Psychologically informed vestibular rehabilitation for persistent dizziness

Herdman, David

Awarding institution:
King's College London

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David Herdman

Institute of Psychiatry, Psychology & Neuroscience
Health Psychology Section
King's College London

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Abstract

Vertigo or dizziness is not a disease, but rather a leading symptom of various underlying conditions. These include disorders of the vestibular system, which is responsible for our sense of balance and motion. Many people with vestibular system disorders experience persistent dizziness that can be particularly hard to treat. Vestibular rehabilitation therapy (VRT) is an established physiotherapy-based treatment for people with vestibular dysfunction, which tries to reduce dizziness and improve postural and gaze stability. However, the benefits are not universal and do not always correlate with physiological findings. Poor response to VRT may be because there are concomitant psychosocial factors contributing to the chronicity of the symptoms which are not addressed during VRT.

Previous studies have shown a correlation between dizziness and anxiety and depression. Studies of cognitive-behavioural therapy (CBT) for persistent dizziness have to date focused on reducing generalised anxiety to determine if this in turn relieves the dizziness symptoms. This appears to have limited or only short-term success. This may be because although anxiety and depression likely play a role in exacerbating symptoms, mood is only one factor in this multifactorial condition. Understanding a broader range of psychosocial factors specific to vestibular disorders may be needed to provide more tailored and targeted CBT. Combining this with VRT would provide an integrated approach to treating both physiological and psychological features of the disorder.

The central question of this thesis was to see whether we could design and evaluate the feasibility of an empirically derived theory-based 'CBT informed' vestibular rehabilitation intervention for people with persistent dizziness.

The project followed the Medical Research Council framework for developing and evaluating complex interventions. A systematic review of 89 studies using meta-analysis and narrative synthesis identified potentially modifiable psychosocial factors from existing research related to dizziness handicap and symptom severity. A longitudinal survey (n =185 pre diagnosis) was conducted to test the relationship between relevant psychological variables, clinical tests of vestibular deficits and dizziness handicap and subjective symptoms. The cross-sectional results showed that the psychological factors which included distress, negative illness perceptions, and unhelpful cognitive-behavioural responses to symptoms explained >50% of the variance in self-reported handicap and around 30% of the variance in symptom severity. Following diagnosis (n=135) an all-or-nothing erratic pattern of behaviour and experiencing symptoms for a longer time predicted higher dizziness handicap, although baseline 'handicap' was the strongest predictor. The results of the review and the survey, together with patient-public representation, informed the development of an

integrated manual-based programme of 'CBT informed' vestibular rehabilitation, called INVEST, combining cognitive behavioural therapy and physical rehabilitation.

A parallel group randomised controlled pilot-feasibility trial was then conducted, with 40 participants with persistent dizziness who were randomly assigned to receive 6 sessions of INVEST (n=20) or current 'gold standard' VRT (n=20). Participants were individually randomised using a minimisation procedure with allocation concealment. Both interventions were delivered by specialist physiotherapists. Primary feasibility and self-report outcomes were collected at baseline and 4 months post randomisation. A nested qualitative study was also conducted post-intervention to explore the acceptability of the intervention and identify any areas in need of improvement. This study demonstrated excellent acceptability and feasibility. The study met all the a-priori criteria to progress to a full-scale efficacy trial, including 80% of eligible patients participating (pre-defined criteria >70%), 15% therapy and 2.5% trial drop-out rates (criteria <20%), comparable acceptability ratings to current gold standard VRT, and 80% adherence to sessions (criteria >60%). Fifty-nine percent of patients screened met the selection criteria and the enrolment rate was 80%. According to the qualitative data and exploratory treatment effect sizes, the intervention appeared to be both acceptable and potentially beneficial.

This thesis improves our understanding of chronic vestibular symptoms. It provides invaluable information to inform a larger scale trial of an intervention that could potentially improve the quality of life of sufferers, above and beyond standard physiotherapy care. By delivering the intervention as part of physiotherapy, this in turn will improve the access to psychological therapies and use of available resources; reduce the need for patients to see more than one healthcare professional and allow clinicians to respond to patient preference. Improving the outcome of rehabilitation may have additional beneficial social and economic implications as the patient is able to better manage their condition. Given the high prevalence of persistent dizziness in audio-vestibular, neuro-otology, and VRT clinics there is a sufficient need, and number of patients, to run a fully powered RCT.

PhD Publications

Herdman, D., Norton, S., Pavlou, M., Murdin, L., & Moss-Morris, R. (2020). Vestibular deficits and psychological factors correlating to dizziness handicap and symptom severity. *Journal of Psychosomatic Research*, 132, 109969. DOI: https://doi.org/10.1016/j.jpsychores.2020.109969

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Frequently Used Abbreviations

ANOVA	Analysis of Variance
BPPV	Benign Paroxysmal Positional Vertigo
CBT	Cognitive Behavioural Therapy
CBRQ	Cognitive Behavioural Responses to symptoms Questionnaire
CNS	Central Nervous System
CSM	Common Sense Model
DHI	Dizziness Handicap Inventory
GAD-7	Generalised Anxiety Disorders – 7 item scale
IPQ-R	Illness Perceptions Questionnaire - Revised
LTCs	Long-Term Conditions
NHS	National Health Service
PHQ-9	Patient Health Questionnaire – 9 item scale
PHQ-ADS	Patient Health Questionnaire – Anxiety & Depression Scale
PPPD	Persistent Postural Perceptual Dizziness
RCT	Randomised Control Trial
SSRI	Selective Serotonin Reuptake Inhibitor
TFA	Theoretical Framework of Acceptability
VRT	Vestibular Rehabilitation (therapy)

Chapter 1

Introduction

1.1 Chapter Overview

This chapter will introduce important concepts and background concerning persistent dizziness in relation to vestibular disorders. Dizziness and other related vestibular symptoms are described, and vestibular symptom perception is conceptualised within a model of embodied predictive processing. The prevalence and impact of dizziness, and the clinical features of vestibular dysfunction will be discussed with reference to vestibular compensation. The clinical features of functional dizziness are covered along with a discussion of the limited literature on the available treatments including principles of vestibular rehabilitation and cognitive behavioural therapy.

1.2 What are vestibular symptoms?

The brain and the inner ear are in constant communication with the body and one another to achieve balance. This ability to control our balance effectively and maintain spatial awareness is intrinsically important in how we function in everyday life. The peripheral vestibular system consists of the labyrinth of the inner ear and vestibular nerve, and is responsible for estimating head movement and position in space (Hain, 2011). It is closely integrated with somatosensory and visual information so that the central nervous system (CNS) can maintain balance, spatial orientation, and ensure that visual input remains stable whilst we continually move and navigate our environment. It has been labelled as a 'sixth sense' (Goldberg, 2012) as it provides us with information that is essential to our ability to make sense of the world around us.

Vestibular dysfunction can produce a range of unpleasant and disabling symptoms such as disorientation, vertigo (an illusion of movement), unsteadiness, light-headedness, blurred vision (oscillopsia), and autonomic symptoms similar to motion sickness (Bisdorff et al., 2009). International symptom classifications define 'dizziness' as a sensation of distorted spatial orientation, separate to 'vertigo' which is the illusion of movement (Bisdorff et al., 2009). However, patients often use the term 'dizziness' as an all-encompassing and relatively non-specific term that can include any or all of the above symptoms. This thesis adopts the nomenclature of 'dizziness' in this broadest viewpoint to describe an altered perception of orientation and motion in space.

1.3 How do the symptoms come about?

Prior work on vestibular perception and disease has provided us with a fundamental understanding of the circuit-based mechanisms by which vestibular information is processed and generates vestibulo-motor reflexes (Bronstein et al., 2015). Traditionally a key assumption has been that symptom reports closely reflect some physiological dysfunction and that the latter directly causes the experience of symptoms.

This assumption seems to hold quite well when vestibular dysfunction generates rather intense and specific interoceptive sensations that are low in complexity, are clearly localised, and have clear on/off boundaries (Van den Bergh et al., 2017a). Examples of this can be seen during an acute vestibular loss (e.g., peripheral vestibulopathy, aka vestibular neuritis / labyrinthitis) or during an attack of Benign Paroxysmal Positional Vertigo (BPPV). However, for more systemic changes in the body and/or for persistent symptoms, the correspondence between symptoms and parameters indicating vestibular dysfunction is poor (Yip & Strupp, 2018) and only accurate under some conditions.

New developments in the conceptualisation of symptom perception could account for these differences. Within a framework of embodied predictive processing, dizziness is thought to arise because of a mismatch between the internal expectations (predictions) and actual incoming sensory information from the vestibular, visual, and somatosensory systems (Cullen, 2019; Klingner et al., 2016). The brain actively constructs an internal (generative) model of the world and the body reflecting previous experiences, optimised in such a way that sensory inputs are predictable. When this is violated, the mismatch can be used as an error signal to update its internal model, and the failure to encode the precision (the error bars) is experienced as vertigo/dizziness (See Figure. 1). From this perspective, symptoms serve a specific purpose since they also compel us to act in a way to reduce the prediction error (e.g., stop, change our gaze etc).

Many vestibular-related perceptual phenomena can only be understood by acknowledging that perception is not just a reflection of incoming information, but that it is also largely reliant on pre-existing (prior) information (Pezzulo et al., 2019). One only needs to consider the destabilising effect of stepping onto a broken escalator for one such example where a strong categorical 'prior' overrides actual incoming sensory data from your vision ('likelihood'). Sensory perception relies on predictions instead of constantly analysing every bit of sensory data, which would be very inefficient. The brain therefore constructs a meaningful percept based on our sensory experience.

1.4 The vestibular system is a network

Another important consideration is that the vestibular system is more widely distributed throughout the cerebral cortex than other sensory networks (Cullen, 2019). Although these vestibular cortical circuits are not fully understood, the parietoinsular

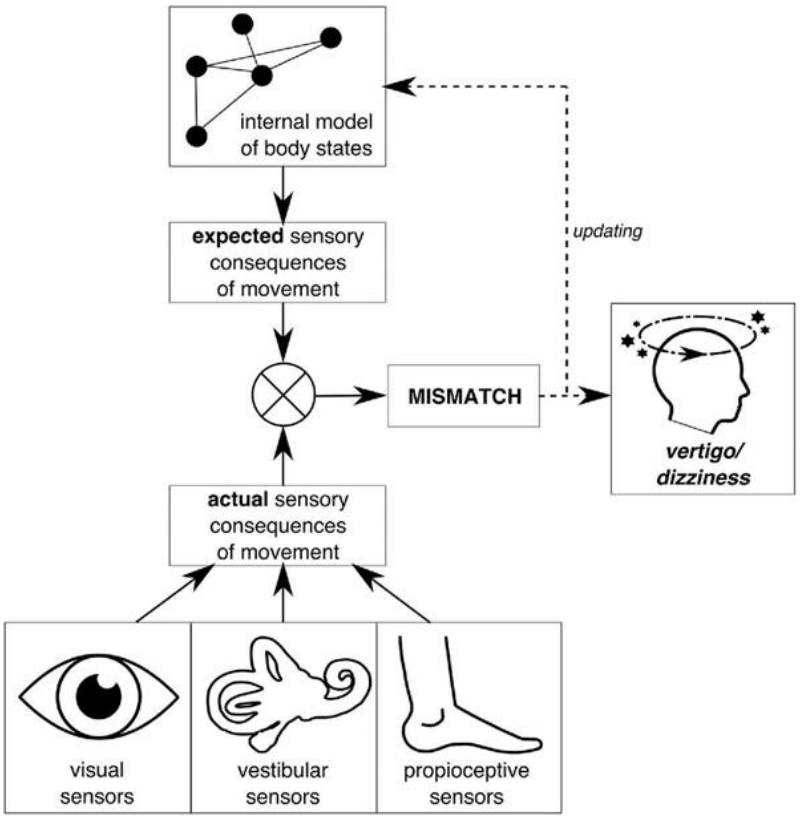

Figure 1. A simplified model of vestibular symptom perception. Used with permission from Schröder et al. (2021)

vestibular cortex (PIVC) is likely involved (Lopez & Blanke, 2011). However, it's existence as a primary vestibular cortex, in the way you can observe a 'visual' or 'auditory' cortical area, has been disputed (Zu Eulenburg et al., 2012). Instead, the 'vestibular network' can be conceived as a composition of spatially distributed brain areas that utilise information about head movements and position for their particular processing task (Klingner et al., 2016). In a system analogous to global air-travel, some areas such as the PIVC may act like 'hubs' for *multisensory* processing, whilst still allowing vestibular information to travel globally.

It is this necessary complexity that allows the balance system to act flexibly in a world full of constantly changing situations. Although not fully understood, the widely distributed vestibular system has the potential to influence multiple neurocognitive, affective, and other psychological functions in health and disease, which could in turn theoretically be improved by vestibular interventions (Cullen, 2019; Gurvich et al., 2013).

The vestibular system does not operate in a purely feedforward manner, however. Instead, the social, cognitive, behavioural, or affective context alters the stages of vestibular processing. For example, the vestibular nuclei can cancel out the earliest vestibular afferent information arising from active movements, comparing the actual sensory feedback with its own expected 'generative' internal model (Brooks & Cullen,

2019). It is therefore thought that salient 'unexpected' head motion is preferentially encoded to the next hierarchical level, most notably the PIVC and ventral intraparietal cortex (VIP), which are both involved in self-motion perception (Dale & Cullen, 2019). This explains why someone's behavioural context/function goal will alter vestibular processing, for example to differentiate between self or environmental motion (e.g., Did I move, or did something move me?) (Cullen & Wang, 2020).

1.5 Dizziness is common and troublesome

'Dizziness' is amongst the most common complaints across all medical settings (Bird et al., 1998; Mendel et al., 2010) and lifetime prevalence of significant dizziness amongst adult community populations may be as high as 30% (Murdin & Schilder, 2015). The one-year prevalence of dizziness amongst a working age community sample in the UK was 23%, with nearly half of those reporting some degree of handicap and 30% had been dizzy for more than 5 years (Yardley et al., 1998d). There is a marked female preponderance amongst people with vertigo/dizziness (Neuhauser et al., 2005). Prevalence of 'vestibular vertigo' increases with age in the general population (Neuhauser, 2007), although the mean age observed in UK neuro-otology clinics tends to be people in their 40's, 50's and 60's (Herdman et al., 2020c). This might reflect older adults who attend falls rather than dizziness services, and/or that many vestibular conditions actually have a peak onset in people middle-aged (Neuhauser, 2013).

Vestibular symptoms are not only common, but they also have profound personal and socioeconomic consequences. For example, age and sex-adjusted health related quality of life is lower in people with dizziness and vertigo compared with those without dizziness (Gopinath et al., 2009; Neuhauser et al., 2008). A large survey (n=4869) of the German general population found that out of the respondents with moderate to severe dizziness or vertigo (n=1003), 41% had taken time off work (Neuhauser et al., 2005). In another multi-country observational registry of 4,294 patients with vertigo, only half were employed (Benecke et al., 2013). Amongst this working patient population, 70% had reduced their workload, 63% had lost working days, 5% had changed and 6% had quit their jobs, because of their vestibular symptoms.

Use of healthcare services amongst patients with vestibular symptoms is high (Benecke et al., 2013), often resulting in costly and/or even unnecessary investigations (Gandolfi et al., 2015; MacDonald & Melhem, 1997). Patients with comorbid anxiety are at particularly high risk of reporting greater subjective impairment and healthcare utilization due to their dizziness including higher frequency of consultations and increased use of medication (Wiltink et al., 2009).

1.6 There are different vestibular syndromes

At the time of writing according to the Bárány Society initiative for the establishment of the International Classification of Vestibular Disorders (ICVD, 2022), there are

internationally accepted diagnostic criteria for Vestibular migraine, Meniere's Disease, Benign paroxysmal positional vertigo (BPPV), Vestibular paroxysmia, Persistent postural-perceptual dizziness, Bilateral vestibulopathy, Hemodynamic orthostatic dizziness/vertigo, Presbyvestibulopathy, Mal de Debarquement syndrome, Superior semi-circular canal dehiscence syndrome, and Vestibular Migraine of Childhood/Recurrent Vertigo of Childhood. There is also soon to be a consensus document on acute unilateral vestibulopathy/Vestibular Neuritis.

Most disorders can be differentiated based on their temporal characteristics, i.e., whether the symptoms appear as an acute onset lasting a day to a few weeks (e.g., acute unilateral vestibulopathy, brainstem or cerebellar stroke), episodic attacks (e.g., BPPV (< 1 min), vestibular paroxysmia (< 1 min), vestibular migraine (5 min to 72 h), Meniere's disease (20 min to 12 h) or persistent symptoms lasting more than 3 months (e.g., bilateral vestibulopathy, functional dizziness, neurodegenerative diseases (cerebellar vertigo, extrapyramidal disorders)) (Strupp et al., 2020).

Some vestibular disorders can also produce accompanying symptoms such as unilateral tinnitus, aural pressure, and hearing loss in the case of Meniere's disease, or headache in the case of Vestibular migraine. Specific triggering and modulating factors can also point towards a particular diagnosis, such as when symptoms are evoked by lying down or turning over in bed (e.g., BPPV), or changes in pressure or loud noises (e.g., semi-circular canal dehiscence).

1.7 Recovery isn't always straightforward

Patients who suffer a sudden loss of vestibular function (e.g., acute unilateral vestibulopathy/Vestibular Neuritis) experience a myriad of symptoms including vertigo, vomiting, and unsteadiness (Halmagyi et al., 2010). Since vestibular disorders create sensory conflict or mismatch in multisensory brain regions, they can also produce frightening out-of-body experiences (Lopez & Elziere, 2018). These symptoms are usually expected to recover within a few days or weeks by a process called 'vestibular compensation' (Curthoys & Halmagyi, 1999), where the altered pattern of neural activity is retuned, and they develop new sensory strategies (Curthoys, 2000; Macdougall & Curthoys, 2012). The restitution of function can be interpreted as the adaptation of one's own cerebral model through adaptation and habituation. It figures as such that this process requires repetitive exposure to dizziness-provoking movements and environments which result in error signals (Allum, 2012).

However, 30-50% of patients report persistent dizziness after one year (Godemann et al., 2005) and several years (Kammerlind et al., 2005) following vestibular neuritis despite the resolution of objective findings. This suggests incomplete compensation even after the remission of the causal neurotologic illness. Such 'functional' dizziness is the most common condition seen in specialist outpatient neurotology clinics (Strupp et al., 2020) and also accounts for half of the patients with vertigo or dizziness referred to

outpatient neurology clinics (Stone et al., 2010). Understanding this heterogeneity in recovery and why these people continue to experience symptoms is a particular challenge and can lead to frustration for clinicians and patients.

Historically vestibular classifications often assumed purely biological causes and mechanisms, whereas others assumed purely psychogenic. However even in prototypical vestibular diseases factors such as anxiety and depression are known to influence recurrence rates, treatment response, and somatic symptom reports that bear little relation to physiological disease indicators (Yip & Strupp, 2018).

Dizziness itself can be a frightening experience which can lead to panic and anxiety regardless of trait anxiety (Pollak et al., 2003). There is a tendency for rapid conditioning, generalization, and avoidance behaviour to develop. Almost half of patients with vestibular symptoms suffer from psychiatric comorbidity, and these patients show more severe psychosocial impairment (Lahmann et al., 2015). In an earlier study of people attending a specialist vestibular clinic, I found that over 30% fulfilled criteria for onward referral due to anxiety and/or depression (Herdman et al., 2020c), far higher than the prevalence in general medicine (10-16%) (King et al., 2008).

Fifty percent of patients develop clinically significant psychiatric disturbance within 3-5 years after an acute vestibular loss (Eagger et al., 1992). Similarly, patients with anxiety disorders often report vestibular symptoms and may even have abnormal testing (Jacob et al., 2009). This reciprocal relationship between vestibular symptoms and anxiety is also thought to maintain symptoms and interfere with normal compensation (Saman et al., 2012).

1.8 Dizziness, balance, and anxiety

There may be several integrated mechanisms concerning dizziness and balance disorders. Our sense of balance and ability to stay upright is dependent on both the physical and psychological aspects (Adkin & Carpenter, 2018). One hundred years ago, French psychologist and pharmacist Émile Coué (Coué, 1922) wrote of how someone could easily walk along a plank placed on the floor, but if that same plank was high up – either in reality or believed so in a hypnotic state – they would be unable to do so because of their fear of falling. Only in recent years has this exact experimental set up been adopted to confirm and understand such observations.

Previous work has focussed on automatic responses to dizziness and imbalance, leading authors to focus on models of classical conditioning, in which triggering events result in behavioural responses (e.g., stiffening posture) that do not revert to normal and lead to permanent symptoms (Brandt, 1996; Staab, 2012). This implies that the unconscious conditioning is more important than, or separate to, the conscious evaluation of dizziness.

However, ignoring the relationship between cognition and behaviour may lead to theoretical flaws and missed therapeutic opportunities. Ellmers et al. (2022) have demonstrated that threats to balance may influence behaviour through both automatic and conscious, fear driven adaptations. Modulating prior expectations of the difficulty of an upcoming balance task has also been shown to manipulate the subjective sense of stability and physical performance (Castro et al., 2022).

Likewise early work by Yardley's group showed that negative beliefs about dizziness were a cause, not a consequence, of restricted involvement in daily activities and were more predictive of chronicity than anxiety or depression alone (Yardley, 1994a; Yardley et al., 2001). There has been limited research since then, which we aim to build on. Chapter 5 will provide a larger systematic overview of the psychological factors related to persistent dizziness.

1.9 Functional neuro-vestibular disorders

The mind-body relationship is a hallmark of persistent physical symptoms and functional disorders since they do not fit the traditional biomedical disease model. The term 'functional dizziness' has emerged to explain a change in the mode of action, unconnected with any perceptible alteration of structure - although even this may be a potentially misleading dichotomy since structural, functional and psychiatric features often overlap and interact with one another (Dieterich et al., 2016). In 2017, the Barany society formally classified a functional neuro-vestibular disorder and called it 'persistent postural perceptual dizziness' (PPPD) (Staab et al., 2017). This unified common features of earlier terms such as 'chronic subjective dizziness', 'phobic postural vertigo', 'visual vertigo', and 'space and motion discomfort'. It also moved towards a more modern understanding of persistent physical symptoms and away from dichotomous 'psychogenic' explanations.

PPPD is characterized by persistent dizziness and/or unsteadiness on most days over a period of 3 months or longer, with the symptoms lasting for hours per day, but not necessarily the whole day (Staab et al., 2017). Patients tend to adopt unhelpful 'high-risk' (fear of falling) postural control strategies such as slowing gait speed and stiffening postural muscles (Dieterich et al., 2001; Krafczyk et al., 1999) and alter the relative weighting of multi-modal (i.e., vestibular, visual, proprioceptive, auditory) perceptual information for orientation (Cousins et al., 2014). The symptoms arise spontaneously but may be worsened by upright posture, active or passive body movements, or exposure to situations where visual information regarding orientation is misleading (Bronstein, 1995; Redfern et al., 2001), such as train stations or supermarkets (Pavlou et al., 2006).

As many as 45% of patients have an 'organic' vestibular disorder before the onset of symptoms, but the condition can also be triggered by other medical or psychiatric events which trigger dizziness. The fact that such an array of precipitants can result in

this stereotyped symptom cluster supports the possibility of a shared pathophysiological mechanism whereby functional disorders emerge and manifest because of 'perceptual dysregulation' in the central nervous system (Edwards et al., 2012; Henningsen et al., 2018; Pezzulo et al., 2019; Van den Bergh et al., 2017a). Within the framework of predictive coding, central processing of incoming sensory information is biased by a mismatch resulting from incorrect internal expectations (such as overly precise threat-related categorical priors) leading to symptom perception (Van den Bergh et al., 2020). In this way, vestibular dysfunction is neither needed or sufficient to explain persistent dizziness, and all symptoms achieve the same levels of absolute 'realness'.

There is emerging empirical validation of this hypothesis (Arshad et al., 2022; Lehnen et al., 2019; Schröder et al., 2021) and a move towards improving positive diagnosis of functional dizziness (Stone et al., 2020). Two recent reviews of neuroimaging studies using magnetic resonance imaging and single photon emission computed tomography in PPPD found consistent evidence for decreased brain structure, function, and connectivity among the areas involved in multisensory vestibular processing regions and spatial cognition (Im et al., 2021; Indovina et al., 2021). There was also increased function and connectivity in prefrontal and attentional/emotional regulatory areas and visual processing regions. These changes are thought to reflect maladaptive and compensatory mechanisms including a shift in multisensory integration to favour visual over vestibular inputs (i.e., visual dependence), and anxiety-related mechanisms (i.e., hypervigilance, stiffening) on postural control (Im et al., 2021; Indovina et al., 2021). The variations and inconsistencies across studies were thought to be accounted for by triggering factors (e.g., peripheral vestibulopathy), personality (e.g., neuroticism) and psychological factors (e.g., anxiety and depression) as they seem to notably modulate brain functional activity and connectivity patterns (Im et al., 2021).

People with PPPD are likely to be the most disabled and distressed by their symptoms as opposed to other kinds of dizziness (Graham et al., 2021). It usually takes a long time to receive a diagnosis and they have a particularly poor prognosis with standard treatment (Popkirov et al., 2018a). Although individual treatments have been recommended, there is a lack of prospective, randomised controlled trials (Popkirov et al., 2018a).

1.10 Medication for chronic dizziness

Since vestibular compensation needs the stimulus of the sensory mismatch to occur, anti-vertiginous drugs may be counterproductive because most are vestibular sedatives. Therefore, vestibular suppressants should only be used for symptomatic relief during acute attacks otherwise they risk prolonging recovery (Strupp et al., 2011). They are not indicated in patients suffering from chronic dizziness or positional vertigo and can produce unwanted effects such as extrapyramidal syndromes. Effective long-term pharmacological treatments do exist for some vestibular disorders, most notably there are anti-Meniere's, and anti-migrainous medications. Although most treatment studies

have focused on reducing the attacks of vertigo (Strupp et al., 2011), rather than persistent physical symptoms.

Some patients with chronic dizziness may benefit from a selective serotonin reuptake inhibitor (SSRI) or other antidepressants, regardless of the presence of comorbid depression (Staab et al., 2002). However, there are only a few small case series and no prospective, randomised placebo-controlled trials (Popkirov et al., 2018b). The mechanisms by which SSRI may help are not understood and the role of serotonin is complex but may influence learning rate and increase underlying neural plasticity to better deal with (potential) loss and aversion (Iigaya et al., 2018). However, just like many people with functional neurological disorders, people with PPPD may be especially sensitive to medication, and 1 in 5 patients in research trials drop-out due to side-effects (Popkirov et al., 2018b).

1.11 Vestibular rehabilitation

There is now widespread consensus that vestibular rehabilitation (VRT), an exercise-based treatment, is the most effective method for improving symptoms of dizziness, postural, gait and gaze instability and nausea due to vestibular dysfunction. It is usually delivered by physiotherapists and consists of graded head, body and eye movement exercises that are performed both in the therapy setting and at home, providing the repeated exposure to movement necessary to achieve neurological adaptation (Hain, 2011; Lacour, 2006; Lacour & Bernard-Demanze, 2014). Goals of VRT typically include reducing vestibular and balance related impairments, improving function, reducing the severity and frequency of symptoms, and enhancing daily activity levels in a wide variety of environmental contexts (Whitney & Rossi, 2000; Whitney et al., 2020). This approach is at least twice as effective in achieving these aims compared with rest and standard medical care (Yardley et al., 1998a). The most recent Cochrane systematic review states that 'there is moderate to strong evidence that [VRT] is safe, effective management for…vestibular dysfunction' (McDonnell & Hillier, 2015).

However, not everybody benefits from VRT (Clendaniel, 2010; Krebs et al., 2003), and outcomes can be negatively affected by psychological factors such as anxiety, depression, and fear of movement (Whitney et al., 2020). For patients with unilateral vestibular deficits, around 25% of subjects do not improve at all, depending on which outcome measures are assessed, and most patients still report 'bothersome' symptoms at discharge even in world renowned therapy centres (Herdman et al., 2012).

Most patients attending vestibular rehabilitation departments have experienced vestibular symptoms for years persisting well beyond the acute compensation period, and many clinical trials of VRT have probably included patients with PPPD unwittingly (Staab, 2011). There is some evidence that the 'habituation' form of VRT can be beneficial specifically for PPPD based on one retrospective study (Thompson et al., 2015) and another small clinical trial (Nada et al., 2019), although the patients who

did not benefit from VRT had a longer duration of PPPD, more complex aggravating factors and a higher DHI score than those who benefitted.

There is also evidence for exercises for specific components of PPPD, such as visually induced dizziness (Pavlou et al., 2013). However, some of the mechanisms by which VRT is thought to bring about change, such as improved vestibular reflexes, have not been supported in relation to chronic dizziness (Millar et al., 2020). Developing theory-based interventions and identifying the reasons why some patients do not respond to rehabilitation is therefore imperative to improve the current situation.

1.12 Cognitive-behavioural therapy for dizziness

The frequent occurrence of psychological symptoms in patients with vestibular disorders has prompted several authors to recommend cognitive behavioural therapy (CBT), which originated as treatment for emotional disorders, in particular depression and anxiety (Beck, 1976). In recent years, it has been increasingly used for LTCs to manage symptoms such as pain (Williams et al., 2020) and fatigue (Moss-Morris et al., 2021). CBT is concerned with how a person thinks (cognition), how they feel (emotion) and what they do (behaviour) in the context of the human body and symptoms (physiology). Change in any one of these domains may bring about changes in another (Beck, 1991).

There are many implicit similarities between vestibular rehabilitation and psychological therapies such as CBT (Staab, 2011). Shared principles include promoting habituation (through repetitive movement), exposure to provocative movements and environments, cognitive reframing and challenging negative beliefs, and enhancing self-efficacy in previously feared situations. As psychological factors appear to make a significant contribution to patient recovery, the benefits of exercise therapy might be enhanced by augmenting these implicit psychological elements.

The strongest evidence in support of CBT for PPPD comes from Edelman et al. (2012) who developed a brief-CBT treatment delivered by a psychotherapist over three weekly sessions. They based the treatment on a model used for panic and anxiety disorders but did include specific psychoeducation and modified techniques of attention allocation, behavioural experiments, and interoceptive exposure with cognitive distancing for functional dizziness (referred to as 'chronic subjective dizziness' in the article, one of the precursors to PPPD). This did bring about large changes in disability, but curiously not anxiety or depression, compared to a waiting-list control group. In addition to the lack of an active control group, the participants had a relatively low average Dizziness Handicap Inventory (DHI) at baseline compared to what would be expected in most tertiary vestibular clinics in the UK, and around 25% did not show a reliable improvement following treatment. Nevertheless, treatment gains were maintained at one- and six months post treatment, and higher levels of DHI at follow up were mainly predicted by higher levels of pre-treatment anxiety (Mahoney

et al., 2013). The potential benefit of CBT for PPPD has also been suggested in retrospective chart reviews (Waterston et al., 2021).

Holmberg et al. (2006) completed a parallel group study comparing 16 participants treated with CBT to 15 participants treated with VRT. Whilst the VRT included habituation exercises, it was not customised and would not represent current best practice. Conversely, they did not attempt to standardise the CBT treatment, and rather chose to base it on analysis of each individual case. As such it was highly dependent on the counselling ability of one person. They found relatively weak CBT effects on dizziness related interference, anxiety, and depression but they did not include a measure of postural control. Moreover, all the effects were lost at 1 year follow up and the test results were similar to those obtained before treatment (Holmberg et al., 2007).

Studies combining VRT and explicit CBT have thus far mainly been small scale trials that have yielded mixed results. Johansson et al. (2001) compared a waiting list control group (n=10) with nine patients receiving 5 weekly group sessions over 7 weeks of combined VRT and CBT. The CBT components were added to promote relaxation, reduce anxiety, and avoidance of feared situations and movements. Improvement in dizziness interference as measured by the Dizziness Handicap Inventory (DHI) was observed, although this was not associated with changes in postural control, anxiety or depression. Unfortunately, the exclusion of an active control group does not allow for direct comparison between components of the intervention.

Andersson et al. (2006) demonstrated moderate effect sizes for measures of dizziness-related handicap in 29 participants with combined CBT and VRT. The CBT components were designed to reduce anxiety and avoidance behaviour and promote relaxation. There was a lack of significant change in general measures of distress and the authors suggested that the CBT components of the study should be expanded accordingly. The study was small and potentially statistically underpowered and they acknowledge several shortcomings such as the lack of long-term follow up, independent assessor, physiotherapy involvement or active control group.

More recently Kristiansen et al. (2019) conducted a small feasibility trial of integrated CBT-VRT delivered by a physiotherapist as a group intervention with eight (2 hour) weekly sessions. However, not many completed all the sessions and on close inspection of the data only one of the original seven recruited participants achieved a reliable change according to the DHI, suggesting that an intensive group-based intervention may not be feasible. Schmid et al. (2018) also delivered a group-based CBT-VRT intervention but found that DHI did not improve in the group with abnormal balance control.

One of the 'third wave' cognitive-behavioural therapies called 'Acceptance and Commitment Therapy' (ACT) has also been combined with VRT and shown

promising results for a reduction in disability in a small pilot study of people with PPPD when conducted by a psychiatrist and psychologist (Kuwabara et al., 2020). ACT shares many similar features to traditional CBT, such as cognitive distancing (aka cognitive diffusion) but places a greater emphasis on developing psychological flexibility (a combination of acceptance, awareness, and behaviour change processes) alongside mindfulness meditation practice.

Multimodal inpatient treatment programmes for PPPD have also started to emerge (Axer et al., 2020; Limburg et al., 2019), but these have the potential to be costly and difficult to replicate in the National Health Service (NHS).

One reason why CBT or integrated CBT-VRT has not been widely adopted, or shown to have mixed results in clinical trials, may be because the CBT interventions were often poorly described, and designed with no theoretical basis of why they might work, choosing to focus on models of anxiety and depression rather than illness beliefs and related behaviours. Furthermore, most of the CBT in previous trials was delivered by psychotherapists. In clinical practice, when treatments are delivered separately many patients turn down psychological therapy as it intuitively may not make sense to them as they present with physical symptoms and/or it may be seen as 'delegitimizing' (Parsons et al., 2012). Likewise, many studies excluded patients based on the presence of abnormal vestibular testing, which limits access to therapies that patients may benefit from.

Allied healthcare professionals with additional training have demonstrated effectiveness in delivering psychological therapies integrated with rehabilitation for other LTCs (Brunner et al., 2013). There is also evidence from a qualitative study of vestibular physiotherapists in the UK who believed that managing anxiety related to dizziness was within their scope of practice, although they acknowledged a lack of training and guidance on this as a barrier (Walker et al., 2018). Although there is a lack of published evidence for the application and effectiveness of such psychologically informed VRT, it may still prove beneficial to explicitly maximize the psychological benefits of rehabilitation drawing from a broader biopsychosocial approach.

Chapter 2

Methods

2.1 Chapter Overview

The details of the methodologies for each of the objectives are contained in the relevant chapters. This chapter will introduce the process model used to guide the steps and stages of the project, as well as providing a broader overview of the research. I will discuss the key questionnaires that were used with reference to relevant validation work done as part of this project. The chapter concludes with an overview of the remaining chapters in this thesis.

2.2 Aims and objectives

The overall aim of the project was to design and evaluate a new cognitive behavioural intervention that could be incorporated with, and had the potential to improve the outcomes of, vestibular rehabilitation for people with persistent dizziness.

This project had the following objectives:

1. To determine the relationships between cognitive, behavioural, and emotional factors to dizziness-related disability (handicap) and severity pre-diagnosis (chapter 3 [published manuscript]) and at three months follow up (chapter 4 [published manuscript]).
2. To identify what psychological factors have also been researched in the literature and their relationship to dizziness-related disability and severity (chapter 5 [manuscript under review]).
3. To use the data collected in 1 and 2 to conceptualise chronic vestibular symptoms within a cognitive-behavioural model outlining potentially modifiable psychosocial factors (chapter 5, part of the systematic review).
4. To use this treatment model to design a new integrated cognitive behavioural therapy and vestibular rehabilitation intervention (the INVEST intervention) for patients with persistent dizziness based on the previous work (chapter 6 and chapter 7 [published manuscript]).
5. To evaluate the feasibility and potential efficacy of INVEST and test multiple methodological components and clinical outcomes simultaneously to inform a fully powered randomised controlled trial (chapter 8 [accepted manuscript, in press] and chapter 9).

2.3 Developing complex interventions

The project followed the UK Medical Research Council's (MRC) guidelines for developing and evaluating complex interventions (Skivington et al., 2021). This describes the four phases of developing, feasibility/piloting, evaluation, and implementation (see Figure 2). The phases are iterative and not mutually exclusive since they all must address a common set of core elements – considering context (how does the intervention interact with its context?), developing and refining programme theory (what is the underpinning programme theory?), engaging stakeholders (how can diverse stakeholder perspectives be included in the research?), identifying key uncertainties (what are the key uncertainties?), refining the intervention (how can the intervention be refined?), and economic considerations (what are the comparative resource and outcome consequences of the intervention?).

The idea originated out of my own clinical experience as a vestibular physiotherapist, together with discussions with Professor Rona Moss-Morris. In the conceptualisation of the project, we involved stakeholders including funders, and the Meniere's Society, to prioritise the research question and expected outcomes, and methods. A patient-participant involvement (PPI) group comprised a diverse group of volunteers with lived experience of chronic dizziness recruited from advertisements in the Meniere's Society magazine, online, and posters in the vestibular clinic at Guy's Hospital. The size and composition of the group fluctuated throughout the project ranging between 6 and 12 people. PPI activities were wide ranging and central to the project, including evaluating outcome measures and study materials, the co-development of the intervention and contents of a treatment manual. At various critical stages we held focus group meetings, email, and telephone exchanges. We also included qualitative feedback of patients who had undergone the intervention.

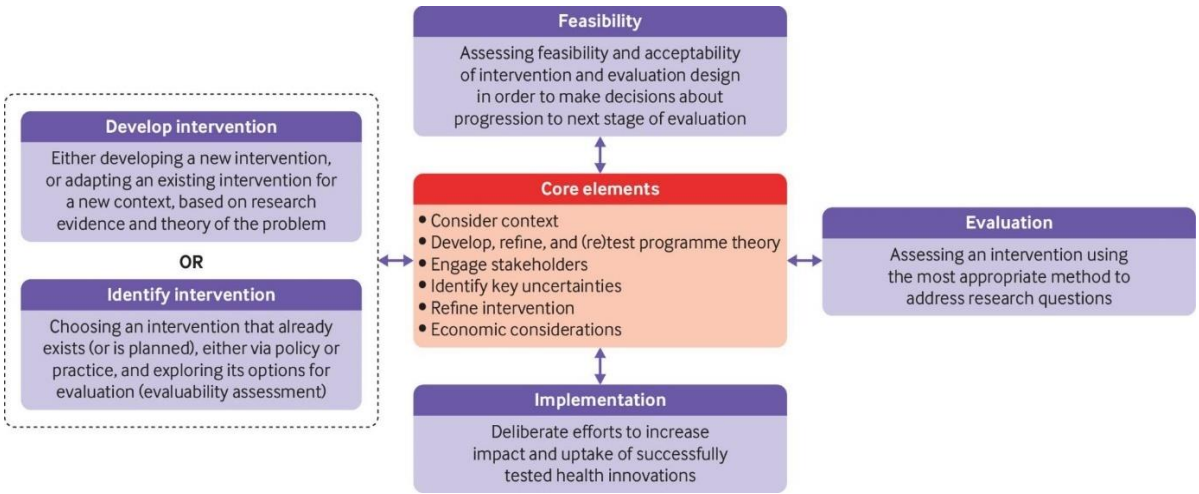

Figure 2. Medical Research Council (MRC) framework for developing and evaluating complex interventions (Skivington et al., 2021)

2.4 Self-report questionnaires

This project utilised many self-report measures to assess the impact of dizziness and the psychological aspects. These are described in their respective chapters, but this section will provide further background on four key measures which were used throughout, with reference to related theoretical models and validation work. All the questionnaires can be found in Appendix A.

Dizziness Handicap Inventory

The Dizziness Handicap Inventory (DHI) (Jacobson & Newman, 1990) was used as a key primary outcome in the empirical chapters. It consists of 25 questions designed to assess the impact of dizziness on everyday life. It is the most widely used instrument to measure dizziness-related disability (Fong et al., 2015) hence the terminology 'handicap' is used for consistency with the literature despite the negative connotations. There are three subgroups representing functional, emotional, and physical aspects of dizziness and unsteadiness. However, subsequent work has not supported the independence of these categories and so it is advisable to use the total score (Koppelaar-van Eijsden et al., 2022; Van De Wyngaerde et al., 2019). The DHI total score is scored out of 100 and can indicate mild (<30), moderate (30-60) and severe (>60) handicap and functional impairment (Whitney et al., 2004). Although the available evidence for a few measurement properties is limited, there is evidence to support good construct validity, responsiveness, and reliability (Koppelaar-van Eijsden et al., 2022; Mutlu & Serbetcioglu, 2013; Tamber et al., 2009). It has been widely adopted in clinical practice and research to evaluate the effects of treatment across conditions (Mutlu & Serbetcioglu, 2013).

Some studies have already found that the DHI score does not correlate significantly with objective clinical tests of vestibular function (Jacobson & Calder, 2000; Jacobson & Newman, 1990; Jacobson et al., 1991; Yip & Strupp, 2018), and its ability to discriminate between vestibular disorders is variable (Mutlu & Serbetcioglu, 2013). Stewart et al. (2018) concluded that this lack of relationship with vestibular system parameters reduces the credibility of the DHI, however, this is misguided since it was not originally designed for this purpose and may better reflect limitations in current vestibular tests. Instead, it adds important and unique pieces of information regarding the personal impact of dizziness (Mutlu & Serbetcioglu, 2013).

Vertigo Symptom Scale

The Vertigo Symptom Scale (VSS) (Yardley et al., 1992a) is a measure of the frequency and severity of dizziness symptoms over the last 12 months. The original questionnaire consists of 34 items and contains two main subscales measuring vertigo-balance (VSS-VER) and autonomic-anxiety (VSS-AA) symptoms. The shortened version of the VSS (VSS-SF) (Yardley et al., 2004b) was used in the cross-sectional study as a measure of symptom severity, consisting of 15 items. Each item is scored on

a 5-point scale (range 0–4), and a measure of symptom severity is obtained by summing the item scores. The total scale score ranges 0–60, higher scores indicating more severe problems. Severe dizziness has been defined as ≥ 12 points on the total scale (Yardley et al., 2004a). The scale also comprises the two subscales relating to vertigo-balance (score ranging 0-32) and autonomic-anxiety symptoms (score ranging 0-28) (Yardley et al., 1998b). However, only the vertigo-balance subscale was used since the autonomic-anxiety symptoms scale is designed to measure symptoms suggestive of anxiety and was therefore removed. It was also felt on review that the autonomic scale contained some items, such 'headache, or feeling of pressure in the head' that might reflect known vestibular disorders such as vestibular migraine, which were not formally recognised at the time the measure was developed. The VSS-SF has shown satisfactory internal consistency (Soderman et al., 2001) and moderate test-retest reliability (Yardley et al., 1998a). However, information on other psychometric properties is limited.

Measures of the key hypothesised perpetuating mechanisms of chronic dizziness

Illness Perceptions Questionnaire

The importance of illness cognitions is highlighted throughout this thesis. Social cognition is the study of how people think about and make sense of themselves and others. Under this broad umbrella, health psychologists have studied people's cognitions about health and illness to understand and predict whether people engage in health and illness-related behaviours. One such model that has emerged is Leventhal's Common Sense Model (CSM) of self-regulation, which describes the dimensions along which lay people make sense of illness, and how these dimensions influence the adoption of behaviours to reduce threats to health (Leventhal et al., 2016). The CSM describes two parallel but linked emotional and cognitive pathways that together influence behaviour, and health related outcomes (Figure 3). People have both cognitive and emotional representations of their illness which drive behavioural and

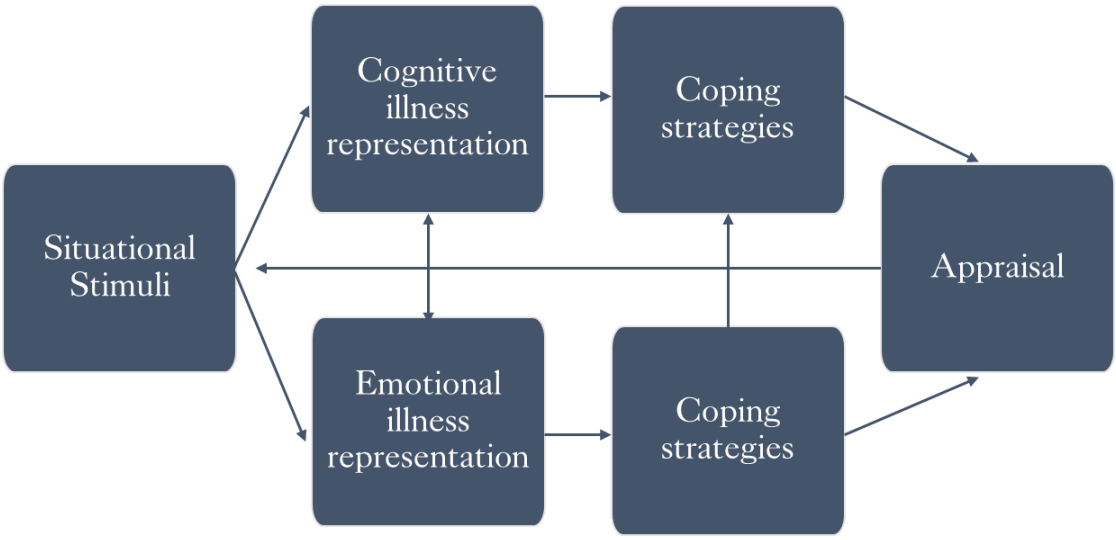

Figure 3. Common-sense self-regulation model (Leventhal et al., 1980)

coping responses. Appraisal of the effectiveness of these responses can further influences their illness cognitions and emotions in a feedback process.

The Illness Perceptions Questionnaire (Weinman et al., 2012) is a self-report measure for assessing the five dimensions of illness cognitions defined in the CSM. This comprises *identity* – the symptoms the patient associates with the illness; *cause* – personal ideas about the cause of illness; *time-line* – the perceived duration of the illness; *consequences* – effects of the illness on quality of life in general and on functional and social aspects of daily life; and *cure/control* – how one controls or recovers from the illness. Patients are required to indicate the degree to which they agree or disagree with statements using a five-point Likert scale (strongly disagree, disagree, neither agree nor disagree, disagree, strongly agree).

The questionnaire was updated to the Illness Perceptions Questionnaire-Revised (IPQ-R) scale by Moss-Morris et al. (2002). This is the version used in this thesis since it addresses minor psychometric problems by splitting cure/control into two dimensions – *treatment* control and *personal* control and adds additional subscales assessing *cyclical timeline* - beliefs about the recurrent or unpredictable nature the condition, *illness coherence* – the degree to which they understand and make sense of their condition, and emotional *representation* – perceptions of distress associated with the condition. Higher scores indicate more negative beliefs, except for the control and coherence subscales where high scores indicate positive perceptions. Scores for the identity scale indicate how many symptoms they attribute to their condition. The tool is widely used and shown to be a good predictor of ongoing physical symptoms in a variety of conditions as well as a mediator of change in severity of symptoms in CBT based symptom interventions (Hagger et al., 2017).

We also used the Brief-Illness Perceptions Questionnaire (B-IPQ) (Broadbent et al., 2006) for the final trial, which includes just eight single-item scales; one to assess each

dimension of consequences, timeline, identity, personal control, treatment control, emotional response, concern, and coherence. Although the brevity of this version is useful to reduce participant burden, it has been criticised for using single items to define constructs (Broadbent et al., 2015). To address this, some studies have used a total negative illness perception score by first cross checking the internal consistency of the total scale (Chilcot & Moss-Morris, 2013; Knoop et al., 2012). We followed a similar procedure.

The original authors recommend these measures be customised for the specific medical problem by replacing the word 'illness' with the specific diagnostic label. Since people with vestibular disorders can have many different conditions and disease labels, we replaced the word 'illness' with 'dizziness' to capture negative dizziness perceptions. For scoring, some items are reverse scored. The authors also recommend that the illness identity scale is modified to make it more relevant. As part of the early patient-public representative meetings, we asked them to freely recall any physical symptoms they may have experienced since the onset of their condition and added these to the identity scale if it did not already appear.

Cognitive-Behavioural Responses to Symptoms Questionnaire

Whilst these overarching illness beliefs are clearly important in understanding individual variations in the impact of an illness, qualitative studies suggest that day-to-day interpretations of symptoms also appear to be particularly important in determining coping behaviours which may enhance the experience of vestibular disorder (Fridberg & Gustavsson, 2019).

Other measures have largely focussed on conditions such as chronic pain to identify specific cognitive behavioural responses such as catastrophising and fear avoidance. The Cognitive and Behavioural Responses Questionnaire (CBRQ) (Moss-Morris & Chalder, 2003) was therefore developed to capture a unique and broad range of cognitive interpretations of symptoms, symptom focusing, and associated behaviours across different long-term conditions (LTCs).

The CBRQ contains four cognitive subscales: *fear avoidance beliefs* (i.e., that activity is harmful for symptoms), *embarrassment avoidance beliefs* (the need to avoid activities due to the potential for symptoms to cause embarrassment), *symptom focusing* (the need to pay careful attention to, and monitor symptoms), and *catastrophising* (negative and inflated beliefs in anticipation of dizziness), and two behavioural subscales; *avoidance of activity* (excessive resting or avoiding activity when experiencing symptoms) and *all-or-nothing* behaviour (where people push themselves to get things done when symptoms allow and then crash). Preliminary analysis indicated that fear avoidance could be further split into two subscales, as was adopted in this thesis, one pertaining to the affective interpretation of symptoms (*fear avoidance beliefs*), but also another

subscale capturing an interpretation of symptoms as signalling *damage* to the body. The result is a 40-item scale measured on a 5-point Likert scale.

Although the CBRQ has been used in other studies (Ali et al., 2017; Artom et al., 2017; Chilcot et al., 2016; Skerrett & Moss-Morris, 2006) and has been validated in chronic fatigue syndrome (Loades et al., 2020), its original development, and psychometric properties as a transdiagnostic measure across different illness groups have not been reported. In a separate paper by our group currently under review (Picariello et al., 2022), data from this cohort went towards evaluating the psychometric properties of the CBRQ across different LTCs. Confirmatory factor analysis revealed that the 6-or 7-factor structure had appropriate fit, with satisfactory internal reliability, good construct validity and sensitivity to change. A shortened CBRQ displayed good psychometric quality so may be an option for future research, although somewhat reduced reliability for some subscales.

Patient Health Questionnaire – Anxiety & Depression Scale

The Patient Health Questionnaire-9 (PHQ-9)(Spitzer et al., 1999) and Generalised Anxiety Depression Scale-7 (GAD-7)(Spitzer et al., 2006) are well validated instruments for depression and anxiety respectively across LTCs, are freely available for use and acceptable to people with dizziness (Herdman et al., 2020c). There is some evidence that these scales can be combined in LTCs resulting in the Patient Health Questionnaire – Anxiety and Depression Scale (PHQ-ADS) (Chilcot et al., 2018; Kroenke et al., 2016).

There are several potential advantages of a single anxiety-depression score in research and clinical practice, stemming from the common co-occurrence of depressive, anxiety, and somatic symptoms. Once again, we conducted secondary data analysis from this cohort of people with vertigo and dizziness to investigate the factor structure, internal consistency, and construct validity of the PHQ-ADS for this population (Herdman et al., 2022). This analysis supported the structural validity of the PHQ-ADS and suggested that a total score appropriately captured distress severity as measured by anxiety-depression. It does not undermine the utility or value of screening for depression or anxiety separately either, since the two-factor model for the PHQ-9 and GAD-7 also had good fit in our analysis and their respective characteristics have already been well established. This is an advantage over scales such as the Hospital Anxiety and Depression Scale which has been found to have inconsistent factor structure in people with dizziness (Piker et al., 2015).

2.5 Methods within this thesis

Systematic review

A systematic review of empirical studies investigating the relationship(s) between psychological factors and dizziness handicap and symptom severity was conducted

following PRISMA guidelines. The emphasis was on drawing on the research evidence base, allowing the creation of a preliminary theoretical model to link psychological factors that may perpetuate dizziness handicap and severity of symptoms - identify potential mechanisms and new treatment targets.

A meta-analysis was conducted to determine the strength of the relationship between dizziness outcomes, anxiety, and depression, but a detailed narrative synthesis allowed a variety of theories and a broad range of relevant literature to also be considered.

Cross-sectional and prospective methods

The identification of causal and contextual factors affecting the relevant outcome is a key step in intervention development. The initial systematic review and expert panel identified that anxiety and depression was likely to be relevant, but that other areas such as illness and symptom beliefs, beliefs about emotions and perfectionism were not represented in the literature. A cross-sectional and prospective study explored the relative contribution of demographic variables, standardised vestibular function testing, and self-reported modifiable psychological factors in explaining the variance in dizziness severity and handicap. Uniquely we recruited patients with vertigo and dizziness who were still on the waiting list to attend a specialist dizziness clinic and again three months after their initial consultation. Hence it was possible to record psychological factors such as illness beliefs before and after diagnosis.

Intervention development

There are a variety of approaches to intervention development. We formally combined existing approaches to intervention development as described by Araujo-Soares et al. (2019) and O'Cathain et al. (2019). A total of 18 iterative actions are identified, displayed in seven domains of intervention development: Conception and Planning, Designing and Creating, Refining, Documenting and Planning for Future Evaluation. To address the 'designing' and 'creating' domain we employed intervention mapping techniques against key therapeutic targets identified from the theoretical model, revising this in accordance with the findings from the cross-sectional and prospective studies, and systematic review. We used a large multidisciplinary team to maximise idea generation and innovation. We also worked with professional stakeholders and PPI to offer solutions and features of the intervention.

Feasibility

The purpose of the feasibility study was to assess predefined progression criteria for a full-scale efficacy trial. The value of feasibility testing is now widely accepted with key guidelines and terms well defined (Thabane et al., 2016). This addressed uncertainties around recruitment, data collection, retention, outcomes, and analysis. It also addressed uncertainties around the intervention itself including the content and delivery.

Since we identified that the content of the intervention may influence the acceptability, we used the Theoretical Framework of Acceptability (TFA) (Sekhon et al., 2017) to guide the evaluation. It defines acceptability as 'a multi-faceted construct that reflects the extent to which people delivering or receiving a healthcare intervention consider it to be appropriate, based on anticipated or experienced cognitive and emotional responses to the intervention' (Sekhon et al., 2017: p.4). The TFA consists of seven component constructs which we measured using quantitative and qualitative methods: affective attitude, burden, perceived effectiveness, ethicality, intervention coherence, opportunity costs, and self-efficacy.

A decision was taken to test the intervention against current gold standard vestibular rehabilitation (Hall et al., 2016). The stems from the view that it is impractical and illogical to construct a placebo therapy which would contain the same characteristics as the treatment for which it serves as a control (Kirsch et al., 2016).

Qualitative methods were embedded to ask participants who received INVEST about their experiences of the intervention to further assess acceptability of the intervention and identify any issues needing revision.

2.6 Thesis overview

This thesis incorporates publications,[1] with additional chapters such as this one to explain methodology, intervention development, and additional qualitative data. A systematic review was conducted initially which informed the selection of psychological measures for the prospective questionnaire study. This is not, however, presented first since this was updated for publication after the publication of those studies. The review is therefore presented afterwards since it includes reference to those studies and proposes the model around which the development of the intervention occurred. The development and components of the intervention are then described, before presenting the protocol and results of the randomised trial used to determine its acceptability and feasibility. The thesis concludes with a final overall discussion.

[1] The format of all published articles has been changed to meet the format of the thesis and all citations merged to appear in the final references section.

Chapter 3 [published manuscript]

Vestibular deficits and psychological factors correlating to dizziness handicap and symptom severity

Authors: David Herdman, Sam Norton, Marousa Pavlou, Louisa Murdin, Rona Moss-Morris

3.1 Abstract

Objective: To determine the relative contribution of demographic variables, objective testing and psychological factors in explaining the variance in dizziness severity and handicap.

Methods: One-hundred and eighty-five consecutive patients on the waiting list to attend a diagnostic appointment in a tertiary neuro-otology clinic with a primary complaint of vertigo or dizziness completed a cross-sectional survey. Primary outcomes were the Dizziness Handicap Inventory and the vertigo subscale of the Vertigo Symptom Scale-Short Form. Psychological questionnaires assessed anxiety and depressive symptoms, illness perceptions, cognitive and behavioural responses to symptoms, beliefs about emotions and psychological vulnerability. Patients also underwent standardised audio-vestibular investigations and tests to reach a diagnosis at appointment.

Results: Objective disease characteristics were not associated with handicap and only the presence of vestibular dysfunction on one test (caloric) was associated with symptom severity. Almost all the psychological factors were correlated with dizziness outcomes. The total hierarchical regression model explained 63% of the variance in dizziness handicap, and 53% was explained by the psychological variables. The regression model for symptom severity explained 36% of the variance, and 30% was explained by the psychological factors. In adjusted models, factors associated with dizziness handicap included age, female gender, distress, symptom focusing, embarrassment, avoidance, and beliefs about negative consequences. Fear avoidance was the only independent correlate in the fully adjusted model of symptom severity.

Conclusion: Self-reported dizziness severity and handicap are not correlated with clinical tests of vestibular

deficits but are associated with psychological factors including anxiety, depression, illness perceptions, cognitive and behavioural responses.

3.2 Introduction

Acute vestibular (inner ear) dysfunction is characterised by vertigo, nausea, vomiting, imbalance and deficits in gaze stabilisation and body representation (Lopez, 2016). Recovery occurs due to central and peripheral compensation that relies on many different neurobiological plastic events, including sensory and behavioural substitutions (Lacour & Bernard-Demanze, 2014; Lacour et al., 2016). Typically, symptoms are expected to last days or weeks but around 50% of patients continue to experience chronic and disabling symptoms of dizziness and balance disturbance often despite resolution of the causal illness (Best et al., 2009; Cousins et al., 2017; Godemann et al., 2005; Heinrichs et al., 2007; Kammerlind et al., 2005).

The functional consequences of a vestibular condition and the degree of compensation can vary among individuals. Except in the case of new acute presentations, objectively measured vestibular deficits show little relationship to vestibular symptoms or handicap (Patel et al., 2016; Yip & Strupp, 2018). Longitudinal studies also show that vestibular testing cannot identify patients who do or do not get better following acute illness (Best et al., 2009; Cousins et al., 2017; Godemann et al., 2005; Okinaka et al., 1993; Palla et al., 2008). Despite this, current clinical practice emphasises the use of vestibular testing and test results to determine the causes and treatment of symptoms.

Less attention is paid to addressing a wider range of psychosocial factors that may be important in determining symptom severity and handicap, especially in patients with symptoms of more than a few weeks' duration. Understanding psychological factors in the perpetuation of symptoms fits with the neurobiological account of vestibular compensation as a process of habituation and relearning that involves multisensory brain regions reliant on behavioural exposure (Lacour & Bernard-Demanze, 2014; Lacour et al., 2016). Exercise therapy that encourages the necessary repeated exposure to movement may benefit from techniques that build on the many implicit psychological elements (Staab, 2011). However, there are currently few studies exploring the relative contributions of psychological factors, beyond anxiety and depression (Cousins et al., 2017; Kirby & Yardley, 2009a; Probst et al., 2017; Yardley et al., 1994).

Although anxiety and depression are consistently correlated with vestibular symptom severity and handicap, other psychological factors such as illness beliefs and behaviours may play a more direct and specific role. For instance, it is common for patients to restrict their head movement to lessen symptoms, although such avoidance behaviours are likely to limit compensation resulting in a more protracted time course. Patients with dizziness frequently endorse negative beliefs about the consequences of dizziness leading to avoidance in a range of physical and social activities (Mendel et al., 1997;

Yardley, 1994a; Yardley et al., 1992b). One longitudinal study found that negative beliefs were a significant predictor of handicap after controlling for symptom severity (Yardley et al., 2001). Fear of bodily sensations and cognitions about these symptoms may also play a mediating role in the relationship between vestibular symptoms and psychopathology (Radziej et al., 2018).

Explanatory models of 'medically unexplained' (functional) vestibular disorders, mainly Persistent postural-perceptual dizziness (PPPD), also propose factors such as anxiety-driven hypervigilance (Popkirov et al., 2018a). Perfectionism and beliefs about expressing emotions also have implications for the development and maintenance of similar clinical problems (Brooks et al., 2017; Moss-Morris et al., 2011; Sibelli et al., 2018; Sibelli et al., 2017). Few studies have looked at whether psychological factors are equally relevant to both medically explained and functional vestibular disorders. New instruments now exist to measure such constructs relevant to persistent physical symptoms but have not been explored in patients with vestibular disorders.

Collectively, these lines of investigation posit roles for psychological variables as predisposing, precipitating, provoking, and perpetuating factors in patients with vestibular and balance disorders. If confirmed, then psychological factors would merit greater attention in diagnostic evaluations, treatment plans, mechanistic research, and healthcare systems design.

The primary objective of this study was to evaluate biopsychosocial factors associated with the variability of dizziness handicap and severity in patients with a range of vertigo and dizziness related disorders including those considered medically explained and unexplained. The following hypothesis were therefore tested:

1. Psychological distress, as measured by depression and anxiety symptoms, would be associated with greater dizziness severity and handicap

2. Negative illness perceptions, higher levels of symptom focussing, and unhelpful interpretation of symptoms (such as catastrophising, damage and fear avoidance beliefs) and greater use of all-or-nothing and avoidance/resting behaviours would all be associated with greater levels of dizziness severity and handicap.

3. Negative self-beliefs and beliefs about expressing emotions would also be associated with greater severity and handicap.

4. The psychosocial variables described in 1 to 3 would explain a greater proportion of the variance in patient-reported handicap or symptom severity than objectively measured vestibular deficits.

3.3 Methods

Study design and participants

Cross-sectional survey of consecutive patients on the waiting list to attend an initial diagnostic appointment in a tertiary multidisciplinary neuro-otology clinic due to symptoms of dizziness and vertigo between March and December 2018. Participants were contacted by phone and sent a patient information sheet and consent form along with the questionnaires and self-addressed envelopes. Consenting participants completed the survey prior to their initial consultation, either online or through the post. Participants were considered eligible providing they were still experiencing dizziness and were over the age of 18. Participants were excluded if they had a) insufficient mastery of the English language to allow them to complete the survey independently, b) a comorbid disease resulting in cognitive impairment, or c) acute severe mental health problems (e.g., psychosis). The study was approved by the NHS Health Research Authority (16/NI/0256).

Self-reported Questionnaires

Dizziness Handicap Inventory

The Dizziness Handicap Inventory (DHI) (Jacobson & Newman, 1990) was used to measure participants' dizziness related handicap. The 25-item self-report scale measures the extent dizziness interferes with physical, functional, and emotional aspects of one's life. Higher scores represent higher levels of dizziness handicap and activity restriction. The internal reliability in this study was excellent (Cronbach's α = 0.92).

Symptom severity

The vertigo subscale of the shortened version of the Vertigo Symptom Scale (VSS) (Yardley et al., 1998b; Yardley et al., 1992a) was used in this study to measure participants' dizziness severity. Cronbach's α was 0.86.

Psychological correlates

Depressive symptoms were assessed using the Patient Health Questionnaire-9 (PHQ-9) (Spitzer et al., 1999) which scores each of the 9 DSM-IV criteria from '0' (not at all) to '3' (nearly every day). Anxiety symptoms were measured using the Generalised Anxiety Disorder Questionnaire-7 (GAD-7) (Spitzer et al., 2006). Both questionnaires have been widely validated in physically ill populations and the internal reliability was excellent in this current study, with a Cronbach's α of 0.92 for anxiety and 0.88 for depression. These two scales can also be summed to give a composite measure of distress (PHQ-ADS) (Kroenke et al., 2016).

The Illness Perception Questionnaire-Revised (IPQ-R) (Moss-Morris et al., 2002) was used to measure participants' illness perceptions (beliefs). In this study, the word illness

was replaced with 'dizziness condition' and the illness identity scale was modified to make it more relevant to patients with vertigo and dizziness as per the authors' recommendations. The IPQ-R measures the key components in Leventhal's common sense self-regulatory model. Illness identity measures the number of symptoms out of a list of 23 that the individual ascribes to their illness. The following dimensions were measured on a five-point Likert scale (strongly disagree – strongly agree) in response to several statements for each item; Timeline (e.g., 'my dizziness is likely to be permanent rather than temporary'), Consequences (e.g. 'my dizziness has major consequences on my life'), Personal Control (e.g. 'I have the power to influence my dizziness'), Treatment Control (e.g. 'there is little that can be done to improve my dizziness'), Illness Coherence (e.g. 'my dizziness is a mystery to me'), Timeline Cyclical (e.g. 'my dizziness symptoms come and go in cycles'), and Emotional Representations (e.g. 'when I think about my dizziness condition I get upset'). The Causal Factors list was removed from the analysis since this was frequently misinterpreted. Cronbach's α for the included subscales ranged from 0.75 to 0.89 indicating good reliability.

The Cognitive-Behavioural Responses to Symptoms Questionnaire (CBRQ) (Skerrett & Moss-Morris, 2006) measured participants' cognitive and behavioural responses to symptoms. The five subscales dealing with cognitive responses are Symptom Focusing (e.g., 'I think a great deal about my symptoms'), Catastrophizing (e.g., 'I will never feel right again'), Damaging Beliefs (e.g., 'symptoms are a signal that I am damaging myself'), Fear Avoidance (e.g., 'I should avoid exercise when I have symptoms') and Embarrassment Avoidance (e.g., 'The embarrassing nature of my symptoms prevents me from doing things'). The two behavioural subscales are All or- Nothing (e.g., 'I find myself rushing to get things done before I crash') and Avoidance/Rest (e.g., 'I stay in bed to control my symptoms'). All items are rated on a 5-point Likert scale ranging from 'strongly disagree' to 'strongly agree'. The last question assesses Causal Attributions and asks patients to describe the nature of their symptoms on a 5-point scale ranging from 'my symptoms are physical' to 'my symptoms are psychological in nature'. Cronbach's α for the subscales ranged from 0.80 to 0.92.

The Psychological Vulnerability Scale (PVS) (Sinclair & Wallston, 1999) measures unhelpful beliefs about oneself. It screens for vulnerability related to perceptions of dependency, perfectionism, negative attributions, and the need for external sources of approval. High scores indicate more maladaptive thinking. The scale's internal reliability in this study was high (α = 0.82).

The Beliefs about Emotions Scale (BAE) (Rimes & Chalder, 2010) was used to assess unhelpful beliefs about emotions. It measures the extent to which the person holds beliefs that it is unacceptable to experience or express negative emotions. High scores indicate more unhelpful beliefs about emotions. The scale's internal reliability was high (α =0.72).

Demographic and clinical data

Demographic factors were measured through a self-report questionnaire and their medical records were accessed to record their diagnosis and vestibular test results.

Postural control

Physiotherapists completed a balance assessment at the initial appointment that included the mini–Balance Evaluation Systems Test (mini BESTest) (King & Horak, 2013), which has four sub-scales measuring different aspects of postural control and higher scores indicate better balance.

Neuro-Otological examination

Participants who attended their appointment in the neuro-otology clinic underwent a thorough standardised clinical history and examination including videonystagmography (VNG). Vestibular function was assessed using the video head impulse test (vHIT) and caloric irrigation. It was not mandatory for all patients to undergo all tests if the diagnosis was apparent on clinical examination (e.g., BPPV) or if there were other medical contra-indications, or if the patient declined the investigation. Findings for every patient were reviewed by the consultant Audiovestibular physician (LM) and patients underwent further testing (such an imaging or VEMPs) or had follow up examinations when required to reach a final diagnosis. The diagnosis was made based on consensus diagnostic criteria and commonly accepted definitions of the Barany Society (Bisdorff et al., 2015).

For vHIT testing, the Otometrics® system was used and all six canals were tested. The patient was instructed to fixate on a target on the wall at eye level approximately 1.5 m in front of them. Following calibration, the examiner made multiple unpredictable head movements over a range of velocities until the system had recorded twenty correct impulses in each direction. The average gain (ratio of the eye movement velocity to the head movement velocity) was calculated according to the software and the traces were checked for accuracy. Abnormal vHIT testing was determined by the presence of pathologic overt or covert corrective eye movements (saccades) and gain below the normative cut-off values.

Caloric testing was performed according to the guidelines from the British Society of Audiology (British Society of Audiology, 2010). The patient was lying reclined with the head at 30˚ and wearing video oculography goggles to prevent fixation. Binaural bithermal irrigations (30 & 44 ˚C) were performed and the resultant vestibular nystagmus was recorded to yield plots of the slow phase vestibular eye movements. The degree of canal paresis was calculated using the Jongkees formula. The caloric test was rated as abnormal when the difference in canal paresis was more than 20% or indicated bilateral vestibular hypofunction. These tests are considered the 'gold standard' since there are no other objective tests considered as sensitive or specific in the frequency

range they assess (Perez & Rama-Lopez, 2003; Zapala et al., 2008). The vHIT also has excellent test-retest reliability (Singh et al., 2019).

Sample size

A prior power calculation using G*Power version 3.1 indicated that a minimum sample size of n = 161 would be sufficient to detect a medium R2 effect size = 0.15, with a power of 0.80 at a two-tailed alpha level of 0.05. This included the variables drawn from the measures above with a multiple linear regression fixed model after including demographic and disease variables.

Statistical methods

When data were missing or unclear, participants were contacted to clarify their response. All data were analysed using SPSS version 25. Bivariate correlations, two-tailed t-tests and ANOVAS were used to examine the relationships between dizziness handicap and severity with the other variables. Two hierarchical regression analyses were conducted to determine whether the psychological variables under investigation predicted dizziness handicap and dizziness severity after controlling for relevant demographic and disease variables. Model improvement was evaluated using ΔF-statistic. Improvement in explained variance was calculated using ΔR2. Statistical significance level was assumed at $p < .05$.

3.4 Results

Participant characteristics

The flow of participants is shown in Figure 4. One hundred and eighty-five participants returned their questionnaires (a 39% response rate of eligible subjects). There was no significant difference in age (mean difference = −0.19, t = −0.12, p = .91) or gender ($x2 = 0.23$, p = .63) between responders and non-responders. Table 1 shows the demographic characteristics of the sample, which was predominantly female (74%) and white British (63%). The median duration of symptoms was 2 years (range 3 months to 32 years) and 32% of participants reported occupational disability directly as a result of dizziness.

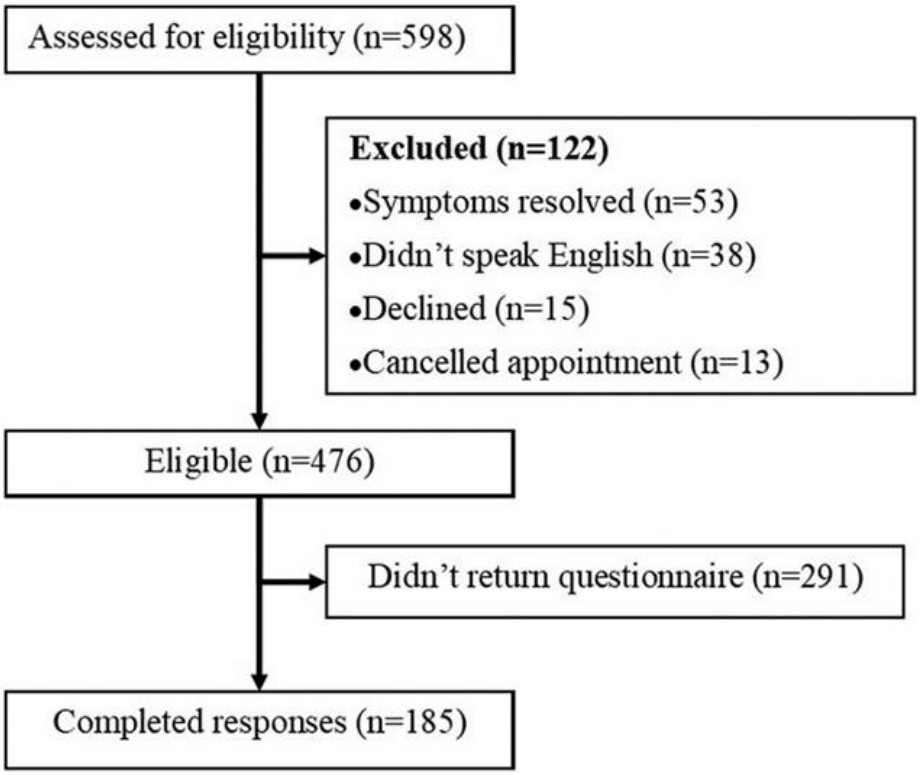

Figure 4. Flow diagram of participants

Table 1 also shows the primary diagnoses and the self-report categorical data for handicap, anxiety, and depression of the sample. Eighty-eight percent of participants met diagnostic criteria for a primary neuro-otological disorder. Eight participants did not attend their appointment and therefore did not receive a diagnosis. Using the standard cut-offs (Spitzer et al., 1999; Spitzer et al., 2006) for the PHQ-9 and GAD-7, 40% had at least moderate symptoms of depression and 28% had anxiety.

Table 1. Sample demographic and disease characteristics

Factor	Sample
Age, mean (SD, range)	53.57 (17.386, 18–90)
Gender female, n (%)	137 (74.1)
Ethnicity, n (%)	
White British	117 (63.2)
White, other	29 (15.7)
Caribbean	10 (5.4)
African	7 (3.8)
White, Irish	6 (3.2)
Asian, other	4 (2.2)
Chinese	2 (1.1)
Mixed, white & Asian	2 (1.1)
Mixed, other	1 (0.5)
Bangladeshi	1 (0.5)
Indian	1 (0.5)
Any other ethnicity	5 (2.7)
Relationship status, n (%)	
Married/civil partnership	69 (37.3)
Single	48 (25.9)
Living with partner	28 (15.1)
Divorced	19 (10.3)
Widowed	14 (7.6)
Separated	7 (3.8)
Highest educational attainment, n (%)	
Post graduate degree	28 (15.1)
University degree	48 (25.9)
Trade/apprenticeship	8 (4.3)
Certificate/diploma	24 (13)
A-levels (advanced higher equivalent)	14 (7.6)
GCSE (secondary/ high school equivalent)	35 (18.9)
No formal education	20 (10.8)
Other	8 (4.3)
Employment status, n (%)	
Retired	58 (31.4)
Employed, full time	55 (29.7)
Unemployed	26 (14.1)
Employed, part time	21 (11.4)
Student	6 (3.2)
Homemaker	5 (2.7)
Other	14 (7.6)
Reduced hours at work due to dizziness, n (%)	31 (16.8)
Unemployed or retired due to dizziness, n (%)	28 (15.1)
Disease characteristics	
Duration of dizziness in months, mean (SD)	48.33 (64.36), median 24
Currently taking medication for dizziness, n (%)	50 (27)

Primary Diagnosis, n (%)	
Unilateral peripheral vestibulopathy	45 (25.4)
Benign paroxysmal positional vertigo	37 (20.9)
Vestibular migraine	38 (21.5)
Persistent postural perceptual dizziness	11 (6.2)
Meniere's disease	9 (5.1)
Central	5 (2.8)
Bilateral vestibulopathy	5 (2.8)
Vestibular schwannoma	3 (1.7)
Semi-circular canal dehiscence	2 (1.1)
Other (unexplained, non-vestibular etc)	22 (12.4)
Dizziness Handicap Inventory, mean (SD)	
Mild handicap (0–30)	39 (21)
Moderate handicap (30–60)	88 (47.6)
Severe handicap (>60)	58 (31.4)
Anxiety (GAD-7), n (%)	
Mild (5–9)	61 (33)
Moderate (10–14)	20 (10.8)
Severe (≥15)	31 (16.8)
Depression (PHQ-9), n (%)	
Mild (5–9)	50 (27)
Moderate (10–14)	37 (20)
Moderately severe (15–19)	22 (11.9)
Severe (≥20)	15 (8.1)

Note: GAD-7 = Generalised Anxiety Disorder-7. PHQ-9 = Patient Health Questionnaire-9.

Demographic association with dizziness handicap and severity

In general, dizziness handicap and severity showed few associations with demographic variables included in Table 1. Younger participants had higher VSS scores ($r = -0.24$, $p < .01$) and females had significantly higher DHI score (mean difference = 10.327, $t = 2.76$, $p < .01$). Greater DHI also showed a small correlation with longer symptom duration ($r = 0.18$, $p = .02$). The only other significant difference detected was between employment status for handicap ($F(6,178) = 2.99$, $p < .01$) and VSS scores ($F(6, 178) = 2.41$, $p = .03$); post hoc tests showed that this difference existed due to higher scores in unemployed participants.

Objective vestibular testing

Forty-four percent (n = 78) of the 176 participants who completed either the caloric or vHIT tests had a relevant vestibular abnormality. Ten percent (n = 15) of the 149 vHIT results and 38.8% (n = 50) of the 129 caloric results were abnormal. The DHI showed no difference in the scores for normal versus abnormal vestibular function parameters (Caloric canal paresis, vHIT abnormality, any other relevant abnormality throughout examination, all $p > .05$). The only difference for VSS severity was that patients with a canal paresis had significantly higher vertigo symptom score; $t(127) = -2.06$, $p = .40$, mean difference – 2.7, 95% CI -5.274 to −0.125.

There was no difference in handicap between patients with (M = 49.55, SD = 22.64) and without (M = 41.81, SD = 23.52) a detectable central/peripheral vestibular disorder; t (175) =1.49, p = .14. The DHI did not differ significantly between ten identified disease groups either. The same lack of association was observed for symptom severity.

Balance control

One hundred and sixty-one participants (87%) completed the miniBESTest. The distribution was negatively skewed (M 23.3, SD 5.64) and 47 (25%) participants achieved the maximum score, indicating normal balance. There was a small to moderate correlation between the DHI and the miniBESTest ($r_s(159) = -0.34$, $p < .01$) and each of its subscales (r_s − 0.16 to −0.32). However, the miniBESTest was not associated with the vertigo symptom scale except for the dynamic gait subscale ($r_s(159) = -0.18$, $p = .025$).

Psychological correlates of dizziness handicap and dizziness severity

Univariate correlates between psychological factors and the DHI and VSS are shown in Table 2, accompanied by descriptive statistics. A full list of intercorrelations can be found in the supplementary material. Dizziness handicap and symptom severity correlated significantly with one another and with all the other psychological variables, except illness beliefs concerning personal control, illness coherence and cyclical timeline and people's beliefs about expressing emotions.

On the CBRQ causal attribution subscale, nobody attributed their symptoms to a 'psychological' cause. The causal attribution scale was not associated with the VSS, although there was a significant difference in handicap across the other categories (F (3, 168) = 5.54, p < .01), and post-hoc analyses showed that participants who recognised 'both physical and psychological factors' reported higher levels of handicap compared to participants who described their symptoms as 'physical' in nature.

Table 2. Correlations between the psychological and outcome variables (dizziness handicap and severity).

	Mean (S.D.)	DHI	VSS-V
Dizziness handicap (DHI)	48.90 (22.733)	–	0.618**
Dizziness severity (VSS-V)	13.87 (7.526)	0.618**	–
Psychological distress			
Depression (PHQ-9)	8.70 (6.597)	0.651**	0.512**
Anxiety (GAD-7)	7.02 (6.054)	0.576**	0.411**
Cognitive-behavioural factors (CBRQ)			
Symptom focusing	19.84 (5.725)	0.418**	0.284**
Catastrophising	11.63 (3.874)	0.542**	0.415**
Damaging beliefs	15.46 (4.015)	0.388**	0.227**
Fear-avoidance	18.94 (4.803)	0.427**	0.463**
Embarrassment-avoidance	16.19 (6.333)	0.586**	0.444**
All-or-nothing behaviour	12.02 (5.185)	0.443**	0.272**
Avoidance/resting behaviour	19.40 (7.188)	0.633**	0.433**
Illness Perceptions (IPQ-R)			
Identity	10.28 (5.382)	0.427**	0.339**
Acute-chronic timeline	18.99 (4.689)	0.345**	0.282**
Consequences	19.31 (5.389)	0.598**	0.443**
Personal control	17.11 (4.358)	−0.054	0.001
Treatment control	16.07 (3.367)	−0.179*	−0.201**
Illness coherence	11.66 (4.595)	0.037	0.004
Cyclical timeline	14.07 (3.564)	0.066	−0.039
Emotional representation	20.82 (5.723)	0.493**	0.286**
Beliefs About Emotions (BAE)	18.42 (7.311)	0.011	0.011
Psychological Vulnerability (PVS)	13.48 (5.919)	0.332**	0.218**

Note: DHI = Dizziness Handicap Inventory. VSS-V = Vertigo Symptom Scale –Vertigo Subscale. PHQ-9 = Patient Health Questionnaire-9. GAD-7 = Generalised Anxiety Disorders-7. CBRQ = Cognitive Behavioural Responses to Symptoms Questionnaire. IPQ-R = Illness Perceptions Questionnaire – Revised. BAE = Beliefs About Emotions. PVS = Psychological Vulnerability Scale.

∗ p < .05. ∗∗ p < .01.

Multivariate regression: factors associated with dizziness handicap

The regression model for dizziness handicap is shown in Table 3. The first step of the model controlled for age, gender and other demographic characteristics that showed univariate associations with handicap (duration and unemployment). Together, these explained 10% of the variance in handicap. For the second stage we created a dummy variable to control for the presence of abnormal vestibular function on either caloric or vHIT testing, although this did not add significantly to the variance explained.

Psychological factors significantly associated with dizziness handicap, with a correlation of ≥0.2 were then included in the hierarchical regression model. The emotional representation variable of the IPQ-R was also excluded from the analyses as it overlaps with depressive symptoms (PHQ-9). Due to collinearity the PHQ-ADS was used as a composite measure of anxiety and depression. Adding these psychological factors significantly improved the model (F change = 19.225, p < .01), explaining a further 53.3% of the variance in handicap. The fully adjusted model explained 62.7% of the variance in dizziness handicap. Female gender remained significant at each stage,

although unemployment was no longer significant after controlling for psychological variables. In the final model, distress, symptom focusing, embarrassment, avoidance behaviour and beliefs about the negative consequences of the condition contributed significant independent variance.

Due to missing data for the balance measure (miniBESTest), a separate hierarchical regression analysis was conducted including patients who had normal and abnormal balance on the miniBESTest as a dummy variable in an additional step although this did not significantly add to the variance explained or change the overall conclusions.

Table 3. Hierarchical regression model for dizziness handicap

Variable	Step 1			Step 2			Step 3		
	B	*SE B*	*β*	*B*	*SE B*	*β*	*B*	*SE B*	*β*
Age	−0.030	0.096	−0.023	−0.039	0.097	−0.030	0.154	0.072	0.117*
Duration	6.609	3.583	0.134	6.993	3.616	0.142	1.192	2.508	0.024
Unemployed	15.975	4.834	0.244**	15.689	4.851	0.240**	0.993	3.386	0.015
Vestibular dysfunction				2.776	3.335	0.060	0.573	2.227	0.012
Distress (PHQ-ADS)							0.565	0.143	0.286**
Physical attribution							1.201	2.576	0.026
Symptom focusing							−0.657	0.292	−0.166*
Catastrophising							0.425	0.508	0.072
Damaging beliefs							0.214	0.386	0.038
Fear-avoidance							0.350	0.298	0.073
Embarrassment-avoidance							0.675	0.245	0.188**
All-or-nothing behaviour							0.294	0.250	0.066
Avoidance/resting							0.758	0.228	0.237**
Identity							0.409	0.230	0.098
Chronic timeline							0.277	0.266	0.057
Negative consequences							0.578	0.292	0.137*
Psychological vulnerability							0.053	0.232	0.014
R^2, (Adjusted R^2)	0.129 (0.109)			0.133 (0.107)			0.665 (0.627)		
F for change in R^2	6.353**			0.693			19.225**		

Note: PHQ-ADS = Patient Health Questionnaire Anxiety and Depression Scale.

∗ p ≤.05. ∗∗ p < .01.

Multivariate regression: factors associated with dizziness severity

The same methods were used to perform a hierarchical regression using the vertigo subscale of the VSS as the dependent variable (Table 4). As with handicap, age and gender were entered together with unemployment, which was the only other associated demographic variable, explaining 11% of the variance. Since caloric paresis was associated with the VSS univariately, this was entered into the second step although

only explained a further 3.5% of the variance. The same selection criteria used for handicap was applied to the relevant psychological factors in the final step which again significantly improved the model (F change = 4.591, p < .01), explaining a further 30% of the variance in symptom severity. The fully adjusted model explained 35.8% of the variance in symptom severity. After including the psychological variables, demographics and caloric paresis were no longer significant.

Table 4. Hierarchical model for symptom severity (vertigo symptom scale-vertigo).

Variable	Step 1			Step 2			Step 3		
	B	*SE B*	*β*	*B*	*SE B*	*β*	*B*	*SE B*	*β*
Age	−0.088	0.036	−0.208*	−0.088	0.036	−0.208*	−0.064	0.035	−0.151
Gender	2.218	1.404	0.135	2.521	1.389	0.153	−0.017	1.298	−0.001
Unemployed	3.832	1.719	0.191*	3.657	1.693	0.182*	−0.620	1.653	−0.031
Caloric paresis				2.791	1.247	0.187*	1.940	1.194	0.130
Distress (PHQ-ADS)							0.112	0.069	0.172
Symptom focusing							−0.262	0.148	−0.192
Catastrophising							0.374	0.257	0.195
Damaging beliefs							−0.124	0.188	−0.069
Fear-avoidance							0.375	0.153	0.240*
Embarrassment-avoidance							0.203	0.118	0.176
All-or-nothing behaviour							0.075	0.122	0.054
Avoidance-resting							0.029	0.110	0.029
Identity							0.023	0.112	0.017
Chronic timeline							−0.037	0.131	−0.024
Negative consequences							0.258	0.146	0.184
Treatment control							−0.097	0.174	−0.043
Psychological vulnerability							−0.155	0.117	−0.120
R^2, (Adjusted R^2)	0.109 (0.088)			0.144 (0.116)			0.443 (0.358)		
F for change in R^2	5.122**			5.014*			4.591**		

Note: PHQ-ADS = Patient Health Questionnaire Anxiety and Depression Scale.

** p ≤.05. ** p < .01.*

3.5 Discussion

Results from this study found that general and dizziness-specific psychological factors were most strongly associated with dizziness handicap and severity in a representative sample of patients due to attend a neuro-otology clinic. The psychological factors were significantly correlated above and beyond objective vestibular deficits or diagnosis. The final model accounted for 63% of the variance in dizziness handicap and 36% of the variance in dizziness severity scores. In the fully adjusted models dizziness handicap was associated with age, gender, distress, symptom focusing, embarrassment, avoidance behaviours, and beliefs about negative consequences. Fear avoidance was

the only factor in the fully adjusted analysis found to be uniquely correlated with dizziness/vertigo severity.

Vestibular deficits were not associated with either dizziness outcome, except for the presence of an abnormal caloric test that showed a small association with symptom severity. The lack of correlation between dizziness handicap and standard vestibular tests is consistent with clinical experience (Bronstein et al., 2010b) and prospective studies (Yip & Strupp, 2018). This does not, however, mean that physiological variables are not important since vestibular compensation is a multi-modal and multi-faceted process (Brandt et al., 1997; Lacour et al., 2016), but that the degree of perceived disability cannot be explained solely by the presence of underlying structural vestibular deficits. Instead, a combination of disrupted visuo-vestibular perception and psychological characteristics is at play. Several functional and structural neuroimaging changes have been identified in patients with chronic dizziness, which reflect and underlie these psychophysiological and psychological features (Lacour et al., 2016).

Worse balance, as measured by the miniBESTest, was associated with higher levels of handicap, although only dynamic gait was slightly correlated with symptom severity. This supports the need to address balance dysfunction during rehabilitation as patients with chronic dizziness often adopt pathological postural strategies characterised by en-bloc movements in an effort to minimise symptoms of dizziness and fear of falling (Best et al., 2015) and cognitive-behavioural therapy alone may not adequately address such balance deficits (Schmid et al., 2018).

Greater psychological distress was significantly associated with both dizziness handicap and severity and, together with the other psychological variables, contributed the most to the variance explained within the adjusted models. This link may exist for a variety of reasons such as the close anatomic connections in the central nervous system (Balaban & Thayer, 2001), less likelihood to engage in behaviours that promote adaptation or greater likelihood to engage in avoidance behaviours (Yardley, 1994b), as well as enhanced perception and increased attention to somatic symptoms (Yardley & Redfern, 2001).

Although the role of psychological factors has been researched in functional vestibular and medically unexplained syndromes, the results of the current study suggest these factors are relevant to disorders regardless of whether they are classified as medically explained or unexplained (functional). This provides support for the DSM-5 (Diagnostic and Statistical Manual of Mental Disorders 5th ed.) concept of somatic symptom disorder ("Somatic Symptom and Related Disorders," 2013), a diagnosis that does not differentiate whether physical symptoms have a medical explanation. The results do give rise to the question in what way dysfunctional cognitions and fears contribute to dizziness. The model of Persistent postural perceptual dizziness (PPPD) already stresses the role of attentional strategies and integrates predisposing and precipitating aetiological factors with perpetuating factors (Popkirov et al., 2018a). Our

results suggest that those mechanisms might also be relevant to the perpetuation of dizziness severity in the presence of 'organic' disease.

Regarding illness perceptions, participants who reported higher levels of handicap and symptoms attributed more somatic symptoms to their condition and had stronger beliefs that their illness would last a long time, have serious consequences, be untreatable and more distressing. Although the nature of this relationship is likely bidirectional, there is evidence that negative illness beliefs and heightened attention to the body increases somatic symptom reports (Pennebaker, 2012) and are associated with maladaptive coping styles (Hagger et al., 2017).

All of the hypothesised dizziness-specific symptom interpretations and behavioural responses were associated with both handicap and symptom severity in the univariate analysis. These results support qualitative research that indicates that vertigo and dizziness are often viewed as intrinsically frightening and potentially stigmatising [18]. Results are also in line with Pothier et al. (2018) who reported a positive correlation between catastrophising and handicap across diagnostic classifications. The current study confirmed that adopting avoidance behaviours in response to symptoms was associated with subjective dizziness. It is also the first study to show that all-or-nothing approach, where people push themselves to keep going until they crash, was also associated with subjective dizziness and handicap. Establishing stable patterns of activity before following a graded exercise programme may therefore be helpful.

Individuals with medically unexplained syndromes who favour a physical disease explanation for their symptoms can experience greater disability (Sharpe et al., 1992). In this sample, however, participants with higher dizziness handicap acknowledged both physical and psychological factors. This does warrant further clarification since it is also possible that participants with high levels of symptoms were simply acknowledging psychological distress as a natural consequence of an undiagnosed and untreated structural disease. However, it does suggest that patients are open to more complex causal models of their illness which likely include an interaction between biological and psychological factors.

Finally, the psychological vulnerability scale was associated with higher handicap and symptoms. The scale reflects cognitive beliefs that are thought to make individuals more fragile under stress conditions (e.g., dependence, perfectionism, and need to be approved by external sources). Those patients may be less protected when faced with the negative experiences of having dizziness that affects their ability to live up to certain high standards. Conversely, negative beliefs about emotions had a near zero correlation with the dizziness outcomes. This could be because the scores typically remained 'neutral' when responses to the individual statements on the BAE were examined, suggesting that negative beliefs about emotions were either not very prevalent or that participants were not willing to disclose.

These results support the argument to expand the make-up and training of healthcare teams that evaluate patients with vestibular and balance problems. Rarely are they constructed to include personnel or resources to identify and treat psychological contributions to patients' morbidity. The results point to the potential benefits of expanding rehabilitation protocols to include interventions that modify symptom-related beliefs and behavioural responses via cognitive behavioural therapy.

Strengths and limitations

This study investigated how illness perceptions and symptom-specific fears and beliefs, and behavioural responses are associated with dizziness handicap and symptom severity, together with objectively measured vestibular function, in a large sample prior to diagnostic evaluation.

However, the cross-sectional data presented in this study does not allow inferring conclusions regarding the direction or causality. Future longitudinal research could examine changes in these constructs following diagnosis and intervention.

The validity of the psychological measures used in this study have been established in a range of samples with other medically explained and unexplained disorders. Although not previously scrutinised within this population, Cronbach's α values for each of the measures in the current study were in the good to excellent range, suggesting the measures have good internal reliability within this cohort. Finally, despite a relatively large sample there was not sufficient power to detect significance of individual factors within the fully adjusted model.

Despite the limitations, this study suggests that efforts to treat chronic dizziness from a purely biomedical perspective, regardless of cause, may be insufficient, and that rehabilitation protocols should consider interventions to modify specific symptoms-related beliefs and fears.

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Declaration of Competing Interest

None.

Supplementary data

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Chapter 4 [published manuscript]

The Role of Prediagnosis Audiovestibular Dysfunction Versus Distress, Illness-Related Cognitions, and Behaviours in Predicted Ongoing Dizziness Handicap

Authors: David Herdman, Sam Norton, Marousa Pavlou, Louisa Murdin, and Rona Moss-Morris

4.1 Abstract

Objective: People with chronic vestibular diseases experience variable degrees of self-perceived disability. However, longitudinal data examining the predictive validity of relevant clinical variables alongside psychological variables are limited. The present study examined whether these factors predict self-reported dizziness handicap 3 months after assessment and diagnosis.

Methods: Patients were recruited from a waiting list of a tertiary neuro-otology clinic and completed standardized mood, cognitive, behavioural, and dizziness handicap questionnaires before and 3 months after their initial consultation and diagnosis. All patients were clinically assessed and underwent comprehensive audiovestibular investigations.

Results: Seventy-three percent of participants responded at follow-up (n = 135, 73% female, mean [standard deviation] age = 54.23 [17.53] years), of whom 88% were diagnosed with a neurotological condition. There were significant improvements in handicap, depression, and anxiety at 3 months. Thirty (22%) of 135 showed clinically meaningful improvement in handicap. The percentage of case-level depression and anxiety remained the same. Negative illness perceptions and symptom responses reduced, although participants still tended to view their condition negatively. Vestibular tests and type of diagnosis were not associated with self-reported handicap. Most baseline psychological variables significantly correlated with handicap at 3 months. When adjusting for baseline handicap and demographics, the baseline psychological variables only explained a significant ~3% of the variance in dizziness

handicap at follow-up, with baseline handicap explaining most of the variance. All-or-nothing behaviour was the most significant predictor.

Conclusions: Tertiary patients with vertigo and dizziness report negative illness perceptions and cognitive and behavioural responses to symptoms that are associated with self-reported handicap over time. Future studies are needed to investigate whether targeting these factors alongside traditional treatment approaches improves handicap in patients with chronic dizziness.

4.2 Introduction

The term "dizziness" refers either to a disturbance of spatial orientation or to a false perception of movement, which is more specifically called "vertigo" (Bisdorff et al., 2009). Dizziness is a common complaint in medicine, and around 20% to 30% of people will experience rotatory vertigo (Mendel et al., 2010; Neuhauser et al., 2008; Yardley et al., 1998d), which may be interpreted as a more specific marker of vestibular disturbance. Vestibular disorders can also be associated with a wide range of physical symptoms such as unsteadiness, unstable vision, motion intolerance, and autonomic symptoms, as well as cognitive symptoms ranging from impaired spatial learning and memory to altered sense of body ownership and embodiment (Lopez, 2016; Lopez et al., 2008; Smith et al., 2005).

These symptoms can result in substantial morbidity and disability, especially in patients with chronic symptoms. One in 10 people of working age report some degree of handicap due to current dizziness (Murdin & Schilder, 2015). A significant proportion of people are sufficiently disabled or distressed to be referred for investigation and management to hospital outpatient clinics. In many patients, a structural vestibular disorder can be identified, although "functional" or "medically unexplained" dizziness syndromes can also occur as primary or secondary conditions (Dieterich & Staab, 2017).

The Dizziness Handicap Inventory (DHI) (Jacobson & Newman, 1990) has been widely adopted in specialist settings to measure self-perceived dizziness-related disability. There is substantial variability in the levels of handicap even in relatively homogenous patient groups (Mutlu & Serbetcioglu, 2013). The level of handicap does not necessarily correlate with deficits on neuro-otological tests measuring the structural integrity of peripheral or central vestibular systems (Palla et al., 2008; Patel et al., 2016; Yip & Strupp, 2018). In contrast, studies have shown strong correlations between the DHI and anxiety, depression, and autonomic arousal (Cousins et al., 2017; Herdman et al., 2020c; Probst et al., 2017; Yardley, 1994a, 1994b; Yardley et al., 1994; Yardley & Redfern, 2001) and pathophysiological mechanisms have been proposed to explain this (Balaban & Thayer, 2001). Patients with prior anxiety and neurotic personality traits may also be more likely to develop secondary functional disorders such as "persistent postural perceptual dizziness" (Chiarella et al., 2016). Although premorbid

and comorbid mental health issues seem to play a role, the evidence to date suggests that they cannot fully explain the extent of the dizziness handicap. Not all patients have mental health disorders, and developing therapeutic treatments based on models of anxiety may be suboptimal (Hudson & Moss-Morris, 2019).

A handful of other studies have explored the role of patients' emotional responses to symptoms and beliefs about their illness in perpetuating handicap and dizziness symptoms. In an early study, Yardley (1994a) found that negative beliefs about the consequences of dizziness including fear of losing control were a significant predictor of dizziness and disability levels over time. Yardley et al. (2001) also found that beliefs about the negative consequences of dizziness at baseline predicted handicap for 6 months and could be effectively reduced with vestibular rehabilitation. Follow-up studies of patients with acute vestibulopathy found a positive relationship between patients' fear of panic-related physical symptoms and handicap (Cousins et al., 2017; Godemann et al., 2005). A recent cross-sectional study (Wolf et al., 2020) measured dizziness-specific cognitions using the Illness Perceptions Questionnaire—Revised (IPQ-R) (Moss-Morris et al., 2002). This study found that negative perceived consequences of dizziness were the strongest correlate of dizziness handicap after adjusting for demographic variables, severity of symptoms, depression, and anxiety. This suggests beliefs about illness may be more important predictors of disability than anxiety, mood, and severity of symptoms, but longitudinal research is needed to confirm this relationship.

In a precursor to the current study, we found that levels of handicap and symptom severity measured with self-report questionnaires before attending a specialist dizziness clinic were not correlated with either health care professional assessed vestibular function or diagnoses (Herdman et al., 2020b). In contrast, psychological factors including distress (anxiety and depression), illness perceptions, and cognitive-behavioural responses to dizziness such as avoidance of activity and focusing on symptoms were significantly correlated with handicap and severity of symptoms. The addition of cognitive-behavioural symptom interpretations is important because interpretations of symptoms may be direct drivers of day-to-day behaviour in people with vertigo and dizziness, which may ultimately lead to handicap. Psychological factors accounted for 53% and 30% of the variance in handicap and symptoms. There is therefore accumulating evidence for a range of common transdiagnostic psychological factors or mechanisms that might contribute to dizziness/vertigo-related disability.

The purpose of the current study was to extend the cross-sectional research by investigating longitudinally and prospectively whether this broader range of psychological factors and responses to symptoms before specialist input are associated with self-reported dizziness handicap 3 months after consultation and diagnosis. This article aims to answer the following questions:

1. Do dizziness handicap, distress, illness perceptions, and cognitive-behavioural responses to symptoms improve after specialist clinical assessment and diagnosis?

2. Are diagnostic category and vestibular test outcomes associated with self-reported handicap 3 months after consultation?

3. Do prediagnosis perceptions of dizziness, cognitive-behavioural responses to symptoms, and emotional factors predict handicap at 3 months after diagnosis?

4.3 Methods

Participants

Consecutive participants were recruited from the waiting list of the multidisciplinary balance clinic at Guy's & St Thomas' NHS Foundation Trust, London, between March and December 2018. Of the 476 eligible patients eligible to participate, 185 completed the baseline questionnaire and were contacted again after 3 months (Figure 5). The original cross-sectional findings are presented elsewhere (Herdman et al., 2020b). The study was approved by the NHS Health Research Authority (16/NI/0256).

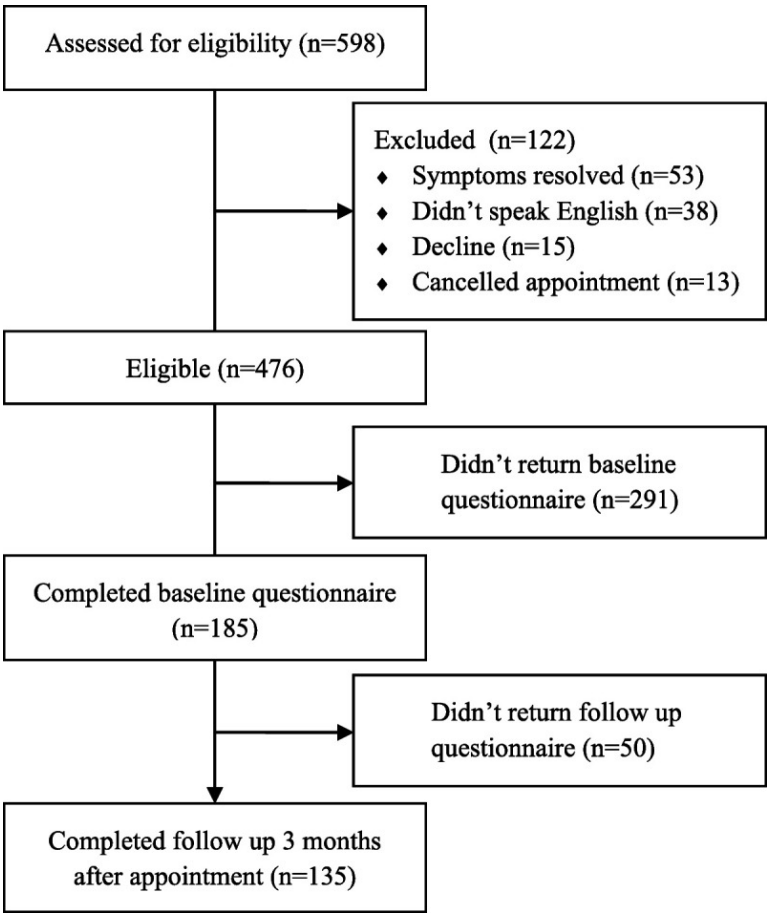

Figure 5. Participant flowchart

Data Collection

People on the waiting list received the questionnaire approximately 1 to 2 months before their initial appointment and completed it either electronically or via mail before they came for their appointment. Participants completed follow-up questionnaires 3 months after their initial diagnostic appointments. Reminders were sent out to nonresponders after 1 month. To facilitate follow-up, £10 expenses were sent to participants on completion of the three-month questionnaires.

Measures

Primary Outcome

DHI (Jacobson & Newman, 1990) is a 25-question scale that measures the extent dizziness causes physical, functional, and emotional disability. Higher scores represent higher levels of handicap and activity restriction.

Predictors

Patient Health Questionnaire (PHQ-9) (Spitzer et al., 1999) is a nine-item scale that measures the frequency of depressive symptoms in the last 2 weeks from "0" (not at all) to "3" (nearly every day). Scores of 10 or more indicate probable depression.

Generalized Anxiety Disorder (GAD-7) (Spitzer et al., 2006) is a seven-item scale that measures the frequency of anxiety symptoms in the last 2 weeks in the same way as the PHQ-9 and also has a cut-off of 10 or more for probable anxiety. For the purposes of analyses, it is also possible to combine the PHQ-9 and GAD-7 to form the Patient Health Questionnaire Anxiety and Depression Scale (Kroenke et al., 2016) as a composite measure of depression and anxiety.

IPQ-R (Moss-Morris et al., 2002) measures illness-related cognitions (beliefs). In accordance with the author's recommendations, the word "illness" was replaced with "dizziness condition" and the illness identity scale was modified to include symptoms relevant to people with vestibular disorders. The first domain measured the number of symptoms that the individual ascribed to their condition (illness identity). The other subscale measured how long they thought it would last (timeline), whether it would result in serious consequences (consequences), whether they believed they had power to influence their condition (personal control) or whether any treatment could improve it (treatment control), whether they understood the condition (illness coherence), whether the dizziness would come and go (cyclical timeline), and whether they had a strong emotional reaction when thinking about their dizziness (emotional representation). Participants are asked to respond to several statements for each domain on a 5-point Likert scale from "strongly agree" to "strongly disagree."

Cognitive-Behavioural Response to symptoms Questionnaire (Ryan et al., 2018) measures patients' cognitive and behavioural responses to symptoms. The five subscales

dealing with cognitive responses are symptom focusing, catastrophizing, damaging beliefs, fear avoidance, and embarrassment avoidance. The two behavioural subscales are all-or-nothing and avoidance/rest.

Beliefs About Emotions Scale (Rimes & Chalder, 2010) measures the extent to which patients believe it is unacceptable to experience negative emotions or to express emotion to others.

Psychological Vulnerability Scale (Sinclair & Wallston, 1999) measures maladaptive cognitive responses related to perceptions of dependency, perfectionism, negative attributions, and the need for external sources of approval.

Clinical Assessment and Treatment

All patients underwent a standardized clinical history and examination followed by a comprehensive vestibular battery to assess both peripheral and central vestibular function to reach a diagnosis. Findings for every patient were reviewed by the consultant audiovestibular physician (L.M.) who made the diagnosis based on consensus diagnostic criteria and commonly accepted definitions of the International Classification of Vestibular Disorders (Bisdorff et al., 2015).

Vestibular function was assessed using the video head impulse test (vHIT), caloric irrigation, and videonystagmography, which are the main laboratory tests that measure different frequency functions of the vestibular organ, its reflexes, and central neural connections (Nelson et al., 2016). Patients underwent further testing (such as imaging or vestibular-evoked myogenic potentials) or had additional examinations when clinically indicated to reach a final diagnosis. Further information on the vestibular testing can be found in the cross-sectional article (Herdman et al., 2020b).

Treatment of benign paroxysmal positional vertigo (BPPV) was carried out on the day; otherwise, patients were referred to see a physiotherapist for vestibular rehabilitation and/or the audiovestibular physician to discuss medical investigation or management. Patients also underwent psychological screening by validated questionnaires, and psychological assessment was recommended if they scored above the relevant threshold. The waiting lists to begin these treatments (other than BPPV) were typically longer than 3 months, although some patients may have been in the early stages of a vestibular rehabilitation program.

Statistical Analysis

Data were analysed using SPSS version 25. Two-sample t tests, $\chi 2$ test, and Fishers exact tests were used to examine the differences between responders and nonresponders. Because duration of dizziness was not normally distributed, this was log transformed for analyses. t Tests and analyses of variance (ANOVAs) explored the differences in handicap and psychological profile according to vestibular testing status

and diagnoses, respectively. Paired-sample t tests showed the change in scores between baseline and follow-up. Bivariate Pearson correlations explored the relationship between the psychological variables and handicap and partial correlations adjusted for baseline handicap. Because of multiple tests, we used the more stringent $p < .001$ to interpret relationships as significant. To assess if type of diagnosis affected the results, $\chi 2$ tests were performed. Hierarchical multiple linear regression was performed to predict DHI at follow-up. A dummy variable for vestibular testing was created to account for whether patients had any evidence of vestibular abnormality on one or more laboratory tests, consistent with diagnostic approaches in the internationally accepted diagnostic criteria of the Barany Society (Bisdorff et al., 2015).

4.4 Results

Participants

One hundred eighty-five consecutive patients completed the baseline questions, and 135 (73%) returned completed questionnaires at 3-month follow-up (Figure 5). There were no significant differences between responders (n = 135) and dropouts (n = 50) for the demographic variables or primary diagnosis (Table 5). For responders, the mean (standard deviation [SD]) duration of illness at baseline was 50.57 (69.161) months, and the median was 24 months (Table 5). Of all the demographic variables and diagnoses, only duration of dizziness at baseline was correlated with handicap at follow-up ($r = 0.28$, $p = .001$).

Table 5. Demographic and Diagnoses for Responders at Baseline and Follow-Up

Baseline Variable	Respondents at Baseline (n = 185)	Respondents at 3 mo. (n = 135)	Statistical Comparison
Age, y			
M (SD)	53.57 (17.386)	54.23 (17.531)	t = −0.850, p = .40, 95%
Range	18–90	18–90	CI = −8.133 to 3.233
Sex: female, n (%)	137 (74.1)	98 (72.6)	$\chi 2 = 0.555$, p = .46
Duration, mo.			
M (SD)	48.33 (64.359)	50.57 (69.161)	U = 3569.5, p = .55
Median	24	24	
Ethnicity, n (%)			
White	152 (82)	114 (84.4)	
Black, minority ethnic	33 (18)	21 (15.6)	$\chi 2 = 1.775$, p = .18
Marital status, n (%)			
Married/civil partnership	69 (37.3)	53 (39.3)	
Living with partner	28 (15.1)	19 (14.1)	
Single	48 (25.9)	35 (25.9)	$\chi 2 = 2.656$, p = .75
Divorced	19 (10.3)	15 (11.1)	
Separated	7 (3.8)	4 (3)	
Widowed	14 (7.6)	9 (6.7)	

Employment, n (%)			
Employed (full time)	55 (29.7)	37 (27.4)	
Employed (part time)	21 (11.4)	15 (11.1)	
Unemployed	26 (14.1)	18 (13.3)	$\chi2 = 6.163$, $p = .41$
Retired	58 (31.4)	48 (35.6)	
Student	6 (3.2)	5 (3.7)	
Home maker	5 (2.7)	4 (3.0)	
Other	14 (7.6)	8 (5.9)	
Education, n (%)			
Postgraduate	28 (15.1)	6 (2.2)	
University	48 (25.9)	33 (24.4)	
Apprenticeship	8 (4.3)	7 (5.2)	
Certificate/diploma	24 (13)	17 (12.6)	$\chi2 = 5.059$, $p = .65$
A-levels	14 (7.6)	11 (8.1)	
GCSE	35 (18.9)	29 (21.5)	
None	20 (10.8)	12 (8.9)	
Other	8 (4.3)	6 (4.4)	
Diagnosis, n (%)			
UPV	45 (25.4)	34 (25.2)	
BPPV	37 (20.9)	27 (20)	
VM	38 (21.5)	28 (20.7)	
Functional (e.g., PPPD)	11 (6.2)	7 (5.2)	
MD	9 (5.1)	6 (4.4)	$\chi2 = 4.467$, $p = .88$
Central	5 (2.8)	5 (3.7)	
BPV	5 (2.8)	4 (3)	
VS	3 (1.7)	3 (2.2)	
SSCD	2 (1.1)	1 (0.7)	
Other	22 (12.4)	16 (11.9)	

Note: M (SD) = mean (standard deviation); GCSE = General Certificate of Secondary Education; UPV = unilateral peripheral vestibulopathy; BPPV = benign paroxysmal positional vertigo; VM = vestibular migraine; PPPD = persistent postural perceptual dizziness; MD = Meniere disease; Central = central nervous system disorders; BPV = bilateral peripheral vestibulopathy; VS = vestibular schwannoma; SSCD = superior semi-circular canal dehiscence.

Do Dizziness Handicap, Distress, Illness Perceptions, and Cognitive-Behavioural Responses to Symptoms Improve After Clinical Assessment and Diagnosis?

Change in Handicap Scores

Baseline handicap measured by DHI was strongly correlated with handicap at 3-month follow-up ($r = 0.83$, $p < .01$). There was a mean (SD) improvement of 7.45 (14.57), which was statistically significant ($t(134) = 5.944$, $p < .001$). The maximum improvement was 52, and the maximum deterioration was 34. According to the clinically meaningful change score of 18 points as described by Jacobson and Newman (10), 3% (n = 4) of participants worsened, 75% (n = 101) stayed the same, and 22% (n = 30) improved. According to the recommended cut-offs, 39% (n = 52) had mild handicap, 36% (n = 49) had moderate, and 18% (n = 34) had severe handicap at follow-up.

Change in Anxiety and Depression Scores

There was a mean (SD) improvement from baseline to 3 months of 1.05 (4.97) on the depression scale (PHQ-9), which was statistically significant ($t(134) = 2.459$, $p = .015$). There was also a significant mean (SD) improvement of 1.04 (4.91) on the anxiety scale (GAD-7; $t(134) = 2.454$, $p = .015$). At 3 months, the proportion of participants who scored above the clinical threshold for suspected depression and anxiety remained the same (Table 6). At baseline, 41% (n = 55) had at least one measure of distress that met the cut-off compared with 37% (n = 50) at follow-up.

Table 6. Number of Participants Meeting Cutoff Scores for Distress Measures

	Baseline	**Follow-up**
Depression (PHQ-9 ≥ 10)	51 (38%)	43 (32%)
Anxiety (GAD-7 ≥ 10)	34 (25%)	33 (24%)
No. distress measures meeting cut-off		
1	25 (19%)	24 (18%)
2	30 (22%)	26 (19%)

Note: PHQ-9 = Patient Health Questionnaire, GAD-7 = Generalized Anxiety Disorder Scale.

Change in Illness Perceptions

When compared with baseline, at 3 months after diagnosis, participants had significantly greater understanding (coherence) of their condition, considered dizziness to have less serious consequences to their lives and had reduced negative emotions in relation to the condition (Table **7)**. For belief in the chronic or cyclical nature of their condition, and personal and treatment control, there were no significant differences in scores. Participants attributed fewer symptoms to their condition (illness identity) at follow-up, although the significance was borderline ($p = .053$).

At follow-up, 56% of participants had not changed their symptom attribution. Twenty-three percent of participants adopted a more psychological attribution, and 21% adopted a more physical attribution for their symptoms. Despite this individual variation in symptom attribution, a McNemar test determined that the difference in the proportion of participants with physical, psychological, and combined attributions at baseline and follow-up was not significantly different ($\chi2(3) = 5.032$, $p = .17$).

Table 7. Comparison of IPQ-R and CBRQ Scores at Baseline and Follow-Up

	Baseline, M (SD)	Follow-Up, M (SD)	Paired *t* Test (95% CI)
IPQ-R			
Illness identity	9.73 (5.24)	8.84 (5.42)	$t = 1.951, p = .053$ (−0.012 to 1.790)
Timeline (chronic)	18.94 (4.79)	18.45 (5.12)	$t = 1.351, p = .18$ (−0.228 to 1.210)
Timeline (cyclical)	13.98 (3.69)	13.52 (3.73)	$t = 1.351, p = .18$ (−0.213 to 1.132)
Consequences	18.83 (5.67)	17.68 (5.75)	$t = 2.714, p = .008$ (0.311 to 1.982)
Emotional representations	20.35 (5.79)	18.64 (6.10)	$t = 4.147, p < .001$ (0.895 to 2.527)
Personal control	17.29 (4.40)	17.70 (4.41)	$t = -1.072, p = .29$ (−1.159 to 0.344)
Treatment control	16.11 (3.47)	16.28 (4.41)	$t = 4.464, p = .64$ (−0.907 to 0.562)
Illness coherence	11.60 (4.62)	14.24 (5.07)	$t = -5.83, p < .001$ (−3.534 to −1.744)
CBRQ			
Symptom focusing	19.17 (5.765)	17.53 (6.374)	$t = 3.512, p = .001$ (0.718 to 2.571)
Catastrophizing	11.41 (3.946)	10.32 (4.001)	$t = 4.174, p < .001$ (0.577 to 1.616)
Damaging beliefs	15.21 (4.074)	13.84 (3.961)	$t = 4.959, p < .001$ (0.824 to 1.917)
Fear avoidance	18.53 (4.731)	17.04 (4.893)	$t = 4.058, p < .001$ (0.763 to 2.215)
Embarrassment avoidance	15.82 (6.268)	14.56 (6.752)	$t = 3.122, p = .002$ (0.461 to 2.057)
All-or-nothing behaviour	11.56 (5.288)	11.03 (5.081)	$t = 1.488, p = .14$ (−0.173 to 1.225)
Avoidance/resting behaviour	18.63 (7.068)	17.50 (7.244)	$t = 2.507, p = .013$ (0.239 to 2.028)

Note: IPQ-R = Illness Perceptions Questionnaire—Revised; CBRQ = Cognitive-Behavioural Response to symptoms Questionnaire; M (SD) = mean (standard deviation); CI = confidence interval.

Change in Cognitions and Behavioural Responses to Symptoms

There was a significant improvement in all the symptom cognitions (Table 7). Avoidance behaviour also significantly improved, although all-or-nothing behaviour did not significantly change.

Are Type of Diagnosis and Audiovestibular Test Outcomes Associated With Self-Reported Handicap 3 months After Consultation?

A one-way Welch ANOVA was conducted to determine if the level of handicap was different for the top 5 diagnostic groups, as the assumption of homogeneity of variances was violated (Levene test, p = .047). DHI scores increased from people with the Meniere disease (M [SD] = 29 [20]), to functional dizziness (M [SD] = 31 [23]), to vestibular migraine (M [SD] = 40 [29]), to BPPV (M [SD] = 41 [32]), to chronic unilateral peripheral vestibulopathy (M [SD] = 42 [21]), in that order. However, the overall test of differences between the groups was not statistically significant (Welch's F(4, 21.763) = 0.690, p = .61).

An independent-samples t test was run to determine if there were differences in handicap between patients with and without vestibular deficits. Patients with normal vestibular function scored 2.7 points (95% confidence interval [CI], −6.49 to 11.89) higher than did patients with abnormal vestibular function, which was not significant (t(128) = 5.81, p = .56). There were no significant differences in dizziness handicap at follow-up when each of the most frequently completed vestibular tests was analysed individually, which included videonystagmography (M = 2.92, 95% CI, −9.837 to 15.676, t(125) = 0.453, p = .65), vHIT (M = −1.03, 95% CI = −15.73 to 13.66, t(109) = −0.139, p = .89), and caloric paresis (M = −4.254, 95% CI = −14.209 to 5.701, t(91) = −0.849, p = .40).

Do Prediagnosis Perceptions of the Dizziness, Cognitive-Behavioural Responses to Symptoms and Emotional Factors Predict Handicap at 3 Months After Diagnosis?

Dizziness Handicap: Bivariate Correlations

Table 8 shows correlations for the psychological variables measured at baseline with the dizziness handicap score at 3 months. Most baseline variables showed moderate to large associations with handicap at 3 months. Baseline anxiety and depression, all of the subscales of the Cognitive-Behavioural Response to symptoms Questionnaire, and the identity, chronic timeline, serious consequences, and emotional representation subscales of the IPQ-R were all significant correlates (p < .001) of handicap at 3 months. The personal control, treatment control, illness coherence, and cyclical timeline subscales of the IPQ-R and the beliefs about emotions scale were not significantly related to handicap. Partial correlations after adjusting for baseline handicap reduced the correlations to nonsignificant except for all-or-nothing behaviour, which continued to be independent predictors of higher levels of handicap at 3 months.

Table 8. Correlations Between Psychological Variables and Dizziness Handicap at Follow-Up

Psychological Variables at Baseline	Dizziness Handicap at 3 mo.	
	Correlation (*r*)	Partial Correlations (*r*) Controlling for Baseline Handicap
Psychological distress		
Depression (PHQ-9)	0.675*	0.244
Anxiety (GAD-7)	0.594*	0.192
CBRQ		
Symptom focusing	0.392*	0.184
Catastrophizing	0.512*	0.145
Damaging beliefs	0.368*	0.066
Fear avoidance	0.411*	0.008
Embarrassment avoidance	0.594*	0.148
All-or-nothing behaviour	0.498*	0.289*
Avoidance/resting behaviour	0.605*	0.196
IPQ-R		
Identity	0.387*	0.088
Chronic timeline	0.366*	0.192
Consequences	0.539*	0.062
Personal control	−0.003	0.031
Treatment control	−0.205	−0.068
Illness coherence	0.069	0.132
Cyclical timeline	0.141	0.183
Emotional representation	0.478*	0.164
Beliefs About Emotions	0.027	0.074
Psychological Vulnerability	0.347*	0.189

PHQ-9 = Patient Health Questionnaire, GAD-7 = Generalized Anxiety Disorder Scale; CBRQ = Cognitive Behavioural response to Symptoms Questionnaire; IPQ-R = Illness Perceptions Questionnaire—Revised.
Partial correlation for association between baseline psychological variables and dizziness handicap at 3-month follow-up, controlling for dizziness handicap at baseline.
**p < .001.*

Regression

Hierarchical multiple linear regression was performed to predict DHI at follow-up (3 months), entering age, sex, and baseline DHI as control variables followed by baseline psychological variables, which were correlated with DHI at follow-up, with a correlation of ≥0.2 (Table 9). Because of collinearity, the emotional representation variable of the IPQ-R was excluded from the analyses as it overlaps with depressive symptoms and PHQ-9 and GAD-7 scores were grouped (Patient Health Questionnaire Anxiety and Depression Scale) as a composite measure of distress (Kroenke et al.,

2016). This model was significant (adjusted R2 = 0.735, ANOVA F = 22.86, p < .001) in which baseline dizziness handicap explained the most variance in handicap at follow-up, although adding the psychological variables still contributed an additional significant 3% to the model.

Table 9. Regression Model of DHI at Follow-Up (3 mo.)

Predictors (Baseline)	Dizziness Handicap (Follow-Up)				
	ΔR^2	Std. Error of the Estimate	R^2 Change	F Change	Sig. F Change
Step 1					
Demographic variables[a]	0.082	25.112	0.103	4.987	.003
Step 2					
Baseline DHI	0.707	14.186	0.613	280.520	.000
Step 3					
Psychological variables[b]	0.735	13.492	0.053	2.056	.022

Note: DHI = Dizziness Handicap Inventory.

[a]*Control variables included age, sex, and duration of symptoms.*

[b]*Psychological variables included The Patient Health Questionnaire Anxiety and Depression Scale, the Psychological Vulnerability, and subscales from the Illness Perceptions Questionnaire—Revised and Cognitive-Behavioural Response to symptoms Questionnaire.*

4.5 Discussion

The study found a significant improvement in dizziness handicap, anxiety, and depression 3 months after an initial consultation in a specialist vestibular clinic. Participants perceived fewer negative consequences, had significantly greater understanding of their illness, and were less emotionally affected by their condition. Participants also reported reductions in unhelpful cognitive-behavioural responses to symptoms (e.g., less symptom focusing, catastrophizing about symptoms, and avoiding activities because of embarrassment and/or fear). There was no change in all-or-nothing ("boom-bust") behaviour. Dizziness handicap at follow-up was associated with symptom duration, but not with any other demographic factor, diagnosis, or vestibular function test. The baseline self-report psychological measures were associated with dizziness handicap at follow-up, although the correlations were no longer significant after adjusting for baseline dizziness handicap except for all-or-nothing behaviour in response to symptoms. The fully adjusted model explained 74% of the variance in dizziness handicap at follow-up with the psychological factors explaining a significant 3%, and baseline dizziness handicap explaining the majority of the variance.

The data suggest that, although there was a significant improvement in handicap after diagnosis, the change was small and self-reported handicap remained relatively stable over the 3-month period of the study. Although the psychological measures improved,

the overall levels indicated that participants still tended to view their condition negatively and the rates of illness distress remained elevated. These participants had received a diagnosis and some treatment, and although these seem to reduce handicap, more is clearly needed to reduce handicap further as many patients were still significantly impaired.

There was no difference in self-reported handicap between the most common diagnoses and between patients with and without evidence of structural vestibular dysfunction. Normally, vestibular reflex function is highly correlated with vestibular perception. For example, when the vestibular system is stimulated (e.g., by irrigating the ear canal with warm water in the case of the "caloric test"), there will be a vestibular ocular reflex response manifest as a spontaneous eye movement (called nystagmus) and reproduction of vertigo. However, standard laboratory tests of vestibular reflex function seem to tell us little of how the patients with chronic disorders are feeling or their daily functioning.

These findings are in accordance with previous findings that neither caloric nor vHIT results predict symptom outcome in vestibular neuritis (Patel et al., 2016). Allum et al. (2017) also demonstrated that recovery occurs both in patients who recover peripheral (caloric) vestibular function and in those who do not. This occurs because of brain plasticity, which is influenced by exposure as observed in individuals who adapt to repeated vestibular stimulating from training (dancers) (Nigmatullina et al., 2015). Neuroimaging studies have also identified a wider vestibular network in the brain (Lopez, 2016) that goes beyond the traditional, lower-level reflex motor circuits measured using standard laboratory testing. These studies have started to find correlates between handicap and vestibular functional architecture (Li et al., 2020) that may help us understand further the relationships between physiology and ongoing symptoms and handicap.

Although more work is needed to understand the biology underpinning ongoing symptoms, the results do point to a number of possible mechanisms that may contribute to the perpetuation of dizziness handicap. The most important predictor in this study appeared to be "all-or-nothing" (or "boom-bust") behavioural responses to symptoms, which was the only item to retain its association with dizziness handicap over time when baseline dizziness was adjusted. This may be because people who engage in this behaviour tend to be quite symptom contingent, so if they are feeling good, they may overdo activity and then crash. This may lead to future negatively conditioned emotional responses to physical activity and dizziness.

Although other psychological variables were correlated with handicap over time, their effect on handicap disappeared when adjusting for baseline handicap. These factors were also relatively stable overtime, so it may be that they contribute to a vicious cycle of handicap whereby understandable cognitive behavioural and emotional responses to the initial symptoms and handicap actually contribute to the severity of the symptoms overtime.

For example, anxiety and depression could influence self-perceived handicap in a number of ways. Anxiety arousal can increase the somatic symptoms induced by balance disorders (Yardley & Redfern, 2001) and exert direct effects on vestibular information processing required for the perception and control of orientation (Balaban & Thayer, 2001). Anxiety and negative affect are also closely related to reporting of physical symptoms and negative attributional processes that can contribute to an escalating cycle of conditioned fear, arousal, and restriction of activity (McAndrew et al., 2014).

The relationship between distress and handicap, however, is imperfect. To adapt successfully to long-standing dizziness and vertigo, people also need to develop relatively accurate and balanced beliefs about symptoms, illness, and treatment. People develop their own "common sense" model of their condition, and sometimes that can be more negative than it needs to be or less accurate in some way (Hagger et al., 2017; Leventhal et al., 2016). In this study, participants who attributed a wider range of symptoms to their condition (higher illness identity), believed that their condition would last a long time, have more serious consequences, and be less likely to respond to treatment had higher levels of dizziness handicap at follow-up.

In their cross-sectional study, Wolf et al. (2020) also found correlations between handicap and illness perceptions, particularly negative perceived consequences. In this study, some but not all of the illness perceptions improved at follow-up, suggesting that somatic experience early in the temporal sequence of the condition is important in the development and maintenance of negative illness perceptions. It is important to note than in some instances, patients' symptom interpretations may indeed be accurate, but the overriding tendency to view symptoms of dizziness negatively seems to be unhelpful. Therefore, it is important to explore how patients think about or understand their condition and have some idea of whether that is an accurate or balanced view or not.

It is not only the overall representation of the illness that is important but also the day-to-day interpretation of symptoms, which seemed to be more consistently associated with self-reported handicap than beliefs about the illness as a whole. This may be because patients tend to focus on the symptoms rather than on more sophisticated or complex representations of their condition (Ghio et al., 2018). Dizziness handicap was higher in patients who focused more on their symptoms, catastrophized about the consequences of experiencing symptoms, believed that their dizziness symptoms were a sign of physical damage, were fearful of activity, and felt embarrassed about their symptoms.

These findings add to previous research that show that patients with dizziness frequently endorse such negative beliefs (Yardley et al., 2001), and that concerns of social embarrassment and being unable to fulfil normal roles contribute to dizziness handicap (Yardley, 1994a). Pothier et al. (2018) also found that catastrophizing about

dizziness explained a significant proportion of the variance in handicap after adjusting for mood. Thus, focusing on physical symptoms may effectively increase those sensations, and if patients perceive dizziness as a sign of imminent threat, their attempts to cope with this possibility may effectively prolong their handicap because restriction of physical movement may hinder natural recovery (compensation) from the initial vestibular dysfunction (Lacour & Bernard-Demanze, 2014) and reinforce negative perceptions.

In this study, patients with greater psychological vulnerability at baseline and follow-up also reported more dizziness handicap. Psychological vulnerability refers to people who base their self-esteem and respect on input from or in relation to others (Sinclair & Wallston, 1999). It could place patients at risk of greater distress and/or maladaptive coping, such as pressure to push on and then crash (all-or-nothing behaviour), because of perceived failure to live up to certain high standards. This could be relevant to people with vestibular diseases because vertigo and dizziness symptoms can markedly interfere with one's ability to achieve goals.

This study uniquely recorded data before diagnosis in a representative sample of patients attending a specialist clinic for dizziness. This was a pragmatic longitudinal design, and interpretation of causality is still limited owing to the imperfect control for confounders that exists outside a randomized trial and without multiple assessment points. Although we statistically adjusted for baseline dizziness handicap, this will not fully control for past exposure. Therefore, some reverse causality could be present in our estimates of the association between baseline variables and 3-month handicap. It was not possible to control for the treatment delivered to patients, although 3-month follow-up was chosen because most patients would have either not yet received treatment or been in the very early stages. It was also not possible to ascertain precisely when they completed the baseline measures before their diagnostic appointments.

Likewise, although the sample was representative, the response rate at follow-up may have affected our ability to detect more meaningful results, meaning some effects may have been underestimated. Nevertheless, our data point to the major relevance of longitudinal change in patients' perceptions, cognitions, and behaviours, as well as their negative affect, in understanding their levels of dizziness handicap regardless of neuro-otological diagnosis or vestibular function status.

4.6 Conclusion

Psychological factors including distress, dizziness-specific cognitions, behavioural responses, and negative illness perceptions before attending a specialist neuro-otology clinic predict ongoing dizziness handicap. The diagnostic process was associated with improvements in dizziness and psychological factors, although the level of distress remained high. Patients still tended to view their condition in a negative way and exhibit unhelpful cognitive and behavioural responses to symptoms. Vestibular function

tests or diagnosis, on the other hand, were not associated with ongoing dizziness handicap. Future studies should investigate whether targeting these factors alongside vestibular rehabilitation improves handicap in patients with chronic dizziness.

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Chapter 5 [manuscript under review]

A systematic review and meta-analysis of psychological correlates of dizziness related disability and symptom severity

Authors: David Herdman, Alicia Hughes, Louisa Murdin, Marousa Pavlou, Rona Moss-Morris, PhD

5.1 Abstract

Vertigo and dizziness are common and highly debilitating symptoms which may be influenced by psychological factors. This meta-analytic systematic review with detailed narrative synthesis examined empirical studies investigating the relationship(s) between psychological factors and dizziness handicap (disability) and symptom severity. CINAHL, EMBASE, Medline, PsychInfo and Web of Science databases were searched, and 89 studies were included. Meta-analysis was performed where there was sufficient data, which found moderate to large weighted positive correlations between handicap and anxiety (pooled correlation r = 0.52; 95% CI = 0.47-0.58), depression (r = 0.55; 0.47-0.62) combined scales (r = 0.57; 0.53-0.60) and autonomic symptoms (r = 0.55; 0.46-0.64). Moderate positive correlations were found between vertigo severity and anxiety (r = 0.35; 0.29-0.40) and depression (r = 0.37; 0.33-0.41). A detailed narrative synthesis identified other psychological variables of which negative illness perceptions and interpretations of symptoms, avoidance behaviour, and sleep disturbance were also consistently related to dizziness outcomes. Several limitations in the methodology across the studies were identified and recommendations for future research are therefore provided.

Highlights

- Many patients with dizziness experience ongoing disability despite standard treatment
- This review included 89 studies of psychological factors related to dizziness
- Moderate to large correlations were found between dizziness and anxiety/depression
- A model of cognitive-behavioural factors is proposed that reflects the current empirical evidence

- Recommendations are provided to improve methodological progress in this literature.

5.2 Introduction

The sensation of 'dizziness' can arise from a disturbed perception of spatial orientation (Bisdorff et al., 2009). Although closely aligned, international symptom classification guidelines define 'vertigo' independent from dizziness to include a false sense of motion such as spinning or rocking (Bisdorff et al., 2009). Taken together, dizziness and vertigo are amongst the most reported symptoms in the population (Mendel et al., 2010). Lifetime prevalence of significant dizziness may be as high as 30% (Murdin & Schilder, 2015). Even though most causes of dizziness can be considered benign in a medical sense, as a chronic symptom it results in significant morbidity with detrimental effects on work, travel, social and family life that can be long lasting or permanent (Bronstein et al., 2010a; Neuhauser et al., 2008; Skoien et al., 2008). When assessed using questionnaires, dizziness is frequently associated with long-lasting emotional distress and disability.

The most common cause of dizziness and vertigo is due to dysfunction of the vestibular system, which encodes self-motion and integrates with visual, and proprioceptive senses. The vestibular sense has widespread connections throughout the cerebral cortex and plays a vital role in our subjective sense of self-motion, orientation, and maintaining stable vision and posture (Cullen, 2012). The symptoms of vertigo and dizziness are therefore frequently accompanied by unsteadiness, unstable vision during head movement, difficulties during complex behaviours such as self-motion perception and navigation, and autonomic symptoms akin to sea sickness.

Vestibular conditions can cause a single episode of vertigo (e.g., acute peripheral vestibulopathy), episodic attacks (e.g., Meniere's Disease), or chronic dizziness and/or balance symptoms (e.g., central or bilateral vestibulopathy, or functional vestibular syndromes). Balance disorders typically exist along a spectrum, whereby some are considered to fit better with a more traditional biomedical disease model than others. Benign paroxysmal positional vertigo (BPPV) is an example of a condition with a relatively well understood biomedical pathomechanism. Nevertheless, anxiety and depression has been found to reduce the efficacy of treatment for BPPV and increase the risk of recurrence (Wei et al., 2018). On the other end of the scale, 'functional' vestibular syndromes such as Persistent Postural Perceptual Dizziness (PPPD) can arise as a consequence of any vestibular disorder since the structural, functional and psychological factors that cause this presentation are interrelated (Staab & Ruckenstein, 2007).

Psychological factors are thought to have the potential to influence the neurobiological adaptations that occur during recovery from acute vestibular system dysfunction. For full 'vestibular compensation' to occur, and thus recovery to occur, the brain needs to

adapt to the new pattern of sensory input and create new motor responses (Lacour, 2006). This process requires exposure to the disrupted signals and sensorimotor activity and so it is essential for patients to engage in movements which may provoke dizziness in order to recover.

Vestibular rehabilitation therapy (VRT) is a behavioural intervention which includes the necessary graded eye, head, and body movements designed to promote central compensation. It is a safe and effective treatment for people with chronic dizziness (Kundakci et al., 2018), although in some of the randomised trials only around 50% of subjects in the intervention group achieve the desired level of subjective improvement (McDonnell & Hillier, 2015).

Although psychological factors are considered relevant, VRT usually focusses on physiological processes and it is unclear what combination of factors are responsible for therapeutic change, which are usually not solely explained by changes in vestibular system function (Millar et al., 2020).

Only a small number of studies have explored incorporating elements of psychological therapies into vestibular rehabilitation, perhaps in part because it is not clear what factors should be targeted (Staab, 2011). Anxiety and depression are prevalent in around 30-50% of patients with chronic dizziness (Best et al., 2006; Lahmann et al., 2015), but some patients may understandably regard focussing on anxiety and depression as being incorrect where comorbidity is either not present or where they view the comorbid distress as a response to the primary dizziness problem (Herdman et al., 2021a).

Logically speaking, novel therapies for chronic dizziness should be designed to address factors identified as important in maintaining symptoms and illness specific disability (Hudson & Moss-Morris, 2019). However, no such agreed theoretical framework exists. Since objective tests of vestibular function used in the diagnostic work-up such as caloric testing, rotatory chair testing or video head impulse tests are inadequate for evaluating the personal *impact* of dizziness and response to treatment (Perez et al., 2003), most studies use self-report scales. The Dizziness Handicap Inventory (DHI) is the most commonly used tool to assess the self-perceived disability imposed by vestibular system disease (Jacobson & Newman, 1990).

In their review of the DHI, Mutlu and Serbetcioglu (2013) cited Pollak et al. (2012) to show the relationship with anxiety and depression, although there are in fact many observational studies that have investigated psychological factors contributing to self-reported dizziness handicap. As multiple similar studies investigating such relationships have accrued and considering the clinical importance of this topic given its centrality to the understanding and treatment of such a common and disabling clinical problem, a review and synthesis of this research has become increasingly important.

A reliable critical synthesis of the field of research would allow clinicians and researchers involved in rehabilitation, neuro-otology, and neuroscience to gain an understanding of psychosocial factors that are linked to chronic vertigo and dizziness to inform the development of a more integrated approach to developing scientifically based and effective assessments and interventions. A review would also help structure research questions and provide clear guidance for future studies.

The aims of this systematic review were to

(1) Narratively summarise the existing empirical findings of the range of psychosocial factors associated with persistent vertigo and dizziness related disability.

(2) Meta-analyse the size of the relationships between psychosocial variables and vertigo and dizziness where sufficient data is available.

(3) Critically evaluate the methodology used, identifying gaps in the evidence and ideas for future research in this area.

5.3 Methods

Eligibility

Studies were included if they met the following criteria:

a) Studies of adults with vertigo/dizziness reporting quantitative, psychometrically valid dizziness outcome measures in conjunction with psychosocial factors typically considered modifiable in the context of psychological approaches. Dizziness outcomes were defined in two ways; (1) dizziness related disability (commonly referred to as 'handicap' in the neuro-otology literature) and (2) severity of dizziness/vertigo symptoms. For the purposes of this review, we did not include general quality of life outcomes since we were interested in illness specific measures.

And one of the following, (b) to (e):

b) Explored bivariate relationships between psychosocial factors and dizziness severity or disability

c) Reported statistical models with psychosocial factors as predictors and dizziness symptoms or interference as outcome variables

d) Tested differences related to dizziness outcomes and different psychological subgroups

e) Evaluated treatments for chronic dizziness looking at psychosocial mediators of outcome in relation to dizziness outcomes

Studies were excluded if they failed to meet minimum inclusion criteria (a) and did not report at least one of the statistical methods described in criteria (b) to (e). Studies were also excluded if they:

a) Were not written in English

b) Were non-empirical, general discussion or theoretical papers

c) Used qualitative rather than quantitative methods

d) Reported only multivariate statistical models with dizziness severity or disability as predictors and psychosocial factors as outcome variables

e) Used only participants with primary mental health disorders

f) Used a general population rather than clinical or patient specific cohorts

Search strategy

Electronic databases (CINAHL, EMBASE, MEDLINE, PsychInfo and Web of Science) were searched for relevant empirical studies published since 1980. Reference sections of included articles were searched, and corresponding authors contacted to identify unpublished studies. Search terms were customised to each database and involved combining key word searches for a list of psychological and dizziness terms and limits were applied to include only papers published in English. A comprehensive list of search terms for each database is included in Appendix C. Duplicates were removed, and titles and abstracts screened according to the inclusion/ exclusion criteria. Studies not meeting these criteria were removed and the full tests of all remaining studies were retrieved and screened by two authors. Just prior to submission (January 2022) the search was updated through PubMed by a single author.

Data Extraction

Extracted data included publication data, study design, sample characteristics, setting, psychological and dizziness outcomes. For over seventy percent of papers double extraction was performed to ensure accuracy of key findings and quality assessment. Where multiple measures were included in a study, consensus was reached amongst the reviewers as to which variables were relevant to the review questions. The reviewers only collected the information from the articles that were relevant to the review question and scored the quality assessment accordingly.

Quality assessment

The 'Checklist for Measuring Quality' developed by (Downs & Black, 1998) was adapted specifically for this review based on consensus amongst the authors (see Appendix D). We also omitted the items that are only relevant to intervention studies. The quality assessment tool contained items assessing reporting, diagnosis, external validity, internal validity – bias, internal validity – confounding, and power. Some items within those categories assessed features specific to longitudinal designs such that the maximum quality score was 16 for cross-sectional studies and 19 for longitudinal or intervention studies. Studies were assessed according to the nature of the data extracted for the purposes of this review, which in some cases did not correspond to the overall study design.

Synthesis

We completed meta-analysis to determine the strength of the cross-sectional relationship between dizziness outcomes and anxiety and depression. The Hunter-Schmidt method was calculated to produce a weighted mean of the raw correlation coefficient and sample sizes (Hunter & Schmidt, 1990) using Stats Direct. Data extracted were from bivariate cross-sectional analyses since studies that reported multivariate models varied substantially with respect to control variables included which limit meaningful interpretation of multivariate analyses across studies. The original three factor subscale of the Dizziness Handicap Inventory (DHI) includes items on physical, functional, and emotional consequences although since the internal validity of this subscale structure has not been supported (Kurre et al., 2010; Tamber et al., 2009) we used the total score for the purposes of meta-analysis.

The multifaceted nature of the review question precluded meta-analysis for the other psychological factors identified due to a combination of the low number of studies for each factor and the heterogeneity in the methods used. Instead, psychological factors were grouped into overarching conceptually or thematically related categories and a narrative synthesis is provided.

5.4 Results

Study characteristics

Initial searches yielded 10,953 results, 1,782 of which were duplicates. After screening titles and abstracts, the full texts of 335 studies were screened for inclusion (Figure 6). Eighty-nine studies were identified as meeting inclusion criteria for this review and are summarised in Table 10. Twenty-three studies were longitudinal, 55 studies were cross-sectional, and 11 studies were clinical trials. The studies included a total of 15,615 participants. The median sample size was 102, ranging from 10 (Gomez-Alvarez & Jauregui-Renaud, 2011) to 1,159 (Obermann et al., 2015). Most studies included patients with mixed vestibular pathologies (n=57), followed by unilateral peripheral

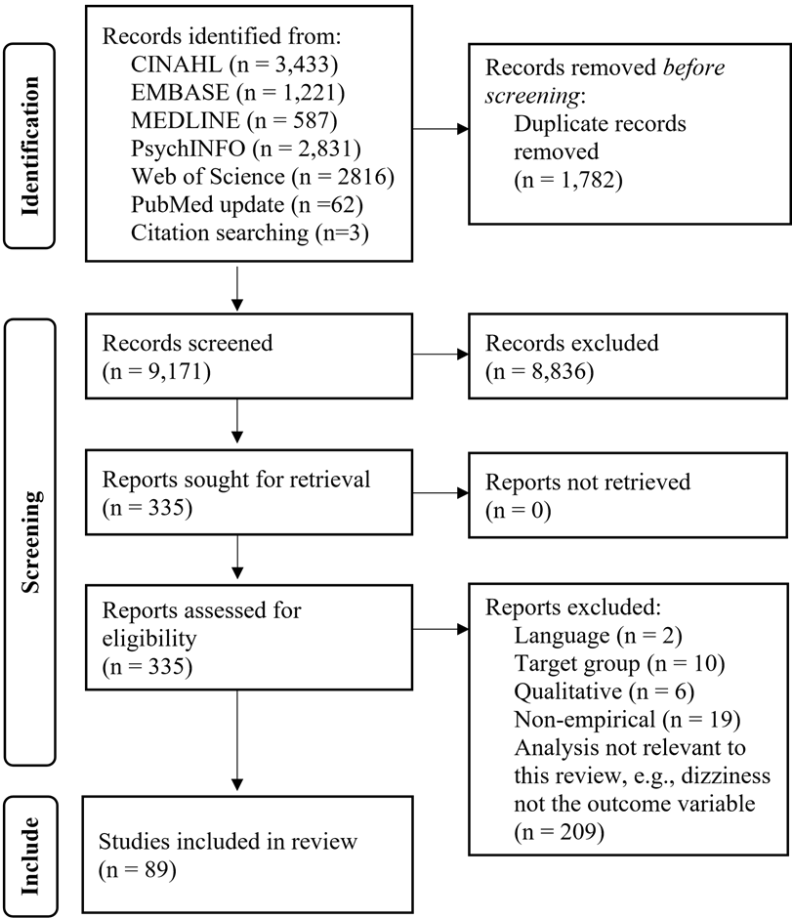

Figure 6. PRISMA Flow Diagram

vestibulopathy (n=9), Meniere's Disease (n=8), functional vestibular disorders (n=6), BPPV (n=4), cervicogenic dizziness (n=3), vestibular schwannoma (n=2), vestibular migraine (n=1) and TBI (n=1). Some studies used the same participants but reported their findings in separate articles so are included in the section relevant to the results presented in each article. Where the same results were reported in different journals, only one article was chosen.

For measures of handicap (illness-related disability), 49 studies used the Dizziness Handicap Inventory, and 15 studies used the Vertigo Handicap Questionnaire. Other outcomes included the UCLA Dizziness Questionnaire (n=3), Vestibular Activities and Participation (n=2), Illness Intrusiveness Rating Scale (n=1), Meniere's Disease Impact Scale (n=1), Subjective impairment visual analogue scale (n=1), Vestibular Disorders Activities of Daily Living scale (n=1), Sickness Impact Profile (n=1) and three studies used study specific handicap scales.

For measures of vertigo and dizziness symptom severity, 27 used the Vertigo Symptom Scale. Two studies used the Situational Characteristics Questionnaire which measures visually induced dizziness, and one study respectively used the Vertigo Dizziness Imbalance symptom scale, Dizziness Symptoms Inventory, and a dizziness visual

analogue scale. One study used a single question item to measure perceived change in vertigo severity.

Four themes were identified which included: (1) 'Distress', incorporating anxiety, depression and affective related factors: (2) 'Personality and resilience factors', incorporating trait characteristics: (3) 'Cognitions' which reflect thoughts and beliefs about an illness, symptom(s) or health behaviour and (3) 'Behaviour and coping', incorporating actions taken by individuals including sleep.

Table 10. Studies included in the systematic review

Ref.	Design	Country	Participants	Sex N (%)	Age Mean (SD)	Setting	Theme	Dizziness measure	Psych measure	Key findings	Quality score
Abe-Fujisawa et al. (2021)	CS	Japan	76 Mixed older adults	M 25 (33%) F 51 (67%)	74.2 (6.3)	Otolaryngology Department	Distress	DHI	HADS	DHI correlated to HADS anxiety (r 0.38, p<.01) but not depression (r 0.18)	11
Arroll et al. (2012)	CS	UK	74 MD	M 20 (27%) F 54 (73%)	57.9 (12.21)	UK patient support group	Distress Cognitions	DHI	CES-D MUIS	Illness uncertainty was correlated with emotional (r=0.256) and functional (r=0.244) handicap, but not with physical handicap. Depression was moderately related with all handicap subscales (r=0.453 to 0.492).	11
Arroll et al. (2016)	CS	UK	66 MdDS	M 4 (6%) F 62 (94%)	52.1 (12.2 years)	2 online patient support web sites	Distress	IIRS MdDS-scale	SSCI	Stigma was association with symptom severity (r=.381) and negatively associated with illness intrusiveness (r=-.588)	11
Bayat et al. (2020)	CS	Iran	130 Mixed	M 52 (40%) F 78 (60%)	range 18-75 (Mean NK)	Balance clinic	Distress	DHI	BAI	Positive correlation between BAI and DHI total (r=0.636), and each of its component scores.	12
(Best et al., 2009)	L	Germany	68 Mixed	NK	NK	NK	Distress	VSS/VHQ	HADS SCL-90R	'Subjective symptoms' correlated positively with acute depressive and anxiety distress as measured by the SCL-90R – limited reporting.	6
Cheng et al. (2012)	CS	Taiwan	79 Mixed	M 44 (56%) F 34 (44%)	68 (15.54)	Outpatient clinic	Distress	DHI	HADS	Anxiety and depression (HADS>8) and total HADS score were significantly associated with different categorical levels of DHI	12
Chiarella et al. (2016)	CS	Italy	19 CSD 22 PVD	M 17 (41%) F 24 (59%)	CSD 34.9 (12.6) PVD 39.5 (11.3)	Neuro-otology clinic	Personality	DHI	NEO-PI-R	Neuroticism positively correlated with DHI in the CSD group (r=0.53) but less so in the PVD group (r=0.37, p=0.09)	10
Cousins et al. (2017)	L	UK	42 AUV	M 24 (57%) F 18 (43%)	50 (SD NK)	Hospital and neuro-otology unit	Distress Cognitions	DHI	HADS BSQ VSS-A	At 10 weeks DHI was correlated with HADS (0.71), VSS-A (0.698) and BSQ (0.583). DHI at 10 weeks was correlated with baseline VSS_A (r=0.351) and BSQ (r=0.529 - although this was no longer sig after adjustment) but was not related with HADS.	13
Cuenca-Martinez et al. (2018)	CS	Spain	64 CGD	M 10 (16%) F 54 (84%)	Lower DHI group = 52.95 (14.00) Higher DHI group = 53.84 (10.84)	Physical therapy service	Cognitions	DHI	TSK PCS	In patients with high DHI, DHI was correlated with kinesiophobia (r=0.36), and pain catastrophising (0.416). There were significant group differences between people with low vs high DHI for both kinesiophobia and pain catastrophising with large effect sizes.	9
Dunlap et al. (2020)	CS	USA	404 Mixed	M 143 (35%) F 261 (65%)	54.0 (17.0)	Balance disorder and physical therapy clinic	Cognitions	VAP	VAAI-9	The VAAI-9 and VAP scores at baseline were correlated (p=0.81, 95% CI 0.77 to 0.84).	11
Dunlap et al. (2021)	L	USA	286 Mixed	Only baseline data provided	Only baseline data provided	Balance disorder and physical therapy clinic	Cognitions	VAP VAS - dizziness	VAAI-9	Fear avoidance at baseline and was significantly associated with VAP (ρ = 0.54), and dizziness VAS at 3-month follow-up (ρ = 0.37). Approximately 38% of the variation in VAP score at follow-up was predicted by 9-item VAAI score, dizziness VAS, and HADS-D score when considered together.	15
Formeister et al. (2020)	CS	USA	70 Mixed	M 36 (51%) F 34 (49%)	56.3 (13.8)	Neuro-otology clinic	Distress	DHI	PHQ9 GAD7	DHI was positively correlated with PHQ9 (adjusted r2 =0.40, p<0.001) and GAD-7 (adjusted r2 = 0.16, p<0.001).	13
Gerretsen et al. (2020)	Trial	Canada	229 Mixed	M 73 (32%) F 156 (68%)	52.3 (15.2)	Interdisciplinary neurotology clinic	Cognitions	DHI	DCS	Exploratory multiple regression analyses revealed that change in DCS scores was a predictor of percentage change in DHI scores in the whole sample and in both study groups.	15
Godemann et al. (2004)	L	Germany	67 Hospitalised AUV	M 29 (43%) F 38 (57%)	52 (14.3)	1 neurological and 7 ENT departments	Behaviour Cognitions Personality	VSS	STAI FKV ACQ	13 of the 67 patients reported continuing dizziness but only 3 had pathological scores on VSS. In linear regression, the predominant factors contributing to severity of vertigo: female sex, dependent personality	9

Ref.	Design	Country	Participants	Sex N (%)	Age Mean (SD)	Setting	Theme	Dizziness measure	Psych measure	Key findings	Quality score
									BSQ PSSI	structure, tendency to evaluate body sensations fearfully (ACQ) and together explained 35% of the variance in vertigo associated symptoms.	
Godemann et al. (2005)	CS	Germany	75 1 year after AUV	M 37 (49%) F 38 (51%)	50 (4.5)	Hospital and neuro-otology unit	Cognitions Distress	VSS	ACQ BSQ STAI-S SCL-90-R	30/75 (40.5%) still experienced the presence of vertigo a year after discharge from hospital. VSS vertigo severity was correlated with all psychological measures (ACQ total 0.45; ACQ somatic crisis 0.42; ACQ loss of control 0.28; BSQ total 0.47; STAI state 0.39; SCL-R-90 Anxiety 0.41; SCL-90-R Phobic avoiding behaviour 0.41). The ACQ and the BSQ together can predict the severity of the vertigo up to 30%, but this did not consider possible confounding.	12
Gomez-Alvarez and Jauregui-Renaud (2011)	L	Mexico	10 AUV	M 5 (50%) F 5 (50%)	39 (14)	2 neuro-otology departments	Distress	DHI	DD DES GHQ-12 SAS HDRS	High number of patients reported symptoms of DD in the acute stages. No correlation was observed between the differences on the DD or DES score and the differences on the DHI or other instruments. Also, no significant correlation was observed between the Hamilton scale difference and any of the other total score differences.	13
Goto et al. (2017)	Trial	Japan	138 PVD	M 33 (24%) F 105 (76%)	61 (14.8)	In-hospital vestibular rehabilitation programme	Cognitions	DHI	SSCS SSAS SDS STAI HADS	The following psychological variables were significantly associated with DHI changes after the intervention: pre- SSAS, pre-SDS, pre-STAI (State and Trait), and pre-HADS. In the multivariate analyses, adjusting for demographics, pre-SSCS was significantly associated with DHI change - specifically the DHI score decreased with increased pre-SSCS.	14
Green Jr et al. (2007)	L	USA	61 refractory MD	M 20 (33%) F 41 (67%)	49 (9.0)	3 academic medical centres and 1 private clinic	Personality	VSS	SOC	VSS was negatively correlated with SOC (-.456, p<.001) (people with weaker sense of coherence had more symptoms). Only the meaningfulness domain was significantly related with vertigo category over time.	13
Grunfeld et al. (2003)	CS	UK	91 Mixed	M 34 (37%) F 57 (63%)	M 50.1 F 40.1	Tertiary referral unit	Distress Personality	VSS	HADS RSE SSQ	Vertigo symptoms were sig correlated with depression (r=0.260) and anxiety (r=0.214) but not with self-esteem or social contacts	11
Hägnebo et al. (1999)	CS	Sweden	50 MD	M 18 (36%) F 32 (64%)	56 (13)	Meniere's Disease patient association	Personality Behaviour	DHI	WOCQ ASI	Together the subscales of the WOCQ accounted for 33% of the variance in DHI functional subscale, with significant contributions from Escape/Avoidance and Distancing that were positively, and Self-Controlling negatively related to functional handicap. The coping strategy did not predict perceived emotional or physical handicap from dizziness. A positive correlated was found between the ASI DHI/emo, but no sig. correlations with the other DHI subscales.	10
Heinrichs et al. (2007)	L	Germany	43 AUV (24) or BPPV (n=19)	M 18 (42%) F 25 (58%)	56.2 (13.5)	Community Hospital	Distress Cognition Behaviour	VAS – subjective impairment	BAI ACQ BSQ MI BDI SCL-90R	Comparing those who continued to complain about dizziness and those who had recovered (70%), neither the ACQ nor BSQ sig differed between the groups at the time of admission. However, 3 months later, both groups differed sig across all dependent variables and the group with persistent dizziness had a higher prevalence of mental disorders. The BSQ was a sig predictor of ongoing dizziness in patients with AUV but not in BPPV.	13
Herdman et al. (2020c)	CS	UK	954 Mixed	M 290 (30%) F 664 (70%)	51.48 (16.0)	Vestibular clinic	Distress	DHI	PHQ9 GAD7	DHI was positively correlated to anxiety (0.611) and depression (0.712,) after adjusting for age and gender	14
Herdman et al. (2020b)	CS	UK	185 Mixed	M 48 (26%) F 137 (74%)	53.57 (17.39)	Vestibular clinic	Distress Cognitions Personality Behaviour	DHI VSS	PHQ9 GAD7 IPQ-R CBRQ PVS BAE	DHI was correlated with PHQ9 (0.651), GAD7 (0.576), all CBRQ items (0.388 to 0.633), illness perceptions only identity (0.427)/chronic timeline (0.345)/consequences (0.598)/Treatment control (-0.179) and Emotion Representation (0.493) but not personal control, illness coherence or cyclical timeline. DHI was not correlated to beliefs about emotions. DHI was correlated to psychological vulnerability (0.322). VSS had the same pattern of significant correlations. In the total regression	15

Ref.	Design	Country	Participants	Sex N (%)	Age Mean (SD)	Setting	Theme	Dizziness measure	Psych measure	Key findings	Quality score
										model, psychological variables explained 53% of the variance in DHI, and 30% for symptoms (VSS). Factors associated with DHI included age, female gender, distress, symptom focusing, embarrassment, avoidance, and beliefs about negative consequences. Fear avoidance was the only independent correlate in the fully adjusted model of symptom severity.	
Herdman et al. (2020a)	L	UK	135 Mixed	M 37 (27%) F 98 (73%)	54.23 (17.53)	Vestibular clinic	Distress Cognitions Personality Behaviour	DHI	PHQ9 GAD7 IPQ-R CBRQ PVS BAE	Baseline psychological variables correlated with DHI at 3 months follow up after initial examination: Depression (0.675) Anxiety (0.594) CBRQ - (0.368 to 0.605) IPQ-R - identity, chronic timeline and emotion representation, psychological vulnerability (0.347), not beliefs about emotions These correlations were not sig after adjusting for baseline handicap other than all-or-nothing behaviour.	18
Hong et al. (2012)	L	Korea	126 Mixed, older adults	M 30 (24%) F 96 (76%)	71.6	4 outpatient hospitals	Distress	K-VADL	BDI STAI BAI	Significant correlations between the K-VADL score and both the BDI (r=0.357) and STAI (r=0.444) scores before and after 3 months general treatment as usual management (r=0.486, r=0.373).	12
Honrubia et al. (1996)	CS	USA	362 Mixed	M 150 (41%) F 210 (58%)	58	Outpatient clinic	Distress	UCLA-DQ	GCS CAS (>30 = distress)	Significant effect of psychologic distress on impairment in daily activities, quality of life, and fear of becoming dizzy, with higher responses for the patients affected by psychologic distress.	8
Horii et al. (2007)	Trial	Japan	41 Mixed	At baseline, n=60 M=22 (37%) F 38 (63%)	Group 1 = 51.5 (2.7) Group 2 = 44.8 (2.8)	Open label trial of fluvoxamine	Distress	DHI	HADS	Post-pre ratio of HADS scores and subjective handicaps showed a significant correlation (r=0.388)	12
Horii et al. (2016)	CS	Japan	29 Mixed	M 9 (31%) F 20 (69%)	Group 1 (HADS<13) = 49.8 (3.9) Group 2 = 49.4 (3.7)	University Hospital, during trial of milnacipran	Distress	DHI	HADS	Significant correlation between HADS Total and Handicap before treatment (r=0.69). DHI was more severe in groups with HADS>12 before treatment (p<0.01)	11
Humphriss et al. (2004)	CS	UK	100 Mixed	M 34 (34%) F 66 (66%)	49.6 (14.3)	University Hospital Neuro-otology practice	Distress	DHI	NQ	Sig correlation between DHI and Nijmegen scores (rho=0.348). Similarly when patients were grouped in accordance with Nijmegen score significance, total DHI scores were sig greater in pts with significant Nijmegen scores.	14
Kamalvand et al. (2017)	CS	Iran	101 Mixed	M 39 (39%) F 62 (61%)	47.76 (13.72)	Outpatient clinic	Distress	VSS	BAI	In this validation study, the Persian version of the VSS-VER was correlated with anxiety (r=0.40)	11
Kammerlind et al. (2005)	L	Sweden	51 3-6 years after a trial for AUV	M 27 (47%) F 24 (47%)	56 (13)	3 x ENT departments	Distress	UCLA-DQ	HADS	The group with remaining symptoms (n=9) had higher level of anxiety and depression compared with no symptom group (n=9) (groups based on the UCLA-DQ).	13
Kammerlind et al. (2011)	L	Sweden	40 AUV	M 23 (55%) F 19 (45%)	54	2 x ENT Departments	Cognitions	UCLA-DQ	HADS DBS	There were no differences between the subjects with substantial remaining symptoms according to the UCLA-DQ at 6 months (n=7) compared with the group with no remaining symptoms after 6 months (n=12) in beliefs, anxiety or depression	17
Ketola et al. (2014)	CS	Finland	547 members of Finnish Meniere's Federation	M 112 (21%) F 434 (79%)	61 (11.1)	Postal sample	Personality	DHI MD-impact scale	SOC-13	Higher SOC scores were related to lower scores in MD-impact and more severe vertigo symptoms.	7
Kim et al. (2018)	CS	Korea	237 Mixed	M 78 (40%) F 169 (60%)	41.73 (12.77)	Otolaryngology Department	Behaviour	DHI	PSQI ISI	The correlation coefficient between DHI and PSQI/ISI were highest in VM (0.491/0.415), then BPPV (0.269/0.306) and in the PVD group only PSQI was correlated (0.330). There were no significant correlations in the MD or PD group.	11

Ref.	Design	Country	Participants	Sex N (%)	Age Mean (SD)	Setting	Theme	Dizziness measure	Psych measure	Key findings	Quality score
Kirby and Yardley (2009a)	CS	UK	358 members of MD support group	M 112 (31%) F 246 (69%)	NK (range 28-90)	Participating in a trial	Distress	VSS	HADS	Vertigo severity at baseline was higher in people with anxiety at baseline (HADS>7) (d=0.25)	9
Kirby and Yardley (2009b)	CS	UK	800 members of MD society	M 296 (37%) F 505 (63%)	60.54 (12.54)	Postal sample, MD society	Distress Cognitions	DHI VSS	IUS SHAI PCL HADS VSS-A	DHI was sig assoc with autonomic arousal (r=0.65), intolerance of uncertainty (r=0.47), health anxiety (r=0.45), PTSD (r=0.65), anxiety (r=0.62), and depression (r=0.71). These remained sig after controlling for demographic variables and symptom severity. In the final model PTSD symptoms contributed most to handicap and the inclusion of PTSD significantly mediated the effect of intolerance of uncertainty on handicap.	12
Kleffelgaard et al. (2017)	CS	Norway	65 Enrolled in RCT 2-6 months following mTBI with dizziness	M 19 (29%) F 46 (71%)	39.2 (12.9)	Outpatient department of Physical Medicine & Rehabilitation	Distress	DHI VSS	HADS	Autonomic arousal (r=0.363), anxiety (0.359) and depression (0.426) correlated with DHI. These remained sig after adjusting for vertigo and post concussive symptom severity, comorbidity, neck pain and balance. Vertigo symptoms also correlated with autonomic symptoms (0.310), depression (0.287) but not anxiety (0.175).	12
Kondo et al. (2015)	CS	Japan	159 Mixed	M 55 (35%) F 104 (65%)	57.4 (16.8)	4 otolaryngology departments	Distress	DHI VSS	HADS	DHI total was correlated with HADS-A (0.555) and HAD-D (0.534). VSS-sf-V was also correlated with HADS-A (0.425) and HADS-D (0.390)	12
Kurre et al. (2010)	CS	Germany	194 Mixed	M 74 (38%) F 120 (62%)	50.6 (13.6)	Tertiary centre	Distress	DHI	HADS	DHI was sig correlated with HADS total (0.59), HADS-A (0.43) and HADS-D (0.66)	12
Kurre et al. (2012)	CS	Switzerland	200 Mixed	M 76 (38%) F (124 (62%)	49.7 (13.5)	Outpatient centre for Vertigo & Balance Disorders	Distress	DHI	HADS	DHI was sig correlated with HADS Total (0.60), HADS-D (0.66) and HADS-A (0.45). The correlations remained sig after adjusting for symptom severity with the VSS (HADS = 0.44, HADS-D = 0.5, HADS-A = 0.3). The correlation values were higher in men.	13
Limburg et al. (2019)	Trial	Germany	72 Inpatients with FVD	M 38 (53%) F 34 (47%)	49 (14.9)	Inpatient setting receiving psychodynamic approach	Cognitions	VHQ	KLC	Hierarchical linear regression analysis showed that VHQ at admission was the only significant predictor of VHQ at follow up; internal-external body-related locus of control did not add predictive value	16
Maarsingh et al. (2011)	CS	Netherlands	415 Mixed (primary care)	M 110 (27%) F 305 (73%)	78.5 (7.2)	Primary care	Distress	DHI	PHQ	Prescence of anxiety and/or depression disorder was associated with higher score on DHI	10
MacDowell et al. (2018)	L	USA	118 Mixed (VRT patients)	M 34 (31%) F 82 (69%)	60.08 (16.20)	Vestibular Rehabilitation Therapy Department	Personality	DHI	PANAS	Retrospective chart review found pts with abnormal affect had higher DHI scores pre and post rehab, but the difference between groups was not statistically significant. Pts with abnormal affect required longer length of therapy intervention that the group with normal affect	10
Mahoney et al. (2013)	Trial	Australia	44 FVD	M 12 (27%) F 32 (73%)	46.7 (12.97)	Neuro-otology clinic (RCT)	Distress	DSI DHI	MINI SBI DASS-21	DHI scores at 6 months post CBT was related to pre-treatment anxiety (r=0.49) but not to severity of pre-treatment dizziness symptoms or avoidance behaviours. Only pre-treatment anxiety was a significant predictor of DHI at 6 months post treatment in the regression model	13
Menant et al. (2020)	CS	Australia	305 Mixed (RCT)	M 112 (37%) F 193 (63%)	67.8 (8.3)	RCT	Distress Personality	DHI	PHQ9 GAD7 Neuroticism subscale from the NEO 5-	Binary analysis showed participants with moderate/severe DHI had significantly elevated symptoms of anxiety, depression, and neuroticism. Severe anxiety (GAD7 >7) was included in the multivariate model and remained significant.	13

Ref.	Design	Country	Participants	Sex N (%)	Age Mean (SD)	Setting	Theme	Dizziness measure	Psych measure	Key findings	Quality score
									Factor Inventory		
Mendel et al. (2001)	CS	Sweden	99 Mixed	M 36 (36%) F 63 (64%)	Male 55 (16) Female 54 (14)	Outpatient audiology clinic	Personality	VHQ VSS SIP	SOC HADS	Patients classified as having strong SOC scored significantly less handicap than patients with weak or moderate SOC. Vertigo symptoms were not significantly different between SOC groups.	11
Micarelli et al. (2019)	CS	Italy	93 CGD	M 42 (45%) F 51 (55%)	43.3 (13.7)	Interdisciplinary disorder clinic	Distress	DHI	TSK HADS	DHI was correlated with Kinesiophobia (TSK-17, r=0.72), and anxiety (HADS-A, r=0.77)	11
Micarelli et al. (2020)	CS	Italy	49 CGD 43 with both CGD and temporomandibular disorders (TMJ)	Male CGD = 23 (47%) TMD/CGD = 20 (47%) Female CGD = 26 (53%) TMD/CGD = 23 (44%)	CGD = 44.3 (14.1) TMD/CGD = 45.1 (16.2)	Centre for Balance and Rehabilitation Research, after enrolling in a local longitudinal cohort study	Distress Cognitions	DHI	TSK HADS	DHI was significantly correlated to TSK-17 in the TMD/CGD group (0.72) and CGD group (0.74). The DHI was also significantly correlated with HADS-Anxiety in the TMD/CGD (0.76) and CGD (0.76) groups.	11
Miura et al. (2017)	CS	Japan	591 (Group B, dizziness, and tinnitus = 75); Group D, dizziness only = 516)	Male Group B = 25 (33%) Group D = 174 (34%) Female Group B = 50 (67%) Group D = 342 (66%)	Group B = 55.23 (16.23) Group D = 55.30 (18.37)	Outpatient hospital	Distress	DHI	HADS	Group B, sig correlations between DHI and sub scores of HADS-A (0.27) but not HADS-D (0.11). Group D, sig correlated between DHI and sub scores of HADS-A (r=0.32) and HADS-D (r=0.28).	12
Miyazaki et al. (2017)	CS	France & Japan	114 MD who underwent MIAUV surgery (French study only)	M 43 (38%) F 71 (62%)	51 (range 23-76)	Hospital	Distress	DHI	SAST	Pre-operatively the SAST and DHI were significantly related, with higher SAST category (SAST>23) scores associated with higher DHI scores	9
Monzani et al. (2001)	CS	Italy	207 Mixed	M 59 (29%) F 147 (71%)	M 52.8 F 55.6	Hospital Centre for Vestibular Testing	Distress	UCLA-DQ	HADS	Anxiety (0.205) and depression (0.182) sub scores were sig correlated with UCLA-DQ total scores. Patients with HADS>8 on either HADS-A or HADS-D had sig higher UCLA-DQ total score, mainly due to higher fear of dizziness sub scores.	12
Nazareth et al. (1999)	L	UK	193 Primary care with dizziness	M 69 (36%) F 124 (64%)	NK	London general practices	Distress Behaviour	Handicap Scale (0-6)	Anxiety and Avoidance Scale (0-6)	Significant univariate predictors of chronic handicapping dizziness included anxiety and avoidance at baseline. On logistic regression, only the presence of vertigo, fainting, and avoidance were identified as independent predictors	8
Obermann et al. (2015)	L	Germany	1159 patients who underwent inter-disciplinary	M 496 (39%) F 776 (61%)	61.1 (16.5)	Tertiary neuro-otology institution	Distress	DHI	ADS STAI	Multivariate analysis did not identify either anxiety or depression as sig risk factor for change in DHI score 2 years following inter-disciplinary treatment.	15

Ref.	Design	Country	Participants	Sex N (%)	Age Mean (SD)	Setting	Theme	Dizziness measure	Psych measure	Key findings	Quality score
			treatment for dizziness								
Pavlou et al. (2004)	Trial	UK	40/45 Mixed	M 12 (30%) F 28 (70%)	Gp 1= 43.8 (3.3) Gp 2 = 43.0 (2.6)	RCT	Distress	VSS SCQ	HADS VSS-A	When collapsing all patients' scores independent of group, improvements in anxiety (HADS-A) only correlated with improvements in visual vertigo (r=0.53). Depression and autonomic/somatic anxiety (VSS-A) symptoms significantly correlated with improvements in both visual (HAD-D r=0.57; VSS_A r=0.70) and global vertigo (HAD-D r=0.36, VSS-A r=0.37)	13
Pavlou et al. (2013)	Trial	UK	45/60 Mixed	M 14 (23) F 46 (77%)	Group 1: 47.5 Group 2: 49.3 Group 3: 46 range 28-73	RCT	Distress	SCQ VSS	BDI BAI VSS-A	When collapsing all patients scores independent of group, anxiety score improvements correlated with SCQ improvement. VSS-A improvements significantly correlated with both SCQ (r = 0.34; P < .05) and VSS-S (r = 0.33; P < .05) improvement.	15
Piker et al. (2008)	CS	USA	63 Mixed	M 18 (29%) F 45 (71%)	55 (14)	Balance clinic	Distress Behaviour	DHI VSS	HADS WOCQ	DHI correlated with autonomic arousal (r=0.52), anxiety (r=0.52) and depression (r=0.60). DHI total score showed weak to moderate positive correlations with all the WOCQ subscales (r=0.25 - 0.49) with the exception of Distancing and Planful Problem Solving. The highest correlation was between the Escape/Avoidance (r=0.49). No coping strategy was negatively correlated with less dizziness handicap.	10
Piker et al. (2015)	CS	USA	205 Mixed	M 80 (39%) F 125 (61%)	56.4 (14.8)	Vestibular disorders laboratory	Distress	DHI	HADS	DHI correlated with anxiety (r=0.542), depression (r=0.623) and total HADS (r=0.654)	12
Pollak et al. (2012)	L	Israel	37 idiopathic BPPV	M 14 (38%) F 23 (62%)	59.2 (14.5)	Outpatient clinic	Distress Cognitions	DHI	IPQ-R IUS STAI	DHI functional and emotional subscales were correlated with IPQ-R consequences scale (r=0.3 for both) and state anxiety (0.4). DHI emotional subscale was also correlated with trait anxiety (0.3) and uncertainty score (0.3). They omitted the physical subscale from analysis. All items of the IPQ-R except for disease predictability and belief in personal control did not change significantly after treatment for BPPV	11
Pothier et al. (2018)	CS	Canada	457 Mixed	M 154 (34%) F 303 (66%)	53.4 (15.4)	Neurotology outpatient clinic	Cognitions	DHI	DCS PANAS	Positive correlation between catastrophising and handicap (r=0.67) and moderate to strong associations across diagnostic classifications. Catastrophising remained independently associated with handicap after positive and negative affectivity was entered into a regression model, accounting for 47.1% of the variance.	11
Probst et al. (2017)	L	Germany	111 Mixed	M 35 (32%) F 76 (68%)	53.55 (15.26)	Centre for Vertigo & Balance Disorders	Distress	VHQ VSS	BDI-II BAI PHQ-15	Vertigo Handicap at T2 was sig correlated with T1 measures of depression (0.54), Anxiety (0.61) and Somatization (0.54). In single mediation models, the effect vertigo symptoms at baseline exerted on vertigo-related handicap at 12 months follow up was significantly mediated by depression, anxiety as well as somatization. When adjusting for other mediators in a multiple mediator model, only depression at 6-month follow up mediated the effect of vertigo symptoms at baseline on vertigo-related handicap at 12-month follow up.	13
Radziej et al. (2015)	CS	Germany	343 Mixed (185 with 'organically explained' & 158 with functional vestibular symptoms)	M 141 (41%) F 202 (59%)	55.96 (16.49)	Tertiary centre for vertigo & balance disorders	Distress	VHQ VSS	CTQ PDS IES	Regression analysis across groups revealed that, regardless of their diagnosis, prior traumatic life events on the PDS checklist, was associated with vertigo symptoms (VSS-V). Beyond that, vertigo-balance symptoms were predicted by the posttraumatic stress symptom clusters 'avoidance' and 'intrusion'. Handicapped activity (VHQ) showed only an association with one of the symptom clusters (avoidance). Amongst childhood events, emotional abuse and emotional neglect showed the strongest link to VS-related variables. Multiple linear regression revealed that the selected trauma measures	14

Ref.	Design	Country	Participants	Sex N (%)	Age Mean (SD)	Setting	Theme	Dizziness measure	Psych measure	Key findings	Quality score
										accounted for 6-9% of the variance in VSS subscales and the VHQ anxiety subscales, but the model predicting restrictions of physical and social activity (VHQ-ACT) failed to reach significance.	
Radziej et al. (2018)	CS	Germany	210 Mixed	M 91 (43%) F 119 (57%)	57.4 (15.6)	Tertiary centre for vertigo & balance disorders	Distress	VSS	BDI-II BAI	Vertigo symptoms were sig correlated to depression (r=0.31) and anxiety (0.35) at baseline	12
Roh et al. (2017)	CS	Korea	456 Mixed (acute 327, chronic 127)	M 146 (32%) F 310 (68%) Acute (111:216) Chronic (35:92)	55.8 (16.97) Acute 65.25 (17) Chronic 54.79 (16.70)	Outpatient dizziness clinic	Distress	DHI	HADS	Acute dizziness (<4 weeks) anxiety (r=0.454), depression (0.513) and total HADS (r=0.521) all correlated with DHI. Chronic dizziness (>4 weeks) anxiety (0.484), depression (r=0.535) and total score (r=0.533) all correlated with DHI. When patients were subdivided by the severity of dizziness symptoms using the DHI, the total and subscale HADS scores were significantly higher in patients with severe symptoms for both acute and chronic dizziness. For both patients with acute and chronic dizziness, patients above the cut-off for anxiety and depression (>7) and total score (>10) had sig higher level of DHI.	12
Saman et al. (2014)	CS	UK	63 untreated VS	M 36 (57%) F 27 (43%)	53.4 (11.8)	Neuro-otology department	Distress	VHQ	VSS-A	Sig correlation between the VHQ and the VSS-A (r=0.59). Hierarchical regression with VHQ as dependent variable showed autonomic and somatic symptoms of anxiety contributed to the model.	13
Saman et al. (2016)	Trial	UK	Exp 1 - 15 post-resection VS Exp 2 – 37 with VS in situ)	M (Exp 1) - 7 (47%) F (Exp 1) - 8 (53%)	Exp 1 - 50.33 (9.24)	2 Hospitals	Distress	VHQ VSS	STAI	Exp 1 - there was a big difference when comparing STAI at baseline and at the peak of the subjective vertiginous response in post-resection patients with a unilateral vestibular deafferentation. Exp 2 - VS in situ patients with balance symptoms had significantly worse state anxiety at the peak vertiginous response than patients without balance symptoms (according to VSS-VER), as did patients with a balance-related handicap. Correlation was found between peak caloric stimulation STAI-Y scores and the VSS-VER (r=0.61) and the VHQ (r=0.63).	10
Schmid et al. (2018)	Trial	Switzerland	32 Mixed - Split into 2 groups (with and without imbalance)	M 9 (28.13) F 23 (71.88)	Dizziness only = 44.8 (12.1) Dizziness and imbalance group = 60.6 (8.3)	Department of Psychosomatic Medicine	Psychological distress and psychiatric disorders	DHI	BSI	Sig correlations existed between the BSI phobic anxiety pre- and post-therapy and DHI for the dizziness only and the dizziness + imbalance groups, although the dizziness only regression showed a higher correlation (r=0.71 vs 0.57)	12
Schmid et al. (2020)	Trial	Switzerland	40 Mixed - receiving group CBT and VR for dizziness split into 2 groups (with and without balance deficit)	M 14 (35%) F 26 (65%)	Dizziness only group - 45.6 (14.0) Balance deficit group - 60.1 (9.9)	Department of Psychosomatic Medicine and ORL - patients undergoing multimodal intervention trial	Psychological distress and psychiatric disorders	DHI	BSI	All items of the BSI were correlated with the DHI, apart from the paranoid ideation scale in the balance deficit group, but the strongest correlations for both groups were for the BSI sub-scores phobic-anxiety (r=0.69/r=0.55) and obsessive/compulsive behaviour (r=0.62/r=0.47). Phobic anxiety was the best correlated variable with DHI scores in both groups, which explained 30% of the variance in DHI in the balance deficit group, and 55% of DHI variance in the dizziness only group	12
Soderman et al. (2001)	CS	Sweden	77 MD	NK	NK	2 x Departments of Otolaryngology and Audiology	Personality	VSS	SOC	Lower SOC was independently associated with vertigo symptom severity after controlling for gender, age, treatment, and duration	10

Ref.	Design	Country	Participants	Sex N (%)	Age Mean (SD)	Setting	Theme	Dizziness measure	Psych measure	Key findings	Quality score
Sugaya et al. (2017a)	CS	Japan	252 Mixed (inpatients)	M 73 (29%) F 179 (71%)	62.6 (16.6)	Inpatient medical centre	Behaviour	DHI	PSQI-J, >5 HADS	The prevalence of sleep disturbance was 65.1% in the participants and women showed a higher score than men. There was a significant interaction between sleep disturbance and sex in DHI total score. The scores were higher in women with sleep disturbance than in men with sleep disturbance. In addition, the presence of sleep disturbance was associated with severe anxiety and depression	10
Sugaya et al. (2017b)	L	Japan	127 Mixed inpatients	M 28 (22%) F 99 (78%)	62.55 (17.23)	Inpatient medical centre	Behaviour	DHI	PSQI-J HADS	1 month after an intensive 5-day in-patient vestibular rehabilitation, some participants (19.7%) recovered from sleep disturbance. Chronic dizziness patients with sleep disturbance at T2 had significantly higher DHI and HADS scores at T2 than patients without sleep disturbance at T2, after adjusting for these scores at T1.	11
Toshishige et al. (2020)	Trial	Japan	37 FVD attending a group CBT study	M 11 (30%) F 26 (70%)	41.1 (13.3)	Dept of Otolaryngology & Department of Psychiatry	Distress	DHI	HADS	Presence or absence of comorbid anxiety disorders (p = .023) was a significant positive predictive factor for improvement of DHI from pre-treatment to 6-month follow-up.	14
Tschan et al. (2008)	CS	Germany	188 Mixed	M 97 (52%) F 91 (48%)	50.5 (14.1)	Outpatient Neurology Department	Distress	VSS	HADS GSI VSS-A	Vertigo related symptoms (VSS-VER) was correlated to autonomic anxiety (r=0.45), HADS-A (0.21), GSI global severity index (0.29), but not to HADS-D (0.18)	12
Tschan et al. (2013)	L	Germany	92 SVD - 65 followed up	M 48 (52%) F 44 (48%)	41.8 (13.3)	Outpatient hospital	Distress	VHQ VSS	GSI VSS-A	Dividing the study participants into 3 categories, whether the VHQ worsened (n=20), stayed the same (n=21) or reduced by 15 or more (n=20) to compare vertigo handicap at baseline to 3 year follow up, found that the patients with increased handicap also increased in distress and differed significantly from the patients with decreased handicap. The patients with increased handicap also had increased vertigo symptoms and autonomic anxiety and differed sig from the patients with decreased handicap.	15
Von Rimscha et al. (2013)	CS	Switzerland	208 Mixed	M 111 (53%) F 97 (47%)	45.2 (11.8)	Interdisciplinary treatment centre for vertigo/dizziness	Personality	DHI	TAS-20 HADS	Sig positive correlation between TAS-20 total score and the DHI total score (r=0.30). Regarding the TAS-20 subfactors, there were significant positive correlations between factor 1 (difficulties in identifying feelings, r=0.29) and factor 2 (difficulties in describing feelings, r=0.29) with the DHI total score as well as the emotional and functional scale scores, whereas factor 3 (externally oriented thinking) did not correlate with DHI.	11
Weidt et al. (2014a)	CS	Switzerland	177 Mixed	M 86 (49%) F 91 (51%)	44.4 (11.9)	Interdisciplinary Centre for Vertigo and Balance Disorders	Distress Cognitions	DHI	PRISM HADS	DHI was correlated with HADS (r=0.56) and PRISM (-0.56)	11
Weidt et al. (2014b)	CS	Switzerland	203 Mixed	M 97 (48%) F 106 (52%)	44.6 (12.0)	Interdisciplinary treatment centre for vertigo/dizziness	Distress	DHI	HADS	DHI was sig correlated with HADS (r=0.58)	12
Wolf et al. (2020)	CS	Germany	419 Mixed	M 194 (46%) F 225 (54%)	53.5 (15.5)	Centre for Vertigo and Balance Disorders	Cognitions Distress	VHQ	IPQ-R VSS-A BAI BDI-II	Significant correlations were found between VHQ and IPQ-R scales Perceived Consequences (r=0.62), Emotional Representations (r=0.46), and weak correlations with Timeline (r=0.24), and Cyclic Timeline (r=0.27). The VHQ also correlated moderately with BDI-II (r=0.54) and BAI (r=0.58). Regression analysis indicated that the most important predictor of VHQ was Perceived Consequences. Emotional Representations showed to be a significant predictor of VHQ as well, although other illness perceptions	16

Ref.	Design	Country	Participants	Sex N (%)	Age Mean (SD)	Setting	Theme	Dizziness measure	Psych measure	Key findings	Quality score
										subscales did not reveal additional explanatory value. Weak significant correlations were found between VSS-VER and IPQ-R scales Personal Control (r=0.35), cyclical timelines (r=0.23) and emotional representations (0.20). The VSS-VER also correlated weakly with BDI-II (r=0.25) and BAI (r=0.31).	
Yan et al. (2020)	CS	China	70 AUV	M 34 (49%) F 36 (51%)	47.2 (17.1)	Department of otolaryngology	Distress	DHI	HADS	DHI score at the acute stage correlated with HADS (total score) across all age groups (r=0.597)	12
Yanik et al. (2008)	CS	Turkey	103 BPPV	M 35 (34%) F 68 (66%)	51.7 (8)	Outpatient clinic	Distress	VSS VDI	BDI	VSS was correlated with BDI (r=0.55) VDI-SS was correlated (low) sig with BDI (r=0.2)	11
Yardley et al. (1992c)	CS	UK	127 Mixed	M 50 (39%) F 77 (61%)	46.5	Neuro-otology clinics	Distress Personality	VHQ	HADS STAI-T VSS-A	Handicap was assoc with autonomic symptoms (r=0.35), trait anxiety (r=0.44) and HADS (r=0.46). Multiple regression showed handicap was assoc with higher levels of autonomic and vertigo symptoms, together with anxiety and depression, which together accounted for 51% of the total variance. Although current emotional state (anxiety state) was highly correlated with level of handicap, there was no direct assoc between anxiety personality and handicap scores after controlling for the other variables.	12
Yardley et al. (1992a)	CS	UK	127 Mixed	M 50 (39%) F 77 (61%)	46.5	Neuro-otology clinics x 2	Distress Personality	VHQ	HADS STAI-T VSS-A	Handicap was assoc with autonomic symptoms (r=0.35), HADS-A (0.41), HADS-D (0.42), trait anxiety (0.44) and somatisation (-.31). Vertigo severity, autonomic arousal and depression combined to explain a total of 42.3% of the variance in handicap scores. After controlling for these variables, the influence of trait and state anxiety did not quite reach significance	11
Yardley and Putman (1992)	CS	UK	84 Mixed	M 30 (36%) F 54 (64%)	48	Audiology department	Distress	VHQ	3-item subscale measuring anxiety and depression	Anxiety/depression was correlated with VHQ (0.54) and all the VHQ subscales.	6
Yardley (1994b)	L	UK	101 Mixed	M 42 (42%) F 59 (58%)	M 51.6 F 45.2	2 outpatient neuro-otology clinics	Distress Behaviour	VSS VHQ	Vertigo Specific Coping Questionnaire MBSS MHLC HADS VSS-A	At both T1 and T2 (7 month), handicap was correlated with HADS (r=0.44, 0.43), autonomic symptoms (r=0.40, 0.47), internal locus of control (MHLC; -0.25, -0.18), distraction (0.17, 0.24), and relinquishing responsibility (0.42, 0.34). In regression analysis at T1, the partial correlations, controlling for all other variables, indicated that vertigo severity, distress, and relinquishing responsibility made the most significant contributions to variance in handicap. The assoc between relinquishing responsibility and handicap remained significant after controlling for locus of control, and so did not appear to be mediated entirely by control beliefs. Another stepwise regression looked at which variables significantly related to residualised handicap at T2, and found that only autonomic symptoms (VSS) and HADS scores at T1 were significantly correlated with residualised T2 handicap	13
Yardley (1994a)	L	UK	101 Mixed	M 42 (42%) F 59 (58%)	M 51.6 F 45.2	Outpatient neuro-otology clinics	Distress Cognitions	VSS VHQ	HADS DBS VSS-A	Raw handicap scores at 7 months correlated with autonomic symptoms (r=0.32) and HADS (r=0.42). Residualised handicap scores at 7 months were still correlated with HADS (r=0.23) but no longer related to autonomic symptoms. Cross-sectional results comparing only T2 measures found residualised VHQ was related to autonomic symptoms (r=0.40) and HADS (0.37). The beliefs questionnaire measures at T2 identified losing control was related to both raw (0.42) and residualised (0.22) handicap. Only serious illness was	10

Ref.	Design	Country	Participants	Sex N (%)	Age Mean (SD)	Setting	Theme	Dizziness measure	Psych measure	Key findings	Quality score
										not related to either raw or residualised handicap. Although belief in the possibility of a severe attack was correlated with raw handicap severity, this relationship was insignificant after controlling for handicap at T1. The relationship between loss of control, autonomic symptoms and handicap remained significant even after controlling for somatization, vertigo severity, anxiety and depression in a multiple regression analysis. A second regression analysis revealed that the sub factor social incompetence accounted for the relationship with handicap and belief in physical danger was unrelated.	
Yardley et al. (1994)	L	UK	101 Mixed	M 42 (42%) F 59 (58%)	M 51.6 F 45.2	Outpatient neuro-otology clinics	Distress Personality	Perceived change in severity of vertigo (single question item)	HADS STAI-T VSS-A	Perceived change in severity of vertigo measured at 7 months by response to a single question was assoc with baseline autonomic symptoms (0.34), emotional distress (r=0.29) but not with trait anxiety. The multiple regression analysis showed that initial levels of vertigo, handicap, trait anxiety and distress were unrelated to perceived change in severity of vertigo, but that perceived change was strongly related to changes in symptoms and psychosocial status between Time 1 and Time 2. Autonomic symptoms at both T1 and T2 proved to be the only longitudinal predictor of perceived change in vertigo severity. Measures of balance function were not related to outcome.	9
Yardley et al. (1998d)	CS	UK	480 Mixed	M 150 (31%) F 325 (68%)	Not reported	4 London primary care centres	Behaviour Distress	Handicap survey questionnaire validated for study	Anxiety & avoidance survey questionnaire (developed for study)	Handicap levels increased significantly with co-morbidity between dizziness, anxiety and avoidance behaviour	9
Yardley et al. (1999)	CS	Mexico	172 Mixed	M 42 (24%) F 130 (76%)	46.5 (14.2)	Outpatient hospital	Distress	VSS VHQ	BAI BDI VSS-A	Handicap VHQ was associated with autonomic symptoms (0.38), BAI anxiety (0.40) and depression (0.38). VSS-VER was assoc with autonomic symptoms (0.46), BAI Anxiety (0.18) but not depression (0.10). In a stepwise multiple regression with VHQ as the dependent variable, the resulting equation explained 24% of the variance in handicap with vertigo, somatic anxiety, and depression being independently related to handicap.	11
Yardley et al. (2001)	L	UK	76 Mixed (33 assigned to treatment)	M 18 (24%) F 58 (76%)	60.2 (13.9)	Primary care RCT	Cognitions Distress	VHQ-8 VSS	DBS VSS-A	Handicap at follow up was associated with baseline belief (r=0.55) and autonomic symptoms (r=0.39). Symptom severity at follow up was associated with baseline beliefs (r=0.35) and autonomic symptoms (r=0.66). A regression showed that baseline handicap, vertigo and autonomic symptoms and beliefs explained nearly 50% of the variance in handicap at 6 months follow up. Baseline beliefs proved to be the only predictor of change in handicap, accounting for 4.4% of the variance in handicap at follow up after controlling for baseline handicap and symptoms	11
Zhu et al. (2020)	L	China	131 BPPV 45 Vestibular Migraine	M BPPV - 43 (33%) VM - 13 (29%) F BPPV 88 (67%)	BPPV - 50.86 (13.47) VM - 49.0 (12.98)	Neurology department	Distress	DHI	HADS	Change in DHI scores in BPPV patients was positively correlated with changes in HADS (r=0.591). Change in DHI in VM patients was also positively correlated with changes in HADS (r=0.556)	13

Ref.	Design	Country	Participants	Sex N (%)	Age Mean (SD)	Setting	Theme	Dizziness measure	Psych measure	Key findings	Quality score
				VM 32 (71%)							

Notes: NK = not known.

Study design: *CS = cross-sectional; L = longitudinal.*

Conditions: *AUV = acute unilateral vestibulopathy; BPPV = benign paroxysmal positional vertigo; CBT = cognitive behavioural therapy; CGD = cervicogenic dizziness; CSD = chronic subjective dizziness; FVD = functional vestibular disorder; MD = Meniere's disease; MdDS = mal de debarquement syndrome; MIAUV = minimally invasive vestibular neurotomy; mTBI = mild traumatic brain injury; PVD = peripheral vestibular disorder; SVD = somatoform vertigo and dizziness; VM = vestibular migraine; VR = vestibular rehabilitation; VS = vestibular schwannoma.*

Psychological instruments: *ACQ = agoraphobic cognitions questionnaire; ADS = general depression scale; ASI = anxiety sensitivity index; BAE = beliefs about emotions scale; BAI = beck anxiety inventory; BDI = beck depression inventory; BSI = brief symptom inventory; BSQ = body sensation questionnaire; CAS = clinical anxiety scale; CBRQ = cognitive-behavioural responses to symptoms questionnaire; CES-D = center for epidemiologic studies depression scale; CTQ = childhood trauma questionnaire; DASS-21 = depression, anxiety and stress scales-21; DBS = dizziness beliefs scale; DCS = dizziness catastrophising scale; DD = depersonalisation - derealisation inventory; DES = dissociative experiences scale; DSM-IV = structured clinical interview; FKV = freiburg coping illness questionnaire; GAD7 = generalised anxiety disorder scale; GCS = generalized contentment scale; GHQ-12 = general health questionnaire; GSI = global severity index; HADS = hospital anxiety & depression scale; HDRS = hamilton depression rating scale; IES = impact of events scale; IPQ-R = illness perception questionnaire revised; ISI = insomnia severity index; IUS = intolerance of uncertainty scale; KLC = body-related locus of control questionnaire; MBSS = miller behavioural style scale; MHLC = multidimensional health locus of control; MI = mobility inventory; MINI = mini international neuropsychiatric interview; MUIS = mishel uncertainty in illness scale; NEO-PI-R = NEO personality inventory; NQ = Nijmegen questionnaire; PANAS = positive and negative affective scale; PCL = PTSD checklist; PCS = pain catastrophizing scale; PDS = posttraumatic diagnostic scale; PHQ = patient health questionnaire; PRISM = pictorial representation of illness and self-measure; PSQI = pittsburgh sleep quality index; PSSI = personality type and disorder inventory; PVS = psychological vulnerability scale; RSE = rosenberg's self-esteem scale; SAS = zung instrument for anxiety disorders; SAST = short anxiety screening test; SBI = safety behaviours inventory; SCL-90R = symptom checklist-90R; SDS = zung self-rating depression scale; SHAI = short-form health anxiety inventory; SOC = sense of coherence scale; SSAS = somatosensory amplification scale; SSCI = stigma scale for chronic illness; SSCS = somatosensory catastrophising scale; SSQ = social support questionnaire; STAI = spielberger state-trait anxiety inventory; TAS-20 = toronto alexithymia scale; TSK = tampa scale for kinesophobia; VAAI-9 = vestibular activities avoidance instrument; VCQ = vertigo coping questionnaire; VSS-A = vertigo symptom scale - autonomic subscale; WOCQ = ways of coping questionnaire.*

Dizziness questionnaires: *DHI = dizziness handicap inventory; DSI = dizziness symptoms inventory; VAS = visual analogue scale; KADL = Korean version of the vestibular disorders activities of daily living scale; SCQ = situational characteristic questionnaire; SIP = sickness impact profile; IIRS = illness intrusiveness ratings scale; University of California Los Angeles dizziness questionnaire; VDI = vertigo dizziness imbalance symptom scale; VHQ = vertigo handicap questionnaire; VSS = vertigo symptom scale; VAP vestibular activities and participation*

Distress

Anxiety & Depression

This section focuses on generic self-report measures of anxiety and/or depressive symptoms.

Group differences

Studies which used recognised cut-off values for anxiety and depression showed that patients who score above these cut-offs have significantly higher handicap (Cheng et al., 2012; Maarsingh et al., 2011; Menant et al., 2020; Miyazaki et al., 2017; Roh et al., 2017; Yardley et al., 1998d; Zhu et al., 2020), vertigo severity (Kirby & Yardley, 2009a) and impairment in daily activities, quality of life, and fear of becoming dizzy (Honrubia et al., 1996; Monzani et al., 2001).

Cross-sectional studies

In terms of correlational studies, the average sample size-weighted correlation (r) with handicap scales was 0.52 (95% CI = 0.47 to 0.58) for anxiety [Figure 7A]; 0.55 (95% CI = 0.47 to 0.62) for depression [Figure 7B]; and 0.56 (95% CI = 0.53 to 0.60) for combined anxiety and depression scales [Figure 7C]. There was significant heterogeneity between studies, but analysis of the dataset run on different subgroups showed large effects for each measurement tool which corresponded to the effect estimation based on the whole dataset. All but one study used the Hospital Anxiety & Depression Scale (HADS) to measure combined anxiety and depression. One study (Yardley & Putman, 1992) used a shortened three-item scale of anxiety and depression but removing this did not change the estimate.

For vertigo severity as measured by the Vertigo Symptom Scale (vertigo subscale) there was also a positive relationship with anxiety (pooled correlation r = 0.35, 95% CI = 0.29, 0.40 and depression (r = 0.37, 95% CI = 0.33, 0.41).

One study also experimentally induced a vestibular stimulus by means of caloric assessment in patients with a vestibular schwannoma and found state anxiety at peak caloric stimulation correlated with the severity of symptoms and overall handicap (Saman et al., 2016).

Longitudinal studies

One study found no prospective correlation between HADS recorded during the acute phase of an acute unilateral vestibulopathy and handicap at 10 weeks (Cousins et al., 2017). Other prospective studies recruited mixed diagnosis samples from outpatient clinics, where the majority would be in the chronic phase, and found that anxiety and depression were correlated with unadjusted handicap scales at three months (Herdman et al., 2020a) and residualised handicap at seven months (Yardley, 1994a, 1994b) but

were not a strong predictor in the respective regression models (Herdman et al., 2020a; Yardley, 1994b).

Only one prospective study conducted mediation analysis and found that anxiety and depression, as well as the presence of other somatic symptoms explained (mediated) the relationship between vertigo symptoms and handicap at twelve months when tested in single mediation models (Probst et al., 2017). When they were explored in a parallel model, depression was the only mediator between vertigo symptoms and handicap to reach significance.

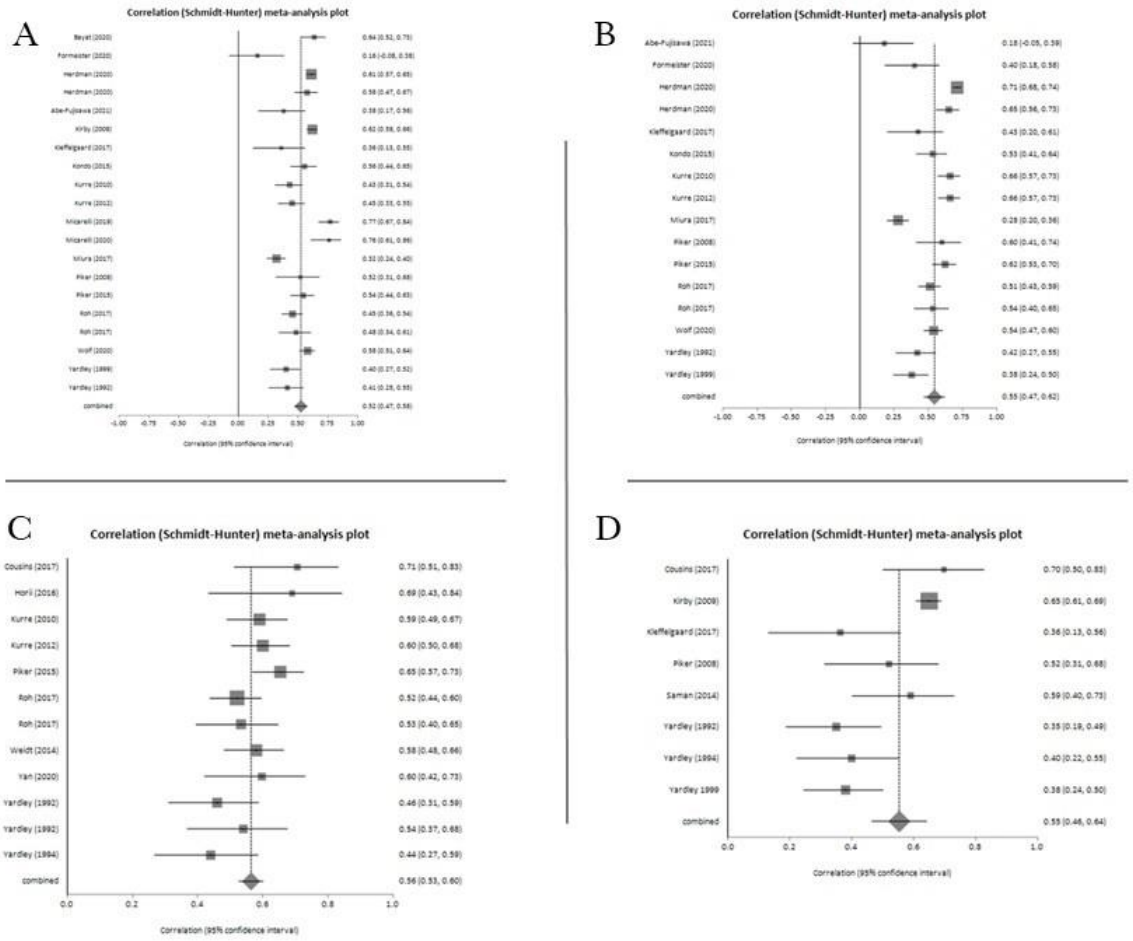

Figure 7. Meta-analysis plots for sample size weighted mean of the correlation coefficient between dizziness handicap and (A) anxiety; (B) depression; (C) combined anxiety-depression; (D) autonomic symptoms

Intervention studies

Studies which have measured anxiety and depression in the context of intervention trials found anxiety and depression remained related to handicap before and after multidisciplinary interventions (Hong et al., 2012) and change in handicap scores was positively correlated with changes in anxiety and depression following standard treatments for Meniere's Disease and BPPV (Zhu et al., 2020), following CBT (Schmid

et al., 2018; Schmid et al., 2020) and anti-depressant medication for chronic dizziness (Horii et al., 2007) and improvement in depression, but not anxiety, correlated with improvements in symptom severity after vestibular rehabilitation (Pavlou et al., 2004).

Higher levels of baseline anxiety were associated with worse handicap outcomes following CBT in one study (Mahoney et al., 2013) although the presence of anxiety was a predictor of long-term effectiveness of CBT in another (Toshishige et al., 2020). Likewise, Goto et al. (2017) found higher baseline anxiety and depression was associated with improved handicap following in-hospitalised vestibular rehabilitation. These differences may be due to the different format and content of the interventions.

Autonomic arousal

Dizziness handicap might also be affected by symptoms related to the somatic manifestations rather than the more explicit cognitive/affective aspects of anxiety, since autonomic arousal may be implicated in enhanced generation and awareness of vestibular symptoms.

Cross-sectional studies

This is demonstrated by the consistent interrelations between the vertigo and autonomic subscale of the vertigo symptom scale (Kleffelgaard et al., 2017; Tschan et al., 2008; Yardley et al., 1992a; Yardley et al., 1999). The average sample size-weighted correlation (r) between autonomic symptoms and dizziness handicap was 0.55 (95% CI = 0.46 to 0.64) [Figure 7D] although there was significant heterogeneity.

One study specifically looked at the role of hyperventilation using the Nijmegen Questionnaire and found a significant positive correlation with handicap (rho = 0.348) (Humphriss et al., 2004).

Longitudinal studies

Prospective studies also found autonomic anxiety symptoms were prospectively associated with perceived change in vertigo symptom severity (Yardley et al., 1994) and handicap (Cousins et al., 2017; Tschan et al., 2013; Yardley, 1994a, 1994b; Yardley et al., 2001). Acute autonomic arousal was also a key predictor of handicap following unilateral vestibulopathy (Cousins et al., 2017). Improvements in autonomic symptoms also correlated with improvements in vertigo symptoms during rehabilitation (Pavlou et al., 2013; Pavlou et al., 2004).

Health anxiety

Health anxiety involves generic worry about illness and greater focus towards somatic sensations. One cross-sectional study found that that dizziness handicap was associated with health anxiety (r=0.45) in members of a patient support group for Meniere's Disease which remained significant after adjusting for demographic, illness and other psychological variables (Kirby & Yardley, 2009b).

Depersonalisation / Derealisation

Derealisation and depersonalisation (DD) include having a sense of unreality and detachment. They can accompany a wide variety of neurological, vestibular and psychiatric conditions as well as occur as a chronic primary mental health disorder (Hunter et al., 2017).

Only one study examined a small clinical sample of patients (n=10) following acute vestibulopathy and found high prevalence of DD but no relationship between the differences on DD symptoms and the differences in symptom severity or handicap over three months (Gomez-Alvarez & Jauregui-Renaud, 2011).

Life Events

Several models have been proposed to explain the relationship between adverse life events and persistent symptoms. These models draw on 'stress-systems' framework and emphasise that potentially traumatic events early in life can alter how an individual responds to current stress and result in other mental health sequelae in a complex way (Kozlowska, 2013).

One cross-sectional study found that patients with either 'medically explained' or 'functional' vestibular symptoms did not differ regarding the number of traumatic experiences (Radziej et al., 2015). Regardless of whether symptoms could be medically explained or not, childhood trauma predicted to some extent the variance of dizziness outcomes, mainly those associated with symptom severity and the emotional subscale of the dizziness handicap inventory, although the multiple regression model predicting activity related handicap failed to reach significance.

Psychiatric morbidity

One cross-sectional study of patients with Meniere's disease who were members of a patient support group found that symptoms of PTSD (post-traumatic stress disorder) were significantly associated with dizziness handicap (r=0.65), which remained significant after adjusting for demographic variables and symptom severity (Kirby & Yardley, 2009b). In their final regression model PTSD symptoms contributed most to handicap.

Schmid et al. (2020) found that BSI sub-scores, a screening tool for psychopathology, were positively correlated when regressed with handicap in a group of participants recruited to a trial of CBT and VR. Phobic anxiety explained 30% of the variance in dizziness handicap in the group of participants with a 'quantified balance deficit' and 55% of the variance with the addition of obsessive/compulsive behaviour and perceived state of health scores in the group who had 'dizziness only'.

Personality and resilience factors

This theme explores whether personality – that is, relatively stable individual differences– have important implications for dizziness outcomes. Although the degree to which individual personality is malleable is unclear, there is good evidence that personality does change in response to new environments and life events (Segerstrom, 2019), suggesting that personality interventions are indeed possible and therefore deserve a place in this review.

Anxiety proneness

Anxiety sensitivity or trait anxiety, hereafter referred to as 'anxiety proneness' is reflected in most models of personality as a relatively stable tendency to perceive situations as dangerous or threatening (Spielberger, 1972). Six studies used the Spielberger's State-Trait Anxiety Inventory – Trait Scale (STAI-T) and one study used the Anxiety Sensitivity Index (ASI) which both measure the more cognitive aspects of anxiety proneness, and one study used the Positive and Negative Affect scale (PANAS) in a way to capture trait dimensions of mood and affect.

Cross-sectional studies

Neither of the two cross-sectional studies found consistent relationships between anxiety proneness and dizziness handicap. In one study, the relationship was no longer significant after controlling for state emotional distress and symptom severity (Yardley et al., 1992a; Yardley et al., 1992c). The other study only found a relationship between anxiety proneness (ASI) and emotional aspects of handicap (r=0.40), but not with the functional or physical subscales (Hägnebo et al., 1999).

Longitudinal studies

One study found that trait anxiety had a near zero correlation with perceived change in symptom severity seven months after attending an outpatient neuro-otology clinic although they only used a single question item to measure perceived improvement (Yardley et al., 1994). Another low-quality study also failed to demonstrate a relationship between anxiety proneness and symptom severity six months after an acute vestibular disorder (Godemann et al., 2004).

Intervention studies

Intervention studies show mixed support for a relationship between anxiety proneness and dizziness handicap either. One study found a medium correlation with an activities of daily living scale before (r=0.44) and three months after (r=0.37) interdisciplinary treatment, but they combined state and trait anxiety (Hong et al., 2012). Another study which combined state and trait anxiety Obermann et al. (2015) did not find a relationship with change in handicap two years after interdisciplinary treatment. A retrospective chart review found that whilst individuals with low positive and high negative trait required longer length of therapy intervention than the group with normal

'trait' affect, there were no significant differences in handicap scores before or after rehabilitation (MacDowell et al., 2018). Only one study found that those high in anxiety proneness at baseline benefitted more from vestibular rehabilitation (Goto et al., 2017).

Psychological vulnerability

Psychological vulnerability is conceptualised as specific ways of emotional processing and cognitive beliefs that reflect dependence on others for one's sense of self-worth that leaves individuals vulnerable to stress and maladaptive functioning (Sinclair & Wallston, 1999).

One study of patients due to attend a specialist clinic for vertigo and dizziness used the psychological vulnerability scale which relates to perceptions of dependency, perfectionism, negative attributions, and the need for external sources of approval. This study found that psychological vulnerability had a small to moderate correlation with handicap at baseline (r=0.32) (Herdman et al., 2020b) and remained correlated three months after the initial consultation (r=0.35) but was no longer significant after adjusting for baseline handicap (Herdman et al., 2020a).

Neuroticism

Neuroticism as a personality trait refers to a stable lifelong tendency to experience negative affect. Watson and Pennebaker (1989) suggested it be seen not just as psychological trait but as a more generic vulnerability to physical illness and psychological distress.

Secondary analysis of a six-month randomised controlled trial for community-living people aged 50 years and older found participants with elevated handicap (DHI 31+) had significantly higher neuroticism (Menant et al., 2020). Another study found a positive correlation between neuroticism and overall handicap in a small sample of patients with functional dizziness (r=0.53) but less so in a group with other peripheral vestibular disorders (r=0.37) (Chiarella et al., 2016).

Sense of coherence

The concept of sense of coherence (SOC) arose from the salutogenic models of health. It is described by Antonovsky as a trait that reflects a coping capacity to deal with life stressors made up of the extent to which events are perceived as making logical sense, to which a person feels they can cope and can find meaning (Antonovsky, 1979).

One study of members of a Meniere's Disease (MD) support group found that lower SOC scores were related to higher scores in MD-impact and more severe vertigo symptoms (Ketola et al., 2014). Soderman et al. (2001) sampled patients with Meniere's disease still experiencing symptoms from otolaryngology departments and also found that lower SOC was independently associated with symptom severity after

adjusting for gender, age, treatment, and symptom duration. One longitudinal sample, also of patients with MD refractory to intervention, found that the mean SOC scores were relatively high but people with lower SOC had more symptoms (r=-.46) (Green Jr et al., 2007). The SOC scores remained fairly stable and only the meaningfulness domain was significantly related with vertigo category over time.

One cross-sectional study used patients with mixed vestibular disorders but found similar results with patients with low self-reported SOC more likely to report higher handicap and other psychosocial impact, despite having similar symptom severity to those patients with high SOC (Mendel et al., 2001).

Alexithymia

Alexithymia refers to a personality trait characterised by difficulty in identifying, labelling and understanding emotions (Sifneos, 1973). It can be measured using the Toronto Alexithymia Scale. Only one cross-sectional study examined its relationship with dizziness handicap in patients with unspecified dizziness attending an interdisciplinary treatment centre and found a small to moderate positive correlation (r=0.30) (Von Rimscha et al., 2013). Specifically, there was a significant positive correlation between dizziness handicap and the subfactors relating to difficulty identifying (r=0.29) and describing feelings (r=0.29) but not with externally oriented thinking (a preoccupation with the minute details of external events).

Self esteem

Self-esteem reflects an individual's positive and negative views towards the self. It is usually viewed as a personality characteristic, although state variations can also exist. One study used Rosenberg's Self-Esteem Scale (RSE) but did not find any linear correlation with symptom severity in patients with dizziness attending a neuro-otology unit with and without vestibular lesions (Grunfeld et al., 2003).

Cognitions

Illness perceptions

The Common-Sense Model of Self-Regulation (CSM) proposes that when faced with a health threat people construct their own lay beliefs about their illness which can influence behavioural and emotional responses, and ultimately their psychological and physical outcome (Leventhal et al., 2016).

Four studies used the Illness Perceptions Questionnaire-Revised which is based on the CSM. Two high quality cross-sectional studies of patients with mixed vestibular disorders attending specialist centres both found that viewing the illness as having serious consequences was strongly related to handicap (r=0.61 & 0.62) (Herdman et al., 2020b; Wolf et al., 2020). Importantly, both studies included regression analysis which showed negative illness perceptions explain some of the variance in dizziness

handicap (Herdman et al., 2020b) and perceived consequences remained the most important correlate even after adjusting for demographic variables and symptom severity (Wolf et al., 2020). Other negative illness perceptions such as viewing the illness as upsetting, chronic, cyclical, and having multiple symptoms were also related to dizziness handicap. However, the degree to which the patient feels control over the illness and its cure was not associated with dizziness handicap in either study. In both studies symptom severity (VSS) had similar patterns of correlations as dizziness handicap, albeit weaker. Pollak et al. (2012) also found that it was the negative perceived consequences subscale was correlated with functional and emotional handicap subscales in people with BPPV.

In their naturalistic follow-up of patients three months after initial diagnostic consultation at a tertiary clinic, Herdman et al. (2020a) once again found that viewing the illness as having serious consequences was the strongest correlate of dizziness handicap at follow up, followed by viewing the illness as upsetting, causing multiple symptoms, and chronic in nature.

Locus of control

Health-related locus of control refers to the degree to which individuals' believe they have the power to control various factors that affect their health in general (Wallston et al., 1978). One study used the Body-Related Locus of Control Questionnaire, with its two subscales covering internal and external body-related locus of control, which refers to the concept of whether a person perceives that he or she has control over bodily symptoms (internal locus of control) or interprets the symptoms as by chance or due to outer influences that cannot be controlled by themselves. This mimics the 'personal control' subscale of the IPQ-R, which like in the previous studies, did not add predictive value to the variance explained in dizziness handicap, this time at 6 months following 'psychosomatic inpatient therapy' for patients with functional vestibular symptoms (Limburg et al., 2019).

The other study used the more commonly used Multidimensional Health Locus of Control (MHLC) questionnaire but only internal locus of control had a weak negative correlation with dizziness handicap at both time points, but was not associated with residualised dizziness handicap seven months after attending a neuro-otology clinic (Yardley, 1994b).

Weidt et al. (2014a) asked participants to depict the distance between their illness and themselves using the PRISM test, which is a measure of a patient's perception of the controllability of their illness. They found that patients with a small self-illness separation (the distance between the illness and the self) had higher levels of dizziness handicap (r=-.56). However, this instrument does not ask specifically about control beliefs and is itself used as an inverse measure of illness intrusiveness.

Interpretations of symptoms

Evidence suggests that interpretations of everyday symptoms, such as catastrophising about symptoms (believing in worst-case scenarios) or viewing symptoms as signs of biological damage are also relevant in physical illness.

Yardley (1994a) identified three common beliefs about dizziness amongst patients presenting to a specialist clinic: concern about losing control, fear of serious illness and anticipation of a severe attack. Belief that dizziness may result in a severe attack was weakly correlated with symptom severity and dizziness handicap, although the latter was no longer significant after adjusting for baseline handicap. There was no correlation with fear of serious illness but there was a correlation between handicap and belief in loss of control (r=0.42) which remained significant after controlling for baseline handicap. This could also be interpreted in line with illness perceptions related to serious consequences.

Yardley et al. (2001) also found that primary care patients with dizziness frequently endorsed the same beliefs about dizziness which were a significant predictor of handicap at six months after adjusting for baseline handicap and symptom severity. One other study used the same measure developed by Yardley to investigate group differences following an acute vertigo episode, but since only seven patients reported remaining symptoms, no firm conclusions could be drawn (Kammerlind et al., 2011).

One study found that pain catastrophising was related to higher dizziness handicap (Cuenca-Martinez et al., 2018), in addition to kinesiophobia (fear of movement), which was also strongly correlated with handicap in another study (Micarelli et al., 2020), although both studies used a diagnosis of 'cervicogenic dizziness' which does not have an accepted classification at this time. Evidence from a mixed diagnosis sample of patients comes from (Pothier et al., 2018) who adapted the well-established Pain Catastrophising Scale for dizziness (DCS) in a large retrospective record review and found a strong positive correlation between catastrophising and handicap (r=0.67) and moderate to strong associations across diagnostic classifications. Catastrophising remained independently associated with handicap after positive and negative affectivity was entered into a regression model, accounting for 47.1% of the variance. Gerretsen et al. (2020) also found that change in DCS scores was a predictor of percentage change in DHI scores following interdisciplinary psychiatric treatment for chronic dizziness.

Herdman et al. (2020b) also found that catastrophising about dizziness was strongly associated with dizziness handicap and symptom severity, using the Cognitive and Behavioural Responses to Symptoms Questionnaire (CBRQ). Other CBRQ cognitive subscales including focussing on symptoms, belief that dizziness is a sign of damage, and belief about the dangers (fear avoidance) and potential embarrassment (embarrassment avoidance) of undertaking activity were also highly correlated with

dizziness handicap and symptom severity. Fear avoidance was the only independent correlate in the fully adjusted model of symptom severity. All of the items remained correlated with dizziness handicap three months later (Herdman et al., 2020a).

Dunlap et al. (2020) evaluated the shortened Vestibular Activities Avoidance Instrument as a measure of fear avoidance and found it too was associated with greater activity limitations and participation restrictions in patients attending specialist balance disorder settings. In their follow up study three months later, baseline fear avoidance continued to be significantly associated with handicap (rs = 0.54), and symptom severity (rs = 0.37) (Dunlap et al., 2021).

Anxiety related cognitions

Catastrophising about symptoms related to anxiety and panic, also known as 'fear of fear', is hypothesised to be an important feature of avoidance behaviour and development of agoraphobia (Chambless & Gracely, 1989). Godemann et al. (2004) found that higher scores on the Agoraphobic Cognitions Questionnaire (ACQ) were associated with continuation of vertigo 6 months after acute vestibulopathy, which also contributed significantly to symptom severity. A year later vertigo symptom severity was still positively correlated with the ACQ total score (r=0.45) and both its subscales referring to physical concern (r=0.42) and loss of control (r=0.28) (Godemann et al., 2005).

One study did not find any difference in baseline anxiety related cognitions between patients with and without remaining symptoms three months following acute vestibulopathy but this study was limited due to the small group size (n=13) (Heinrichs et al., 2007).

Cognitions related to bodily symptoms

Anxiety related cognitions also include the degree to which patients fear physical symptoms commonly associated with anxiety, which at least in the case of the Body Sensations Questionnaire (BSQ) also includes dizziness.

One longitudinal study found that initial fear of bodily sensations was associated with handicap between ten weeks and ten months following unilateral peripheral vestibulopathy (Cousins et al., 2017). However, this correlation was no longer significant after adjusting for other baseline variables such as autonomic arousal and the extent to which the individual relies on visual input for spatial orientation (visual dependency).

Amongst the three studies that have looked at the relationship between fear of bodily sensations and symptom severity, one found a positive correlation (r=0.47) which contributed to the variance in symptom severity one year following unilateral peripheral vestibulopathy (Godemann et al., 2005). The other two studies also found

group differences between patients with and without remaining symptoms, but the BSQ was either not significantly related to symptom severity in multivariate analysis after six months (Godemann et al., 2004) or only in patients with a confirmed vestibulopathy after three months (Heinrichs et al., 2007) although both studies are limited due to small sample sizes.

One study used the Somatosensory Catastrophising Scale (SSCS) which is broader and perhaps more relevant to dizziness populations than the BSQ, in that it attempts to measure the degree to which participants pay attention to, interpret, and characterise general physical symptoms negatively. Thus, it is not only limited to anxiety related symptoms. This study reported that improvement in perceived handicap was worse in patients who catastrophised their bodily sensation before an inpatient vestibular rehabilitation programme (Goto et al., 2017).

Illness uncertainty

The model of uncertainty in illness is based upon a cognitive appraisal model and refers to an 'inability to determine the meaning of illness-related events' (Mishel, 1990, p. 256). Vestibular disorders are often characterised by unpredictable symptoms, unknown aetiology and diagnostic uncertainty that may be a key risk factor in adjustment to the condition.

One cross-sectional study measured illness uncertainty using the Mishel Uncertainty in Illness Scale amongst members of a UK Meniere's Disease support group and found it was weakly related with emotional (r=0.26) and functional (r=0.24) dizziness handicap, but not with physical handicap (Arroll et al., 2012).

Another cross-sectional study measured how members of a Meniere's Disease support group might interpret and respond to uncertainty using the Intolerance of Uncertainty Scale, which reflects the beliefs about the necessity of being certain and attempts to control future events. They found Intolerance of Uncertainty correlated with handicap (r=0.47) although the strength of the association was reduced after controlling for PTSD symptoms in the regression model (Kirby & Yardley, 2009b).

Stigma

The perceived stigma associated with vestibular conditions may contribute to poorer health outcomes. Corrigan et al. (2006) described a theoretical model of self-stigma as a process by which public attitudes lead to personal responses and ultimately, self-stigmatization.

One cross-sectional study recruited participants from a website for people with 'Mal de debarquement' (MdDS), a functional vestibular syndrome whereby individuals may feel stigmatised since they have a condition which is medically unexplained and largely invisible to those around them despite the constant sensation of rocking and swaying it

provokes. This study found stigma was association with symptom severity (r=0.38) but appeared to be negatively associated with illness intrusiveness (r=-0.59) (Arroll et al., 2016).

Coping and Behaviour

Coping

Coping is a key element of Lazarus and Folkman's (1984) transactional theory of stress and reflects the ways in which individuals interact with stressors in an attempt to return to normal functioning. Consistent with neurobiological models of 'compensation' following vestibular dysfunction, all of the studies in this section supported the role of avoidance behaviour also contributing to dizziness handicap (Heinrichs et al., 2007; Herdman et al., 2020a, 2020b; Nazareth et al., 1999; Piker et al., 2008; Yardley, 1994b; Yardley et al., 1998d), although the role of other styles of coping and the relationship over time is less clear.

Two cross-sectional studies used the Ways of Coping Questionnaire (WOCQ) to measure different coping styles, and both found the highest positive correlation with handicap was with escape/avoidance. Piker et al. (2008) found that dizziness handicap showed weak to moderate positive correlations with all the other WOCQ subscales (r=0.25 to 0.49) except for distancing oneself and planful problem solving. However, the other study did find that distancing, along with avoidance and less use of efforts to regulate one's feelings and actions, were associated with the functional subscale of dizziness handicap in members of a patient support group for Meniere's Disease (Hägnebo et al., 1999).

One longitudinal study found that coping strategies that 'relinquished responsibility' (a type of avoidance) away from personal mental or physical effort (which included a broad range of strategies such as asking advice from others, sleeping, watching TV, eating, and drinking) and distracting attention away from the symptoms positively correlated with handicap at baseline and seven months after attending a neuro-otology clinic, although it was relinquishing responsibility that made the most significant independent contribution to variance in dizziness handicap even after adjusting for distress, control beliefs and demographics (Yardley, 1994b). However few subjects reported often using such strategies and coping style did not have a significant longitudinal relationship with residualised dizziness handicap scores. Another longitudinal study of low quality did not find any difference between symptom severity and coping styles according to the Freiburg Coping Illness Questionnaire six months following an acute vestibulopathy (Godemann, et al., 2004).

Herdman et al. (2020b) evaluated patients on a waiting list for a specialist dizziness clinic and found that both avoidance and all-or-nothing behaviour were positively correlated to dizziness handicap (r=0.63; r=0.43) and vertigo severity (r=0.44; r=0.27) according to the 'Cognitive-Behavioural Responses to Symptoms Questionnaire'. In

their follow up study 3 months after consultation, neither strategy significantly changed, and both remained related to dizziness handicap, although only all-or-nothing behaviour retained its significance after adjusting for baseline dizziness handicap (Herdman et al., 2020a).

Sleep

Three studies explored relationships between dizziness handicap and sleep disturbance, which is a common target of treatment within cognitive-behavioural therapy. Two cross-sectional studies used translated versions of the Pittsburgh Sleep Quality Index (PSQI) to measure the quality and patterns of sleep. Sugaya et al. (2017a) found a positive correlation between handicap and sleep disturbance only in female participants. Kim et al. (2018) found that the relationship between sleep and handicap was strongest in patients with vestibular migraine (VM), followed by vestibular neuritis (AUV) and benign paroxysmal positional vertigo (BPPV), but not for patients with Meniere's Disease (MD), 'psychogenic dizziness' and another mixed pathology group. They also found that that Insomnia Severity Index (ISI) was correlated to handicap only in subjects with VM and BPPV.

One study examined the relationship between improvements in dizziness handicap and improvements in sleep disturbance following a short inpatient vestibular rehabilitation programme for participants with mixed peripheral vestibular disorders and comorbid sleep disturbance and found that participants with persistent sleep disturbance after 1 month had significantly higher handicap than participants without sleep disturbance even after adjusting for these scores at baseline (Sugaya et al., 2017b).

Methodological quality of included studies

The full quality assessment of the included studies is included in Appendix E. Interpreting the individual rating scores can be misleading since a high score may still hide a significant flaw or bias in one area. We therefore provide a summary of the quality assessment of studies included in this review to enable a clear understanding of the evidence base and potential sources of bias in this review.

There were several limitations that were consistently identified in the included studies. Most articles (87%) did not provide a sample size justification. Context regarding the sample size determination of an experiment is important for interpreting and determining the meaning of the experiment's results. Researchers should be more transparent in how they determine their sample sizes and carefully consider if they are suitable.

Secondly, not many studies adequately reported their recruitment methods. Consecutive sampling or random selection were the dominant types of recruitment, but 48% did not identify the source population for patients and describe how the patients were selected in enough detail to allow replication. Even more (85%) did not state the

proportion of those asked who agreed to participate, making it difficult to determine whether the participants were representative.

Thirdly, there were inconsistencies in reporting of participant characteristics. A number of vestibular and balance disorders are associated with chronic dizziness, and most studies combined data across a range of diagnoses. This is not necessarily a problem, but not all studies adequality described the different diagnoses present in their sample and 52% did not refer to a diagnostic classification so it is difficult to compare findings and the extent to which the results can be generalised to other samples. Using samples based on self-report diagnosis is particularly problematic due to the frequent misdiagnosis present outside specialist centres.

Fourthly, correlation without confounding is a major concern because it results in biased estimation of relationships. Seventy-eight percent of studies did not adequately adjust for confounding in the analysis and 85% of longitudinal studies did not take losses of participants to follow-up into account. Studies that attempted to apply statistical correction methods, such as regression models, were not without fault either. The decision on what factors were considered as confounders was applied inconsistently and, in many cases, relevant illness (e.g., mood, diagnosis etc) and demographic factors (e.g., age and gender) were not included. A few studies also used automated selection procedures that may have resulted in inappropriate exclusions or inclusions in their models.

Finally, most studies did not refer to a psychological theory or hypothesis as a rationale for selecting psychological factors, making it difficult to derive a consistent picture of how constructs are theoretically related, and which may be most important. Future research should attempt to minimise these limitations.

5.5 Discussion

The review shows preliminary evidence that dizziness related handicap and symptom severity are associated with several key psychological variables. By far the most frequently investigated psychological factors were measures of anxiety and depression and the meta-analysis found weighted positive correlations which were large in magnitude for handicap and moderate for symptom severity. A large weighted positive correlation was also found between autonomic anxiety and handicap. There was emerging evidence for the role of sense of coherence, negative illness beliefs, symptom specific cognitions, and associated behaviours such as avoidance, all-or-nothing coping, and sleep disturbance. The psychological factors which were consistently related to dizziness outcomes are summarised in Figure 8 with their hypothesised relationships drawn from some existing models. A complex interplay of many of these factors probably exists in a single patient and a few of these putative mechanisms will be unpacked with reference to the wider literature in the following discussion.

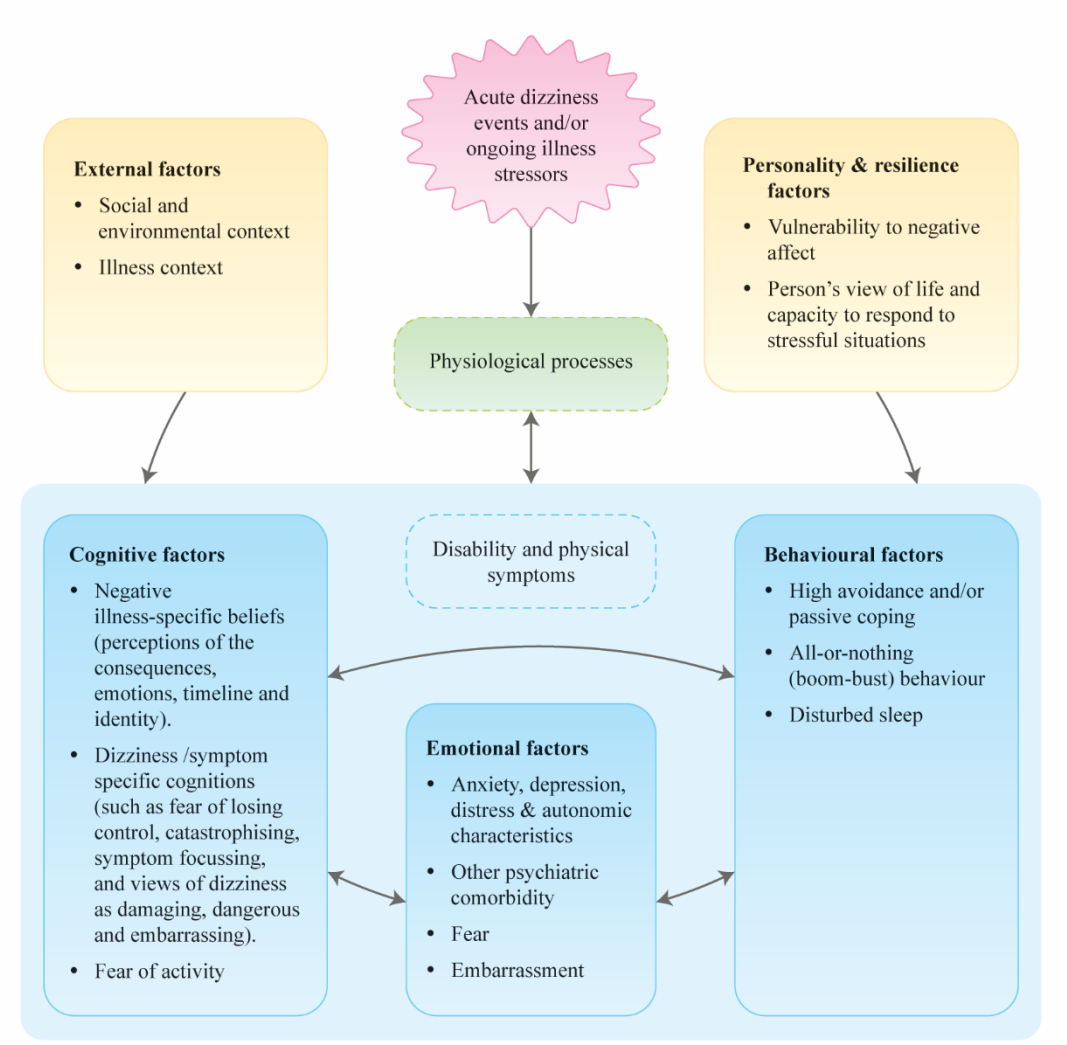

Figure 8. Psychological factors associated with self-report dizziness outcomes. These factors can contribute to dizziness directly and indirectly through cognitive, behavioural, and emotional responses to the illness

Anxiety and depressive symptoms were consistently associated with worse dizziness outcomes. In many cases, the association seems to be bidirectional in that the symptom burden increases the risk of anxiety/depression and vice versa. A large number of potential reasons could account for this association, ranging from chance occurrence, biological processes, behavioural links such as physical inactivity, and psychosocial factors (Jacob & Furman, 2001). The (neuro)biological mechanism that is often cited is the overlap in the neural structures involved in both dizziness and anxiety (Balaban & Thayer, 2001). Links to physiological stress response systems have also been investigated in animals models and in humans during laboratory vestibular stimulation (Saman et al., 2012).

Although Cousins et al. (2017) did not include measures of stress axis activation in their study, they did find that the Hospital Anxiety and Depression Scale in the acute phase of vestibular dysfunction did not predict handicap at 10 weeks. This could be due to the expert assessment and reassurance provided in this study but could support evidence that an acute vestibular stress response (albeit measured by self-report) is not

always disadvantageous and may even be adaptive in certain situations. Rather it was the distress experienced beyond this period that was associated with worse outcomes.

The review also found that dizzy patients who reported higher levels of autonomic anxiety symptoms reported greater concurrent handicap. Elevated arousal may be implicated in greater perception of autonomic symptoms, since it is often associated with hyperventilation which itself induces somatic symptoms including dizziness (Yardley & Redfern, 2001). One included study found a high incidence of hyperventilation (23%) in patients attending vestibular assessment (Humphriss et al., 2004). Again there are relevant biological vestibular-autonomic links, and acute vestibular lesions can disrupt breathing rhythm which then fails to adapt (Jáuregui-Renaud et al., 2005). Hyperventilation itself is used as a clinical test since it can reveal a latent vestibular asymmetry, although the lack of correlation between hyperventilation and vestibular function observed by Humphriss et al. (2004) refutes a purely biologically driven conclusion. Another theory is that people may misinterpret symptoms of hypervigilance and anxiety as a sign of illness, particularly because they overlap with vestibular symptoms and illness schema, creating a vicious cycle of arousal and restriction of activity (Moss-Morris & Petrie, 1999; Van den Bergh et al., 2017b).

These core features of physiological arousal, hyperventilation, and misattribution feature in most models of persistent physical symptoms. A more unifying conceptualisation of these findings comes from models of embodied predictive processing, broadly based on the idea that vestibular perception is reliant on pre-existing (prior) information and predictions, rather than detailed accurate information (Klingner et al., 2016). Factors such as negative affect may lead to overly precise threat-related categorical priors that go on to dominate conscious experience and result in 'perceptual dysregulation' (Van den Bergh et al., 2020; Van den Bergh et al., 2017b). This is a compelling theory since it helps to understand how dizziness can become uncoupled from vestibular function, but requires additional testing and empirical evidence (Seemungal & Passamonti, 2018).

This review also found evidence that other psychiatric conditions are also associated with worse handicap when present, most notably PTSD. Although self-report screening tools tend to overestimate the prevalence of psychological disorders, one can also point to evidence of studies that have used more in-depth methods such as clinical interviews. These studies also tend to find that patients with psychiatric comorbidity have higher handicap and dizziness symptoms (Best et al., 2006; Lahmann et al., 2015; Limburg et al., 2017; Limburg et al., 2016). However, it is not clear what combination of psychiatric and vestibular disorder, if any, appears worse (Teggi et al., 2010).

Most of the factors identified in this review can themselves be influenced by intrapersonal, or predisposing, characteristics. Although there was supportive evidence for the role of health anxiety and neuroticism (vulnerability to negative affect), surprisingly this review did not find evidence for the role of trait anxiety. Whilst it is

part of the personality dimension of neuroticism, trait anxiety is characterised by a stable perception of environmental stimuli as threatening (Gidron, 2013). This could be due to findings that show an acute vertigo attack is sufficient to cause extreme anxiety irrespective of premorbid anxiety (Pollak et al., 2003). It could also reflect the neuroanatomical and functional distinction between state and trait anxiety (Saviola et al., 2020).

There was more compelling evidence for the role of 'sense of coherence', best summarised as a person's coping capacity to respond to stressful events. The salutogenic model suggests that the successful application of resources to deal with stressors is due to a combination of behavioural and perceptual mechanisms (Antonovsky, 1979). In line with this hypothesis, Ketola et al. (2014) found that people with higher coherence had less restriction with activities such as exercise which typically promote vestibular compensation. Likewise, Mendel et al. (2001) found those people with strong SOC also had less emotional distress, better sleep, and psychosocial functioning that those with weak SOC. Although considered a stable personality trait, research has shown that interventions can influence SOC and Super et al. (2016) suggest that this can be achieved by empowerment and reflective practices.

The theoretical foundation of alexithymia, on the other hand, is more controversial. This is in part due to the methodology of giving people you suspect cannot self-assess their emotions a questionnaire to ask if they can self-assess their emotions. As a result several studies suggest that the TAS-20 is confounded by general distress (Marchesi, 2015), although the only study in this review to look at alexithymia did not adjust for this.

It is also important to consider that these background factors and a person's previous experience can help shape their thoughts (meta-cognitions) related to dizziness. However, some of the instruments used to operationalise cognitions in this review, such as the Agoraphobic Cognitions Questionnaire (ACQ) and Body Sensations Questionnaire (BSQ), were initially developed to assess fear of symptoms typically associated with anxiety arousal. Although this includes dizziness it is not clear whether the 'fear' is illness specific. Nevertheless, reacting to somatic symptoms in a fearful or catastrophic manner appears to be related to worse handicap and there is good quality evidence that negative illness cognitions predict worse dizziness handicap.

A clearer understanding of the role of cognitions is informed by the emerging literature on illness representations based on the common-sense model of self-regulation (CSM), which refers to the idiosyncratic beliefs people have about their illness that guide behavioural coping and emotional responses (Leventhal et al., 2003). This review showed that negative illness perceptions, such as viewing the illness as chronic, cyclical, or as having serious consequences or multiple symptoms, was associated with dizziness handicap. Most notably the studies that used the IPQ-R self-report inventory based on the CSM showed that beliefs that dizziness could result in serious consequences had

the strongest correlation with dizziness outcomes. One interpretation of this could be that strong beliefs in the negative consequences of the illness promote avoidance behaviours and threat appraisals in a very direct way (Hagger et al., 2017).

On the other hand, illness perceptions related to personal control were not related to dizziness in the literature. This construct reflects the belief that health is or is not in one's control and is similar to the concept of health locus of control which derives from social learning theory. This may be a limitation of the questionnaire method, which does not account for the proposed multi-level concepts and their behavioural interactions (Leventhal et al., 2016). For example, the perception that health is not in one's control could make individuals less motivated to learn self-management skills but could also indicate trust in medical professionals and adherence to treatments recommended by them.

Whilst overarching illness beliefs are clearly important in understanding individual variations, this review identified growing evidence that everyday interpretations of symptoms, such as catastrophising (believing in worst case scenarios) and kinesiophobia (fear of movement), could be even more relevant when determining coping behaviours which may enhance the experience of symptoms. According to the fear-avoidance model originally developed for chronic pain, fear of actual or anticipated symptoms or the view that symptoms could signal tissue damage often lead to avoidance behaviour and hypervigilance towards the body (Vlaeyen & Linton, 2012). Included studies also suggested avoidance behaviours could be driven by social embarrassment, a recurrent theme in qualitative studies (Yardley et al., 1992b). Greater strategic (and/or automatic) attention towards symptoms (symptom focussing) could also increase perceived severity of symptoms, disrupt orientation and balance control.

If coping behaviours mediate the effects of illness perceptions on outcomes, it is unsurprising that this review found that the tendency to use avoidance (versus approach) related coping was associated with worse outcomes. This is consistent with neurobiological models since recovery in vestibular disorders is known to occur only through active exposure to the movements and environments that provoke dizziness, meaning that avoidance is an understandable but particularly maladaptive strategy for these conditions. The other relevant response identified was all-or-nothing behaviours (boom or bust), where people push themselves to get things done when symptoms allow and then crash (Moss-Morris, 2005). Such inconsistent behaviour may reflect a belief that activity levels should be dictated by symptom experience and may also reflect a fear of the aftermath of activity, which in turn could negatively influence physiological processes over time.

The role of other coping behaviours on dizziness outcomes was sometimes contradictory, however. This could reflect limitations in 'coping' classifications in measures such as the WOCQ which measures coping in response to general stress rather than being illness specific (Schwartz et al., 1999). It is also important to note that the

adaptiveness of coping behaviours is context dependent. For example, distraction may be helpful therapeutically to improve balance control under conditions of perceived postural threat (e.g. fear of falls) (Johnson et al., 2020), but could be detrimental if used as a constructive avoidance behaviour.

The included studies also reveal a recent rise in numbers of sleep studies in this area and support a relationship with dizziness handicap. Both of the included studies found similarly high prevalence of sleep disturbance amongst patients with chronic dizziness (>65%) (Kim et al., 2018; Sugaya et al., 2017a), which may not be representative of other treatment centres. It appears that patients with conditions such as vestibular migraine could be particularly susceptible to sleep disturbance, consistent with literature that suggests a bidirectional association between headache severity and sleep disturbance (Houle et al., 2012; Odegard et al., 2011). They also found that sleep disturbance was associated with anxiety and depression, but to what extent this accounted for the relationship with handicap wasn't explored.

This review has therefore successfully identified several psychological variables that are related to dizziness handicap and severity. This is valuable for identifying potential targets for therapy. However, there are important limitations which should be considered. As already stated, future studies should address the methodological flaws highlighted earlier in this review. Many studies were limited to cross-sectional analysis and when using self-report inventories even longitudinal studies struggle to evaluate the dynamic mechanisms underlying these predictive relationships (Leventhal et al., 2016). Causality cannot therefore be assumed. Similarly, whilst it is logical and appropriate to identify therapeutic strategies to target such factors, these data do not prove that therapies targeted at these factors will be successful. Identifying specific targets for intervention at the individual level must account for the rich personal context and illness-related variables. However, this review provides a platform from which further research can investigate these possible contributing factors or mechanisms and gives rise to a number of testable hypotheses. The capacity to address the physical and psychological consequences of vertigo and dizziness will be a necessary step for improving outcomes in vestibular rehabilitation.

Chapter 6

Intervention Development

6.1 Chapter Overview

Dizziness can be considered a phylogenetically based aversive stimulus (Treisman, 1977), belonging to a basic 'embodied' motivational system that urges the individual to act and to restore the body's equilibrium, rather than just a sensory or emotional experience (Kaski et al., 2021). CBT techniques proceed from the view that an individual's interpretation, evaluation, and beliefs about their health condition and coping repertoire, with respect to dizziness and disability, will affect the degree of emotional and physical disability associated with dizziness. In this respect it can directly affect the condition itself, rather than focus on just the emotional consequences. Further improvements in vestibular rehabilitation treatments may require a paradigm shift toward more integrated approaches. It should be noted, however, that the usage of the term 'cognitive behavioural therapy', like 'physiotherapy', varies widely and does not represent one single intervention. Mechanisms underpinning the primary outcome must be theoretically and empirically defined. This chapter outlines the approach taken to develop methods to target the specific correlates of dizziness handicap and integrate these methods into an existing VRT approach. The intervention techniques and components will then be described in detail.

6.2 Intervention Mapping

Interventions to change health-related behaviours will have a greater chance of effectiveness if they are grounded in appropriate theory (Araujo-Soares et al., 2019; O'Cathain et al., 2019). The Medical Research Council guidelines for the development of complex interventions also emphasise the importance of developing interventions that are grounded in theory and empirical evidence (Skivington et al., 2021). This step represents the 'designing' and 'creating' stages of the taxonomy suggested by O'Cathain et al. (2019) and follows intervention mapping techniques as described by Bartholomew Eldredge et al. (2016).

The CBT formulation model (see chapter 5, Figure 8) provided us with a framework for translation into a CBT-VRT treatment protocol. We mapped evidence-based CBT and VRT treatment principles onto the biopsychosocial correlates of dizziness handicap and symptoms outlined in the model (see Table 11). This was carried out in meetings with supervisors and focus group/intervention mapping meetings with a large section of the Health Psychology Department at King's College London, who had a broad range of experience and expertise in developing CBT interventions for LTC's.

Table 11. Intervention mapping

Key determinant	Description of therapeutic aim	VRT techniques	CBT techniques
Lack of knowledge	*To provide an explanation for their symptoms, that helps them engage with therapy*	Information provision	Guided self-discovery Manual
Predisposing factors			
View of the world as manageable, understandable, and meaningful	*To be able to apply resources to deal with the dizziness problem* *To encourage a flexible dispositional orientation.*	Education	Empowerment and reflection processes Formulation Goal setting
Perfectionism		-	Cognitive restructuring Behavioural experiments Self-compassion
Physiological			
Threat-related balance	*To normalise balance and gait*	Graded balance exercises	Distraction techniques & cognitive aspects of balance control (threat appraisal)
Motion intolerance	*To reduce any motion triggered dizziness*	Habituation exercises	Exposure
Behaviour			
Avoidance	*To increase activity*	Graded exercise	Graded exposure
All-or-nothing	*To stabilise activity and rest, and gain control over symptoms*	Pacing	Activity monitoring and scheduling Sleep techniques
Sleep	*To improve sleep*	Information about sleep hygiene	
Cognitive			
Fear avoidance beliefs	*To re-engage in activity*	Graded exercise Habituation	Behavioural 'in-vivo' exposure with response prevention

			Identifying unhelpful thoughts and coming up with alternatives
Symptom focusing	*Reduce attention to the body/ symptoms*	-	Distraction
	To re-engage in activity		
Embarrassment avoidance			Behavioural experiments
Catastrophising	*To reduce threat appraisal and catastrophic cognitions*		Cognitive restructuring Behavioural experiments
Negative illness beliefs	*To develop adaptive illness beliefs*		Cognitive restructuring
		Education	Psychoeducation with self-guided worksheets
			Cognitive restructuring
Emotion			
Autonomic anxiety	*To reduce autonomic symptoms*	Breathing control exercises	Breathing and relaxation techniques Cognitive restructuring
Anxiety	*To reduce anxiety related to dizziness*		Behavioural experiments
		Exercises to build balance confidence	Behavioural activation
Depression	*To boost mood*	Exercise	

6.3 Implementation strategies

Normalisation Process Theory (Murray et al., 2010) suggests that interventions should consider implementation strategies from the earliest stages of development. Since physiotherapists are already providing rehabilitation for people with PPPD which is readily accepted, we wanted to design an intervention that could be deliverable within this context, with some additional therapist training. This was not designed as a mental health intervention, but rather an integrated CBT informed vestibular treatment targeted specifically to dizziness.

The timing of sessions to be 60 minutes for the initial consultation, and 30-minute follow-ups, was chosen to reflect current practice within the host neuro-vestibular therapy department. Likewise, six sessions was chosen to reflect the minimum number of sessions of VRT usually offered. The following section provides definitions and details of the core intervention components and related techniques.

6.4 Intervention techniques and components

Initial assessment interview

The initial assessment includes assessing patients' current symptoms, triggering factors and their beliefs about their condition. A semi-structured interview was developed to help gather information on the cognitive, behavioural, and psychophysiological aspects of the dizziness complaint and their role in the maintenance of the complaint and the consequences and interference caused by the problem (see Appendix F). It also includes information regarding the predisposing factors and other areas of life stressors. Any suicidal ideation should also be risk-assessed (Herdman et al., 2020c). Caution is exercised, however, in eliciting certain types of material such as information relating to memories of trauma and abuse. This material can be traumatic and out of scope within the context of physiotherapy and if trauma seems like a key issue, onward referral may be indicated.

Socratic Dialogue

The therapist can elicit, via guided discovery, specific cognitions, and beliefs about the dizziness, as well as help patients modify behaviour and cognition. One method of doing so is socratic dialogue, which depends on the use of questions and summary statements to 'clarify meaning, elicit emotion and consequences, as well as to gradually create insight or explore alternative actions' (James et al., 2010 [p. 85]). It takes practice but is considered a core feature of the cognitive-behavioural method, which differs from physiotherapy in the premise that questioning which allows the subject to reach their own conclusions will be more beneficial than direct information-giving.

Formulation

One significant part of the assessment process is to facilitate the development of a shared view of the patient's problem that identifies and frames the problem in terms of the relationship between physical symptoms, thoughts, feelings, and behaviour. The patient's own words are used, and the focus is on identifying possible perpetuating factors as therapeutic targets. Patients with chronic dizziness do not usually conceive of their problem as 'psychogenic' (Herdman et al., 2021a), and they may not talk about fear. The discussion should be geared toward the patient's perception of their dizziness problem. The formulation starts with the trigger and the physical symptoms. The therapist can paraphrase their personal story in terms of harmfulness ('I can understand that you feel that it might be better not to lie down flat', or 'I understand that you expect that moving your head might further harm you') rather than using words fear and anxiety. Making the origins of their fear clearer to the patient can be helpful when presenting the formulation and rationale for treatment.

Figure 9 shows a formulation from a participant in the trial, who developed PPPD following an acute vestibulopathy ('vestibular neuritis'). The formulation depicts graphically, in her own words, how the physical sensations were interpreted as threatening, with the potential for physical harm and embarrassment. Related to this

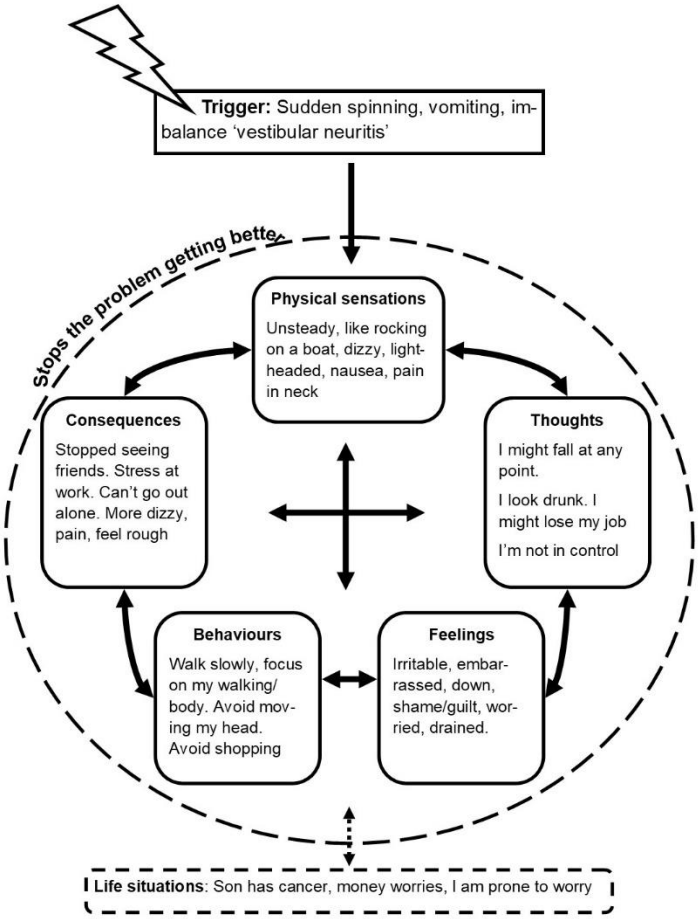

Figure 9. Formulation of a patient who took part in INVEST.

were feelings of depression, anxiety, guilt, and shame. These feelings were also influenced by external social factors. To control this, she engaged in behaviours such as walking slowly, consciously processing her gait and focusing on her symptoms, and avoiding activities that might trigger dizziness. The potential consequences and conflict between the current ways of coping and her desired future (e.g., seeing friends) is highlighted. It is also highlighted how these coping strategies stop the problem getting better, leading to more engrained symptoms over time. Formulation typically includes education since it can correct any misconceptions that have occurred and helps to reframe the dizziness problem. The goals of the education part are discussed below.

There was a concern amongst the PPI group that such a formulation could be seen to blame the patient for their condition, so this is proactively addressed as part of the education (below) and by explaining that they are not to blame for the dizziness or the hardships they have experienced as a result. Instead, with practice, they can make some automatic behaviours more likely than others and have more control over their future actions and experiences.

Education

The specific goal of education is to address unhelpful illness beliefs (addressing the identity, consequences, and timeline illness representations) and provide an explanation for their symptoms, which is credible and integrates the idiosyncrasies or personalised nature of the patient's dizziness problem (illness coherence). The general point is that the patients' coping behaviours are intuitive, normal defensive responses to dizziness, which may have been adaptive in acute dizziness but have lost their efficacy as the dizziness persisted. The aim is to engage the patient in this behavioural treatment that helps them disengage from unhelpful behaviours. This implies more than just reassurance, although unambiguously providing new information that dizziness is not a sign of damage can be helpful. The therapist acknowledges that avoidance is a normal form of defence in an abnormal situation of dizziness/imbalance but that after a while, this response loses its adaptiveness and becomes counterproductive.

During intervention development we discussed various stories and metaphors to help patients reconceptualise their dizziness experience. Examples like the ballet dancer learning to pirouette, or the fisherman staying out at sea to acquire their sea-legs, help the patient recognise the need to expose themselves for their balance system to adjust. Other examples, such as stepping onto a broken escalator, show that dizziness is not always a sign of injury and can be a product of our previous experiences. Specifically harnessing mindsets about non-life-threatening symptoms as a positive signal that the treatment is working has been found to dramatically improve outcomes to interventions such as oral immunotherapy (Howe et al., 2019). In this regard, we explain that dizziness is a sign of their body adjusting and that trying dizziness-provoking exercises help to seed their brain to build a new adaptive response.

Aspects of the manual specifically addressed illness 'identity' representations by asking the patient to re-evaluate different explanations for their symptoms, for example symptoms that might be attributable to a relapse vs normal fluctuation, symptoms related to decondition, anxiety or depression. This method has previously been used to treat chronic fatigue in people with multiple sclerosis (Moss-Morris et al., 2013).

Best et al. (2015) found that a psychoeducational intervention which included information on the impact of threat on balance led to normalisation of pathological balance behaviour (e.g., anxious anticipatory contraction of the antigravity muscles) in a small pilot study. Again, we include examples of experiments of people walking on a high platform to help patients make predictions about the effect of threat and understand that consciously processing balance is a normal but nonetheless counterproductive response. Table 12 provides a list of some of the analogies used in the manual.

Table 12. Analogies of 'everyday' dizziness used in the manual

Examples	Brief Description
The railway illusion	The experience of sitting in a train on a platform and not knowing whether you are moving, or the train next to you is moving.
Travel sickness	The experience of motion sickness when travelling due to sensory mismatch.
The broken escalator	The experience of momentary imbalance when stepping onto a broken escalator.
Walking the plank	Eliciting threat related balance reactions in healthy people by asking them to walk along a gangway at height

Balance Therapy & Attention Allocation

One of the key features of functional dizziness is how much just in everyday life our sense of postural threat changes our perception. When we stand there is a natural body sway, but when raised in the air we stiffen up. Just the awareness of being in a situation where there is greater risk of postural failure causes a change in the perception of how much we are moving by 2-10%, and the reactivity of postural control reflexes to our natural sway (Cleworth et al., 2018).

Patients with functional dizziness typically exhibit patterns of postural control adjustments typically seen in normal individuals at height or in patients with fear of heights and fear of falling, even at ground level. This results in co-contraction of ankle musculature with or without increased truncal sway (Holmberg et al., 2009; Odman & Maire, 2008). Patients also adopt a gait pattern akin to 'walking on ice', characterised by shorter steps, wider base of support, reduced speed, and increased time spent with both feet on the ground (Schniepp et al., 2014). These changes correlate with changes in conscious attention to movement (Ellmers et al., 2022). Overall, these observations support the hypothesis that postural changes with PPPD reflect a maladaptation of

high-risk postural control strategies triggered by an initial stimulus that persists due, in part to, excessive self-observation and anxiety (Popkirov et al., 2018a).

Authors such as Wuehr et al. (2017) and Johnson et al. (2020) have shown that distracting attention can normalise such threat related postural adjustments. As a result, this serves as the first 'behavioural experiment' in the intervention. Patients are asked to perform a task that triggers imbalance, such as standing with eyes closed or walking, and perform a distracting task. Popkirov et al. (2018a) suggests that tasks such as asking the patient to name a number written on their back or testing eye movements during stance may be more effective than mental distraction such as listing months backwards. However, in our clinical experience, mental distraction may be required to sufficiently capture attention. The one caveat is patients who may struggle with mental arithmetic, since the performance anxiety may itself cause someone with an anxious temperament to stiffen up (Hainaut et al., 2011). Another consideration is dual task interference, which can naturally impair postural control, proportional to the difficulty of each task particularly in people already prone to falling (Ghai et al., 2017). We therefore advise that the task is agreed between the therapist and patient, something that will sufficiently capture attention, but achievable without causing additional anxiety (e.g., stating alternate letters of the alphabet). Just like other behavioural experiments, the aim should be made explicit so the patient is encouraged to keep their attention on the task and be reassured that they are not being 'tested'.

Observation of a reduction of body sway through distraction or the normalisation of gait, and/or reduction in perception of body sway, can be explained as proof of cognitive influences and the counter-intuitive effects of consciously processing balance, which can then serve as a starting point for treatment. Gait re-education then concentrates on alternative or exaggerated gaits (e.g., taking larger strides, walking faster, backwards) that are used to promote automatic movement. Manipulating speed can be another powerful experiment, since Brandt et al. (1999) were the first to identify that even patients with an acute vestibular disorder balance better when running than when standing or walking slowly, since the automatic spinal locomotor programme suppresses destabilising vestibular input. Asking the patient to run can not only demonstrate what they are capable of but can act as disconfirming evidence against the very strategies (e.g., walk slowly and carefully) they have adopted to control their symptoms. However, running should come later as part of the 'in-vivo' experiments for any patient with a high degree of fear, since they may interpret bodily signals to infer a high probability of causing harm thus triggering a conscious fear response (Ellmers et al., 2022). The early sessions should focus on normalising gait to show evidence of reversibility and achieve early 'buy-in' to therapy.

The therapist should also observe the patient's eyes during walking to examine threat related gaze behaviour (Staab, 2014). Although this has not received as much attention in the literature, there is evidence of how individuals visually scan their walking path when fearful of falling (Ellmers & Young, 2019). People tend to become more

hypervigilant towards immediate threats to balance and reduce visual exploration (reductions in the number of gaze transfers) (Ellmers & Young, 2019). This is consistent with observations of people with PPPD, who typically 'freeze' their gaze to the ground (Staab, 2014), motivated to avoid falling and consciously controlling every step, at the expense of transferring gaze further ahead. Examples of exercises here may include encouragement to transfer their vision elsewhere (e.g., throwing a ball in the air when walking), as well as cognitive dual tasks when stepping over obstacles (postural threats) to 'disengage' from performing conscious visual planning.

There are other ways we can measure alterations in postural control, such as the sensory organisation test, which manipulates the visual and proprioceptive inputs to balance. Patients with PPPD tend to perform worst with their eyes closed, and particularly when the visual environment is misleading (Söhsten et al., 2016). This supports the findings of people with chronic dizziness after an acute vestibular event who exhibit 'visual dependence', due to an over-weighting of visual cues for spatial orientation (Cousins et al., 2014). As a result, exercises that reduce reliance of vision for postural control, such as standing on a soft balance pad with eyes closed, should be incorporated. This can also include distraction tasks to eliminate any potential threat responses and reduce perception of sway.

Goal Setting

A recent review has questioned the validity of the SMART acronym (e.g., Specific, Measurable, Achievable, Realistic, Timebound) for setting physical activity goals (Swann et al., 2022). We had also reflected on the limitations of the SMART acronym in our group discussions. Traditionally VRT goals may be symptom or impairment driven. For example, one might set a SMART goal to reduce dizziness by 50% or improve the persons balance score by a set amount within 6 weeks of VRT. In addition to the valid criticisms of this approach by Swann et al. (2022), it is also our experience that such goals usually only serve the therapist rather than patient. We decided not to include SMART goals and instead to draw on relevant CBT theory and clinical experience.

Rather we considered 'valued' goals, enhancing the *motivational* quality of goal setting. A patient's main goal may still be to 'get rid of the dizziness', which the therapist must validate, whilst asking 'And if we got rid of the dizziness, what might that look like?'. The therapist also enquires about the persons' 'values' (i.e., what is important to them) to prioritise and modify goals. For example, if their activity goal is a 10-minute walk, and their values are friends and family, they could invite a family member along or arrange to meet up with a friend.

Here the only real requirement is to identify meaningful and realistic goals to regain a sense of control and purpose, and to restore hope. The patient is asked to break down the goal into achievable steps and to identify available resources (within themselves or

the environment) to reach those new goals. This approach draws on the salutogenic model to build sense of coherence, and implementation theory to allow early goal attainment. Setting functional goals redirects the focus of attention from dizziness and physical symptoms toward daily life activities with the emphasis on the possibility of change away from disability status. As the patient is invited to formulate their goals in the manual, goal setting reinforces the notion that active participation is an essential part of treatment. The goals are monitored and re-evaluated if needed as therapy progresses.

Activity Planning & 'Pacing'

There are many diary formats which can be used to gather information on patient problems. Many patients will have already attempted to monitor their symptoms to identify potential triggers and avoid dizziness. Whilst understandable, actively tracking symptoms has been shown to result in greater symptom reporting, symptom severity, and slower recovery from injury (Ferrari, 2015; Ferrari & Russell, 2010).

The use of diaries in CBT is rather to help patients to make discoveries about their current coping strategies and any potential trade-offs. Subsequent forward planning of activities, so called 'behavioural activation', has been a core part of CBT for depression since its inception. For people with dizziness, it may also start the process of developing explorative behaviour, a prerequisite for exposure-based treatment (Vlaeyen & Crombez, 2020).

We considered a Weekly Activity Schedule to be sufficiently general to monitor patient activity levels. The patient is asked to record their activity, and sleep, for one week. We didn't include symptom ratings, as this begins the process of changing the person's mindset away from avoiding symptoms and towards exploring various ways to reach valued activities. Nevertheless, some patients may still find that they make therapeutically beneficial discoveries about their symptoms using activity monitoring.

On review of the diary, the therapist and patient can work collaboratively to identify different patterns that may exist. Things to look out for here include avoidance behaviour, and all-or-nothing behaviour, which consists of fluctuating patterns of activity. It is also a useful time to consider the balance of activities, such as any trade-off between pleasurable activities and accomplishment tasks (e.g., everyday chores).

As part of behavioural activation for people with depression, you may make a schedule of activities hour to hour throughout the day and encourage the individual with depression to follow the schedule and not necessarily their mood. But when working with people with dizziness who are not necessarily clinically depressed, we felt we didn't have to do the activity scheduling on an hour-to-hour basis during the day. We can instead simply plan a few activities during the week and make people make some

decisions about when they do them. The idea is still that we want the patient to follow the activity plan rather than their symptoms, focussing on habit and consistency.

For someone engaging in all-or-nothing behaviour, it is important that they consider what activity is achievable on a bad day. This is then repeated daily, which may at first look like they are doing less activity but allows them to stabilise their symptoms and acknowledge achievement and accomplishment tasks which are not contingent on their symptoms. Activities can then be gradually increased (graded) accordingly and help break any conditioned response to those activities.

For someone engaging in a lot of avoidance behaviours, they may already do lots of accomplishment tasks (e.g., tasks that allow them to get through the day), but they don't do many pleasurable things. For this patient, the activity plan may include one or two pleasurable things during the day, or small tasks that help them overcome avoidance that may also provide a good mood boost.

The activity planning can be easily combined with their goals, since activity scheduling is more likely to be successful if patients are engaging in things that are meaningful to them. The therapist can also ask about any potential obstacles, and problem-solving techniques may help in finding ways to overcome them, avoiding the tendency of the therapist to offer an immediate solution (Eccleston & Crombez, 2007).

Autonomic anxiety

The review highlighted a relationship between symptoms of autonomic anxiety and dizziness. During postural changes, autonomic reflexes maintain homeostasis. Vestibular stimuli appear to play a part in these autonomic responses (Yates, 1998; Yates et al., 2002). In healthy subjects, both caloric vestibular stimuli (Jauregui-Renaud et al., 2000) and rotation of the head (Jáuregui-Renaud et al., 2001) can induce an increase in breathing frequency. Yardley et al. (1998c) also found greater increases in respiration rate following head movement among patients with vestibular disorders who complained of more somatic symptoms. Once again, the disruptions during an acute vestibular event (e.g., the influence of vestibular stimuli on the control of the breathing rhythm) may prevail during the chronic stage (Jáuregui-Renaud et al., 2005). This may be one reason why there is such a high prevalence of breathing pattern disorder, and specifically hyperventilation, in people with chronic dizziness (Humphriss et al., 2004). This in turn can create a vicious cycle, since hyperventilation can also cause dizziness, and even 'unmask' vestibular signs in people with compensated vestibular deficits (Califano et al., 2011).

Little is known about the best type of breathing exercise. Jáuregui-Renaud et al. (2007) found that supplementing VRT with exercises which focussed on paced breathing rhythm saw a greater reduction in handicap scores. Slow deep breathing (SDB) is commonly employed in the management of pain (Jafari et al., 2020), but the underlying

mechanisms remain equivocal and the effects on dizziness are not known. Lucy Yardley's group also included controlled breathing as part of their internet- and booklet-based vestibular rehabilitation protocols (van Vugt et al., 2019; Yardley et al., 2012).

We decided to include diaphragmatic breathing in addition to referencing other common relaxation strategies, such as Progressive Muscle Relaxation, which the patient may find helpful to reduce tension. However, these were not mandatory since we also recognised the possibility of some of those exercises to heighten awareness of physical symptoms for people with LTCs, possibly because of increased focus on the body or breath during the relaxation exercise.

Behavioural Experiments In-Vivo

At some point people with increased levels of dizziness-related fear will need to expose themselves to the situations they have identified as dangerous or threatening. One way to approach this is with 'in-vivo' exposure with response prevention, which creates prediction errors and opportunities to discover that the anticipated harm signalled by dizziness is overestimated (Vlaeyen & Crombez, 2020). Exposure with response prevention has a long tradition in the treatment of anxiety disorders, but it also has been applied successfully mainly in patients with chronic musculoskeletal pain (Glombiewski et al., 2018), but also in conditions such as irritable bowel syndrome (Boersma et al., 2016) and tinnitus (Fuller et al., 2020).

In this treatment, patients perform an activity to challenge the validity of their catastrophic expectations. These expectations take the form of 'if P, then Q' predictions, which are subsequently tested during a behavioural experiment. For example, a person with PPPD may expect that walking across a train platform will inevitably cause them to lose their balance, resulting in either injury or embarrassment: '*If I walk across the train platform* (P), *then I will fall over* (Q1) *and I will die* (Q2).' A behavioural experiment is designed to create the opportunity to falsify the prediction. After the therapist models the activity, the experiment is carried out and evaluated. In contrast to VRT, the exposure is developed to critically challenge idiosyncratic beliefs and expectations and to encourage explorative behaviours. Patients are asked to judge activities on the degree of anticipated harmfulness of the activity, since it is the overestimation of harm, rather than the dizziness itself, which is challenged.

Outcome studies have shown that such exposure treatments are especially effective in reducing symptom-related fear and the perceived harmfulness of physical activity (den Hollander et al. 2016, Leeuw et al. 2008, Linton et al. 2008a, Woods & Asmundson 2008), but have not been used in the treatment of vestibular disorders.

They have been suggested to outperform more traditional CBT in terms of reducing movement-related disability, but a major concern is that more patients drop-out partly

because they are not convinced about the benefits or want to avoid exposure sessions (Glombiewski et al., 2018) . As a result, we only introduce in-vivo exposure in the third session, once sufficient understanding and therapeutic alliance has formed, and the patient has already started to engage in non-dizziness-based goals.

Competent delivery of exposure in vivo is very challenging as there are many intricacies inherent in delivering it successfully and ways to strength inhibitory learning (Morley & Eccleston, 2004). To address this, I developed specific worksheets to guide the patient (and therapist) through creating a fear hierarchy, to formulating a hypothesis and experiment, and de-briefing. The therapist pays special attention to the use of any safety-seeking behaviours which can act as exceptions to the rule, preventing the desired decrease in anxiety or correction of catastrophic thinking. As therapy progresses, it becomes important to focus on generalisation in as many different situations as possible.

Cognitive Therapy

Section seven of the manual introduces traditional cognitive restructuring techniques to address unhelpful dizziness illness perceptions and day-to-day cognitive interpretations of symptoms. We hypothesised that patients with PPPD will relate better to the dizziness-specific thinking styles presented in the self-help resource and therefore engage more with the treatment. This part of the manual was designed as more of a self-help tool for the patient to work through at their own pace as these explicit thought challenging techniques would typically fall outside the scope of physiotherapy. Patients may have thinking styles that reflect biases commonly observed in people with anxiety or depression (Burns, 1999). For some individuals with dizziness however, the experience of depression for example can be subtly different from depression in a mental health setting. This can be reflected in the cognitive features of negative thoughts. Whereas primary depression is often marked by a degree of self-denigration, patients with dizziness and depression focus their negative thoughts on the dizziness rather than core aspects of the self. These thinking patterns are still amenable to change using traditional cognitive restructuring techniques.

For instance, our review identified that elevated dizziness handicap occurs when individuals have pessimistic illness beliefs concerning the timeline and consequences. Some individuals may demonstrate specific biases in the way they appraise those potential threats. For example, they may think in ‘black and white’ terms and use absolute statements such as ‘I will never be able to exercise again’ or ‘I am stuck with this forever’. The cross-sectional work also identified the importance of beliefs about symptom interpretations and having unrealistically high personal standards (psychological vulnerability). The manual, therefore, introduces patients to the concept of biased thinking patterns, self-monitoring of thoughts using thought diaries, thought challenging techniques, and the generation of alternative thoughts. Time during session 4 or 5 can be used to go through an example of the thought restructuring exercise,

although this part of the manual is designed to be predominantly for the patient to work through at their own pace as these explicit thought challenging techniques would typically fall outside the scope of physiotherapy.

Identifying barriers to recovery and sleep management

This section refers to session 5 (section 8 of the manual) which uses principles from problem-solving therapy to prompt patients to consider any other potentially relevant factors that may be impeding progress (D'Zurilla & Goldfried, 1971). Links with worrying thoughts and avoidance are reinforced and patients are encouraged to go back to review earlier sections of the manual. Patients are also referred to strategies for perfectionism and related self-critical responses and other ways to build self-compassion.

Our review identified that poor sleep would exacerbate dizziness handicap for some people. Sleep hygiene including stimulus control techniques (Bootzin et al., 1991) are therefore introduced here. If the patient has more severe problems with insomnia, information about sleep restriction therapy is provided (Spielman et al., 1987).

Relapse Management and Prevention

For some people, their dizziness can increase from time to time and/or increase at the beginning of therapy as their activity levels increase. The likelihood of this generating catastrophic thinking can be managed proactively by explicitly generating the expectation that this is a normal response to therapy because of re-engaging with movements that have been relatively neglected. When explaining the rationale for treatment, dizziness flare-ups can be normalised as an integral part of the treatment rather than a signal of impending treatment failure. If a patient does experience a significant dizziness flare-up this can be reframed as a good preparation for relapse prevention because it allows the patient to prepare for potential increases in dizziness that may occur after the treatment period.

The final session summarises skills learnt, and progress made over the preceding six sessions. This creates an opportunity to check for residual unhelpful beliefs about dizziness and to challenge them. The reversal of any remaining avoidance behaviours tied to target fears is also undertaken. It also puts in place relapse prevention protocols to sustain improvements into the future.

Vestibular Rehabilitation Exercises

Vestibular rehabilitation consists of eye, head, and postural exercises of progressive complexity (see Table 13). Individual exercises are selected based on identifying symptoms and the presence of triggers and a functional 'objective' assessment incorporating different balance tasks (Klatt et al., 2015).

VRT for PPPD is usually focussed on habituation exercises (Popkirov et al., 2018a). Habituation refers to the idea that repeated exposure to a provocative stimulus (e.g., head movements) will lead to a reduction of the motion-provoked symptoms (Norré & De Weerdt, 1980). This approach has been adapted since its initial description to improve compliance and tolerance. Exercises are now chosen according to the provoking movements and the observed tolerance of the patient, which usually consist of around 3 sets of 5 cycles, performed twice a day (Clendaniel, 2010). Specific stimuli and exercises have also been developed for people who experience environmental and other visually triggered symptoms, using similar principles of exposure usually incorporating optokinetic stimuli (Pavlou et al., 2013; Pavlou et al., 2004).

People with vestibulopathy also report a lack of visual acuity with head movement, since the 'vestibulo-ocular reflex (VOR)' cannot stabilise the gaze. Gaze stability ('fixation' or 'adaptation') exercises work to either improve the VOR (adaptation) or lead to an eye movement that substitutes for the deficient VOR. These exercises can still be prescribed for people with PPPD who report head movement provoked dizziness regardless of VOR function, since they include repetitive head movements which is likely to be the beneficial component (Millar et al., 2020). However, since the habituation principles described previously are usually more tolerable, VOR adaptation exercises are usually only included when required for gaze instability.

For balance retraining, instead of performing the same exercises repetitively, patients should be challenged by a multitude of exercise variations (See Table 13) (Klatt et al., 2015). For the purposes of the VRT 'control' group, the therapists was encouraged to utilise these principles and refer specifically to the clinical practice guidelines from the Academy of Neurologic Physical Therapy (Hall et al., 2016), which recommends a home exercise program of gaze stabilization exercises consisting of a minimum of 3 to 5 times per day for a total of at least 20 minutes daily, and balance exercises for a minimum of 20 minutes daily, for individuals with chronic vestibulopathy.

Table 13. Exercises typically included in vestibular rehabilitation. Adapted from Klatt et al. (2015)

Habituation exercises for motion provoked dizziness *(progressing amplitude, speed, position, and number of repetitions according to patient tolerance)*
Turn head from side to side Move head up and down Seated trunk flexion-extension Turn around 180˚/360˚
Habituation exercises for visually induced dizziness *(performed sitting, standing, then walking, progressing duration, and adding head movements)*
Exposure to complex visual patterns Exposure to optokinetic stimuli and optokinetic scenes (e.g., YouTube videos)
Gaze stability exercises for visual blurring *(with variations to the stance position, stance surface, distance of the target, and the background from plain to complex)*

Performing head exercises while fixating a stationary target (VORx1)
Performing head exercises while fixating a moving target (VORx2)
Performing head exercise while fixating a stationary target with eyes closed (remembered target)
Performing eye rotation toward one target, followed by a head rotation towards the same target (gaze shift 'substitution' exercise)
Balance and gait abnormalities *(variations including foot stance, surface, eyes open, eyes closed, static vs dynamic head movements, and additional dual tasks)*
Standing, feet together, eyes closed
Standing, feet together, eyes closed, moving head side to side
Standing on a balance cushion, feet apart, eyes closed
Walking and turning head
Practice walking in circles, pivot turns, up slopes, stairs, around obstacles

6.5 The Manual

The manual was created to support and guide the above therapy. It was created with the assistance of PPI representatives as previously described. The aim of the manual was to structure and standardise the therapy whilst allowing individual case conceptualisation. Parts of the manual and worksheets were adapted from existing resources (Burgess & Chalder, 2019; Greenberger & Padesky, 2016; Moss-Morris et al., 2013; Williams et al., 2011).

We worked with a graphic design company who also made sure that whilst the design aided reading by reducing glare, it was also sufficiently contrasting to pass relevant accessibility standards. The manual was deemed readable, but it must be noted that most of the PPI group were well read and had achieved a higher education qualification.

6.6 Session Guide

Table 14 provides a brief outline of the session guides. The first session lasts 60 minutes and follow up sessions last 30 minutes to fit in with existing VRT timetables. The sessions are spaced fortnightly apart to allow sufficient time for the patient to complete tasks and experience change. As therapy progresses (after session 3), there is scope for sessions to be spaced further apart. This should be negotiated between the patient and therapist. For example, one person may want to wait 3 or 4 weeks before their final appointment to see how they manage without as frequent contact.

Sessions 1 and 2 are best adhered to in the order specified. However, the degree of emphasis on the CBT treatment principles outlined in the manual will differ on a patient-by-patient basis, according to their cognitive-behavioural profile. Session 1 focusses on socialisation to the treatment model, psychoeducation and addressing functional balance deficits. Session 2 introduces goal setting and activity planning

techniques, and habituation exercises customised to the needs of the patient. The structure and emphasis given across sessions 3–6 will vary depending on the needs of the patient (their case conceptualisation). The manual explicitly informs patients that they may find some sections more relevant and useful than others.

Table 14. Session Guide

Summary & therapist's role	Relevant manual content	Linked homework tasks
Session 1: Defining the problem		
• Initial interview to complete a cognitive-behavioural analysis of the problem with special attention to the persons illness beliefs and situational triggers. • A balance assessment is conducted, including a behavioural experiment to demonstrate the effects of hypervigilance and attention switching on dizziness and balance. • The therapist and patient create a shared formulation in the form of a vicious cycle to help them make sense of their condition and provide a rationale for treatment. • Education is provided in a way that the patient can view their condition as a common condition that can improve with exposure.	Section 1: Dizziness and balance Section 2: What keeps the dizziness problem going? Section 5: Symptom management (activity diary)	• The manual is introduced at the end of the session, and the patient is invited to read the first 2 chapters. • The patient is also asked to keep an activity diary for 1 week • One or two balance exercises are prescribed according to the examination, usually including distraction
Session 2: Dealing with the problem		
• The session starts with recapping key information from session ONE and the therapist reminds the patient of their personalised vicious cycle. The activity log is reviewed so that associations with avoidance and safety behaviours can be identified, and information on the negative consequences of these behaviours can be discussed. • The rationale for graded exercises as treatment is reinforced as a way of lessening the impact of dizziness and improving psychological wellbeing. The patient formulates specific treatment goals, and these are incorporated into an activity plan. • Specific vestibular and balance exercises are introduced and prescribed according to the specific complaints.	Section 3: Steps to recovery Section 4: Physiotherapy for dizziness Section 5: Symptom management	• To complete activity plan for the week • To complete specific vestibular and balance exercises prescribed.
Session 3: Overcoming avoidance		
	Section 5: Symptom management	• Further behavioural experiments are agreed as

• A review of the goals set in session 2 is conducted and response to therapy. By now the patient should have experienced some reduction in their symptoms and be performing exercises regularly. • Alternative strategies for responding to dizziness and associated symptoms are demonstrated when needed, such as breathing control. These strategies can be used in the case of high levels of autonomic symptoms and/or overwhelming vertigo and dizziness during therapy. • A hierarchy of various fear-eliciting physical movements and activities from daily life is made and from this individually tailored practice tasks are developed. These take the form of a series of behavioural tests during which irrational expectations are challenged. At first an experiment is be performed in clinic.	Section 6: Overcoming avoidance	homework. Emphasis is given on generalising the fear exposure to different settings and without therapist supervision.
Session 4: Managing negative thoughts • A review of response to session 3 and homework is conducted. • If agreed in the last session, session 4 can be given to a pre-planned in-vivo experiment (e.g., this might include meeting up outside the clinic, or walking to a local train station or supermarket). • Otherwise, the session can focus on the 'thoughts' component of the vicious cycle. The idea of unhelpful thinking patterns related to vertigo and dizziness are introduced. • A review of vestibular/balance exercises can also be conducted to progress the home exercise programme.	Section 6: Overcoming avoidance Section 7: Challenging dizzy thoughts	The patient is invited to work through section 7 of the manual to develop skills to identify, and modify, biased thinking.
Session 5: Identifying barriers to recovery • This session allows the patient and therapist to recap and review progress made over the previous four sessions, to identify any ongoing blocks to recovery. • Further areas of need are identified (e.g., a self-assessment of whether additional support for managing sleep is encouraged).	Section 8: Overcoming blocks to recovery and dealing with setbacks	• Sleep therapy if needed • Increased activity and exercises are strongly encouraged with a focus on generalisation by practicing

Further sign-posting information is provided depending on the problems identified. • Further exercises targeting feared or provocative situations are prescribed.		exercises in as many different situations as possible. Visual desensitisation exercises can be prescribed if needed.
Session 6: Planning for the future • The patient reflects on progress made over the 6 sessions by identifying new skills and knowledge learnt and successfully completed between sessions. Specific action plans are developed to implement when an acute flare up or managing setbacks.	Section 9: Planning for the future	• The patient completes the relapse management plan and sets goals for the next 3 months.

6.7 Patient-Public Involvement

The content and design of the seven sessions were presented to the PPI group in different focus group meetings. Patient representatives did not receive active CBT / VRT treatment from the perspective of a patient in therapy. Specifically, their role was to provide initial feedback on the relevance of the sessions and to review the manual. They were also presented with several CBT-based manuals produced by the Health Psychology Department at King's College London and asked to provide feedback on their design.

Patient representative comments largely centred on the layout of the manual and considerations to support accessibility. Due to difficulty with visual orientation, they did not want any wavy or zig-zag lines across the page. They also struggled with processing of images and colours that deviated from natural properties (e.g., conflicting strong colour pallets). They also disliked glare from blank white space. A simple four-colour palette was therefore used throughout, with no stark black or white to avoid glare. The manuals were also printed on a soft grey paper stock. Text was set in dark blue rather than black.

We also highlighted summaries and key points at their suggestion since these are often the bits people are more likely to read. They wanted notes pages at the end of each section to allow them to keep notes/questions as they go. The text was made larger so they could easily read it. Wiro-binding was preferred to allow the manual to lay flat, making it easier to annotate by hand.

One person poignantly described how the process of obtaining a diagnosis and navigating the medical system for many years had been 'dehumanising', disempowering, and that they were often talked down to. For this reason, there was a strong desire for patients to be depicted as real humans, and for the manual to have a mature look without being trivialised by cartoons. Photographs were also treated to the same simple monotone.

The content of the problem-solving section of the manual was informed extensively by our patient representatives' experience with chronic dizziness. For example, they wanted information to be included about work related issues since this had been information they had found hard to come by.

The content of the psychoeducation was acceptable, and I checked their understanding. Metaphors can be helpful in putting complex topics in simple, familiar terms. However, some metaphors failed since patient representatives sometimes failed to understand them or misunderstood the meaning. Metaphors were therefore used with care in the manual since they can provide the illusion of knowledge, and their explicit meaning was made clear.

6.8 Using the 'PPPD' Label

There has been some concern raised by medical professionals over whether using the term PPPD is readily accepted or helpful for people with functional dizziness. We carried out a separate qualitative study of people who had recently been diagnosed with PPPD (Herdman et al., 2021a). The interviews were thematically analysed following the recommendations of Braun and Clarke (2006), and four themes were identified. There was often a sense of relief about acquiring a label which was seen as validating. The diagnosis allowed them to re-evaluate illness-beliefs, with the diagnosis giving greater perception of control and willingness to approach rehabilitation. Many participants reflected difficulty understanding the nomenclature of PPPD, finding the terminology itself problematic or confusing, potentially misinterpreting 'persistent' as meaning 'poor prognosis' for example. Finally, the participants recognised psychological distress as a natural consequence of the symptoms but were less likely to make psychological attributions as to the cause of PPPD.

PPPD as a term is beneficial in that it is descriptive and aetiologically neutral. However, there is some redundancy in it, particularly since all symptoms will be 'perceptual'. Some patients find it confusing and the terminology does not easily engender sympathy or understanding from others. Most of the other negative consequences of a 'PPPD' label can be explained by the observation that much of healthcare uses labels without exploring their meaning with the individual. Some patients may have had PPPD triggered by another vestibular disorder and not readily accept an alternative 'new' label to their disorder. We therefore reference PPPD explicitly in the manual as a valid diagnosis, but the rest of the manual focuses on the dizziness problem to allow an individual narrative of PPPD to develop and for the patient to use a label they feel fits their illness or problem best.

There are also misconceptions amongst healthcare professionals about PPPD as to whether it is a diagnosis of exclusion or derogatory 'waste-basket syndrome' (Hain, 2022). Neither is true. The term PPPD was therefore not made explicit in the inclusion criteria, to get around these misconceptions amongst doctors potentially identifying patients for the study. Since the beginning of the trial however, there has been much better recognition and understanding of PPPD such that we do not believe this would now be a significant barrier to recruitment in a future trial.

6.9 Training & Supervision

There are many intricacies of performing a psychologically informed treatment. INVEST requires thorough knowledge of behavioural neuro-otology, as well as general and specific competencies in the application of the CBT components drawn from the competency framework for the delivery of psychological interventions to people with persistent physical health problems (Roth & Pilling, 2018). I received eight-day introductory level training in CBT from the Oxford Cognitive Therapy Centre. As part

of the trial, I received weekly or fortnightly supervision with my supervisor RMM, which included role-play and feedback on audio-recorded sessions with patient consent. This mainly consisted of recordings of challenging sessions or ones where I had more queries. The audiotapes were listened to by me and my supervisor before supervision.

6.10 Trial components

Some trials of VRT for chronic dizziness have been designed to include placebo or 'sham' exercises (Kundakci et al., 2018). However, there are both practical and conceptual problems with attempts to extend the placebo concept from the medical setting to the therapeutic setting. As Kirsch et al. (2016) explains this is both impractical and illogical. Impractical because it is impossible to construct a placebo that is believable and has the same characteristics as the treatment for which it serves as a control. If the control group contained the same psychological properties as the intervention (i.e., the therapist used the same words and procedures), it would no longer be a control condition. Instead, it would be the treatment. The conceptual problem is that we are specifically targeting the factors that produce medical placebo effects (e.g., therapeutic relationship, expectancy etc) as theoretically legitimate mechanisms of therapeutic change.

The other consideration was that for practical reasons I was going to deliver the intervention, which would bias the study. Although this is not necessarily an issue, since the focus of the trial was to continue to develop and refine the intervention, with an emphasis on feasibility and acceptability rather than efficacy. Nevertheless, to counter-act this obvious limitation, and to address the lack of suitable 'control', we decided to use a senior specialist grade physiotherapist who had experience in managing people with PPPD, to offer customised vestibular rehabilitation conforming to international 'gold-standard' guidelines (Hall et al., 2016).

6.11 Summary

This chapter discussed how the evidence presented in the previous chapters informed the development of an integrated CBT-VRT treatment for persistent dizziness. The process of developing the intervention following an intervention mapping framework (Bartholomew Eldredge et al., 2016) and input from stakeholders including PPI to enhance intervention acceptability. The intervention techniques and components were then discussed in detailed along with the guided self-management manual. The protocol for the feasibility testing of the intervention will be presented in the next chapter.

Chapter 7 [published manuscript]

Protocol for a randomised controlled feasibility study of psychologically informed vestibular rehabilitation for people with persistent dizziness: INVEST trial

Authors: David Herdman, Sam Norton, Marousa Pavlou, Louisa Murdin, and Rona Moss-Morris

7.1 Abstract

Background: Dizziness is a common complaint that often persists and leads to disability and distress. Several cognitive and behavioural responses may contribute to the neurobiological adaptations that maintain persistent vestibular symptoms. This paper will present the protocol of a two-arm parallel group feasibility randomised controlled trial designed to determine whether a fully powered efficacy trial is achievable by examining the feasibility of recruitment, acceptability and potential benefits of an integrated cognitive behavioural therapy and vestibular rehabilitation (CBT-VR) treatment for people with persistent dizziness.

Methods: Forty adult patients will be recruited from a tertiary vestibular clinic with persistent movement–triggered dizziness for 3 months or longer who have moderate–high levels of dizziness handicap. Participants will be 1:1 randomised, using a minimisation procedure, to six sessions of either CBT-VR (intervention arm) or VR only (control arm). Measures will be collected at baseline and 4 months post randomisation. The primary feasibility outcomes include descriptive data on numbers meeting eligibility criteria, rates of recruitment, numbers retained post randomisation, treatment adherence and an acceptability questionnaire. Treatment effects on self-report outcomes will be estimated to determine that 95% confidence intervals for the effects are consistent with anticipated effects and minimum clinically important differences, and to provide information needed for the power calculation of an efficacy trial. A nested qualitative study will be conducted post-intervention (intervention group only) to explore the acceptability of the intervention and identify any areas in need of improvement.

Discussion: If a trial of CBT-VR is feasible, acceptability data will be used to enhance the intervention if needed and refine the multicentre RCT protocol. Future studies will need to consider the training required for other physiotherapists to deliver the intervention.

Trial registration: ClinicalTrials.gov, ISRCTN 10420559

7.2 Introduction

Vertigo and dizziness are common complaints in the general population and are often caused by vestibular disorders [1]. Dizziness as a symptom can persist in patients with vestibular disorders even after the recovery of the acute crisis and lead to functional vestibular syndromes [2, 3]. It is frequently accompanied by unsteadiness and a range of other unpleasant and disabling symptoms such as blurred vision, nausea, pallor, psychological complaints, and cognitive deficits in spatial navigation, memory, attention, executive function and body schema [4].

Vestibular rehabilitation (VR) is an exercise-based treatment recommended for people with persistent dizziness and balance symptoms [5]. VR aims to facilitate the ability of the central nervous system to 'compensate' and restore normal function [6]. The exercises are based on principles of habituation and adaptation/substitution, in addition to balance retraining [7]. Patients are expected to carry out a home-based exercise programme over a number of weeks or months with graded exposure to dizziness-provoking stimulus as core to the intervention. However, in some of the randomised trials, only around 50% of subjects in the intervention group achieve the desired level of subjective improvement in dizziness symptoms [8]. In clinical practice, around 25% do not improve at all depending on which outcome measure is used, and the majority continue to report 'bothersome' symptoms [9].

Since psychological factors are intrinsically linked with recovery from balance disorders, a combination of cognitive behavioural therapy (CBT) and VR has been recommended for a long time now [10]. Indeed, physiotherapists working in vestibular rehabilitation consider managing aspects of anxiety within their scope of practice but acknowledge the need for tailored training and guidance [11]. Tailored training requires an evidence-based manualised CBT treatment capable of synergistically targeting mental and physical health aspects of dizziness. In a systematic review, four randomised controlled trials (RCTs) reported improvement in dizziness following therapy, combined with VR or relaxation techniques [12]. However, the sample sizes were small, and the effects on dizziness outcomes tended to be weak, with one study evaluating long-term effects finding similar results to those obtained before treatment [13]. The components of the therapy were not described in detail and did not involve a strict manual.

Since then, Edelman et al. [14] found reductions in dizziness outcomes, avoidance and safety behaviours, but not depression or anxiety in a short 3-session psychological intervention compared with a waiting list control. These effects were maintained after 6 months, although higher levels of anxiety predicted higher levels of disability [15]. A recent feasibility study evaluated a group intervention based on traditional VR and a model of CBT based on panic anxiety. Only one participant experienced a meaningful improvement in pre and posttreatment scores on the subjective dizziness outcome suggesting CBT based on panic, and/or group-based treatment may not be the best protocol [16].

These studies highlight that there is no agreed theoretical framework or manualised treatment protocol, which sufficiently integrates the psychological and self-management needs of patients with chronic dizziness. This makes it difficult to replicate interventions but also raises important theoretical implications when CBT protocols are based on empirical cognitive–behavioural models of depression and anxiety. In these models, emotions are conceptualised as primary mental health disorders rather than a reaction to objectively challenging symptoms. These protocols also fail to address illness-specific behavioural self-management techniques.

For individuals experiencing persistent dizziness, a CBT protocol which remains contextually anchored to their experience of living with dizziness may ultimately promote better engagement with rehabilitation and improve health outcomes. We conducted a theoretical modelling prospective study which revealed the importance of a variety of illness-specific cognitive and behavioural factors in the experience of dizziness-related disability [17, 18]. This was drawn together with a review of the literature to develop a model specific to dizziness (article in preparation), and we then used intervention mapping techniques [19, 20] to design an intervention and detailed manual which integrated CBT methods into traditional VR.

The aim of this study is therefore to evaluate the feasibility of the manualised 'INVEST' (integrated CBT-VR) protocol, for participants with persistent dizziness, as part of the preparation for a full-scale randomised controlled trial.

Primary objectives

The following are the primary objectives:

- To determine the recruitment rate
- To assess retention of participants by estimating follow-up rates
- To assess the acceptability of the intended self-report outcome measures for a future definitive trial (i.e., questionnaire feedback, completion rates, item-level missing data, floor/ceiling effects and estimates of variance)
- To explore the level of acceptability of the interventions through a survey and by measuring percentage of patients completing each of the interventions

- To formulate a suitable method to measure physiotherapist fidelity for a future multicentre trial

Secondary objectives

The following are the secondary objectives:

- To explore treatment effects on self-report outcomes to determine that 95% confidence intervals for the effects are consistent with anticipated effects and minimum clinically important differences
- To estimate key elements that would inform a power calculation to inform a power calculation for an efficacy study
- To qualitatively explore patient perceptions of the credibility, acceptability and usefulness of the intervention and identify areas of improvement for a future full-scale trial

7.3 Methods

Design

This feasibility randomised control trial with nested qualitative study will be composed of two-armed, parallel groups, to gather preliminary information on the intervention (INVEST) and the feasibility of conducting a full-scale trial.

Setting

Participants will be recruited and treated at the audio-vestibular and physiotherapy service at St George's University Hospitals NHS Foundation Trust. Questionnaires and outcome assessment will be done online.

Sample size

In agreement with current recommendations for pilot study sample size, 20 participants will be included in each group [21, 22]. Given a sample size of 40, assuming participation rates of 33% and drop-out rates of 20%, it will be possible to estimate 95% confidence intervals for the participation and drop-out rates within a maximum interval of ± 9% and ± 16% respectively.

Participants

Participants will be recruited who must meet all the following criteria:

- Patients attending the neuro-otology balance clinic at St George's University Hospitals Foundation Trust with symptoms of chronic dizziness (≥ 3 months) made worse by movement of the self and/or the environment

- Have a vestibular diagnosis[1]
- Dizziness Handicap Inventory (DHI) ≥ 40
- Aged ≥ 18 years
- Not currently participating in vestibular rehabilitation or psychological treatment (talking therapies)
- Able to provide consent and willing to comply with the proposed training and testing regime

Participants will be excluded if they meet one or more of the following exclusion criteria:

- Patients with vestibular migraine or other headache/migraine disorder with ≥ 3 headaches a month and/or MIDAS (Migraine Disability Assessment) ≥ 6 since they would not usually be suitable for vestibular rehabilitation until their headaches are under control
- Patients with active Meniere's disease or BPPV (benign paroxysmal positional vertigo) since they would not usually be suitable for vestibular rehabilitation until their vestibular function is stable
- Patients with central vestibular disorders (excluding migraine and functional disorders), other neurological disorders, bilateral vestibulopathy or acute severe mental health illness since these conditions would interfere in the outcome of rehabilitation
- Patients with acute orthopaedic disorders influencing balance control and gait
- Insufficient grasp of written/spoken English or have special communication needs

Flow of recruitment and participant timeline

Patients will be approached to participate by the Audiovestibular Physicians in the vestibular clinic who will complete the initial screen (see Figure 10). Interested potential participants will be given a participant information sheet and contacted by the principal investigator (DH) for telephone screening to make sure they meet all the inclusion criteria (e.g., DHI criteria). Consent forms and baseline questionnaires will be completed online. Participants will then be randomised to either the intervention group or the control group. Follow-up data will be collected at 4 months post randomisation, and data will be anonymised. On completion of the postintervention measures, a subsample of participants will be invited to take part in the qualitative interview.

[1] The early trial register stated that patients with ongoing investigations would be excluded. Due to the current restrictions on vestibular testing during the COVID-19 pandemic, we will not exclude patients based on this criterion. Nevertheless, we still expect the Audiovestibular Physicians to make a reasonable clinical diagnosis based on the Barany classification and to defer recruitment should investigations be essential to make a diagnosis.

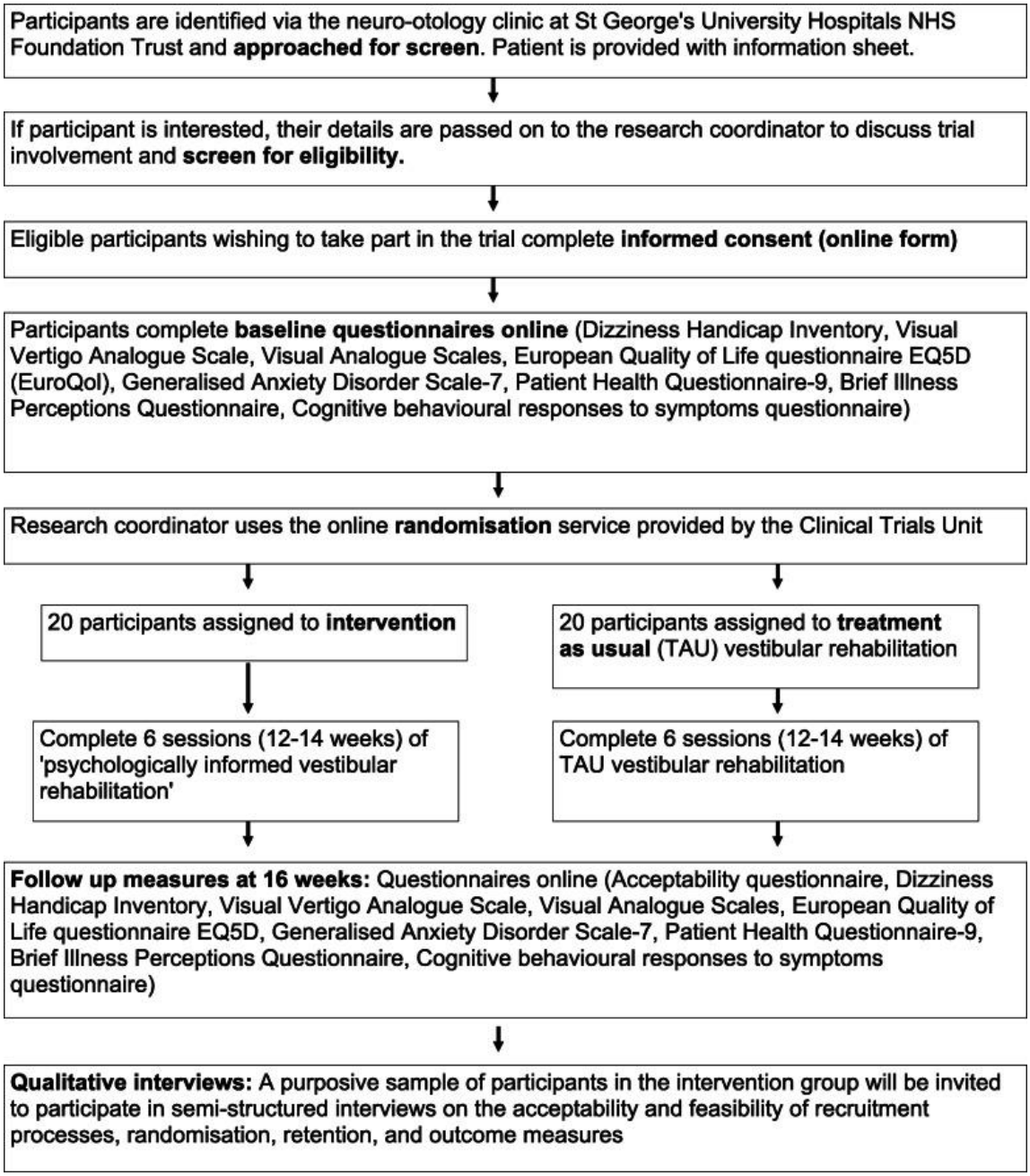

Figure 10. Trial flowchart

Randomisation and blinding

Participants will be randomised consecutively, and physicians will be blinded to allocation sequence. Randomisation will be completed by the King's Clinical Trials Unit via an online electronic system using a minimisation procedure with a probability of 0.8 to assure similar distribution of selected participant factors between trial groups, to include three dichotomous outcomes: gender (male/female), age (18–60/over 60) and dizziness handicap (DHI score 40–59/≥ 60).

Interventions

INVEST intervention

The treatment is a tailored integrated cognitive behavioural therapy–vestibular rehabilitation (CBT-VR)–based intervention with therapist support. The purpose of this intervention is to target individual's dizziness beliefs and cognitive–behavioural responses to symptoms in order to facilitate vestibular rehabilitation. The development of the intervention was systematic, based on findings of a review and prospective studies, with substantial input from patient and public representatives and a multidisciplinary team of health psychologists, physiotherapists and audiovestibular physicians. The structure and content of the manual was drafted based on previous CBT interventions developed by the department of Health Psychology at King's College London [23–25], and other sources [26–28].

Participants will be provided with a structured therapy manual including worksheets. This will be accompanied by six sessions with the primary researcher (DH) who has experience in working with patients with severe dizziness as a physiotherapist and has received some basic training in CBT. In accordance with CBT-VR principles, participants will be encouraged to complete tasks and exercises between sessions. The first session will be structured around education and include an individual formulation and cognitive behavioural analysis of the dizziness problem. The general point of the first session is that the patient's behavioural responses are a normal defensive response to the aversive stimuli, which may have been adaptive in acute dizziness but have lost their efficacy as the dizziness has persisted.

The participant is guided towards sections of the manual that may be more relevant to their own problem. It includes the following components:

1. Education: Educational content about persistent dizziness is provided, and participants are given space to develop their own case formulation to help make sense of their experiences from a psychophysiological perspective.
2. Goal setting: Worksheets allow participants to set goals for therapy. Specific functional goals are encouraged that redirect the focus of attention toward daily life activities and are broken down into achievable steps.
3. Activity monitoring: Worksheets help participants to identify avoidance and/or all-or-nothing behaviours, establish activity tolerance levels and identify discriminative stimuli eliciting dizziness behaviours. Participants are encouraged to adopt a consistent and balanced approach to activities through planning activity diaries.
4. Distraction techniques: Education about distraction with in-session behavioural experiments to demonstrate the effects of symptom focusing and attention switching on dizziness and balance

5. Reattribution of symptoms: Participants are encouraged to identify symptoms and reattribute them to either symptoms of their condition, medication, deconditioning, stress and anxiety or depression.
6. Relaxation techniques: The link between autonomic anxiety and dizziness is presented and relaxation methods introduced including diaphragmatic breathing, progressive muscle relaxation and guided imagery relaxation.
7. In vivo exposure: Participants identify avoidance and safety behaviours and establish a dizziness-related fear hierarchy followed by graded exposure to fear eliciting activities in a series of behavioural tests during which catastrophic expectations are challenged.
8. Cognitive therapy: The link between thoughts and behaviours is presented, participants encouraged to identify their own thoughts, and worksheets to restructure the dizziness-related beliefs and behavioural experiments designed to challenge maladaptive beliefs.
9. Problem solving: A review of information and strategies implemented so far, and review of progress are made with additional information on fear beliefs, perfectionism, managing financial and work-related stress, and sleep problems. Sleep restriction therapy is recommended where appropriate.
10. The potential for dizziness flare-ups is managed proactively by attempting to alter the patient's expectations and reduce the likelihood of catastrophising throughout therapy. The patient reflects on progress made over the 6 sessions and develops a relapse management toolkit.

Although originally, all sessions were designed to be face to face, to be consistent with current service provision due to the COVID-19 pandemic, we will not discriminate against people who cannot attend in person, and instead offer them the same therapy over video consultation software.

The first session will last 1 h, while the remaining five sessions will last 30 min. This is consistent with current physiotherapy clinical practice. Table 15 includes a summary of content for the sessions. As a general rule, participants may need sessions once a fortnight initially, but the time between sessions becomes more spaced out as therapy progresses, and they become more independent, for 12–14 weeks.

Table 15. Summary of content for the sessions

Summary of sessions	
Session	**Content**
1	Understanding the problem (formulation) Familiarisation with workbook Symptom control techniques Homework: activity monitoring
2	Review activity diary Goal setting Physiotherapy exercises

Summary of sessions	
Session	**Content**
	Activity planning Homework: activity and rest goal setting
3	Review sleep, activity and rest goal sheet In vivo behavioural experiments Homework: behavioural experiments & exposure training
4	Review homework Review of beliefs and cognitions Progress physiotherapy exercises Homework: thought diary
5	Review thought diary Review of progress and problem solving Homework: depending on identification of ongoing problems (e.g., sleep therapy, etc.)
6	Planning for the future Relapse management

Treatment as usual

Treatment as usual will be vestibular rehabilitation, consisting of specific exercise techniques to target identified impairments or functional limitations, delivered by a senior specialist vestibular physiotherapist at St George's Hospital. The physiotherapy will be consistent with the latest evidence-based Clinical Practice Guidelines [29]. Participants will also be asked to complete a home exercise programme. The session duration and schedule will be the same as the intervention with the first session lasting an hour and follow-up appointments 30 min, up to six sessions between 12 and 14 weeks.

Clinical supervision

DH has attended training to deliver low-intensity CBT techniques and will undergo further training with role-played sessions with feedback from RMM. Ongoing supervision will be provided by RMM. Shared reflection of recorded sessions will be discussed in line with the core competency framework for delivering psychological therapies in long-term conditions [30].

Intervention fidelity

The therapist delivering the intervention sessions will follow the detailed and structured manual developed for the patients. With permission from the participants, sought on the consent form, therapy sessions will be video recorded and assessed for fidelity during supervision by RMM.

Primary feasibility outcomes

Feasibility will be assessed by collecting descriptive data on recruitment and retention rates and willingness to be randomised according to the Consolidated Standards of Reporting Trials feasibility and pilot trial guidelines [31]. The following will be recorded:

- Suitability of eligibility criteria: number of people excluded from the trial and for what reasons. This will allow us to assess whether the criteria are appropriate.
- Willingness to participate: the proportion of eligible patients who agree to participate.
- Retention rates: the proportion of participants who were randomised that completed follow-up assessment as well as recording of attendance at therapist sessions. If participants drop out, we will attempt to contact them to find out the reasons.
- Time needed to collect and analyse data: time sheets will record the duration of collection and analysis of the data.
- Acceptability/satisfaction of the intervention: This will be evaluated at follow-up using a questionnaire based on the component constructs in the theoretical framework of acceptability [32]. It will take the form of eight statements using a five-item Likert response scale (strongly agree to strongly disagree):
    - I feel positive about the treatment.
    - The amount of effort required to participate in the treatment was acceptable.
    - The treatment fits with my values.
    - The treatment made sense to me.
    - The time involved in engaging in the treatment was acceptable to me.
    - The treatment was effective to help me manage my condition.
    - I was able to perform the activities required to participate in the treatment.

Self-report outcomes

Dizziness handicap

The Dizziness Handicap Inventory (DHI) [33] consists of 25 questions designed to assess physical, functional and emotional aspects of dizziness-associated disability and 'handicap'. For each question, the participant can choose 'yes', 'no' or 'sometimes', and the total score ranges from 0 to 100 with higher scores indicating more severe handicap and activity restriction. With high test–retest reliability and low error of measurement scores, the DHI has been widely adopted in clinical practice and trials to evaluate the effects of vestibular rehabilitation with mixed dizziness diagnoses [33–35].

Visually induced dizziness

The Visual Vertigo Analogue Scale (VVAS) [36] is a nine-item visual analogue scale that rates the intensity of dizziness during daily situations typically inducing 'visually induced dizziness' (ViD) such as 'walking through a supermarket aisle' or 'watching action television'. Intolerance of visual motion is a common symptom for people with chronic vestibulopathy induced by dynamic visual input and has been shown to be a negative prognostic indicator [2, 37]. The VVAS shows validity and responsiveness to change [38].

Dizziness interference

Dizziness interference will be calculated using a visual analogue scale. Participants will answer the question, 'Over the past week, what percentage of the time has dizziness interfered with your activities?' by drawing a vertical line across a 10-cm line with 20% increments. Test–retest reliability for this tool is excellent [39].

Health-related quality of life

The European Quality of Life questionnaire EQ5D (EuroQol) [40] measures health-related quality of life for clinical and economic appraisal. The first part of the instrument is a self-reported description of the subject's health using a five-dimensional classification. It contains five items, each with three response choices. The answers are converted into a score ranging up to 1.00, indicating high health-related quality of life. The second part is a self-rated valuation of the subject's health using a vertical VAS in the form of a thermometer ranging from 0 (worst imaginable state of health) at the bottom to 100 (best imaginable state of health) at the top. The test–retest and inter-rater reliability has been established for patients with dizziness and disequilibrium [41] and has been used to assess cost effectiveness of vestibular rehabilitation [42].

Balance

The trial register entry (ISRCTN 10420559) includes a blinded mini-Balance Evaluation Systems Test (mini-BESTest) [43, 44]. Due to the COVID-19 local restrictions, it is no longer possible for participants to attend in person for this test, and it has therefore been removed from the protocol. When possible, the physiotherapists will be encouraged to complete and record this assessment as part of their initial evaluation.

Self-report outcomes: process variables

The following self-report outcomes will be targets for the intervention so will also be assessed:

Illness perceptions

The Brief Illness Perception Questionnaire (B-IPQ) [45] is a nine-item scale where each item assesses one dimension of illness perceptions. In accordance with the recommendations from the authors, the word 'illness' will be replaced by 'dizziness condition' in order to reflect the specific dizziness illness–related perceptions of participants. It may be possible to compute an overall score which represents the degree to which the illness is perceived as threatening or benign. The internal consistency of this score will be checked.

Cognitive and behavioural responses to dizziness

The Cognitive and Behavioural Responses to Symptoms Questionnaire (CBRQ) [46] assesses patients' cognitive and behavioural responses to the experience of symptoms. The five subscales dealing with cognitive responses are symptom focusing (e.g., 'I think a great deal about my dizziness'), catastrophising (e.g., 'I will never feel right again'), damaging beliefs (e.g., 'dizziness is a signal that I am damaging myself'), fear avoidance (e.g., 'I should avoid exercise when I have dizziness') and embarrassment avoidance (e.g., 'The embarrassing nature of my dizziness prevents me from doing things'). The two behavioural subscales are all or nothing (e.g., 'I find myself rushing to get things done before I crash') and avoidance/rest (e.g., 'I stay in bed to control my dizziness'). High scores indicate more unhelpful responses, and the reliability and validity have recently been established for patients with dizziness [47].

Anxiety and depression

Depressive symptoms will be assessed using the Patient Health Questionnaire-9 (PHQ-9) [48], and anxiety will be assessed using the Generalised Anxiety Disorders-7 Questionnaire (GAD-7) [49]. These questionnaires have been widely validated in physically ill populations, and higher scores indicate more severe symptoms.

Other treatments

Participants will be asked whether they have received any pharmacological, psychological or exercise-based treatment in addition to INVEST since starting the study.

Adverse events

Information about occurrence of serious adverse events since the start of the study will be reported according to good clinical practice guidelines. Adverse events will be flagged up to the trial management team, and participants will be contacted to further assess the adverse event and its relationship to the study.

Sociodemographic and clinical characteristics

Sociodemographic characteristics including gender, age, ethnicity and level of education will be collected at baseline via self-report. Clinical characteristics will include the diagnosis and will be verified at baseline according to their clinical records. The clinical diagnoses will be made by an Audiovestibular Physician based on the Barany diagnostic criteria.

Qualitative interviews

Qualitative methods will be used in order to obtain a more comprehensive understanding of acceptability of the trial requirements and therapy approach, therapy outcomes and feedback on the intervention.

The sample will be recruited from the feasibility trial, and the study will be nested within the main trial. Participants will be asked for additional consent to be interviewed. When each of these participants completes their trial intervention and their post-therapy assessment, a decision will be made as to whether to contact them for interview. After the first 10 interviews, sampling will become increasingly purposive with the aim of interviewing a sample with maximum variation. We will seek variation in terms of demographics, and attitudes towards therapy as gleaned from responses to Likert scale questions described previously. The sample will not be selected to be representative of the trial participants but to include people likely to hold different viewpoints.

Interviews will be scheduled as soon as possible after completion of the post-therapy questionnaire assessments. The interview will consist of a series of open-ended questions relating to expectations of the interventions, how participants found the therapy and any changes they had experienced. All interviews will be recorded and transcribed verbatim.

Analysis plan

Descriptive statistics of patients approached, screened, eligible, consented and randomised will be computed to address the primary objectives. Reasons for non-consent, exclusion and drop-out, at each stage of the study, will be recorded and reported. Adherence to the intervention will be reported using descriptive statistics to include the mean number of sessions completed, a breakdown on the number of participants completing each session and mean duration of the sessions. To account for uncertainty due to sampling error, all estimates will be presented with 95% CIs. The standard deviation of the key self-reported outcome and the correlation between the baseline and follow-up assessments of the outcomes will be computed to inform the sample size for a future efficacy trial.

To address acceptability, a mixed methods approach will be used, drawing on both the quantitative and qualitative findings to determine any intervention-specific issues, including whether the number and pacing of sessions seemed sufficient.

The psychometric adequacy of the self-report instruments used will be assessed to address the secondary outcomes. Floor and ceiling effects will be considered as a key indicator of potential sensitivity of the scale to detect changes. Reliability will be assessed using Cronbach's alpha, with a minimum acceptable cut-off at $\alpha = 0.70$, but preferably at $\alpha = 0.80$ or higher, particularly for the key variables. Non-completion of individual items will be checked to ensure that there are no potentially problematic items for this patient population.

An analysis of covariance (ANCOVA) approach will be performed to estimate the postintervention mean difference in outcomes: dizziness disability (handicap), dizziness severity, dizziness interference, depression and anxiety. Given the feasibility nature of the trial, the statistical significance of any post-randomisation group differences will not be assessed; instead, effect sizes and CI will be estimated and used for interpretation. Each analysis will adjust for the baseline level of the outcome variable and factors used in the minimisation procedure. Group allocation will be included as an indicator variable following the intention-to-treat principle.

Finally, to qualitatively explore the acceptability and usefulness of the intervention from the perspective of the participants, the semi-structured qualitative interviews will be transcribed verbatim and analysed using inductive thematic analysis with the use of NVivo software. Thematic analysis revolves around identifying recurrent themes and patterns from the interviews and developing a coding manual [50].

Progression criteria

To inform the decision whether to proceed to a full-scale efficacy trial, the following a priori criteria will be used. We will deem the trial appropriate to progress if (1) ≥ 70% of eligible patients participate; (2) drop-out rate is < 20%; (3) there are comparable acceptability ratings to the TAU based on the quantitative and qualitative data and (4) ≥ 60% adhere to sessions. The 'Stop' criteria will consider if (1) < 30% of eligible patients participate; (2) drop-out rate is > 40%; (3) < 60% of participants report acceptability of the intervention according to quantitative and qualitative data and (4) < 30% adherence to sessions. Stop criteria will also consider if there are irreconcilable serious adverse events attributed to the intervention (e.g., due to behavioural experiments, in vivo exposure, etc.). Where the assessment outcome falls between the 'Go' and 'Stop' criteria, the trial committee will consider the data and identify steps needed to progress to a full-scale trial. The trial committee will consider the data presented and make a judgement about whether the methodology and intervention were delivered as intended. We will also use the experience from clinicians and participants to further optimise the intervention and manual.

7.4 Discussion

In recent years, there has been a greater demand to integrate cognitive approaches and enhance the behavioural aspects of vestibular rehabilitation. This protocol represents such an integrated treatment designed specifically to manage the problems associated with the maintenance of persistent dizziness. It represents a theory driven and scientific approach designed following the Medical Research Council guidance [20] for developing and evaluating complex interventions. It has also been designed to be delivered by vestibular physiotherapists, which offers a pragmatic solution to the problem accessing psychological treatment interventions tailored to the specific problems associated with dizziness.

This study is limited because it is single site and includes only one treating therapist in the intervention arm. Future studies will need to consider the training required for other physiotherapists to deliver the intervention. There may be restrictions on participants attending in person due to the current pandemic. As a first step, this study will identify unique challenges that occur in the recruitment and retention of patients and will be able to examine the acceptability of this treatment to patients in terms of whether its content was relevant and useful. This will allow the researchers to further refine the intervention, consider the most suitable training needs for therapists, and substantially inform the design of a future large-scale trial powered to detect the efficacy of integrated CBT and VR treatments for the management of persistent dizziness, accompanied by a longer follow-up to assess any sustained effects of the intervention on outcomes.

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Chapter 8 [manuscript in press]

The INVEST trial: A randomised feasibility trial of psychologically informed vestibular rehabilitation versus current gold standard physiotherapy for people with Persistent Postural Perceptual Dizziness

8.1 Abstract

Objective: To determine the feasibility and acceptability of conducting a randomised controlled trial of cognitive-behavioural therapy informed vestibular rehabilitation (the INVEST intervention) designed for persistent dizziness.

Methods: A two-armed parallel groups randomised feasibility study of INVEST vs. a time matched gold standard vestibular rehabilitation (VRT) control. Participants with PPPD (persistent postural perceptual dizziness) were recruited from a specialist vestibular clinic in London, UK. Participants were individually randomised using a minimisation procedure with allocation concealment. Measures of feasibility and clinical outcome were collected and assessed at 4 months.

Results: Forty adults with PPPD were randomised to six sessions of INVEST (n=20) or gold standard VRT (n=20). Overall, 59% of patients screened met the inclusion criteria, of which 80% enrolled. Acceptability of INVEST, as assessed against the Theoretical Framework of Acceptability (TFA), was excellent and 80% adhered to all 6 sessions. There were small to moderate treatment effects in favour of INVEST across all measures, including dizziness handicap, negative illness perceptions, symptom focussing, fear avoidance, and distress (standardised mean difference $[SMD]_g$ = 0.45; SMD_g = 0.77; SMD_g = 0.56; SMD_g = 0.50, respectively). No intervention-related serious adverse events were reported.

Conclusions: The study results give strong support for the feasibility of a full-scale trial. Both arms had high rates of recruitment, retention, and acceptability. There was

promising support of the benefits of integrated cognitive behavioural therapy-based vestibular rehabilitation compared to gold standard vestibular rehabilitation. The study fulfilled all the a-priori criteria to advance to a full-scale efficacy trial.

Trial registration number: ISRCTN10420559

Key Messages

What is already known on this topic – Persistent Postural Perceptual Dizziness (PPPD) is a common and disabling functional neuro-vestibular disorder. A course of vestibular rehabilitation is usually advised but many patients remain disabled by symptoms.

What this study adds –Combining elements of cognitive-behavioural therapy with vestibular rehabilitation demonstrated excellent acceptability, feasibility, and signals of greater reductions in dizziness in people with PPPD than current gold-standard treatment.

How this study might affect research, practice or policy – the study met criteria to advance to a full-scale trial with further considerations to optimise the intervention.

8.2 Introduction

Persistent Postural Perceptual Dizziness (PPPD) is a complex functional neuro-vestibular disorder characterised by persistent dizziness, non-spinning vertigo and/or unsteadiness (Staab et al., 2017). It is thought to be a long-term maladaptation to neuro-otological, neurological or medical illness, and/or psychological distress. Since its international classification by the Bárány Society (Staab et al., 2017), PPPD is increasingly recognised as the single most common vestibular syndrome in specialised outpatient clinics (Strupp et al., 2020) and likely represents the vast majority of patients referred to vestibular rehabilitation (Staab, 2011). People living with PPPD have poor quality of life, severe dizziness handicap and an elevated risk of anxiety and depression (Azzi et al., 2021; Sui Lin & Prepageran, 2021).

Tailored treatment strategies have been recommended, including pharmacotherapy with selective serotonin reuptake inhibitors (SSRI), physiotherapy (vestibular rehabilitation) and cognitive-behavioural therapy (CBT), but there is a lack of prospective, randomised controlled trials or information on prognosis or outcomes (Popkirov et al., 2018b). Vestibular rehabilitation therapy (VRT) is an established exercise-based treatment for people with structural vestibular disorders (McDonnell & Hillier, 2015) that is usually recommended for people with PPPD (Nada et al., 2019; Thompson et al., 2015). However the exercises must be carefully graded to avoid intolerable symptom provocation and psychological factors are known to negatively affect outcome (Whitney et al., 2020). There is limited evidence in favour of CBT in PPPD (Edelman et al., 2012), although one study reported short-term relief (Holmberg

et al., 2007). However, there are better results when CBT is adapted to target illness specific factors such as anxiety-related postural behaviour (Best et al., 2015). There are also promising multidisciplinary programs (Axer et al., 2020; Limburg et al., 2019), but these can be costly and difficult to replicate. Due to their similarities, there has been a desire to combine CBT and VRT for a long time (Beidel & Horak, 2001; Staab, 2011; Yardley & Redfern, 2001), but no theory-driven, evidence-based intervention with a standardised treatment manual currently exists. To date there are only a few case reports and pilot studies (Andersson et al., 2006; Johansson et al., 2001; Kristiansen et al., 2019; Kuwabara et al., 2020; Schmid et al., 2018). Moreover, previous trials do not test interventions against current best practice.

To address this gap, we developed a combined CBT-VRT intervention based on existing research data and theoretical modelling of the psychological factors that contribute to dizziness handicap (Herdman et al., 2020a, 2020b). Based on those findings and working in partnership with patient representatives we developed a patient manual. We believe there is a better chance of acceptability and success when the intervention can remediate specific perpetuators of dizziness and be integrated within a physiotherapy programme.

The aim of this study was to evaluate the feasibility and acceptability of the integrated CBT and VRT (INVEST) intervention and trial methodology, for people with PPPD, as part of the preparation for a full-scale randomized controlled trial. Specific objectives were to determine the recruitment and retention rate, to test the utility of a range of outcome measures, levels of acceptability, assess adherence and to collect outcome data to explore treatment effects and estimate key elements that would inform a large-scale trial. For a breakdown of the detailed study objectives and predefined progression criteria, please see the published protocol (Herdman et al., 2021b).

8.3 Methods

Design

Two-armed parallel groups randomised controlled single centre feasibility trial with online assessment before randomisation (T0) and at follow-up four months post-randomisation (T1). Participants in the INVEST arm were also invited to participate in a qualitative interview after T1 (results will be reported elsewhere). There were no changes from the published research protocol, which contains more detailed methods and intervention specifics (Herdman et al., 2021b).

Setting

An outpatient tertiary (specialist) setting at St George's University NHS Foundation Trust in urban London, United Kingdom. Recruitment was between November 2020 and August 2021 but was discontinuous due to the status of clinics during the COVID-19 pandemic.

Participants

Adults (aged 18 or over) with persistent movement triggered dizziness for ≥ 3 months due to a vestibular diagnosis (according to the international classification of vestibular disorders), scoring ≥ 40 on the Dizziness Handicap Inventory (DHI), able to read and speak English, and willing and able to take part in the study were eligible.

Patients were excluded if they had another active condition which could interfere with their ability to participate in physiotherapy, including ≥ 3 headache/migraines a month, severe mental health disorder, another neurological disorder, acute orthopaedic disorders affecting balance and gait, and active Meniere's disease or Benign Paroxysmal Positional Vertigo (BPPV). We also excluded patients with central (such as strokes, intracranial tumors, degenerative disorders and metabolic conditions, but not including functional dizziness/PPPD or vestibular migraine)(Choi & Kim, 2017) or bilateral vestibulopathy (according to the Barany criteria (Strupp et al., 2017)).

Participants were identified by Audio-Vestibular Physicians and/or on referral to the vestibular physiotherapy department. Pre-screening excluded patients with active BPPV or unrelated audio-vestibular disorders that do not require vestibular physiotherapy. Potential participants were screened for eligibility via telephone and sent a participant information sheet by email or post according to their preference. Participants enrolled by completing an online consent form. No compensation was provided for taking part.

Sample Size Determination

The intended sample size was 40 assuming participation rates of 33% and drop-out rates of 20%, to estimate 95% confidence intervals for the participation and drop-out rates within a maximum interval of ± 9% and ± 16% respectively.

Randomisation

The random allocation sequence was generated using a minimisation procedure with a probability of 0.8 to assure similar distribution of selected participant factors between trial groups, which included three dichotomous outcomes: gender (male/female), age (18–60/over 60) and dizziness handicap (DHI score 40–59/≥ 60). Participants were randomized consecutively in the order in which they were referred to the study, and all staff and patients were blinded to allocation sequence. Randomisation was implemented independently by King's Clinical Trials Unit via an online electronic system.

Interventions

The interventions are detailed in the protocol (Herdman et al., 2021b). Each arm was delivered by a different senior specialist grade physiotherapist (DH and KF).

INVEST

In brief, INVEST included six-sessions of individual CBT-informed VRT aimed specifically at dizziness (not depression or anxiety) with a patient manual and therapist support. The initial session was 60 minutes, follow-up appointments were 30 minutes, and all were led by the same physiotherapist (author DH) who had additional training in CBT. There was a focus on transparency in communication which started with a shared cognitive-behavioural formulation and psychoeducation. Exercises were customised and focussed on normalising any maladaptive postural strategies (e.g., 'high-threat' postural control) early on, and habituation. Exercises were performed in clinic and at home. Other techniques included goal setting, activity planning and graded exercise, attention allocation and relaxation techniques, cognitive therapy focussed on illness beliefs, exposure in-vivo with behavioural experiments for dizziness related fear, relapse management and prevention.

Vestibular rehabilitation (control)

The six-sessions of individual VRT were time-matched to the INVEST protocol. The VRT represented 'gold standard' treatment based on evidence-based Clinical Practice Guidelines (Hall et al., 2016) and recommendations for people with PPPD (Popkirov et al., 2018b) to promote graded habituation to movement and visual stimuli. Participants were provided with a customised exercise programme, performed in clinic and at home, which included a range of general exercises (e.g., walking programmes) and more specific adaptation, habituation, visual desensitisation, static and dynamic balance exercises.

Measures

Sociodemographic and Clinical Data

Self-reported sociodemographic data were collected at baseline. Clinical data were extracted from medical records at T0 and T1. A diagnosis of PPPD was based on the latest Barany classification (Staab et al., 2017). Since it is common for people with PPPD to have other vestibular disorders or conditions which provoke dizziness, relevant co-existing conditions were extracted from each participant's medical records. Results of any vestibular laboratory function testing were also extracted and interpreted according to their respective normative values.

Feasibility Outcomes

Numbers of eligible people recruited, willingness to be randomised and retention rates were collected. Acceptability was evaluated at follow-up using an eight-item scale to assess the constructs in the theoretical framework of acceptability (Sekhon et al., 2017).

Self-report Outcomes

Participants completed all self-report measures online at T0 and T1 independently at home including measures of Dizziness Handicap (DHI) (Jacobson & Newman, 1990), visually induced dizziness (Visual Vertigo Analogue Scale [VVAS]) (Dannenbaum et al., 2011), dizziness interference (Percentage of time symptoms interfere with life [%TSI]) (Hall & Herdman, 2006) and health status (European Quality of Life questionnaire [EQ5D]) (Group, 1990). All scales are previously well-validated in people with chronic dizziness (see protocol for details) (Herdman et al., 2021b).

Putative process variables measured included negative dizziness specific illness perceptions (Brief Illness Perception Questionnaire [B-IPQ]) (Broadbent et al., 2006), cognitive and behavioural responses to dizziness (Cognitive and Behavioural Responses to Symptoms Questionnaire [CBRQ]) (Picariello et al., 2022), depression (Patient Health Questionnaire-9 [PHQ-9]) (Spitzer et al., 1999), anxiety (Generalized Anxiety Disorders-7 [GAD-7]) (Spitzer et al., 2006) and combined distress (Patient Health Questionnaire Anxiety and Depression Scale [PHQ-ADS]) (Herdman et al., 2022). Internal consistency for all outcome measures was acceptable (Cronbach alpha all ≥0.7).

At T1, participants were asked to self-report any other new treatments started during the study and a record of any adverse events was updated throughout.

Balance

All participants completed either the mini–Balance Evaluation Systems Test (mini-BESTest) (Franchignoni et al., 2010) or a hybrid balance assessment T0 and T1. The hybrid balance assessment consisted of the Mini-BESTest excluding those items that were not possible to conduct virtually. The most difficult item to measure was reactive postural control since this requires a therapist to provide an external perturbation. To account for this missing data, patients were dichotomised as demonstrating either normal or abnormal balance control based on the available data and therapist judgement.

Statistical Analysis

Questionnaires were completed online and there was no question item missing data. Descriptive statistics were used to summarise the number of patients approached, screened, eligible, consented, and randomised. Reasons for non-consent, exclusion, and dropout, at each stage of the study, were recorded. Similarly, descriptive statistics were computed to report adherence to the intervention.

Internal consistency of the measures was assessed using the Cronbach alpha coefficient at both T0 and T1. The EQ5D health state utility score was calculated from individual health profiles using the value set for England (Devlin et al., 2018). Mean and standard deviations (SD) are provided for all self-report outcomes by visit and by treatment.

Estimates of treatment effect at T1 were based on an analysis of covariance (ANCOVA) to estimate the postintervention mean difference. The analysis adjusted for the baseline level of the outcome variable, baseline DHI, age and sex. Group allocation was included as an indicator variable following the intention-to-treat principle. Given the feasibility nature of the trial, with a small sample size not powered to detect between group differences, the statistical significance of any post-randomisation group differences was not assessed; instead, effect sizes were calculated as standardised mean differences using Hedge's g (SMDg) applying the small sample bias correction factor (Hedges & Olkin, 1985).

8.4 Results

Participant Flow and Feasibility outcomes

Participant flow is shown in Figure 11. After a pre-screen conducted by the medical team, 85 people were approached and 35 (41%; 95%CI 31% to 52%) excluded due to ineligibility (reasons in Figure 1). Seven (14%; 95%CI 6% to 26%) eligible patients did not want to be randomised, three (6%) because of concern about being in a trial. Another three patients were not recruited as they were untraceable or unavailable after initial screening. Forty out of 50 eligible participants (80% enrolment rate; 95%CI 66% to 90%) were recruited and randomly assigned to the INVEST intervention (n=20) or gold standard VRT (control; n=20).

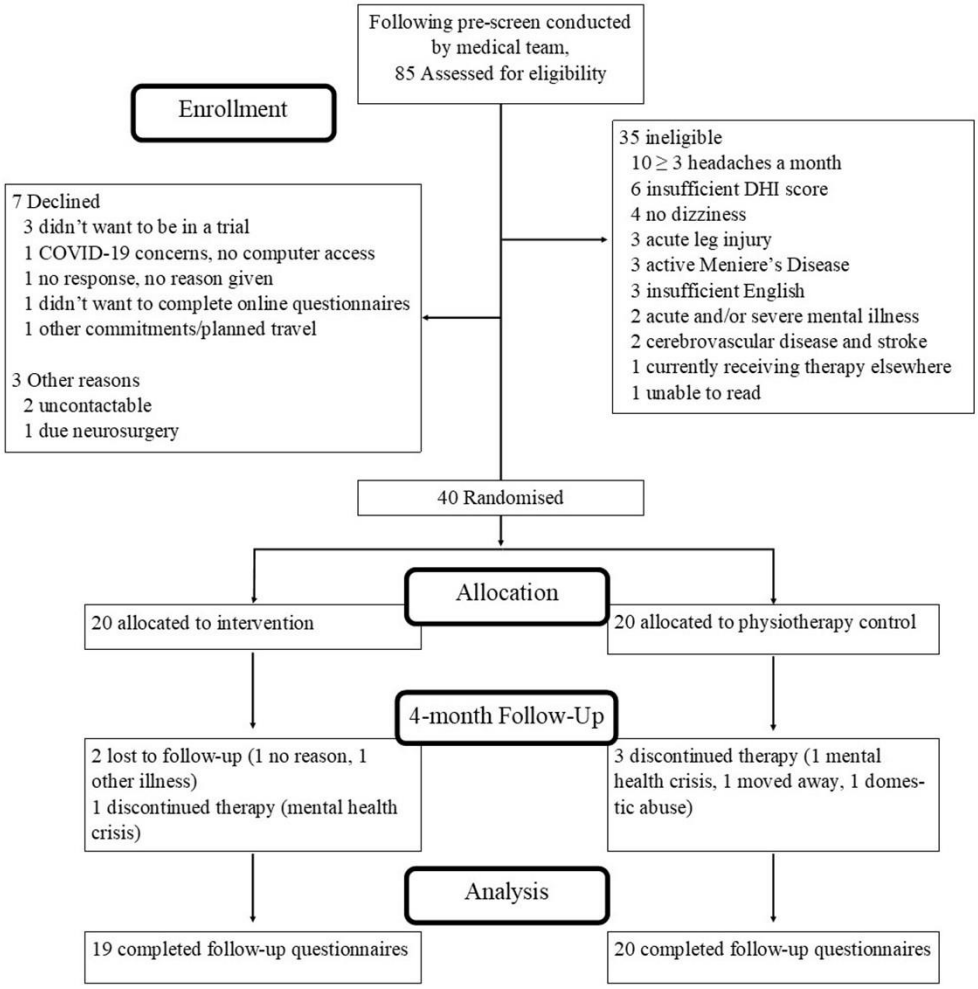

Figure 11. Participant flow-diagram.

Note: DHI = Dizziness Handicap Inventory

Drop-out rate from INVEST was 15% (95%CI 3% to 38%) and 20% for VRT (6% to 44%). One participant lost to follow-up did not complete the follow-up questionnaires (trial drop-out 2.5%; 95%CI <1% to 13%).

Baseline Characteristics

Table 16 shows baseline characteristics by group. The groups were equally distributed for age, sex, sociodemographic, clinical (including dizziness handicap), and psychological characteristics at baseline. The mean age was 44.5 years (SD = 17, range 19-79) and 32/40 (80%) were women. Median symptom duration was 2 years (IQR 46.5 months; range 5 months to 21 years).

Table 16. Baseline characteristics

	Group allocation	
	INVEST	VRT
Age (mean, SD)	44.60 (16.96)	44.30 (17.44)

Sex (n, %)		
Female	16 (80%)	16 (80%)
Male	4 (20%)	4 (20%)
Ethnicity (n, %)		
White	13 (65%)	15 (75%)
Mixed or Multiple ethnic groups	1 (5%)	1 (5%)
Asian or Asian British	2 (10%)	2 (10%)
Black, African, Caribbean, or Black British	4 (20%)	1 (5%)
Other ethnic group	0 (0%)	1 (5%)
Education (n, %)		
Higher Education	13 (65%)	12 (60%)
College, vocational level 3, and equivalents	3 (15%)	5 (25%)
High school, vocational level 2, and equivalents	0 (0%)	2 (10%)
Qualifications at level 1 and below	1 (5%)	0 (0%)
Other qualifications: level unknown	1 (5%)	0 (0%)
No qualifications	2 (10%)	1 (5%)
Employment status (n, %)		
Employed	13 (65%)	10 (50%)
Unemployed	4 (20%)	7 (35%)
Student	1 (5%)	2 (10%)
Retired	2 (10%)	1 (5%)
Clinical variables		
Diagnosis (n, %)		
Persistent Postural Perceptual Dizziness	20 (100%)	20(100%)
Illness duration, months (median, IQR)	24 (95)	21 (32)
Another related condition/trigger (n, %)		
Vestibular migraine	9 (45%)	8 (40%)
Clinical features of anxiety	9 (45%)	5 (25%)
Unilateral peripheral vestibulopathy	5 (25%)	8 (40%)
BPPV	2 (10%)	6 (30%)
Meniere's/Migraine overlap	0 (0%)	1 (5%)
Meniere's Disease	1 (5%)	0 (0%)
Vestibular testing abnormalities (n, %)		
Unilateral vestibular dysfunction	6 (30%)	2 (10%)
Normal vestibular function testing	11 (55%)	11 (55%)
On SSRI/SNRI medication (n, %)	1 (5%)	2 (10%)
Dizziness Handicap Inventory (mean, SD)	63.80 (17.84)	65.10 (14.76)

Note: SD = standard deviation; IQR = inter-quartile range; BPPV = Benign Paroxysmal Positional Vertigo; SSRI = selective serotonin reuptake inhibitors; SNRI = Serotonin-norepinephrine reuptake inhibitor. VRT = vestibular rehabilitation.

Acceptability

Figure 12 shows responses to the acceptability questionnaire. More than 80% of participants in both arms rated 'agree' or 'strongly agree' in favour for each domain. Participants in the INVEST arm tended to have slightly stronger positive opinions compared to the control arm.

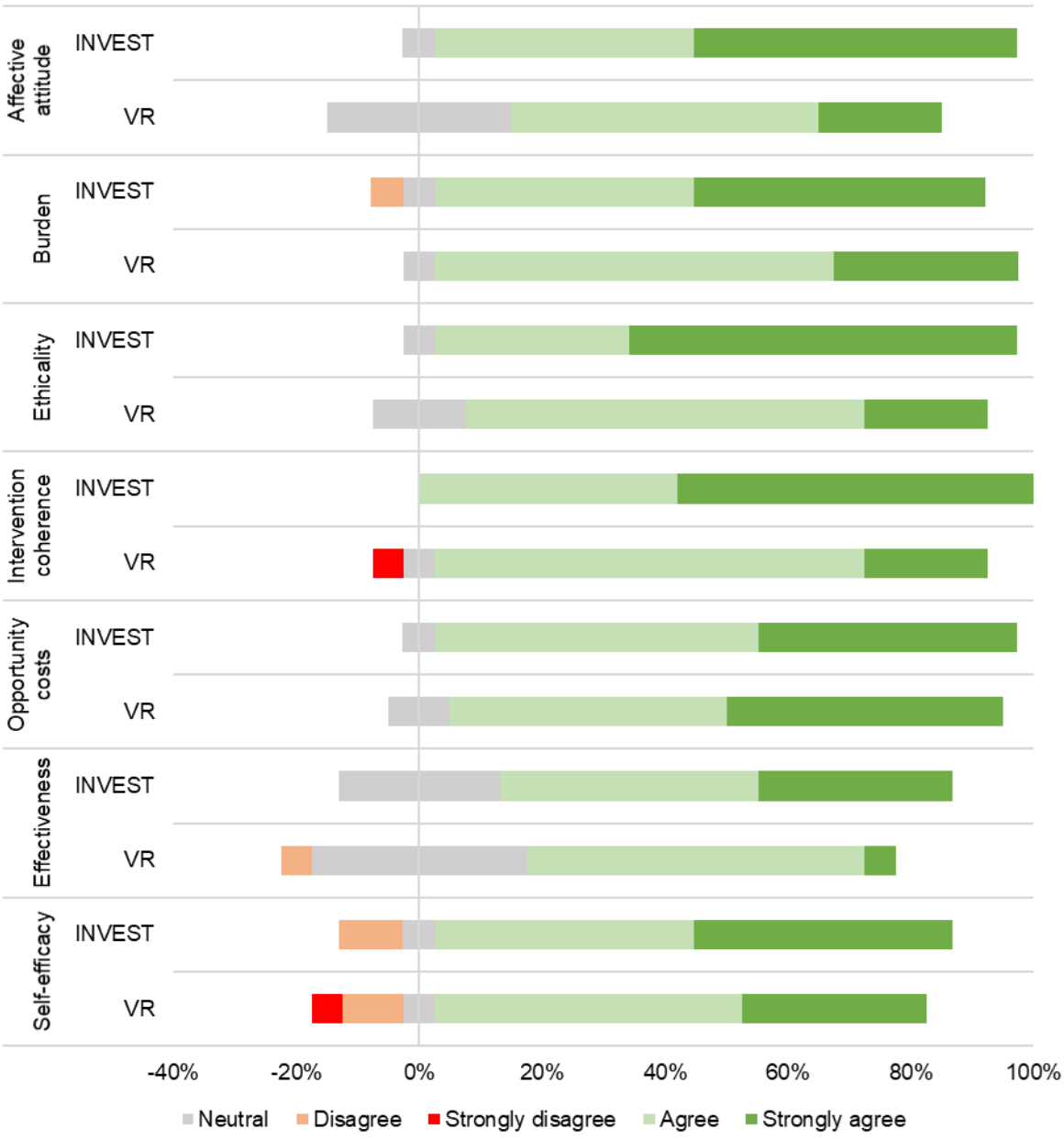

Figure 12. Likert scale acceptability data according to group allocation
Note: INVEST = integrated intervention. VRT = gold standard vestibular rehabilitation

Outcomes

Twelve participants (60%; 95%CI 36% to 81%) in the intervention compared to seven (35%; 95%CI 15% to 59%) in the control group achieved a reliable improvement according to the dizziness handicap (>18 point reduction). Nine participants (45%; 95%CI 23% to 68%) in the intervention had a 'reliable recovery' as defined by a DHI score below 30, compared to one (5%; 95%CI <1% to 25%) in the control group.

Table 17 provides prescores and postcores for the self-report questionnaires and estimates of treatment effect at T1 adjusted for baseline levels, dizziness handicap, age,

and sex. Figure 13 shows a forest plot with confidence intervals to visualise the estimates of the treatment effects and their uncertainty. On average, all outcomes improved from baseline for both groups. Between-group differences at T1, adjusting for baseline level, typically demonstrated small to moderate effects in favour of INVEST for all dizziness and qualtiy of life related outcomes. In terms of putative mechanisms, reductions in negative illness perceptions showed the largest effect (SMDg 0.77) with moderate effects on distress and almost all symptom interpretation variables suggested greater reductions in catastrophising, beliefs that symptoms cause damage, and embarresment and fear avoidance. INVEST did not appear to have greater benefit for all-or-nothing behaviour (SMDg 0.04).

At baseline (T0), 16 participants (80%; 95%CI 56% to 94%) in the intervention group and 17 participants (85%; 95% 62% to 97%) in the control group were identified to have a abnormal balance scores. At follow-up only one participant (5%; 95%CI <1% to 25%) in the intervention group and five participants (25%; 95%CI 9% to 49%) in the control group were identified to still have abnormal balance. Thirty-three participants completed the mini-Bestest at baseline (intervention Mean 23.1, SD 3.57; control Mean 24, SD 3.56) and 25 participants completed it at T1 (intervention Mean 27.6, SD 1.12; control Mean 26.9, SD 1.85).

Table 17. Means of outcome measures at each assessment and post-randomisation treatment effects

Outcome Measure	INVEST Intervention		VRT Physiotherapy Control		Adjusted Mean Difference[a]		
	Baseline (n=20) Mean (SD)	Follow-up (n=19) Mean (SD)	Baseline (n=20) Mean (SD)	Follow-up (n=20) Mean (SD)	Mean difference (SE)	95% CI for difference	Hedge's *g* (95% CI)
Dizziness handicap							
DHI	63.80 (17.84)	37.16 (23.84)	65.10 (14.76)	48.80 (19.44)	10.04 (6.48)	-3.14 to 23.21	0.45 (-0.12, 1.02)
Visually induced dizziness							
VVAS	54.33 (20.97)	30.41 (24.29)	54.44 (20.47)	38.33 (22.45)	5.45 (5.90)	-6.557 to 17.46	0.23 (-0.26, 0.71)
Dizziness interference							
%TSI	57.00 (29.80)	29.32 (26.08)	65.50 (27.32)	39.70 (27.64)	8.05 (9.12)	-10.50 to 26.60	0.29 (-0.36, 0.95)
Health state							
EQ-5D-5L index value	0.52 (0.25)	0.67 (0.27)	0.50 (0.26)	0.58 (0.25)	-0.06 (0.06)	-0.19 to 0.07	0.23 (-0.22, 0.67)
EQ VAS	47.75 (23.33)	57.79 (24.30)	48.90 (26.58)	50.45 (24.57)	-6.85 (6.71)	-20.50 to 6.80	0.27 (-0.25, 0.80)
Negative dizziness perceptions							
B-IPQ	55.75 (10.78)	32.79 (15.39)	57.40 (7.37)	46.20 (14.27)	11.73 (4.75)	2.07 to 21.39	0.77 (0.16, 1.39)
CBRQ domains							
Fear avoidance	14.45 (4.37)	8.74 (4.59)	15.35 (4.67)	11.55 (4.51)	2.34 (1.21)	-0.13 to 4.81	0.50 (-0.01, 1.01)
Catastrophising	9.30 (3.25)	5.42 (4.25)	10.30 (3.26)	7.60 (3.41)	1.30 (1.11)	-0.955 to 3.56	0.33 (-0.22, 0.88)
Damage beliefs	11.95 (3.32)	7.53 (4.77)	12.15 (3.28)	9.80 (3.59)	2.07 (1.01)	0.14 to 4.13	0.48 (0.02, 0.94)
Embarrassment avoidance	14.65 (5.48)	8.58 (6.70)	14.05 (5.35)	10.80 (4.80)	2.82 (1.60)	-0.44 to 6.07	0.47 (-0.05, 1.00)
Symptom focussing	16.40 (4.86)	10.84 (5.47)	17.80 (4.46)	14.70 (4.79)	2.95 (1.41)	0.08 to 5.81	0.56 (0.04, 1.09)
All-or-nothing behaviour	8.50 (4.40)	6.53 (4.41)	7.85 (4.90)	6.45 (4.17)	0.17 (1.14)	-2.14 to 2.48	0.04 (-0.47, 0.55)
Rest/Avoidance behaviour	14.75 (6.79)	8.16 (5.96)	13.25 (6.63)	10.35 (5.02)	3.08 (1.56)	-0.10 to 6.26	0.55 (0.00, 1.09)
Depression							
PHQ9	10.35 (6.10)	5.37 (5.06)	12.50 (8.07)	9.65 (7.32)	2.62 (1.70)	-0.84 to 6.08	0.41 (-0.11, 0.93)
Anxiety							
GAD7	7.10 (4.95)	4.47 (4.12)	10.40 (6.992)	8.30 (6.58)	2.02 (1.64)	-1.32 to 5.35	0.36 (-0.21, 0.93)
Distress							
PHQ-ADS	17.45 (10.39)	9.84 (8.90)	22.90 (14.47)	17.95 (13.47)	4.52 (3.24)	-2.08 to 11.11	0.39 (-0.16, 0.93)

a. Adjustment for multiple comparisons: Bonferroni.

Notes: VRT = vestibular rehabilitation; DHI = Dizziness Handicap Inventory; VVAS = Visual Vertigo Analogue Scale; %TSI = Percentage Time Symptoms Interfere with normal activities; EQ-5D-5L = European Quality of Life questionnaire (EuroQol); EQ VAS = EuroQol Visual Analogue Scale; B-IPQ = Brief Illness Perceptions Questionnaire; CBRQ = Cognitive Behavioural Responses to Symptoms Questionnaire; PHQ9 = Patient Health Questionnaire 9 item scale; GAD7 = Generalised Anxiety Disorders 7 item scale; PHQ-ADS = Patient Health Questionnaire – Anxiety and Depression Scale

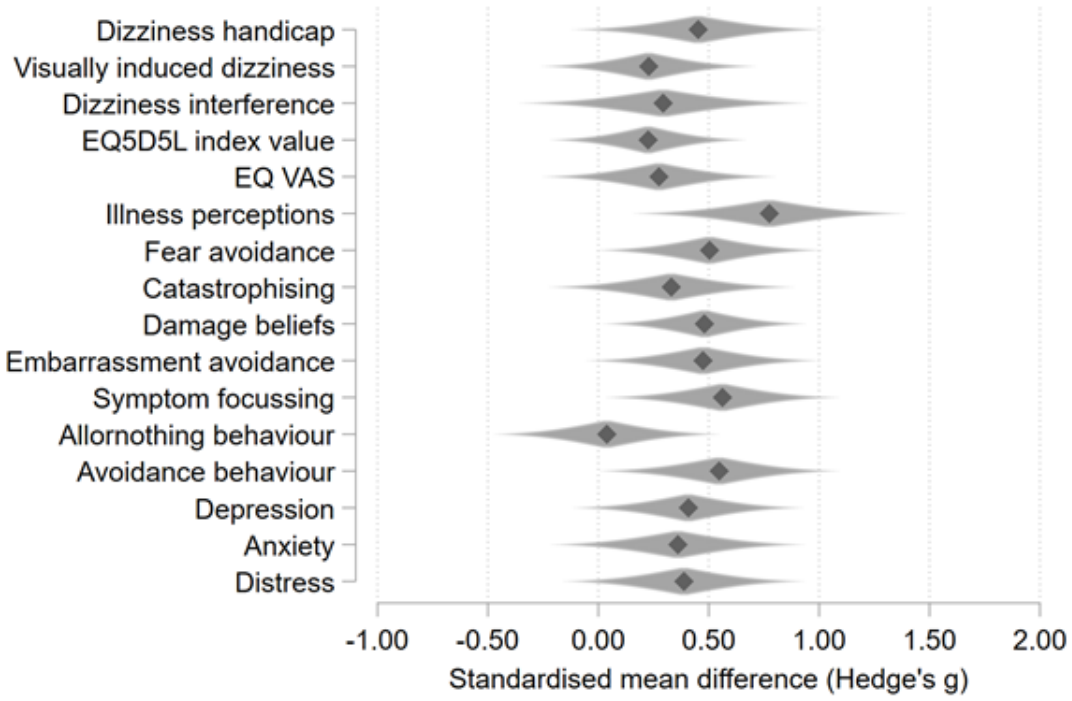

Figure 13. Treatment effect sizes and confidence intervals.
Note: Dizziness interference = %TSI, EQ5DL = EG-5D-5L = European Quality of Life questionnaire (EuroQol); EQ VAS = EuroQol Visual Analogue Scale

Adherence to INVEST

All participants completed the first two sessions. Eighty percent (n=16; 95%CI 56% to 94%) completed all six sessions. One participant missed session six due to other commitments. Two dropped out after the second session and one dropped out after 3 sessions. Seventy-five percent of sessions were conducted in person and 25% remotely. Most sessions adhered to the prescribed duration except for exposure in-vivo (usually session 3) which usually lasted 45 minutes. One participant from each group had a relapse of BPPV (Benign Paroxysmal Positional Vertigo) which was successfully treated with a single canalith repositioning procedure. Table 18 lists other treatments started during the trial.

Adverse events

Table 18 lists adverse events for each group. One participant from each group had exacerbation of migraines which could reasonably be attributed to exercise. Otherwise, participants did not attribute any other adverse incident to intervention related activity.

Table 18. Other treatments started during the trial and any adverse events

	INVEST	VRT
Other treatments started during trial (n)		
Amitriptyline / Nortriptyline	0	2
SSRI/SNRI	1	1

Talking therapies	0	2
Betahistine	0	1
Fertility treatment	1	0
Pelvic adhesiolysis	1	0
Physiotherapy for pain condition	1	1
Herbal supplements	1	0
Adverse events (n)		
BPPV relapse	1	1
Migraine flare	1	1
Mental illness	1	1
Injurious fall, not related to exercise	1	0
Traumatic family event	1	0
Victim of domestic abuse	0	1
Hospital admission, unrelated condition	0	1

Note: VRT = gold-standard vestibular rehabilitation

8.5 Discussion

This study aimed to establish the feasibility and acceptability of a large RCT and potential benefits of a theory-based CBT-informed VRT intervention when compared to current gold-standard VRT for people with PPPD. The study met all the a-priori criteria to progress to a full-scale efficacy trial, including 80% of eligible patients participating (pre-defined criteria >70%), 15% therapy and 2.5% trial drop-out rates (criteria <20%), comparable acceptability ratings to current gold standard VRT, and 80% adherence to sessions (criteria >60%). Fifty-nine percent of patients screened met the selection criteria and the enrolment rate was 80%. This translates to roughly two patients screened for every one participant. Given the high prevalence of PPPD in audio-vestibular, neuro-otology, and VRT clinics there are sufficient patients to run a fully powered RCT. High rates of recruitment and retention point to an INVEST RCT being acceptable

According to the acceptability survey and exploratory treatment effect sizes, the intervention appeared to be both acceptable and beneficial. All treatment effects favoured the INVEST intervention. Treatment effects for dizziness handicap were clinically meaningful and a larger proportion of the intervention group achieved a reliable improvement (60%) vs. the control group (35%). Although these treatment effects cannot be taken as evidence for efficacy, they compare favourably to similar published studies (Andersson et al., 2006; Johansson et al., 2001; Kristiansen et al., 2019; Kuwabara et al., 2020; Schmid et al., 2018). However, given the small sample size uncertainty in these estimates was considerable. The findings still provide a strong signal for efficacy that supports the justification for a full-scale efficacy study. Participants in this current study had a high level of dizziness handicap and a median illness duration of 2 years, which is usually associated with a poor prognosis (Whitney et al., 2020), indicating that these may be important factors to consider as treatment effect modifiers in a full scale trial. Since treatment effects were not universal, we believe there is further scope for the intervention to be improved. Suggestions for some of the improvements will be presented in the detailed qualitative analysis to follow.

Between-group comparison for putative process variables suggests that INVEST is changing the proposed mechanisms of action as intended. This is particularly true for negative beliefs about dizziness and the way in which patients attend to and appraise dizziness as threatening or embarrassing. Reductions in avoidance and resting in response to symptoms was also greater in INVEST. It was surprising that all-or-nothing behaviour did not show a larger treatment effect, since our previous prospective data found this to be a strong predictor and was a predominant feature of INVEST (Herdman et al., 2020a, 2020b). Both groups improved so this may also reflect similarities between the interventions in terms of pacing and graded exercise.

The gait and balance outcome measures were sometimes difficult to execute when completed over video. The Mini-BESTest was suboptimal since it is only validated for face-to-face evaluation. Further, many people with PPPD exhibit features of 'functional gait disorder' (Schniepp et al., 2014). As discussed by Nicholson et al. (2019), the unique clinical aspects of functional disorders means that the usual prioritization of 'objective' or 'subjective' measures may not be appropriate when it comes to measuring balance and gait. For example, due to temporal variability in balance performance, objective snapshot tests such as gait speed may not accurately reflect the general state of the disorder. Likewise, since attention, and therefore clinical examination, can modify gait performance in people with PPPD, clinical assessment may not reflect actual performance outside of this context. Objective measures such as posturography have shown merit in PPPD although again this requires face-to-face evaluation, and the cost is a significant barrier. The advent of wearable motion sensors may be a useful compromise and other clinical tests of dynamic gait performance may be more practical. INVEST did appear to simultaneously improve postural control, and a dichotomised outcome has been adopted in other studies (Schmid et al., 2018), but the lack of blinding and validity is a limitation. Improvements in balance observed in the INVEST group could be because the balance exercises were focussed on allocating attention away from consciously controlling balance and fear driven adaptations to balance control (Wuehr et al., 2017), so a measure that could reliably evaluate this would be preferable.

There were no reported serious adverse incidents attributed to the intervention. The risk profile appears similar to standard vestibular rehabilitation. There was a single mental health-related adverse event in both groups, which both patients attributed to external factors rather than to trial interventions. There were no adverse reactions to any behavioural experiment. Other social external traumatic events occurred, which may reflect the presence of social risk factors associated with persistent functional symptoms. Interestingly one participant from each group also had a reoccurrence of BPPV. This provides another benefit of such an intervention being delivered by a physiotherapist or multidisciplinary team, because such conditions can be easily identified and treated quickly, minimising the impact of symptom relapse.

Limitations in our study must be noted. This was a single site RCT, and the first author, who led the INVEST development, was the physiotherapist delivering it in this trial. To try and counteract this, the person delivering the standard VRT arm was also a senior physiotherapist specialising in VRT. A full-scale multi-centre efficacy trial will need to consider the level of training and supervision required for a range of physiotherapists to deliver it successfully. We did not use a standardised diagnostic schedule to ascertain clinically significant psychiatric comorbidity as a basis for study exclusion. This may have led to inclusion of inappropriate patients, particularly people with post-traumatic stress disorder who require specialist CBT programmes. For practical purposes during the pandemic, we allowed flexibility in the mode of delivery between face to face and virtual appointments. Whilst this likely reflects the way services will continue to operate, there is a lack of evidence to say if this affects outcomes and some participants had a strong perception that face-to -face was better. Therefore, future studies may need control for delivery mode. Our control group represented current gold VRT, although we suspect there may have also been some treatment contamination as both therapists worked in the same department. Future trials will need to consider ways to reduce such contamination, such as cluster randomisation, or spatially separating trial arms. As with all behavioural trials, participants and therapists could not be blinded to treatment group, which may have introduced bias. However, the trial information provided made it clear both were treatments for PPPD with no expectation that one was better than the other. Likewise, it is difficult to tightly control the therapy being delivered when the treatment requires a tailored approach. Using the patient manual was one such approach, although digitalising aspects of the intervention remains an option in the future. Sessions were also audiotaped for supervision and fidelity purposes. More work is needed to ensure fidelity of the standard care arm in a larger trial. Most outcomes were subjective patient-reported outcomes completed online, which may be influenced by many factors, including lack of blinding. However, we argue that it is impractical and illogical to construct a placebo therapy which would contain the same characteristics as the treatment for which it serves as a control. Instead, as was a strength of this study, comparison to a current gold standard therapy should be used when evaluating treatment efficacy and that the only person that needs to be blinded is the statistician.

8.6 Conclusions

Preliminary trial findings support the acceptability and feasibility of INVEST, a CBT-informed VRT intervention aimed at dizziness for people with PPPD. Estimates support medium treatment effects and potential benefits compared to gold standard VRT in a small group of patients that have high levels of dizziness related disability and a poor prognosis with the current available treatment. Findings strongly support the need for a multicentre randomised trial of the INVEST intervention.

8.7 Acknowledgements

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Additional information not contained in manuscript

The recommended target total sample size for an efficacy trial is 352 (176 per group) based on this providing 90% power to detect a difference of at least 6 points on the DHI, at the 5% significance level. This calculation assumes that the ANCOVA approach will be used where the baseline level of the outcome is controlled for as part of the analysis (r01=.4). A difference of 6 points on the DHI was chosen as this agrees with estimates of the minimum clinically important difference (MCID) using the standard error of the measurement and the .3 of a standard deviation rule based on the feasibility study where the SD was 19.1 and reliability was .9. We recommend that this sample size is inflated to account for anticipated loss to follow up.

Chapter 9

Qualitative findings

9.1 Introduction

As in other areas of research, mixed method designs are advocated when designing and testing complex interventions because they provide a better understanding of research issues than either qualitative or quantitative approaches alone (Borglin, 2015). In such designs, qualitative methods are used to explore and obtain insight and in-depth understanding of the topic of interest (Patton, 2002).

Embedding qualitative work within the feasibility trial reflects the Medical Research Council (MRC) process evaluation guidelines 'evaluation' and 'feasibility' phase. The MRC specifically discourages using a purely quantitative approach, since it is rarely adequate to answer questions beyond effectiveness (Skivington et al., 2021). According to the MRC process model, it is important to understand how an intervention induces change, as well as details on the most important enablers and constraints on its delivery. This section also refers to the 'Refining' domains of intervention development outlined by O'Cathain et al. (2019), where qualitative research with those receiving the intervention is recommended as part of testing for feasibility and acceptability. This can then inform the other phases and domains, since these process models are iterative, such that evaluation can lead to new insights into improving theories about how to intervene and you are encouraged to make changes to the intervention if possible.

We therefore embedded a qualitative study into the INVEST feasibility study. The objectives were to understand, from the patient perspective, the 1) levels of acceptability, 2) potential reasons for success or failure of the intervention, and 3) to provide additional insight into any areas of INVEST needing adaptation.

9.2 Methods

Recruitment

As part of the initial consent procedure, participants were asked if they would also consent to being interviewed at follow-up by an independent researcher. Participants who were randomised to receive INVEST and had previously consented to interview were therefore approached by an independent researcher (i.e., a researcher not involved in developing or delivering INVEST and not known to the participants) after they had completed follow-up questionnaires. Recruitment was purposive, meaning that participants were chosen to achieve maximum diversity of the population being

studied but not necessarily a statistically representative sample (Patton, 2002). The sampling criteria were designed to ensure representation of different ages, sex/gender, ethnicity, therapy outcome (and acceptability), language, and education. Participants could represent more than one of these criteria, and we planned to recruit a minimum of 10, and more if the participant was likely to hold a different viewpoint or represent a unique demographic.

Interviews

The interviews were semi-structured and conducted one to one over the phone with the independent researcher. The researcher was another physiotherapist who was a specialist in vestibular and neurological rehabilitation. She received training from the Health Psychology Section at King's College London on how to conduct qualitative interviews, which included role play and feedback. Feedback on the first two recordings was also provided to check fidelity against the topic guide and discussion with myself throughout to check against potential themes and any specific questions for that individual (e.g., their views as an under-represented group, such as male sex).

Topic guide

The topic guide (see Appendix H) was designed to address the components of the Theoretical Framework of Acceptability (TFA) (Sekhon et al., 2017), as described in previous chapters. We used questions recommended by the TFA authors and adapted for people with dizziness. The TFA defines the domain 'ethicality' as 'the extent to which the intervention has good fit with an individual's value system' (Sekhon et al., 2017, p. 8). This particular item was difficult to interpret with regards to INVEST which we did not anticipate would result in such issues. We therefore expanded this domain to enquire as to whether the intervention was sensitive to cultural and individual differences, and the acceptability of the intervention being delivered by a physiotherapist as compared with a psychologist.

Since we also wanted to explore reasons for change, we used open ended questions to explore components of the intervention that they considered important and changes in thoughts, feelings, and behaviours. This part drew from the Common-Sense Model of Self-Regulation (CSM) (Leventhal et al., 2003), which focuses on beliefs about dizziness and coping procedures.

Analysis

All interviews were transcribed verbatim by an external company. Since we wanted to assess the applicability of the TFA and CSM to this intervention, a framework analysis approach was taken which permitted the use of an existing framework of codes (the TFA and CSM) and the inductive identification of additional codes (Ritchie et al., 1994). The data presented represents the initial analysis of transcripts performed by one researcher (myself) against a-priori codes based on the seven constructs outlined in the

TFA as suggested by the TFA authors (Sekhon et al., 2017), and constructs outlined in the CSM. I followed the stages as outlined by Gale et al. (2013). After first familiarising myself with the data (transcripts), I then went through each transcript line-by-line and applied a code (a label) describing what I had interpreted in the passage as important. Some lines therefore had more than one type of 'code'. Some codes were pre-defined, consistent with the TFA and CSM constructs, whilst other 'open' codes referred to anything that might be relevant (e.g., outcomes or experiences, emotions, beliefs etc). These codes were then reviewed, and any 'open' codes were grouped together into relevant categories to form a working analytical framework. The transcripts were then indexed again using these categories and codes. Using N-VIVO 12 Pro (QSR International Pty Ltd, 2020) allowed data to be easily compared within and between cases, and illustrative quotations to be compiled for each category from each transcript. Data were compared back against initial notes made by the interviewer and myself during the early familiarisation stage to make sure that data had retained the original meanings.

9.3 Results

Participant characteristics

Eleven participants consented and completed the interview, who represented a diverse sample of people who had completed the INVEST arm (see Table 19). Participants ranged from experiencing a complete resolution of dizziness, to no change. Unfortunately attempts to contact participants who had dropped out of the intervention were unsuccessful despite multiple efforts.

Table 19. Participant characteristics for qualitative interviews

Variable	Details
Female (*n*, %)	8 (73%)
Age (mean, *SD*, range [years])	39 (14; 20-68)
Ethnicity (*n*, %)	
White, British	5 (45%)
White, other	2 (18%)
Black, African, Caribbean, or Black British	3 (27%)
Asian or Asian British	1 (9%)
English not first language (n, %)	3 (27%)
Educational attainment (n, %)	
No formal education	2 (18%)
Outcome on handicap inventory	
Improved	8 (73%)
Not improved	3 (27%)

Themes associated with the TFA

Affective Attitude

Participants felt universally positive about the intervention. They felt this was something that should be rolled out and made available to other patients.

'I would highly recommend it to anyone that's suffering with these kind of symptoms like I was, it has changed my life.' (Interview 5)

'I think that's a very good method. I think it's something that should be made more like mainstream.' (Interview 11)

Participants felt positive that they were listened to, and their experiences validated.

'There's no comparison to the point I was to where I am now. And I said, no, just the confirmation that is partly real…, (chuckles) I mean it's a physical thing that happened to you and it's real.' (Interview 10)

Burden

The time and energy needed to engage in such a therapy programme was frequently raised as an important, yet ultimately worthwhile, barrier.

'I put lots of effort in doing this. And I don't know whether everybody would do it, to be honest. But for me, if you want to get over the problem, then you have to work on it.' (Interview 1)

'I thought it was quite a lot of effort because it did require to do quite a lot of things on top of like work and everything like that. It was quite a lot. But yeah, no, it was okay.' (Interview 4)

Perceived Effectiveness

Participants felt it had been effective both at reducing symptoms and increasing function and participation.

'I feel so much better. I feel more like me. My husband said that he has me back.' (Interview 9)

'I try many, many times in these 10 years to start jogging again and I never succeeded. And since I've been with this physiotherapy … now I'm jogging between two to three times a week …it's a massive, massive achievement.' (Interview 1)

All three participants interviewed who did not improve on their handicap scores still felt INVEST helped in other ways, such as gaining more control, understanding their condition better, and allowing them to participate in activities again.

'I thought it was pretty effective... it's sort of given me a much better toolkit for dealing with dizziness.' (Interview 8)

'I think actually that was probably quite useful, that's probably one of the best parts in that it's only recently obvious that I've discovered that there might be more kind of how I react to things in the emotional manner..., so I found that quite useful.' (Interview 3)

'Yeah, like it made me feel more comfortable in some situations, definitely. But I would say my symptoms are still very bad even when I'm kind of exposing myself in the situations... So yeah, I think it's definitely helped me to stay calm when my symptoms come.' (Interview 4)

Two of the people who showed no improvement on their handicap scores identified other primary somatic complaints (complex chronic pain/fibromyalgia and chronic fatigue syndrome) as the main barrier to further progress.

'it's just unfortunate that I've got three painful things... I started to deteriorate quite rapidly, and I think my kind of conclusion was that a lot of it is related to that pain. (Interview 3)

'...one of the things that I had was dizziness and fatigue...the dizziness is more manageable now...fatigue is still ongoing, so I haven't managed to sort of get over that... I always find that I sometimes crash' (Interview 8)

The third participant identified ongoing health anxiety in the context of further medical investigations.

'I don't know, because I'm still kind of sceptical about my diagnosis. I feel like that's probably stumping my progress...why did [neurologist] want me to have an MRI if I've already got a diagnosis...maybe I've got something else. It's not PPPD...The ongoing investigations are causing me to have worry about what potentially could be wrong with me.' (Interview 4)

Ethicality

Nobody raised any ethical, cultural, or moral concerns. Seeing a physiotherapist was preferential over a psychologist. This was partly due to the associated stigma, but also that the participants put a lot of value on the need for the therapist to have knowledge about the diagnosis and balance system.

'So, if I had been sent to a psychologist, I would have been cross. Because I would have said to myself, they think I'm mad, they think I'm making this up' (Interview 10)

'… there was a lot of different exercises and movements that I had to do. And I don't think I would have same knowledge from someone that is just working on the brain side of things.' (Interview 5)

Intervention coherence

All participants were initially motivated to participate due to the potential to improve their symptoms, with some expressing desperation after suffering for many years.

'To be honest because I've had this problem for over 10 years. And to get rid of it, I was prepared to try absolutely anything.' (Interview 1)

Some participants were pessimistic about the potential for physiotherapy to help their condition initially, but this did not influence refusal to participate or engagement with treatment.

'I really thought, physiotherapy is not going to do anything for this...I expect something like you know, chemicals like a pill or injection. And I was a bit dubious about something not medical... Then when I start, the first thing I can say is good because I have someone now I can talk to, I can express my feelings…' (Interview 10)

'I'll be honest I thought I was a bit like, I thought it'd be a bit needy like embarrassing, but he explained like it's normal all these things I'm experiencing.' (Interview 11)

The intervention was seen as universally coherent. Participants were able to see the benefits of using an integrated approach and the need to address psychological factors together with physical rehabilitation.

'I think it did because sometimes when you do just physiotherapy with the physiotherapist, they don't understand what you go through mentally as well, and they don't sometimes understand, you know, how difficult it is for you to perform certain activities. And I can see now that I have some mental barrier's that I need to break.' (Interview 1)

'I guess so as soon as kind of certain things have pointed out to me like for example, I was concentrating too much on not falling over… it kind of clicked instantly with some of the things which did make a difference.' (Interview 5)

'I didn't realise quite so much like how much psychological stuff would be in there as well, kind of making me realise how much my brain had to really learn things which was interesting.' (Interview 6)

'And the physiotherapy is not just the physical exercises as I say, it's also cutting down the walls you erected yourself.' (Interview 10)

Opportunity costs

Flexibility over appointment times and method of delivery was seen as an important contributor to improve the opportunity costs.

'…having a very demanding job, it's difficult to take that time off, so that's the only thing I would say. So, you have to try and find a time in the day, normally for me it was in the evening to try and get the session in.' (Interview 3)

'…he was really considerate of if I had other appointments or other things going on, then it wouldn't be the following week, we'd kind of build it in so there wasn't too much pressure…' (Interview 6)

Almost all participants had a very strong preference for face-to-face appointments. This was either down to their own experiences, or personal preference.

'I mean I know for a fact if I had just gone over Zoom, I probably wouldn't have come out the other end with quite the knowledge and the coping mechanisms…I felt like, as for the virtual ones, I kind of just felt the same. Whereas each time I went away from the face-to-face, I felt like we achieved the next step.' (Interview 5)

'I think it was actually more beneficial to be there with like face-to-face consultation rather than doing over the video. I mean, I'm more a person, you know, I'm a face-to-face person.' (Interview 1)

One participant who had chosen for all sessions to be done online also expressed a preference for face-to-face in the future.

'I really prefer to do it face-to-face …trying to do the exercises while you're on the screen was probably harder that if I've been able to do it face-to-face but it didn't hamper the experience too much…' (Interview 6)

Some also reflected that the act of commuting to the appointment was partly therapeutic, and that being face-to-face allowed them to push themselves more.

'… that was another way of me pushing myself out of my comfort zone to get on the underground and go on the escalators on my own…I needed to get myself there, and just go and deal with the situation…Because there was a point where I had to kind of stand in an open space and stand there for like five minutes, which felt like the longest time of my life. If I was on the phone to someone, I wouldn't have probably done that.' (Interview 7)

Self-efficacy

Some participants drew on support of close family members and friends to complete therapeutic activities and attend appointments.

> *'Yeah, I was lucky though because I had somebody, I had my mum that dropped me off.' (Interview 11)*

> *'It was really good for me because I- I...didn't like train stations with the platforms...- and I did it slowly and my husband took me in the car first and then- and then I ended up going on my own and that gave me a lot of confidence.' (Interview 9)*

For some people the restrictions during the COVID-19 pandemic made therapeutic activities difficult.

> *'The only challenging bit was when doing exercises became...doing bigger exercises like the things that I would find challenging, some of them were harder to do because obviously COVID, like going on escalators and lifts, and working from home isn't something that I would encounter a lift or an as escalator, so I'm kind of having to actively seek that out, rather than- usually that would be part of my day-to-day life.' (Interview 6)*

Behavioural exposure was seen as the most challenging task to complete independently. Working collaboratively with the therapist to set realistic goals was seen as important.

> *'So, I think when you're with a therapist...you think, "Yeah, yeah, I can probably..." When you're away from that situation...you might feel... I'm not going to necessarily be able to do that. So, I think it's knowing within yourself and being honest throughout the whole process in terms of what is realistic and what's not.' (Interview 7)*

Patients consistently identified the therapeutic alliance as a strong facilitator to allow them to engage in fearful activities again.

> *'I remember him saying...look at yourself, look at where you are. And you need someone to tell you that...I would not have been able to do it, just by doing, doing the exercises, you know, they send you a video and you do the exercise and all like... It would not have been, not at all.' (Interview 10)*

> *'I think the physiotherapist is the most important part of it...I thought I had- I had to because he had put so much time into me, and I felt- I felt I had to do it. And it made me much happier...' (Interview 9)*

Mechanisms of change

To support the quantitative process variables, the interview schedule also asked about illness beliefs and what mechanisms of change were important to participants.

Illness coherence

Following the intervention, participants were able to make better sense of their condition and the relevant perpetuating factors.

'I guess changing my mindset on it…So, it was kind of definitely being more aware of what it is and how to overcome the sensations…And obviously, when you worry about many things and think about anything constantly, things get worse in your body.' (Interview 5)

'One thing that kind of sticks out is if you don't move your head at all then your brain's reacting to - it's hypersensitive to that and so by not doing it, I was kind of making it worse and now I'm kind of like okay, a little bit of dizziness is fine but the more I kind of carry on doing things and the more I, I don't know, walk and talk and move like a normal human then the less that I'm going to feel dizzy, so I think that kind of massively helped.' (Interview 6)

Consequences:

Participants had both less fearful anticipated consequences and experienced less concrete physical, psychological and social consequences from dizziness.

'I used to think, 'Oh, I might feel unwell. I might be unwell for a long time.'…And so, that's what I've learnt, that it is not the end of the world. I need to carry on because nothing bad is going to happen.' (Interview 1)

'Did it actually harm me in that situation? If not, then, you know, keep moving forward with it. … And I have noticed that I've changed a lot that I'm not panicking as much as I were, or I can overcome my symptoms a lot more quickly than I probably could have in the past.' (Interview 7)

Emotions

Participants experienced less negative emotional representations about dizziness. Specifically, they reported less fear, depression, and anxiety.

'Before I was feeling scared but for now I don't feel scared, it's gone.' (Interview 2)

'… [I was] probably suffering quite a bit with depression and was just crying, not really know what to do with myself. I think speaking to someone, a professional that is medically trained is much better than just talking to family that don't really understand. They just think I'm being silly and it's all in my

head. So, I feel like I've definitely moved forward a lot, and I'm much happier as well. (Interview 7)

'Before it was kind of anxiety, worrying, depression, yeah. Am I ever going to get better? Those kind of, yeah, downward spiral of thoughts...' (Interview 8)

Timeline:

Participants experienced both a reduction in the objective duration of symptoms and perceived temporal features of the health threat.

'If you're this dizzy, then take a moment, it will pass, you know it will pass. And it really helped because you don't restraint or constrict on yourself so much. And it's been unbelievable.' (Interview 10)

'Doing this treatment has made me kind of realise, okay, it's not necessarily a permanent part of your life and there are ways to kind of manage it.' (Interview 6)

Identity:

Participants did not explicitly state a change in symptoms they attributed to the disorder, but rather suggested they had developed a more adaptive *interpretation* of symptoms.

'I feel like just trying my best to, just sort of allow my body to adapt to it. That's one of the parts that I'm, your brain has to adapt to it so sometimes being dingey is what you need to feel.' (Interview 11)

Cure/control

Participants frequently made statements about regaining a sense of control over their symptoms, and a shift in beliefs from having an unmanageable illness to a condition that they could recover from.

'I know that there's things out there and I understand that, like it's not all in my head because there was a point in my life where I thought it was all in my head and I know that it's not, and that it can get better.' (Interview 11)

Coping procedures

Following INVEST, participants were able to change their appraisal and enactment of coping procedures. They reported less avoidance and/or all-or-nothing behaviours, in addition to not using safety behaviours.

'I'm doing a lot more like I'm back in actual gym now and I'm not afraid that I'm going to fall over like social settings as well. I feel like I'm not avoiding going out or being in a social environment thinking that people are looking at me because I was swaying left and right. (Interview 5)

'I found it hard to speak to people because it didn't, (coughs) I was falling over and erm, and he took me to [supermarket]and made me go and speak to people. Erm, and then I could see that people weren't really bothered.' (Interview 9)

Manual

The manual was seen as a helpful and important contribution, allowing people to reflect on their progress, and helping them to lean about their condition.

'I think even just having that manual has just been really helpful for me. I mean it is my bible really, to be honest. And even when I started reading it, there was a lot of stuff I didn't know about myself and how the body kind of works, and what happens when you're in certain situations.' (Interview 7)

'Reading through that as well as talking to him like made me realise why the symptoms have carried on so long and how the brain kind of reacts at stuff, so it kind of explained quite a lot actually.' (Interview 6)

One participant initially found the size of the manual quite daunting and having a learning disorder made it more challenging.

'It was quite a big work but it's a little bit overwhelming when I was handed it. I felt, oh gosh, I've got homework to do as well. (Laughter)... and I'm also dyslexic so if I read something, I kind of have to read it five times in order to get all of the information out of it' (Interview 5)

Recommendations

Although the participants engaged well with INVEST, some had suggestions on how to optimise it. For those participants with less education, they wanted other options to accompany the manual such as videos.

'Maybe if they can make like a virtual version of it...even audio book...that would be a good idea... Yeah, it would have been easier to sort of remember the stuff.' (Interview 11)

Three participants felt that six sessions wasn't enough, and two felt that the timing of the sessions (30 minutes) felt rushed.

'But for me, I was thinking like it's too quickly because I've done just, if I've not forgot, like six sessions... because you've been for the dizziness like eight years, seven years...like to go shopping I'm [still] scared.' (Interview 2)

'Half an hour went very quickly...Sometimes, I thought I could have probably benefitted from a longer session rather than just half an hour.' (Interview 7)

9.4 Discussion

The key finding of this study was that INVEST was highly acceptable, providing further support to the quantitative measure of acceptability. Participants felt positive about the intervention having taken part. It was seen to be both effective in reducing dizziness and improving quality of life, something that participants found useful for themselves, and the wider population of people with dizziness. The participants understood the intervention and how it works and appreciated the need to address mental and physical aspects of rehabilitation. There was consensus that INVEST was effortful, but that this effort was a worthwhile investment. INVEST was also challenging, and the therapeutic alliance was critical in helpful participants perform the behaviour(s) required of them.

The qualitative data also supported the evidence that INVEST changed the proposed mechanisms of action as intended. Participants expressed a more adaptive mindset about dizziness, paying less attention to it, and appraising dizziness as less threatening or embarrassing. Participants had less avoidance behaviours and were able to appraise their coping repertoire differently, viewing exposure as critical. The previous quantitative methods did not show a change in all-or-nothing behaviour whereas there was some limited support for the benefit of pacing activities in the interviews, even if participants were more likely to cite approach related behaviours as critical.

The high intensity of therapy related activities and graded exposure in vivo allowed participants to work towards valued goals and overcome dizziness-related fears. However, the qualitative data suggested this intensity may make it unsuitable for some patients with other somatic conditions. With this in mind it may be worth excluding those for whom complex chronic pain or fatigue is the *dominant* problem. Alternately simultaneously probing for possible co-morbidities may be needed and addressed alongside the balance disorder, since it has been shown that dizziness often appears alongside localised pain (Malmström et al., 2020). However, there is little evidence about if or how pain or fatigue affect recovery. It is also important to collaborate with the multidisciplinary team, since in at least one case a suggestion for further medical investigation brought about a break in the therapeutic alliance and further health related anxiety.

Participants provided valuable suggestions to improve the intervention, such as providing video and audio materials or an alternative manual for people with lower educational attainment. We believe that the manual could also be made shorter, and/or alternative versions could be made for those who need or prefer more plain English.

There was also a preference for more than six sessions, and/or a three-month check-up. Six sessions would be considered brief therapy even for people with acute structural vestibular disorders and it would be reasonable to extend this to eight.

The data also pointed towards the importance of the therapist. Participants felt that it was important for the physiotherapist to have specialist knowledge about the balance system, and that trust in the physiotherapist allowed them to engage in fearful activities again. This has important implications for the training of future providers, and analysis of a future efficacy trial since there may be important 'therapist effects'. This might also be an issue around therapist supervision which is a requirement of delivering psychological interventions but not provided routinely for physiotherapy.

Implications for practice

This study provides insightful implications to improve INVEST and clinical practice. Given that both INVEST and VRT take effort, it may be helpful to be candid with patients about the potential burden of treatment procedures early on to utilise problem solving techniques and address the expectation of patients to be actively involved. INVEST is specific to dizziness, and patients with other primary somatic complaints may benefit from trans-diagnostic approaches better. Physiotherapists should be trained to understand the effect of the therapeutic relationship and ways to enhance it. The patient manual should be adjusted accordingly, and we think it is a good idea to provide multimedia options. Finally, more sessions may be needed in some cases and the timing of sessions should be sufficient to achieve the necessary aims of behavioural experiments.

Strengths & Limitations

A strength of this study was that the interviews were conducted by a separate researcher who was not known to the patient. The patient was encouraged to be open and honest with the express aim to improve the treatment in the future. The participants also represented a diverse yet representative sample of people who had undergone the treatment, in terms of demographic and clinical variables.

The analysis of data was conducted by a single author (myself), who was the person delivering INVEST, which represents a significant source of bias. Using some pre-existing theoretical constructs deductively limits bias to some degree and makes sure that potentially important categories are not missed. However, given more time and resources, a rigorous thematic analysis could provide more trustworthy and insightful findings (Braun & Clarke, 2006). This should also be repeated with at least two researchers coding the transcripts, which we were unable to fulfil at the time of writing. Another limitation is that we were unable to interview participants who had dropped out of the intervention. Therefore, there may be problems with the acceptability of the intervention that we are not aware of.

9.5 Conclusion

All the participants described having had at least some benefit from the intervention, which ranged from greater insight and coping to complete symptom resolution. They

endorsed this approach, albeit finding it effortful and challenging, for people with PPPD. INVEST legitimised their illness experience, providing clarity on the role of psychological factors and ways of coping. The participants identified several components of the intervention that they considered important, and those were generally related to the understanding that they had gained which allowed them to adopt adaptive illness beliefs and approach activities again. They appreciated that it was delivered by a specialist clinician and were able to provide general and specific insights to allow INVEST to be optimised.

Chapter 10

Final Discussion

10.1 Introduction to the chapter

This thesis presented a series of studies beginning with a cross-sectional and prospective study, and systematic review, which collectively informed a cognitive-behavioural model of dizziness, and the development of an integrated cognitive-behavioural therapy informed vestibular rehabilitation treatment (INVEST). A randomised feasibility study was conducted with the primary aim of determining the feasibility of testing the intervention in a randomised controlled trial (RCT). A qualitative study of patients receiving the intervention was embedded into the feasibility study, providing insights into how the intervention worked, and how to optimise it prior to an RCT. Feasibility was demonstrated with high rates of participant recruitment, retention and intervention acceptability. The clinical outcomes were promising, suggesting that an appropriately powered RCT has a reasonable chance of demonstrating clinical and health-economic effectiveness.

This chapter will draw together the research findings with a synthesis of the included studies. The theoretical implications of this thesis will then be discussed in the context of planning for future research and a fully powered randomised controlled effectiveness trial, before appraising the limitations of this research and providing an overall conclusion.

10.2 Summary of the overall purpose of the dissertation and main findings from each of the included articles

The overall aim of the project was to design and evaluate a new cognitive behavioural intervention that could be incorporated with, and had the potential to improve the outcomes of, vestibular rehabilitation for people with persistent dizziness. The project followed the broad MRC guidelines for developing complex interventions, but to optimise the intervention development a taxonomy of approaches were identified and adopted.

To determine the contribution of cognitive, behavioural, and emotional factors to dizziness-related handicap and severity, a cross-sectional study surveyed 185 people with vertigo/dizziness who were on the waiting list to attend a specialist vestibular clinic. This study found the following:

- Psychological distress, as measured by depression and anxiety symptoms was associated with greater dizziness severity and handicap.
- Negative illness beliefs were associated with greater levels of dizziness severity and handicap, which included attributing more somatic symptoms to the condition and having stronger belief that the condition would last a long time, have serious consequences, be untreatable, and more distressing.
- Negative interpretations of symptoms (embarrassment, catastrophising, fear avoidance beliefs, symptom focussing, and belief that dizziness represented damage) were correlated with higher levels of dizziness severity and handicap.
- Greater use of avoidance and all-or-nothing behaviour was associated with greater level of dizziness severity and handicap.
- Psychological vulnerability (reflecting greater levels of dependence, perfectionism, and the need for external sources of approval) was associated with higher handicap and symptoms. Negative beliefs about expressing emotions, however, was not associated with the dizziness outcomes.
- These psychological variables were significantly correlated above and beyond objective vestibular deficits or diagnosis. The final model accounted for 63% of the variance in dizziness handicap and 36% of the variance in dizziness severity. In the fully adjusted model dizziness handicap was associated with age, gender, distress, symptom focusing, embarrassment, avoidance behaviours, and beliefs about negative consequences. Fear avoidance was the only factor in the fully adjusted analysis found to be uniquely correlated with dizziness severity.

One hundred and thirty-five participants responded when we surveyed them again three months after their initial diagnostic consultation. This study found the following:

- There were significant improvements in dizziness handicap, distress, illness perceptions and cognitive-behavioural responses to symptoms after clinical assessment and diagnosis.
- However, some variables showed little change over time. Any improvements tended to be small in magnitude and still reflected largely unhelpful illness beliefs and symptom responses, and heightened levels of distress as measured by anxiety and depression. These findings show that patients can continue to experience heightened levels of handicap, negative cognitive and behavioural responses to symptoms and psychological distress despite usual care.
- All-or-nothing behavioural responses predicted dizziness handicap at follow up, but baseline handicap was the strongest predictor and accounted for much of the variance at follow up. Most participants had experienced dizziness for a long time, which might indicate that somatic experience and illness schema developed earlier in the temporal sequence of the condition is important in the development and maintenance of such negative illness responses.

- Having a longer duration of dizziness was associated with greater dizziness handicap at follow up, but there was no other relationship between handicap and any other demographic factor, diagnosis, or vestibular function test.

Although the systematic review was started first and helped to inform what psychological factors to consider in the prospective study, it was then revised in line with these findings to formulate a cognitive-behavioural 'theoretical' model. This included 89 studies that had explored the relationship between dizziness handicap/severity and modifiable psychological factors. This study found the following:

- Across the literature, moderate to large correlations existed between self-reported dizziness handicap and symptoms, and measures of anxiety, depression, and autonomic symptoms.
- There was emerging evidence for the role of sense of coherence – in its general sense in relation to life roles and values as defined in salutogenesis. There was also evidence for negative illness beliefs, particularly belief that dizziness results in serious consequences, and interpretating symptoms in a fearful or catastrophic manner. Avoidance behaviours were also consistently related to dizziness outcomes, and sleep disturbance may be another important behavioural factor. These factors may contribute to dizziness directly and indirectly through cognitive, behavioural, and emotional responses to the illness, and a complex interplay of them may exist in a single patient.

To design a cognitive behaviourally informed physiotherapy treatment for chronic dizziness (INVEST), we included a diverse range of experts to map these psychological and illness specific determinants to CBT techniques and worked collaboratively with the target population to develop a treatment manual.

To evaluate the feasibility and potential efficacy of the intervention and test multiple methodological components and clinical outcomes simultaneously to inform a fully powered randomised controlled trial, a protocol for a feasibility randomised controlled trial was designed and published, which included a-priori progression criteria for a fully powered trial. This trial improved on previous feasibility trials as it included an active control group which represented current best practice, and the timing and duration of therapy was mapped onto current VRT to improve longer term implementation. We also employed an external randomisation procedure.

A two-armed parallel groups randomised feasibility study recruited people with PPPD from a specialist vestibular clinic at St George's Hospital NHS Foundation Trust. Participants were randomised to receive either the INVEST or a gold standard VRT which served as the active control. This study found the following:

- 80% of eligible patients agreed to participate

- Only one patient dropped out of the trial, and 15% dropped out of therapy
- Acceptability of INVEST was excellent according to quantitative and qualitative data
- 80% of patients adhered to all 6 sessions
- Small to moderate treatment effects in favour of INVEST were found across all measures

The study therefore provided strong support for the feasibility of a full-scale trial. Both arms had high rates of recruitment, retention, and acceptability. There was promising support of the benefits of integrated CBT based VRT compared to VRT alone. The study fulfilled all the a-priori criteria to advance to a full-scale efficacy trial.

A qualitative study was embedded which interviewed eleven participants who had received the intervention. This study supported the main conclusions that INVEST was acceptable and worked as intended, whilst suggesting we may be able to optimise the intervention further prior to a full RCT. Specifically this included:

- Making explicit the effort required to participant in INVEST
- Providing more sessions
- Considering the impact of other somatic complaints
- Simplifying the manual, and providing multi-media options

The qualitative study also made it clear that patients agreed with the need to address mental and physical aspects of rehabilitation, but they would rather see a physiotherapist with knowledge of the balance system. The therapeutic alliance was critical in allowing participants to carry out the activities required of them, and this therefore has implications for the training of future physiotherapists.

10.3 Main points of integrated discussion

Functional dizziness is a common cause of disability and distress, for which there is a lack of evidence to support a limited number of treatment options. A combined CBT-VRT intervention has been the ambition of behavioural neuro-otology for the last 20 years, but this presents the first theory driven manualised intervention, meticulously designed to target the key perpetuators of dizziness related disability, and the first to be compared against current gold standard VRT. In identifying unique factors related to dizziness handicap and severity, the thesis has broadened the view of psychological correlates beyond anxiety and depression, providing new treatment targets and a firmer scientific basis on which to develop a more focussed approach to integrated VRT.

The key components of the intervention were education about dizziness and balance disorders, demonstrating to the patient how their perception of dizziness and stability can be adjusted according to the degree of conscious attention, using vestibular exercises to develop strategies that normalise movement and habituate to triggering

stimuli, direct exposure to fearful activities and movements while helping them prevent unhelpful responses, thought restructuring to develop more adaptive illness beliefs and interpretations, and developing a long-term personalised symptom management plan.

The finding that many participants were not resistant to acknowledging a role for psychological factors in their problem after treatment may be significant in the intervention success and points to the validity of this model. Participants felt the intervention was highly credible. The research identified several factors that may mediate a good treatment outcome and influence levels of dizziness related handicap. These were: (i) changes in illness beliefs; (ii) normalised locomotor control, (iii) changes in threat value and responses to symptoms including avoidance beliefs, symptom focussing and catastrophising, (iv) reduction of distress as measured by anxiety and depression, and (v) extinction of avoidance related behaviours. It is notable that INVEST appeared to improve levels of distress, which is in contrast to other CBT trials where they did not get greater change on these measures, which might be due to targeting the specific illness-related thoughts and behaviours perpetuating fear and anxiety rather than generalised anxiety/panic beliefs. Together they may represent an overall shift away from threat related perceptual mechanisms towards a more flexible and adaptive generative model. However, a large effectiveness trial with embedded process analysis is needed to confirm if these factors are indeed mediating change in dizziness outcomes. The timing, duration, and method of treatment delivery are other elements of the intervention that warrant further exploration.

The intervention cohort improved despite an average symptom duration of two years. Given the evidence that symptom chronicity is associated with a poor prognosis (Dieterich et al., 2016), and that years of chronicity usually imply a higher degree of maladaptation, with severe disability and more engrained illness beliefs, it is possible that if the intervention was delivered earlier in the course of dizziness, it may be even more effective. This is why all healthcare professionals managing dizziness and balance disorders should be aware of the psychophysiological mechanisms summarised in this thesis. It is particularly encouraging that good outcome with treatment can be achieved for people who have a poor prognosis with the current available treatment.

This thesis explored a specific physiotherapy intervention, although physiotherapy is only one of a few different treatment approaches and professional groups that may be effective for people with persistent dizziness. Other potentially effective treatments available include psychological therapy, multidisciplinary rehabilitation, and medication. Given the heterogeneity of patients with persistent dizziness, it follows that a variety of different treatments are necessary to suit the needs and preferences of people with this diagnosis. It is also not clear whether a combination of these treatments may be complimentary. The preference of the patients who participated in this trial was to see a balance disorder specialist so a priority in future work is to identify criteria that predict which patients are most likely to benefit from this intervention, and ways to identify patients who may need supplementary or alternative treatment. For example,

who may benefit from the addition of SSRI medication and methods to identify people who need specific mental health intervention.

The thesis introduced intervention components, such as behavioural exposure in-vivo with response prevention, which have not been used for dizziness before. This is therefore the first study that suggests such modalities may be viable techniques in the management of persistent dizziness. However, intervention components labelled as either 'CBT' or 'VRT' cannot be assumed to be equivalent homogenous entities since they may still vary regarding several factors such as the nuances of communication, mode of delivery, the duration of intervention, and setting to name but a few. It is also not clear whether those components are the most effective methods for targeting those specific treatment targets. Further research into any of those modalities (e.g., the most effective breathing techniques etc), including research into new technologies such as virtual reality and digital technologies, may provide further additive treatment effects and/or efficiency of delivery. For example, the ACTIB trial showed that CBT could be delivered effectively by a web-based self-management tool for people with irritable bowel, helping to standardise the intervention and increase access to CBT for people with LTCs whilst reducing therapist input (Everitt et al., 2019).

Progression to a definitive trial

The findings of this thesis support the progression from the feasibility study to a pragmatic multicentre RCT. Additionally, the findings should inform the design of this trial. A key methodological consideration for a future trial will be to avoid contamination between treatment arms, which will need to consider the possibility of spatially separating trial arms or conducting a cluster randomisation (Magill et al., 2019).

Refining, standardising, and documenting the intervention so that it is both implementable and reproducible in a clinical trial will be a challenging but necessary step. This will involve recruiting and training other physiotherapists to deliver the intervention as planned. In the meantime, the intervention can be implemented in practice and monitored at St George's Hospital.

Given the high satisfaction ratings and positive treatment outcomes, a future trial should aim to reproduce as closely as possible the conditions of the study intervention. Training will need to be standardised, and it is my intention to produce a therapist manual to accompany the patient manual. Treatment fidelity can then be checked against pre-defined criteria as outlined in this manual. An addition that may improve the intervention is adding another two follow up sessions and extending sessions to 45 minutes. This is realistic, since many outpatient neurological/vestibular physiotherapy services in the UK already provide longer sessions and the restrictions on the number of sessions is usually self-imposed.

Another addition may be to refine the treatment manual and provide options, including multi-media, to improve its accessibility. Finally, the method of delivery (face-to-face, virtual, or blended delivery) will need to reflect the desire for maximum effectiveness, standardisation, but also implementation in clinical practice.

A future study should consider secondary mediation analysis to explore potential treatment mechanisms. The measures used in the feasibility study appear appropriate for such an analysis since they were responsive to change. However, a pragmatic tool to measure balance and postural control may need to be considered which doesn't rely on unblinded therapists.

Reproducing the study intervention in a pragmatic multicentre trial will be challenging due to local/regional differences in service provision, waiting lists, and availability and willingness of staff. However, informal discussions with UK physiotherapists and potential collaborators makes me believe that many people would come forward and it would be possible.

10.4 Disciplinary implications

Chronic dizziness has been an area in which medicine has struggled to make sense of a person's suffering, where patients feel neglected and abandoned. The treatment of all illness starts with a conceptualisation of the symptoms. Our ability to peer into the balance system, to examine the vestibular organ and measure and make sense of vestibular system function have persuaded us that dizziness is nothing more and nothing less than a bit of the vestibular apparatus gone awry.

My own journey to this point involved a steadily increasing understanding that vestibular medicine often does not serve patients with persistent dizziness well, particularly where a person's suffering isn't accompanied by any abnormal test results. What drew me to a career in vestibular rehabilitation has always been the interface of psychology, neurology, and neuro-otology, but what frequently transpires is confusion, uncertainty, and fragmentation between these areas.

There has been tremendous advances in the understanding and diagnosis of vestibular disorders over the last few decades (Welgampola et al., 2017), and appreciation of the many patient groups who might benefit from VRT (Dunlap et al., 2019). However, this belies an underlying reality that VRT has in fact felt in a state of stagnation. Rehabilitation technologies are being proposed but without any theoretical underpinning of how they might work, just replicating current techniques. The pioneers of VRT brought about innovative and effective exercises for vestibular dysfunction based on the proposed mechanisms thought to contribute to recovery (Herdman, 1998). VRT research is now more concerned with answering questions about what works for whom, rather than how. If we want better treatments for

persistent dizziness, we need to identify the processes of change and the modifiable factors known to have an impact on outcomes.

This thesis supports evidence that with our current measurements, there is often no clear association between the severity of the original vestibular dysfunction and the subsequent long-term disability. This does not mean that the illness should not be taken seriously or imply that the patient's symptoms are not real or dismiss the physical. Elucidating the factors associated with dizziness is clinically pertinent, especially considering its high prevalence, as well as the considerable social, occupational, and cognitive impairment imparted.

For many people, 'no medical cause' for their vestibular symptoms can be determined. But when services broaden their concept of vestibular and balance disorders to include peripheral, central, and behavioural factors, it is possible to improve diagnostic rates and accuracy (Staab, 2013). In fact, PPPD has become the most common disorder identified in specialist dizziness clinics (Strupp et al., 2020). Successful integrated care opens-up an expanded list of therapeutic strategies and yields insights into mechanisms of balance function that might otherwise be overlooked.

Without addressing these issues, offering integrated rehabilitation and physiotherapy, and addressing the fear and despair that patients experience when facing such challenging problems, we can make the patient's situation worse. All vestibular illnesses have a perceptual and psychosocial component, which is to say that our experience of symptoms can be very subjective and influenced by a variety of non-medical factors. This thesis identified how the impact of dizziness is influenced by our beliefs, such as how serious we think the cause and consequences might be, our mood, and the ways we try to cope and manage dizziness. By addressing all these factors, psychological approaches can not only address mental health consequences but can reduce and even cure symptoms.

When assessing a patient with persistent vestibular symptoms, a physiotherapist needs to explore all these other factors that could be important in ameliorating the symptoms. This includes identifying any current mood disorders, such as depression, as well as anxiety disorders, which may be maintaining or exacerbating the problem. Focusing on the symptoms is an understandable but unhelpful means of perpetuating problems. This is often driven by fear of what the symptoms may represent, so also understanding the person's views on the illness, how serious they believe it is, and whether they believe it to be controllable or not are all important to elicit and address.

The clinical trial was designed to assess feasibility, so we cannot interpret clinical outcomes as controlled evidence for effectiveness. However, given the lack of quality clinical trials for PPPD, the feasibility study outcome data make a significant contribution to the evidence base that supports the use of specialist physiotherapy for PPPD.

In the future physiotherapists will need to be willing to employ an expanded list of therapeutic strategies alongside traditional VRT to include cognitive and behavioural interventions. The profession will need to adapt to meet the needs of the patients, liberated from a narrow view of health and illness, and unconstrained by services and professional boundaries that seek to separate the mind and the body. We do, however, also need clearer pathways where further psychology or psychiatry expertise is needed. This involves developing assessments which will help ascertain when onward referral to a psychologist would be indicated either before or after the treatment.

10.5 Limitations not already addressed

Researchers who take a person-based approach (Yardley et al., 2015) to intervention development may view this research too limited by its focus on quantitative methodology in the early stages of development. In their view, including quantitative studies in the synthesis of data and conducting qualitative research in the development phase allows the intervention to be more relevant and engaging. We did consult with PPI and stakeholders who already have in depth understanding and access to users' perspectives, and there has been limited qualitative studies to draw from in comparison to the vast evidence cited in this thesis. We also embedded a qualitative study into the feasibility study, basing views on actual use of the intervention.

Other approaches that undertake specific actions such as the Behaviour Change Wheel (Michie et al., 2011) may be more comprehensive than the methods used here. The related Behaviour Change Technique Taxonomy (BCTT) tool was developed to help interventionists systematically design and describe theory-based health behaviour change interventions. However, we suggest are too prescriptive for developing complex interventions for conditions that target more than just change in behaviour. INVEST uses broader therapeutic strategies than those defined in the behaviour change taxonomy, including the integral components of the therapeutic relationship. Nevertheless, a lack of transparency or clarity about specific intervention components is a valid criticism of interventions in health psychology, so each component of INVEST has been described in detail in this thesis, which includes how the techniques were used in the context of the therapist sessions and the self-management manual.

The specific limitations of the studies are discussed in detail in the relevant chapters. The cognitive-behavioural factors considered in this thesis require further experimental support. Likewise, the systematic review highlighted many methodological shortcomings in the literature to date suggesting that the overall quality of evidence supporting the proposed model is poor, and a more comprehensive model of dizziness handicap that seeks to elucidate the mind-body interaction is still needed. Furthermore, the systematic review was not pre-registered, which was an oversight on my part.

Another limitation is that all studies were set in specialist vestibular departments of tertiary level hospitals, where the patient population seen in this setting is likely to be

biased towards more severe cases of dizziness, and the findings presented may have limited generalisability outside this setting. This is especially relevant in the interpretation of the feasibility study, due to the single recruiting centre.

A limitation that warrants repeating is that I delivered the INVEST intervention, and I have a particular interest in PPPD and a vested interest in this approach. This would not be the case if the intervention is rolled out across the NHS. The training needs of other physiotherapists will need to be considered in the next iteration alongside developing a broader implementation strategy.

10.6 Conclusions from the dissertation overall

This thesis provided evidence that symptom severity and dizziness handicap are not correlated with the results of clinical vestibular tests but corresponded with psychological factors such anxiety and depression and several illnesses specific cognitive and behavioural responses. A CBT-informed physiotherapy was designed to address these factors by focussing on beliefs about symptoms, attention allocation, and fear extinction amongst other things. The intervention achieved high rates of acceptability, recruitment, and retention, and promising treatment effects compared to current best practice, which warrants progression to a multicentre randomised efficacy trial.

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Appendix A: Questionnaires

Questionnaires

Thank you for agreeing to participate in this study. We are very interested in finding out more about the symptoms of your dizziness and the impact these symptoms have had on your life.

This booklet contains a number of questions about the nature of your symptoms, how you manage these symptoms and the impact these have had on your physical health and quality of life

There are no **right** or **wrong** answers to these questions. We are most interested in your **own** personal views rather than those of your family or the people who are treating you.

- **We ask you to answer the questions as honestly and as quickly as possible.**

- **If you find it hard to keep your mind on the statements, take a short break.**

This questionnaire is completely CONFIDENTIAL

It will only be seen by the researcher and not by any of the staff who look after you.

As soon as you have completed the questionnaire, please return it in the stamped, self-addressed envelope provided.

Thank you very much for your time

Questions About You

Full Name:	
DOB:	DD/MM/YYYY
Gender:	
How long have you experienced dizziness? (try to be as accurate as possible)	

1. Do you still have symptoms of dizziness? (please tick one box)

☐ Yes ☐ No

2. Do you speak English?

☐ Yes ☐ No

If you answered NO to either question 1 or 2, please STOP *and return the consent form and questionnaire using the self-addressed envelope provided. If you answered YES, please continue.*

3. What is your ethnicity? (tick one)

Black or Black British

☐ Caribbean

☐ African

☐ Any other black background

White

☐ British

☐ Irish

☐ Any other white background

Asian or Asian British

☐ Indian

☐ Pakistani

☐ Bangladeshi

☐ Any other Asian background

Mixed

☐ White & Black Caribbean

☐ White & Black African

☐ White & Asian

☐ Any other mixed background

Other ethnic groups

☐ Chinese

☐ Any other ethnic groups

4. What is your current relationship status? (tick one)

☐ Single

☐ Living with a partner

☐ Separated

☐ Married/civil partnership

☐ Widowed

☐ Divorced

5. What is the highest level of education you have completed? (tick one)

☐ No formal education
☐ GCSE/O level or equivalent
☐ A level or equivalent
☐ Certificate/diploma
☐ Trade/apprenticeship
☐ University degree
☐ Post graduate (e.g. Masters or higher)
Other (please specify) ……

6. Which of the following best describes your current job status? (tick one)

☐ Employed outside the home, full-time *(go to Q6a)*
☐ Employed outside the home, part-time *(go to Q6a)*
☐ Unemployed *(go to Q6b)*
☐ Homemaker
☐ Retired
☐ Student
☐ Other

6(a) Have you reduced your hours of work because of your dizziness?

☐ Yes ☐ No

6(b) Are you unemployed because of your dizziness?

☐ Yes ☐ No

7. Are you currently taking medication which has been prescribed by a doctor for dizziness? (tick one)

☐ Yes ☐ No

The following section asks you specific questions about your experience of dizziness

Dizziness Handicap Inventory (DHI)

The purpose of this questionnaire is to identify difficulties that you may be experiencing because of your 'dizziness'. Please circle 'Yes' *or* 'Sometimes' *or* 'No' to each question. Answer each question only as it pertains to your dizziness problem.

P1	Does looking up increase your problem?	Yes	Sometimes	No
E2	Because of your problem, do you feel frustrated?	Yes	Sometimes	No
F3	Because of your problem, do you restrict your travel for business or recreation?	Yes	Sometimes	No
P4	Does walking down the aisle of a supermarket increase your problems?	Yes	Sometimes	No
F5	Because of your problem, do you have difficulty getting into or out of bed?	Yes	Sometimes	No
F6	Does your problem significantly restrict your participation in social activities, such as going out to dinner, going to the movies, dancing, or going to parties?	Yes	Sometimes	No
F7	Because of your problem, do you have difficulty reading?	Yes	Sometimes	No
P8	Does performing more ambitious activities such as sports, dancing, household chores (sweeping or putting dishes away) increase your problems?	Yes	Sometimes	No
E9	Because of your problem, are you afraid to leave your home without having someone accompany you?	Yes	Sometimes	No
E10	Because of your problem have you been embarrassed in front of others?	Yes	Sometimes	No
P11	Do quick movements of your head increase your problem?	Yes	Sometimes	No
F12	Because of your problem, do you avoid heights?	Yes	Sometimes	No
P13	Does turning over in bed increase your problem?	Yes	Sometimes	No
F14	Because of your problem, is it difficult for you to do strenuous homework or yard work (gardening)?	Yes	Sometimes	No
E15	Because of your problem, are you afraid people may think you are intoxicated?	Yes	Sometimes	No
F16	Because of your problem, is it difficult for you to go for a walk by yourself?	Yes	Sometimes	No
P17	Does walking down a sidewalk (pavement) increase your problem?	Yes	Sometimes	No
E18	Because of your problem, is it difficult for you to concentrate?	Yes	Sometimes	No
F19	Because of your problem, is it difficult for you to walk around your house in the dark?	Yes	Sometimes	No
E20	Because of your problem, are you afraid to stay home alone?	Yes	Sometimes	No
E21	Because of your problem, do you feel handicapped?	Yes	Sometimes	No
E22	Has the problem placed stress on your relationships with members of your family or friends?	Yes	Sometimes	No
E23	Because of your problem, are you depressed?	Yes	Sometimes	No
F24	Does your problem interfere with your job or household responsibilities?	Yes	Sometimes	No
P25	Does bending over increase your problem?	Yes	Sometimes	No

Vertigo Symptom Scale (VSS)

Please circle the appropriate number to indicate about how many times you have experienced each of the symptoms listed below during the past year.

The range of responses are:

0	1	2	3	4
Never	A few times	Several times	Quite often (every week)	Very often (most days)

How often in the past month have you had the following symptoms:

V1	A feeling that either you, or things around you, are spinning or moving, lasting less than 20 minutes	0	1	2	3	4
A2	Hot or cold spells	0	1	2	3	4
V3	Nausea (feeling sick), vomiting	0	1	2	3	4
V4	A feeling that either you, or things around you, are spinning or moving, lasting more than 20 minutes	0	1	2	3	4
A5	Heart pounding or fluttering	0	1	2	3	4
V6	A feeling of being dizzy, disoriented or 'swimmy', lasting all day	0	1	2	3	4
A7	Headache, or feeling of pressure in the head	0	1	2	3	4
V8	Unable to stand or walk properly without support, veering or staggering to one side	0	1	2	3	4
A9	Difficulty breathing, being short of breath	0	1	2	3	4
V10	Feeling unsteady, about to lose balance, lasting more than 20 minutes	0	1	2	3	4
A11	Excessive sweating	0	1	2	3	4
A12	Feeling faint, about to black out	0	1	2	3	4
V13	Feeling unsteady, about to lose balance, lasting less than 20 minutes	0	1	2	3	4
A14	Pains in the heart or chest region	0	1	2	3	4
V15	A feeling of being dizzy, disoriented or 'swimmy', lasting less than 20 minutes	0	1	2	3	4

Illness Perceptions Questionnaire – Revised (IPQ-R)

YOUR VIEWS ABOUT YOUR ILLNESS

Listed below are a number of symptoms that you may or may not have experienced since your condition. Please indicate by circling *Yes* or *No*, whether you have experienced any of these symptoms since your condition, and whether you believe that these symptoms are related to your condition.

	I have experienced this symptom *since my condition*			This symptom is *related to my condition*	
Pain	Yes	No	_____	Yes	No
Sore throat	Yes	No	_____	Yes	No
Nausea	Yes	No	_____	Yes	No
Breathlessness	Yes	No	_____	Yes	No
Weight loss	Yes	No	_____	Yes	No
Fatigue	Yes	No	_____	Yes	No
Stiff joints	Yes	No	_____	Yes	No
Sore eyes	Yes	No	_____	Yes	No
Wheeziness	Yes	No	_____	Yes	No
Headaches	Yes	No	_____	Yes	No
Upset stomach	Yes	No	_____	Yes	No
Sleep difficulties	Yes	No	_____	Yes	No
Dizziness	Yes	No	_____	Yes	No
Loss of strength	Yes	No	_____	Yes	No
Imbalance	Yes	No	_____	Yes	No
Blurred vision	Yes	No	_____	Yes	No
Sensitivity to light or sound	Yes	No	_____	Yes	No
Hearing difficulties	Yes	No	_____	Yes	No
Difficulty concentrating	Yes	No	_____	Yes	No
Speech problems	Yes	No	_____	Yes	No
Sensitivity or fullness in the ears	Yes	No	_____	Yes	No
Motion sickness	Yes	No	_____	Yes	No
Tinnitus (noises in the ear)	Yes	No	_____	Yes	No

We are interested in your own personal views of how you now see your dizziness. Please indicate how much you agree or disagree with the following statements about your dizziness by ticking the appropriate box.

VIEWS ABOUT YOUR SYMPTOMS		STRONGLY DISAGREE	DISAGREE	NEITHER AGREE NOR DISAGREE	AGREE	STRONGLY AGREE
IP1	My dizziness will last a short time					
IP2	My dizziness is likely to be permanent rather than temporary					
IP3	My dizziness will last for a long time					
IP4	This dizziness will pass quickly					
IP5	I expect to have this dizziness for the rest of my life					
IP6	My dizziness is a serious condition					
IP7	My dizziness has major consequences on my life					
IP8	My dizziness does not have much effect on my life					
IP9	My dizziness strongly affects the way others see me					
IP10	My dizziness has serious financial consequences					
IP11	My dizziness causes difficulties for those who are close to me					
IP12	There is a lot which I can do to control my symptoms					
IP13	What I do can determine whether my dizziness gets better or worse					
IP14	The course of my dizziness depends on me					
IP15	Nothing I do will affect my dizziness					
IP16	I have the power to influence my dizziness					
IP17	My actions will have no effect on the outcome of my dizziness					
IP18	My dizziness will improve in time					
IP19	There is very little that can be done to improve my dizziness					
IP20	My treatment will be effective in curing my dizziness					

VIEWS ABOUT YOUR SYMPTOMS		STRONGLY DISAGREE	DISAGREE	NEITHER AGREE NOR DISAGREE	AGREE	STRONGLY AGREE
IP21	The negative effects of my dizziness can be prevented (avoided) by my treatment					
IP22	My treatment can control my dizziness					
IP23	There is nothing which can help my condition					
IP24	The symptoms of my condition are puzzling to me					
IP25	My dizziness is a mystery to me					
IP26	I don't understand my dizziness					
IP27	My dizziness doesn't make any sense to me					
IP28	I have a clear picture or understanding of my condition					
IP29	The symptoms of my dizziness change a great deal from day to day					
IP30	My symptoms come and go in cycles					
IP31	My dizziness is very unpredictable					
IP32	I go through cycles in which my dizziness gets better and worse.					
IP33	I get depressed when I think about my dizziness					
IP34	When I think about my dizziness I get upset					
IP35	My dizziness makes me feel angry					
IP36	My dizziness does not worry me					
IP37	Having this dizziness makes me feel anxious					
IP38	My dizziness makes me feel afraid					

CAUSES OF MY ILLNESS

We are interested in what you consider may have been the cause of your illness. As people are very different, there is no correct answer for this question. We are most interested in your own views about the factors that caused your illness rather than what others including doctors or family may have suggested to you. Below is a list of possible causes for your illness. Please indicate how much you agree or disagree that they were causes for you by ticking the appropriate box.

POSSIBLE CAUSES		STRONGLY DISAGREE	DISAGREE	NEITHER AGREE NOR DISAGREE	AGREE	STRONGLY AGREE
C1	Stress or worry					
C2	Hereditary – it runs in my family					
C3	A Germ or virus					
C4	Diet or eating habits					
C5	Chance or bad luck					
C6	Poor medical care in my past					
C7	Pollution in the environment					
C8	My own behaviour					
C9	My mental attitude e.g. thinking about life negatively					
C10	Family problems or worries caused my condition					
C11	Overwork					
C12	My emotional state, e.g. feeling down, lonely, anxious, empty					
C13	Ageing					
C14	Alcohol					
C15	Smoking					
C16	Accident or injury					
C17	My personality					
C18	Altered immunity					

In the table below, please list in rank-order the three most important factors that you now believe caused YOUR illness. You may use any of the items from the box above, or you may have additional ideas of your own.

The most important causes for me:-

1. _____
2. _____
3. _____

Cognitive Behavioural Response to Symptoms Questionnaire (CBRQ)

Please indicate how much you agree or disagree with the following statements about your current symptoms by ticking the appropriate box.

	VIEWS ABOUT YOUR SYMPTOMS	STRONGLY DISAGREE	DISAGREE	NEITHER AGREE NOR DISAGREE	AGREE	STRONGLY AGREE
FA1	I am afraid that I will make my symptoms worse if I exercise					
FA2	My symptoms would be relieved if I were to exercise					
FA3	Avoiding unnecessary activities is the safest thing I can do to prevent my symptoms from worsening					
FA4	The severity of my symptoms must mean there is something serious going on in my body					
FA9	Even though I experience symptoms, I don't think they are actually harming me					
FA10	When I experience symptoms, my body is telling me that there is something seriously wrong.					
FA12	Physical activity makes my symptoms worse					
FA14	Doing less helps symptoms					
FA15	Symptoms are a signal that I am damaging myself					
FA16	I am afraid I will have more symptoms if I am not careful					
FA17	I should avoid exercise when I have symptoms					
C1	I worry that I may become permanently bedridden because of my symptoms					
C2	I think that if my symptoms get too severe they may never decrease					
C4	My illness is awful and I feel that it overwhelms me					
C6	I will never feel right again					
SF1	When I experience symptoms, I think about them constantly					
SF2	I worry when I am experiencing symptoms					
SF3	When I am experiencing symptoms it is difficult for me to think of anything else					
SF5	I think a great deal about my symptoms					
SF9	My symptoms are always at the back of my mind					
SF12	I spend a lot of time thinking about my illness					
EA1	I am embarrassed about my symptoms					
EA2	I worry that people will think badly of me because of my symptoms					
EA3	The embarrassing nature of my symptoms prevents me from doing things					
EA4	I avoid social situations because I am scared my symptoms will get out of control					
EA5	I am ashamed of my symptoms					
EA6	My symptoms have the potential to make me look foolish in front of other people					

We are interested in how you respond to or manage your symptoms at the moment. Listed below are a number of different responses that people may have to their symptoms.

Please indicate how often you respond in the following ways by ticking the appropriate box. Choose the most accurate answer for YOU, not what you think "most people" would say or do.

	MANAGING SYMPTOMS	**Never**	**Sometimes**	**Quite often**	**Very often**	**All the time**
L2	I stay in bed to control my symptoms					
L3	When I experience symptoms, I rest					
L4	I tend to avoid activities that make my symptoms worse					
L7	I tend to nap during the day to control my symptoms					
AL1	I tend to overdo things when I feel energetic					
AL2	I find myself rushing to get things done before I crash					
AL3	I tend to overdo things and then rest up for a while					
AL4	I tend to do a lot on a good day and rest on a bad day					
L9	I sleep when I'm tired in order to control my symptoms					
L10	I avoid making social arrangements in case I'm not up to it					
L11	I avoid exerting myself in order to control my symptoms					
AL5	I'm a bit all or nothing when it comes to doing things					
L13	I avoid stressful situations					

Which of the following best describes the nature of your symptoms (please tick one):

My symptoms are physical	My symptoms are mainly physical	Both physical and psychological factors are involved in my symptoms	My symptoms are mainly psychological	My symptoms are psychological in nature

Patient Health Questionnaire (PHQ-9)

Over the last 2 weeks, how often have you been bothered by any of the following problems? (please circle one answer for each question)

		Not at all	Several days	More than half the days	Nearly every day
1	Little interest or pleasure in doing things	0	1	2	3
2	Feeling down, depressed, or hopeless	0	1	2	3
3	Trouble falling or staying asleep, or sleeping too much	0	1	2	3
4	Feeling tired or having little energy	0	1	2	3
5	Poor appetite or overeating	0	1	2	3
6	Feeling bad about yourself — or that you are a failure or have let yourself or your family down	0	1	2	3
7	Trouble concentrating on things, such as reading the newspaper or watching television	0	1	2	3
8	Moving or speaking so slowly that other people could have noticed? Or the opposite — being so fidgety or restless that you have been moving around a lot more than usual	0	1	2	3
9	Thoughts that you would be better off dead or of hurting yourself in some way	0	1	2	3

Generalized Anxiety Disorder (GAD-7)

Over the last 2 weeks, how often have you been bothered by any of the following problems? (please circle one answer for each question)

		Not at all	Several days	More than half the days	Nearly every day
1	Feeling nervous, anxious or on edge	0	1	2	3
2	Not being able to stop or control worrying	0	1	2	3
3	Worrying too much about different things	0	1	2	3
4	Trouble relaxing	0	1	2	3
5	Being so restless that it is hard to sit still	0	1	2	3
6	Becoming easily annoyed or irritable	0	1	2	3
7	Feeling afraid as if something awful might happen	0	1	2	3

Psychological Vulnerability Scale (PVS)

For each statement please tick a box to rate how well it describes you

	1 Does not describe me at all	**2**	**3**	**4**	**5 Describes me very well**
1) If I don't achieve my goals, I feel like a failure as a person					
2) I feel entitled to better treatment from others than I generally receive					
3) I am frequently aware of feeling inferior to other people					
4) I need approval from others to feel good about myself					
5) I tend to set my goals too high and become frustrated trying to reach them					
6) I often feel resentful when others take advantage of me					

Beliefs About Emotions Scale (BAE)

Please read each statement below and decide how well it describes how you have felt over the past three months. Please indicate your answer by placing a tick under the column that best describes what you think. To decide whether a given answer has been typical of your way of looking at things over the past three months, simply keep in mind what you are like most of the time.

	Totally agree	Agree very much	Agree slightly	Neutral	Disagree slightly	Disagree very much	Totally disagree
1. If I have difficulties I should not admit them to others.	6	5	4	3	2	1	0
2. I should be able to control my emotions.	6	5	4	3	2	1	0
3. If I am having difficulties, it is important to put on a brave face.	6	5	4	3	2	1	0
4. If I show signs of weakness, then others will reject me.	6	5	4	3	2	1	0
5. I should not let myself give in to negative feelings.	6	5	4	3	2	1	0
6. To be acceptable to others, I must keep any difficulties or negative feelings to myself.	6	5	4	3	2	1	0

THANK YOU. YOU HAVE NOW FINISHED.

Appendix B: Patient Information Sheet for survey

Version Number 2 IRAS Project ID: 211535 07/02/2017

Guy's and St Thomas' NHS
NHS Foundation Trust

KING'S College LONDON

INFORMATION SHEET FOR PARTICIPANTS

REC Reference Number: 16/NI/0256

ID No:

YOU SHOULD KEEP THIS INFORMATION SHEET FOR YOUR RECORDS

Title of study

Factors which affect quality of life and severity of symptoms in chronic dizziness

Invitation Paragraph
You are being invited to take part in a research study, which forms part of an educational degree (PhD). Before you decide whether you want to take part, it is important for you to understand why the research is being done and what your participation will involve. Please take time to read the following information carefully and discuss it with family members or friends if you wish. Please contact me if anything is unclear or if you would like more information. Take time to decide whether or not you wish to take part.

What is the purpose of the study?
Research has shown that many people go to hospital or visit their family doctor (GP) because of dizziness. For some people the dizziness will get better quickly, but other people may be left with more chronic symptoms. Dizziness is complex and is affected by many different things. We are interested in finding out more about the nature of your symptoms, how you manage these symptoms and the impact these have had on your physical health and your well-being. Learning about how these things may be linked will help us to manage them more effectively in the future.

Why have I been invited to take part?
You have been approached about this study because you are on the waiting list to attend the balance clinic at Guy's Hospital. We are inviting those people who still experience symptoms of dizziness to take part in the project.

Do I have to take part?
No. It is up to you to decide whether to take part or not. If you wish to participate we will ask you to complete and return the questionnaires. If you do not respond we may send you the questionnaires again with a reminder. We want to make sure you have the opportunity to take part if you have forgotten. However, you can ignore this if you do not want to take part.

What will happen to me if I take part?
We will want you to complete a questionnaire before your appointment and 3 months afterwards. We will also want to collect the results from your routine balance test. We will want to use anonymous data collected during the project for future ethically approved research.

If you agree to take part, you can complete the questionnaires and send it back to us using the prepaid self-addressed envelope provided. If you would prefer to complete an electronic version, you can access it using this web address: https://kings.onlinesurveys.ac.uk/dizziness-study

The questionnaires should take around 20-40 minutes to complete. The research is in no way connected with the treatment you will receive in the balance clinic. The survey will ask you some standard questions about how you view your dizziness, how it affects your life, how you manage with dizziness alongside other more general difficulties. If you have missed a question out, then researcher may contact you to clarify.

By completing and returning the questionnaire, you will be agreeing to your anonymous data being used for future research, and for the researchers to collect the routine balance test results from your medical records.

Incentives
We would very much appreciate your help. To thank you for your time completing the survey we will offer £10 once you have completed and returned the final survey. The money can be posted to your home address or you can collect it in person if you prefer. At the conclusion of the study, we will also provide you with a newsletter summarizing the main findings.

What are the possible benefits of taking part?
There may not be any immediate or direct benefits to you by taking part. However, some people may find it helpful or interesting to think about their illness and how it affects them. Your participation will help us to develop a better idea of how we can help people who experience persistent dizziness. At the end of the project, we will send you a newsletter describing the major findings and alerting you to any research publications we have generated from the project.

What are the possible risks of taking part?
The risks involved in participating are very small. It is possible that you might find it upsetting reflecting on questions in relation to your own experiences of dizziness. If you get upset, you can take a break or decide not to continue with the survey. The questionnaire content will not be used for clinical purposes and we are not able to provide individual feedback. However, should you become distressed during or after completing the questionnaire you will have the opportunity to contact the research team (see the contact details below). At this time, you may be asked if you would like relevant information about other agencies, such as patient support groups. The researchers may also encourage you to talk to your family doctor (GP) for more information. At this stage if the principle investigator is concerned about your distress then he will ask your permission for the lead supervisor (Rona Moss-Morris, a HPCP registered Psychologist) to contact you directly. She will then asses your circumstances over the telephone and suggest appropriate referral through the GP.

Will my taking part be kept confidential?
Yes. All information about your participation in this study will be kept confidential in accordance with the Data Protection Act 1998. Once the survey is complete, your name will be kept separately from the survey data, and a linking participant ID number will be used, making it anonymised. Your information will be stored on secure computers, locked

within offices and in locked file cabinets, and will only be available to members of the research team. This information will only be used for the purposes of the current study. Your study data will be retained for 4-years and subsequently disposed of securely. Your responses to our questions will remain completely confidential unless you tell us something to indicate that your own health and safety are currently in danger. Your anonymised data will be shared with other researchers and may be used for other research purposes. Please note you can request withdrawal of your data from the study up until it has been analysed. If you wish to withdraw from the study at any stage, this will remain anonymous and in no way will affect the quality of treatment you receive in the future.

How is the project being funded?
The project is funded by a National Institute for Health Research (NIHR) Clinical Doctoral Research Fellowship Award.

What will happen to the results of the study?
The results will be used to help the researchers better understand persistent dizziness and develop future treatments. The study will be presented at scientific conferences and be written up for publication in scientific journals. It will not be possible to identify you from any of the data. We will provide you with a summary sheet of the results.

Who should I contact for further information?
If you have any questions or require more information about this study, please contact me using the following contact details:

Name: Mr David Herdman
Job title: Principal Investigator (Clinical Doctoral Research Fellow)
Telephone number: 020 7188 0188 Email address: david.herdman@kcl.ac.uk
Address: Department of Health Psychology, Institute of Psychiatry Psychology & Neuroscience (IoPPN), Kings College London, 5th Floor Bermondsey Wing, Guys Campus, London SE1 9RT

What if something goes wrong?
If you have a concern about any aspect of this study, you should ask to speak to Professor Rona Moss-Morris on 020 7188 0178 or email rona.moss-morris@kcl.ac.uk who will do her best to answer your questions. If you remain unhappy and wish to complain formally, you can do this through the Guy's and St Thomas' Patients Advice and Liaison Service (PALS) on 020 7188 8801, pals@gstt.nhs.uk. The PALS team are based in the main entrance on the ground floor at St Thomas' Hospital and on the ground floor at Guy's Hospital in the Tower Wing.

If something does go wrong and you are harmed during the research you may have grounds for legal action for compensation against Guy's and St Thomas' NHS Foundation Trust and/or King's College London but you may have to pay legal costs. The normal National Health Service complaints mechanisms will still be available to you (if appropriate).

Thank you for reading this information sheet and for considering taking part in this research.

Appendix C: Search Terms for Systematic Review

Search Terms:

CINAHL:

Vestibular search terms:	*Psychosocial search terms:*
(MH "Labyrinth Diseases+")	anxiety N3 cognition*
(MH "Dizziness")	"Alexithymia"
(migrain* N6 (vertigo or dizz* or vestibul* or spinning))	(MH "Affective Symptoms+/DI/PP/PF/PR/RH/TH/SS")
((vertigo or vestibulopath* or dizziness* or vestibular or balance*) and (disorder or hypofunction* or dysfunction* or impair* or disability* or pathology* or disturbance* or syndrome* or symptom*))	(MH "Psychosocial Aspects of Illness+")
	catastrophi*
	"attentional bias"
	(MH "Anxiety+")
	(MH "Depression")
	TX psychologic N2 (aspect or factor*)
	"coherence"
	(MH "Sleep") OR (MH "Sleep Deprivation") OR (MH "Stress+")
	(MH "Anticipatory Anxiety")
	"acceptance"
	(health or illness or core or cognitive) N6 (representation* or belief* or bias*)
	(health or illness or coping) N3 (behavio?r* or ability* or strateg*)
	psychologic* N2 (aspect* or factor*)

EMBASE:

Vestibular search terms	*Psychosocial search terms*
exp vestibular disorder/co, di, dm, rh, th [Complication, Diagnosis, Disease Management, Rehabilitation, Therapy]	exp anxiety/
exp vertigo/co, di, dm, rh, th [Complication, Diagnosis, Disease Management, Rehabilitation, Therapy]	exp catastrophizing/
exp dizziness/co, di, dm, rh, th [Complication, Diagnosis, Disease Management, Rehabilitation, Therapy]	exp depression/

exp vestibular neuronitis/co, di, dm, rh, th [Complication, Diagnosis, Disease Management, Rehabilitation, Therapy]	exp psychological aspect/
exp vestibular migraine/co, di, dm, rh, th [Complication, Diagnosis, Disease Management, Rehabilitation, Therapy]	(psychologic* adj2 (aspect or factor*)).tw.
((vertigo or vestibulopath* or dizziness or vestibular or balance*) and (disorder or hypofunction* or dysfunction* or impair* or disability* or pathology* or disturbance* or syndrom* or symptom*)).ti.	exp coping behavior/
exp neurotology/	coherence.tw.
neuro-otology.tw.	(anxi* adj3 cognition*).tw. [mp=title, abstract, heading word, drug trade name, original title, device manufacturer, drug manufacturer, device trade name, keyword]
	alexithymia/ or alexithymia.tw.
	exp emotional stress/
	sleep disorder/ or insomnia/
	fear/ or anticipatory anxiety/
	acceptance.tw.
	((health or illness or coping) adj3 (behavio?r* or ability* or strateg*)).tw.
	((health or illness or core or cognitive) adj6 (representation* or belief* or bias)).tw.
	exp avoidance behavior/

MEDLINE:

Vestibular search terms	*Psychosocial search terms*
labyrinthitis/ or exp vestibular diseases/	exp Anxiety/co, px, rh, th [Complications, Psychology, Rehabilitation, Therapy]
dizziness/ or exp vertigo/	catastrophi*.tw.
Vestibular Neuronitis/	affective symptoms/ or depression/
(Vertigo or vestibulopath* or dizziness or ((vestibular or balance*) and (disorder or hypofunction* or dysfunction* or impair* or disability* or pathology* or disturbance* or syndrome* or symptom*))).ti.	((health or illness or core or cognitive) adj6 (representation* or belief* or bias)).tw.
exp Endolymphatic Hydrops/co, px, rh, th [Complications, Psychology, Rehabilitation, Therapy]	psychosocial.tw.
(migrain* adj6 (vertigo or dizz* or vestibul* or spinning)).tw.	"Sense of Coherence"/
Neurotology/	alexithymia.tw.

neuro-otology.tw.	Attention/co, pp, px, th [Complications, Physiopathology, Psychology, Therapy]
	(anxiety adj3 cognition).tw.
	Stress, Psychological/pp, px, rh [Physiopathology, Psychology, Rehabilitation]
	Fear/
	avoidance.tw.
	(psychologic* adj2 (aspect or factor)).tw.
	((health or illness or coping) adj3 (behavio?r* or ability* or strateg*)).tw.
	emotional.mp. and (stress or distress or exhaustion or pressure or tension).tw. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
	sleep/ or sleep deprivation/
	acceptance.tw.

PsychInfo:

Vestibular search terms	*Psychosocial search terms*
exp labyrinth disorders/	exp Anxiety/
exp vertigo/	exp fear/
exp menieres disease/	exp cognitive processes/
((Vestibular adj2 migraine) or (Migraine adj2 dizziness)).tw.	catastrophi*.tw.
((vertigo or vestibulopath* or dizziness* or vestibular or balance*) and (disorder or hypofunction* or dysfunction* or impair* or disability* or pathology* or disturbance* or syndrome* or symptom*)).tw.	exp "depression (emotion)"/
	exp coping behavior/ or ((health or illness or coping) adj3 (behavio?r* or ability* or strateg*)).tw.
	(anxiety adj3 cognition).tw.
	((health or illness or core or cognitive) adj2 (representation* or belief* or bias)).tw.
	exp Alexithymia/
	exp attention/
	acceptance.tw.
	sleep/ or sleep deprivation/ or sleep disorders/
	psychosocial.tw.
	(psychologic* adj2 (aspect or factor*)).tw.
	exp psychosocial factors/

Web of Science:

Vestibular search terms	*Psychosocial search terms*
(vestibular)	(anxiety)
((vertigo or vestibulopath* or dizziness or vestibular or balance) and (disorder or hypofunction* or dysfunction* or impair* or disabilit* or pathology* or disturbance* or syndrome* or symptom* or chronic*))	(depression)
((migrain* and (vertig* or dizz* or vestibul* or spinning or lightheaded*)))	("coping behaviour")
((meniere* OR (ENDOLYMPHATIC and HYDROPS) or (LABYRINTH and HYDROPS)))	((health or illness) near/3 belief*)
	("sense of coherence")
	(anxiety near/3 cognition*)
	(alexithymia)

Appendix D: Quality Assessment Criteria

Reporting

1. Is the hypothesis/aim/objective of the study clearly described?

2. Are the main outcomes to be measured clearly described in the Introduction or Methods section? If the main outcomes are first mentioned in the Results section, the question should be answered no.

3. Are the characteristics of the patients included in the study clearly described? In cohort studies and trials, inclusion and/or exclusion criteria should be given. In case-control studies, a case-definition and the source for controls should be given.

4. Are the distributions of principle confounders in each group of subjects to be compared clearly described? A list of principal confounders is provided. Yes =2, Partially =1, No =0

5. Are the main findings of the study clearly described? Simple outcome data (including denominators and numerators) should be reported for all major findings so that the reader can check the major analyses and conclusions (This question does not cover statistical tests which are considered below)

6. Does the study provide estimates of the random variability of the data for the main outcomes? In non-normally distributed data the inter-quartile range of results should be reported. In normally distributed the standard error, standard deviation or confidence intervals should be reported. If the distribution of the data is not described, it must be assumed that the estimates used were appropriate and the question should be answered yes.

7. Have the characteristics of patients lost to follow-up been described? This should be answered yes where there were no losses to follow-up or where losses to follow-up were so small that findings would be unaffected by their inclusion. This should be answered no, where a study does not report the number of patients lost to follow-up.

8. Have actual probability values been reported (e.g. 0.035 rather than <0.05) for the main outcomes except where the probability value is less than 0.001? If odd ratio and confidence intervals are provided, then the answer should be yes.

Diagnostic Accuracy

9. Are the methods used to confirm a diagnosis of a vestibular disorder appropriate? Do they reference clinical testing protocol and/or diagnostic classification

External Validity

10. Were the subjects asked to participate in the study representative of the entire population from which they were recruited? The study must identify the source population for patients and describe how the patients were selected. Patients would be representative if they comprised the entire source population, an unselected sample of consecutive patients, or a random sample. Random sampling is only feasible where a list of all members of the relevant population exists. Where a study does not report the proportion of the source population from which the patients are derived, the question should be answered as unable to determine.

11. Were those subjects who were prepared to participate representative of the entire population from which they were recruited? The proportion of those asked who agreed should be stated. Validation that the sample was representative would include demonstrating that the distribution of the main confounding factors was the same in the study sample and the source population.

Internal Validity – Bias

12. If any of the results of the study were based on 'data dredging', was this made clear? Any analysis that had not been planned at the outset of the study should be clearly indicated. If no retrospective unplanned subgroup analysis were reported, then answer yes.

13. In trials and cohort studies, do the analyses adjust for different lengths of follow-up of patients, or in case-control studies, is the time period between the intervention and outcome the same for cases and controls? Where follow-up was the same for all study patients the answer should be yes. If different lengths of follow-up were adjusted for by, for example, survival analysis the answer should be yes. Studies where differences in follow-up are ignored should be answered no.

14. Were the statistical tests used to assess the main outcomes appropriate? The statistical techniques used must be appropriate to the data. For example non-parametric methods should be used for small sample sizes. Where little statistical analysis has been undertaken but where there is no evidence of bias, the question should be answered yes. If the distribution of the data (normal or not) is not described it must be assumed that the estimates used were appropriate and the question should be answered yes.

15. Were the main outcome tools used accurate (valid and reliable)? For studies where the outcome measures are clearly described, the question should be answered yes. For

studies which refer to other work or that demonstrates the outcome measures are accurate, the question should be answered yes.

Internal Validity- Confounding

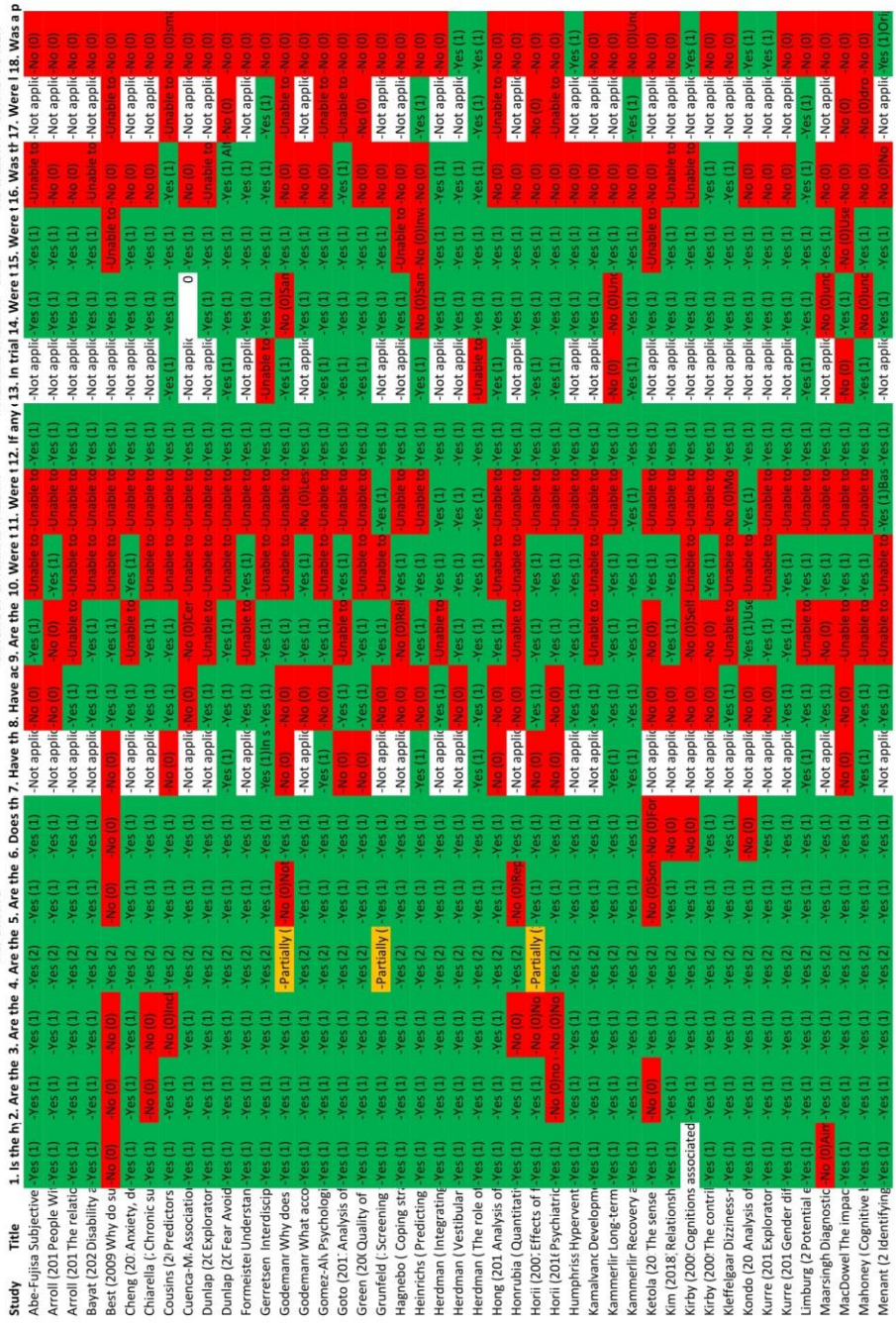
16. Was there adequate adjustment for confounding in the analyses from which the main findings were drawn? This question should be answered no for trials if: the main conclusions of the study were based on analyses of treatment rather than intention to treat; the distribution of known confounders in the different treatment groups was not described; or the distribution of known confounders differed between the treatment groups but was not taken into account in the analysis. In non-randomised studies if the effect of the main confounders was not investigated or confounding was demonstrated but no adjustment was made in the final analysis the question should be answered no.

17. Were losses of patients to follow-up taken into account? If the numbers of patients lost to follow-up are not reported, the question should be answered as unable to determine. If the proportion lost to follow-up was too small to affect the main findings, the question should be answered yes.

Power

18. Was a power calculation conducted? Original wording: Did the study have sufficient power to detect a clinically important effect where the probability value for a difference being due to chance is less than 5%? Sample sizes have been calculated to detect a difference of x% and y%.

Appendix E: Quality Assessment Scores



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Appendix F: Initial INVEST interview guide

The Initial Interview

Aim: To gather information on the cognitive, behavioural, and psychophysiological aspect of the dizziness complaints. To get a better understanding of the role of dizziness-related fear in the maintenance of the complaint and their interference with daily life activities.

Topics:

Symptoms & illness specific triggers

- Describe your current symptoms (affective quality, severity, triggers, course over the day)
- When did the dizziness start for the first time? (experiential learning, traumatic event)
- What is the course of your dizziness over time, from start until now? (Changes, intermittent episodes, number of episodes, slowly increasing)
- What are the things that can worse or ease the dizziness (safety cues, eliciting cues)?
- What do you do to cope with these symptoms?
- Do you have control over the dizziness?
- What do you do when the dizziness increases? (escape, avoidance, behaviour, pacing)
- What are you not doing anymore due to the dizziness? (avoidance behaviour)
- What are the consequences of your dizziness problem for your daily life activities? (work, household, leisure, social contacts, intimacy, family)
- What has changed in your life since you have been dizzy? (Negative reinforcement)

Illness specific triggers

- Is there something specific to [dizziness condition] that you are finding challenging at the moment? Tell me more about that? How do you cope with that?

Illness beliefs

- What do you think is going on in your body? What do you think causes your dizziness? (Catastrophic interpretations)
- Has your illness affected how you see yourself and if so how?
- Why do you think these are the causes of your dizziness ? (Verbal and observational learning)
- What is the cause of your dizziness as indicated by your doctors? (Verbal learning)

Treatment beliefs, self-management behaviours

- Talk me through the treatment recommendations for [dizziness condition]
- How are you managing your medicines/ prescribed treatments?
- How do other people respond when they observe you are dizzy? (social reinforcers)
- What would happen if you were continue with your valued activities despite dizziness? (Threat value of dizziness)

- What do you think will happen to your dizziness in the near future? (Expectancies)

Feelings

- How does living with the dizziness make you feel?

Therapy goals

- How would you like your life to be different? Why is this important to you?
- What do you wish to attain with this treatment? (Goals)

Appendix G: Patient Information Sheet for the INVEST Feasibility Trial

INFORMATION SHEET FOR PARTICIPANTS

IRAS No: 276707

YOU WILL BE GIVEN A COPY OF THIS INFORMATION SHEET

Title of project

INVEST feasibility study

Invitation Paragraph

I would like to invite you to participate in this research project which forms part of my PhD research. The study is being sponsored by King's College London. Before you decide whether you want to take part, it is important for you to understand why the research is being done and what your participation will involve. Please take time to read the following information carefully and discuss it with others if you wish. Ask me if there is anything that is not clear or if you would like more information.

What is the purpose of the project?

The treatment of dizziness consists of exercises called 'vestibular rehabilitation'. There is evidence to show that not everyone improves with this treatment. New studies suggest that it may be better to look at each person individually and teach them to think about their dizziness and the way they move in a different way. Using the latest evidence from neuroscience and psychology to enhance the usual vestibular exercises.

The purpose of the project is to assess whether it is possible to conduct a full-scale main study. To do this we will collect important information that is needed to design a larger study alongside interviews with participants.

Why have I been invited to take part?

You are being invited to participate in this project because you have experienced dizziness for at least three months. In order to ensure that you are eligible to take part in the study, you will need to complete a screening questionnaire. Once you return the completed screening questionnaire, a member of the research team will notify you of your eligibility.

What will happen if I take part?

For each participant, the study will take 4 months overall and your participation will involve the following:

- Complete online questionnaires (about 20-30 minutes)
- You will then be randomly allocated (like the flip of a coin) by a computer to one of two treatment approaches. You will not be able to choose the treatment group you

are assigned to. The study is conducted this way to ensure that the results are reliable. Below we provide more detail of what each approach will include
- You will be asked to complete the same questionnaires online after 4 months to see if there has been any change.

The two treatments are:

1) Treatment as usual physiotherapy
The role of being in this group is very important to the study because the outcomes gained by this group set the bar to know whether the new treatment is any better or not. You will receive six sessions of vestibular rehabilitation offered by the physiotherapist you would normally see at St George's Hospital. The treatment will be delivered as normal and is not determined by the study.

2) Psychologically informed physiotherapy
If you are allocated to this group, you will be asked to attend six sessions with a trained physiotherapist over a 4-month period. The first session will be 1 hour and the rest 30 minutes each. The exercises in each group will be similar but the treatment in this group will include a manual, which includes information about dizziness and all factors that can contribute to it. You will be assisted to increase your physical activity levels and look at different ways to manage your physical symptoms and emotional well-being.

At the end of the treatment you may be asked to be interviewed by a researcher. The researcher will be a different person to your therapist. They will ask you questions about your experience of the trial, such as your level of satisfaction and what you learnt. You can choose for the interview to take place over video or over the phone. Taking part in the interview is not essential to your participation in the study.

As part of participation you will be asked to provide permission for us to access:
- Hospital records: Information from your health records will allow us to confirm information about your test results and diagnosis.
- Video: You may be asked if it would be OK if some of your treatment sessions were video recorded. The purpose of these videos is to make sure that your physiotherapist is delivering the rehabilitation in the ideal way. The videos will not be used for any other purpose and will be destroyed after that use. If you are not comfortable with this, you can just decline the invitation. It will not affect your treatment in any way nor your relationship with your physiotherapist or any of the research team.

We will also inform your GP (General practitioner) of your involvement if you choose to take part.

Do I have to take part?

Participation is completely voluntary. You should only take part if you want to and choosing not to take part will not disadvantage you in anyway. Once you have read the information sheet, please contact us if you have any questions that will help you make a decision about taking part. If you decide to take part we will ask you to sign a consent form and you will be given a copy of this consent form to keep.

Costs

There are no costs to participants associated with the project. Any travel expenses will be reimbursed.

Do I have to come to the hospital?
You will be offered a face-to-face session in the clinic, but you can choose to attend your consultation online via a video call. The website is called 'Attend Anywhere', which is an NHS based video service currently being used at St George's Hospital. Video calls are secure, and no data is stored. You will be given a link to a private video room that only authorised clinicians can enter.

If you attend a face-to-face session you may be asked to wear a mask and the physiotherapist may be wearing some protective clothing, consistent with whatever the hospital guidelines and transmission-based precautions are at the time of your appointment.

What are the possible risks of taking part?

- Participation will require some time commitment to attend physiotherapy and complete the questionnaires and tests as described above.
- Participants in both groups will undergo a clinical examination and be prescribed movements to do in the clinic and at home. It is possible these may worsen your dizziness, especially in the short term. If you do experience any increase in dizziness, please bring this to the attention of your treating physiotherapist.
- There may be a risk of transmission of COVID19 from attending the hospital setting.
- It is possible that completing the questionnaires may cause you some distress, but this is rare. If the questions cause you any concerns or upset you, please stop answering the questions and speak to the researcher.
- Some of the questionnaires ask you about your mood. Depending on your responses, the researcher may ask you questions to assess your safety and make appropriate support services available to you if needed.

What are the possible benefits of taking part?

- If randomised to the usual physiotherapy care group, you will be greatly helping the study by your experience setting the benchmark by which we will know whether the physiotherapy protocol being studied is any better or not. Without this, our study would not be possible.
- If randomised to the new treatment group, you will receive physiotherapy treatment from a qualified physiotherapist who has taken the extra training required for them to deliver it.
- We anticipate the results of this research will allow us to:
    - Improve the knowledge we have about this health condition.
    - Inform health care practitioners about better ways of managing people with the same health condition as you.
    - Inform a future research study in persistent dizziness.

How will you use information about me?

Your data will be processed in accordance with the General Data Protection Regulation 2018 (GDPR).

- We will need to use information from you and your medical records for this research project. This information will include your name, hospital number and contact details. People will use this information to do the research or to check your records to make sure that the research is being done properly.
- People who do not need to know who you are will not be able to see your name or contact details. Your data will have a code number instead.
- All collected data, video-recordings, and interview transcripts (if you decided to take part in the interview) will be identified by the code number. This is to ensure your anonymity and confidentiality. Your personal information, such your name and contact details will be stored separately from all the collected data.
- The data will be stored securely in a locked cabinet in the Health Psychology department of King's College London. Electronic files will be locked, and password protected.
- Your study data will be retained for 4-years and subsequently disposed of securely. Everything discussed during the sessions or interview will remain completely anonymous unless you tell us something to indicate that your own health and safety is currently in danger.
- We will write our reports in a way that no-one can work out that you took part in the study.

Where can I find out more about how my information is used?

You can find out more about how we use your information
- at www.hra.nhs.uk/information-about-patients/
- by contacting King's College London's Data Protection Officer, Mr Albert Chan at info-compliance@kcl.ac.uk

What if I change my mind about taking part?

You are free withdraw at any point of the project, without having to give a reason. Withdrawing from the project will not affect you in any way. Your rights to access, change or move your information are limited, as we need to manage your information in specific ways in order for the research to be reliable and accurate. If you withdraw from the project, we will keep the information about you that we have already obtained. To safeguard your rights, we will use the minimum personally-identifiable information possible.

How is the project being funded?

This project is being funded by the National Institute for Health Research (NIHR). You can find more information about them by visiting their website (www.nihr.ac.uk).

What will happen to the results of the project?

The results of the project will be summarised in my PhD thesis. The study will also be presented at scientific meetings and written up for publication in scientific journal. We will make sure we write the reports about the study in a way that no-one can work out that you took part in the study. We will write to you at the end of the research and inform you of the publication of the main results of the research. This is likely to be in 2021.

Who should I contact for further information?

If you have any questions or require more information about this project, please contact me using the following contact details:

David Herdman
Email: david.herdman@kcl.ac.uk

What if I have further questions, or if something goes wrong?

If this project has harmed you in any way or if you wish to make a complaint about the conduct of the project you should first contact the research team at King's College London using the details below for further advice and information. If something does go wrong and you are harmed during the research you may have grounds for legal action for compensation against King's College London and/or NHS Trust, but you may have to pay your legal costs. The normal National Health Service complaints mechanisms will still be available to you (if appropriate).

Professor Rona Moss-Morris
Email: rona.moss-morris@kcl.ac.uk
Tel: +44 (0)207 188 0178

You can also contact St George's University Hospitals NHS Foundation Trust Patient Advice and Liaison Service (PALS):

Email: pals@stgeorges.nhs.uk
Tel: +44 (0)208 725 2453

Thank you for reading this information sheet and for considering taking part in this research.

Appendix H: Topic Guide for post INVEST Qualitative study

IRAS No: 276707 V1 04/02/2020

INVEST feasibility study

Qualitative Interview Schedule

Introduction

When you started the INVEST study you kindly agreed to be contacted to take part in an interview about your experiences of the INVEST programme. Are you still happy to complete the interview? I would like to record the conversation we have today so that I can refer back to it at a later date, is that ok?

Before we start there are a few things I'd just like to mention:

What we talk about will be used as part of the study, but anything said will remain anonymous, we're going to ensure this by not using your real name when we type up the interview.

If I ask a question that you don't want to answer that is absolutely fine, just say so and I'll ask you a different question. If at any point you would like to have a break or stop participating, then please just tell me and we will stop the interview.

I'm contacting a number of people who are taking part in the study to find out about their experience of the INVEST study. Anything you can tell me about your experiences including good and bad points would be useful.

Do you have any questions before we start? Are you happy to continue?

Right before I start the interview, I will do a brief introduction, please just ignore me.
Can I press record?

This is Participant number: xxxxx. Today is the Date: xxxx

Section A: Trial feasibility & acceptability

1. I am going to ask you some questions about your experience of taking part in the actual trial?
Prompts:

- Why did you agreed to take part in the study?
- What were your expectations of the study and were they met?
- What was your experience of completing the online questionnaires? Were they too long or was this ok? Were there some questions you didn't like?
- Did you find any difficulties with the process of attending the face-to-face or video sessions (e.g., getting time off work, childcare arrangements etc.)?
- Was there anything that could be different to make it easier for you to participate?

2. Now I'm going to ask you about your experiences you have had with the physiotherapist doing the therapy (acceptability)

- **Attitude** – do you feel positive about the treatment, what do you think of this method as a treatment option for managing dizziness?
- **Burden** – was the amount of effort required to participate in the treatment acceptable
- Intervention coherence – did the treatment make sense
- **Ethicality** – did this fit with you values, any ethical or moral concerns, what do you think about seeing a physiotherapist for this as opposed to, for example, a psychologist. Any side effects?
- **Opportunity costs** – what about the time involved in engaging in the therapy – e.g., using the manual, face to face or video, online questionnaires, exercises between session. Issues with adherence, reasons for drop out.
- **Perceived effectiveness** – was it effective in helping to manage your condition
- **Self-efficacy** – were you able to perform the activities required to participate in the treatment. Were there any barriers for you engaging in the exercises/tasks?

Section B: Intervention
Now just a few questions about the outcome of the therapy

1. How is your dizziness compared to before you started the trial? Why do you think that is? E.g., What do you think was the most important factor for you getting better, or not improving?
3. Can you tell me about anything that you feel has changed as a result of the study?
Prompts:
- Symptoms
- Thoughts / Beliefs
- Feelings
- Behaviours

4. Did you learn anything new about managing your condition compared to before the trial?
5. Did you try physiotherapy for this in the past? If so, how did this therapy differ?

Closing and Ending
Thank you very much for sharing your thoughts and experiences with me today.

What you've told me will really help us to understand patients' experiences and hopefully to improve our treatments for dizziness.

Before we finish, is there anything else you want to tell me? Is there anything you want to ask me?