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ORIGINAL RESEARCH

Couples' Attributions for Work Function Changes in Prodromal Huntington Disease

Nancy R. Downing · Janet K. Williams · Jane S. Paulsen

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Abstract People who have tested positive for the expanded Huntington disease (HD) gene who are not yet diagnosed (pre-HD) and their companions report subtle changes in ability of people with pre-HD to do their jobs. However, it is not known whether they attribute these changes to HD. Semi-structured telephone interviews were analyzed from seven persons with pre-HD at different estimated points from diagnosis and six companions. Data were analyzed using qualitative analysis methods. Participants made attributions related to health, work, and temperament. Only one participant attributed a change to HD. The process of forming attributions was demonstrated through symptom monitoring and comparison of participants with pre-HD to others with and without HD. Participants also expressed uncertainty regarding how to make attributions. Attributions influence coping procedures, including whether to seek and accept medical treatment. In persons with prodromal HD the relationship between attributions and use of coping strategies for symptoms that interfere with job functioning is unknown.

Keywords Huntington disease · Common sense model · Qualitative research

N. R. Downing · J. K. Williams
University of Iowa College of Nursing,
Iowa City, IA, USA

J. S. Paulsen (✉)
Departments of Psychiatry and Neurology,
University of Iowa Carver College of Medicine,
1-305 MEB,
Iowa City, IA 52242, USA
e-mail: jane-paulsen@uiowa.edu

Introduction

When people notice changes in their functional abilities, such as memory problems or fatigue, they try to find explanations for these changes. The attributions they choose to explain these changes influence how they cope with these changes. Leventhal and colleagues created the Common Sense Model of Illness Representation to describe this phenomenon (Leventhal et al. 1980). The model is based on the observation that when people perceive somatic changes it is common sense to assign attributions to them and to select appropriate coping procedures based on theses attributions (Leventhal et al. 1998). The Common Sense Model has been used widely to describe how people form illness representations, also called illness perceptions. The Common Sense Model has been used extensively to describe illness representations in persons with multiple sclerosis (Lerdal et al. 2009), heart failure (Jurgens et al. 2009), hypertension (Chen et al. 2009), and myocardial infarction (Broadbent et al. 2009). In the present analysis, the Common Sense Model (CSM) is used to explore illness representations in HD gene-expanded individuals and their companions in the preclinical stages of HD.

Huntington disease is an autosomal dominant neurodegenerative disease characterized by progressive motor, cognitive, and behavioral decline. Age of diagnosis is based on classic motor symptoms and is typically during middle age; death occurs approximately 17–20 years after diagnosis (Myers 2004). The disease involves a trinucleotide (CAG) expansion of the huntingtin gene on chromosome 4p16.3 (OMIM 2009). Prevalence of HD in North America is approximately 1 in 10,000 individuals (Walker and Raymond 2004). Until recently, the period prior to clinical diagnosis has been referred to as "presymptomatic." However, current data suggest this term is a misnomer since

functional changes prior to clinical diagnosis may represent symptoms of impending HD; thus, a more appropriate term for the period prior to clinical diagnosis is "prodromal" (Paulsen 2010).

According to the Common Sense Model, assigning attributions is an active process, beginning with an appraisal stage in which symptoms are first noticed. At this point symptoms may be subtle and nonspecific. The appraisal stage may be longer for symptoms that are mild or ambiguous and may result in making benign attributions instead of threatening ones (Leventhal et al. 1998). As symptoms progress, they may become more severe and distinctive, at which time people are more likely to attribute them to illness. The active process of assigning attributions to perceived changes involves both emotional and cognitive processes and is based on the "symmetry rule": People seek labels for perceived symptoms; given labels, people seek symptoms to confirm the labels (Leventhal et al. 1998). This phenomenon was demonstrated in a study in which students with normal blood pressure, when told they had hypertension, proceeded to identify symptoms of high blood pressure such as headaches, palpitations, tension, dizziness, and flushing (Baumann et al. 1989).

People form illness identities based on information from three sources: perceived symptoms, external sources (including healthcare providers, family, and media), and past experience with the illness (Leventhal et al. 1984). Past experiences with an illness may lead to inaccurate illness attributions. For example, persons who have had past experience with influenza, cancer, hypertension, depression, or schizophrenia, either in themselves or a close family member, held illness beliefs that were not always consistent with medical knowledge (Godoy-Izquierdo et al. 2007).

Illness representations are important because they influence the coping procedures people select. Coping procedures are defined as "the cognitive and behavioral actions we take (or do not take) to enhance health and to prevent, treat...and rehabilitate from illness" (Leventhal et al. 1998). This includes whether to seek medical care and accept treatment.

A group of researchers used the CSM to explore illness perception in persons diagnosed with HD and their spouses. In one study, patients' acceptance of HD was positively related to their mental health (Helder et al. 2001). The authors maintain that acceptance allows people with serious illnesses to use more effective coping mechanisms. In another study (Kaptein et al. 2007), persons with HD and their spouses had similar illness representations; however, spouses reported more symptoms than persons with HD and attributed more symptoms to HD. Persons with HD attributed many of their symptoms to stress. Also, spouses who believed less in a cure for HD had a more positive impact on their partners' vitality and social functioning. At the same time, persons with HD who had a stronger belief in control over their illness reported a higher quality of life. In another study, spouses seldom used denial as a coping method, and denial decreased as the severity and duration of HD symptoms increased (Helder et al. 2002). In total, these studies indicate that acceptance of HD is related to better outcomes and it is not necessary, or even desirable for members of a couple to have the same illness representations.

The studies discussed above were conducted by the same team of researchers and involved persons already diagnosed with HD. No data could be found that explore illness representations in prodromal HD. It has been possible for people at risk for HD to receive definitive genetic testing to determine whether they will develop HD since the gene expansion was discovered in 1993 (The Huntington's Disease Collaborative 1993). While clinical diagnosis of HD is not made until a person displays distinctive motor signs (e.g., chorea, dystonia, and impaired voluntary movements) (Hogarth 2003), recent research indicates subtle changes in cognition, behavior, and motor control can appear years before clinical diagnosis (Paulsen et al. 2008). Neuroimaging studies indicate structural and functional changes in the brain also occur prior to clinical diagnosis (Kloeppel et al. 2009; Paulsen 2009; Paulsen et al. 2008).

The changes that occur in prodromal HD may affect a person's ability to maintain day to day functions, including work functions. Family members may be the first to notice functional changes; in some cases they feel as though they are the only ones who notice these changes (Williams et al. 2007). For this reason, they are useful informants in HD research. It may be especially important to include family as informants in disease processes that involve cognitive loss (Davis 2001), as HD does. Furthermore, insight is impaired in persons with clinically diagnosed HD and it is not clear at what point impairment begins (Hoth et al. 2007). This means family members may provide important information in addition to participants with pre-HD regarding functional changes in prodromal HD. Family members are also impacted by HD (Williams et al. 2009) and therefore influence the care persons with HD receive.

Family members report changes in behavior, activities, relationships, and physical abilities in HD gene-expanded individuals prior to clinical diagnosis; however, they don't necessarily know whether the changes are related to HD (Williams et al. 2007). Individuals with pre-HD and their companions also report subtle changes in work function; changes in work function may be one of the most reliable indicators of functional changes in the prodromal stages of HD (Paulsen et al. 2009).

While persons in the prodromal phase of HD and their companions report subtle changes in work function, it is not clear whether they attribute these changes to HD. Attributions have consequences because they influence the coping

procedures people choose, including whether to seek treatment for noticed changes. While clinicians might recognize changes in work function related to prodromal HD, it is not known whether individuals with pre-HD or their companions do. The purpose of the current analysis is to explore the attributions of persons with pre-HD and their companions make for perceived changes in work function. The answer to this question may provide insight into whether these individuals would recognize potential benefits of treatments, lifestyle adaptations, or support to maximize function and wellbeing in the prodromal period.

Methods

The data in the current analysis were obtained from semi-structured telephone interviews with individuals with pre-HD and their companions. The entire interview was part of a larger mixed methods study designed to create a tool to measure work function in prodromal HD. Work function was defined in the study as the cognitive, behavioral, and physical ability to perform expected tasks related to paid or unpaid work. The work function study is part of PREDICT-HD 2.0, a multi-site longitudinal study designed to identify and track markers of HD during the prodromal period (Paulsen et al. 2006).

Participants

Participants were identified in collaboration with the PREDICT-HD 2.0 coordinators. Purposeful criterion sampling (Sandelowski 1995) was used to recruit individuals with HD gene expansion at different points from HD diagnosis as estimated using age and CAG-repeat number: far (≥15 years), mid (9–15 years), and near (≤9 years) (Langbehn et al. 2004). Participants are not identified in the results according to estimated proximity to diagnosis in order to preserve anonymity.

A sample size of at least six individuals with pre-HD and six companions was considered adequate to discern meaningful results from the qualitative interviews (Morse 1994). Potential participants were persons who have previously indicated their willingness to participate in HD-related research. All participants who were contacted agreed to participate. Nine individuals with pre-HD and eight companions agreed to participate. Two participants with pre-HD were later identified as having received a clinical diagnosis of HD since their last classification and they and their companions were excluded from the current analysis. Thus, 13 individuals were included in the final analysis: seven persons with pre-HD and six companions. Participants with pre-HD included in this analysis include: four individuals classified as far from diagnosis, two midway from diagnosis, and one near diagnosis. Companions were spouses or significant others of three persons classified as far from diagnosis, two midway from diagnosis, and one near diagnosis. Age range of participants with pre-HD was 30–59 years; median age was 44.5 years. Six of the seven participants with pre-HD were female. The University of Iowa Institutional Review Board approved the study.

Procedure

The interviewer (NRD) contacted prospective participants by telephone and asked them whether they were willing to participate in a study about changes in work function in "presymptomatic HD." After participants returned mailed consent documents, the interviewer contacted them for semi-structured telephone interviews at a pre-arranged time. Participants responded to questions regarding changes in individuals with pre-HD work function including changes in physical skills, keeping track of information, getting places on time, relationships at work, quality of work, mood, and interest in work (see Table 1 for sample questions). The interviewer also asked participants to describe how individuals with pre-HD manage changes in work function and whether they have advice regarding work function for others with presymptomatic HD. The interviewer asked participants to comment on any other issues related to work function that were not asked in the interview. The interview did not include questions regarding attributions for noticed change. Interview length varied according to how many changes in work function participants described and continued until participants had nothing more to add. Average interview length was 26 min.

Data Analysis

Data were analyzed using qualitative descriptive techniques (Sandelowski 2000). One coder (NRD) initially coded the data using NVivo 8 software (QSR International 2000). The coder did not use a conceptual framework during the initial analysis; rather, the theme "attribution" was identified in the data. Following the identification of the attribution

Table 1 Sample of Semi-Structured Interview Questions

What kinds of physical skills do you use in your job? What changes have you noticed?
How are things going when you need to keep track of information?
What about getting along with other people at work?
People sometimes say that people with presymptomatic HD don't feel as well as they used to, and that can make it more difficult to do their jobs. What is your experience?

theme, the initial coder conducted a literature search on attribution and selected the Common Sense Model as a framework for further data analysis. An expert in HD research and a researcher who uses the Common Sense Model in healthcare research confirmed the appropriateness of using the model in this context. The initial coder then returned to the data and coded again looking for themes related to the Common Sense Model. The study was not designed using the Common Sense Model; however, the coder found data related to attributions as well as the active process of making attributions for noticed functional changes and decided to use the Common Sense Model as a framework. Codes were created for attributions and for active processes.

After coding the data, another member of the research team (JKW), an expert in HD, assessed reliability of the coding. The two investigators discussed differences until 100% agreement was reached. The remaining two members of the research team assisted in assessing the validity of the data analysis conducted by the initial coder and the HD expert. The entire team discussed validity issues including: definition of coding terms, clarification of themes, descriptive validity (the themes identified by the initial coder and verified by the expert were apparent in the data to the other team members), and interpretive validity (the interpretations of the data made by the initial coder and the expert made sense to the other team members) (Sandelowski 2000). Discussion continued until 100% agreement was reached.

Finally, content analysis (Morse and Niehaus 2009) was conducted in order to count the number of participants who made attributions and the number of participants who attributed a change to HD. Content analysis also allowed the researchers to compare responses between persons with pre-HD and their companions.

Results

Most participants volunteered attributions for noticed changes in work function. Ten of the 13 participants volunteered attributions: six out of seven persons with pre-HD and four out of six companions. Two participants—partners in a married couple—denied noticing changes. Although participants were asked specifically about work function, several talked about changes at home as well, including irritability and memory issues. Some participants might have been influenced by external illness labels when making attributions. For example, a companion stated, "Just by the nature of it being 'presymptomatic,' it doesn't seem to be affecting her yet." On the other hand, consistent with the Common Sense Model symmetry rule, a participant with pre-HD expressed the tendency to attribute changes to HD based on the label: "So you don't know if it's the Huntington's or just knowing that you're positive for Huntington's?"

Only one participant, however, attributed changes to HD. This change was not work-related (spouse was choking on pills and food). Three categories of attributions for reported functional changes were identified: health-related, work-related, and temperament-related. In addition, participants also provided insight into some of the active processes they used to make, or try to make, attributions for noticed functional changes. Four themes related to active processes were identified: symptom monitoring, comparison to others with HD, comparison to others without HD, and comparison to past self. Participants also expressed uncertainty regarding how to make attributions for noticeable changes in function and whether they were related to HD.

Attributions

Health-Related Attributions

Health-related attributions for functional changes were common among both participants with pre-HD and companions. However, only one companion attributed a spouse's functional changes to HD. Several other health-related attributions were mentioned, with aging being the most common.

Aging After discussing subtle changes in memory, physical ability, or relating to others in the work environment, participants indicated they attributed many noticed changes to aging. For the most part, individuals with pre-HD minimized or normalized these changes, indicating they were not considered very severe: "Oh my gosh, [it's] not the beginning of the end, it's just getting old"; "So, normal little age quirks that show up [that] you notice with one's body when you're 30 and 40 and 50." A participant attributed a change in his attitude toward coworkers to aging: "I look around at people my age and...you've known so many people that have come and gone; after awhile it kind of seems to be kind of a numbness—you don't get engaged to people." A participant wondered whether his memory changes were attributable to aging: "[U]sually if I just wrote a couple of words I would remember everything. Now I am not quite so sure. And...my recall of events is not as good....I don't know what that is, 'cause I am older."

Companions also attributed functional changes to aging, including increased use of visual reminders such as "to-do" lists and decreased socializing at work. One companion normalized his partner's memory problems by attributing them to aging, which affects everyone. "[O]ur memory is not the same as when we are in our fifties as it was in our forties or thirties."

Other Health-Related Issues Several participants with pre-HD and companions offered other health-related attributions for noticed functional changes including allergies, ankle and wrist injuries, accidents, menopause, and other health conditions: "I'm always moody. I think that is a lot with between menopause, whatever that is that you go through, and things"; "She said she had an [automobile] accident...has trouble with her short term memory sometimes." One participant drew a distinction between physical changes due to injuries versus those that might be attributed to HD: "I have had injuries. I lost a ligament in my right foot.... I broke my left wrist skiing last winter, so it's a little stiff....Nothing from Huntington's." Even when behavior was notably different it was not attributed to HD: "Yeah, my concentration is less than it used to be…especially if I'm reading....It's just harder to stick with the book....[S]o we kind of joke around that I have adult ADD"; "I would say she has gotten moodier....she's having a terrible menopause. You know, versus when she was younger....If I had to put my finger on one definitive cause...I would think that would be it."

Work-Related Attributions

Participants with pre-HD made attributions for changes in their work function related to their work situation, including problems with supervisors or coworkers, having a bad day, and being overworked: "I got my first negative review ever this year, that I ever had in my life, but then I asked around and I found out that they gave every single person a negative review"; "There is one [coworker] that aggravates the death out me because she is completely incompetent. So she and I kind of carefully avoid each other"; "I get really irritated, real fast....[T]hat probably has a lot to do with all the studying for all the [job-related] exams"; "I used to remember stuff all the time. But, I didn't have so much on my plate....I didn't finish [an assignment] on time. And, I don't know if that's just because I have so many things going on...." In fact, if a participant with pre-HD mentioned HD, the person disclaimed it as an attribution: "[S]omebody might say, 'Well, that is just Huntington's'; and it's not Huntington's....The guy having a scream attack because he is having a bad day is just okay." Companions also noted changes in their partners were due to work situations and overload: "She don't put up with as much crap...I think she might have too much on her with school and work";

> I mean there's sometimes where she's had a tough day or something where she might email me and kinda joke around that she needed another job or something like that but it has more to do with just trying to vent off steam about having a bad day.

Temperament-Related

The behavior of many of the individuals with pre-HD was described in terms of their pre-existing personalities. Most of the time, problems noted in work function were not considered changes, but rather a reflection of their temperaments: "No changes; I am chronically late"; "I've never been a super coordinated person anyway." Companions also talked about their partners in terms of innate temperaments: "He's always kept a lot to himself"; "He's never been on time in his life." Again, if a companion mentioned HD, often it was to deny it as an attribution:

> She being a perfectionist, certain things really piss her off....Like, she's obsessively clean and neat; so if you bring a leaf in, you better take it out. So I don't think it has nothing to do with Huntington's. It's just being a witch [laughs].

Even when companions acknowledged behavior had changed, they attributed it to temperament. "[s]he gets much more stressed out, like, easier... [but] she has always been high strung." Participants with pre-HD also attributed change in behavior to a change in temperament:

> I definitely have a lot more confidence when it comes to arguing [at work]. Before I think I would have swept it under the rug...and now I'll just battle with them....I've gotten the confidence to be sort of obnoxious.

Active Processing

According to the Common Sense Model, forming illness representations is an active process involving both cognitive and emotional pathways. Participants in this study provided insight into the active processes they used for evaluating changes they noticed in their or their partners' work function abilities. Participants displayed active processing through monitoring symptoms and comparing individuals with pre-HD to others with and without HD. Not all active processes resulted in an attribution. Several participants expressed uncertainty regarding the meaning of some functional changes.

Symptom Monitoring

Participants monitored symptoms, compared current functional abilities with past functional abilities, and paid attention to things that were "not like" themselves. Four participants talked about monitoring for HD symptoms. Two participants with pre-HD monitored themselves and were both concerned about safety: "I pay attention to things....I don't want to work at a time when my

performance compromises my safety. That is something I kind of think about"; "And if my working is really starting to be different because of Huntington's, then I really need to probably get out of the working world." A companions discussed monitoring his partner: "[T]he awareness is always there, yeah.... You always watch them for little tell-tale signs." One participant also had a supervisor monitor her for changes: "[S]he literally put a tickler in her calendar for once a year to stop and think about me and see if I seemed any different." Symptom monitoring did not necessarily result in making an attribution. A participant noticed a change but expressed uncertainty regarding its meaning: "I've noticed every once in awhile I find myself dragging maybe a foot....You know when I noticed I thought, 'I wonder if this is how it is?'....[I]t was odd though, 'cuz it's not like me. And I didn't know why."

Comparison to Others with HD

Participants compared changes to what they had observed in others affected by HD. Again, they did not necessarily make attributions after making comparisons. Comparing his partner's behavior with that of her father who had HD, a companion stated, "[S]he is quite stubborn and you know, her father, he was very stubborn." Two persons with pre-HD expressed reluctance to attribute changes to HD because the age of clinical diagnosis for their parents was older than their current age: "[T]he knowledge I got from genetic counseling was that age of onset would be most likely the same as my mom's....[S]o that is pretty far away for me, you know, to be panicking about 20 years away"; "Because Huntington's in my family tends to come later in life....I'm thinking maybe that kind of thing...might have been just too early for me to see."

Comparison to Others Without HD

Participants also compared changes in function with others who do *not* have HD. In one case, a participant with pre-HD attributed changes to an illness she and her mother (who does not have HD) both have, even though the comparison didn't match very well: "...[M]y mother...has rheumatoid arthritis, but she has never had it to...this extent, and her symptoms, I guess, aren't like mine." Companions also compared partners to themselves: "[S]he remembers way more than I do, that's for sure"; "The only yardstick you have is yourself....[and] *I* don't feel as good as I did 10 years ago either."

Uncertainty

The majority of participants (four persons with pre-HD and four companions) expressed uncertainty at least once point during the interview regarding how to make attributions for noticed changes. "I hide in my room a lot more....I don't know why—it doesn't make any sense. Everybody's nice...."

> [I] notice that I have periods of time when I 'ruminate?' I can't shake it. It can be a problem. I don't know what that is either. I think I might have always done it; now I am becoming aware of it. But I'm not sure.

All but one of the participants who expressed uncertainty wondered whether changes were related to HD. "Sometimes I feel tired a lot....I don't know if that is part of it [HD] or not";

> [S]ometimes if I get too tense I'll just go to a mall, put on my shoes and walk for an hour....'cause that relaxes me....[A]nd I'm not a good relaxed person. I don't know if it has anything to do with if Huntington's is coming on...I don't know. I really don't.

Companions stated: "[I]t's hard to know what's normal aging and what might be HD"; "I don't know if it's part of the HD or if that's just she is stubborn"; "I think she just sleeps more than what she used to. And again, I don't know how much of what she does is HD-related...."

> [I]t's really hard to distinguish, you know, what one thing would be versus another. Every time something comes up and I think...that [HD] might be a direct cause, then I think, no...it would have to be obvious, you know what I mean?

For two participants with pre-HD, uncertainty itself was problematic: "I mean, if they could just tell me what in the world it is, I mean, if...they said it's because of the HD, I'd be like, 'Okay, fine. Thank you.'"

> I think a lot of it is...the uncertainty of what is going to happen in the future. Because, at some point, it *is* going to start to affect me. And at some point it is going to start to affect my ability to do my job. And every now and then I think about that, and you know, it worries me.

Although participants were not specifically asked to do so, they made attributions for changes they had noticed in the functioning of individuals with pre-HD. Participants also demonstrated the use of active processes in either deciding or not deciding to make attributions for noted functional changes. Participants expressed discomfort with uncertainty regarding how to make attributions for noted changes and uncertainty regarding when changes will affect their ability to do their jobs.

Comparison of Couples' Responses

Responses of persons with pre-HD and their companions were remarkably similar. The person with pre-HD who did not note changes was the spouse of one of the companions who also did not note changes. The other companion that did not note changes was the significant other of a person who noted only that she received a negative job review for the first time, to which she attributed a supervisor who gave everyone negative reviews. Her companion stated she had bad days at work but did not elaborate; he also denied she had any changes in work function.

The companion of the woman who attributed irritability to menopause also attributed her moodiness to menopause. Both members of another couple attributed the woman's irritability to having too much to do. In another couple, both the man with pre-HD and his wife attributed his memory changes to aging. The wife of the companion who attributed his wife's choking to HD did not specifically attribute changes in herself to HD but did express a lot of uncertainty regarding whether changes she experienced were related to HD.

Discussion

The findings in this analysis provide preliminary evidence that people with pre-HD and their companions might notice changes in functional abilities in prodromal HD and make attributions for these changes. However, in this sample, most did not attribute noticed changes to HD. The tendency to make attributions for changes illustrates the basic assumption in the Common Sense Model that people assign labels to somatic changes. According to the symmetry rule of the Common Sense Model, it would thus be logical to assume participants would also attribute at least some noticed changes to HD because one member of the couple had tested positive for the HD gene expansion. Furthermore, participants were asked to participate in HD-related research. Despite this, only one participant attributed a change in his spouse's functioning to HD. Instead, most changes in function were attributed to aging and other health conditions, work environment, and temperament. These findings appear to violate the Common Sense Model symmetry rule. This may partly be explained by the label "presymptomatic" that was used during the study. Cover letters, consent documents, and the interview script all referred to "presymptomatic HD." If persons are told they or their companions are presymptomatic it may not seem logical to attribute changes to HD. This is illustrated by the companion who stated that "by the nature of it being 'presymptomatic'" it didn't seem to be affecting his partner yet.

Curiously, many of the attributions that were volunteered by participants were factors over which they have little or no control, such as aging and temperament. People faced with an uncontrollable disease might prefer to attribute changes to things over which they have more control; however, this did not seem to be the case. On the other hand, aging and temperament may be seen as more "normal" phenomena, which may decrease the perception of their severity. They may represent examples of normalization and social comparison which occur when forming illness representations (Leventhal et al. 1980; Leventhal et al. 1985). Normalization involves attributing changes to non life-threatening causes and using social comparison to attribute changes to things "normal" people experience. For example, it has been found that older people often compare their physical changes to others and attribute them to aging, even when they may actually be related to illness (Leventhal 1984).

Aging is related to subtle functional changes, including mild physical and cognitive decline and decreased social activity (Buchman et al. 2009; Caserta et al. 2009). Persons who are middle-aged may be more likely than older persons to use avoidance and delay seeking medical care when their symptoms are ambiguous (Leventhal et al. 1993). This could have consequences for prodromal HD; if people attribute changes to aging they may be less likely to seek medical treatment that could potentially improve their functional abilities. Also, in the Common Sense Model, representations of the self often overlap with illness representations (Leventhal et al. 1999). Thus, the untangling of temperament and subtle functional changes related to prodromal HD may be complicated. It is possible that only when one confronts a change that is "not like" the individual with pre-HD does one begin to question non HD-related attributions.

Another way to interpret the tendency to avoid making HD-related attributions is denial. Denial is a recognized coping procedure in diagnosed HD which might be helpful because treatments are limited and there is no cure (Decruyenaere et al. 2003). People sometimes use denial when an illness threatens family roles or creates fear (Leventhal et al. 1995). In some spouses of persons with HD, the threat of family role changes creates distress and can lead to avoidance coping, a form of denial (Decruyenaere et al. 2003). Spouses experience disruption in their own work and recreational lives when HD symptoms affect their partners (McCabe et al. 2008). Some couples cope by minimizing the impact of HD on their lives (Richards 2004). Thus, there is evidence that family members use denial after HD diagnosis; this may be relevant in prodromal HD as well.

While participants provided insight into the active processes they used to make attributions, not all active

processes resulted in illness attributions. This is not surprising given that most changes were vague and ambiguous. HD attributions might increase as functional changes become more distinct and severe, as illustrated by the husband who attributed his wife's choking to HD.

The amount of uncertainty expressed by participants is troubling. For persons at risk for HD who have not been tested, vague symptoms and uncertainty of their meaning can create high levels of distress (Duncan et al. 2008). Thus, it might be beneficial for individuals with pre-HD to attribute subtle functional changes to HD if it facilitates more effective coping. For example, although there is no cure for HD, treatments for some symptoms exist (Adam and Jankovic 2008), including medications to improve mood, ability to focus (Grimbergen and Roos 2003), and irritability (Ranen et al. 1996). These symptoms have the potential to interfere with important aspects of quality of life such as work and relationships. Evidence of structural brain changes in prodromal HD suggests it may be important to target affected brain regions with medications prior to significant structural damage (Hannan 2005). Attributing functional changes to HD prior to diagnosis may facilitate earlier treatment that could improve functioning and allow individuals with pre-HD and their companions more time to plan for the future.

Individuals with pre-HD and their companions may benefit from more information regarding changes to expect in prodromal HD and what treatments are available to help maintain functional abilities longer. The term "presymptomatic" should be avoided as it creates the misconception that there are no symptoms prior to HD diagnosis. Changing patients' illness perceptions in other disease processes such as diabetes (Lawson and Harvey 2009) and myocardial infarction (Broadbent et al. 2009; Petrie et al. 2002) have led to improved health outcomes for patients and their spouses. There may be benefits, therefore in helping persons with pre-HD and their companions understand subtle changes they might expect in prodromal HD.

Furthermore, acceptance of chronic illness and relinquishing control may be empowering for some people (Aujoulat et al. 2008). Patients who are uncertain how to make attributions for noticed physical changes have expressed dissatisfaction with healthcare provider interactions (Frostholm et al. 2005). In some illness processes, communication between patients and their healthcare providers improved when providers assessed patients' illness representations (de Ridder et al. 2007). Therefore, people in the prodromal stages of HD might benefit from direct discussions with healthcare providers regarding their illness perceptions, including how they make attributions for functional changes.

Of course, there are ethical considerations in attributing subtle functional changes to HD prior to diagnosis, including the risk of psychological harm (Bloch et al. 1992; Witjes-Ane et al. 2007) and risk of exposing individuals with pre-HD and their families to discrimination (Penziner et al. 2008). On the other hand, another ethical consideration is the right of people to have access to new information regarding a disease that impacts them. They also have the right to self determination regarding what to do with this information.

This study has some important limitations; therefore the findings should be considered preliminary. First, this is an analysis of findings that the larger study was not designed to elicit. Participants volunteered attributions for noticed changes and were not asked to do so. Therefore, it is possible more participants would have attributed some changes to HD if they had been asked to make attributions. Next, it is important to keep in mind that the subtle functional changes noted by participants in the current study could indeed be attributed to things other than HD. After all, people who are gene positive for HD also age, and they have other illnesses and circumstances that may contribute to noticeable changes in work function. Family members have expressed frustration when healthcare providers do not adequately address health issues other than HD (Williams et al. 2007). It is important to sort out which changes are related to HD and which changes are related to other factors in order to determine the most appropriate interventions.

Participants were also only asked about changes regarding work function, which might have biased results. Employment is related to financial well-being, social and personal well-being (Warr 2008). Therefore, it is possible that participants might have greater incentive to avoid acknowledgement of work function changes since loss of employment is threatening. Recent research indicates changes that occur prior to diagnosis have the potential to impact work function (Paulsen et al. 2009), indicating some of the changes noted by participants in this study might, in fact, be related to pre-HD, and might become more problematic over time. Further research in this population should include assessment of noticed changes in broader areas of function including at home and in social and recreational activities.

Another limitation is that the results of this analysis might only apply to people who have chosen to have the predictive test for HD. Persons who undergo predictive HD testing might use denial and minimization less frequently than those who forgo testing (Decruyenaere et al. 2003). Couples who choose to undergo testing might also have more positive relationships prior to testing (Quaid and Wesson 1995). The similarity in responses of persons with pre-HD and their companions in this study illustrate this last point. In sum, couples who undergo genetic testing and participate in research together might communicate with

each other in more positive ways than persons who forgo testing and do not participate in research. Thus, the results presented here cannot be generalized all persons at risk for HD and their companions. However, the potential merits and risks of interventions that include assessment of illness representations and education on what to expect in prodromal HD merit further study.

Despite these limitations, the current analysis has important implications, including the need to clarify when symptoms of HD actually begin and to sort out changes that are related to prodromal HD versus other factors. If individuals with pre-HD and their companions do not recognize that functional changes might be related to HD, they might miss opportunities to seek interventions to treat or delay symptoms that have the potential to interfere with daily function and negatively impact their quality of life.

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