Author's response to reviews

Title: Coping, Adaptation, and Perceived Risk to Children: A Survey of Parents Affected with Bipolar Disorder

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Author's response to reviews: see over
Dear Editors:

Thank you for allowing us to respond to these thoughtful reviews. We hope that we have adequately addressed the concerns and suggestions of the reviewers.

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Response to reviewer 1

1. Consider omitting coping from title as it is not a dependent variable.

*Change made, thank you.*

2. Abstract is long, consider condensing e.g. omitting first sentence.

*Abstract is edited.*

3. Results section in abstract could be condensed for brevity. However the addition of the direction of correlation would be informative.

*The abstract includes the positive and negative nature of the associations.*

4. In the results section in the abstract, data can inform interventions for parents with BPD rather than individuals with BPD as it is not clear that results are generalizable in this way.

*This change has been made.*

5. As ‘adaptation’ is only measured by self report it may be better introduced as subjective adaptation. (discretionary)

*All assessments of perceptions can be described as subjective, although this modifier is not used in the published literature, just as “objective” is not used to modify an assessment of a medication level. Social science assesses cognitions and affect that are inherently subjective. In the published literature using the PAS, the construct is described as “adaptation to a condition or risk.”*

6. Last sentence of first paragraph under ‘justification’ is confusing, could be beneficial to clarify what ‘negative factors’ refer to (predictors of symptom remission?). (discretionary)

AND…

7. Second paragraph under ‘justification’, first sentence, contributing factor to
what? Well being or negative symptoms? (discretionary)

These two paragraphs have been edited.

8. On page 6 it would be helpful to know the working definition of ‘well-being’ in the study and how this might differ from ‘quality of life’. (discretionary).

Well-being here is used to mean psychological well-being that can be represented by several different assessments, including quality of life. We clarified “psychological well-being” and defined psychological well-being and quality of life.

9. It would be helpful to elucidate whether coping and adaptation are entirely separate variables, given that it is mentioned that aspects of coping are measured when measuring adaptation using self-report. (minor essential revision)

Whether variables were tested as independent or dependent variables originates from theoretical and empirical work based on the Transactional Model of Stress and Coping. Coping (successful or not) is the way we manage stress from illness or risk and when it is effective, this facilitates adaptation. These are distinct concepts and as such are assessed using scales with items that distinguish them. The TMSC model has been used extensively in the chronic illness literature and proves a robust justification for hypotheses that test the relationships among the variables.

The PAS measures adaptation in four domains, one of which is coping efficacy. This is an assessment of whether coping “worked”. When it does, you are better adapted. This reflects how the concept has been defined and is assessed. Coping and adaptation are separate constructs that are most often significantly correlated.

10. It is helpful that the final paragraph in the background section sets out the scope of the investigation. However, it appears that coping and adaptation are set out as dependant variables when in the method section coping is given as a predictor variable. Also, I wondered if there had been a directional hypothesis on the effect of ‘living with’ BPD on the dependent/outcome variables (minor essential revision)

The aims and hypotheses have been clarified.

11. On page 8, it is helpful that measures used are referenced, however it would be helpful to be clear about whether satisfactory validity and reliability have been previously established in the references given, as well as the info given about reliability in the current sample. (discretionary)

While this would be helpful to include, we deleted this information from the manuscript based upon need to manage the length.

12. Later it is mentioned that some measures have not been validated for use in studies using a population with mental illness. It would be helpful to clarify this here, in relation to each measure. (discretionary)

We now specify which measures have not been validated in a population with
mental illness.

13. Why are two measures of illness perceptions used? (minor compulsory revision)

We clarified the terminology describing the measures as they assess distinct aspects of illness perceptions. The Brief IPQ measures illness severity, which is an independent variable. We also included questions on perceived current mania and depressive symptoms, and confidence in diagnosis as distinct illness appraisals—these were analyzed as confounder variables.

14. I was left unclear about whether a correction for multiple analyses had been applied. (discretionary)

No correction was made as is customary for social science studies of this size.

15. In results/discussion section I was unclear whether it was a new finding that coping tended to be active/social OR blame/denial in character. This is interesting and maybe worthy of more discussion. (discretionary)

This is not a novel finding (and in fact was an expected finding), and a statement to this effect was added to the results section.

16. In limitations section there is no comment about whether the sample was representative of the population of bipolar parents. Sampling bias may be operating as there is a preponderance of mother rather than fathers, despite an more equal gender split being found in bipolar disorder (Kendell and Zealley, 1993). A lack of racial diversity also. (major compulsory revision)

We have added several additional statements to the limitations section that includes a statement about the lack of diversity and preponderance of mothers in our sample. The limitations section already includes a statement about the likely lack of generalizability to all bipolar parents.

17. Table 3. If there were information about clinical ranges for scores or scores expected in normal population, this would help the reader interpret this information. (discretionary)

Assessments of psychosocial perceptions do not have clinical “cut-offs,” like scales of anxiety or depressive symptoms. They are assessments of cognitions, affect and behavior pertaining to health and not assessments of mental health states where a cut off is used to discern a clinically meaningful finding.

18. In discussion it would be interesting to hear consideration of why particular variables were predictors rather than outcomes e.g. could coping also be construed as an outcome? (discretionary)

See response to 9.

19. I would be interested in the author’s thoughts about whether dispositional optimism is related to the concept of attributional style and the associated literature? (discretionary)
Yes, optimistic individuals are more likely to accept attributions that they do not control than are less optimistic individuals.

20. In the discussion or conclusions section, it would be helpful to know more about how genetic counseling could be useful or is indicated. Would the authors agree that risk perceptions evident in this sample are realistic and show a strength in the appraisals in this area? (discretionary)

We did not attempt to define a “reasonable” risk perception in this population. The risk question was qualitative, asking participants to compare their child’s risk to the general population risk. Based on past research and clinical experience, we are not surprised that the population identified the risk to their children as increased over the population level, which based on empiric data is accurate. We describe responses to an open-ended question that give some support for rational risk attribution. We have clarified the language about the usefulness of genetic counseling in the manuscript.

Response to reviewer 2

Major Compulsory Revisions

(1) The question posed by the authors is not clear. The only hypothesis stated is at the end of the introduction, “greater adaptation would be correlated with lower perceived risk to children”. Is this the study’s primary aim? This implies that adaptation is the predictor; however, this is listed under outcomes in the methods. The authors should clearly articulate in numerical form what their primary aims are and then address those aims in a systematic way throughout the manuscript. If the latter is the primary aim, the rationale for this is not sufficiently explained in the introduction.

We have revised the aims and hypotheses and mapped the analytic approach and results to each question.

(2) One qualitative study is described indicating that parent’s degree of concern for their children was related to their own subjective perception of their own well being. How was parent’s degree of concern measured in this study?

The study referenced was a qualitative interview study; themes related to degree of concern emerged from the interviews, but no direct measure was used. Exploratory studies such as this one are commonly used to identify themes to be studied in quantitative follow-up survey studies.

(3) Parent degree of concern is not the same as parent’s perception of genetic transmission to their children.

We agree, and this is the reason for including genetic and environmental aspects to the measure of the familial recurrence of bipolar disorder, as well as an open-ended question about causation.

(4) The rationale for including this type of measure needs to be explained, or I would consider omitting this from the manuscript. I am unaware of any literature to suggest that parent’s perception of genetic transmission of mood
disorder to their children is associated with their own well being, nor is there any described here, and I have concerns regarding the soundness of this hypothesis. I am not surprised these variables were not correlated.

*Descriptive data arose from an exploratory study of bipolar parents that suggested the hypothesis that parental wellbeing is related to perceived risk to children. We revised the introduction and aims related to parental perception of risk.*

The high genetic loading of bipolar disorder is well established in both the scientific community and general public. I would presume most parents with bipolar disorder would adequately understand that their children are at an increased risk of developing mood disorders compared to the general population. If this was not the case, this may reflect the education of the parent, which is not measured in this study or reported at least. Major revisions need to be made to the introduction, rational and objective of this manuscript. Once a clear objective is described, the title of the manuscript should be revised to reflect this.

*The research hypotheses and aims have been clarified and the title revised. Significant revisions have been made to the introduction and the reporting of the risk perception data.*

(2) The discussion of this manuscript needs to be revised in the following areas:

a. The primary aim of this paper is not articulated as to identify interventional targets for bipolar disorder, nor is it placed to do so. Please omit this from the first line of the conclusions. This research has the potential to inform more research on potential targets for intervention.

*This suggested edit has been made.*

b. The data from this study does not suggest that interventions targeted at enhancing coping may improve adaption. Please revise, as this is over interpreting the results. This study did not examine the impact of any interventions. There are other areas where this is described. Please revise accordingly.

*Findings from this study indicate a positive relationship between effective coping and more positive adaptation. That suggests that interventions aimed at enhancing coping may facilitate adaptation. We modified to language to highlight the need for research on interventions targeted at enhancing coping to improve adaption.*

c. It is not clear why the authors are suggesting the following: “One may speculate that dispositional pessimism is not only a risk factor for mental illness, but is perhaps a manifestation of the disorder”. The finding that dispositional optimism is negatively correlated with depressive symptoms does not suggest the latter statement. This is over interpreting the results. *We removed this statement.*
(3) The limitations of this study are not adequately described.

a. The authors comment on the self-referral nature of this study and how this limits the generalizability of their sample, however, the reasons for this are not adequately described. There is a major concern of selection bias here, not only because subjects may be interested in the research question (a special population), but because there is a high possibility that the sample may not even have bipolar disorder. In addition, the authors present no information on education or socio-economic status (SES) of the parents or any other comorbid diagnoses which has large implications for family functioning and parents psychosocial functioning. If education or variables of SES are not available, the authors need to address the potential impact this has on their findings and how it relates to external validity.

While we have no way to confirm that the respondents have bipolar disorder, the respondents perceive themselves to be affected. Thousands of surveys of disease populations are conducted in social science annually in the US, funded by the federal government. Studying perceptions of living with disease contributes to understanding about the lived experience of the condition beyond objective assessments of disease, which reveal little about how people cope. There is no a priori reason to expect that individuals without bipolar disorder would mis-represent themselves as affected and take the survey, just as there would be no reason to expect that an individual who does not have type I diabetes would access and choose to take a survey on the effects of type I diabetes. We added additional statements in the limitations section of the survey to address this concern and to confirm that we do not have data on SES/education level.

(5) Is there any information on the location of the subjects (i.e., Country, State, Province)? Please include if it is available. If this information is not available, the authors need to comment on this in the limitations section.

Data about state has been added to the results and the table of sample characteristics by reporting the total number of states represented from across the United States.

(5) The scores on the scales are difficult to interpret as there are no cut off scores reported. If there are cut off scores available, report these in the measures section. Are there any other comparisons of adaptation scores rather than Klinefelter syndrome? This is not a helpful comparison as the symptom presentation and etiology of these two disorders are very different despite a shared high genetic loading.

The PAS mean adaptation score was lower than found in eight studies of chronic illnesses where it has ranged from 2.66 to 4.23 (SD ranged from 0.6 to 1.2). Alpha scores of reliability have ranged from 0.83 to 0.97. There are no normative scores on this scale. By definition, if you are adapting to something, you have a condition or risk so there are not norms for the general population. Comparisons can only be made to values from other chronic disease populations. We removed the specific reference to Klinefelter syndrome and
added the broader range of means.

(6) Were there any associations between the demographic variables and perceived risk to children or adaptation? If the authors have these data, they should report this as the demographic information also has important treatment implications (e.g., racial differences).

We added a clarification that none of the demographic data collected showed any correlation with perceived risk to children and no demographic confounders were significant in the adaptation regression.

Minor Essential Revisions

1) The tables are poorly formatted and difficult to interpret. Please add in the unit of analysis in Table 1 and justify all tables so the alignment is straight. It is unnecessary to describe the measures in Table 3. Please remove this information and replace with the measure name. Measure descriptive information

The tables have been edited and re-formatted. Thank you for this suggestion.
RESPONSE TO REVIEWER 3

This manuscript would be improved with more concise writing – for example the Background section is not focused specifically on the research question and it is not clear when reading through it where it is heading.\textit{We have revised the background section to be more concise and targeted.}

The methods are weak given that a selection bias (those who want to fill out the survey) is in operation likely. Also you have no way of confirming that the respondents have the disorder you are studying. There is essentially no objective measures and no detail regarding the clinical course which would directly influence the perceptions of coping and adaptation (ie psychotic features? Quality of remission? ).

\textit{As with all opt-in studies, especially online surveys, we expect selection bias for those most interested in participating in research, telling others about their condition and having the cognitive capacity to access and complete the survey. This bias is stated as a limitation. No assumption is made that we recruited a generalizable sample of individuals with bipolar disease. Those who self-select are often those affected by a condition who can best articulate what the disease experience is like and how it has affected them. The way we make it more objective is to frame the analyses on accepted theoretical models that predict relationships between the independent variables, giving rise to testable hypotheses.}

\textit{While we have no way to confirm that the respondents have bipolar disorder, the sample perceive themselves to be affected. Thousands of surveys of disease populations are conducted in social science annually in the US, funded by the federal government. Studying perceptions of living with disease contributes to understanding about the lived experience of the condition beyond objective assessments of disease, which reveal little about how people cope. There is no a priori reason to expect that individuals without bipolar disorder would mis-represent themselves as affected and take the survey, just as there would be no reason to expect that an individual who does not have type I diabetes would access and choose to take a survey on the effects of type I diabetes. We added additional statements in the limitations section of the survey to address this concern.}

Specific comments:

Background:

A recent meta-analysis estimated that the heritability of bipolar disorder is estimated at 85% (Bienvenu et al.,). Furthermore there are more relevant references to show that the birthrate in families with a parent with bipolar disorder is close to that of the general population.

\textit{We updated the reference on the heritability of bipolar disorder, and removed the section describing the birthrate to streamline the background section.}

The research question was not succinctly asked in such a way that it directly mapped on to an analytic approach.
We have revised the research questions and mapped the analytic approach and results to each question.

Methods:

There should be much more detail collected to characterize the illness course and current clinical state for interpretation.

We did not collect data about the illness course or current clinical state, other than the questions about current manic or depressive symptoms, time since diagnosis, and confidence in diagnosis.

Statistical Analysis:

The mediation hypothesis should be explicitly stated.

This has been added.

Furthermore, age of the offspring would likely affect the perception of risk (ie if younger increased, if older and well decreased).

The age of the offspring do not affect the parental perception of risk. This has been clarified in the results section. The inclusion criteria required a child of 30 years or less. The cross-sectional nature of the survey and qualitative formulation of the risk question do not lend themselves to determination of the longitudinal assessment of whether perceived risk decreases with child's age.

Furthermore, the nature of the parent illness would affect the perception and the lack of detail on this limit any analysis or conclusions.

While this is true, the sample is sufficiently large to capture a variety of expression of bipolar with the stated bias that respondents were sufficiently capacitated to complete the survey. While individual illness course may be reflected in responses, the sample represents a scope of experiences.

Results:

The majority of patients endorsed a perceived genetic/familial aspect to their illness and the vast majority (87%) endorsed a greater perceived risk to children − therefore the main finding seems hat patients are knowledgeable about the heritability of this disorder. Also with little variation in perceived risk it is curious how this can have a differential affect on coping.

Conclusions:

The main conclusion here seems to be that patients (assuming they have bipolar disorder) seem to be well aware of the familial/genetic component and those that endorse better adaptation, tend to endorse social coping and a positive outlook − however, this is not really new and not really informative regarding the important and interesting psychological associations between parents and children.

For clinicians, these data are indeed not surprising as they reinforce clinical experience. However, there is little published data about coping with bipolar as a
whole (i.e., distinct from studies of coping with specific states or symptoms, such as mania or hypomania) or psychological wellbeing, unrelated to illness manifestations or predictors of remission, for individuals with bipolar disorder. We agree that there are interesting psychological associations between parents and children. In this study, we also measured parental coping with perceived risk to children; that data is reported in a separate manuscript.