Health Research Authority

Evaluating Models of care, best practice and care pathways for women who are dependent on drugs and their infants, from preconception to 18 months postnatal.

Stepping Stones Study

PROTOCOL VERSION NUMBER AND DATE

Protocol Version 3.0 October 2022

RESEARCH REFERENCE NUMBERS

IRAS Number: 309152

SPONSORS Number: RE18563

FUNDERS Number: NIHR130619

SIGNATURE PAGE

The undersigned confirm that the following protocol has been agreed and accepted and that the Chief Investigator agrees to conduct the study in compliance with the approved protocol and will adhere to the principles outlined in the Declaration of Helsinki, the Sponsor's SOPs, and other regulatory requirement.

I agree to ensure that the confidential information contained in this document will not be used for any other purpose other than the evaluation or conduct of the investigation without the prior written consent of the Sponsor

I also confirm that I will make the findings of the study publicly available through publication or other dissemination tools without any unnecessary delay and that an honest accurate and transparent account of the study will be given; and that any discrepancies from the study as planned in this protocol will be explained.

For and on behalf of the Study Sponsor:

Signature:

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Date:

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Name (please print):

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Position:

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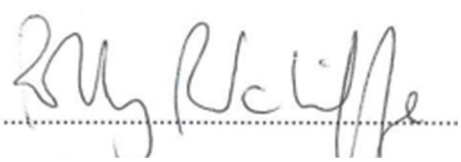
Chief Investigator:

Signature: 

Date:

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Name: (please print):POLLY RADCLIFFE

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LIST of CONTENTS

KEY STUDY CONTACTS

Insert full details of the key study contacts including the following

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Sponsor	
Joint-sponsor(s)/co-sponsor(s)	Full contact details including phone, email and fax numbers of ALL organisations assuming sponsorship responsibilities as a joint- or co-sponsor/s (If applicable)
Funder(s)	NIHR
Key Protocol Contributors	Full contact details including phone, email and fax numbers (If applicable)
Committees	Full contact details including phone, email and fax numbers

STUDY SUMMARY

Study Title	Evaluating Models of care, best practice and care pathways for women who are dependent on drugs and their infants, from preconception to 18 months postnatal
Internal ref. no. (or short title)	Stepping Stones Study
Study Design	A multi-method, longitudinal, qualitative case study design that will identify models of best practice for women who experience drug dependence in pregnancy and how these can be optimised to meet the needs of mothers and their infants. The study will be co-produced with health and social care professionals and service users through the Expert Advisory and Co-production group to ensure findings and recommendations are directly relevant and implementable.
Study Participants	Women who are dependent on one or more of the following drugs: prescribed opioids (e.g., methadone), illicit opioids (e.g., heroin), benzodiazepines, cocaine/crack or amphetamines, who are pregnant or post-partum. GPs, midwives, health visitors, social workers and substance use treatment staff
Planned Size of Sample (if applicable)	Four case study sites 40 pregnant/postpartum women 88 GPs, midwives, health visitors, social workers and substance use treatment staff 16 service managers, commissioners and policy makers

Follow up duration (if applicable)	Qualitative data will be collected via serial, in-depth interviews with each study participant at 4/5 months intervals from early pregnancy through to 18 months postnatally.
Planned Study Period	30 months
Research Question/Aim(s)	Aim: To undertake an in-depth longitudinal study of women's experiences of health and social care from early pregnancy to up to 18 months after the baby is born to determine how best to meet their health and social needs and those of their babies Questions: 1. What are key candidate models of multidisciplinary care for women who are dependent on drugs from preconception through to 18 months postnatal? (Phase 1) 2. What is best practice across health and social care for optimising outcomes and reducing inequalities for women who are dependent on drugs around childbirth? (Phase 1) 3. How do women who are dependent on drugs experience services and their care journeys, and how do these experiences impact on engagement and outcomes for women and their infants? (Phase 2) 4. What is the optimal service model for women who are dependent on drugs (from preconception up to 18 months postnatal), to foster good parenting and to provide a safe, stable and nurturing caregiving environment for the mother, infant and family as a whole? (Phase 3) 5. What are the optimal best practice insights for the future care of women who are dependent on drugs and their infants, to maximise engagement with services, maternal and infant outcomes, and to prevent out-of-home care placements? (Phase 3)

FUNDING AND SUPPORT IN KIND

FUNDER(S)	National Institute of Health Research, Health Service and Delivery Research Programme

ROLE OF STUDY SPONSOR AND FUNDER

King's College London is the sponsor for this research programme and will assume overall responsibility for the initiation and management of the study. King's College London, as research Sponsor indemnifies its staff, research participants and research protocols with public liability insurance. These policies include provision for indemnity in the event of a successful litigious claim for proven non-negligent harm.

The programme is funded by the NIHR Health Services & Delivery Research. The NIHR HS&DR will monitor progress and be informed of all changes to the protocol. The NIHR HS&DR will be sent all outputs at least 28 days before publication/dissemination. All published outputs will acknowledge funding and include the following disclaimer:

'This project is funded by the National Institute for Health Research (NIHR) Health Services Delivery and Research programme (project reference NIHR130619). The views expressed are those of the author(s) and not necessarily those of the NIHR or the Department of Health and Social Care.'

Decisions about the study design, conduct, data analysis and interpretation, manuscript writing, and dissemination of results will be made by the Chief Investigator and study management groups (see below) and will not be within the responsibility of the sponsor or funder.

ROLES AND RESPONSIBILITIES OF STUDY MANAGEMENT COMMITEES/GROUPS & INDIVIDUALS

King's College London will be the project sponsor and the host organisation, with Radcliffe as the Chief Investigator. Subcontracts will be put in place between King's College London and other partner organisations, detailing the budget resources allocated, the responsibilities and the expected contributions of each party. There will also be site agreements between the sponsor and the NHS Trusts for the recruiting hospitals. Ethical approval and NHS R&D approvals will be obtained before the commencement of Phase 2

Expert Advisory and Co-Production Group

We have established an Expert Advisory and Co-Production Group (EACG) to inform discussions and decisions on the scope and focus of our literature review, the conduct of our qualitative field work, synthesis and interpretation of preliminary findings, candidate service models, and plans to translate and disseminate findings. The group will include women with lived experience of addiction (who will be supported by peer support workers) and will meet virtually approximately four times per year for the duration of the project.

Chair

Professor Julie Taylor, Professor of Child Protection, School of Nursing, University of Birmingham.

Members

Joanne Lacey, Regional Lead (London), Institute of Health Visiting, North East London NHS Foundation Trust and Fellow of Institute of Health Visiting

Virginia Wright, Parents, carers & families (alcohol and drugs) lead, Public Health England

Maggie Boreham, Principle Public Health Specialist, London Borough of Hackney

Sally Egan, Director & Child Health Commissioner, NHS Lothian Maternal and Child Health Planning, Policy and Performance Directorate (retired)

Professor Pat Hoddinott, Chair in Primary Care, Nursing Midwifery and Allied Health Professions Research Unit, University of Stirling

Jo Daubney Specialist Midwife. Professional Midwifery Advocate, Gloucestershire Hospitals NHS Foundation Trust

Karen Erskine, Drug Liaison Midwife, James Paget James Paget University Hospitals NHS Foundation Trust

Professor Tessa Parkes, Research Director for the Salvation Army Centre for Addiction Services and Research at the University of Stirling

Four service users will be supported remotely and/or in person by peer mentors from the Addiction Service User Research Group, KCL and the Drug Research Network, Scotland to ensure they can fully participate in the EACG.

The Study Steering Committee (SSC) will provide independent oversight of the study on behalf of the study sponsor. The SSC will meet (in person) a minimum of once yearly (to be decided by the Committee according to NIHR guidelines and outlined in the Charter). The SSC comprises independent members to provide oversight of the project and ensure that the project is conducted to the standards set out in the Department of Health's Research Governance Framework for Health and Social Care (82) and the Guidelines for Good Clinical Practice. The SSC will comprise members of the EACG who meet the NIHR criteria of independence.

The Data Monitoring and Ethics Committee (DMEC) comprises independent members and will provide oversight of the safety, rights and well-being of the study participants and will make recommendations to the Steering Committee regarding ethical or safety concerns. The DMEC will meet annually just before the SSC. The project team will provide the DMEC with a comprehensive report, the content which will be agreed in advance by the Chair of the DMEC.

The Programme Management Group (PMG) will meet at least twice a year and will report to the SSC at their meetings. The Programme Management group includes all co-investigators to ensure milestones are achieved, oversee progress, trouble shoot if problems arise, plan the next stage and agree timelines.

A Core Programme Group (CPG) will meet at least every month (and more frequently as needed) to oversee day-to-day running of the programme. The CPG includes the Chief Investigator (Radcliff), Co-Lead Scotland (Cheyne), research fellows and research assistants and will draw on other expertise in the team when needed.

The Chief Investigator has overall responsibility for the study and will oversee all study management. The Chief Investigator will be responsible for monitoring of safety outcomes and reporting arrangements. The data custodian will be the Chief Investigator.

The project therefore has a clear management structure with the most appropriately qualified research team member taking responsibility for each aspect, and representation from the most relevant stakeholders.

PROTOCOL CONTRIBUTORS

In preparation for the Stage 1 application, we have taken the views of mothers who use or who have used drugs (from London & Edinburgh), lead practitioners from specialist services (Hackney, Glasgow & Edinburgh), and the Nursing, Midwifery and Allied Health Professionals Research Unit, University of Stirling, 'People in Research' Group to seek their opinions on our study design. Feedback included strong support for the potential impact of the research to explore women's experiences of perinatal care pathways and to identify best practice models of care to improve outcomes for families and the use of innovative methods.

In preparation for the Stage 2 application, we sought the views of safeguarding midwives (from London, Leeds, Glasgow and Ayrshire), substance use practitioners, children and family social workers, and public health experts in the London Borough of Hackney, a manager of residential substance use treatment service which accepts pregnant and postpartum women and their children; and a fellow of the Institute of Health Visiting. All professionals consulted approved the qualitative longitudinal design of the research with mothers and stated that the study will fill an urgent gap in knowledge. Safeguarding midwives and substance use practitioners stated that the recruitment target is feasible and approved the inclusion of online focus groups with professionals and telephone/online interviews with individual managers, commissioners, and policy makers.

Professionals consulted were enthusiastic about the role of the Expert Advisory and Co-production Group (EACG) that will guide the conduct of the study and made recommendations to invite representatives of professional and service user groups to take part. We received funds from the NIHR London Research Design Service for consultation with service users. We conducted online and telephone consultations with three mothers in community and residential treatment and members of the Addiction Service User Research Group, KCL. Mothers gave advice on retaining women in the study by sending regular text or Whattsap messages. They also suggested that women would positively enjoy drawing Timelines to map significant events in their personal lives and to help them recall episodes of drug use, relapse and engagement with treatment. Mothers also recommended the use of audio or video diaries in order for women to record events as they occur and as a stimulus for discussion in the in-depth, serial interviews. Members of the Addiction Service User Research Group, KCL approved the

plan to reimburse research participants for their time. They also gave positive feedback on the involvement of experts by experience in the EACG. They suggested that service users should be supported by peer support workers in order to fully participate in the EACG and the dissemination activities, a recommendation which we have included and has been costed in the proposal.

KEY WORDS:

Perinatal mental health

Drug use in pregnancy

Parental drug use

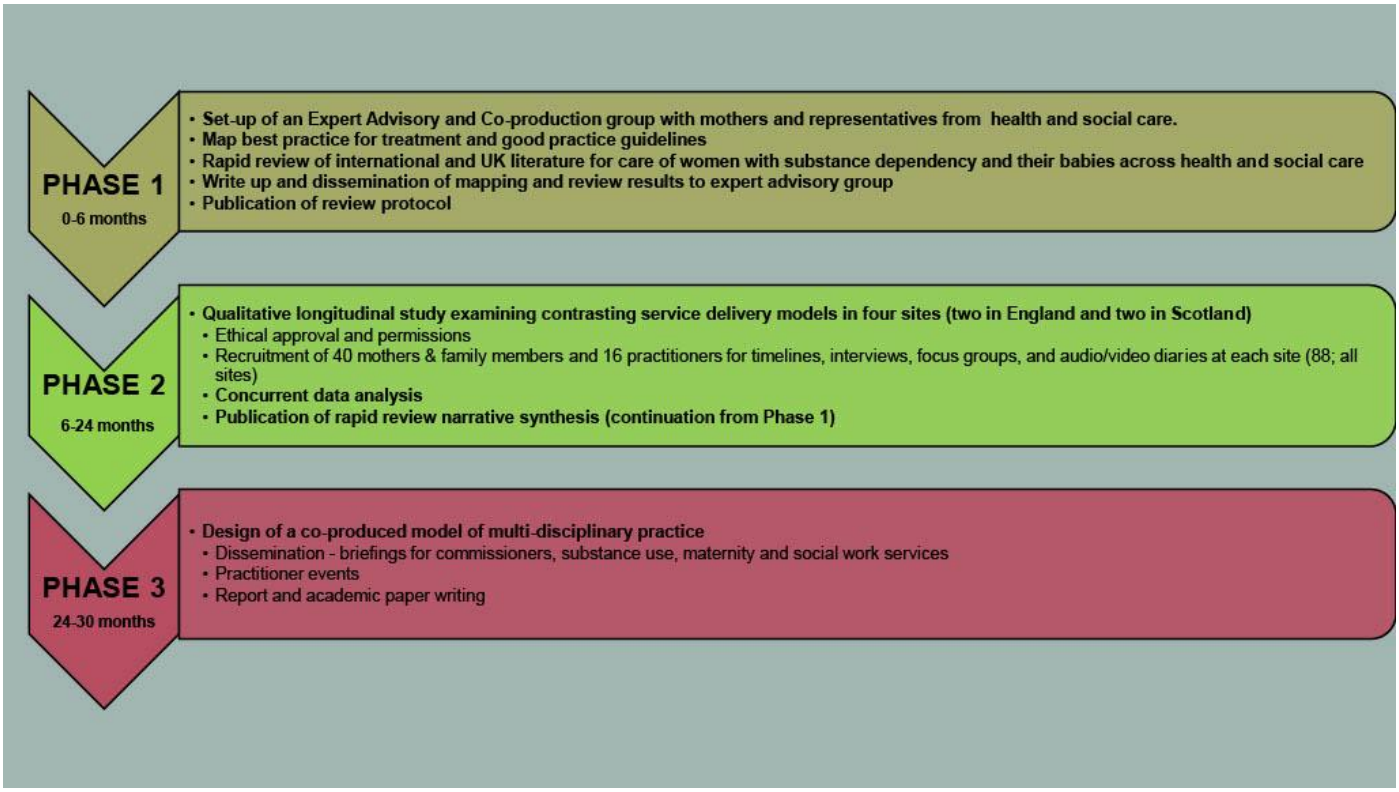
Integrated interventions

Neonatal care

Vulnerable populations

STUDY FLOW CHART

Evaluating Models of care, best practice and care pathways for women who are dependent on drugs and their infants, from preconception to 18 months postnatal.


PHASE 1
0-6 months
• Set-up of an Expert Advisory and Co-production group with mothers and representatives from health and social care.
• Map best practice for treatment and good practice guidelines
• Rapid review of international and UK literature for care of women with substance dependency and their babies across health and social care
• Write up and dissemination of mapping and review results to expert advisory group
• Publication of review protocol
PHASE 2
6-24 months
• Qualitative longitudinal study examining contrasting service delivery models in four sites (two in England and two in Scotland)
• Ethical approval and permissions
• Recruitment of 40 mothers & family members and 16 practitioners for timelines, interviews, focus groups, and audio/video diaries at each site (88; all sites)
• Concurrent data analysis
• Publication of rapid review narrative synthesis (continuation from Phase 1)
PHASE 3
24-30 months
• Design of a co-produced model of multi-disciplinary practice
• Dissemination - briefings for commissioners, substance use, maternity and social work services
• Practitioner events
• Report and academic paper writing

STUDY PROTOCOL

Evaluating Models of care, best practice and care pathways for women who are dependent on drugs and their infants, from preconception to 18 months postnatal.

1. BACKGROUND

There is growing recognition of the impact on children of parental drug use (Advisory Council on the Misuse of Drugs, 2003; Scottish Executive, 2004) with studies emphasising the enduring problems for children beginning in utero with delays in physical, cognitive and socioemotional development (Huestis & Choo, 2002; Vaughn, Ollie, McMillen, Scott, & Munson, 2007). As a result of the increased risk of serious harms and poor outcomes, a high proportion of children of mothers who use substances become the subject of child protection procedures (Brandon et al., 2012). It is estimated that between 50% and 80% of children in foster care are from households with at least one substance using parent (Besinger, Garland, Litrownik, & Landsverk, 1999; Fernandez & Lee, 2013). Just over 50% of adults receiving substance use treatment are parents or live with children (Grella, Hser, & Huang, 2006; National Treatment Agency for Substance Use, 2012). For women receiving opioid substitution treatment, this figure rises to over 80% (Lundgren et al., 2013). A recent analysis of the National Drug Treatment Monitoring System for England (Hay, 2018) suggests that over 140,000 children in England live with mothers and/or a father receiving treatment for opiate use.

The 'inverse care law', described nearly 50 years ago by Tudor Hart (1971), occurs when 'the availability of good medical care tends to vary inversely with the need for it in the population served'. This circumstance particularly applies to pregnant women and mothers who are dependent on drugs because their complex health and social needs are related to histories of abuse, mental health problems, poor physical health, drug-related violence and crime, social exclusion, homelessness and poverty. These mothers and their infants are highly vulnerable yet are at risk of falling through the gaps in particular, during maternity care that is more focussed on healthy pregnant women and women with physical or mental health problems. While women who use drugs require normal maternity care and often have physical or/ and mental health problems they often do not 'fit' into standard care pathways. Substance use in pregnancy is thus a multifaceted public health problem (Manning, Best, Faulkner, & Titherington, 2009) with many confounding factors and implications for the long-term health and wellbeing of both mothers and children (National Institute for Health and Social Care Excellence, 2010 (2018); World

Health Organization, 2014). This study addresses an urgent gap in knowledge regarding the kinds of multidisciplinary support and care that can provide good outcomes for this group of women and their children in the first eighteen months of life.

A rapid scoping review prepared for this application identified 19 largely North American published studies of interventions for pregnant women who are dependent on drugs. These included i. therapeutic interventions delivered in the context of substance treatment services, ii. integrated, multidisciplinary services and iii care coordination and patient navigation programmes. The review found that therapeutic interventions delivered alongside substance use treatment for pregnant and post-natal women who are dependent on drugs have some benefit in helping new mothers to develop sensitivity to their babies' and to improve attachment. The review found that the therapeutic relationship between patient/service user and service provider is key in delivering good outcomes for women and that the provision of food, housing and welfare benefits support encouraged women's engagement with and retention in services. Studies suggest that women who are dependent on substances engage more readily with strengths-based and trauma informed services. It is not clear however which combination of services are most important in delivering which outcomes for women or their children and how relevant these findings are to the UK context.

2. RATIONALE

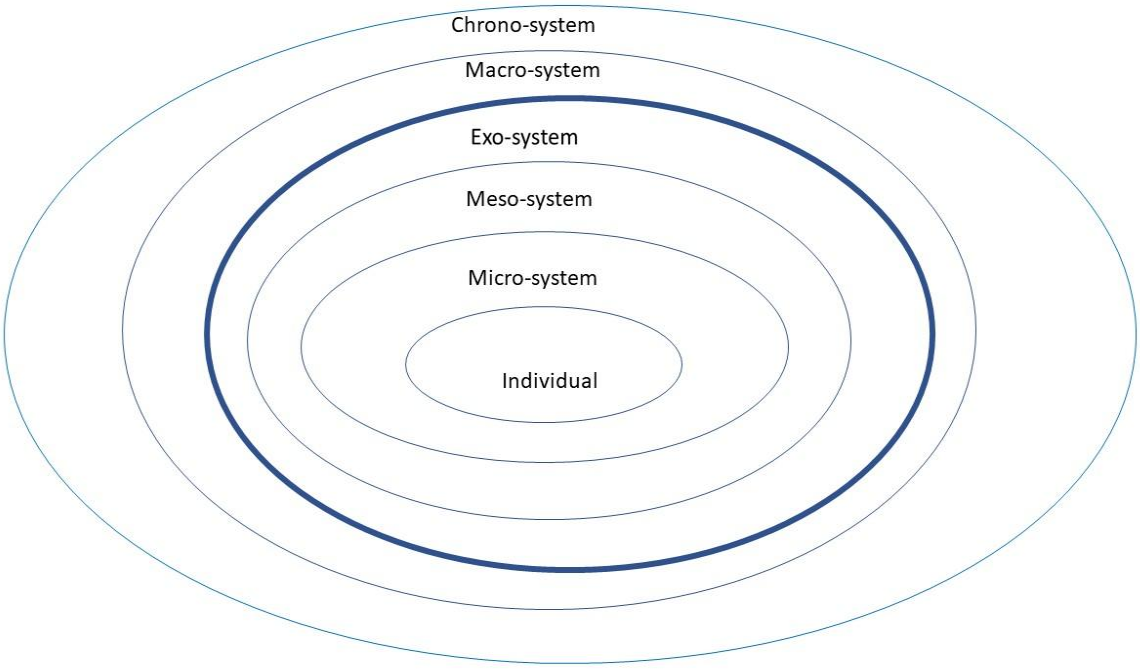
There is little evidence of the lived experiences of women receiving multidisciplinary and integrated treatment services in the UK and whether their needs and their infants needs are met. We do not yet know if current services are acceptable to women, and if they lead to better or worse outcomes for families. Existing recommendations for woman-focused care, coordinated multidisciplinary family support plans, and integrated care pathways (Knight et al., 2018; National Institute for Health and Social Care Excellence, 2010 (2018); NHS England, 2016; Scottish Government, 2017; World Health Organization, 2014) have been inconsistently implemented across the UK. Despite a proliferation of UK and international good practice guidance on managing drug dependence around childbirth (Knight et al., 2018; National Institute for Health and Social Care Excellence, 2010 (2018); NHS England, 2016; Scottish Government, 2017; Tarasoff et al., 2018; World Health Organization, 2014), there is little robust UK evidence for preconception care; which models of care deliver the best outcomes for women and their children, and virtually no evidence

that women with who are dependent on drugs themselves have been involved in efforts to improve or redesign services (National Institute for Health and Social Care Excellence, 2010 (2018)).

Wide disparities in both the numbers of babies admitted to neonatal units for Neonatal Abstinence Syndrome (NAS) (NHS Digital, 2019) and out-of-home placements in different local authority areas indicate regional concentrations of disadvantage and diversity of clinical practice (Bywaters, 2015). Although the severity of neonatal drug withdrawal symptoms is not linked to maternal dose of opioid use during pregnancy (Cleary et al., 2010), opioid use in pregnancy and its effects can be particularly stigmatising (Nichols, Welborn, Gringle, & Lee, 2020) women can be made to feel responsible for their babies' condition (Chandler et al., 2013; Radcliffe, 2011). While it is unclear whether a diagnosis of NAS influences child protection decision making (Canfield, Radcliffe, Marlow, Boreham, & Gilchrist, 2017; Gilchrist & Taylor, 2009), evidence suggests that removal of infants at birth and involvement with the child protection system may heighten risk of intentional and non-intentional deaths among women (Knight et al., 2018; Thumath et al., 2020). Evidence of a rise of repeat removals of infants in England and Wales (Alrouh et al., 2019; Broadhurst & Mason, 2013) and in kinship care arrangements in Scotland (Hill, Gilligan, & Connelly, 2019) from mothers who are dependent on drugs and other complex needs, suggests there is an urgent need to identify how services can work across disciplinary boundaries to consistently support this vulnerable group of mothers to care for their children. UK evidence is thus required of 1) best practice models for coordinated and integrated/multidisciplinary care that have the potential to interrupt the transmission of adversity across generations and 2) women's views and experiences of different models of care and how services could be improved. The literature on models of perinatal care for women who are dependent on drugs is overwhelmingly North American. We have found no qualitative studies that have tracked women with who are dependent on drugs and their children longitudinally in their journeys through pregnancy and postnatally.

3. THEORETICAL FRAMEWORK

The five interlocking domains of influence described in Bronfenbrenner's (1993) ecological model of human development will provide a theoretical framework for our study. These are micro systems; meso systems; exo-systems; macrosystems and the chrono system. The latter, temporal domain encompasses changes over time both within individual lives and within the environments in which individuals are located, conceptualising how changes in time may relate to and affect outcomes. Bronfenbrenner considered that these varied domains are nested inside one another, like Russian dolls. He proposed that research designs should investigate the interrelationships between the nested domains (1993: 38).



Our empirical study of women who are dependent on drugs will entail Qualitative Longitudinal Research (QLR), an approach that is located within a qualitative, interpretivist tradition (Ritchie and Lewis 2003). QLR uses a range of qualitative methods to explore individual experiences as they unfold over time. The emphasis of QLR has been to capture critical moments as well as "the motivations and experiences of biographical change" (Morrow & Crivello, 2015). In this study we will aim to understand the events and experiences for mothers and their infants' in their contact and interactions with services and support (substance use, maternity services, child safeguarding services and systems of care and surveillance)

in order to identify characteristic health and social care journeys. Informed by the rapid review, this longitudinal study will enable us to describe and to evaluate models of care, best practice and care pathways for women who are dependent on drugs and their infants.

4. RESEARCH QUESTIONS/AIMS

Aim

To undertake an in-depth longitudinal study of women's experiences of health and social care from early pregnancy to up to 18 months after the baby is born to determine how best to meet their health and social needs and those of their babies.

Research questions

1. What are key candidate models of multidisciplinary care for women who are drug dependent from preconception through to 18 months postnatal? (Phase 1)

2. What is best practice across health and social care for optimising outcomes and reducing inequalities for women who are drug dependent around childbirth? (Phase 1)

3. How do women who are drug dependent experience services and their care journey, and how do these experiences impact on engagement and outcomes for women and their infants? (Phase 2)

4. What is the optimal service model for drug dependent women (from preconception up to 18 months postnatal), to foster good parenting and to provide a safe, stable and nurturing caregiving environment for the mother, infant and family as a whole? (Phase 3)

5. What is the optimal best practice guidance for the care of drug dependent mothers and their infants, to maximise engagement with services, maternal and infant outcomes, and to prevent out-of-home care placements? (Phase 3)

Objectives

1. Establish an Expert Advisory and Co-Production Group to guide the conduct of the study and to co-produce the research outputs.

2. Review the international evidence on models of care and care pathways for women who are dependent on drugs and their infants from preconception through to 18 months postnatal and clinical and good practice guidance on the treatment and care of mothers who are dependent on drugs and their infants.

3. Critically evaluate women's experiences of health and social care, their care journeys and outcomes for the family, from confirmation of pregnancy to 18 months postnatal.

4. Co-produce an optimal service model for women who are dependent on drugs, child and family-centred care and insights for future care and practice guidance to optimise outcomes for mothers, infants and the family.

Outcomes

Reduce morbidity and improve long-term health and wellbeing. This research will identify optimal best practice and ways in which care may better meet the needs of women who are dependent on drugs and their babies. Conception to 18 months post birth is a crucial time period in the development of the infant in terms of physical, mental and social development. Improving care and support will positively impact the health and wellbeing of women, improve their parenting skills and confidence, increase engagement with support services and reduce the number of infants removed from parental care. Longer term this study has the potential to reduce the number of children of drug using mothers who require long term care and interrupt the transmission of adversity across generations.

Improve efficiency, effectiveness and consistency of services. Currently UK services for care of drug dependent women and babies are characterised by variation and this means that many of the UKs most vulnerable mothers and babies receive unpredictable quality care. We will highlight best practice, co-produce an optimal service model for women, child and family-centred care and develop evidence based good practice guidance to improve outcomes for mothers, infants and the family. This will benefit all those concerned with service design and delivery, facilitating more consistent, efficient effective safe and patent focussed services.

Generate new knowledge. The research addresses gaps in knowledge and the lack of an evidence base for effective care models for women who are dependent on drugs and their babies**.** It will be of interest to policy makers and commissioners at a local level (e.g., Alcohol & Drug partnerships,

Integrated Joint Boards (Scotland) & national level (e.g., Scottish Government Health and Social Care Directorates, Public Health England, Scottish Government Drug Policy Unit, NICE) and internationally (e.g., WHO). Our research will provide vital evidence of what works in what contexts in relation to mothers using drugs and will inform national and local health and social care policy.

5. STUDY DESIGN/METHODS

This is a multi-method study involving rapid and systematic reviews, longitudinal, qualitative case studies and co-production. The study will be conducted in three phases described below.

PHASE 1 (0-6 Months)

Aims

1. To identify key candidate models of multidisciplinary care for women who are drug dependent from preconception through to 18 months postnatal.

2.To identify best practice across health and social care for optimising outcomes and reducing inequalities for women who are drug dependent around childbirth.

Objectives

1. Establish an Expert Advisory and Co-Production Group (EACG) to guide the conduct of the study and to co-produce the research outputs.

2. Review the international evidence on models of care and care pathways for women who are dependent on drugs and their infants from preconception through to 18 months postnatal and clinical and good practice guidance on the treatment and care of mothers who are dependent on drugs and their infants.

Design

Co-production and Patient Public involvement and rapid and systematic review.

Phase 1 part one. We will establish an Expert Advisory and Co-Production Group to inform discussions and decisions on the scope and focus of the literature review, the conduct of our qualitative field work, synthesis and interpretation of preliminary findings, candidate service models, and plans to translate and disseminate findings. The composition of the EACG is described above. We have funds for four service users in both Scotland and England to be supported remotely and/or in person by peer mentors

from the Addiction Service User Research Group, KCL and the Drug Research Network, Scotland to ensure they can fully participate. The group will meet online (using MS Teams) eight times (approximately quarterly) across the project commencing with a half day research framing event.

Our approach to co-production will be informed by the 5 step Co-production Star http://www.coproductionscotland.org.uk/resources/the-co-production-star/ as follows:

1. **Map It**– explore existing and new forms of co-production of services in similar areas, our phase one rapid review will identify existing good practice nationally and internationally.
2. **Focus It**– all stakeholders contribute to developing a theory of change, ensuring relevance of outputs to service commissioners/ policy makers and practitioners.
3. **People It**– Our EACG will involve people who are willing and able to co-produce outcomes and insights for care and services which are a priority for them or in their area.
4. **Market it**– we will develop a range of outputs to ensure maximum reach and impact. For each output we will consider appropriate social marketing and behaviour change tools to maximise impact (Phase 3)
5. **Grow It**– We will seek further funding to support implementation of the optimised model at scale across the UK (Phase 3).

Our co-production plan is depicted in Appendix 1.

Phase 1 part two Rapid evidence review

This review will focus on the implementation and effectiveness of multidisciplinary and integrated models of care delivery for women who are dependent on drugs and their infants. We will review the evidence using rapid review methods (Khangura, Konnyu, Cushman, Grimshaw, & Moher, 2012). Rapid review is ‘a type of knowledge synthesis in which components of the systematic review process are simplified or omitted to produce information in a short period of time’ (Khangura et al., 2012). Rapid review is characterised by engagement with knowledge-users throughout the review process, expedited timeframe, and the extent of synthesis (Hartling et al., 2015).

The rapid review will be carried out in consultation with the EACG, using the ACTIVE framework for involving people in the systematic review process (Pollock et al (2019). The group will be integral to establishing the parameters of the search, as well as offering feedback and interpretation of the review results. The Rapid review will be conducted in two stages:

1. We will conduct a scoping review of current clinical guidance, and related policy documents in the UK to map suggested 'best practice', using the JBI (Joanna Briggs Institute) Scoping review methodology and Prisma-Scr Findings will inform the development of our rapid systematic review protocol, determine the scope of the review, and develop the conceptual framework.
2. Secondly, we will carry out a rapid mixed methods systematic review of the primary research evidence on models of care. Integrated findings will be used to establish which type of services, interventions and approaches to care are best suited to this population, to improve outcomes for mothers and their infants. The EACG will provide interpretation of, and validate, the review findings and identify how results and recommendations will be used.

1, Scoping review mapping clinical guidelines, and policy documents that address the needs of women who are drug dependent during the perinatal period.

This review will employ the Joanna Briggs Scoping review methodology and will be registered on the Open Science Framework repository (https://osf.io/ja69n/)

A predefined systematic search strategy will be followed to identify relevant documents meeting specified inclusion and exclusion criteria. This will include conducting a search of the following electronic databases, Social Care Online, PsycINFO, CINAHL, Scopus and Trip using an agreed in advance combination of keywords and mesh terms. Key government, statutory and non-statutory web-based platforms such as NICE, Sign, ScotPhO, etc. will also be searched using these keywords. The expert advisory group and coproduction group (EACG) of key stakeholders will be consulted to advise of any further documents to be considered. Other external UK-based leading experts such as those within the Perinatal Mental Health Networks will also be contacted. References within policy documents will be checked to identify any other relevant documents.

All documents will be independently screened by title / abstract / executive summary by one reviewer, with 25% being verified by a second reviewer. Documents will then be independently screened in full text by one reviewer with 25% verified by a second reviewer. Any disagreements between reviewers at each stage of the selection process will be resolved through discussion, or with an additional reviewer. Reasons for exclusion will be recorded.

Data will be independently extracted by one reviewer using a predefined template, with 25% verified by a second reviewer.

Reporting: Findings will be reported in narrative, as well as a being presented in tabular form. A timeline diagram will also be used to illustrate the points at which the document is relevant to a woman's perinatal journey. The PRISMA-ScR will be used to ensure that reporting is transparent and complete. We will present our findings for discussion and feedback with our EACG. This process will help to identify best practice models for optimising outcomes and reducing inequalities for women who are dependent on drugs with the aim of swiftly feeding back important conclusions to key policymakers and practitioners working with women/mothers who use substances.

2.Rapid mixed method systematic review:

This rapid systematic review aims to answer the following research questions, and will identify, critically appraise and synthesis all relevant research studies:

1. What are the range of interventions and approaches that have been developed for women who are dependent upon drugs (illicit and prescribed opioids; stimulants and benzodiazepines) in the perinatal period?
2. What approaches have been shown to be most effective at meeting the needs of women who are dependent upon drugs (illicit and prescribed opioids; stimulants and benzodiazepines) in the perinatal period?
3. To what extent do women who use drugs (illicit and prescribed opioids; stimulants and benzodiazepines) in the perinatal perceive these approaches to meet their needs?

This systematic review is concerned with both effect and experience thus lending itself to a mixed method systematic review methodology (JBI ch8.) There has been an increase in mixed method systematic reviews with service providers and policy makers appreciating the usefulness in bringing together all types of research evidence relating to a specific health problem, or topic. As such, much has also been written about different methodological approaches to analysing, synthesising and reporting mixed method reviews (Hong et al., 2017; Pearson et al., 2015; Sandelowski et at., 2012). In this review we will adopt a convergent segregated mixed methods approach as outlined in the Joanna Briggs Institute (JBI) manual chapter 8 for mixed evidence synthesis (Lizarondon et al., 2020) .This approach reflects the range of research questions that have been posed and will allow us to identify and fully appraise the different types of available data, before considering how they relate to each other, and what overall insight they may offer together in answering the research questions.

We will use an adapted version of the preferred reporting items for systematic reviews and meta-analysis protocols (PRISMA-P). An application has been submitted to PROSPERO for the registration of this rapid mixed methods systematic review

Inclusion/exclusion criteria:

The inclusion and exclusion criteria will be developed in consultation with the EACG and refined by two reviewers. Publications will be English language, year range 1990-2022, and will include both peer reviewed articles and grey literature. All study designs will be included, quantitative (RCT (parallel and crossover), cohort studies, cross sectional, and case-control) qualitative and mixed methods if the study seeks to address perinatal substance use. Studies not reporting on interventions for perinatal substance use will be excluded.

Search strategy:

The search will have a limited scope based on topic and end-user needs, focusing on established electronic databases, specifically Medline, PubMed, Global Health, Psych Info, CINAHL, ASSIA and EMBASE. The initial search string will utilise keywords to describe publications which report on care and intervention models for perinatal substance use that meet the relevant eligibility criteria. These search terms will be expanded and modified to include MESH terms and bullion phrases as appropriate for each specific data-base.

A separate combination of keywords will be used to search for qualitative literature, and as well as searching the listed electronic databases we will also include searches of google scholar and other internet based platforms that may help to identify unpublished or grey literature.

Record selection and extraction:

Relevant electronic records will be exported and stored in EndNote. Duplicate records will be removed. The titles and abstract review of a sample (50 records) will be carried out by two reviewers against the inclusion and exclusion criteria. The search criteria will then be clarified as needed. After this process, a single reviewer will review the title and abstract of all results against the inclusion/exclusion criteria in order to accelerate the review process. A second reviewer will independently screen a sample of 25%. Disagreements will be resolved through discussion, and if required a third reviewer will be mediate.

All publications eligible for inclusion will be retrieved in full, and a full text review will be conducted by a single independent reviewer, with 25% screened by a second reviewer. Any disagreements will be resolved by discussion and if required screened by a third independent reviewer. Forward and backward citation tracking will be applied to included studies and any further identified studies will be screened in the same way as described above, first by title and abstract, and then in full text using pre-defined inclusion and exclusion criteria.

A modified version of the Template for Intervention Description and Replication (TIDieR) will be used to extract data, as it provides a comprehensive framework for the description of interventions in both systematic reviews as well as primary reports (Hoffmann et al., 2014).

Assessing quality and risk of bias.

We will use the mixed methods appraisal tool (MMAT) (Pluye et al., 2014) to assess quality and risk of bias in the included studies and present the findings in a table.

Data synthesis and analysis:

We will start by categorising high-level findings in a table (e.g., study population, intervention type, outcome / effect, and recommendations). The studies will then be grouped and analysed according to study designs (Quantitative, qualitative, and mixed methods). They will be further grouped by intervention type, within these categories. We will describe in narrative the types of interventions identified before proceeding to analysis of the evidence or each.

Quantitative studies

Where possible, we will pool information pertaining to study/intervention, impact, and outcomes, conducting meta-analysis where appropriate using forest plots. We anticipate there may be high levels of heterogeneity in both study design and outcome measures and that this may not be possible for many of the included studies. We will also describe the available data using narrative synthesis.

Mixed Method Studies

Where appropriate / possible data will be extracted and analysed separately for the quantitative and qualitative components of the study. If data reported is not sufficient to be extracted in this manner a narrative synthesis will be provided.

Qualitative studies:

Findings of qualitative studies can be unclear. We will extract author reported themes, subthemes, and conclusions. Where possible we will undertake thematic content analysis to identify common categories and themes within these categories using the research questions as a-priori headings. Should the data reported not be rich enough for such an analysis, a narrative synthesis will be provided.

Synthesis:

The findings from each part of the data analysis and synthesis will be brought together in narrative synthesis. The qualitative, quantitative, and mixed method findings will be compared and juxtaposed for similarities and differences. The review questions will be used as a priori headings to present an overall synthesis of the different data, and analysis.

Reporting

We will use the JBI mixed method systematic review reporting guidance (JBI 2020) together with PRISMA – P to ensure transparent and full reporting of our methodology, analysis and findings.

Outputs: Preliminary results of the review will be shared with the EACG members in a Webinar to seek feedback from members on key findings. The draft review report will then be shared with members of the Expert Advisory Group and their views solicited on the interpretation of results and how findings can feed into the development of good practice case studies and models in Phase 2 and 3 of the study.

PHASE TWO (6-24 months)

Aim

To explore how women who are drug dependent experience services and their care journey, and how these experiences impact on engagement and outcomes for women and their infants.

Objective

To critically evaluate women's experiences of health and social care, their care journeys and outcomes for the family, from confirmation of pregnancy to 18 months postnatal.

Design

A longitudinal case study involving qualitative data collected in four case study sites.

We currently know little about models of integrated/multidisciplinary maternity and postnatal care for women who use drugs and their children in the UK, how substance use, maternity services and child protection services work together to deliver care and how women experience and negotiate these care pathways over time. Our study is informed by the work of critical social work theorists who have highlighted how social work systems in the UK have increasingly developed to manage institutional risk rather than to enable families to flourish within supportive networks and who seek to reimagine child protection (Featherstone, Gupta, Morris, & White, 2018).

METHODOLOGY

Our objective in this study is to provide a detailed understanding of how women navigate health, social care, and maternity services from preconception to up to 18 months postnatally and their experiences of barriers and enablers to effective care. Our study will adopt Qualitative Longitudinal Research (QLR), an approach that seeks to explore lives in depth as they develop in time (Farrall, Hunter, Sharpe, & Calverley, 2016) and to make sense of experiences as they unfold over time' (Miller, 2015). QLR is applicable to research questions that "relate to the life course, trajectories, and critical moments, as well as the motivations and experiences of biographical change" and for better understanding of the complexities of processes associated with change (Morrow & Crivello, 2015). The QLR approach advances how individual trajectories are mediated through a cultural turn, exploring detailed textures of social life (Neale & Flowerdew, 2003). By adopting a QLR approach we aim to understand the events and experiences in participants' lives that they identify as significant, focusing on those relevant to their health and care needs in pregnancy and postnatally. This is a resource intensive approach involving a highly vulnerable population.

Data will be collected through:

I. Timelines drawn by women over the course of the study, enabling the researcher to reflect on the procession of time and change alongside the participant (Gramling & Carr, 2004; Monico, Ludwig, Lertch, & Mitchell, 2020)
II. Serial qualitative interviews x 5 (Murray et al., 2009) with women will be conducted to capture longitudinal data (200 interviews)

III. Audio/video diaries in order to capture responses to events and experiences as they happen and as a stimulus for in-depth interviews (Bernays, Rhodes, & Jankovic Terzic, 2014; Gramling & Carr, 2004).

IV. Routinely collected NHS prescribing data. With permission from participants, these data will be collected from the Prescribing Information System (PIS) in Scotland and the English Prescribing Dataset (EPD).

V. Focus groups and individual interviews will be conducted with professionals (28 interviews)

6. STUDY SETTING

We will carry out a qualitative longitudinal study over an 18-month period in four contrasting sites in order to map women's trajectories across the care pathways. This will include two sites in England (Homerton University Hospital/City and Hackney CCG and Leeds and York Partnership NHS Foundation Trust, Leeds University Teaching Hospital and NHS Leeds CCG) and two in Scotland (NHS Greater Glasgow and Clyde and NHS Ayrshire and Arran).

In order to select sites, heads of maternity services in Scotland and England were contacted, and information was requested about service models for women/mothers who use drugs and their infants and on contextual factors such as number of annual births, prevalence of drug use in the general and maternal population, admission to neonatal units for NAS and removal of infants associated with drug use. We selected sites with a high prevalence of drug use and deprivation with differing approaches to providing integrated services for women/mothers who use drugs and their families. We aimed to select sites with higher/lower than average numbers of removal of infants and higher/lower than average numbers of babies admitted to neonatal units for NAS. An issue in describing sites is that recording and reporting of data may be incomplete and is not always comparable between Scottish and English sites. We also considered issues such as the service's interest in participating in the study and the need to avoid sites where other research involving the same population is currently ongoing.

<u>Leeds</u>

Leeds Teaching Hospitals NHS Trust is a large maternity service in the north of England, with around 10,000 annual births. Data extracted from the Maternity Booking Data System indicate that of 10,184 maternity bookings in 2019, 70 women were recorded to be a current user of any substances (0.7%)(Goldsborough, 2020). In Leeds Teaching hospitals, all babies born to women identified as using substances are admitted for observations and/or treatment to Transitional Care (for babies requiring extra care). In 2018, 54 babies were admitted with their mothers to Transitional Care of whom 16 received pharmacological treatment for neonatal abstinence syndrome (0.16 % all babies born). Rates

of infant removals appear to be high. In 2018, 58 new-born babies under one week (0.6% of births) and 84 babies under three months (0.9%) became looked after under interim and full care orders by Leeds City Council, Children Services. Drug related deaths in the general population in Leeds are 5.5 per 100,000.

Hackney

Homerton Hospital NHS Foundation Trust is a medium sized maternity service in the South East of England, with around 5,500 annual births. In 2018, 33 babies (0.6%) were born to mothers who used opioids/crack and who were at risk of developing neonatal abstinence syndrome (London Borough of Hackney, 2019). 19 babies were diagnosed with NAS of whom 13 received pharmacological treatment in the neonatal intensive care unit (0.22 of all babies born). The Joint Strategic Needs Analysis for the London borough of Hackney (2019) reports a particularly high incidence of substance use among young women in the borough with 11 new presentations to substance use treatment services as pregnant in 2018/19, 5% of all new presentations compared to 3% nationally. Rates of infant removals is lower. In the London Borough of Hackney, in 2019/20, 8 babies became looked after within 10 days of birth (0.16%) and 13 babies (0.3%) became looked after within three months' of birth. Drug related death in the general population in Hackney are 7.3 per 100,000.

Glasgow

NHS Greater Glasgow and Clyde is a large maternity service in west central Scotland, with around 11,600 annual births, serving a population with high levels of socio-economic deprivation. Glasgow has a high prevalence of drug use in the general population (2.1% compared to 1.7% for Scotland) and a higher rate of drug related deaths (35.6 per 100,000 compared to 24.3 in Scotland) (Scottish Public Health Observatory, 2021). Despite this, maternities affected by drug use are reported to be relatively low at 7.8 per 1000 births and the number of babies affected by maternal use of drugs is reported to be 0.6 per 1000 live births. However, these data may be affected by inaccurate reporting. The rate of child protection associated with drug use is 5.3 per 10,000, however, these data includes all ages.

Ayrshire & ARRAN

NHS Ayrshire and Arran is a small/medium maternity service in the southwest of Scotland (3,107 annual births) serving a mixed urban and rural population with areas of high socioeconomic deprivation. Prevalence of drug use in the general population is 1.7%, drug related deaths are high (34.5 per 100,000) (Scottish Public Health Observatory, 2021). 16.7 per 1000 maternities are reported to be affected by drug use with 2.8 per 1000 babies reported to be affected by maternal use of drugs. The rate of child protection associated with drug use is 7 per 10,000.

Table 1. Summary description of research sites

Site	location	Maternity service	Drug related deaths	Babies affected by drug use	Infant removals
Leeds	North of England	Large	low	0.16%	Higher
Hackney	London	medium	medium	0.22%	Lower
Glasgow	Central Scotland	Large	Very high	0.06 (incomplete data)	Not comparable
Ayrshire	S/W Scotland	Small/medium	Very high	0.28%	Not comparable

Already it is clear that these sites vary in whether they focus on the antenatal or postnatal period, whether they primarily target the wellbeing and protection of the child and/or the support and psychoeducation of the mother; and in the range of biomedical, psychosocial and practical treatment services and support they provide. Furthermore, the sites differ substantially in terms of policy contexts, socio-economic status, geography, culture, ethnicity of service users, availability of illicit drugs and types of polydrug use. Understanding how these wider contextual factors affect models of care, care pathways and outcomes for families will be a key focus of this study.

7. SAMPLE AND RECRUITMENT

We will recruit 40 women who are dependent on drugs (n=10 per site) from maternity services and substance use treatment services approximately 30 of whom will be recruited to the study prenatally and 10, between 2 and 9 months postnatally. This sampling strategy acknowledges that women who use drugs may present late to maternity services and after the window of 10 weeks within which the NHS recommends the first booking appointment takes place and enable us to sample women whose babies are suffering from more severe Neonatal Abstinence Syndrome. In the two sites in England, some women who use and who are in treatment for drugs use in pregnancy are referred postnatally to

a residential detoxification and rehabilitation centre in Plymouth. We will also recruit women postnatally from this service in England.

Additionally, we will also enrol GPs, midwives, health visitors, social workers and substance use treatment staff (21 per site) for online focus group (or individual interviews if preferred) interviews in each site. We will also seek to enrol 16 other key informants e.g., service managers, commissioners, policymakers (4 per site) for individual interviews.

Sample

In each area 10 women will be recruited from maternity services and substance use treatment services. Approximately 30 of whom will be recruited to the study prenatally and 10 women, between 2 and 9 months postnatally.

Eligibility Criteria

- Pregnant or between 2 and 9 months postnatal
- Dependent on one or more of the following drugs: prescribed opioids (e.g. methadone), illicit opioids (e.g. heroin), benzodiazepines, cocaine/crack or amphetamines. (And alcohol where it is used in conjunction with these drugs)
- Over the age of 18
- Speak English and can provide informed consent.

Exclusion Criteria

- Uses alcohol without drugs
- Under the age of 18
- Does not speak English

Focus group interviews will also be conducted with professionals in each site who will include staff working in maternity, child health, primary care, addiction (NHS/3rd Sector), sexual health, social services and other agencies (e.g., housing, police). We will also recruit service managers, commissioners, and policy makers for individual interviews in each site.

Recruitment

1. Specialist midwives at participating antenatal services and key workers at substance use treatment services will inform eligible women about the study and give them a flyer with a comic strip explaining what the study is about and including a QR code link to the study website and/or a business card with the study logo and a QR code. The study website will house a short video which will explain what the study is about and what would be involved in participating. The website will also have a page with information about what will happen in relation to participant data and our privacy statement. If women are interested in hearing more, midwives and key workers will obtain permission from the potential participant to pass their contact details on to the research team who will later call the potential participant to discuss the aims of the study in more detail, explain what taking part involves and arrange a time to meet the potential participant face-to-face to go through the participant information sheet and obtain informed consent. Prospective participants may also contact the research team via a contact sheet on the study website. The researcher will check with the midwife or key worker if there are any known risks in visiting the participant in her own home.
2. The *Participant Information Sheet* (PIS) will be given to potential participants by the researcher, prior to eliciting informed consent.

In addition, the researcher will verbally explain the study to potential participants.
The researcher will: assure potential participants of confidentiality, what to expect during the study and give them contact details in case of complaint or need for further information; will inform potential participants that participation is voluntary and that they can withdraw at any time without affecting their care. The limitations to confidentiality will be explained to potential participants, i.e., that any disclosures of significant risk of future harm to self or others will be disclosed to their key worker or the duty worker in the service where the interview is taking place or relevant authorities.

The researcher will give potential participants the opportunity to ask any questions about participating in the research. If the potential participant is still interested, the researcher will invite them to participate.

3. Obtaining Informed Consent

Written informed consent will be obtained from the participant by the researcher. The *consent form* (see Consent Form) will be counter-signed by the researcher. The consent form includes agreement for

audio-recorded interviews and professional transcribing; long term storage of data; and optional consent for access to GP and/or prescribing records.

At this time, the researcher will take the following contact information from the participant to enable the researcher to remind participants of their interview time (see Contact Details Form):

- contact details – mobile, house phone, email, address (ask participant their preferred contact method for contact and for sending summary report if requested);
- In addition, participants will be asked for their consent for researchers to liaise with the service from which they were recruited if it is not possible to contact them through the contact details they have provided.

If the participant can do the in-depth interview at the same appointment as obtaining written consent, then the researcher will conduct the first interview. If this is not possible, the researcher will arrange a future date/time for the researcher to conduct the interview.

Researchers should check participants' preferred method of communication and contact times by referring to the contact details form. If there are no instructions regarding participants' preferred methods and times for contact, researchers should establish these on your first contact.

The researcher will contact the participant in advance of the interview to remind them of the appointment. In addition, they will contact them the day before by text or WhatsApp message and on the date agreed to remind them. As the researcher will have gained participants' consent to liaise with their service-provider, the researcher will also be able to track participants that they are unable to reach through the contact details provided, or in the event of not showing up to an agreed appointment for interview.

Identification numbers will be allocated to participants for research purposes. The researcher will state the unique participant ID, NOT the participant's name or other personally identifiable information.

Preference is to interview women in a hospital or other health care setting, in children's centres or in local services, depending on the availability of interview rooms in each site. If none of these is possible, then interviewers could conduct interviews in women's homes or by telephone. If the first interview is scheduled to take place in a woman's home, two researchers should attend.

Data Collection

In the first four months of field work, researchers will recruit, develop rapport and conduct initial Timelines, and base line interviews, introducing the idea of audio diaries for subsequent research engagement. They will maintain regular contact with research participants between interviews with weekly text and WhatsApp messages. Where possible researchers will conduct in person in depth qualitative interviews and Timelines but these methods can also be conducted online or by telephone should this be necessary.

Timelines

Prior to the base line interview, participants will be invited to draw a **Timeline**, a visual data-collection method, depicting a chronology of events including the development of drug use, engagement with treatment, access to support and health services, intimate and family relationships, previous pregnancies, and other events they identify as significant for themselves and their families. The creation of Timelines will enable the charting of processes and change through time from the participants' perspective, including access to, engagement with, and drop out of services; substance use and relapse; the events around the birth, children becoming looked after and changes in treatment and care. Because Timelines place health conditions and behaviours in the context of a person's life history, they have been found to be particularly helpful to visualise substance use and treatment journeys (Berends & Savic, 2017) and to facilitate the recollection and sequencing of personal events as well as revealing the multiplicities of lived experience (Sheridan, Chamberlain, & Dupuis, 2011). With the agreement of the research participant, the researcher will retain the Timeline drawings and diagrams for systematic analysis and will make careful fieldnotes following the Timeline drawing exercise and interviews.

Data from GP records

With women's permission we will access their GP records. Information will be drawn from these records on mental and physical health and substance use treatment. These data will be added to the Timelines.

Serial Qualitative Interviews

In-depth serial interviews with mothers of approximately 60 minutes will be conducted at baseline, followed by four further interviews (at four/five-month intervals). Life history interviews will be followed by interviews that will elicit women's changing experience of treatment and services over time. Care trajectories will be mapped in relation to access and engagement with services.

Audio/ Video Diaries

Participants will also be invited to record short, weekly audio or written diaries on their own mobile phones or in notebooks in order to capture their responses to critical events as they happen. Researchers will send text and Whattsap messages reminding participants of the option to record their diary entries. Like visual Timeline data, the audio diaries will provide a stimulus for and will augment the insights revealed by in-depth interview data. The diaries will be captured by researchers in field notes (Bernays et al., 2014) and will not be transcribed. While pregnant women who use drugs may be considered difficult to engage in research our experience is that once rapport is established women are often keen to talk about their experiences. Three mothers with experience of drug dependence, who were consulted individually about the proposed research methods, reported that in their view, women receiving treatment for drug use would be comfortable both drawing Timelines in order to describe key events in their lives to researchers and to record audio and/or video diaries.

Focus groups and individual interviews with staff.

In each site, networks of professionals (GPs, midwives, social workers, substance use staff, and health visitors) providing services to the women taking part in our study will be identified at the initial site set up meetings and by snowball sampling thereafter. Staff will be contacted via work address or email, sent information about the study and invited to take part in online focus group (or individual) interviews using MS Teams. Informed consent will be sought from each participant using a verbal consent protocol prior to the start of the interviews.

Three focus groups of 6-8 practitioners will take place in each site. In-depth telephone/online individual interviews will also be conducted with four key informants (e.g., service managers, commissioners) in each site. Focus group and stake holder interviews will be used to test a range of archetypal 'journey maps' derived from timeline analysis and interviews with women in each service and will thus serve to verify the development of applied models of care and best practice. Topics will include: the nature and extent of family planning and sexual/reproductive healthcare for women experiencing Substance Use Disorder; the booking appointment, midwifery and obstetric care and involvement with specialist services; substance use treatment and care; comorbidities and blood borne virus care; extent of wider social support (e.g., assistance with housing, welfare benefits, transport etc); breastfeeding advice and support; preparation and support for caring for a baby with Neonatal Abstinence Syndrome (NAS); pre-birth safeguarding; delivery of care within the labour ward/postnatal ward and the neonatal unit; post-

birth discharge planning and child protection/parenting support interventions. Funds have been allocated for the transcription of focus group and individual interviews.

Data Analysis

Data collection and analysis will run concurrently. Timelines will be analysed for both content and form and in relation to the interview data at the time they were produced. Timelines, interview data and field notes will be thematically coded to map out women's relationships to services in order to build archetypal 'journey maps' which will be presented as case studies for discussion at professional service focus groups and stake holder interviews. In turn, information and interpretation from the focus group will guide recommendations for models of care, intervention, and best practice.

Focus groups and individual interviews will be transcribed by an approved contractor. Transcripts will be checked for accuracy and anonymised. A coding framework will be collaboratively developed by the research team and tested by each researcher independently coding a sample of transcripts. Transcripts, timelines and field notes will be uploaded to Nvivo. Thereafter, researchers will independently code interview transcripts and field note data, using Braun and Clarke's (2006) method of thematic analysis. Analysis will be oriented to mapping women's relationships to services in order to build archetypal 'journey maps' which will be presented as case studies for discussion at professional service focus groups and stake holder interviews. In turn, information and interpretation from the focus group will guide recommendations for models of care, intervention, and best practice.

Phase 2 case studies will compare local care pathways, policies and protocols between sites and in relation to the good practice models identified in Phase 1, furnishing a descriptive typology of the research sites. Reviews of evidence and of clinical and good practice guidance will thus inform the empirical research in which we will address women's experiences and responses to the arrangements of services and models of care. The coproduction and dissemination activities in Phase 3 will draw on findings of both Phases 1& 2.

PHASE THREE (24-30 months)

Aim

1. To determine the optimal service model/s for drug dependent women (from preconception up to18 months postnatal), to foster good parenting and to provide a safe, stable and nurturing

caregiving environment for the mother, infant and family as a whole.

2. To identify/ create the optimal best practice guidance for the care of drug dependent mothers and their infants, to maximise engagement with services, maternal and infant outcomes, and to prevent out-of-home care placements.

Objective

To co-produce an optimal service model for women who are dependent on drugs, child and family-centred care and insights for future care and practice guidance to optimise outcomes for mothers, infants and the family.

Design

Phase 3 will involve Coproduction and public involvement as described in Phase 1. Specifically Phase 3 will involve the following stages of co-production:

- Market it– we will develop a range of outputs to ensure maximum reach and impact. For each output we will consider appropriate social marketing and behaviour change tools to maximise impact.
- Grow It– We will seek further funding to support implementation of the optimised model at scale across the UK.

Throughout the study, analytic summaries of emergent themes and case studies will be prepared for discussion in the Expert Advisory and Co-Production Group, which will inform subsequent data collection and the analytic focus. Our Expert Advisory and Co-Production Group will develop and oversee our long-term engagement plan. The co-production approach will provide a pathway to dissemination and uptake of findings. Members of the group will discuss and debate study findings and will co-produce outputs including policy briefings and infographics for different stakeholders to communicate key messages from our research, so they are relevant and accessible.

We will seek further funding to support implementation of the optimised model at scale across the UK and to undertake an evaluation using implementation science approaches. We will advocate for ring fenced NHS funding to support improved joined-up services for women who are dependent on drugs and their babies including through the establishment of an All-Party Parliamentary Group on perinatal care pathways for women who use drugs.

8. ETHICAL AND REGULATORY COMPLIANCE

The study will not be initiated before the protocol, consent forms and participant information sheets have received approval / favourable opinion from the Research Ethics Committee (REC), the respective National Health Service or other healthcare provider's Research & Development (R&D) department, and the Health Research Authority if required. Ethical approval is not required for Phase one but the systematic review protocol will be submitted to PROSPERO as required by NIHR. Application for ethical approval for Phase two will be made to the Health Research Authority, and to King's College London following the award of the grant. Before any site can enrol participants into the study, the Chief Investigator/Principal Investigator or designee will ensure that appropriate approvals from participating organisations are in place. Specific arrangements on how to gain approval from participating organisations are in place and comply with the relevant guidance.

Should a protocol amendment be made that requires REC approval, the changes in the protocol will not be instituted until the amendment and revised informed consent forms and participant information sheets (if appropriate) have been reviewed and received approval / favourable opinion from the REC and R&D departments. A protocol amendment intended to eliminate an apparent immediate hazard to participants may be implemented immediately providing that the REC are notified as soon as possible and an approval is requested. Minor protocol amendments only for logistical or administrative changes may be implemented immediately; and the REC will be informed.

For any amendment to the study, the Chief Investigator or designee, in agreement with the sponsor will submit information to the appropriate body in order for them to issue approval for the amendment. The Chief Investigator or designee will work with sites (R&D departments at NHS sites as well as the study delivery team) so they can put the necessary arrangements in place to implement the amendment to confirm their support for the study as amended.

Research will be conducted in accordance with the ethical principles that have their origin in the Declaration of Helsinki, 1996; the principles of Good Clinical Practice and the UK Department of Health Policy Framework for Health and Social Care, 2017. GDPR regulations will be adhered to.

Our project involves research on drug use, child welfare, health, relationships and other highly sensitive topics with vulnerable children and families, and frontline health and social care professionals, in contexts that could cause distress both to the research participants and the researchers. The research subject matter, proposed methods, and vulnerable study population, raise important ethical and governance issues which require careful management, planning and review. We describe potential risks to participants and researchers below. We will obtain NHS Research Ethics Committee approvals to conduct this study and we will be guided by UK Research Ethics and Governance Frameworks which provide guidelines on the principles, conduct and regulatory requirements of health and social care research (e.g. Health Research Authority 2017; RCUK updated 2017).

KEY ETHICAL CONSIDERATIONS involved in research with drug-using parents include: informed consent, confidentiality, child and adult protection, handling sensitive topics, researcher safety and wellbeing, anonymity and data protection. These are briefly discussed below:

We will ensure that recruitment into the study is entirely voluntary and written or audio recorded verbal informed consent is obtained from all research participants. We will monitor consent on an on-going basis. Likewise, our field work in designated health and social care services will require written agreement from senior managers as well as informed consent from practitioners located within each service.

POTENTIAL RISKS TO PARTICIPANTS

Confidentiality will be important to maintain, given the sensitive nature of issues around drug-related behaviour, parenting and child welfare. However, confidentiality cannot be guaranteed 100% and we will explain the limits of confidentiality, for example, when a child or vulnerable adult is at risk of significant harm. In our experience, when these topics are discussed openly and sensitively, most parents and family members understand these limits, agree with the required responsibilities and actions of the research team and consent to take part in the study. Our researchers will adhere to local (site specific) inter-agency child and adult protection procedures and will be supported by trained and experienced members of the research team.

Participation is voluntary and participants are notified in the oral consent process that there is a potential risk that they may experience some emotional upset. Participant distress will be minimised through monitoring and termination of the interaction at that point, if necessary. All participants will be supplied with materials providing signposting to relevant support services. Any answers suggesting a risk of harm to participants or others will be shared with participant's key worker/s or support services. If it is necessary emergency services would be informed. Such breaches of confidentiality will only take place

as a last resort, and the researcher will inform the participant about who they have contacted, and the information disclosed. In the Oral consent process participants are made aware of the researchers' duty of care if we feel participants experience undue distress or say that they will harm themselves or someone else

POTENTIAL RISKS TO RESEARCHERS

This project involves lone working, home visits and fieldwork in contexts which may pose a risk to the researcher because of, for example, exposure to environmental hazards, distressing information, and disturbing or risky situations. These risks can be mitigated by implementing a robust fieldwork protocol which includes procedures to carefully plan, manage and conduct the field work as well as vigorous arrangements for researcher debriefing, support and supervision. In addition, research governance arrangements (e.g., NHS research passports for researchers, indemnity insurance, reporting of adverse events) and our data management plan (Appendix A) will be included in our ethics application. This will include, for example, compliance with GDPR legislation and the Data Protection Act (2018). Funds have been included in the budget for personal alarms for researchers.

9. Dissemination and Implementation

We have developed this proposal through extensive engagement with multiple stakeholders, including addiction, public health, maternity services, and service user representatives. As evidenced by our letters of support, there is enthusiasm to engage in this study and maternity services have indicated that there is an urgent need to develop the evidence base to inform service development in this area. Our interdisciplinary research team has substantial relevant academic and professional experience to engage in the short and longer term with NHS and Social care service providers, third sector and with service users. We will disseminate our findings to the following audiences:

Academic and Research Community: We have included funds for the publication of two open access papers which will be published in high impact journals such as Addiction and Child Abuse and Neglect in order to ensure global reach and access. We will also disseminate findings via specialist conferences such the International Society for the Study of Addiction and BASPCAN - British Association for Child Protection. Research and practitioner networks will also be utilised to disseminate reports e.g. the Institute of Health Visiting, the Scottish Alcohol and Drugs Delivery Reform Group, Public Health

Research Networks, NSPCC Caspar, research in practice. A study report will also be written and showcased at the commissioner/practitioner dissemination event.

Policy makers, commissioners and service providers: Representatives from Public Health England and the Scottish Government (Safer Communities Directorate, Directorate for Children and Families and Health and Social Care Directorates), The Royal College of Midwives, Royal Colleague of Paediatrics and Child Health (RCPCH), Royal College of Obstetrics and Gynaecology, addiction charities and programmes (Trevi House, Scottish Drug Forum, Action on Addiction), Adfam, the Institute for Health Visiting and the Maternal Mental Health Alliance will be invited to join our Expert Advisory Group to advise on effective dissemination to policy makers and service providers. They will assist in the development of lay summaries for our outputs and advise on key policy recommendations. We will also publish our research in professional journals to improve reach and impact. Existing practitioner networks will be utilised to disseminate findings including e.g. With Scotland and Social Work Network, National Safeguarding Midwife Network.

Service users and the lay public: four women with lived experience of drug use will be invited to join our Expert Advisory and Co-Production Group to ensure service users have a voice throughout the study and in the dissemination of findings. Funds have been allocated to reimburse these four women for taking part in the Expert Advisory and Co-Production Group, for Scottish mothers to be supported by peer support practitioners working for Drugs Research Network, Scotland and for English mothers to be supported by peer support practitioners working for the Addiction Service User Research Group, King's College. These are organisations which involve and represent people who use drugs on drug-related issues and in research. Peer support practitioners will ensure that service users are involved in the translation of our findings for the lay public and can also disseminate our findings through their media bulletins and websites. We will also work with King's College and the University of Stirling Press Offices to design and implement a media strategy. Service users will be invited to take a lead role in planning and delivering our expert event which is designed to disseminate study findings.

Expert Events (half day events): We will host half day 'Expert Events' at the end of the study in Scotland and England to which our policy leads will be invited to speak to share best practices, showcase exemplars and case studies from the four research sites. We will establish co-creative solutions to problems of implementation, as well as identify barriers to service enhancement and delivery for specific organisations and communities of practice social care from early pregnancy to up to 18 months after

the baby is born. By adopting need to be adapted to each practitioner group. Such a forum can also be used to test out tentative interventions and assess the acceptability and feasibility of proposed models/interventions. These half day events in Scotland and England will include service user and public perspectives, and contribute to briefings and media, and practitioner events.

Provider/practitioner dissemination workshops (half day events): Best practice summaries and briefings, carefully narrated through collaboration with media and communications experts, will be shared through multiple media. This will be targeted to service providers, practitioners, commissioners, and service users. These products will be launched with the project report in half day practitioner workshops (one in Scotland and one in England) to promote the adoption of the findings and engage stakeholder in mobilising knowledge into local practices. The practitioner events will have a focus on practice and will provide the opportunity for service providers from the case study sites to join with the research team and co-production group in mobilizing knowledge and engaging practitioners from across the UK in adopting study findings. These national workshops will take place in easily accessible locations to maximise participation.

DATA MANAGEMENT AND STORAGE:

All researchers and study site staff will comply with the requirements of the Data Protection Act (2018) and the principles of GDPR with regards to the collection, storage, processing and disclosure of personal information and will uphold the Act's core principles. A detailed data management plan is included as **Appendix A**. All researchers will have completed their institution's training on GDPR and Confidentiality Quiz and HRA Good Clinical Practice online training.

Audio files and other electronic data will be encrypted and stored on a password protected network drive hosted by King's College, London and the University of Stirling. Passwords only known by the immediate research team.

Consent will be sought to audio-record the interviews, and researchers will explain the purpose of these recordings. Only participant identification numbers will be stored on the audio files and on the transcribed files.

Audio Recordings

Encrypted digital recorders will be used to record interviews and focus groups securely. Following an interview or focus group data will be transferred from the digital recorder to central databases as soon as possible, and once the recording is checked, it will be deleted from the digital recorder. Audio-

recordings will not be stored on the recording device. Audio files will be downloaded onto a secure server at each participating site (King's College London, University of Stirling). As soon as the audio-file is checked on the server, it will be deleted from the recording device.

To ensure confidentiality of participants, the researcher will assign unique participant IDs to the transcripts, known only to appropriate members of the research team. The researchers at each site will read transcripts and de-identify any identifiable information. Researchers will pseudonymise all sensitive and special category data using a series of codes. Personal information will be deleted from the transcripts to ensure that names of participants and others, names of services, service providers or cities are de-identified or where appropriate pseudonyms used.

Electronic sound files of interviews will be archived in a secure location for a minimum period of 7 years following the end of the study. Names/contact details of consenting participants linking them to the datasets will be stored in password protected files on a secure server at each of the sites (King's College London, University of Stirling). Only the researchers will have access to these data. When necessary, encrypted emails will be sent containing relevant attachments between research teams. Researchers from King's College London, the University of Huddersfield and the University of Stirling will view the anonymised transcripts for the purpose of analysis.

In accordance with UK Data Service guidelines and General Data Protection Regulations (GDPR), all participants will be given (and encouraged to retain) an information sheet outlining the goals and motivation for the project, who is undertaking it, the funder, and plans for dissemination and use of results. The information sheet also explains the confidentiality and anonymity processes planned and that participants' responses will not allow them to be identified.

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11. APPENDICIES

Appendix 1 Coproduction plan