

## The Fear of Tinnitus Questionnaire

Citation for published version (APA):

Fuller, T. E., Cima, R. F. F., Van den Bussche, E., & Vlaeyen, J. W. S. (2019). The Fear of Tinnitus Questionnaire: Toward a Reliable and Valid Means of Assessing Fear in Adults with Tinnitus. Ear and Hearing, 40(6), 1467-1477. https://doi.org/10.1097/AUD.0000000000000728

Document status and date: Published: 01/01/2019

DOI: 10.1097/AUD.000000000000728

**Document Version:** Publisher's PDF, also known as Version of record

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## The Fear of Tinnitus Questionnaire: Toward a Reliable and Valid Means of Assessing Fear in Adults with Tinnitus

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**Objectives:** The purpose of this study was to assess the factor structure, reliability, and validity of the Fear of Tinnitus Questionnaire (FTQ); a brief self-report measure of people's fears about potential cognitive, emotional, behavioral, and social consequences of living with tinnitus.

**Design:** Five hundred eighty-eight Dutch-speaking adults with tinnitus completed an online battery of questionnaires measuring tinnitus-related distress, fear, catastrophizing, and quality of life. The sample was randomly split into two to perform exploratory and Bayesian confirmatory factor analyses. A subsample of participants (n = 144) completed the battery of questionnaires a second time after a 2-week interval to calculate test-retest reliability and conduct a Bland-Altman analysis. Convergent and concurrent validity of the FTQ was assessed with the complete data set and measures of tinnitus-related distress as the outcome.

**Results:** Exploratory factor analyses indicated that single- and three-factor FTQ models were both valid solutions. Posterior predictive *p* values for the Bayesian confirmatory factor analyses ranged between 0.51 and 0.53 indicating that the respective models were an excellent fit for the data. The FTQ showed excellent test-retest reliability (average value intraclass correlation coefficient (ICC) = 0.92; 95% confidence interval, 0.89–0.95) and in the Bland-Altman analysis, satisfactory agreement between participants' scores after a 2-week interval. Furthermore, the FTQ demonstrated good internal reliability ( $\alpha = 0.83$ , 95% confidence interval, 0.81–0.85) and added statistically significant amounts of variance to models predicting tinnitus-related distress and interference in daily life.

**Conclusions:** The FTQ has good psychometric properties and can be used to assess people's fear of tinnitus in research or clinical settings. Further work to establish the reliability and validity should be conducted and include an examination of a version of the FTQ that uses Likert-type response scales which might offer improved sensitivity.

Key words: Fear, Questionnaire, Reliability, Tinnitus, Validity.

(Ear & Hearing 2019;40;1467-1477)

## INTRODUCTION

Subjective tinnitus is defined as the perception of sound in the absence of an observable or objectively measureable internal or external source. Approximately 15 to 20% of the adult population reports tinnitus (Davis & El Refaie 2000; Kim et al. 2015) which can be considered to have two main elements—the

Supplemental digital content is available for this article. Direct URL citations appear in the printed text and are provided in the HTML and text of this article on the journal's Web site (www.ear-hearing.com). perceived sound and the reaction to it (Hoare et al. 2014). Subjective tinnitus (hereafter simply referred to as tinnitus) is frequently, but not always, associated with hearing loss and other otological conditions such as hyperacusis and otosclerosis (Langguth et al. 2013). It is as of yet unclear how acute tinnitus develops into chronic debilitating complaints. While many people experience tinnitus only a small proportion (i.e., 1-3%; Davis & El Refaie 2000; Fujii et al. 2011; Kim et al. 2015) report severe levels of distress, interference in social and occupational functioning, difficulties with concentration, memory dysfunction, sleep problems, lower quality of life, and severe psychological suffering (Andersson & Edvinsson 2008; Joo et al. 2015; Tyler & Baker 1983). The psychological consequences associated with living with bothersome tinnitus include increased chances of higher levels of depression and anxiety symptoms (Sullivan et al. 1988; Zöger et al. 2006; Langguth 2011; Pinto et al. 2014; Pattyn et al. 2016), and lower self-esteem (Krog et al. 2010). These potential undesirable consequences combined with troubling thoughts about possible physical or psychological damage in all likelihood add to the conviction in many patients that tinnitus is something to fear.

Fear is an emotional state or response triggered by existing or imminent threats to one's health or safety. It is usually considered adaptive as it motivates people to try to minimize or prevent harm from occurring. However, persistent efforts to avoid or escape (perceived) threatening situations where (increased) tinnitus is expected can be considered maladaptive. That is, in the long term, the level of distress and disability might worsen as a consequence, for example, of people avoiding or withdrawing from social or work situations that they fear might worsen the tinnitus.

#### Fear Avoidance Model of Tinnitus

The fear avoidance (FA) model has, as its name suggests, the concepts of *fear* and *avoidance* as its central tenets. It is a cognitive behavioral model that was developed in an attempt to better understand and predict the distress and interference in mood and daily life activities of people living with chronic pain (Lethem et al. 1983; Vlaeyen & Linton 2000, 2012; Vlaeyen et al. 2016). The similarities between chronic pain and chronic tinnitus (Møller 1997, 2000) have proved informative for the understanding of tinnitus suffering, and hence the FA model has recently been applied to tinnitus (Cima et al. 2011a; Kleinstäuber et al. 2013). [See Cima et al. (2011a) or Kleinstäuber et al. (2013) for a visual representation of the FA model of tinnitus.] The FA model of tinnitus is predictive in that it suggests directional relationships between the respective variables within it. The model offers the possibility to formulate hypotheses about how the perception of tinnitus triggers cognitive, emotional, and behavioral responses leading to distress and interference in some people but not others. Specifically,

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it predicts that through catastrophic (mis)interpretations about the sound such as that it is a sign of impending harm (Andersson & McKenna 2006), the subsequent fear of the tinnitus leads to behavioral strategies to avoid instances of worsened tinnitus (Hallberg et al. 1992; Budd & Pugh 1996). The model also predicts that people who interpret the tinnitus sound as benign will not have fearful reactions and therefore will not engage in avoidant, escape, or safety behaviors.

# Development of and Previous Research with the Fear of Tinnitus Questionnaire

Tinnitus-related fear as a concept has thus far received relatively little attention in the research literature and was not specifically measured until the Fear of Tinnitus Questionnaire (FTQ; Supplemental Digital Content 1, http://links.lww.com/EANDH/A526) was developed. It is a brief self-report measure that specifically aims to measure levels of fear of experiencing tinnitus and possible consequences associated with it. The FTQ was based on two measures of fear associated with pain [the Tampa scale for Kinesiophobia (TSK; Roelofs et al. 2007) and the Pain Anxiety Symptoms Scale (McCracken et al. 1992)] was designed to produce a total score indicative of fear of tinnitus, and to be used as an outcome measure rather than a diagnostic tool. Higher scores on the FTQ are intended to indicate higher levels of tinnitus-related fear.

Recent research in a clinical sample has found that high levels of fear of tinnitus are indeed associated with higher tinnitus distress and poorer quality of life (Cima et al. 2011a, 2017). Comprehensive mediation analyses indicated that reductions in tinnitus-related fear explain the improvements in quality of life and decrease in tinnitus-related distress following specialized CBT for tinnitus (Cima et al. 2017).

In addition in a large randomized controlled trial, results showed that the FTQ total score changed over time and between two clinical treatment groups (Cima et al. 2012). While the FTQ total score appears to work as intended, researchers have not previously examined its psychometric properties. Further, we do not know if the FTO is actually a unifactorial measure, has redundant items, or has subscales that explain/predict tinnitus-related distress. The purpose of this study was thus to investigate the psychometric properties of the FTQ by examining the validity and reliability of the factor structure and the ability of the measure to predict levels of tinnitus related distress. It extends existing research examining data from the FTQ (e.g., Cima et al. 2011a) by using data from a sample of people experiencing tinnitus but not participating in a clinical trial. No hypotheses were made regarding the test-retest reliability or the number of underlying factors likely to be identified in the FTO. We expected that the FTQ total score would positively correlate with health anxiety, illness behavior, and physical and mental health. Finally, based on the FA model (Lethem et al. 1983; Vlaeyen & Linton 2000, 2012; Vlaeyen et al. 2016), fear of tinnitus, as measured by the FTO, was predicted to be associated with measures of tinnitusrelated disability, distress, and intensity.

#### MATERIALS AND METHODS

A brief overview of the participants and procedures are provided here as they have been described in detail in a previous publication (Cima et al. 2011a). Ethics approval to conduct the study was given by the Ethics Committee of the Faculty of Psychology and Neuroscience at Maastricht University, The Netherlands (approval number: 72\_04\_07\_2008).

#### **Participants**

Advertisements placed on the websites of the Dutch Association for Hearing Disorders (www.nvvs.nl) and the Dutch Tinnitus Platform (www.tinnitus.nl) between November 2008 and April 2009 invited visitors to the website to participate in a large cross-sectional study that included a battery of tinnitus-specific, general health, and mood-related measures. Participants who experienced, but were not necessarily bothered or distressed by, tinnitus were eligible to participate in this study. We have chosen to select a broad range of participants to maximize generalizability. We included a broad variety of tinnitus sufferers, including variety concerning their experienced level of distress.

Seven hundred ninety-one respondents requested additional information about the study and consent forms. Of the 615 participants who returned their consent form and completed at least one questionnaire, 588 completed the FTQ. For the purposes of this study, data from the 588 participants who completed the FTQ were retained for analysis. That is, there were no missing data on the FTQ, but potentially on other questionnaires (see Table 5). (Note: not all participants provided response to questions about age, gender, tinnitus duration, and other demographic variables, and thus the sample size numbers do not always equal 588.) The sample included 230 women with a mean age of 50 years (range 18-80 years; SD 11.33 years) and 342 men with a mean age of 53.5 years (range 17-81 years; SD 53.46). Most of the participants were employed and approximately 80% were not currently receiving any treatment for tinnitus. Tables 1 and 2 report demographic characteristics for the

 
 TABLE 1. Participant characteristics and P values from Chisquare tests indicating EFA and CFA sample equivalence

		Sample		
Characteristic	EFA, n (%)	CFA, n (%)	Total, n (%)	p
Gender				
Female	118 (41.69)	112 (38.75)	230 (40.20)	0.47
Male	165 (58.31)	177 (61.25)	342 (59.80)	
Education				
Primary	33 (11.66)	39 (13.49)	72 (12.59)	0.52
Secondary	66 (23.32)	63 (21.80)	129 (22.55)	
Vocational training	55 (19.43)	62 (21.45)	117 (20.45)	
Higher education	129 (45.56)	125 (43.25)	254 (44.40)	
Location of tinnitus				
Left ear	66 (23.32)	58 (20.07)	124 (21.67)	0.39
Right ear	34 (12.01)	48 (16.60)	82 (14.34)	
Both ears	146 (51.59)	143 (49.48)	289 (50.52)	
Middle of head	37 (13.07)	40 (13.84)	77 (13.46)	
Hearing loss				
Present	140 (49.46)	135 (46.71)	275 (48.07)	0.51
Employed				
Yes	174 (30.42)	179 (31.29)	353 (61.71)	0.49
No	115 (20.10)	104 (18.18)	219 (38.29)	
Currently receiving treatment				
Yes	70 (12.23)	55 (9.62)	125 (21.85)	0.11
No	283 (49.48)	289 (50.52)	447 (78.15)	

CFA, confirmatory factor analysis; EFA, exploratory factor analysis.

TABLE 2. Descriptive statistics of participants' tinnitus and age by sample with p values from independent samples *t*-test indicating equivalence between groups

		Sample		
Characteristic	EFA, n = 283	CFA, n = 289	Total	р
Tinnitus duration Years (SD)	5.42 (1.49)	5.59 (1.43)	5.51 (1.46)	0.17
Tinnitus severity VAS (SD)	5.59 (2.00)	5.82 (1.97)	5.71 (1.99)	0.24
Age Years (SD)	51.30 (12.07)	52.84 (11.14)	52.07 (11.63)	0.11

Missing age-related data for n = 16 participants.

CFA, confirmatory factor analysis; EFA, exploratory factor analysis; VAS, visual analog scale.

samples used for exploratory and confirmatory factor analysis (CFA), respectively.

#### Procedure

After giving consent, participants received unique login codes via e-mail to access the online assessment by questionnaires. The measures used in this study were selected because of their sound psychometric properties, relevance for the research question, and frequent use in tinnitus research as outcome measures. The battery of questionnaires which were all presented in Dutch was hosted on the "Emium" website (www. emium.nl; Janssen 2008). The order of questionnaires and questions was fixed, with questionnaires being presented as a self-contained set of questions with specific instructions and clear demarcations of the start and end points. The questionnaires were presented in the following order: Short Form-36, Tinnitus Catastrophizing Scale (TCS), Tinnitus Questionnaire (TQ), Illness Attitudes Scale (IAS), FTQ, Tinnitus Disability Index (TDI), demographic questions, and visual analog scales measuring tinnitus intensity. Participants were able to read and complete them independently but were not required to complete the questionnaires in one sitting (although they were instructed to complete them within a day) and hence could login and out as they wished.

For the purpose of establishing the test-retest reliability of the FTQ, a subset of 250 participants were invited exactly 2 weeks after first completing the questionnaire battery to do so a second time. Over half (n = 144, 58%) of these participants who were not undertaking treatment for tinnitus-related distress

(according to self-report) completed all the measures in the questionnaire battery a second time.

The complete sample (N = 588) was randomly split precisely in half to perform the exploratory and confirmatory factor analyses on two separate subsamples. We chose to split the sample in half for cross-validation purposes in accordance with the recommendations of Gerbing and Hamilton (1996). They found—by using Monte Carlo methods—that EFA contributes to model specification before cross-validation using CFA and was hence a useful method in identifying a model that could be evaluated with CFA.

#### Measures

The Cronbach alphas for the questionnaires listed below are presented in Table 3 with values shown for the respective samples.

FTQ (Cima et al. 2011a). The FTQ is a 17-item self-report measure designed to assess respondents' level of fear regarding their tinnitus. Items in the questionnaire are presented as a series of statements (e.g., "I am afraid that my tinnitus will become worse") for which respondents are asked to indicate if it is applicable to their current situation. Each statement receives a score of 1 when applicable and 0 when not applicable. The maximum is 17 with higher scores reflecting higher levels of tinnitus related fear. The total score is the sum of all applicable statements and provides an overall rating of fear of tinnitus. The FTQ has shown to be sensitive to change following treatment (Cima et al. 2012).

TCS (Cima et al. 2011a) assessed thinking the worst about tinnitus (i.e., catastrophizing). The TCS is a 13-item measure based on the Pain Catastrophizing Scale (Sullivan et al. 1995). Respondents use a five-point scale ranging from 0 to 4 to indicate the degree to which statements (e.g. "It's terrible and I think it's never going to get any better") applies to them. Higher scores indicate higher levels of catastrophizing and with the maximum being 62.

TQ (Hallam et al. 1988; Meeus et al. 2007) is a widely used measure of tinnitus-related distress. It contains 52 items, has a three-point scale representing "true" "partly true," and "not true" that respondents use to indicate levels of agreement with the respective items (e.g., "It's unfair that I have to suffer with my noises") and a maximum score of 84 (minimum is zero). Higher scores indicate greater levels of tinnitus-related distress. The TQ has six subscales within the measure that includes emotional and cognitive distress, sleep disturbance, intrusiveness, auditory and perceptual difficulties, and somatic complaints

TABLE 3. Internal consistency for	or questionnaires	used by respective samp	les
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		Sample $\alpha$ (95%	CI)	
Measure	Entire Sample (N = 588)	Test-Retest (n = 144)	EFA (n = 294)	CFA (n = 294)
FTQ	0.83 (0.81–0.85)	0.84 (0.80-0.88)	0.82 (0.79–0.85)	0.84 (0.81–0.86)
TCS	0.93 (0.93–0.94)	0.94 (0.93–0.95)	0.94 (0.92–0.95)	0.93 (0.92-0.94)
TQ	0.94 (0.93–0.95)	0.95 (0.94–0.96)	0.94 (0.93–0.95)	0.94 (0.93-0.95)
TDI	0.89 (0.87–0.90)	0.89 (0.86–0.92)	0.90 (0.88–0.92)	0.87 (0.85–0.90)
IAS	0.85 (.83–0.86)	0.88 (0.86–0.91)	0.85 (0.83–0.87)	0.85 (0.82–0.87)
SF-36 PH	0.91 (0.90–0.92)	0.91 (0.89–0.92)	0.92 (0.91–0.92)	0.90 (0.89–0.92)
SF-36 MH	0.93 (0.92–0.93)	0.93 (.92–0.95)	0.93 (0.91–0.94)	0.93 (0.91–0.94)

CFA, confirmatory factor analysis; EFA, exploratory factor analysis; FTQ, Fear of Tinnitus Questionnaire; IAS, Illness Attitude Scale; SF-36 PH, Short Form-36 Physical Health; SF-36 MH, Short Form 36 Mental Health; TCS, Tinnitus Catastrophizing Scale; TDI, Tinnitus Disability Index; TQ, Tinnitus Questionnaire.

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attributable to the tinnitus. The TQ has high internal consistency, convergent, and discriminant validity and is sensitive to change (Baguley et al. 2000; Zeman et al. 2012).

TDI (Cima et al. 2011b) was used to measure tinnitus-related functional impairment. The TDI is a 7-item measure that uses an 11-point Likert-type scale (range from 0 to 10; maximum score 70) to assess the degree to which respondents' daily life activities (e.g., social activities) are adversely affected by tinnitus. Higher scores indicate higher levels of disability. The respective anchors are "no disability" and "total disability". The TDI has sound psychometric properties and is strongly correlated with measures of quality of life and psychological distress associated with tinnitus (Cima et al. 2011b).

Three questions were used to measure the intensity of participants' tinnitus. [Note that, at the time of data collection, the Tinnitus Magnitude Index (Schmidt et al. 2014) had not been developed yet.] The respective questions, ("How do you judge your tinnitus when it was at its worst? How do you judge your tinnitus when it was at its least? How did you judge your tinnitus when it was at its least? How did you judge your tinnitus when it was at its usual intensity?") had to be answered using visual analog scales with the endpoint anchors "not intense at all" and "the most intense imaginable." Participants were instructed to "click" on a point on the respective horizontal lines that was indicative of the tinnitus severity. Although it was not visible to respondents, the points along the horizontal line represented scores for 0 to 100. Although it is not usual practice to do so, scores for each question were averaged to produce a single "tinnitus intensity" score to be used in the subsequent analyses.

IAS (Kellner 1987) is a 29-item measure of fears, beliefs, and attitudes related to health. The original version includes nine subscales although a factor analysis of the Dutch version indicated that a two-factor solution provided the best model fit. The two scales-health anxiety (items 2-4, 6, 13-17, 19, and 21) and illness behavior (items 23-25 and 27-29)(Speckens et al. 1996) are used in this study. The IAS asks respondents to report on a five-point Likert-type scale the frequency with which they experience particular health-related thoughts or behaviors. The response options range from "no" (zero points) to "most of the time" (four points) with higher scores indicating poorer greater levels of health anxiety and illness behavior, respectively. The maximum scores for Health Anxiety and Illness Behavior subscales are 48 and 24, respectively. The IAS has been demonstrated to possess stable test-retest reliability of the subscales (Kellner 1987; Fava et al. 1988) and has good concurrent, convergent, and discriminant validity (Stewart & Watt 2000).

The Short Form-36 (SF-36; Hays et al. 1993; Ware Jr et al. 1998; Mösges et al. 2008) is a widely used self-report measure of health. It has 36 items that can form two independent global scores that relate to physical and mental health, respectively (McHorney et al. 1993). Higher scores (maximum equals 100 and minimum equals 0) indicate better quality of life. The SF-36 has been translated into Dutch, validated and is considered appropriate for use with people with or without a chronic health condition (Aaronson et al. 1998). Although an SF-36 total score is sometimes calculated, it is not recommended by the developers of the tool (Lins & Carvalho 2016), and hence we use the two summary health scores.

**Demographics** • Participants responded to a brief set of demographic questions asking for details of their gender, age, education, duration of tinnitus, hearing loss, current treatments, employment status, and history of absences from work due to illness.

## Analysis

R version 3.2 and the "userfriendlyscience" package (Peters 2014) were used to calculate confidence intervals and estimates of reliability for the questionnaires. IBM SPSS Statistics version 24 was used for exploratory factor analysis, to calculate correlations and hierarchical regressions to inform judgments about concurrent and convergent validity and test-retest reliability. We used IBM SPSS AMOS version 24 to conduct Bayesian CFA. In all analyses where data were missing, pairwise deletion was used instead of imputation methods.

We adopted a conservative approach in testing the assumption that the FTQ is best interpreted as a single-factor measure.

Data used in the analyses are held in a repository managed by Maastricht University and available upon request.

Exploratory Factor Analysis • The FTQ items have binary response options, in which case it is not advised to use traditional factor analytic methods (Lee & Song 2003) as it can lead to "biased standard errors and significance tests, overestimation of the number of factors, and underestimation of the factor loadings" (Woods 2002). Given this, tetrachoric correlation matrices were used instead and were calculated using a macro specifically developed for the purpose by Lorenzo-Seva and Ferrando (2012) in IBM SPSS Statistics version 24. The assumptions and conditions regarding the suitability of the data set were examined before conducting analyses. Specifically, the ratio of sample size to number of variables, Kaiser-Meyer-Olkin (KMO) measure of sampling adequacy and Bartlett test of sphericity were checked. Principal Axis Factoring with oblique rotation was conducted on the tetrachoric correlation matrices. The number of factors to extract was determined by examining the scree plot, Kaiser criterion (i.e., eigenvalues > 1) and parallel analysis. Parallel analysis (Horn 1965) is an accurate and robust (Zwick & Velicer 1986) Monte Carlo simulation technique that estimates the number of possible factors to be retained. The simulations/calculations are based on random data sets that have the same number of variables and sample size as the real correlation matrix being analyzed. Factor loadings of 0.32 or greater were considered sufficient (Comrey & Lee 1992). However, we did not automatically discard items with loadings that were below, but still close to, this cutoff value because it was possible that items could be theoretically or conceptually similar to other items on a subscale.

Confirmatory Factor Analysis • To conduct CFA for dichotomous data, IBM SPSS AMOS version 24 offers Bayesian CFA. This method has several advantages over a frequentist approach. For example, it can provide a good performance in small samples; avoid inadmissible model parameter values; offer computational advantages and intuitive interpretation of the results, and allow for the specification of background knowledge through the specification of prior knowledge of parameters/distributions (Hoofs et al. 2017). We did not however specify a prior distribution, because with a large sample size selecting priors makes little difference to outcomes and as a result used the Amos default uniform distribution setting (Arbuckle 2013). A cutoff was set at 1.1 for convergence of the model (Hu & Bentler 1999) and posterior predictive p was used for assessing goodness of fit of model. Posterior predictive p is a Bayesian estimate of goodness of fit where the closer the posterior predictive p values are to 0.50 the better the model fit is considered (Muthén & Asparouhov 2012; Gelman 2013).\*



Fig. 1. Bland-Altman plot for FTQ total score at time points 1 and 2 (n = 144). LOA: Limits of agreement defined as the mean difference of the scores at time points one and two  $\pm 1.96$  SD of the differences. FTQ, Fear of Tinnitus Questionnaire.

Test-Retest Reliability • A subset of 144 respondents not undertaking treatment completed the questionnaires again after a 2-week interval for the purposes of collecting data for test-retest analysis. We used two methods to assess test-retest reliability. First, the intraclass correlation coefficient using a two-way mixed model with absolute agreement was calculated. Second, we conducted a Bland-Altman analysis (Bland & Altman 1986) to examine the level of agreement between the FTO total scores at the respective time points. The Bland-Altman analysis is comprised of three steps: first, a single sample t-test to determine if the mean difference between the scores at the two time points was significantly different from zero (from a statistical perspective); second, plotting these differences between the two FTQ total scores by the average FTQ total scores across the two time points (see Fig. 1); and finally, conducting a simple linear regression to examine if proportional bias was present.

*Convergent validity* was investigated by examining the correlations between the total score of the FTQ with measures of tinnitus related distress and impairment (i.e., TQ and TDI), the health anxiety and illness behavior scales of the IAS, and the physical and mental health summary measure scores of the SF-36.

To test *concurrent validity* (the relationship between a distinct but related concept/measure) of the FTQ, we examined the unique contribution of tinnitus-related fear in accounting for variability in tinnitus distress (i.e., TQ) and tinnitus-related disability (i.e., TDI) using hierarchical regression. The criterion for multicolinearity—variance inflation factor—was set at 2. The FTQ total scores were entered into the regression analyses after controlling for age, gender, education, self-reported tinnitus severity, health anxiety and illness behavior, and mental and physical health as measured by the SF-36.

## RESULTS

#### **Sample Equivalence**

Chi-square and independent samples *t*-tests using IBM SPSS statistics version 24 revealed that the samples used for the exploratory and confirmatory factor analyses were equivalent. There were no significant differences on gender, highest

completed level of education, age, duration of tinnitus, whether they had hearing loss, the severity of the tinnitus, the location of their tinnitus, employment status and whether they were currently receiving treatment (Tables 1 and 2).

## **Exploratory Factor Analysis**

Exploratory factor analysis was conducted using principal axis factoring on a random selection of half of the participants (n = 294) on the FTQ with oblique rotation. The KMO measure of sampling adequacy was poor (KMO = 0.121) indicating that some items might need to be removed from the analysis. Bartlett test of Sphericity  $\chi^2(136) = 4289.05$ , p < 0.001, however, was sufficient indicating that the correlations between items were large enough to proceed with factor analysis. There was variation between the prespecified methods for determining how many factors to extract. Specifically, six factors had eigenvalues larger than 1; the scree plot (Supplemental Digital Content 2, http://links.lww.com/EANDH/A527) indicated that retaining up to four factors could be appropriate and parallel analysis indicated that three factors would be optimal. Given this variation, we examined four possible factor structures, with one to four retained factors, in attempt to identify the most parsimonious solution.

A single-factor solution accounted for 30.6% of the variance, while two-, three-, or four-factor solutions accounted for 43.3, 54.9, and 62.3% of the variance, respectively. Inspection of the two-factor pattern matrix (Supplemental Digital Content 2, http://links.lww.com/EANDH/A527) revealed that the first factor contained 11 items and the second contained 4 items. Item 12 did not load on either factor (factor loadings of 0.18 and -0.01, respectively) while item 16 loaded on both factors (0.50 and 0.31, respectively). This type of cross-loadings is undesirable (Costello & Osborne 2005). Based on this, we considered removing items 12 and 16. Item 6 showed a loading of 0.28 which we considered to be close to the cutoff. In addition, as this item did not have cross-loadings, we decided to retain it in the analysis. However, this item should be treated with some caution. The three-factor pattern matrix (Table 4) yielded three factors with eight, four, and three items, respectively. Item 12

TABLE 4. Pattern matrix of exploratory factor analysis—three-factor solution for the Fear of Tinnitu	2 Questionnaire (n = 2	294)
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		Factor			
Item Number and Statement	Future Consequences	Somatic	Deterioration		
(5) I am afraid that my tinnitus will drive me crazy	0.796	0.154	-0.022		
(8) I am afraid the moment will come that my head cannot withstand tinnitus anymore	0.595	0.153	0.148		
(9) My mental condition will become severely affected by my tinnitus	0.731	0.066	-0.026		
(10) I am afraid that tinnitus will stop me from ever having a normal life again	0.696	-0.076	-0.090		
(11) I am afraid that I will never be able to experience silence again because of tinnitus	0.701	-0.141	0.049		
(13) I am afraid I will not be able to do anything anymore because of my tinnitus	0.503	0.110	0.113		
(14) It worries me to think I may never be able to learn how to cope with this condition	0.860	-0.053	-0.090		
(15) It would be terrible if my tinnitus proved a life-long condition	0.434	-0.090	0.142		
(16) I am concerned that tinnitus may be a risk to my physical health	0.505	<u>0.312</u>	0.009		
(3) I fear that my tinnitus is the result of a tumor	-0.089	0.876	0.037		
(4) Even though my tinnitus is getting worse. I do not think it points to a serious disease	-0.142	0.729	-0.021		
(17) I am afraid that tinnitus may be a preliminary sign of brain hemorrhage or similar	0.160	0.579	0.150		
(6) The fact that I have tinnitus does not mean that my health is at risk	0.105	0.282	-0.058		
(1) I am afraid that my tinnitus will deteriorate my hearing	-0.137	0.005	0.986		
(2) I am afraid that my tinnitus will become worse	0.131	-0.066	0.452		
(7) I am afraid my tinnitus will leave me deaf	-0.077	0.108	0.890		
(12) I am afraid that loud noises will aggravate my tinnitus	0.102	-0.013	0.149		
Eigen values	5.20	2.17	1.96		
% of variance	30.57	12.75	11.55		
α	0.80	0.53	0.67		
(95% CI)	0.77–0.84	0.45–0.61	0.60-0.73		

Bold indicates factor loadings above or close to the cutoff of 0.32. Underlined values indicate cross-loading of items on two or more factors.

Cl, confidence interval.

did not load on any factor and item 16 loaded on two factors, which indicates that they do not appear to be measuring the same construct and thus could be removed from further analysis. Again, item 6 had a loading of 0.28 but showed no cross-loadings and was conceptually similar to other items on the subscale. In the three-factor solution, the third factor comprised the three items that showed the lowest loadings on the factor with 11 items in the two-factor solution. A *four-factor solution* (see Supplemental Digital Content 2, http://links.lww.com/ EANDH/A527) yielded two factors with four items, one factor with three and one factor with only one item, respectively. Four items (8, 13, 14, and 16) loaded on two factors. Item 12 did not load on any of the possible four factors.

Of these possible solutions, overall, it appeared that the single-factor and three-factor solutions were more defensible than

	TABLE 5.	Descriptive	statistics	from	outcome	measures
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	Mean (95% CI)	SD	Min-Max	N
FTQ	4.66 (4.44-4.88)	2.76	0–12	588
TCS	21.75 (20.8-22.71)	11.78	0–52	588
TDI	248 (235–261)	154	0–648	581
TQ	47.09 (45.57-48.62)	18.83	5–96	588
SF-36 PH	45.97 (45.10–46.85)	10.77	17–79	588
SF-36 MH	45.39 (44/48-46.31)	11.26	23–78	588
IAS-HA	9.43 (8.89–9.97)	6.67	0–36	588
IAS-IB	11.97 (11.60–12.35)	4.59	3–26	588
Tinnitus intensity	5.71 (5.51–5.91)	1.99	0–10	385

FTQ, Fear of Tinnitus Questionnaire; IAS-HA, Illness Attitude Scale-Health Anxiety; IAS-IB, Illness Attitude Scale-Illness Behavior; SF-36 PH, Short Form-36 Physical Health; SF-36 MH, Short Form-36 Mental Health; TCS, Tinnitus Catastrophizing Scale; TDI, Tinnitus Disability Index: TO, Tinnitus Questionnaire: Tinnitus Intensity = averace tinnitus intensity. the two- or four-factor solutions. First, the four-factor solution was clearly less coherent with a factor containing only a single item and four items showed cross-loadings. Second, the threefactor solution actually groups the items with lowest loadings from the two-factor solution neatly to form a meaningful third factor. A single-factor solution accounts for a little over 30% of the variance and is most consistent with the intention of the developers of the FTQ. On the other hand, a three-factor solution accounts for a greater proportion of variance (55%) and has clusters of items that could theoretically represent meaningful subscales of fears relating to: future consequences (items: 5, 8, 9, 10, 11, 13, 14, 15); hearing and tinnitus-related deterioration (items: 1, 2, 7); and, somatic conditions (items: 3, 4, 6, 17). The internal consistency of these potential FTQ subscales was  $\alpha = 0.80$  (95% confidence interval, CI, 0.77–0.84) for *fu*ture consequence,  $\alpha = 0.67$  (95% CI, 0.60–0.73) for hearing and tinnitus-related *deterioration*, and  $\alpha = 0.53$  (95% CI, 0.45–0.61) for *somatic*-related fears.

#### **Confirmatory Factor Analyses**

Given the results of the exploratory factor analysis, three models were tested: two single-factor models (one model with and one model without items 12 and 16) and a three-factor model (items 12 and 16 removed) with the subscales of *future consequences, deterioration,* and *somatic-related fears,* using the confirmatory sample (n = 294).

Because the FTQ items have binary response options, the asymptotic distribution function was used, as this does not require the assumption of normality to be met. Bayesian estimation was conducted to generate estimates of the goodness of fit (posterior predictive p values) for the respective models. The posterior predictive p values for the respective models for the

single-factor solution were as follows: FTQ-17 item = 0.53, FTQ-15 item = 0.52 and for the three-factor solution: FTQ-15 item = 0.51. Detailed results from the Bayesian analysis showing the regression weights, intercepts, and variance are provided in Supplemental Digital Content 3, http://links.lww. com/EANDH/A528. The posterior predictive p values indicate that each of the respective models provides an excellent fit with the data from the confirmatory sample. No other indices of goodness of fit were calculated due to the binary nature of the data.

The following results all relate to the 17-item version of the FTQ.

#### **Test-Retest Reliability**

The average value intraclass correlation coefficient for the FTQ total score was excellent (0.92; 95% CI, 0.89–0.95) and statistically significant (F[144, 144] = 13.82, p < 0.001) indicating that respondents' total FTQ scores were very similar when completed after a 2-week interval.

The Bland-Altman analysis revealed a more a slightly more complex picture in the data over time. Specifically, the single sample *t-test* indicated that the mean difference (0.56 between the scores) was significantly different from zero (t[144] = 3.46. p < 0.01) which suggests that there is limited agreement between the FTQ scores at the respective time points. However, from a clinical perspective, an average difference of <1 is unlikely to be meaningful. Subsequent simple linear regression analysis predicting the difference between the FTQ total scores based on the average FTQ total score indicated that there was no proportional bias in the data (F[1, 143] = 0.05, p > 0.05, with an  $R^2 = 0.00$ ). This means that there was no systematic variation in the FTQ total scores at time one and two over the range of possible scores. For example, the differences in total FTQ scores between time point one and two did not increase with increasing average FTQ total score.

The Bland-Altman plot (Fig. 1) reveals that 8 of the 144 participants' results were outside the upper and lower limits of agreement (defined as the mean difference of the scores at time points one and two  $\pm 1.96$  SD of the differences). Overall, this result suggests that there was satisfactory agreement between participants' scores after a 2-week interval.

## **Convergent Validity**

Tables 5 and 6 respectively show the descriptive statistics and bivariate correlations for the FTQ, and target variables tinnitus disability (TDI), tinnitus intensity, health anxiety, illness behavior, overall physical and mental health, and tinnitus-related distress (TQ). Table 6 reveals that virtually all the variables are positively and significantly correlated with each other but that the strength of association varies. Specifically, the FTQ total score is strongly correlated (i.e.,  $r \ge 0.7$ ) with the TCS and TQ total scores, moderately correlated (i.e., r = 0.3 to .7) with TDI, SF-36 MH, IAS-HA, IAS-IB and weakly (i.e.,  $r \le 0.3$ ) correlated with SF-36 PH. These results suggest that someone who is more afraid of tinnitus (i.e., high scores on the FTQ) is more likely to also experience higher levels of: (a) tinnitus related distress; (b) catastrophizing; (c) health anxiety; (d) tinnitus interference in daily life; (e) illness behavior; (f) tinnitus intensity; (g) mental health problems; and (h) physical health problems.

 TABLE 6. Pearson's correlation coefficients of the Fear of

 Tinnitus Questionnaire total score with included measures

	FTQ	TCS	TDI	TQ	SF-36 PH	SF-36 MH	IAS-HA	IAS-IB
TCS	0.72*							
TDI	0.53*	0.51*						
TQ	0.75*	0.77*	0.71*					
SF-36 PH	0.16*	0.16*	0.33*	0.25*				
SF-36 MH	0.40*	0.38*	0.43*	0.48*	0.41*			
IAS-HA	0.43*	0.36*	0.21*	0.29*	0.03	0.24*		
IAS-IB	0.33*	0.28*	0.48*	0.45*	0.45*	0.43*	0.24*	
Tinnitus intensity	0.45*	0.51*	0.48*	0.59*	0.20*	0.29*	0.15*	0.34*

There was no missing data on all questionnaires/scores except for TDI (n = 581) and Tinnitus Intensity (n = 385). We used pairwise deletion rather than imputation of values for missing data.

FTQ, Fear of Tinnitus Questionnaire; IAS-HA, Illness Attitude Scale-Health Anxiety; IAS-IB = Illness Attitude Scale-Illness Behavior; SF-36 PH, Short Form-36 Physical health; SF-36 MH, Short-Form-36 Mental health; TCS, Tinnitus Catastrophizing Scale; TDI, Tinnitus Disability Index; TQ, Tinnitus Questionnaire; Tinnitus Intensity = average tinnitus intensity.  $^{\circ}p < 0.01$ .

#### **Concurrent Validity**

A series of four hierarchical regressions (see Tables 7 and 8) were conducted to examine the respective proportions of variance explained in the TDI, TQ, SF36-PH, and SF36-MH by the FTQ total score. In each regression, age, gender, education, and average tinnitus intensity were the first group of variables entered into the model. The second block of variables entered into the model included mental and physical health, health anxiety, and illness behavior except in the instance when SF36-PH and MH, respectively, were used as the dependent variable. Multicollinearity was a problem between the FTQ and TCS (variance inflation factor = 2.64). Given that the regression equations were run omitting TCS from the models. The FTQ

 
 TABLE 7. Summary of hierarchical regression analysis for predicting tinnitus-related disability and distress

	Т	innitus-rela	ated Outcom	e	
	Interfere Activitie	Interference in Activities (TDI)		Distress (TQ)	
	$\Delta R^2$	β	$\Delta R^2$	β	
Step 1 Control variables*	0.24††		0.39††		
Step 2 SF-36 PH SF-36 MH IAS-HA IAS-IB	0.15††	0.08 0.21† 0.03 0.23†	0.15††	-0.04 0.23†† 0.13†† 0.22††	
Step 3 FTQ total score	0.06††	0.33†	0.17††	0.55††	
Total <i>R</i> ² n	0.45† 385		0.71†† 385		

 $\Delta R^2$  = change in amount of variance explained at each step in the regression model.  $\beta$  values indicate how much the Tinnitus Questionnaire and Tinnitus Disability Index scores change by when the FTQ total score changes by one standard deviation.

Control variables included age, gender, education, average tinnitus intensity.

 $^{\dagger}p < 0.01.$  $^{\dagger\dagger}p < 0.001.$ 

FTQ, Fear of Tinnitus Questionnaire; IAS-HA, Illness Attitude Scale-Health Anxiety; IAS-IB, Illness Attitude Scale-Illness Behavior; SF-36 PH, Short Form-36 Physical Health; SF-36 MH. Short Form 36 Mental Health: TDI. Tinnitus Disability Index: TQ. Tinnitus Questionnaire.

TABLE 8. Summary	of hierarchical	regression	analysis for
variables predicting	mental and phy	sical health	(i.e., SF-36
subscales)			

		Health Outcome				
	Mental	Health	Physica	l Health		
	$\Delta R^2$	β	$\Delta R^2$	β		
Step 1 Control variables*	0.09††		0.07††			
Step 2 SF-36 PH SF-36 MH IAS-HA IAS-IB	0.20††	0.26† NA 0.15† 0.26†	0.20††	NA 0.27† -0.13† 0.32†		
Step 3 FTQ total score Total <i>R</i> <sup>2</sup> n	0.03†† 0.33†† 385	0.23†	0.00 0.27†† 385	-0.09		

 $\Delta R^2$  = change in amount of variance explained at each step in the regression model.  $\beta$  values indicate how much the SF-36 Mental health and SF-36 Physical health scores respectively change by, when the FTQ total score is changes by one standard deviation. Control variables included age, gender, education, average tinnitus intensity.  $^7p < 0.01$ .

<sup>t†</sup>ρ < 0.001

FTQ, Fear of Tinnitus Questionnaire; IAS-HA, Illness Attitude Scale-Health Anxiety; IAS-IB, Illness Attitude Scale-Illness Behavior; NA, Not applicable; SF-36 PH, Short Form-36 Physical Health; SF-36 MH, Short Form 36 Mental Health; TDI, Tinnitus Disability Index; TQ, Tinnitus Questionnaire.

total score significantly contributes to explaining variance in tinnitus-related distress and impairment as measured by the TQ and TDI, respectively (Table 7). The model however accounted for less variance of TDI as a dependent variable (adjusted  $R^2 = 0.43$ ; F[9, 375] = 33.72, p < 0.001) compared with TQ as a dependent variable (adjusted  $R^2 = 0.70$ ; F[9, 375] = 102.07, p < 0.001) indicating that the FTQ total score is better suited for explaining distress rather than tinnitus-related disability. The models also predict a larger proportion of variance in mental health (adjusted  $R^2 = 0.31$  F[8, 376] = 22.72, p < 0.001) compared with physical health (adjusted  $R^2 = 0.25$  F[8, 376] = 17.37, p < 0.001) as measured by the SF-36 (Table 8).

#### **Response Distributions**

A post hoc, exploratory examination of the response frequency on the FTQ items revealed that five statements (numbers 3, 4, 6, 13, and 17) had poor discriminatory power. Approximately 85% of participants indicated that items 4, 6, and 13, and 94% indicated items 3 and 17 did not apply to their current situation.

#### DISCUSSION

The main purpose of this study was to examine the reliability, validity, and factor structure of the FTQ. One- and three-factor solutions were assessed as providing a good fit for the data set. The single-factor solution accounted for approximately 30% of variance, compared with 55% of the variance for the three-factor solution. The internal consistency of the single-factor solution was excellent, whereas the internal consistency of the three-factor solution was variable, with all items of one sub-scale (*somatic*—items 3, 4, 6, 17) having poor internal consistency and discriminatory power (i.e., most participants indicated that these items did not apply to their current situation). There

were similarities though in the themes of the subscales identified in the Pain Anxiety Symptoms Scale (McCracken et al. 1992; Roelofs et al. 2004) and TSK (Roelofs et al. 2007). For example, the *somatic* subscale in the three-factor model of the FTQ has similar items to the *somatic focus* subscale in the TSK (Roelofs et al. 2007). Results indicated that the FTQ (17-items) total score gives stable results over a 2-week period (i.e., has good test-retest reliability) and contributes significantly to models predicting tinnitus-related distress as measured by the TQ and tinnitus-related impairment as measured by the TDI.

Assessing the convergent validity of the FTQ was, at the time of data collection, made difficult by the absence of alternative measures of tinnitus-related fear. In lieu of that, the IAS-Health Anxiety subscale was used to provide an indication of convergent validity. The moderate correlation might result from two key differences between the FTQ and IAS. Specifically, the FTQ aims to measure fear of a specific, present or imminent threat to health (i.e., tinnitus), whereas the IAS-Health Anxiety subscale assesses levels of worry regarding possible aversive future health concerns that are more general in nature, or potentially even not relevant to respondents. In contrast, strong correlations were found between the FTQ and the TQ, the TCS and the TDI. The TQ, which is a measure of global tinnitus-related distress, does not purport to measure tinnitus-related fear specifically. The strong correlation with the TQ could be due to many items on the FTQ relating to consequences of having tinnitus—albeit with a focus on being afraid of rather than actually experiencing them which is in accordance with the FA model. The strong correlation between the TCS and FTQ total scores is consistent with the FA model, which predicts that higher levels of catastrophizing results in higher levels of fear. The relatively weak correlations between the FTQ total score and measures of physical health and illness behavior are also logically consistent (although not explicitly predicted a priori) as a high level of fear of an outcome or condition does not necessarily mean that one already has poor physical health and that one acts in a sick role.

To our knowledge, the FTQ is the first questionnaire designed to solely measure tinnitus-related fear. The Tinnitus Fear Avoidance Cognitions and Behaviors Scale (T-FAS; Kleinstäuber et al. 2013) assesses tinnitus fear-related cognitions and avoidance behaviors. It includes a subscale-cognitions-that contains items that might have a strong conceptual overlap to those found on the FTQ. For example, both measures include items regarding the fear that tinnitus will adversely affect quality of life. In their study examining the properties of the T-FAS, Kleinstäuber and colleagues found, with a sample of participants seeking treatment, that the cognitions subscale was moderately (r = 0.57) correlated with the Tinnitus Handicap Inventory total score and with measures of anxiety (r = 0.46) and anxiety sensitivity (r = 0.45). The pattern of results reported here is consistent with these results even though the samples differ (that is, a web-based compared with treatment seeking sample).

#### Limitations

There are several potential limitations of the current study that need to be considered. First, the data were collected from questionnaires presented in a fixed-order online that might have attracted a specific sample of participants. In addition to there being potential-order effects and participants being self-selected, participants did not undergo any examination or assessment of hearing loss, tinnitus or verification of other characteristics and could have completed the questionnaires over a period of days even though they were instructed to complete it within a single a day. If respondents' tinnitus fluctuated over time, this might have influenced how they responded to the measures. While these issues might limit the degree to which the findings can be generalized to other samples, participants would have had little to gain by exaggerating or minimizing their symptoms or experience with tinnitus. The sample actually shares some similarities with that reported by Kleinstäuber et al. (2013) (e.g., the ratio of female participants, and the average age and education level of participants was nearly identical). The samples differ though on the proportion of participants reporting hearing loss (approximately 48% in this sample compared with 75%) and duration of living with tinnitus (5 compared with 8 years).

Although it is not necessarily a methodological limitation of this study, it is possible that the format of the response options (dichotomous) of the FTQ affected the measure's ability to distinguish between respondents with varying degrees of fear of tinnitus that in turn affects the measure's concurrent (predictive) validity. That is, a Likert-type scale offers respondents greater choice in how much fear, they might report for a specific item rather than simply indicating whether they experience any fear. Debate exists in the literature regarding the optimal number of response options to survey questions and the impact that less compared with more options has on results (Clark & Watson 1995). Some have argued that there are extensive problems (e.g., relating to accuracy of response choice and reliability) with dichotomous response options that affect validity and discriminating power (Comrey 1988; Preston & Colman 2000). Others have argued that reliability and test-retest reliability in particular are not affected by dichotomous data (Matell & Jacoby 1971; Preston & Colman 2000). In this case, the dichotomous response options did not affect test-retest reliability.

Given that approximately 90% of participants indicated that 5 of the 17 items were irrelevant to them, possibly reflects that participants already had a good level of knowledge about the etiology of tinnitus. This seems possible given that they had on average experienced tinnitus for over 5 years and were visiting websites where information about tinnitus was available. In addition, participants might suffer only a mild-to-moderate level of tinnitus fear. By way of comparison, after extensive CBT treatment, participants had an average FTQ total score of 4.20, 12 months after baseline measurements (Cima et al. 2012) compared with participants in this study who had an average FTQ score of 4.66. Similarly, Swedish patients in a recent online cohort study had a mean total FTQ score of 4.00 (Müller et al. 2016). Regardless, the FTQ total score has been shown to be a variable that has explanatory power in predicting/detecting treatment response in a clinical trial of stepped care specialized CBT for tinnitus (Cima et al. 2012).

Last, it is noted that we did not predict, a priori, the strength of association between measures and that there is a high level of intercorrelation between the FTQ and the TQ. This high level of intercorrelation is likely to be because there is, in part, conceptual overlap between the FTQ and TQ. The TQ includes several subscales (e.g., emotional distress, somatic complaints) which produce a global assessment of tinnitus distress severity. In line with this, a recent mediation analysis demonstrated how reductions in tinnitus-related fear (as measured with the FTQ) partially explain why specialized cognitive behavioral therapy (CBT) is effective in improving quality of life and reducing tinnitus-related distress (Cima et al. 2017).

## **Future Directions**

Given these issues with the FTQ, it might be informative for future research to examine the effect of changing response options to a five-point Likert-type scale on the factor structure and ability to contribute to models predicting tinnitus-related distress or impairment, respectively. (A study addressing this is currently in progress.) It would also be informative to examine

- the effect of the mode of completing the FTQ (e.g., online compared with by pen and paper) as there is conflicting research about whether it matters or not (Bowling 2005; van de Looij-Jansen & de Wilde 2008; Walt et al. 2008; Wang et al. 2013),
- 2. how the T-FAS and FTQ relate in samples of people with tinnitus seeking and not seeking treatment, respectively,
- validate the FTQ in different populations of people with tinnitus who might interpret their tinnitus differently from those in the sample recruited for this study, and
- 4. the reliability and validity of a 15-item version of the FTQ.

To assess face validity, it would also be of interest to examine what people with tinnitus think of the actual measure. Do they, for example, actually think in terms of being *afraid* or *fearful* of particular consequences or risks associated with their tinnitus and or hearing loss? In addition, examining and revising the wording of item 6 would be useful, because this item showed a rather weak factor loading and low relevance to participants' current situation. A qualitative approach similar to one used to examine the subscales of the Tampa Scale of Kinesiophobia (Bunzli et al. 2015) might yield valuable information in this regard.

Finally, future research could specifically examine the somatic subscale items to investigate their relevance for individuals with tinnitus considering the duration of the complaints. People with recent onset tinnitus might be relatively naive about it and find the items on the somatic subscale to be applicable. In contrast, those who have experienced tinnitus for a longer period may be reasonably well informed about the etiology and possible implications of tinnitus and thus are aware that most of the items are unlikely to be true. In addition, the effects of excluding item 6 from the FTQ based on its low factor loading on the psychometric properties need further study.

#### CONCLUSIONS

The results of this study indicate that the Dutch version of the FTQ has good psychometric properties and that for tinnitus research purposes the FTQ total score is a valid and informative outcome measure. The three-factor version might be useful for clinicians, because it provides specific information on fear-related thoughts that could subsequently form a part of the focus during treatment.

### ACKNOWLEDGMENTS

This research was supported by funding from SWOL Limburgs Fonds voor Revalidatie and Adelante, Centre for expertise in Rehabilitation and Audiology, R. F. F. Cima is funded through The Netherlands Organization

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for Scientific Research (NWO) through the Innovational Research Incentives Scheme: Veni (Project number 451-15-032). J. W. S. V. is supported by the "Asthenes" long-term structural funding–Methusalem grant (METH/15/011), funded by the Flemish Government, Belgium. The funders are not involved in any aspect of the project's protocol, collection, and analysis of data, and nor have they had input regarding the interpretation and dissemination of the study's results.

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Received January 26, 2018; accepted February 13, 2019.

The authors have no conflicts of interest to disclose.

T. E. F. analyzed data and wrote the article. R. F. F. C. designed the study, collected the data, and provided critical revision. E. V. d. B. provided statistical advice and critical revision. J. W. S. V. provided critical revisions and study oversight.

\* For further discussion of posterior predictive p values see Meng (1994) and Rubin (1984).

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