Author's response to reviews

Title: "I am pregnant and my husband has diabetes. Is there a risk for the foetus?" A qualitative study of questions asked by email about the role of genetic susceptibility to diabetes

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Author's response to reviews: see over
Amsterdam, September 23rd 2010

Dear members of the Editorial Team,

We would like to thank you for the opportunity to revise our manuscript, originally entitled “Five persons in my family have diabetes. Does this have to do with inheritance?” A qualitative study of patients and family members searching for online information about the role of inheritance in diabetes”.

The comments of the reviewers have been very helpful in revising the manuscript and we believe it has substantially improved. Please find below the response to each of the comments.

There is one specific issue we would like to present to the Editorial Team. Reviewer 1 advised to opt for another quotation in the title for better reference to the results of the study. Therefore, we suggest a revised title:

“I am pregnant and my husband has diabetes. Is there a risk for the foetus?”
A qualitative study of questions asked by email about the role of genetic susceptibility to diabetes.

However, we will leave it to the decision of the Editorial Team whether a quotation in the title is preferred. As an alternative title, we suggest:

Patients and family members inquiring the role of genetic susceptibility to diabetes. A qualitative study of questions asked by email.

We are looking forward to receive the final editorial decision.

On behalf of the authors,

Sincerely yours,

Mrs. Suzanne C.M. van Esch, MA

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REVIEWER’S COMMENTS

Reviewer: David Whitford

Reviewer’s report:
Thank you for the opportunity to review this paper. I found the use of content analysis of an otherwise unexplored database of interest. However, the major problem that I had with this paper was that it failed to address a clearly enunciated research question and therefore lacked a clear focus in the discussion. I think the authors should rethink what question they are really answering and discuss the results in relation to that question.

We thank Prof. David Whitford for his adequate review and useful comments. Please find below the response to each of the comments.

Major Compulsory Revisions
1. The research question needs to be more clearly defined. The aim of the study is expressed as exploring the information needs of patients and family members searching for online information about the role of inheritance in diabetes. In fact, this is not what is described because the internet users had already accessed information from the website. What is explored are the areas of uncertainty or the difficult to understand concepts or the unaddressed information needs of users. This needs to be made more explicit in the research question.

Similarly the abstract needs to reflect an adapted research question.

It is correctly noticed that we only indirectly got insight in the information needs of people searching for online information about the role of inheritance in diabetes. Assuming visitors read the provided information on the website, indeed the emailed questions address areas of uncertainty, difficult to understand concepts or undressed information. However, due to limitations of using secondary data, we do not know whether people actually did read the provided information. Some questions suggest they did not, and in that case the question reflects people’s initial information need on diabetes and inheritance.

We are thankful for this comment that gave us the opportunity to be more specific in our description of the available data. Throughout the whole manuscript, the utilisation of secondary data in this study is pronounced. We rephrased the research question, but we did not change its focus dramatically (see also our comments under point 6):

“In the Netherlands, the National Genetic Research and Information Center provides online information about the genetics of diabetes (all subtypes). Thereby, the Center offers website visitors the opportunity to ask a question per email. We used this data base with emailed questions to gain insight into people’s need of (additional) information about the role of inheritance in diabetes. After all, depending on whether or not visitors read the provided information, the emailed questions reflect visitors information needs, unaddressed issues, areas of uncertainty or difficult to understand concepts. Our research interest is in developing a profile of questioners, as well as the discovery of themes and tendencies in the emailed questions. Findings from this study will contribute to better understanding of specific information needs of online consumers about genetics and
diabetes. The results may help to tailor existing clinical and public (online) health information to the needs of an increasing population at risk for diabetes.”

Accordingly, the background information (including the research question) in the abstract is revised:

“Diabetes Mellitus is a global health problem. Personalised genomics seem promising in enabling individualised treatment and primary prevention. In the Netherlands, the National Genetic Research and Information Center provides online information about the genetics of diabetes and thereby offers website visitors the opportunity to ask a question per email. The current study aims at exploring people’s need of (additional) information about the role of inheritance in diabetes. Results may help to tailor existing clinical and public (online) health information to the needs of an increasing population at risk for diabetes.”

2. The title does not reflect the results of the study. The results of the study suggest people are not asking about inheritance in general but about risk to self or offspring – this is different to what the title suggests.

As we formulated the title, we searched for an appealing quote. The initial quote “Five persons in my family have diabetes. Does this have to do with inheritance?” refers to 37.2% of the questions concerning ‘inheritance in general’.

The reviewer is right that with this quote we ignored the study’s main results (indeed, most emailed questions refer to inheritance in relation to reproductive choices and worry about offspring’s health). Therefore, we suggest a revised title:

“I am pregnant and my husband has diabetes. Is there a risk for the foetus?”
A qualitative study of questions asked by email about the role of genetic susceptibility to diabetes.

However, as we indicated in the letter to the Editorial Team, we will leave it to their decision whether a quotation in the title is preferred. As an alternative title, without quotation, we suggest:

Patients and family members inquiring the role of genetic susceptibility to diabetes. A qualitative study of questions asked by email.

3. The methods need to be clarified in a few areas. The use of content analysis in this context is interesting. Classical content analysis would assume that the codes of interest have already been discovered and described but in this case it appears that they were identified by a look at the data. Is this the case? In addition, classical content analysis would usually allow independent coding by multiple coders and then examine the degree of inter-observer variation through use of a Kappa statistic rather than discussion within the research team. Was this done and what were the results? This would add to the reliability and external validity of the results. It may seem pedantic but the authors are describing thematic content analysis rather than a pure classical content analysis.
The reviewer’s observation is right: in our analysis, themes and tendencies of interest emerged from the data. We did not use classical content analysis and therefore a Kappa statistic was not calculated. Data are analysed following the Grounded Theory and inter-coder agreement was reached by discussion in the research team. Additional literature research learned that this approach is applicable in computer-mediated convenience samples. We extensively revised the Method section (thereby also considering the comments of reviewer 2) and described the process of data analysis as follows:

“[…] In this study, we adopted an iterative and inductive approach which is argued to be applicable in computer-mediated convenience samples[23]. Two researchers (SvE and research assistant) double-coded all email questions using qualitative data indexing software (Kwalitan 5.0[24]). The labelled data were analysed in line with the Grounded theory approach, which means that emerging themes and tendencies were identified and categorised[25]. Ambiguities were resolved and categories were reduced to major themes in discussion with two senior researchers and re-reading the emails[26]. The researchers reckon with the fact that secondary content analysis may impede the guidelines of Grounded Theory regarding ongoing data generation and the saturation principle[27].

After qualitative classification, data were quantified in order to develop participants’ profile (by age, gender and family status) and observe the distribution of coding labels within the emerged categories (‘type of diabetes inquired’, ‘topics inquired’, ‘expressed worry’, and ‘type of information requested’). Illustrative quotes are presented as summaries of the questions’ quintessence, paraphrasing the original Dutch formulation as much as possible. Participants’ identification number (#), gender, age in years, and family status are included after each quote to help the reader identify the background of its source.”

4. I would find it useful to have some indication of what is meant by some terms of degree in the results e.g. 'mainly' women - is that 51% or 90% - I know it is in the table but it is difficult referring back and forth all the time. The use of qualitative quotes is useful.

We agree that the used indications like ‘most’ and ‘mainly’ are open to multiple interpretation. We added percentages in the text and refer to tables at the beginning of each Results section. For example:

“As shown in Table 1, it appeared that most people asking questions via the website were relatively young; 54.8% was ≤30 years. Mainly women (82.7%) inquired the role of inheritance in diabetes. Almost half of the questions were asked by diabetes patients; 29.6% by relatives and the remaining by partners of diabetes patients.”

5. The discussion on limitations of the study and generalisability needs to be expanded. What are the implications that a younger, female group of internet users have asked these questions and that they are more about risk to offspring than personal risk or risk to other family members.
In the Discussion section, we indicate that our finding (young women in need of genetic information, because they worry about offspring’s health) resembles other research outcomes.

“[…] This is in line with previous research indicating that younger Internet users and women are most likely to search for online genetic information[28, 29]. Yet, the online population is expanding and becoming more representative in terms of race, age, income, and educational attainment[30, 31].”

And:

“Finally, data reveal that the majority of queries concern topics related to (future) pregnancy and family planning. This finding is in line with the relatively young age of questioners and overrepresentation of women in our study. It is known that the phase of reproduction generates an active search for genetic information[28, 39]. Women have been found to search for genetic information, because they worry about the health of their (future) offspring[40]. These results resemble our finding that in most queries worry about the diabetes related health of (future) offspring is expressed.”

We believe these results not merely imply a limitation of the study, but rather an outcome to take into account when utilising genetic information in (online) public health initiatives.

However, we acknowledge the reviewer’s concern about the study’s generalizability and added to the Limitations section:

“We are aware that people submitting e-mail questions via the Internet may represent a selective group (in our study young, predominantly female Internet users) and we cannot exclude selection bias.”

The issue that questioners worry more about risk to offspring than personal risk is addressed in the concluding remarks:

“Considering the high prevalence of T2DM and GDM, more effort seems needed to explain the multifactorial aetiology (and with it the risk of familial clustering). Opportunities to delay or prevent T2DM and GDM onset by adopting a healthy lifestyle[43, 44] should be emphasised. To optimise health behaviour, these efforts should take public perceptions about inherited predisposition and primary prevention into account[45, 46].”

6. The discussion should be based more around the revised research question. 1) Why are users of the site asking these questions? 2) Are these questions more difficult to understand from the website? 3) What conclusions do the authors draw about the utility of the email approach? 4) What do these questions contribute to the explanation of genetic risk?

To answer the first two questions, content analysis of the provided information on the website would be required. We looked at the web content, but decided not to include a systematic content analysis into this study. After all, the main deficiency of our study sample (inherently to secondary analysis) is that it does not reveal whether visitors actually did read the provided information. For that matter, findings would not be reliable.
We inform the reader about this limitation in the Discussion section:

“However by utilising secondary data analysis, we were unable to further expand our understanding by posing additional questions for example related to the amount of visitors actually reading the information provided, the degree of understanding and perceived utility of the expert answers received.”

To address the reviewer’s 3rd and 4th question, we added a paragraph in the Discussion section in which the email approach is addressed:

“In addition of public information, individuals may wish to receive personalised (risk) information or advice. Utilising an email approach often requires more detailed information from the person than currently provided. As an alternative, clinicians and public health providers could compile a list of frequently asked questions (and answers) about diabetes and inheritance and incorporate it into (web based) diabetes family education.”

Discretionary Revisions
Improvements to areas of grammar e.g. change the passive 'Understanding perceptions, ideas and concerns is required to develop education and improve health information meeting the needs and knowledge of the public' to the active "In order to meet the health information needs of the public, health professionals must understand their perceptions, ideas and concerns.'

The manuscript is checked on correct use of English language.
Reviewer: Susan Sullivan-Bolyai

We are grateful to Dr. Susan Sullivan-Bolyai for her feedback on our manuscripts. Hereafter we will respond to each of the comments.

Reviewer's report:
A) Background section should include ROL regarding how often people retrieve/use internet to answer medical-related questions for themselves or loved ones.

We thank the reviewer for pointing at this missing information. We believe the discourse in the Background section has substantially improved. Two paragraphs are added:

“Health consumers have become increasingly interested in genetic information[10, 11]. This increasing interest is coupled with a growing trend in consumer uses of the Internet for health-related purposes. Statistics reveal that eight in ten American internet users (approximately 113 million adults) searched online for health information in 2006[12]. In the Netherlands, 93% of the population has access to Internet. Of all persons using the Internet, 54.0% looked for information about health and medicines at least once in three months in 2009[13].

While consumers recognise great potential in the Internet for health communication on human genetics[14], health professionals and genetic experts acknowledge that the translation of genomic information will be a challenge[15]. Information has to be adjusted to the (genetic) literacy levels of target audiences[16] and has to serve the public's genetic information needs[17]. Since the growing popularity of Internet use, indeed there are a lot of studies evaluating genetic web content[18, 19], readability[20], and effect on behavioural outcomes[10].”

B) Methods: Omitted 2ndary content analysis and briefly its benefits and weaknesses and internet use (pros/cons).

As indicated in reply on the first comment of reviewer 1, throughout the whole manuscript the utilisation of secondary data in this study is pronounced. The pros and cons of secondary content analysis as a data collection and analysis method are addressed. We made clear in the Data analysis section, that:

“The advantage associated with secondary content analysis is its convenience and cost-effectiveness. In other words, it provides the maximum use of data without the need of recruitment, data collection, and participants’ endeavour. In this study, we adopted an iterative and inductive approach which is argued to be applicable in computer-mediated convenience samples[23]. […] The researchers reckon with the fact that secondary content analysis may impede the guidelines of Grounded Theory regarding ongoing data generation and the saturation principle[27].”

Limitations are further described in the Discussion section:
“However by utilising secondary data analysis, we were unable to further expand our understanding by posing additional questions for example related to the amount of visitors actually reading the information provided, the degree of understanding and perceived utility of the expert answers received. On the other hand, it appeared that the available 172 queries generated a study sample that was rich enough to emerge categories reflecting interesting themes and tendencies to describe.”

C) Missing: Explanation of IRB process & ethical considerations esp. with internet use: how did you protect confidentiality of participants etc.

We agree with the reviewer that information about ethical aspects and confidentiality was lacking in the original manuscript. We added information about the IRB process at the end of the Data source section:

“Since January 2005, the National Genetic Research and Information Center has systematically registered personal queries from website visitors. This data base with emailed questions is designed for administrative purposes, as well as monitoring the quality of the web content. For the current study, the Center handed over data concerning ‘diabetes and inheritance’ to the researchers and consented with the research objectives and methods. Given its observational, non-invasive character, the study was deemed not to be subject to approval of the Medical Ethical Committee of the VU University Medical Center. Evidently, subjects’ confidentiality was respected.”

Regulations about the participants’ confidentiality and consent are explained in the Study sample section:

“Data were derived from a sample of 265 e-mailed questions related to diabetes and inheritance (administered between January 2005 and November 2009). The National Genetic Research and Information Center assigned an identification number (#) to each email. The researchers received the emails without name and email address, to protect confidentiality of participants. A list with identifications numbers and corresponding information about questioners’ gender, age, and personal versus professional interest was enclosed.

It is not possible to ask informed consent of participants in secondary analyses, and we therefore excluded all questioners (n=11) that opted not to provide any personal information.”

D) Missing: sample and inclusion/exclusion criteria although you mention it in analysis section (where it doesn’t belong).

The text in the Methods section is reshuffled and refined. Now, in the Study sample section we explained exclusion criteria and data management.

“In addition, two exclusion criteria were applied. Fifty-eight e-mails did not relate to genetics and inheritance, but concerned diabetes (treatment) in general. Secondly, in this study we were primarily interested in (additional) information needs of ‘private’ health
consumers, aiming at tailoring (online) information about diabetes and genetics. Thirty-eight questions were asked by students and health care professionals, and therefore were excluded from the sample. From the 158 e-mails left, fourteen contained two questions. In total, 172 queries were included in this study.”

E) Missing: data management (you have it under analysis; how you cleaned the data and linking it back to inclusion/exclusion criteria.

Data management is described in the ‘study sample’ section. See our comment above.

F) Give n for each result instead of saying 1/5, ½.

We decided to present most results in percentages. Also, indications like ‘most’ or ‘mainly’ are clarified, for instance:

“As shown in Table 1, it appeared that most people asking questions via the website were relatively young; 54.8% was ≤30 years. Mainly women (82.7%) inquired the role of inheritance in diabetes.”

In some sentences, the amount of participants or questions is mentioned:
“Fourteen participants (8.2%) inquired about T1DM as well as T2DM, […]”

G) Alert reader to 'Table 1' at the beginning of results and then do a brief overview of who the participants were.

We referred to corresponding Tables at the beginning of each new paragraph.

“As shown in Table 1, […]”
“Table 2 displays the topics of interest.”

H) I am curious, when someone asks a question, who answers it for them? I would include this information when you are describing the online site.

In the original version of the manuscript, it was already mentioned that:
“Apart from reading the information on the website, visitors are offered the possibility to submit a question per e-mail, that is answered by an expert in the field.”

I) First paragraph in Discussion section belongs in 'Results'.
Accordingly, we deleted the summary of the most important results in the Discussion section.
J) Discussion should compare results to previous empirical literature, is it similar, different and if so why. I found that lacking in this section.

We reflect and elaborate on our results using previous empirical findings. Some additional literature search is conducted. For a better overview, the study outcomes in the Discussion section are described in the same order as the Results are presented.

Since we extensively revised the Discussion section, we would ask the reviewer to assess the complete text in the revised manuscript.

K) I think the most common questions/concerns posed could be incorporated into diabetes family education where the clinician could say 'here is a list of the most common questions asked online'....and then have the patient/family member check off which ones they are concerned about and discuss.

We thank the reviewer for this comment. We used this recommendation as a concluding remark in the section with Practical implications:

“In addition, clinicians and public health providers could compile a list of frequently asked questions (and answers) about diabetes and inheritance and incorporate it into (web based) diabetes family education.”