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Rethinking Strategies for Positive Newborn Screening Result Delivery

Submission date	Recruitment status	[X] Prospectively registered
15/01/2018	Suspended	[X] Protocol
Registration date	**Overall study status**	[_] Statistical analysis plan
17/01/2018	Completed	[X] Results
Last Edited	**Condition category**	[_] Individual participant data
31/10/2022	Genetic Diseases	

Plain English summary of protocol

Background and study aims
Each year about 800,000 babies in the UK have a blood test taken (called newborn bloodspot screening (NBS)) to screen for specific conditions, which if treated early will improve the child's health and well-being. In 2015-16, over 10,000 babies were identified as being affected or healthy carriers of a gene for one of the conditions screened for, which include sickle cell disease, cystic fibrosis, metabolic conditions and hypothyroidism. When a positive result occurs, a variety of ways are used to deliver the result but many parents complain about the approaches used. The aim of this study is for parents and health professionals to work together to design interventions to facilitate effective communication of positive NBS results to parents by health professionals. This study includes four phases.

Who can participate?
Health care professors in NBS and parents of children who have received a NBS+ result in the previous 3-12 months.

What does the study involve?
In the first phase of the study, staff are invited to participate in telephone interview lasting 30-45 minutes. In the second phase of the study, staff are observed for 60 minutes on up to 5 occasions and are also invited to take part in semi-structured interviews lasting 60 minutes, participant in a staff feedback event lasting 120 minutes, and work in co-design working groups each lasting 180 minutes. Parents and carers are invited to participate in one hour filmed narrative interviews, feedback events (120 minutes), a staff and parent even (180 minutes) and in the co-designed working groups. In the third phase of the study, staff receive training and are invited for semi-structure interviews. They are also observed for one hour up to five occasions. Parents and carers are invited to one hour semi-structured interviews. In the fourth phase of the study includes stakeholder meeting lasting 180 minutes.

What are the possible benefits and risks of participating?
There will be no direct benefits or anticipated risks for those taking part in the study.

Where is the study run from?
This study is being run by City, University of London (UK) and takes place in hospitals in the UK.

When is the study starting and how long is it expected to run for?
March 2015 to March 2020

Who is funding the study?
National Institute for Health Research (UK)

Who is the main contact?
Dr Jane Chudleigh (Public)
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Study website
https://www.city.ac.uk/news/2017/september/rethinking-strategies-for-pos

Contact information

Type(s)
Public

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Additional identifiers

EudraCT/CTIS number

IRAS number

ClinicalTrials.gov number

Secondary identifying numbers
36339

Study information

Scientific Title

Rethinking Strategies for Positive Newborn Screening Result (NBS+) Delivery (ReSPoND): A process evaluation of co-designing interventions to minimise impact on parental emotional well-being and stress

Acronym
ReSPoND

Study objectives
Can parents and staff co-design interventions to improve delivery of positive Newborn Screening Result (NBS) results to parents that can be successfully implemented into routine practice in a cost effective manner?

Ethics approval required
Old ethics approval format

Ethics approval(s)
London-Stanmore Ethics Committee, 01/01/2018, 17/LO/2102

Study design
Non-randomised; Both; Design type: Process of Care, Complex Intervention, Other, Qualitative

Primary study design
Interventional

Secondary study design
Randomised controlled trial

Study setting(s)
Hospital

Study type(s)
Treatment

Participant information sheet
Not available in web format, please use the contact details below to request a patient information sheet

Health condition(s) or problem(s) studied
Genetic screening in newborn

Interventions
This study contains four phases. Phase one is planned for six months. The second phase is conducted after six to 12 months.

Phase 1 (0-6 months):
This involves a conducting a national survey using semi-structured telephone interviews with all 13 NBS-laboratories (NBSL) in England and representatives of clinical teams(n=40) for each of the four condition specific group (CSGs). These will determine current approaches used for

communication of NBS+ results from laboratory to parents for each CSG and inform selection of case study sites using a predefined sampling framework. Quantitative data is analysed using descriptive statistics, qualitative data are analysed using content analysis.

PHASE 2: uses Experience-based Co-design in two selected study sites (from Phase 1)
1. Non-participant observation of 20 staff delivering NBS+ results to parents. 15 semi-structured interviews with staff (NBSL staff/Nurses/Consultants/Health Visitors/Midwives/Genetic Counsellors) involved in communicating NBS+ results.
Staff(n=15) meets to identify priorities for improving delivery of NBS+ results. Data will be analysed for themes to inform co-design working groups co-designed working groups (CDWGs) (stage 3) (6-12 months).
2. Filmed, interviews with 20 parents (ensuring representation of CSGs) exploring experiences of receiving NBS+ results to identify key themes. Parents (n=20) view a composite film of the interviews to ensure it is representative of their experiences and to identify emerging issues. Themes identified from parent interviews are made into a 30-minute composite film (6-12 months).
3. Joint staff-parent event in each study site to share experiences and view composite film. Mixed staff-parent focus groups to identify joint priorities for improving delivery of NBS+ results. Thematic analysis is done to identify joint priorities. (12-15 months)
4. Parents and staff from both study sites come together in 4 co-designed working groups (6-8 members each) to produce co-designed interventions for improving delivery of NBS+ results to parents (15-18 months).

PHASE 3 (18-27 months) uses two selected study sites (Phase 1).
20-30 staff involved in delivery of NBS+ results in the study sites are trained to implement the co-designed interventions for the four CSGs concurrently. Success criteria is defined and monitored on a weekly basis during implementation.

A parallel process evaluation underpinned by Normalisation Process Theory is conducted. Non-participant observation of 20-30 staff delivering NBS+ results to parents and semi-structured interviews with 20-25 parents and 20-25 staff to identify healthcare resources required for delivery of the interventions, staff and parental experiences and factors that influence implementation. These qualitative data is also used to determine suitable outcome measures for a future evaluation study. Factors parents identify as influencing experiences during delivery of NBS+ results are compared with the content of measures such as GAD7, PHQ9, Parenting Stress Index, EQ5D and ICECAP-A to determine where most overlap occurs.

Observation and interview data are used to determine how the co-designed interventions impact on parents and which outcomes and healthcare resources are important to evaluate in a future evaluation study. A cost analysis using the NHS perspective, compares costs associated with current communication practices and the new co-designed interventions. For both, resources required are defined and combined with unit costs to produce a total costs.

PHASE 4 (27-30 months):
Key stakeholders(n=10) (NBS co-ordinators/NBSLs staff/health visitors/midwives/parents) meet and the nominal group technique used to reach consensus about the need for, and potential design, of an evaluation study of the co-designed interventions.

Intervention Type
Other

Primary outcome measure

Production of co-designed, evaluated interventions for the communication of initial, positive NBS results to parent measured during the process evaluation and health economic analysis during months 18-27.

Secondary outcome measures
Phase 1:
Description of current communication practice measured during interviews with staff at 4-6 months.

Phase 2:
Co-designed interventions for the four condition specific groups produced during the co-design working group at 6-18 months.

Phase 3:
1. The cost of current communication strategies and costs associated with the co-designed interventions measured during the health economic evaluation during months 18-27
2. The acceptability and feasibility of the of the co-designed interventions measured during the process evaluation during months 18-27
3. Choice of potential outcomes measures (GAD 7 PHQ 9 PSI EQ5D and ICECAP-A) for use in a future evaluation study measured during the process evaluation and economic evaluation during months 18-27

Phase 4:
Need for and design of a future evaluation study measured during the focus group during months 27-30.

Overall study start date
01/03/2015

Completion date
31/12/2020

Eligibility

Key inclusion criteria
Parents:
Parents of children who have received a NBS+ result in the previous 3-12 months including true positives, false positives and children who later have a cystic fibrosis screen positive, inconclusive diagnosis (CFSPID). This time frame has been chosen as the focus for this research based on feedback from parents of children who have previously received a NBS+ result. It has also been demonstrated that positive NBS can impact on child-parent relationships during the first year of life.

Health professionals:
1. Staff employed in NBS laboratories and involved in the processing of NBS+ results
2. Staff who have been involved in communicating NBS+ results to parents in the last 6 months.

Participant type(s)
Patient

Age group

Adult

Sex
Both

Target number of participants
Planned Sample Size: 160; UK Sample Size: 160

Key exclusion criteria
Parents:
1. Parent of children who have received a negative NBS result
2. Parents of children with co-morbidities that are likely to influence their perception of receiving the positive NBS result
3. Parents whose baby has died prior to being approached to be involved in the study
4. Inability of parents to understand and give informed consent
5. Parents whose recruitment is contraindicated on psychosocial grounds (identified by their health visitor or specialist nurse)

Health professionals:
1. Staff who have not been involved in communicating positive NBS results to parents in the last 6 months
2. Staff who have personal experience of receiving a positive NBS result

Date of first enrolment
01/04/2018

Date of final enrolment
30/09/2020

Locations

Countries of recruitment
England

United Kingdom

Study participating centre
Addenbrooke's Hospital
Hills Road
Cambridge
United Kingdom
CB2 0QQ

Study participating centre
Birmingham Children's Hospital
Steelhouse Lane
Birmingham

United Kingdom
B4 6NH

Study participating centre
Bristol Royal Infirmary
Upper Maudlin Street
Bristol
United Kingdom
BS2 8HW

Study participating centre
Southmead Hospital
Southmead Road
Westbury-on-Trym
Bristol
United Kingdom
BS10 5NB

Study participating centre
St Helier Hospital
Wrythe Lane
Carshalton
Carshalton
United Kingdom
SM5 1AA

Study participating centre
Queen Mary's Hospital for Children
Wrythe Lane
Carlshalton Surrey
Carshalton
United Kingdom
SM5 1AA

Study participating centre
St James' University Hospital
Beckett Street
Leeds
United Kingdom
LS9 7TF

Study participating centre
Alder Hey Children’s Hospital
E Prescot Road
Liverpool
United Kingdom
L12 2AP

Study participating centre
Great Ormond Street Hospital
Great Ormond Street
London
United Kingdom
WC1N 3JH

Study participating centre
St Thomas' Hospital
Westminster Bridge Road
Lambeth
London
United Kingdom
SE1 7EH

Study participating centre
Royal Manchester Children's Hospital
Oxford Road
Manchester
United Kingdom
M13 9WL

Study participating centre
Royal Victoria Infirmary
Queen Victoria Road
Newcastle upon Tyne
United Kingdom
NE1 4LP

Study participating centre
John Radcliffe Hospital
Headley Way

Headington
Oxford
United Kingdom
OX3 9DU

Study participating centre
Queen Alexandra Hospital
Southwick Hill Road
Cosham
Portsmouth
United Kingdom
PO6 3LY

Study participating centre
Sheffield Children's Hospital
Western Bank
Sheffield
United Kingdom
S10 2TH

Study participating centre
Leeds Children’s Hospital
Leeds
United Kingdom
LS2 9NS

Sponsor information

Organisation
City, University of London

Sponsor details
Northampton Square
London
England
United Kingdom
EC1V 0HB

Sponsor type
University/education

Website
http://www.city.ac.uk/

ROR
https://ror.org/04489at23

Funder(s)

Funder type
Government

Funder Name
National Institute for Health Research

Alternative Name(s)
National Institute for Health Research, NIHR Research, NIHRresearch, NIHR - National Institute for Health Research, NIHR (The National Institute for Health and Care Research), NIHR

Funding Body Type
Government organisation

Funding Body Subtype
National government

Location
United Kingdom

Results and Publications

Publication and dissemination plan
Parents involved in the study and those who form the advisory group will be sent a summary of the research findings. The Programme Manager for the NHS Sickle Cell and Thalassaemia Screening Programme and the Programme Manager for the NHS Newborn Blood Spot Screening Programme have been approached and provided their support with regard to the implementation of these guidelines in the future. Therefore, our study findings will be disseminated on the national NBS websites (https://www.gov.uk/topic/population-screening-programmes/newborn-bloodspot, https://www.gov.uk/topic/population-screening-programmes/sickle-cell-thalassaemia) so that it may be available to staff such as health visitors, midwives and clinical nurse specialists who will be involved in the delivery of the initial positive NBS result.

Additionally, the findings will be disseminated via the website of the relevant charities and support groups associated with these conditions all of whom have been contacted and provided their endorsement for this study (CF Trust, Sickle Cell Society, British Thyroid Foundation, National Society for Phenylketonuria, CLIMB).

In addition, results will be disseminated by the usual academic routes at relevant national and international conferences and published in relevant international peer reviewed journals (including the NIHR HS&DR journal).

Intention to publish date
30/09/2021

Individual participant data (IPD) sharing plan
The data sharing plans for the current study are unknown and will be made available at a later date.

IPD sharing plan summary
Data sharing statement to be made available at a later date

Study outputs

Output type	Details	Date created	Date added	Peer reviewed?	Patient-facing?
Protocol article	protocol	04/09/2019	12/09/2019	Yes	No
Other publications	Health professionals' experiences	01/10/2020	07/10/2020	Yes	No
Other publications	Process evaluation	27/08/2021	01/09/2021	Yes	No
Interim results article	Assessment of current practice	12/12/2020	31/10/2022	Yes	No
Results article		01/07/2022	31/10/2022	Yes	No
HRA research summary			28/06/2023	No	No