions

RB: Conceptualization, Writing – original draft, Writing – review & editing, Formal analysis, Methodology, Visualization, Data curation, Investigation. MR: Conceptualization, Writing – original draft, Writing – review & editing, Data curation, Formal analysis, Methodology, Visualization, Investigation. IB: Conceptualization, Data curation, Methodology, Writing – original draft, Writing – review & editing, Investigation, Supervision, Funding acquisition, Project administration, Resources, Validation. EB: Conceptualization, Data curation, Methodology, Writing – original draft, Writing – review & editing, Funding acquisition, Investigation, Project administration, Supervision, Resources, Validation.

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Conflict of interest

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Supplementary material

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References

1. Bui AL, Coates MM, Matthay EC. Years of life lost due to encounters with law enforcement in the USA, 2015–2016. *J Epidemiol Community Health*. (2018) 72:715–8. doi: 10.1136/jech-2017-210059

2. Blair IV, Danyluck C, Judd CM, Manson SM, Laudenslager ML, Daugherty SL, et al. Validation of the brief perceived ethnic discrimination questionnaire–community version in American Indians. *Cult Divers Ethn Minor Psychol*. (2021) 27:47. doi: 10.1037/cdp0000419

3. Gone JP. Redressing first nations historical trauma: theorizing mechanisms for indigenous culture as mental health treatment. *Transcult Psychiatry*. (2013) 50:683–706. doi: 10.1177/1363461513487669

4. Krieger N. Embodying inequality: a review of concepts, measures, and methods for studying health consequences of discrimination. *Int J Health Serv*. (1999) 29:295352. doi: 10.2190/M11W-VWXE-KQM9-G97Q

5. Williams DR, Mohammed SA. Discrimination and racial disparities in health: evidence and needed research. *J Behav Med*. (2009) 32:20–47. doi: 10.1007/s10865-008-9185-0

6. Jones ML., Galliher RV. Daily racial microaggressions and ethnic identification among Native American young adults. *Cultural Diversity and Ethnic Minority Psychology*. (2015) 21:1

7. D'Amico EJ, Dickerson DL, Brown RA, Klein DJ, Agniel D, Johnson C. Unveiling an 'invisible population': health, substance use, sexual behavior, culture, and discrimination among urban American Indian/Alaska native adolescents in California. *Ethn Health*. (2021) 26:845–62. doi: 10.1080/13557858.2018.1562054

8. Whitbeck LB, Hoyt DR, McMorris BJ, Chen X, Stubben JD. Perceived discrimination, and early substance abuse among American Indian Children. *J Health Soc Behav*. (2001) 42:405–24. doi: 10.2307/3090187

9. Brown RA, Dickerson DL, Klein DJ, Agniel D, Johnson CL, D'Amico EJ. Identifying as American Indian/Alaska Native in urban areas: Implications for adolescent behavioral health and well-being. *Youth and Society*. (2021) 53, 54–75.

10. Dickerson DL, Brown RA, Klein DJ, Agniel D, Johnson C, D'Amico EJ. Overt perceived discrimination and racial microaggressions and their association with health risk behaviors among a sample of urban American Indian/Alaska Native adolescents. *Journal of Racial and Ethnic Health Disparities*. (2019) 6, 733–742. doi: 10.1007/s40615-019-00572-1

11. Johansson P, Jacobsen C, Buchwald D. Perceived discrimination in health care among American Indians/Alaska natives. *Ethn Dis*. (2006) 16:766–71.

12. Findling MG, Casey LS, Fryberg SA, Hafner S, Blendon RJ, Benson JM, et al. Discrimination in the United States: experiences of native Americans. *Health Serv Res*. (2019) 54:1431–41. doi: 10.1111/1475-6773.13224

13. Keeys M, Baca J, Maybank A. Focus: preventive medicine: race, racism, and the policy of 21st century medicine. *Yale J Biol Med*. (2021) 94:153.

14. Ozier EM, Taylor VJ, Murphy MC. The cognitive effects of experiencing and observing subtle racial discrimination. *J Soc Issues*. (2019) 75:1087–115. doi: 10.1111/josi.12349

15. Brondolo E, Kelly KP, Coakley V, Gordon T, Thompson S, Levy E, et al. The perceived ethnic discrimination questionnaire: development and preliminary validation of a community version 1. *J Appl Soc Psychol*. (2005) 35:335–65. doi: 10.1111/j.1559-1816.2005.tb02124.x

16. Contrada RJ, Ashmore RD, Gary ML, Coups E, Egeth JD, Sewell A, et al. Ethnicity-related sources of stress and their effects on well-being. *Curr Dir Psychol Sci*. (2000) 9:136–9. doi: 10.1111/1467-8721.00078

17. Robertson DL. Invisibility in the color-blind era: examining legitimized racism against indigenous peoples. *Am. Indian Quart*. (2015) 39:113–53. doi: 10.5250/amerindiquar.39.2.0113

18. House LE, Stiffman AR, Brown E. Unraveling cultural threads: a qualitative study of culture and ethnic identity among urban southwestern American Indian youth parents and elders. *J Child Fam Stud*. (2006) 15:393–407. doi: 10.1007/s10826-006-9038-9

19. Contrada RJ, Ashmore RD, Gary ML, Coups E, Egeth JD, Sewell A, et al. Measures of ethnicity-related stress: psychometric properties, ethnic group differences, and associations with well-being 1. *J Appl Soc Psychol*. (2001) 31:1775–820. doi: 10.1111/j.1559-1816.2001.tb00205.x

20. Juntunen CL, Barraclough DJ, Broneck CL, Seibel GA, Winrow SA, Morin PM. American Indian perspectives on the career journey. *J Couns Psychol*. (2001) 48:274–85. doi: 10.1037/0022-0167.48.3.274

21. Skewes M., Blume A. (2019). Understanding the link between racial trauma and substance use among American Indians. Available at: https://psycnet.apa.org/fulltext/2019-01033-008.html

22. Silva EO, Gillmann CJ, Tate KL. (2018). Confronting institutional discrimination in a color-blind world. Qualitative Sociology Review, 14, 84–108. doi: 10.18778/1733-8077.14.1.05

23. Brown RA, Dickerson DL, D'Amico EJ. Cultural identity among urban American Indian/Alaska native youth: implications for alcohol and drug use. *Prev Sci*. (2016) 17:852–61. doi: 10.1007/s11121-016-0680-1

24. Solomon TGA, Starks RRB, Attakai A, Molina F, Cordova-Marks F, Kahn-John M, et al. The generational impact of racism on health: voices from American Indian communities: study examines the generational impact of racism on the health of American Indian communities and people. *Health Aff*. (2022) 41:281–8. doi: 10.1377/hlthaff.2021.01419

25. Leveque D. M. (1994). Cultural and parental influences on achievement among native American students in Barstow unified School District. Paper presented at the national meeting of the Comparative and International Educational Society, San Diego, CA.

26. Montgomery D, Miville ML, Winterowd C, Jeffries B, Baysden MF. American Indian college students: an exploration into resiliency factors revealed through personal stories. *Cult Divers Ethn Minor Psychol*. (2000) 6:387–98. doi: 10.1037/1099-9809.6.4.387

27. Krieger N. Racial and gender discrimination: risk factors for high blood pressure?. *Social science and medicine*. (1990) 30:1273–1281. doi: 10.1093/oxfordjournals.aje.a115571

28. Whitbeck LB, McMorris BJ, Hoyt DR, Stubben JD, LaFromboise T. Perceived discrimination, traditional practices, and depressive symptoms among American Indians in the upper Midwest. *J Health Soc Behav*. (2002) 43:400–18. doi: 10.2307/3090234

29. Willeto AA, Sanderson PR, Barger SD, Teufel-Shone NI. "If you're down, you know, get up, be proud of yourself, go forward": exploring urban southwest American Indian individual resilience. *Am Indian Alaska Native Mental Health Res*. (2023) 30:53–81. doi: 10.5820/aian.3001.2023.53

30. Masten AS. Ordinary magic: resilience processes in development. *Am Psychol*. (2001) 56:227. doi: 10.1037/0003-066X.56.3.227

31. Danyluck C, Blair IV, Manson SM, Laudenslager ML, Daugherty SL, Jiang L, et al. Older and wiser? Age moderates the association between discrimination and depressive symptoms in American Indians and Alaska Natives. *J Aging Health*. (2021) 33:10S–7S. doi: 10.1177/08982643211013699

32. Danyluck C, Blair IV, Manson SM, Laudenslager ML, Daugherty SL, Brondolo E. Discrimination and sleep impairment in American Indians and Alaska natives. *Ann Behav Med*. (2022) 56:969–76. doi: 10.1093/abm/kaab097

33. Spielberger CD, Reheiser EC. Assessment of emotions: anxiety, anger, depression, and curiosity. *Appl Psychol Health Well Being*. (2009) 1:271–302. doi: 10.1111/j.1758-0854.2009.01017.x

34. Krieger N. Discrimination and health. *Soc Epidemiol*. (2014) 1:36–75. doi: 10.2190/HS.44.4.b

35. Peters BJ, Overall NC, Cameron LD, Hammond MD, Low RS, Girme YU. Do habitual emotional suppression measures predict response-focused situational suppression during social interactions? *Emotion*. (2020) 20:1005. doi: 10.1037/emo0000620

36. Burkley M, Burkley E, Andrade A, Bell AC. Symbols of pride or prejudice? Examining the impact of native American sports mascots on stereotype application. *J Soc Psychol*. (2017) 157:223–35. doi: 10.1080/00224545.2016.1208142

37. Carter RT, Forsyth J. Reactions to racial discrimination: emotional stress and helpseeking behaviors. *Psychol Trauma Theory Res Pract Policy*. (2010) 2:183–91. doi: 10.1037/a0020102

38. Langer CL, Furman R. Exploring identity and assimilation: research and interpretive poems. *Forum Qual Soc Res*. (2004) 5:2. doi: 10.17169/fqs-5.2.609

39. Fiske ST, Cuddy AJC, Glick P, Xu J. A model of (often mixed) stereotype content: competence and warmth respectively follow from perceived status and competition. *J Pers Soc Psychol*. (2002) 82:878–902. doi: 10.1037/0022-3514.82.6.878

40. Cuddy AJC, Fiske ST, Glick P. The BIAS map: behaviors from intergroup affect and stereotypes. *J Pers Soc Psychol*. (2007) 92:631–48. doi: 10.1037/0022-3514.92.4.631

41. Fiske ST, Cuddy AJC, Glick P. Universal dimensions of social cognition: warmth and competence. *Trends Cogn Sci*. (2007) 11:77–83. doi: 10.1016/j.tics.2006.11.005

42. Fiske ST, Xu J, Cuddy AC, Glick P. (dis)respecting versus (dis)liking: status and interdependence predict ambivalent stereotypes of competence and warmth. *J Soc Issues*. (1999) 55:473–89. doi: 10.1111/0022-4537.00128

43. Tighe S. 'Of course we are crazy': discrimination of native American Indians through. *Crim Justice, Justice Policy Journal*. (2014) 11:10–11.

44. Brondolo E, Blair IV, Kaur A. Biopsychosocial mechanisms linking discrimination to health: a focus on social cognition In: B Major, JF Dovidio and BG Link, editors. The Oxford Handbook of stigma, discrimination, and health, vol. *1*. New York, NY: Oxford University Press (2018)

45. Brondolo E, Kaur A, Flores M. Structural racism and health in the age of COVID-19: a selective review with policy implications. *Soc Issues Policy Rev*. (2023) 17:34–61. doi: 10.1111/sipr.12095

46. Brondolo E, Thompson S, Brady N, Appel R, Cassells A, Tobin JN, et al. The relationship of racism to appraisals and coping in a community sample. *Ethn Dis*. (2005) 15:14–9.

47. Colorado Commission of Indian Affairs. Available at: https://ccia.colorado.gov/ (Accessed May 5, 2021).

48. Norris T, Vines PL, Hoeffel EM. The american indian and Alaska native population: 2010. Washington, DC: US Department of Commerce, Economics and Statistics Administration, US Census Bureau (2023).

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T'ah kóó hóniidló, we're still HERE! Mining legacies, Indigenous health and innovative solutions

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In the world today, there are many unknowns especially with rising environmental concerns. However, one of the most important is an irreplaceable and shared resource, water or as the Diné (Navajo) refer to as, Tó. Throughout the world many Indigenous communities are facing water challenges, from lack of and access to adequate infrastructure, water rights, climate change and water contamination issues due to a variety of sources including anthropogenic sources like mining, especially, on the Navajo (Diné) reservation. This article aims to bring forth awareness of the long-standing water contamination issues in Diné communities and to shed light on innovative solutions being developed through current research efforts. Specifically, Dr. Tsosie's research aims to optimize a handheld point of use filter unit with a filter casing design that is customizable to a community and/or household through an easily removable and exchangeable cartridge system. Despite all the challenges and legacies of mining faced by not only Diné communities but many Indigenous communities, We Remain and We Are Still Here.

KEYWORDS

water filtration, Navajo Nation, water, health, mining legacies

1 Introduction

As a Diné scientist, I often reflect back on growing up on the Diné reservation and living without electricity and running water. This experience has given me a perspective that is much different than the average US citizen. I remember a time when my brothers and I would have to haul water from the local windmills for our livestock and our family's everyday household needs. Unknowingly, we were drinking from contaminated water sources. Over the next few decades, I began to educate myself about the potential connections between contaminated water sources and their impact on human health after witnessing fellow community members succumb to a wide variety of health conditions with possible links to environmental contaminants. These events have given me the motivation to pursue remedies for ground and surface water contaminations in the form of point-of-use filters.

Since this study is aimed at addressing these contamination issues within a Diné community, it is important to practice proper protocols of Indigenous Research Methodologies (IRM). IRM is a conceptual framework, based on an Indigenous ontological foundation and epistemological approach (1–7). The approach is often developed by the researcher through recognizing their Indigenous worldview and approaching their research from a holistic, respectful and inclusive manner with the community in which they are conducting research. IRM begins by approaching the research at the community level first and understanding the perspectives and concerns of the community. In this study, IRM was applied by using Diné

Worldviews to frame the scientific approaches and applying respectful collaborations with the community (8). The process of establishing respect and trust from the community is a priority. Developing these relationships was accomplished by hosting listening sessions and receiving community input. The protocols of gaining the respect and approval of the community to conduct the research on water sources was vital to the collaboration. Many of the concerns that arose from the community listening sessions were about human health impacts and impacts on livestock.

2 Background

2.1 History of the Diné and mining

The Navajo reservation (Diné Bikéyah) is located is the U.S. southwest and occupies the states of Arizona, New Mexico and Utah. It spans 27,000 square miles of "unparalleled beauty" (9). The population has grown to 399,494 enrolled tribal members with more than 166,000 that live on the reservation (10). The vast majority of the Navajo reservation is located on the Colorado Plateau (11). Considering the multiple campaigns by the U.S. government and military to remove the Diné from their homelands, we remain within our four sacred mountains that we hold reverent, a direct result of the resilient mindsets of our ancestors. The Diné did not go quietly and resisted removal for many years. The U.S. Army used tactics commonly labeled as "scorched earth policies" that destroyed more than 4,000 peach trees and 11,000 acres of both corn and beans (3).

The most prevalent and devastating issues, are the impacts and legacies from mining. The discovery of abundant natural resources on Diné lands, created an influx of prospectors and mining operations extracting coal, vanadium, uranium (U), copper, sand and gravel (12–22, 68, 69). These mining operations employed many Diné citizens who were not informed of the potential health hazards (12, 13). As these operations closed down, some were abandoned without proper cleanup. According to the Navajo Abandoned Mines Lands department, there are currently 273 coal sites, 33 copper mines and over 1,000 non-coal (uranium) abandoned mines, some have been and are being reclaimed (23). To date, several efforts are actively working with clean-up, education, and outreach. These efforts are difficult to address, expensive and require collaboration from local communities, tribal, state and federal agencies.

2.2 Metal impacts on human health

Numerous health effects that arise from exposure to heavy metals and metalloids including arsenic, uranium, lead, mercury, manganese, and others (12, 16, 18–22, 24, 25). These complex mixtures naturally occur in the environment and individuals are commonly exposed to these metals by inhalation, ingestion and in some cases through dermal contact. Comprehensive public health studies on the Navajo Nation report preliminary results that indicate chronic kidney disease, diabetes, high blood pressure and autoimmune disease are higher in Navajo communities with a higher number of uranium mines (26, 27). Initial exposure models indicate that environmental exposures, including living within 0.8 kilometer of a uranium mine site and coming in contact with wastes are significant predictors of kidney disease and diabetes (26, 27).

Uranium exposure has been linked to increases in cancer mortalities and has been demonstrated to have radiological effects in various organs such as bone, kidney, brain, liver, lung, intestine and reproductive systems (28–32). These effects are mainly due to uranium's radiological (alpha emitter) and chemical properties. Thus, uranium toxicity results from both chemical and radiological toxicity (33). A link has been established between birth defects and adverse pregnancy outcomes for women living in close proximity of abandoned mines (30, 34–36). Some of these exposures occur from occupational exposure and through exposure to mine tailings. Some studies have also indicated that acute exposure to uranium can be chemically toxic to the kidney and chronic exposure can be genotoxic (37).

Similarly, arsenic is also an element of concern throughout the world due to its toxicity. According to the World Health Organization, arsenic persists in groundwater and natural water sources and is highly toxic in its inorganic form (38). The common form of exposure is through ingestion and can lead to numerous adverse health effects in humans such as neuropathy, developmental disabilities, numerous skin disorders, hypertension, various cancers (skin, lungs, bladder and kidney), cardiovascular disease and diabetes (38–42). Some studies suggest that there are synergistic effects through co-contaminates such as arsenic and selenium that naturally occur in the environment and are associated with other types of health problems and cancers (30, 38, 41, 43).

While both uranium and arsenic have established maximum contaminate limits (MCL) in drinking water, vanadium currently does not have any set standards in the U.S. Carcinogenicity of vanadium has not been identified in drinking water standards, health advisories list, nor health hazards lists. The exception is vanadium pentoxide which has an inhalation risk and standards that vary from 0.02 to 0.1 mg/m^3, depending on the organization OSHA, NIOSH and the American Conference of Governmental Industrial Hygienists (44). In 2009, the U.S. EPA assessed the carcinogenicity of vanadium and concluded that an MCL cannot be specified. This is because there are no human data and inadequate animal studies with links to increased cancer mortalities and the potential carcinogenicity of soluble inorganic vanadium compounds (44, 60). However, chronic exposure to vanadium compounds can affect the respiratory system and causes irritation and more serious effects like bronchitis and pneumonitis (45). Determining exactly how vanadium contributes to respiratory related illnesses remains for the most part unknown. This area of vanadium research requires more *in vitro* and *in vivo* studies to determine the mechanisms of how it relates to respiratory illnesses. Additional studies are required to determine the overall effect of vanadium on both the environment and human health. Since vanadium concentrations are highest in soil, background levels similar to arsenic and uranium are created. Therefore, it is difficult to pinpoint the origin of the contamination in the environment.

2.3 Metal impacts on the environment

Water, a precious resource, is globally impacted because of anthropogenic sources like mining. While the geographic area of the

Navajo Reservation has a natural abundance of metal and metalloids like uranium, vanadium and arsenic, the disturbance of these ores by mining has mobilized these elements, increasing levels in ground and surface waters. Numerous studies by US EPA, DOE, USGS and academic institutions have reported the extent of uranium contamination (8, 14–23, 31, 34–36, 39, 40, 42–59). Arsenic and vanadium are rarely mentioned in these studies although they pose an additional threat to human health. Anthropogenic activities do contribute to sources of metal contamination in water, soil and air (32). Aside from the greatest concern for water source contamination, the extent of soil and vegetation contamination is understudied. There is a growing concern for soil and vegetation contamination because of the potential implication that it can have harmful effects on livestock, traditional plants and medicines. If a soil is contaminated with uranium, arsenic and vanadium, then there is an increased possibility that the vegetative cover is also contaminated by uptake into the roots and other parts of the plant. El Hayek *et.al* demonstrated the uptake of uranyl in roots of *Brassica juncea* (46). Other studies have shown uptake of arsenic in chokecherries and other indigenous plants (70, 71). Currently, a series of studies are being conducted within Diné communities focusing on livestock, vegetation and agricultural products and the uptake of contaminating metals, so that these mechanisms are better understood, and a plan can be developed to address these community and tribal concerns. At the top of the list are concerns for the Diné livelihoods of ranching and agricultural economies.

2.4 Historical and current efforts to address mining legacies

In the last 30 years, there have been numerous efforts to understand the extent of contamination in soil and water within the boundaries of the Navajo Nation. To date, some of the most concerning sites have been reclaimed to a certain extent (23, 39, 40). The remediation efforts are on-going by tribal and federal agencies. Additionally, researchers like myself, are conducting studies to further understand the extent of contamination and attempting to provide Diné communities with answers (8, 47–49, 58, 59). Some studies are investigating livestock and wildlife (50–52). Others are exploring innovative approaches to temporarily address the water contamination (53–56, 61–63).

In 1994 and through 2000, the USEPA Region 9, U.S. Army Corp of Engineers (USACOE), Navajo Nation EPA Superfund Program (NNEPA), Bechtel Environmental and the U.S. Department of Energy (USDOE) Remote Sensing Laboratory investigated issues related to abandoned mines and the exposure to radiation and toxic metals that led to the Abandoned Uranium Mines Project. The overall objective of the investigation was to determine whether abandoned uranium and vanadium mines or related mine features posed a significant risk to human health and to identify areas or features requiring action to reduce exposure. It was determined that there were multiple pathways of exposure included water, soil, groundwater and combined pathways (57).

In 2013, Dias da Cunha *et.al* conducted studies on radionuclides in four small communities. They presented results that identified 19% of the water sources tested had uranium concentrations that exceeded MCL for total uranium (47). Furthermore, 14% of the water sources tested exceeded MCL for U^{238} and 17% for U^{234} (47). The Ingram group at Northern Arizona University have been involved in a number of studies that identified water sources exceeding the established MCL for both uranium and arsenic (8, 48–50). Out of New Mexico, the University of New Mexico Center for Native Environmental Health Equity groups has a long history of studying impacts of uranium, mining on human health, as well as a number of other environmental impacts (53). There are numerous research groups conducting studies on the impacts of mining on the Diné people.

One interesting area of study is the development of innovative solutions to address the water quality issues that these communities are facing. A collaboration between Navajo Technical University and New Mexico Institute of Mining and Technology, the Navajo Nation Water Purification Project (N^4WPP), is currently developing desalinization units to provide an alternative and temporary solution for contaminated water sources (54). Researchers at the University of Arizona are also developing a similar technology, a solar powered desalinization unit that implements hollow fiber membranes to treat brackish waters (55). The University of Austin is collaborating with Diné citizens to impregnate silver nanoparticles with pine resin, used in traditional Diné pottery, as a means to remove bacteria from water (56). Collaborative research through the Center for Native Environmental Health Equity and Oklahoma State University is currently investigating the occurrence of microplastics and other products in open dump sites (53).

3 Development of innovative filtration technologies

Similarly, the Tsosie research group is developing a handheld water filtration unit for use by community water haulers. This project began as a vision and involved multiple partners from tribal communities to students. Tribal communities involved in this project, were involved by bringing forth concerns for their water sources. Past mining efforts to geologic conditions created challenging water insecurities and contamination issues that these communities continue to deal with. The point of use (POU) filter is designed with the user in mind. The filter is a handheld unit that is meant to travel with the user to unregulated water sources located, at times, miles apart. In the spring the availability of water is more widespread but as late fall approaches, the water becomes sparce. The POU filter will purify the water collected at these unregulated water sources, such as windmill wells. The filter ensures safe drinking water for livestock, for agricultural and household purposes. However, it is currently advised that these unregulated water sources should not be used for human consumption because these water sources are not regulated by the tribe.

In 2022, Dr. Tsosie collaborated with five mechanical engineering students, two students from civil engineering at Montana State University to design, develop and produce the handheld unit. An additional unit design feature is the customizable cartridge system. The unit has removable cartridges that allows for different solid phase extraction (SPE) materials to be changed and customized to the site. For example, the system could be used on the Navajo Nation, where the waters are high in arsenic, uranium and vanadium, and in Montana where the biggest challenge is nitrates and biological contaminates. The resin types can range from silica polyamine composites (SPCs) (8, 64–67), ion exchange resins, and other SPE materials that are currently on the market. SPCs are ideal since they are materials that can be regenerated. Thinking beyond just the Diné nation, the possibility of this filter being used to restore drinking water in communities facing similar contamination issues is something to aspire to for the well-being of future generations. This SPC-POU filter could have potential applications for elements of concern beyond just arsenic, vanadium and uranium. Examples include microplastics and per-and polyfluoroalkyl substances. This project is working on bench top studies to identify any unresolved issues with the unit. This could include issues with flow, increased pressure drops across the system, the potential need for a solar powered pump, and an indicator that identifies SPE materials are nearing saturation. In addition, studies are aimed at identifying additional SPE materials for selective removal of elements of concern.

These innovative technologies, led by Diné citizens, are addressing water challenges on the Navajo Nation and thereby addressing the health and environmental disparities being experienced by fellow tribal members. While the impacts and legacies from mining are great, there are also great minds attempting to resolve these issues. The process may be slow but progress is being made.

4 Conclusion

Throughout history Indigenous peoples have experienced so many hardships and setbacks, but we remain and we are still here. Through raising our voices and sharing our stories, we are able to bring change to our communities. Indigenous and non-Indigenous researchers alike are making a difference in our communities. Through efforts to bring awareness, promoting health and by creating innovative solutions, these communities are able to lessen the disparity gap. The examples brought forth in this perspective are a small portion of issues that are impacting Indigenous communities. The realm of environmental justice pertaining to Indigenous communities spans multiple areas, including water, soil, air, plants, animals, Indigenous data sovereignty, law, health, tribal rights and much more.

Incorporating IRM is vital to working *with* and *in* Indigenous communities. As the field of Indigenous scholars that are incorporating IRM into their research and practices is growing, it also becomes imperative that Indigenous Knowledge's and worldviews are equitably included in academic spaces. There is an opportunity for Indigenous researchers like myself to demonstrate that Indigenous and western science/knowledge's can cooperatively resolve problems in communities impacted by environmental injustices.

The intent and main motivation of the POU filter project was first and foremost the quality of life of the Diné people especially those living in close proximity of abandoned mines. It has been 38 years since the operations of these mines ceased and yet the Diné people continue to thrive despite the increased cancer mortalities, respiratory illness, diabetes and other diseases linked to the lingering effects of mining. The examples of new innovative solutions are a clear way forward. Finally, it does not matter who is at fault, but the efforts leading the charge to create change is more important.

WE ARE A RESILIENT & PROUD PEOPLE!

Data availability statement

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding author.

Author contributions

RT: Writing – review & editing, Writing – original draft.

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Conflict of interest

The author declares that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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References

1. Getty GA. The journey between western and indigenous research paradigms. *J Transcult Nurs*. (2009) 21:5–14. doi: 10.1177/1043659609349062

2. Chilisa B. Indigenous Research Methodologies. Thousand oaks, CA: SAGE Publications (2012).

3. Smith LT. Decolonizing methodologies: Research and indigenous peoples. *2nd* ed. London; New York: New York: Zed Books; New York: Distributed in the USA exclusively by Palgrave Macmillan (2012).

4. Kovach M. Indigenous methodologies: Characteristics, conversations and contexts. Toronto: University of Toronto Press Inc. (2012).

5. Wilson S. Research is ceremony: Indigenous research methods. Black Point, N.S: Fernwood Pub (2008).

6. Kirkness VJ, Barnhardt R. First nations and higher education: the four R's--respect, relevance, reciprocity, responsibility. *J Coll Univ Stud Hous*. (2016) 42:94.

7. Porsanger J. An essay about indigenous methodology. *Nordlit: Tidsskrift i litteratur og kultur*. (2004) 8:105. doi: 10.7557/13.1910

8. Tsosie R.L. Addressing long-standing water contamination issues in Diné communities using indigenous and Western scientific approaches [dissertation/master's thesis]. Missoula (MT): University of Montana. (2020).

9. Navajo Nation. Navajo nation history. Available at: http://www.navajo-nsn.gov/history.htm (Accessed November 20, 2019).

10. Lonas L. Navajo nation becomes largest tribe in US after pandemic saw climb in enrollment. US: The Hill.

11. ADWR. Arizona Department of Water Resources (ADWR). Arizona water atlas: volume 2 eastern plateau planning area. (2009).

12. Eichstaedt PH. If you poison us: Uranium and native Americans. *1st* ed. Santa Fe, N.M: Red Crane Books (1994).

13. Markstrom C, Charley P. Psychological effects of the technological/human-caused environmental disasters: examination of the Navajo and uranium. *Am Indian Alsk Native Ment Health Res*. (2003) 11:19–45. doi: 10.5820/aian.1101.2003.19

14. 66th Congress. Bureau of Indian Affairs appropriations. (1919).

15. Ascarza W. Mine tales: four corners area was great producer of vanadium. Arizona Daily Star Tucsoncom. (2016). Available from: https://tucson.com/news/local/mine-tales-four-corners-area-was-great-producer-of-vanadium/article_d31fc02e-79c8-538c-bdf4-e1708981bd22.html (Accessed November 20, 2019).

16. Brugge D, Goble R. The history of uranium mining and the Navaho people. *Am J Public Health*. (2002) 92:1410–9. doi: 10.2105/AJPH.92.9.1410

17. Chenoweth WL. Historical review uranium-vanadium production in the northern and western Carrizo Mountains, apache county, Arizona. *Open-File Report*. (1985):85–113.

18. Pasternak J. Yellow dirt: An American story of a poisoned land and a people betrayed. 1st free press. *Hardcover* ed. New York, NY: Free Press (2010).

19. Voyles TB. Wastelanding: Legacies of uranium mining in Navajo country. Minneapolis, MN: University of Minnesota Press (2015).

20. U.S. Environmental Protection Agency. Federal actions to address impacts of uranium contamination in the Navajo nation five-year plan summary report. (2013). Available at: http://www.epa.gov/region9/superfund/navajo-nation/pdf/NavajoUraniumReport2013.pdf (Accessed November 20 2019).

21. DeLemos JL, Brugge D, Cajero M. Development of risk maps to minimize uranium exposures in the Navajo Churchrock mining district. *Environ Health: Global Access Sci Source*. (2009) 8:29–43. doi: 10.1186/1476-069X-8-29

22. International Workshop on Environmental Contamination from Uranium Production Facilities and their Remediation (2004 : Lisbon, Portugal) Environmental contamination from uranium production facilities and their remediation : proceedings of an international workshop on environmental contamination from uranium production facilities and their remediation / organized by the International Atomic Energy Agency and held in Lisbon. (2005) 11–13.

23. AML/UMTRA Department. Navajo nation abandoned mine lands. (2024). Available at: https://aml.navajo-nsn.gov (Accessed February 6, 2024).

24. Girard JE. Analysis of water and wastewater: radioactive substances In: Principles of environmental chemistry, vol. *274*. *1st* ed. Sudbury, MA: Jones and Bartlett Publishers, Inc. (2005)

25. Tchounwou PB, Yedjou CG, Patlolla AK, Sutton DJ. Heavy metals toxicity and the environment. *EXS*. (2012) 101:133–64. doi: 10.1007/978-3-7643-8340-4_6

26. Shuey C. Uranium exposure and public health in New Mexico and the Navajo nation: A literature summary. (2008).

27. Hund L, Bedrick EJ, Miller C, Huerta G, Nez T, Ramone S, et al. A Bayesian framework for estimating disease risk due to exposure to uranium mine and mill waste on the Navajo nation. *J Royal Statistical Society Series A: Statistics in Society*. (2015) 178:1069–91. doi: 10.1111/rssa.12099

28. U.S. Environmental Protection Agency. Basic information about radionuclides in drinking water. Available at: http://water.epa.gov/drink/contaminants/basicinformation/radionuclides.cfm (Accessed November 20, 2019). (2013).

29. Darolles C, Broggio D, Feugier A, Frelon S, Dublineau I, de Meo M, et al. Different genotoxic profiles between depleted and enriched uranium. *Toxicol Lett*. (2010) 192:337–48. doi: 10.1016/j.toxlet.2009.11.009

30. Yazzie M, Gamble S, Civitello E, Stearns DM. Uranyl acetate causes DNA single strand breaks in vitro in the presence of ascorbate (vitamin C). *Chem Res Toxicol*. (2003) 16:524–30. doi: 10.1021/tx025685q

31. Harmon ME, Lewis J, Miller C, Hoover J, Ali AS, Shuey C, et al. Residential proximity to abandoned uranium mines and serum inflammatory potential in chronically exposed Navajo communities. *J Expo Sci Environ Epidemiol*. (2017) 27:365–71. doi: 10.1038/jes.2016.79

32. Zhang P, Yang M, Lan J, Huang Y, Zhang J, Huang S, et al. Water quality degradation due to heavy metal contamination: health impacts and eco-friendly approaches for heavy metal remediation. *Toxics*. (2023) 11:828. doi: 10.3390/toxics11100828

33. Priest N. Toxicity of depleted uranium. *Lancet*. (2001) 357:244–6. doi: 10.1016/S0140-6736(00)03605-9

34. Shields LM, Wiese WH, Skipper BJ, Charley B, Banally L. Navajo birth outcomes in the Shiprock uranium mining area. *Health Phys*. (1992) 63:542–51. doi: 10.1097/00004032-199211000-00005

35. Lewis J, Gonzales M, Burnette C, Benally M, Seanez P, Shuey C, et al. Environmental exposures to metals in native communities and implications for child development: basis for the Navajo birth cohort study. *J Soc Work Disabil Rehabil*. (2015) 14:245–69. doi: 10.1080/1536710X.2015.1068261

36. Hoover JH, Erdei E, Begay D, Gonzales M, NBCS Study TeamJarrett JM, et al. Exposure to uranium and co-occurring metals among pregnant Navajo women. *Environ Res*. (2020) 190:109943. doi: 10.1016/j.envres.2020.109943

37. Kopacek J, Ondrias K, Sedlakova B, Tomaskova J, Zahradnikova L, Sedlak J, et al. Type 2 IP3 receptors are involved in uranyl acetate induced apoptosis in HEK 293 cells. *Toxicology*. (2009) 262:73–9. doi: 10.1016/j.tox.2009.05.006

38. World Health Organization. Arsenic. (2018). Available at: https://www.who.int/news-room/fact-sheets/detail/arsenic (Accessed November 20, 2019).

39. Navajo nation environmental protection agency. AUM (2024). Available at: https://navajoepa.org [Accessed February 6, 2024].

40. United state Environmental Protection Agency. Navajo Nation and AUMs (2024). Available at: https://www.epa.gov/navajo-nation-uranium-cleanup (Accessed November 20, 2019).

41. Xu H, Zhou X, Wen X, Lauer FT, Liu KJ, Hudson LG, et al. Environmentally relevant concentrations of arsenite induce dose-dependent differential genotoxicity through poly(ADP-ribose) polymerase inhibition and oxidative stress in mouse thymus cells. *Toxicol Sci*. (2016) 149:31–41. doi: 10.1093/toxsci/kfv211

42. Brugge D, Buchner V. Health effects of uranium: new research findings. [Internet]. (2011) 26:231–249. Available at: https://pubmed.ncbi.nlm.nih.gov/22435323/

43. Cooper KL, Dashner E, Tsosie R, Cho YM, Lewis J, Hudson LG. Inhibition of poly(ADP-ribose)polymerase-1 and DNA repair by uranium. *Toxicol Appl Pharmacol*. (2015) 291:13–20. doi: 10.1016/j.taap.2015.11.017

44. Hoover J, Gonzales M, Shuey C, Barney Y, Lewis J. Elevated arsenic and uranium concentrations in unregulated water sources on the Navajo nation, USA. *Expo Health*. (2017) 9:113–24. doi: 10.1007/s12403-016-0226-6

45. Ding W, Liu W, Cooper KL, Qin XJ, de Souza Bergo PL, Hudson LG, et al. Inhibition of poly(ADP-ribose) polymerase-1 by arsenite interferes with repair of oxidative DNA damage. *J Biol Chem*. (2008) 284:6809–17. doi: 10.1074/jbc.M805566200

46. U.S. Environmental Protection Agency. Provisional peer-reviewed toxicity values for vanadium and its soluble inorganic compounds other than vanadium pentoxide (CASRN 7440-62-2 and others). (2009).

47. Dias DC, Henderson H, Thomson B, Hecht A. Ground water contamination with U, U, U, ra and pb from past uranium mining: cove wash, Arizona. *Environ Geochem Health*. (2014) 36:477–87. doi: 10.1007/s10653-013-9575-2

48. Credo J, Torkelson J, Rock T, Ingram JC. Quantification of elemental contaminants in unregulated water across Western Navajo nation. *Int J Environ Res Public Health*. (2019) 16:2727. doi: 10.3390/ijerph16152727

49. Jones L, Credo J, Parnell R, Ingram JC. Dissolved uranium and arsenic in unregulated groundwater sources – Western Navajo nation. *J Contemp Water Res Educ*. (2020) 169:27–43. doi: 10.1111/j.1936-704X.2020.03330.x

50. Jani C. Ingram; Lindsey Jones; Jonathan Credo; Tommy Rock uranium and arsenic unregulated water issues on Navajo lands. *J Vac Sci Technol A*. (2020) 38:031003. doi: 10.1116/1.5142283

51. Lister A, Froyum J, Ingram JC. Determination of uranium in sheep hearts from the Navajo nation. *J Vet Med Animal Sci*. (2021) 4:1055. doi: 10.47739/2378-931X/1263

52. Liu Z, Lin Y, Hoover JH, Beene D, Singer N, Charley P. Individual level spatial-temporal modeling of exposure potential of livestock in the cove wash watershed. *Arizona Annals of GIS*. (2023) 29:87–107. doi: 10.1080/19475683.2022.2075935

53. Center for Native Environmental Health Equity. PRO (2024). Available at: https://hsc.unm.edu/pharmacy/research/areas/native-eh-equity/ (Accessed February 6., 2024).

54. Navajo nation water purification project. N4WPP. (2024). Available at: https://www.nmt.edu/restore/ (Accessed February 6., 2024).

55. University of Arizona. Navajo water purification (2024). Available at: https://chee.engineering.arizona.edu/news-events/chemical-engineers-create-desalination-bus-navajo-nation (Accessed February 6., 2024).

56. Rowles LS, Tso D, Dolocan A, Kirisits MJ, Lawler DF, Navid B. Saleh integrating Navajo pottery techniques to improve silver nanoparticle-enabled ceramic water filters for disinfection. *Environ Sci Technol*. (2023) 57:17132–43. doi: 10.1021/acs.est.3c03462

57. U.S. Environmental Protection Agency. Abandoned uranium mines project, Arizona, New Mexico, Utah - Navajo lands, 1994-2000, project atlas. U.S: Environmental Protection Agency, region 9 (2000).

58. Athas W. Drinking water arsenic and cancer risk in New Mexico. *New Mexico Epidemiol.* (2010) 1:1–4.

59. Blake JM, Avasarala S, Artyushkova K, Ali A-MS, Brearley AJ, Shuey C, et al. Elevated concentrations of U and co-occurring metals in abandoned mine wastes in a northeastern Arizona native American community. *Environ Sci Technol*. (2015) 49:8506–14. doi: 10.1021/acs.est.5b01408

60. Costigan M, Cary R, Dobson S. Vanadium pentoxide and other inorganic vanadium compounds. (2001).

61. Rosenberg E, Pinson G, Tsosie R, Tutu H, Cukrowska E. Uranium remediation by ion exchange methods: a critical review. *Johnson Matthey Technol Rev*. (2016) 60:59–77.

62. Moore-Nall A, Tsosie R. REE data support Permian Phosphoria formation sourced oil as a source of metals for U-V mineralization in the northern Bighorn Basin. *Fortschr Mineral*. (2017) 7:232. doi: 10.3390/min7120232

63. Tsosie Ranalda, Rosenberg Edward "Uranium capture from high sulfate and nitrate waste streams with modified silica polyamine composites." *Proceedings of the international water conference (IWC)* (2017).

64. Tutu H, Bakatula E, Dlamini S, Rosenberg E, Kailasam V, Cukrowska EM. Kinetic, equilibrium and thermodynamic modelling of the sorption of metals from aqueous solution by a silica polyamine composite.(report). *Water SA*. (2013) 39:437. doi: 10.4314/wsa.v39i4.1

65. Allen JJ, Rosenberg E, Johnston E, Hart C. Sol-gel synthesis and characterization of silica polyamine composites: applications to metal ion capture. *ACS Appl Mater Interfaces*. (2012) 4:1573–84. doi: 10.1021/am201761m

66. Hughes MA, Nielsen D, Rosenberg E, Gobetto R, Viale A, Burton SD, et al. Structural investigations of silica polyamine composites: surface coverage, metal ion coordination, and ligand modification. *Ind Eng Chem Res*. (2006) 45:6538–47. doi: 10.1021/ie0601448

67. Allen J, Berlin M, Hughes M, Johnston E, Kailasam V, Rosenberg E, et al. Structural design at the polymer surface interface in nanoporous silica polyamine composites. *Mater Chem Phys*. (2011) 126:973–82. doi: 10.1016/j.matchemphys.2010.11.053

68. Chenoweth WL, Malan RC. The uranium deposits of northeastern Arizona. In: Monument Valley (Arizona, Utah and New Mexico). Eds. H. L. James. New Mexico Geological Society 24th Annual Fall Field Conference Guidebook. (1973):139–149.

69. Stokes WL. Carnotite deposits in the Carrizo Mountain area, Navajo Indian reservation, apache county, Arizona and San Juan county, New Mexico: USGS prof. Paper 690. New Mexico: Geological Survey Circular 11 (1951).

70. El Hayek E, Torres C, Rodriguez-Freire L. Effect of calcium on the bioavailability of dissolved uranium(VI) in plant roots under circumneutral pH. *Environ Sci Technol*. (2018) 52:13089–98. doi: 10.1021/acs.est.8b02724

71. Kant J, Larson G, Burckhard S, Berdanier B, Meyers R. Contemporary use of wild fruits by the Lakota in South Dakota and implications for cultural identity. *Great Plains Res*. (2015) 25:13–24. doi: 10.1353/gpr.2015.0011

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The adaptation process of a safety planning intervention to promote connectedness and reduce distress in Diné adolescents and adults during the COVID-19 pandemic

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Introduction: Indigenous connectedness is an impetus for health, well-being, self-confidence, cultural preservation, and communal thriving. When this connectedness is disrupted, the beliefs, values, and ways of life that weave Indigenous communities together is threatened. In the Spring of 2020, the COVID-19 virus crept into Tribal Nations across the United States and exacerbated significant health-related and educational inequities. The first case of COVID-19, or *Dikos Ntsaaígíí-19* in the *Diné* (Navajo) language, arrived in the Southwest in March 2020. The virus quickly spread amongst Diné communities and contributed to immediate school closures. These closures created significant disruption to connectedness on the Navajo Nation.

Methods: As part of the Community Based Participatory Research process, our team worked with a Community Advisory Board (CAB) to facilitate a longitudinal cohort study titled "Project SafeSchools" and, most critically, to culturally adapt mental health interventions to be implemented as a part of this study. This paper describes the process our team used to select, adapt, and test Safety Planning and Caring Contacts interventions to reduce elevated rates of depression, anxiety, and suicidal ideation amongst Diné adolescents and adults. CBPR was the primary approach used to engage with Navajo Nation communities and the adaptation process was guided by a scoping study of frameworks for adapting public health evidence-based interventions (EBI) and was guided by the cultural adaptation process. Our team met virtually several times in 2021 and 2022 as the parent launched and as the pilot randomized clinical trial called "+Connection is Medicine" started. When Safety Planning and Caring Contacts messaging was selected, the study team, which consisted primarily of Navajo research personnel led by a Navajo community-based principal investigator (PI) and allied PIs with extensive experience in implementation science, infectious disease prevention, and CBPR, were adapted and presented to CAB members. This CAB also included a youth advisory council who participated in the field testing and further adaptation process.

Results: The use of the CAB allowed for a collaborative workgroup effort to examine the feasibility and acceptability of using safety planning to help reduce suicide risk factors with Navajo adolescents and adults. Most importantly, this CAB collaborative with researchers to further tailor safety plan intervention materials to align with Navajo values related to connectedness to relatives, community, mental health resources, and the land. In addition, the caring contacts messaging was aligned with safety plans to provide culturally sensitive messages that would be shared with randomized participants.

Discussion: Mental health stigma is highly common in reservation-based communities. In Indigenous communities, mental illness has association with not living well or not living culturally aligned further complicates the likelihood of at-risk community members contacting resources available in their communities. By using an Indigenous approach that restores connectedness, and reminds participants of their belonging, +CiM researchers were able to develop enhanced versions of safety plans to use in their pilot randomized controlled trial.

KEYWORDS

COVID-19, safety planning, mental health, American Indian, stress

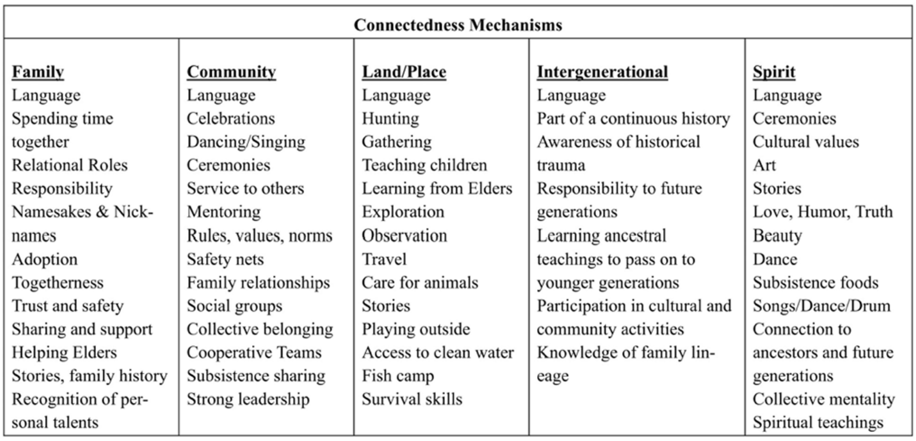
Introduction

> "Before me peaceful, behind me peaceful, under me peaceful, over me peaceful, all around me peaceful." ~ A Diné Teaching.

Dr. Jessica Ulrich, an Inupiaq researcher, presents a context for this paper – "Indigenous peoples are not trapped in a traumatic past (1, p.1)." We start this paper with this perspective to purposefully celebrate Indigenous resilience. Ullrich (1) goes on to describe how Indigenous people have always overcome hardships and provides a framework for how to represent the interplay of Indigenous connectedness and resilience. It is through this resilience that we identify vital protective factors of Indigenous flourishment as we strive to build peaceful and healthier Indigenous communities while also supporting these communities after the COVID-19 pandemic. A prominent feature of healthy Indigenous communities is connectedness, evidenced by the observation of thriving Indigenous communities. The communities that maintain their old ways of living and knowledge are often the communities that demonstrate higher resilience in times of hardship. It is also these communities that model how Indigenous values can serve as protective factors for well-being. To "live in a good way" means that the Indigenous person knows who they are and where they come from. They also live in balance with their children, partners, family, community, and the earth. Ulrich's Indigenous Connectedness Framework depicts essential connectedness mechanisms that are unique to Indigenous peoples: Family, Community, Land/Place, Intergenerational, and Spirit (see Figure 1 for connectedness mechanisms). Indigenous connectedness is an impetus for health, mental well-being, self-confidence, stability, cultural preservation, self-identity and communal thriving. When this connectedness is disrupted, the glue that holds Indigenous communities together is threatened. We utilized this framework as way to address the mental health stigma common in Indigenous communities by conceptualizing a traditional value related to belonging within a family and community, which has proven to be protective of mental health status (2–4). This approach allowed the +Connection is Medicine research team to think of mental health solutions from an Indigenous perspective and to tailor interventions designed to foster connectedness and restore belonging for adolescents and adults impacted by the COVID-19 pandemic.

Background

In the Spring of 2020, the COVID-19 virus crept into Tribal Nations across the United States (U.S.) and exacerbated significant health-related and educational inequities. Tribal Nations were tasked with protecting and advocating for their sovereignty and demonstrating self-determination to help ensure the survival of Indigenous peoples and their ways of knowing. Early in the pandemic, mortality was the highest amongst American Indian and Alaska Native (AI/AN) people (5). During this time, most public schools across the U.S. stopped in-person learning in the Spring of 2020 to reduce the transmission of the virus among child populations. The first case of COVID-19, or *Dikos Ntsaaígíí-19* in the *Diné* language, arrived in the Southwest in March 2020. Amongst the Navajo Nation communities the virus spread quickly resulting in immediate school closures across the reservation. During the 2020–2021 academic school year, most schools on the reservations remained closed for in-person learning, and many switched to virtual learning, as a result of Tribal Nations having sovereignty over their lands. The switch entirely to online learning was challenging and has had lasting impacts on child learning (25, 26). Schools on the reservation were known to provide critical nutritional, physical, and social–emotional support to students and families before the pandemic. The reservation-based schools did not only focus on the youths' academic instructions but served as an access point for nutritious breakfast and lunch, healthcare, and connection to peers and other adults who provide



Connectedness Mechanisms				
Family	**Community**	**Land/Place**	**Intergenerational**	**Spirit**
Language Spending time together Relational Roles Responsibility Namesakes & Nicknames Adoption Togetherness Trust and safety Sharing and support Helping Elders Stories, family history Recognition of personal talents	Language Celebrations Dancing/Singing Ceremonies Service to others Mentoring Rules, values, norms Safety nets Family relationships Social groups Collective belonging Cooperative Teams Subsistence sharing Strong leadership	Language Hunting Gathering Teaching children Learning from Elders Exploration Observation Travel Care for animals Stories Playing outside Access to clean water Fish camp Survival skills	Language Part of a continuous history Awareness of historical trauma Responsibility to future generations Learning ancestral teachings to pass on to younger generations Participation in cultural and community activities Knowledge of family lineage	Language Ceremonies Cultural values Art Stories Love, Humor, Truth Beauty Dance Subsistence foods Songs/Dance/Drum Connection to ancestors and future generations Collective mentality Spiritual teachings

FIGURE 1
Mechanisms of Indigenous Connectedness offered by Ulrich.

critical social support to the students. While schools and families mounted tremendous efforts to overcome these disruptions (e.g., delivery of meals, school buses equipped with Internet hotspots and delivery of school supplies to students), school closure contributed to a reduction in access to critical resources especially those who were in need most (6, 7). In response to this our team of Indigenous and allied researchers pivoted to serving the community by conducting formative research which provided foundational knowledge on the mental health and psychosocial impacts of COVID-19 and school closures on Diné youth and adult wellbeing.

In the Spring of 2021, our team launched the "Project SafeSchools" (PSS), a prospective cohort study on the Navajo Nation and another Southwest Tribe. The study aims were to: (1) understand the barriers and facilitators to school re-opening and in-person school attendance from the perspective of multiple stakeholders in Diné and Apache communities; and (2) evaluate the educational, social, emotional, physical, and mental health impacts of returning to in-person learning for caregivers and youth ages 4–16 (8). This study was approved by the Navajo Nation Health Research Board, agency councils, and local chapter governments. We also had the support from the Navajo Nation Department of Diné Education and the local Indian Health Service units.

As part of the Indigenous Community Based Participatory Research (ICBPR) framework, our community-based study team leads worked with local Community Advisory Boards (CABs) which consisted of tribal community members and partners to help in the design of the PSS study and provide ongoing feedback (9). Further, the ICBPR included frequent engagement with tribal governing agencies, such as chapter houses, tribal councils, and tribal public health offices (10). Preliminary results from the initial baseline surveys indicated high levels of distress among caregivers and their youth. Our CABs urged action and in response, our team pivoted to selecting and adapting evidence-based interventions to address distress in our study population. Thus, a pilot randomized controlled trial called +Connection is Medicine (+CiM) on the Navajo Nation was created. This paper aims to describe the process our team used to select, adapt, and test evidence-based interventions that can be leveraged to promote connectedness and reduce stress for the families enrolled in the PSS cohort.

Methods

The primary objective of the +CiM intervention was to reduce elevated rates of depression, anxiety, and suicidal ideation in our parent study pool through the adaptation of Safety Planning and Caring Contacts interventions which have successfully been used in communities that have experienced highly traumatic events, including veterans and Indigenous people. A key component of this adaptation was the culturally tailoring of this intervention to prioritize Indigenous connectedness as a conceptual framework. In this paper, we focus on the adaptation process that took place on the Navajo Nation, specifically, within the communities of Shiprock, NM, Chinle, AZ, and Tuba City, AZ. The adaptation process primarily focused on the experiences of reservation-based Navajo families. The overall study was approved by the Johns Hopkins Bloomberg School of Public Health Institutional Review Board (IRB #00020570) and the Navajo Nation Human Subjects Research Review Board (NNR-22.445).

Science of adaptation

Furthermore, we used the steps identified by Escoffery et al. (11) to organize how our research team adapted our +CiM intervention to promote connectedness and reduce distress for vulnerable study participants. The adaptation process is based on a scoping study of frameworks for adapting public health evidence-based interventions (EBI) and was guided by the cultural adaptation process of Ward et al. (12). This approach offers researchers strategies on how to organize adaptation of existing EBIs and has been commonly used by public health teams. The stages include the following: (1) assess the community, (2) understand the EBI(s), (3) select the EBI, (4) consult with experts, (5) consult with stakeholders, (6) decide what needs adaptation, (7) adapt the original program, (8) train staff, (9) test the adapted materials, (10) implement, and (11) evaluate (see Figure 2 for a detailed description of each category). Steps for adaptation for each Tribal Nation will be discussed within the context of each category below. We do not present steps (10) and (11) as those are reported once the study has been completed.

Step name	Step descriptions
1.Assess community	• Identify behavioral determinants and risk behaviors of the new target population using focus groups, interviews, needs assessments, and logic models • Assess organizational capacity to implement the program
2.Understand the intervention	• Identify and review relevant EBPs and their program materials • Understand the theory behind the programs and their core elements
3.Select intervention	• Select the program that best matches the new population and context
4.Consult with experts	• Consult content experts, including original program developers, as needed • Incorporate expert advice into program
5.Consult with stakeholders	• Seek input from advisory boards and community planning groups where program implementation takes place • Identify stakeholder partners who can champion program adoption in new setting and ensure program fidelity
6.Decide what needs adaptation	• Decide whether to adapt or implement original program • Theater test selected EBP using new target population and other stakeholders to generate adaptations • Determine how original and new target population/setting differ in terms of risk and protective factors • Identify areas where EBP needs to be adapted and include possible changes in program structure, content, provider, or delivery methods • Retain fidelity to core elements • Systematically reduce mismatches between the program and the new context
7.Adapt the original program	• Develop adaptation plan • Adapt the original program contents through collaborative efforts • Make cultural adaptations continuously through pilot testing • Core components responsible for change should not be modified
8.Train staff	• Select and train staff to ensure quality implementation
9.Test the adapted materials	• Pretest adapted materials with stakeholder groups • Conduct readability tests • Pilot test adapted EBP in new target population • Modify EBP further if necessary
10.Implement	• Develop implementation plan based on results generated in previous steps • Identify implementers, behaviors, and outcomes • Develop scope, sequence, and instructions • Execute adapted EBP
11.Evaluate	• Document the adaptation process and evaluate the process and outcomes of the adapted intervention as implemented • Write evaluation questions; choose indicators, measures, and the evaluation design; plan data collection, analysis, and reporting • Employ empowerment evaluation approach framework to improve program implementation

FIGURE 2
Stages of Implementation Science.

Assessing the community needs

The Johns Hopkins Center for Indigenous Health (JHCIH) has a successful three-decade relationship working with federally recognized tribes in the Southwest. JHCIH has a long history of scaling public health studies in reservation and rural communities. JHCIH's earliest work focused on collaborating with the Navajo Nation, the communities of focus in this study. JHCIH remains committed to addressing elevated rates of mental distress across these Tribal Nations. Over the years, the JHCIH has successfully recruited members of these two tribal communities to serve as research staff and eventually as principal investigators of major studies. This approach has helped researchers to understand the main drivers of poor health and systemic issues that exacerbate infectious diseases and allowed this research team to be well-equipped to shift their focus to supporting the mental health of PSS study participants. This history combined with initial data from the PSS Cohort study led to our team quickly responding to the mental health needs of the Diné and Apache children and adults most in need.

Understanding and selecting the EBIs

Given the complexities of working within an ongoing pandemic with changing public health mitigation approaches, our team sought brief interventions that were both feasible and impactful. Our teams in both tribal communities have a long history of working on suicide prevention including the development of the award-winning Celebrating Life Program [CL; (13)]. This work combined with Navajo teachings on connections to others which had been undermined by the pandemic, led our team to select interventions that have been shown to reduce distress and social isolation. Our team selected both the Safety Planning Intervention (14) and Caring Contacts (15) interventions as priority evidence-based programs to adapt. Both these interventions are brief and require minimal in-person contact, which was key given the changing nature of the COVID-19 pandemic, particularly on Tribal lands.

Select the intervention

Safety planning interventions (SPI) are a crucial component of suicide prevention strategies and are considered an evidence-based intervention that has successfully been used with Indigenous communities by researchers at JHCIH (16). The SPI is a brief intervention that directly targets suicide risk with demonstrated efficacy (27). The SPI aims to provide people with an individualized set of steps that can be used progressively to both reduce risk and maintain safety when distressing thoughts emerge. In the SPI, safety plans are developed collaboratively between providers and at-risk individuals. The developers of SPI have also developed and tested an enhanced version of safety planning known as SPI+, which includes a series of brief telephone calls after discharge that focus on assessing risk, revising the safety plan, and facilitating connections to care. The SPI+ is conceptualized to target three proximal targets and underlying mechanisms of suicide risk including increasing coping, decreasing access to lethal means, and increasing engagement in care (17). This enhanced version was utilized in the +CiM study.

Similarly, the Caring Contacts intervention is a low-cost, low-intensity mental health program designed to reduce suicide risk, which involves sending periodic, brief, non-demanding messages of care and concern to individuals who have exhibited suicide risk (18, 19). It has been shown in numerous studies to reduce the risk of suicide and associated thoughts and behaviors (15). The underlying mechanisms thought to drive change include instilling a sense of connectedness and social support, which are well-documented protective factors against mental illness and suicidal thoughts and behaviors. These messages are perceived as a supportive presence that may disrupt feelings of isolation and hopelessness, common factors contributing to suicidal thoughts.

Consult with experts

Research staff from the CL program were consulted to provide their thoughts and feedback on both the intervention adaptation and study design. Many of the CL staff have nearly 20 years of experience in the mental and behavioral health field, and several serve as expert trainers in suicide prevention interventions. Key questions for experts included, how to broaden the target of these interventions to not only focus on suicide risk but to apply it to broader mental distress. Navajo Nation CAB members provided their thoughts and feedback on the appropriateness of these interventions in their communities at the time. Finally, our team engaged two suicide and mental health experts, both clinical psychologists who have worked closely with Indigenous communities, at the JHCIH, who also provided their feedback on the appropriateness and potential adaptation of the selected interventions.

Consult with stakeholders

The research team utilized community-based participatory research methods to engage community input. We relied heavily on the parent study's (PSS) CAB members from the participating communities to provide us with valuable input throughout all stages of the adaptation process for +CiM. We met virtually for CAB meetings with advanced notification to maximize attendance and availability of CAB members. CAB members were members of the tribal nations, community partners, physicians, educators, and parents. For +CiM on the Navajo Nation, we recruited adolescent youth between the ages of 12–16 to serve on our youth advisory board, mostly to aid with the adaptation of our safety plan intervention for youth. Prior to CAB meetings, proposed adaptations of the intervention materials were sent to members for review. During CAB meetings, the project PIs, one of which is a member of the Navajo Nation, facilitated the discussion about the culturally tailoring process and proposed procedures while study coordinators recorded feedback and responses. The PIs and coordinators met during weekly meetings to discuss and synthesize CAB member feedback.

Decide what needs adaptation

Based on these notes during these CAB meetings, the feedback was discussed as a team and themes were identified based on discussion and direct feedback during these sessions. Cultural responsivity and sensitivity were the primary areas recommended for adaptation, as recommended by CAB members. This included thinking through procedures for how to introduce the concepts of mental health and suicidality with AI/AN people who have cultural taboos against discussions of death and loss of life. In addition, CAB members from both communities suggested the use of culturally tailored messaging for the Caring Contacts materials. Therefore, two distinct types of Caring Contact messages were developed, one set for each tribal community.

Adapt the original program

The recommendations from the CAB members from both communities were directly incorporated into existing safety plan materials from Stanley and Brown (14). Additional adaptations included the creation of a safety planning guide created specifically for use in reservation-based communities and incorporated cultural safety practices. This included helping participants to connect with not just family members and local and national crisis resources, but also to spaces in their community that make them feel safe (e.g., a traditional dwelling, an outdoor space or a family gathering area). Safety plans were individualized for adolescents and youth and were field tested with members of our community partners and youth advisory council that consisted of Navajo adolescents. Safety plans have been utilized in other community-based suicide and other violence prevention and activities and were adapted through input from local teams that work on suicide prevention in the local communities (20, 21).

Train staff

After the adequate approvals were obtained from institutional and tribal review boards, the research team went into formal and informal training procedures to train field staff on safety plans which became known as "Coping Plans" for adult participants and "Helping Plans" for youth participants. As timing was a major factor with the study, we opted for using our existing PSS research team who were already trained in responsive research practices and conducting suicide prevention interventions. An in-person meeting was conducted with community-based teams with the project leads training staff on safety planning, standardization of conducting the plan, and methods for data collection. Following the in-person meeting, a community-based investigator provided on-going in person support to teams which included role play with life and retroactive feedback and direct coaching. In addition, training videos were recorded to allow team members access to a pre-recorded standardized training session with a participant.

Test the adapted materials

Once the study team was trained through in-person role play scenarios that included a structured feedback sessions from study team leads in which a fidelity checklist was used to track key implementation steps, the study team was ready to implement the safety plans with participants. The training fidelity checklist was used as a guide for ongoing fidelity checks to maintain the standardization of safety plan administration. The team then moved into active recruitment and consenting of participants.

Results

The COVID-19 pandemic revealed many challenges in delivering mental health interventions to vulnerable Indigenous populations. This included the complete halting of in-person programming, limited mental health personnel, and an overworked healthcare system that shifted all attention to pandemic mitigation. What was learned quickly with the adaptation of the safety planning intervention with +CiM was that EBIs can be culturally adapted and modified quickly and effectively to meet the urgent mental health needs of AI/AN communities. In precision public health, we think of how to quickly initiate action to meet the health care needs of a community experiencing inequity. An essential part of providing effective mental health support to reservation-based families on the Diné is understanding the beauty and intricacy of Indigenous connectedness and how disruptions can lead to increased community stress. Using a precision public health approach to determine an area of need and applying an EBI within an Indigenous conceptual framework shows that researchers can effectively respond to community crises in culturally responsive manners. To accomplish this, there must be existing relationships with tribal communities.

Strong indigenous community partnerships

Strong community partnerships, developed over time and following CBPR processes, in the two tribal communities played a vital role in improving health outcomes and promoting overall well-being in our study participants. This specifically included transparency throughout the research process, which included frequent reporting to CAB members and community partners. Health is not solely determined by access to healthcare services or individual behaviors; it is influenced by a wide range of social, economic, environmental, and cultural factors. By fostering collaboration between healthcare providers, community organizations, local government, and residents, strong community partnerships can address these underlying determinants of health and create sustainable improvements.

Indigenous community health workers

The community health worker (CHW) model is a strategy that involves trained individuals from within the community who act as a bridge between healthcare providers and the community members they serve (22–24). CHWs are tribal community members who live and work in tribal communities, who are trained to provide basic healthcare services, health education, and support to underserved populations, particularly in rural or disadvantaged communities. When it comes to improving mental health in rural communities, the CHW model can be highly effective. The use of CHWs has proven to be an effective public health approach to addressing mental health needs in these two tribal communities. CHWs establish strong connections with community members, gaining their trust and understanding of their unique cultural, social, and economic contexts. These paraprofessionals engage with individuals and community leaders to identify mental health challenges, stigma, and barriers to care. CHWs act as navigators, helping individuals access mental health services in their communities. They assist with making appointments, accompany individuals to appointments if needed, and help navigate the complex healthcare system. They provide information about available mental health resources, such as counseling centers, support groups, or helplines. CHWs often have similar backgrounds or life experiences as the community members they serve. This shared experience allows CHWs to offer empathetic and culturally responsive peer support, reducing the stigma associated with mental health and creating a safe space for individuals to share their struggles. CHWs can also advocate for the needs of their community, raising awareness among policymakers and healthcare providers about the mental health challenges faced by rural populations. CHWs maintain ongoing contact with individuals to ensure they are connected to appropriate mental health services and to provide continued support and encouragement. They can conduct regular check-ins, reinforce treatment plans, and assist with medication adherence, if applicable. This continuity of care improves treatment outcomes and reduces the risk of relapse.

Overall, the CHW model offers a comprehensive approach to improving mental health in rural tribal communities. By leveraging their local knowledge, cultural sensitivity, and close relationships with community members, CHWs can address the unique challenges faced by rural populations, reduce barriers to care, and promote mental well-being. Care coordination is crucial in mental healthcare as it plays a vital role in ensuring effective and efficient delivery of care to patients. It involves the seamless integration of various healthcare professionals, services, and resources to create a comprehensive and well-coordinated plan of action. By facilitating communication, collaboration, and information sharing among providers, care coordination promotes continuity, avoids duplication of efforts, reduces medical errors, and enhances patient outcomes. It helps to optimize the use of healthcare resources, improve patient satisfaction, and increase the overall quality and safety of care. Ultimately, care coordination serves as a critical link that connects patients to the right care at the right time, making it an essential component of a patient-centered and efficient healthcare system.

Discussion

During the COVID-19 pandemic, it was apparent early on that the mental health of adolescents and adults would be greatly impacted due to the constant exposure to stress, loss of life, and major disruption to social networks. When working with communities that have experienced elevated stressors linked to a natural disaster or pandemic, the adaptation of mental health-related interventions must take precedence in recovery efforts. This adaptation paper highlights how researchers engaged tribal community member partners can come together to develop solutions to respond to elevated rates of mental health crises in culturally responsive ways. This paper also highlights the importance of building strong working relationships with tribal public health programs, schools, and tribal governments to plan sustainable community-based interventions. There were limitations to implementing safety plan interventions, without the cultural adaptations, on the Navajo Nation. Therefore, we present in this paper a model for how to enhance and adapt exiting EBIs to pilot across Indigenous communities. When there are limited mental health solutions in reservation-based communities, research teams must strategize how to use what works and how to further tailor these solutions to the everchanging needs and demands of Indigenous communities. It is the intent of the researchers to track and report how these EBIs worked on the Navajo Nation and more papers will be published sharing the preliminary findings.

Data availability statement

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding author.

Author contributions

JA-B: Conceptualization, Funding acquisition, Methodology, Writing – review & editing, Writing – original draft, Validation, Supervision, Resources, Project administration. VB: Writing – review & editing, Writing – original draft. KD-C: Writing – review & editing, Writing – original draft. LJ: Writing – review & editing, Writing – original draft. SA: Writing – review & editing, Writing – original draft. RB: Writing – review & editing, Writing – original draft. LH: Writing – review & editing, Writing – original draft. EH: Writing – original draft, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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References

1. Ullrich JS. For the love of our children: an indigenous connectedness framework. *AlterNative*. (2019) 15:121–30. doi: 10.1177/1177180119828114

2. Nelson V, Derrett S, Wyeth E. Indigenous perspectives on concepts and determinants of flourishing in a health and well-being context: a scoping review protocol. *BMJ Open*. (2021) 11:e045893. doi: 10.1136/bmjopen-2020-045893

3. de Gouveia J, Ebersöhn L. Wellbeing as positive outcome of a social connectedness pathway to resilience: an indigenous psychology perspective In: Handbook of quality of life in African societies (2019). 71–95.

4. Bullen J, Hill-Wall T, Anderson K, Brown A, Bracknell C, Newnham EA, et al. From deficit to strength-based aboriginal health research—moving toward flourishing. *Int J Environ Res Public Health*. (2023) 20:5395. doi: 10.3390/ijerph20075395

5. Arrazola J, Masiello MM, Joshi S, Dominguez AE, Poel A, Wilkie CM, et al. COVID-19 mortality among American Indian and Alaska native persons — 14 states, January–June 2020. *MMWR Morb Mortal Wkly Rep*. (2021) 69:1853–6. doi: 10.15585/mmwr.mm6949a3

6. Nicola T, Gable A, Ash J. The response of rural districts to the Covid-19 pandemic. Cambridge, MA: Center for Education Policy Research (2020).

7. Running Bear C, Terrill WPA, Frates A, Peterson P, Ulrich J. Challenges for rural native American students with disabilities during COVID-19. *Rural Special Educ Quart*. (2021) 40:60–9. doi: 10.1177/8756870520982294

8. Allison-Burbank JD, Ingalls A, Rebman P, Chambers R, Begay R, Grass R, et al. Measuring the effects of the COVID-19 pandemic on Diné and White Mountain apache school personnel, families, and students: protocol for a prospective longitudinal cohort study. *BMC Public Health*. (2022) 22:1481. doi: 10.1186/s12889-022-13208-7

9. Begay R, Archuleta S, Allison-Burbank JD, Begaye V, Howe L, Jim L, et al. Indigenous community-based participatory research approach and safe return to in-person learning for Diné youth and families. *Am J Public Health*. (2024) 114:S350–3. doi: 10.2105/AJPH.2024.307576

10. Christopher S, Saha R, Lachapelle P, Jennings D, Colclough Y, Cooper C, et al. Applying indigenous community-based participatory research principles to partnership development in health disparities research. *Fam Community Health*. (2011) 34:246–55. doi: 10.1097/FCH.0b013e318219606f

11. Escoffery C, Lebow-Skelley E, Udelson H, Böing EA, Wood R, Fernandez ME, et al. A scoping study of frameworks for adapting public health evidence-based interventions. *Transl Behav Med*. (2019) 9:1–10. doi: 10.1093/tbm/ibx067

12. Ward EA, Iron Cloud-TwoDogs E, Gier EE, Littlefield L, Tandon SD. Cultural adaptation of the mothers and babies intervention for use in tribal communities. *Front Psych*. (2022) 13:807432. doi: 10.3389/fpsyt.2022.807432

13. Cwik MF, Tingey L, Maschino A, Goklish N, Larzelere-Hinton F, Walkup J, et al. Decreases in suicide deaths and attempts linked to the White Mountain apache suicide surveillance and prevention system, 2001–2012. *Am J Public Health*. (2016) 106:2183–9. doi: 10.2105/AJPH.2016.303453

14. Stanley B, Brown GK. Safety planning intervention: a brief intervention to mitigate suicide risk. *Cogn Behav Pract*. (2012) 19:256–64. doi: 10.1016/j.cbpra.2011.01.001

15. Skopp NA, Smolenski DJ, Bush NE, Beech EH, Workman DE, Edwards-Stewart A, et al. Caring contacts for suicide prevention: a systematic review and meta-analysis. *Psychol Serv*. (2023) 20:74–83. doi: 10.1037/ser0000645

16. Cwik MF, Barlow A, Goklish N, Larzelere-Hinton F, Tingey L, Craig M, et al. Community-based surveillance and case management for suicide prevention: an American Indian tribally initiated system. *Am J Public Health*. (2014) 104:e18–23. doi: 10.2105/AJPH.2014.301872

17. Boudreaux ED, Stanley B, Green KL, Galfalvy H, Brown GK. (2021) a randomized, controlled trial of the safety planning intervention: research design and methods. *Contemp Clin Trials*. (2021) 103:106291. doi: 10.1016/j.cct.2021.106291

18. Luxton DD, Kinn JT, June JD, Pierre LW, Reger MA, Gahm GA. Caring letters project: a military suicide-prevention pilot program. *Crisis*. (2012) 33:5–12. doi: 10.1027/0227-5910/a000093

19. Reger MA, Luxton DD, Tucker RP, Comtois KA, Keen AD, Landes SJ, et al. Implementation methods for the caring contacts suicide prevention intervention. *Prof Psychol Res Pract*. (2017) 48:369–77. doi: 10.1037/pro0000134

20. Bagwell-Gray ME, Loerzel E, Dana-Sacco G, Messing J, Glass N, Sabri B, et al. From myPlan to ourCircle: adapting a web-based safety planning intervention for native American women exposed to intimate partner violence In: Indigenous health equity and wellness: Routledge (2022). 168–85.

21. Brockie T, Decker E, Barlow A, Cwik M, Ricker A, Aguilar T, et al. Planning for implementation and sustainability of a community-based suicide surveillance system in a native American community. *Implement Sci Commun*. (2023) 4:1–12. doi: 10.1186/s43058-022-00376-1

22. O'Keefe VM, Cwik MF, Haroz EE, Barlow A. Increasing culturally responsive care and mental health equity with indigenous community mental health workers. *Psychol Serv*. (2021) 18:84–92. doi: 10.1037/ser0000358

23. Roland KB, Milliken EL, Rohan EA, DeGroff A, White S, Melillo S, et al. Use of community health workers and patient navigators to improve Cancer outcomes among patients served by federally qualified health centers: a systematic literature review. *Health Equity*. (2017) 1:61–76. doi: 10.1089/heq.2017.0001

24. Spencer MS, Rosland AM, Kieffer EC, Sinco BR, Valerio M, Palmisano G, et al. Effectiveness of a community health worker intervention among African American and Latino adults with type 2 diabetes: a randomized controlled trial. *Am J Public Health*. (2011) 101:2253–60. doi: 10.2105/AJPH.2010.300106

25. Groetzinger K. *Navajo Families without Internet Struggle to Home School during COVID-19 Pandemic*. Guam: National Public Radio (NPR), (2020).

26. Ferguson M, Rhodes K, Loughhead M, McIntyre H, Procter, N. The Effectiveness of the Safety Planning Intervention for Adults Experiencing Suicide-Related Distress: A Systematic Review. *Archives of suicide research: official journal of the International Academy for Suicide Research*, (2022). 26:1022–1045. doi: 10.1080/13811118.2021.1915217

27. Stanley B, Brown GK, Brenner LA, Galfalvy HC, Currier GW, Knox KL, et al Comparison of the Safety Planning Intervention With Follow-up vs Usual Care of Suicidal Patients Treated in the Emergency Department. *JAMA psychiatry*, (2018). 75, 894–900. doi: 10.1001/jamapsychiatry.2018.1776

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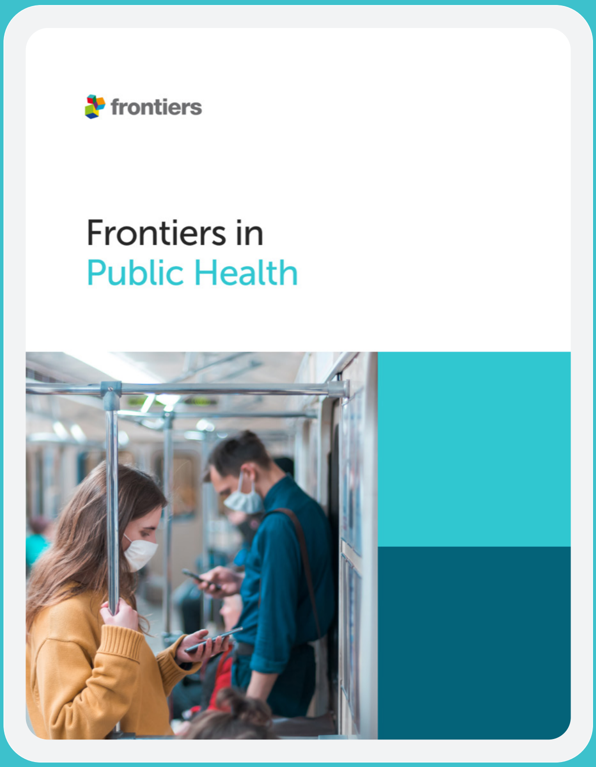

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