Author's response to reviews

Title: Assessing Disease Disclosure in Adults with Cystic Fibrosis: The Adult Data for Understanding Lifestyle and Transitions Survey

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Author's response to reviews: see over
Dear Dr. Shipley,

I would like to thank you and the reviewers for your thorough reviews of the manuscript we submitted to *BMC Pulmonary Medicine* entitled, “Assessing Disease Disclosure in Adults with Cystic Fibrosis: The Adult Data for Understanding Lifestyle and Transitions Survey” (MS: 8525232753933284). We have addressed reviewers’ comments regarding the manuscript and addressed all of the concerns. Below we have outlined each of the decisions we made about the manuscript in response to the reviewers’ comments and hope that you find them satisfactory.

**Reviewer: 1**

**Minor Essential Revisions:**

1) **RESULTS** (under the “Disclosure by lung function” heading): the authors state “disclosure to others may be influenced by severity of lung disease” and they show significant differences in the extent of disclosure as a function of airway limitation. It appears from the graphs that the extent to which the categories apply (e.g. percent of the “not applicable” category) may account for a large portion of these findings, particularly for the Supervisor/Employer/Teachers and Co-workers categories. This makes sense given that severe airway restriction might influence one’s ability to work. So a remaining, unanswered (yet, closely related) question is: among those to which these categories ARE applicable, do the rates of disclosure vary according to the extent of airway limitation? Running these additional follow-up analyses that exclude those who selected “N/A” may shed some additional light on these findings and control for this potential confound. **We appreciate the reviewer’s comment and would like to clarify that analyses only included participants for whom the analysis was applicable. The N/A category was not included in analyses but was used in the figure to accurately represent the sample sizes. The chi-square analyses in the results section are based on the number of participants for whom it applied. For example, with co-workers, only n=561 had co-workers; thus, this was the sample used for analyses. If the participant chose N/A, they were not included in the analysis. We have clarified this in the statistical analysis section via the following sentence: “Between-group analyses were conducted using Chi-square tests and only participants for whom the question was appropriate (e.g. only those who had co-workers were included in the analysis regarding differences in disclosure to coworkers based on disease severity).”**

2) **RESULTS** (under the “Challenges discussed with partner” heading). I think these results are interesting in terms of the influence of gender on issues discussed with partner prior to a commitment; however, it is not clear how these analyses relate to the stated objectives of this paper (the impact of disclosure on
relationships and/or the “how adults with CF perceived the disclosure process within specific personal and professional relationships”). The authors are encouraged to better link these analyses/results to the disclosure process in the introduction, in terms of the statistical analyses performed (e.g. gender differences in disclosure) and/or conceptually by making these linkages more explicit. Alternatively, the authors may consider dropping these analyses. Based on the comments, we have altered this section to focus more on gender effects on disclosure with some details of gender influence on discussions with significant others.

Discretionary Revisions

3) ABSTRACT/BACKGROUND/DISCUSSION: An important finding of this paper was that disclosure appears to vary by the severity of lung disease; however, this is not mentioned in the abstract or as an objective in the background. The authors are encouraged to clarify this point in those sections. Thank you for this suggestion, changes have been made to both the abstract and the background. The abstract now includes: Results also indicated that disclosure may be influenced by severity of lung disease, with CF individuals having normal or mild lung disease based on FEV1 being less likely to disclose their diagnosis to both co-workers (p< 0.01) and supervisors/teachers (p< 0.01).

4) RESULTS: The authors are encouraged to consider adding effect sizes to their statistical analyses. Effect sizes have now been added to the results section, as well as one sentence regarding calculation of effect sizes in the statistical analysis section (“Effect sizes were calculated from Cramer’s phi to Cohen’s d to ease interpretation, with 0.20, 0.50, and 0.80 representing small, medium, and large effects, respectively.”)

5) DISCUSSION (last paragraph), minor point: The authors state that “‘preventative disclosure’, as it is termed, may have a positive influence on relationships by increasing awareness and reducing misperception” and rightfully point out that given some potentially negative effects that guidance and suggestions for disclosure may be necessary. It might be helpful to acknowledge that by definition Preventive disclosures include both concealment and disclosure (p. 246 Joachim & Acorn, 2000) for this very reason. We thank the reviewer for pointing this out. The reference to this has been added and the citation provided. The section now reads: Thus ‘preventative disclosure’ [5], as it is termed, may have a positive influence on relationships by increasing awareness and reducing misperceptions. By definition however preventative disclosure requires an analysis of the balance of potential positive and negative effects of disclosure vs continued concealment. Given that we found some negative effects of disclosure reported for supervisors/employers/teachers or romantic partners, it may be important for caregivers to provide guidance and suggestions for disclosure in these social contexts.

Reviewer 2

Major Compulsory Revisions:
There are relatively few studies of disease disclosure in CF. However, there are relevant studies from a number of other chronic conditions. Although different in their etiology, conditions like HIV, arthritis, epilepsy and mental health or mood disorders share some features with CF like being largely invisible to others (i.e., people may not be aware that an individual has a chronic condition), the potential for disability with everyday activities, an uncertain disease course, and periods of wellness interrupted by episodes of impairment or more serious illness flares. These commonalities are relevant to self-disclosure and have been examined in research with other conditions, especially disease disclosure in the workplace. This paper would be strengthened if the introduction and discussion were situated within this larger body of research and the study findings were discussed in terms of the new data they add to the broader research area. Currently, there are only 9 studies referenced in the paper, 4 of which relate to disclosure. In particular, previous studies link disclosure to need (e.g., illness severity), perceived support from others, and concerns about stigma and potential discrimination. Thank you for this recommendation and specific recommended citations. We have made significant changes to the background and discussion to include reference to previous research in other diseases. In addition, the recommended citations have all been added.

Descriptive results are erroneously reported as reflecting a significant causal relationship between variables. However, these relationships weren’t tested. For example, one of the headings in the results section is, “Effect of disclosure on relationship”. This section states that “relationships with relatives or close friends were most likely to be positively affected by disclosure” and “…a negative effect was most likely to be reported for romantic partners…”. The first paragraph of the discussion states, “… this survey is the new insight into patterns of disclosure in adults and its subsequent effects on relationships” and “The decision to disclose CF was shown to be influenced by several factors…, which are most likely linked with self-perceived risk/benefit evaluations of disclosure”. This does not accurately capture the data or the findings. Causal language throughout the results and discussion sections should be changed and replaced with descriptive language and a report of significant associations. For example, rather than “relationships with relatives or close friends were most likely to be positively affected…” the findings should read, “relationships with relatives or close friends often were perceived as being positively affected…”. A “negative effect was most likely to be reported” should be changed to “a negative effect was reported more frequently for…”. Stating that “our finding that disclosure has a neutral or positive effect on most relationships…” should be changed to “Our finding that disclosure was often perceived as having no effect or a positive effect on…” Thank you for this important suggestion. The title in the results section has been changed from “Effect of disclosure on relationship” to “Perceived effects of disclosure on relationships.” All causal language in the results and discussion has been replaced.

As noted earlier, the discussion section would benefit from placing the results within the context of other research on disease self-disclosure to emphasize the complexity of this area and its relevance. For example, the authors note that workplace issues surrounding disclosure may relate to time off for health care utilization and fear of rejection. Elsewhere, they discuss that individuals may be
more likely to disclose if they “risk detection”. Other research on self-disclosure highlights that perceived need (e.g., disease severity, need for treatment) and perceiving others as supportive consistently relates to disclosure. Some research has also linked this support to outcomes (e.g., less work stress, more help from others, fewer workplace problems). Putting the findings from this study into this broader context would help develop areas where there is additional research needed in CF. This should be highlighted in the discussion. Thank you. Please see the response to comment number 1.

4) The tone of the conclusion paragraph should be altered. The authors write, “those with mild lung disease are more likely to conceal their CF diagnosis from supervisors, employers, teachers and co-workers”. This suggests an active attempt to hide one’s disease from others, which may or may not be true. Data from other research suggests that those with mild conditions may feel their condition doesn’t limit or interfere with activities and therefore, there is no need or reason to disclose. Without additional research into the reasons people have for disclosing or not disclosing, a more neutral interpretation or a range of interpretations should be provided. The authors could note that future research should examine in detail reasons for disclosing or not disclosing. Thank you for this distinction. We have changed the concluding paragraph to reflect that more mild patients may not feel the need to disclose (rather than are more likely to conceal.) We have also addressed this specifically in the discussion: “Specifically, patients with mild disease may consider their symptoms less obvious and their bodily appearance/function more ‘normal’ in comparison to those with more severe disease and, therefore, feel less need to risk disclosure of their condition and the supposed consequences.”

Minor Essential Revisions:

5) The measures section of the paper is incomplete. It should include all measures that are described in the results. This includes demographic information, disease severity questions, and questions related to topics discussed with potential partners. We have better clarified the nature of our 55 item survey, which included demographic and disease severity questions, as well as the questions of interest for this manuscript regarding disclosure and challenges faced by participants once disclosure had occurred.

6) The authors note that responses to medical and screening questions were needed to be included in the final dataset. More information on the response rate would be helpful in understanding the sample and its generalizability (e.g., number of respondents overall; percentage of respondents with complete data). We have included that a total of 8% of the US adult CF population based on the CF Foundation registry completed the survey. We have also now correctly noted sample sizes in individual analyses as no one was excluded completely from these analyses. We have deleted that sentence accordingly.

7) Were questions on education, income, and marital status asked of survey respondents? If so, they would be informative in understanding the current
sample and should be included. **We have added information about education and marital status into Table 1.**

8) Figure 2 consists of three separate bar graphs that are difficult to compare. It would be helpful if they were combined into a single graph. The bottom would consist of each comparison group (e.g., relatives) and three bars (mild/moderate/severe airway limitation). This would enable the reader to more easily assess potential differences in disclosure reports. **Figure 2 now has separated out the original figure into several smaller figures based on the person to whom disclosure occurred. We feel that this figure, suggested by the reviewer, better captures the ability to examine differences based on disease severity. Thank you for this suggestion.**

Discretionary Revisions

9) The last sentence in the first paragraph of the introduction should be clarified. It may be mis-interpreted to mean that over 45% of the population in 2007 had CF and not, among those with CF, 45% were aged 18 years and older. **Thank you for pointing this out. We have now changed the text accordingly.**

10) In the Methods section, it would be preferable to change the heading from “Patients” to “Participants” or “Respondents” as this is a community health survey asking about the lives of adults with cystic fibrosis not a clinical study focused on health care treatment or services for patients. **We have changed the heading to “Participants.”**

11) The Procedure section of the methods should come before a description of the Measures. **We have moved the procedure section before the Measures section.**

12) What is the n for the section on “Challenges discussed with partner”? This could be noted in the text and Table 2. **We have added the sample size in both the text and table.**

In sum, we have attempted to address all of the concerns and questions raised by the reviewers and believe these changes have greatly improved the paper. Thank you for the opportunity for resubmission.

Sincerely,

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