



# Diagnostic uncertainty in patients, parents, and physicians: a compensatory control theory perspective

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## ABSTRACT

Medical diagnoses offer a structure by which psychological uncertainty can be attenuated, allowing patients to diminish psychological threats and focus on health prognosis. Yet when no diagnosis can be made, patients may experience *diagnostic uncertainty* – perceiving the medical field as unable to provide an accurate explanation of the cause of their health problems. This review examines the psychological threat that diagnostic uncertainty imposes on individuals' need for control and understanding, and the resulting consequences experienced by patients, parents of pediatric patients, and physicians. Using compensatory control theory as a framework, we propose a taxonomy of behaviors that people may adopt in order to regain control in the face of diagnostic uncertainty and to reaffirm that the world is not random and chaotic. To manage diagnostic uncertainty, people may bolster their personal agency, affiliate with external systems they see as acting in their interest, affirm clear connections between behaviors and outcomes, and affirm nonspecific epistemic structure. Diagnostic uncertainty is approached from the perspectives of patients, parents of pediatric patients, and physicians, demonstrating how each group responds in order to maintain a sense that the world has structure and is not random. Discussion centers on moderators, limitations, and implications for clinical practice.

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A patient experiences symptoms with no known origins and goes through multiple inconclusive tests over a period of months. A parent tries to simultaneously understand the symptoms being experienced by their child and the multiple unclear diagnoses offered by the physician. A physician is confronted with multiple inconclusive tests in their attempt to diagnose a rare or poorly understood medical condition or is trying to diagnose a patient with an undifferentiated symptom, such as abdominal pain, that could be a symptom of multiple, common conditions. In all of these cases, the patient, parent, or physician is experiencing diagnostic uncertainty, which has been defined as the 'subjective perception of an inability to provide an accurate explanation of the patient's health problem (Bhise et al., 2018, p. 111).' The consequences of diagnostic uncertainty are vast and detrimental, as it is associated with emotional distress, depression and anxiety, disability, pain intensity, and pain catastrophizing (Geisser & Roth, 1998; Serbic et al., 2016). Furthermore, the search for a correct diagnosis places a burden on health services, as previous studies have linked diagnostic uncertainty to diagnostic error, over-testing, unneeded surgeries, and increased health-care costs (see Bhise et al., 2018 for systematic review).

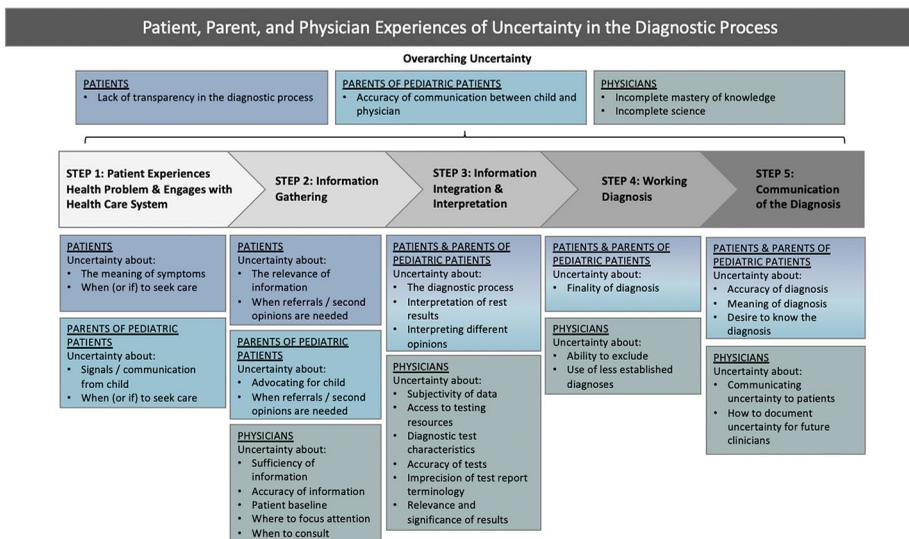
In this paper, we examine diagnostic uncertainty as a situation that is characterized by a lack of control. The term diagnostic uncertainty describes both patients' perceptions that an accurate

diagnosis or explanation of illness is absent, as well as the perception that the given disease label is incorrect (see Pincus et al., 2018 for definition and discussion). Meyer et al. (2021) recently proposed a model of diagnostic uncertainty, specifically noting the overarching uncertainties experienced by patients and clinicians through five steps of the diagnostic process. In the first step, patients experience health problems and engage with physicians as part of the healthcare system. In the second step, patients gather information about the health problem, including information provided by physicians. In the third step, this information is integrated and interpreted by patients and physicians. In the fourth step, a working diagnosis is proffered. And in the fifth and final step, the diagnosis is communicated (Meyer et al., 2021).

At each step, there are different aspects of the diagnosis that patients and physicians may feel uncertain about. For patients, these uncertainties include a lack of transparency in the diagnostic process; and for physicians, they must cope with incomplete mastery of knowledge and incomplete science. We adapted their model to include parents of pediatric patients. When it is a child that is experiencing diagnostic uncertainty, the parents may experience an additional, overarching uncertainty regarding the accuracy of communication between their child and the physician. In Figure 1, we elaborate on the uncertainties that patients, physicians, and parents, respectively, may experience at each step of the diagnostic uncertainty process.

Unspecific symptoms may draw out the diagnostic process, as they often result in the frequent diagnosis, and misdiagnosis, of the common ‘unexplained’ medical conditions, such as chronic fatigue syndrome, fibromyalgia, and irritable bowel syndrome (Fitzcharles & Boulos, 2003; Mueller et al., 2016), further contributing to patients’ diminished perceptions of control. Left feeling uncertain about the future, patients and their families may become desperate for the stability provided by a diagnosis (Gahl & Tifft, 2011). Yet when no diagnosis can be made, individuals must find ways to manage the threat to personal control that diagnostic uncertainty imposes.

Although there are many theoretical frameworks that detail the role of psychological control in living with health threats, we focus on compensatory control theory (CCT) (Kay et al., 2009; Landau et al., 2015) as a framework. CCT is built upon the premise that it is psychologically stressful and anxiety provoking to perceive the world as random (Janoff-Bulman, 1992; Pennebaker & Stone, 2004). In order to prevent these feelings of chaos and randomness, people are motivated to perceive



**Figure 1.** Note. Figure adapted from model of patient and clinician experiences of uncertainty in the diagnostic process by Meyer et al. (2021) as informed by the Diagnostic Process Model of the National Academies of Sciences, Engineering, and Medicine (2015).

the world as ordered and structured (Kruglanski & Webster, 1996; Landau et al., 2004; Landau et al., 2006). In situations when personal control is low, people question their core epistemic beliefs that the world is structured and orderly. Individuals may first attempt to assert personal control over their situation by taking behavioral steps, however if that fails to change their circumstances and they cannot restore control, people may then turn to external means of imbuing the world with order (Landau et al., 2015).

We thus propose that compensatory control theory is particularly well suited to explain people's responses to the experience of diagnostic uncertainty, where their understanding and ability to control their health and their future is threatened. Studies drawing from CCT have shown how people draw on larger systems (e.g., government, religion) to provide control when they are in situations of control deprivation (Kay et al., 2008); accordingly, the theory may enable better understanding of how people confront control threats within the medical system. In the compensatory control process, the first step is being in a state of low personal control. In medical situations, this can occur from a reduction of one's personal control during the experience of health problems for which one has not received a clear diagnosis and treatment plan. This experience of low control is generally interpreted as a threat to seeing the world as structured and orderly (Kay et al., 2008; Landau et al., 2015). In typical compensatory control experiments, people are randomly assigned to experience a threat to personal control (vs. experiencing general negativity) and are then assessed on relevant outcome measures. People with diminished control were more likely to see clear patterns in pattern recognition tasks, (Whitson & Galinsky, 2008) and to show greater belief in external systems, ranging from belief in God to trust in government to broader social belief systems (e.g., Axt et al., 2020; Kay et al., 2008, 2010; Ma et al., 2019; Stanley et al., 2020) compared to people whose control was not diminished. These broader belief systems can substitute for personal control and enable a person to maintain a perception of an ordered world when one's personal control is low. This experimental approach may provide, as we outline below, a novel perspective on how to help people address the lack of control they may experience during periods of diagnostic uncertainty when confronting health threats.

As diagnostic uncertainty has not been examined yet through the lens of social psychological theory (see Klein et al., 2015 for discussion), the goal of the paper is to propose an overarching framework for understanding what may seem to be otherwise disparate behaviors engaged in by people confronting diagnostic uncertainty. Drawing on the CCT framework, we propose a taxonomy of behaviors that people may adopt in order to regain control when experiencing diagnostic uncertainty. As the reach of undiagnosed disease extends beyond just the patient, the framework is applied from the perspectives of patients, parents of pediatric patients, and physicians. These three unique perspectives are reviewed, addressing the control diminution that diagnostic uncertainty causes each group. Issues for future research are then considered, specifically focusing on potential moderators, that is gender and socioeconomic status. Lastly, limitations and further implications for use in clinical practice are discussed. Before developing the compensatory control framework further, we first outline other perspectives of control and coping, their potential relation to diagnostic uncertainty, and why the compensatory control process provides a novel perspective for researchers and practitioners.

### **Sustaining perceived control in response to health threats**

Within health psychology, one of the most important contributions to understanding how people cope with chronic health threats is Shelley Taylor's (1983) theory of cognitive adaptation. This theory, which introduced a series of mechanisms by which individuals psychologically manage diagnosed health threats, is an important starting point from which to understand health threats that lack clear diagnoses. The theory of cognitive adaptation centers around three motivations people have: a quest for meaning, an attempt to augment personal self-esteem, and an effort to restore mastery, both over the event and in life in general. It is this effort to restore mastery that may be

most challenging when a patient is experiencing diagnostic uncertainty. In one classic paper, Taylor and colleagues (1984) interviewed women with breast cancer and their families to assess the women's psychological adjustment. It was observed that by making causal attributions to arrive at an understanding of why their cancer originated, people could maintain a greater sense of mastery or control. The vast majority of respondents formed an explanation as to what caused their breast cancer, even though the true cause was not clearly identified (Taylor et al., 1984). From a compensatory control theory perspective, the women's attempts to identify causal explanations for their medical condition can be viewed as an attempt to restore a feeling of order when their perception of control was diminished (Kay et al., 2010).

Most of the breast cancer patients also demonstrated some attempt at psychological control, although their control beliefs varied. Many patients also reported the belief that 'others' could at least somewhat, if not largely, control their cancer. The 'other' agency in which these patients attributed control varied: for the vast majority it was physicians or treatment and for some it was God or a therapist (Taylor et al., 1984). External agents were relied upon to exert control over the situation when personal control was diminished; in doing so, people were, to some extent, able to restore their perception of the world as predictable and non-random. This seminal study identified the importance of attributions in patients' attempt to bolster a perception of control over their diagnosed chronic illness – and we would argue that the same motivations apply to those lacking a diagnosis for their health ailment. It also anticipated an important facet of compensatory control theory, that people will use external systems (e.g., physicians, religion) to assert control when personal control is threatened (Kay et al., 2008).

A second classic approach to understanding control maintenance that is relevant to understanding diagnostic uncertainty stems from research on individuals' adaptive responses to threats via primary and secondary control. People utilize primary control to change the environment to fit their individual needs (Rothbaum et al., 1982). Exerting primary control over one's situation successfully also enables people to see the causal connections between themselves and their outcomes. By contrast, secondary control enables people to align the self with their existing environments (Rothbaum et al., 1982). Four specific types of secondary control have been proposed: *illusory control* is exerted by aligning oneself with the idea that chance is in one's favor; *vicarious control* is utilized when people align themselves with powerful others with whom they may share control; *predictive control* occurs when people feel that they are able to predict outcomes and manage their expectations; and *interpretative control* relies on finding meaning or understanding through attributions (Rothbaum et al., 1982). This model argues that 'inward behaviors' such as submission, passivity, or withdrawal have adaptive purposes in that they promote secondary control. Thus, these behaviors do not lead people to relinquish control but rather to bolster their perceptions of control, facilitating improved adjustment (Rothbaum et al., 1982). As Skinner (1996) noted, the assertions of primary and secondary control are reactions to the loss of control, and are therefore psychological actions people take to compensate for this loss. The dual-process model of assimilative and accommodative coping (Rothermund & Brandstädter, 2003) draws similar distinctions in ways that people engage in compensatory activities to actively diminish threats through assimilative mode of coping that transforms a negative experience (by having it conform with the person's goals), or to adapt to the constraints of the threat through accommodative processes (through disengagement from blocked goals; Rothermund & Brandstädter, 2003). In response to health threats, individuals may not have the ability to exert primary control to change their circumstance through assimilative coping; rather, they must adapt to the situation, in which case secondary control efforts or accommodative coping may enable better coping when control is lost – a compensatory process.

A revised definition of secondary control has been proposed that is particularly relevant to the present discussion, which posits that secondary control is exerted when individuals adjust facets of the self and accept their current circumstance as is (Morling & Evered, 2006). In their review, Morling and Evered (2006) do not emphasize secondary control as promoting a sense of perceived control; rather, they theorize that the direct consequence of secondary control is the experience of

greater fit within one's environment, which, potentially may facilitate a sense of control in later stages of psychological processing. Secondary control, then, has been identified as adaptive for coping in situations that threaten an individual's sense of subjective control. Much research on secondary control centers on how secondary control behaviors facilitate coping in relation to sickness, stress, grief, and misfortune (Morling & Evered, 2006). For example, children diagnosed with leukemia who largely employed secondary coping methods, defined as attempts to adjust themselves to their current condition, were found to manage better than the children who did not partake in secondary coping methods (Weisz et al., 1994). As diagnostic uncertainty stems from health threats largely out of individuals' direct control, secondary control mechanisms may play an important role in aiding adjustment.

As secondary control has primarily been studied in health psychology as a response to clearly diagnosed health threats, diagnostic uncertainty raises additional issues to consider. In response to health threats, especially those without a clear diagnosis, many instances of secondary control may be usefully considered as compensatory control behaviors (Landau et al., 2015). For patients and families experiencing diagnostic uncertainty, their personal ability to enact change within the diagnostic process is limited, leading to a sense of control diminution. The use of secondary control behaviors can be conceptualized as a compensatory reaction to illness that is particularly acute for those facing diagnostic uncertainty. Instances of secondary control, specifically of vicarious control, elicited in response to diagnostic uncertainty, enable people to compensate for feelings of lost control. We return to this point and elaborate on its implications for how both patients and parents of pediatric patients address their diagnostic uncertainty.

Although research on control maintenance in relation to diagnosed disease provides insight as to how illnesses affect control perception, research is limited on control maintenance and diagnostic uncertainty, which presents an additional, epistemic threat to control and understanding (beyond the inherent threat to physical health). For example, patients experiencing diagnostic uncertainty may not be able to establish a sense of mastery over their condition by making causal attributions in the same manner as people with a clear diagnosis (e.g., women with breast cancer in Taylor, 1984). Compensatory control theory may thus offer additional theoretical leverage to the strategies discussed in these classic approaches to control in order to better understand how people cope with diagnostic uncertainty.

### Compensatory control strategies

Possessing a high level of perceived control is correlated with greater adjustment, healthier behaviors, and improved emotional functioning (Tangney et al., 2004). As such, an important aspect of adaptive coping among those facing diagnostic uncertainty is to restore a sense of perceived control. Such situations that threaten perceived control evoke negative arousal, eliciting compensatory responses (Kay et al., 2009). Compensatory control theory postulates that in situations that threaten one's sense of control, people enact four actions to reestablish control: they bolster personal agency, or strengthen the perception that they possess the abilities and resources necessary to achieve certain outcomes or goals; they affiliate with external systems perceived to be acting in one's interest, thus influencing the outcome on one's behalf; they affirm clear connections between behaviors and outcomes, establishing a specific structure on which to rely; and they affirm non-specific epistemic structure, or seek out or prefer structured interpretations of the world unrelated to the control-reducing event (Landau et al., 2015). Further clarification of each of the four compensatory behaviors, as outlined in Landau and colleague's (2015) review, are presented in Table 1. These compensatory responses will vary proportionally to the degree that a person is invested in the domain in which the threat occurs (Landau et al., 2015). Health is a domain vital to life; thus, strong compensatory responses are evoked when control in this domain is threatened. Diagnoses are associated with treatment plans and expected outcomes, enabling the prospect of a timeline. Thus, diagnoses offer a structure by which uncertainty can be reduced, allowing individuals to

**Table 1.** Compensatory Control Behaviors.

Behavior	Description
Bolstering personal agency	Viewing oneself as having the innate capacity to obtain specific desired outcomes.
Affiliating with external systems perceived to be acting in one's interest	Viewing powerful external systems as asserting control over, or intervening in, one's personal outcomes on one's behalf.
Affirming clear connections between behaviors and outcomes	Entrusting that certain actions are associated with, and will result in, specific outcomes in a given domain.
Affirming nonspecific epistemic structure	Maintaining the idea that the world is structured in a simple, clear, and consistent manner such that any intentional act has a probable chance of success.

Note. Behavior descriptions are adapted from Landau et al. (2015).

diminish the imposing threat. The question we address, then, is how is it that individuals manage the control diminution caused by diagnostic uncertainty. Using compensatory control theory as a guide, we approach diagnostic uncertainty from three different perspectives, to understand how patients, parents of pediatric patients, and physicians respond in order to regain control.

### *The perspective of patients*

Living with a disease without a diagnosis affects more than just a patient's physical health. An analysis of illness narratives written by adult and pediatric patients applying to the Undiagnosed Disease Network (2021) revealed patients' accounts of frustration with nameless illness, feelings of irritation as though they must constantly explain themselves to doctors and other people, and beliefs that they have exhausted all other diagnostic alternatives (Spillmann et al., 2017). Patients and their families experience desperation, seclusion, and loneliness, culminating in an experience marked by a sense of suffering and loss in their lives (Gahl & Tifft, 2011; Spillmann et al., 2017). Research adopting a qualitative approach to studying individuals' experiences of chronic back pain supports this sentiment, as people reported that having visible evidence made their disease feel more credible; as such, those experiencing diagnostic uncertainty felt a sense of guilt, and sought further treatment and diagnosis (Serbic & Pincus, 2013). Without a diagnosis, treatment plans may be more tenuous, leaving many aspects of the future uncertain. Lacking knowledge of what is happening within their bodies and potentially feeling let down by healthcare providers, patients experiencing diagnostic uncertainty face a double-edged sword that threatens both their health and their faith in the medical system. With no tangible plan to focus on and an unclear outcome, patients enduring diagnostic uncertainty are left with a search for diagnosis that leaves them struggling to engage in other aspects of their lives (Pincus et al., 2018).

Compensatory control theory identifies behaviors that may be particularly effective to restore personal feelings of authority when control is threatened. A specific control appraisal that may be utilized by patients facing diagnostic uncertainty is the affirmation of nonspecific epistemic structure, as perceived control is entrenched in the idea that the world follows a clear, stable structure (Landau et al., 2015). In other words, believing that events in the world, such as becoming ill, occur as expected and for a reason, may lessen patients' internal feelings of disorder. Although this belief may imply an unfavorable outcome of suffering from disease, the structure it provides may nevertheless be beneficial. Indeed, the directors of the Undiagnosed Disease Program at the NIH reported that, 'In general, individuals who have lived with rare diseases would rather know that they have a serious disease than continue with no diagnosis (Gahl & Tifft, 2011, pp. 1904–1905).'

In the realm of disease prognosis, while it is intuitive for people to prefer optimistic prognoses over non-optimistic but structured ones, research supports the notion that people prefer structured prognoses over optimistic ones (see Landau et al. (2015) for discussion). For example, one study (Rutjens et al., 2013) examined the appeal of stage theories in conditions of reduced control. In an experimental design, participants were given two theories describing the progression of

Alzheimer's Disease: a stage theory detailing consistent deterioration of health, and a continuum theory emphasizing variation in progression, thus implying hope of maintained health for an unknown amount of time. When compared to those primed with negativity, participants primed with randomness preferred the stage theory over the continuum theory, even when the stage theory depicted a more pessimistic prognosis. When people feel threatened by randomness, as may be the case during situations of diagnostic uncertainty, the order-restoring capacity of stage theories is attractive (Rutjens et al., 2013). Other research confirms that negative feedback on health screenings is not devalued when it is expected (Renner, 2004); that is, health feedback, even if it is negative, can help people when it provides a diagnosis that reduces uncertainty. This finding is consistent with compensatory control theory, as control diminution resulted in a desire to establish epistemic structure, leading to a preference for the predictable outcome, even when the alternative held more hope. These results suggest that, in some cases, people would rather face predictable declines in health than an uncertain timeline. This interpretation is viewed from the CCT perspective as independent of patients' personal agency or their estimated probability of obtaining certain outcomes, instead reinforcing a structure that compensates for personal lack of control (Landau et al., 2015), and can restore a perception that the world is orderly and structured.

CCT posits that the use of one compensatory control behavior does not limit the effectiveness of another; they are ultimately substitutable (Kay et al., 2009). Therefore, when faced with threat to personal control (as occurs with health threats), people may employ multiple compensatory control behaviors in unison to elicit the greatest perception of control possible. As a result, in addition to adopting a preference for nonspecific epistemic structure, many patients will also affiliate with external systems that they perceive as acting in their interest. External systems create the illusion that there is less disorder in the world, attracting the support of those whose control perception is threatened (Kay et al., 2008). Joining in the power of a greater system allows for the assertion of vicarious control, diminishing personal feelings of lost authority (Rothbaum et al., 1982). Among patients, the most straightforward example of this is placing trust in medical professionals and facilities. Accordingly, diagnostic uncertainty may lead to greater utilization of healthcare services as people seek the certainty and reassurance that comes with diagnosis (Pincus et al., 2018; see also Holt et al., 2015). By aligning themselves with physicians and hospitals acting on their behalf, patients may establish vicarious control via these agents, potentially reducing the helplessness that their illness imposes on them. However, in the case of diagnostic uncertainty, placing trust in the medical system can further feelings of frustration if it too is unable to provide answers (Lillrank, 2003). Consequently, the experience of delayed diagnosis can result in a marked loss of confidence in the healthcare system (Neville et al., 2019; Rode, 2005).

Religion also offers the comfort of a governing external system; as such, patients will often bolster their faith in times of control diminution. In their research, Kay and colleagues (2008) found that after recalling an event in which they had insufficient control, participants reported an increased belief in a controlling God. Religion allows for a sense of vicarious control by placing the ungovernable situation in the hands of an omnipotent higher power. Hence, this behavior is observed in the first-person accounts of many patients managing severe medical conditions with uncertain prognoses such as spinal cord injuries (Bulman & Wortman, 1977) and permanent disabilities (Dalal & Pande, 1988). By affiliating with governing external systems such as one's medical team or religion, and believing that they are acting in one's own favor, patients are able to reestablish a greater sense of control.

Although patients are the sole physical experiencer of the illness, diagnostic uncertainty threatens feelings of control in other key populations. Loved ones, specifically parents of pediatric patients, take on much of the burden that diagnostic uncertainty imposes (Graungaard & Skov, 2007). Additionally, medical professionals are heavily impacted, as they share responsibility for the diagnostic decision-making, which is increasingly challenging under conditions of uncertainty (Hatch, 2016). These populations not only offer unique perspectives of the control diminution caused by diagnostic

uncertainty, but they also exhibit analogous compensatory approaches in pursuit of the same goal: regaining a sense that the world is orderly and controlled.

### ***The perspective of parents<sup>1</sup> of pediatric patients***

A child that becomes ill is dependent on parents or caregivers to serve as advocates to help the child navigate through the healthcare system (Eminson, 2007). For parents of children born with severe disabilities, satisfaction with the diagnostic process was found to be strongly correlated with certainty of the diagnosis and length of the process; increased certainty of diagnosis was associated with greater satisfaction (Graungaard & Skov, 2007). A qualitative study of parents of adolescents presenting with chronic pain found that in response to uncertainty relating to their child's condition or prognosis, parents experienced a sense of helplessness and distress (Jordan et al., 2007). Parents worry that aspects of their child's condition are being overlooked, and fear for their child's future (Spillmann et al., 2017). Adolescents experiencing medically unexplained symptoms and their parents reported difficulty accepting a lack of diagnosis, as they believed that naming the disease would correspond with strategies and solutions to address the condition (Moulin et al., 2015). Moreover, qualitative studies of parents with children experiencing diagnostic uncertainty have revealed a desperation for medical explanation so intense that parents report that they would prefer the discovery of a serious pathology (rather than continued uncertainty), as it would validate their child's pain, provide care options, and help friends and family understand the condition (Jordan et al., 2007; Moulin et al., 2015). A systematic review and meta-analysis of illness uncertainty with pediatric patients and caregivers found positive coping strategies to be associated with reduced uncertainty, suggesting a relationship between how people cope and their experience of uncertainty (Szulcowski et al., 2017). Yet, when diagnostic uncertainty cannot be abated, parents may be left with no tangible action plan or probable outcome, culminating in diminished perceptions of control. Due to parents of pediatric patients' unique role as executors of healthcare decisions for their children, as well as the extensive research on how parents manage diagnostic uncertainty (e.g., Szulcowski et al., 2017), we focus on parents as caregivers, recognizing that adult caregivers may experience similar psychological responses.

Compensatory control theory suggests multiple compensatory strategies that parents may adopt in order to combat this lack of control. One method is to bolster personal agency, which, for parents, can be observed in their relentless search for a diagnosis for their child. This behavior is common among parents with children experiencing diagnostic uncertainty, as they devote themselves and their time vying for resources for their child in an attempt to assert control over the situation (Jordan et al., 2007). For some parents, this may entail scouring books or the Internet for diagnostic research to bring to the attention of physicians, advocating for further medical testing, or participating in medical consultations (Moulin et al., 2015). Expanding efforts to identify the cause of the disease allows parents to perceive an increased likelihood of their child being successfully diagnosed or treated, in turn moving their sense of control closer to baseline (Landau et al., 2015).

Like patients, parents of pediatric patients may also compensate for lack of control by affiliating with external systems seemingly acting on their behalf. They embrace the same external systems as patients, as they experience similar control diminution. Accordingly, many parents turn to religion and the medical system for comfort. Kay et al. (2008) found that people experiencing insufficient control align themselves with the belief that God is acting on their behalf. This conviction may allow parents to experience a sense of vicarious control, placing the ungovernable situation in the hands of perceived higher powers (Rothbaum et al., 1982), and reasserting the belief that control exists – even if it is not exhibited by personal control. Similarly, parents may attempt to embrace the structure of the medical system to reassert a perception of an orderly world. In their review of pediatric diagnostic uncertainty, Pincus et al. (2018) postulate that the interactions between pediatric patients, their parents, and physicians are key events in adjustment; diagnostic uncertainty hinders clear explanations, causing communication to become ambiguous and

unsettling. By facilitating a communicative relationship with their child's medical team, parents may build a trusting relationship in which they can rely. When interviewed, parents of acutely ill, hospitalized children expressed that communication with their child's medical team was vital to having a positive hospital experience; parents who were unable to form a satisfactory relationship with the hospital staff reported feeling powerless or out of control (Roden, 2005). In another study of parents engaged in a difficult diagnostic process, the prevailing expectations for communication with the physicians were identified as empathy and active inclusion in the process; if these expectations were not met, it added to the uncertainty and lack of control of the experience of diagnostic uncertainty – but if upheld, parents reported being satisfied, even if the health prognosis was negative (Graungaard & Skov, 2007). Building relationships with external systems effectively eases parents' anxiety by allowing them to develop a sense of security and a perception of predictability in an uncontrollable environment. By entrusting that medical professionals and religious powers will intervene in their child's care, parents may be better able to manage psychologically with the threat their child's diagnostic uncertainty poses. Yet, as we review in the next section, doctors' experience with diagnostic uncertainty may likewise be characterized by a lack of control.

### **The perspective of physicians**

Lack of diagnosis is not only a liability for families, but for medical professionals as well (Tift & Adams, 2014). In his book *Better: A Surgeon's Notes on Performance*, the surgeon Atul Gawande (2007) reminds readers of the humanity of medical professionals, who may, at times, seem superhuman. Doctors go into medical practice believing the field is characterized by cunning diagnosis and technical ability; however, much of medical knowledge is incomplete, and there are no fixed steps to approaching illness (Gawande, 2007). For most physicians, rare and undiagnosed diseases fall outside the realm of familiar medical practice, leaving them in unprecedented territory. Similarly, patients with undifferentiated symptoms that could relate to multiple and common conditions present a challenge for physicians, and indeed, these undifferentiated symptoms are one of the largest sources of diagnostic errors (Singh et al., 2013). Caring for patients without clear diagnoses presents many challenges to healthcare professionals, as they often need additional time to decipher signs and symptoms, decide if further testing is necessary, confer with providers in other specialties, and follow up on inconclusive test results (Bhise et al., 2018; Spillmann et al., 2017). When they are not able to successfully address their patients' diagnostic uncertainty, physician's own sense of control can be threatened as well, and with this, their perception that their world is structured and orderly.

Several responses that medical professionals may engage in to address this control deficit are consistent with compensatory control theory. For example, medical professionals can affirm clear contingencies between actions and outcomes within the diagnostic process. Doctors believe, based on their training and experience, that there is typically a reliable link between symptoms and test results; thus, after exhausting diagnostic tests and repeatedly receiving inconclusive results, doctors may conclude that patients are somatizing or overexaggerating their symptoms (Tift & Adams, 2014). Although this conclusion provides an explanation as to why the doctor could not assign diagnosis and may restore his or her sense of control, the assumption that patients are reporting their symptoms out of proportion could lead to feelings of abandonment among patients (Gahl & Tift, 2011). In a qualitative study of adolescents facing diagnostic uncertainty, the feeling that their physician did not believe them was most acute when it was suggested that their symptoms might have a psychological origin (Moulin et al., 2015). Feeling as though their doctor has failed to diagnose them, some patients reported feeling let down by the healthcare system, or even reported a belief that the physicians were incompetent (Moulin et al., 2015). This, in turn, may adversely affect the trust that patients bestow in their physicians in an attempt to regain a sense of control themselves, highlighting the interconnection between patients' and physicians' compensatory control processes.

The NIH reported that patients in their Undiagnosed Diseases Program often do not lack diagnoses, but rather have too many of them (Gahl & Tifft, 2011). That is, in some cases in which physicians are unable to provide an accurate diagnosis to explain a patient's symptoms, they may assign partial or multiple diagnoses (Tifft & Adams, 2014). Diagnostic uncertainty has been linked in previous studies with increased hospitalizations and referrals, heightened healthcare costs, over-testing, and diagnostic variation (physicians assigning multiple diagnoses to the same patient; see Bhise et al., 2018 for discussion). Medical professionals' tendencies to assign multiple diagnoses or diagnoses that ultimately prove incorrect may serve the psychological function of bolstering personal agency, as they are utilizing the knowledge and resources that they do have to provide diagnoses that account for observed symptoms, even if the diagnosis does not prove to be accurate. Even if the physician is uncertain, naming the disease satisfies the patient and provides them the structure associated with diagnosis and potential treatment options, as we have noted earlier (Gahl & Tifft, 2011). Thus, this behavior feels consistent with their Hippocratic Oath, and doctors may feel justified in these well-intended actions, which may also have the effect of restoring their own sense of control and competency. Yet, the increased perception of control this behavior instills may ultimately be misleading to patients (and fellow healthcare professionals) as it could draw out the length of the diagnostic process, exacerbating diagnostic uncertainty among those involved.

Doctors' tendencies to assign multiple or partial diagnoses can manifest in further problems, as it often leads to the misdiagnosis and overdiagnosis of the typical 'unexplained' medical conditions such as chronic fatigue, fibromyalgia, and irritable bowel syndrome (Mueller et al., 2016). Specifically, there has been a general trend of overdiagnosis of fibromyalgia (FM) among the undiagnosed population, of which the chief symptoms include musculoskeletal complaints, widespread pain, and other non-specific symptoms (Fitzcharles & Boulos, 2003). Recent studies suggest that FM is being overly diagnosed, while other conditions are being overlooked by physicians. In one such study of 76 patients with referrals expressing FM, 34% had their diagnoses upheld after in-depth physical examinations and evaluation of symptoms (Fitzcharles & Boulos, 2003). Inaccurate diagnoses may be caused by many factors; nevertheless, it makes clear the difficulty of the diagnostic process. The stigmatization of diagnostic uncertainty stemming from the lack of medical understanding surrounding rare conditions may further the lack of control these conditions already cause patients and physicians alike, leading to additional distress and feelings that the world is random and unpredictable. Resulting attempts by physicians to abate this state may only prolong the path to diagnosis when the research and resources available are insufficient. Subsequently, the patient-physician relationship may suffer (Mueller et al., 2016).

It should certainly be noted that doctors are working within a medical system that at times constrains their options. Medical protocols are designed to reduce unnecessary surgery and tests, which in the aggregate is a problem (Kaplan, 2019). However, in some instances, these protocols may prevent doctors from prescribing, and patients from obtaining, needed treatment. Genome sequencing and other advanced technologies that assist in the diagnosis and discovery of rare diseases are currently not readily available to most clinical providers (Cope et al., 2020). Although the medical system as a whole may provide an external structure that doctors can rely on to reduce uncertainty via adherence to protocols (affirming external structure), it may also cause distress when it limits a physician's ability to further the search for a diagnosis for a sick patient.

Mysterious, undiagnosed illnesses fall outside the boundaries of commonly practiced medicine. Being a successful physician, Gawande (2007) argues, requires a willingness to recognize shortcomings, and an ability to change. Doctors confronting diagnostic uncertainty in patients must acknowledge the inevitability of failure and frustration, though this recognition stands in stark contrast to the diagnosis, treatment, and understanding of disease for which they have been trained, and to which they typically experience (Gahl & Tifft, 2011). The uncommon nature of rare diseases has made clear the difficulties doctors and patients encounter within the diagnostic process. Patients and their families are often simply grateful for any personalized care oriented toward a diagnosis, even if the efforts fail (Gahl & Tifft, 2011). To best serve their patients, doctors treating patients managing diagnostic uncertainty may benefit from acknowledging the associated uncertainty and lack of

control that they themselves are experiencing. Accordingly, some researchers have suggested that uncertainty management is an essential skill that should be a part of educational curriculum for general practitioners (O’Riordan et al., 2011). Recognizing the importance of managing uncertainty may allow physicians to change certain compensatory control behaviors that could hinder the diagnostic process, and allow them to acknowledge with greater transparency the desire to assert control over an inherently uncontrollable situation that they, as well as their patients, must at times confront. Table 2 provides examples of the compensatory control behaviors that may be exhibited by patients, parents, and physicians.

### Issues for future research

In response to the national health problem of undiagnosed disease, the NIH established the Undiagnosed Disease Program (UDP) in 2008 with the intent of furthering the understanding of novel disease expression and providing diagnoses for individuals who have spent years facing diagnostic uncertainty. This mission was substantiated on a global scale following two international conferences, where representatives from 18 countries established the Undiagnosed Disease Program International (UDPI) in 2014 (Taruscio et al., 2015). Organizations such as the UDP and the Undiagnosed Disease Network (2021) are leading the push for advances in etiology research, addressing the need

**Table 2.** Examples of Compensatory Control Behaviors Exhibited in Patients, Parents of Pediatric Patients, and Physicians in Response to Diagnostic Uncertainty.

Behavior	Application
<b>Bolstering personal agency</b>	<p><b>Parents</b></p> <ul style="list-style-type: none"> <li>Conducting their own diagnostic research, advocating for further medical testing, partaking in their child’s medical consultations</li> </ul> <p><b>Physicians</b></p> <ul style="list-style-type: none"> <li>Using the knowledge and resources available to provide one or multiple diagnos(e)s that explain the observed symptoms, even if they are uncertain</li> </ul>
	<p><b>Patients</b></p> <ul style="list-style-type: none"> <li>Seeking reassurance and authority of healthcare provider and medical system</li> <li>Strengthening beliefs in controlling religious figures</li> </ul> <p><b>Parents</b></p> <ul style="list-style-type: none"> <li>Placing authority and trust in medical facilities and physicians as an external system</li> </ul>
<b>Affirming clear connections between behaviors and outcomes</b>	<p><b>Physicians</b></p> <ul style="list-style-type: none"> <li>Concluding that patients are somatizing symptoms after receiving inconclusive test results</li> </ul>
<b>Affirming nonspecific epistemic structure</b>	<p><b>Patients</b></p> <ul style="list-style-type: none"> <li>Preferring structured prognoses over optimistic but unpredictable ones</li> <li>Adopting structures that are otherwise discordant with their interests, such as seeing themselves as fated to suffer from disease</li> </ul>

for improved diagnosis and care of individuals facing rare disease (Taruscio et al., 2015). Though research into the pathophysiology of new and rare diseases is vital to reforming these diagnostic measures, acknowledging the role that compensatory control behaviors play in the process may also improve the experience for patients, parents of pediatric patients, and doctors alike. However, individuals' experiences with diagnostic uncertainty may be moderated by other factors such as gender and socioeconomic status. Research is warranted into how these moderating factors may affect individuals' control perceptions in response to diagnostic uncertainty, potentially influencing the effectiveness of compensatory control strategies.

## Moderators

### Gender

Gender may moderate both physician's and patient's response to diagnostic uncertainty – and thus, the extent to which they engage in the compensatory control strategies outlined in this paper. Patients may respond differently to diagnostic uncertainty based upon the gender of their physician (Blanch et al., 2009; Cousin et al., 2013). In a simulated setting, physician-expressed uncertainty resulted in decreased patient satisfaction only for female physicians; furthermore, physician-expressed uncertainty in the field setting led to decreased patient satisfaction only when the physician was female and the patient was male (Cousin et al., 2013). Cousin and colleagues posit these results may be the consequence of negative stereotypes held against female physicians, particularly by male patients, and that signs of uncertainty may confirm their negative views, resulting in dissatisfaction. Beyond dissatisfaction, one implication is that for patients who have these negative stereotypes, they may engage to a greater extent in compensatory strategies with female physicians – seeking alternative means to reassure themselves that the world is predictable and orderly (see also Ma et al., 2019).

However, other research has not clearly confirmed this gendered pattern – and so it is an open question for future research. In a study assessing analogue patients' reactions to medical students' expressions of uncertainty, patients reacted negatively to both genders, but were somewhat less satisfied with male medical students than with female medical students when they expressed uncertainty (Blanch et al., 2009). Differences in these studies' results may be due to systematic differences in samples; the participants in the Blanch et al. (2009) study were typically younger and more educated than that of the Cousin et al. (2013) own study. This could have implications for the studies' results, as high expectations could lead participants to have negative reactions to both male and female medical students' uncertainty. Furthermore, a younger and better educated sample may not have the same prevalence of gender stereotypes (Cousin et al., 2013).

Gender may also affect the way that physicians handle diagnostic uncertainty. A survey of general practitioners (GPs) suggests that anxiety stemming from diagnostic uncertainty may produce gender specific reactions; female GPs' concern about bad outcomes resulted in increased test ordering and the use of more 'primary care heuristics,' while male GPs with heightened anxiety increased test ordering but used fewer 'primary care heuristics' (Schneider et al., 2010). However, other studies have found null effects among gender differences in doctor's and children's experience of diagnostic uncertainty (Lindley et al., 2014; Tanna et al., 2020). Thus, there are many open questions in the field of physician-expressed uncertainty (see Cousin et al., 2013 for discussion); there are also methodological limitations stemming from the vignette methodology as tools for measuring physician uncertainty have not been well validated. As such, different operationalizations of uncertainty may yield divergent findings, with corresponding implications for compensatory control processes.

### Socioeconomic status

Social class is closely related to perceived sense of control, with lower SES individuals in the United States typically possessing a lower sense of personal control when compared to individuals of middle

and high SES (Eom et al., 2018; Kraus et al., 2009). This accentuates the importance of SES in control processes, leading to the question of how SES may affect the use of compensatory control behaviors in situations that threaten control.

The lack of control experienced by patients with diagnostic uncertainty may be exacerbated among lower SES individuals due to the consequences that a lack of resources may have on health, more generally, which may prompt them to engage in compensatory strategies to a greater extent. In many low SES communities, healthcare is not as widely accessible due to scarcity of resources (Tiftt & Adams, 2014). When individuals in these communities become ill, it adds further socioeconomic burden to an already costly situation. A systematic review of cost-of-illness studies found most rare diseases are associated, both directly and indirectly, with significant financial burden (Angelis et al., 2015). This economic burden not only adds to the stress of diagnostic uncertainty, further exacerbating symptoms and hindering coping, but may also play a role in the vast number of misdiagnoses. In some cases, no diagnosis equates to no services for patients; consequently, doctors may assign partial or incorrect diagnoses in order to give patients access to necessary medical care (Tiftt & Adams, 2014). This pressure physicians feel to give a diagnosis may contribute to misdiagnoses, which can prolong patients' experiences of uncertainty within the diagnostic process, further diminishing their senses of personal control, triggering compensatory control processes.

### **Limitations, implications, and conclusions**

The examination of compensatory control behaviors as mechanisms to manage the control threat imposed by diagnostic uncertainty has revealed how a wide range of people adapt. These behaviors, when utilized by patients and parents, can be helpful in reducing perceptions of control diminution and restoring perceptions of order and predictability to the world. Additionally, CCT may provide a lens through which medical professionals may better comprehend the experiences of their patients, as well as their own behaviors, potentially reducing stigmatization of patients lacking clear diagnoses. Nevertheless, the analysis presented in this paper has an important limitation that should be noted, for it also serves as an opportunity: The integration of compensatory control theory and diagnostic uncertainty is conceptual, and not empirical. Although the framework of CCT is derived from extensive experimental research, there has yet to be empirical research that has applied CCT in contexts with patients, parents, or physicians who are enduring a difficult diagnostic process. It is our goal in this paper to outline the connections between the literatures as they illustrate the potential utility of the CCT approach for those studying diagnostic uncertainty in these populations, spurring future research and application. One avenue for this research is to further integrate CCT with the different stages of diagnostic uncertainty outlined in [Figure 1](#) by testing whether certain compensatory strategies (i.e., bolstering personal agency, affiliating with external systems perceived to be acting in one's interest, affirming clear connections between behaviors and outcomes, and affirming nonspecific epistemic structure) are more likely to be used at particular stages (i.e., patient experiences health problem and engages with healthcare system; information gathering; information integration and interpretation; working diagnosis; communication of diagnosis). Mapping the CCT strategies onto the stages (as well as the different roles of patient, parent, and physician) through empirical investigation would be of both theoretical and applied importance.

The present analysis has implications for reducing stigma among those who do not have a diagnosis for their illness. In their systematic review, Bhise et al. (2018) emphasize the positive impact that clinicians embracing diagnostic uncertainty as a norm could have. Acknowledging diagnostic uncertainty and effectively implementing measurement and management guidelines could help medical professionals deviate from standard protocol and potentially more effectively allocate the employment of clinical resources in cases of uncertainty. This acknowledgment may potentially reduce the compensatory control behaviors outlined in this paper that may exacerbate stigma among patients and parents of pediatric patients. At the same time, institutional efforts to improve diagnostic

capacity will reduce diagnostic uncertainty. The expansion and establishment of programs such as the NIH's Undiagnosed Disease Program are vital to this cause, with their comprehensive, multidisciplinary tactics serving as a model. The goal of this program is to research and discover new and rare diseases; through their efforts they have been able to successfully provide diagnoses and treatment for individuals whom medicine had previously failed to diagnose (Gahl & Tifft, 2011). The continued expansion and popularization of such programs directed towards rare and undiagnosed diseases can greatly improve diagnoses and thus reduce the number of people dealing with diagnostic uncertainty.

Ultimately, simply naming a rare illness is not the goal of patients, parents, and practitioners. Rather, most patients strive to manage their symptoms and maintain the best quality of life possible (Bhise et al., 2018; Spillmann et al., 2017). Receiving a diagnosis can be conducive to this, allowing for a straightforward prognosis and treatment plan. However, there are gaps in medical knowledge, in which lie diseases that cannot currently be diagnosed. These patients face health threats, extensive waiting, and diminished psychological control. Much importance lies in learning about the behaviors and processes that contribute to these patients' experiences; and thus, greater understanding is needed of the behaviors people engage in to compensate for the lack of control associated with diagnostic uncertainty.

The prevalence, and resulting consequences, of diagnostic uncertainty in medical practice suggests the utility of integrating perspectives from different fields of psychology to help understand this issue. Diagnostic uncertainty not only threatens one's health, perceptions of control, and belief that the world is an orderly and predictable place, but it also threatens one's faith in the medical system when it cannot provide needed answers. Overall, the healthcare system is not currently equipped to provide answers to many patients confronting diagnostic uncertainty, resulting in frustration among all involved. Neither medical professionals nor patients and their families are satisfied by the diagnostic measures in place (Gahl & Tifft, 2011). The present paper offers compensatory control theory as a social psychological framework for understanding the responses individuals within the system make in attempt to psychologically manage the experience when a diagnosis cannot be made. Working to improve the system for those who depend on it when the origins of their maladies are undiagnosed mysteries may enable patients and their families to obtain a greater sense of psychological control when it is most needed.

## Note

1. We use the term parents to refer to both parents and other primary caregivers of ill children. We also note that this analysis likely applies to caregivers of geriatric patients who have diminished capacity to communicate with their clinicians. Although the focus of the present review is on caregivers of pediatric patients, similarities and differences between them and caregivers of other patients with difficulty communicating is an important topic for further research.

## Disclosure statement

No potential conflict of interest was reported by the author(s).

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