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Understanding self-management behaviors in symptomatic adults with uncertain etiology using an illness perceptions framework

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Abstract

The self-management behaviors of individuals with medical conditions that have an unknown etiology have not been studied. This study assesses the relationship between illness perceptions and various illness self-management behaviors in patients undergoing clinical genomic sequencing to identify a genetic cause for their condition. Hierarchical linear regression, Poisson linear regression, and logistic regression were used to assess the effect of illness perceptions (i.e., perceived consequences, timeline, personal control, treatment control, identity, concern, understanding, emotional impact, and causal beliefs as measured by the Brief Illness Perceptions Questionnaire) on healthcare use, prescription medication use, and doctor recommended supplement use, respectively (n = 200). Analyses revealed that (1) illness identity beliefs were positively associated with healthcare use (β =.20, p =.04), (2) both treatment control beliefs (B =. 03, p =.02) and genetic causal beliefs (B =.17, p =.049) were positively associated with prescription medication use, and (3) both timeline beliefs (OR =1.23, p =.02) and emotional impact (OR =1.20, p =.02) were positively associated with doctor recommended supplement use. These findings can be used to inform the development of guidelines for treating patients who are seeking a genetic diagnosis for their illness.

Keywords

illness perceptions; self-management; genomic sequencing; healthcare utilization; medication use; supplement use

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Ethical approval: All procedures performed in studies involving human participants were in according with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. This article does not contain any studies with animals performed by any of the authors. Informed consent was obtained from all individual participants included in the study.

In the context of chronic diseases, self-management broadly refers to strategies patients use to manage the actual or potential impact of their disease (Richard & Shea, 2011). It is characterized as a "cluster of daily behaviors" to manage health (Glasgow & Anderson, 1999, p. 2090). The self-management behaviors of individuals with well-defined medical conditions (e.g., diabetes) have been studied extensively. Furthermore, a patient's lifestyle behaviors (e.g., diet, exercise) and adherence to treatments (e.g., medications and other therapies) are a common target of self-management interventions (e.g., Lorig et al., 2001). Patients may also consult with healthcare professionals about potential treatments or strategies for managing their symptoms (Richard & Shea, 2011), although these self-management behaviors are less frequently studied.

In contrast, the self-management behaviors of individuals with unexplained medical conditions are harder to characterize. These individuals often face misdiagnosis, diagnostic delay, and uncertainty in treatment options (Nettleton et al., 2005; The Lancet, 2009; van der Kloot et al., 2010). Research reveals that these factors contribute to extensive testing and consultations with specialists. Other work shows that individuals engage in these behaviors — seeking an explanation or diagnosis —in order to more clearly understand what to expect for the future and how to manage a health concern (Koopman & Schweitzer, 1999; Lewis, Skirton, & Jones, 2010). Although individuals' beliefs about their illness (such as those concerning the severity of the illness and its impact on daily life) have been found to contribute to self-management behaviors across a variety of populations (Bratzke et al., 2015), individuals who cannot identify an explanation, diagnosis, or appropriate treatment options for their condition are likely to experience increased levels of frustration and uncertainty as a result (Nettleton et al., 2005), and we hypothesize that they may also rely more heavily on their beliefs about their illness because they lack the information they need for self-management.

Accordingly, the present study informs prior research on self-management by investigating the association between illness beliefs and self-management behaviors in a population of patients who are undergoing whole exome sequencing (WES) to identify a potential genetic explanation for their condition. Unlike individuals with medically unexplained symptoms (MUS) who have symptoms with no known physical cause (Gormley, 2014), and therefore are dismissed by medical professionals or made to feel like it is "all in the mind" (Nettleton et al., 2005), this population experiences symptoms that are suspected to be genetic in nature, but has yet to receive a definitive genetic explanation for them. This uncertainty about what caused their condition can shape the beliefs patients bring with them into the clinical encounter and influence the self-management behaviors they utilize as they seek a genetic diagnosis. Just as a clinical diagnosis provides valuable information to those with MUS, a genetic explanation may help clarify the potential of recurrent risk, implications for family planning, and inform illness management decisions of people who are affected by a disease process that strongly appears to be genetic in origin, although that genetic cause has not yet been identified.

Illness Beliefs

Leventhal's common-sense model (CSM) of self-regulation (1984, 2003) provides a theoretical framework for understanding how individuals' beliefs about their illness (referred to as *illness perceptions*) can influence their self-management behaviors. This theoretical framework states that upon experiencing a health threat, individuals engage in an iterative coping process based on the cognitive and emotional representations of that threat. The effectiveness of this coping process is evaluated, and the cognitive and emotional representations of the health threat are re-appraised based on the evaluation of this coping behavior. The CSM identifies five core domains of illness perceptions individuals hold about a health threat: identity, cause, consequences, timeline, and control (Leventhal et al., 2003). *Identity* refers to how individuals define their health threat and the symptoms associated with that health threat. *Cause* refers to the factors individuals believe led to the health threat. *Consequences* are the expected outcomes and perceived impact of the health threat. *Timeline* is the duration of the health threat, and is usually conceptualized as being of limited duration (acute) or as persisting for a longer and possibly indefinite amount of time (chronic). Finally, *Control* refers to individuals' beliefs about the extent to which personal or medical actions can help control the health threat. These cognitive representations work in parallel with emotional representations to direct coping behaviors (Leventhal et al., 1984).

Self-management behaviors have been identified as one manifestation of coping responses to health threats (Leventhal et al., 2003), and the framework of illness perceptions provided by the CSM has been used to predict illness self-management behaviors for a variety of populations with well-defined medical conditions, including asthma, chronic obstructive pulmonary disease, and cardiovascular disease (French et al., 2006; Hagger & Orbell, 2003; Kaptein et al., 2008; Petrie et al., 2007). For instance, a meta-analytic review (Hagger & Orbell, 2003) found that perceived controllability is significantly associated with active coping behaviors, such as adherence to diet and drug regimens. Previous literature assessing behavioral outcomes in patient populations has found that timeline beliefs and emotional impact were associated with medication adherence in end-stage renal disease patients (O'Connor et al., 2008); greater perceived consequences predicted healthcare use among adults with congenital heart disease (Schoormans et al., 2014); identity beliefs, control beliefs, perceived consequences, and understanding predicted attendance at cardiac rehabilitation following an acute myocardial infarction (French et al., 2006); and timeline beliefs, identity beliefs, and causal beliefs were associated with medication adherence among asthma patients (Kaptein et al., 2008). Other research has found positive associations between illness identity, emotional impact, and perceived consequences with healthcare utilization (Frostholm et al., 2005; Jackson et al., 2006). As noted, however, the illness perceptions framework is generally utilized in populations with conditions that have clear treatment options, implications for family, and prognoses. Little is known about how individuals with medical conditions with uncertain etiology perceive their illness and its impact on their attempt to manage their conditions in spite of this uncertainty. Therefore, applying the CSM illness perceptions framework to individuals undergoing diagnostic genomic sequencing can help us understand how illness perceptions are associated with illness self-management in this population.

Current Study

The current study assesses the association between illness perceptions and various self-management behaviors (health care use, prescription medication use, and supplement use) among individuals who are seeking a genetic explanation for their presenting health condition. In the context of symptomatic, chronic disease management, these three self-management behaviors – health care use, prescription medication use, and supplement use – indicate people's attempts to manage their health and therefore serve as useful targets for analysis to understand illness self-management decisions.

Participants were adult patients who were having diagnostic WES as part of a study evaluating use of genomic sequencing in clinical care. These participants have all received a clinical diagnosis and are currently seeking a genetic explanation for their medical condition, which can provide valuable information regarding the potential for recurring risk and implications for family planning. In light of the iterative nature of illness perceptions and coping processes (Petrie & Weinman, 2012), understanding the illness perceptions people hold *before* receiving results from a genetic test, as well as the concurrent links between these illness perceptions and illness self-management, can provide valuable insight into how pre-existing illness perceptions may contribute to behavioral responses to subsequent results from these tests (Lerman et al., 2002).

This cross-sectional study assesses the relationship between illness perceptions, healthcare use, prescription medication use, and doctor recommended supplement use before receiving genomic sequencing results. Among this population of symptomatic individuals who are seeking a genetic etiology for their condition, we expected that some illness perceptions may be more strongly associated with these self-management behaviors than what has been reported in the literature on other populations. Specifically, we hypothesized that illness identity, beliefs about treatment control, and emotional impact would be independently, positively associated with both healthcare use and prescription medication use. Research on supplement use in healthy populations suggests that supplements are viewed as a health management strategy that is distinct from other "medical" strategies, such as medication use, and one that allows individuals to exert some degree of personal control over their health (Nichter & Thompson, 2006). Consequently, we hypothesized that only beliefs about personal control would be independently, positively associated with supplement use. We hypothesized that no other illness perceptions (i.e. consequences, timeline, concern, and cause) would be associated with any of the outcomes.

Methods

Participants

Participants were adult patients (n = 223) participating in the North Carolina Clinical Genomic Evaluation by NextGen Exome Sequencing (NCGENES) study at the University of North Carolina at Chapel Hill (UNC). NCGENES investigates the clinical use of diagnostic genomic sequencing in patient populations affected by a health concern that appears to have a genetic cause, although that cause has not yet been identified. Their health concerns were related to potentially heritable cancers, cardiogenetic conditions,

neurodevelopmental disorders, congenital malformations, retinal disease, and other conditions such as hemophilia and mitochondrial disorders. The study used whole exome sequencing (WES), a form of next generation genomic sequencing (Lohmann & Klein, 2014), to identify explanatory genetic variants. Children were also sequenced in the study and their parents participated in study assessments, but their data were not included in current analyses. Adult patient participants were recruited from the UNC Cancer and Adult Genetics Clinic and Vidant Medical Center (Greenville, NC) by referral from a physician identifying them as having a condition that is highly suspected to have a genetic cause. Participants were excluded if they did not speak English or Spanish or if they had already received a genetic diagnosis through prior testing. Of the eligible participants contacted for the study (n = 724), 347 agreed to receive WES (46.9%) and completed the baseline assessment between September 2012 and July 2014. Parent respondents were excluded from the analysis, so the current study is based on data from the subset of 223 adult patients who have completed the first telephone survey as of July 2014.

Procedures

All participants completed an intake questionnaire mailed to their home prior to meeting with a genetic counselor to enroll in the study. At the enrollment meeting, participants met with the genetic counselor, learned about WES, completed informed consent procedures, and provided a blood sample; no personalized genetic counseling was provided at this time. Two weeks later, participants completed a structured telephone interview conducted by trained research assistants (Time 1 interview) followed by a mailed questionnaire (Time 1 questionnaire). The intake questionnaire, Time 1 telephone interview, and Time 1 questionnaire were all completed before receiving WES results and are considered baseline data. Data collected at subsequent follow-up assessments (i.e., two weeks after return of diagnostic WES results and three months and six months after that) were not used in the present study, which was focused on understanding illness perceptions people hold before receiving results from a genetic test and their concurrent associations with illness self-management. Demographic information was obtained from the intake questionnaire and data on illness perceptions, healthcare utilization, prescription medication use, and supplement use was collected during the Time 1 telephone interview. All procedures were approved by the Institutional Review Boards of the University of North Carolina at Chapel Hill and Vidant Medical System.

Measures

Illness perceptions were assessed with the Brief Illness Perceptions Questionnaire (IPQ) (Broadbent et al., 2006). This 9-item self-report scale measures cognitive and emotional illness perceptions. Each item corresponds to one illness perceptions domain including (1) consequences, (2) timeline, (3) personal control, (4) treatment control, (5) identity, (6) concern, (7) understanding, (8) emotional impact, and (9) causal beliefs. Responses for items 1–8 are scored on a scale from 0 to 10. Higher values indicate more negative beliefs for items 1, 2, 5, 6, and 8. Higher values indicate more positive beliefs for items 3, 4, and 7.

Item 9 is one open-ended question assessing causal beliefs by asking participants to “List in rank-order the three most important factors that you now believe caused your health

concern." Two variables were extracted from responses to this question. First, we identified participants who most strongly endorsed a genetic causal model for their illness by dichotomizing responses to indicate those who listed a genetic or hereditary cause as the most important factor (1) and those who did not (0). Second, we identified participants who were most uncertain about the cause of their condition (indicated by the fact that they did not have a genetic nor non-genetic explanation) by dichotomizing responses to indicate those who responded "don't know" and provided no further answers to the causal beliefs question (1) and those who provided at least one answer (0). These two dichotomous variables were used to measure causal beliefs.

Healthcare utilization was measured with one question from the National Health and Nutrition Examination Survey (NHANES) asking "During the past 12 months, how many times have you seen a doctor or other health care professional about your health at a doctor's office, a clinic, hospital emergency room, at home or some other place? Do not include times you were hospitalized overnight" (Center for Disease Control and Prevention, 2013). Response options were: "None," "1," "2 to 3", "4 to 9," "10 to 12," and "13 or more" and responses were analyzed as a continuous variable.

Prescription medication use and *supplement use* were measured with two questions created for the NCGENES study: (1) "How many medications prescribed by a doctor do you take each day or on most days?" and (2) "How many vitamins, minerals, herbs, or other supplements recommended by a doctor do you take each day or on most days?" Participants' responses for prescription medication use were analyzed as a continuous variable. Their responses for doctor recommended supplement use were positively skewed and kurtotic (mean=1.57, SD=1.91, range=0–13, skew=2.31, kurtosis=8.24); therefore this variable was dichotomized to indicate those who took one or more supplements (1) and those who took no supplements (0).

Covariates. Research has linked age, gender, certain medical conditions, and disease severity to healthcare utilization, medication use, and supplement use (Satia-Abouta et al., 2003; Schoormans et al., 2014), therefore all analyses controlled for age, gender, diagnostic category, and physical functioning, as well as education and income. Covariates measured at enrollment include age at enrollment (continuous), gender (female=1), income (continuous), and education (more than 4 year college=1), physical functioning, and diagnostic category. Physical functioning was measured through the Karnofsky Performance Status scale, scored on a scale of 1 to 8 and coded so that higher scores reflect better physical functioning (Karnofsky et al., 1948). This variable accounts for differences in symptoms and physical functioning present in our population that may confound the relationship between illness beliefs and self-management. Diagnostic category is a label assigned to each participant corresponding to his or her clinical diagnosis (e.g., cancer, cardiogenetics, neurodevelopmental disorders, congenital malformations), which also informs the panel of genetic variants to be examined during WES. This variable was used to control for differences in healthcare and self-management experiences inherent to the condition. Indicator codes with cancer as the reference group (0) were used because this was the largest and most homogenous group in terms of pre-existing healthcare needs.

Sample

Based on z-scores >|3.3| (Tabachnick & Fidell, 2001), nine outliers (six for Timeline and three for prescription medication use) were detected. Analyses were conducted with and without the outliers, and all nine cases were removed from the sample because they were found to have a biasing effect on the results. Fourteen cases did not provide relevant answers to the causal beliefs question and were removed from analyses. As a result, the final sample size for all analyses was 200.

Analysis plan

Descriptive statistics of IPQ scores were used to evaluate illness perceptions in this sample. Hierarchical multivariate linear regression was used to evaluate the association between IPQ items and healthcare utilization. Poisson linear regression was used to evaluate the association between IPQ items and prescription medication use in order to account for the use of count data. Hierarchical logistic regression was used to evaluate association between IPQ predictors and doctor recommended supplement use. All analyses were conducted using SPSS 22.0 (IBM Corp. Released 2013. Armonk, NY).

Results

Descriptive statistics on sample demographics can be found in Table 1. The mean age for the sample was 46 years old. Seventy-one percent of the sample was female, 80% were non-Hispanic white, 59% had an annual household income of $60,000 or more, and 46% had a 4-year college education or more. Most of the sample has a medical condition classified as “other” (38%), followed by cancer (36%), neurodevelopmental disorders (14%), cardiogenetic disorders (11%), and congenital malformations (1%). The “other” category was used for conditions that did not appear frequently in the sample and included conditions such as retinal disease, hearing loss, hemophilia, or mitochondrial disorders. More than half the sample (53%) had fewer than 10 healthcare visits in the previous 12 months. The mean number of prescription medications taken was 3.7, and 66% of the sample reported taking supplements recommended by a doctor. Overall, participants exhibited moderate illness perceptions scores (means ranged from 4.2–7.9), except for timeline beliefs (mean=9.0, SD=1.88). More than half the sample endorsed a strong genetic causal model for their illness (52.0%), and 13% were most uncertain about the cause of their illness, indicating they “don’t know” what caused their illness and provided no further responses. Bivariate correlations of illness perceptions and outcome variables can be found in Table 2.

Healthcare utilization

Table 3 presents the results of the multivariate linear regression of IPQ variables on healthcare utilization in the previous 12 months. Adding the IPQ variables did not significantly improve the variance explained by the model (R^2 change=.07, F(10, 180)=1.69, p=.09). In the final model, being female (β=.30, t(198)=4.26, p<.001) significantly predicted healthcare use. Relative to participants with cancer, having a neurodevelopmental disorder (β=−.22, t(198)=−2.62, p=.009), a congenital disorder (β=−.14, t(198)=−2.02, p=.045), or having a disorder categorized as “other” (β=−.13, t(198)= −2.19, p=.03) was associated with significantly fewer healthcare visits. As expected, stronger identity beliefs, indicating greater

symptom severity, were associated with greater healthcare utilization (β=.20, t(198)=2.04, *p*=.04).

Prescription medication use

Table 4 presents the results of the Poisson regression model of IPQ variables on prescription medication use. Adding the IPQ variables significantly improved the model fit compared to the reduced model with only the covariates ($X^2(10)=19.79$, *p*=.03). In the final model, age was positively associated with prescription medication use (B=0.01, Wald $X^2(1)=20.31$, *p*<.001), as was being female (B=.41, Wald $X^2(1)=19.00$, *p*<.001). Relative to participants with cancer, having a cardiogenetic condition (B=.83, Wald $X^2(1)=44.76$, *p*<.001) or a condition classified as "other" (B=.40, Wald $X^2(1)=13.25$, *p*<.001) was associated with greater prescription medication use. Better physical functioning was negatively associated with prescription medication use (B=−.02, Wald $X^2(1)=30.05$, *p*<.001). As expected, stronger treatment control beliefs were associated with greater prescription medication use (B=.034, Wald $X^2(1)=5.69$, *p*=.02). Furthermore, those who most strongly endorsed a genetic cause for their health concern had higher prescription medication use than those who did not most strongly endorse a genetic cause for their health concern (B=.17, Wald $X^2(1)=3.88$, *p*=.049).

Doctor recommended supplement use

Table 5 presents the results of the logistic regression of IPQ variables on supplement use. Adding the IPQ variables significantly improved the model fit compared to the reduced model with only the covariates ($X^2(10)=20.64$, *p*=.024). No covariates were significantly associated with doctor recommended supplement use after controlling for all other variables in the model. Unexpectedly, those with higher timeline scores, indicating they believed their health concern would last a long time, had 1.23 times the odds of using supplements recommended by a doctor than those with lower timeline scores (95% CI=[1.03, 1.46]; *p*=.02). Additionally, those who reported that their health condition caused a higher emotional impact had 1.20 times the odds of using supplements recommended by a doctor than those who reported a lower emotional impact (95% CI=[1.03, 1.40]; *p*=.02).

Discussion

This study extended research on illness perceptions—previously focused on patient populations with relatively well-defined conditions —to an adult patient population undergoing genomic sequencing to find a genetic cause for their medical condition. We hypothesized that illness identity, beliefs about treatment control, and emotional impact would be positively associated with healthcare use and prescription medication use, and that only beliefs about personal control would be positively associated with supplement use.

Unlike prior research, our findings indicated a relatively weak role for illness perceptions compared to demographic and medical variables in these self-management behaviors. In each analysis, only one or two IPQ variables were significantly associated with the outcome of interest after controlling for all other covariates, and these associations differed across outcomes. Although only illness identity beliefs were positively associated with healthcare utilization, both treatment control beliefs and genetic causal beliefs were positively

associated with prescription medication use, and both timeline beliefs and emotional impact were positively associated with doctor recommended supplement use. This suggests that illness perceptions may offer some insight into various self-management behaviors in this population (people with unexplained medical conditions seeking a genetic explanation through WES) beyond that of demographic and medical factors, but their influence is unique to the type of health behavior in question.

In accordance with the chronic nature of their conditions, the mean timeline score in our sample indicated that most individuals believed their condition would last a long time. In addition, more than half the sample endorsed genetic causal beliefs, and an eighth of the sample expressed they “don’t know” what caused their health concern. This speaks to the unique circumstances of the patients in our study, all of whom were seeking to confirm a potential genetic cause for their health concern.

Consistent with literature on illness perceptions (Leventhal et al., 1984), symptom appraisal (i.e., identity) was the most salient illness belief linked with healthcare utilization, suggesting that symptom appraisal is important for deciding how to manage a health concern, specifically whether or not to consult a professional. However, contrary to the theory and other literature on healthcare utilization (Frostholm et al., 2005), emotional impact was not significantly associated with healthcare use in this sample. Similarly, perceived consequences were not significantly associated with healthcare use, despite significant bivariate correlations with healthcare use. This finding may partly be attributed to high correlations among perceived consequences, identity beliefs, concern, and emotional impact in this sample; whereas, in theory, each item represents an independent illness perceptions domain, these results suggest that perceived consequences, concern, and emotional impact may influence healthcare use through similar mechanisms.

This study also found that beliefs about treatment control and genetic causal beliefs were significantly associated with prescription medication use. This finding is consistent with other evidence regarding the joint effects of illness perceptions and treatment beliefs on behavior, which has suggested that beliefs about treatment, not just illness, are important contributors to health self-management behaviors (French et al., 2013; Rajpura & Nayak, 2014). Similar to the previous discussion, these findings also suggest that perceived consequences, identity beliefs, and emotional impact may influence prescription medication use through similar mechanisms, or they can be accounted for through other demographic or medical variables in this sample, and therefore were not independent predictors of prescription medication use.

Our finding linking endorsing a genetic causal model for a health concern with greater prescription medication use supports the importance of considering etiological models of disease when examining self-management behavior. Marteau and Weinman (2006) argue that the strategies that individuals employ to cope with a health concern will in part depend on what the individuals believe caused the health threat. These researchers apply this framework to theorize about behavioral responses to DNA risk information, claiming that individuals who believe their condition is genetic do not adhere to lifestyle recommendations because they believe that the genetic nature of the disease makes it

uncontrollable and/or only responsive to biological treatment. The positive association between genetic causal beliefs and prescription medication use is a compelling contribution to Marteau and Weinman's argument.

No known research has applied a validated measure of illness perceptions to examine the relation between illness perceptions, as identified by the CSM, and supplement use a patient population such as ours; therefore, this study extends the illness perceptions literature to include a distinct self-management behavior patient populations may use to manage their health. Furthermore, doctor recommended supplement use represents a unique type of self-management behavior because, although it is a helpful health management recommendation, it may not carry the same medical connotation that healthcare use and prescription medication use does (Nichter & Thompson, 2006). In this analysis, the results revealed that timeline beliefs and emotional impact were significantly associated with doctor recommended supplement use. When individuals believe their health concern will last for a long time, they may be more willing to explore unconventional or alternative therapies to manage their health, such as supplement use. These findings also highlight effects of emotional impact on the likelihood of taking doctor recommended supplements. This association is consistent with the theoretical and empirical literature on illness perceptions, as well as other research examining the role that emotions play in decision-making (Slovic et al., 2005). Notably, timeline beliefs and emotional impact were also the only two variables significantly associated with supplement use in bivariate correlations, suggesting that the effect of these variables on doctor recommended supplement use cannot be accounted for by other demographic or medical variables. This observation further supports the notion that supplements represent a type of self-management behavior that is uniquely driven by particular beliefs about illness.

In sum, this study found that five different illness perceptions variables were uniquely associated with healthcare use, prescription medication use, and doctor recommended supplement use among symptomatic adults undergoing diagnostic genomic sequencing, and different illness perceptions predicted each outcome. The primary limitation in this study is its cross-sectional nature, which precludes us from drawing causal inferences on the relationship between these illness perceptions and the target outcomes. The reciprocal nature of illness perceptions and behavior also makes it challenging to determine the directionality of influence. Furthermore, the low response rate of eligible participants limits the generalizability of these findings to other similar populations. We also know little about participants' history of seeking information about their presenting health condition, including how long they had been seeking an explanation for its cause. Because their medical history may provide an important context for their current illness perceptions, future research should include key features of patients' medical history.

Another limitation relates to the measurement of our key variables. The Brief Illness Perceptions Questionnaire uses only one or two items to measure each domain identified by the CSM. Although this brief measure has been validated in other studies, one item may not be sufficient to assess complex phenomena such as illness perceptions. Our study used the shortened version of the measure in consideration of participant burden when completing the surveys, but future research should consider using the full measure to more thoroughly

assess illness perceptions. Furthermore, although we contend that healthcare use, prescription medication use, and supplement use broadly represent actions taken by individuals to manage their health, our measures did not distinguish the reasons why individuals sought health care services or took medication or supplements, limiting our inferences about self-management behavior in this sample.

Nonetheless, this study provides useful insight into the self-management behaviors of individuals with medical conditions for which there is no clear etiology. Understanding how illness perceptions contribute to self-management behaviors among these individuals can help guide patient-provider interactions surrounding diagnostic testing and direct health decision-making. This information may also be used to guide the development of guidelines for treating patients with illness that have a suspected, but unclear, cause. This study provides information that can be followed up by further research to build a knowledge base needed to help improve health outcomes of individuals facing medical conditions with uncertain etiology.

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Table 1

Participant characteristics, July 2012 to July 2014 (*n*=200)

	%(n)	*M(SD)*	Observed Range
Age		46 (14.7)	18 – 78
Female	70.5 (141)		
Non-Hispanic White	79.5 (159)		
Annual income $60,000 or more	59.0 (118)		
Four year college education or more	45.5 (91)		
Diagnostic category			
Cancer	36.0 (72)		
Cardiogenetics	11.0 (22)		
Neurodevelopmental disorders	14.0 (28)		
Congenital malformations	1.0 (2)		
Other	38.0 (76)		
Healthcare visits			
0	0.5 (1)		
1	1.5 (3)		
2 to 3	16.0 (32)		
4 to 9	35.0 (70)		
10 to 12	13.5 (27)		
13 or more	33.5 (67)		
Prescription medication use		3.7 (3.3)	0 – 14
Use doctor recommended supplements	66.0 (132)		
Illness perceptions			
Consequences		6.4 (2.98)	0 – 10
Timeline		9.0 (1.88)	2 – 10
Personal control		4.2 (3.05)	0 – 10
Treatment control		6.2 (3.15)	0 – 10
Identity		5.6 (3.10)	0 – 10
Concern		7.4 (2.87)	0 – 10
Understanding		7.9 (2.47)	0 – 10
Emotional impact		5.2 (3.04)	0 – 10
Cause: genetic	52.0 (104)		
Cause: unknown	12.5 (25)		

Table 2

Bivariate correlations of illness perceptions, healthcare use, prescription medication use, and doctor recommended supplement use (*n*=200)

	1	2	3	4	5	6	7	8	9	10	11	12	13
1. Consequences	--	0.017	−0.104	0.017	0.598***	0.631***	−0.034	0.597***	−0.133	0.131	0.22**	0.235**	−0.004
2. Timeline		--	-0.226**	−0.151*	0.07	0.089	−0.063	0.072	0.161*	0.049	−0.076	0.11	0.194**
3. Personal control			--	0.354***	−0.185**	−0.091	0.248***	−0.188**	−0.091	−1.09	0.079	−0.004	0.059
4. Treatment control				--	−0.053	0.057	0.216**	0.055	−0.153*	−0.111	0.118	0.112	0.004
5. Identity					--	0.574***	−0.141*	0.553***	0.018	0.098	0.167*	0.219*	−0.002
6. Concern						--	−0.017	0.573***	−0.137	0.184**	0.186	0.222**	0.072
7. Understanding							--	−0.065	0.089	−0.154*	0.092	0.14*	0.115
8. Emotional impact								--	−0.138	0.182**	0.175*	0.223**	0.139*
9. Cause: genetic									--	−0.393***	−0.063	0.049	0.113
10. Cause: unknown										--	0.026	0.026	0.016
11. Healthcare use											--	0.236**	0.052
12. Prescription medication use												--	0.464
13. Supplement use													--

* p < .05.

** p < .01.

*** p < .001

Table 3

Hierarchical linear regression of healthcare use on illness perceptions (*n*=200)

Independent variable	B	SE	β	*t*	p
Consequences	0.052	0.039	0.132	1.348	0.179
Timeline	−0.018	0.043	−0.03	−0.410	0.683
Personal Control	0.022	0.029	0.056	0.762	0.447
Treatment Control	0.012	0.029	0.031	0.411	0.681
Identity	0.077	0.038	0.204	2.042	0.043
Concern	0.025	0.038	0.062	0.659	0.511
Understanding	0.001	0.035	0.001	0.019	0.985
Emotional impact	−0.018	0.035	−0.046	−0.500	0.617
Cause: genetic	−0.043	0.176	−0.018	−0.246	0.806
Cause: unknown	0.158	0.264	0.044	0.598	0.551

Note: R^2=0.261; R^2 change=0.07; (F(10, 180)=1.69, p=0.085)

Table 4

Poisson linear regression of prescription medication use on illness perceptions (*n*=200)

Independent variable	B	SE	Wald	df	p
Consequences	0.005	0.020	0.067	1	0.800
Timeline	0.022	0.024	0.897	1	0.340
Personal Control	0.011	0.014	0.635	1	0.430
Treatment Control	0.034	0.014	5.694	1	0.020
Identity	−0.009	0.019	0.254	1	0.610
Concern	0.017	0.020	0.744	1	0.390
Understanding	0.031	0.019	2.715	1	0.100
Emotional impact	0.012	0.018	0.472	1	0.490
Cause: genetic	0.172	0.088	3.878	1	0.049
Cause: unknown	0.213	0.128	2.746	1	0.100

Note: Likelihood ratio $X^2(19)=208.61$, $p<0.001$

Table 5

Logistic regression of supplement use on illness perceptions (*n*=200)

Independent variable	OR	95% CI		p
Consequences	0.89	0.76	1.05	0.173
Timeline	1.23	1.03	1.46	0.021
Personal Control	1.11	0.98	1.25	0.115
Treatment Control	0.99	0.87	1.11	0.811
Identity	0.91	0.77	1.07	0.236
Concern	1.08	0.92	1.27	0.341
Understanding	1.07	0.93	1.24	0.341
Emotional impact	1.20	1.03	1.40	0.019
Cause: genetic	0.50	0.24	1.06	0.490
Cause: unknown	0.67	0.22	2.06	0.672

Note: Model $X^2(19)=31.42$, p=0.04; Nagelkerke $R^2=0.201$