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Exploring parents' experiences of their infant's 'cystic fibrosis screen positive, inconclusive diagnosis' (CFSPID) designation through interviews

Submission date
18/03/2021

Recruitment status
No longer recruiting

Registration date
19/04/2021

Overall study status
Completed

Last Edited
09/06/2023

Condition category
Neonatal Diseases

[ ] Prospectively registered
[ ] Protocol
[ ] Statistical analysis plan
[ ] Results
[ ] Individual participant data
[ ] Record updated in last year

Plain English summary of protocol

Background and study aims:
Within the first few days of a newborn baby's life, a small blood sample is taken to test them for genetic health conditions. This is called newborn bloodpsot screening or NBS. The aim of NBS is to identify conditions early, so that prevention or treatment can start as soon as possible. However, new NBS technology can pick up cases where doctors aren't sure if the baby will develop a condition or not. One example of this is 'cystic fibrosis screen positive, inconclusive diagnosis' or CFSPID. Babies with CFSPID have a positive NBS result for cystic fibrosis, but usually do not have any symptoms and usually remain well. Currently, we do not know very much about how CFSPID affects families emotionally, although some research suggests that families do not cope well with the uncertainty of CFSPID. The aim of this study is to interview parents of children with CFSPID, to find out about their experiences. It is hoped that having more knowledge about this experience will help us to improve things for other families in the future.

Who can participate?
Parent(s) of children with CFSPID will be told about the study by their own clinic and can contact the researcher if they want to take part. Parents must be able to understand the study and be willing to talk about their experiences. Parents will not be interviewed if their child has passed away or is seriously ill.

What does the study involve?
The study will involve interviews with parents about their experiences. The interviewer will have a set of questions about their experiences, but the parents are in control of how the interview goes and what they choose to discuss. The interviews will be around 1-2 hours long depending on parents' responses. The interviews will be recorded (just the voices) and typed up, changing names and removing any information that could identify individuals. The study team will then permanently delete the voice recording and analyse the typed interviews for anything significant about CFSPID. A method called 'grounded theory' will be used with the aim to bring together the content from all the interviews to create a theory about how people experience

CFSPID. To test the theory as it develops, parents may be asked to take part in a second interview to provide their feedback on the theory so far. This interview will be run and analysed in the same way as the first interview. Parents do not have to take part in a second interview if they do not want to.

What are the possible benefits and risks of participating?
There are no specific benefits to participating although in our experience the study team have found that many people value the opportunity to talk about their experiences.

This study involves people talking about events and topics that they may find upsetting, and so there is a risk that they will become distressed. The interviews have been designed to minimise this risk and the interviewer is trained in how to interview people sensitively. The participants have full control over the interview and may choose to pause or end the interview at any time. The participants will be signposted to sources of support if they are distressed.

Where is the study run from?
The University of Manchester (UK)

When is the study starting and how long is it expected to run for?
September 2019 to May 2023

Who is funding the study?
The study is being carried out as part of the researcher's PhD, which is funded by a scholarship from The University of Manchester (UK)

Who is the main contact?
Faye Johnson, faye.johnson@manchester.ac.uk

Contact information

Type(s)
Scientific

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Additional identifiers

Clinical Trials Information System (CTIS)
Nil known

Integrated Research Application System (IRAS)
278871

Protocol serial number
IRAS 278871

Study information

Scientific Title
Exploring parents’ experiences of CFSPID designation: interview study

Study objectives
This is a qualitative study to explore parents' experiences of receiving a newborn genetic screening result of 'cystic fibrosis screen positive, inconclusive diagnosis' (CFSPID). Nascent work in this area suggests that uncertain screening results such as CFSPID can have a negative psychological impact on parents. The aim of this study is to explore this further, in order to improve the experience for parents of children with CFSPID, and more broadly to inform debates regarding expanded genomic screening.

Ethics approval required
Old ethics approval format

Ethics approval(s)
Pending, HRA and NHS REC

Study design
Qualitative interview study

Primary study design
Observational

Study type(s)
Other

Health condition(s) or problem(s) studied
Cystic fibrosis screen positive, inconclusive diagnosis (CFSPID)

Interventions
This qualitative study will use grounded theory methodology. Parents will be asked to take part in interviews to capture their experiences. The researcher will have a set of topics, questions, or tasks to help explore the area of interest, but the order and precise wording of questions will be determined by the interviewee’ responses. Interview questions are left as open as possible so participants may respond freely and authentically. The length of the interview will be determined by the length of the participants’ responses. As the study progresses and the grounded theory is developing, additional questions will be asked of participants to follow areas

of importance and test the emerging theory. Some participants may be contacted to request follow-up interviews to capture their views. This will be determined by 'theoretical sampling' – sampling driven by decisions to test the emerging theory. As the researcher begins to build the grounded theory, interviews will explore emerging ideas and test the emerging theory. Interviews will be carried out by in a mutually convenient location of the participant's choice (this is likely to be the participant's home if possible), or over video conferencing or phone. Interviews will be audio recorded for transcription and analysed using grounded theory methodology.

Intervention Type
Other

Primary outcome(s)
Parent experiences of their infant's 'cystic fibrosis screen positive, inconclusive diagnosis' (CFSPID) designation using audio-recorded and transcribed participant interview responses which will then be analysed for themes pertaining to the research question collected at a single timepoint

Key secondary outcome(s))
There are no secondary outcome measures

Completion date
31/05/2023

Eligibility

Key inclusion criteria
1. The parent or parents of an infant diagnosed with CFSPID following newborn bloodspot screening
2. Aged ≥18 years
3. Able to understand the purpose and implications of the research study
4. Willing and able to talk about their experiences with a researcher. Where participants are speakers of languages other than English, the research team will arrange a translator.

Participant type(s)
Carer

Healthy volunteers allowed
No

Age group
Adult

Lower age limit
18 years

Sex
All

Total final enrolment

Key exclusion criteria
1. Parent of a child who has died or has significant diagnosis of another disease or health issue
2. Barriers to fully understanding the research and communicating with the researcher

Date of first enrolment
01/04/2021

Date of final enrolment
31/05/2023

Locations

Countries of recruitment
United Kingdom

England

Study participating centre
The University of Manchester
Oxford Road
Manchester
United Kingdom
M13 9PL

Sponsor information

Organisation
University of Manchester

ROR
https://ror.org/027m9bs27

Funder(s)

Funder type
University/education

Funder Name
University of Manchester

Alternative Name(s)
University of Manchester in United Kingdom, University of Manchester UK, The University of Manchester, UoM

Funding Body Type
Government organisation

Funding Body Subtype
Universities (academic only)

Location
United Kingdom

Results and Publications

Individual participant data (IPD) sharing plan
The datasets generated during and/or analysed during the current study are not expected to be made available due to the sensitive nature of the raw data - transcripts of confidential interviews, discussing sensitive topics, containing potentially identifiable information. The subsequent results publication(s) will contain suitable excerpts of data in the form of anonymised quotations.

IPD sharing plan summary
Not expected to be made available