Peer Review File

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Reviewer A

Comment 1: It would be helpful to include a few sentences about Sickle Cell Disease in general and common symptoms, and data that are monitored/collected by HCP. This would give non-experts an overview and to contextualise the data and categories identified in the results as missing.

Reply 1: Thank you, this makes a lot of sense, and we agree that the manuscript was previously missing this. We have included this in the methods section as demonstrated below.

Changes in the text:

Methods: "For the focus-group, individuals aged 18 and over with lived experience of SCD were recruited on a voluntary basis. We focused on SCD, as this is a rare condition subject to considerable day-to-day variability, with several key considerations impacting quality-of-life, not all of which being routinely observed within the health record, including hydration, the onset of sickle cell crises, infection, swelling, side effects from medication, including opiates, and issues with visual acuity."

Comment 2: In the method section it is unclear how the initial access to the individuals with SCD co-designing study materials was achieved and for how long they were involved.

Reply 2: We agree, and have since amended the methods to provide additional details regarding when and how access to patients was provided, as shown below. It is important to point out that one of those involved in providing materials for the focus-group also subsequently contributed to the writing of the paper [ZGS].

Changes in the text:

Methods: "Following introductions by a UK-based national sickle cell charity (the Sickle Cell Society), an advertisement poster was co-designed by individuals living with SCD and disseminated on the social media pages (LinkedIn and Twitter) of the Sickle Cell Society"

"To address these core questions, a semi-structured topic guide informed by existing literature [26] was co-designed with a patient research partner [ZGS] who the authors engaged with via the Sickle Cell Society."

Comment 3: Further, It would have good to report the demographic composition of the focus group and survey participants in the method section as it is later discussed that BAME individuals have higher rates of SCD, and the age might have impacted study results.

Reply 3: Unfortunately we did not collect demographic details as part of the focus group, as we did not have any exclusion criteria in mind during this initial phase of exploration and hypothesis generation. We were happy to speak to people from the age of 18 upwards, from any ethnicity. As a result we did not collect this data. We accept that this is a limitation in terms of generalizability, as it could be argued that certain age groups may be more or less willing to share personal health data, and therefore that the themes uncovered and further explored in the survey may have been subject to

change. Therefore, we have listed this as a key limitation within the discussion.

Changes in the text:

Discussion: “This may have been further compounded by advertising both the focus group and survey via social media, and requiring respondents to take part digitally. We had a high number of respondents aged 35 or under, and the age breakdown of those involved in our study may not be reflective of the population norms for SCD. Added to this is the limitation that we did not collect demographic data as part of the initial focus group, which informed the latter survey. Therefore, we cannot be certain that we achieved a balanced mix of views from those of varying ages, ethnicities and socio-economic status, which may have subsequently impacted our ability to delve into certain themes, and prevented exploration of these themes during the subsequent survey.”

Comment 4: While the study and interpretation of the results are on a solid base reporting level, however, they could be enhanced by contextualising their finding with literature critically examining digital interventions, chronic illness and datafied practices, or critical user studies such as:
https://doi.org/10.1146/annurev-anthro-102116-041244
https://doi.org/10.1177/205520762211095
https://www.jstor.org/stable/26652332.

Reply 4: We agree completely and have compared our findings in light of the papers suggested, in addition to others. Namely we have demonstrated the value for such interventions, especially in marginalized groups, but contrasted this against the risks associated with sharing health data via digital health.

Changes in the text:

Discussion: “This finding therefore suggests a disparity between what is currently being measured and therefore valued by clinicians in condition management, and what is valued by “empowered” patients [28,29]. This suggests a key role for such data in filling gaps in our understanding of poorly understood conditions from the patients’ unique perspective, as opposed to simply providing “biocapital” for others [30,31], including science and technology industries to harness and exploit, as suggested elsewhere [32,33].”

Discussion: “Co-design between patients and HCPs may help broaden current understanding and better align these stakeholder perspectives and expectations with regard to data collection and sharing. This requirement to better understand the experiences of patients and how digital health may play a role in broadening understanding has been well documented among lower-income or culturally marginalized individuals [38]. This research therefore provides a valuable addition to the literature, particularly in light of the struggles those living with sickle cell disorder face in being heard [39].”

Discussion: “Prior studies have shown that willingness to share health data among those with rare disorders comes with specific requirements in order to respect their privacy, choices and needs for information regarding the use of their data [7], with such conditions viewed as safeguards preventing data from being misused, surreptitiously extracted or used to serve agendas that benefit research and industry as opposed to patients [53,54].”

Comment 5: It would be good to include a discussion or forward looking section on how the data derived from this study could be used for digital health interventions or apps to support patients with SCD on a daily basis and how this could support also long term decision making with their HCP.

Reply 5: We have done as suggested and included a brief section at the end of the discussion.

Changes in the text:

Discussion: Future research should examine how the findings presented here can be put into practice. Respondents in both our focus group and survey were clear in their belief that more should and can be done to understand the full spectrum of what it is like to live with a rare condition, and that digital health can play a valuable role in filling this gap. What is unclear is how such a technology would need to look, the features required, and how feasible collection of such data is on a routine basis. Key to this is considering not only willingness to provide data, but also the quality of such data collected in real-world "non-controlled" settings, and how influential this is for healthcare providers required to make decisions regarding their patients' care.."

Comment 6: Overall, written in a clear and accessible manner, a few minor mistakes and colloquial forms can be found in the paper (e.g. didn't instead of did not). A grammar and spell check should fix this.

Reply 6: We have been through the manuscript, removing informal language and tidying up typos etc.

Comment 7: Line 53 & 66: poor awareness: of what and by whom?

Reply 7: Fair. We have removed both mentions owing to the words required to spell out awareness of natural history, symptoms and progression among HCPs and the wider public. Given the limitation in both the key findings and abstracts this seemed to make most sense.

Changes in the text:

Abstract: "Those living with the rare condition SCD were supportive of collecting and sharing data to foster research and improve understanding and outcomes."

Key findings: "Those living with the rare condition sickle cell disorder were supportive of collecting/ sharing personal health data with HCPs, charitable organisations and pharmaceutical companies; to improve understanding of day-to-day condition impact."

Comment 8: Line 88 "Manage their personal health data": people manage their health with or through data, not their health data.

Reply 8: We agree. Since changed as demonstrated below.

Changes in the text: "People living with long-term conditions are increasingly using digital health technologies (DHTs) to record and monitor health data, using it to manage their personal health [1-3]; encompassing a broad range of information such as medication adherence, health and lifestyle

practices, outcomes and experiences."

Comment 9: Line 99: "natural history": meaning unclear.

Reply 9: We used this term in the most scientific sense, referring to the natural history of condition progression, as denoted in natural history studies. We have since added the term progression to clarify. If this remains unclear, we can always remove the full sentence if required?

Changes in the text: "The wider scale collection, curation and analysis of personal health data may provide unrivalled opportunities in advancing current understanding of the natural history and progression of health conditions that may otherwise remain poorly understood [8,9]."

Comment 10: Line 123: "RWD": needs explanation. abbreviation?

Reply 10: Good spot, since corrected.

Changes in the text: "However, with patients not just the recipients of care, but rather experts with lived experience of these largely misunderstood conditions, the potential exists to expand usage of digitally derived real-world data (RWD)"

Comment 11: Table 1: Duplicate last row.

Reply 11: Thank you for spotting this, not quite sure what happened there. Corrected as suggested.

Comment 12: Line 330: "what data is shared and by whom:" colon indicates missing quote or similar?

Reply 12: We have checked the earlier versions of the manuscript and it is simply a typo.

Comment 13: Line 370-378: Some percentages do not match the figures, needs checking. Also consistency if decimal points are used or not.

Reply 13: Apologies, this is a failure in us conveying what was done here. When we said the following: "The largest opportunity reported by respondents was within the pharmaceutical industry where 71% of those willing to share health data were yet to do so, followed by charitable organisations (70%) and digital health interventions (65.2%)."

What we have done is taken the proportion of those who HAVE previously shared data and divided it through by the proportion who WOULD share data in the future. For example, in the case of the pharmaceutical industry 19.1% HAVE, 66% WOULD, 19.1/66 = 28.9% of those who WOULD, HAVE. Therefore, 71.1% of those who WOULD, HAVE NOT. We have since made this much clearer to the reader.

Changes to the text: "Across all organisational types, the proportion of respondents willing to share data was consistently greater than the proportion who had previously shared data with these organisation types. The largest opportunity reported by respondents was within the pharmaceutical industry where 19.1% of respondents had previously shared data, and 66% were willing to do so in

the future; suggesting that 71% of those willing to share health data were yet to do so. This was followed by charitable organisations where 70% of those willing to share data were yet to do so (21.3% shared to date compared to 70.2% willing to share) and digital health interventions (65.2%."

Reviewer B

1. Ref 10 is missing in the Main Text.

Response: Apologies, this must have been omitted either while editing the paper following the first round of revisions, or during our final round of internal review. Either way this reference was not needed here, and has therefore been removed. All other reference numbers have been updated to reflect the omission of the reference from the manuscript.

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2. Column headers are required for Table 2.

Response: Done

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3. There are two different legends in the file: Supp Table 1.docx. Please check and revise.

Response: Done

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4. Please check the word "emotitionally" in Figure 1.

Response: This has since been amended.

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5. Supplementary Figure 2 is not a figure. Please rename it as a Supplementary file or Appendix.

Response: Done, now named Supplementary File 1

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6. "For example, a 2021 study exploring the reasons why those living with rare disorders shared their personal health data online found that participants had several concerns about privacy, but that the motivation for sharing despite this risk was that it could lead to new developments, thereby helping themselves and others [56]."

The published year of Ref 56 is 2018. Please check whether revision is needed.

Response: Apologies, the Haeusermann paper is the correct paper, we however erroneously mis-labelled the year. I have reflected this change in the manuscript.

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7. Ref 25 was cited right after Ref 22 without Ref 23, 24 cited in between. Please renumber the references to meet the consecutive standard.

Author response: Apologies, this has now been completed.