Integrating wider aspects of life with sickle cell disease into the trial protocol
Initial analysis from focus group and suggested next steps

Aim

The acute pain crisis (APC) trial under preparation by the pain group is aiming to compare approaches to managing APCs. This requires careful consideration of endpoints: is this through pain scores, time to treatment, time in hospital, or the use of quality of life instruments? Is it through economic analysis? All of these are potentially valuable, but they may miss **wider priorities that matter to people living with SCD**, which are potentially impacted by APC care.

Methods

Group composition

The focus group took place on 22nd November 2023. Participants were invited through their haemoglobinopathy co-ordinating centres (HCCs). The invitation is included as Appendix 1. It is worth noting that this group is composed of members who are already involved in their HCCs, and often have other advocacy roles. The focus group included five people living with SCD, two parents of children with SCD, and one advocacy group leader. One member was from Bristol and the rest were from London. The discussion was facilitated by a Consultant Psychologist with experience in focus group methodology (KA). Two organisers and one researcher (SH) were also present but did not actively participate, except one occasion when asked a direct question (SH).

Questions

This focus group aimed to explore elements of life beyond hospital care. The following prompts were used to lead discussion, but participants were free to discuss as other topics arose:

- How does education affect people with sickle cell disease and their health outcomes?
- What are the barriers to educational achievement for children and young people with sickle cell disease?
- How do work environment conditions affect people with sickle cell disease?
- How does unemployment and job insecurity affect people with sickle cell disease?
- How does housing influence outcomes of people with SCD?
- How does social inclusion and non-discrimination affect people with sickle cell?
- What are the challenges faced by people with sickle cell who are discriminated against in accessing healthcare services?

Analysis

The focus group discussion was audio recorded and computer aided transcription was used to produce a rough transcript (Whisper AI). The rough transcript was checked, corrected and formatted by SH.

SH undertook reflexive thematic analysis within an interpretivist framework.[1] I used an experiential orientation – meaning that I did not seek to critique or go beyond the

meanings offered up by participants. I adopted an inductive approach to coding, meaning that I devised a system of coding from the focus group data, rather than starting with a pre-defined set of codes. I developed these codes into themes based on significance and relevance to the focus group aim, rather than frequency. The results below reflect an initial analysis, which is yet to be completed.

Results

Discussion within the group illuminated the intersecting social, systemic, and structural barriers that negatively impact health for people living with sickle cell disease in the UK. Their stories provide critical context for improving care and designing inclusive clinical trials. My approach to analysis specifically focuses on aspects most relevant to trial design; there is more that could be learned from the discussion if other questions are brought to it. I outline some of these points for further exploration after the main themes.

Theme 1: Isolation, exclusion and missing out

Participants noted elements exclusion from different parts of life: sports at school, activities with friends, and a wider sense of missing out. Some of this was directly due to hospital visits, some was action aiming to avoid crises (e.g. avoiding swimming), but some exclusion was because of undue fear, borne of ignorance about the condition.

One mother of a person with SCD reported how school experiences intersected with Covid19 isolation:
"That reclusive side of him is still there…he hardly comes out of the house"

Suggested research endpoints: Involvement in activities and hobbies you enjoy; ability to join in activities with friends and loved ones.

Theme 2: Confidence in self management of sickle cell disease

Subtheme: Knowledge about rights and channels of support

"There is an opportunity for more education for people with sickle to learn how to cope with their condition"

Part of this also involved advocating for your own rights:
"it's important to really know your rights and have the right support, and have an [employing] organisation who respect that".

Suggested research endpoints: Self-confidence in managing condition, advocating for self and accessing help

Theme 3: Relationships with employers

A difficult decision for participants was whether to disclose your status as a person with SCD to employers.

One participant chose to do so: “I wasn’t afraid to let people know about my condition and what I needed when I needed”, but found it burdensome to not have this taken seriously “it’s like you get sick of having to explain yourself constantly.”

A different participant chose not to disclose, but this also led to problems. She spoke of the passive-aggressive comments that she got after returning from a work trip, in which she’d been asked to run up a mountain to collect other members of staff. “There’s a lot of trauma involved in being in that position and being on the spot in the moment and feeling brave enough and empowered enough and supported enough to say something to an employee who might be showing you passive aggression”. Eventually she lost her role at this company.

Suggested research endpoints: Relationship with employer or stability at work; confidence or enjoyment of work and education

Theme 4: Compounding trauma and intersecting disadvantage

Example excerpts:
"And then you obviously, when you experience more and more of these things, the trauma and the anxiety and the anger and the frustration and then if you take it on the next person, then they are like, well look at this angry black person.”

“If you're a sickle cell sufferer, especially one that has regular crises, how do you manage to stay in employment, have good finance, to then deal with everything from housing, bills, heating, transport, medication, because we don't get the free prescription, unless you've got another condition that's on the specific list... If you don't have parents, you don't have good siblings, or you don't have good friend network, you don't know what's in the community, you don't have good education, it starts to pile up all the negative aspects that are building up within your life and then it's like what route am I taking now?”

“The detrimental impact of unemployment, joy security has that cycle of stress causing a crisis, money worries causing a crisis, that impact that unemployment and job insecurity has on family and relationships”

Suggested research endpoints: Financial stability; relationships with loved ones; self-esteem

Points raised in discussion which are not expanded on here:

The impact of underinvestment in SCD research, care and education
Racism as an intersectional factor – e.g. you experience difficulties at work both because of stigma against SCD and anti-Black racism
Gratitude for healthcare provision in the UK
Disbelief of pain by teachers and healthcare professionals
Relationship with schools and lack of understanding by education professionals

How might this be taken forward?

If the trial planning group felt that some of factors were valuable for the trial, then they could be developed into potential measurable endpoints.
We could then hold a second online focus group (online to facilitate greater geographical diversity) and circulate the draft endpoints in advance. The second focus group could then help refine these. The key question for the focus group would be: "How can we make sure what we are measuring in the trial reflects what matters most to you?"

We would explain that we are not specifically setting out to address these issues in an intervention, but that they may well be markers for holistic health, or be influenced by the care intervention (e.g. patients may grow in knowledge of rights and support by being around other patients or specialist staff).

Reference

1. Byrne D. A worked example of Braun and Clarke's approach to reflexive thematic analysis. *Qual Quant*. 2022;56(3):1391-1412. doi:10.1007/s11135-021-01182-y

Appendix 1: Invitation to participants for focus group

NHS Health and Race Observatory Sickle Cell Project
Patient Focus Group
Social Determinants of Health, Equity and Diversity

Background
Social determinants of health are the conditions in which people are born, grow, live, work, and age. Examples include access to education, economic stability, social support, and the neighbourhoods where people live. They include where we are born, where we live and learn, where we work and play, and how we age. Understanding and dealing with these factors can make a big difference in the health and quality of life for people with sickle cell disease. It can also help make sure everyone has a fair chance to be healthy.

Objective
Sickle cell disease mostly affects people from minority ethnic communities in the UK. These are groups of people with different backgrounds, customs, and beliefs. Their views on chronic illness and what kinds of treatment they find acceptable are influenced by their cultures. Additionally, many people with sickle cell disease are in the lower economic groups of society. This means they might not have as much money or resources. It's clear that these social factors affect how sickle cell disease affects people. Our goal is to figure out how these factors might impact who joins, stays in, and does well in clinical trials.

Focus Group
You are invited to participate in a patient focus group, which will be held in parallel with the UK Forum on Haemoglobin Disorders Meeting.
Date: Wednesday 22 November 2023
Time: 1pm to 5pm
Venue: Cavendish Conference Centre
22 Duchess Mews
London
W1G 9DT

NHS Health and Race Observatory Sickle Cell Project Lead Investigators:
Prof Paul Telfer, Dr Kofi Anie, Dr Sanne Lugthart, Carol Burt (PPV Representative), Dr Stuart-Smith, Dr Kotsiopoulou