Supplementary Appendix 1: Ethically conscious framework for public involvement at the research design stage

Supplement to: Harding, S., Samways, B., Dillon, A., Butcher, S., Boyd, A., Mukherjee, R., Cook, P.A., McQuire, C. Establishing a national linked database for fetal alcohol spectrum disorder (FASD) in the UK: multi-method public and professional involvement to determine acceptability and feasibility.

The framework items presented below follow recommendations from: *Pandya-Wood R, Barron DS, Elliott J. A framework for public involvement at the design stage of NHS health and social care research: time to develop ethically conscious standards. Res Involv Engagem. 2017;3:6.*

1) ***Allocating sufficient time for public involvement***

Our PPI activities took place over a 13-month period, to allow time for meaningful and detailed discussions with different stakeholder groups. We designed our discussion guides for the workshops that involved people with FASD and their supporters to minimise the number of questions asked within the timeframe, while still ensuring that all important topics were covered. This was to allow as much time as possible for contributors to process the information for each question and contribute without excessive time pressure. We allowed 4 hours (including breaks) for our clinical workshop to allow plenty of time for discussion.

2) ***Avoiding 'tokenism'***

In the accompanying article, we describe precisely how our public contributors were involved in shaping our ideas for development of a national FASD database, what their suggestions were, and how these suggestions will be incorporated as we develop our future research plans.

3) **Registering of research design stage public involvement work early with NHS Research and Development (R&D) Trust Office**

We sought advice from the National Institute for Health and Care Excellence Research Design Service South West (NIHR RDS) on the conduct and reporting of this PPI work. We did not register this public involvement work with NHS R&D Trust Offices. This was because involvement of the public was not due to occur NHS premises (therefore NHS indemnity issues were not relevant) and because our funding came from University of Bristol research institutes rather than, for example, the RDS (Research Design Service) Public Involvement Fund. However, involving NHR R&D offices at an earlier stage is certainly something that we will consider for future PPI work.

4) ***Communicating clearly from the outset***

All contributors were provided with written information about the format and purpose of our PPI activities in advance and were given email, telephone and X/Twitter contact details to enable direct messaging to the project team, should they want to discuss anything further. This information was tailored to be as clear as possible for each stakeholder group.

i Workshops for people with FASD and their caregivers: We developed information sheets in collaboration with the National Organisation for FASD (SB), and an adult with FASD and their parent. This ensured that the language used was as accessible as possible to contributors. We made use of visual prompts, which we co-designed with the National Organisation of FASD, to provide a summary of our planned research, and provided pictures of who would be facilitating the workshops. At the start of each workshop the facilitators (CM and SB) introduced themselves, explained the purpose and format of the meeting, including considerations around confidentiality, and provided the attendees with opportunities to ask any further questions.

ii Clinical stakeholders: Relevant stakeholders, identified through our mapping and stakeholder identification processes (described in more detail in the main manuscript) were contacted by the project team via email or webform, explaining the purpose and format of involvement opportunities, which included telephone/video call and hybrid workshop formats. They were invited to express their interest in (or decline) involvement and were provided with the contact details of the project team to discuss any further queries.

iii Other professional stakeholders (e.g. policy/data contacts). All were approached by the project team via email or webform in the first instance. Emails explained the purpose of this PPI project, the format of involvement opportunities, and the contact details of the project team for further information.

5) ***Entitling public contributors to stop their involvement for any unstated reason(s)***

All contributors were informed that they could stop their involvement at any point and that they did not have to answer any questions that they did not want to.

6) ***Operating 'fairness of opportunity'***

We carried out stakeholder mapping to create a comprehensive summary of all key public and professional stakeholder groups. We tailored our approach, in collaboration with the National Organisation of FASD, to maximise fairness of opportunity for all stakeholder groups. This included enabling adults with FASD to bring a supporter with them to a workshop, if preferred; scheduling discussion groups for times that were most suitable for maximising participation. For example, workshops with people with FASD and their supporters took part in the evening to avoid excluding those who worked during the daytime; scheduling the clinical workshop to take place as a 'pre-conference' event, coinciding with the FASD in the UK conference to maximise in-person attendance at the conference location. We provided contributors with multiple ways

to contact the project team (email, telephone, social media) to suit contact preferences.

7) ***Differentiating between public involvement activities and qualitative research methods***

We made it clear to all contributors that this was a PPI project and not a qualitative research study. We communicated this through our written materials (e.g. information sheets, email templates) and verbally, during meetings/workshops. For example, we used the terms 'research advisory group' or 'workshop' to refer to group discussions, rather than focus groups. Our discussion guides focused on gathering information to inform the design of future proposals to establish a National Database for FASD, rather than seeking to answer research questions.

We referred to National Institute for Health and Care Research (NIHR) guidance, which outlines the key differences between PPI and qualitative research [1] and consulted with the NIHR Research Design Service South West to ensure that our project adhered to definitions of PPI, rather than straying into qualitative research.

8) ***Working sensitively***

We are aware that FASD can be an emotive topic, particularly for those living with FASD and their caregivers. Therefore, we designed our question guides for PPI activities to be as sensitive as possible to minimise the risk of causing emotional distress to stakeholders. This included making sure that all of our discussions focused on the design, acceptability, feasibility and perceived risks/benefits of a FASD database only, rather than seeking information about the lived experience of FASD. Discussion guides for public workshops were developed in collaboration with the National Organisation for FASD, and an adult with FASD and their parent. This ensured that the language and content were appropriate for our lived experience contributors. All contributors were assured that discussions were confidential and that they did not have to answer any questions that they did not want to. Workshops with lived experience contributors were co-facilitated by the project lead (CM) and lead of the National Organisation of FASD (SB), both of whom have extensive experience of carrying out interviews/workshops with public contributors, including people living with FASD. Adults with FASD were able to bring a supporter with them to the online workshop, if preferred. This further ensured that our PPI activities were conducted sensitively. Smaller groups were deemed appropriate for meaningful collaboration with people with FASD, smaller sessions are most appropriate to allow interviewers to ensure the participants are following the discussion and have ample opportunity to respond in a low stress environment given their cognitive and neurodevelopmental challenges.

9) ***Being conscious of confidentiality***

We informed all stakeholders that discussions would be confidential, that these may be recorded by the project team for note-taking purposes, that anonymised content from the PPI activities may also be used in project outputs, and that they could opt out of being included in the recording and/or any outputs.

10) ***Valuing, acknowledging and rewarding public involvement***

All public contributors were reimbursed for their time with shopping vouchers of their choice. Professional contributors were reimbursed for all travel expenses and provided with lunch and refreshments during the workshop. Lived experience and professional contributors both commented on the value of these involvement opportunities for providing the opportunity to meet/network with others in similar situations. We have acknowledged the valuable contribution of our public and professional stakeholders in all related outputs and continue to include them in updates/discussions/opportunities related to development of this database, as appropriate.

[1] National Institute for Health Research. A brief guide to patient and public involvement and qualitative methods within health and social care research. Available at https://www.rds-se.nihr.ac.uk/wp-content/uploads/RDS_Guide_to_PPI.pdf. Accessed 19.06.2024.

Supplementary Appendix 2

Data proforma: UK National FASD Database v1 (20.03.23) - short form*

__Note: this version of the data capture proforma was unintentionally abbreviated, such that information about neurodevelopment and growth were captured as free text under Q33 rather than standardised tick box form. 3 out of 6 participating clinics used this version of the form.__

Thank you for your interest in our project, which investigates the feasibility of establishing a UK National FASD Database. As part of this process, we would like to understand what data on FASD is available. To support this, we would be grateful if you could complete this form. We expect that it will take no longer than 15-20 minutes to complete. You do not have to answer any questions that you do not want to.

For this exercise, we are asking for you to indicate whether you collect certain information during FASD assessments, and whether you would potentially share this information (subject to appropriate governance procedures being in place). We are not requesting any information about individual patients. The goal is to understand the nature of the assessments and data collected in clinic. This exercise is just to gauge willingness to share data and that there is no commitment at this stage.

If you are attending the FASD Database Workshop on 29th March 2023, either online or in-person, we would be very grateful if you could complete this form by Monday 27th March 2023. This will help us to shape the workshop based on what you tell us.

The responses on this form will be used to help inform the next stages of our proposal, as we develop plans for the UK FASD Database. They may also form the basis of outputs from this project. Any information that we include in future outputs will be anonymised. You can opt out if you do not want any of your responses included in project outputs. Please email cheryl.mcquire@bristol.ac.uk if you wish to opt out.

Many thanks again for your support with this project

Below is an example of a data form. We would be very grateful if you could indicate whether you currently collect data on each of the measures below and, if not, whether you would consider collecting this data in the future. We also ask whether you might be willing to share this data. Please note that any processes for sharing data will be secure and will follow strict governance measures with trusted data managers.

1.Name of clinic:	**2.Date completed:**
3.Name of respondent(s):	**4.Respondent(s) email:**
5.Which diagnostic criteria do you use (e.g. SIGN 156, Canadian, 4-Digit Code, other (please specify):	
6.How many FASD assessments do you carry out each year (estimate if not sure):	
7.How many people referred for FASD assessment do you have data on (estimate if not sure):	
8.How do you store the assessment information of people with FASD (e.g. paper records, electronic, other [please specify]):	
9.Date range for the data you hold on FASD (e.g. 2000 – present [estimate if not sure]):	
10.Who is responsible for data sharing approvals in your organisation (e.g. NHS Caldicott Guardian, clinic director, R&D team, other (please specify):	
Do you share data on the person with FASD with other organisations (e.g. GP, education, social care, other [please specify): Yes/no	

If you answered 'yes' to the previous question what data do you share? (e.g. FASD diagnosis, needs assessments, education, health and care (EHC) plan):								
	Data currently collected?		**Would you consider collecting this data in the future?**		**Would you be willing to share this data (subject to robust governance)?**		**Which measures do you currently use? (put NA if not applicable)**	**Notes**
	Yes	**No**	**Yes**	**No**	**Yes**	**No**		
11.Participant ID / NHS number	☐	☐	☐	☐	☐	☐		
12.Year of diagnosis	☐	☐	☐	☐	☐	☐		
13.County	☐	☐	☐	☐	☐	☐		
14.Date of Referral	☐	☐	☐	☐	☐	☐		
15. The data collected is:								
• Retrospective	☐	☐	☐	☐	☐	☐		
• Prospective	☐	☐	☐	☐	☐	☐		
• Both (e.g. re-assessment)	☐	☐	☐	✓	☐	☐		

	Data currently collected?		**Would you consider collecting this data in the future?**		**Would you be willing to share this data (subject to robust governance)?**		**Which measures do you currently use? (put NA if not applicable)**	**Notes**
	Yes	**No**	**Yes**	**Yes**	**No**	**Yes**		
16.Reason for referral: E.g., Behavioural issues, Learning difficulties, Issues with the law, Developmental delays/or delays to meet developmental milestones, Transition to adulthood, Other_____	☐	☐	✓	☐	☐	☐		
17.Source of Referral For example, Social Agency, Medical Referral, Education System, Legal System, Self, Other _____	☐	☐	☐	☐	☐	☐		
18.Current Living Situation E.g.: Independent, with biological mother / father, with kin, foster care, adoptive parent(s), group home, homeless, in custody, etc	☐	☐	☐	☐	☐	☐		
19.Gender	☐	☐	☐	☐	☐	☐		
20.Date of Birth	☐	☐	☐	☐	☐	☐		

Sibling and parent diagnosis								
	Data currently collected?		Would you consider collecting this data in the future?		Would you be willing to share this data (subject to robust and appropriate governance)?		Which measures do you currently use?	Notes
	Yes	No	Yes	No	Yes	No		
21.Has a sibling been diagnosed with FASD?	☐	☐	☐	☐	☐	☐		*If so, specify if this is biological sibling, foster/adopted/unknown*
22.Has a biological parent been diagnosed with FASD?	☐	☐	☐	☐	☐	☐		
Prenatal alcohol exposure								
	Data currently collected in assessments?		Would you consider collecting this data in future assessments?		Would you be willing to share this data (subject to robust governance)?		Which measures do you currently use?	Notes
	Yes	No	Yes	No	Yes	No		
23.Prenatal alcohol exposure	☐	☐	☐	☐	☐	☐		

Sentinel facial features								
	Data currently collected in assessments?		Would you consider collecting this data in future assessments?		Would you be happy to share this data?		Which measures do you currently use?	Notes
	Yes	No	Yes	No	Yes	No		
24.Palpebral fissure length	☐	☐	☐	☐	☐	☐		
25.Philtrum smoothness	☐	☐	☐	☐	☐	☐		
26.Upper lip thinness	☐	☐	☐	☐	☐	☐		
27.Total number of sentinel facial features present	☐	☐	☐	☐	☐	☐		
28.How do you measure the facial phenotype e.g. manual with ruler, 2D photographic screening, 3D photographic screening, other (please specify)								
29.Do you retain images from facial screening and, if so, in what format?								

29.Are patients are sent for genetic testing?	**Yes/No/Unsure:**
If yes:	
30.Which organisation carries out the testing?	
31.Do you have data for the results of this?	
32.Is this data is held by the organisation that does the testing or by your clinic/pathway?	

33.Please use the section below to let us know about any other information that you hold if relevant to FASD

Information you collect	**Would you be willing to share this data (subject to appropriate governance)?**	**Which measures do you currently use for this?**

Data proforma: UK National FASD Database v2 (28.03.23) - full version*

Note: this is the full version of our data capture proforma that includes previously omitted items on neurodevelopment and growth. *3 out of 6 participating clinics used this version of the form.

Data Form

UK National FASD Database

Thank you for your interest in our project, which investigates the feasibility of establishing a UK National FASD Database. As part of this process, we would like to understand what data on FASD is available. To support this, we would be grateful if you could complete this form. We expect that it will take no longer than 15-20 minutes to complete. You do not have to answer any questions that you do not want to.

For this exercise, we are asking for you to indicate whether you collect certain information during FASD assessments, and whether you would potentially share this information (subject to appropriate governance procedures being in place). We are not requesting any information about individual patients. The goal is to understand the nature of the assessments and data collected in clinic. This exercise is just to gauge willingness to share data and that there is no commitment at this stage.

If you are attending the FASD Database Workshop on 29th March 2023, either online or in-person, we would be very grateful if you could complete this form by Monday 27th March 2023. This will help us to shape the workshop based on what you tell us.

The responses on this form will be used to help inform the next stages of our proposal, as we develop plans for the UK FASD Database. They may also form the basis of outputs from this project. Any information that we include in future outputs will be anonymised. You can opt out if you do not want any of your responses included in project outputs. Please email cheryl.mcquire@bristol.ac.uk if you wish to opt out. Many thanks again for your support with this project.

Below is an example of a data form. We would be very grateful if you could indicate whether you currently collect data on each of the measures below and, if not, whether you would consider collecting this data in the future. We also ask whether you might be willing to share this data. Please note that any processes for sharing data will be secure and will follow strict governance measures with trusted data managers.

1.Name of clinic:	**2.Date completed:**
3.Name of respondent(s):	**4.Respondent(s) email:**
5.Which diagnostic criteria do you use (e.g. SIGN 156, Canadian, 4-Digit Code, other (please specify):	
6.How many FASD assessments do you carry out each year (estimate if not sure):	
7.How many people referred for FASD assessment do you have data on (estimate if not sure):	
8.How do you store the assessment information of people with FASD (e.g. paper records, electronic, other [please specify]):	
9.Date range for the data you hold on FASD (e.g. 2000 – present [estimate if not sure]):	
10.Who is responsible for data sharing approvals in your organisation (e.g. NHS Caldicott Guardian, clinic director, R&D team, other (please specify):	
Do you share data on the person with FASD with other organisations (e.g. GP, education, social care, other [please specify): Yes/no	

If you answered 'yes' to the previous question what data do you share? (e.g. FASD diagnosis, needs assessments, education, health and care (EHC) plan):								
	Data currently collected?		Would you consider collecting this data in the future?		Would you be willing to share this data (subject to robust governance)?		Which measures do you currently use? (put NA if not applicable)	Notes
	Yes	No	Yes	No	Yes	No		
11.Participant ID / NHS number	☐	☐	☐	☐	☐	☐		
12.Year of diagnosis	☐	☐	☐	☐	☐	☐		
13.County	☐	☐	☐	☐	☐	☐		
14.Date of Referral	☐	☐	☐	☐	☐	☐		
15. The data collected is:								
• Retrospective	☐	☐	☐	☐	☐	☐		
• Prospective	☐	☐	☐	☐	☐	☐		
• Both (e.g. re-assessment)	☐	☐	☐	☐	☐	☐		

	Data currently collected?		Would you consider collecting this data in the future?		Would you be willing to share this data (subject to robust governance)?		Which measures do you currently use? (put NA if not applicable)	Notes
	Yes	No	Yes	Yes	No	Yes		
16.Reason for referral: E.g., Behavioural issues, Learning difficulties, Issues with the law, Developmental delays/or delays to meet developmental milestones, Transition to adulthood, Other_____	☐	☐	☐	☐	☐	☐		
17.Source of Referral For example, Social Agency, Medical Referral, Education System, Legal System, Self, Other _____	☐	☐	☐	☐	☐	☐		
18.Current Living Situation E.g.: Independent, with biological mother / father, with kin, foster care, adoptive parent(s), group home, homeless, in custody, etc	☐	☐	☐	☐	☐	☐		
19.Gender	☐	☐	☐	☐	☐	☐		
20.Date of Birth	☐	☐	☐	☐	☐	☐		

Sibling and parent diagnosis								
	Data currently collected?		Would you consider collecting this data in the future?		Would you be willing to share this data (subject to robust and appropriate governance)?		Which measures do you currently use?	Notes
	Yes	No	Yes	No	Yes	No		
21.Has a sibling been diagnosed with FASD?	☐	☐	☐	☐	☐	☐		*If so, specify if this is biological sibling, foster/adopted/unknown*
22.Has a biological parent been diagnosed with FASD?	☐	☐	☐	☐	☐	☐		
Prenatal alcohol exposure								
	Data currently collected in assessments?		Would you consider collecting this data in future assessments?		Would you be willing to share this data (subject to robust governance)?		Which measures do you currently use?	Notes
	Yes	No	Yes	No	Yes	No		
23.Prenatal alcohol exposure	☐	☐	☐	☐	☐	☐		

Sentinel facial features								
	Data currently collected in assessments?		Would you consider collecting this data in future assessments?		Would you be happy to share this data?		Which measures do you currently use?	Notes
	Yes	No	Yes	No	Yes	No		
24.Palpebral fissure length	☐	☐	☐	☐	☐	☐		
25.Philtrum smoothness	☐	☐	☐	☐	☐	☐		
26.Upper lip thinness	☐	☐	☐	☐	☐	☐		
27.Total number of sentinel facial features present	☐	☐	☐	☐	☐	☐		
28.How do you measure the facial phenotype e.g. manual with ruler, 2D photographic screening, 3D photographic screening, other (please specify)								
29.Do you retain images from facial screening and, if so, in what format?								

Neurobehavioural Assessment								
	Data currently collected in assessments?		Would you consider collecting this data in future assessments?		Would you be happy to share this data?		Which measures do you currently use?	Notes
	Yes	No	Yes	No	Yes	No		
30. Brain Domain Assessment Results (please indicate for each of the domains below)	☐	☐	☐	☐	☐	☐		
Motor Skills	☐	☐	☐	☐	☐	☐		
Neuroanatomy / Neurophysiology	☐	☐	☐	☐	☐	☐		
Cognition	☐	☐	☐	☐	☐	☐		
Language	☐	☐	☐	☐	☐	☐		
Academic Achievement	☐	☐	☐	☐	☐	☐		
Memory	☐	☐	☐	☐	☐	☐		
Attention	☐	☐	☐	☐	☐	☐		
Executive Function, including impulse control	☐	☐	☐	☐	☐	☐		
Affect Regulation	☐	☐	☐	☐	☐	☐		
Adaptive Behaviour, Social Skills, Social Communication	☐	☐	☐	☐	☐	☐		
IQ	☐	☐	☐	☐	☐	☐		
31. Growth								
Height	☐	☐	☐	☐	☐	☐		
Weight	☐	☐	☐	☐	☐	☐		

Diagnosis								
	Data currently collected in assessments?		Would you consider collecting this data in future assessments?		Would you be happy to share this data?		Which measures do you currently use?	Notes
	Yes	No	Yes	No	Yes	No		
32. Diagnosis **e.g.:** FASD with/ without Sentinel Facial Features; At Risk for Neurodevelopmental Disorder and FASD, Associated with Prenatal Alcohol Exposure No FASD diagnosis Other	☐	☐	☐	☐	☐	☐		
	Data currently collected in assessments?		Would you consider collecting this		Would you be happy to share this data?		Which measures do you currently use?	Notes
			data in future assessments?					
	Yes	No	Yes	No	Yes	No		
32. Other Associated Features (From a checklist) e.g. Sleep problems; Sensory sensitivities; Attachment, etc.	☐	☐	☐	☐	☐	☐		
33. Co-morbidities (from a checklist) e.g.: ADHD; Genetic disorder Intellectual disability, etc.	☐	☐	☐	☐	☐	☐		
34. Other diagnoses previously given and accepted (from a checklist)	☐	☐	☐	☐	☐	☐		
35. Medical Health Diagnosis History (from a checklist) e.g. Gastrointestinal, Musculoskeletal, mental health problems etc.	☐	☐	☐	☐	☐	☐		

	Data currently collected in assessments?		Would you consider collecting this data in future assessments?		Would you be happy to share this data?		Which measures do you currently use?	Notes
	Yes	No	Yes	No	Yes	No		
36. A list of all current medications	☐	☐	☐	☐	☐	☐		
37. Whether and which substances are currently being used/misused by the individual being assessed?	☐	☐	☐	☐	☐	☐		
38. Whether there is any intervention currently access for the above.	☐	☐	☐	☐	☐	☐		
39. Wider issues currently being experienced by the individual being assessed? (from checklist including) • Legal problems: Victim • Legal problems: Offender • Custody issues/family court • Employment problems • Support living needs • Jail • School expulsion/suspension	☐	☐	☐	☐	☐	☐		

	Data currently collected in assessments?		Would you consider collecting this data in future assessments?		Would you be happy to share this data?		Which measures do you currently use?	Notes
	Yes	No	Yes	No	Yes	No		
40. Which recommendations were made for the individual the individual being assessed? (from a checklist) e.g. • Coaching Support (individual or group) • Communication strategies • FASD assessment/early intervention Etc.	☐	☐	☐	☐	☐	☐		
41. Any additions, recommendations or comments.	☐	☐	☐	☐	☐	☐		

42. Are patients sent for genetic testing?	**Yes/No/Unsure:**
If yes:	
43.Which organisation carries out the testing?	
44.Do you have data for the results of this?	
45.Is this data is held by the organisation that does the testing or by your clinic/pathway?	

46.Please use the section below to let us know about any other information that you hold if relevant to FASD

Information you collect	Would you be willing to share this data (subject to appropriate governance)?	Which measures do you currently use for this?