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Research Report

The Hold me Tight Program for Couples Facing Huntington's Disease

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Abstract.
Background: A positive predictive genetic test for Huntington's disease (HD) can be a life-changing event for both carriers and their partners, leading to lower wellbeing and increasing the risk for separation and divorce. The 'Hold me Tight' program (HmT), based on emotionally focused couples' therapy, aims at strengthening the couple bond by targeting attachment needs.
Objective: This study investigates whether the HmT program helps couples strengthen their relationship, as an investment in a future where the disease will affect life in many ways.
Methods: In a multiple baseline design using three baselines of varying length, 15 couples of presymptomatic HD-carriers and their partners were included. In three consecutive groups, couples underwent the intervention (an adapted version of the 8-session HmT program) in four weekly sessions and completed self-report questionnaires throughout the study period of 19 weeks (17 measurements). Attachment style was assessed at baseline, resilience at baseline and at the end of the follow-up, while relationship satisfaction and wellbeing were measured weekly. A multi-level model was applied to the data.
Results: Over the course of the study, wellbeing and relationship satisfaction significantly improved; resilience, however, did not. Furthermore, all three outcome measures were moderated by attachment style, with more securely attached individuals showing better outcomes.
Conclusion: HmT improved wellbeing and relationship satisfaction of couples facing HD. Due to these improvements and high patient acceptability rates, this program could become a standardized procedure in HD care. The program could be adapted for other populations, e.g., couples facing other genetic neurological disorders.

Keywords: Huntington's disease, hold me tight, resilience, psychological, couples

INTRODUCTION

Huntington's disease: Characteristics, epidemiology, and genetic background

Huntington's disease (HD) is a rare neurodegenerative hereditary disease characterized by involuntary motor activity (*choreatic movements*) as well as psychiatric, behavioral, and cognitive symptoms, such as depression, apathy, and dementia [1]. Approximately 9 per 100,000 people are affected by HD in the European population [2]. The mean age of onset is between 40 and 45 years [3]. Patients become increasingly dependent on others due to the progression of their symptoms, leading to death after an average of 17–20 years. Pneumonia and suicide are leading causes of death [1, 4].

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The underlying cause of HD is a trinucleotide repeat expansion of the Huntingtin gene (*HTT*) on the short arm of chromosome 4. A person having 40 or more CAG repeats on this location will develop HD at some point in life, with more CAG repeats being associated with earlier onset. Persons with 36–39 repeats might develop HD at a later stage in life ('reduced penetrance') [5]. Due to an autosomal-dominant inheritance pattern, offspring of Huntington's disease gene expansion carriers (HD-carriers) have a 50% chance of inheriting the mutant *HTT* gene [5].

Persons at-risk for HD can assess their carrier status by getting a predictive genetic test. They face the difficult decision whether they want to know their carrier status and thus whether or not they will develop HD in the future. International recommendations on predictive testing for HD include psychosocial support and counselling to be available before the test commences, so that persons at risk can make an informed decision about taking the test and appropriate care can be given after a positive test result [6].

Psychosocial factors in Huntington's disease

The predictive testing guidelines also urge caution towards the consequences the test may have for the family, such as family planning by the affected person and their spouse. The median age to get tested at is 37 years, an age at which people often have a committed partnership and children [7]. Not only the affected person, but also the spouse may experience significant distress after receiving a positive test result [8, 9].

Overall, relationship satisfaction of couples in which one partner is affected by HD is lower than in the general population, and also lower than in other neurodegenerative diseases [10]. There is an elevated rate of divorce and separation [11, 12]. In couples, non-affected partners indicate lower relationship satisfaction than their HD-carrier spouses [10, 11]. On the one hand, this may be because people with HD display impaired social cognition, such as low Theory of Mind, thus limiting their possibility to empathize with their partner's feelings towards the relationship. A recent meta-analysis has shown that this is especially the case with negative emotions, such as anger, disgust, and fear, even before the onset of motor symptoms [13]. On the other hand, the non-affected partner may try to compensate for unmet needs by being overly supportive and engaging in excessive caregiving. This impacts the relationship satisfaction of both partners: the HD-carriers may feel like they are not being given enough space, while the non-affected partner may feel a sense of never-ending obligation towards their affected spouse [9].

Resilience, describing a personality trait that is protective against distress, is an important construct in healthcare. Patients who are more resilient are able to communicate more effectively with healthcare providers, thus benefitting their treatment outcomes [14]. In general, resilience in people with neurodegenerative diseases is related to higher independence in activities of daily life, and resilience was found to improve mental health. Conversely, having less psychological distress improved resilience in people with HD [15]. Regarding partnership, a study with persons with neurodegenerative diseases (including HD) found that the length of the current partner relationship determined resilience in dealing with the problems arising with the disease. Married couples and long-term couples, as well as couples who raised children together were found to be more resilient [14]. Family members generally care for and support HD-carriers, which has a favorable impact on the HD-carrier's resilience, but this can also elicit feelings of guilt and resentment among HD-carriers, which have a negative impact on resilience [16].

The wellbeing of both partners is endangered after a positive test result. Partners experience 'disenfranchised grief', meaning that they too have feelings of imminent loss or grief, but this cannot be publicly acknowledged or mourned because they are not the mutation carriers and therefore have 'no right' to display their grief [9]. Partners are therefore often found to be more negatively affected on an emotional and cognitive level than the HD-carriers themselves. Coping with the test result, both negative and positive, is reportedly difficult for those who underwent the test. Many children of mutation carriers, however, anticipate and prepare for HD before even knowing their mutation status [17]. Partners experience more fearful cognitions and have less effective coping styles than HD-carriers, such as withdrawing [8, 9, 18]. A typical behavior a partner may consequently engage in is overprotection. This may result in less self-efficacy, less distress-reduction, and more depression in the patient, thus undermining patient wellbeing [18–22].

Attachment theory and implications for HD-carriers

Growing up with a parent who has HD may negatively impact the attachment style of offspring [23, 24]. Attachment style can be seen as a two-dimensional construct, consisting of attachment anxiety

and attachment avoidance. Anxiously attached individuals focus on negative emotions and relational threats. They show an intense fear of separation and a strong desire of appreciation and attention from their significant other because they have low self-worth [25]. Hence, a core characteristic is their negative view of themselves [26]. Persons who are high on attachment avoidance typically express distrust towards their partners because they think that they will be undependable when needed. They value being self-reliant, and a core characteristic is subsequently their negative view of others [26, 27]. Persons are considered to be more insecurely attached when they score higher on attachment anxiety, attachment avoidance, or both (see [26], for more details).

In offspring of HD patients, higher levels of attachment anxiety are found than in the general population [23]. The timing of their parent's onset of symptoms as well as the number of adverse childhood experiences (especially those related to psychiatric problems of a parent) predicted a more insecure attachment style of offspring of HD patients [23, 24]. Children of parents with HD describe a negative impact of the disease on family dynamics, with both parents being greatly affected by the onset of HD, and inadequate, inconsistent parenting styles, creating conflict-ridden relationships between children and parents [28]. Insecure attachment can lead to poorer mental health [23, 29], which can further impede the quality of life and wellbeing of these persons. Resilience and relationship satisfaction are typically higher for persons with a more secure attachment style [30, 31]. Although a person's attachment style originates in childhood and is relatively stable throughout life, a secure partner relationship in which attachment needs are fulfilled can contribute to more trust in the partner's emotional availability and support and to a greater sense of attachment security [32]. Couples who face a future with HD may benefit from an attachment-based intervention in which they explore and understand each other's emotional needs. They may feel more secure in their relationship, experience more quality of life, and feel more confident that their partner will be supportive throughout the disease process. Persons with insecure attachment styles may have more to gain from relationship/attachment-focused interventions.

To the knowledge of the authors, for presymptomatic HD-carriers, no studies have been done on interventions using an attachment theory approach, even though there is evidence that this can improve clinical outcomes [33]. By taking a more systemic approach and addressing attachment needs of carriers as well as their partners, clinical outcomes would likely improve more than in individual therapy.

Fig. 1. Seven Transforming Conversations [35].

The HmT intervention

An empirically validated approach to address relational problems is the Hold me Tight relationship enhancement program (HmT). HmT is derived from Emotionally Focused Therapy based on attachment theory, and seeks to improve partner relationships [34]. It consists of eight sessions: one introduction session and seven 'transforming conversations' for couples (see Fig. 1). These conversations between partners can be imagined as a step-by-step program aimed at examining typical results of miscommunicated or unmet relationship needs and recognizing these needs in each other, ultimately leading to improved and strengthened relationships. It can be used in a group therapy setting, or with just the two partners present. Materials include video clips, exercises, role plays, and various other tools [35]. The effectiveness of HmT has been proven in non-clinical populations, with large size effects for improving trust and intimacy, and reducing anxiety [36]. Recent reviews have shown large effect sizes in various areas, such as marital satisfaction and target complaint reduction, thus strengthening HmT's position as an evidence-based therapy [37, 38]. Next to improving relationship satisfaction, HmT also succeeds in

strengthening personal resilience [39]. Furthermore, a special HmT program for cardiac patients has been developed (Healing Hearts Together, ICEEFT, 2019; [40]), which is currently being evaluated (clinicaltrials.gov: NCT03938116). Special adaptations for the cancer population have also been made [41]. Therefore, it is reasonable to assume the HmT program can be adapted successfully for an HD population. This would additionally benefit the area of psychological interventions in HD, as there is a great scarcity of methodologically sound studies [42]. HmT has the potential to offer a much-needed systemic approach by helping couples to explore and discuss what they need from each other after a positive predictive genetic test. This may strengthen their bond and help couples prepare for future difficult situations due to the onset and progression of HD. Furthermore, future generations in families with HD may profit: parents with strong emotional bonds raise children who are more securely attached, leading to fewer psychological problems [32, 43]. Lastly, by learning how to identify their own and each other's emotional needs, these couples may be able to apply these skills in their interactions with their children.

Aims and scope of this study

In this study, the aim is to examine whether the "Hold me Tight" relationship enhancement program has favorable effects on relationship satisfaction, resilience, and wellbeing in couples where one partner has the HD mutation. To this end, three hypotheses were put forward:

1. Relationship satisfaction is higher after having participated in the 'Hold me Tight' program.
2. Resilience is higher after having participated in the 'Hold me Tight' program.
3. Psychological wellbeing is higher after having participated in the 'Hold me Tight' program.

As part of a secondary analysis, the effect of attachment style on the proposed relations was investigated. Specifically, it was expected that:

4. Attachment style is a moderator on the relation between intervention and outcomes: A more insecure attachment style (i.e., more attachment anxiety, attachment avoidance, or both), is associated with larger pre-post-differences in relationship satisfaction, resilience and/or wellbeing.

METHODS

Recruitment and sample

Participants were recruited at the department of Clinical Genetics at the Leiden University Medical Centre (Leiden, The Netherlands) by screening for all persons who had enrolled for predictive genetic testing in the years 2017–2020 and had received an unfavorable result on their predictive genetic test (i.e., 36 or more CAG repeats in the *HTT*-gene). Recruitment was also done via professional networks of health care providers working with the HD-population and via the Dutch HD-patient organization (*Vereniging van Huntington*). Prospective participants were couples where one partner was an HD-carrier and the other was not. Further inclusion criteria were having a partner relationship for $\geq$ 12 months and the mutation carrying partner being between 18 and 65 years of age. Participants were excluded if one or both of the partners were not fluent in Dutch, had manifest psychopathology (defined as a score $\geq$ 63 on the Global Severity Index (GSI) of the Brief Symptom Inventory (BSI)), or if the HD-carrier showed motor symptoms of HD or had received a clinical diagnosis of HD prior to entry in the HmT program.

Based on similar previous studies [44], methodological aspects [45, 46], and other considerations regarding feasibility, available financial funding, expected effect size, and drop-out rates, it was determined that 18 couples would be the ideal sample size (with an acceptable range of 15–21 couples). Prospective participants were informed via e-mail and telephone call until the number of included couples needed for the study was reached. This resulted in informing 38 couples (see Fig. 2). After informed consent, 23 couples agreed to participate and were enrolled in the study. A total of 15 couples completed the entire study, resulting in a drop-out rate of 35%. Some couples joined the study after the first measurements, but before they started the intervention. Every couple that started the intervention completed the intervention.

The Medical Ethical Trial Committee of the Leiden University Medical Center, after receipt of necessary documents, ruled that this study was not subject to the human subjects act (#NL71670.058.20).

Design

Since HD is a rare disease, it would be difficult to fulfill reasonable power criteria in a parallel randomized clinical trial (RCT). To solve this problem,

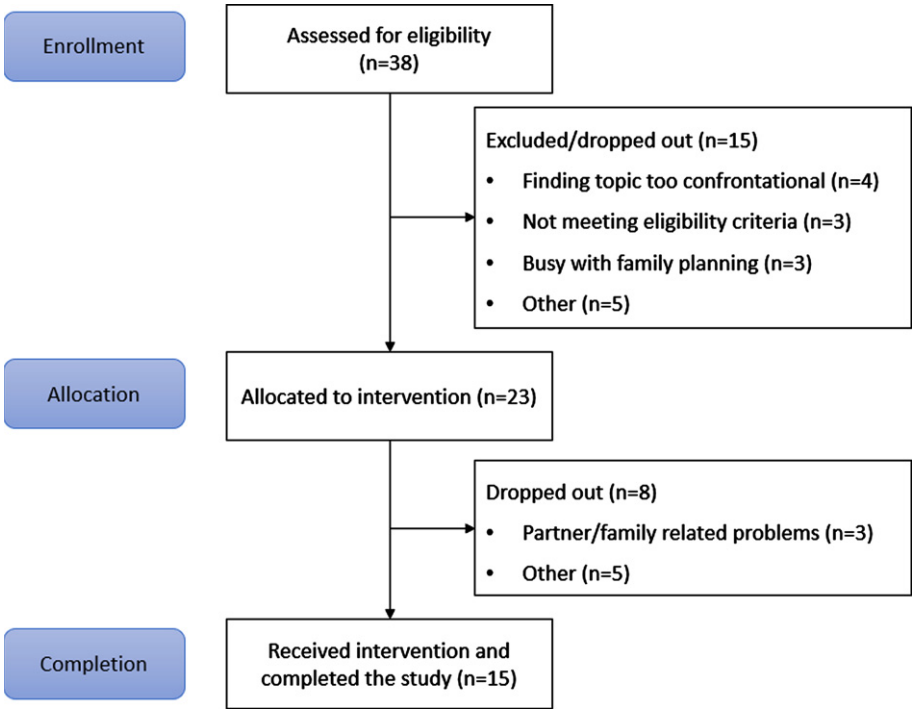


Fig. 2. CONSORT-style flowchart of participants (n = number of couples).

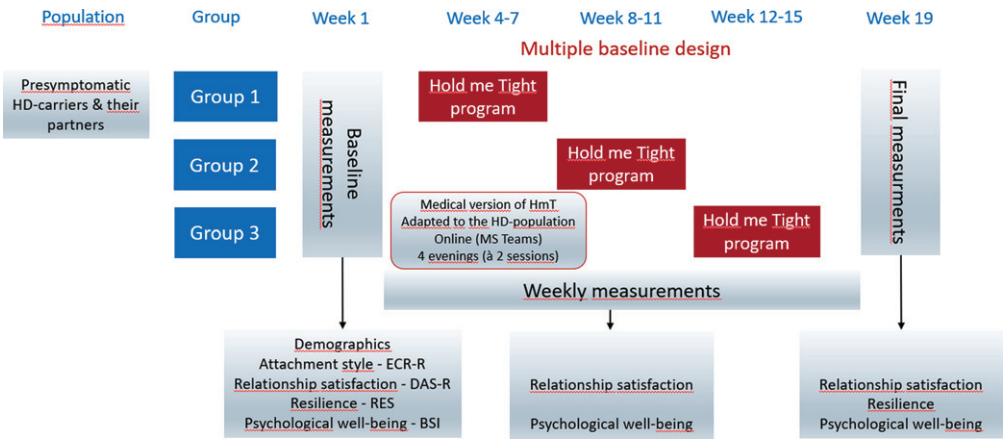


Fig. 3. Visualization of the study design.

a multiple baseline design was used. This is a special cross-over design in which couples switch from the control to the intervention group at different time points and it is, in general, more powerful than a parallel cluster RCT [47]. Specifically, three groups of different baseline lengths were used. Each group consisted of 4–6 couples. After 3, 7, or 11 weeks respectively, the groups started the intervention (see Fig. 3).

By using a multiple baseline design, we were able to effectively maximize the power of our statistical conclusions. By staggering the start of the intervention, a large pool of comparisons is created that has all the advantages of an ABA design combined with elements of an RCT (e.g., when the first group started the training phase, the other two were control groups). Furthermore, this design is robust against historical events. For example, if a hopeful medical finding for

HD would have been discovered during our study period, this would have affected all three groups in their three different study stages, meaning we could have estimated the influence of this event and then recalculated our study effect without this event.

Couples were first blindly distributed to the groups. To increase the feasibility and prevent drop-outs, the couple's availability was taken into account—for example, one couple wanted to do the intervention *before* they had their baby, which was due soon, so groups were reshuffled in order to accommodate this (semi-randomized approach). As this was only necessary for 3 couples, allocation can still be considered random enough to prevent systematic variation.

Procedure

Participants started the study at the same time with baseline measurements (T1), filled in weekly measurements (T2-T16), and completed the study at the same time with final measurements (T17). At baseline, participants were asked about demographic and biographic variables, attachment style, relationship satisfaction, resilience, and psychological wellbeing. Repeated measures were weekly assessments of relationship satisfaction and psychological wellbeing. Relationship satisfaction, resilience, and psychological wellbeing were assessed at T17. All questionnaires were sent out using the Castor Electronic Data Capture System (Castor EDC).

The participating couples received the Dutch version of the book 'Hold me Tight' [48] two weeks before the intervention of the group started. Two trained and experienced psychologists (Dr. L. B. van der Meer (LvdM) and Dr. A. C. Brouwer-Dudok de Wit) carried out the HmT program in the first group; the program for the consecutive groups was performed by LvdM, with practical support of TMP and AD, a master student who was involved in the project. The eight distinct sessions (introduction and seven 'conversations') were held on four evenings of 4 hours each for each baseline-group. Due to national COVID-19 restrictions, the intervention was done via Microsoft Teams. As described in a recent publication [49], each couple sat together in front of one screen. To adjust the HmT program to the HD population, and in line with the Healing Hearts Together program, an additional session was added between the introduction and the first conversation, to focus on HD. In this session, all HD-carriers were grouped together separately from their partners, who were also grouped together, consecutively, both with one of the psychologists (LvdM). Thus, a safe atmosphere was created to talk about the hardships and experiences of facing a future with HD as a consequence of the positive test result, with people who are in the same situation. In line with the Healing Hearts Together program and other HmT-programs for a medical population, this session replaced the third conversation ('Revisiting a Rocky Moment'; see Fig. 1).

Because the content of the HmT 'conversations' can be extremely intimate (such as 'raw spots', physical intimacy, and forgiveness), the couples completed the remaining exercises separately from the other couples (by muting themselves and turning off their cameras) after receiving instructions and introductory materials. The couples were not obliged to share their experiences with the other couples. Between meetings, participants were asked to read chapters of the HmT-book [48] and to follow up on what was done in the sessions, by exercises or assignments provided by the trainers.

To assess how participants perceived the program, the psychologists asked for feedback at the end of each evening, and written comments were given by participants during the evaluation after completion of the HmT program.

Measures

Dyadic adjustment scale

The Dyadic Adjustment Scale (DAS) is a questionnaire used to assess relationship satisfaction in cohabiting couples [50]. It consists of 32 items (with 2–6 answer categories each) measuring various areas of adjustment towards each other, such as dyadic satisfaction, dyadic cohesion, dyadic consensus, and affectional expression. In this study, the full range was used to compute a total score. Respondents can obtain a score of up to 151, with higher scores indicating higher relationship adjustment, meaning both that the individual has adjusted to being part of a couple, as well as the dyad functioning well as a couple. A Cronbach's $\alpha = 0.888$ was estimated in the study sample, in line with previous studies, ($\alpha = 0.915$ [51]), indicating high reliability. The construct validity is also very high, amounting to an $r = 0.86$ Pearson correlation with other well validated scales measuring marital adjustment [50]. This questionnaire was filled out every week.

Resilience evaluation scale

Resilience was measured using the Resilience Evaluation Scale (RES) [52]. It consists of 9 items

(5-point Likert scale), representing the constructs of self-confidence (3 items) and self-efficacy (6 items). Respondents can score between 0 and 36 points, with higher scores indicating more psychological resilience. The participants filled in this questionnaire at baseline and at the end of the study (T17). The estimated reliability was $\alpha = 0.883$ in the study sample.

Brief symptom inventory

The Brief Symptom Inventory (BSI) is a self-report scale for psychological symptoms [53], which was developed from the SCL-90-R. It consists of 53 items on a 5-point Likert scale. Higher scores indicate more symptoms and higher intensity of symptoms. The BSI is a very broad measure; it comprises 9 primary symptom dimensions, as well as three global indices of distress: the Global Severity Index (GSI), the Positive Symptom Distress Index (PSDI), and the Positive Symptom Total (PST). For this study, the GSI was considered the most useful index, as it reflects both the number of symptoms as well as the intensity of distress. Participants were asked to fill this questionnaire in every week. In the study sample, the GSI had a reliability of $\alpha = 0.938$.

Experiences in Close Relationships – revised

Adult attachment style was assessed at baseline, using the Experiences in Close Relationships – revised [54]. This 36-item self-report questionnaire measures attachment anxiety and attachment avoidance (18 items each, 7-point Likert scale). Scores range from –54 to 54 per subscale, with higher scores representing higher attachment anxiety or avoidance. The attachment anxiety scale had a reliability of $\alpha = 0.76$ and the attachment avoidance scale reached $\alpha = 0.915$.

Participants' experiences

Oral feedback on the program was asked from participants after each evening, and written feedback was collected via Qualtrics XM Experience Management Software after completion of the HmT program.

Statistical analysis

All the questionnaires were sent out and filled in electronically and the data were stored using Castor EDC.

Continuous variables were described using mean and standard deviation while categorical variables were described using percentages. Baseline differences among the three intervention groups were analyzed using one-way ANOVA to ensure that group allocation was indeed random and that all three groups were comparable at baseline. Admittedly, with groups of 10 persons an ANOVA is underpowered, but it was decided to include this information, as crass differences would have been detected. Intraclass correlation of each outcome at baseline was determined to assess how much of the variation in the individuals' scores stemmed from random variation. High intraclass correlation would signify that participants filled out their questionnaires with almost exactly the same scoring pattern every week.

Since the RES was only filled in at baseline and at the end of the study, the intervention effect was evaluated using paired *t*-test once to assess pre-post differences.

Linear mixed effects models were used to analyze the changes of GSI and DAS total scores over time and evaluate the effect of the intervention while capturing within subject correlations. The main effects of time (in weeks), intervention (time-varying variable intervention indicator variable) and their interaction were included as fixed effects. Within-subjects correlations over time were modelled using random intercepts and random slopes terms and both model specifications were compared using likelihood ratio tests. Significance was assumed at $\alpha = 0.05$ (two tailed).

Linearity of the time effect was checked by running a regression with only the time variable. Normality was assessed through QQ-plots.

The potential moderation effect of attachment was evaluated including an interaction term in the linear mixed effect models for GSI and DAS. For the RES, classical moderation analysis utilizing regression analyses was done, as outlined by Baron and Kenny [55]. Note that these procedures are analogous, just that linear mixed effects models can deal with time-sampled data, and classical moderation analysis is described with only 3 parameters (one independent variable, one dependent variable, and one (or more) moderators).

Analyses were performed in IBM SPSS version 25.0 [56] and R version 3.6.1 [57]. SPSS was used for descriptive analyses, comparisons at baseline, and pairwise *t*-tests for the RES data. The R package *lme4* was used for mixed effect modeling of the GSI and the DAS data [58]. For other functions, the packages *psych* [59], *foreign* [60], *ggplot2* [61], *lmerTest* [62], and *jtools* [63] were used.

Table 1
Demographic and biographic characteristics of participants

Age (y)	41.73 (10.77)
Duration of relation (in y)	16.10 (11.00)
Marital status (per couple)	
Married	12
Documented civil union	1
Undocumented civil union	2
Couples with children	11
Average number	2.18 (1.14)
Education	
Lower secondary education	7
Intermediate secondary education	1
Higher secondary or professional education	11
University degree	11
Job status	
Employed	29
Studying	1
Time since gene test (y)	3.8 (3.17)
Calculated years to onset	17.4 (8.37)
Comorbidities	7
Neurological	1
Oncological	2
Psychiatric	4

Calculated years to onset according to Langbehn et al. [3]. Values represent mean (standard deviation) or absolute numbers if not followed by a standard deviation.

Table 2
Descriptive statistics for all questionnaires over the course of the study

	T1	T17
DAS	105.8 (16.3)	113.3 (13.3)
RES	25.23 (3.27)	26.2 (6.65)
GSI	0.38 (0.29)	0.24 (0.24)
ECR-R; Attachment anxiety	–1.52 (0.63)	
ECR-R; Attachment avoidance	–1.73 (0.76)	

Values represent means (standard deviations). Attachment anxiety and avoidance were only measured once, as they represent trait variables. DAS, Dyadic Adjustment Scale; RES, Resilience Evaluation Scale; GSI, General Severity Index of the Brief Symptom Inventory; ECR-R, Experiences in Close Relationships – Revised.

Table 3
Coefficients in the DAS model

	b	95% *CI*	*SE*	*p*	
(Intercept)	–6.25	[–11.860; –0.666]	2.76	0.03	*
Week	0.61	[0.397; 0.814]	0.11	< 0.0001	***
Phase	2.8	[1.468; 4.137]	0.68	< 0.0001	***
Week*phase	–0.27	[–0.374; –0.164]	0.05	< 0.0001	***

Values represent unstandardized regression coefficients estimated from the multilevel models. $^{*}p < 0.05$, $^{**}p < 0.01$, $^{***}p < 0.001$.

RESULTS

Demographic and biographic variables

The study sample consisted of 30 participants (15 couples). Participant characteristics are presented in Table 1. Overall, the study sample appears to be representative of the population that is seen in a setting of predictive genetic testing for HD in the center where the study was conducted.

Relationship Satisfaction (via DAS)

At baseline, the average relationship satisfaction score was 105.8 out of 151 ($SD = 16.3$), representing a medium-to-high relationship satisfaction; see also Table 2. No significant differences in relationship satisfaction were found between the three groups ($F = 0.764$, df = 2, $p = 0.476$). Partners scored higher on relationship satisfaction than HD-carriers at baseline ($M = 108.9$ vs. $M = 103.36$). The length of the relationship barely correlated with relationship satisfaction ($r = 0.078$).

The intraclass correlation of the null-model was $r^2 = 89.2\%$, indicating a high level of within-individual correlation of relationship satisfaction scores over time, i.e., a high stability in the answer patterns of each person. Linearity and normality assumptions were fulfilled.

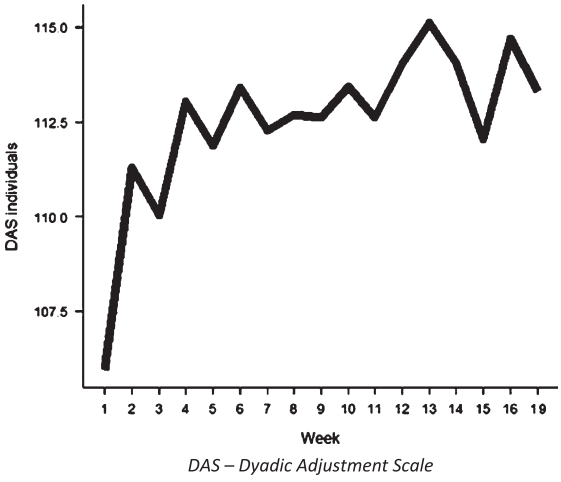


Fig. 4. Course of relationship satisfaction (DAS) over time (study weeks).

At the end of the study (week 19), after all participants of the three groups had completed the intervention, the mean relationship satisfaction score was $M = 113.3$ ($SD = 13.3$). Both time and group-phase (pre-intervention, intervention, or post-intervention), as well as their interaction, were significant predictors of relationship satisfaction (see Table 3 and Fig. 4). Group-phase, which represents the intervention effect, had a strong contribution to relationship

Table 4
Coefficients in the GSI model

	b	95% *CI*	*SE*	*p*	
(Intercept)	0.145	[0.036; 0.254]	0.054	0.012	*
Phase	–0.070	[–0.110; –0.030]	0.020	0.001	***
Week	–0.013	[–0.019; –0.007]	0.003	< 0.001	***
Week*phase	0.006	[0.003; 0.009]	0.002	< 0.001	***

Values represent unstandardized regression coefficients estimated from the multilevel models. $^{*}p < 0.05$, $^{**}p < 0.01$, $^{***}p < 0.001$.

satisfaction, with $b = 2.8$, $p < 0.001$. The correlation between slopes and intercepts was $r = -0.45$.

Partners had higher scores for relationship satisfaction than HD-carriers ($M = 113.9$ vs. $M = 112.7$).

Resilience (via RES)

At baseline, participants had a mean resilience score of 25.23 ($SD = 3.27$, see Table 2). There was no significant difference between the three groups at baseline ($F = 0.863$, df = 2, $p = 0.433$). With a mean of $M = 26.2$ (SD = 6.65) in the second and last measurement, after all participants had completed the intervention, resilience did not improve significantly over the course of the study ($t = –0.669$, 95% CI = [–3.92, 1, 99], $p = 0.509$).

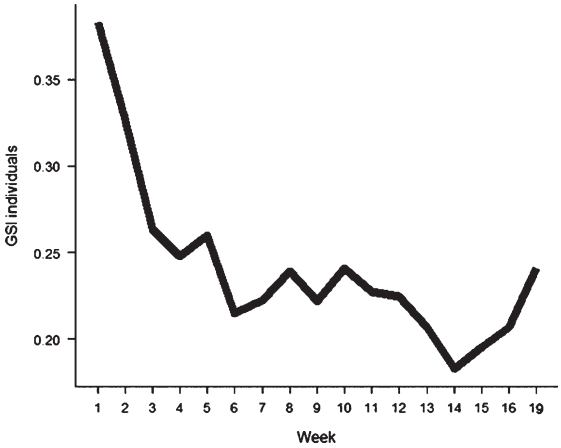
Wellbeing (via GSI)

At baseline, participants had a mean wellbeing score of 0.38 ($SD = 0.29$). The three groups were not systematically different in level of wellbeing ($F = 0.41$, df = 2, $p = 0.668$). Both time and group phase, as well as their interaction, significantly predicted wellbeing (see Table 4 and Fig. 5). The intraclass correlation was $r^2 = 86.2\%$, indicating a high correlation of scores within individuals over time. Linearity and normality were fulfilled.

The GSI score improved over the course of the study, with an average score of $M = 0.24$ (SD = 0.24) in week 19 (See Table 2 and Fig. 5). The subject-specific slopes and intercepts correlated with $r = –0.66$: persons who scored higher on wellbeing at baseline showed a smaller increase in wellbeing over the course of the study than persons with lower wellbeing scores at baseline.

Influence of attachment

As measured at baseline, participants scored $M = –1.52$ (SD = 0.63) on Attachment Anxiety and $M = –1.73$ (SD = 0.76) on Attachment Avoidance. This shows that the sample was mostly securely



GSI, General Severity Index of the Brief Symptom Inventory

Fig. 5. Course of wellbeing (GSI) over time (study weeks).

attached. A correlation of $r = 0.5$ was observed between the subscales, meaning these subscales are not independent of each other.

For attachment anxiety, persons with higher anxiety had lower relationship satisfaction ($b = –9.46$, 95% CI = [–16.67; –1.50], $p = 0.02$), but the moderation effect was not significant at $b = 0.36$ (95% CI = [–0.69, 1.38], $p = 0.5$). Persons with higher attachment avoidance were shown to have lower relationship satisfaction ($b = –7.1$, 95% CI = [–13.4; –0.75], $p = 0.03$), but there was also no significant moderation effect (interaction $b = 0.16$, 95% CI = [–0.69; 1.00], $p = 0.72$).

Attachment anxiety did not moderate the relation between intervention phase and resilience (interaction effect: $b = –0.437$, 95% CI = [–1.75; 0.87], $p = 0.499$). Persons with higher attachment anxiety had lower resilience at the end of the study ($b = –4.13$, 95% CI = [–8.14, –0.12], $p = 0.044$). However, attachment avoidance moderated the course of resilience (interaction term: $b = 1.606$, 95% CI = [0.61; 2.61], $p = 0.003$), changing the sign of the main effect of time (without moderation: $b = –0.326$, 95% CI = [–1.22; 0.57], $p = 0.461$; with moderation $b = 0.362$, 95% CI = [–51; 1.24], $p = 0.403$) and enlarged the contribution of the main effect of attachment avoidance (without moderation: $b = 0.043$, 95% CI = [–3.83; 3.91], 0.982; with moderation: $b = 1.769$, 95% CI = [–1.71; 5.25], $p = 0.306$). The moderation had an explained variance of 56.2% (vs. 16.6% of explained variance of the model without moderation).

Attachment anxiety significantly predicted wellbeing, with persons with lower levels of attachment

anxiety experiencing better wellbeing ($b = 0.18$, 95% CI = [0.05; 0.31], $p < 0.01$). There was no moderating effect of attachment on the course of wellbeing in the different experimental phases (interaction effect: $b = –0.006$, 95% CI = [–0.04; 0.02], $p = 0.68$). Lastly, attachment avoidance was significant on top of the already presented general wellbeing model (main effect: $b = 0.13$, 95% CI = [0.01; 0.24], $p = 0.036$). The moderating effect was significant, with persons with lower levels of attachment avoidance experiencing lower wellbeing ($b = –0.034$, 95% CI = [–0.06; –0.01], $p < 0.01$).

Participant comments

The reactions to the intervention were highly positive. Participants commended the online setting. Several persons said it was pleasant to meet "companions in fate" (Dutch: *lotgenoten*) or "people who are in the same boat", and that they enjoyed contact with other couples going through similar hardships. The setting and atmosphere were described as safe and trustworthy. Participants said they thought the intervention was useful and worthwhile and some mentioned that they found it interesting to hear about scientific approaches behind relationship therapy and HD. Some did note that they thought the sessions were too frequent and they were tired in the evenings, while others said they enjoyed these evenings.

DISCUSSION

This study indicates that the 'Hold me Tight' relationship enhancement program effectively improves relationship satisfaction and wellbeing for couples facing a future with HD. In the HmT program, couples take time to talk with each other about their unmet attachment needs and discuss what they both emotionally need from each other, especially in difficult times. Participants' comments after completing the program show that talking about their emotional needs, in a way that is different from their usual interactions, helps couples perceive more trust in each other. This may explain their increased satisfaction in the relationship, which may also contribute to more wellbeing. This study extends previous works on the usefulness of HmT in medical populations [40, 41] and is the first of its kind for a hereditary neurodegenerative disease. Furthermore, it adds to the scarce literature about interventions addressing the family and partnership dynamics in a genetically transmitted condition. Moreover, it underlines that an online HmT intervention can be successful, which makes it more applicable in a population with a rare condition such as HD, as a group may be formed from persons throughout the country.

Both relationship satisfaction and wellbeing were higher after having completed the HmT program than at baseline, confirming hypotheses 1 and 3. Despite the potential 'learning effect' from filling in the same questionnaires weekly, causing high correlation of the repeated scores of an individual, significant improvements in these outcome measures could be detected.

In this study, no significant increase in resilience was found, so hypothesis 2 was not confirmed. As resilience is considered to be a trait-like characteristic and would not be expected to vary from week to week, it was only measured twice. The study was probably underpowered to find effects with this kind of analysis. There were 30 participants in total, which is enough in a multiple baseline design, but the resilience measurements were not subject to the multilevel models that exploited the advantages of the multiple baseline design. This means that even if there were an effect, it would not have been detected. Studies with larger samples might be able to find significant improvements in resilience after completing a HmT program.

In contrast to expectations (hypothesis 4), persons with a more insecure attachment style were not found to benefit more from the intervention. This may be explained by the levels of attachment anxiety and avoidance in the sample being too low to detect any noteworthy patterns. It is still possible that the hypothesis is true; more robust studies might be able to show this. Nonetheless, the intervention is useful for participants regardless of their attachment style.

Partners did not have lower relationship satisfaction than HD-carriers, in contrast to expectations based on previous studies [9, 10]. The length of the relationship did also not seem to play a role in satisfaction ratings (as previously found in [14]). Perhaps the expectations people have towards relationships have changed in the last 15 years—maybe certain abilities and tasks in everyday life used to be more associated with one gender than with the other, which is why the partner may have felt additionally helpless when the HD-carrier shows signs of decline. Nowadays, with more gender parity, people learn more abilities associated with the other gender. Thus, people may feel less overwhelmed with managing everyday tasks when one partner cannot participate anymore.

In total, this study advances the understanding of the value of attachment and strong relationships in a rare genetic adult-onset condition. Family life is an important environmental factor in HD: from growing up with the omnipresence of HD, to needing to disclose your status to a partner, to worrying about transmitting it to your child. This is why HmT is a well-fitting intervention for HD: it focusses on the very attachment needs (triggered by low relationship satisfaction) that are often left unmet when a partner's health starts deteriorating and when relationship fears arise that are difficult to share, both with each other and with others (see [9]). Arguably, carrying out this intervention in the presymptomatic phase is also beneficial, because, as mentioned above, HD patients have been shown to have lower Theory of Mind and sensitivity towards their partner's needs [13]. By doing HmT this early, couples are investing in their sensitivity towards each other in the future. Having learned to explore and discuss their emotional needs in this early stage, they are likely more successful at navigating the difficulties lying ahead of them. Furthermore, HmT can foster sensitive parenting, thereby improving the parent-child relationship in families with HD, and allowing children in such families a healthy development [23]. An interesting avenue for future research would be to determine the role of the mutation-carrying parent's age of onset on resilience and attachment. In the present study, no information on this was gathered, but previous studies indicate that being exposed to the parent's disease process at a younger age increases the risk for insecure attachment and may therefore negatively influence resilience and wellbeing [23, 24, 64].

HmT is an easily implementable intervention. It is highly feasible in practical terms, as it can be done online or in person and does not need any special materials, apart from Sue Johnson's book and materials from the EFT website. It can be effectively carried out online [49]. From the participants' comments, it can be concluded that it has a high face validity. Furthermore, by strengthening wellbeing and relationship satisfaction, HmT should also contribute to better communication with healthcare providers, more independence in everyday activities, and should sustainably lower distress [9, 14, 15].

As a result, it can be assumed that HmT could also be effective in other genetic neurological conditions, such as cerebral autosomal dominant arteriopathy with subcortical infarcts and leukoencephalopathy, hereditary cerebral amyloid angiopathy, familial amyotrophic lateral sclerosis, and familial dementias. Future research with these populations is recommended. Moreover, longer follow-up would be needed, to see whether the favorable effect of the HmT program is long-lasting, or whether there should be a refresher program to consolidate the positive outcomes of HmT after some time. Given the limited time and resources, this was not feasible in the present study. It will be interesting to see the results of the Healing Hearts Together trial (clinicaltrials.gov: NCT03938116, [40]), which has a very similar setup to the present study and will evaluate the effects at 6 months post intervention. Conradi and colleagues found small-to-medium effects in a non-medical sample at 3.5 months post intervention, so it might be expected that this would also be the case in the present sample [44].

A limitation of the study is the high dropout-per-timepoint rate, which is partly explained by the fact that recruitment was continued until the needed number was likely to be reached. It is noteworthy that none of the participants dropped out once they had started the HmT program. Couples invested four weekly evenings in the program, accepting practical consequences (rescheduling work, cooking, having dinner, finding a private place to talk, etc.), and expressed in general that this was well worth the effort.

Participants also told us that it was often tiring to have a roughly 3.5-hour intervention after work, and occasionally they were distracted by children or pets. On the other hand, participants also gave highly positive feedback. Most participants mentioned that they enjoyed exchanging experiences, advice, and thoughts with other couples, therefore, keeping a group setting for future interventions is recommended. A large part of the sample also mentioned that they especially liked the session where the HD-carriers and the partners had separate group conversations. Participants rated the usefulness of the intervention highly.

Another limitation to the study may have been the online setting. Participants may have felt less commitment to the study than they would have if they had physically needed to attend. However, the online setting was also an advantage in disguise. Since HD is such a rare disease, it would be difficult to find enough participants from one region. By moving the intervention online, couples from all over the Netherlands were able to participate. Lastly, a recent study has shown that emotionally focused therapy can easily be adapted to an online setting, making it an effective tool to manage relational discord even at a distance [49].

To summarize, based on the present study, the 'Hold me Tight' relationship enhancement program appears to effectively support couples facing a future with HD. Both personal wellbeing and relationship satisfaction within the couple can be improved by applying this program, regardless of their attachment style. This program could be a valuable addendum to standard psychological care after receiving an unfavorable result in genetic testing in neurodegenerative disorders.

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CONFLICT OF INTEREST

The authors report no conflict of interest.

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