


# Identifying strategic research priorities of stakeholders for fetal alcohol spectrum disorder in Aotearoa

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## ABSTRACT

Fetal alcohol spectrum disorder (FASD) is under-researched in Aotearoa New Zealand (NZ). There is a pressing need to establish a research agenda that is aligned with Te Tiriti o Waitangi and relevant to both stakeholders and end-users, to improve the outcomes and support for people affected by FASD. We conducted hui and interviews to identify consensus on research priorities among key stakeholders for FASD in NZ, including whānau and caregivers, health care workers, educators, academics and clinicians. The hui focused on three main topics: prevention; assessment and diagnosis; and intervention. Hui transcripts were analysed to identify priorities. Once the research questions or priorities were identified we sent a follow-up survey to participants asking them to rate each research question on their importance and relevance. Four hui and ten interviews ( $n = 52$ ) were conducted across the North Island between April and July 2021. We identified 20 research priorities from the hui and interviews. Based on participant feedback ( $n = 18$ ), the most important and relevant priority was: 'What can we do to encourage success for people with FASD throughout their lives?'. The priorities identified through the workshops and hui will be valuable in guiding future research and policies relating to FASD.

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## Introduction

Fetal alcohol spectrum disorder (FASD) describes the neurological and physical effects of prenatal exposure to alcohol (PAE; Brown et al. 2019). FASD is a lifelong disability that impacts many areas of development including physical and cognitive development (Cook et al. 2016). Without proper support, individuals with FASD face challenges across multiple domains, including long-term health, education, and social outcomes for the affected individual, their families and whānau, and society (O'Leary et al. 2013; Rangmar et al. 2015).

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Global prevalence of FASD is estimated at 8 in 1000 (Lange et al. 2017); however, the prevalence of FASD is likely higher in countries and populations where regular or heavy alcohol consumption is a feature of mainstream culture (Lange et al. 2017; Popova et al. 2017), for example, the estimated prevalence of FASD in South Africa is 111.1 per 1000 (95% CI 71.1–158.4), but populations with higher drinking norms (such as rural communities) have reported estimates as high as 259 per 1000 (May et al. 2016).

In Aotearoa New Zealand (NZ), approximately 80% of adults report having consumed alcohol in the past 12 months (Ministry of Health 2021). Population surveys have repeatedly reported relatively high rates of alcohol use in pregnancy. Analysis of the Growing Up in New Zealand cohort ( $n = 6822$ ) found 71% of women reported drinking before pregnancy or becoming aware of their pregnancy, 23% during the first trimester, and 13% after the first trimester (Rossen et al. 2018). The odds of consuming alcohol during pregnancy were higher for women who were Pākehā or Māori. There is extensive evidence of the teratogenic effects of prenatal alcohol exposure that can lead to serious changes in neurodevelopment and a wide range of congenital abnormalities (Chu et al. 2022). It is therefore crucial to understand the factors that cause or encourage alcohol consumption during pregnancy.

The Ministry of Health estimate 3 to 5 in every 100 live births are affected by alcohol (Ministry of Health 2022). The estimated annual cost of FASD to the country, including the use of services and support, is \$690 million: around \$15,000 for every individual with FASD (Easton et al. 2016), however, this is likely an underestimate as the prevalence of FASD is unknown and certain costs, such as incarceration, were not considered (McCormack et al. 2021). Early identification of FASD (Streissguth et al. 2004) and multidisciplinary interventions (Reid et al. 2015) is a protective factor for adverse life outcomes, including school disruption, justice involvement and confinement, and is essential to ensure the needs of individuals with FASD and their families and whānau are appropriately managed and supported (Petrenko et al. 2014). Accessing a diagnosis of FASD in NZ is a long and difficult process, due to the lack of trained health professionals and resources to support diagnosis. Therefore, people are often left unsupported on long waiting lists, up to four years, to be assessed for FASD (Health and Safety Developments 2015). Identifying effective interventions for FASD can amount to significant long-term benefits for the individual, as well as improved societal outcomes in terms of economic productivity, health, education, and criminal justice (Hopkins et al. 2008) and addressing wider socio-cultural determinants of health and health inequity. However, there are very few FASD informed interventions available in NZ. There is a pressing need to develop evidence-based interventions to address PAE and support those living with FASD, policies and practices that address the needs, and support diagnosis, of individuals with FASD, and those of their families, whānau and caregivers.

### ***Establishing research priorities***

Increasingly researchers are engaging in activities that involve non-research stakeholders in the process of identifying and prioritising research needs (Bryant et al. 2014; Razavi et al. 2020). Importantly, engaging stakeholders in priority setting research can improve uptake and implementation (Bryant et al. 2014) and therefore achieve better outcomes for the community. A wide variety of approaches have been used in priority setting activities, including Delphi studies (Bryant et al. 2014), the Nominal Group

technique (Scott and Deadrick 1982), priority setting partnership (Cowan 2010), focus groups, workshops, and stakeholder surveys and questionnaires (Bryant et al. 2014).

In NZ, Māori are the tangata whenua (indigenous people of NZ). Colonisation of NZ resulted in significant trauma, that has ongoing effects for health and equitable outcomes for Māori (Barnes and McCreanor 2019). The Te Tiriti o Waitangi (The Treaty of Waitangi) is an agreement between Māori and Pākehā established following colonisation that ensures the protection, partnership and equal treatment of Māori (Walker et al. 2006; Orange 2012). The treaty ensures the efforts of equity in health outcomes (Walker et al. 2006), yet Māori often experience poorer health outcomes. For Māori hauora (health) is holistic incorporating the balance of taha tinana (physical health), taha hinen-garo (mental health), taha whānau (family), and taha wairua (spiritual dimension; Durie 1985). Whānau (family) incorporates extended family members that is important to well-being. Connection with whenua (land) is also critical for wellbeing and haurora. Any research into FASD in NZ needs to align with Te Tiriti o Waitangi to ensure that Māori experiences are heard, protected, and included in the research process. Importantly, the research priorities identified need to reflect the needs of Māori with FASD and ensure they are relevant and useful to both stakeholders and end-users and to inform the practices of wider systems that may be ill-equipped to meet these needs. These research priorities need to ensure equitable research focusing on all those with lived experience of FASD.

Establishing research priorities from those with experience or expertise of FASD can help guide researchers to addressing key questions and make a difference for those affected by FASD. However, few attempts have been made to identify research priorities for FASD stakeholders. For example, Australian researchers engaged the general population, those working in organisations that support people with FASD and caregivers of people living with FASD, in online surveys and workshops, to identify research priorities (Finlay-Jones et al. 2020). This approach, however, primarily drew on those interested in FASD prevention, rather than supporting people with FASD and their families.

### ***FASD action plan 2016***

In NZ in 2016 the FASD Working Group was established to create a national action plan about FASD. The FASD Working Group consisted of representatives from government agencies (such as Ministry of Health, Education, and Justice) and submissions were made by those affected by FASD and experts in the field (FASD Working Group 2016). The FASD action plan recognises that there is a paucity of evidence-based interventions that provide support for individuals with FASD and reduce the incidence of FASD. Despite highlighting the weaknesses in the underlying evidence base, the action plan largely neglected the need for evidence-based treatments of FASD fit for NZ, and a broad evidence base to inform policy and practices. Instead, the plan prioritised prevention through alcohol and drug support, sexual and reproductive health care, and education. Crucially, the plan also lacked a focus on Te Tiriti o Waitangi and lacked representation by stakeholders affected by FASD, particularly Māori and whānau of people with FASD, and its focus on those already affected by FASD.

Although, there is no clear information available on the prevalence or outcomes of FASD for Māori, the burden of FASD could be greater for Māori due to the ongoing

effects of colonisation (Ministry of Health 2019). International studies suggest indigenous people have higher rates of FASD (Mutch et al. 2015; Palmetter et al. 2021), however, there are limitations in sampling methods that could bias these studies. A recent review of interventions for those with FASD found very few if any had a Kaupapa Māori approach or consider Te Ao Māori and the interventions that exist largely focus on prevention (Espiner et al. 2022). These findings highlight the need to ensure that priorities for FASD research in NZ include the voices and experiences of Māori.

In order to address gaps in the FASD Action Plan 2016, we conducted a series of multi-stakeholder hui to identify health delivery research priorities for FASD in NZ that are important, in the first instance, to individuals and whānau affected by FASD, and also to health workers, educators, clinicians, advocacy groups, and researchers. This is the first and most important step in addressing the evidence gap in FASD research in Aotearoa NZ that considers the unique policy environment and the place of Te Tiriti o Waitangi in ensuring health equity.

## Method

To ensure meaningful stakeholder engagement, particularly the engagement with Māori whānau, it was important that we approached the priority setting activity in a way that incorporated Kaupapa Māori research methods, Āta – Growing respectful relationships (Pohatu 2004), Experience Based Co-Design (Green et al. 2020), and Kaupapa Māori Action Research (Kerr et al. 2010), and ways of doing things. Therefore, we conducted hui and interviews to explore questions or concerns that different stakeholder groups had about PAE and FASD. These hui were led by a Māori research and clinician. Ethics approval for the study was obtained from the Health and Disability Ethics Committee (20/STH/230).

To ensure that Māori knowledge and ways of understanding the world were at the forefront of our project, we drew on the expertise of Māori researchers and organisations involved in the research with experience conducting research and supporting Māori health in NZ. This partnership helped establish key relationships between researchers and with key FASD stakeholders in the community. We ensured the study design and interpretation of the findings was co-led by Māori and non-Māori researchers, to ensure proper tikanga (traditions and values) and customs was respected (Walker et al. 2006). Māori researchers led data analysis and interpretation, to ensure the experiences of Māori were understood by all those involved in the research (Walker et al. 2006).

## Participants

We recruited participants from the following stakeholder groups:

- Whānau and caregivers of individuals living with FASD or suspected FASD;
- Health workers, support workers, social workers, and other front-facing staff involved in the care of people with FASD;
- Teachers;

- Clinicians and researchers with an interest in FASD or working with people with FASD.

Participants were recruited through advocacy and provider networks using purposive sampling. We used targeted recruitment methods to ensure that Māori were represented across stakeholder groups and oversampled whānau and caregivers to ensure their views were captured. Hui location were selected based on the presence of Māori FASD leaders within the communities and the established relationships to organisations and individuals who might be interested in attending the hui. Information about the hui were circulated by FASD leaders through their networks and via word of mouth. Whānau and caregivers of people with FASD were recruited through established networks and organisations such as FASD-CAN and Hāpai te Hauora. Academics and clinician that were experts or had significant experience with FASD were approached directly by the research team via email. Unlike previous research priority setting for FASD (Finlay-Jones et al. 2020) we did not include the general population to amplify the voices of those affected by FASD.

### ***Inclusion criteria***

Participants were eligible to participate if they were:

- Aged 18 years or over;
- Able to speak and read English;
- Reside in New Zealand; and
- Belong to one or more of the stakeholder groups described above.

Whānau and caregivers were eligible if they were personally involved in the care of an individual who has been diagnosed with FASD or who is suspected of having FASD. A *koha* (reimbursement) of \$50 per person was provided to participants in the whānau and caregivers hui as a recognition of their time and travel costs to attend the session.

### ***Baseline data***

Baseline information was collected prior to the hui via a baseline survey. The baseline information included socio-demographic information (age, gender, ethnicity, occupation); stakeholder group, previous experience with FASD; and what questions they thought it was important for the research agenda to address.

### ***Hui***

Hui and workshops were facilitated by a Māori researcher and clinician and caregiver of children with FASD who was experienced in collaborative research (AM). We opened the hui with a *mihi* (formal welcome), *karakia* (chant/prayer), and *whakawhanungatanga* (relationship building) amongst the group. The facilitator introduced the purpose of the hui and the research and gave an overview of FASD.

The hui was broken into three main topics, with a workshop presentation throughout the hui on Te Whare o Oro (a Māori neurological-based model for understanding FASD). The three main topics were: prevention; assessment and diagnosis; and

intervention. Within these topics we also explored the lifelong prognosis of FASD and the needs of Māori. The discussion followed a semi-structured interview guide administered by the facilitator. At the end of the hui the facilitator closed the session with a karakia.

### **Interviews**

Interviews were offered to participants that were unable to attend the hui. Interviews were conducted by a Pākehā (New Zealand European) interviewer (JM) and followed the same facilitation guide used in the hui. Interviews were conducted one-on-one or as group interviews.

### **Analysis**

Hui and key informant interviews were transcribed verbatim and analysed qualitatively by two members of the research team (JD, JM). One of the researchers was a Māori woman and the other was a Pākehā woman. The researchers familiarised and coded the data individually, identifying issues or concerns raised in the focus groups and interviews. Potential themes were constructed from the codes, which were then reviewed collaboratively by the researchers. Together the researchers grouped and redefined potential themes, narrowing down the potential themes and framing these themes to research questions (Terry et al. 2017).

Baseline demographics were summarised and presented in a table. For continuous data, variables were summarised as mean (standard deviation) and median (range), while categorical variables were summarised as frequency and percentage.

### **Validation**

Once the research questions or priorities were identified we sent a follow-up survey to the hui and interview participants via email. Participants were asked to rate each research question on their importance (from not important to very important) and relevance (from not relevant to very relevant). To determine the top priorities, we summed the scores across participant on importance and relevance (with 1 representing not important/relevant and 5 very important/ relevant). All missing data was treated as not important (i.e. 1). Participants were also asked to select their five most important questions from the list provided, and rate how closely the research priorities matched their own priorities.

### **Results**

Four hui were conducted across the North Island between 14 April 2021 and 16 July 2021. The number of participants at each hui ranged from 8 to 18. We also conducted interviews with ten participants who could not attend the hui.

Table 1 describes the participants by stakeholder group. Two of the hui mixed different stakeholder groups, while one hui only included academics and clinicians and the other only included whānau.

**Table 1.** Hui and interview participant characteristics.

	Research/clinical <i>n</i> = 10	Whānau <i>n</i> = 20	Education <i>n</i> = 6	Health <i>n</i> = 16
<b>Age (<i>M</i>, range)</b>	50.9 (30–69)	53.0 (25–71)	48.1 (41–69)	47.25 (23–62)
<b>Gender</b>				
Female	8	17	3	15
Male	2	6	3	1
<b>Ethnicity</b>				
Māori	2	8	4	11
Pacific	0	3	0	0
Non-Māori, Non-Pacific	8	12	2	5
<b>Education</b>				
Secondary School	–	6	–	–
Trade qualification	–	2	–	–
UG	1	4	1	5
PG	9	9	5	8
Other	–	2	–	3
<b>First Heard FASD</b>				
In the last year	–	2	–	1
2–3 years	–	1	–	3
4–5 years	–	1	1	1
5–10 years	3	6	3	4
10 or more years	7	10	2	7

Many participants indicated that they belonged to more than one of the stakeholder groups identified. For example, health workers reported having relatives with FASD and caregivers worked in the health or education sector. None of the whānau participating in the study were biological parents. Whānau included foster and adoptive parents, whāngai (informal adoption within whānau) and extended family member (e.g. grandparents).

### Research priorities

We identified 20 research priorities from the hui and interviews. The priorities were classified into the following categories: systems (i.e. relating to government or social systems), workforce/community (i.e. relating to the FASD workforce or broader community), and whānau (i.e. relating to the individual and whānau living with FASD), as per Table 2.

### Whānau

Priorities were categorised under whānau where the issue raised directly affected individuals living with FASD or whānau of individuals living with FASD. Whānau priorities also included issues that related to preventing FASD, for example, understanding what factors other than alcohol exposure contribute to FASD and how to support whānau decision making around alcohol use in pregnancy. Other priorities included a better understanding of FASD across the lifespan, especially around the challenges of adults with FASD, the impact of diagnosis of FASD on the whānau and what whānau need to support individuals with FASD.

### Workforce and community

Priorities were categorised under workforce/community if the issues raised primarily affected the workforce and community. Priorities could be further subdivided in this

**Table 2.** Research priorities classification.

Systems	Workforce/community	Whānau
What can we do to encourage success for people with FASD throughout their lives?	What does the frontline workforce know about FASD?	What are the factors that determine the consequences of parental alcohol exposure?
What is the cost of FASD to society?	What does the frontline workforce/community need to support people through alcohol-free pregnancy?	How does the diagnosis of FASD impact whānau of people with FASD?
What is the prevalence of FASD in the general population and specific populations?	What support can we provided for mothers during pregnancy to support with alcohol dependence?	What factors can be used to support whānau decision making around alcohol use in pregnancy?
Incorporation of FASD information into school health education	Identifying effective, evidence-based intervention for FASD	What does FASD look like across the lifespan?
Establishing consistent messaging and pathways to diagnosis and intervention for people with FASD.	What Kaupapa Māori approaches are existing (within health, education and social sectors) that can be applied to FASD intervention and assessment	What factors can be used to support whānau of people with FASD in their role as carers?
Exploring the inception of alcohol into Māori culture.	Understanding the attitudes towards FASD	
Understanding how the NZ drinking culture and alcohol environment contributes to FASD.	Understanding the attitudes towards prenatal alcohol exposure	
What are the barriers to recognition and support for people with FASD?		

category as relating to prevention of FASD and alcohol exposed pregnancies or relating to supporting individuals with FASD. Workforce/community interventions relating to prevention included empowering the workforce and community to support people through alcohol-free pregnancies and how to support women with alcohol dependence through pregnancy and understanding attitudes and behaviours towards prenatal alcohol exposure. Workforce/community interventions relating to support included understanding the needs of the frontline workforce to enable them to support people with FASD (i.e. knowledge of FASD and barriers to accessing resources), understanding attitudes and behaviours towards FASD, identifying effective, evidence-based interventions for FASD, and exploring Kaupapa Māori approaches to FASD intervention and assessment.

**Systems**

Priorities were categorised under systems if they raised issues that related to government or social systems, such as social norms and health and education systems. These priorities included issues around culture, such as understanding how the drinking culture and alcohol environment contribute to FASD and exploring historical and contemporary inception of alcohol into Māori culture. Priorities also included quantifying the prevalence of FASD – in the general population or specific populations such as Youth Justice, Corrections, Oranga Tamariki – Ministry for Children, and Alternative Education – and the cost of FASD to society. At the level of governance, priorities included understanding barriers to recognition and support for people with FASD and establishing consistent standards for FASD (for messaging, diagnosis, and referral pathways), as well as incorporating FASD into school curriculum. Finally, participants raised priorities around adverse outcomes and encouraging success for people with FASD throughout their lives.



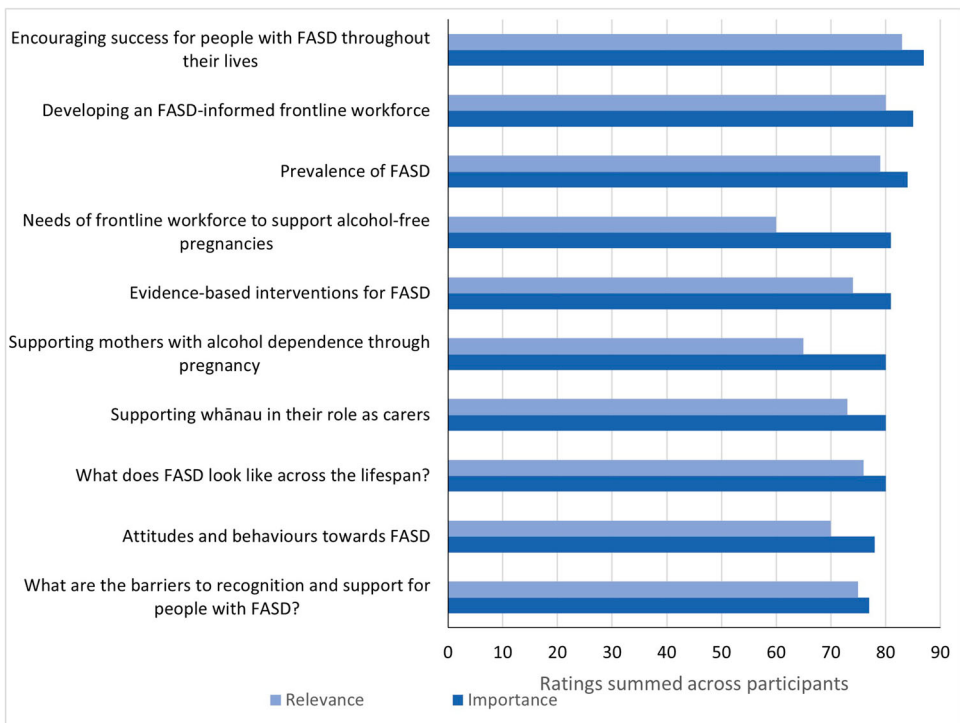
**Feedback on priorities**

Eighteen participants provided feedback on the priorities identified in the hui. The largest group of respondents were Whānau ( $n = 10$ ), followed by academics and clinicians ( $n = 4$ ). The remaining participants were from social work, social services, and health ( $n = 4$ ).

The highest scoring priority on both importance and relevance was ‘What can we do to encourage success for people with FASD throughout their lives?’. All participants rated this priority as Very Important ( $n = 15$ ) or Important ( $n = 3$ ), and the majority rated it as Very Relevant ( $n = 13$ ) or Relevant ( $n = 4$ ).

The ten priorities with the highest sum scores can be found in Figure 1. The items that scored high on both importance and relevance were (i.e. 75 or above in both categories): Encouraging success for people with FASD throughout their lives; Developing an FASD-informed frontline workforce; Prevalence of FASD; What does FASD look like across the lifespan; and Barriers to recognition and support for FASD. Supporting mothers with alcohol dependence and Needs of the frontline workforce to support alcohol-free pregnancies were rated high in importance, but low in relevance. The lowest rated priorities were around factors contributing to FASD and cultural factors contributing to FASD (i.e. NZ drinking culture, alcohol environment, and inception of alcohol to Māori culture).

When asked to select their five most important priorities, the following priorities were endorsed by the most people: Identifying effective, evidence-based intervention for FASD



**Figure 1.** Summed scores for top ten most important priorities rated by participants.

( $n = 12$ ); What can we do to encourage success for people with FASD throughout their lives ( $n = 10$ ); What factors can be used to support whānau of people with FASD in their role as carers ( $n = 8$ ); What are the barriers to recognition and support for people with FASD ( $n = 7$ ); and What is the prevalence of FASD in general and specific populations ( $n = 7$ ). Most participants reported that the priorities matched their own closely or very closely ( $n = 15$ ), with only two participants reporting that the priorities matched their own moderately closely.

Seven participants identified additional priorities that they felt were missing. These included perspectives from whānau and caregivers and those living with FASD; identifying barriers within the systems and redesigning them to better support people affected by FASD; Hauora (health and wellbeing) models to support whānau impacted by prenatal alcohol exposure and FASD; mental health of caregivers and how to provide support to carers; incidences of FASD in relation to adverse outcomes (e.g. suicide, drug addiction, care and protection); understanding of knowledge, attitudes, and behaviours of the education workforce and ensuring they have the knowledge to provide support for children with FASD.

## Discussion

We aimed to identify priorities for stakeholders regarding FASD research in Aotearoa by engaging in hui and a priority setting exercise. We identified 20 issues or priorities for FASD research in Aotearoa which were largely endorsed by our participants. An important priority for our participants was understanding what we can do to encourage success for people with FASD throughout their lives. Participants also emphasised the need for evidence-informed intervention and treatment, as well as the need for information on the prevalence and effects of FASD in order to better inform research and policy.

The main priority of stakeholders reflects the need for strengths-based FASD research. Despite, several studies exploring the lived experiences of people with FASD research mainly focuses on identifying the challenges and difficulties they experience (Domeij et al. 2018). A recent review identified that strengths based research around FASD is scant, however, people with FASD are aware of their FASD and can identify their strengths such as perseverance and a desire to help others (Flannigan et al. 2021). People living with FASD identified that having positive support networks and people was key to success (Flannigan et al. 2021). Yet, very little research has been conducted in line with the stakeholder's priorities of encouraging success. Therefore, future research needs to explore what success looks like for people with FASD and identify ways to foster this success.

Stakeholders identified several research priorities focusing on workforce development, including identifying knowledge gaps and upskilling key professionals to support those with FASD. Professionals working in education, justice and health professions recognise there is a lack of awareness, knowledge and training to support people with FASD (McCormack et al. 2022). Internationally, health and education professionals have some awareness and understanding of FASD, but often feel unprepared to support those living with FASD (Payne et al. 2005; Turner 2006; Passmore et al. 2018). In NZ, professionals working in the justice sector emphasised the need of official diagnosis and support for people with FASD (Gibbs 2022). Yet the lack of knowledge and

professional training made it difficult to support people with FASD, so justice professionals had self-sought training and education opportunities outside their organisations (Gibbs 2022). People with FASD and their caregivers often have negative experiences when accessing support leaving them feeling unsupported (Salmon 2008; Salmon and Buetow 2012). These negative experiences stem from professionals lack of knowledge and recognition of FASD, and the stigmatising beliefs held by professionals (Salmon 2008; Salmon and Buetow 2012). Therefore, several research priorities identified by stakeholders focused on upskilling professionals and improving the support available for those living with FASD.

Exploring the experiences of Māori with PAE and FASD were a key dimension of the study and the unique perspective of Māori were evident in the research priorities identified by participants. In particular, Māori participants highlighted the importance of exploring the inception of alcohol into Māori culture and its role in colonisation, as well as the need to identify existing Kaupapa Māori approaches that could be applied to intervention and assessments of FASD and developing Kaupapa Māori interventions where those were absent. Additionally, Māori participants emphasised the importance of a hauora and whānau approach, rather than interventions that only focus on the individual. These priorities are consistent with previous literature that show a lack of NZ research on FASD with Kaupapa Māori or Te Ao Māori view (Crawford et al. 2020; Espiner et al. 2022). The one study that has explored the effects of FASD with a Te Ao Māori view demonstrate the importance of understanding the effects of FASD within Māori culture and customs (Crawford et al. 2020). Likewise, there is some international evidence to suggest that the ongoing effects of colonisation and mistreatment could contribute to prenatal alcohol consumption in indigenous communities (Johnston and Boyle 2013; Gonzales et al. 2021). Taking such an approach and considering historical factors could improve the understanding and interventions to support Māori with FASD.

Compared to priority setting exercises in Australia, our participants appear to place less emphasis on the determinants of PAE and prevention of PAE. In the Australian FASD priority setting activity the top priorities identified by participants concerned mainly PAE (Finlay-Jones et al. 2020), comparatively our participants focused on understanding and support for those with FASD. This may be due to differences in the processes and makeup of our participants, the Australian study utilised a broader participant group and surveys to identify the initial priorities (Finlay-Jones et al. 2020). Most participants (62%) in Finlay-Jones et al. (2020) reported that they were interested in FASD prevention. By comparison, the majority of our participants (78%) were either supporting someone with FASD (as a caregiver or other whānau) or worked for organisations providing services or support for people with FASD (such as Ministry of Education, mental health and community organisations), the remaining were experts in FASD research. While our participants shared a lot of thoughts about prevention of FASD in the hui, priorities relating to prevention were not given high importance or relevance in the validation survey. Instead, our participants tended to focus on improving outcomes for individuals with FASD and their whānau. A similar exercise conducted in Canada (Flannigan 2016) focused on four main areas: FASD Prevention in the preconception period, Interventions for children and youth, Youth and adults with FASD, and Knowledge translation.

Our study has established key research priorities of stakeholders with lived experience of FASD in NZ. These priorities provide direction and guidance for researchers on the

key issues or concerns that should be focused on, such as the strengths, success and developing interventions and supports for those living with FASD. The research priorities may not capture the view of all stakeholders with lived experience of FASD in NZ, therefore, a larger validation study could be captured to provide insight on the relative importance of these factors. Nevertheless, these research priorities show a need for future Kaupapa Māori research and research based in NZ that responds to the needs and can help provide support to those living with FASD in NZ.

### ***Strengths and limitations***

Although generality of findings may be limited by the small sample size, we were able to generate a depth of information from our participants using the hui and workshop approach. The workshop participants were from a range of roles – from caregiving for people living with FASD to those from service agencies providing care and support – and participants provided diverse perspectives on the research priorities for FASD. We deliberately oversampled caregivers and whānau ensuring that the perspectives of those with lived experience of FASD are a part of the research agenda. The workshops were facilitated by a Māori practitioner and carer, ensuring that they had relevance to Māori and the feedback elicited was relevant to the political, social, and cultural context of NZ.

Despite several attempts to engage participants in the validation exercise, we were unable to reach the majority of participants who attended the hui. The low response rate may be due to a number of factors, including the timing of the survey (several months after the initial hui) and significant contribution to research and burdens of the FASD community in NZ, such as time commitments and lengthy questionnaires. Using different methods of communication, such as telephone calls, may have improved participants responses. Due to the low response rate from other groups, the voices of carers were amplified in the feedback, and priorities may not fully reflect the priorities of the FASD community as a whole. An additional Delphi process may be required to expand the reach of the survey and develop a prioritised agenda for FASD research reflecting the views of a large group of stakeholders. The criteria used to prioritise research may be expanded in further research to take into account other factors including feasibility, urgency, equity, and impact of the identified priorities, and the extent to which the priorities advance the health needs of Māori. Likewise, despite recruitment efforts our participants did not include biological caregivers, and our participants were mainly female and resided in the North Island of New Zealand. Participation of biological caregivers in FASD research is often low, due to the stigma or shame associated with FASD (Corrigan et al. 2017), and the potential that FASD often goes undiagnosed. Therefore, the research priorities may not reflect the view of all stakeholders with lived experience of FASD in New Zealand, but rather those included in this sample. Biological parents could provide further priorities around the prevention of PAE, however, it is difficult to recruit biological parents for research. Despite this, our findings are an important first step in establishing the key research issues for those with lived experiences of FASD and further research should ensure the priorities are consistent across other stakeholders in New Zealand.

## Conclusion

Our findings are a complement to the FASD Action Plan developed by the FASD Working Group (FASD Working Group 2016). The priorities identified through the workshops and hui will be valuable in guiding future research and policies relating to FASD. Further research is needed to evaluate the priorities so they can be used to establish a 5–10 year FASD research agenda that is responsive to the needs of those affected or working with people living with FASD. NZ needs more research focusing on FASD for Māori within a Te Ao Māori view.

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