Author's response to reviews

Title: Exploration of knowledge and understanding in patients with primary adrenal insufficiency: A mixed methods study.

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Version: 2 Date: 05 Jul 2017

Author’s response to reviews:

Dear Dr. Shipley,

Thank you very much for giving us the opportunity to revise our manuscript. We would also like to thank the reviewers for their helpful comments and suggestions. We have now addressed all the points raised by the reviewers and amended the manuscript accordingly. Please see our point-by-point responses below. All changes to the manuscript are track-changed except changes to references.

The reviewers comments have improved our manuscript significantly and we do hope that you find our paper is acceptable for BMC Endocrine Disorders.

Kind regards

Ms. Lisa Shepherd

On behalf of all authors

Reviewer reports:

Birgit Harbeck (Reviewer 1): Please include all comments for the authors in this box rather than uploading your report as an attachment. Please only upload as attachments annotated versions of
manuscripts, graphs, supporting materials or other aspects of your report which cannot be included in a text format.

Thank you, this has been noted.

I have no further comments, all points of criticism were taken up.

Kathleen E. Bethin (Reviewer 3): The authors have made significant improvements to the manuscript. However, there are several issues that still need to be resolved.

Table 4 is mislabeled and cited. It should now be Table 3.

Thank you, the table has now been relabelled as Table 3 and corresponding text amended to cite table 3.

Lines 55-57 need to be rewritten the grammar is incorrect-

'Whilst those who have experienced a previous adrenal crisis are at greater risk of a subsequent episode [6,7]. The occurrence of adrenal crisis in PAI has been reported as 5.2-8.3 crisis/100 patient years [6,7,8].'

Thank you, this has now been amended to read.

‘The occurrence of adrenal crisis in PAI has been reported as 5.2-8.3 crisis/100 patient years [6,7,8]. About 55% of women and 52% of men reported one or more adrenal crises since diagnosis.[7]. Whilst those who have experienced a previous adrenal crisis are at greater risk of a subsequent episode [6,7].’

The authors need to provide a rationale for aiming for 15 subjects- citing a reference is not sufficient.

Thank you, the rationale for aiming for 15 participants has now been given and the sample/participants section now reads as follows.

‘Pragmatic purposive sampling was adopted from two different demographic location hospitals in a single tertiary NHS Trust in UK. It was planned to recruit up to 15 participants, or until data saturation was achieved [12,13]. It has been posited by Guest et al.(2006) that data saturation occurs by the time 12 interviews have been analysed [13]. Therefore the planned recruitment number allowed for attrition and allowed for further data to be gathered until data saturation was achieved. However, data saturation occurred when ten patients had participated in the study.’

The authors need to clarify how many different providers were involved in educating the patients and how many different providers failed to document education of the patient in the records. Since one of the main conclusions of the manuscript is that providers are not documenting this is crucial. It would also be important to note if there is a difference in documentation between physicians and nurses.
Thank you, most patients were seen by several health care professionals from the endocrinology team during their care. It is not known how many healthcare providers were involved in educating the patients as this information was not captured during the study and this has now been added as a limitation of the study.

The following comment has also been added for clarification to the ‘documentation of the education provided to patient’s’ section as below;

'Documentation of the education provided to patients

This study showed that documentation in the medical notes regarding the education provided to patients with PAI was ‘good but detail inadequate’.

The authors statement on lines 223-225 is an over interpretation of their data."However, length of time since diagnosis was not related to increased knowledge as demonstrated in the verbatim quote from participant 8." The quote from participant 8 is "...the only thing I've ever been told is if you're abroad on holiday and you have stomach problems you know, if you have diarrhoea or something like that, then take an additional tablet but that's as much as I've been told..."(P8)." This quote does not provide data to support the authors statement on lines 223-225.

Thank you, a further verbatim quote to support this statement has been added to this section.

“No, no, not at all. Funnily enough I did not realise you could, you should or you needed to, to be honest”(P8)

The section starting on line 282 needs to be re-written. This sections starts out "It was apparent that there were three support systems, family provided substantial support when it came to the management of the participants' condition and also when seeking a diagnosis for the condition." The three support systems need to be listed and then the authors can describe the 3.

Thank you, this section has been re-written and the paragraph now reads as follows.

'Three support systems; family, healthcare professionals and voluntary were identified as important to and relied upon by the participants. Family provided substantial support when it came to the management of the participants' condition and also when seeking a diagnosis for the condition. The diagnosis was often only established after perseverance from family in seeking medical help. Whilst two participants felt their family should have a better understanding of the condition, most participants relied upon the opinion and assertiveness of family during intercurrent illnesses. This was related to the administration of emergency treatment when seeking urgent medical attention.'

Data needs to be provided to support the authors statement that "Findings from the present study confirm that participants received many differential diagnoses."

Thank you, the following quotes have now been inserted to support this statement and this section now reads as follows.
‘All of the respondents described the length of time to diagnosis as a problem, receiving many differential diagnoses, despite seeking medical attention on several occasions prior to diagnosis. This is demonstrated in the below verbatim quotes.

“…I used to go the football…and half way through the first half I had to sit down. If I used to go the pub I used to have to sit down… I was too tired to stand up.” (P5)

“I just kept looking at them thinking why are my hands brown? Why are these brown on me?” (P6)

“The Doctor did a glandular fever test which was clear and then he decided I was depressed.” (P1)

“…they kept saying it was the effect of septrin or was I pregnant or had I got anorexia (laughter) and all sorts of different things…”(P7)’

They also need to provide data to support Finally, there also appears to be inconsistency in terms of health professional knowledge, and advice given.

Thank you, this statement is supported by a quote from P6 in the section on ’stop taking their steroids’ as below,

‘Worryingly two participants had been advised to stop taking their steroids either by a healthcare professional or friend at some point since their diagnosis.

“…I couldn’t hold anything down, she [G.P] said don’t worry it won’t hurt you to miss them for once cause you’ve got bits of steroid going round your body… but she shouldn’t have told me that.” (P6)’

A further quote to support this has been inserted in the section adjust medication to now read as below,

‘Respondents informed HCPs of their condition, and also relied upon them for advice and extra medication during dental and surgical procedures. They saw the HCP as the ‘expert’ and were guided by their recommendation, even if they knew this to be incorrect.

“Yes I did have to tell them…when they knew I got Addison’s disease they said that I would have to go over night ‘cause I would have to have steroids before I went into surgery. I’d have to stay in that extra bit longer probably just as extra day to boost me up…” (P9)

“I think it depends on which Doctor you see as well. Like when I’ve been to clinic and I like to see one Doctor now rather than each time I come to clinic to see a different Doctor and they tell you different things and I don’t think they know that much about it…”(P7)

Lines 578-580 do not make sense- "It has been found that glucocorticoid replacement impacts on patients self-perceived outcomes, such as physical activity or family life"
Thank you, this section has now been amended to read as follows,

‘The present study describes the initial disbelief and shock participants feel when receiving the diagnosis. They begin to establish ways of dealing with the diagnosis and treatment regimens and incorporate this into their lives. Forss et al. (2012) also found that glucocorticoid replacement regimens impacted on patients’ lives and their self-perceived outcomes, such as physical activity or family life [34].