

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

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<b>Registration date</b> 19/04/2021	<b>Overall study status</b> Completed	<input type="checkbox"/> Statistical analysis plan <input type="checkbox"/> Results
<b>Last Edited</b> 09/06/2023	<b>Condition category</b> Neonatal Diseases	<input type="checkbox"/> Individual participant data <input type="checkbox"/> 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 bloodspot 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](mailto:faye.johnson@manchester.ac.uk)

## Contact information

### Type(s)

Scientific

### Contact name

Ms Faye Johnson

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# Additional identifiers

## EudraCT/CTIS number

Nil known

## IRAS number

278871

## ClinicalTrials.gov number

Nil known

## Secondary identifying numbers

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

## Secondary study design

Cross sectional study

## Study setting(s)

Other

## Study type(s)

Other

## Participant information sheet

Not available in web format, please use contact details to request a participant information sheet.

### **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's 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 measure**

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

### **Secondary outcome measures**

There are no secondary outcome measures

### **Overall study start date**

30/09/2019

### **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  $\geq 18$  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

**Age group**

Adult

**Lower age limit**

18 Years

**Sex**

Both

**Target number of participants**

20

**Total final enrolment**

15

**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**

England

United Kingdom

**Study participating centre**

**The University of Manchester**

Oxford Road

Manchester

United Kingdom

M13 9PL

**Sponsor information****Organisation**

University of Manchester

**Sponsor details**

Oxford Road  
Manchester  
England  
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M13 9PL  
+44 (0)1612755436  
FBMHethics@manchester.ac.uk

**Sponsor type**

University/education

**Website**

<https://www.manchester.ac.uk/>

**ROR**

<https://ror.org/027m9bs27>

## **Funder(s)**

**Funder type**

University/education

**Funder Name**

University of Manchester

**Alternative Name(s)**

The University of Manchester, University of Manchester UK, University of Manchester in United Kingdom, UoM

**Funding Body Type**

Government organisation

**Funding Body Subtype**

Universities (academic only)

**Location**

United Kingdom

## **Results and Publications**

Publication and dissemination plan

The full study report will be in the form of Faye Johnson's PhD thesis which will be accessible through The University of Manchester. We also plan to publish in a high-impact peer-reviewed journal. Due to the nature of qualitative research, raw data will not be available.

**Intention to publish date**

31/05/2024

**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