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Factors Associated with Perceived Uncertainty among Parents of Children with Undiagnosed Medical Conditions

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Abstract

Uncertainty is a pervasive characteristic of illness. Yet little is known about the individual or situational factors that contribute to perceptions of uncertainty. The present study aims to examine the factors that contribute to perceived uncertainty among parents of a child with an undiagnosed condition. Two hundred sixty-six parents of a child, or children, affected by an undiagnosed medical condition for at least two years completed an electronically administered mixed-methods survey assessing theoretical predictors of perceived uncertainty. Multivariate linear regression analyses were used to identify the relationship of key variables to perceived uncertainty. Parents' perceived control and optimism were negatively associated with uncertainty ($B = -4.044$, $p \leq 0.001$, $B = -0.477$, $p \leq 0.05$). Subjective disease severity was positively associated with perceived uncertainty ($B = 1.797$, $p \leq 0.05$). Our findings suggest that parents who experience greater uncertainty feel less control over their child's medical condition, which may lead to less effective coping and poorer adaptation. Parents who are less optimistic or who perceive their child's disease as more severe may benefit most from interventions that target situations where parents perceive the least control, thereby enhancing coping and ultimately, adaptation.

Keywords

uncertainty; rare diseases; perceived control; parents; undiagnosed

INTRODUCTION

Uncertainty pervades health care and arises from various sources including absent, vague or complex health information and indeterminate future outcomes [Han et al., 2011]. Mishel's Theory of Uncertainty in Illness suggests that when an event is perceived as uncertain it is because an individual is unable to determine the meaning of illness-related events. This occurs when an individual is unable to assign definite values to objects and events and/or is unable to accurately predict outcomes because sufficient cues are lacking [Mishel, 1988]. Central to this definition is that the uncertainty of an event must be appraised; it is neither inherently good nor bad. Indeed, some parents report personal growth as an outcome of uncertainty and recognize that an uncertain outcome leaves open the possibilities for a positive outcome in their child [Clarke-Steffen, 1993; Cohen, 1993; Rosenthal et al., 2001]. However, research has also demonstrated that the consequences of parental uncertainty can

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include anxiety, depression, cognitive disturbances or helplessness [Jessop and Stein, 1985; Grootenhuis and Last, 1997a; Grootenhuis and Last, 1997b], which have significant health costs [Mitchell and Hauser-Cram, 2008; Raphael et al., 2010]. These qualitative studies on the outcomes of perceived uncertainty suggest that it may hinder or facilitate a parent's adaptation to a health threat in their child.

In spite of its impact on individuals, families and populations, there has been little research to identify individual or disease characteristics that contribute to perceived uncertainty. The Transactional Model of Stress and Coping provides a conceptual framework for identifying these variables. In this model, an individual's response to an event (such as the realization that his or her child has a medical condition), is based on his or her appraisals. Appraisals consist of an individual's evaluation of whether the event is an opportunity or a threat, as well as the individual's assessment of what can be done about the situation. Appraisals contribute to the individual's choice of coping strategies, which mediate adaptation [Lazarus and Folkman, 1984]. Here, we considered perceived uncertainty, perceived control and disease severity as appraisals. We identified other variables from the research literature, dispositional optimism and time elapsed since diagnosis, that we hypothesized contribute to perceptions of uncertainty and that likely interact with constructs in the Transactional Model.

Research has demonstrated the important role of perceived personal control in understanding a patient's response to a health threat [Thompson et al., 1993; Steptoe and Wardle, 2001]. Strategies that have been identified as helping individuals obtain greater control over a health condition, such as gathering information about a diagnosis, prognosis, or recurrence risk of disease [Meissen et al., 1991; Mennie et al., 1993; Michie et al., 1997], are rarely available to parents of children without a diagnosis. Thus, perceptions of control may be predicted to be lower in parents of children with an undiagnosed medical condition and a critical factor influencing parents' experience of uncertainty.

Dispositional optimism has long been demonstrated to influence responses to a wide variety of adverse or difficult situations, including heath threats [Scheier et al., 1989; Allison et al., 2000]. Similarly, optimism appears to play a role in protecting against poor mental and physical health among caregivers of children experiencing a health threat [Ekas et al., 2010; Tifferet et al., 2010]. Carroll and Arthur found a non-significant, negative trend between optimism and perceived uncertainty among patients scheduled to receive an implantable cardioverter defibrillator (ICD) [Carroll and Arthur, 2010]. Similar to health behavior theories, the Transactional Model does not include personality factors likely to affect appraisals and coping. Nonetheless, the identification of traits that contribute to perceived uncertainty may allow clinicians to focus intervention efforts on individuals who are likely to adversely experience greater uncertainty.

Among parents of children with diagnosed conditions, disease characteristics that increase uncertainty include unknown etiology and prognosis and unpredictable symptoms or episodes [Jessop and Stein, 1985; Cohen, 1993; Horner, 1997]. Objectively measured disease severity has also been associated with maternal uncertainty in a small sample of mothers of hospitalized children [Tomlinson et al., 1996]. These results are consistent with the predictions of the Transactional Model and suggest that disease-related characteristics may also be associated with uncertainty in parents of children whose illness is undiagnosed.

The period of time prior to receiving a diagnosis has been identified as one of great uncertainty for parents [Lenhard et al., 2005]. Retrospective and longitudinal investigations have suggested that time may modify the appraisal of uncertainty [Graungaard and Skov, 2007] and parents' desire for a diagnosis [Rosenthal, et al., 2001; Makela et al., 2009]. Time

passage affects perceptions of a health threat and can be hypothesized to influence appraisals and coping. We sought to clarify the nature of the relationship between perceived uncertainty and the amount of time a child has been sick and undiagnosed.

The aims of the present study were to examine whether perceived control, dispositional optimism, disease severity and the amount of time elapsed since a child was identified as sick were associated with perceived uncertainty about a child's undiagnosed medical condition. These variables were chosen based on the relationships predicted by the Transactional Model of Stress and Coping and the empiric literature. We hypothesized that (i) less perceived uncertainty would be associated with greater perceived personal control; (ii) less perceived uncertainty would be associated with greater dispositional optimism; (iii) less perceived uncertainty would be associated with greater disease severity; (iv) less perceived uncertainty would be associated with greater time elapsed since a child was identified as sick; (v) higher perceptions of control, greater dispositional optimism, less disease severity and greater time elapsed since child became sick, would predict lower perceptions of uncertainty.

MATERIALS AND METHODS

Participants and Procedures

Mothers and fathers older than18 years of age with a living child or children affected by a medical condition that remained undiagnosed for more than two years who could read and write English, were invited to participate. Only one parent per household was eligible. A sample size calculation revealed that 225 respondents would provide 80% power (at an alpha level of 0.05) to detect a 3% effect of a single key variable on the primary outcome variable, uncertainty, in a linear regression model where the variance of all other predictors in the model was estimated at 10%.

Participants in the present study were recruited from disease advocacy organizations, patient support groups, online message boards and blogs that were identified through online searches using keywords such as “undiagnosed,” “syndromes without a name” “parents of chronically ill children” and discussions with parents of undiagnosed children who were known to the authors. Websites, forums and organizations that hosted the invitation were reviewed for their legitimacy and the appropriateness of their content before posting. These websites and organizations addressed issues relevant to the undiagnosed pediatric disease communities in Australia, Canada, Ireland, New Zealand, the UK and the U.S.A. Individuals who met the eligibility criteria completed an electronically-administered survey. Recruitment occurred between October 2009 and July 2010. The study was approved by the National Human Genome Research Institute Institutional Review Board and was granted a waiver of written informed consent.

Measures

Sociodemographic characteristics—Data were collected on respondent's age, sex, race, ethnicity, marital status, country of residence, educational level, and income. Additionally, data were collected on how many children without a diagnosis the participant had and each child's age.

Parents Perception of Uncertainty in Illness Scale—The 31-item Parents' Perception of Uncertainty in Illness Scale (PPUS) measures the uncertainty parents experience related to their child's illness [Mishel, 1983]. Each item has five response options ranging from 1 (strongly disagree) to 5 (strongly agree), yielding scores from 31 to 155 with higher scores indicating greater perception of uncertainty. In this population, the full scale

Cronbach's alpha was 0.86. The PPUS is composed of four dimensions of uncertainty consistent with theoretical and empirical predictions. The four domains include: Ambiguity (the absence or vagueness of cues concerning the planning or carrying out of care for the child [13 items]), Lack of Clarity (receiving or perceiving information about the child's treatment and the system of care [9 items]), Lack of Information (the absence of information concerning the diagnosis and the seriousness of condition [5 items]), and Unpredictability (the inability to make daily or future predictions concerning symptoms and illness outcome [4 items]). The reliability coefficients from the present study for the four subscales are: 0.80, 0.76, 0.52, and 0.53, respectively. Based on their poor internal consistency, data from the Lack of Information and Unpredictability sub-scales were excluded from analyses based on PPUS subscales, but they were included in analyses based on the total PPUS scale. The Cronbach's alpha for the PPUS including two subscales combined and all four subscales are, respectively, 0.85 and 0.86.

Perceived personal control—Participants were asked, “On a scale of 1 to 7, how much control do you feel you have over your child's undiagnosed medical condition?” where 1 = `no control' and 7 = `a lot of control.' This item was modified from an item previously used to assess parents' perception of control over their child's medical condition [Lipinski et al., 2006].

Dispositional optimism—Dispositional optimism was measured using the Life Orientation Test - Revised (LOT-R) [Scheier et al., 1994]. This is a ten-item measure in which four items are filler. In the original scoring of the LOT-R, 0 = strongly disagree and 4 = strongly agree. Items were summed for a composite score in which a higher score indicated greater optimism. The Cronbach's alpha was 0.84.

Medical history—For each undiagnosed child, respondents were asked at what age they first realized their child's health was compromised (prenatally, at <6 years of age, 6 – 12 years, 13 – 18 years) and how long s/he has known each child's health was different (1 year or less, 2 – 5 years, 6 – 10 years, 11 – 15 years, over 15 years, or the respondent could indicate the exact amount of time he or she has known). Respondents were asked to assess the severity of each undiagnosed child's medical condition on a seven point scale from `very mild' (1) to `very severe' (7).

Open-ended questions—To better understand responses to items that quantitatively measured constructs and because these constructs have primarily been explored through semi-structured interviews, we included open-ended questions after some of the closed questions. After the item regarding perceived control, parents were asked to elaborate on the ways they felt they did and did not have control over the child or children's undiagnosed medical condition. After assessing their severity of their child's disease, we invited parents to describe the features of each child's condition and which day-to-day activities were affected by the condition.

Data analysis

Quantitative analysis—All analyses were carried out using PASW 19.0. To determine whether sociodemographic, medical or other variables were associated with uncertainty, we used Pearson's correlation coefficient to examine associations between continuous outcome and predictor variables, two-sided Student's t-test to examine associations between continuous outcome and binary variables and one-way analysis of variance (ANOVA) when the variable had three or more categories. Because almost all respondents were White non-Hispanic females, race, ethnicity and gender were not included as covariates. Four items with multiple response categories were collapsed into fewer categories before multivariate

analyses were performed because of the small percentage of respondents in some categories: time elapsed since parent determined that the child was ill (2 categories: <6; ≥6 years), relationship status (2 categories: currently in committed relationship; not in a committed relationship), country of residence (2 categories: U.S.; non-U.S.), and, the highest level of education completed (4 categories: elementary school, high school/GED, technical school; some college; college graduate; graduate school).

Multiple linear regression was performed using stepwise backwards elimination to determine the relevant contribution of the independent variables on the variance in uncertainty and its components as measured by the subscales. We used a $p<0.05$ criterion for determining the statistical significance of independent variables. The relationships of independent to the dependent variable (perceptions of uncertainty) were assessed in models that controlled for an *a priori* selected set of covariates, and one covariate that was tested for inclusion in the model. The independent variables assessed for their relationships with perceptions of uncertainty were: optimism measured by the LOT-R score, perceived personal control (PPC) score, perceived disease severity and the amount of time a child has been sick and undiagnosed. The *a priori* set of covariates included in each model were: respondent's age, highest education level attained (a categorical variable), current country of residence and whether the respondent was currently in a committed relationship. These variables were selected *a priori* for inclusion as covariates because they had either been identified in the literature as likely to contribute to perceptions of uncertainty, or are part of the Transactional Model of Stress and Coping and thus were predicted to contribute to perceptions of uncertainty. One additional variable that has not been previously shown in the literature to be a confounder, but was identified by our study team as a potential confounder, was tested for inclusion in the model. This variable was a measure of the respondent's number of children without a diagnosis. It was included in the model as a confounder if it satisfied a $p<0.20$ criterion in a bivariate test of association with the dependent variable. Final models contain independent variables with a $p \leq 0.05$ and all potentially confounding variables. Although child's age was considered as a possible confounding variable, it was not included in any model because of its co-linearity with the independent variable measuring the amount of time the child has been sick and undiagnosed. Co-linearity was not found when analyzing other predictor variables. Only respondents for whom there were complete data were included in each regression analysis. Available cases for individual regressions ranged from 195 – 203 (73 – 76% of full sample).

Qualitative analysis—Responses to open-ended questions were analyzed with NVivo. A preliminary codebook was developed based on previously published research in this population that used qualitative methods. As text was coded, it was compared to the established codes and emerging codes were added when an appropriate code could not be identified. Through this analysis a final codebook was developed. A second investigator coded 25% of responses and inter-rater consistency was evaluated to ensure reliability of findings. When significant differences in coding of a response were found, data and codes were discussed. If the coders agreed that the definition of a code required further clarification or if a new code emerged from the discussion, then codes and definitions were amended as appropriate. Coded text was analyzed to make inferences about the meaning(s) underlying respondents' responses. When key themes were identified, they were cross-referenced to other codes to further interpret and explain respondents' experiences.

RESULTS

Participants

Of the 391 individuals who started the survey, 39 self-declared as ineligible; 17 indicated they were eligible, but their responses to an open-ended question indicated otherwise; and 69 individuals indicated that they were eligible, but completed none of the survey. Thus, responses from 266 individuals were included in this analysis. A response rate cannot be calculated because we do not know the total number of individuals who had access to the web link but chose not to complete the survey.

Participant characteristics

Table I shows the sociodemographic data on the sample. Participants were mostly White, non-Hispanic, middle-aged, had undergraduate or graduate degrees, in a committed relationship and female.

Descriptive results

The distributions of responses to the psychological variables are shown in Table II. Here we analyze these values in relation to normative data from other populations. Mean PPUS total and subscale scores were significantly higher in this sample than among parents of diagnosed children ($p<0.001$) (data not shown) [Mishel, 1997]. The mean perceived personal control score was 2.96 (range 1–7). Mean LOT-R scores in this sample were significantly lower than scores reported for college students ($p<0.001$) and individuals awaiting coronary artery bypass surgery ($p<0.001$) (data not shown) [Scheier, et al., 1994]. The perceived disease severity scale mean was 5.23 (range 1 – 7). No respondents endorsed `very mild' (1) to describe their child's disease. Neither the mean perceived personal control nor disease severity scores were significantly different than those obtained using similar questions among parents of children with rare disorders (data not shown) [Lipinski, et al., 2006].

Bivariate analyses

Bivariate analyses in Table III indicate that lower perceived personal control was associated with greater perceived uncertainty as measured by the total PPUS scale and two subscales, ambiguity and lack of clarity. Greater dispositional optimism was associated with less perceived uncertainty as measured by the total and two subscales of the PPUS. Mean PPUS scores for parents whose child was undiagnosed for <6y were significantly higher than parents whose child was undiagnosed for ≥6y. Greater perceived disease severity was associated with greater perceptions of uncertainty. In addition to these hypothesized relationships, neither highest education achieved nor respondent age were associated with mean PPUS scores.

Multivariate analyses

Uncertainty full scale—As shown in Table IV, individuals who perceived less personal control over the child's undiagnosed medical condition, were less optimistic, and perceived their child's disease to be more severe had significantly higher uncertainty scores. These three variables explained 23% of the variance (adjusted R^2) in overall uncertainty. Greater time elapsed since the child was identified as sick was not included in the final model because it did not satisfy the criterion $p \leq 0.05$ and none of the sociodemographic variables were statistically significantly associated with the full scale.

Ambiguity subscale—Individuals who perceived less personal control over the child's undiagnosed medical condition, were less optimistic, and perceived their child's disease to

be more severe had significantly higher scores in ambiguity. These three variables explained 19% of the variance (adjusted R^2) in ambiguity. Greater time elapsed since the child was identified as sick was not included in the final model because it did not satisfy the criterion $p \leq 0.05$ and none of the sociodemographic variables were statistically significantly associated with this subscale.

Lack of clarity subscale—Individuals who perceived less personal control over the child's undiagnosed medical condition and were less optimistic had significantly higher scores in lack of clarity. These two variables explained 18% of the variance (R^2) in lack of clarity. Greater time elapsed since the child was identified as sick was not included in the final model because it did not satisfy the criterion $p \leq 0.05$ and none of the sociodemographic variables were statistically significantly associated with this subscale.

Qualitative analyses

Two hundred and 209 individuals, respectively, responded to the questions asking respondents to identify areas where they do and do not have control over their child's medical condition. The percentage agreement for the text coded from this question ranged from 79.7 – 99.8% and Cohen's kappa ranged from 0.71 – 0.99. In identifying areas where they do have control, many parents implicitly identified the strategies they use to cope with their child's condition and the uncertainty associated with it. Specifically, four themes emerged from the analysis: information and decision-making about health care, advocacy, child's comfort and self-care. Information and decision-making about health care included areas such as monitoring the child's condition and maintaining accurate records, increasing one's knowledge about medicine and making testing or treatment decisions (e.g. “Most things that have been done to her have directly impacted her in a negative way. We take the control back and do what we think is right and she does much better.”). Advocacy included activities such as voicing the child's needs to a health care provider, presenting research to health care providers and encouraging health care providers to care for their child (e.g. “He has made me learn to fight and become persistent in the health care world to gain access to the care my child needs.”). Child's comfort was composed of activities such as maintaining the child's routine and environment, controlling his/her diet, and providing emotional support to the child (e.g. “I am in control of their daily environment to ensure it is safe, loving and helps them to be the best they can be.”). Self-care activities were those focused on the parent and were often done with the goal of helping the child. These included maintaining a positive attitude and praying (e.g. “I see a difference in my child when I am positive.”).

The themes that emerged from the question about things over which parents do *not* have control were: disease, future, medical care, and isolation. The theme of disease included disease stability, the child's limitations and information about the diseases (e.g. “When she has bad days there is nothing I can do to make it better or stop.”). The theme of future included short- and long-term planning, recurrence risks for the respondent and the respondent's family, and concerns about who will care for child after respondent is not able to (e.g. “We don't have control over what the future holds for anyone anyway, but with an undiagnosed child you cannot really plan or dream because those hopes and dreams can be dashed in a second.”). When respondents described not having control over medical care, they described experiences such as disagreeing with a health care provider's decision, negative experience with health care providers, and the lack of treatment available (e.g. “All of my son's doctors seem happy to wait until he gets very sick again before taking action.”). The theme of isolation included isolation due to a lack of models on which to base expectations, not knowing what to tell others and inability to gain the acceptance of others (e.g. “You can't get support from others who have been on the same journey when you don't

know what the journey is."). Over 10% of the respondents (22) indicated that they have no control over *anything*.

DISCUSSION

Uncertainty has been suggested as both a barrier and a facilitator of adaptation to a health threat. Yet there is limited information on key constructs associated with perceptions of uncertainty. This is particularly true among parents when the uncertainty arises from a health threat to their child [Jessop and Stein, 1985; Grootenhuis and Last, 1997a; Grootenhuis and Last, 1997b]. The present study contributes data on the relationships between optimism, disease severity, time elapsed since a child was identified as sick and perceptions of uncertainty in a mixed methods study with a large sample size, and replicates the previously described relationship between perceived uncertainty and personal control [Lipinski, et al., 2006].

Although increasing perceived control has been proposed as a goal of genetic counseling [Shiloh et al., 1997; Berkenstadt et al., 1999], it has infrequently been studied as a variable in understanding a parent's response to their child's health condition. Our bivariate and multivariate analyses indicate that when parents perceive greater uncertainty, they perceive less control over their child's condition. The cross-sectional study design does not permit us to determine whether greater uncertainty leads to lower perceived control or *vice versa*. Our findings replicate those of Lipinski et al. [2006] who investigated the relationship between perceived control and uncertainty among parents of children with rare chromosomal abnormalities. We extend this work by demonstrating this relationship among the parents of children without a diagnosis. Genetic counseling has been identified as a potential point of intervention to improve outcomes under uncertain conditions [Baty et al., 2006; Lipinski, et al., 2006]. For example, genetic counselors may be able to assist parents in indentifying areas where they do have (some) control. With training, genetic counselors may use interventions that coach parents in using more effective coping strategies. Such interventions include Coping Effectiveness Training, in which individuals identify the controllable and uncontrollable aspects of their situation and are assisted in identifying coping strategies that are predicted to best match the controllability of the stressor [Chesney et al., 1996].

As hypothesized, there was a significant negative association between optimism and perceived uncertainty in bivariate analysis that remained significant when controlling for other variables in regression analysis. This is consistent with the limited research on the relationship between these variables [Carroll and Arthur, 2010]. There are several possible mechanisms underlying this relationship. It is possible that optimists perceive less uncertainty because of the optimist's general expectation for a positive outcome. Expecting a positive outcome may preclude one from perceiving a situation as having an uncertain outcome. It is also possible that optimists perceive a situation as less uncertain because they attend less to the uncertainty. Further research could replicate or refute our findings and identify the mechanism(s) that underlie this association. These results suggest that clinicians screen more pessimistic parents to better target interventions towards those individuals most in need of support. However, we are unaware of clinical screening tools for levels of optimism. As the LOT-R contains only 10 items, it may be incorporated into a clinic's pre-visit paperwork without significantly burdening patients or clinicians and used to target services.

Subjective disease severity was significantly associated with overall perceptions of uncertainty in bivariate and multivariate linear regression analyses. The association between subjective disease severity and perceived uncertainty suggests that if a child's illness is perceived as worse, a parent may believe that the uncertainty may have a more negative

resolution. This interpretation is consistent with previous findings which have suggested that even after receiving a diagnosis, parents have long lasting concerns about their child's health, cognitive abilities and lifespan [Stewart and Mishel, 2000; Lenhard, et al., 2005]. It is also possible that the association between perceived disease severity and uncertainty may reflect parents' perceptions that disorders about which little is known are more serious. Clinicians may help parents by asking about their perceptions of the severity of the child's disease. If there exists significant differences in the parent's and clinician's perceptions, further clarification may reduce the perceived uncertainty of the situation.

Contrary to our hypothesis, the significant relationship between time elapsed and perceived uncertainty in the bivariate analysis was not found in multivariate linear regression. A previous study that used qualitative interviews suggested that time may modify parents' appraisals of uncertainty [Graungaard and Skov, 2007]. The hypothesized relationship may not have been found in our study because of measurement differences (perceived uncertainty versus emotional response to uncertainty). Indeed, as the time a child has been ill increases, so may a parent's awareness of the extent of ignorance about his or her condition.

In contrast to previous findings, in this population neither the respondent's age [Lipinski, et al., 2006] nor their level of education [Mishel, 1997] was associated with perceptions of uncertainty in bivariate and multivariate analyses. Mishel [1983] hypothesized that parents with greater education would have more skills at accessing information about a disorder. The lack of association in our sample between education and perceived uncertainty may be explained by the contrast to previous studies that have investigated parents of diagnosed children, where it may be assumed that more information is available. Alternatively, previous studies have relied on small samples that were not generalizable [Mishel, 1997]

Our qualitative results indicate that parents feel control over some aspects of their undiagnosed child's health care but little control over others. For those areas in which parents perceived control, they often identified both problem- and emotion-focused strategies that they used to cope with the situation (e.g. maintaining accurate records, advocating for their child, keeping a positive outlook). The areas that parents identified and the coping strategies they used (when described) are similar to the findings from studies of parents of undiagnosed children that relied exclusively on qualitative methods [Rosenthal, et al., 2001; Graungaard and Skov, 2007; Lewis et al., 2010].

Limitations

There are several limitations to the present study. Respondents were more often White, middle-aged, well educated, and female. Although not representative of the general population, these characteristics do mirror many characteristics of individuals who use the internet to obtain information about health issues [Anker et al., 2011]. Participants in the present study were recruited from disease advocacy and support organizations, online message boards and blogs; thus, the present study likely has biases in ascertainment, response and recall. Because data were not obtained from non-respondents, we do not know whether respondents were significantly different from non respondents in any of the measured variables. The low mean LOT-R score suggests that there was a significant bias towards pessimism in this sample. We used single items to measure perceived personal control and disease severity. Both constructs are likely multi-dimensional, thus the complex nature of the relationships between these variables was likely not fully captured.

Conclusions

Our study has identified individual and disease characteristics that contribute to perceptions of uncertainty. Parents who perceive less control over their child's health perceive greater

uncertainty; this is particularly true for less optimistic parents and parents who feel their child's disease is more severe. Further studies are necessary to elucidate the causal pathways among these variables. Identifying parents who are less optimistic and those who perceive their child's disease as more severe may allow clinicians to focus interventions on those parents in the greatest need of support. The Transactional Model and previous research indicate that parents who report greater perceived uncertainty and less perceived control may use less effective coping strategies. Parents of undiagnosed children may benefit from interventions designed to help them feel less uncertain by clarifying what is known about their child's condition and emphasizing those areas over which parents do have control as gathered from our qualitative analysis: gathering information to enhance health care decisions, advocating for their child, ensuring the child's comfort and caring for themselves.

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Table I

Sociodemographic and clinical characteristics of sample (n = 266)

Variable	n (%)
Gender	
Female	228 (97)
Male	8 (3)
Ethnicity	
Not Hispanic/Latino	222 (96)
Hispanic/Latino	10 (4)
Race	
American Indian/Alaska Native	1 (0.4)
Asian	6 (3)
Native Hawaiian or other Pacific Islander	0
Black of African American	2 (1)
White	210 (91)
More than one race	12 (5)
Relationship status	
Committed relationship	218 (83)
Not committed relationship	38 (17)
Residency	
USA	180 (82)
Not USA	38 (18)
Highest level of education attained	
Elementary school, High school/GED, Technical college	27 (12)
Some college	52 (24)
College graduate	82 (37)
Graduate school	59 (27)
Annual household income ($)	
<30,000	13 (15)
30,000 – 50,000	14 (16)
50,001 – 70,000	24 (27)
>70,000	38 (42)
Age of respondent (mean (range))	40y (22 – 68y)
Age of 1st child described (mean (range))	9.5y (2m – 31y)
Number of undiagnosed children	
1	188 (86)
2	25 (11)
3	5 (2)
4	1 (0.4)
Length of time 1st child described has been sick	
<6 years	112 (51)
≥6 years	107 (49)

Table II

Means and standard deviations of key variables and primary outcomes.

Variable (scale range)	Mean (SD)	Range
Perceived personal control (1–7) (General perceptions of control over child undiagnosed condition)	2.96 (1.6)	1 – 7
Perceived disease severity (1–7) (General perceptions of disease severity)	5.23 (1.3)	2 – 7
Life Orientation Test – Revised (0 – 24) (optimism)	12.8 (4.7)	0 – 24
Parents' Perception of Uncertainty in Illness Scale (31 – 155)	107.8 (15.4)	62 – 152
Ambiguity subscale (13 – 65)	45.8 (8.1)	20 – 65
Lack of Clarity subscale (9 – 45)	29.6 (6.0)	15 – 45
Lack of information subscale (5 – 25)	18.5 (3.2)	8 – 25
Unpredictability subscale (4 – 20)	14.0 (2.9)	7 – 20

SD, standard deviation

Table III

Bivariate associations between the primary outcome, sociodemographic, and medical variables.

Parents' Perception of Uncertainty in Illness Scale						
	Total Scale		Subscales			
	Uncertainty		Ambiguity		Lack of clarity	
ANOVA	*F*(d.f)	p value	*F*(d.f)	p value	*F*(d.f)	p value
Time elapsed since child identified as sick (0–5y vs. 6+y)	**4.287 (1)**	**0.040**	2.397 (1)	0.123	3.037 (1)	0.083
Number of undiagnosed children (1 – 4)	1.544 (3)	0.204	0.648 (3)	0.585	**1.463 (3)**	**0.023**
Pearson correlation	Coefficient	p value	Coefficient	p value	Coefficient	p value
Perceived personal control	**−0.394**	**<0.001**	**−0.319**	**<0.001**	**−0.389**	**<0.001**
Dispositional optimism	**−0.209**	**0.001**	**−0.254**	**<0.001**	**−0.222**	**0.001**
Disease severity	**0.188**	**0.006**	**0.272**	**<0.001**	0.061	0.380

Statistically significant associations (p≤0.05) are **bold**

Table IV

Multiple variable linear regression analyses predicting uncertainty.

	Outcome variable		
	Total scale	Subscales	
Predictor variable	**Uncertainty** **B** **CI**	**Ambiguity** **B** **CI**	**Lack of clarity** **B** **CI**
Perceived personal control (1 – 7)	−4.044[b] −5.245 – −2.844	−1.642[b] −2.293 – −0.990	−1.441[b] −1.924 – −0.964
Optimism (0 – 24)	−0.477[a] −0.884 – −0.070	−0.290[c] −0.509 – −0.070	−0.231[c] −0.388 – −0.074
Disease severity (1 – 7)	1.797[a] 0.314 – 3.279	1.377[b] 0.589 – 2.165	—

Table includes all predictor variables that remained in the model after backwards step elimination. The confounding variables respondent's age, country of residence, highest education completed, and whether respondent is in a committed relationship are included as covariates in all models. Data not shown because relationships were not statistically significant. B, unstandardized regression coefficient, CI, 95% confidence interval.

[a] $P \leq 0.05$,

[b] $P \leq 0.001$,

[c] $P \leq 0.01$