

# The Lililwan Project: study protocol for a population-based active case ascertainment study of the prevalence of fetal alcohol spectrum disorders (FASD) in remote Australian Aboriginal communities

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## ABSTRACT

**Introduction:** Anecdotal reports suggest that high-risk drinking in pregnancy is common in some remote Australian communities. Alcohol is teratogenic and may cause a range of lifelong conditions termed ‘fetal alcohol spectrum disorders’ (FASD). Australia has few diagnostic services for FASD, and prevalence of these neurodevelopmental disorders remains unknown. In 2009, Aboriginal leaders in the remote Fitzroy Valley in North Western Australia identified FASD as a community priority and initiated the Lililwan<sup>1</sup> Project in partnership with leading research organisations. This project will 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.

**Methods and analysis:** This is a population-based active case ascertainment study of all children born in 2002 and 2003 and residing in the Fitzroy Valley. Participants will be identified from the Fitzroy Valley Population Project and Communicare databases. Parents/carers will be interviewed using a standardised diagnostic questionnaire modified for local language and cultural requirements to determine the demographics, antenatal exposures, birth outcomes, education and psychosocial status of each child. A comprehensive interdisciplinary health and neurodevelopmental assessment will be performed using tests and operational definitions adapted for the local context. Internationally recognised diagnostic criteria will be applied to determine FASD prevalence. Relationships between pregnancy exposures and early life trauma, neurodevelopmental, health and education outcomes

<sup>1</sup>Lililwan is a Kimberley Kriol word meaning ‘all of the little ones’ (Kimberley Kriol is a local language spoken by many Aboriginal people in the Fitzroy Valley).

## ARTICLE SUMMARY

### Article focus

- To establish the need for prevalence data on FASD in remote Australia and for improved awareness and diagnosis of FASD.
- To describe the protocol used in Australia’s first population-based study of FASD prevalence using active case ascertainment in remote Aboriginal communities.
- To demonstrate a process of community consultation and clinical research that respects the priorities, language and culture of Aboriginal communities.

### Key messages

- Accurate prevalence data on FASD and other health and developmental outcomes will inform prevention, service provision and policy in child health, education and justice.
- This research will provide immediate and direct benefits to participants and the broader community, including a feasible and transferable model of FASD diagnosis and a model for culturally responsive research with Aboriginal communities.

will be evaluated using regression analysis. Results will be reported according to STROBE guidelines for observational studies.

**Ethics and dissemination:** Ethics approval has been granted by the University of Sydney Human Research Ethics Committee, the Western Australian Aboriginal Health Information and Ethics Committee, the Western Australian Country Health Service Board Research Ethics Committee and the Kimberley Aboriginal Health Planning Forum Research Sub-committee. Results will

ARTICLE SUMMARY

Strengths and limitations of this study

- The study was a response to a local community initiative and followed extensive community consultation.
- The population-based active method of case ascertainment will provide the most accurate prevalence data for diagnoses on the entire FASD spectrum and other health and developmental outcomes.
- Standardised and locally developed clinical assessments whose interpretation is less biased by culture and language have been chosen carefully with cross-cultural considerations in mind and are considered valid for the purpose of the study.
- There are no normative data for Aboriginal children for the assessments used in the study.
- Study findings may not generalise to all children born in the Fitzroy Valley following the introduction of community-led alcohol restrictions in 2007, after which time FASD prevalence may have decreased.

be disseminated widely through peer-reviewed manuscripts, reports, conference presentations and the media.

INTRODUCTION

The Fitzroy Valley is 2500 km North of Perth and 400 km east of Broome, in the remote West Kimberley region of Western Australia, with Fitzroy Crossing town at its centre. The Valley is home to approximately 4500 people, 80% of whom are Aboriginal, belonging to five language groups (Bunuba, Walmajarri/Wangkatjungka, Nyikina and Gooniyandi). There are 45 distinct communities ranging from larger communities (300 people) in the Fitzroy Crossing town site to small Aboriginal cattle station communities with as few as 10–20 people depending on the season.

In 2007, these communities were in crisis with high rates of alcohol abuse, alcohol-related harms, violence and crime. During that year, there were 55 deaths (13 being suicides), with a coronial enquiry finding alcohol to be a factor in many deaths (June Oscar, personal communication, 2010). In the face of resistance from those with a vested interest in the sale of alcohol, brave local women led their community to successfully lobby for restrictions on take-away sales of full strength alcohol such that: *the sale of packaged liquor, exceeding a concentration of ethanol in liquor of 2.7% at 20°C, is prohibited to any person, other than a lodger (as defined in Section 3 of the Liquor Control Act 1988)*. The benefits of these restrictions for the community were both immediate and enduring. One year after introducing the restrictions, an independent evaluation showed a 28% reduction in alcohol-related police tasks, a 36% reduction in alcohol-related presentations to hospital, a 14% increase in school attendance and a reduction in the sale of pure alcohol from the Crossing Inn from 104 l/day to 23 l/day.<sup>1</sup> A new state of sobriety had emerged in

Fitzroy Crossing, and a number of remote satellite communities in the Fitzroy Valley took steps to become ‘dry communities’, with complete alcohol bans. The community became aware of a range of conditions called fetal alcohol spectrum disorders or ‘FASD’ and recognised the signs and symptoms in many children. In 2009, members of Nindilingarri Cultural Health Services and Marninwarntikura Women’s Resource Centre, in collaboration with community members, health and education professionals, developed a strategy to overcome FASD and early life trauma in the Fitzroy Valley. They called the strategy *Marulu*. This is a word from the Bunuba language meaning ‘precious, worth nurturing’, which reflects how they feel about children. The three priorities of the strategy are to prevent FASD, diagnose FASD and support affected families. Building on existing relationships, the community invited The George Institute for Global Health and the Sydney Medical School, University of Sydney, to partner with them in the Lililwan Project to enable FASD diagnosis and determine prevalence. Prior to commencing this work, extensive community consultation was undertaken to identify potential risks and benefits, resulting in well-informed and widespread community support.<sup>2</sup>

Fetal alcohol spectrum disorders

Alcohol is a teratogen and exposure in utero can cause a spectrum of lifelong physical and neurocognitive abnormalities termed ‘fetal alcohol spectrum disorders’ (FASD), including specific diagnoses of fetal alcohol syndrome (FAS), partial fetal alcohol syndrome (pFAS) and neurodevelopmental disorder—alcohol exposed (ND/AE).<sup>3</sup> The teratogenic effects of alcohol were reported in the medical literature in 1968,<sup>4</sup> and the term ‘FAS’ was used in 1973 to describe a pattern of characteristic facial features, growth deficiency and central nervous system (CNS) impairment in babies exposed to alcohol in utero.<sup>5</sup> There is increasing knowledge about the effects of antenatal alcohol exposure on brain structure<sup>6,7</sup> and function, including memory, cognition, executive function, gross and fine motor control, sensory processing, language and behaviour.<sup>6,8,9</sup> Neurodevelopmental disabilities associated with FASD significantly impact quality of life, social and economic participation. Families caring for affected individuals report very high levels of caregiving stress, particularly if the child has problems with executive functioning.<sup>10</sup> There is often an overlay of guilt that FASD could have been prevented.<sup>10</sup> Individuals diagnosed with FASD frequently develop secondary disabilities associated with high rates of disrupted education (61%), including mental health problems (90%), trouble with the law (60%) and substance abuse (39%).<sup>11</sup> Compounding the tragedy for individuals and families, service use and loss of productivity for an individual with any diagnosis on the FASD spectrum costs up to US\$22 000 per year.<sup>12</sup> For people with FAS, the lifetime cost may reach US\$2.5

million.<sup>12</sup> When welfare and justice costs are added, the economic burden is more substantial. Prevalence data on FASD are needed to document the extent of the problem and inform prevention strategies and provision of health, education and justice services.

### Alcohol use in pregnancy

Australia has among the highest annual per capita consumption of pure alcohol, at 10–12.49 l.<sup>13</sup> In Fitzroy Crossing immediately prior to alcohol restrictions being implemented in 2007, the annual per capita alcohol consumption was estimated at 27 l (over three times the national average) or 35 cans of beer per week for every person in Fitzroy Crossing.<sup>14</sup>

Alcohol use in pregnancy is common. Of pregnant women in Moscow, 52% reported alcohol consumption in the most recent month of their pregnancy.<sup>15</sup> In France, 52.2% of women surveyed drank at least once during their pregnancy and 13% of those who drank consumed five or more drinks per occasion in a 'binge' pattern.<sup>16</sup> Large studies in the USA show that approximately 12%–13%<sup>17 18</sup> of women drink in pregnancy, one in 50 (2%) of all women surveyed drinking in a 'binge' pattern during pregnancy. Importantly, these international figures may underestimate the problem: a limitation of surveying women during pregnancy is under-reporting, whereas in retrospective surveys, women report higher consumption.<sup>19</sup>

In Australia, alcohol consumption is common among women of childbearing age. Of women aged 18–24 years, 11% drink at risky/high-risk levels on a weekly basis.<sup>20</sup> National surveys show differences in drinking patterns between Indigenous and non-Indigenous Australian women.<sup>21</sup> Indigenous women are less likely to drink alcohol than non-Indigenous women, but those who do drink are more likely to do so at hazardous levels.<sup>21</sup> In a survey of almost 5000 non-Indigenous Western Australian women between 1995 and 1997, 60% reported drinking in pregnancy, and 4% reported drinking at 'binge' levels of five or more standard drinks per occasion during pregnancy.<sup>22</sup> In one large national survey, 20% of Indigenous Australian women reported drinking in pregnancy,<sup>23</sup> but population-based data, particularly from remote settings, are scarce. Focus group work with Indigenous women suggests that reasons for drinking include lack of knowledge about harms to the fetus, unemployment, having a partner who drinks, domestic violence, loss of traditional land and culture and the legacy of the 'stolen generation' when children were forcibly removed from their mothers by government authorities between 1909 and 1969 (Heather D'Antoine, personal communication, 2011).

### FASD prevalence

International estimates of prevalence of FASD vary markedly depending on study methods. Surveillance or record review studies report a lower prevalence (often due to under-reporting) than clinic-based studies (in

which there may be selection bias), while active case ascertainment studies report the highest prevalence.<sup>24</sup> Most studies report only FAS or partial FAS because the presence of physical signs makes these conditions easier to identify than ND/AE, in which dysmorphism may be subtle or absent.<sup>24 25</sup> The effect of study method on reported prevalence is evident from a review of FAS prevalence studies to 2009. Passive surveillance and clinic-based studies report a median prevalence of 0.27 and 1.9 cases per 1000 people, respectively, while active case ascertainment studies report a median of 8.5 cases per 1000 people.<sup>24</sup> The demographics of the study population and inclusion of the entire FASD spectrum also influences prevalence rates. The few active case ascertainment studies that included all diagnoses on the FASD spectrum report a median prevalence of 19.0 cases per 1000 people.<sup>24</sup> These findings suggest that active case ascertainment studies are likely to be most accurate and that assessing the full range of diagnoses on the FASD spectrum in an entire population in high-risk communities is likely to result in high prevalence figures.

In Australia, the prevalence of FASD is unknown, and there are few FASD screening programmes or diagnostic clinics.<sup>26 27</sup> Under-ascertainment is likely due to lack of screening of children at risk and under-diagnosis of FASD.<sup>28</sup> In Western Australia, fewer than 50% of health professionals routinely ask about alcohol use during pregnancy and only 12% know the four diagnostic features of FAS.<sup>29 30</sup> Australian researchers have attempted to estimate the prevalence of FAS or FASD. In Western Australia, FAS prevalence was reported as 0.18 cases per 1000 West Australian live births between 1980 and 1997 (with rates 100 times higher in the Indigenous population).<sup>31</sup> In an audit of the Victorian Birth Defects Register, the prevalence of FAS was between 0.01 and 0.03 cases per 1000 live births in Victoria between 1995 and 2002.<sup>32</sup> A retrospective medical record and paediatric letter review of children born between 1990 and 2000 in Darwin, Northern Territory, estimated the overall prevalence of FAS to be 0.68 per 1000 children seen in the Royal Darwin Hospital during the study period (and 1.87 per 1000 Indigenous children).<sup>33</sup>

An active surveillance study involved paediatricians notifying newly diagnosed (incident) cases of FAS to the Australian Paediatric Surveillance Unit each month between 2001 and 2004. A prevalence of 0.06 per 1000 live births over the study period was found.<sup>34</sup> Significantly higher birth prevalence and incidence rates were reported in the Indigenous population.<sup>34</sup> Limitations of Australian studies include likely under-diagnosis and under-reporting, limited data on prenatal alcohol exposure, variable data on which diagnoses were made, inconsistent use of diagnostic criteria and limited access to remote populations and therefore under-representation of this high-risk group.

Although attempts to ascertain FASD prevalence in Australia have been made, there is a need for a rigorous population-based active case ascertainment study with



high external validity, using internationally recognised diagnostic criteria for the entire spectrum of FASD diagnoses.

### Evidence gaps and the significance of this work

Accurate FASD prevalence data are needed to plan prevention, diagnostic and management strategies.<sup>35</sup> Accurate data on fetal alcohol exposure is essential to make a FASD diagnosis and examine the relationship between quantity and timing of exposure, and physical and neurocognitive outcomes. Diagnosing all conditions on the FASD spectrum (FAS, pFAS, ND/AE) is important because significant learning and behavioural problems, such as problems in memory and in higher level cognitive and language skills, can result from antenatal alcohol exposure in the absence of characteristic facial features and growth restriction seen in FAS.<sup>36</sup> In high-risk communities, particularly where retention of traditional culture relies on stories and songs passed on orally, there is an urgency to address the devastating effects of FASD and to determine the most effective prevention strategies.<sup>37 38</sup> Clinical studies with active case ascertainment will provide the highest and most accurate rates of FASD prevalence.<sup>24</sup>

In Australia, addressing FASD has become a focus of the Prime Minister's Science, Engineering and Innovation Council and the Australian Human Rights Commission and a Parliamentary Inquiry into the prevention, incidence and management of FASD is currently underway.<sup>37 39 40</sup>

### Study aims

The aims of the Lirilwan Project are as follows:

- ▶ Establish the prevalence of FASD and other health and developmental problems in all children born in 2002 and 2003 and residing in the Fitzroy Valley.
- ▶ Determine relationships between pregnancy exposures and neurodevelopmental outcomes.

Additionally, the researchers undertake to provide individual, multidisciplinary health management plans for each participant and to deliver education and family support to increase the benefit of the study to participants and their communities. Data will also be used in service planning and prevention programmes.

### Hypotheses

It is hypothesised that:

- ▶ Using validated and locally developed measures, we will identify higher prevalence rates of FASD, developmental delay, behavioural problems, hearing impairment and educational underachievement in this population than predicted by current Australian data.
- ▶ We will identify modifiable risk factors for adverse health and educational outcomes.

## METHODS AND ANALYSIS

### Study design

The Lirilwan Project is a population-based active case ascertainment study of the prevalence of FASD.

### Setting

This study will take place in the remote Fitzroy Valley of North Western Australia, including Fitzroy Crossing town and approximately 45 remote communities representing the language groups of the Bunuba, Walmajarri/Wangkatjungka, Gooniyandi and Nyikina peoples.<sup>41</sup> The Fitzroy Valley is approximately 2500 km North of Perth and 400 km East of Broome and includes communities within a radius of 200 km from Fitzroy Crossing town. The total population of the Fitzroy Valley is 4500, approximately 80% being Aboriginal.<sup>41</sup>

### Study timeline

The project is being carried out between October 2009 and June 2014 in two stages. Stage 1 includes community consultation, recruitment and interviews with parents/carers using a diagnostic questionnaire and will occur between 2009 and 2011. Stage 2 involves interdisciplinary clinical assessments, diagnostic case conferences and feedback of management plans to participants and health and education providers and will occur during 2011 and 2012. Data entry and analysis, preparation of reports and publication of manuscripts will occur between 2012 and 2014.

### Participants

Participants will be all children born between 1 January 2002 and 31 December 2003 and living in the Fitzroy Valley in 2010 or 2011. They will be identified from the Fitzroy Valley Population Project<sup>41</sup> and Communicare (electronic population health program) databases. This age group was selected as being sufficiently old to participate in assessment for FASD and health, educational and behavioural outcomes, yet young enough to benefit from recommended interventions in the health, school and home settings. These children will likely have environmental and alcohol exposures similar to those living in remote communities elsewhere in Western Australia, the Northern Territory, South Australia, Queensland, Victoria and New South Wales. Findings may also be relevant to remote or low socioeconomic communities internationally.

### Recruitment

Parents or carers of children in the cohort will be visited by 'community navigators' (local Aboriginal people working on the research team) who will explain the purpose of the study using participant information statements and consent forms, read to parents/carers in plain English, Kimberley Kriol or in one of the local Aboriginal languages as required. Pictorial aides have been developed to assist with explanation of the study rationale, methods and clinical assessment processes. With assistance from local community navigators, researchers will obtain written consent for participation in the study at three points in time: for parent/carer participation in stage 1 interviews using a diagnostic questionnaire, prior to interdisciplinary clinical

assessment (stage 2) and prior to sharing management plans with health and education providers, and making relevant referrals.

Due to inconsistent recording in the remote setting, participants' birth dates will be documented using multiple sources: midwife birth notification forms, hard copy medical records and Communicare electronic health records. Birth dates will be confirmed with parents/carers for accuracy. Families are very mobile within the Fitzroy Valley and often leave the Valley for climatic, cultural or personal reasons.<sup>41</sup> Participants will be located using community navigator advice on their current location.

### Diagnostic criteria, outcome measures and statistical analysis

Analyses will be carried out in the context of the study aims and hypotheses described above. The Canadian guidelines for the diagnosis of FASD<sup>42</sup> will be used to assign specific diagnoses along the FASD spectrum based on comprehensive interdisciplinary clinical assessment. Diagnostic criteria are listed at [table 1](#).

The Canadian guidelines were chosen following feedback from local health professionals and cultural leaders regarding the diagnostic terminology and practicability of use in a remote clinical setting. Due to the unique cultural and language context of the study population, specific clinical assessments whose interpretation is less biased by culture and language were chosen.

Prevalence of FASD (including diagnoses of FAS, partial FAS and ND/AE), and other health and selected developmental diagnoses, will be calculated as the proportion of children diagnosed in the total population of children in the age group and expressed per 1000. Prevalence will be reported in all children in two age groups (those born in 2002 and 2003); hence, CIs will not be calculated.

In children diagnosed with FASD, associations between their diagnosis and education and health outcomes will be examined. In children exposed to alcohol in utero associations between all pregnancy exposures (including alcohol and other adverse antenatal events) and early life trauma (including household overcrowding, emotional or physical trauma), and health and developmental outcomes (including speech and language, visual motor integration, cognition and executive function) will be examined.

Descriptive analyses will be conducted to obtain frequencies and means for variables of interest, followed by univariate analyses to assess whether exposures are significantly related to outcomes and to inform multivariate analyses. Multivariate models will then be built if appropriate, with inclusion of other variables as potential confounders. Logistic regression models will be built for binary outcome variables and linear models for continuous outcome variables. Potential confounders will be selected for inclusion in regression models based on the published literature or

if the variable is significantly correlated with the outcome and is a main risk factor in the univariate analysis ( $p < 0.2$ ).

When statistical power allows, plausible effect modification will be tested by adding an interaction term to the full model containing all possible confounders. If effect modification is found (a significant interaction variable  $p < 0.05$ ), further analyses will be stratified by that variable.

All analyses will be conducted using SAS V.9.2. Results will be reported according to STROBE guidelines for reporting observational studies.<sup>44</sup>

Normative population data are not available for Australian Aboriginal children for any of the clinical assessments used in the study. Clinicians have consulted extensively to choose assessments that minimise cultural and linguistic bias. Population norms for facial features of FAS are available for Caucasian and African-American populations. The African-American Lip-Philtrum Guide<sup>45</sup> will be used for assessing lip thinness and philtrum smoothness in Aboriginal children, and the Hall population reference data will be used for palpebral fissure length.<sup>43</sup> For growth parameters, WHO charts will be used, as they are most relevant to this population.

### Data collection

Data will be collected in two stages.

#### Stage 1: Characterising the cohort by administration of a diagnostic questionnaire and medical note review

A comprehensive diagnostic questionnaire has been developed for use in interviews with parents/carers. Content is based on a literature review of FASD diagnostic criteria and existing diagnostic checklists<sup>42 45-49</sup> with consideration of potential antenatal and environmental influences on child development,<sup>50-53</sup> maternal risk factors for birth defects and risk factors for FASD.<sup>54-57</sup>

The diagnostic questionnaire includes 114 questions about child demographics, schooling, language, place of residence and living conditions, prenatal exposures (including alcohol quantified using the AUDIT-C<sup>58</sup>), neonatal history, early life trauma (relevant to some remote Aboriginal communities), health and educational outcomes and family characteristics. It was developed by paediatricians in plain English and modified by local language experts at the Kimberley Interpreting Service to ensure content and language appropriateness. Reliability testing was conducted on a pilot questionnaire, which was then revised before formal reliability testing, which confirmed high inter- and intra-rater reliability. Community navigators will assist researchers to administer the questionnaire in plain English, Kimberley Kriol or local Aboriginal languages as required (Bunuba, Walmajarri/Wangkatjungka, Gooniyandi and Nyikina). Participant medical records will be comprehensively reviewed for information

**Table 1** Diagnostic criteria based on the Canadian guidelines for the diagnosis of fetal alcohol spectrum disorders<sup>42</sup>

| Diagnostic category                      | Fetal alcohol syndrome   | Partial fetal alcohol syndrome   | Neurodevelopmental disorder—alcohol exposed                             |
|--|--|--|---|
| Diagnostic criteria                      | Requires all four criteria below to be met   | Requires confirmed prenatal alcohol exposure, the presence of two of the three characteristic facial anomalies at any age and CNS criteria to be met   | Requires confirmed prenatal alcohol exposure and CNS criteria to be met |
| Prenatal alcohol exposure                | Confirmed or unknown   | Confirmed  | Confirmed   |
| Facial anomalies                         | Presence of all three of the following facial anomalies at any age: <ul style="list-style-type: none"> <li>▶ Short palpebral fissure length (<math>\leq 2</math> SDs below the mean using the Hall charts<sup>43</sup>)</li> <li>▶ Smooth philtrum (rank 4 or 5 on the UW Lip-Philtrum Guide*)</li> <li>▶ Thin upper lip (rank 4 or 5 on the UW Lip-Philtrum Guide)</li> </ul>   | Presence of any two of the following facial anomalies at any age: <ul style="list-style-type: none"> <li>▶ Short palpebral fissure length (<math>\leq 2</math> SDs below the mean)</li> <li>▶ Smooth philtrum (rank 4 or 5 on the UW Lip-Philtrum Guide)</li> <li>▶ Thin upper lip (rank 4 or 5 on the UW Lip-Philtrum Guide)</li> </ul> | No anomalies required   |
| Growth deficit                           | Prenatal or postnatal growth deficit indicated by birth length or weight $\leq 10$ th percentile adjusted for gestational age or postnatal height or weight $\leq 10$ th percentile  | No deficit required  | No deficit required   |
| Central nervous system (CNS) abnormality | Significant CNS dysfunction (evidence of impairment in three or more of the following CNS domains): <ul style="list-style-type: none"> <li>▶ Hard and soft neurological signs; seizure disorder; gross and fine motor functioning; articulation, phonology and motor speech</li> <li>▶ Cognition (IQ or uneven cognitive profile)</li> <li>▶ Memory</li> <li>▶ Executive functioning and abstract reasoning</li> <li>▶ Communication (expressive and receptive language)</li> <li>▶ Attention deficit/hyperactivity +/- other behavioural problems; abnormal sensory processing</li> <li>▶ Visual motor integration</li> <li>▶ Adaptive behaviour/social skills/social communication</li> <li>▶ Academic achievement</li> <li>▶ CNS structure (including head circumference <math>\leq 3</math>rd percentile or other structural CNS abnormality)</li> </ul> |  |   |

\*The University of Washington (UW) Lip-Philtrum Guide is a 5-point Likert scale with representative photographs of lip and philtrum combinations with ranks 1–3 within normal limits and ranks 4 and 5 outside normal limits.

related to the pregnancy, birth, and lifelong health and developmental outcomes documented at the time of review. Hard copy medical records, Communicare community health records and midwife notification data will be reviewed.

**Stage 2: Comprehensive interdisciplinary clinical assessments**

A trial of stage 2 interdisciplinary health and neurodevelopmental assessments will be conducted in Fitzroy Crossing for six children to pilot clinical assessment measures and processes. After the pilot phase, direct testing of each child will be conducted over three non-consecutive days (with additional data gathered from

teachers and parents through questionnaire when relevant). This is detailed in table 2.

Day 1 assessments (including audiology and ophthalmology) will be conducted over a 2-week period (between 1 week and 5 months before the day 2 and 3 clinical assessments). Information from teachers will be requested prior to day 2 and 3 assessments; however, for logistical reasons, this information may not be immediately available. Day 2 and 3 assessments will not always be conducted in the same order for logistical reasons. Interdisciplinary case conferencing will be used to assign specific diagnoses of FASD (including FAS, pFAS and ND/AE) and other health and developmental problems.

**Table 2** Clinical assessments and outcome measures conducted over three non-consecutive days, by discipline

| Discipline           | Assessment   | Outcome measures  |
|----------------------|--|---|
| Day 1*               |  |   |
| Community navigator  | Cultural/language/spiritual health/social assessment†  | Impact of cultural and social determinants on child health and ability to participate in the Lirilwan Project   |
| Audiology            | Tympanometry   | Tympanic membrane mobility/middle ear function  |
|                      | Audiometry   | Conductive and sensorineural hearing loss   |
|                      | Video-otoscopy   | Tympanic membrane integrity or abnormality  |
|                      | LiSN-S assessment for Central Auditory Processing Disorder <sup>59</sup>   | Auditory processing abilities   |
| Ophthalmology        | Culturally appropriate optotypes   | Visual acuity   |
|                      | Nidek Autorefractor  | Refractive status   |
|                      | Clinical eye examination   | Ocular mobility, strabismus, nystagmus  |
|                      | Colour photograph of front of the eye  | Structure of the iris and cornea  |
| School teacher       | Colour photograph of the retina and optic nerve  | Optic nerve size, retinal vascular status   |
|                      | ASEBA teacher report form <sup>60</sup>  | Educational, competence and behavioural profile from teacher perspective  |
| Child health nurse   | Sensory profile: school companion <sup>61</sup>  | Sensory processing profile in the school setting  |
|                      | Speech/language qualitative questionnaire  | Speech, language and literacy abilities in the school setting   |
|                      | Anthropometric measurements  | Height, weight, body mass index, head circumference, abdominal circumference, mid-arm circumference             |
| Day 2‡               |  |   |
| Child psychology     | Universal Non-verbal Intelligence Test <sup>62</sup>   | Cognitive profile, using non-verbal assessment of memory and reasoning, in symbolic and non-symbolic modalities |
|                      | ASEBA child behaviour checklist/6–18 <sup>60</sup> (carer questionnaire)§  | Competence in school, activities and social skills and behavioural problems profile, from carer perspective     |
|                      | Children's colour trails <sup>63</sup>   | Aspects of executive function   |
|                      | Digit span subtest of the Wechsler Intelligence Scale for Children-IV <sup>64</sup>  | Aspects of executive function and short-term auditory memory  |
| Occupational therapy | Bruininks-Oseretsky Test of Motor Proficiency—age 4–21 years (BOT2) <sup>65</sup>  | Fine motor precision, fine motor integration, manual dexterity and overall manual coordination                  |
|                      | Buktenica Test of Visual-Motor Integration—age 2–100 years, with Visual Perception and Motor Coordination subtests <sup>66</sup> | Visual motor skill, perception and integration  |
|                      | Short sensory profile (carer questionnaire) <sup>67</sup> §  | Sensory processing in the home setting  |
|                      |  |   |

Continued

Table 2 Continued

| Discipline   | Assessment   | Outcome measures   |
|--|--|--|
| Day 3‡   |  |  |
| Speech and language: screening assessments developed locally for Kimberley Kriol speakers specifically for this project, based on consultation with clinicians and language consultants and review of literature including Gould <sup>68</sup> | Video recording of children participating in free play/activity for 10–15 min<br>Interactive story telling using a culturally familiar story with moral theme (conducted in Kimberley Kriol)   | Social communication and peer interaction skills<br><br>Scores of comprehension using Blank Levels of questioning, expressive language and degree of prompting required (eg, repetition/rephrasing of question)  |
|  | Non-word repetition task   | Working phonological memory, literacy skills and phonological awareness  |
|  | Sequencing and narrative discourse activity (conducted in Kimberley Kriol)<br>Oromotor assessment  | Sequencing, grammar, recount, higher level expressive language<br>Articulation, phonology and motor speech function  |
| Validated screening test used in English-speaking children   | Clinical Evaluation of Language Fundamentals Screening Test (CELF 4), Australian language adaptation—age 5–21 years <sup>69</sup>  | Screening test for risk of a language disorder   |
| Paediatrics  | Complete physical, neurological and dysmorphology examination (including palpebral fissure length measurement and use of UW Lip-Philtrum Guide <sup>45</sup> )<br>Interpretation of anthropometric data using WHO child growth standards <sup>70</sup> | Identification of dysmorphologies specific to antenatal alcohol exposure and physical health status (including common dental, skin, ear and respiratory disease)<br>Evidence of growth restriction, microcephaly |
| Physiotherapy  | Bruininks-Oseretsky Test of Motor Proficiency—age 4–21 years (BOT2) <sup>65</sup><br><br>Quick Neurological Screening Test—II (Revised edition)—age 5 and above <sup>71</sup>  | Bilateral coordination, balance, running speed and agility, upper limb coordination, overall body coordination, strength and agility<br>Soft neurological signs  |

\*Day 1 assessments will occur over a 2-week period for the entire cohort, not consecutively with day 2 or 3 assessments.

†Community navigators will conduct an informal non-standardised assessment of cultural, language and social issues and relay any relevant information to the clinical team for consideration prior to assessment and at the time of case conferencing.

‡For logistical reasons, day 2 and 3 assessments will not always occur on consecutive days or in the order presented here.

§The ASEBA Child Behaviour Questionnaire and Short Sensory Profile (carer questionnaire) will be interpreted in the local language and some items altered for cultural congruence.

FASD diagnoses will be based on categorisation of dysmorphology, growth restriction, alcohol exposure, CNS structure and domains of neurodevelopmental impairment as outlined in table 1. To avoid bias, clinicians will be blinded to alcohol exposure until after they have scored and determined outcomes for facial anomalies, growth deficit and CNS abnormalities.

### Communicating results to participants and local service providers

A composite interdisciplinary report and management plan will be compiled during the interdisciplinary case conference. The assessment findings will be communicated to parents and carers (with the assistance of a community navigator if appropriate) and, with parental consent, information about individual children will be provided to relevant teachers and health professionals.

Professional development sessions will be carried out in the schools of all participating communities. When a diagnosis on the FASD spectrum is made, follow-up

information and support will be provided to families by an experienced educator and psychologist employed through Marninwarntikura Women’s Resource Centre. Strategic planning by community leaders for ongoing support and services will occur as appropriate as part of the broader FASD strategy.

### Ethics and dissemination

This study of child health in a predominately Aboriginal population requires particular consideration of cultural and language issues. Ethics approval has been granted for all stages of this study (stage 1 administration of diagnostic questionnaires, a trial of stage 2 clinical assessments and stage 2 clinical assessments) by the University of Sydney Human Research Ethics Committee (approval numbers 12527, 13187, 13551), the Western Australian Aboriginal Health Information and Ethics Committee (approval numbers 271-01/10, 319-10/10, 344-04/11), the Western Australian Country Health Service Board Research Ethics Committee (approval numbers 2010:01, 2010:28, 2011:04) and the Kimberley



Aboriginal Health Planning Forum Research Subcommittee (approval numbers 2010-001, 2010-001, 2010-001). A memorandum of understanding that clearly defined the project's underlying principles, scope of work, and roles and responsibilities of each organisation was signed between Fitzroy Valley community organisations (Marninwarrtikura Women's Resource Centre, Nindilingarri Cultural Health Services) and research organisations (The George Institute, Sydney Medical School).

Dissemination of the results of this study will occur on numerous levels. Locally, individual health summaries and management plans will be explained and provided to the child's parent/carer and with their consent to schools and the local health service. Group results of prevalence and other key findings will be shared with participant communities during a series of community feedback meetings. Manuscripts based on this work will be submitted to leading journals in the field and results will be presented at scientific meetings in Australia and internationally. Australian government agencies and policy makers have been engaged through the study group's presentation to a Parliamentary Inquiry into FASD incidence and prevention. The impact of study results at a national level will be enhanced through the research team's strong links with the Australian Human Rights Commission, who profiled the community's strategy to overcome FASD in the 2010 Social Justice Report.<sup>37</sup>

## DISCUSSION

A key strength of this study is that it resulted from a local community initiative and followed extensive community consultation. The population-based active method of case ascertainment will provide accurate prevalence data for diagnoses on the entire FASD spectrum and other health and developmental outcomes. Relationships between modifiable exposures and outcomes will inform prevention, service provision and policy for child development, healthcare, education and justice. Ethical, language and cultural issues were paramount at all stages of study conception and design. Local community leaders are represented as Chief Investigators on the study and approximately 30% of the research team are local Aboriginal people. The language used to gain consent for participation and during the study will be informed and often delivered by local language experts. Comprehensive assessment results will feed into interdisciplinary health summaries and management plans that can be used by families, health and education services to improve outcomes for these children in a timely fashion.

Limitations of the clinical assessments include lack of normative data for Aboriginal children. To mitigate this, assessments were chosen carefully with cross-cultural considerations in mind and are considered valid for the purpose of this study. Additionally there are no normative data for Australian Aboriginals for the facial features of FAS, so existing data from African-American popu-

lations will be used. Study findings may not generalise to children born in the Fitzroy Valley following the introduction of community-led alcohol restrictions in 2007, after which time FASD prevalence may have decreased.

This work will provide accurate data on the health and developmental status of an entire population of children living in the Fitzroy Valley. Accurate estimation of the prevalence of diagnoses on the FASD spectrum will inform the level of health, education and other services required in this population. Prevention strategies will be based on detailed knowledge of antenatal exposures and environmental impacts on child health and development.

The Liliwan Project provides a model for effective community consultation, adaptation of clinical and research techniques to suit complex cultural and language conditions and provision of short- and long-term benefit to communities that participate in clinical research. Australia's Social Justice Commissioner described this project as 'an example of researchers reciprocating both the spirit and intent of the community by working to address the challenges of FASD in genuine partnership—one where research is done with the community and not just about the community',<sup>2</sup> adding that 'a process guided by a relationship underpinned by meaningful, respectful engagement and collaboration will always be more effective and successful than one that is not'.<sup>37</sup>

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