Describing the experience of Indigenous peoples with prenatal alcohol exposure and FASD: a global review of the literature to inform a Kaupapa Māori study into the experiences of Māori with FASD

Emma Espiner, Freeman Apou, Elizabeth Strickett, Andi Crawford, Maria Ngawati

ABSTRACT

AIM: This paper reports the findings of a literature review in answer to the research question: “What are the strengths and weaknesses of the existing research into the experience of prenatal alcohol exposure and Foetal Alcohol Spectrum Disorder in Indigenous communities?”

METHOD: MEDLINE (Ovid), psychnfo, CINAHL Plus and Web of Science, EMBASE, Informit databases were searched using key words to identify relevant literature. Given the anticipated scarcity of research relevant to our study, no geographic or chronologic limitations were placed on the searches. Studies which were solely descriptive were excluded, but reviews were included. The data analysis was informed by a Kaupapa Māori positioning and the ‘CONSIDER’ statement on reporting of health research involving Indigenous people.

RESULTS: Thirty-four papers met the inclusion criteria. Four main categories of papers were found: protocols/methods development, evaluation of interventions to reduce alcohol-exposed pregnancy, research seeking to understand alcohol use in pregnancy and interventions to improve the experience of people with FASD and their families. Indigenous peoples of Australia, Canada and North America were the participants of the papers found in this review, with only one research study found from Aotearoa New Zealand.

CONCLUSION: The existing literature on FASD in Indigenous communities internationally is heavily skewed towards the development and evaluation of interventions to reduce alcohol-exposed pregnancies. There is also a focus on studies which aim to understand and describe the variables which lead to alcohol use among Indigenous communities, and the relationship with alcohol use in the perinatal period. In the last ten years, a number of protocols/methods development for FASD-related interventions in Indigenous communities have been published. There is one published study from Aotearoa in the scientific literature into the experience of Māori with FASD.

Foetal Alcohol Spectrum Disorder (FASD) is one of the key causes of developmental disability and is characterised by severe neurological impairments that significantly impact general health and wellbeing. People who live with FASD are at risk of developing a number of secondary challenges which include but are not limited to: disrupted school experiences, trouble with the law, mental health problems, suicide and addictions. These challenges occur more frequently when difficulties are not understood, and when children and whānau are unsupported early in life. Due to significant and lifelong learning and behavioural needs, individuals with FASD typically require much more support and appropriate interventions in order to function to their potential.

Rationale for undertaking this research

FASD is a poorly understood but increasingly recognised problem in Aotearoa. The increasing prevalence of FASD has significant implications for health equity, addiction services, education, the justice system and public health. Māori are disproportionately affected, which has implications under Te Tiriti o Waitangi (Treaty...
of Waitangi) that promises tino rangatiratanga (Māori sovereignty—ultimate control and authority) and equity to Māori.\textsuperscript{15}

To understand the aetiology of FASD as well its wide-ranging secondary harms, the historical and contemporary context of the lives of whānau affected by FASD must be examined. Recent Doctoral research in Aotearoa suggests that colonisation, as well as historical and contemporary trauma, create a context for tamariki (children) Māori to be over-represented in a clinical sample of tamariki experiencing FASD.\textsuperscript{16} Furthermore, these tamariki were found to experience challenges in many areas (eg low socio-economic status, a high prevalence of adverse childhood experiences (ACEs), parental learning difficulties, multiple placements) and despite presenting with significant neurological challenges they often did not meet the criteria for health and education services.\textsuperscript{16}

As with other Indigenous populations, Māori in Aotearoa have also experienced trauma due to colonisation, and subsequently experience greater levels of inequity than non-Māori.\textsuperscript{17,18} An over-representation of FASD in international research also suggests a higher prevalence of FASD in First Nation populations,\textsuperscript{19,20} and a need to consider colonisation and historical trauma having a direct effect on the health and social context of First Nation populations.\textsuperscript{21} In fact, in terms of prevalence, it is suggested that it may be more important to focus on the intergenerational trauma of colonisation rather than associating FASD prevalence with First Nation populations.\textsuperscript{22,23,24} An increased risk of alcohol abuse in First Nation populations may be more likely to be related to depression and trauma arising from past and present processes of colonisation, rather than race or culture. Of more concern is research suggesting that First Nations people of Canada experience a higher risk of suicide and involvement in justice and care and protection systems.\textsuperscript{25} It is likely that First Nations people from all nationalities who experience FASD will require support and intervention that is designed to address these inequities.\textsuperscript{25}

As the impact of FASD on whānau (family) is increasingly being recognised, front line service providers are doing their best to respond, but are continuing to experience systemic barriers within health, education and social services that disadvantage their clients pre- and post-diagnosis.\textsuperscript{13,26} The dearth of research into the experiences of Indigenous peoples with FASD, and the lack of validated interventions, contributes to the challenges for whānau and service providers seeking culturally safe and effective ways of working together to minimise the negative consequences of FASD.

As part of this research project into the experiences of whānau Māori with FASD, a literature review has been designed to inform the development of the research protocol. This review is intended to broadly sample the existing literature on the Indigenous experience of FASD, and any culturally safe interventions which have been developed, implemented and/or evaluated.

### Methods

A semi-systematic meta-narrative review was employed due to the utility of this method for our broad research question. A meta-narrative examines overlapping fields of study on a topic by elucidating the complementary or contrasting strategies employed. While narrative reviews are informative, and allow for greater unstructured searching, they also have a greater risk of selection bias.\textsuperscript{27} By using structured standards as set out by PRISMA guidelines, we can eliminate bias, and have a greater strength to the overall discussion.\textsuperscript{28} PRISMA is the gold standard for examining RCTs in a systematic fashion, but is also used when evaluating interventions.\textsuperscript{29} Additionally, the limitations of studies will reflect the need for greater understanding of the experiences of Māori with FASD and how future research can facilitate this.

MEDLINE (Ovid), psychINFO, CINAHL Plus and Web of Science, EMBASE, Informit databases were searched using key words to identify relevant literature. Given the anticipated scarcity of research relevant to our study, no geographic, chronicologic or authorship limitations were placed on the searches. Studies which were solely descriptive were excluded, but reviews were included. Other sources included supplied research from members of the research team, grey literature and the references of existing reviews on the topic. The data analysis was informed by a Kaupapa Māori positioning and the ‘CONSIDER’ statement on reporting of health research involving Indigenous people.\textsuperscript{30,31}

### Results

Seven hundred and fifteen papers were identified from initial searching. Duplicate articles were removed, and the remaining abstracts were reviewed using inclusion and exclusion criteria resulting in 28 articles for study inclusion following full article review. One additional paper was
identified for inclusion from a systematic review (Table 2). Five additional papers were identified through peer review by Andi Crawford. Studies were from the United States of America (n=17), Australia (n=13), Canada (n=3) and Aotearoa (n=1).

Combining a Kaupapa Māori analysis with the recommendations of the ‘CONSIDER’ statement on reporting of health research involving Indigenous people, the included literature varied significantly in terms of its alignment with best practice research principles when working with Indigenous peoples.\(^{30,31}\) Researcher positionality was rarely mentioned. Most authors avoided deficit framing and attempted social determinants-based analyses of findings. Author definitions of culturally appropriate methods and interventions demonstrated poor consistency, varying from retro-fitted “consultation” where an attempt is made to introduce an Indigenous framework or intervention halfway through a project, to mature partnerships between researchers and Indigenous leaders where communities identify project aims, contribute to methods development, co-design interventions and facilitate evaluation.

The literature included in this review was organised into four categories, outlined below. The categories are: protocols/methods development; interventions to reduce alcohol-exposed pregnancy; research into alcohol use in Indigenous communities; and interventions to improve the experience of Indigenous people with FASD and their families.

### Protocols/methods development

The research team chose to include papers (n=12) to

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<tr>
<th>Table 1: Literature search inclusion and exclusion criteria.</th>
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<tr>
<td><strong>Population</strong></td>
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<td><strong>Intervention</strong></td>
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<td><strong>Comparison</strong></td>
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<td><strong>Outcomes</strong></td>
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<td><strong>Inclusion criteria</strong></td>
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<td><strong>Exclusion criteria</strong></td>
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Table 2: Literature analysed (34 papers).

<table>
<thead>
<tr>
<th>Lead author, date</th>
<th>Title</th>
<th>Country</th>
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<tbody>
<tr>
<td>Andersson E., 2019</td>
<td>Jandu Yani U for All Families’ Triple P-positive parenting program in remote Australian Aboriginal communities: A study protocol for a community intervention trial</td>
<td>Australia</td>
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<tr>
<td>Crawford A., 2020</td>
<td>Cognitive and social /emotional influences on adaptive functioning in children with FASD: Clinical and cultural considerations</td>
<td>New Zealand</td>
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<td>Bridge P., 2011</td>
<td>Ord Valley Aboriginal Health Service’s fetal alcohol spectrum disorders program: Big steps, sold outcome</td>
<td>Australia</td>
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<tr>
<td>Doherty E., 2019</td>
<td>Antenatal care for alcohol consumption during pregnancy: Pregnant women’s reported receipt of care and associated characteristics</td>
<td>Australia</td>
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<tr>
<td>Finlay-Jones, 2020</td>
<td>Community Priority setting for Fetal Alcohol Spectrum Disorder Research in Australia</td>
<td>Australia</td>
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<tr>
<td>Fitzpatrick J P., 2012</td>
<td>The Liliwan Project: study protocol for a population-based active case ascertainment study of the prevalence of fetal alcohol spectrum disorders (FASD) in remore Australian Aboriginal communities</td>
<td>Australia</td>
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<tr>
<td>Gibson S, 2020</td>
<td>Influences on drinking choices among indigenous and non-indigenous pregnant women in Australia: A qualitative study</td>
<td>Australia</td>
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<tr>
<td>Hamilton S L, 2020</td>
<td>“That thing in his head”: Aboriginal and non-Aboriginal caregiver responses to neurodevelopmental disability diagnoses</td>
<td>Australia</td>
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<tr>
<td>Hansen J D, 2015</td>
<td>Development and implementation of CHOICES Group to Reduce Drinking, Improve Contraception, and Prevent Alcohol-Exposed Pregnancies in American Indian Women</td>
<td>USA</td>
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<tr>
<td>Hansen J D, 2015</td>
<td>Importance of social support in preventing alcohol-exposed pregnancies with American Indian communities</td>
<td>USA</td>
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<tr>
<td>Hansen J D, 2013</td>
<td>Prevention of alcohol-exposed pregnancies with American Indian women</td>
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<tr>
<td>Hansen J D, 2017</td>
<td>Impact of the CHOICES Intervention in Preventing Alcohol-Exposed Pregnancies in American Indian Women</td>
<td>USA</td>
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<tr>
<td>Hansen J D, 2015</td>
<td>The oglala sioux tribe CHOICES program: Modifying an existing alcohol-exposed pregnancy intervention for use in an American Indian community</td>
<td>USA</td>
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<tr>
<td>Hansen J D, 2020</td>
<td>Acceptability of an eHealth Intervention to Prevent Alcohol-Exposed Pregnancy Among American Indian/Alaska Native Teens</td>
<td>USA</td>
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<tr>
<td>Hansen J D, 2012</td>
<td>Development of a Media Campaign on Fetal Alcohol Spectrum Disorders for Northern Plains American Indian Communities</td>
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<tr>
<td>Keightley M, 2018</td>
<td>Investigating a theatre-based intervention for Indigenous youth with fetal alcohol spectrum disorder</td>
<td>Canada</td>
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<tr>
<td>Ma G X,</td>
<td>Fetal alcohol syndrome among Native American adolescents: A model prevention program.</td>
<td>USA</td>
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Table 2 (continued): Literature analysed (34 papers).

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<tr>
<th>Lead author, date</th>
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<tr>
<td>Ma G X, 1998</td>
<td>The use of qualitative approach in fetal syndrome prevention among American Indian youth</td>
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<tr>
<td>Ma G X, 1998</td>
<td>Native American adolescents' views of fetal alcohol syndrome prevention in schools</td>
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<td>Massis K B, 1991</td>
<td>A comprehensive local program for the prevention of fetal alcohol syndrome</td>
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<td>Mascotti P, 2006</td>
<td>Preventing fetal alcohol spectrum disorder in Aboriginal communities: a methods development project</td>
<td>Australia</td>
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<tr>
<td>May P A, 1989</td>
<td>A macro-level fetal alcohol syndrome prevention program for native americans and Alaska natives – description and evaluation</td>
<td>USA</td>
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<tr>
<td>May P A, 2008</td>
<td>Enhanced case management to prevent fetal alcohol spectrum disorders in Northern Plains communities</td>
<td>USA</td>
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<tr>
<td>Montag A C, 2015</td>
<td>Preventing Alcohol-Exposed Pregnancy Among an American Indian/Alaska Native Population: Effect of a Screening, Brief Intervention, and Referral to Treatment Intervention</td>
<td>USA</td>
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<tr>
<td>Montag A C, 2017</td>
<td>Tailoring an Alcohol Intervention for American Indian Alaska Native Women of Childbearing Age: Listening to the Community</td>
<td>USA</td>
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<tr>
<td>Morton Ninomiya M E, 2017</td>
<td>Reconciling community-based indigenous research and academic practices: Knowing principles is not always enough</td>
<td>Canada</td>
</tr>
<tr>
<td>Pei J, 2019</td>
<td>Exploring the contributions and suitability of relational and community-centred fetal alcohol spectrum disorder (FASD) prevention work in First Nation communities</td>
<td>Canada</td>
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<tr>
<td>Plaisier K J, 1989</td>
<td>Fetal Alcohol Syndrome prevention in American Indian communities of Michigan's Upper Peninsula</td>
<td>USA</td>
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<tr>
<td>Reid N 2021</td>
<td>Yarning about fetal alcohol spectrum disorder: Outcomes of a community-based workshop</td>
<td>Australia</td>
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<tr>
<td>Shrestha U, 2019</td>
<td>Community perceptions of alcohol exposed pregnancy prevention program for American Indian and Alaska native teens</td>
<td>USA</td>
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<tr>
<td>Wagner B, 2017</td>
<td>The development of a culturally appropriate school based intervention for Australian Aboriginal children living in remote communities: A formative evaluation of the Alert Program</td>
<td>Australia</td>
</tr>
<tr>
<td>Wagner B, 2018</td>
<td>Study protocol for a self-controlled cluster randomised trial of the Alert Program to improve self-regulation and executive function in Australian Aboriginal children with fetal alcohol spectrum disorder</td>
<td>Australia</td>
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<tr>
<td>Wagner B, 2020</td>
<td>School-based intervention to address self-regulation and executive functioning in children attending primary schools in remote Australian Aboriginal communities</td>
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describing protocols and methods development due to their relevance in relation to the broader project that this literature review forms part of—a Kaupapa Māori study into the experience of whānau Māori with FASD. This subset of papers included the most nuanced descriptions of co-design methods and engagement with Indigenous communities, which is directly relevant to the methodology of the broader research project. For example, the Marulu Strategy is the result of Aboriginal leaders in the Fitzroy Valley, Australia, engaging strategic partners to address FASD in the region. A key part of the strategy, the Lililwan Project, is a population-based active case ascertainment study of all children born in 2002 and 2003 in the Fitzroy Valley. The context for this study is a national lack of diagnostic services for FASD and a consequent lack of quality prevalence data. The aim is to establish the prevalence of FASD and other health and developmental problems in school-aged children residing in the Fitzroy Valley, providing data to inform FASD prevention and management. The research is intended to contribute specifically to a feasible and transferable model of FASD diagnosis, and a model for culturally responsive research with Aboriginal communities.

Another intervention in the Marulu Strategy is described by Wagner et al (2018), who published a study protocol to evaluate a school-based programme designed to improve self-regulation and executive function in Aboriginal children with FASD. The programme being evaluated, the “Alert Program”, was designed by occupational therapists and was not specifically created for Indigenous children, but has been adapted. Anderson et al (2019) describe the adaptation of a parenting programme for the community, arising from the needs identified in the Lililwan study. The “Triple P-positive parenting program” looked at enhancing the confidence of local practitioners and carers in strategies to manage complex child behaviours, and to ameliorate challenging behaviours of the children with FASD in their care.

Outside of the literature relating specifically to the Lililwan Project, Masotti et al (2006) describe a three-year methods development project based on Participatory Action Research (PAR) principles in Canada, specifically aimed at exploring and addressing questions of collaboration between academia and Indigenous communities, in the development of a suite of interventions to prevent FASD. Morton, Ninomiya & Pollock (2017) analyse a community driven research project into FASD prevention as a method of exploring dilemmas and strategies for implementing Indigenous research guidelines. They provide a strongly nuanced interrogation of the positionality of the researchers relative to the community partners, and a discussion of the challenges of applying best practice Indigenous research principles when the aims of the Indigenous community themselves are in conflict with those principles, or when academic and community priorities are at odds with one another.

In the USA, Montag et al (2017) describe the iterative modification process for the adaptation of an intervention to reduce risky alcohol consumption among Indigenous women of childbearing age, finding that iteration was required for cultural congruence. This subset of papers also includes two descriptions of the protocols used to adapt the ‘CHOICES’ programme for Indigenous communities—‘CHOICES’ is outlined in more detail under the interventions category. In Australia, Reid et al (2021) found that collaboration in the form of co-design with community is vital to support the adaptation of appropriate evidence-based practices for neurodevelopmental assessment services to suit local context. This was the result of a workshop for community members (n=87) in a remote north-west Queensland community. Finlay-Jones (2020) reports on the priorities identified during two community surveys (responses: n=146 and n=45) and a consensus workshop with 21 community members. The authors recommend a national network of researchers to progress research to address community priorities. These priorities were organised into three categories—understanding influences on prenatal alcohol exposure, preventing prenatal alcohol exposure, promoting maternal and child health, and improving FASD diagnosis and management.

### Interventions to reduce alcohol-exposed pregnancies

This is the largest subset of papers (n=13) included in this review, including one randomised controlled trial, with the remainder broadly defined as qualitative evaluations. Many of the studies (n=5) describe the outcomes from adaptations of the ‘CHOICES’ intervention for different Indigenous communities. ‘CHOICES’ is an intervention funded by the Centres for Disease Control and Prevention in the United States for women who are not pregnant, but could become pregnant, and who are drinking alcohol at risky levels. The intervention uses motivational inter-
viewing combined with contraceptive advice.\textsuperscript{41} A randomised controlled trial found that 69% of women in the intervention group reduced their risk of an alcohol-exposed pregnancy compared to 54% in the control group.\textsuperscript{44} The intervention has been adapted for several Indigenous populations.\textsuperscript{45–48}

Masis et al (1991) describe a programme funded by the Indian Health Service targeted at prevention of alcohol-exposed pregnancy which incorporates training clinicians and members of the community, baseline screening of children suspected to have been alcohol-exposed in pregnancy, and alcohol consumption screening of pregnant women in prenatal clinics.\textsuperscript{49} This programme had high acceptance ratings among Indigenous women participants (n=39), and contributed positively to alcohol consumption reduction and increased awareness of the risks of alcohol-exposed pregnancy. Montag et al (2015) assess the efficacy of a culturally targeted screening, brief intervention and referral to treatment intervention when compared with assessment for risky drinking alone among Indigenous women of child-bearing age (n=247). The authors found that participation in assessment alone was equivalent to being randomised into the intervention group, with respect to reduced risky drinking.\textsuperscript{50} A 2012 paper by Hanson et al describes the development of a media campaign on fetal alcohol spectrum disorders, which was intended to be culturally and linguistically appropriate for the Indigenous communities in the Northern Plains, USA.\textsuperscript{51} The success of this media campaign was attributed to the strong community engagement in the development of the campaign and the design of the dissemination strategy. The evaluation suggests the campaign increased knowledge, but it was not possible to identify a corresponding decrease in harmful alcohol use.

In May et al (2008), the same Indigenous Northern Plains community was the setting for research into the efficacy of enhanced case management to prevent alcohol use in pregnancy among Indigenous women (n=131).\textsuperscript{52} Data were collected at six-month intervals from six to 72 months after enrolment, showing reduced alcohol use overall and reduced binge drinking. Seventy percent of women who were not pregnant at the time of follow-up were protected from having a child with FASD by not drinking, using birth control, or both. Other outcomes measured included pregnancies which progressed to normal deliveries (76%) and positive resolution of issues relating to custody disputes, the criminal justice system, and education. A 2018 study authored by Pei et al reports on the initial findings of relational, trauma-informed and community-based approaches for programmes aimed at the prevention of future alcohol and drug-exposed births.\textsuperscript{53} The authors found that the intervention, delivered through the Parent-Child Assistance Program (PCAP) via mentors providing three-year home visitation services, was effective and well-suited for use in Indigenous communities. May et al (1989) describe a successful intervention aimed at providing Indigenous communities in the USA with the knowledge, skills and strategies to initiate prevention measures.\textsuperscript{54} From 2011, Bridge reports on the decrease in alcohol consumption among pregnant Indigenous women in the Ord Valley in Australia, following FASD education delivered via antenatal clinics and educational sessions delivered to the wider community.\textsuperscript{55} Ma et al (1998) describe a model prevention program to improve Indigenous adolescents’ knowledge of FASD, which demonstrated knowledge change at a greater magnitude than attitude change.\textsuperscript{56}

Two systematic reviews from the USA on this topic by Montag et al (2012) and Symons et al (2018) were identified during the course of our research, and these were consulted to inform the discussion but were not included in the analysis due to the study design being part of our exclusion criteria.\textsuperscript{57,58}

Alcohol use in Indigenous communities in relation to the risk of FASD

These studies (n=5) describe perceptions of alcohol use among Indigenous peoples.\textsuperscript{59–63} In their survey of 1,363 pregnant women, Doherty et al (2019) found that Aboriginal women were more likely to be questioned about their alcohol use, and to receive advice and/or referrals to appropriate services than non-Indigenous pregnant women in Australia.\textsuperscript{59} Gibson, McCarthy & Muggli (2020) interviewed 14 pregnant Aboriginal women and 14 pregnant non-Indigenous Australian women, exploring influences on their decision making with respect to alcohol use during pregnancy, finding that women understood alcohol consumption was harmful in pregnancy, but that they were generally unable to describe the nature of that harm. Non-Indigenous women were more likely to prioritise individual choice in their decision making to continue consuming alcohol during pregnancy, while Aboriginal women filtered their decision making through their experiences of
having seen the harm from alcohol use in pregnancy in their communities, and the perception of responsibility for the baby growing inside them. The authors noted the impact of a woman’s social environment on her ability to abstain while pregnant, and the disproportionate adverse circumstances experienced by Aboriginal women compared with non-Indigenous women.⁴⁰

Hanson & Jensen (2015) identified the important influence of positive social support in preventing alcohol-exposed pregnancies in Indigenous communities in the USA, through interviews with 58 Indigenous adults, emphasising the cultural and historic significance of collectivism. This support often came from elder female relatives, such as grandmothers and “aunties” who were identified by participants as having a special role in connecting Indigenous youth and young adults with culture and appropriate behaviours, influencing decisions such as remaining abstinent from alcohol while pregnant. Culturally relevant group activities such as beading, sewing, sweats and community powwows were suggested by participants as important to include in the design of any prevention programme.⁴¹ Ma et al (1998) surveyed Indigenous sixth, seventh and eighth grade school children about attitudes towards, and awareness of Foetal Alcohol Syndrome (FAS was the umbrella term at the time) risk factors and prevention strategies. The study revealed limited prevention programmes and a reliance on immediate family and peers as the most important influences on these issues.⁴² Plaisier (1989) noted in a study of 29 Indigenous women and six Indigenous health workers in the USA that Indigenous health workers were a positive and effective source of advice about alcohol consumption in pregnancy for the women in their communities, but that the lack of resources limited their ability to reach all Indigenous women who might be at risk for alcohol-exposed pregnancies.⁴³

**Interventions to improve the experience of Indigenous people with FASD and their families**

Four papers described interventions targeted at improving the experience of people with FASD. Keightley et al (2018) describe a qualitative evaluation of a theatre-based intervention using artistic media to facilitate social and emotional awareness in Indigenous youth from Canada (n=3) with FASD. The focus on Indigenous youth was incidental, as the inclusion criteria specified only that participants have a confirmed diagnosis of FASD, are in the age range between 9–14 years old, and experience significant difficulties in social functioning. Culturally appropriate activities were incorporated into the intervention retrospectively once the ethnicities of the participants were confirmed, with local First Nations community partners providing guidance and facilitation of activities where required. There was no discussion of the researchers’ positioning or critical reflection on the socio-cultural impacts of ethnicity on the experience of Indigenous youth with FASD, apart from an acknowledgement of the importance of developing interventions that respect “unique cultural factors” for Indigenous youth with FASD.⁴⁴ Wagner et al (2017, 2020) report on the school-based intervention described in the protocols section, finding that that adapted, culturally appropriate programme improved self-regulation among the participants, and that the intervention was congruent with participants’ cultural expectations.⁴⁵,⁴⁶

Hamilton (2020) discusses the importance of addressing cultural differences, as well as stigma, guilt and shame in a paper looking into Aboriginal and non-Aboriginal caregiver responses to neurodevelopmental disability diagnoses.⁴⁷ In the sole paper included from Aotearoa, Crawford et al (2020) report on the findings from a study into the experience of 39 (82% Māori) children and their families with FASD compared with a comparison group with respect to adaptive functioning.⁴⁸ This study aimed to understand impairments underlying poor adaptive functioning of children with FASD within their cultural context, with the aim of better understanding their needs and identifying potential targets for early intervention. This study was notable for its prioritisation of power-sharing with Indigenous participants and researchers in its stated aims and methods.

**Discussion**

The literature pertaining to FASD in Indigenous communities is dominated by a focus on prevention of alcohol-exposed pregnancies. While understandable from a population health prevention level, this highlights a concerning gap in the literature of the investigation and development of appropriate interventions to support Indigenous people with FASD and their families.

There are three outcomes from this review which are relevant to the research team’s broader project looking into the experience of whānau Māori with FASD. First, the findings regarding the methods of engagement with Indigenous commu-
nities which were most successful and empowering for the communities. Second, the evidence for FASD prevention initiatives in Indigenous communities. Finally, the concordance with assumptions made by this research team with respect to our study background, aims and principles.

Overwhelmingly, and unsurprisingly, engagement between community and researchers was most successful in the instances where the community had first determined there was a need to act on FASD, and where community leaders were engaged in the research from the beginning. The researchers who described the most success in terms of the depth of their relationships with community members, and respect for cultural imperatives, were those who experienced the most success in the outcomes of their research. While it was encouraging that a majority of the papers included outlined the importance and nature of their relationships with local Indigenous leaders, there was a notable lack of Indigenous researchers involved from the academic and clinical perspective. There was a spectrum of engagement with the principles outlined in the ‘CONSIDER’ statement against which the research was assessed for cultural safety; a small number of researchers explicitly stated the precise nature of their engagement and governance/power-sharing arrangements with community, though most did not link their relationships with community to sovereignty in research efforts, or control of the research process. Few researchers reflected on their positionality as researchers. The vast majority avoided deficit blaming and described a broad understanding of the social determinants linked to alcohol use among Indigenous communities.

The evidence for successful FASD prevention initiatives in Indigenous communities is linked strongly to the quality of engagement between those researching/designing the prevention initiatives and the community itself. As outlined above, outcomes were most emphatically positive where Indigenous community members were deeply involved in the instigation, development and dissemination of interventions. In some cases, this involved training members of the community—both clinical and non-clinical—in delivering an intervention, for example mentoring for Indigenous women of child-bearing age who were at risk of alcohol related harm. Knowledge gain was an outcome frequently measured among the studies designed to improve understanding of FASD, and researchers have reported success at improving community awareness of FASD.

Researchers consistently expressed frustration at the lack of health system action in terms of providing consistent, relevant information for Indigenous communities, and indifference from the alcoholic beverages industry to the harm caused. Behavioural change and other outcomes such as improvements in education for Indigenous women of child-bearing age, resolution of issues with custody arrangements and the justice system were less commonly assessed by researchers. The long-term follow-up required to assess these outcomes, and the frequently noted lack of funding for research into FASD was commented upon by several researchers who expressed a desire to extend their research into these outcomes but who were unable to due to lack of resources. One study mentioned the increased risk of death among women with severely FAS affected children, with 75% of women in a particular study deceased six years after giving birth to a child with severe FAS. This statistic highlights the broad and significant impact of FASD from those affected—to their parents and wider communities—and the need to think beyond linear prevention efforts to interventions which can benefit entire whānau.

This review specifically sought to evaluate the existing evidence regarding the experience of Indigenous people with FASD. Accordingly, prevalence studies and other epidemiological research, which did not also explore the experience of Indigenous people, were excluded. However, some of the papers which were excluded were consulted in order to confirm concordance with assumptions made by this research team with respect to our study background, aims and principles. From this part of the review, we found that barriers to a FASD diagnosis are experienced by Indigenous people globally, that these barriers directly impact the quality of prevalence data and prevent adequate funding and resource allocation. There appears to be little change in this respect from the oldest paper included, from 1989, to the present. Of the papers included in this review, there is broad agreement among the authors that the prevalence rates of FASD are likely to be higher for Indigenous peoples than non-Indigenous, which has also been discussed in our introduction. Frustratingly, without increased workforce capacity to diagnose those suspected to have FASD, services cannot be designed or allocated and people with FASD and their whānau, rendering them invisible to our health system.

Interventions to support whānau Māori who are experiencing FASD must be delivered across
prevention and management/support services and initiatives. Research must be conducted in accordance with the ‘CONSIDER’ statement, and must be based in Kaupapa Māori methodology. Whilst there is international evidence of FASD prevention initiatives in Indigenous communities, less is known about interventions aimed at supporting Indigenous people who experience FASD. However, there is little evidence in either prevention or support interventions in Aotearoa. In Aotearoa, interventions must address inequities caused by colonised systems. It is recommended that Kaupapa Māori research is urgently required in FASD prevention and services to ensure whānau Māori receive equitable and effective services and support.

Conclusion

The existing literature on FASD in Indigenous communities internationally is heavily skewed towards the development, implementation and evaluation of interventions to reduce alcohol-exposed pregnancies. There is also a focus on studies which aim to understand and describe the variables which lead to alcohol use among Indigenous communities, and to the relationship with alcohol use in the perinatal period. A number of protocols/method development for FASD-related interventions in Indigenous communities have been published, but few have been fully reported upon or evaluated.

There was a single published study from Aotearoa in the scientific literature into the experience of Māori with FASD which met our inclusion criteria. This lack of research contributes to ignorance about the extent of the impact of FASD on Indigenous communities, which in turn hinders efforts to design interventions to reduce the incidence of FASD, and to support those with FASD and their families. In practice, there are opportunities to apply the findings of this review to our own research, at the same time acknowledging that while there are similarities among Indigenous communities globally, we cannot assume generalisability from one Indigenous culture to another, and Māori have unique strengths and challenges in responding to a diagnosis of FASD in the whānau, which are specific to the Aotearoa socio-cultural context.
COMPETING INTERESTS
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