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Rare but heard: Using Asynchronous Virtual Focus Groups, Interviews and Roundtable discussions to create a Personalised Psychological Intervention for Primary Sclerosing Cholangitis – a protocol

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Complete List of Authors:	Ranieri, Veronica; Tavistock and Portman NHS Foundation Trust, Research & Development; University College London, Science and Technology Studies Kennedy, Eilis; Tavistock and Portman NHS Foundation Trust, Research & Development Walmsley, Martine; PSC Support Thorburn, Doug; Royal Free London NHS Trust, Department of Hepatology McKay, Kathy; Tavistock and Portman NHS Foundation Trust, Research & Development
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3 Rare but heard: Using Asynchronous Virtual Focus Groups, Interviews and
4 Roundtable discussions to create a Personalised Psychological Intervention for
5 Primary Sclerosing Cholangitis – a protocol
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11 V. Ranieri PhD MSc MA BA,^{1,2} E. Kennedy MB BCh BAO MRCPsych^{1,3}, M. Walmsley
12 BSc,⁴ D. Thorburn MB ChB MD FRCP,⁵ 6 K. McKay PhD BA LLB,^{1,6}
13
14
15
16
17

18
19 ¹Research & Development Unit, Tavistock Centre, Tavistock & Portman NHS Foundation
20 Trust, 120 Belsize Lane, NW3 5BA, United Kingdom
21
22

23 ²Department of Science & Technology Studies, University College London, Gower Street,
24 London WC1E 6BT, United Kingdom
25
26

27 ³Research Department of Clinical, Educational & Health Psychology, University College
28 London, Gower Street, London WC1E 6BT, United Kingdom
29
30

31 ⁴PSC Support, 3 Gary O'Donnell Drive, Didcot, OX11 6BT, United Kingdom
32
33

34 ⁵Sheila Sherlock Liver Centre & UCL Institute of Liver and Digestive Health, Royal Free
35 Hospital, Pond Street, London NW3 2QG, United Kingdom
36
37

38 ⁶Institute of Psychology, Health and Society, Waterhouse Building, Block B, Brownlow
39 Street, University of Liverpool, Liverpool L69 3GL, United Kingdom
40
41
42
43
44
45

46 Address for correspondence: Veronica Ranieri PhD, Research & Development Unit,
47 Tavistock Centre, Tavistock & Portman NHS Foundation Trust, 120 Belsize Lane, NW3
48 5AS, United Kingdom (e-mail: vranieri@tavi-port.nhs.uk).
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ABSTRACT

Introduction Primary Sclerosing Cholangitis (PSC) is a rare and chronic disease characterised by inflammation and fibrosis of the liver's bile ducts. There is no known cause or cure for the illness, which often progresses to end-stage liver disease requiring liver transplantation. Symptoms of PSC can be very burdensome on those living with the illness, leading to restrictions in daily living, as well as a greater risk of colorectal and biliary tract cancers. Limited voices from lived experience suggest that living with PSC can cause considerable psychological distress. This study, therefore, aims to explore how the illness impacts the psychological wellbeing of those living with the illness, and those supporting them. It also aims to create a personalised psychological intervention to support all groups.

Methods and analysis This project will take a layered qualitative approach to understanding the ways in which people experience living with PSC within their day-to-day lives. There will be two stages to this study, which will pilot a unique methodological process using online resources. The first stage will consist of Asynchronous Virtual Focus Groups (AVFGs) with those living with PSC and those who provide support for those diagnosed with PSC, and narrative interviews with both groups and health professionals. The second stage will comprise a roundtable discussion where the researchers and health professionals will devise a personalised psychological intervention to help support those living with PSC and their supporters. The study duration is expected to be 18 months.

Ethics and dissemination The proposed study has been approved by the UK Health Research Authority and London – Queen Square Research Ethics Committee as application 18/LO/1075. Results from the AVFGs and narrative interviews will be submitted for peer-reviewed publication. The findings of the study will also be presented nationally to PSC and medical communities, and a summary of the findings will be shared with participants.

ARTICLE SUMMARY

Strengths and limitations:

- This study will employ innovative qualitative research methods, such as the use of asynchronous virtual focus groups (AVFGs), to capture the lived experiences of those with a rare illness spread over a large geographic space who would not be able to participate in a study like this otherwise.
- A triangulated perspective will be used to explore the experiences of those living with PSC, caregivers and health professionals, with a view to improving the support provided to all.
- A personalised psychological intervention, that can be tailored to fit individuals' personal circumstances, will be created in response to analysing these experiences.
- However, involvement in AVFGs is dependent on participants' literacy, computer-literacy and access to technology.
- Although online qualitative approaches can target difficult-to-access populations, they cannot replace face-to-face human contact and interaction.

INTRODUCTION

Primary Sclerosing Cholangitis (PSC) is a rare and chronic disease characterised by the narrowing and blockage of the liver's bile ducts from inflammation and fibrosis.¹ Recent estimates suggest that the incidence of PSC is 0.68 per 100,000 person-years.² It disproportionately affects men, and symptoms include jaundice, fatigue, and itching, among others.^{1,3} However, these symptoms vary: some people are asymptomatic when first diagnosed, whereas others have a significant symptom burden.⁴ In addition to this variation in symptoms, the speed and aggressiveness with which the disease can progress may differ between individuals, with little indication from the literature as to why.¹ Furthermore, there is no curative treatment for PSC, with liver transplant offered as an option for those reaching end-stage liver disease, with those living with the illness displaying a greater incidence of colorectal and biliary tract cancers in addition to other complications.^{5,6}

In the absence of curative treatment, recent research has begun to examine the impact of living with PSC on individuals' psychological wellbeing.⁷ Similar to other long-term physical illnesses, individuals living with PSC tend to report a greater number of depressive symptoms, and a greater level of impairments pertaining to quality of life and wellbeing compared to the general population.⁷⁻⁹ Voices from lived experience reveal that the uncertainty with which the disease may progress to an eventual need for a liver transplant, and whether and when that transplant may be possible, can lead to considerable distress and anxiety regarding the future.^{10,11} In addition to such distress, individuals with PSC also report feeling socially isolated due to the rarity of the illness.^{8,12} Consequentially, such difficulties emphasise a need for access to emotional support that addresses this population's unmet needs and reinstalls hope in the future.¹⁰

A need for greater access to support is likely also felt by those supporting individuals living with PSC, whether informally or formally.⁷ Partners and families supporting those living with PSC may need to adjust and prepare for caring for a serious illness. Due to the rarity of the illness, few healthcare professionals will have experience of how PSC can present and progress, and so treating the presenting individual may result in unexpected challenges for them, especially in finding appropriate and effective pathways of care for their patient.¹¹ In light of these difficulties, the proposed study will aim to understand and chronicle the impact of PSC on the psychological wellbeing of those diagnosed with it and their caregivers, and on

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3 healthcare professionals' ability to support both. The study will seek to narrate the experiences
4 of various people impacted by a PSC diagnosis, with a view to creating a personalised
5 psychological intervention for all.
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10 *Research questions*

- 13 1. What are the experiences of people living with PSC in terms of their mental health and
14 emotional wellbeing?
- 15 2. What are the experiences of partners and family members of people living with PSC in
16 terms of their mental health and wellbeing?
- 17 3. What are the experiences of healthcare professionals in working with people living with
18 PSC and finding appropriate and effective pathways of care?
- 19 4. What would a framework of 'Personalised Psychological Intervention for PSC' for
20 people with PSC and their partners and families look like?
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29 *Objectives*

- 30 1. To explore the experiences of people living with PSC in terms of their mental health
31 and emotional wellbeing.
- 32 2. To explore the experiences of family members and partners of people living with PSC
33 in terms of their mental health and emotional wellbeing.
- 34 3. To explore the experiences of health professionals working with people living with PSC
35 and finding appropriate and effective pathways of care.
- 36 4. To develop a framework of a 'Personalised Psychological Intervention for PSC'.
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48 **METHODS and ANALYSIS**

49 *Procedure*

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51 This project will take a layered qualitative approach to understanding the ways in which people
52 experience living with PSC within their day-to-day lives. There will be two stages to this study,
53 which will also pilot a unique methodological process using online resources. The study
54 duration is expected to be 18 months.
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3 **Stage One** answers Research Questions 1, 2, and 3 and will comprise of:
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7 1. Asynchronous virtual focus groups (AVFGs)(Research Questions 1 and 2)

8 AVFGs are focus groups held online over a time period that is longer than a usual focus
9 group, usually one question per week over a period of several weeks. Similar to a
10 traditional focus group, they still have set time limits and the ability for participants to
11 engage with each other. They have been previously used successfully with vulnerable
12 and diverse populations.¹³ As PSC is so rare, conducting face-to-face research is
13 difficult, so AVFGs are a good way of accessing as wide a group as possible.¹⁴
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20 Separate AVFGs will be conducted with both people living with PSC and family
21 members to allow participants to be as open and honest in their responses as they wish.
22 AVFGs will be limited to 6-9 participants per group. Due to their online nature, more
23 than one AVFG will be held at the same time. Four questions will be asked over four
24 weeks to guide the discussion. Within the AVFG, one question will be posed per week
25 and the participants can post as many responses as they wish each week; both in
26 response to the question posed and what other responders have said. After a week, the
27 previous question will be closed and the next one will be opened; this will be clearly
28 stated. In this way, participants can respond when it suits them, with the responses being
29 as long or as short as they feel comfortable with. The AVFGs will be moderated twice
30 daily by researchers KM and VR to ensure participants' wellbeing, and every
31 participant will have access to clear contact details for the research team and helplines
32 (ie. Samaritans) should a participant become upset. All questions will focus on
33 understanding the mental health and wellbeing (including feelings of hopelessness,
34 anxiety, powerlessness, and resilience) of people with the disease and their partners and
35 family members, and how these feelings have changed since diagnosis and potential
36 changes in severity of symptoms.
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51 2. Narrative interviews (telephone/online/face-to-face - more in-depth experience)
52 (answers Research Questions 1-3)
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56 Narrative interviews are crucial to unpacking the themes identified from the AVFGs to
57 inform the development of a personalised psychological intervention. Narrative
58 interviews will be conducted with:
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5 a) A purposive sample of participants living with PSC and family members who
6 participated in the AVFGs, and

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8 b) Healthcare professionals recruited either via PSC support or from asking those living
9 with PSC who took part in AVFGs if they are willing to provide the name and contact
10 details of their healthcare practitioner. As PSC is uncommon, it is expected that not
11 every healthcare practitioner will be experienced in treating it, which can lead to
12 feelings of stress when trying to determine the best pathway of care for their patient.
13 Healthcare professionals will not have access to any data disclosed during interviews
14 and focus groups with participants who have PSC, and vice versa.
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22 **Stage Two** answers Research Question 4, and is based on the findings from Stage One.
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25 Practitioners from hepatology, gastroenterology and mental health will be invited to a
26 presentation of the themes and narratives uncovered from the data in Stage One. Following this
27 presentation, they will be asked to partake in an audio-recorded roundtable discussion on the
28 findings of the focus groups and interviews. From these findings, they will be asked to help co-
29 construct a Personalised Psychological Intervention. After the completion of this roundtable,
30 the researchers will write up the conclusions of the discussion and will send these to the
31 practitioners for confirmation.
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39 *Sample*

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41 Participants will consist of those with lived experience of PSC, family members/partners/carers
42 of people living with PSC, and health professionals who have experience working with people
43 living with PSC. This study aims to empower those living with PSC and their loved ones and
44 those who care for them to make their voices heard. As such, we are keeping our inclusion
45 criteria general so as to include as many people who would like to participate as possible.
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50 Participants must:

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- 53 • Be aged 18 years or more
 - 54 • Be UK residents
 - 55 • Self-identify as someone living with PSC **or** a partner, family member or carer of a
56 person living with PSC **or** a health professional working with people living with PSC
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5 Due to the rarity of this disease and the lack of extant research exploring experiences of PSC,
6 it is difficult to specify the size of our sample. The study will aim to include a range of 37-62
7 participants overall. This range acknowledges that recruitment of individuals with PSC may be
8 challenging given the rarity of the illness, but also ensures enough participation for data
9 saturation. For the AVFGs we aim to run 2 – 4 focus groups consisting of 6 – 9 participants in
10 each. This will include: 1 – 2 focus groups of people living with PSC and 1 – 2 focus groups
11 of partners and family members of people living with PSC. From these, we plan on purposively
12 sampling up to ten individuals to take part in narrative interviews. Additionally, we aim to
13 interview 5 – 10 health professionals.
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22 *Setting*

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24 Participants will be recruited using the PSC Support network. PSC Support is a charity that
25 plays a key role in connecting people living with PSC to each other and wider PSC
26 communities, as well as providing access to support and information. PSC Support will
27 promote the study, as well as guide us on the appropriateness and relevance of the study design.
28 The study will be advertised via PSC Support's media presence (ie. Facebook and Twitter). All
29 interested people can contact the researchers (KM and VR) directly to get more information
30 about the study. All study specific activities (e.g. recruitment etc.) will be undertaken by the
31 researchers who are based in the Tavistock and Portman NHS Foundation Trust. As PSC is a
32 rare disease, participants will be recruited from across the United Kingdom to allow greater
33 participation.
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43 Asynchronous Virtual Focus Groups (AVFGs) will be hosted on an existing Virtual Learning
44 Environment (VLE) (ie. Moodle or Blackboard).^{15,16} VLEs are suitable for collecting this type
45 of data as they enable researchers to host focus groups on their own custom-made site which
46 is secure, confidential, and a safe environment for research participants.¹⁷ For example, this
47 allows the researchers to create ground rules for discussion which detail what style, tone, and
48 content of messages would be appropriate for discussions to ensure the well-being of all
49 participants.¹⁷ AVFGs also allow the researchers to access participants regardless of location,
50 which is likely to be an obstacle with rare diseases such as PSC. When the AVFGs are ongoing,
51 the researchers will check the content twice daily to ensure safety for all parties involved.
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3 All interviews are expected to last a maximum of one hour. Many healthcare professionals face
4 considerable time constraints in their workplace and so the interview will be constructed to
5 take as little time as possible. Participation is entirely voluntary. Given the potential distance
6 of participants, these will be conducted over the phone or via Telemedicine, depending upon
7 the person's preference. The opportunity to talk face-to-face or via email will be offered to
8 local participants should they prefer to do so. Participants will be asked to give verbal audio
9 recorded consent and consent to their interview being audio-recorded. The questions within the
10 interview will focus on similar themes as the AVFGs, but the narrative interview format will
11 allow for a deeper exploration of people's experiences. Should any participant become
12 distressed during the interview, they will be asked if they would like to discontinue the
13 interview or the recording. At the conclusion of each interview, the researcher will debrief with
14 the participant to check how they are feeling and offer contact details for helplines if needed.
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25 *Analysis*

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29 The data collected from Stage One will be transcribed, coded and de-identified, and then
30 analysed thematically and narratively using NVivo as the data management system.¹⁸ The first
31 researcher will go through the transcripts from the AVFGs and the different interviews and
32 compare themes and narratives between and within the three different groups (people with
33 PSC, partners and family members of people with PSC, healthcare professionals). Themes and
34 narratives will be double-checked by a second researcher, and any disagreements discussed
35 and resolved.
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43 Findings will then be taken to the PSC Support Study Steering Committee to discuss in terms
44 of what has been found, and whether anything has been missed by the researchers and would
45 be an important inclusion in terms of creating the Personalised Psychological Intervention
46 framework. Following this, the group of gastroenterology, hepatology and mental health
47 professionals will then be invited to attend a face-to-face roundtable meeting to discuss ideas
48 and issues that could ground a 'Personalised Psychological Intervention for PSC' framework
49 of recommendations, in conjunction with those with lived experience. This innovative
50 framework of recommendations could help tailor interventions to not only the individuals
51 diagnosed with PSC, but also their partners and families, to ensure they received the best
52 support for their physical and mental health. Further, although the details would be personalised
53 to each individual depending on their symptoms and severity, the framework of these
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3 interventions could then be shared to the wider healthcare professional community so they
4 would be better prepared to support a patient diagnosed with PSC. Again, this framework will
5 then be shared with the PSC Support steering group to ensure that it is pragmatic and practical
6 in terms of what people with PSC need in terms of care and support in their everyday lives.
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10 11 12 **ETHICS and DISSEMINATION**

13 14 15 *Ethical Approval*

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17 This study was approved by the Health Research Authority and received a Favourable Opinion
18 from London - Queen Square NHS Research Ethics Committee under application number
19 18/LO/1075.
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23 24 25 *Informed consent*

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27 Informed consent will be obtained for each participant. All participants will be provided with
28 an information sheet indicating the purpose of the study and will have an opportunity to ask
29 questions. Consent for participants partaking in the AVFGs will be recorded online. Formal
30 consent will also be recorded for all those taking part in narrative interviews. Should interviews
31 be carried out over telephone or via Telemedicine, participants will be asked to give verbal
32 consent which will be audio-recorded. Signed and written consent will be sought from those
33 taking part in face-to-face or email interviews. Consent from all members taking part in a
34 roundtable discussion will be audio-recorded.
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41 42 43 *Data protection and patient confidentiality*

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45 Information provided in confidence will be anonymised and not be used or disclosed in a form
46 that might identify the participant without his or her express written consent. Participants will be
47 given the opportunity to choose their own pseudonym and these will be held in password protected
48 databases on an encrypted NHS server at the Tavistock and Portman NHS Foundation Trust. Only
49 the Chief Investigator and researchers will have access to it, and only anonymised data will be
50 shared with anyone else. Only participants' unique pseudonyms will be linked to the raw data
51 collected (e.g. audio recordings). Audio files will be identified by pseudocodes, however it is
52 possible that names of people and/or places may be mentioned during the interviews. All digital
53 data files (including audio files) will be assigned a pseudocode and uploaded onto a secure NHS
54 server as soon as possible after data is collected, and the files deleted from the memory disk or
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3 hard drive. Memory disks and hard drives containing data will be held in locked cabinets if it is
4 not possible to upload the files immediately following data collection. Data will be uploaded onto
5 the secure server, only accessible by authorised researchers, and will be transferred to a trusted
6 transcription company using secure data transfer methods. All members of the research team will
7 be reminded of their duty to observe the NHS code of ethics regarding patient confidentiality.
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10 11 12 13 *Dissemination*

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17 The findings of the study will be submitted for publication in peer-reviewed journals, and will be
18 presented nationally. All intellectual property rights relating to the data arising from the study shall
19 be held by the Tavistock & Portman NHS Foundation Trust. Participants who consent to receive
20 the study findings will be sent an electronic or hard copy interim report and a final lay summary
21 of the findings, depending on their preferred method of communication. They will also be
22 provided with the details of where to access the online publication of the full study report.
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29 **Authors' contributions:** EK and KM constructed the protocol. KM and VR drafted this protocol
30 article. It was subsequently reviewed by all authors (EK, DT, MW and RS).
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34 **Funding statement:** This work was supported by PSC Support grant number 220318VR.
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38 **Competing interests statement:** MW is the Chair of Trustees at PSC Support and an individual
39 living with PSC. DT is a member of the PSC Support Expert Panel. No other authors have
40 competing interests.
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44 **Patient and Public Involvement:** The study was co-designed with PPI involvement from PSC
45 Support, and will be overseen by a steering committee composed of individuals living with PSC
46 or caring for relatives living with PSC. The study will be advertised by PSC Support and its
47 findings will be evaluated and disseminated by members of PSC Support.
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53 **Word Count:** 3,873 words (including references)
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Complete List of Authors:	Ranieri, Veronica; Tavistock and Portman NHS Foundation Trust, Research & Development; University College London, Science and Technology Studies Kennedy, Eilis; Tavistock and Portman NHS Foundation Trust, Research & Development Walmsley, Martine; PSC Support Thorburn, Doug; Royal Free London NHS Trust, Department of Hepatology McKay, Kathy; Tavistock and Portman NHS Foundation Trust, Research & Development
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11 V. Ranieri PhD MSc MA BA,^{1,2} E. Kennedy MB BCh BAO MRCPsych^{1,3}, M. Walmsley
12 BSc,⁴ D. Thorburn MB ChB MD FRCP,⁵ 6 K. McKay PhD BA LLB,^{1,6}
13
14
15
16
17

18 ¹Research & Development Unit, Tavistock Centre, Tavistock & Portman NHS Foundation
19 Trust, 120 Belsize Lane, NW3 5BA, United Kingdom
20
21
22

23 ²Department of Science & Technology Studies, University College London, Gower Street,
24 London WC1E 6BT, United Kingdom
25
26

27 ³Research Department of Clinical, Educational & Health Psychology, University College
28 London, Gower Street, London WC1E 6BT, United Kingdom
29
30
31

32 ⁴PSC Support, 3 Gary O'Donnell Drive, Didcot, OX11 6BT, United Kingdom
33
34

35 ⁵Sheila Sherlock Liver Centre & UCL Institute of Liver and Digestive Health, Royal Free
36 Hospital, Pond Street, London NW3 2QG, United Kingdom
37
38

39 ⁶Institute of Psychology, Health and Society, Waterhouse Building, Block B, Brownlow
40 Street, University of Liverpool, Liverpool L69 3GL, United Kingdom
41
42
43
44
45

46 Address for correspondence: Veronica Ranieri PhD, Research & Development Unit,
47 Tavistock Centre, Tavistock & Portman NHS Foundation Trust, 120 Belsize Lane, NW3
48 5AS, United Kingdom (e-mail: v.ranieri@ucl.ac.uk)
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ABSTRACT

Introduction Primary Sclerosing Cholangitis (PSC) is a rare and chronic disease characterised by inflammation and fibrosis of the liver's bile ducts. There is no known cause or cure for the illness, which often progresses to end-stage liver disease requiring liver transplantation. Symptoms of PSC can be very burdensome on those living with the illness, leading to restrictions in daily living, as well as a greater risk of colorectal and biliary tract cancers. Limited voices from lived experience suggest that living with PSC can cause considerable psychological distress. This study, therefore, aims to explore how the illness impacts the psychological wellbeing of those living with the illness, and those supporting them. It also aims to create a personalised psychological intervention to support all groups.

Methods and analysis This project will take a layered qualitative approach to understanding the ways in which people experience living with PSC within their day-to-day lives. There will be two stages to this study, which will pilot a unique methodological process using online resources. The first stage will consist of Asynchronous Virtual Focus Groups (AVFGs) with those living with PSC and those who provide support for those diagnosed with PSC, and narrative interviews with both groups and health professionals. Both the AVFGs and the narrative interviews will be analysed using thematic narrative analysis. The second stage will comprise a roundtable discussion where the researchers and health professionals will devise a personalised psychological intervention to help support those living with PSC and their supporters. The study duration is expected to be 18 months.

Ethics and dissemination The proposed study has been approved by the UK Health Research Authority and London – Queen Square Research Ethics Committee as application 18/LO/1075. Results from the AVFGs and narrative interviews will be submitted for peer-reviewed publication. The findings of the study will also be presented nationally to PSC and medical communities, and a summary of the findings will be shared with participants.

ARTICLE SUMMARY

Strengths and limitations:

- This study will employ innovative qualitative research methods, such as the use of asynchronous virtual focus groups (AVFGs), to capture the lived experiences of those with a rare illness spread over a large geographic space who would not be able to participate in a study like this otherwise.
- A triangulated perspective will be used to explore the experiences of those living with PSC, caregivers and health professionals, with a view to improving the support provided to all.
- A personalised psychological intervention, that can be tailored to fit individuals' personal circumstances, will be created in response to analysing these experiences.
- However, involvement in AVFGs is dependent on participants' literacy, computer-literacy and access to technology.
- Although online qualitative approaches can target difficult-to-access populations, they cannot replace face-to-face human contact and interaction.

INTRODUCTION

Primary Sclerosing Cholangitis (PSC) is a rare and chronic disease characterised by the narrowing and blockage of the liver's bile ducts from inflammation and fibrosis.¹ Recent estimates suggest that the incidence of PSC is 0.68 per 100,000 person-years.² It disproportionately affects men, and symptoms include jaundice, fatigue, and itching, among others.^{1,3} However, these symptoms vary: some people are asymptomatic when first diagnosed, whereas others have a significant symptom burden.⁴ In addition to this variation in symptoms, the speed and aggressiveness with which the disease can progress may differ between individuals, with little indication from the literature as to why.¹ Furthermore, there is no curative treatment for PSC, with liver transplant offered as an option for those reaching end-stage liver disease, with those living with the illness displaying a greater incidence of colorectal and biliary tract cancers in addition to other complications.^{5,6}

In the absence of curative treatment, recent research has begun to examine the impact of living with PSC on individuals' psychological wellbeing.⁷ Similar to other long-term physical illnesses, individuals living with PSC tend to report a greater number of depressive symptoms, and a greater level of impairments pertaining to quality of life and wellbeing compared to the general population.⁷⁻⁹ Voices from lived experience reveal that the uncertainty with which the disease may progress to an eventual need for a liver transplant, and whether and when that transplant may be possible, can lead to considerable distress and anxiety regarding the future.^{10,11} In addition to such distress, individuals with PSC also report feeling socially isolated due to the rarity of the illness.^{8,12} Consequentially, such difficulties emphasise a need for access to emotional support that addresses this population's unmet needs and reinstalls hope in the future.¹⁰

A need for greater access to support is likely also felt by those supporting individuals living with PSC, whether informally or formally.⁷ Partners and families supporting those living with PSC may need to adjust and prepare for caring for a serious illness. Due to the rarity of the illness, few healthcare professionals will have experience of how PSC can present and progress, and so treating the presenting individual may result in unexpected challenges for them, especially in finding appropriate and effective pathways of care for their patient.¹¹ In light of these difficulties, the proposed study will aim to understand and chronicle the impact of PSC on the psychological wellbeing of those diagnosed with it and their caregivers, and on

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3 healthcare professionals' ability to support both. The study will seek to narrate the experiences
4 of various people impacted by a PSC diagnosis, with a view to creating a personalised
5 psychological intervention for all.
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10 *Research questions*

- 13 1. What are the experiences of people living with PSC in terms of their mental health and
14 emotional wellbeing?
- 15 2. What are the experiences of partners and family members of people living with PSC in
16 terms of their mental health and wellbeing?
- 17 3. What are the experiences of healthcare professionals in working with people living with
18 PSC and finding appropriate and effective pathways of care?
- 19 4. What would a framework of 'Personalised Psychological Intervention for PSC' for
20 people with PSC and their partners and families look like?
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29 *Objectives*

- 30 1. To explore the experiences of people living with PSC in terms of their mental health
31 and emotional wellbeing.
- 32 2. To explore the experiences of family members and partners of people living with PSC
33 in terms of their mental health and emotional wellbeing.
- 34 3. To explore the experiences of health professionals working with people living with PSC
35 and finding appropriate and effective pathways of care.
- 36 4. To develop a framework of a 'Personalised Psychological Intervention for PSC'.
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48 **METHODS and ANALYSIS**

49 *Procedure*

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52 This project will take a layered qualitative approach to understanding the ways in which people
53 experience living with PSC within their day-to-day lives. There will be two stages to this study,
54 which will also pilot a unique methodological process using online resources. The study
55 duration is expected to be 18 months.
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3 **Stage One** answers Research Questions 1, 2, and 3 and will comprise of:
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7 1. Asynchronous virtual focus groups (AVFGs)(Research Questions 1 and 2)

8 AVFGs are focus groups held online over a time period that is longer than a usual focus
9 group, usually one question per week over a period of several weeks. Similar to a
10 traditional focus group, they still have set time limits and the ability for participants to
11 engage with each other. They have been previously used successfully with vulnerable
12 and diverse populations.¹³ As PSC is so rare, conducting face-to-face research is
13 difficult, so AVFGs are a good way of accessing as wide a group as possible.¹⁴
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20 Separate AVFGs will be conducted with both people living with PSC and family
21 members to allow participants to be as open and honest in their responses as they wish.
22 AVFGs will be held on a virtual learning environment (VLE) platform and will be
23 limited to 6-9 participants per group. Due to their online nature, more than one AVFG
24 will be held at the same time. Participants will be asked to answer a different question
25 each week for four weeks, relating to their experiences of receiving a diagnosis of PSC,
26 their experiences with healthcare professionals, what helps or hinders their wellbeing,
27 and what advice they would share with someone recently diagnosed with PSC. one
28 question will be posed per week and the participants can post as many responses as they
29 wish each week; both in response to the question posed and what other responders have
30 said. After a week, the previous question will be closed and the next one will be opened;
31 this will be clearly stated. In this way, participants can respond when it suits them, with
32 the responses being as long or as short as they feel comfortable with. The AVFGs will
33 be moderated twice daily by researchers KM and VR to check the content of what is
34 posted by participants. Participants will be asked to follow a series of common
35 'netiquette' guidelines, for instance asking participants to respect other participants'
36 points of view and refraining from using all caps. Should the moderators see evidence
37 of severe psychological distress, researchers KM and VR will make direct contact with
38 participants and redirect them towards the appropriate support. Every participant will
39 also have online access to clear contact details for the research team and helplines (ie.
40 Samaritans) for the duration of the study.
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58 2. Narrative interviews (telephone/online/face-to-face - more in-depth experience)
59 (answers Research Questions 1-3)
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5 Narrative interviews are crucial to unpacking the themes identified from the AVFGs to
6 inform the development of a personalised psychological intervention. Narrative
7 interviews will be conducted with:
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12 a) A purposive sample of participants living with PSC and family members who
13 participated in the AVFGs, and
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15 b) Healthcare professionals recruited either via PSC support or from asking those living
16 with PSC who took part in AVFGs if they are willing to provide the name and contact
17 details of their healthcare practitioner. As PSC is uncommon, it is expected that not
18 every healthcare practitioner will be experienced in treating it, which can lead to
19 feelings of stress when trying to determine the best pathway of care for their patient.
20 Healthcare professionals will not have access to any data disclosed during interviews
21 and focus groups with participants who have PSC, and vice versa.
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29 **Stage Two** answers Research Question 4, and is based on the findings from Stage One.
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32 Practitioners from hepatology, gastroenterology and mental health will be invited to a
33 presentation of the themes and narratives uncovered from the data in Stage One. Following this
34 presentation, they will be asked to partake in an audio-recorded roundtable discussion on the
35 findings of the focus groups and interviews. From these findings, they will be asked to help
36 co-construct a Personalised Psychological Intervention. After the completion of this roundtable,
37 the researchers will write up the conclusions of the discussion and will send these to the
38 practitioners for confirmation.
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46 *Sample*

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48 Participants will consist of those with lived experience of PSC, family members/partners/carers
49 of people living with PSC, and health professionals who have experience working with people
50 living with PSC. This study aims to empower those living with PSC and their loved ones and
51 those who care for them to make their voices heard. As such, we are keeping our inclusion
52 criteria general so as to include as many people who would like to participate as possible.
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57 Participants must:

- 58 • Be aged 18 years or more

- Be UK residents
- Self-identify as someone living with PSC **or** a partner, family member or carer of a person living with PSC **or** a health professional working with people living with PSC

Due to the rarity of this disease and the lack of extant research exploring experiences of PSC, it is difficult to specify the size of our sample. The study will aim to include a range of 37-62 participants overall. This range acknowledges that recruitment of individuals with PSC may be challenging given the rarity of the illness, but also ensures enough participation for data saturation. For the AVFGs we aim to run 2 – 4 focus groups consisting of 6 – 9 participants in each. This will include: 1 – 2 focus groups of people living with PSC and 1 – 2 focus groups of partners and family members of people living with PSC. From these, we plan on purposively sampling up to ten individuals to take part in narrative interviews. Additionally, we aim to interview 5 – 10 health professionals.

Setting

Participants will be recruited using the PSC Support network. PSC Support is a charity that plays a key role in connecting people living with PSC to each other and wider PSC communities, as well as providing access to support and information. PSC Support will promote the study, as well as guide us on the appropriateness and relevance of the study design. The study will be advertised via PSC Support's media presence (ie. Facebook and Twitter). All interested people can contact the researchers (KM and VR) directly to get more information about the study. All study specific activities (e.g. recruitment etc.) will be undertaken by the researchers who are based in the Tavistock and Portman NHS Foundation Trust. As PSC is a rare disease, participants will be recruited from across the United Kingdom to allow greater participation.

Asynchronous Virtual Focus Groups (AVFGs) will be hosted on an existing Virtual Learning Environment (VLE) (ie. Moodle or Blackboard).^{15,16} VLEs are suitable for collecting this type of data as they enable researchers to host focus groups on their own custom-made site which is secure, confidential, and a safe environment for research participants.¹⁷ For example, this allows the researchers to create ground rules for discussion which detail what style, tone, and content of messages would be appropriate for discussions to ensure the well-being of all participants.¹⁷ AVFGs also allow the researchers to access participants regardless of location,

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3 which is likely to be an obstacle with rare diseases such as PSC. When the AVFGs are ongoing,
4 the researchers will check the content twice daily to ensure safety for all parties involved.
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8 All interviews are expected to last a maximum of one hour. Many healthcare professionals face
9 considerable time constraints in their workplace and so the interview will be constructed to
10 take as little time as possible. Participation is entirely voluntary. Given the potential distance
11 of participants, these will be conducted over the phone or via Telemedicine, depending upon
12 the person's preference. The opportunity to talk face-to-face or via email will be offered to
13 local participants should they prefer to do so. Participants will be asked to give verbal audio
14 recorded consent and consent to their interview being audio-recorded. The questions within the
15 interview will focus on similar themes as the AVFGs, but the narrative interview format will
16 allow for a deeper exploration of people's experiences. Should any participant become
17 distressed during the interview, they will be asked if they would like to discontinue the
18 interview or the recording. At the conclusion of each interview, the researcher will debrief with
19 the participant to check how they are feeling and offer contact details for helplines if needed.
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30 *Analysis*

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33 The data collected from Stage One will be transcribed, coded and de-identified, and then
34 analysed thematically and narratively using NVivo as the data management system.¹⁸ The first
35 researcher (VR) will go through the transcripts from the AVFGs and the different interviews
36 and analyse these using thematic narrative analysis. This will involve comparing themes and
37 narratives both chronologically and between the three different groups (people with PSC,
38 partners and family members of people with PSC, healthcare professionals). Themes and
39 narratives will be double-checked by a second researcher (KM), and any disagreements
40 discussed and resolved between the two researchers.
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50 Findings will then be taken to the PSC Support Study Steering Committee to discuss in terms
51 of what has been found, and whether anything has been missed by the researchers and would
52 be an important inclusion in terms of creating the Personalised Psychological Intervention
53 framework. Following this, the group of gastroenterology, hepatology and mental health
54 professionals will then be invited to attend a face-to-face roundtable meeting to discuss ideas
55 and issues that could ground a 'Personalised Psychological Intervention for PSC' framework
56 of recommendations, in conjunction with those with lived experience. This innovative
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3 framework of recommendations could help tailor interventions to not only the individuals
4 diagnosed with PSC, but also their partners and families, to ensure they received the best
5 support for their physical and mental health. Further, although the details would be personalised
6 to each individual depending on their symptoms and severity, the framework of these
7 interventions could then be shared to the wider healthcare professional community so they
8 would be better prepared to support a patient diagnosed with PSC. Again, this framework will
9 then be shared with the PSC Support steering group to ensure that it is pragmatic and practical
10 in terms of what people with PSC need in terms of care and support in their everyday lives.
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18 *Patient and Public Involvement*

19 The study was co-designed with PPI involvement from PSC Support, and will be overseen by a
20 steering committee composed of individuals living with PSC or caring for relatives living with
21 PSC. The study will be advertised by PSC Support and its findings will be evaluated and
22 disseminated by members of PSC Support.
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29 **ETHICS and DISSEMINATION**

30 *Ethical Approval*

31 This study was approved by the Health Research Authority and received a Favourable Opinion
32 from London - Queen Square NHS Research Ethics Committee under application number
33 18/LO/1075.
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40 *Informed consent*

41 Informed consent will be obtained for each participant. All participants will be provided with
42 an information sheet indicating the purpose of the study and will have an opportunity to ask
43 questions. Consent for participants partaking in the AVFGs will be recorded online. Formal
44 consent will also be recorded for all those taking part in narrative interviews. Should interviews
45 be carried out over telephone or via Telemedicine, participants will be asked to give verbal
46 consent which will be audio-recorded. Signed and written consent will be sought from those
47 taking part in face-to-face or email interviews. Consent from all members taking part in a
48 roundtable discussion will be audio-recorded.
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58 *Data protection and patient confidentiality*

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3 Information provided in confidence will be anonymised and not be used or disclosed in a form
4 that might identify the participant without his or her express written consent. Participants will be
5 given the opportunity to choose their own pseudonym and these will be held in password protected
6 databases on an encrypted NHS server at the Tavistock and Portman NHS Foundation Trust. Only
7 the Chief Investigator and researchers will have access to it, and only anonymised data will be
8 shared with anyone else. Only participants' unique pseudonyms will be linked to the raw data
9 collected (e.g. audio recordings). Audio files will be identified by pseudocodes, however it is
10 possible that names of people and/or places may be mentioned during the interviews. All digital
11 data files (including audio files) will be assigned a pseudocode and uploaded onto a secure NHS
12 server as soon as possible after data is collected, and the files deleted from the memory disk or
13 hard drive. Memory disks and hard drives containing data will be held in locked cabinets if it is
14 not possible to upload the files immediately following data collection. Data will be uploaded onto
15 the secure server, only accessible by authorised researchers, and will be transferred to a trusted
16 transcription company using secure data transfer methods. All members of the research team will
17 be reminded of their duty to observe the NHS code of ethics regarding patient confidentiality.
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31 *Dissemination*

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34 The findings of the study will be submitted for publication in peer-reviewed journals, and will be
35 presented nationally. All intellectual property rights relating to the data arising from the study shall
36 be held by the Tavistock & Portman NHS Foundation Trust. Participants who consent to receive
37 the study findings will be sent an electronic or hard copy interim report and a final lay summary
38 of the findings, depending on their preferred method of communication. They will also be
39 provided with the details of where to access the online publication of the full study report.
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46 **Authors' contributions:** EK and KM constructed the protocol. KM and VR drafted this protocol
47 article. It was subsequently reviewed by all authors (EK, DT, and MW).
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51 **Funding statement:** This work was supported by PSC Support grant number 220318VR.
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55 **Competing interests statement:** MW is the Chair of Trustees at PSC Support and an individual
56 living with PSC. DT is a member of the PSC Support Expert Panel. No other authors have
57 competing interests.
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Word Count: 3,963 words (including references)

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Standards for Reporting Qualitative Research (SRQR)*

<http://www.equator-network.org/reporting-guidelines/srqr/>

Page/line no(s).

Title and abstract

<p>Title - Concise description of the nature and topic of the study Identifying the study as qualitative or indicating the approach (e.g., ethnography, grounded theory) or data collection methods (e.g., interview, focus group) is recommended</p>	1
<p>Abstract - Summary of key elements of the study using the abstract format of the intended publication; typically includes background, purpose, methods, results, and conclusions</p>	2

Introduction

<p>Problem formulation - Description and significance of the problem/phenomenon studied; review of relevant theory and empirical work; problem statement</p>	3-4
<p>Purpose or research question - Purpose of the study and specific objectives or questions</p>	4

Methods

<p>Qualitative approach and research paradigm - Qualitative approach (e.g., ethnography, grounded theory, case study, phenomenology, narrative research) and guiding theory if appropriate; identifying the research paradigm (e.g., postpositivist, constructivist/ interpretivist) is also recommended; rationale**</p>	6-7
<p>Researcher characteristics and reflexivity - Researchers' characteristics that may influence the research, including personal attributes, qualifications/experience, relationship with participants, assumptions, and/or presuppositions; potential or actual interaction between researchers' characteristics and the research questions, approach, methods, results, and/or transferability</p>	N/A
<p>Context - Setting/site and salient contextual factors; rationale**</p>	8-9
<p>Sampling strategy - How and why research participants, documents, or events were selected; criteria for deciding when no further sampling was necessary (e.g., sampling saturation); rationale**</p>	7-8
<p>Ethical issues pertaining to human subjects - Documentation of approval by an appropriate ethics review board and participant consent, or explanation for lack thereof; other confidentiality and data security issues</p>	10-11
<p>Data collection methods - Types of data collected; details of data collection procedures including (as appropriate) start and stop dates of data collection and analysis, iterative process, triangulation of sources/methods, and modification of procedures in response to evolving study findings; rationale**</p>	5-8

Data collection instruments and technologies - Description of instruments (e.g., interview guides, questionnaires) and devices (e.g., audio recorders) used for data collection; if/how the instrument(s) changed over the course of the study	5-8
Units of study - Number and relevant characteristics of participants, documents, or events included in the study; level of participation (could be reported in results)	N/A
Data processing - Methods for processing data prior to and during analysis, including transcription, data entry, data management and security, verification of data integrity, data coding, and anonymization/de-identification of excerpts	9-10
Data analysis - Process by which inferences, themes, etc., were identified and developed, including the researchers involved in data analysis; usually references a specific paradigm or approach; rationale**	9-10
Techniques to enhance trustworthiness - Techniques to enhance trustworthiness and credibility of data analysis (e.g., member checking, audit trail, triangulation); rationale**	N/A

Results/findings

Synthesis and interpretation - Main findings (e.g., interpretations, inferences, and themes); might include development of a theory or model, or integration with prior research or theory	N/A
Links to empirical data - Evidence (e.g., quotes, field notes, text excerpts, photographs) to substantiate analytic findings	N/A

Discussion

Integration with prior work, implications, transferability, and contribution(s) to the field - Short summary of main findings; explanation of how findings and conclusions connect to, support, elaborate on, or challenge conclusions of earlier scholarship; discussion of scope of application/generalizability; identification of unique contribution(s) to scholarship in a discipline or field	N/A
Limitations - Trustworthiness and limitations of findings	N/A

Other

Conflicts of interest - Potential sources of influence or perceived influence on study conduct and conclusions; how these were managed	11
Funding - Sources of funding and other support; role of funders in data collection, interpretation, and reporting	11

*The authors created the SRQR by searching the literature to identify guidelines, reporting standards, and critical appraisal criteria for qualitative research; reviewing the reference lists of retrieved sources; and contacting experts to gain feedback. The SRQR aims to improve the transparency of all aspects of qualitative research by providing clear standards for reporting qualitative research.

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**The rationale should briefly discuss the justification for choosing that theory, approach, method, or technique rather than other options available, the assumptions and limitations implicit in those choices, and how those choices influence study conclusions and transferability. As appropriate, the rationale for several items might be discussed together.

Reference:

O'Brien BC, Harris IB, Beckman TJ, Reed DA, Cook DA. **Standards for reporting qualitative research: a synthesis of recommendations.** *Academic Medicine*, Vol. 89, No. 9 / Sept 2014
DOI: 10.1097/ACM.0000000000000388

For peer review only