Chapter 6

Suicidality in Huntington's disease: a qualitative study on coping styles and support strategies

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Submitted

Abstract

Background: Huntington's disease (HD) mutation carriers are at increased risk of suicidal ideation, suicide attempts, and completed suicide. However, research is lacking on coping strategies and treatment options that can be offered to suicidal HD mutation carriers. This study explores how HD mutation carriers cope with suicidality, how their partners support them, and their ideas and wishes regarding how relatives and healthcare professionals can help them in coping with suicidality.

Methods: This qualitative study included 11 HD mutation carriers who experienced suicidal ideation or attempted suicide and 3 of their partners. They participated in a focus group discussion or an individual in-depth interview. Two independent researchers fragmented the transcribed interviews, coded these fragments, and grouped them under themes.

Results: HD mutation carriers used four main strategies to cope with suicidality, including talking about suicidality, employing self-management activities, using medication, and discussing end-of-life wishes. Partners, relatives, and healthcare professionals can support suicidal HD mutation carriers in each of those four strategies.

Limitations: Although data saturation was reached within this sample, we included only a limited and selected group of HD mutation carriers and their partners.

Conclusion: Despite the absence of a turnkey solution for suicidality in HD, healthcare professionals can play an important role in supporting suicidal HD mutation carriers by providing an opportunity to talk about suicidality, providing psychoeducation on self-management, prescribing medication, and discussing end-of-life wishes. Future HD-specific intervention studies could investigate the effect of combining these treatment strategies into one holistic approach.

Introduction

Huntington's disease (HD) is an autosomal dominant progressive neurodegenerative disease.[1] An expanded CAG repeat on the short arm of chromosome 4 causes a mutant polyglutamine chain in the huntingtin protein.[2] The disease is characterised by motor dysfunction, cognitive decline, and neuropsychiatric symptoms. Although some of these symptoms can be treated, the disease cannot be cured.[3]

An important psychiatric sign of HD is the increased risk of suicide, as first described by George Huntington in 1872.[4] More recent studies showed that HD patients were 2-8 times more likely to die by suicide than individuals from the general population,[5-8] which accounts for up to 11.4% of all deaths in motor symptomatic HD patients.[5-24] In addition, lifetime suicide attempts have been reported by 3.2-17.7%[7;9;10;12;13;17;18;25-31] of the mutation carriers, compared with only 2.7% in the general population.[32] Furthermore, 8-34% of the HD mutation carriers experienced some form of suicidal ideation in the month prior to the interview,[25;28;31;33-36] compared with 0% of the first-degree non-carrier relatives.[33] These numbers emphasise the frequent occurrence of suicidal ideation and suicide attempts (together referred to as 'suicidality') in HD. Suicidality is associated with lower quality of life[37;38] and an increased occurrence of completed suicide.[39;40]

Both suicidal ideation and completed suicide occur most frequently in early-to-middle disease stages.[5;7;8;11;31;34] In addition, depression is reported to be the strongest association of suicidality in HD.[26;28;29;31;33;41] Mutation carriers with other psychiatric symptoms like irritability/aggression,[28;31] obsessive and compulsive symptoms,[30] and a previous suicide attempt[31;41] as well as those who used antidepressants[33] or benzodiazepines,[31] were also more likely to experience or develop suicidality.

Despite the high frequency of suicidality in HD, limited information is available about coping with or the treatment of suicidality in HD, comprising only a few case descriptions of suicidal HD patients who were pharmacologically treated.[42-44]

Therefore, we conducted a qualitative study among HD mutation carriers who had experienced suicidal ideation or attempted suicide and among their partners. The primary aim was to examine how HD mutation carriers cope with suicidality and their ideas and wishes regarding how relatives and healthcare professionals can help them cope with suicidality. An additional aim was to explore how the spouses of HD mutation carriers support their partners with regard to suicidality.

Methods

Recruitment procedure and participants
This qualitative study was conducted among HD mutation carriers recruited between February and May 2014 and who had previously participated in a Dutch prospective cohort study on behavioural problems and psychiatric disorders in HD.[45]

Of all mutation carriers who 1) had participated in the last follow-up measurement of the aforementioned cohort study, 2) had given consent to be contacted for future research, and 3) were known not to have died (n = 90), 40 had reported the presence of thoughts of death, suicidal ideation or a previous suicide attempt at one of the measurements of the cohort study. Of these 40 HD mutation carriers, 11 consented to participate in the present qualitative study (Figure 1). If possible, participation in a focus group discussion was the preferred study method.[46;47] Eight mutation carriers participated in a focus group discussion (divided over 2 groups of 4 participants each) and 3 in an individual in-depth interview.

The focus group discussions were analysed first, followed by the individual interviews. Data saturation was reached after these focus group discussions, as no new understandings/ information on support strategies emerged from the individual interviews.[46]

All participants were asked whether they had a partner and, if so, whether he/she knew about their suicidal thoughts. After consent, we contacted their partner for participation in a focus group discussion. Of the 4 partners of participants that we could contact, 3 were willing to participate.

This study was approved by the Medical Ethical Committee of the LUMC and all participants provided written informed consent.

Focus group/individual interview for HD participants
Before the focus group discussion/individual interview, each HD participant had an individual intake appointment to explain: the purpose of the study, the topic list, and the course of the focus group discussion/individual interview. In addition, the presence and severity of depressed mood and suicidality in the last month were assessed by the items 'depressed mood' and 'suicidal ideation' of the Problem Behaviours Assessment (PBA).[25] Also, lifetime suicidality was assessed by the Columbia-Suicide Severity Rating Scale (C-SSRS).[48]

The actual focus group discussions/individual interviews were conducted by a psychiatrist

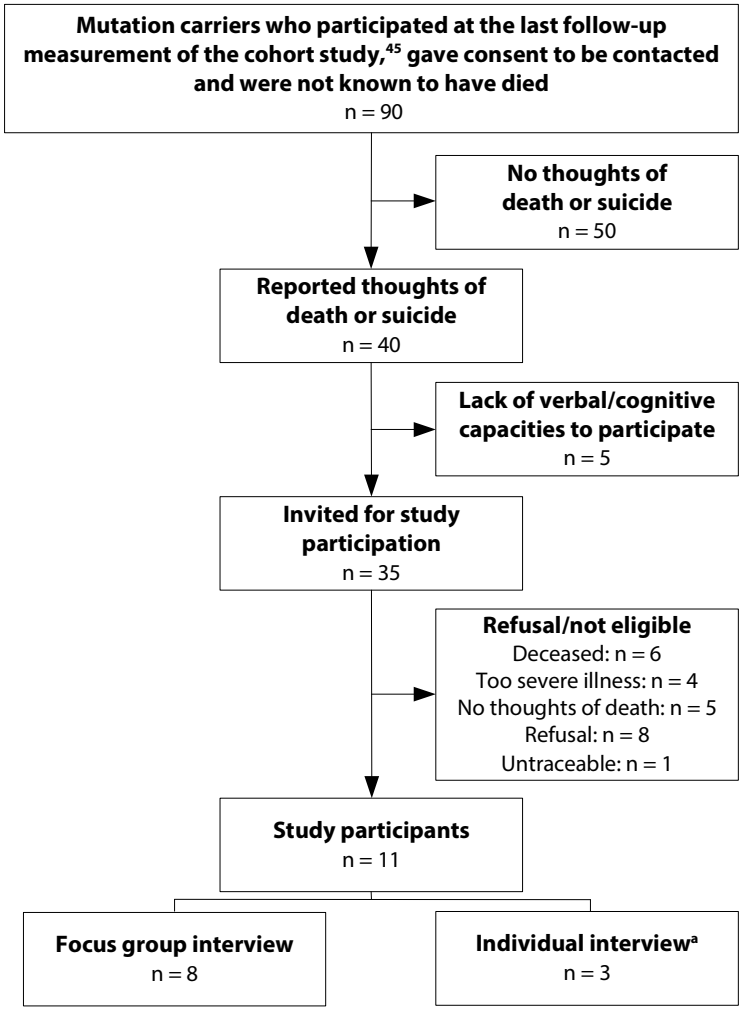

Figure 1. Selection of HD study participants
[a] Two participants were considered too ill to participate in a focus group interview and one participant did not want to talk about her experiences in a group.

(moderator/interviewer) with longstanding experience in the study and treatment of HD mutation carriers (EvD). This researcher was specifically trained in moderating focus groups for the present study. He was also the treating psychiatrist of one of the participants of the focus group discussion. The first author (AAMH), who is experienced in interviewing HD mutation carriers, conducted the intake appointment and was present as note taker during the focus groups and individual interviews. The focus groups lasted 1.5-2 h (including a short break),

and the individual interviews lasted 15-60 min each. During the focus groups and individual interviews, we mainly referred to 'thoughts of death', and explained that this term could cover the whole spectrum of suicidal thoughts: ranging from not wanting to live anymore should the disease become more severe, to actual suicide attempts. The moderator/interviewer ensured that the whole range of thoughts of death was discussed. To introduce this sensitive topic, the focus group discussion/individual interview started with an open question about how participants felt about this study actually being conducted. Subsequent questions were based on what the participants said themselves. A topic list was used as a reminder for the moderator/interviewer and consisted of 3 main items: how the mutation carriers themselves coped with suicidality; how relatives could support suicidal mutation carriers; and how healthcare professionals could help suicidal mutation carriers.

Focus group for partners

The focus group discussion for partners lasted ± 80 min (including a short break). The session started with an open question regarding how participants felt about this study being conducted and subsequent questions depended on the participants' responses. The topic list was based on results of the mutation carrier focus groups and individual interviews, and covered the following items: discussion of suicidal thoughts with partner; support of suicidal partner; and advice for future partners of suicidal HD mutation carriers.

Data analysis

All focus group discussions and individual interviews were recorded on tape and transcribed ad verbatim. The analyses started with fragmenting the transcripts of the HD mutation carriers and open coding of the fragments by two independent researchers (AAMH: medical PhD student and AH: physician). The initial code list included only 3 codes: coping themselves, support from relatives, and help from healthcare professionals, as these were the items on the topic list. Codes were grouped into themes; this involved independent and close examination and comparison of the transcripts by the two researchers. After the coding of each transcript, these researchers discussed their fragments, codes, and themes until consensus was reached, constantly taking into account the previous transcripts. Examples of new codes emerging from the data were 'listening' and 'trust' which were grouped as 'factors influencing talking', which was grouped under the major category 'talking about suicidality'. Having discussed all the transcripts of the HD participants, both researchers independently recognised that all codes and themes which described coping and support strategies could be grouped into four major categories. Subsequently, they independently read and interpreted the fragments that belonged to these four categories. Thereafter, the partner focus group transcripts were coded and interpretive reading was conducted to analyse what the partners had said about these

four major categories. Atlas.ti 7.5 (Scientific Software Development GmbH, Berlin, Germany) was used to code the interviews.

Results

Study population

The disease stage of the participants ranged from self-reported pre-motor symptomatic to participants who had to live in a nursing home. There were three male mutation carriers and one male partner, the age of the participants ranged from 32-71 years, and the number of CAG repeats ranged from 41-47. Table 1 presents the scores on the 'depressed mood' and 'suicidality' items of the PBA.

Table 1. Clinical characteristics of the study participants with Huntington's disease.

Participants	Lifetime thoughts of death[a]	Lifetime thoughts of suicide[a]	Lifetime suicide plan[a]	Lifetime suicide attempt[a]	PBA depressed mood severity[b]	PBA suicidality severity[b]
M1	yes	yes	no	no	0	0
M2	yes	yes	no	no	2	1
M3	yes	yes	yes	no	2	0
M4	yes	yes	no	no	3	1
M5	yes	yes	yes	yes	0	0
M6	yes	yes	yes	yes	3	1
M7	yes	yes	no	no	3	1
M8	yes	yes	no	no	3	0
M9	yes	yes	no	no	0	0
M10	yes	yes	no	no	3	3
M11	yes	no	no	no	0	0

M indicates mutation carrier; PBA, Problem Behaviour Assessment

[a] Lifetime thoughts of death, thoughts of suicide, suicide plan, and attempts as assessed by the Columbia Suicide Severity Rating Scale lifetime.[48]

[b] PBA depressed mood and suicidality severity scores in the last month (range 0-4), with higher scores indicating more severe depressed mood or suicidality.[25]

Support strategies for suicidality in HD

First of all, participants indicated that there is no turnkey solution for suicidality in HD. They mentioned several ways of support that can be grouped in the following categories: 1) talking about suicidality, 2) self-management, 3) medication use, and 4) discussing end-of-life wishes.

1. Talking about suicidality

All participants stressed that they had a great need to talk about their suicidality, even though this was considered difficult and the threshold to do so was high. This is illustrated by one of the participants who said: *"Someone who thinks about death already has difficulty expressing this ... and has feelings of self-disapproval because of this thought [….] But my experience is that it really helps when you eventually do talk about everything - with all your sorrows and misery. This always gives relief."*

Table 2 shows factors that, according to the participants, could facilitate or complicate talking about suicidality. Listening, taking the thoughts seriously and not trivialising these thoughts were the most important factors in helping them to express their feelings to a relative or healthcare professional. The preferred person to talk to varied per participant and included their partner, a family member, friend, care manager, psychologist, or hypnotherapist. Some participants stressed that this person should not be too close, and they preferred someone who is *"a relative outsider"*, because they did not want to hurt their partner or children by telling them about their suicidal thoughts. However, the spouses mentioned that they did want their partner to tell them about suicidal ideation, as was stated by a female spouse: *"I don't want him to think about that all on his own - I think that's far too lonely"*, although the spouses admitted that their partner's expression of suicidal ideation was also a burden on their own lives. When the person to talk to was less close (either a professional or a non-professional), a trustful relationship needed to be established before the mutation carriers felt 'close' enough to talk about suicidal ideation, although the extent of this relationship differed per participant. Furthermore, some participants found it difficult when family members who experienced the same disease process did not want to talk about the disease at all, as illustrated by one participant: *"You'd think you'd have a good relationship with someone […] somebody who lives the same sort of life as you, but who's really very different."*

Participants stated that positive experiences with talking to someone about suicidal thoughts resulted in a decrease of suicidal ideation, and in relief, improved self-esteem, and increased ability to see the positive things in life. A few participants who read up on the disease since the moment they became aware of their genetic status recognised that they had now learned to think more positively when they felt depressed for a longer period, or when someone pointed

out the good things in life. However, most other participants felt that the 'helping person' ignored their suicidal thoughts when they simply responded by pointing out the positive things in life. This difficulty experienced by persons trying to give support is illustrated by one of the partners who said: *"If you even make a small reference to something positive […] this works in a counterproductive way [….] what you're really trying to do is cheer the other person up"* and sometimes it is better *"not go into it"* but *"just be there [for your partner]"*. Also, the mutation carriers themselves mentioned that non-verbal signs of support, like someone putting an arm around you, can be very helpful. Negative experiences with talking about suicidal ideation resulted, for example, in withdrawing from the contact *"which made me feel lonely and yes, that made everything worse."* Others mentioned that they had a negative experience with too much talking about suicidality to too many different healthcare professionals. One mutation carrier attributed this to the fact that she had not yet learned *"to let go"*. The delicate balance between actively facing HD/suicidality on the one hand and taking your mind off suicidality/HD on the other was also important with regard to self-management.

Given the importance of talking about suicidality, the moderator specifically asked participants whether others should actively address the presence of suicidal thoughts. The mutation carriers pointed out that it is best to ask about their mood when others see that they are sad or doing less well; also, when the mutation carrier indicates a depressed mood, questions about suicidality could be asked (Table 2). Partners acknowledged that it is difficult to find the right moment to talk, e.g. due to a busy daily life, and because they did not want to introduce this painful topic when their partner was feeling well. Given the emotional burden for both mutation carriers and partners, one of the partners advised other partners to try and talk about the suicidality during multiple conversations, as a person can only handle a certain amount each time. One participant could talk with her partner about suicidality very well because he *"allowed me to talk about it and he took the time to discuss it."* Talking about suicidality was also difficult for the partners themselves, particularly when they had to decide on the amount of control they wished to exercise in order to prevent suicide. Partners themselves also needed to talk about suicidality with others, which one partner did by joining a peer support group, whereas another partner had negative experiences with such a group because of all the unpleasant stories she heard about what was still to come.

Most participants agreed that healthcare professionals could (or even should) ask about suicidal thoughts, even though they might feel *"taken by surprise"*. This topic should be introduced delicately by asking about sad feelings and, if present, follow-up questions should be asked (Table 2). In case of suicidal thoughts, healthcare professionals should guarantee further possibilities to talk about suicidality, e.g. by referral to a psychologist. Both mutation carriers

Table 2. Factors mentioned by participants that facilitate or complicate talking about suicidality to others (professionals and non-professionals).

Influencing factors	Example quotes
Reaction to expression of suicidal ideation	
Facilitate: Listening, taking thoughts seriously, accepting/allowing thoughts, showing empathy/ understanding, naming and asking questions (like: *"Why now?"*), embracing someone, being there for someone, holding hands, giving someone the time to see positive things themselves	*"For me it's important that other people take me seriously and don't immediately say things like: 'well, life is worth living' or 'look outside, the sun's shining' [….]For me it helps when someone just listens to what I'm saying and … that I can express my … well, my darkest thoughts."*
Complicate: Trivialising, neglecting, denying, panicking, bringing someone further down, distancing, only focusing on practicalities or distraction, giving advice that cannot be put into practice at home	*"I love horses and then they [the psychologist] say: 'Well, you should take up horse riding again.' Then I feel OK for a moment, but at home there's no money for this. So you feel good for a moment, but afterwards ... you can't do anything about it."*
Personality & relationship characteristics	
Facilitate: Feeling a 'click', trusting and feeling safe with someone, being able to be honest with someone, 24/7 availability to be called or reached, knowing about personal history, HD knowledge (HD-specific therapist), no explaining necessary, can be an outsider	*"with thoughts of death … that you actually only need to press a button and then you can talk to someone immediately"* *"just one person that you can be totally honest with"*
Complicate: Very close person whom you will hurt	*"Because at the moment you want to talk about this matter - to my wife - and then I know, oh yeh, that will hurt my wife and that's just what you're not going to do."*

Circumstances	
Facilitate: Taking time, good environment (e.g. in a forest), regular appointments (with healthcare professional), visiting someone to talk to	*"Well, it's like you're a sort of onion - and all the layers need to be peeled off [….] bit by bit actually - and that's what you can't do if you're only once with the doctor."*
Complicate: Costs (for healthcare professionals), limited time (for healthcare professionals)	*"I think that if you ask that question you need to have a lot of interest and concern for the patient … but doctors' appointments are usually only 10 minutes, 20 minutes, or half an hour, but in this sort of a case you should be willing to run over the usual allotted time."*
Follow-up	
Facilitate: Referral to psychologist/psychiatrist/ care manager, regular follow-up meetings, asking how someone is getting on, calling someone up	*"I think that when he [the neurologist] notices this, then he should refer you to a psychologist […] or a care manager, which for me is the ideal solution. That's where it really all comes together."*
Complicate: Not following-up	*"I told my sisters and brothers that I … had suicidal thoughts. But … they didn't do anything about it [....] I thought they might have at least agreed to take it in turns to call me. Once a week, or something like that."*
Moment to address	
Facilitate: When others see you are doing less well, following-up on sad/depressed thoughts	*One participant: "Well he [the professional] could ask something like: Do you have depressing thoughts? And when someone confirms this then he [the professional] should keep on questioning and if someone says 'no' then maybe he/she doesn't have thoughts about death."* *Other participant: "Yes, usually it stops after that question. Indeed, they ask do you have depressing thoughts and when you answer yes or no they usually beat about the bush or something. Maybe they should take it one step further."*
Complicate: Out of the blue, when you come to the doctor for something non-HD and non-mood related.	*"Look, when you walk in happily about something else and they look like they're thinking - 'oh that's a Huntington's patient' - then you shouldn't do it [ask about suicidal ideation]."*

and spouses emphasised that healthcare professionals should have adequate knowledge on HD and *"be familiar with the whole picture"*. A combination of individual therapy for the mutation carrier and therapy sessions that partners could also attend was preferred by the participating spouses.

2. Self-management

Participants mentioned many different activities that they used to cope with suicidality and with HD itself. These activities could result in increased self-esteem and decreased stress and thoughts of death. Most of these activities could be grouped as either 1) actively facing suicidality/HD (e.g. talking about suicidal ideation/HD, gaining information on HD, realising that you are not the only one, contacting an HD support centre, making a crisis plan, setting future goals, healthy living, and hypnotherapy), or 2) taking their mind off it, mainly by exercise and relaxation (e.g. yoga, meditation, going outside, cycling, walking, distraction, focusing on daily life, social contacts, family life, taking rest, and creative therapy). *"The art"* of *"balancing"* these elements was considered important, since activities on both sides of this spectrum could become too extreme (Figure 2) and then worsen their feelings. Reducing suicidal thoughts and getting back in balance after too much confrontation with HD or thoughts of death (e.g. in family, by rumination, listening to sad music or watching films or TV shows about death) could be achieved by taking medication, calling someone, taking rest, letting go, taking distance, focusing on yourself, or having someone who can take you out. At the same time, for some participants, negative experiences were induced by too much avoidance and not wanting to face the disease which resulted in destructive behaviour like taking drugs and alcohol, aggression, or isolating oneself. At that point, participants said they need people who can *"pull you out of the mud, so to speak."* One of the participants gave the following advice: *"Don't stay at home worrying… It is a cliché, but it actually does help: go out for a walk with someone, or go cycling! Or … I don't know … if you stay at home in front of the TV all day, or behind the computer … if you isolate yourself, in my experience, it'll only get worse and worse."*

It was advised to employ such self-management strategies and create an environment of people you can talk to before the disease starts, as it is more difficult to do this when the disease becomes more severe or when someone is depressed. Coping styles witnessed in their own family also influenced how participants dealt with the disease themselves. Several participants criticised the coping styles of their family members, e.g. when they refused to talk about it and/or avoided everything that had to do with HD.

Figure 2. Balance between actively facing Huntington's disease (HD)/suicidality and taking one's mind off it.
Self-management strategies (see Results section) can be grouped into one of these categories; our participants considered it important to aim to keep both sides in balance.

3. Medication use

Almost all participants who used medication (mostly antidepressants) were positive about its effects on their mood and suicidal thoughts. Only one woman reported that she stopped using antidepressants because she always felt unwell after taking these pills. In her opinion the pills suppressed her feelings, and she emphasised that the *"the instruction leaflet already states […] suicidal tendencies."*

The reluctance of some participants to start taking medication was recognised by the participants who were currently using medication from the period before they started. Reasons for this reluctance were: potential side-effects and being unable to separate these from disease symptoms, fear of emotional suppression, and *"wanting to do it on your own strength"*. Some participants needed time and discussion with relatives to decide about starting medication. Participants who used medication for a longer period of time mentioned that they did not want to quit, because of its effectiveness.

Many participants who used medication combined this with talking to someone: the importance of this was stressed by one participant who described an appointment with a neurologist as follows: *"when I mention I feel depressed, [the neurologist says]: 'Well - here's the medication', whereas in fact he could say something like: 'Take this medication, and try and see a psychologist in a couple of days.' People might not always want to do this, but offering this, yes that can help some people [….] I think it often helps to literally say these things out loud to a psychologist or psychiatrist."* Partners recognised the positive effects of medication for various HD symptoms, and one partner stated *"without all of that [HD medication] I couldn't have coped for so long."*

4. Discussing end-of-life wishes

Some participants had a wish and/or advance directive for euthanasia in case the disease became very severe: in the Netherlands, under strictly controlled conditions, this is legal. The prospect of such a way out could help in coping with suicidal thoughts, as explained by one of the participants: *"I kind of made my own crisis plan: what to do if I think I want to jump in front of a train. Because … I don't want to end up that way. So I made arrangements about this [euthanasia directive] and that helps."* Most participants had seen examples of HD in their family and clearly knew what they did not want to experience (such as admission to a nursing home), but some also mentioned that their views might change. Also, some participants in an early stage of the disease did not know whether or not they would want euthanasia, or had not yet thought about this. Some found it difficult to decide at what point you need to think about end-of-life wishes: this was illustrated by one participant who was recently clinically diagnosed and

still fully occupied with accepting the functional decline he experienced: *"When should I start thinking about that [euthanasia directive]? [….] I don't want to face it yet, because I feel reasonably OK […] but there's a point when you'll go further downhill, and then … then you can't properly think about it."* These participants did not want the doctor to actively address euthanasia, or they considered it a topic that should first be discussed with their partner. However, another participant, who had thought about euthanasia since her genetic diagnosis, would like the doctor to address this. Also, one partner thought it was normal that the doctor had asked about euthanasia when his wife was admitted to a nursing home, since they had discussed this together since the beginning of the disease.

Those who expressed a wish for euthanasia emphasised the importance of this being their own choice. One participant described both her positive and negative experiences with healthcare professionals as follows: *"I was always used to the fact that healthcare professionals […] are focused on making sure you won't end your life [….] but at the moment it does help me that I can just be very honest about it and yes … that people understand. That maybe I don't want to continue living at some point in time … and, yes that this is my own choice and not … not someone else's."* The fact that the doctor might not want to carry out their request at the time they have reached this point, which some participants had witnessed in their close surroundings, worried them as *"you have no guarantee"*. Also, one partner expressed such doubts, but acknowledged she had no knowledge about the legal requirements for euthanasia. Once a participant had the confidence that his/her wish would be carried out, this provided some relief. Positive euthanasia experiences in the family strengthened this confidence, and further thoughts about end-of-life wishes were also influenced by family experiences. One participant realised that in an advanced disease stage he might not understand his situation and he wondered *"So who is euthanasia most important for? For me? ... Or for my wife and children?"*

Discussion

This qualitative study shows that HD mutation carriers use various strategies to cope with suicidality including talking about suicidality, employing self-management activities, using medication, and discussing end-of-life wishes. Relatives and healthcare professionals can support suicidal mutation carriers in each of those four strategies.

Coping styles and support strategies

In older non-HD populations with a death wish or suicidal ideation, physicians were less likely to consider treatment and avoided further conversations about it.[49;50] They felt they could not

help[50;51] and considered suicidal ideation in older people more normal and rational.[50] Although healthcare professionals treating HD might have similar feelings, our study participants emphasised the urgent need to talk about their suicidality, either with a healthcare professional or a relative. They also stressed that questions about suicidality should be asked in case of a depressed mood, which is the most important association and predictor of suicidality in HD.[26;28;29;31;33;41] Similar to non-HD populations, the majority of patients do not easily disclose their suicidal thoughts to healthcare professionals[52;53] and general practitioners only asked questions about suicide in 36% of patients with depressive symptoms.[54] Furthermore, both general practitioners and neurologists thought that less than 50% of the patients with suicidal ideation needed psychiatric assessment.[55] It was previously suggested that suicidal persons communicate their thoughts more often to relatives than to healthcare professionals.[56] The participating spouses stressed they wanted their partners to tell them about suicidal thoughts and wanted to be involved in the treatment. A two-hour education program for caregivers of suicidal patients was shown to result in improved caring ability, such as better communicative skills and a more positive attitude.[57] Also, it was shown that the involvement of a significant other in the treatment of suicidal ideation resulted in greater improvement on suicidality, depression, and hopelessness measures.[58]

Applying self-managements strategies while keeping the balance between actively facing HD/suicidality and taking one's mind off, was another strategy that helped mutation carriers to cope with suicidality. Several of the self-management strategies mentioned by the participants (e.g. relaxation and exercise) have been shown effective in reducing suicidal ideation[59-61] and/or depressive symptoms[62-66] in non-HD populations, including patients with a chronic illness.[63] Within HD, positive effects of relaxation[67;68] and exercise[69-72] on several clinical outcomes have been reported, including on depression.[70]

Case reports on treatment for suicidality in HD have only focused on pharmacotherapy;[42-44] positive effects were highlighted by our study participants who used medication. In non-HD populations, antidepressants (most frequently mentioned by our participants) have been shown effective in reducing suicidal ideation in patients with major depressive disorder.[73] Within HD, one case report showed that mirtazapine could reduce suicidal ideation[43] and a HD case series demonstrated the effectiveness of lithium in reducing suicidality.[42] Lithium was not specifically mentioned by our study participants, but in both non-HD unipolar and bipolar patients treatment with lithium resulted in a reduced suicide risk.[74] Pharmacotherapy for depression is recommended in guidelines for non-HD suicidal patients,[52;75] whereas in HD undertreatment of depression is likely[76] and there is insufficient evidence to guide treatment choice.[77]

Finally, discussing end-of-life wishes helped some participants to cope with suicidality; however, this is not one of the treatment strategies recommended in general non-HD guidelines on suicidality and, moreover, is illegal in many countries. It has been estimated that 7% of the affected HD patients in the Netherlands die by euthanasia or physician-assisted suicide.[78] In non-HD populations, euthanasia is often considered to be inconceivable for psychiatric patients with a death wish.[79] It was previously recommended that HD physicians address end-of-life wishes,[80] even though they often do not initiate this conversation.[81] In the present study, not all study participants wanted their physician to actively address euthanasia. As reported previously, [80;82;83] these patients do have thoughts about their future which are mainly based on experiences in their family[82] and are usually discussed first with the family.[80] It was beyond the scope of the present study to disentangle the best moment to address these thoughts. If a patient has a wish for euthanasia, a letter of intent,[81] repeated conversations,[80;84] a trusting relationship[80] and informing patients about legal criteria/requirements[80] were previously suggested strategies that could be offered by physicians. Although not specifically mentioned by participants in our study, this might have resulted in more peace of mind for some of our participants with a wish for euthanasia.

Strengths and limitations

This is the first study to investigate attitudes towards, and coping and support strategies for suicidality in HD, which included HD mutation carriers in various disease stages. Also, the use of focus group discussions facilitated the observation of differences of opinion between participants about the coping and support strategies that were proposed.

Some limitations need to be addressed. First, we included a small and selected group which limits the generalisability of the findings. Although data saturation was reached, some participants mentioned that their family members had more avoiding ways of dealing with the disease, but did not want to participate in research in general. Second, because only three partners participated, their data were used only to determine whether they had similar ideas about treatment strategies as the mutation carriers themselves. Third, the moderator and researchers who analysed the interviews had received medical education, which might have unintentionally focused their attention on strategies that are similar to those used in current clinical practice. Fourth, generalisability of the results to other countries might be limited, especially since (under specific circumstances) euthanasia is permissible in the Netherlands, whereas this is illegal in many other countries.

Clinical recommendations

Despite the absence of a turnkey solution for suicidality in HD, healthcare professionals can

play an important role in all four strategies, which are largely in line with general non-HD suicide guidelines. All healthcare professionals who treat HD mutation carriers should ask about suicidal ideation, especially when symptoms of depression are present, and refer them to a (preferably HD-related) psychologist or psychiatrist for regular sessions to talk about suicidality. Active and non-judgmental listening, and taking their thoughts seriously, will strongly facilitate talking about suicidality. The application of the different self-management strategies might be enhanced with psychoeducation, which should also address the frequent occurrence of suicidality in HD so that patients realise that they are 'not the only one'. If the physician considers prescribing medication, this should always be combined with talking about suicidality. Furthermore, both the effectiveness and side-effects have to be addressed, and time should be allowed for the patient to decide on starting medication. Significant others should also be involved in the treatment and (often) need to receive psychoeducation from healthcare professionals,[52;85] which must at least contain guidance on talking about suicidality and suggestions for support strategies for this person him or herself. If a patient has a wish for euthanasia, and euthanasia is permissible in their country, the healthcare professional should ensure repeated conversations about this topic and show a commitment of best intentions.[80;81]

Recommendations for further study

Future studies on the treatment of suicidality in HD should ideally investigate a combination of the proposed support strategies; this was shown to be effective in elderly populations[86;87] in whom a program of psychoeducation, access to a care manager, behavioural activation, and pharmacotherapy or psychotherapy resulted in lower rates of suicidal ideation and improved quality of life compared with the treatment-as-usual group.[87;88] Also, HD-specific studies should investigate whether involvement of a significant other in the experimental treatment leads to a greater decrease in suicidality for mutation carriers and less stress for caregivers.

Conclusion

This study indicates that the best practice for suicidality in HD is talking about suicidality, self-management strategies, using medication, and discussing end-of-life wishes. Future HD-specific intervention studies should investigate a combined approach of these treatment strategies to establish clinical evidence and improve guidance for the treatment of suicidality in HD.

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