Reviewer’s report

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

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Reviewer: Per Dahlqvist

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The paper describes a qualitative study on semi-structured interviews of 10 PAI-patients from a single tertiary UK centre. The research question on PAI-patients' knowledge and understanding of their disease and application of this is important. The authors conclude that the patients experience a transitional journey of adapting to the diagnosis and that patients do not always apply the knowledge of their disease in intercurrent illness.

Major comments:

- The study's qualitative approach to the research question may lead to new knowledge, but should be interpreted with caution and as hypothesis generating especially considering the very limited number of patient interviews from a single centre. This considerably hamper the generalizability of the results, which must be more clearly stated in the discussion, conclusion and abstract. Furthermore, the paper is very long, especially the discussion which needs to be condensed and more focused on the data collected in the study.

- Line 83-85: The authors planned to recruit 15 participants or "until data saturation was achieved", but present data from 10 patients. This is confusing and the authors should 1) present the scientific basis for the figure 15 and 2) present why the data collection stopped at 10 patients. Why was this figure considered sufficient? Is some kind of power analysis possible in a qualitative study?

Minor comments:

Background:

- Line 48-49: The authors state in the background that "A stressful event also includes trauma and psychological distress.". As far as I know there is no consensus or evidence to support that psychological distress causes a "...need to increase their GC dose...". To justify this statement the authors refer to the study by White and Arlt, in which 1% of the 767 patients self-reported that "anxiety/psychological upset/distress" was the factor leading to an unplanned emergency hydrocortisone injection. The statement on psychological distress as a "need to increase GC" should be removed unless considerably more support from the literature can be found and referred to.
- Line 66-67: The authors refer to the study by Hahner et al [9] on educated AI patients (including 221 PAI-patients) and then state that "data on PAI is lacking" - this is very strange and needs to be rephrased. Furthermore, I suggest moving the sentence on line 63-64 to be the final sentence of the section since this better conclude the background and ties on to the aim.

Methods:

- Table 1: #2 - the word "shock" may be interpreted in two very different ways in this setting and is confusing. Do the authors mean circulatory shock (i.e. the general medical meaning of shock), highly relevant to this patient group or psychological stress, which may be considered a reason to increase GC dose? This need to be clarified both regarding the way the word was used in the patient interviews and which meaning is considered in the manuscript.

Results:

The results section is very long with several example citations of the patients' answers. The section should be condensed.

- Line 146: Two male and eight female patients were interviewed. This is a very limited number and also does not represent the gender distribution of PAI (M-40/F-60). This needs to be adressed at least as a limitation in the discussion.

- Table 5: The authors clearly describe several important participant demographic, which is very important in such a small study. Since the aim is seek to learn more about patients knowledge, their own collection of information on AI and application of their knowledge it is reasonable to believe that the patients' level of education at the time of the interview may be an important factor. This should be included in table 5.

- Line 198: Here it is stated that time since diagnosis was not associated with increased knowledge. This is a quantitative statement and the authors should present data to support it.

The interviewed patients' age and time since diagnosis is highly variable, which may be considered a strength of the study making the data more generalizable. It would be interesting if the authors tried to find more associations between interview data and age/time since diagnosis.

- Line 313-354: In the collection and interpretation of data from medical records some discrepancies were noted versus patient interview data but it is stated that overall documentation of sick day rules and steroid card was good. It is not clear what scientific question the authors wish to explore in this section. Please clarify the aim of the medical record part of the study.

Discussion:
The discussion on the "journey" of the patient is interesting and focused on the data. Other parts of the discussion is very long and speculative and should be condensed considerably and focused on the aims and data of the present study.

- Line 358: This sentence needs to be rephrased to emphasize that "The study revealed that a majority of the interviewed patients with PAI at this tertiary UK centre..."

- Line 370-374: The authors state that the present study "discovered" that also family, HCP and voluntary require knowledge of the condition and therapy including dose adjustments. This would by most professionals in medicine/endocrinology be considered common knowledge which can hardly be claimed to have been "discovered" in this study, possibly "confirmed" or "showed". Please rephrase.

- Line 462 (and later): This section on medical records is very long and may be condensed. The authors conclude several times that documentation of the education in the medical records requires improvement and standardization. However, it is not explicit what the patients would gain from this. Do the authors advocate that "info about sick-rules", "steroid card", "jewelry" and "injection kit" is always documented in the medical records and hypothesize that this may enhance the patients' education and understanding of their condition? This may very well be true, but if this is the authors case the authors should present this hypothesis more clearly and preferably some literature/data to support it. Regarding generalizability of these findings it is likely that traditions and legislations regarding medical documentation/records vary significantly between different countries. This is likely to further hamper generalizability of these findings, which should be highlighted in the discussion.

Conclusion:

Line 564: The final conclusion of the study is that "...there is a need for HCP to be educated and promote diagnosis and therapy". This is obviously important, but really way to obvious and a matter of course to be considered a conclusion of this study. If the authors which to keep this sentence in the paper it needs to be moved from the conclusions section.

Are the methods appropriate and well described?
If not, please specify what is required in your comments to the authors.

Unable to assess

Does the work include the necessary controls?
If not, please specify which controls are required in your comments to the authors.

Unable to assess

Are the conclusions drawn adequately supported by the data shown?
If not, please explain in your comments to the authors.
Yes

Are you able to assess any statistics in the manuscript or would you recommend an additional statistical review?
If an additional statistical review is recommended, please specify what aspects require further assessment in your comments to the editors.

Not relevant to this manuscript

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Please indicate the quality of language in the manuscript:

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