Author’s response to reviews

Title: Moral dilemmas and conflicts concerning patients in a vegetative state/unresponsive wakefulness syndrome: shared or non shared decision making? A qualitative study of the professional perspective in two moral case deliberations

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the response letter will be submitted in the attach files

Dear editor,

Thank you for your comments on our manuscript ‘Moral dilemmas and conflicts concerning patients in a vegetative state /unresponsive wakefulness syndrome; shared or non-shared decision making? A qualitative study on two moral case deliberations’. (METH-D-17-00139)

You have summarized the most important. In this reply letter we address the comments of the reviewers as summarized by you. In the manuscript we write the adjustments in italics. In this reply letter we highlight the adjustments with • and write the actual changes in italics and refer to the lines in the manuscript.

Reviewer 1:

Comment 1: I do not think this is a study of two moral deliberations. It is a study of the relationships of care providers and families and the sources of disagreements over prolonged treatment of these patients.
Reply: We agree with the reviewer that we did not study the process of Moral Deliberations (MD's). The aim of the study was to make an inventory of the professional perspective on conflicts concerning long-term care for patients in a vegetative state/unresponsive wakefulness syndrome (VS/UWS) in Dutch nursing homes. In order to clarify that the perspective is among health care professionals (HCP) (including the perceived patient’s/family issues) we adjust several sections in the manuscript:

- Subtitle: We add 'the professional perspective in': A qualitative study of the professional perspective in two moral case deliberations.(line 5-6)

- Abstract part aim: We change the line 'To explore the dilemmas and conflicts experienced by professionals in long-term care' into: 'To make an inventory of the professional perspective on conflicts in long-term care of patients in VS/UWS.(line 42-43)

- Abstract part methods: We add 'transcripts': A qualitative study of transcripts on 2 Moral Deliberations (MD's) in 2 cases of patients in VS/UWS in long-term care facilities.(line 44)

- Background: We change the line 'Our study aimed to explore the conflicts and ethical dilemmas experienced by professionals of patients in VS/UWS in Dutch long-term care settings' into: 'The aim of our study is to make an inventory of the professional perspective on conflicts concerning patients in VS/UWS in Dutch long-term care settings.(line 126-127)

- Methods: We add 'of the professional perspective' and 'transcripts': We present an explorative, qualitative study of the professional perspective in two cases of patients in VS/UWS residing in Dutch nursing homes, using transcripts of two moral deliberations (MD's).(line 132-134)

- Results, analysis: We add 'transcripts': Analysis of the transcripts on two MD's.(line 231)
Comment 2: It is a serious limitation not to have the family perspectives. At one point I thought it was fatal to the paper, but I think it is still important to learn what care providers think the family thinks. Still, the authors need to be careful not to overstate their knowledge of what the families actually think, since this is all second-hand and may reflect the perspectives of the professionals and their frustrations, etc., more than actual family perspectives.

Reply: We agree with the reviewer we would like direct information of the families. Given the nature of an MD, information from the family is indirect. Thus we invited the families to give their perspectives but they refused to participate in interviews. Therefore, it is in these cases impossible to obtain direct information from the families of these patients in VS/UWS. We recommended in future studies in-depth interviews with families of other patients in order to learn from their perspectives. In a new and already started study we use the results of this analysis in the interview guide for those in-depth interviews with families. To highlight this next step we add a line:

- Recommendations: We have started interviewing families of other patients in VS/UWS using the themes found in this study in the actual interview guide.

Further, we agree with the reviewer that we did not have the actual perspective of the families. Therefore, a possibility on bias occurs as the professionals interpret what families are thinking and telling. In the limitations section we add text to elaborate on this possible source of bias:

- Limitations: The lack of direct information from them might result in a bias regarding the information of families, as professionals have told what they think families think, feel and tell. We think here two different aspects are at stake:

  o Firstly, the information on what the families communicated seems quit reliable since multiple sources/professionals in the MD -nurses, psychologists, social workers, physicians, speech therapists and physiotherapists- mentioned what the family told them. Sometimes these expressions were quoted literally, as the members stressed in the MD that they paraphrased the family.

  o Secondly, we acknowledge a possibility that the professionals expressed their own perspectives and frustrations, instead those of the families. The ethicist that chaired the MD's made sure that all aspects concerning the families were sincerely discussed by everyone involved.
We agree with the reviewer that it is important to understand how care providers reflect and interpret how the families think. The way professionals experience families' perspectives might contribute to the conflicts. We agree with the reviewer that this study gives a first insight. Therefore we add some lines:

• Limitations: In the MD the participants mentioned how they thought the families thought and felt. Although this is indirect information we still think that it is important to understand how care providers reflect and interpret how the families think and feel. Given the prolonged contacts between the professionals and the families conflicts are likely to happen as the patients have been treated many years and the families visit their relative daily and participate in daily care. The way professionals experience families' perspectives might contribute to the conflicts.(line 548-554)

Comment 3: "Family members" and "families" are used a bit indiscriminately and as if they are interchangeable people. More precision here is warranted.

Reply: the terms "Family members" and "families" are explained in the following sentences:

• Results: ‘Family member’ and ‘family’ are purposefully distinguished. The family of a patient is not just one person but consists of a group of individuals. Each individual has its own perspectives, feelings and thoughts, even if the family agrees to one point of view. With ‘family’ we address a family as a whole, with 'family member' we refer to one family member in order to see possible differences within a family.(line 191-195)

Comment 4: I'm surprised the authors did not need institutional ethics review, although I am not familiar with the standards in the Netherlands.

Reply: As already explained in the material and methods section, the study was evaluated by the accredited regional medical research ethics committee. According to the Dutch Medical Research Involving Human Subjects Act (1998), the study did not meet criteria for medical scientific interventional research. Therefore, no additional ethical evaluation was needed. The committee also stated that since maximal anonymity was secured, consent of the family or the participants of the MD was not required. Furthermore, we consulted an independent ethicist who confirmed that no formal consent of the families was needed since the families did not participate in the study and anonymity was optimized. Anonymity was optimized by
anonymization of several aspects like the patient’s names, time after incident, age, family relationships and other potentially identifying information, as recommended by Saunders et al.(Saunders, Kitzinger et al. 2015) (line173-183)

Comment 5: References to "best interest," "advance directives," and "advance care planning" require definition. In the U.S. best interests and respect for autonomy through honoring advance directives are two very different ways of determining what should be done. "Best interests" is an objective standard; honoring an advance directive respects the subjective wishes/values of the patient. I could not follow what was meant by "advance care planning."

Reply: The words ‘in the best interest’ of the patient as used in this manuscript are not intended as the legal term used as an objective standard in the US. In order to prevent misunderstanding we use the words 'the most appropriate treatment for the patient' if in the earlier text was written 'in the best interest'.

Discussion: To decide what is the most appropriate treatment for the patient after consulting the family.(line 448)

Recommendations: If no AD is available the physician has to make the critical medical decision after consulting the family about what is the most appropriate treatment for the patient.(line 561-562)

We agree with the reviewer that the definitions of Advance Directive (AD) and of Advance Care Planning (ACP) are lacking. We therefore clarify these definitions:

- Results: Definition of Advance Directive (AD): Oral or written directives from the patient on his subjective wishes and values on treatment decisions.(Emanuel 1993)(line 186-187)

- Results: Definition of Advance Care Planning (ACP): The process of developing a valid expression of wishes of patients during several meetings.(Emanuel, Danis et al. 1995) It
is a decision making process from the patient, or his family with the treating physician in anticipation of end of life. (line 188-190)

Comment 6: The failure of the system to provide adequate expert opinion (neurologists) to determine the condition and prognosis of these patients is quite shocking. The paper does not, in my view, address this adequately and treats it more as something that would be helpful in coming to resolution rather than as an essential part of medical care.

Reply: We agree with the reviewer that an adequate expert opinion is essential in order to be able to provide adequate medical care for these patients. However such expertise is not widespread, for multiple reasons. The first reason is the small number of these patients in long-term care. We have already mentioned this in the limitation section in a systematic review of prevalence studies of VS/UWS, the Netherlands had the lowest prevalence-rate of VS/UWS (0.2/100 000) of the world. In a Dutch long-term care prevalence study, only 3 patients in VS/UWS were found beyond 10 years of onset. (line 525-528) The second reason for few experts is that the diagnosis of VS/UWS is difficult to make. As we have already mentioned in the discussion section that several international studies throughout the previous decades show a misdiagnosis rate of around 38-40%. (line 405-406) Up to now structured behavioural assessments like the Coma Recovery Scale-revised (CRS-r) are the most commonly used test to confirm the level of consciousness. (Kalmar and Giacino 2005, Gerrard, Zafonte et al. 2014) We mentioned in the result section that in these two cases an expert opinion by an in these patient experienced Elderly Care Physician (ECP), using CRS-r assessment, was proposed, yet in only one case the family gave their consent for a second opinion by an expert. In that case the diagnosis was confirmed by CRS-r. (line 490-491)

In order to stress more the necessity of expertise we add the following sentence:

• Recommendations: Given the complexity and the small numbers of these patients and the high percentage of misdiagnosis adequate expertise, including on the use of CRS-r is, absolutely a necessity. (line 563-564)

Reviewer 2:

Comment 1: As these are second-hand reports about families, it is difficult to know what they indicate about the families' actual visions, beliefs, and experiences.
Reply: See also comment 5 of Reviewer 1. We agree with reviewer 2 that during a MD the experiences of the family are seen through the eyes of the HCP. It is, therefore, not known what the actual visions, beliefs, and experiences of the families are. Therefore a possibility on bias occurs as the professionals with their own difficulties and frustrations interpret the perceived family issues. This comment is already addressed in the reply on comment 2 of reviewer 1. We add changes to highlight this:

• Limitations: (line 539-543), (line 543-547) and (line 548-554)

We agree with the reviewer that we do not know the actual visions, beliefs and experiences of the families. Therefore we already recommended interviews with families of other patients in VS/UWS, in order to learn from their perspectives.(583-584)

Comment 2: These are really two case reports of what was said in the MD process, based on notes. They would have been sounder methodologically had they interviewed the participants or the families.

Reply: See also response to comment 1 of reviewer 1. We agree with the reviewer that in the study we used the transcripts of the MD's. We have already addressed this in comment 1 of reviewer 1. Therefore we add 'transcripts':

• Abstract part methods: (line 42-43)

• Methods: (line 133)

• Results part analysis: (line 231)

To react on the comment of reviewer 2 regarding the methods we add the reason to use MD's instead of interviews of the individual professionals:
• Strengths: We used transcripts of the MD's instead of interviews of the participants because in a MD one is able to explore differences in interpretations that might lead to differences in behaviour, while in interviews one can only study the facts told by participants. (line 511-514)

As already mentioned in the methods (line 152-154) and in the limitation section (line 537-538) we intended to interview the families of these patients but they refused to participate.

Comment 3: Do the notes introduce bias in the themes?

Reply: We think the bias in the texts are limited as they are verbatim. In order to explain this we add some lines:

• Methods: The first three authors consented on the transcripts of the MD's. (line 156-157)

• Limitations: Audio taping of the MD might have been a better method but we do not think it introduced a bias in themes as the first three authors all observed or chaired the MD and agreed on the notes. They had consented on the transcripts of the MD's. After the transcripts were written they independently coded the transcript and finally they had consensus over the themes. (line 531-535)

Comment 4: Did the structure of the process of the MD limit what participants thought they could say?

Reply: We have no indication that the structure of the MD limited what participants thought they could say, on the contrary. The way the MD's are chaired, in a save atmosphere, gives all opportunity for the participants to express themselves freely. This is considered vital in a MD. Subsequently, we saw a wide range of sometimes deviating facts and opinions. In order to address this comment we add the following lines:
• Strengths: A strength of a MD is that all participants have an equal input and can address their feelings and opinions from their own perspective. This is stressed in the beginning of the MD by the independent ethicist. In these MD's the ethicist gave equal opportunity to all participants to express themselves freely. The ethicist created a safe atmosphere and encouraged all members of the MD to share their knowledge, ideas and opinions. The wide range of facts, including, deviating facts and opinions, that were put forward underline the save atmosphere and freedom to speak. (line 516-523)

Comment 5: How might interviews with the families or participants in the MD process have introduced new themes?

Reply: As mentioned in the reply on reviewer 2 (second part of comment 2),

• Strengths: we purposefully used a MD to explore differences in interpretations that might lead to differences in behaviour, while in interviews one can only study the facts told by participants. (line 511-514)

We do not know whether new themes might have emerged if we had chosen to interview the participants of the MD. Recently, in another study, we have performed interviews in a national cohort study with all physicians of these patients regarding treatment decisions. (line 586-588) Preliminary results suggest similar themes and also inner contradictory feelings and thoughts.

We agree with the reviewer that we would like to know whether interviews with families would introduce new themes. Since we did not have the opportunity to learn the actual perspective of the families we do not know whether new themes would have been introduced in interviews with those families in this study.

Comment 6: More generally, it is unclear how these two reports might generalize, or even what the next steps might be after this "pilot." Some discussion of what this was a pilot "for" might have been helpful.
Comment 6a: It is unclear how these two reports might generalize?

Reply: Generalization is not a goal of this qualitative study. We aimed to make an inventory of the professional perspective of the difficulties experienced by HCP in case of long-term care for patients in VS/UWS. Further research may validate our results, although, as already mentioned in the limitation section, the total population in the Netherlands is very small. Therefore we think these 2 cases represent a significant sample of the whole population of VS/UWS patients in Dutch long-term care, in particular after 10 years of onset.

Although we have studied the Dutch situation, the care for these patients generate similar problems all over the world. Like the high rate of misdiagnosis, the many harsh conflicts as illustrated in the background section in the Terrie Schiavo or the Englaro cases. Given the complexity of those patients, we agree with reviewer 1 the lessons as we have learned from our study, and already recommended are relevant too for those patients and their families and professionals treating them.

Comment 6b: Some discussion of what this was a pilot "for".

Reply: This qualitative inventory is a first step to develop recommendations for medical practice, investigations and education as we have already written in the recommendations:

- An expert to assist in establishing the diagnosis, prognosis and subsequent treatment goals and plans.
- In support of the professionals and to make them aware of dilemmas we recommend the introduction of MD's.
- Education of all professionals regarding: diagnosis, prognosis, appropriate treatment plans and medical ethical considerations of patients in VS/UWS; excellent communication skills, insight in the effects of inner-contradictory feelings and thoughts.
• We also recommend organizations dealing with these patients to provide stability in the teams and support their personnel, also in facilitating MD’s during the process.(line 576-577)

In order to stress more for whom the results might be helpful we add the following lines:

• Recommendations: For the patients this might result in the most appropriate treatment.(line 578) For the families support by experts, more uniformity in diagnosis, prognosis and treatment goals might prevent conflicts.(line 578-580) For the professionals the above mentioned recommendations can facilitate their work.(line 583-584)

Comment 6c: What might the next steps be after this "pilot?"

Reply: As next steps we have already recommended several future studies. (line 584-585) In an on-going study we have recently performed interviews with the responsible Elderly Care Physicians (ECP's) of a cohort of all patients in VS/UWS in the Dutch nursing homes. Currently we are analysing these data. We have started interviewing families of other patients in VS/UWS to learn from their perspectives. With a special focus on the themes and contradictory feelings and thoughts found in this study. To clarify on the next steps we ad 'for future studies' and we add lines on recent studies:

• Recommendations: We recommend for future studies, in-depth interviews with families and with physicians further exploring their role in the decision making process and in dilemmas.(line 584-585)

• Recommendations: We have already performed such interviews with all the Elderly Care Physicians (ECP's) of a cohort of all patients in a VS/UWS in the Dutch nursing homes. Currently we are analysing these data. We have started interviewing families of other patients in VS/UWS using the themes found in this study in the actual interview guide.(line 587-590)
Literature


