Author's response to reviews

Title: Effect of guided self-determination youth intervention integrated into outpatient visits versus treatment as usual on glycemic control and life skills: a randomized clinical trial in adolescents with type 1 diabetes

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Author's response to reviews: see over
Dear Editors-in-Chief Doug Altman, Curt Furberg and, Jeremy Grimshaw,

Thank you for giving us the opportunity to re-submit the manuscript. We have revised the manuscript according to the points raised by the three reviewers. The revised text is marked with yellow color in the manuscript and below explained in details.

On behalf of the authors,

Gitte R. Husted

**Title:** Effect of guided self-determination youth intervention integrated into outpatient visits versus treatment as usual on glycemic control and life skills: a randomized clinical trial in adolescents with type 1 diabetes.

Comments to Authors
Reviewer 1: Jayashree Gopal
1. As the method used is specific for psychotherapy, I was not able to assess how well validated this method is for this age group in other chronic disease management settings.

**Authors comment:**
*Guided Self-Determination-Youth is not a method for psychotherapy. The method is originally developed through programmatic qualitative research in adult diabetes care (reference number 30). Three grounded theories identified why barriers to empowerment seldom were – or were not - overcome in the interaction between health care providers (HCP) and patients. The method was developed to overcome these barriers and is based on theories such as empowerment, self-determination theory, and life skills theory. These theories have been used in youth before.*

2. Did the lesser number of visits by the participants and parents in the intervention group impact on outcome? There is some mention of this, maybe a little further discussion on why this may have happened and what it implies for the applicability of this method in the real world would be of benefit to clinicians.

**Authors comment:**
*The intervention group did not receive a lesser number of visits than the control group. In fact they needed more visits to complete the eight sessions in order to work through all reflection sheets. We have clarified this in the result section, page 18. We have added our recommendation for how to handle this in the real world in limitations page. 25.*

3. To clarify how the authors plan to determine whether the unchanged HbA1C in the intervention group is a coincidence or not (mentioned under discussions).

**Authors comment:**
*Our point in the discussion section was to tell that whether our finding of an unchanged average HbA1c level in the GSD-Y group during the trial period represents a true difference from the increase in the control group or is a coincidence cannot be said. (Page 21).*
Comments to Authors:
Reviewer 2: Sue Channon
Background: The background is well described capturing the challenges and descriptions of interventions for young people with diabetes and their families.

Methodology:
This intervention has been adapted from a successful intervention with adults. The theoretical underpinning of the intervention is well described in the protocol paper and briefly captured here. However there appear to be significant differences between this intervention and the adult study:
Procedural: A two-hour group intervention delivered outside clinic setting by researcher and HCP to a triadic intervention for adolescent, parent and HCP in clinic.
Developmental stage: it was not clear how the intervention had been adapted from work with adults to work with young people and their parents in a triad. With the youngest participant being 13 years old it struck me that with an intervention that focusses on autonomy-support and shared decision making (within personal and professional relationships) and competency that there may well be young people for whom this isn’t consistent with their personal developmental stage and also the developmental stage of their family.
To understand the “fit” between intervention and participants it is crucial to understand the adaptations that were made in particular in relation to developmental stage and to include parental role.

Authors comment:
We adjusted and developed the Guided Self-Determination-Youth version based on the part of participatory action research (reference number 19,20) that involves participants through on-going dialogues, joint discussions, and reflections. We involved 22 adolescents (13-18 years, 10 girls), parents and 9 interdisciplinary HCPs through practical training session in real outpatient during 18 months. GRH participated in all training sessions with HCPs, patients and parents and observed how the participants reacted to GSD-Y. This was followed by individual interviews evaluating their perception of using the method. GSD-Y was deemed acceptable and feasible for all participants based on dialogues, observations and reactions showing that GSD-Y was efficient in that sense that it had the expected theoretical influence on interactions within dyads and triads. We found that GSD-Y was able to work in different ages because HCPs were able to take the individual developmental stage and the individual family's resources into account. The adjustment and development did not alter the original content and aim of GSD as it only resulted in a reduction in number of reflection sheets from 21 to 18, 1 hour outpatient visit, and minor vocabulary changes such as operationalizing words (they: my mother, father, friends). We have concretised the summary of this in the paper. (Page 6).

Delivery of intervention: HCPs were tested in relation to their delivery of GSDY prior to trial but there is no mention of ongoing measurement of fidelity to the intervention through the progress of the trial. With practitioners delivering both the intervention and the control usual care this seems to be important in terms of establishing whether any result can be ascribed to delivery of the intervention.

Authors comment:
Fidelity was very important concerning two core parts of GSD-Y: correct use of reflection sheets and verbal communication. A major source to fidelity assessment was the filled in reflection sheets
per se, because they showed which person-specific challenges or difficulties were expressed and acknowledged in the progress of the mutual decision-making and problem solving. Sheets filled in by each patient/parent were saved during and after the intervention. Fidelity concerning the verbal communication was additionally measured during the trial. 37 outpatient visits distributed on 11 adolescents and parents and 9 HCP’s were digitally recorded and these data are a part of data for the qualitative evaluation of GSD-Y accepted for publication in Journal of Advanced Nursing 8 March 2014, expected to be available in August 2014. We have added this information to the method section. (Page 13).

I think the idea was that the intervention was delivered as part of routine care – Could the authors clarify what routine contact is as 8 sessions for 30-45 minutes in 12 months (as per control group) seems unlikely to be routine levels of contact? Who would they normally see in routine contact?(just one practitioner?)

Control group – was there any attempt to understand what was covered in these sessions? There needs to be a description of usual care.

Authors comment:
Usually adolescents with poorly regulated diabetes are seen more often than adolescents with well-regulated diabetes in Denmark. Some are seen every fortnight while others are seen every month or every second month. It is the individual HCP who decides how often it is necessary. Adolescents normally see a physician once or twice a year and a diabetes nurse at the other visits. In standard care they always see the same HCPs.

Usual care in outpatient clinic consists of measurement of HbA1c, a talk about the result of this, a talk about possible ways to achieve better regulation, measurement of weight and height.

We have added a short description of usual care and concretised that the control group was seen as often as the intervention group during the trial. (Page 10).

We used a case report form to capture what happened during control sessions for both groups and we registered who participated at each session. This information has been added to the method section. (Page 13)

Were the sheets sent out in advance or completed in waiting area?

Authors comment:
Reflection sheets were handed out at the end of every outpatient session followed by an explanation to ensure that adolescents and parents were aware of how to fill them in before attending next time. This information has been added to the description of GSD-Y intervention in the method section. (Page 9).

Was there a particular reason why dietetic involvement if it was to happen would only be in the first two sessions for the intervention group but “as needed” for the control group?

Authors comment:
We have concretised that dietician involvement were not only related to the first two sessions, but the reflection sheets used in the two first session helped to identify if a dietician was needed. For instance, in one sheet in these sessions, patients mark if they believe that they 1) do not take the amount of insulin needed for the food they eat, 2) do not adjust their insulin dosis, when their blood sugar is high, or 3) eat too much when their blood sugar is low. Moreover they are asked whether
they wish to change this within the first month, within the first ½ year or later. Or if they wish to continue with this pattern. The referral could afterwards take place during the entire trial. There are no specific guidelines indicating when adolescents can be referred to dieticians and therefore adolescents in the control group could be referred according to usual practice which means that it is the individual HCPs who decides if it is necessary. (Page 10).

I wasn’t clear if all sessions were triadic and parents had an additional two sessions? In the intervention section reference is made to “before each visit the adolescents and parents….” and then later there is reference to the parental two sessions - this could be made clearer.

Authors comment:
Not all sessions were triadic. Parents were allowed to participate in their adolescent’s session except session 2. At the remaining sessions the participants made individual decisions whether the adolescents would like to have sessions alone. Parents were offered 2 parental GSD-Y sessions alone using their own reflection sheets.
This has now been concretised in the method section. (Page 9-10).

Outcomes
The secondary outcomes match the intervention - I think with teenagers it makes it particularly challenging to have HbA1c as the primary outcome in a 12 month study with a psychosocial intervention (rather than say the perceived competence or another variable that is at the heart of the intervention) but it is rare to find a study that doesn’t so it fits with expectations of studies in diabetes. In terms of the writing I felt the section on the secondary outcomes section and the measures would benefit from a re-write - it was a bit jumbled and bringing them together more closely might make it easier to read (and potentially save some words). I think the definition of improvement in life skills as a composite measure also needs some greater clarity.

Authors comment:
The section on the secondary outcomes and the description of the scales applied has been re-written and gathered in one section. (Page 11-12).
The definition of improvement in life skills has been re-written and described the conditions which contribute to developing life skills with reference to Self-determination theory by Deci and Ryan, reference number 32. (Page 12).

Results
Visits-sessions: I wasn’t clear quite what was being described - did they come to clinic but not receive the intervention? What defines a completed session? I got thoroughly confused through this section – which may be use of terminology – visits/sessions? This needs clarification although I got the general message that it didn’t go as planned and that the intervention went more off plan that the control group.

Authors comment:
Prior to the trial, we had planned which reflection sheets could be talked about in one visit. However, it turned out that sometimes more visits were needed for working with this content. Accordingly we decided to differentiate between “session 1-8” containing the originally planned content and “visits” saying the number of times the adolescents actually met with their HCP. This is now concretised in the method section (page 9) and in the result section (page 18).
Who was in charge of determining whether parents attended or not? (not sure this is clear in methods).

Authors comment:
HCPs were in charge of determining whether parents had attended or not. HCPs filled in a Case Report Form at every visit to capture this information described in the method section. (Page 13).

In the section on the dietician does the sentence “Each GSDY adolescent completed 1 to 6 visits” mean 1-6 visits to the dietician?

Authors comment:
We have concretised this in the result section. It means 1-6 visits to the dietician. (Page19).

Looking at the numbers it would seem that each practitioner worked with around 5 each in the intervention group? The results control for practitioner but this wasn’t really addressed either in the results or the discussion.

Authors comment:
We have informed at the beginning of the result section that equal numbers of GSD-Y and control adolescents were allocated to each HCPs – approximately 10 adolescents to each HCP and at page 16 we have concretised that each HCP completed different number of patients. We have reflected upon this difference in connection to the discussion of the limitations of the study. (Page 25).

Discussion
Amotivation: the definition of amotivation is slightly buried in the text earlier in the piece – given that it is discussed in detail I think it needs to be clearer or reiterated in more detail actually what it is. It is a little confusing as it is a negative term – does a reduction in amotivation mean they are more motivated? Is this the beginning of a process of engagement?

Authors comment:
Being amotivated is defined in the method section which describes the TSRQ scale, secondary outcomes. (Page 11). Amotivation is the sense of having no intention to change or feel unable to change. A reduction in amotivation means they get less amotivated for taking their insulin, less amotivated for measuring blood sugar and following diets and exercising. Amotivation has been connected with helplessness and is according to Levesque et al. 2007 (reference number 41) incompatible with self-determination: “People who are amotivated lack motivation and are therefore not self-determined. They do not behave in a purposeful manner. They experience no meaningful relation between what they are doing and themselves.” We have extended the description of amotivation and also elaborated further on the decreased amotivation in the discussion. Decreased amotivation might be the first sign of beginning a process of engagement and a sign of developing life skills. (Page 23).

It is good to put the results in context of three other trials with similar types of findings - I wasn’t sure if the authors made a little too much of the fact that their intervention HbA1c was unchanged (when others went up) but they were tentative so I think it was legitimate.
There is little discussion of the fact that 11 young people did not complete the intervention of which 7 were patient withdrawals - active and passive – compared to 3 non-completers in the control group. Was there a sense that this was quite a demanding intervention?

Authors comment:
We agree that the intervention may seem demanding since the numbers of non-completers were so high in the intervention group. This is added to the discussion section (page 22) and to the limitations of the study (page 25).

The deviation from the original protocol is included and the rationale for not following the group approach used in the adult study is well and honestly justified. Given the level of differences between this and the adult study intervention I wouldn’t go so far as to attribute the different outcome on HbA1c to the completion of the two reflection sheets – seems a bit extreme so I would modify that sentence.

Authors comment:
We have modified the sentence. (Page22).

I think the writing gets much harder to follow when the discussion considers amotivation. I didn’t understand why amotivation indicates that the adolescents felt motivated to identify the reasons for self-management in which they believed – this needed to be clarified.

Authors comment:
As prior mentioned we have modified this sentence and referred to Self-Determination theory as described by Deci and Ryan, reference number 32.

In the description of the measure earlier in the paper autonomy is different to amotivation but somehow the young people are now being described as autonomously motivated which I didn’t understand as I didn’t think there was a significant impact on autonomy.

Authors comment:
We have re-written this section (page 23) and agree that we did not see a significant impact on TSRQ autonomy after Holm’s correction.

Why does a lack of autonomy support in the parents and HCP responses indicate they are applying pressure? It may be that the parents’ responses on questionnaires don’t indicate an increase in autonomy support but they are demonstrating this by enabling the children to attend appointments alone? What is the appropriate amount of autonomy given that there is clear evidence of the positive impact of continued parental involvement across adolescence? It is also a very valid point that maybe the reduced amotivation relates to the sheer fact of higher number of visits – indicating the HCPs commitment to them and their care?

Authors comment:
We have re-written this part of the discussion (page 23). Based on Self-Determination theory, Niemic (2006), reference number 44, Leonard et al.’s study (2005), reference number 45 and the study by Christian (1999) reference number 46, we argue that parents who promote controlled motivation for diabetes self-management may contribute to resistance and amotivation instead. We agree that we do not know how much parents should continue to be involved across adolescence.
We doubt whether higher number of visits per se contributed to reduction in amotivation. None of the other scales were influenced by this difference between the groups. (Page 23).

The possible spillover ie contamination between arms is recognised but it also needs to be acknowledged that with no measurement of the fidelity of the intervention there is no way of knowing what was actually occurring in any of the sessions either intervention or control?

Authors comment:
We have added the information regarding the fidelity assessment which took place during the trial in order to ensure that GSD-Y was delivered as intended in the GSD-Y group. However, we are aware, that we do not know what happened in the control group except we know that the reflection sheets were not used here. (Page 13 & 24).

I appreciate this is the quantitative paper but a few headlines about the qualitative experience of the participants would be of interest.

Authors comment:
Our paper presenting the findings from the qualitative evaluation of GSD-Y was accepted for publication 8 March 2014 in Journal of Advanced Nursing and we expect it to be published in August 2014.

Comments to Authors:
Reviewer 3: Andrew Vickers
The main problem with the statistical reporting is that the authors do not provide the main estimates of interest, that is, a difference between group means or proportions along with a 95% C.I. This is a basic reporting requirement of CONSORT. It isn't enough to say that "the difference was non-significant", it is important to know the direction of the effect size and whether the trial has excluded a clinically relevant effect.
It also appears that the statistical model did not include baseline score as a covariate, which dramatically increases statistical power.

Authors comment:
There were a few errors in the last description of the statistical analysis which we had missed. They might suggest that we had adjusted all analysis with the baseline value of log(HbA1c). Actually all 14 primary analyses (primary outcome and the important secondary outcomes) were adjusted by the baseline value of the dependent variable and the stratification variable health care person (HCP). Since the main effect of the follow-up indicator and the interaction between the follow-up indicator and the intervention indicator were insignificant in all analyses the model was reduced accordingly. The enclosed table 2 gives the P value of the main effect of the intervention and the quantities that you have requested. The primary outcome and each of the important secondary outcomes are now presented with the main estimates of interest with 95% CI so that the estimated magnitude and direction of the effect size may now be seen. Clearly they were needed. Thank you for pointing it out.