Respiratory, birth and health economic measures for use with Indigenous Australian infants in a research trial: A modified Delphi with an Indigenous panel

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Abstract

Background: There is significant disparity between the respiratory health of Indigenous and non-Indigenous Australian infants. There is no culturally accepted measure to collect respiratory health outcomes in Indigenous infants. The aim of this study was to gain end user and expert consensus on the most relevant and acceptable respiratory and birth measures for Indigenous infants at birth, between birth and 6 months, and at 6 months of age follow-up for use in a research trial.

Methods: A three round modified Delphi process was conducted from February 2018 to April 2019. Eight Indigenous panel members, and 18 Indigenous women participated. Items reached consensus if 7/8 (≥80 %) panel members indicated the item was 'very essential'. Qualitative responses by Indigenous women and the panel were used to modify the 6 months of age surveys.

Results: In total, 15 items for birth, 48 items from 1 to 6 months, and five potential questionnaires for use at 6 months of age were considered. Of those, 15 measures for birth were accepted, i.e., gestational age, birth weight, Neonatal Intensive Care Unit (NICU) admissions, length, head circumference, sex, Apgar score, substance use, cord blood gas values, labour, birth type, health of the mother, number people living in the home, education of mother and place of residence. Seventeen measures from 1-to 6 months of age were accepted, i.e., acute respiratory symptoms (7), general health items (2), health care utilisation (6), exposure to tobacco smoke (1) and breastfeeding status (1). Three questionnaires for use at 6 months of age were accepted, i.e., a shortened 33-item respiratory questionnaire, a clinical history survey and a developmental questionnaire.

Conclusions: In a modified Delphi process with an Indigenous panel, measures and items were proposed for use to assess respiratory, birth and health economic outcomes in Indigenous Australian infants between birth and 6 months of age. This initial step can be used to develop a set of relevant and acceptable measures to report respiratory illness and birth outcomes in community based Indigenous infants.

Background

Undetected respiratory illness is common in Indigenous children leading to high rates of chronic illness and hospitalisation (1–4). Indigenous Australian children aged 0 to 4 years are hospitalised for respiratory illness at two times the rate of other Australian children (2). Rates are higher for certain respiratory diseases in some regions, such as bronchiectasis in Northern Queensland (5). A combination of social, historical, and cultural contexts contribute to the high, and unacceptable rates of disease (2). Addressing the social determinants of health will see the greatest improvements, though clinical care must be improved simultaneously (6).

Infancy is a critical time to detect, and effectively manage respiratory illness. While up to 6 episodes of self-limiting respiratory illness is normal in the first year of life (7, 8)), untreated chronic symptoms commonly lead to lifelong disease, poor quality of life and early death (6). Respiratory illnesses can be

split into upper and lower. Lower respiratory infections are usually more serious and a leading cause of poor health and death in infants (2). The most common symptom for an acute lower respiratory illness is cough. If cough persists for more than four weeks duration, it may indicate an underlying chronic respiratory illness (9). Measuring chronicity and severity of acute respiratory symptoms, particularly wet cough, is important to detecting underlying chronic illness (10).

Despite respiratory illness being a leading contributor to the total burden of disease among Indigenous children, there is scarcity of community level data (2). One single urban centre study with 180 Indigenous children under five years of age used monthly interviews over 12 months to measure acute respiratory illness (11). One in five children experienced at least one episode of chronic cough (12). More than half of the children identified with chronic cough were diagnosed with an underlying lung disease, mostly protracted bacterial bronchitis, asthma and bronchiectasis (12). A second study in remote Indigenous communities with 651 children under six years of age using observations to measure illness reported a point prevalence for cough of 39% (3). In national parent reported data from 2012–2013 asthma prevalence is 15% as compared to 9% in non-Indigenous children (13).

As well as limited data, inconsistent measures have been used to capture respiratory illness. There are no standard measures for respiratory symptoms or illness specifically developed for Indigenous children (2). In research trials, respiratory symptoms are typically collected via parent-reported questionnaires, interviews, or symptom diary cards (14). Parent-reported measures are valuable and clinically relevant with wide reach at relatively low cost. However parent-report is reliant on accurate recall and health literacy and response rates can be low (15). Cough is the main outcome collected via parent-report for respiratory illness (14). Reliability of parent reported cough for children is reported to be good for daytime cough and poor for nocturnal cough (14). Accuracy of parent reported wheeze is reported to be low (16). In a recent study with 889 Indigenous children and young people aged 3 to 25 years the prevalence of parent-reported common respiratory illnesses was compared to medical records for agreement (15). Agreement was moderate for asthma and bronchiectasis, and poor for pneumonia. Factors likely to have contributed to these discrepancies include health literacy, language, ease of identifying disease and undiagnosed disease (15). Gold standard measures for detecting respiratory illness are clinical assessment including observation and objective tests such as spirometry and/or x-ray (17), though these measures can be impractical for trials due to the ongoing and fluctuating nature of symptoms as well as being costly, time intensive and burdensome for families.

Culturally safe, effective measures for detecting respiratory illness in Indigenous infants needs further development to improve respiratory health outcomes (2). Accurate data is vital to enable us to understand the current state of Indigenous infant health, to acknowledge progress, and to determine how to reduce inequalities between Indigenous and non-Indigenous children (18). There is an entrenched lack of trust from Indigenous Australians in health care professionals and systems (19), medical research (20) due to historical and current policies (including the Stolen Generations) which requires intense consultation with Indigenous leaders, consumers and topic experts to ensure that cultural safety of Indigenous peoples is paramount in research (20). The purpose of this study was to systematically

consult a group of Indigenous academics, clinicians and women on the most accurate, culturally safe, and feasible respiratory health measures for use with Indigenous mothers and infants for a research trial.

Method

Study design

A modified Delphi with an Indigenous expert panel was used. The Delphi method is a culturally acceptable method of gaining consensus and has been used in other areas of Indigenous health research (21, 22). The consensus process was completed between February 2018 and April 2019. The Delphi technique is a method used to collect opinions from a group of experts to achieve consensus on a particular research question (23). Repeated questionnaires are used to facilitate independent, gradual and considered opinions (24). Modified versions involving group discussion may be used where feasibility and operational aspects are solved through group problem solving (25–27). In this study, discussion was also an opportunity for dialogue on cultural safety considerations. This study was conducted in the context of identifying Australian Indigenous culturally acceptable measures for use in a trial to assess infant respiratory symptoms and illness. The measures would be used to follow up infants born to mothers enrolled in the SISTAQUIT® (Supporting Indigenous Smokers To Assist Quitting) smoking cessation trial (Australian New Zealand Clinical Trials Registry trials (ACTRN12618000972224).

Participants

An Indigenous expert panel participated in the three round Delphi process and Indigenous women provided feedback on the 6-month surveys. Using a snowball recruitment strategy, a list of potential expert panel members known to study Investigators were invited to participate by email and asked to share the invitation with colleagues. The list included 12 academics and 20 peak bodies. Panel members were emailed the full SISTAQUIT study protocol and invited to participate if they had research or clinical expertise in Indigenous infant health. Panel members were also asked to invite colleagues who may be interested. Members continued to be invited until a minimum quota of seven was reached (14,15,17). Eight Indigenous advisory panel members participated. Panel members included, 1) Postdoctoral researcher in acute respiratory illness with Indigenous children, 2) Principal Research Fellow in mothers and babies health, 3) representative of HealthInfoNet, 4) Associate Professor at an Indigenous research unit 5) Representative of Indigenous Allied Health Australia (IAHA), 6) Obstetrician, 7) Paediatrician and 8) Representative of The Congress of Aboriginal and Torres Strait Islander Nurses and Midwives (CATSINaM). Indigenous women who were all mothers of young children were recruited through known networks of Indigenous research assistants in Hunter New England and the Mid North Coast of New South Wales.

Description of the modified Delphi method used

A three round modified Delphi with teleconference and two repeat questionnaires was used. An overview of the consensus process is presented in Fig. 1. Round one involved a group discussion with the

Indigenous expert panel and rounds two and three used repeat online questionnaires. Feedback from 18 Indigenous women on potential respiratory questionnaires for use at 6 months of age were gathered between rounds two and three.

Review of literature

A search of the literature was completed to identify outcome measures used with Indigenous Australian infants up to 6-months of age. Outcomes of interest were 1) birth outcomes related to adverse impact of exposure to tobacco in-utero (as per broader study), 2) respiratory symptoms and illness, 3) health care utilisation, and 4) developmental outcomes. Keywords were used to search electronic databases including HealthInfoNet, Google Scholar, ScienceDirect, Cochrane Library and CINAHL. Reference lists and grey literature were searched. Known experts in the field were contacted and asked of knowledge on measures used in clinical practice.

Round one: Teleconference

The first teleconference was used to provide an overview of the study; and to seek preferences for the Delphi process i.e. online questionnaires or interviews. During this call, participants were also asked to share knowledge on potential measures and were given guidance on the information required by the panel to support decision-making.

Questionnaire development

The questionnaire of potential outcomes included items on types of outcome measures, mode and frequency of data collection and acceptability of existing surveys for use at 6-months of age. Potential birth outcome measures were derived from a Cochrane review on smoking cessation interventions used during pregnancy (28), acute respiratory symptoms from a survey used in a longitudinal study on respiratory symptoms in Indigenous children (11) and items on health care utilisation from a systematic review and a cost-consequence analysis (29, 30). Two additional items on breastfeeding and exposure to environmental tobacco smoke were added from the respiratory symptoms survey(11). Potential questionnaires identified for use from a literature review? at 6 months included two respiratory screening tools, 1) a 50-item respiratory questionnaire (31) and 2) an 18-item respiratory adapted into Creole (32) as well as a clinical assessment form developed for the purpose of the larger study. A development screening tool with an adapted version for remote Indigenous communities was also identified (33, 34). A Respiratory Paediatrician (JM) and Health Research Economist (SD) provided expertise on respiratory health and health care utilisation items respectively.

Round Two: Questionnaire

An online questionnaire delivered on REDCap software was used. The questionnaire consisted of three sections with 58 items. Participants were also asked for feedback on 4 existing questionnaires for use at 6 months of age. In total, participants took approximately 30 minutes to complete. In section one, participants were asked multiple choice and open-ended questions on birth outcome measures and

potential modes of data collection. Consensus was pre-determined for multiple-choice items as 80% agreement (26, 35). Items progressed to round three if agreement was between 50 and 80% and omitted if below 50%. Additional items suggested for inclusion in qualitative responses were added to the round 3 questionnaire. In section two, participants were asked to rate respiratory symptoms and health care utilisation items using a 4-point Likert scale (very essential, somewhat essential, non-essential and unsure) as to whether each item should be collected in the trial. As above, consensus was pre-determined as 80% agreement (using 'very essential' only). Items progressed to round three if agreement was 50 to 80% and omitted if below 50%. In the final section, participants were asked for qualitative feedback on 4 potential questionnaires for use at 6-months: two respiratory, one developmental and one clinical assessment form. Qualitative responses were synthesised and used to modify questionnaires.

Feedback from Indigenous women

Two focus groups were held by Indigenous research assistants to gain feedback from 18 Indigenous women on two respiratory questionnaires. Both focus groups were conducted in regional areas of New South Wales. Questions used to gather feedback on acceptability include: 1) Are the questions easy to understand? 2) Is the language appropriate? 3) What do you think of the length of the questionnaire? 4) Would you feel comfortable answering this questionnaire? Women provided feedback verbally and in writing. Feedback was used to modify questionnaires.

Round Three: Questionnaire

The round 3 survey was sent via email to the panel and took participants approximately forty-five minutes to complete. Additional information was provided as requested by participants in round 2 to aid decision making. Participants were asked to reply 'yes' or 'no' for inclusion of each birth outcome measure and rate respiratory symptoms and health care utilisation items that had not reached consensus in round 2 using the same 4-point Likert scale. Participants were provided summary points of the panel's qualitative feedback as well as the modified versions of the three questionnaires and asked to reply 'yes' or 'no' for the acceptability of the modified versions. A space was also available for qualitative feedback. Consensus was achieved by combining 'very essential' and 'somewhat essential' responses at 80% agreement. A rule was enacted to accept items with highest frequency where 80% was not reached. This rule was not pre-determined and enacted due to the timeline of the larger study. Results were presented to the panel for final comment.

Results

Round One: Teleconference

Four of the eight panel members attended a group teleconference and three members were interviewed individually by SP. The panel agreed to participating in online questionnaires rather than interviews to increase flexibility in participation for future rounds. The panel recommended qualitative feedback be included as well as the rating of items.

Birth Outcomes

Round One: Teleconference

Birth outcomes discussed as important included birth weight, small for gestational age, head circumference Apgar score, delivery at less than 37 weeks gestation, stillbirth, Neonatal Intensive Care Unit (NICU) admissions and sex. Panel members considered it essential to limit women's burden to answer surveys straight after birth by using discharge summaries or data linkage.

Round Two: Questionnaire

Seven measures at birth (birth weight, gestational age, Apgar score, NICU admissions, sex length, head circumference) were presented for consensus. Three items reached consensus and four progressed to round three (Table 1). The panel suggested an additional seven outcomes in qualitative responses including substance use in pregnancy, cord blood gas values, labor type (induction, spontaneous), birth type (caesarean, vaginal), health of the mother, number people living in home, educational attainments of the mother and place of residence. Seven members (> 80%) indicated the best mode of data collection to be hospital discharge summary.

Round Three: Questionnaire

Eleven items were presented for consensus (Table 1). All 11 items in round 3 reached consensus (Table 1). A total of 15 items were accepted as essential items to collect.

Table 1
Consensus for birth outcomes

Items	Round 2 n = 8	Round 3 n = 8	Consensus
Gestational age	7	-	✓
Birth weight	7	-	✓
NICU admissions	7	-	✓
Length	6	7	✓
Head circumference	4	8	✓
Sex	5	8	✓
Apgar score	5	8	✓
Substance use in pregnancy	-	8	✓
Cord blood gas values	-	8	✓
Labour (induction, spontaneous)	-	8	✓
Birth type (caesarean, vaginal)	-	8	✓
Health of mother	-	8	✓
Number people living in home	-	8	✓
Education	-	8	✓
Place of residence	-	8	✓
Total:	7	11	15

Data collection from one to 6 months of age for respiratory symptoms and health service utilisation

Round One: Teleconference

Panels members were asked to consider the best mode of data collection from the mothers of the infants from one to 6 months of age. Options discussed included phone call, face-to-face, text message, online diary using phone application or weblink. The panel recommended phone calls or face to face (with use of text message to organise time/venue). The panel advised that women were unlikely to use a mobile phone application to report data. The panel recommended gaining feedback from Indigenous women on their preference for the modality of data collection i.e. phone call, face-to-face, email, mobile phone application. Options discussed for personnel to collect data included an on-site research facilitator (a volunteer for the service who would be aiding the main trial) or other female health worker with a trusted relationship with the woman. The panel members advised additional information would be required to

form a decision on the inclusion of respiratory items and requested input from Respiratory Paediatrician (JM) as required to support decision making.

Round Two: Questionnaire

Forty-eight items were presented in total for consideration. Five items were presented on how data should be collected (frequency, number of survey questions, modality, personnel to collect data and reimbursement amount) (Table 2). Two items reached consensus, 1) frequency of data to be collect monthly rather than fortnightly and 2) modality of collection for women to choose their preference. Three items progressed to round three (number of survey questions, personnel to collect data, reimbursement amount). Forty-three items were presented on acute respiratory symptoms, health care utilisation, exposure to tobacco smoke and breastfeeding status. Of the 43 items, one item reached consensus (exposure to tobacco smoke). Twenty-eight items progressed to round three and 16 items were omitted (Table 3).

Table 2

Frequency, number of questions, mode, personnel to collect data and reimbursement

Items	Round 2 n = 8	Results 3 n = 8	Consensus
Frequency of data collection: Fortnightly Monthly	0 7	- 7	١
Number of questions: 1 to 5 6 to 10 11 to 15 16 to 20	0 4 1 2	- 5* 1 2	١
Modality: Phone call Survey Phone app Email Women's preference Women randomised to different modality	3 2 2 1 7 1	- - - - 7 -	١
Who should collect data: Research facilitator (based on site, Indigenous or non-Indigenous) Indigenous researcher (based at research institution) Non Indigenous researcher (based at research institution) Research facilitator, if not possible, Indigenous researcher Unsure	6 5 2 - 1	2 3 - 3* -	١
Reimbursement to mother, amount per survey: $15 voucher Baby bundle (value of $15) $10 voucher $5 voucher Research site to choose either $15 or $ baby bundle	3 3 1 0 -	1 2 - - 5*	.١

* Rule enacted, highest frequency accepted if consensus not achieved in Round 3

Table 3

Consensus for outcomes for acute respiratory symptoms, health care utilisation, and exposure to tobacco and breastfeeding status from 1 to 6 months of age

Item	Round 2 n = 8	Round 3 n = 8	Consensus
Has your baby had wheeze or whistle in the past 4 weeks?	4	7	ו
Has your baby had a moist or wet cough in the past 4 weeks?	6	7	ו
Has your baby had a dry cough in the past 4 weeks?	6	7	ו
Has your baby had shortness of breath in the past 4 weeks?	4	7*	ו
Has your baby had an earache in the past 4 weeks?	4	7*	ו
Has your baby had a runny nose in the past 4 weeks?	4	7*	ו
Does your baby have a cough today?	6	5*	ו
Have you been worried about your baby's health for any reason in the past 4 weeks?	5	7*	ו
If yes, what have you been worried about?	4	8*	ו
Has your baby been hospitalised in the past 4 weeks?	6	7*	ו
If yes, what were the reasons your baby went to hospital?	5	7*	ו
If yes, how many days was your baby hospitalised?	6	7*	ו
Has your baby been to see a doctor at any time in the past 4 weeks?	5	7*	ו
If yes, what were the reasons?	5	7	ו
Has your baby been given medications in the past 4 weeks?	6	7*	ו
Has exposure to tobacco smoke changed?	7	-	ו
Has breastfeeding changed in the past 4 weeks?	6	8*	ו
Any out of pocket expenses to care for your baby’s sickness?	4	3	ן
Has your baby had any feeding difficulties in the past 4 weeks?	4	3	ן
Has your baby had a fever/temp/feel hot in the past 4 weeks?	2	-	ן
Has your baby had chills in the past 4 weeks?	1	-	ן
Has your baby vomited in the past 4 weeks?	1	-	ן

* Rule of combining ‘very essential’ and ‘somewhat essential’ enacted

Item	Round 2 n = 8	Round 3 n = 8	Consensus
Has your baby had diarrhea in the past 4 weeks?	1	-	⌋
Has your baby had irritability in the past 4 weeks?	0	-	⌋
Has your baby had increased tiredness in the past 4 weeks?	0	-	⌋
Has your baby had unsettled sleep in the past 4 weeks?	0	-	⌋
Has your baby had fast breathing in the past 4 weeks?	4	0	⌋
How many days has your baby had the cough for?	6	6	⌋
Are you worried about your baby's cough becoming worse?	5	1	⌋
What is your baby's cough like in daytime?	5	0	⌋
What is your baby's cough like in night time?	5	0	⌋
Total number of days the baby was in hospital.	3	-	⌋
Anything else that affects your family getting health care for your baby?	4	3	⌋
If yes, how many times has the baby been to the doctor?	3	-	⌋
Total number of days baby was in hospital	3	-	⌋
Amount of time spent from work/home to get health care for baby?	3	-	⌋
How many hours per week have been spent getting health care for your baby?	1	-	⌋
Has your baby been given antibiotics in the past 4 weeks?	6	1	⌋
What is the name of the hospital?	0	-	⌋
Has any person in the baby's household had a respiratory illness?	2	-	⌋
Has your baby seen any other health professional?	5	4	⌋
How many times has your baby been to see the health professional?	3	5	⌋
Reason (s) baby seen by other health professional	3	7	⌋
Total	43	28	17
* Rule of combining ‘very essential’ and ‘somewhat essential’ enacted			

Round Three: Questionnaire

Thirty-one items were presented in total. Of the three items presented on how data should be collected, number of questions was 5 to 10, site to choose personnel to collect data and site to choose $15 gift card or $15 baby bundle. Of the 28 measures to be collected presented in round three, 17 were accepted. Five items reached consensus by achieving a response frequency of ≥ 80% and twelve items reached consensus through enacting the rule to combine votes for ‘very essential’ and ‘somewhat essential’. Items accepted include seven acute respiratory symptoms, two general health items, six items on health care utilisation, one item on exposure to tobacco smoke and one item on breastfeeding status. Additional recommendations from the panel were to provide families and health providers with education on detecting and managing chronic cough, and to ensure adequate follow up of infants with chronic cough.

Measures for respiratory illness and development for 6 months old infants

Round One

Teleconference
Five measures were discussed, 1) 50-item parent report respiratory symptom screening questionnaire (31), 2) 18-item respiratory screening questionnaire adapted into Creole (32), 3) a clinical assessment form developed for the purpose of the larger study, 4) Ages and Stages Questionnaire (ASQ) (33) and 5) an adapted version of ASQ for remote Indigenous communities, ASQ-TRAK (34)). Participants were not aware of any other suitable measures or existing surveys.

Round Two: Questionnaire

Of the five assessments tools, none reached consensus for use in the existing form. Qualitative feedback from the panel recommended a shorter length questionnaire. The questionnaire adapted into Creole language from the Torres Strait was not considered suitable for most Indigenous women. Participants recommended specific language changes or inclusion of definitions for words such as 'posset', 'wheeze' and, 'rattles/ruttles'. Minor feedback was received on the clinical assessment form including a recommendation to ask more broadly about a child’s respiratory health and then use prompts for specific respiratory conditions, e.g. bronchitis.

Five of eight participants indicated it was important to collect developmental outcomes at six months and five of eight indicated that the ASQ and ASQ TRAK were suitable tools. Key feedback on how the data should be collected included: a health professional should complete it with the woman and infant, the health professional must be familiar with working in Indigenous communities, and the questionnaire should be completed prior to a clinical assessment and the results provided to the clinical assessor.

Community responses from focus groups:

Overall feedback from the Indigenous women indicated a preference for the 50-item questionnaire compared to the 18-item questionnaire adapted into Creole. There was an overwhelming consensus to shorten the length and clarify certain terms, such as ‘posset’ and ‘rattly breathing’. Similar to the

Indigenous panel, women advised that the Creole language was only suitable for Indigenous people who speak Torres Strait Creole. Women also recommended a simpler layout, particularly if surveys are to be parent completed.

Round Three

Based on the feedback gathered from participants, several changes were made to the 6 months of age questionnaires presented in round three. The 50-item questionnaire was reduced to 33- items. The clinical assessment form was reduced to one page and included growth parameters, immunisations, respiratory illnesses since birth, other significant illness since birth, and current medications. The clinical assessment form was recommended to be completed with information extracted from the clinical notes and parent report. A consensus from participants, 8/8 (100%), was achieved for use of the three assessment tools in their amended form.

Discussion

A modified Delphi process was completed with eight Indigenous experts, and focus groups were conducted with 18 Indigenous women about culturally safe measures for Infant respiratory health. To our knowledge, this is the first consensus-based study on measures for detecting respiratory illness in Indigenous Australian infants. Measures that reached consensus included 15 measures at birth, 17 measures from 1 to 6 months of age, and three questionnaires to be used at 6 months of age. The preferred mode for data collection differed for the different time points. Consensus was reached that birth measures should to be collected via a hospital discharge summary, 1 to 6 month measures via parent report with mode decided by woman i.e. phone call, mobile phone application, or online survey and 6 months of age measures collected using parent report questionnaires completed with a trusted health professional in conjunction with clinical notes.

All measures at birth (15/15) were accepted for inclusion. The high rate of inclusion might be due to the standard nature of measures and minimal burden to participating women, as data would be collected via discharge summary. Measures collected from 1 to 6 months consisted of mainly acute respiratory symptoms (7/17) and health care utilisation (6/17). The result was a monthly self-report survey with a minimum of 13 and maximum 17 items. Of the 17 items, only five items were accepted for inclusion in rounds two and three. These five items were ‘wheeze/whistle’, ‘moist/wet/cough’, ‘dry cough’, ‘reasons for seeing a doctor’ and ‘change in exposure to tobacco smoke’. The remaining 12 items were included based on a rule to combine ‘very essential’ and ‘somewhat essential’ votes. The five initial items may indicate items the panel considered most essential to measure; they also are well aligned with the literature. Wheeze is the most reliable symptom to detect asthma (36) and wet cough for bronchiectasis (4, 6, 37). Seeing a doctor may indicate severity, and exposure to environmental tobacco smoke during infancy doubles the risk of hospitalisation for respiratory illness in infancy (38), so an important variable to collect.

Two potential respiratory questionnaires for use at 6 months of age were presented to the panel, 1) a 50-item questionnaire developed for British infants (39) and 2) a 20-item questionnaire adapted for Torres Strait Islander infants (32). It was consistent between the panel and women in the focus groups that Torres Strait Creole is not suitable for most Indigenous women, though a questionnaire with fewer items was preferred. The language of the 50 item questionnaire was largely understood and accepted by women, which is unsurprising as it stems from the ISAAC protocol which has been tested in 97 countries (40). The 50-item questionnaire was ultimately shortened to 33 items based on feedback. A developmental screening measure, the Ages and Stages questionnaire (41) as well as the adapted version for remote Indigenous communities (34) were also presented to the panel. Interestingly all panel members indicated inclusion of a measure on child development, when not typically measured in studies on respiratory health. The strong interest to include a developmental measure raises the question of what other measures may be important, and perhaps more meaningful to Indigenous communities. Other less commonly reported measures in child respiratory studies include child parent quality of life (42, 43) and child functioning (44).

This study had several limitations. The involvement of Indigenous women was limited. Women participated in one focus group to provide feedback on one type of measure (6 months of age respiratory questionnaires); we did not obtain final feedback from women on changes made to the questionnaire recommended by the expert panel (removal of 17 items). The measures identified in this study may be more confidently used if greater end user involvement had occurred (45). While we strongly acknowledge the importance of end-user involvement, the focus here was to gain expert consensus from Indigenous academics and clinicians on essential respiratory measures, future studies should place emphasis on pre-testing the identified measures with end-users from a range of communities. A second limitation was that findings may not be generalisable to the diversity of Indigenous peoples of Australia. While panel members were from different regional, remote and urban communities, the number of panel members was relatively small (8) and women were from NSW communities only. A third and important limitation was that the measures identified focus on a rather short period in a child's life, birth to 6 months of age. The 6 months age range was of focus as it is the follow-up period of the larger trial. As many chronic respiratory illnesses only develop later in childhood and are uncertain in infancy, e.g. asthma and bronchiectasis, accepted measures for use throughout childhood are needed. Lastly, if further rounds of consensus were completed the number of items may have been reduced, which can result in higher response rates for trials (46). An important consideration to be examined if pre-testing of measures.

The strength of this study was the engagement of Indigenous experts from several disciplines to work together and identify a comprehensive set of respiratory measures in the context of cultural safety for Indigenous infants. Knowledge was generated with Indigenous academics, clinicians and women to optimise the cultural safety of data collection in a trial examining infant respiratory outcomes. The measures identified are for a number of time points in the first 6 months of life using a range of sources (medical records, parent report and observation). A range of sources is important given the known pitfalls of relying on any one of these sources alone (15).

A modified Delphi process may be a useful method to systematically involve Indigenous people in decisions for trials. The Delphi has been used in other areas of Indigenous health research including to develop mental health guidelines (21) and data collection strategies for maternity experiences (22). Other high-level consultative methods to develop measures for use with Indigenous people have also been used. A recent example is the development of a survey for the Mayi Kuwayu Study, a national longitudinal study on adult Indigenous Australian well-being (47). Consultation was completed with 165 Indigenous peoples attending 24 focus groups across Australia from 2014 to 2017. Pilot testing of the survey was completed with 160 and 209 Indigenous participants. A second example is the Healing the Past by Nurturing the Future study, a study in part to develop a measure to identify complex trauma experienced by Indigenous parents (48). Consultation includes four large-scale co-design workshops across three States with Indigenous parents, service providers, community leaders, researchers and wider community members. Comprehensive consultation is expected from conception to conclusion in research with Indigenous peoples (49). With varying methods and approaches for consultation, a Delphi methodology is one approach that can provide a systematic, transparent and feasible process for expert consensus in trials.

The Indigenous panel that participated in the consensus process made two important unexpected recommendations that may aid more accurate data collection and increase recruitment and retention in trials. The first was to provide education to participating families and health providers on respiratory symptoms and management pathways. This recommendation aligns with a recent qualitative study with 40 Indigenous community members reporting 70% considered chronic cough normal in children (50). By providing culturally appropriate definitions on respiratory terms such as wheeze and wet cough, and information on the importance of seeking treatment, the accuracy of parent report may improve and lead to better disease detection and optimal treatment (51). The second recommendation was to provide adequate follow up of participating infants. Cough guidelines recommend children aged 14 years or less with a chronic cough of 4 weeks should have a chest radiograph and spirometry test (when age appropriate) (52). In research studies on infant respiratory health, we have opportunity and ethical responsibility (49) to ensure that children receive adequate treatment during and on study completion. Studies designed with a reciprocal approach including assured access to quality treatment may improve retention rates, as in a recent study on incidence of respiratory illness in Queensland (12).

This is a preliminary step in developing a set of standard measures to detect respiratory illness in community based Indigenous infants. Future research is needed to test the validity and the reliability of the identified measures for use in trials and practice. Additional considerations for pre and pilot testing these measures may include information for families to combat the normalisation of respiratory illness (53), flexible mode of delivery given the many other needs and problems Indigenous families experience (20), and trusted and skilled interviewers to ensure cultural safety.

Conclusions

A modified Delphi process with Indigenous multi-disciplinary experts determined culturally safe measures to identify respiratory illness in Indigenous infants from birth to six months of age. We set out to develop a set of measures that would meet the needs of families, clinicians and researchers that were culturally safe and feasible. In total, 15 items for birth, 17 items from 1 to 6 months and 3 surveys for use at 6 months of age were identified. Future studies are required to assess the validity and reliability of and participation in surveys using these relevant and acceptable measures.

Abbreviations

IAHA -, Indigenous Allied Health Australia

Australian Indigenous HealthInfoNet

CATSINaM - The Congress of Aboriginal and Torres Strait Islander Nurses and Midwives

NICU - Neonatal Intensive Care Unit

ASQ - Ages and Stages Questionnaire

Declarations

Ethics approval and consent to participate

This consultation process was part of a larger study, SISTAQUIT (Supporting Indigenous Smokers To Assist Quitting): a Cluster Randomised Controlled Trial to Improve Strategies for the Management of Smoking Cessation in Pregnant Aboriginal and/or Torres Strait Islander Women. The consultation was approved by The University of Newcastle Human Research Ethics Committee (HREC Ref H-2015-0438) and several other HRECs. The HRECs approved that a committee would be convened to consult on the collection of the infant outcomes, and the recommended outcome measures were later accepted into the protocol by the ethics committees. Ethical approval for participation of Indigenous women in focus groups was obtained as part of a separate study from The University of Newcastle HREC (REF H-2017-0247) and the New South Wales Aboriginal Health and Medical Research Council (AH&MRC) (1303/17) HREC. Written consent was obtained from women.

Consent for publication

Consent for publication obtained from women who attended focus groups using institution consent forms. A copy of the manuscript was sent to all panel members for review prior to submission. Written consent was obtained from all panel members for publication.

Availability of data and materials

The datasets used and/or analysed during the current study available from the corresponding author on reasonable request.

Competing interests

The authors declare that they have no financial or non-financial competing interests.

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Authors' contributions

All authors contributed to the design of the study including surveys used during the study. SP collected and analysed the results and wrote all manuscript drafts. JM provided expertise on respiratory health items. KH provided expertise on respiratory health and cultural guidance. BB, GG, JM, and KH provided supervision to PhD candidate SP throughout study. All authors read, edited and approved the final manuscript.

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Figures

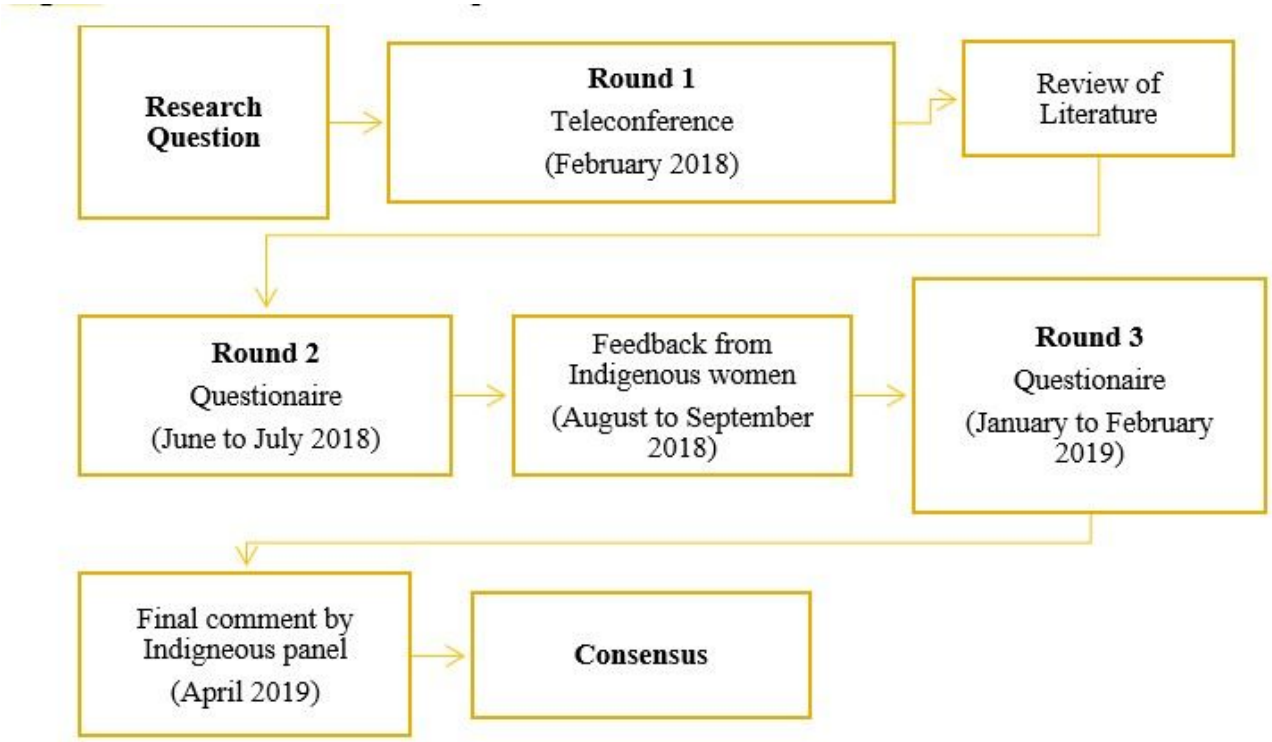

Figure 1

Overview of consensus process