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

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ABSTRACT

Introduction: Fetal alcohol spectrum disorder (FASD) is one of the leading non-genetic causes of developmental disability worldwide and is thought to be particularly common in the UK. Despite this, there is a lack of data on FASD in the UK.

Objective: To conduct public and professional involvement work to establish stakeholder views on the feasibility, acceptability, key purposes, and design of a national linked longitudinal research database for FASD in the UK.

Methods: We consulted with stakeholders using online workshops (one for adults with FASD [and their supporters] N=5; one for caregivers of people with FASD N=7), 1:1/small-team video calls/email communication with clinicians, policymakers, data-governance experts, third-sector representatives, and researchers [N=35]), and one hybrid clinical workshop (N=17). Discussions covered data availability, benefits, challenges, and design preferences for a national pseudonymised linked database for FASD. We derived key themes from the notes and recordings collected across all involvement activities.

Results: Our tailored, multi-method approach generated high levels of stakeholder engagement. Stakeholders expressed support for a pseudonymised national linked database for FASD. Key anticipated benefits were the potential for: increased awareness and understanding of FASD leading to better support; new insights into clinical profiles leading to greater diagnostic efficiency; facilitating international collaboration; and increased knowledge of the long-term impacts of FASD on health, social care, education, economic and criminal justice outcomes. Given the rich data infrastructure established in the UK, stakeholders expressed that a national linked FASD database could be world-leading. Common stakeholder concerns were around privacy and data-sharing and the importance of retaining space for clinical judgement alongside insights gained from quantitative analyses.

Conclusions: Multi-method and multidisciplinary public and professional involvement activities demonstrated support for a national linked database for FASD in the UK. Flexible, diverse, embedded stakeholder collaboration will be essential as we establish this database.

INTRODUCTION

Background

Fetal alcohol spectrum disorder (FASD) is one of the leading non-genetic causes of developmental disability worldwide [1]. It is thought to be particularly common in the UK, affecting an estimated 1.8 – 3.6% of children in the general population [2, 3]. Caused by prenatal alcohol exposure, FASD is associated with neurodevelopmental impairments, poor physical and mental health, substance misuse, and social problems across the lifespan [4-7]. Early diagnosis and support can significantly improve outcomes for those affected, as well as incurring significant cost savings for society [5, 6].

The full continuum of FASD consists of the subtypes 'FASD with sentinel facial features' (also known as Fetal Alcohol Syndrome [FAS]) and 'FASD without sentinel facial features'[8]. Individuals who have FASD with sentinel facial features represent a minority of cases (approximately 10%) and present with small palpebral fissure length, smooth philtrum, and thin upper lip. Since most individuals with FASD do not have these recognisable facial features, FASD has been described as a mostly 'invisible' or 'hidden' disability [3, 8-10].

The invisibility analogy is consistent also with the lack of accessible data on FASD in the UK. This inaccessibility presents a significant barrier when attempting to achieve important FASD research, policy and healthcare goals [11, 12]. There are several reasons why UK data on FASD are currently lacking. First, standardised diagnostic codes for the full FASD continuum (FASD with and without sentinel facial features) have not been readily available or used by clinicians. For example, healthcare providers in England are required to use the Systematized Nomenclature of Medicine Clinical Terms (SNOMED CT) for capturing clinical information [13]. Although SNOMED CT does include a code for the full spectrum of FASD, only a code for fetal alcohol syndrome (i.e. FASD with sentinel facial features) is currently available for use in one of the main electronic clinical systems used in UK care settings - EMIS Web (EMIS Group plc©). Since approximately 90% of individuals with FASD do not present with sentinel facial features [3, 14] FASD has been significantly underreported in routine data.

In April 2024, new SNOMED CT codes for FASD were introduced. These are Fetal Alcohol Spectrum Disorder with sentinel facial features (disorder); Fetal Alcohol Spectrum Disorder without sentinel facial features (disorder); and at increased risk for fetal disorder (finding)[15-17]. These codes represent a step forward in terms of representing the full FASD spectrum using nomenclature consistent with the latest National Institute for Health and Care Excellence (NICE) and Scottish Intercollegiate Guidance Network (SIGN) guidelines [8, 11]. However, further work is required to ensure that these codes are available for use in all major clinical coding systems.

The historic lack of relevant, accessible diagnostic codes for FASD are not the only barrier to the recording of FASD in routine data sources. Usage statistics show that SNOMED CT codes for FAS and prenatal alcohol exposure have not been commonly used in practice [18]. Research investigating the reporting of FASD using Hospital Episodes Statistics (based on ICD-11 coding [19]) has similarly found high levels of underreporting as FASD is not easily detectable at birth, is not often a primary reason for hospitalisation, and under-recognition

of FASD means that it is less likely to be coded as a co-existing condition [20, 21]. In addition, many clinicians report a lack of knowledge and confidence in making a FASD diagnosis and information on prenatal alcohol exposure is often missing or inaccurate, further complicating diagnosis. Perceived stigma, commonalities and comorbidities with other neurodevelopmental disorders, and the lack of a clearly defined care pathway have been cited as additional reasons for underdiagnosis and misdiagnosis of FASD [10, 13, 22-25].

There is a potential solution to addressing this critical FASD 'data gap'. In 2019 the Scottish Intercollegiate Guidance Network (SIGN) published landmark guidance on children and young people with prenatal alcohol exposure [8]. Since this publication, there has been a swathe of complimentary policy and guidance publications in the UK. Many are now in effect nationally, including a National Institute for Health and Care Excellence (NICE) Quality Standard for FASD [11] and Department of Health and Social Care (DHSC) Health Needs Assessment for FASD [14], among others [26, 27]. These publications represent a unified call for a step-change in provision for FASD, including increased prevention, awareness, understanding, diagnosis, and support [8, 11, 14, 26, 27]. In parallel, there is a drive for digital transformation across the UK to improve health and social care services. This includes the Government's 'Data Saves Lives' policy, which envisages a landscape of regional 'secure data environments' linking together electronic health and care records to enable analyses for the public good [28-34]. Data on FASD are currently being collected in some NHS and private clinical settings across the UK and publication of the aforementioned FASD guidance is anticipated to lead to a significant increase in diagnostic provision [26], with several new FASD clinics due to open in the near term. This could make a register utilising local NHS data feasible for the first time.

International exemplars are available. FASD databases in Australia, Canada and the USA have catalysed advances in diagnosis, treatment, prevention and support [35-40]. We aspire to achieve the same in the UK. Moving beyond international exemplars, established UK data linkage infrastructure offers potential for an electronic longitudinal linked database for FASD. This would be the first of its kind in the world. As highlighted by Health Data Research UK (HDR UK) "The UK is in a unique position to realise the potential of [routinely collected] health data, thanks to the NHS and its cradle-to-grave records for a population of over 65 million people" [41]. In this context, we have a crucial and timely opportunity to establish a world-leading data infrastructure for FASD in the UK that is efficient and standardised, can be linked to routine data, and which would be pivotal in informing, monitoring progress against, and achieving, (inter)national policy and research objectives [10, 11, 26, 42].

Aim and scope

Stakeholder support is crucial for the success of large-scale data research projects, such as this [43]. One way to achieve this is through early, sustained, and meaningful patient and public involvement (PPI) [44, 45]. In this paper, we follow the National Institute for Health and Care Research definition of PPI, as follows: "*research being carried out 'with' or 'by' members of the public rather than 'to', 'about' or 'for' them"* [46]. The work that we present in this paper represents the earliest stages of the research cycle model, namely: prioritising/shaping research, research design and grant development [46].

The primary aim of this PPI project was to work with relevant public (adults with FASD and their caregivers) and professional (clinicians, data experts, third sector representatives, researchers and policymaking) stakeholders to establish the feasibility, acceptability, key purposes, and design of the first pseudonymised linked national longitudinal research database for FASD in the UK.

METHODS

Approach:

Our PPI activities were designed with consideration of the UK Standards for Public Involvement in Research (UKSPI) [47] and reported according to the GRIPP2 (Guidance for Reporting Involvement of Patients and the Public) checklist [48]. The project team included individuals from academic, clinical, third sector and data governance sectors.

Approvals and ethical considerations:

Although formal ethics committee approvals were not required for this PPI project [49], we adopted an 'ethically conscious' approach, following principles outlined in Pandya-Wood et al. [50]. Supplementary Appendix 1 provides a detailed summary of how our approach aligned with each of the elements proposed in the ethically conscious framework for public involvement [50]. All stakeholders were informed of the purpose of the PPI activities and were told that their participation was voluntary. Workshop contributors were informed that their anonymised data may be used in project outputs, and that they had the right to opt out of being included in recordings and outputs.

Stakeholder identification and engagement planning

Three members of the project team (CM, AD and SH) conducted stakeholder mapping to identify and prioritise key groups and contacts for PPI. We used and expanded upon methods outlined in NHS Stakeholder Analysis guidance [51]. First, we developed a list of all of the people and groups likely to hold information relevant to, and be affected by, the development of a UK National Database for FASD. This included public stakeholders (people with FASD and their supporters) and professional stakeholders (third sector, clinical, academic, data providers/specialists, and policy representatives). Specific contacts were identified through the project team's networks and online searches. Next, we assigned priorities to our stakeholder involvement activities. For this, we considered the traditional stakeholder mapping dimensions of power, influence and the extent to which stakeholders are affected by the project or change[51]. In addition to 'who' to include we considered 'when' to include stakeholders. For example, early engagement with public contributors, clinicians, policy, and data governance experts was deemed to be important. In contrast, we decided that engagement with data contacts for potential linkage of our FASD database was not as time-sensitive. Next, we considered 'how' best to involve different stakeholders. Following USKPI principles, we sought to provide flexible and inclusive opportunities for PPI. This included consideration of the capacity/availability of key stakeholders, their access/support needs/preferences, and an understanding of the project constraints including budget, timescales and logistics (e.g. for arranging in-person meetings). Finally, we determined 'what' to include in our discussions with each stakeholder group based on, for example, whether we were seeking technical information (e.g. from data/academic contacts), or to gain an understanding of the acceptability and potential impact of a national

FASD database (e.g. from public/clinical stakeholders). Below we describe our PPI approach for each of our stakeholder groups. Figure 1 provides a visual summary of our involvement activities over time.

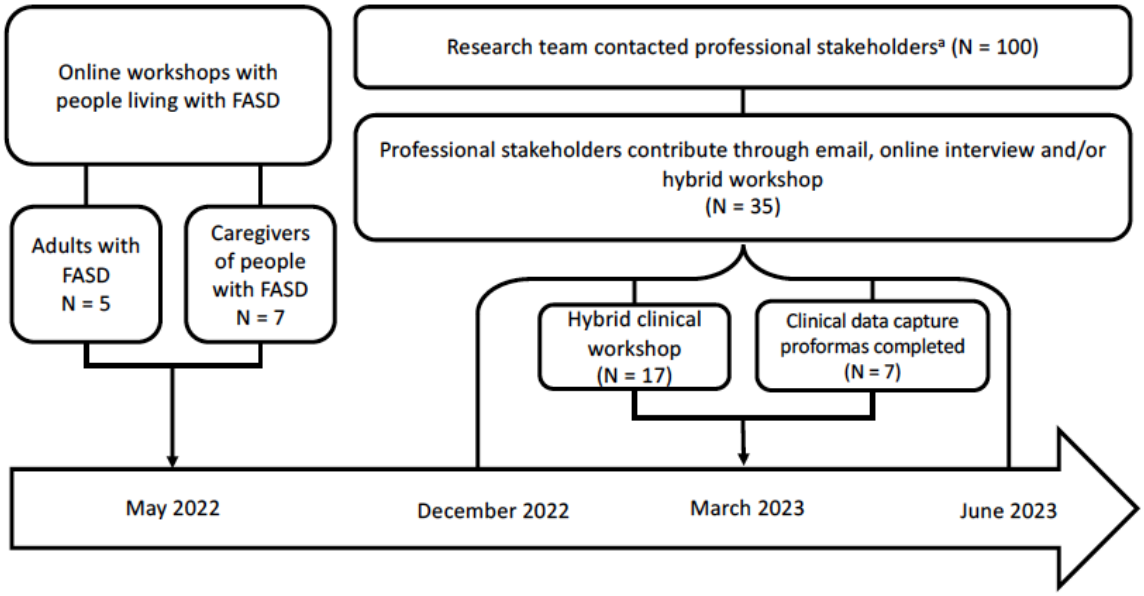

Figure 1: Summary timeline of all public involvement activities related to the development of a National Database for Fetal Alcohol Spectrum Disorder (FASD) in the UK. [a] Professional stakeholders included third sector, clinical, academic, data providers/specialists, and policy representatives.

'Lived experience' stakeholders: Adults with FASD and their caregivers

The format, materials, identification of public contributors, and delivery of our PPI activities with 'lived experience' stakeholders (adults with FASD and their caregivers) were designed and delivered in collaboration with the National Organisation for FASD, an adult with FASD and their birth mother. Online workshops were deemed to be the most appropriate format for involving stakeholders with lived experience. Contact with adults with FASD and their caregivers was supported by the National Organisation for FASD, who assisted with the design and distribution of easily accessible visual information sheets and adverts promoting the PPI opportunity. Adverts and information sheets were circulated by the National Organisation for FASD in May 2022 directly through their social media channels (Twitter and Facebook) and through emails to members of their FASD expert committee including: one educational specialist; two psychiatrists involved in FASD diagnosis; one paediatrician; two leads of local FASD support networks; one complex disability charity (the Seashell Trust); one community interest company/social enterprise (Much Laughter CIC); one independent consultant and trainer supporting vulnerable children and young people; one birth mother of an adult with FASD; and seven adults with FASD. These contacts responded directly to the project team with expressions of interest from people with lived experience of FASD and/or circulated information about this PPI opportunity more widely to relevant contacts. Members of the project team also circulated details of the opportunity through social media (X/Twitter). Recruitment numbers were limited to five adults with FASD and seven caregivers with FASD. This was primarily due to budget constraints — we had a limited amount available through public engagement funding and wanted to ensure that those who

contributed to this PPI work were appropriately reimbursed. Also, 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.

Stakeholders with lived experience who were interested in taking part contacted the study team via email or telephone to confirm participation and were given easy access information about the proposed database and the opportunity to ask further questions before agreeing to take part. Adults with FASD were invited to have a caregiver with them for support during the online workshop if they wished. All contributors with lived experience were reimbursed for their contribution following the NIHR payment guidance for researchers and professionals [52].

We carried out two one-hour online workshops using videoconferencing software in May 2022, one for adults with FASD and their supporters (N=5 adults with FASD), and one for caregivers of people with FASD (N=7). Workshops were facilitated by CM and SB and sought to explore the views and experiences of people with FASD and their caregivers with regards to: a) data sharing and consent for research, particularly in relation to views on a national FASD database and use of their personal data; b) research priorities; c) procedures for the study; d) whether they would like to be contacted to be involved in the study in the future as a contributor or PPI/steering group member; and e) any other aspects that contributors deemed relevant.

Professional stakeholders: third sector, clinical, academic, data providers/specialists, and policy representatives

Professional stakeholders were contacted by members of the project team via email or webform, provided with information about the PPI purpose and activities, and offered the opportunity to be involved through email, 1:1/group videocalls and/or a four-hour hybrid workshop. Videocall and email discussions were tailored to maximise relevance for each stakeholder and covered topics including data availability, consent and governance issues, and data storage/linkage opportunities. Informal notes were made of these discussions by members of the project team (CM, BS and AD). Between December 2022 and June 2023, we contacted 100 professional stakeholders, of whom 35 engaged in videocalls and/or email communication. We hosted the hybrid PPI workshop at the University of Salford in March 2023. This workshop took place the day before the 'FASD in the UK' conference (held at the same location) to maximise the potential for in-person workshop attendance. We provided in-person attendees with refreshments and travel reimbursement. The workshop was designed primarily for clinicians and included opportunities for: networking; an introduction to our proposal for a pseudonymised national linked longitudinal database for FASD; presentations from the Director of the UK Longitudinal Linkage collaboration (AB) and lead for the Canadian National Database for FASD (Professor Jocelynn Cook); breakout and full-group discussions on data sharing and governance; clinical data capture; presentation of a draft data pipeline for a national FASD database; and discussion of perceived benefits and challenges of establishing and using a national database for FASD in the UK. Seventeen stakeholders attended the workshop (ten in-person and seven online attendees consisting of 16 clinical and one third-sector representative). To assess the availability and format of

clinical data on FASD, clinical stakeholders were also invited to complete a proforma, based on the Canadian National Database for FASD data capture form (templates shown in Supplementary Appendix 2). The proforma included a list of measures relating to FASD including demographic information, FASD symptomology, and assessment of parental alcohol exposure. For each measure, contributors were asked whether this type of data was currently being collected in clinics, whether they would consider collecting it in the future and if they would be willing to share the data (subject to robust governance). There were two versions of this form – a short version and full version. Both are presented in Supplementary Appendix 2. The short version of the data capture proforma was unintentionally abbreviated and as a result information about neurodevelopment and growth were captured as free text under Q33 rather than in standardised tick box form. Three out of six participating clinics used this short version of the form and three out of six clinics used the full version of the form, which included additional tick box responses for the neurodevelopmental and growth domains.

Synthesis

Workshop/meeting notes and recordings: Two members of the project team (SH and CM) familiarised themselves with all of the notes and recordings collated for PPI activities. Following familiarisation, they independently identified potential themes, met to discuss similarities and differences in these themes, and worked together to refine themes until consensus was reached. Additionally, members of the wider project team who were involved in the PPI activities (SB, BS, AD) checked and approved the interpretations of the themes and summaries. For example, SB checked the interpretation of themes for those with lived experience of FASD, as she co-facilitated these groups.

Data capture proforma: SH extracted information from the clinician-completed data extraction proforma into an Excel spreadsheet. This information was then imported into Stata software and summarised quantitatively using descriptive statistics. As previously mentioned, there were two versions of the data capture proforma and these differed in terms of how information on neurodevelopmental and growth data was captured. To overcome this, we synthesised evidence on these clinical criteria by generating binary variables about data availability and willingness to share data (yes/no). Findings from the data capture forms are summarised separately under the "Data availability" section below.

RESULTS

Public and professional contributors' discussions on the feasibility and acceptability of a pseudonymised national linked database for FASD were grouped according to the following three themes: i) perceived benefits, ii) perceived challenges, iii) recommendations for database design. These results are summarised in Table 1 and described further below. Under the 'data availability' subheading at the end of this section, we describe the availability and format of FASD data, based on the data capture proformas completed by clinical contributors.

Table 1: Summary of themes across all public and professional involvement activities

Theme	Sub-theme (if applicable)

Benefits of establishing a national database for FASD in the UK *'advantages all around'*	• Advancing FASD research and policy • Increased awareness, understanding, and support for FASD • Informing clinical practice • Supporting people living with FASD 'to be heard'
Challenges of establishing a national database for FASD in the UK *'a mishmash of data'*	• Data sharing, governance, privacy considerations • Establishing the most appropriate model of consent • Existing data are inconsistent, unreliable, inaccessible • Difficulties of reducing complex diagnostic data • Limited in-clinic resources for data input
Recommendations for database design *'numbers don't always tell the truth'*	• Needs to be manageable for busy clinicians/staff • Should include confirmed and unconfirmed FASD cases • Should enable input of qualitative, as well as standardised quantitative data • Should include a 'full picture' of FASD

Benefits of establishing a national database for FASD in the UK

Across all groups, public and professional contributors were supportive of a national database for FASD in the UK, speaking of '*advantages all around'.* Key anticipated benefits included advancing research and policy, informing clinical practice, increasing diagnostic efficiency, and the potentially '*endless possibilities*' for prompting increased understanding, awareness, and support for FASD. In the excerpt below, a clinician recalls a phrase that they heard from people living with disabilities to express these ideas:

> *'The message was "no data, no problem, no actions" …any attempt to bring data together allows us all to speak from something other than our opinions, because that doesn't quite cut it… being stronger together would let us have a voice more importantly the individuals with that lived experience and exert their right to be heard through having data collected about them with them' (Clinician P6, main workshop)*

In addition, people living with FASD described the burden of having to explain their condition to others and noting that:

> *'[there is] so little proper joined up understanding of FASD' (Caregiver of person with FASD, online workshop)*

> *'trying to explain my FASD is not easy for someone who hasn't got a clue, [it] would save me a lot of stress to explain it.' (Adult with FASD)*

They suggested that a national database for FASD might help with this, shedding light on this often 'hidden' condition, and facilitating improved access to support.

> *'I feel like I've been in the shadows for a while. If it's going to help the situation, open doors, then maybe it's best to share [the data]' (Adult with FASD)*

'My son has so much more difficult time than those with ADHD, autism, etc… why is FASD still not making a difference in what support [he gets]?' (Caregiver of child with FASD)

However, during the online workshop, it became clear that the relatively abstract concept of an anonymised database may be difficult to grasp for some adults with FASD. For example, some mistakenly thought that the database would be available to healthcare practitioners when they went for medical appointments and that this was how the database would help others to understand their condition. This demonstrated that further refinement of our materials and involvement activities with people with FASD will be needed to fully understand their views and preferences for a national database for FASD in the UK.

Academic contributors were enthusiastic about the potential for a national database for FASD to illuminate the long-term outcomes and costs associated with FASD, potential opportunities for intervention, and cost-benefit analyses of improved prevention, diagnosis, and support. Policy contributors noted clear alignment with contemporary FASD and data-transformation policies.

Clinical stakeholders discussed the potential benefits of a national FASD database for providing novel insights into clinical profiles, which in turn could lead to improvements in FASD diagnosis processes:

'I can see how this database will be a potential way of structuring and really helping develop assessment more nationally'. (Clinician, P4, workshop breakout session)

Stakeholders noted that these clinical insights could lead to significant gains in the efficiency, and therefore capacity for, FASD diagnosis, which is currently a time-consuming and complex process, and one which has an extensive waiting list. For example, if the data could help identify which brain domains are most commonly impaired, this could lead to a minimum data set for assessment, thus streamlining the process, as described in this exchange:

'…some of these factors are really robust…the [Canadian] data really show those brain domains that are always, always, well 80% impaired…that's pretty powerful' (Researcher, P17, workshop)

'What [P17] just said about the [commonly affected brain] domains was so powerful…one of the questions that comes up constantly is "we can't do multidisciplinary assessment"…if there are three [neurological] domains that come up constantly we could be able to make a diagnosis, particularly the ones that come up that are easy to [assess]…if that doesn't come up then go to multidisciplinary assessment because we have to make a cost-effective model which actually works…these big datasets allow you to say "look this is what it's actually showing"' (Clinician, P9, main workshop)

Taken together, these discussions highlighted the potential to support a tiered assessment model to enable people with FASD to achieve a robust diagnosis, more quickly and cost-

effectively, with more '*complicated*' cases being referred for further assessment. While this '*slim[med] down*' model was broadly endorsed by clinical stakeholders, some also raised concerns that these data-driven insights could lead to an overly-simplistic approach for what can be considered a complex disorder. These views are described in more detail in the *challenges* section below.

Challenges in developing a national database for FASD in the UK

While contributors were supportive of a national database for FASD overall, they also anticipated some challenges, explaining:

> *'I think the reason we're all thinking challenges is because we want to happen and therefore we're trying to anticipate any hiccups on the way.' (Clinician P5, main workshop)*

Clinical stakeholders expressed the importance of ensuring that any variables included on a future National FASD Database are not viewed as the only or most important factors to be considered in clinical assessment:

> *'One of the challenges about [data standardisation] might be to...accidentally imply...that people can only assess with certain tools...Because this database can become a really powerful and effective tool we don't want to just accidentally go down the road, that if it's not on the database, it doesn't matter.' (Clinician P4, main workshop)*

Contributors from the clinical workshop noted that data recording processes were highly variable across settings, and that missing data were common, often because clinicians do not have the time and resources to collect and input the data. Data were recorded in paper and electronic format and those who use paper records expressed concerns about the cost and resources required to convert data to electronic records:

> *'we have [two electronic] database[s]…and…paper folders…I'm the only person who's [adding to the electronic records] so it's not going very fast…Some things are added. Some things aren't. It's a weird mishmash…there's no kind of real reason to what's on there.' (Clinician P7, breakout room 2)*

Establishing an appropriate model of consent was discussed as another key consideration. Clinical, academic and data governance contributors expressed a strong preference for a default (opt-out) consent model on the basis of supporting inclusive research. This approach leans towards societal 'public goods' to ensure inclusion of marginalised and harder-to-reach individuals with FASD and the sustainability of the database, and may conflict with individual rights for self-determination. Clinicians from settings with existing FASD data explained that while they did have consent to contact some patients for future research studies, obtaining consent from these individuals would be time-consuming and potentially challenging as, for example, individuals with FASD are more likely to experience unstable living arrangements, and/or be involved with the care or criminal justice system. Academic contributors advised that a default (opt-out) model of consent would be important to minimise sampling bias and to ensure that participants who were 'hard to reach' and who, arguably, may be most in need would not be excluded.

While lived experience contributors expressed a preference for a default consent model overall, stating they would 'feel fine about [data sharing] with reassurance of confidentiality' (Caregiver of person with FASD); three out of 12 contributors (two adults with FASD and 1 caregiver) expressed reservations and indicated that they may prefer an opt-in model:

'I'd just rather have someone ask. We've never had someone ask what we would prefer. To know it was used behind my back. Would like to give permission for research. Would bug me if they didn't ask.' (Adult with FASD)

Furthermore, our workshop discussions with adults with FASD and their caregivers suggested that the perceived acceptability of onward linkage depended on the type of linked data. Among the 12 lived experience contributors that we met with, all were comfortable with onward linkage to health, education, and employment data. All of the adults with FASD (some of whom had experience with the criminal justice system) stated that linkage to criminal justice data would be acceptable. In contrast only 5 out of 7 caregivers of people with FASD deemed this to be acceptable. As one caregiver stated:

'*Because of what my* children have been through before adoption, it was pretty horrific, wouldn't want that info to be available to others, court records are sealed.'

(Caregiver of person with FASD, online workshop)

Similarly, although the majority of adults with FASD (4 out of 5) and their caregivers (5 out of 7) expressed that they would be comfortable having their FASD records linked to social care data, some expressed concerns:

'[It] almost feels like someone looking into your past and interrogating it'

(Adult with FASD, online workshop)

Caregivers of people with FASD suggested that it would be useful to ask stakeholders about preferred consent models for specific data types as we develop this linked data resource further.

All contributors emphasised the need for robust data governance processes. Contributors with data linkage expertise also expressed the importance of ensuring that there would not be a risk of reidentification, particularly as the number of cases of FASD in a national database may be small initially. Some clinicians from private settings stated that they would need additional support from data governance/legal experts when developing data sharing protocols, as they did not have a dedicated information governance team:

'I want to share my data. I don't have a problem, but the legal responsibilities that I have are really high…and because I'm not part of the NHS or a bigger culture it literally does stop with me, so I suppose that's something I would need assistance with.' (Clinician P10, main workshop)

Together, these discussions show that stakeholders were supportive of the database overall and deemed it to be in the public interest. However, ongoing engagement with those living

with FASD and their caregivers in the design and communication of consent processes would be required to deliver this data resource in an acceptable, transparent and accessible way. Additionally, clinicians highlighted resource constraints that could impact the feasibility of data collection and input. Stakeholders' suggestions on how to address some of these challenges will be addressed further in the next section.

Recommendations for the design of a national database for FASD in the UK

Public contributors discussed the design elements that they felt were important to consider when designing a national database for FASD in the UK. Key points were that data collection should be standardised so far as possible, but should also leave room for qualitative input; that contributing to the database needs to be manageable for clinic staff and may require further resources to support data input; that the database should provide a 'full picture' of the lives of those with FASD, including strengths and difficulties; and that data should include both confirmed and unconfirmed FASD cases. These points are described further below.

First, clinicians noted that a degree of data standardisation and harmonisation would be valuable, stating:

> *'you need to get consensus on what assessment will you use for what domains and have as much standardisation [as possible]'. (Clinician P6, main workshop).*

Standardisation was also seen as important for facilitating comparison with international FASD datasets:

> *'I think we can absolutely do international collaborations…the more data you have, the better we are' (Clinician P17, main workshop)*

Data governance and clinical contributors also discussed the feasibility of sharing FASD data between different countries in the UK, for example between England and Scotland. Due to difficulties with 'cross-border' data sharing processes/infrastructure, contributors felt it was most feasible, in the first instance, to collate FASD data independently in England and Scotland, and then determine whether this approach can be adapted over time. It was agreed that a standardised data collection protocol would help ensure that data are comparable if it is possible to merge English and Scottish FASD datasets in the future. This could include the use of a standardised data capture proforma (such as that provided in Supplementary Appendix 2).

Clinicians recommended that consent for the database should be flexible to allow for any future projects:

> *'here the challenge… is prospective use of the data because things change over time and it's good that that permission has some flexibility in it, which allows an important new data set which we haven't thought of today' (Clinician P9, main workshop)*

People with lived experience of FASD expressed that they wanted to ensure that any linked data would provide a full picture of the lives of those with FASD, including both strengths

and difficulties. For example, caregivers of people with FASD highlighted that people with FASD may be 'vulnerable' to victimisation and noted that any linkages to criminal justice data should capture the cases in which people with FASD feature as victims, as well as perpetrators, to not give a distorted picture of their lives.

Clinicians and researchers highlighted there would be value in including data from cases where patients had been assessed for FASD but did not meet criteria, suggesting this could provide an important comparison group:

> *'there's something about capturing those who would deemed sufficiently at risk to warrant an assessment but ultimately it was deemed not to be'. (Clinician, P3, main workshop)*

> *'I think the exposed group that don't meet criteria is a really important comparator' (Clinician, P17, main workshop)*

Additionally, clinicians described the importance of retaining flexibility and space for clinical judgement in their practice. They discussed the complexity of FASD diagnosis and expressed concerns that a solely quantitative dataset within a national FASD database could lead to the loss of important details from qualitative notes:

> *'It would help that if there was some qualitative aspect or element [to the database]… a lot of the really juicy data that we get comes from the developmental history [and] triangulating between questionnaires, clinical observation, direct assessment' (Clinician, P4, breakout room)*

> *'[When doing clinical testing you have] to look at the quality of what [those being assessed] do and how they do it as much as the numbers themselves, numbers don't always tell the truth...sometimes you have to go with your gut' (Clinician P9, main workshop)*

As described in the *challenges* section above, clinicians reported having limited time for data input. Therefore, efficient data input procedures and consideration of additional support for initial data input was deemed an important database development.

Data availability

Six clinical stakeholders completed our FASD data capture proformas, based on the Canadian National Database for FASD (templates provided in Supplementary appendix 2) [36]. This exercise was designed to assess the format and availability of FASD data in UK clinics and its potential compatibility with international databases. Overall, there was a large degree of alignment in the data collected in clinics with the measures in the data capture proformas; additionally, clinicians mostly agreed that, if these measures were not currently being collected, they would consider collecting this in the future. Stakeholders expressed a willingness to share personal identifier, demographic and clinical data, subject to robust governance processes. More details about the availability of different types of demographic, personal identifier, and clinical data are presented in **Error! Reference source not found.** and summarised in the text below.

Table 2 Summary of data capture proformas showing the availability of data from 6 clinics.

	Demographic data and personal identifiers (9 items)			Sentinel facial features (3 items)			Prenatal alcohol exposure (1 item)			Neurodevelopmental assessment (1 item)[a]	
Clinic	Currently collect (% of items)	*Would consider collecting (% of items)	Willing to share (% of items)	Currently collect (% of items)	Would consider collecting (% of items)	Willing to share (% of items)	Currently collect (yes/no)	Would consider collecting (yes/no)	Willing to share (yes/no)	Currently collect (yes/no)	Willing to share (yes/no/)
1	67%	100%	78%	100%	NA	100%	Yes	NA	Yes	Yes	Yes
2	100%	NA	100%	100%	NA	100%	Yes	NA	Yes	.	.
3	44%	100%	.	100%	NA	.	Yes	NA	.	Yes	.
4	78%	.	100%	100%	NA	100%	Yes	NA	Yes	Yes	Maybe[b]
5	0%	100%	100%	.	100%	100%	No	Yes	Yes	Yes	Yes
6	56%	100%	89%	100%	NA	100%	Yes	NA	Yes	NA	NA

[a] Responses for the neurodevelopmental data are summarised as a binary response under 1 item (corresponding to yes/no), as respondents included information on this domain as both free text and tick box responses. [b] This respondent answered that data sharing would need to be discussed further with the team, as they were currently doing some research from the data and therefore, indicated that they may wish to defer data transfer until this was completed. NA = not applicable, "." = all measures missing,

Demographic information and patient characteristics

The demographic variables included in our data capture proforma were: NHS number, year of diagnosis, country, date of referral, reason for referral, source of referral, current living situation, gender and date of birth. Table 2 shows that a large portion of these variables were being collected by the clinics that completed this form. In clinics where these measures were not collected, clinicians stated that they would be willing to collect these measures in the future. Clinics indicated that they would be willing to share 78 – 100% of demographic and patient characteristics data items, subject to robust data governance. Measures which some clinics were reluctant to share included NHS number, reason for referral and date of birth.

Sentinel facial features

Sentinel facial features included palpebral fissure length, philtrum smoothness and upper lip thinness. As shown in Table 2 all of the clinics who answered this question (n=5) currently collected data on all of these measures. The remaining clinic indicated that they would consider collecting these facial phenotype measures in the future. Clinics expressed a willingness to share this data. All the clinics who answered this question (n=5) stated that they would be willing to share data on all 3 of these measures.

Prenatal alcohol exposure

Five of the 6 clinics surveyed collected data on prenatal alcohol exposure. The clinic that did not collect these data stated that they would be willing to collect in the future. Across the clinics there was a willingness to share this data.

Neurodevelopmental assessment

Four of the 5 clinics who answered this question currently collect data on variables regarding neurodevelopment. The clinic that did not collect these data indicated that this measure was not applicable in their setting, as it was a clinic for children aged 2 years and under. This clinic stated that they would refer to other clinicians for further assessment when the child had reached an older age, when needed. Regarding the sharing of this data, two of the three clinics that answered this question stated that they would be willing to share, and one clinic stated that they may be willing to share but had some reluctance due to the clinic doing research on the data.

DISCUSSION

Our multidisciplinary, multi-method public and professional involvement activities indicated support from a broad range of stakeholders for the development of a pseudonymised national linked database for FASD in the UK. For people living with FASD and third-sector representatives, the main anticipated benefit was increased understanding, awareness, and support for FASD. Lived experience stakeholders expressed an urgent need for more data and research and communicated that a national FASD database could play an important part in achieving this. Clinicians explained that a national database for FASD could provide important new insights into clinical profiles and that this had the potential to support more informed and efficient diagnosis. Researchers suggested that a linked FASD database could provide important novel insights into health, criminal justice and social outcomes associated with FASD over time, including the potential to identify opportunities for interventions to improve these outcomes. Policy-makers reported that the establishment of a national database for FASD in the UK would align well with current priorities including FASD and data transformation policies. All contributors expressed the importance of robust data governance and consent processes.

Key perceived challenges were the resource implications of harmonising, collating, and inputting existing data, which were described as highly variable in terms of availability and format. Clinicians were supportive of an element of standardisation for prospective data collection but expressed the importance of retaining space for clinical judgement and qualitative data alongside the potential insights gained from quantitative data analysis. Results from our clinical data capture proformas indicated that most of the clinics that responded captured the data necessary to establish a national linked database for FASD, including data on prenatal alcohol exposure, growth, neurodevelopmental outcomes, sentinel facial features and personal identifiers. Clinicians who did not currently collect these data indicated that they would be willing to collect and share such data in the future, subject to robust governance.

Although the UK has seen a recent influx of policy and guidance on FASD in the UK [8, 11, 14, 26, 27], the data necessary to fully understand the impact of, and plan effective service provision for FASD, is lacking. FASD databases have been established in the United States, Canada and Australia. These have been instrumental in understanding the characteristics and needs of those living with FASD in their populations, and for informing policy and practice [35-37, 39, 40]. Accordingly, these offer useful exemplars for the establishment of a UK equivalent, and for illustrating the impact that this may achieve. Our public and professional involvement work demonstrated that UK clinicians were broadly in favour of data standardisation, following a data capture method based on the proforma used by the Canadian National Database for FASD [36]. Furthermore, a recent international survey indicated that 91% of clinicians were in favour of adopting a unified approach for FASD assessment [53]. This offers further potential for future international collaborations and comparisons, if a FASD database were to be established in the UK.

Current international examples of FASD databases have not been linked to routinely collected national health, social care and administrative data sources, instead relying on retrospective reporting and follow up surveys to assess outcomes among those living with

FASD. Given the UK has a rich infrastructure of routine data[41], the development of a national linked database has the potential to provide world-leading insights into an array of long-term health, social care, economic, education and criminal justice outcomes associated with FASD. This could transform (inter)national policy, prevention, and service provision for FASD.

Public involvement and engagement have been recognised as an essential step in gaining public support in big data research, and for ensuring that such endeavours reflect public views and priorities [54-58]. Following best practice guidelines, we adopted a flexible, multimethod approach to make our public involvement work as inclusive and accessible as possible for different stakeholder groups, and involved contributors at the earliest stages of our project [47, 57]. However, we also experienced some challenges. Some adults with FASD and their supporters appeared to find the concept of a pseudonymised national database for FASD difficult to grasp, with some mistakenly thinking that their data would go directly to their GP, for example. Making the concept of a national linked database clearer will be something which we will take forward with future work, for example co-developing, with lived experience stakeholders, a clear and concise way of communicating this concept. Similarly, Teodorowski and colleagues (2023) highlighted that big data can be an abstract and complex topic to discuss with the public, especially among 'seldom-heard' groups, including those with disabilities [54]. This issue is likely to be compounded in the case of FASD, as individuals with FASD often face challenges with abstract reasoning and cognitive processing [12]. The literature suggests that animations and visualisations may make discussions around big data more accessible for the public [54]. This is something that we plan to use to a greater extent in future PPI work on this topic. Following the lived experience workshops our project team reflected that for future PPI activities with this group, it would be useful to take more time to describe what a database is in accessible language. This could include what a database might be used for and why it might be important/certain challenges associated with databases and linked data. It must be noted, however, that people with FASD may be particularly suggestible [59] . Therefore, a balance will need to be struck between providing sufficient information for contributors to understand what is being discussed, while avoiding unwittingly influencing responses. Ongoing collaboration with people with FASD will be key in developing materials for our PPI and research activities in order to enhance engagement and understanding, while minimising unintended consequences.

Given the varied responses of each group, flexible, diverse, and embedded involvement of a range of stakeholders will be essential as we seek to develop a national database for FASD in the UK. Consistent with existing literature on public views on the use and linkage of patient data [60], our stakeholders gave mixed opinions about preferred consent models. While clinicians, data governance, academic and most individuals living with FASD expressed that a default 'opt-out' model for this pseudonymised database would be acceptable, some expressed concerns about their data being included without their explicit consent. Given the spectrum of opinion on preferred models of consent among stakeholders, ongoing stakeholder engagement with a greater number of contributors will be essential as we seek to design a consent model that effectively balances the need to ensure a sustainable resource, that incurs minimal participant burden, and which provides transparency in the process of consent, security, and use of relevant data.

The work presented in this paper has many strengths. We adopted a multi-disciplinary approach gaining perspectives from a range of stakeholders including adults with FASD, caregivers of people with FASD, clinicians, representatives from the third sector, data specialists, academics, and policy stakeholders. Each group offered different perspectives and ideas on the benefits, challenges and preferred design of the database. It was strongly felt among the project team that we would have missed important points if we had not captured data/views from all these groups. In order to reach these groups, we used a multi-method approach, encompassing a hybrid workshop, online 1:1 and group meetings, and email conversations. This multi-format approach enabled more contributors to join and reduced the potential burden of travel/time commitments, which may have been barriers to inclusion for some. Additionally, there was a range of skill sets within the project team, including researchers' data expertise, quantitative and qualitative expertise, specialist clinical, and lived-experience insight. This enabled a broad understanding of the barriers and facilitators that the stakeholders presented to us.

Nevertheless, it is important to note that the number of public contributors involved in this project was relatively low (N = 5 adults with FASD and N = 7 caregivers of people with FASD). This was due to project resource constraints including a limited budget for stakeholder reimbursement. Furthermore, our method for identifying public contributors through the National Organisation for FASD and the project teams' networks will have favoured people with FASD and their caregivers who were already known within these networks. While it is recognised that PPI work is not expected to be generalisable [61], we acknowledge that the summaries and conclusions presented in this article reflect the views of this specific group of contributors. Therefore, the themes and conclusions presented in this work should be considered preliminary. Future work should seek to increase the number of public contributors and provide greater opportunities for 'harder-to-reach' individuals with FASD. This should include individuals from FASD within especially marginalised groups such as those with chronic disabilities, those from ethnic minority backgrounds and those in contact with the criminal justice or care system.

The findings presented in this article provide high-level, descriptive summaries of our discussions with public and professional contributors. This approach is appropriate for PPI projects such as this. Unlike qualitative research, PPI projects do not seek to undertake formal data analysis, but rather are based on a process of 'sense-checking' outputs and illustrating how PPI contributions have informed development of future research proposals [61]. Future, complementary qualitative research may be useful to gain a more in-depth understanding about the lived experience of people with FASD in relation to the development and implementation of this data infrastructure. Another potential limitation is that our workshops may have suffered from contagion whereby opinions from some attendees may have spread throughout the group from one member to others [62]. Dominant voices may have led the conversation and others may not have felt they could share their views freely. To mitigate this, facilitators actively invited contributions from less vocal members of the group, however it is possible that opinions were still influenced by more dominant group members.

CONCLUSIONS

Our multi-method and multidisciplinary public and professional involvement activities indicated support from a broad range of stakeholders for the development of a pseudonymised national linked database for FASD in the UK. Our stakeholders reported that a national FASD database could have far-reaching benefits including facilitating advances in research and policy, improving prevention, and increasing awareness, understanding and effective support for FASD. Importantly, this resource would provide a step-change in increasing the accessibility and visibility of FASD in key public health data sources. Our contributors also highlighted some challenges mainly regarding the practicalities of using the database and data governance issues, and made recommendations for important design features. The perceived benefits and challenges of the database varied by stakeholder group demonstrating that flexible, diverse, embedded stakeholder collaboration will be essential as we seek to establish this database. Given its relatively sophisticated routine data infrastructure, the UK has the potential to develop a world-leading resource to support advancement of FASD knowledge, policy and service provision.

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Ethics statement

These activities were classed as public involvement work, rather than research. In this work, members of the public were acting as specialist advisors, providing valuable knowledge and expertise based on their experience of a health condition or public health concern. Public contributors received written details of the planned activities and were given the opportunities to discuss their involvement with the project team before they participated in each session. However, review by a research ethics committee and formal processes for obtaining informed consent were not needed (NIRH involve – Do I need to apply for ethical approval to involve the public in my research? | INVOLVE).

Data access statement

Data is not available as this paper is a write up of PPI insights and not data.

Conflicts of interest

There are no conflicts of interest to declare.

Abbreviations

DHSC: Department of Health and Social Care

FAS: Fetal alcohol syndrome

FASD: fetal alcohol spectrum disorder

NHS: National Health Service

NICE: National Institute for Health and Care Excellence

OHID: Office for Health Improvement and Disparities

SNOMED CT: Systematized Nomenclature of Medicine Clinical Terms

UK: United Kingdom

UKSPI: UK Standards for Public Involvement in Research

SIGN: Scottish Intercollegiate Guidelines Network

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