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Consensus Statement

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Recommendations to address respondent burden associated with patient-reported outcome assessment

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Patient-reported outcomes (PROs) are increasingly used in healthcare research to provide evidence of the benefits and risks of interventions from the patient perspective and to inform regulatory decisions and health policy. The use of PROs in clinical practice can facilitate symptom monitoring, tailor care to individual needs, aid clinical decision-making and inform value-based healthcare initiatives. Despite their benefits, there are concerns that the potential burden on respondents may reduce their willingness to complete PROs, with potential impact on the completeness and quality of the data for decision-making. We therefore conducted an initial literature review to generate a list of candidate recommendations aimed at reducing respondent burden. This was followed by a two-stage Delphi survey by an international multi-stakeholder group. A consensus meeting was held to finalize the recommendations. The final consensus statement includes 19 recommendations to address PRO respondent burden in healthcare research and clinical practice. If implemented, these recommendations may reduce PRO respondent burden.

Millions of individuals provide PRO data regularly in a variety of settings[1]. The substantial benefits of utilizing PRO data for various purposes, including healthcare research, clinical practice, regulatory purposes and value-based healthcare decisions have been demonstrated and extensively documented[2–8]; however, the completion of PROs places a potential burden on respondents (patients), especially if responses are requested on a regular basis[9–14]. Respondent burden is the degree to which a respondent perceives their participation in a task as difficult, time consuming or emotionally stressful[15].

With regard to the completion of PRO measures, there are several factors that may influence respondent burden, including patient characteristics (such as literacy levels and cognitive impairment)

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and features of the chosen measure (including length, wording, content, sensitivity of items and formatting)[1,11,12]. Response burden could also be linked to the mode of administration of PROs (whether electronic, paper or any other format) and frequency of collection in both healthcare research and clinical practice settings[16,17]; however, these factors are likely to be inter-related and their associations with respondent burden may be nuanced and context-specific. For instance, recent studies have shown that the administration of longer PRO measures may not necessarily be associated with a perception of increased respondent burden, especially if respondents have a clear understanding of the purpose of collection and how their data would be utilized[1,17,18].

Failure to address respondent burden may lead to poor PRO completion rates, missing PRO data or trial participant withdrawal[17]. The US Food and Drug Administration (FDA) advises that sponsors should consider missing data and poor PRO completion rates as possible indicators of inappropriate respondent burden, item content or response options[11]. The implication of such missing data is that poor quality or nonrepresentative PRO information could be deemed as not sufficiently robust to evaluate treatment benefit, inform clinical care or regulatory decision-making. Therefore, to optimize PRO assessments, it is important that the potential benefits of PRO collection are weighed against the potential burden on respondents[19–22].

Given that there are no international guidelines that address this critical issue, the aim of this international effort was to develop consensus-based recommendations to facilitate the minimization of respondent burden for individuals completing PROs in both healthcare research and clinical practice.

Methods

The recommendations were developed through an international Delphi and consensus process as described in the COMET Handbook v.1.0 (ref. 23). The steering group (O.L.A., S.C.R., J.R., P.K. and M.J.C.) oversaw the design and conduct of the study.

Ethical approval

Ethical approval was granted by the University of Birmingham Ethical Review Board (ERN_22-0276). Study information was electronically provided to participants before survey completion and before the consensus meeting. Delphi participants provided electronic informed consent and written consent was obtained from consensus meeting delegates.

Generation of candidate recommendations

Twenty-six candidate recommendations were initially generated based on the findings of a comprehensive literature review conducted and published in 2022 by members of the steering group[1]. In brief, PubMed was searched on the 22 November 2021 to identify eligible studies (further details are provided in Supplementary Information). There were no restrictions on study design or language of publication. Title, abstract and full-text screening was conducted independently by two reviewers. Analysis of the qualitative data was performed using the framework method[24].

International Delphi process

The stakeholders for this project consisted of trialists, PRO-focused clinical researchers and statisticians, patient partners and advocates, healthcare professionals, journal editors, policymakers, industry experts and other professionals who are involved in the implementation of PROs for healthcare research, drug approvals and clinical practice. These individuals were identified through personal networks and suggestions from known experts.

In 2023, the steering group sent invitations to 168 international stakeholders to participate in an online Delphi process to vote on the candidate recommendations and propose additional recommendations. Stakeholder grouping and characteristics are described in the Supplementary Information.

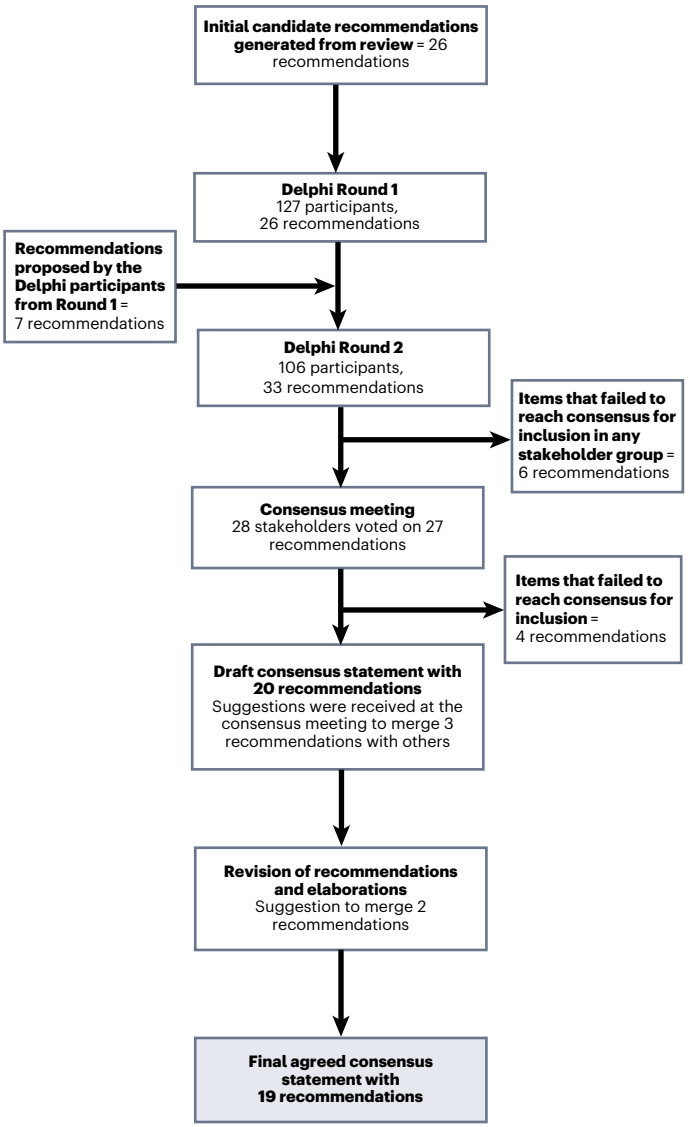

Fig. 1 | Development of the recommendations. The flow chart illustrates the process, which culminated in 19 recommendations on how to address respondent burden associated with PRO assessment.

The two online Delphi surveys were delivered using the Delphi-Manager software (v.5.0) developed and maintained by the COMET initiative. The invitation emails provided information about the study, who to contact for further information and a link to the consent form and survey for those who wished to take part. Voting on the importance of the 26 candidate recommendations was anonymous and scored using a nine-point scale (1–3, not important; 4–6, important but not critical; and 7–9, important and critical). A total of 127 responses were received for Round 1 of the Delphi survey and 106 responses (83% of participants from Round 1) were received for Round 2. The Delphi participants had the option in Round 1 to provide qualitative feedback on the suitability of each recommendation, suggest modifications and propose additional recommendations. The feedback was reviewed and seven additional recommendations were proposed and taken forward to Round 2. Participants were subsequently sent a document detailing how the feedback from Round 1 was addressed by the steering group. Anonymized item-level Round 1 ratings by stakeholder group

Table 1 | The PRO respondent burden recommendations

Number	Recommendation	
Rationale and schedule for PRO assessment		
1	Involve patients, clinicians and other relevant stakeholders in the formulation of the PRO research question(s) or clinical objectives to ensure that they are important and relevant.	
2	Consider the degree of burden that any data collection may impose on respondents and carefully balance this with the quantity and quality of data required.	
3	Ensure that patients and clinicians and other relevant stakeholders are involved in decisions about the PRO assessment schedule and the frequency of assessment.	
Measure selection		
4	Review the literature to identify relevant concept(s) of interest.	
5	Qualitative and quantitative methods may be used to obtain input from patients and clinicians on selecting or developing PRO measures so that they are fit for purpose.	
6	Consider the complexity of the format of PRO measures and their instructions.	
7	Consider the literacy level of respondents.	
8	Ensure that the selected PRO measures are culturally and linguistically relevant for the target population.	
9	Consider the length of PRO measures and decide whether the use of a relatively longer measure is justified.	
10	If selecting more than one PRO measure, avoid overlapping constructs.	
11	Consider the recall periods for measures as longer timeframes may be burdensome for some respondents.	
Measure delivery		
12	Ensure respondents understand why the data are being collected, who will have access, how it will be used and why it is important for them to complete the PRO measures.	
13	Provide clear instructions, training and support for respondents on the completion of PROs as needed.	
14	Provide training and guidance for research staff and clinicians in clinical practice so that they understand the value of PROs and respondent burden.	
15	Specify the level or type of support that can be provided to respondents to facilitate the completion of PRO measures.	
16	Offer flexible modes of administration to meet the needs of target populations and underserved groups.	
17	Where possible, consider the use of ePROs, which may help reduce respondent burden, but must be balanced with the needs and preferences of the target population.	
18	If developing new ePRO systems or modifying an existing one for a new context of use, involve patients and clinicians in the co-design of the ePRO system.	
19	Explore the functionality of ePROs with diverse representatives from the target population where possible.	

were also shared with the Delphi participants for their consideration before voting in Round 2. The participants were informed that their Round 1 responses would be retained if they did not complete Round 2. Delphi participants who agreed to be named are listed in Supplementary Information.

Prespecified threshold for inclusion of recommendations

For inclusion, a recommendation was required to meet the prespecified threshold of ≥70% of the Round 2 Delphi participants rating it as 'important and critical' (7–9) and ≤15% rating it as 'not important' (1–3). Recommendations that achieved consensus were reviewed and ratified at the consensus meeting. Recommendations that did not achieve overall consensus but were rated by ≥70% of any stakeholder group as 'important and critical' (7–9) were discussed at the consensus meeting. A summary of qualitative feedback from the Delphi participants on these recommendations was presented at the meeting. Recommendations that did not meet any of the above criteria were proposed for exclusion.

International consensus meeting

The aim of the meeting was to reach consensus on the content of the recommendations. Following Round 2, the steering group collated and reviewed the ratings and the qualitative feedback from the Delphi participants. They proposed the inclusion or exclusion of recommendations based on the Delphi data and sent these to the consensus meeting delegates ahead of the meeting (Supplementary Information).

A consensus meeting was hosted online via Zoom by the University of Birmingham, UK, in September 2023. The meeting was attended by 36 international delegates who had participated in the Delphi study. The delegates were selected in a manner that ensured good representation across stakeholder groups. There were 28 voters and 8 nonvoters. The nonvoters were members of the steering group and experts from institutions already represented. The delegates consisted of 12 trialists/academic researchers/statisticians, 7 industry experts, 6 regulators/policymakers, 5 healthcare professionals, 5 patients/patient advocates and members of the public and 1 journal editor (Supplementary Information).

Delegates discussed the importance of the recommendations that met the prespecified threshold for consensus (overall) as well as the recommendations that only reached consensus in one or more stakeholder groups. The wording and explanatory text of recommendations were also discussed as required. Following group discussion, delegates were invited to vote anonymously on the candidate recommendations using the Zoom Poll tool. The voting options were to include or exclude with response options of 'yes', 'no' or 'further discussion required'. Supplementary Information provides further details of how the voting was conducted specifically for each recommendation.

Final consultation

Following the consensus meeting, delegates were sent the draft recommendations for their comments and suggestions on the wording and approval of the final version. Supplementary Information provides further information on the methods.

Results

Following the Delphi surveys, 19 recommendations achieved overall consensus and were proposed by the steering group for inclusion; 8 recommendations only achieved consensus in one or more stakeholder groups and required discussion at the consensus meeting; and 6 recommendations did not reach consensus in any stakeholder group and were proposed for exclusion. Further details of the voting and decisions at the consensus meeting can be found in Fig. 1 and Supplementary Information. The 19 recommendations were ratified for inclusion during the consensus meeting (there was consensus to merge two of these recommendations). Of the eight recommendations that were individually discussed, two were voted in for inclusion as standalone recommendations; and there was consensus to merge two other recommendations with recommendations that had been ratified. Two recommendations were merged based on suggestions received on the initial draft of the manuscript. Further details are provided in Supplementary Information.

The final consensus statement provides 19 recommendations for consideration by anyone involved in designing and implementing PRO assessment strategies for healthcare research and clinical practice (Table 1). An elaboration describing each recommendation with supporting evidence is presented below. The recommendations are presented in accordance with the categories from the published review[1], namely (1) rationale and schedule for PRO assessment; (2) measure selection; and (3) measure delivery.

Rationale and schedule for PRO assessment

Recommendation 1: involve patients, clinicians and other relevant stakeholders in the formulation of the PRO research question(s) or clinical objectives to ensure that they are important and relevant. Effective involvement of patients, clinicians and other relevant stakeholders in the formulation of PRO research question(s) and clinical objectives can help ensure the assessment of outcomes that are relevant and valued by all stakeholders in healthcare research and clinical practice[25–27]. Stakeholder involvement will vary depending on the context of PRO use. For example, patients, caregivers and clinicians can provide valuable perspectives both in healthcare research and in clinical practice settings, while early input from regulatory agencies may be particularly useful in clinical trials of investigational medicinal products. PRO assessments may be perceived as less burdensome if the research questions or clinical objectives are considered relevant and important by patients and other relevant stakeholders[28,29].

Recommendation 2: consider the degree of burden that any data collection may impose on respondents and carefully balance this with the quantity and quality of data required. The rationale for collecting PRO data for healthcare research and clinical practice should be evidence-based and should demonstrate that the data collection justifies the burden and potential risks of data collection, such as the time required, emotional angst, distress or fatigue[30].

Recommendation 3: ensure that patients, clinicians and other relevant stakeholders are involved in decisions about the PRO assessment schedule and the frequency of assessment. Consultation with patients, clinicians and other relevant stakeholders will help ensure that the PRO assessment schedule captures clinically relevant periods during treatment or clinical management[25]. The schedule of PRO assessments, including overall duration of assessment, will depend on the research or clinical objective; however, the potential respondent burden should be considered, while maximizing the collection of clinically relevant data[31]. The assessment schedule may not necessarily be tied to clinic visits; considerations for the mode of administration are described in Recommendation 16 (ref. 25).

The frequency of PRO administration should consider disease trajectory and balance this with respondent burden. Data should only be collected if they are essential to addressing the research objective or informing patient care. In a clinical practice setting, patients with a stable disease/condition may require less frequent PRO administration. Long-term monitoring may be burdensome and may lead to reduced PRO completion rates, but may be warranted in some instances (for example, for chronic disease monitoring or real-world evidence generation)[32].

Consider the time points for assessing PRO measures within an allowable window and their relationship to clinical events (for example, treatment, clinical assessments and other assessments). Depending on the research or clinical objective, it may not be necessary to deliver all PRO measures at every time point[28]. A modular approach could be taken, in which different assessment frequencies are selected to reduce patient burden[9]. For example, more general quality-of-life aspects (for example, social or emotional well-being) may be assessed less frequently than the presence and severity of symptoms.

Measure selection

Recommendation 4: review the literature to identify relevant concept(s) of interest. To minimize burden, the concept(s) measured by the PRO measure should be relevant to the target population, disease setting and context of use (healthcare research or clinical practice). A literature review and/or surveys or qualitative work (Recommendation 5) can be conducted to identify concept(s) of interest.

Recommendation 5: qualitative and quantitative methods may be used to obtain input from patients and clinicians on selecting or developing PRO measures so that they are fit for purpose. Patient, clinician and other stakeholder input may be obtained using qualitative and/or quantitative methods, including interviews, focus groups and surveys[28,33]. Patient engagement and involvement is helpful to inform selection of PRO measures that capture meaningful outcomes, while reducing burden[26,34]. This may help avoid overly paternalistic approaches that are clinician- or researcher-driven[35].

Recommendation 6: consider the complexity of the format of PRO measures and their instructions. PRO measures with greater complexity that require more cognitive effort to understand, such as those with complicated instructions, phrasing and reverse response options, may be more burdensome for respondents[11,17,33]. Discussions with patients from the target population may be used to explore these issues and ascertain the level of burden that may be associated with the PRO measures being considered[33]. The use of multiple measures with different formats may further increase complexity and should be avoided if possible.

Recommendation 7: consider the literacy level of respondents. Where possible, promote inclusion of individuals with all levels of reading, writing and problem-solving abilities[11,35]. Ensure that PRO content and training is easy to understand for respondents with different literacy levels and educational experience by conducting relevant readability assessments (for example, Flesch–Kincaid grade level or SMOG (simple measure of gobbledygook) index score)[35]. It is recommended that PRO items be at the reading level of 11–12 years of age or lower; however, this criterion should be contextualized to the intended target population and justified[16].

Recommendation 8: ensure that the selected PRO measures are culturally and linguistically relevant for the target population. If PRO measures are translated into other languages, ensure that they

have undergone linguistic validation with cognitive debriefing[27,36]. Linguistic validation is the testing of translated PRO measures with patients or lay individuals who are representative of the cultural group intending to use the measure to check understandability, interpretation and cultural relevance of the translation[37].

Recommendation 9: consider the length of PRO measures and decide whether the use of a relatively longer measure is justified. Measure length is often considered as a contributing factor to PRO respondent burden; however, measure length should be balanced with patient and clinician input on what outcomes are most relevant to the population and context[30]. Relatively shorter measures may reduce respondent burden and increase patients' willingness to complete forms[38]; however, brevity should not outweigh the utilization of PRO measures with appropriate measurement properties (reliability and validity) to assess outcomes that are relevant to key stakeholders, the research question(s)/PRO objectives and purpose of collection[30,39,40].

There is evidence that the length of a PRO measure may not necessarily be associated with respondent burden[16,18,30] and high response rates could be achieved with administration of relatively longer PRO measures if they are meaningful to respondents[41]. Furthermore, patients may prefer longer forms to shorter versions if they capture concepts that matter to them and can meaningfully inform care[1,17,18]. Ultimately, evaluations of PRO measure length should consider the context of use of the data, the views of those living with the condition and those responsible for using the data. Early patient involvement in selection of the measures is crucial (Recommendation 5).

Linked to the issue of PRO measure length is estimated completion time. The needs of the target population (for example, age, disease severity and comorbid conditions) and aspects of design (for example, mode and place of PRO measure administration), may impact overall completion times. Relevant stakeholder input should be sought on the anticipated completion time and its appropriateness in terms of the research or clinical context and the patient population. For instance, a PRO measure that generally requires more time to complete might not be suitable for use in a busy outpatient clinic. The same PRO measure might be appropriate for use if completed remotely, before clinical appointments. In terms of patient population, a PRO measure that requires less time to complete may be preferable for patients with osteoarthritis of the hand. In a research context it has been suggested that completion time of baseline PRO assessments should ideally be limited to 20 min and 10–15 min for subsequent assessments[10,22].

Recommendation 10: if selecting more than one PRO measure, avoid overlapping constructs. The use of more than one PRO measure requires careful consideration to avoid duplication, overlap or redundancy of constructs[9,42]. The administration of several PRO measures may lead to respondent burden and a higher likelihood of missing data in those measures administered later, particularly if the constructs overlap. For research purposes, it is advisable that measures to support the primary and/or secondary outcomes are prioritized over those supporting exploratory outcomes.

Recommendation 11: consider the recall periods for measures, as longer timeframes may be burdensome for some respondents. When selecting PRO measures, it is important to consider the recall period (for example, 'In the last 7 days…') and whether characteristics of the disease/condition will affect the respondents' ability to recall the information easily and correctly[11,43]. The majority of PRO measures will often have a specified validated recall period, which should not be changed without consultation and approval from the instrument developer. If multiple recall periods have been validated by developers for a particular measure, then input from relevant stakeholders, including clinicians and patients, is recommended to decide which is most appropriate for respondents and the disease/condition of interest[43].

Measure delivery

Recommendation 12: ensure that respondents understand why the data are being collected, who will have access, how it will be used and why it is important for them to complete the PRO measures. It is important to inform patients about why PRO data are being collected, making it clear how the data they report could help improve their own care in clinical practice and the future treatment of patients in healthcare research[10,26,44]. Perceptions of the intrusiveness of items and their usefulness may influence respondents' perception of burden[14]. Explanations of the importance of PRO collection and the challenge of missing PRO data, may encourage respondents to complete PROs on a regular basis[26]. This recommendation applies not only in healthcare research settings, where informed consent is formally obtained, but also in clinical practice where PROs are being used as part of standard care (and patients typically do not sign consent forms).

Recommendation 13: provide clear instructions, training and support for respondents on the completion of PROs as needed. It is important that patients are provided with clear instructions on how to provide their PRO responses and be given ongoing support as needed. This may enhance the quality and completeness of the data collected.

Recommendation 14: provide training and guidance for research staff and clinicians in clinical practice so that they understand the value of PROs and respondent burden. PROs may be perceived to be burdensome by research personnel, clinical teams and research ethics committees, particularly if there are numerous measures or participants are very ill[12,26]. Qualitative interviews suggest that trialists may be reluctant to collect PROs due to the perceived respondent burden, even when participants may be willing to complete them[26]. Appropriate training for staff might help alleviate their concerns and avoid an overly paternalistic approach, and may help them address any questions raised by participants regarding PRO collection. It may also help them provide information on the importance and value of data collection, which may motivate participants to complete PRO measures.

For clinical trials, site manuals or protocols should provide specific guidance on PRO administration and management and highlight the importance of facilitating adherence and completeness of data[31].

Recommendation 15: specify the level or type of support that can be provided to respondents to facilitate the completion of PRO measures. For respondents unable to complete PRO measures on their own, consider and specify what help can be provided to support completion by the respondent (for example, holding a pen, assistance with a telephone or computer keyboard, scrolling/turning pages or reading out text)[11]. Responses to the PRO questions should be decided by the patient and not an assisting person.

Recommendation 16: offer flexible modes of administration to meet the needs of target populations and underserved groups. Modes of PRO administration may include paper, mobile device applications (apps), web-based completion, telephone interviews, interactive voice response, audio-computer-assisted interviews and other modes[10,28,36]. The needs of the target population and their individual preferences should be considered, such as paper or electronic delivery and whether multiple modes are needed to reach all respondent groups[36]. For example, in older people or respondents with low literacy or visual impairment, interactive voice response, provision of grip-pens or interviewer-administered PRO measures could be considered to reduce the burden[10]. Patients can also provide feedback on the acceptability of a bring-your-own device versus provisioned devices and additional options such as tablets or paper versions in the waiting room for those who cannot complete PROs electronically at home. Practical ways to reduce the burden at clinic or study visits should be considered[28].

Table 2 | PRO resources to support the collection of PROs in healthcare research and clinical practice

Resource title	Focus	Purpose
ISOQOL recommends minimum standards for patient-reported outcome measures used in patient-centered outcomes and comparative effectiveness research[52].	Research	Selecting PRO measures
Guidelines for inclusion of patient-reported outcomes in clinical trial protocols: the SPIRIT-PRO extension[53].	Research	Protocol guidance
SPIRIT-PRO extension explanation and elaboration: guidelines for inclusion of patient-reported outcomes in protocols of clinical trials[56].	Research	Explanation and elaboration
SPIRIT-PRO PROtocol reporting template: a template based on recommendations for writing clinical trial protocols with patient-reported outcomes[54].	Research	Protocol template
'Give us the tools!': development of knowledge transfer tools to support the involvement of patient partners in the development of clinical trial protocols with PROs, in accordance with SPIRIT-PRO extension[57].	Research	Patient-focused PRO tool
Patient-reported outcome assessment must be inclusive and equitable[35].	Research and clinical practice	Equity, diversity and inclusion considerations for PRO assessment
Ethical considerations for the inclusion of patient-reported outcomes in clinical research: the PRO ethics guidelines[22].	Research	Ethical guidance
Recommendations for including or reviewing patient-reported outcome end points in grant applications[58].	Research	Grant writing guidance
The use of PRO measures in oncology studies[10].	Research and drug approval	Regulatory guidance (oncology setting)
Guidance for industry, FDA staff and other stakeholders. Patient-focused drug development: methods to identify what is important to patients[59].	Research and drug approval	Regulatory guidance
Guidance for industry, FDA staff and other stakeholders. Patient-focused drug development: collecting comprehensive and representative input[60].	Research and drug approval	Regulatory guidance
Core patient-reported outcomes in cancer clinical trials guidance for industry[9].	Research and drug approval	Regulatory guidance
Patient-focused drug development: selecting, developing, or modifying fit-for-purpose clinical outcome assessments: guidance for industry, FDA staff and other stakeholders[34].	Research and drug approval	Regulatory guidance
Patient-focused drug development: incorporating clinical outcome assessments into endpoints for regulatory decision-making. Guidance for industry, FDA staff and other stakeholders[61].	Research and drug approval	Regulatory guidance
The PROTEUS guide to implementing patient-reported outcomes in clinical practice: a synthesis of resources[55].	Clinical practice	Guidance for the use of PROs in clinical practice
Best practices for the electronic implementation and migration of patient-reported outcome measures[49].	Research	Best practices for migrating and implementing PRO measures
Updated recommendations on evidence needed to support measurement comparability among modes of data collection for patient-reported outcome measures: a good practices report of an ISPOR Task Force[45].	Research and clinical practice	Evidence requirements for measurement comparability of modes of data collection

ISOQOL, The International Society for Quality of Life Research.

Consider the implications of using different modalities when preparing data for analyses. If multiple modes are used for data collection to minimize burden and facilitate diversity and adherence, consider how data from different sources will be integrated. For more information on measurement comparability, see the updated recommendations from The Professional Society for Health Economics and Outcomes Research (ISPOR) Task Force on measurement comparability between modes of data collection for PRO measures[45].

Recommendation 17: where possible, consider the use of ePROs, which may help reduce respondent burden, but must be balanced with the needs and preferences of the target population. With patient populations who have access to and are comfortable with electronic devices, the use of electronic PROs (ePROs) may offer additional functionality, which could help reduce burden and improve adherence in healthcare research and clinical practice[46]. This could include allowing completion on their own devices, with real-time reminders, notifications and responses from the research or clinical team, either to thank them for completion or to respond to issues identified on the PRO, depending on the context of use. Furthermore, ePROs make the use of innovations such as computerized adaptive testing possible. ePROs may also facilitate symptom monitoring between visits[12]; however, patients may face barriers to using digital services, including a lack of digital skills/low computer literacy or lack of access to reliable information technology infrastructure. Estimates suggest that 37% of the world's 7.8 billion population are digitally excluded, with older people, people on low incomes and other marginalized groups most likely to be affected[35]. It is important that these potential barriers and the preferences of the patient population in terms of mode of collection (as described Recommendation 16) are carefully considered with

patient input when developing PRO strategies, to ensure that PRO assessments are as inclusive and equitable as possible[35].

Recommendation 18: if developing new ePRO systems or modifying an existing one for a new context of use, involve patients and clinicians in the co-design of the ePRO system. Ensure that the patients providing input to the development or modification of the ePRO system include representatives from the target population and are diverse in terms of computer literacy and internet access, considering attributes as appropriate to the research question or clinical context[47]. Examples may include but are not limited to: sex; gender; socioeconomic background; race/ethnicity; age; health literacy; computer literacy and internet access; and disease characteristics. There may be country-specific regulatory expectations and requirements that may also need to be considered when developing ePRO systems[48]. The Electronic Clinical Outcome Assessment Consortium has published best practices for the electronic implementation and migration of paper PRO measures to ePROs[49].

Recommendation 19: explore the functionality of ePROs with diverse representatives from the target population where possible. Several ePRO features may facilitate completion and help to minimize burden[46]. Patient involvement in the study co-design and usability testing with the target population can be used to identify appropriate formats[50,51]. Depending on the context and where permissible, consider providing the following elements in the platform: estimated completion time, progress tracker, graphical results that are easy to interpret, positive messaging/reminders, completion rate and a thank you message after completion[42,46]. In terms of the format consider using underlining and capitalization where appropriate, easy-to-read fonts and font sizes, one question per screen, back/next buttons and location, branching logic and adaptive web design (where multiple versions of a web page are created to fit different devices)[11,42,49]. Seek patient preferences on how they receive requests and reminders to complete ePRO measures (for example, emails and/or text messages).

Discussion

Discussions about respondent burden frequently arise when health researchers, trialists and clinical teams are considering the use of PROs for healthcare research or clinical practice. This consensus statement provides accessible information in the form of consensus-based recommendations for addressing PRO-related respondent burden. The 19 recommendations are organized into three categories: rationale and schedule of assessments, measure selection and measure delivery.

The use of these recommendations by stakeholders such as trialists, researchers, clinicians and healthcare providers may facilitate the identification of factors that could influence PRO-related respondent burden and support the formulation of mitigating efforts appropriate for the context of use. Research and clinical teams are encouraged to seek input from PRO experts and utilize these recommendations, as well as the resources we have highlighted in Table 2, when considering the implementation of PROs for healthcare research or clinical practice.

While the recommendations are nonmandatory and not all may be relevant to every context, stakeholders are urged to use and reference this consensus statement to demonstrate explicitly how they have considered and addressed the issue of respondent burden. The consensus statement may also be a useful reference for those involved in scientific and ethical review of protocols and supporting materials such as peer reviewers, funding panels and ethics committees.

Although the recommendations focus on PROs in adult patients within healthcare research and clinical practice, they could be considered for use in other settings or populations such as pediatric populations and measures such as patient-reported experience measures; however, further considerations may be relevant in these contexts beyond the scope of the present work.

These recommendations have some limitations. First, while the initial review that informed the generation of the candidate recommendations was comprehensive (with 89 articles included[1]), only one database (PubMed) was searched. There is a possibility that some relevant articles might have been missed and that some potential recommendations were not identified; however, the international Delphi participants had the opportunity to provide qualitative feedback and propose additional recommendations not identified by the review during Round 1 of the Delphi process.

Second, these recommendations do not consider burden from the perspective of research or clinical staff; separate recommendations are needed to address burden concerns for these stakeholders.

Addressing PRO respondent burden could help to ensure the collection of more representative and high-quality PRO data to inform regulatory decisions and patient care. This work is complementary to existing resources to support the collection of high-quality PROs in healthcare research and clinical care. These include the resources available on the PROTEUS trials and practice website[52–55], equity, diversity and inclusion in the collection and utilization of PROs[35], PRO ethics guidelines[22], the US FDA guidance[11] and European Medicines Agency guidance[10] (Table 2).

The use of the recommendations in this consensus statement and related guidance could lead to high-quality PRO data collection that carefully considers the needs of respondents, promoting inclusive data collection. The impact of these recommendations, when implemented in different clinical contexts, should be evaluated in future research.

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Author contributions

O.L.A., M.J.C., S.C.R., J.R. and P.K. were responsible for concept and design, acquisition and data analysis. O.L.A. and M.J.C. had full access to the data in the study and are responsible for the integrity of the data and the accuracy of the data analysis. All authors were responsible for interpretation of data. O.L.A. and M.J.C. were responsible for drafting the manuscript. All authors were responsible for critical revision of the manuscript for intellectual content.

Competing interests

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Additional information

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