

ORIGINAL RESEARCH

Further development of the intolerance of uncertainty model of GAD: a case series

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Abstract

Intolerance of uncertainty (IoU) is important in the development and maintenance of worry and generalized anxiety disorder (GAD; Dugas *et al.*, 1997). However, it remains unclear why some people respond so negatively to uncertainty and have poor clinical outcomes. We adapted the IoU model to include the influence of developmental and/or attachment factors, and their possible importance to intolerability of uncertainty and associated hypothetical worries. Seven consecutive GAD referrals for CBT were naturalistically treated with the novel approach. All participants completed the 7-item Generalized Anxiety Disorder Scale (GAD-7; Spitzer *et al.*, 2006), the Penn State Worry Questionnaire (PSWQ; Meyer *et al.*, 1990), as well as a novel 10-item Premonition Bias Questionnaire (PBQ; C. Chigwedere *et al.*, unpublished). From pre- to post-treatment, results for both GAD ($p=.001$) and worry ($p=.005$) improved significantly. Clinically significant change or a post-treatment score within the normal population range were observed for both the GAD-7 and PSWQ. The change in believability of worry, measured on the PBQ was also significant from pre- to post-treatment ($p=.008$). Overall, the novel approach may be an alternative approach to treating GAD, with some potential, both as an adjunctive or standalone treatment. However, this is a small case series and the presented novel approach requires empirical support and evaluation in larger experimental studies.

Keywords: case series; CBT; GAD; intolerance of uncertainty

Introduction

Generalized anxiety disorder (GAD) is financially, socially and personally burdensome and highly prevalent (McManus *et al.*, 2009; Ruscio *et al.*, 2017). It is associated with distressing worry about almost anything (Dugas and Robichaud, 2007). Cognitive behavioural therapy (CBT) offers an effective treatment of GAD, with a range of models including worry avoidance (Borkovec, 1994; Borkovec *et al.*, 2004), metacognition (Wells, 1995; Wells, 2010), emotion regulation (Mennin *et al.*, 2002) and Intolerance of Uncertainty (IoU) (Buhr and Dugas, 2002; Dugas *et al.*, 1997; Dugas *et al.*, 1998; Dugas and Robichaud, 2007). However, many people do not achieve expected clinical improvements (Borkovec and Costello, 1993; Borkovec *et al.*, 2002). For example, the proportions of those achieving the standardized recovery criteria of a Penn State Worry Questionnaire (PSWQ; Meyer *et al.*, 1990) score of 47 or less may only be 46% at end of treatment and 57% after a year (Hanrahan *et al.*, 2013). These proportions from Hanrahan *et al.* represent outcomes of gold standard randomized trials, with all the associated supervision and exclusion criteria, not routine clinical outcomes, which would most likely be

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less positive. The IoU model is an integrative approach that draws on theories and interventions from a range of other models, to present an elegant evidence-based approach (Dugas and Robichaud, 2007). Its central hypothesis is that worry may be associated with intolerance of uncertainty, ‘a trait of the individual, characterized by a predisposition to react negatively to an uncertain event or situation, independent of its probability of occurrence and its associated consequences’ (Ladouceur *et al.*, 2000; p. 934).

The ubiquity of uncertainty makes worry diffuse, in both its triggers and content. Dugas and Robichaud (2007) describe a multi-faceted, uncertainty-focused approach to the understanding and treatment of GAD, detailing the extensive research evidence. The four main processes include (1) intolerance of uncertainty, (2) positive beliefs about worry, (3) negative problem orientation and (4) cognitive avoidance. Treatment consists of six modules of (1) psychoeducation, (2) uncertainty recognition and behavioural exposure to real-life situations, (3) re-evaluation of beliefs about worry as useful, (4) problem solving, (5) imaginal exposure to hypothetical worries and (6) relapse prevention.

Based on clinical observation, available theory and discussions with colleagues and patients, we have adapted the IoU model to include a greater role of developmental factors, which have been shown to be associated with GAD (Cassidy, 1995; Cassidy *et al.*, 2009). For example, attachment style developed in childhood relationships with primary caregivers, and described as insecure (avoiding closeness), anxious-ambivalent (clinging and showing distress when separating from carers) or disorganized (inconsistent displays) (Bowlby, 1973), differentiated GAD sufferers from those with panic disorder and normal controls. Specifically, while insecure attachment styles were associated with both disorders (i.e. panic and GAD) compared with controls (Cassidy *et al.*, 2009; Muris *et al.*, 2000), only ambivalent attachment predicted GAD, while both insecure and anxious-ambivalent styles predicted panic disorder (Muris *et al.*, 2001). As such, targeting developmental factors may improve CBT treatment outcomes for some patients. For example, in an additive study (Newman *et al.*, 2015), compared with CBT plus supportive listening (CBT+SL), adding interpersonal and emotion processing therapy (CBT+I/EP) interventions to CBT predicted greater symptom change in GAD participants with avoidant attachment styles. However, greater change at post-treatment was observed for those with an anxious or angry attachment style receiving CBT+SL than CBT+I/EP. Although not specifying attachment styles, we adapted the major focus of intervention from targeting hypothetical worry through imaginal exposure to specific current triggers of uncertainty. We hypothesize that such triggers are associated with developmental experiences through stimulus–stimulus and stimulus–response associative learning. Worry behaviours reduce affect associated with uncertainty, effectively reinforcing worry and maintaining IoU as described by others (Dugas and Robichaud, 2007).

We propose that the worry in GAD may not be about anything (Dugas and Robichaud, 2007) but rather, specific to schema domains (Young *et al.*, 2003). Furthermore, it may be triggered by elements in the environment, which may be reminders of negative developmental experiences (i.e. conditioned stimuli). Such conditioned stimuli become associated with the schema (e.g. failure, abandonment), with a conditioned response of fear. Such conditioned stimuli will be situation-specific triggers of memory traces from learning contexts (Baddeley, 1996), ranging from observed environmental elements to internal sensory and cognitive states, including uncertainty, which activate schemas. Worry may be an avoidance response as described by others (Borkovec *et al.*, 2004; Dugas and Robichaud, 2007), but perhaps, it is the schema activated by situational triggers that is avoided.

To give an illustrative example of the above worry hypothesis, a young man in CBT treatment described a history of public humiliation, including physical bullying and negative comments about his appearance in school. He interpreted such bullying as evidence that he was not good enough and was a failure as a person. Accordingly, he endorsed defectiveness and failure on a self-report measure of schemas. During the contextualization exercise (described below),

such bullying was conceptualized as the unconditioned stimulus (UCS), paired with fear and distress as the unconditioned responses. Elements present in the context, such as being observed, physical sensations and uncertainty may have become the conditioned stimuli (CS), which became associated with the UCS, acquiring its qualities, so that they were able to trigger the fear independently of the UCS. He was anxious and hypervigilant, looking out for the presence of those triggers or anything resembling them. Although collaborative CBT work allowed him to become explicitly aware that such triggering elements were safe, they implicitly represented the possibility of intolerable failure or defectiveness, leading to fear, worry, and escape and avoidance measures. He responded as though his failure and defectiveness predictions were correct, so that he worried and tried to identify possible hypothetical future scenarios, their likely outcomes and solutions. Unfortunately, attending to such future scenarios prevented him from cognitively, affectively and behaviourally engaging with the trigger events. Such engagement would have increased the chances of not only tolerating the trigger events, but also identifying problem-solving solutions.

The above example highlights how worry may involve a perceptual priming for the schema and its triggers (Baddeley, 1996). It may involve the retrieval of over-general, implicit memories (Conway and Pleydell-Pearce, 2000; Williams *et al.*, 2000). Such memories may inform the meanings given to current events, their predicted outcomes and possible solutions. Such predictions may then be experienced as worry. The retrieved memories and associated meanings may give worries a quality of reality, which we have called a *premonition bias*. When asked about the quality of their worries, we have observed that GAD patients generally describe them as believable, as though they are realities that will come true and need to be prepared or planned for. Thus, premonition bias may represent this quality of reality or believability of worry hypotheses in GAD. This differs from concepts such as thought–action fusion, which relate to the magical power of one’s mental or physical actions to cause specified outcomes. We hypothesize that it is because of this premonition bias that actions to prevent predicted outcomes, including worrying, may be positively perceived.

We proposed an adaptation to the IoU model in a three-phase worry cycle of (1) a fear phase, involving *orientation* to the trigger, its *appraisal* and *fright* responses, (2) an anxiety phase, involving *flight-oriented* responses, *premonition bias*-informed appraisals of the flight efforts, and (3) a reinforcement phase, involving *worry behaviours* and *consequences* stages (Fig. 1; Chigwedere and Wilson, 2021). Five treatment modules: (1) socialization, (2) uncertainty tolerance training, (3) contextualization, (4) problem-solving and (5) mindfulness were also described (Chigwedere and Wilson, 2021). The derived treatment approach has also been incorporated into the CBT treatment in a non-inferiority trial (Timulak *et al.*, 2018), and the current naturalistic case series.

Method

Current study

The current study was conducted in the naturalistic context of routine practice in a CBT hospital-based in- and out-patient clinic. Seven patients were recruited for treatment by the authors (C.C. and J.M.). The motivation for the novel treatment approach was an effort to improve outcomes for GAD patients using theory, clinical observation and therapist self-practice (Chigwedere, 2019) evidence. As such, this was a clinically motivated attempt to evaluate our novel model (Chigwedere and Wilson, 2021) in a more structured approach than the piecemeal, adjunctive approach we had applied in usual routine practice. As such, the current study reports the first application of our novel model as a stand-alone intervention for GAD, not as a pseudo-additive or augmentation intervention of the IoU model of Dugas and Robichaud (2007).

The case series approach was chosen over other designs (e.g. an experimental case study approach) because of the novelty of the model and intervention. For example, the order of the modules is untested, making an exploratory case series an appropriate approach. The aim was

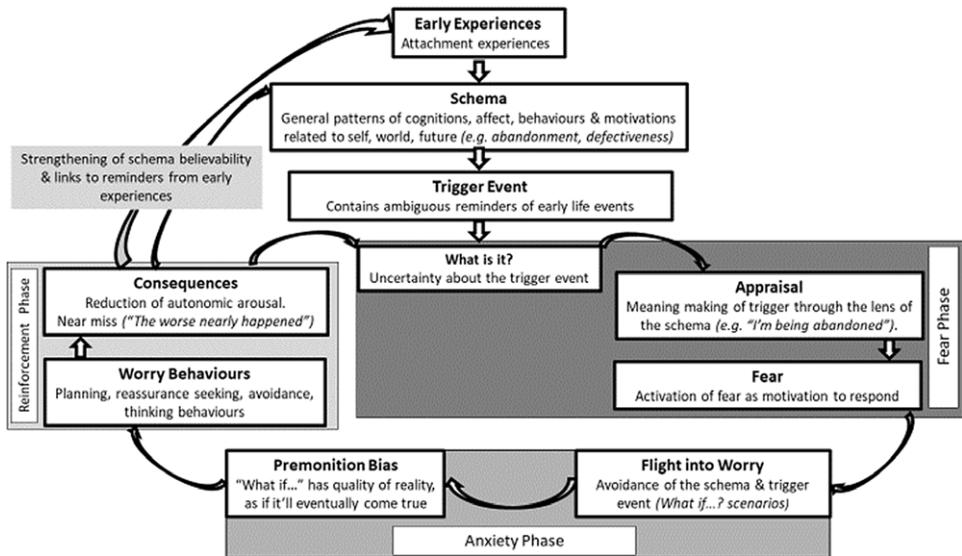


Figure 1. A three-phase model of worry maintenance (from Chigwedere and Wilson, 2021).

simply to evaluate pre- and post-treatment changes on standardized measures of GAD (i.e. measures of anxiety and worry), and a novel concept we have called premonition bias. Although still only a theoretical concept, we have devised a measure, the Premonition Bias Questionnaire (PBQ; C. Chigwedere, unpublished) (see Table 2), and this case series offered opportunity to further test its utility along with standard GAD measures.

The main aim of this case series was to investigate the acceptability and feasibility of the novel model. The in- and out-patient service from which all participants were recruited runs an anxiety disorders treatment programme, informed by the IoU model (Dugas and Robichaud, 2007). Treatment is usually in group or individual formats or a combination of both. The novel model applied in the current case series originally resulted from the clinical observations of the first author, including reflective self-application (Chigwedere, 2019), along with discussions with the second author and other colleagues and patients. This case series provides an opportunity to evaluate and report on the acceptability of the model and treatment as a standalone approach, not as an adjunctive approach as was the case in a previous non-inferiority trial (Timulak *et al.*, 2018).

Recruitment and participants

Seven consecutive referrals (age range 23–57 years) to CBT for GAD/worry were included. All participants had a consultant psychiatrist-confirmed DSM-5 (APA, 2013) diagnosis of GAD, which was arrived at through a semi-structured interview. GAD symptom criteria included excessive and uncontrollable worry about a range of events, accompanied by at least three cognitive or affective/physical symptoms (APA, 2013). The diagnosis was further confirmed at CBT assessment by an experienced CBT therapist (the authors). Criteria for the case series were a PSWQ (Meyer *et al.*, 1990) score of 47 or above and a General Anxiety Disorders Scale (GAD-7; Spitzer *et al.*, 2006) score greater than 10 at baseline. However, Participant 6 was included, despite scoring below 10 on the GAD-7. This participant had recently completed a group treatment programme, where he had scored above 10 on the GAD-7. He described being functionally affected by GAD symptoms. Due to the hypothesis of the developmental or attachment factors in the model, schemas were assessed at baseline only and

Table 1. Participant schemas and developmental factors

Participant	Age	Hypothesized schemas	Developmental factors
1	24	Failure/defectiveness-shame/self-subjugation	Physical and verbal bullying, parental neglect of physical needs
2	52	Defectiveness and shame/failure /enmeshment/ self-sacrificing	Paternal emotional abuse/maternal death
3	26	Abandonment/self-sacrificing	Death/loss
4	38	Enmeshment/ defectiveness and shame/failure/ abandonment	Parental disharmony/lack of validation/ parentification
5	38	Defectiveness and shame/self-subjugation/ unrelenting standards	Parental criticism/comparison with others/ high expectations
6	52	Defectiveness and shame/mistrust and abuse/ unrelenting standards	Parental anger and abuse/teacher and peer bullying/
7	42	Defectiveness/mistrust and abuse/unrelenting standards	Never failed as a child/high-achieving family

Table 2. Premonition Bias Questionnaire

To complete this questionnaire, think back over the last two weeks or so. It may be helpful to recall the last time you were particularly worried and anxious. Using the scale below, indicate the extent to which each statement is a true representation of you. Please enter the appropriate number (1–6) next to each statement as indicated in the example below:

Example: 1. 6 I have a tendency to worry about many different things.

1	2	3	4	5	6
Not at all true of me	Rarely true of me	Sometimes true of me	Often true of me	Very often true of me	Always true of me

1. _____ I have a tendency to worry about many different things and situations.
2. _____ When I start to worry, I find it difficult to stop without actively doing something to distract myself or take my mind off it.
3. _____ When I am worrying, my fears can feel like realities that will eventually come true.
4. _____ When I am worrying about something bad happening (e.g. to someone I care about), not doing something about it (e.g. thinking it through, distracting, etc.), says something negative about me as a person.
5. _____ When I am worrying, I feel as though not doing so, would leave me unprepared.
6. _____ When I am worrying, I feel as though by doing so, I am finding the best solution.
7. _____ Bad things do happen, so worrying about them means that I am taking them seriously.
8. _____ When I am worrying, I feel as though I am helping to prevent the bad thing from happening.
9. _____ When I am worrying, I usually avoid picturing mental images of the thing I fear, because doing so makes it feel more real, believable and likely to happen.
10. _____ When I am worrying, my discomfort seems to support the sense that my fears are real, and to justify the need to worry.

all participants scored high on at least two schemas on the Young Schema Questionnaire (YSQ; Young, 2005) (see Table 1).

Participants’ details have been changed to protect their identities. They were aware that the approach was novel and unevaluated, but had been trialled in routine practice. As such, if treatment was not successful, participants could be offered further treatment using the Dugas and Robichaud (2007) protocol with one of the authors or a different therapist if preferred.

Participant 1 (P1)

P1 was a 23-year-old female university student. She described a history of childhood neglect and bullying. She also described a range of worries with physical and cognitive symptoms and

co-morbid depression. She described a history of physical and verbal bullying in school, as well as childhood neglect (e.g. wearing soiled clothes to school), which exacerbated the bullying. She had a diagnosis of GAD and depression, describing difficulty tolerating uncertainty. She worried excessively about anything, including minor discomforts or illnesses, university work, failure and social situations, particularly possible embarrassment from predicted inappropriate social behaviours.

Participant 2 (P2)

P2 was a 52-year-old married businessman and father of three with GAD and a lifelong history of worry about anything and insomnia, worsening over 18 months in response to work stress. Although the stressful events had resolved, he continued to worry about his employees and financial health of his business. He also worried about his family and his own health, judgement by others, personal finances, failure, and the future in general (e.g. his family's well-being if he died). He described parental loss and anger in childhood, as well as unfavourable comparisons with siblings.

Participant 3 (P3)

P3 was a 26-year-old employed female, who described a history of GAD and worry from a young age. She had managed this using a range of measures, but could no longer control it, particularly the tiredness, sleeplessness, physical tension, headaches and gastric symptoms accompanying the worry. She described close family relationships with good attachments. However, she described childhood accidental death of a close relative, leading to fears of loss of loved ones that continued into adulthood.

Participant 4 (P4)

P4 was a 38-year-old self-employed father of two, with a worsening 4-month history of worry and depression, though he had 'always been a worrier'. He described little parental validation in childhood, with parental relationship disharmony as well as maternal mood instability and withholding of affection, resulting in apprehension, guilt and worry about failure. He worried about the future, finances, his marriage, and his family's health.

Participant 5 (P5)

P5 was a 35-year-old married corporate executive and mother of one, with a history of GAD. She described childhood parental criticism, low self-esteem and compensatory perfectionism. She worried about a range of events including her work, health, finances, friendships and social interactions, marriage and her family's health, and the impact of her mental health on them. She feared being perceived as a failure and not meeting her potential. She described physical tension, headaches and middle insomnia.

Participant 6 (P6)

P6 was a 45-year-old married and employed father of two grown-up children, with a history of low mood and perfectionism. He worried 'about almost anything' including his health, finances, relationships, work and future. He also described worrying about being reprimanded by his employer and customers, getting a criminal record and becoming unemployed. He also worried about a range of minor events that presented the possibility of being reprimanded.

Participant 7 (P7)

P7 was a 57-year-old corporate executive and father of two now adult children and one teenager, with a history of worry and sleep problems. He described a high-achieving middle class family background and a fear of poverty and failure. He worried about finances, his marriage, his safety and that of his home, with a fear of heart attack, stroke or insanity from the anxiety. This was exacerbated by his child's illness and some business difficulties, all of which were now resolved.

All participants described uncontrollable worry and unsuccessful efforts to stop it. This was accompanied by a range of affective responses including a general sense of apprehension, guilt and shame. They described a range of sensations from a general inability to relax, degrees of fluctuating anxiety from feeling 'panicky' to physical tension, headaches, sleep disturbance, nausea, chest tightness, 'butterflies' and shortness of breath. They described sweating, shakiness, increased heart-rate, gastric discomfort and loss of appetite. They engaged in a range of behaviours including procrastination, sleeping, avoiding socializing, seeking reassurance, overworking, avoiding risk, trying to do things perfectly and engaging in distraction, along with worrying and thinking about every possible negative outcome. The worrying was circular and often described as leaving participants feeling 'frozen', but was experienced as a positive thing to do, although causing distress. They all described their worries as having a quality of reality as though they would eventually come true, with one participant stating that, '*When I am worrying, the possible becomes the eventual.*'

Measures

Penn State Worry Questionnaire (PSWQ; Meyer et al., 1990)

The PSWQ is a 16-item measure of the generality, excessiveness and uncontrollability of pathological worry. It is widely used in both clinical and research contexts, and has high test-re-test reliability and internal consistency (Molina and Borkovec, 1994). The thresholds for the PSWQ are 16–39 (low); 40–59 (moderate); and 60–80 (high).

The 7-item Generalized Anxiety Disorder Scale (GAD-7; Spitzer et al., 2006)

The GAD-7 is a widely used measure of GAD, both in research and routine clinical practice, with good sensitivity and specificity (Spitzer *et al.*, 2006; Swinson, 2006). Thresholds are 5–9 (mild), 10–14 (moderate) and >15 (severe), with >10 denoting GAD requiring further evaluation.

The Premonition Bias Questionnaire (PBQ; C. Chigwedere, unpublished)

The PBQ (see Table 2) is a novel, 10-item measure of premonition bias (i.e. the believability or quality of reality of worries). Each item is rated on a 1 (not at all typical of me) to 6 (typical of me) Likert-style scale. Although still requiring a full test of its psychometric properties the PBQ was tested in a small group of GAD participants ($n=22$), it appeared to demonstrate high internal reliability (Cronbach's $\alpha=.96$) and it could discriminate 11 GAD cases (mean=43.09, $SD=9.73$) from 11 normal controls students (mean=19.00, $SD=4.67$; $F_{1,21}=54.803$, $p=.0001$) (Chigwedere, unpublished).

Young Schema Questionnaire (YSQ; Young, 2005)

The long form of the YSQ (YSQ-L) is a 293-item measure of emotional schemas (Young *et al.*, 2003). All items are rated on a 6-point Likert-style scale from 0 (not at all true of me) to 6 (totally true of me). It is used in both clinical and research settings. Although Young *et al.* (2003) describe 18 schemas in five domains, in five independent samples ($n=1564$), the YSQ was found to contain three factors with adequate test-re-test reliability, internal consistency, and good convergent and

discriminant validity with measures of depression, low self-esteem, personality disorder and psychological distress (Schmidt *et al.*, 1995).

Intervention

The treatment was an individualized, novel approach designed to be used flexibly in response to patient needs in a modular format. For this reason, it will be described as a two-phase, five-module treatment. The modules are made up of groups of interventions applied together for all individuals. Although described separately, the modules can run concurrently or overlap, depending on patient need. Each phase targets one hypothesized worry-maintenance factor (i.e. fear, anxiety and reinforcement). As reinforcement is a function of safety-strategies in response to worry, we have not allocated it a distinct treatment phase. As such, two treatment phases will be described.

Fear phase

This phase targets fear, the high arousal, phasic response to imminent threat (LeDoux and Pine, 2016). Neuroimaging studies have found that mindfulness not only reduced activation of neural structures associated with GAD but also led to positive structural changes (e.g. increased integrity of the uncinate fasciculus and reduced density of the amygdala) (Hözel *et al.*, 2013). We hypothesize that this phase targets similar structures, but also results in similar impacts to habituation in exposure therapy and inhibitory learning (Craske *et al.*, 2014).

Module 1: Socialization to the model and self-monitoring

This module targets the hypothesized fear phase of worry. We hypothesized that increasing tolerance of environmental triggers and associated uncertainty would reduce fear, as well as the use of cover and overt worry behaviours as coping strategies. Examples of triggers included P7 thinking about work, and P1 speaking to a peer about university reading material. As discussed below, such events may have triggered a sense of ‘not knowing’, leading to an orienting response, with allocation of attentional resources to appraise and make sense of the triggering event. This may result in conditioned responses of fear, accompanied by negative appraisals, both of which motivate worry as an unhelpful coping strategy. Worry may give a sense of preparation for predicted negative outcomes and reduces autonomic arousal, thereby reinforcing the use of the worry response in future. The non-occurrence of the feared outcomes may strengthen belief in the usefulness of worry, all of which prevent tolerance of uncertainty. The rationale is then, that it may be important to identify the worry trigger and its associated uncertainty, and then to problem-solve *them*, rather than the hypothetical prediction.

Following a thorough assessment, Module 1 specifically aims at socialization to the model and sharing of the treatment rationale. Worry is described as an understandable but unhelpful safety-behaviour. Thus, participants were taught to identify and discriminate worry triggers from the worry response of hypothesizing and predicting outcomes. End of treatment goals were set and self-monitoring started.

Participants engaged in self-monitoring, but unlike the approach proposed by Dugas and Robichaud (2007), self-monitoring was practised concurrently with uncertainty tolerance training (UTT; Module 2). Self-monitoring employed a novel *Worry/Uncertainty Diary* (see Table 3), which separates perceived triggers and their uncertainty from hypothetical worries. A difference between the current approach and that of Dugas and Robichaud (2007) was the identification of specific triggers to be tolerated, not current problems to be problem-solved. As with any other safety behaviour, hypothetical worrying was to be identified and resisted. Engagement with any future-focused worry was only practised to demonstrate the

Table 3. Uncertainty/worry monitoring diary

Date/time	Cue event/situation	Feelings	Current uncertainty	Hypothetical worry
	What was triggered your worry (e.g. where were you? What were you thinking, feeling, etc.?)	How did this affect your feelings physically, emotionally, etc.?	What did this make you uncertain of in that moment (e.g. 'I don't know...')?	What 'what if...' type thoughts did you notice? What did you fear might happen because of this event?

futility of worry, how it increases anxiety and the need to tolerate triggers of worry. Instead of exposure to feared consequences, participants practised UTT as Module 2.

Module 2: Uncertainty tolerance training

This module continued self-monitoring and worry recognition training but included UTT exercises. In UTT, patient and therapist collaboratively identified a worry event that had occurred. The therapist guided the participant to imagine the event, describing it in detail 'so that I can see it in my mind's eye and hear it in my mind's ear with you'. Affective and cognitive responses were collaboratively identified, before the participant was guided to mentally hold the image, resisting any meaning-making or worry. Although resisting engaging with cognitions (worry) was initially difficult, participants soon became more adept at it, tolerating the image and feeling 'calmer'.

Guided discovery in reflecting on the exercise identified the futility and anxiety-maintaining effect of worrying, as well as its avoidance of the situational trigger. The UTT was practised between sessions with different triggers as per learning theory principles (Craske *et al.*, 2014). The reflection component was important because it targeted negative appraisals of the trigger. For example, patients often describe the sense that worry is protective or that the worst will indeed come true unless they worry, as described by others, or without worrying, fear would continue in an unending and catastrophic spiral (Wells, 1995). Such positive and negative appraisals can be restructured in the post-UTT reflection, thereby increasing learning and reducing reinforcement.

In the follow-up session, between-session practice was reviewed and UTT was repeated. When participants demonstrated an ability to engage with a worry trigger, an uncertainty statement, which made explicit 'the unknown' (i.e. the uncertainty) associated with the trigger event was collaboratively identified. Uncertainty statements combine the factually known (i.e. the trigger) with what is unknown (i.e. the uncertainty). An example of an uncertainty statement from P1 was 'I know that my friend has read material that I have not, I don't know what this says about me.'

While attending to the uncertainty statement, participants were encouraged to engage with the image of the trigger, thereby increasing tolerance of, not just the trigger but also the associated uncertainty. We propose that explicitly stating the uncertainty is an important component of tolerating it. Compared with holding the trigger alone, participants often described an increase in arousal and distress once the uncertainty statement was incorporated, but still, reached a point of tolerance. Again, this was practised between sessions. As participants became more adept with UTT, they could apply it to 'live' events.

As participants increasingly tolerated triggers and uncertainty, problem-solving was introduced. This involved collaborative identification of options for responding to the unknown. For example, P6 worried about getting work done. At the point of tolerance of the uncertainty, the therapist asked 'We know there are X tasks to complete, we don't know how you are going to get them all done, so what options do we have right now as you look at that

list?). In doing this exercise, participants begin to practise targeting and problem-solving the real problem (i.e. the current uncertain event), helpfully solving *it*, not a hypothetical future outcome.

Anxiety phase

Anxiety is an apprehensive response to an uncertain, unpredictable and prolonged or distal threat, which may involve different but overlapping structures to those associated with fear (LeDoux and Pine, 2016). Anxiety-specific interventions may be required and this phase contextualizes anxiety and targets its appraisals.

Module 3: Contextualization

As we hypothesize that anxiety-related appraisals of worry and its triggers are informed by developmental and attachment experiences, Module 3 aims to identify possible developmental contexts of worry. Baddeley (1996) proposed that environmental triggers may activate memory traces of negative life events from the past. Individuals may have a lowered threshold for perceiving reminders of those past events and triggering of memory traces elements in the present environment (Brewin *et al.*, 1996). The activated memories and their meanings may be over-general, implicit and schematic (Williams *et al.*, 2000). They may then inform the meanings attributed to events in the present environment, leading to predictions of outcomes and planning of actions. Such predictions and plans may be experienced as worry, which is associated with uncertainty what will happen, how or when. Such uncertainty activates anxiety, especially if predicted outcomes hold negative implications for the individual, which is the case with emotional schemas (Young *et al.*, 2003). For example, P1 had a failure schema, which was triggered by hearing about a colleague's reading; an ambiguous environmental event, which possibly signalled the threat of failure. Attentional resources were allocated towards evaluating the trigger, leading to a 'flood of thoughts' and *appraisals* of imminent failure. This triggered a *fight-flight* fear response ('panicking'). As fear is motivational a *flight* into a 'quest for certainty', involving efforts 'to know what is to come' resulted in a *premonition bias*, as though '*I really will fail*'). Consequent *worry behaviours* included more thinking and reassurance seeking. This reduced autonomic arousal and *reinforced* the cycle.

As per Young *et al.* (2003), we hypothesized that P1's response was a learnt response with a developmental context. As verbal challenging methods risk increasing worry and rumination – the worrier's usual safety-strategy – imagery rescripting (Arntz and Weertman, 1999) was preferred. The starting point was usually a UTT exercise (see Module 2). The affect associated with images was identified before the therapist asked, '*What is your earliest memory/experience of feeling like this?*'. The participant was encouraged to return to an identified early event in imagination, describing the scene in detail, including sensory elements through all five senses (e.g. *Who is there? What do you see, ... feel ... smell ... hear, etc.*). The therapist confirmed participant's age in the image, then asked: '*You are ... years old and ... is happening. What message are you taking from this?*'.

The stated interpretation or 'message' from the event was considered to represent the schema or meaning internalized by the child (Young *et al.*, 2003). Schemas are hypothesized to arise from unmet childhood emotional needs (Young *et al.*, 2003), which the therapist identified by asking: '*You're XXX years old. XXX is going on and giving you the sense that ... [schema] and you feel ... What would you need, so as not to feel like that?*'.

After this, the patient was asked to imagine entering the scene as an adult and attempt to meet the child's unmet emotional need, verbally (e.g. validation), physically (e.g. a hug in imagination only) and behaviourally (e.g. imaginal removal to a safe place). In doing so, alternative/helpful schemas were identified. The therapist could request permission to also enter the scene and

more robustly meet the child's needs if needed. The recent event could be returned to in imagination and re-evaluated with the new learning from the rescripting exercise.

Collaborative reviewing identified the new learning about worry and its context, as well as the received messages and ways to test them and reinforce helpful ones. Such confirmation and disconfirmation provided the context for change-focused cognitive behavioural techniques in follow-up sessions. For example, P6 undertook a behavioural experiment and resisted doing a piece of work perfectly, despite worrying that he would be reprimanded. P4 resisted checking and practised tolerating the arousal to see if he could cope and if disaster occurred. Chair-work (Young *et al.*, 2003) could be introduced and was conceptualized as a method of encouraging *perspective taking*, as a bridge between the imagery rescripting and real-life practice. Between-session practice included UTT, behavioural experiments, exposure, assertiveness and other CBT techniques.

Module 4: Problem-solving

This was a more focused problem-solving intervention than the one used in Module 2. The rationale was that if participants could learn a helpful approach to problem-solving, they would be less likely to resort to worrying. Problem-solving approach has been ably described elsewhere (Dugas and Robichaud, 2007). As such, it will not be detailed here.

Module 5: Mindfulness

Due to its non-judgemental engagement with triggers, UTT bears some similarity to mindfulness. However, in UTT the anchor is a situational conditioned stimulus that may be a trigger of a schema, while in mindfulness the anchor is usually a neutral event (e.g. the breath). It may not be necessary to continue daily UTT, making mindfulness a helpful maintenance and relapse prevention intervention. As such, participants were encouraged to engage in daily mindfulness, while occasionally deliberately engaging with UTT.

Statistical analyses

Statistical analyses were conducted with SPSS version 24 (SPSS, Inc., Chicago, IL, USA). Although the sample size was small, Bland and Altman (2009) describe the robustness of the *t*-test for small sample sizes as well as the Wilcoxon signed rank test (*W*), noting that *W* and other non-parametric tests cannot produce a $p < 0.05$ with samples of $n < 6$. As such, the Shapiro–Wilk test (*W*) of normality of distribution confirmed the normality of the GAD-7 ($W_1 = .904$; $d.f. = 7$; $p = .36$) and PSWQ ($W_1 = .859$; $d.f. = 7$; $p = .15$) but not PBQ ($W_1 = .800$; $d.f. = 7$; $p > .04$).

Clinically significant change (CSC) was calculated as per Jacobson and Truax (1991) for the PSWQ and GAD-7, but not for the PBQ, whose thresholds are not yet known. A 7-point reduction in PSWQ scores at post-treatment and/or a total score < 47 is clinically significant (Akbari *et al.*, 2015). For the GAD-7, a total score below 10 and/or a 6-point drop from baseline denotes clinical significance (Bischoff *et al.*, 2020). On the PBQ, scores of 1–3 (i.e. 10–30 summed totals) are likely to represent normal population responses, so a score of 30 was set as an arbitrary threshold.

Results

Table 4 reports the raw scores of the seven participants on the three scales (PBQ, GAD-7 and PSWQ). All but two participants (P1 and P6) achieved clinically significant change of greater than a 6-point reduction in scores, and were within the normal population range on the GAD7. P1 was at the cut-off for GAD, while P6 was below it at baseline. On the PSWQ, all but one participant (P4) achieved clinically significant change of greater than the 7-point score reduction at post-treatment. Although all pre-treatment PSWQ scores were in the

Table 4. Outcome measures at pre- and post-treatment

Participant	PBQ		GAD7		Change	PSWQ		Change
	Pre	Post	Pre	Post	RCI	Pre	Post	RCI
1	47	27	10	7	-3 (-.98)	66	47	-19 (-3.14)
2	44	28	17	4	-13 (-4.23)	61	38	-23 (-3.79)
3	58	30	14	5	-9 (-2.93)	69	48	-21 (-3.46)
4	41	31	14	5	-9 (-2.93)	50	53	3 (.49)
5	38	27	15	5	-10 (-3.26)	64	53	-11 (1.81)
6	46	28	5	0	-5 (-1.62)	61	39	-22 (3.62)
7	40	19	12	3	-9 (-2.93)	50	37	-13 (-2.14)

RCI, reliable change index; PBQ, Premonition Bias Questionnaire; PSWQ, Penn State Worry Questionnaire; GAD-7, Generalized Anxiety Disorder 7-item scale.

clinical range (>47), at post-treatment, four (1, 2, 6 and 7) were now in the normal range. P4 scored 1 point higher than the pre-treatment score. On the PBQ, all but one participant (P4) had moved from the hypothesized clinical to the normal range. Overall, the treatment appears to have been effective in improving worry. A Wilcoxon signed rank test was run and found a significant change in believability of worry thoughts from pre- to post-treatment as measured by the PBQ. As such, the mean of ranks at post-treatment (mean=27.14; $SD=3.89$) was significantly lower than that at pre-treatment (mean=44.86; $SD=6.64$) ($W_1=-2.360$; $p=.008$). Paired-samples t -tests found significant reductions in both GAD and worry from pre- to post-treatment. As such, GAD-7 (mean difference=8.29; $SD=3.30$; $CI=5.23-11.34$; $t_6=6.639$; $p=.001$) and PSWQ (mean difference=15.14; $SD=9.21$; $CI=6.63-23.66$; $t_6=4.350$; $p=.005$) scores significantly improved from baseline.

Discussion

The case series describes the application of a novel five-module adaptation of the intolerance of uncertainty (IoU; Dugas and Robichaud, 2007) treatment of GAD. We have briefly described the adapted explanation of worry, as outlined in Chigwedere and Wilson (2021), and here, described the treatment of seven patients. The results suggest a reduction in GAD, worry and our hypothesized novel concept of premonition bias.

Most patients showed improvements on all measures, despite the treatment did not including imaginal exposure to hypothetical future-focused worrying. Others too, have reported positive results without scripted imaginal exposure (Mennin *et al.*, 2002). The UTT approach described here bears some resemblance to mindfulness, which has been found to impact neural structures and reduce worry (Hözel *et al.*, 2013). Non-judgmental engagement with images of worry triggers may have the same effects. However, by engaging with specific triggers the approach may be akin to inhibitory learning (Craske *et al.*, 2014), allowing learning of alternative meanings, not just habituation, which was not the goal of UTT.

The use of rescripting and identification of schemas may contextualize standard cognitive behavioural techniques. Doing so may improve engagement with cognitive therapy techniques, by couching them as methods to reclaim one's life from the schema. However, P4 did not improve on the PSWQ and scored higher than at baseline. This might suggest that some patients may not improve with this approach. Some patients did not reach the normal range on the PSWQ. However, except for P4 they all achieved clinically significant change. Also, only 50% of GAD patients have been shown to reach non-clinical ranges at post-treatment (Borkovec *et al.*, 2002). Interestingly, GAD7 and PBQ scores showed greater reductions than PSWQ scores, all reaching hypothesized non-clinical ranges. This may support the assertions

of others that worry is a normal experience that only becomes dysfunctional because of its appraisal (Wells, 1995). Perhaps, positive beliefs about worry may start off simply as responses to worry (Wilson, 2020; Wilson and Hughes, 2011), then become dysfunctional with time. Premonition bias and schemas may explain the initial drive towards worry, and targeting them may help reduce worry, as observed with the seven participants described here and by others (Newman *et al.*, 2015).

Worry may be an ineffective problem-solving approach, which identifies hypothetical solutions to hypothetical scenarios. It may result in dissociation of limbic and cortical structures (Trompe *et al.*, 2012). The current approach potentially recruits cortical structures by engaging them in targeting present moment worry triggers, not hypothetical future scenarios. For example, those with GAD have been shown to have reduced connectivity between the frontal cortex and limbic structures, with increased amygdala density and activation, as well as reduced uncinate fasciculus (UF) integrity, hippocampal density (Trompe *et al.*, 2012) and prefrontal cortex (PFC) activation during threat (Price *et al.*, 2011). The UF may be analogous to an information super-highway between limbic and cortical structures. Importantly, worry reduced when along with other structures, UF integrity was restored with mindfulness (Hözel *et al.*, 2013).

Furthermore, retrieved memories inform how the present moment is made sense of, and accordingly, future predictions and goals (Baddeley, 1996). Several structures, including the PFC are involved in retrieval of episodic memories (Baddeley, 1996). Compared with normal controls, GAD patients' PFC activation is significantly reduced (Price *et al.*, 2011). Such PFC deactivation and exaggerated amygdala activation support conclusions about dissociation of cortical and limbic structures. Although currently only a hypothesis, it is possible that our UTT tasks may reduce such dissociation by encouraging patients to engage with both the situational trigger and an explicitly stated uncertainty statement. It is possible that engaging with the explicitly stated uncertainty statement interferes with memory retrieval processes. Through non-judgemental engagement with the trigger and uncertainty statement, the PFC may be prevented from retrieving autobiographical memories, making associations and activating the schema for meaning making (appraisal). As such, with UTT, appraising the present moment through the lens of the schema may become difficult. The PFC may be forced to engage with, encode and appraise the same present moment event and elements activating the amygdala, not the retrieved memory. This may not only result in reduced structural dissociation, but encouragement of the development of new appraisals and encoding of new specific episodes. Implicit and over-general memory may be reduced (Williams *et al.*, 2000). Although this explanation is only theoretical and in need of more research, Williams *et al.* (2000) found an increase in memory specificity with mindfulness, a similar approach to UTT.

Although encouraging, important limitations and caveats are worth noting in interpreting these results. These are the results of a small naturalistic case series and as such, are difficult to generalize. Furthermore, the cases were treated by two closely associated therapists, with the majority being treated by one therapist. It is unclear if this approach can be learnt and applied effectively by the wider therapeutic community. Of note, premonition bias is a novel, unevaluated concept derived from clinical observation and was measured using a novel scale that still requires comprehensive validation. Both have only been used by our group in clinical practice. However, PBQ scores reduced in line with GAD-7 scores, suggesting a degree of construct validity. Furthermore, the only patient who did not improve on the PSWQ (P4) was also the only one to not score within the arbitrary normal range on the PBQ. More work is required on both the concept and the measure.

In conclusion, although its empirical support is limited, the current approach offers an interesting and potentially helpful approach to the conceptualization and treatment of IoU associated with GAD. Further research and studies with larger samples will be important, and a study incorporating these principles is being written-up. A larger study applying the current

approach is also being designed. In future it will also be important to evaluate it against existing models.

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Data availability statement. The authors confirm that the data supporting the findings of this study are available within the article [and/or its supplementary materials].

Author contributions. **Craig Chigwedere:** Conceptualization (lead), Data curation (lead), Formal analysis (lead), Investigation (equal), Methodology (lead), Project administration (lead), Validation (lead), Visualization (lead), Writing – original draft (lead), Writing – review & editing (lead); **Judy Moran:** Data curation (supporting), Investigation (equal), Methodology (supporting), Project administration (supporting), Validation (supporting), Visualization (supporting), Writing – original draft (supporting).

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