THE DISAPPEARANCE OF A SIGNIFICANT OTHER: CONSEQUENCES AND CARE

Lonneke I.M. Lenferink

The work described in this dissertation was made possible by:

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Cover design by Klaas-Jelmer Sixma, www.studioklaes.nl
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Printing: Ridderprint BV, www.ridderprint.nl

Interior design: Joppe Klein, persoonlijkproefschrift.nl

ISBN: 978-94-034-0597-1 (paperback)
ISBN: 978-94-034-0596-4 (e-book)

rijksuniversiteit
groningen

The disappearance of a significant other

Consequences and care

Proefschrift

ter verkrijging van de graad van doctor aan de
Rijksuniversiteit Groningen
op gezag van de
rector magnificus prof. dr. E. Sterken
en volgens besluit van het College voor Promoties.

De openbare verdediging zal plaatsvinden op

donderdag 24 mei 2018 om 16.15 uur

door

Lonneke Ingrid Maria Lenferink

geboren op 2 oktober 1988
te Zwolle

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This dissertation is dedicated to the 655 family members and friends of missing or deceased persons who contributed to realisation of this dissertation.

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General introduction

Parts of this general introduction are based on: Lenferink, L. I. M., de Keijser, J., Wessel, I., & Boelen, P. A. (submitted). Achterblijvers na vermissing. In J. de Keijser, P.A. Boelen, G.E. Smid (Eds), *Handboek Complexe Rouw*. Amsterdam: Uitgeverij Boom

"Even after seven and a half years, whenever I see a Toyota Camry the same colours as Dad's, I check the number plate…When this sort of thing happens, your world changes in an instant: one minute that person is there: the next they're not. From then on your life isn't the same. And the real challenge with this kind of event is that it's uncommon, so the police and the community don't generally know how to respond. It can be very isolating."

(In 2008, Jason's father was last seen driving in his car on a road just outside his hometown (Missing Persons Advocacy Network, 2017))

"You still want to know. Even though we are almost 100% sure that we will not find her, not alive anyway. It is difficult, you know, because when there is an article in the newspaper saying "a body was found somewhere" or we hear in on the TV or the radio or whatever, then you are immediately on edge. And then you follow that news very intensively at such a time, so that sense of uncertainty remains a sensitive issue."

(About 30 years earlier Angie's teenaged sister disappeared on a night home alone leaving only traces of a crime behind)

"It is just as surreal and mind numbing as the first day because there are no answers. I try to exist day to day but the cold fact remains. I cannot give my husband a dignified burial. I cannot find his remains. I wake with Warren constantly in my thoughts. I go to bed the same. It keeps me awake at night. I cannot close this out. I cannot grieve effectively."

(Warren started a hiking trip about 8 years ago and never returned (Missing Persons Advocacy Network, 2017))

These phrases illustrate the psychological impact of the long-term disappearance of a significant other. Exploring consequences of, and care after the long-term disappearance of a significant other is the overarching aim of this dissertation. A missing person is defined as: "Anyone whose whereabouts is unknown whatever the circumstances of disappearance. They will be considered missing until located and their well-being or otherwise established" (Association of Chief Police Officers, 2010, p. 15). In the following, we summarize rates and types of missing persons, followed by a description of (the impact of) confrontation with the disappearance of a significant one, also termed "ambiguous loss" (Boss, 1999). Secondly, an overview is given of bereavement research that served as fundament of this dissertation. Lastly, an outline of the dissertation is presented.

1. THE DISAPPEARANCE OF A SIGNIFICANT OTHER

1.1 Rates of missing persons

In 2014, at least 80 persons[1] were daily reported as missing to the Dutch police. Fortunately, the majority of the missing persons was reconnected with their families within 48 hours (Schouten, van den Eshof, Schijf, & Schippers, 2016). In the Netherlands, yearly the whereabouts of about 100 missing persons remain unknown for more than one year (van Leiden & Hardemen, 2015). These rates of reports of missing persons and the occurrence of long-term missing persons are comparable to countries with similar political climates, for instance Germany and Australia (Schouten et al., 2016).

The long-term disappearance of a loved one can be considered an unnatural loss, and, as such, is a relatively rare phenomenon in the Netherlands compared with other types of unnatural losses. To illustrate this, in 2015 suicidal death, fatal traffic accident, and homicide occurred 1871, 621, and 120 times, respectively (Centraal Bureau voor de Statistiek, 2017a, 2017b, 2017c). Worldwide, disappearances of persons in armed conflict or due to disasters are more common. For example, 1,717 persons were unaccounted for one year after the 9/11 World Trade Center attacks in 2001 (Boss, 2004) and 2,668 persons were still reported as missing two years after the Tsunami in Japan 2011 (see Matsubara et al., 2014). The death of over 14,000 persons during the war in Bosnia Herzegovina in 1992-1995 was still unconfirmed after 10 years (ICRC, 2007). And it has been estimated that 27,000 enforced disappearances took place in Mexico since the beginning of the "war on drugs" initiated in 2006 (Amnesty International, 2016). In the Syrian refugee crisis, millions of people were separated from family and friends (Amnesty International, 2014).

1. Based on at least 30.000 reports to the Dutch police of missing persons in 2014.

1.2 Types of missing persons

In the Netherlands, about 2.9% of all the registered long-term missing persons cases is presumably caused by disasters or accidents, 3.3% caused by war, and 10.6% disappeared while traveling abroad (van Leiden & Hardemen, 2015). These types of disappearances are categorised as *unintentional absence* on a missing continuum (see Figure 1; Biehal, Mitchell, & Wade, 2003). In 10.5% of the Dutch missing person cases, the missing person is presumably killed or kidnapped (i.e., *forced* on the missing continuum). In 23.9% of the cases, the disappearance concerns an asylum seeker who has been separated from others and lost contact with significant others (i.e., *drifted* on the missing continuum). In 6.1% of the cases, the missing person presumably left voluntarily (i.e., *decided* on the missing continuum).

Figure 1. Missing continuum (Biehal et al., 2003).

These rates should, however, be interpreted with caution, because in most of the registered cases (i.e., 42.7%) the presumed reason/cause of disappearance was not registered. The registrations of missing persons were also based on different databases; saliently, most data originated from a database of a Dutch television show about missing persons and a database of the Dutch police. It is highly likely that the rates of disappearances are biased, due to a lack of a systematic national registration system of missing persons. Furthermore, the rates of disappearances by war and voluntarily missing person cases are most likely higher, because these types of disappearances are under-registered in Dutch databases (Leiden & Hardemen, 2015).

1.3 Ambiguous loss

The disappearance of a loved one is also termed “an ambiguous loss” (Boss, 1999). Boss defines two types of ambiguous loss: 1) psychological absence, but physical presence of a significant other (e.g., parent with dementia) and 2) physical absence, but psychological presence of a significant other (e.g., the disappearance of a spouse). Boss (2006, p. 7) presumes that ambiguous loss is “the most stressful kind of loss due to ambiguity”. Others have described that relatives of missing persons live “in a constant state of anxiety and restlessness” (Hollander, 2016, p. 299) and that an ambiguous loss is “cruel in its unending torment” (Betz & Thorngren, 2006, p. 359). However, these statements are all based on clinical experience, rather than empirical evidence.

Although it is not sufficiently empirically tested, there are several reasons to assume that the grief process following the long-term disappearance of a loved one differs from the grief process after the death of a loved one. Firstly, not knowing whether the loss is temporary or permanent, may lead to preoccupations with the (circumstances of the) disappearance, which in turn may complicate recovery from loss (Boss, 2006). Secondly, holding on to hope for the return of the (remains of the) missing loved one, while at the same time dealing with the absence of the person may prevent people to move on in life, reinforcing prolonged grief (PG) reactions (Heeke, Stammel, & Knaevelsrud, 2015). Furthermore, leaving those left behind without a burial ceremony or site to visit may also contribute to a disturbed grief process (Castle & Phillips, 2003). Fourthly, the absence of specific legislation that enables families of missing persons to obtain the legal right to manage the missing person's affairs[2], may result in considerable financial consequences. For instance, the loss of a missing person's income while continuation of a missing person's mortgage, taxes, and insurances, likely also burdens those left behind. In addition, relatives of missing persons may struggle to find their way through complex legal and ownership issues, with limited professional support available (Blaauw & Lähteenmäki, 2002; Holmes, 2008). Lastly, social marginalization, stigmatization, and lack of social support may be additional stressors that contribute to elevated psychopathology levels post-disappearance (Hollander, 2016; Quirk & Casco, 1994; Robins, 2010).

In sum, the long-term disappearance of a significant other outside the context of armed conflict or disaster appears to be a relatively rare phenomenon. It is conceivable that maladjustment to the loss is more common among relatives of missing persons than relatives of deceased persons because of disappearance-related stressors that may complicate the grief process. Below we present an overview of bereavement research, which is fundamental to this dissertation.

2. BEREAVEMENT RESEARCH

2.1 Grief work and beyond

After a century of research, a debate on which manifestations of grief (if any) should be considered pathological is still ongoing (see for instance Maciejewski & Prigerson, 2017; Stroebe, Schut, & Stroebe, 2007, Stroebe, Stroebe, Schut, & Boerner, 2017a; Zisook, Pies, & Corruble, 2012). The origin of the debate seems to date back to what Stroebe and Schut (1999) called the 'grief work hypothesis'. Freud (1917) is, according to Stroebe, Schut, and Boerner (2017b), the first who described mourning as 'inner labour'. Freud's idea was that mourners need to accept the reality of the loss and detach themselves from the deceased, which is accompanied with inner pain. Those who failed to do their grief work, most likely because of an ambivalent relationship with

2. With the exception of the declaration of presumed death that can be requested in the Netherlands after five years (Ministry of justice and security, 2017).

the deceased, may present pathological grief reactions. These reactions resemble melancholia, including painful dejection, social withdrawal, and decrease in activity. The importance of grief work has also been emphasized in multiple theories that have dominated the literature in the 20th century (see for an overview Stroebe et al., 2017a). For instance, Kübler-Ross' (1969) prescriptive grief stages (i.e., denial, anger, bargaining, depression, and acceptance) and the more descriptive task model of Worden (1991, 2008; i.e., accept the reality of the loss, process the pain of grief, adjust to a world without the deceased, and retain connection to the deceased) are examples of widespread models of grief processes.

However, the adaptive role of grief work has not been empirically supported (Bonanno, 1998; Bonnano & Kaltman, 1999). One of the first examples of a grief model that moves beyond describing tasks, stages, or phases of grief is the dual process model (Stroebe & Schut, 1999, 2010). According to this model, grief is a process of oscillation between confrontation and avoidance of loss-oriented stressors (i.e., aspects of the loss itself) and restoration-oriented stressors (i.e., secondary stressors resulting from bereavement). The dual process model of coping with bereavement describes processes that may occur in general bereavement (Maccallum & Bryant, 2013), in an attempt to explain what adaptive coping with bereavement means (Stroebe & Schut, 2001a). Since the mid 1990's, also another line of research arose that was not focused on normative reactions to the death of a significant other, but rather on debilitating non-normative grief reactions, labelled as "prolonged grief (PG)". Together with the development of the 19-item Inventory of Complicated Grief (ICG) in 1995 by Prigerson et al. research into disturbed grief reactions was initiated and new theoretical ideas were generated. Before we will describe these, we will elaboarte on PG a bit more.

2.2 Prevalence, consequences, and comorbidity of prolonged grief

Different labelling, for instance complicated or traumatic grief, has been used across studies to refer to debilitating non-normative grief reactions that may merit clinical attention. Since the introduction of the term prolonged grief disorder (PGD), this term has found strong consensus among some leading researchers in the field (Prigerson et al., 2009). Because there is no empirical evidence indicating that persistent grief reactions qualitatively differ from acute normative grief reactions (Bryant, 2014), the term PGD is most frequently used in this dissertation to refer to debilitating non-normative grief reactions. Before going into detail about the theory and treatment of PGD, the prevalence, consequences, and comorbidity of PGD are discussed.

2.2.1 Prevalence of prolonged grief

Examples of normative grief reactions are yearning for the deceased, experiencing difficulty accepting the loss, and difficulty engaging in activities following the loss (Prigerson et al., 2009).

When these acute grief reactions persist at least 6 months following the death and cause significant distress and impairments in daily life it may be defined as PGD (Prigerson et al., 2009). PGD will likely be included in the forthcoming 11th edition of the International Classification of Diseases (ICD-11; Maercker et al., 2013), albeit with symptom criteria that are likely to differ from the description put forth by Prigerson et al. (2009). The fifth edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5) included Persistent Complex Bereavement Disorder as condition for further study (APA, 2013). PGD and PCBD seem to cover the same diagnostic entity, based on resemblance in prevalence, sensitivity, and specificity rates, and predictive validity (Maciejewski, Maercker, Boelen, & Prigerson, 2016). They differ from each other with respect to the time criterion (6 months post-loss for PGD vs. 12 months post-loss for PCBD), number of symptoms (7 for PGD vs. 16 for PCBD), and possible symptom profile heterogeneity (48 ways to meet PGD criteria vs. 37,650 ways to meet PCBD criteria; Lenferink & Eisma, 2018).

The results of a meta-analysis suggest that 9.8% of people confronted with the non-violent loss (e.g., illness) of a significant other at least 6 months earlier is at risk for developing PGD (Lundorff, Holmgren, Zachariae, Farver-Vestergaard, & O'Connor, 2017). Experiencing the death of someone significant that happens unexpectedly and/or has a violent cause heightens the risk of elevated grief-related distress (Boelen, de Keijser, & Smid, 2015; Currier, Holland, & Neimeyer, 2006; Goldsmith et al., 2008; Kaltman & Bonanno, 2003). For instance, prevalence rates of PGD among people confronted with a disaster-related loss vary from 14% to 76% (see for an overview Kristensen, Weisæth, & Heir, 2012) and from 25% to 78% for people bereaved by suicide (see for an overview Linde, Treml, Steinig, Nagl, & Kersting, 2017). The disappearance of a significant other also seems to be a risk factor for poor adjustment (Kristensen et al., 2012). For instance, a study among Colombian people confronted with disappearances due to political repression 7 years earlier, showed prevalence rates of 23% for PGD, 69% for depression, and 67% for posttraumatic stress disorder (PTSD). These rates did not significantly differ from Colombian people exposed to the death of a significant other in the same violent context (Heeke et al., 2015). A comparative study, in the context of war, showed that Bosnian women confronted with the disappearance of their spouse reported significantly higher so-called "traumatic" grief and depression levels than Bosnian women confronted with the death of their spouse (Powell, Butollo, & Hagl, 2010). Another study showed higher depression levels among Bosnian adolescents whose fathers disappeared in war than Bosnian adolescents whose fathers were killed (Zvizdic & Butollo, 2001).

2.2.2 Consequences and comorbidity of prolonged grief

PG levels are associated with a host of detrimental health outcomes in bereaved people, including sleep disturbances (Germain, Caroff, Buysse, & Shear, 2005; McDermott et al., 1997), hypertension, cardiac problems (Prigerson et al., 1997), an increased risk for suicidal ideation and behaviour

(Maciejewski et al., 2016; Prigerson et al., 1997), an increased risk for mortality (Stroebe & Schut, 2001b), reduced quality of life (Boelen & Prigerson, 2007; Maciejewski et al., 2016; Silverman et al., 2000), impairments in work and social functioning (Bui et al., 2015; Kristensen, Weisæth, Hussain, & Heir, 2015; Maciejewski et al., 2016; Prigerson et al., 1997), and an increased risk of comorbid mood and anxiety symptoms (Newson, Boelen, Hek, Hofman, & Tiemeier, 2011; Shah & Meeks, 2012).

Factor analytic studies have shown that PG shows overlap with, yet is distinguishable from depression, anxiety, and posttraumatic stress (PTS) in bereaved people (Boelen & Prigerson, 2007; Boelen & van den Bout, 2005; Boelen, van den Bout, & de Keijser, 2003a; Chen et al., 1999; O'Connor, Lasgaard, Shevlin, & Guldin, 2010; Ogrodniczuk et al., 2003; Prigerson et al., 1996). In addition, studies, using latent class analysis have shown that people who are primarily suffering from elevated PG levels following the death or disappearance of a relative are distinguishable from people with elevated PG plus comorbid depression and/or PTS symptoms (Boelen, Reijntjes, Djelantik, & Smid, 2016; Boelen, Spuij, & Reijntjens, 2017; Djelantik, Smid, Kleber, & Boelen, 2017; Heeke, Stammel, Heinrich, & Knaevelsrud, 2017; Lenferink, de Keijser, Smid, Djelantik, & Boelen, 2017; Nickerson et al., 2014).

PG symptomatology shares some commonalities with PTS (including re-experiencing and avoidance symptoms; Heeke et al., 2017; Maercker & Znoj, 2010; Nickerson et al., 2014; O'Connor et al., 2010) and depression symptoms (e.g., sad mood, sense of emptiness, fatigue, and suicidal ideation/behaviour; Boelen et al., 2003a, 2016; Djelantik et al., 2017; MacCallum, Malgaroli, & Bonanno, 2017; Shear et al., 2011). It has been argued that re-experiencing symptoms in PGD differ from those in PTSD with respect to the emotional valence of the memories (Maercker & Znoj, 2010): for PGD re-experiencing of both positive and negative memories, often simultaneously, whereas for PTSD re-experiencing of negative memories is key. Furthermore, avoidance in PGD is aimed at averting painful thoughts and feelings related to the loss, whereas avoidance in PTSD is used to prevent thoughts and memories related to the traumatic event and the recurrence of threat or danger. Depression coincides with inactivity, whereas yearning in PGD seems to be associated with passivity as well as activity (Stroebe & Schut, 1999). In addition, a sad mood in depression is related to low self-esteem, whereas a sad mood in PGD is related to longing for the deceased (Shear et al., 2011).

In sum, since the mid 1990's research on bereavement has focused on PGD. One out of ten people confronted with a non-violent death of a significant other is at risk to develop PGD. Experiencing the disappearance of a significant other heightens the risk of elevated grief-related distress. PGD symptoms resemble normative grief reactions, but differ from these reactions regarding the intensity and duration. A diagnosis of PGD may apply to people experiencing grief reactions that cause significant distress and impairments in daily life following the death of a

significant other 6 months earlier. Elevated PG levels are associated with a host of detrimental physical and mental health outcomes. PG symptoms overlap with, yet are distinguishable, from PTS and depression symptoms.

3. THEORY OF DISTRESS POST-LOSS

Literature on PGD, PTSD, and depression in relatives of missing persons is scarce. To illustrate this, based on a literature review seven quantitative studies have previously examined the psychological consequences for people exposed to the disappearance of a significant other in the context of armed conflict (Heeke & Knaevelsrud, 2015). None of these studies examined correlates of distress post-disappearance that are amendable to change in treatment. Gaining insights into such correlates is relevant for developing psychological treatment for relatives of missing persons in need of support. Previous studies have primarily focused on the associations between sociodemographic and armed conflict-related stressors on the one hand and psychopathology levels on the other hand (Heeke & Knaevelsrud, 2015). To our knowledge, literature on severity and correlates of PGD, PTSD, and/or depression in people exposed to the disappearance of a significant other *outside* the context of armed conflict was absent. In this dissertation, we therefore relied on theories about variables associated with psychopathology in bereaved people that are amendable to therapeutic change.

Because of the limitations of the grief-work hypothesis, theories were generated regarding processes involved in the onset and maintenance of distress after bereavement (Boelen, van den Hout, & van den Bout, 2006a; Bonanno & Kaltman, 1999; Maccallum & Bryant, 2013; Shear & Shair, 2005). One of these theories is Boelen et al.'s (2006a) cognitive behavioural theory of PG. This theory is used as fundament in this dissertation because it attempts to enhance knowledge about underlying processes of PG, rather than normative grief, and specifies how these processes could be assessed and targeted in treatment.

Because the disappearance of a significant other is inherently linked to uncertainties (e.g., not knowing whether the person is alive or dead) that are uncontrollable (Boss, 2006), more than natural losses (e.g., caused by illness), ambiguous losses may give rise to repetitive thinking. This can be understood from the goal-discrepancy theory stating that repetitive thinking focused on goals that have not (yet) been attained (e.g., one feels sad when one wants to feel happy; one misses a loved one that one wants to be close to). What follows is that people with more extreme or unattainable goals may be more inclined to get entangled in repetitive thinking (Ehring & Watkins, 2008; Martin & Tesser, 1989). In this dissertation, we therefore also zoom in on several intrapersonal thinking processes that are expected to relate to disappearance-related distress and could be targeted in treatment.

3.1 Cognitive behavioural theory of prolonged grief

Inspired by cognitive-behavioural models of PTSD (Ehlers & Clark, 2000) and psychopathology (Beck, 1976), a cognitive-behavioural model of PG was developed (Boelen et al., 2006a). This model identifies three underlying but changeable processes that are assumed to play an important role in the onset and maintenance of PG: (a) insufficient integration of the loss into autobiographical knowledge, (b) negative cognitions, and (c) avoidance behaviour.

The first process, insufficient integration of the loss, reflects incongruence between factual knowledge that the loss has happened and experiencing a subjective sense of uncertainty about the permanence of the separation. This sense of uncertainty about the irreversibility of the loss is also coined 'a sense of unrealness' and is often expressed in phrases such as "I know that s/he is dead, but it feels as if it did not happen" (Boelen, 2010, 2017). Following the death of a loved one, most bereaved people naturally adapt to the factual knowledge that the separation is irreversible. The information about the deceased, the relationship with the deceased, and the related thoughts, memories, and feelings are processed and integrated with existing autobiographical knowledge. During this process meaning making takes place, resulting in "a coherent story about the relationship with the deceased with a beginning and an end" (Boelen et al., 2006a). It has been argued that some people have difficulties integrating the loss into autobiographical knowledge. This may result in intrusive memories, ruminative thoughts, feelings of shock, and yearning once the bereaved person is confronted with loss-related stimuli. This could be a variety of loss-related stimuli; all that was once related to the presence of the deceased is now related to his/her absence (Boelen et al., 2006a). Elevated levels of unrealness have been associated with elevated post-loss psychopathology levels (Boelen, 2010, 2017).

The second process includes negative cognitions about the loss and/or its sequelae. These negative cognitions include catastrophic misinterpretations of one's own grief reactions (e.g., "If I let go of my emotions, I will go crazy"), negative beliefs about the self (e.g., "I am ashamed of myself, since s/he died"), life (e.g., "Life has got nothing to offer me anymore"), and future (e.g., "I don't have confidence in the future"). Cross-sectional and longitudinal studies provided support for the associations of negative cognition with PG, PTS, and depression levels on the other hand (Boelen, van Denderen, de Keijser, 2016; Boelen, van den Bout, & van den Hout, 2003b, 2003c, 2006b; Horsch, Jacobs, & McKenzie-McHarg, 2015).

According to the cognitive-behavioural model of PG, the use of avoidance strategies is the third process involved in the onset and maintenance of PG (Boelen et al., 2006a). Avoidance strategies include anxious avoidance and depressive avoidance. Anxious avoidance represents avoidance of situations, places, or people that are associated with the deceased out of the belief that confrontation with reminders of the loss is unbearable. Depressive avoidance represents withdrawal from previous meaningful activities because of the belief that these activities are not

meaningful anymore since the loss. Avoidance behaviours have been linked, concurrently and longitudinally, to elevated psychopathology levels following loss (Boelen & Eisma, 2015; Boelen & van den Bout, 2010).

3.2 Emotion regulation strategies

According to the response styles theory (Nolen-Hoeksema, 1991), responses to affective states are of more importance for maintaining mood disorders than the affective state itself. The way bereaved people respond to a depressed mood, including depressive rumination, was first examined in the 1990s by Nolen-Hoeksema and colleagues (Nolen-Hoeksema, McBride, & Larson, 1997; Nolen-Hoeksema, Parker, & Larson, 1994). Depressive rumination is defined as "repetitively and passively focusing on symptoms of distress and on the possible causes and consequences of these symptoms" (Nolen-Hoeksema, Wisco, & Lyubomirsky, 2008, p. 400). Ironically, in an attempt to understand and solve problems, people start to ruminate; however, by fixating on the problems, rumination impedes active problem solving (Nolen-Hoeksema et al., 2008). Several studies indicated that people who were more inclined to depressive rumination (e.g., "When I feel down, sad, or depressed I think 'Why do I react this way?'") reported higher levels of depression concurrently and longitudinally following the loss of a significant other (Nolen-Hoeksema et al., 1994, 1997).

Similar to depression research (Feldman, Joormann, & Johnson, 2008), in bereavement research studies on affect regulation strategies are mainly focused on the maladaptive role of rumination (Eisma et al., 2015; Morina, 2011; Nolen-Hoeksema et al., 1994, 1997). Knowledge about the effect of positive affect regulation strategies on psychopathology levels is largely lacking, despite previous studies indicating that experiencing/expressing positive mood fosters recovery from loss (Bonnano & Keltner, 1997; Bonanno, Mihalecz, & LeJeune, 1999; Keltner & Bonanno, 1997; Tweed & Tweed, 2011). The Response to Positive Affect (RPA) questionnaire was developed by Feldman et al. (2008) to provoke more research into positive affect regulation strategies, including dampening and enhancing of positive affect. More dampening has been related to increased depression levels (Raes, Smets, Nelis, & Schoofs, 2012); more enhancing has been related to lower depression levels (Nelis, Holmes, & Raes, 2015). The RPA parallels the Ruminative Response Scale, which is often used to assess depressive rumination (Eisma & Stroebe, 2017; Treynor, Gonzales, & Nolen-Hoeksema, 2003). Raes and colleagues found that positive affect regulation strategies, assessed with the RPA, are concurrently and longitudinally linked to depression scores over and above depressive rumination (Raes, Daems, Feldman, Johnson, & Van Gucht, 2009; Raes et al., 2012, 2014).

As more recently argued (Eisma et al., 2015; Eisma & Stroebe, 2017), rumination about the causes and consequences of the loss may be of more relevance to bereaved people than rumination

about the causes and consequences of a depressed mood. Similar to depressive rumination, grief rumination (e.g., "How frequently in the past month did you ask yourself why you deserved this loss?") has also been linked concurrently and longitudinally to elevated psychopathology levels post-loss (Eisma et al., 2015). In the rumination-as-avoidance hypothesis it has been argued that rumination might be a cognitive avoidance style. For instance, by repeatedly thinking about how the loss could have been prevented, the bereaved person avoids thinking about the irreversibility of the loss, which may interfere with acceptance of the loss (Boelen, 2006; Eisma et al., 2013; Eisma & Stroebe, 2017; Stroebe et al., 2007).

Accepting and embracing one's own suffering, coined 'self-compassion', has been identified as a factor preventing people to get entangled in ruminative thinking. A self-compassionate attitude is linked to engagement with, rather than avoidance, of distressing thoughts, memories, and feelings (Leary et al., 2007; Thompson & Waltz, 2008). Based on a finding that self-compassion was significantly and negatively associated with the avoidance PTSD cluster, but not with other PTSD clusters, Thompson and Waltz (2008) concluded that self-compassion might be "a natural process of exposure to trauma-related stimuli" (Thompson & Waltz, 208, pp. 558). Cross-sectional studies have shown that people with higher levels of self-compassion are less inclined to ruminate (Neff, 2003; Svendsen, Kvernenes, Wiker, & Dundas, 2016). The findings of two other cross-sectional studies suggested that depressive rumination might mediate the associations between self-compassion and depression and anxiety levels (Krieger, Altenstein, Baettig, Doerig, & Holtforth, 2013; Raes, 2010). However, the adaptive role of self-compassion in bereaved people remains to be studied.

In conclusion, to enhance our understanding of clinical correlates of psychological symptoms in relatives of missing persons, several theories drawing from research in PG, depression, and PTSD are relevant. First, the cognitive behavioural theory of PG has repeatedly shown to be relevant for our understanding of how negative cognitions and avoidance behaviours are related to post-loss distress and how these variables could be targeted in treatment. Drawing from depression research, ruminating about one's sad mood (i.e., depressive rumination), but also ruminating about the causes and consequences about the loss (i.e., grief rumination) have been identified as risk factors for distress in bereaved people. Lastly, based on depression and PTSD research, indicating the protective role of adaptive regulation of positive affect and self-compassion, it seems worthwhile to explore how these clinical correlates relate to PG, PTS, and depression levels post-loss.

4. TREATMENT OF DISTRESS POST-LOSS

To the best of our knowledge, only one study examined treatment effects for relatives of missing persons. In this quasi-randomised controlled trial (Hagl, Rosner, Butollo, & Powell, 2015), 57 women whose husbands were missing following the war in Bosnia Herzegovina, as well as 62 women whose husbands were killed in war were allocated to one of two conditions. In the first condition people received dialogical exposure group therapy, using an empty-chair method derived from Gestalt therapy to start a direct dialogue with the deceased/missing husband. The second condition consisted of a supportive group in which people talked *about*, instead of *to*, the deceased/missing husband. In addition, elements of CBT including targeting unhelpful thoughts and behavioural elements were added to both conditions. Although the findings of this trial indicate that both conditions seem effective in reducing PTSD and so-called "traumatic grief" levels (i.e., yielding small to moderate effect sizes), the trial was limited in several ways. First, it had some methodological drawbacks, for instance, the use of a quasi-randomization procedure and the measure used to assess grief reactions had psychometric weaknesses. Second, the generalizability of the findings to people confronted with a disappearance not related to the Bosnia-Herzegovina war is limited due to unique features of this study sample (e.g., exposure to other war-related stressors and specific type of disappearance, and low literacy levels).

Based on overviews of treatment effects for bereaved people at least two conclusions can be drawn. Firstly, PG can successfully be treated, but there is limited evidence that PG can be prevented (Boelen & Smid, 2017; Doering & Eima, 2016; Linde et al., 2017; Mancini, Griffin, & Bonanno, 2012; Wittouck, van Autreve, de Jaegere, Portzky, & van Heeringen, 2011). For instance, a meta-analysis reviewed the effectiveness of nine preventive interventions and five PG treatments. Overall, they concluded that PG treatments, but not preventive interventions, yielded significant reductions in PG levels at post-treatment and follow-up compared with pre-treatment (Wittouck et al., 2011). Treatment is therefore most effective when targeted at people experiencing clinically relevant PG levels.

Secondly, cognitive behavioural therapy (CBT) has shown to be most effective in reducing grief-related distress (Boelen & Smid, 2017; Currier, Holland, & Neimeyer, 2010; Doering & Eisma 2016; Mancini et al., 2012). CBT is therefore to date the treatment of choice for bereaved people in need of professional support (see for instance Dutch guidelines for treatment of disturbed grief in Boelen & van den Bout, 2017). CBT includes exposure, cognitive restructuring, and behavioural activation, targeted at the three processes of the cognitive-behavioural model of PG (Boelen et al., 2006a).

Some argue that, unlike treatments for bereaved people, treatment for relatives of missing persons should not focus on "closure" or "coming to terms with the loss", because this may

provoke resistance in relatives of missing persons (Boelen & Smid, 2017; Boss, 2006; Glassock, 2006). Instead, treatment should focus on learning how to manage persistent negative thoughts and feelings related to the disappearance, for instance with mindfulness training (Boss, 2006). These recommendations rely on clinical experiences, rather than empirical evidence.

Mindfulness-based interventions (MBIs) have shown to be promising for bereaved people (O'Connor, Piet, & Hougaard, 2014; Thieleman, Cacciatore, & Hill, 2014). In a controlled pilot study, elderly bereaved people with clinically relevant PGD, PTSD, and/or depression levels received 8 weekly group sessions of mindfulness based cognitive therapy immediately (n = 12) or after about 7 months of waiting (n = 18). Those who started immediately with the intervention showed a significantly larger reduction in depression levels from pre-treatment to 5 months post-treatment (but non-significant differences for PGD and PTSD levels) compared with people in the waiting list control condition (O'Connor et al., 2014). In an uncontrolled trial, mindfulness-based treatment coincided with significant reductions in depression and PTSD levels (grief reactions were not assessed) from pre- to post-treatment in a treatment-seeking bereaved sample (n = 42; Thieleman et al., 2014).

Trials among people with depressive symptoms have shown that repetitive thinking, including rumination and worry, is one of the most important mechanisms of change in MBIs (see for an overview Gu, Strauss, Bond, & Cavanagh, 2015). Given the assumption that relatives of missing persons are, more than bereaved people, inclined to repetitive negative thinking, adding elements of mindfulness to CBT might be specifically beneficial for relatives of long-term missing persons.

5. OUTLINE OF THE DISSERTATION

Exploration of consequences of, and care after, the disappearance of a significant other was fueled by experiences from relatives of missing persons, researchers, and professionals working with families of missing persons, suggesting that grief following ambiguous loss differs in severity, nature, and treatment from grief following the death of a significant other (Betz & Thorngren, 2006; Boss, 2004, 2006; Giesen, 2003). Yet, empirical evidence related to these claims is limited. We argue that it is important to take further steps in this unexplored field to offer guidelines to professionals supporting relatives of missing persons in order to optimize care for people whose significant other is missing.

In **Chapter 2**, a systematic review is presented in which we summarize empirical studies examining the prevalence and correlates of psychological symptoms in relatives of missing persons. In addition, we explore the empirical evidence related to the claim that the disappearance of a loved one is "the most stressful kind of loss" (Boss, 2006, p. 7) by summarizing the results of studies comparing psychopathology levels between relatives of missing and deceased persons.

Studies comparing psychopathology levels between these groups have so far exclusively focused on losses in the context of war or state terrorism (i.e., armed conflict). In this context, exposure to additional potential traumatic events likely affect psychopathology levels (Johnson & Thompson, 2008). This raises the question to what extent do the results of comparative studies in the context of armed conflict generalize to relatives of missing and deceased persons outside this context? In **Chapter 3**, we address this question by comparing PG and PTS levels between people confronted with the disappearance and homicidal loss of a significant other.

Studies examining correlates of distress post-disappearance that are amendable to change in treatment are lacking. Therefore, we aim to identify correlates of disappearance-related distress that could be target in treatment. We focus on cognitive-behavioural and emotion regulation processes that are assumed to play an important role in the onset and maintenance of distress following the disappearance of a significant other.

The associations between cognitive-behavioural variables and post-loss psychopathology levels have mainly been examined in people confronted with a non-violent (e.g., illness) loss of a loved one. However, a previous study has shown that these cognitive-behavioural variables mediate the effect of violent loss (i.e., accident, homicide, and suicide) on PGD, depression, and PTSD levels (Boelen et al., 2015). In **Chapter 4**, we examine whether cognitive-behavioural variables are also related to distress in relatives of missing persons.

Based on previous findings, indicating that positive affect regulation strategies are linked to depression scores over and above rumination, it has been argued that positive affect regulation strategies are at least as important as ruminating about negative affect in depression (Raes et al., 2009, 2012, 2014). Although the detrimental effect of depressive rumination in bereaved people has repeatedly been shown, the role of positive affect regulation strategies has never been examined. In **Chapter 5**, we explore to what extent positive affect regulation strategies are linked to psychopathology levels following the loss of a loved one, over and above rumination. We examine this in two samples separately: a) a sample of people confronted with the recent death of a significant other, and b) a sample of people confronted with the long-term disappearance of a significant other.

According to the rumination-as-avoidance hypothesis (Stroebe et al., 2007), rumination is viewed as a way of avoiding painful aspects of a loss. Self-compassion, is viewed as a way of being naturally exposed to painful inner experiences (Thompson & Waltz, 2008), and may work as an antidote to rumination. In **Chapter 6**, we test (1) the prediction that greater self-compassion is related to lower PG, PTS, and depression levels in relatives of missing persons and (2) to what extent grief rumination mediates these associations.

Adaptive coping strategies are further explored in an interview-study. During the data-collection phase of the studies included in Chapter 3-6, it was salient that a significant proportion of

relatives of missing persons did not meet criteria for clinically relevant levels of psychopathology. Insights into 1) the course of functioning over time in retrospect, and 2) coping strategies considered helpful by these non-clinical relatives of missing persons are gained in **Chapter 7**.

In **Chapter 8,** it is argued in a study protocol of a pilot randomised controlled trial why CBT with elements of mindfulness may be appropriate for reducing psychological distress among relatives of missing persons in need of support. In **Chapter 9** the feasibility and preliminary effectiveness of this treatment approach is evaluated.

The main findings from the preceding chapters are summarized in **Chapter 10**. The findings are discussed and integrated in relation to bereavement research and beyond. Furthermore limitations, clinical implications, and recommendations for future research are provided.

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2

Toward a better understanding of psychological symptoms in people confronted with the disappearance of a loved one: A systematic review

Lenferink, L.I.M., De Keijser, J., Wessel, I., De Vries, D., & Boelen, P.A. (in press). Toward a better understanding of psychological symptoms in people confronted with the disappearance of a loved one: A systematic review. *Trauma, Violence, & Abuse.* doi: 10.1177/1524838017699602

ABSTRACT

Objective The disappearance of a loved one is claimed to be the most stressful type of loss. The present review explores the empirical evidence relating to this claim. Specifically, it summarizes studies exploring the prevalence and correlates of psychological symptoms in relatives of missing persons, as well as studies comparing levels of psychopathology in relatives of the disappeared and the deceased.

Method Two independent reviewers performed a systematic search in Psychinfo, Web of Science, and Medline, which resulted in 15 studies meeting predefined inclusion criteria. Eligible studies included quantitative peer-reviewed articles and dissertations that assessed psychopathology in relatives of missing persons.

Results All reviewed studies were focused on disappearances due to war or state terrorism. Prevalence rates of psychopathology were mainly described in terms of posttraumatic stress disorder and depression and varied considerably among the studies. Number of experienced traumatic events and kinship to the missing person were identified as correlates of psychopathology. Comparative studies showed that psychopathology levels did not differ between relatives of missing and deceased persons.

Conclusions The small number of studies and the heterogeneity of the studies limits the understanding of psychopathology in those left behind. More knowledge about psychopathology post-disappearance could be gained by expanding the focus of research beyond disappearances due to war or state terrorism.

Keywords: missing persons, trauma, depression, grief, stress

Systematic reviews have shown that the death of a significant other can lead to serious mental health issues, including depression, posttraumatic stress disorder (PTSD), and complicated grief[1](Kristensen, Weisæth, & Heir, 2012; Lobb et al., 2010; van Denderen, de Keijser, Kleen, & Boelen, 2015). One of the risk factors for developing psychological symptoms following the loss of a significant other is the type of loss (Kristensen et al., 2012; van Denderen et al., 2015). Unnatural, sudden, and violent losses, such as homicide and suicide are associated with increased risk of psychopathology (Boelen, de Keijser, & Smid, 2015; Currier, Holland, & Neimeyer, 2006).

A unique type of loss is the disappearance of a loved one, also referred to as an 'ambiguous loss[2]' or 'unconfirmed loss' (Boss, 1976; Powell, Butollo, & Hagl, 2010). Disappearances of persons affect thousands of people around the world yearly, especially in the context of war and/or state terrorism[3] (i.e., acts of cruelty conducted by a state against its own people Aust, 2010, p. 265). A frequently cited assumption (e.g., Betz & Thorngren, 2006; Heeke & Knaevelsrud, 2015) originating from family stress theories (Betz & Thorngren, 2006) and family systems theories (Carroll, Olson, & Buckmiller, 2007), is that "Ambiguous loss is the most stressful loss because it defies resolution and creates confused perceptions about who is in or out of a particular family" (Boss, 2004, p. 553).

Recently, Heeke and Knaevelsrud (2015) presented a brief overview of seven quantitative studies focusing on psychopathology after the disappearance of a loved one due to war and state terrorism. They concluded that: a) PTSD, depression, and complicated grief symptoms are common following the disappearance of a loved one and b) these symptoms are more severe compared to symptoms observed in people confronted with the death of a loved one. However, there are some limitations that preclude firm conclusions. From a methodological perspective, the review may not give a complete and valid overview of the existing literature, because it lacked a systematic approach (e.g., no systematic search strategy, specifics about study selection

1. We use the term "complicated grief" throughout the paper to denote a pattern of adaptation to the death/disappearance of a significant other "that involves the presentation of certain grief-related symptoms at a time beyond that which is considered adaptive" (Lobb et al., 2010, p. 674). See Lobb et al. (2010, p. 674) for examples of grief-related symptoms. In previous studies the terms "prolonged grief disorder" or "traumatic grief" were used interchangeably to refer to complicated grief. In recent literature, grief-related distress may also be referred to as "persistent complex bereavement disorder" in accord with the fifth edition of the Diagnostic Statistical Manual of Mental Disorders (APA, 2013).
2. Boss distinguished two types of ambiguous loss: the first denotes when the loved one is physically present, but psychologically absent (e.g., due to dementia) and the second when the loved one is psychologically present, but physically absent (e.g., when someone is reported as missing). Note that within this review the term ambiguous loss refers to the physical disappearance of a loved one.
3. There is no consensus on the definition of state terrorism (Aust, 2010). Within this paper we use "disappearances due to state terrorism" to refer to disappearances that are probably caused by political repression. Studies included in this review that referred to 'political repression' in their text were referred to as 'state terrorism' in the current review. Studies that referred to 'war' in their text were referred to as 'disappearance due to war' in the current review.

criteria, and quality assessment of the reviewed studies). Furthermore, the evidence does not unequivocally support Heeke and Knaevelsrud's (2015) conclusions. For instance, indices of psychopathology were only significantly higher among relatives of missing persons compared to relatives of deceased persons in three out of five comparative studies (Powell et al., 2010; Quirk & Casco, 1994; Zvizdic & Butollo, 2001). Moreover, in these three studies some but not all indices of psychopathology differed significantly. All in all, Boss' (1976, 2004) claim that the disappearance of a loved one is the most stressful type of loss does not seem to rest on a solid empirical basis.

The current review provides a systematic overview of the scientific research on psychological symptoms in people confronted with the disappearance of a loved one. Our review complements Heeke and Knaevelsrud's (2015) review in that we used a systematic approach, in order to prevent selection bias of the reviewed studies and to guarantee replicability. Given the large number of people who are confronted with a disappearance due to war and state terrorism, it is important to give a systematic overview of current state of the literature regarding psychological symptoms in relatives of missing persons. This may contribute, among others, to 1) knowledge about the nature and severity of psychopathology in relatives of missing persons, 2) the identification of risk factors for psychopathology, and 3) directions for future research. In the following, we address three objectives. First, we aimed to summarize the studies examining prevalence rates of psychological symptoms in relatives of missing persons. Secondly, we sought to describe correlates of psychological symptoms. Our third goal was to enumerate the results of studies exploring differences in severity of psychopathology among relatives of disappeared people compared to relatives of deceased people.

METHOD

Inclusion criteria

Quantitative studies published in peer-reviewed academic journals and dissertations of which the abstract is indexed in scientific literature databases were included. The studies needed to report about psychological symptoms in spouses, family members and/or friends of missing persons. A missing person is defined as: "Anyone whose whereabouts is unknown whatever the circumstances of disappearance. They will be considered missing until located and their well-being or otherwise established" (Association of Chief Police Officers, 2010, p.15).

An article was excluded if it (1) was a qualitative or case study, (2) did not include data of participants (e.g., a literature overview), (3) was focused on participants of whom a relative had returned after a period of disappearance, (4) was focused on ambiguous loss in terms of being physically present but psychologically absent (e.g., dementia patients; cf. Boss, 1976), or (5) concerned relatives of persons whose loved one was absent, but not missing (e.g., foster care). A

protocol of the review can be obtained in the PROSPERO register (Lenferink, de Keijser, Boelen, & Wessel, 2015). The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines were followed (Moher, Liberati, Tetzlaff, Altman, & PRISMA group, 2009).

Search strategy

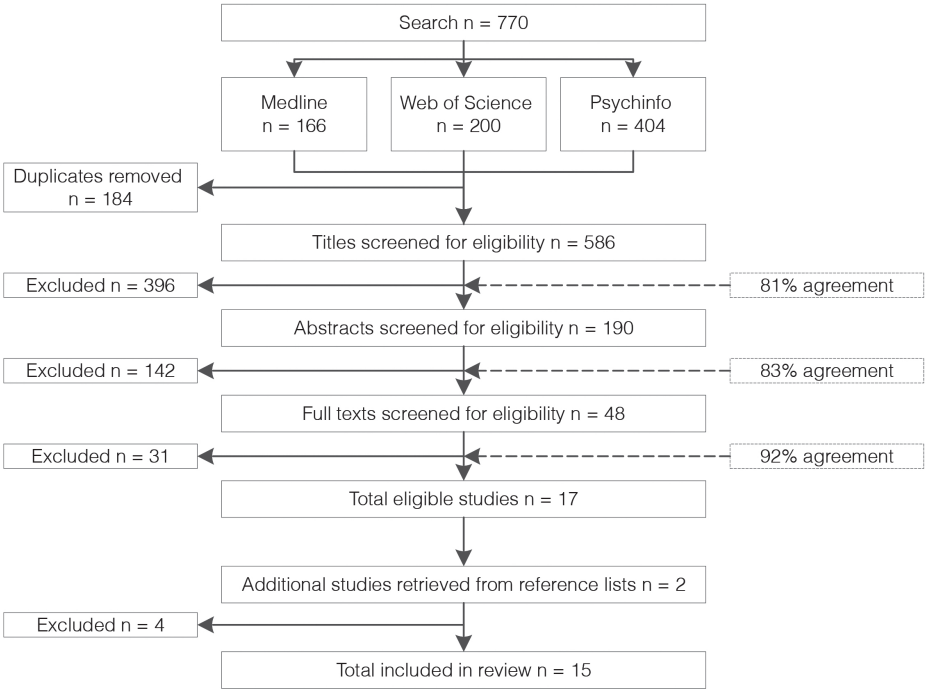
Three topics structured the search terms: 1) missing persons, 2) people who are left behind, and 3) psychological symptoms. Because of the different words that can be used for each of the three search topics we entered multiple search terms (at least 12 per topic) to be as complete as possible. See Appendix Figure A1 for the search terms. Three electronic literature databases (Psychinfo, Web of Science, and Medline) were searched in June 2015. No date or language restrictions were applied in the search strategy.

Study selection

The consecutive steps for the selection of studies that were performed independently by two reviewers are displayed in Figure 1. In sum, the search terms in three databases resulted in 770 hits. After removal of duplicates the remaining articles were screened first by title, second by abstract, and lastly by full-text based upon the in- and exclusion criteria. Finally, the raters screened the reference lists of the eligible studies (n = 17) for additional studies meeting the inclusion criteria. As for interrater reliability, the percentages of absolute agreement between the raters ranged from 81% to 92%. In case of disagreement, consensus was reached through discussion. The databases were again searched in March 2016 for recently added literature, which resulted in zero eligible studies.

Two dissertations (Boss, 1976; Munczek, 1996) initially deemed eligible for inclusion were eventually excluded from the current review, because reading the full-text revealed that they provided the basis for published articles that were already included (Boss, 1977; Munczek & Tuber, 1998). Likewise, one study (Hagl, Rosner, Butollo, & Powell, 2014) was excluded because it appeared to be a clinical trial of which the relevant data were reported in another study (Powell et al., 2010) that was already included in the review. In addition, a language barrier necessitated discarding a study published in Croatian after including it based upon the abstract (Bek, Buzov, & Bilić, 2001).

Figure 1. Study selection



Synthesis of results

First, current and/or lifetime prevalence rates of psychopathology in relatives of missing persons are described for each study. The prevalence rates reflect the percentages of participants scoring beyond the established clinical threshold of the specific measure used. Second, results of correlation analyses or regression analyses, t-, Chi-square and /or F-tests to assess correlates of psychological symptoms are summarized. Third, Cohen's *d* effect sizes were computed for the studies that compared the severity of psychological symptoms between relatives of missing persons and relatives of deceased persons. According to Cohen (1988) effect sizes of $d = 0.2$ to 0.5 are small, $d = 0.5$ to 0.8 medium, and $d = \geq 0.8$ large.

Quality assessment of the included studies

Performing a quality assessment of observational studies in systematic reviews is one of the criteria in the PRISMA guidelines for reporting systematic reviews (Moher et al., 2009). We assessed the quality of the included studies using the Systematic Assessment of Quality in Observational Research (SAQOR; Ross et al., 2011), a checklist that is developed for (Ross et al., 2011) and previously used in quality assessment of psychiatric observational studies (e.g., Kohrt et al., 2014).

The SAQOR evaluates the quality of studies based on meeting criteria in six domains: (i) Sample, (ii) Control/Comparison Group, (iii) Quality of Exposure/Outcome Measurements, (iv) Follow-Up, (v) Distorting Influences, and (vi) Reporting Data. Each domain consists of multiple criteria. The domain is rated as "adequate", "inadequate", "unclear", or "not applicable" based on the frequency of fulfilled criteria of the subsequent domain. An overall quality rating – high, moderate, low, or very low - of each study is then determined based on frequency of adequate domains. As recommended by the authors of the tool (Ross et al., 2011) and consistent with authors using it (e.g., Kohrt et al., 2014) we slightly adapted the SAQOR to fit our specific population (see Appendix B for more details).

RESULTS

Quality assessment

Three studies met SAQOR criteria for high quality (Heeke, Stammel, & Knaevelsrud, 2015; Pérez-Sales, Durán- Pérez, & Herzfeld, 2000; Zvidic & Butollo, 2001), six studies for moderate quality (Baraković, Avdibegović, & Sinanović, 2013, 2014; Campbell & Demi, 2000; Navia & Ossa, 2003; Powell et al., 2010; Reisman, 2003), two studies for low quality (Basharat, Zubair, & Mujeeb, 2014; Munczek & Tuber, 1998), and four studies for very low quality (Boss, 1977, 1980; Clark, 2001; Quirk & Casco, 1994). See Appendix B for more details about the quality assessment of the 15 studies. The four very low quality studies are not addressed in the following.

Characteristics of the included studies

All 11 low to high quality studies used a cross-sectional design. The sample sizes varied from 14 to 225 relatives of missing persons. Two studies (Baraković et al., 2013, 2014) relied on a sample of women (n = 120) whose male family member disappeared during the war in Bosnia Herzegovina 15 to 18 years earlier. Another sample included women from Bosnia Herzegovina (n = 56) whose husbands disappeared during the war on average 7 years earlier (Powell et al., 2010). A fourth study included a sample of adolescents (n = 201) whose fathers had been missing since 3 to 4 years in the context of war in Bosnia Herzegovina (Zvizdic & Butollo, 2001). Two studies were conducted in the United States of America among a small sample of family members of men listed as Missing-In-Action (MIA; Campbell & Demi, 2000) (n = 20) or Prisoner Of War (POW; Reisman, 2003) (n = 14) for over 25 years ago. Two studies were executed among relatives of disappeared persons in Colombia; one due to state terrorism 13 years earlier (n = 73; Heeke et al., 2015) and another due to economic extortive kidnapping (n = 46; Navia & Ossa, 2003). Navia and Ossa (2003) did not provide information on the amount of time that had passed since the disappearance. In Chile, Pérez-Sales et al. (2000) studied people (n = 75) whom a relative disappeared due to state terrorism more than

20 years earlier. Children (*n* = 16) whose fathers disappeared 9 years ago, on average, due to state terrorism were the subjects of a study in Honduras (Munczek & Tuber, 1998). The last study was focused on Pakistani family members (*n* = 225) who lived for 1 to 9 years with the disappearance of a loved one due to state terrorism (Basharat et al., 2014).

Prevalence of psychological symptoms (objective 1)

In four unique samples, described in five articles, prevalence rates of psychological symptoms were reported (Baraković et al., 2013, 2014; Heeke et al., 2015; Navia & Ossa, 2003; Pérez-Sales et al., 2000). Due to the heterogeneity of the studies (e.g., studies varied in terms of instruments used to assess symptoms and in terms of ethnic background of study samples), the prevalence rates of psychological symptoms are reported separately for each study. See Table 1 for a summary of the characteristics and the main findings of the studies.

The two studies of Baraković et al. (2013, 2014) relied on the same sample and showed current self-rated prevalence rates of 88% for depression, 65% for mild to severe anxiety complaints, 56% for PTSD, and 43% for somatic complaints. Heeke et al.'s (2015) study showed that 69% reported current depression, 67% PTSD, and 23% complicated grief based on interviews. Interview-based current prevalence rate of PTSD was 39% in a study of Navia and Ossa (2003). A final study reported considerably lower interview-based current and lifetime prevalence rates of PTSD (1%/3%), depression (3%/17%), anxiety (1%/5%), and complicated grief (7%/27%) (Pérez-Sales et al., 2000).

Table 1. The characteristics of the studies

Citation and quality	Country of study	Sample description	Outcomes of interest	Measures	Prevalence rates of psychopathology	Correlates of psychological symptoms
Baraković et al., 2013 Moderate	Bosnia Herzegovina	120 women with and 40 without a missing family member due to war 15-18 years earlier	Depression; Anxiety; Somatic symptoms	Beck Depression Inventory; Hamilton Anxiety Rating Scale; Somatic Symptom Index	88% current mild - severe depression; 65% current mild to severe anxiety symptoms	Women with a missing son experienced the most severe depression, anxiety, and somatic symptoms compared to women with a missing husband, father, or brother
Baraković et al., 2014 Moderate	Bosnia Herzegovina	See Baraković et al., (2013)	Anxiety; Depression; PTSD	Hamilton Anxiety Rating Scale; Beck Depression Inventory; Harvard Trauma Questionnaire	56% current PTSD	Women with a missing son or husband experienced more severe PTSD symptoms compared to women with a missing father or brother; number of experienced traumatic events was significantly associated with increased levels of PTSD, depression, and anxiety
Basharat et al., 2014 Low	Pakistan	225 persons with a missing family member due to state terrorism 1-9 years earlier	Anxiety; Depression; Stress; Coping strategies	Depression Anxiety and Stress Scale; Brief COPE	-	Being female, older, a spouse of a missing person, and inclined to use emotion-focused coping strategies were associated with increased levels of depression, anxiety, and stress; subjects whose loved one disappeared 1-3 years ago were significantly more distressed than those whose loved one disappeared 3 to 6 years or 6 to 9 years ago, but the latter group was significantly more distressed than those whose loved one disappeared 3.1 to 6 years ago

Table 1 (continued). The characteristics of the studies

Citation and quality	Country of study	Sample description	Outcomes of interest	Measures	Prevalence rates of psychopathology	Correlates of psychological symptoms
Campbell & Demi, 2000 Moderate	United States of America	20 adult children of men listed as MIA over 25 years earlier	PTSD; Complicated grief; Family functioning	Impact of Event Scale; Bereavement Experience Questionnaire Short Form; Family Hardiness Index	-	Gender and age were not associated with complicated grief and PTSD; sense of family 'control' and 'commitment' were negatively associated with PTSD avoidance; sense of family 'control' and 'challenge' were negatively associated with complicated grief
Heeke et al., 2015 High	Colombia	73 family members/friends of disappeared persons on average 13.4 (*SD* = 6.9) years earlier and 222 family members/friends of killed persons on average 12.1 (*SD* = 7.3) years earlier due to state terrorism	Depression; Extent of hope; Complicated grief PTSD	Hopkins Symptom Checklist - Depression subscale; Single item developed; Clinical Structured Interview for Prolonged Grief Disorder; PTSD Checklist - civilian version	69% current depression; 67% current PTSD; 23% current complicated grief	Gender, age, years of education, and time since loss were not associated with complicated grief; number of experienced traumatic events was positively associated with complicated grief, the association disappeared when partialling out PTSD and depression; extent of hope that the missing loved one is still alive explained unique proportion of the variance in complicated grief

Table 1 (continued). The characteristics of the studies

Munczek & Tuber, 1998 Low	Honduras	16 children whose fathers disappeared on average 112 months earlier and 11 children whose fathers were killed on average 49 months earlier in the context of state terrorism	PTSD; Depression; Anxiety	Post-traumatic stress reaction checklist child version; Child Behavior Inventory	-	-
Navia & Ossa, 2003 Moderate	Colombia	46 family members of victims of economic extortive kidnapping for unknown duration and 113 whose relative was released for 2-15 months	Family coping strategies; Family functioning; General psychological distress; PTSD	Family Coping Oriented Personal Evaluation Scale; Family Assessment Device; Global Severity Index of the Symptom Checklist-90-R; Clinician Administered PTSD Scale-DX	39% current PTSD	Family coping strategies (e.g., seeking spiritual support and avoidance) were not associated with PTSD and general psychological distress. Three aspects of family functioning (family roles, behavior control, and general family functioning) were positively associated with general psychological distress

Table 1 (continued). The characteristics of the studies

Citation and quality	Country of study	Sample description	Outcomes of interest	Measures	Prevalence rates of psychopathology	Correlates of psychological symptoms
Pérez-Sales et al., 2000 High	Chile	75 family members of enforced disappeared persons and 44 family members of persons killed in the context of state terrorism more than 20 years earlier	PTSD; Depression; Complicated grief; Anxiety disorders	Psychiatric State Examination (10th edition)	1%/5% current and lifetime anxiety disorders; 3%/17% current and lifetime depression; 7%/27% current and lifetime complicated grief; 1%/3% current and lifetime PTSD	-
Powell et al., 2010 Moderate	Bosnia Herzegovina	56 women whose husband disappeared and 56 whose husband were killed in war on average 7.4 years earlier	General psychological distress; Complicated grief; PTSD	General Health Questionnaire subscales somatic symptoms, anxiety, insomnia, social dysfunction, and, depression; UCLA Grief Inventory; Impact of Event Scale	-	Not prewar or wartime stressors, but postwar stressors were uniquely associated with complicated grief and depression next to type of loss
Reisman, 2003 Moderate	United States of America	14 adult children of men listed as MIA/POW and 70 adult children of men listed as KIA over 25 years earlier	PTSD	Impact of Event Scale	-	-

Table 1 (continued). The characteristics of the studies

Zvidic & Butollo, 2001 High	Bosnia Herzegovina	201 adolescents whose father disappeared, 208 whose father was killed, and 407 adolescents of the control group all in the context of war 3-4 years earlier	Depression	Birleson depression scale for children	-	Number of experienced traumatic events was associated with increased depression levels

Note. In the third column of the table, time since disappearance is reported as was done in the respective studies; Not all studies reported mean and standard deviation (SD) of the time since disappearance; MIA = Missing In Action; POW = Prisoners Of War; KIA = Killed In Action; PTSD = posttraumatic stress disorder; - = not applicable, because the study did not report prevalence rates (based on established criteria) or correlates of psychopathology.

Correlates of psychological symptoms among relatives of the disappeared (objective 2)

Gender. Three studies examined whether psychopathology levels varied as a function of gender (Basharat et al., 2014; Campbell & Demi, 2000; Heeke et al., 2015). To begin with, Basharat et al. (2014) found that females were significantly more stressed (d = 0.41), depressed (d = 0.52), and anxious (d = 0.38) than males. A second study reported that gender was not significantly associated with complicated grief severity (Heeke et al., 2015). A third study reported that gender was unrelated to severity of complicated grief and PTSD (Campbell & Demi, 2000).

Age. Three studies explored the association between age and psychological symptoms (Basharat et al., 2014; Campbell & Demi, 2000; Heeke et al., 2015). The first showed that older participants were significantly more generally distressed than younger participants (Basharat et al., 2014). Two further studies reported non-significant associations between age and psychopathology in terms of complicated grief and PTSD (Campbell & Demi, 2000; Heeke et al., 2015).

Kinship. In two samples the difference in severity of psychopathology according to type of kinship was examined (Baraković et al., 2013, 2014; Basharat et al., 2014). Women with a missing son experienced significantly higher levels of PTSD, depressive, anxiety, and somatic symptoms compared to women with a missing husband, brother, or father (Baraković et al., 2013, 2014). Basharat et al.'s (2014) study showed that spouses were significantly more distressed than parents and siblings. In addition, parents were significantly more distressed than siblings.

Time since disappearance. The association between time since disappearance and psychopathology was studied twice (Basharat et al., 2014; Heeke et al., 2015). Basharat et al. (2014) showed that participants whose loved one disappeared 1-3 years earlier reported significantly higher levels of general distress than those whose loved one disappeared 3 to 9 years ago. However, in the same study, participants whose loved one disappeared 6 to 9 years ago experienced significantly higher levels of general distress than participants whose loved one disappeared 3 to 6 years ago. Heeke et al.'s (2015) study reported a negative correlation (r = -.31, p < .01) between time since disappearance (in months) and complicated grief severity, but time since disappearance was not a significant predictor of complicated grief after controlling for other variables (e.g., depression).

Educational level. The association between educational level and psychopathology was only examined by Heeke et al. (2015). They found education (in terms of number of years of education) to be unrelated to complicated grief.

Number of experienced traumatic events. Four studies assessed the number of traumatic events the relatives of missing persons had been exposed to (Baraković et al., 2014; Heeke et al., 2015; Powell et al., 2010; Zvizdic & Butollo, 2001). Baraković et al. (2014) found an increase in the number of experienced traumatic events to be associated with increased levels of depression (r

= .61, $p < 0.001$), PTSD ($r = .58$, $p < 0.001$), and anxiety ($r = .44$, $p < 0.001$). Zvizdic and Butollo (2001) found that more exposure to several war-related events (e.g., loss of home) and postwar-related events (e.g., family problems) were both also associated with depression ($r = .26$, $p < .01$ and $r = .26$, $p < .01$). A third study performed 11 regression analyses with type of loss (disappearance versus death), number of prewar, wartime, and postwar stressors as predictors and several psychological symptoms as outcome variables. Number of postwar, but not prewar or wartime stressors were associated with one of the complicated grief subscales (defined as traumatic grief) ($t = 3.03$, $p < .01$) and depression ($t = 2.37$, $p < .01$) next to type of loss (Powell et al., 2010). Finally, Heeke et al.'s (2015) study showed that an increase in the number of experienced traumatic events was associated with complicated grief ($r = .30$, $p < .01$); this association disappeared, however, when gender, severity of PTSD, and depression were partialled out.

Family functioning. Two studies examined the association between perceived functioning of the family and psychopathology (Campbell & Demi, 2000; Navia & Ossa, 2003). Navia and Ossa (2003) found no significant associations between five family coping strategies (e.g., seeking spiritual support and passive appraisal) and PTSD and general psychological distress in a subgroup ($n = 18$) of their sample. This subgroup included one family member per missing person in order to handle the within-family clustering effect. In addition, within the same subgroup of their sample a significant association was found between general psychological distress and aspects of family functioning (i.e., family roles ($r = .52$, $p < .05$, i.e., the way in which family members allocate responsibilities), behavior control ($r = .52$, $p < .05$, i.e., the way in which families provide clear standards and rules of behavior), and general family functioning ($r = .50$, $p < .05$)). A higher score on family functioning was indicative of unhealthier family functioning. No significant association was found between PTSD and family functioning. Campbell and Demi (2000) reported that individuals experienced less PTSD symptoms of the avoidance cluster when they felt their family was cooperative in solving problems ($r = -.47$, $p < .05$) and is in control over dealing with adverse life events ($r = -.49$, $p < .05$). Furthermore, individuals reported less complicated grief when they viewed their families as active in managing challenging situations ($r = -.50$, $p < .05$) and in control over dealing with these situations ($r = -.62$, $p < .05$).

Coping strategies. The association between use of coping strategies and psychopathology was examined in only one study (Basharat et al., 2014). That study showed that greater use of emotion-focused coping strategies (e.g., seeking sympathy from others) was associated with increased levels of depression ($r = .38$, $p < .001$), anxiety ($r = .24$, $p < .001$), and stress ($r = .41$, $p < .001$). Greater use of problem-focused coping strategies (e.g., thinking about dealing with the problem) was associated with decreased levels of depression ($r = -.48$, $p < .001$), anxiety ($r = -.35$, $p < .001$), and stress ($r = -.26$, $p < .01$).

Extent of hope that the missing person is alive. The association between extent of hope that the loved one is still alive and psychopathology was explored in one study. Extent of hope explained 5% of the variance in complicated grief above gender, depression and PTSD severity, number of experienced traumatic events, and time since disappearance (Heeke et al., 2015).

Summary of the results of correlational studies. No clear conclusions can be drawn about the association between psychopathology and gender or age. The results with respect to the association between kinship and psychopathology were consistent across two samples. More specifically, spouses and parents of missing persons seem to be the most affected compared to siblings. Based on one study, years of education was unrelated to complicated grief. The two studies that explored the association between time since disappearance and symptom severity yielded contrasting results. The results of four studies indicated that number of traumatic events that people had been exposed to was significantly associated with increased levels of psychopathology. Two studies assessed the association between family functioning and psychological symptoms and found inconsistent results. Using emotion-focused coping strategies more frequently and problem-focused less frequently coping strategies were related to higher psychopathology levels based on a single study. One study showed a positive association between extent of hope that the loved one is still alive and psychopathology.

Disappearance versus death (objective 3)

Six studies, all cross-sectional studies, compared relatives of disappeared to deceased persons, all in the context of state terrorism or war. Homicide was the cause of death in all cases, with an exception of the study of Heeke et al. (2015) of which 93% of the deceased persons were victims of homicide and the other 7% died due to another reason related to state terrorism (e.g., illness). A summary of the studies is offered in Table 2.

PTSD. In four studies the severity or prevalence of PTSD symptoms did not differ significantly between relatives of victims of disappearance to relatives of homicide victims (Heeke et al., 2015; Munczek & Tuber, 1998; Powell et al., 2010; Reisman, 2003). A fifth study showed that lifetime prevalence rate of PTSD since the loss was significantly higher among homicidally bereaved individuals (9.0%) than individuals confronted with the disappearance of a family member (1.3%) (Pérez-Sales et al., 2000).

Depression. One study showed that spouses of men who disappeared had significantly more severe depressive symptoms than homicidally bereaved spouses (Powell et al., 2010). This difference had a medium effect size (d = 0.67). Another study reported that those of whom a loved one disappeared also experienced significantly higher depression levels than homicidally bereaved individuals (d = 0.22) (Zvizdic & Butollo, 2001). Three other studies did not find significant

differences in severity or prevalence of depression between relatives of victims of disappearance or homicide (Heeke et al., 2015; Munczek & Tuber, 1998; Pérez-Sales et al., 2000).

Anxiety. Anxiety symptom severity did not differ significantly between children or spouses of men who were victims of disappearance or homicide (Munczek & Tuber, 1998; Powell et al., 2010). Lifetime prevalence rates of anxiety disorders since the loss was significantly higher for the relatives of the disappeared (5.3%) compared to the homicidally bereaved (0.0%) (Pérez-Sales et al., 2000).

Complicated grief. Women whose husbands disappeared reported significantly higher levels of "traumatic grief" compared to homicidally bereaved women (Powell et al., 2010). This difference can be interpreted as a medium effect (d = .79). Within the same study no significant differences were found between groups in terms of "existential grief" (Powell et al., 2010). It remains unclear in this paper how traumatic and existential grief were defined and how they differed from each other. A second study also failed to show significant differences in severity or prevalence of complicated grief between relatives of the disappeared compared to homicidally bereaved family relatives (Heeke et al., 2015). Pérez-Sales et al. (2000) reported non-significant differences in prevalence rates of complicated grief between family members of persons who disappeared or were victims of homicide.

Social dysfunction and somatic symptoms. One study examined the differences in severity of social dysfunction and somatic symptoms between wives whose husbands disappeared or were victims of homicide. No significant differences were shown for the two groups (Powell et al., 2010).

Summary of the results of comparative studies. In sum, results across six studies, conducted in the context of war or state terrorism, indicated that relatives of disappeared persons and relatives of homicide victims overall did not significantly differ in severity or prevalence rate of psychopathology. Only four out of 24 comparisons across 6 studies yielded significantly higher levels of severity or prevalence of psychological symptoms in the disappeared group compared to the homicide group and these differences had a small to medium effect size (see Table 2).

Table 2. Effect size estimates for differences in psychological symptoms between relatives of victims of disappearance and homicide

Citation	Description of the sample of relatives of enforced disappeared persons	Description of the comparison group of homicidally bereaved relatives	Outcomes	Significantly higher levels of psychopathology for disappeared group	Effect size *(Cohen's d)*	Significantly higher current/ lifetime prevalence rates for disappeared group
Heeke et al., 2015	73 family members and friends of enforced disappeared persons	222 family members and friends of deceased persons (93% were homicidally bereaved)	Depression	N	-0.02	N
			PTSD	N	0.01	N
			Complicated grief	N	0.07	N
Munczek & Tuber, 1998	16 children of enforced disappeared fathers	11 children whose father was a homicide victim	Depression	N	-	
			PTSD	N	-	
			Anxiety	N	-	
Pérez-Sales et al., 2000	75 family members of disappeared persons	44 family members of homicide victims	Depression		-	N/N
			PTSD		-	N/N
			Complicated grief		-	N/N
			Anxiety disorders		-	N/Y
Powell et al., 2010	56 female spouses of victims of disappearance	56 female spouses of homicide victims	Depression	Y	0.67	
			PTSD	N	0.08	
			Normal grief	N	0.17	
			Traumatic grief	Y	0.79	
			Existential grief	N	0.27	
			Social dysfunction	N	0.22	
			Somatic symptoms	N	-0.05	
			Anxiety and insomnia	N	0.13	
Reisman, 2003	14 adult children of men listed as MIA/POW	70 adult children of men listed as KIA	PTSD	N	0.21	

Table 2 (continued). Effect size estimates for differences in psychological symptoms between relatives of victims of disappearance and homicide

Zvidic & Butollo, 2001	201 adolescents whose father disappeared	208 adolescents whose father was a homicide victim	Depression	Y	0.22

Note. N = no; Y = yes; - = the study did not report means, standard deviations, and/or other estimates that are necessary to compute the effect size; empty cells represent studies that did not compare mean scores or prevalence rates of psychopathology between the two groups; MIA = missing-in-action; POW = prisoner of war; KIA = killed-in-action; PTSD = posttraumatic stress disorder.

DISCUSSION

The present study reviewed research relevant to Boss' (2004) frequently cited claim that the disappearance of a loved one is more traumatic than other types of loss. The first aim was summarizing the results regarding prevalence rates of psychological symptoms in relatives of missing persons. Prevalence rates of psychopathology were mainly described in terms of PTSD and depression and varied widely across a small number of studies (i.e., 1 to 67% for PTSD, 3 to 88% for depression, 1 to 65% for anxiety, 7 to 23% for complicated grief, and 43% for somatic complaints). Several things stand out from this summary of prevalence rates. Only five out of 11 studies assessed prevalence rates of psychological symptoms. Furthermore, these five studies varied in (a) instruments used to assess psychopathology (e.g., none of the studies used the same instrument), (b) ethnic background of study samples, (c) composition of study samples (e.g., female family members or children of missing persons and relatives of missing persons due to war versus due to state terrorism), and (d) methodological quality. Therefore, no general conclusive statements can be made about which psychological symptoms are most common among relatives of missing persons.

The second aim was exploring correlates of psychological symptoms in relatives of missing persons. Compared to other background characteristics, the type of kinship as correlate of psychopathology yielded the most consistent results. Spouses and parents of missing persons seem to be the most affected compared to siblings. However, only two studies explored the association between type of kinship and psychopathology. The association between other demographic variables – gender, age, educational level, and time since disappearance – and psychopathology was conflicting. The results of these correlational studies are roughly in accord with earlier studies among bereaved individuals. Reviews (Kristensen et al., 2012; Lobb et al., 2010; Stroebe & Schut, 2001) on grief-related distress indicate that the relationship between gender, educational level, age, and time since loss on the one hand and psychopathology on the other hand is conflicting. The relationship between kinship to the deceased and psychopathology is more consistent and indicates that parents and spouses are most strongly affected by a loss.

It should be noted that our systematic review does not warrant drawing firm conclusions about the association between background characteristics and psychopathology because, taken together, the evidence is not sufficient. The composition of the majority of the study samples precluded the examination of the association between some of the background characteristics and psychopathology. For example, some samples consisted of solely females, which precluded studying the association between psychopathology and gender (Baraković et al., 2013, 2014; Powell et al., 2010). Others only included one type of kinship (e.g., children of missing persons) and therefore did not allow study the potential differences in severity of psychopathology across

different familial relationships (Campbell & Demi, 2000; Munczek & Tuber 1998; Powell et al., 2010; Reisman, 2003; Zvidic & Butollo, 2001).

Furthermore, four studies (Baraković et al., 2014; Heeke et al., 2015; Powell et al., 2010; Zvizdic & Butollo, 2001) yielded evidence that the number of traumatic events participants had experienced is associated with increased levels of psychological symptoms. This is in line with previous trauma research indicating that cumulative trauma heightens the risk of development of PTSD (e.g., Johnson & Thompson, 2008; Kolassa et al., 2010; Wilker et al., 2015). More specifically, several studies showed that being exposed to traumatic events and the loss of a loved one heightens the risk of development of complex psychopathological symptom patterns characterized by comorbid symptoms of complicated grief, PTSD, and depression (cf. Mutabaruka, Séjourné, Bui, Birmes, Chabrol, 2012; Nickerson et al., 2014). Relatives of missing persons with comorbid symptoms may benefit most from interventions targeting different types of symptoms, such as brief eclectic psychotherapy and cognitive behavioral therapy (cf. Smid et al., 2015).

It has been postulated that holding on to hope for the return of the missing loved one may be associated with less emotional distress (Wayland, Maple, McKay, & Glassock, 2016). However, inconsistent with this notion, the single empirical study (Heeke et al., 2015) examining this linkage actually showed that the extent of hope that the loved one was still alive was associated with higher levels of complicated grief. The present review revealed mixed results regarding the use of coping strategies and family functioning as correlates of psychopathology. No further conclusions can be drawn about to what extent psychopathology is related to these constructs, since these potential associations were only explored in three studies, which yielded inconclusive results, and differed from each other in many ways (e.g., sample composition and measures used).

The third aim of the present systematic review was exploring whether the disappearance of a loved one is associated with more severe distress compared to the death of loved one (Boss, 2004). The claim that people who experience an ambiguous loss suffer from more severe psychological symptoms than individuals who experience another type of loss does not seem to be supported by the comparative studies reviewed here. Only four out of in total 24 comparisons across six comparative studies revealed that relatives of missing persons suffered from significantly more severe psychological symptoms compared to homicidally bereaved individuals. This is in contrast with the conclusion drawn in Heeke and Knaevelsrud's (2015) overview. Although there is overlap in the reviewed studies of the overview of Heeke and Knaevelsrud (2015) and our review, incompleteness of this former overview and different weighing of the empirical evidence may explain the difference in conclusion drawn. For instance, Heeke and Knaevelsrud (2015) omitted some comparative studies in their review (e.g. Munczek & Tuber, 1998). In addition, they elaborated on significant findings of a study of Quirk and Casco (1994) that we rated as very low quality and was therefore not discussed.

It is possible that methodological drawbacks of the reviewed studies are responsible for the lack of differences in psychopathology between relatives of the disappeared and the deceased. One of the major methodological shortcomings was the small sample size of the studies, increasing the risk of Type II error. Furthermore, all comparative studies were conducted in the context of war or state terrorism. Both groups were traumatized by different stressors, which might have made it difficult to distinguish between the effect of the loss of a loved one and the effects of other traumatic experiences. Lastly, we believe it is important to emphasize that studies conducted in the context of war-related disappearances may not be comparable to disappearances in the context of state terrorism. Our conclusion therefore needs to be interpreted with caution.

To the best of our knowledge, the current study is the first providing a systematic review of empirical findings about the nature and severity of psychological symptoms in relatives of the disappeared. However, this present review has primarily revealed gaps in the literature on psychological symptoms in relatives of missing persons. The field of psychological research in relatives of missing persons could benefit from at least two types of improvements: 1) expanding the focus of research to comparing relatives of missing persons in the context of war and state terrorism as well as focusing on relatives of missing persons outside the context of war and state terrorism and 2) employing more rigorous research methods.

Expanding the focus of research

The results of our systematic review indicate that with no exception, the existing literature is focused on disappearances in the context of armed conflict (e.g., war or state terrorism). It would be interesting to study to what extent the experience of the disappearance of a loved one due to war differs from the disappearance due to state terrorism. Loosing a significant other in war due to lack of protection by the state may not have the similar impact as experiencing the disappearance of a significant other due to violations conducted by the state. In the latter case, it is possibly more likely that the missing person is still alive, but those left behind may avoid searching for the missing person out of fear for prosecutions or reprisals. In addition, professional support may be less available in the case of disappearances due to state terrorism because, among others, professionals are at risk to become a victim of state terrorism as well. Enhancing knowledge about the similarities and differences in psychological consequences but also assessments of needs and barriers to care for relatives of missing persons in the context of war and state terrorism may be relevant for developing support tailored to the needs of people with different types of enforced disappeared relatives.

Disappearances outside the context of war and state terrorism may occur in a wide range of circumstances varying in intentionality (e.g., deliberately leaving without informing relatives versus being murdered and hidden, or accidentally drowning) (Biehal, Mitchell, & Wade, 2003). As an

example, consider the incidence rate of persons reported as missing to the Australian police. This rate (1.55 per thousand yearly) is higher than the number of reports of sexual assault, homicide, and unarmed robbery combined (Henderson, Henderson, & Kiernan, 2000). Expanding the focus of research from disappearances due to war or state terrorism to other types of disappearances is of interest for at least two reasons.

First, the type of disappearance might be a risk factor for the development of psychopathology, as the type of death is a risk factor for the development of psychological symptoms in the bereaved (Lobb et al., 2010). For example, violent deaths are associated with increased complicated grief symptoms (Kaltman & Bonnano, 2003; Lobb et al., 2010). This might also be the case for disappearances that are probably caused by an act of violence (e.g., kidnapping) compared to accidental disappearances (e.g., drowning accident). Enhancing our knowledge about severity and correlates of psychopathology related to different types of disappearances may facilitate identifying people at risk for developing post-disappearance psychopathology.

Second, studying post-disappearance psychopathology outside of the context of war or state terrorism could verify to what extent the results of our review are applicable to relatives of missing persons in general. With the exception of studies that are focused on relatives of men listed as MIA/POW, inherent to the studies about relatives of enforced disappearances due to war or state terrorism are the confounding effects of the: 1) exposure to additional potential traumatic events (e.g., torture and death of family members) and 2) ethnic background of the participants. It has been repeatedly shown that cumulative trauma heightened the risk of development of psychopathology (e.g., Johnson & Thompson, 2008). Previous research also support that intensity of grief-related distress differs as function of ethnic background (Laurie & Neimeyer, 2008; Oltjenbruns, 1998). Studies in Western societies outside the context of war and state terrorism could explore to what extent the results of our review are applicable to relatives of missing persons without the confounding effects of cumulative trauma and ethnic background.

Methodological improvements for future research

Our review revealed several methodological drawbacks in the literature. First, only 5 out of 11 studies assessed prevalence rates of psychopathology. Other studies did assess psychopathology but used non-validated measures and/or measures without established cut-off criteria for determining “caseness” of psychological disorders (e.g., Munczek & Tuber, 1998). Future research should use validated measures, preferably clinical structural interviews, for assessing the severity of psychopathology among those left behind in order to gain more insights into the extent of the mental health issues. Noteworthy is that the majority of the studies reporting interview-based prevalence rates focused on PTSD and depression. Prevalence rates of complicated grief, conceptualized and measured in different ways, were only studied twice (Heeke et al., 2015; Pérez-

Sales et al., 2000). Two other studies (Campbell & Demi, 2000; Powell et al., 2010) did use a measure assessing grief manifestations, but these measures were not validated or lacked an established cut-off. Likewise, studies including measures of complicated grief are not self-evident in the literature about psychopathology following the death of a loved one (Kristensen et al., 2012; van Denderen et al., 2015). It has been repeatedly shown that complicated grief shows overlap with, yet is distinguishable from depression and anxiety (e.g., Boelen & van den Bout, 2005; Prigerson et al., 1996). In order to encourage further research into complicated grief, Persistent Complex Bereavement Disorder (PCBD) has been included as condition for further study in the DSM-5 (APA, 2013). Expanding the study of PCBD to relatives of missing persons might yield more insight into normal and disturbed grief processes of individuals who experience this unique type of loss.

A second methodological drawback of existing studies focused on psychopathology in relatives of missing persons is that only one study examined multiple potential correlates of psychopathology in relatives of missing persons simultaneously in one regression model (Heeke et al., 2015). Results of zero-order correlational analyses might be spurious due to confounding effect of other variables. Two other studies also performed regression analysis but also included participants of the comparison group in the analysis and as a result these analyses do not reflect the unique effect of the disappearance (Baraković et al., 2014; Powell, et al., 2010). More research that examines potential associated variables of psychopathology simultaneously is needed to gain more insights into psychological variables underlying the occurrence and maintenance of psychopathology. Up until now, correlational studies primarily focus on the association between psychological symptoms and background characteristics and number of experienced traumatic events. Theories about complicated grief and PTSD following the death of a loved one highlight the importance of cognitive and behavioral variables, as well as attachment styles, in the development and persistence of complicated grief and PTSD symptoms (e.g., Boelen, van den Hout, & van den Bout, 2006; Ehlers & Clark, 2000; Maccallum & Bryant, 2013; Shear & Shair, 2005). It would be interesting to study whether these variables are also associated with psychological symptoms following the disappearance of a loved one. This may help to identify individuals at risk for development of psychological symptoms following the disappearance of a loved one and to develop interventions to target these variables in treatment.

Third, all reviewed studies used a cross-sectional design. Longitudinal studies are needed to gain knowledge about prospective risk factors for the development and maintenance of psychopathology in relatives of missing persons.

Fourth, although most of the reviewed studies consisted of clustered data (i.e., multiple relatives of the same missing person) none of the studies used statistical techniques that could handle this clustering. Ignoring the nested structure of the data heightens the risk of spurious significant results. Using multilevel modelling could solve this problem in future research.

Fifth, the results of the comparative studies are highly likely biased due to again a lack of use of validated measures and solely use of mainly non-probability samples that consisted of relatives of enforced disappearances that were exposed to additional traumatic events that might interact with psychopathology. Moreover, due to non-probability sampling methods the generalizability of the results to relatives of missing person within the same context is also limited. Comparative studies using probability sampling methods outside the context of war and state terrorism could give more insights into the unique potential differential effect of the disappearance and the death of a loved one on severity and correlates of psychological symptoms in those left behind.

Sixth, multiple studies were excluded from the current review because they did not distinguish between relatives of missing persons and relatives of formerly missing persons (e.g., Greif & Hegar, 1991) or individual confronted with other potential traumatic experiences (e.g., Al Obaidi & Atallah, 2009). Future research could benefit from subgroup-analyses to explore the potential differential effect of the disappearance of a loved one.

Conclusion

The small number of studies and heterogeneity of the studies limits understanding of the complex and unique experience of those left behind after the disappearance of a loved one. However, based on the findings of our review we conclude (with caution) that: 1) prevalence rates of psychopathology, mainly described in terms of PTSD and depression, varied considerably among the studies, 2) spouses, parents, and those who are more exposed to additional traumatic events are vulnerable for the development of psychological symptoms post-disappearance of a loved one, and 3) the severity of these symptoms does not significantly differ from homicidally bereaved individuals in the context of war and state terrorism. Researchers are challenged to further explore this under-researched field to gain more insights in the nature, prevalence, and correlates of psychopathology in relatives of missing persons.

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ACKNOWLEDGEMENTS

We would like to thank MSc. Sander Sprik for his assistance with the development of the search strategy, MA. Liesbeth E. A. Rischen-Sindram for translating a French article, and Marjolijn Dijkhuis for translating a Spanish article.

PROSPERO registration number CRD42015016757

APPENDIX A

Figure A1. Search terms

("missing family member*" OR "missing father*" OR "missing mother*" OR "missing people" OR "gone missing" OR "missing child*" OR "missing person*" OR "missing in action" OR "people go missing" OR "unconfirmed loss*" OR "ambiguous loss" OR "kidnap*" OR "forced disappearance" OR "disappeared person*") AND ("families" OR "family" OR "child*" OR "relative" OR "relatives" OR "wife" OR "wives" OR "husband*" OR "spouse*" OR "parent*"OR "son*" OR "daughter*") AND ("mental health" OR "stress*" OR "trauma*" OR "disorder*" OR "psychology*" OR "symptom*" OR "emotion*" OR "distress" OR "impact" OR "dysfunction" OR "experience" OR "grief" OR "griev*")

APPENDIX B.

Table B1. Systematic quality assessment of the studies

	Sample							Control/comparison group			Out-come	Follow-up		Distorting influences		Data reporting		Quality
Citation	Representative	Source	Method	Power calculation	Inclusion/Exclusion	Inclusion criteria	Identifiable	Source	Matched	Statistical control	Outcome	Number lost	Reason for loss	Traumatic events	Other	Missing data	Clarity Accuracy	
Baraković et al., 2013	N	N	N	N	Y	Y	Y	Y	N	N	Y	n/a	n/a	N	N	N	N	**M**
Baraković et al., 2014	N	Y	Y	N	Y	Y	N	Y	N	N	Y	n/a	n/a	N	N	N	N	**M**
Basharat et al., 2014	N	Y	Y	N	N	n/a	n/a	n/a	n/a	n/a	Y	n/a	n/a	N	N	N	N	**L**
Boss, 1977	N	N	N	N	N	n/a	n/a	n/a	n/a	n/a	N	n/a	n/a	n/a	N	N	N	**V**
Boss, 1980	N	Y	Y	N	N	n/a	n/a	n/a	n/a	n/a	N	n/a	n/a	n/a	N	N	N	**V**
Campbell & Demi, 2000	N	Y	Y	N	Y	n/a	n/a	n/a	n/a	n/a	Y	n/a	n/a	n/a	N	N	N	**M**
Clark, 2001	N	Y	Y	N	N	n/a	n/a	n/a	n/a	n/a	N	n/a	n/a	n/a	N	N	N	**V**
Heeke et al., 2015	N	Y	Y	N	N	Y	Y	Y	N	Y	Y	n/a	n/a	Y	Y	Y	N	**H**
Munczek & Tuber 1998	N	Y	Y	N	N	Y	Y	Y	N	N	N	n/a	n/a	N	N	N	N	**L**
Navia and Ossa, 2003	N	Y	Y	N	Y	Y	N	Y	N	N	Y	n/a	n/a	N	N	N	Y	**M**
Pérez-Sales et al., 2000	Y	Y	Y	N	N	Y	Y	Y	N	N	Y	n/a	n/a	N	N	N	Y	**H**
Powell et al., 2010	N	Y	Y	N	Y	Y	Y	Y	N	N	N	n/a	n/a	Y	N	N	N	**M**
Quirk & Casco, 1994	N	Y	Y	N	N	Y	Y	N	N	N	N	n/a	n/a	N	N	N	N	**V**

Table B1 (continued). Systematic quality assessment of the studies

Reisman, 2003	N	Y	Y	N	Y	Y	N	Y	N	N	Y	n/a	n/a	N	N	N	N	**M**
Zvidic and Butollo, 2001	N	Y	N	N	N	Y	Y	Y	N	Y	Y	n/a	n/a	Y	Y	N	Y	**H**

Note. N = no; Y = yes; n/a = not applicable; H = high; M = moderate; L = low; V = very low. For Sample, the 'representative' criterion was met if the study determined a base sample across multiple sources that matched the target population and used random sampling to arrive at the sample. The 'source' criterion was met if the study included a description of where the sample was drawn from. The 'method' criterion was met if the recruitment or selection procedure of participants was explicitly stated. The 'sample size' criterion was met if a power calculation was reported and the sample size was in accord with the power calculation. The 'inclusion/exclusion' criterion was met if a description and justifications was given of in-/exclusion criteria. To achieve a score of 'Adequate', a minimum of 3 Sample criteria had to be met. For Control/comparison group, all types of control/comparison groups were taken into account. The 'inclusion' criterion was met if a control/comparison group was included. The 'Identifiable' criterion was met if the control/comparison group was identifiable and a distinction was made between the groups in the study. The 'Source' criterion was met if a description was given of the source and recruitment of the control/comparison group. The 'Matched' criterion was met if matching or randomizing techniques were applied and described. The 'Statistical control' criterion was met if statistical differences between the groups were controlled for except for the primary outcomes or when it was described that there were no statistical differences between the groups. To achieve a score of 'Adequate' a minimum of 3 Control/Comparison criteria had to be met. Studies without a control/comparison group were rated 'not applicable' for this domain. For Outcome, 'outcome' criterion was met if the paper clearly stated what measures were used for which purposes and these measurements were methodologically sound. To achieve a score of 'Adequate' in this category, studies must have met the criterion. For Follow-Up, all study designs were marked as "not applicable", due to cross-sectional design. For Distorting Influence, controlling for "Traumatic events" was used as criterion for the current review since the majority of studies were conducted in the context of war and state terrorism. This criterion was met if studies in the context of war and state terrorism controlled for exposure to traumatic events. "Other" criterion was met if the study controlled for other confounding variables and gave a clear description of how and why they controlled for other variables. To achieve a score of 'Adequate' in this category, studies in the context of war and state terrorism must have met both of these criteria. Studies outside the context of war or state terrorism must have met the "Other" criterion in order to achieve a score of 'Adequate'. For Data Reporting, 'Missing Data' criterion was met if the paper reported about how missing data were dealt with. The 'Clarity Accuracy' criterion was met if data were clearly and accurately presented (e.g., appropriate use of statistics). To achieve a score of 'Adequate', studies must have met both criteria. A final quality level was computed for the studies as follows: high 3 or more adequate in applicable domains; moderate 2 adequate in applicable domains; low 1 inadequate in applicable domains; very low none adequate in applicable domains.

3

Prolonged grief and post-traumatic stress among relatives of missing persons and homicidally bereaved individuals: A comparative study

Lenferink, L.I.M., van Denderen, M.Y., de Keijser, J., Wessel, I., & Boelen, P.A. (2017). Prolonged grief and post-traumatic stress among relatives of missing persons and homicidally bereaved individuals: A comparative study. *Journal of Affective Disorders*, *209*, 1-2.

ABSTRACT

Background

Traumatic loss (e.g., homicide) is associated with elevated prolonged grief disorder (PGD) and posttraumatic stress disorder (PTSD). Several studies comparing relatives of missing persons with homicidally bereaved individuals showed inconsistent results about the difference in PGD- and PTSD-levels between the groups. These studies were conducted in the context of armed conflict, which may confound the results. The current study aims to compare PGD- and PTSD-levels between the groups outside the context of armed conflict.

Methods

Relatives of long-term missing persons (n = 134) and homicidally bereaved individuals (n = 331) completed self-report measures of PGD and PTSD. Multilevel regression modelling was used to compare symptom scores between the groups.

Results

Homicidally bereaved individuals reported significantly higher levels of PGD (d = 0.86) and PTSD (d = 0.28) than relatives of missing persons, when taking relevant covariates (i.e., gender, time since loss, and kinship to the disappeared/deceased person) into account.

Limitations

A limitation of this study is the use of self-report measures instead of clinical interviews.

Conclusion

Prior studies among relatives of missing persons and homicidally bereaved individuals in the context of armed conflict may not be generalizable to similar samples outside these contexts. Future research is needed to further explore differences in bereavement-related psychopathology between different groups and correlates and treatment of this psychopathology.

Keywords: Missing persons; Homicidal loss; Grief; Trauma

Boss (2006) claimed that the disappearance of a loved one is the most traumatic loss, because the whereabouts of the missing person are uncertain. It is important to verify this assumption, as it may contribute to psychopathology through stigmatization (Feigelman et al., 2009). Furthermore, insight into different psychological responses to different types of loss may help determining the necessity of tailored interventions.

Studies comparing symptom-levels of prolonged grief disorder (PGD)[1] and posttraumatic stress disorder (PTSD) between relatives of missing persons and homicidally bereaved individuals are scarce and inconclusive. One study showed a significantly higher lifetime-prevalence of PTSD post-homicide (9.0%) than post-disappearance (1.3%), whereas PGD-rates did not differ (Pérez-Sales et al., 2000). Conversely, another study found equal PTSD-levels, but significantly higher PGD-levels post-disappearance than post-homicide (d = .79) (Powell et al., 2010). A third study showed no group-differences in PGD and PTSD (Heeke et al., 2015).

These studies were conducted in armed conflict. Additional stressors (e.g., homelessness) may confound the results. Consequently, their generalizability to a context other than armed conflict is questionable. Therefore, we compared PGD- and PTSD-levels of relatives of missing persons and homicidally bereaved individuals outside armed conflict, using data from (on-going) research-projects.

Data were available from 134 relatives of missing persons[2] who had been missing for more than 3 months and 331 people confronted with the homicidal loss of a loved one at least 6 months earlier. Table 1 summarizes sample characteristics. A local review board provided ethics approval.

The Inventory of Complicated Grief (ICG) assessed PGD-symptoms (Boelen et al., 2003). Participants rated the frequency of 19 grief reactions during the last month on 5-point scales (anchors 0="never" and 4="always"). In keeping with prior research, scores >25 indicated probable PGD.

The 20-item PTSD Checklist for DSM-5 (PCL-5) assessed the DSM-5 PTSD-criteria in relatives of missing persons (Blevins et al., 2015). Participants rated on scales of 0 ("not at all") to 4 ("extremely") to what extent they experienced PTSD symptoms during the preceding month. We considered scores >1 on at least one B- and C-cluster-item and two D- and E-cluster-items as indicative of probable PTSD.

The PTSD Symptom Scale–Self-Report (PSS-SR) assessed the DSM-IV PTSD-criteria in homicidally bereaved individuals (Foa et al., 1993). Respondents briefly described an event

1. PGD, also referred to as complicated grief or persistent complex bereavement disorder, is characterized by yearning for the deceased and intense sorrow and is distinguishable from normal grief in terms of higher intensity and longer duration.
2. A missing person is defined as: "Anyone whose whereabouts is unknown whatever the circumstances of disappearance. They will be considered missing until located and their well-being or otherwise established" (Association of Chief Police Officers, 2010, pp. 15).

that bothered them most during the past month and rated the severity of 17 PTSD-symptoms during the preceding month on scales ranging from 0="not at all" to 3="five or more times per week/almost always". Scores of >1 on at least one re-experiencing, three avoidance, and two hyperarousal symptoms indicated probable PTSD.

Items that referred to "death" (ICG) or "stressful experience" (PCL-5) were replaced by references to the disappearance. Psychometric properties of the measures are adequate (Blevins et al., 2015; Boelen et al., 2003; Foa et al., 1993; $\alpha >$ .90 for all measures in the current samples).

Multilevel regression-analysis was used to deal with the nested data (i.e., multiple relatives of the same missing/deceased person completed questionnaires). Type of loss was included as independent variable and PGD- or PTSD-scores as dependent variables. Gender, time since loss, and kinship to the disappeared/deceased relative (i.e., spouse, parent, child, or sibling versus other) were included as covariates. An adapted PTSD score[3] for relatives of missing persons was used in the group-comparison. Because the PTSD measure in the homicidally bereaved sample could be related to other events than the homicide, we repeated the analysis for the subgroup of people explicitly referring to the homicidal loss in their event-description (n = 169).

3. To be able to compare the PTSD levels between the samples we transformed the 5-point scale (of the PCL-5) into a 4-point scale (in accord with the PSS-SR). Furthermore, items 10 to 12 of the PCL-5 were not covered in the PSS-SR and were therefore excluded. Item 1, 11, and 12 were not sufficiently covered in the PCL-5 and were therefore also assessed in the sample of relatives of missing persons.

Table 1. Characteristics of the participants

	Relatives of missing persons (*n* = 134)	Homicidally bereaved individuals (*n* = 331)
Gender, *N (%)*		
Men	45 (33.6)	113 (34.1)
Women	89 (66.4)	218 (65.9)
Age (years), *M (SD)*	57.8 (14.2)	52.7 (15.4)
Educational level, *N (%)*		
Low	32 (23.9)	-
Middle	44 (32.8)	-
High	58 (43.3)	-
Lost relative is, *N (%)*		
Partner/spouse	18 (13.4)	25 (7.8)
Child	41 (30.6)	157 (48.8)
Parent	14 (10.4)	41 (12.7)
Sibling	31 (23.1)	52 (16.1)
Other family member	28 (20.9)	28 (8.7)
Other	2 (1.5)	19 (5.9)
Number of years since loss, *M (SD)*	15.5 (17.0)	6.9 (6.5)
Type of disappearance, *N (%)*		
Criminal act	44 (32.8)	n.a.
Voluntarily	33 (24.6)	n.a.
Accident	33 (24.6)	n.a.
No specific suspicion	24 (17.9)	n.a.
Unique victims	89 (66.4)	254 (76.7)
Recruitment via *N (%)*		
Editorial office of a Tv-show about missing persons	36 (26.9)	n.a.
Peer support organizations	30 (22.4)	172 (52.0)
Non-governmental support organization	21 (15.7)	136 (41.1)
Family or friends	35 (26.1)	n.a.
Other	12 (9.0)	23 (6.9)
PGD severity, *Grand mean (SE)*	25.44 (1.40)	39.77 (0.81)
PTSD severity, *Grand mean (SE)*	13.71 (1.04)	19.96 (0.74)

Note. – = data were not available; n.a. = not applicable; PGD = prolonged grief disorder; PTSD = posttraumatic stress disorder.

In relatives of missing persons, prevalence rates were 47.0% and 23.1% for probable PGD and PTSD, respectively. These rates were 83.1% for PGD and 31.4% for PTSD in homicidally bereaved individuals. Homicidally bereaved individuals scored significantly higher than relatives of missing persons on PGD-symptoms ($F(1, 243.05) = 45.37$, $p < .001$, $d = 0.86$) and PTSD-symptoms ($F(1, 291.57) = 5.79$, $p < .05$, $d = 0.28$). Similar findings for PTSD-symptoms emerged when including the subgroup of $n = 169$ homicidally bereaved individuals ($F(1, 187.00) = 7.72$, $p < .01$, $d = 0.41$).

Our findings that PGD-levels and PTSD-levels are higher post-homicide than post-disappearance contrast with prior studies. This indicates the lack of generalizability of findings from studies in the context of armed conflict to populations outside these contexts. Additional war-related stressors in situations of armed conflict may account for differences. Furthermore, non-Western samples were used in previous studies. Grief may differ across cultures (Rosenblatt, 2008). In contrast to previous studies (Heeke et al., 2015; Pérez-Sales et al., 2000) we used self-reports instead of interviews, which may overestimate symptom-levels.

In contrast to relatives of missing persons, homicidally bereaved individuals need to deal with facts about the violent cause of death and the (presumed) perpetrator, which might be an explanation for the higher PGD- and PTSD-levels post-homicide than post-disappearance. Clearly, before conclusions can be drawn, future research should confirm the present findings. The large difference in PGD-levels suggests that PGD should be more central to treatments for people post-homicide relative to post-disappearance. Future research should further explore differences in psychopathology-symptoms and their correlates after different types of loss to inform intervention development.

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4

Cognitive-behavioral correlates of psychological symptoms among relatives of missing persons.

Lenferink, L.I.M., de Keijser, J., Wessel, I., & Boelen, P.A. (submitted). Cognitive-behavioral correlates of psychological symptoms among relatives of missing persons.

ABSTRACT

Background

The disappearance of significant others is associated with an increased risk of prolonged grief disorder (PGD), posttraumatic stress disorder (PTSD), and major depressive disorder (MDD). Enhancing knowledge about cognitive-behavioral correlates of PGD, PTSD, and MDD may generate valuable information for developing interventions for relatives of missing persons. We aimed to examine whether prior findings, indicating that cognitive-behavioral variables are related to distress among bereaved individuals, generalize to relatives of missing persons.

Methods

Relatives of missing persons (*n* = 134) completed self-report measures of negative cognitions, avoidance behaviors, PGD, PTSD, and MDD. Multilevel analysis was used.

Results

Cognitive-behavioral variables explained 40% to 60% of the additional variance in PGD, PTSD, and MDD levels over and above sociodemographic variables.

Conclusion

Similar to bereaved individuals, relatives of missing persons who tend to engage in negative cognitions and avoidance behaviors are more likely to experience elevated psychopathology levels. Addressing cognitive-behavioral variables in treatment may be beneficial.

The long-term disappearance of a significant other is a unique type of loss, due to uncertainty about the whereabouts of the missing loved one. Not knowing whether the separation is temporary or permanent may complicate the grieving process (Boss, 2006). There is evidence that similar to people confronted with other potential traumatic losses (Kristensen, Weisæth, & Heir, 2012), relatives of missing persons have an increased risk of the development of symptoms of prolonged grief disorder (PGD)[1], posttraumatic stress disorder (PTSD), and major depressive disorder (MDD) (Heeke & Knaevelsrud, 2015). For example, a study among people confronted with the disappearance of a family member or friend due to political repression showed that interview-based rates of PGD (23.3%), PTSD (67.1%), and MDD (68.5%) are comparable to homicidally bereaved individuals (Heeke, Stammel, & Knaevelsrud, 2015). PGD symptoms (e.g., intense sorrow and preoccupation with the deceased loved one) differ from normal grief reactions in that they last longer and are more intense (Maciejewski et al., 2016). In addition, the hallmark of PGD (i.e., longing for the deceased) is distinct from key symptoms of PTSD (i.e., intrusion symptoms) and MDD (i.e., sadness), which has been supported by factor analytic research (e.g., Prigerson et al., 1996).

Literature regarding correlates of psychological symptoms, including symptoms of PGD, PTSD, and MDD, in relatives of long-term missing persons is scarce and is primarily focused on sociodemographic correlates (Heeke & Knaevelsrud, 2015; Lenferink, de Keijser, Wessel, de Vries, & Boelen, 2017). Moreover, studies exploring correlates that are amendable to therapeutic change are lacking. Gaining insights into these correlates is relevant for developing psychological treatment for relatives of missing persons in need of professional support.

Several theories emphasize the importance of negative cognitions and avoidance behaviors in the development and persistence of PGD, PTSD, and MDD (e.g., Beck, 1987; Ehlers & Clark, 2000; Shear & Shair, 2005). Multiple studies, based on a cognitive behavioral model of PGD (Boelen, van den Hout, & van den Bout, 2006), showed that negative cognitions and avoidance behavior are related to symptom levels of PGD, but also PTSD and MDD concurrently and longitudinally in people who experienced the death of a loved one (Boelen, de Keijser, & Smid, 2015; Boelen & Eisma, 2015; Boelen & van den Bout, 2010; Eisma et al., 2013; van der Houwen, Stroebe, Schut, Stroebe, & van den Bout, 2010). Negative cognitions may include a negative self-view (e.g., "Since (--) is dead, I feel worthless"), negative view on one's own life (e.g. "My life is meaningless since (--) died"), a pessimistic view on the future (e.g. "I don't have confidence in the future"), and catastrophic misinterpretations of one's own grief reactions (e.g. "Once I would start crying, I would lose control"). Avoidance behavior includes both anxious avoidance and depressive avoidance.

1. PGD resembles persistent complex bereavement disorder as recently included as condition for further study in the fifth edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5,American Psychiatric Association [APA], 2013; see Maciejewski, Maercker, Boelen, & Prigerson, 2016).

Anxious avoidance refers to avoidance of loss-related stimuli out of fear that confrontation with these stimuli will be unbearable. Depressive avoidance behavior includes withdrawal from different social/recreational activities fueled by the belief that these activities are pointless and/or unfulfilling (Boelen et al., 2006).

To the best of our knowledge, cognitive-behavioral correlates of psychopathology in relatives of missing persons have not been studied previously. As noted, knowledge about these correlates may inform development and refinement of treatment interventions. Accordingly, the aim of this study – conducted in the Netherlands – was to examine whether prior findings, indicating that cognitive-behavioral variables are related to emotional distress after the death of loved one generalize to relatives of long-term missing persons. We expected that negative cognitions (about one's self, life, future, and catastrophic misinterpretations of one's own grief reactions) and anxious and depressive avoidance behaviors would explain amounts of variance in symptom levels of PGD, PTSD, and MDD over and above sociodemographic variables.

METHOD

Recruitment

The present data come from 134 participants in an ongoing research project on the correlates and treatment of psychopathology in relatives of missing persons (Lenferink, Wessel, de Keijser, & Boelen, 2016). Inclusion criteria were: a) being a nuclear or extended family member, spouse, or friend of a person who has been missing for at least 3 months, b) fluent in Dutch language, and c) being at least 18 years of age. The following definition of a missing person was used: "Anyone whose whereabouts is unknown whatever the circumstances of disappearance. They will be considered missing until located and their well-being or otherwise established" (Association of Chief Police Officers, 2010, p. 15). Paper-and-pencil questionnaire data were collected between July 2014 and January 2016. Participants were recruited via invitation letters sent by representatives of a Dutch television show about missing persons (26.9%) and peer support organizations (22.4%), and referral via Victim Support the Netherlands — a non-governmental organization for victim support — (15.7%). Others were approached through family members or friends (26.1%) or other recruitment-procedures (e.g., media-attention) (9.0%). These recruitment rates were based on the responses to an item about how the respondents were referred to this study. We cannot rule out that participants could have received multiple invitations to participate from different sources. Approval for conducting the study was obtained from a local ethics review board. All participants gave written informed consent.

Participants

Table 1 summarizes the characteristics of the sample. The majority of the participants were women (66.4%). On average, the participants were 57.8 (SD = 14.2) years of age and the disappearance took place 15.5 (SD = 17.0) years earlier. Most of the participants experienced the disappearance of a child (30.6%) and most participants (32.8%) presumed that their relative disappeared due to a criminal act (e.g., kidnapping, homicide). More than half (61.2%) of the participants assumed that their missing loved one was dead. The 134 participants represented 89 (66.4%) unique missing persons.

Table 1. Characteristics of the participants (*n* = 134)

Women, *N (%)*	89 (66.4)
Age (in years), *M (SD)*	57.8 (14.2)
Educational level, *N (%)*	
Primary to moderate	76 (56.7)
High	58 (43.3)
Lost relative is, *N (%)*	
Partner/spouse	18 (13.4)
Child	41 (30.6)
Parent	14 (10.4)
Sibling	31 (23.1)
Other family member	28 (20.9)
Other	2 (1.5)
Number of years since disappearance, *M (SD)*	15.5 (17.0)
Presumed cause of disappearance, *N (%)*	
Criminal act	44 (32.8)
Voluntarily	33 (24.6)
Accident	33 (24.6)
No specific suspicion	24 (17.9)
Belief about whereabouts of missing person, *N (%)*	
Alive	25 (18.6)
Dead	82 (61.2)
Doubt	27 (20.1)
Unique missing persons	89 (66.4)

Measures

Dependent variables

The 19-item Inventory of Complicated Grief (ICG) was used to assess self-rated symptoms of PGD (Prigerson et al., 1995). Although the original 29-item ICG-r was administered (Boelen, van den Bout, de Keijser, & Hoijtink, 2003; Prigerson & Jacobs, 2001), we only included the frequently used 19 items of the ICG (e.g., Hargrave, Leathem, & Long, 2012) in order to prevent content overlap with items of other measures used within this study. Participants rated how frequently they experienced grief reactions during the last month, on a scale ranging from 0 ("never") to 4 ("always"). Total scores range from 0 to 76. Items that referred to death were adapted to disappearance (e.g., "Ever since he/she has been missing it is hard for me to trust people"). The ICG has adequate psychometric properties (Prigerson et al., 1995). Cronbach's alpha in the current study was .92.

The 20-item PTSD Checklist for DSM-5 (PCL-5) was used to assess PTSD severity (Blevins, Weathers, Davis, Witte, & Domino, 2015; Boeschoten, Bakker, Jongedijk, & Olff, 2014). The PCL-5 represents the DSM-5 PTSD criteria (APA, 2013). Participants rated to what extent they experienced PTSD symptoms during the preceding month on a scale ranging from 0 ("not at all") to 4 ("extremely") (e.g., "How much were you bothered by feeling distant or cut off from other people?"). Total scores range from 0 to 80. The wording 'the stressful experience' in the instruction and the items was replaced by 'the events associated with the disappearance'. The psychometric properties of the PCL-5 are adequate (Blevins et al., 2015). Cronbach's alpha in the current study was .95.

The 30-item Inventory of Depressive Symptomatology – Self-Report (IDS-SR) was used to assess MDD severity (Rush, Gullion, Basco, Jarrett, & Trivedi, 1996). Participants were instructed to choose one out of four options (range 0 - 3) indicating how frequently they experienced a symptom (e.g., "Feeling sad") during the past 7 days. A total score was obtained by summing up 28 out of the 30 items (range 0 – 84). The IDS-SR showed good psychometric properties (Rush et al., 1996). Cronbach's alpha in the current study was .92.

Independent variables

The Grief Cognitions Questionnaire (GCQ) was used to assess negative cognitions related to the disappearance (Boelen & Lensvelt-Mulders, 2005). Four of its nine subscales were assessed: self (six items; e.g. "I am ashamed of myself, since he/she has been missing", range 0 - 30), life (four items; "Life has got nothing to offer me anymore", range 0 - 20), future (five items; "My wishes for the future will never be fulfilled", range 0 - 25), and catastrophic misinterpretations of one's own grief reactions (four items; "If I let go of my emotions, I will go crazy", range 0 - 20). Participants rated their agreement with each item on 6-point scales (anchors: 0 = disagree strongly and 5 = agree

strongly). Words that referred to death in the items were adapted to refer to the disappearance. The GCQ has adequate psychometric properties (Boelen & Lensvelt-Mulders, 2005). Cronbach's alphas of the subscales were .90 (self), .93 (life), .92 (future), and .93 (catastrophic misinterpretations).

The Depressive and Anxious Avoidance in Prolonged Grief Questionnaire (DAAPGQ) was used to assess avoidance behaviors (Boelen & van den Bout, 2010). The 9-item DAAPGQ consists of two subscales; depressive avoidance (five items; e.g., "I develop very few new activities since he/she has been missing, because I am unable to do so.", range 0 - 25) and anxious avoidance (four items; e.g., "I avoid to dwell on painful thoughts and memories connected to his/her disappearance.", range 0 - 20). Participants rated their agreement with each item on 6-point scale with anchors "not at all true for me" (0) to "completely true for me" (5). Words that referred to death in the items were adapted to refer to the disappearance. The DAAPGQ has adequate psychometric properties (Eisma et al., 2013). Cronbach's alpha was .92 for depressive avoidance and .75 for anxious avoidance.

Sociodemographic characteristics

Gender, age, kinship (categorized as child/spouse versus other), number of years since disappearance, and educational level (categorized as primary to moderate versus high) were registered. The presumed cause of disappearance and belief about the whereabouts of the missing loved one were also assessed. Presumed cause of disappearance was categorized as: voluntary, victim of criminal act, victim of accident, and no (specific) suspicion. Belief about the whereabouts of the missing loved one was categorized as: I think (s)he is alive, I doubt whether (s) he is alive, and I think (s)he is not alive.

Statistical analyses

Multilevel regression analyses were used to deal with the nested structure of the data. The nested structure of our data involves a two-level hierarchy: the participant (level 1, henceforth referred to as the participant-level), is nested within a social network/family of other relatives of the same missing person (level 2, henceforth referred to as the family-level). Intraclass coefficients were estimated to assess the proportion of variance at the family-level, based on random intercept-only models with symptom levels (i.e., sum scores) of PGD, PTSD, or MDD as the respective dependent variable. In case no variance could be explained by the family-level, single-level regression analyses were performed.

First, regression analyses were performed to examine whether each sociodemographic variable had a main effect on symptom levels of PGD, PTSD, and/or MDD. To this end, series of univariate analyses were performed with each of the sociodemographic variables as the independent variable, using dummy variables for the categorical variables, and symptom levels of PGD, PTSD, or MDD as the dependent variable.

Subsequently, the sociodemographic variables with a significant main effect on the indices of psychopathology were entered to the first model with symptom levels of PGD, PTSD, or MDD severity scores as dependent variable. The sum scores of the cognitive-behavioral variables were, next to the sociodemographic variables, entered to a second model. The explained proportion of variance at each level was computed for each model. A Bonferroni-correction was used for the main analyses to correct for multiple tests, resulting in an alpha of .02 (.05/3).

RESULTS

For PGD, 18.4% of the variance was at the family-level (level 2) and 81.6% at the participant-level (level 1). For PTSD, 18.1% of the variance was at the family-level and 81.9% at the participant-level. For MDD, the variance at the family-level was 0% and 100% at the participant-level, thus a single-level model was used in further analyses with respect to MDD.

Fixed main effects of sociodemographic variables on symptom levels of PGD, PTSD, and MDD

The results of the univariate regression analyses are displayed in Table 2. Women reported significantly higher levels of MDD (B = 5.29, SE = 2.47, p = .034), but not PGD and PTSD, than men. Time since loss was significantly and inversely related to PGD (B = -0.29, SE = 0.08, p < .001), PTSD (B = -0.22, SE = 0.09, p = .019), and MDD (B = -0.16, SE = 0.07, p = .019), indicating that psychopathology levels decrease with the passage of time. Also type of kinship had a significant main effect on symptom levels of PGD, PTSD, and MDD. Those who experienced the disappearance of a child or spouse reported higher PGD (B = 11.24, SE = 2.35, p < .001), PTSD (B = 9.91, SE = 2.85, p = .001), and MDD (B = 6.20, SE = 2.33, p = .009) levels compared with people whose parent, sibling, or more distant relative disappeared. Those who believed the missing loved one would be still alive reported significantly higher PGD (B = 8.97, SE = 3.42, p = .010) and PTSD (B = 8.23, SE = 4.03, p = .043) levels, not MDD levels, than those who believed that the missing loved one was deceased. Effects of age, educational level, and the type of disappearance (e.g., voluntarily missing versus presumed victim of a criminal act) were not statistically significant.

Table 2. Fixed main effects of sociodemographic variables on PGD, PTSD, and MDD in relatives of missing persons (n = 134); each parameter results from a univariate regression

	PGD		PTSD		MDD	
Variable	B	SE	B	SE	B	SE
Gender	1.83	2.62	4.62	3.04	5.29*	2.47
Educational level	-2.40	2.63	-0.93	3.09	2.55	2.39
Time since loss (in years)	-0.29***	0.08	-0.22*	0.09	-0.16*	0.07
Kinship	-11.24***	2.35	-9.91**	2.85	-6.20**	2.33
Presumed reason of disappearance						
Voluntarily vs. criminal act	5.94	3.53	3.85	4.19	1.65	3.18
Voluntarily vs. accident	-3.88	3.84	-4.19	4.55	-1.61	3.39
Voluntarily vs. no specific suspicion	3.06	4.00	-1.90	4.75	0.04	3.70
Whereabouts of missing loved one						
Dead versus alive	8.97*	3.42	8.23*	4.03	1.60	3.15
Dead versus doubt	4.51	3.21	2.57	3.80	0.26	3.06

Note. Gender was coded as 0 = men, 1 = women; educational level as 0 = primary to moderate, 1 = other, kinship as 0 = child or spouse, 1 = other; "Voluntarily" was the reference category for "Presumed reason of disappearance; "Dead" was the reference category for "Whereabouts of missing loved one"; * = $p < .05$; ** = $p < .01$; *** = $p < .001$.

Cognitive-behavioral correlates of PGD, PTSD, and MDD

First, the sociodemographic variables that yielded significant main effects on symptom levels of PGD, PTSD, or MDD were entered simultaneously in the first model for PGD, PTSD, or MDD, respectively. In the second model, the cognitive-behavioral variables were added. Individual variance inflation factors were all below 5, suggesting no cause for concern about multicollinearity. The results are displayed in Table 3.

The sociodemographic variables explained 20.1% of the variance in PGD at the participant-level. The type of kinship explained a unique proportion of the variance. The explained variance in PGD at the participant-level increased to 64.3% by adding the cognitive-behavioral variables to the second model. Catastrophic misinterpretations of one's own grief reactions and depressive avoidance behavior were uniquely associated with PGD severity when taking into account the other variables.

The sociodemographic variables explained 11.9% of the variance in PTSD at the participant-level. Type of kinship explained a unique proportion of the variance in PTSD. The explained variance in PTSD at the participant-level increased to 70.9% by adding the cognitive-behavioral variables to the second model. Misinterpretations of one's own grief reactions and depressive avoidance were significantly associated with PTSD levels when taking into account the other variables. Because the PTSD measure included two PTSD avoidance symptoms, which may artificially inflate the strength of its relationships with indices of anxious avoidance and depressive avoidance, we

repeated the multilevel analysis for PTSD, excluding the two PTSD avoidance symptoms (items 6 and 7). This analysis yielded similar results, indicating that misinterpretations of one's own grief reactions and depressive avoidance (B = .90, SE = 0.20, $p < .001$), but not anxious avoidance (B = 0.41, SE = 0.22, $p = .07$), were significantly associated with PTSD levels when taking into account the other variables.

The sociodemographic variables explained 10.9% of the variance in MDD. The explained variance in MDD increased to 69.4% by adding the cognitive-behavioral variables to the model. Misinterpretations of one's own grief reactions and depressive avoidance behavior were uniquely associated with MDD levels when taking into account the other variables. Repeating the analysis with excluding three items (item 8, 19, and 21) of the depression measure that may overlap with the indices of anxious avoidance and depressive avoidance yielded similar results, indicating that depressive avoidance was significantly related to depression when taking into account the other variables (B = .77, SE = 0.16, $p < .001$).

Table 3. Results of multiple (multilevel) regression models predicting levels of PGD, PTSD, and MDD in relatives of missing persons (*n* = 134)

	PGD				PTSD				MDD			
	Model 1		Model 2		Model 1		Model 2		Model 1		Model 2	
Variable	B	SE	B	SE	B	SE	B	SE	B	SE	B	SE
Gender	n.a.	n.a.	n.a.	n.a.	n.a.	n.a.	n.a.	n.a.	5.61	2.41	4.88**	1.47
Time since loss	-0.17	0.08	-0.06	0.05	-0.12	0.09	0.05	0.05	-0.14	0.07	0.02	0.04
Kinship	-9.07***	2.39	-3.31	1.68	-8.05**	2.94	-0.18	1.78	-4.89	2.37	0.66	1.49
Whereabouts of missing loved one[a]												
Dead vs alive	5.62	3.24			5.70	3.79			n.a.	n.a.	n.a.	n.a.
Dead vs doubt	2.02	3.02			-0.14	3.71			n.a.	n.a.	n.a.	n.a.
Cognitive variables												
Self			-0.03	0.22			-0.05	0.23			-0.07	0.20
Life			-0.14	0.33			0.36	0.35			0.39	0.29
Future			0.32	0.25			0.40	0.27			0.28	0.23
Misinterpretations			0.90***	0.22			0.71**	0.23			0.52*	0.20
Behavioral variables												
DA			0.61**	0.20			0.91***	0.21			0.94***	0.18
AA			0.37	0.23			0.55	0.24			-0.07	0.20
R^2 level 1, %	20.13		64.31		11.92		70.87		10.85		69.36	
R^2 level 1 change, %			44.18				58.95				58.51	
R^2 level 2, %	20.22		65.40		12.47		72.15					
R^2 level 2 change, %			45.18				59.68					
X^2 Change	38.05***		105.70***		25.13***		145.78***		5.23**		39.15***	

Note. Misinterpretations = catastrophic misinterpretations; DA = depressive avoidance; AA = anxious avoidance; n.a. = not applicable: these variables were not included in the model, because the univariate analyses showed no main effect of this variable on the outcome measure; [a] = The variance at level 2 could only be estimated in the second model by reducing the number of independent variables. In order to retain the maximum number of (significant) relevant predictors in the model, the dummy coded 'whereabouts of missing loved one' was deleted from model 2 for PGD and PTSD; Gender was coded as 0 = men, 1 = women, kinship as 0 = spouse/child, 1 = other; * = $p < .02$ (A Bonferonni-correction was used resulting in an alpha of .02 (.05/3)); ** = $p < .01$; *** = $p < .001$.

DISCUSSION

Our aim was to explore the generalizability of a cognitive behavioral model of disturbed grief (Boelen et al., 2006) for explaining PGD, PTSD, and MDD levels in relatives of long-term missing persons. Consistent with our hypothesis, the cognitive-behavioral variables explained a significant proportion of the variance in symptom levels of PGD, PTSD, and MDD, over and above relevant sociodemographic variables. Catastrophic misinterpretations of one's own grief reactions (but not negative cognitions about one's self, life, and future) and depressive avoidance behavior explained a unique proportion of the variance in PGD, PTSD, and MDD when taking the shared variance of other variables into account. Our findings are roughly in line with previous studies indicating that catastrophic misinterpretations of one's own grief reactions and depressive avoidance were most strongly associated with indices of psychopathology (Boelen et al., 2015; Boelen, van Denderen, & de Keijser, 2016; Boelen & van den Bout, 2010).

Taken together, the results indicate that relatives of missing persons, similar to people bereaved by the death of a loved one, experience elevated PGD, PTSD, and MDD levels when they engage in catastrophic misinterpretations of one's own grief reactions (e.g., "If I let go of my emotions, I will go crazy") and tend to withdraw from different social/recreational activities from the belief that these activities are pointless. These findings also suggest that relatives of missing persons, with elevated psychopathology symptoms, may benefit from addressing these negative cognitions and avoidance behavior in treatment. Cognitive behavioral therapy (CBT) may be the most obvious treatment option, since it has shown to be effective for treatment of post-bereavement complaints (Currier, Holland, & Neimeyer, 2010). For example, CBT could enhance awareness of one's own misinterpretation of the grief reactions and could transform these maladaptive cognitions into more adaptive cognitions (e.g., by cognitive restructuring). Furthermore, CBT could promote relatives of missing persons to reengage in activities that were perceived as fulfilling prior to the disappearance (e.g., by behavioral reactivation). However, the effectiveness of a CBT-based intervention for relatives of missing persons has only been evaluated once (Hagl, Rosner, Butollo, & Powell, 2014). Future research should further evaluate and optimize interventions for relatives of missing persons in need of professional support.

With respect to the sociodemographic correlates of CBT, our univariate analyses showed, among others, a main effect of belief about the whereabouts of the missing person. That is, participants who believed their missing loved one would be still alive reported significantly higher PGD and PTSD levels, not MDD levels, than those who believed that the missing loved one died. This finding is reminiscent of previous findings that the amount of hope that the missing loved one would be still alive was associated with increased PGD levels, but not with PTSD (C. Heeke, personal communication, October 31, 2017) and MDD levels (Heeke et al., 2015). However,

a failure to reject the null-hypothesis renders a finding inconclusive: the absence of a statistically significant effect may indicate that hope that a missing person is alive is less important for MDD levels or that our study was not able to detect the effect. Future research may shed more light on this issue.

More generally, it may be important to focus on the function of hope in treatment for relatives of missing persons (Lenferink et al., 2016). Although holding on to hope that the missing relative is alive may in some cases be more realistic (e.g., kidnapping cases or voluntarily missing persons), for those who maintain hope against all odds, holding on to hope may block acceptance of the irreversibility of the separation. Using exposure techniques in treatment might yield favorable effects, similar to effects of exposure for disturbed grief after bereavement (cf. Bryant et al., 2014).

It was salient that at the family-level (level 2) zero variance in MDD was estimated, contrasting estimated variances at the family-level in PGD and PTSD about 18%. In other words, MDD symptoms may predominately be intrapersonal and thus independent from other relatives of the same missing person. A previous study among bereaved parents also showed a relatively low amount of variance in MDD explained by the family-level compared with the variance levels in PGD (Wijngaards-de Meij et al., 2005). Clearly, this conclusion must remain tentative pending additional research, given that our sample represented a relatively small number of relatives of the same missing person.

Several further limitations need to be taken into account while interpreting the results of the current study. Self-report measures were used, which may lead to overestimation of symptom levels (Engelhard et al., 2007). Furthermore, the items of ICG are not identical to the proposed criteria of persistent complex bereavement disorder for the DSM-5 (APA, 2013) or PGD criteria for the 11th edition of the International Classification of Diseases (Maercker et al., 2013), thus the results of the current study may not directly speak to (future) studies using these criteria (Maciejewski et al., 2016). In addition, we used a convenience sample mostly recruited via (peer) support organizations. It may be that our sample constitutes individuals with more pervasive psychopathology levels compared with individuals who do not seek (peer) support (de Groot & Kollen, 2013). Some caution is therefore warranted when generalizing the current findings to the population. Another limitation of the study sample is that the average time since disappearance was 15.5 years. Although time since disappearance was not strongly related to the outcome measures, we cannot rule out that our results may not be generalizable to people whose relative disappeared more recently. Lastly, it must be noted that the cross-sectional design precludes drawing conclusions about causality.

To conclude, this correlational study, focusing on cognitive-behavioral correlates of psychopathology post-disappearance for the first time, showed the generalizability of the cognitive-behavioral model of psychopathology post-loss to relatives missing persons. Our

findings are consistent with other studies among bereaved samples, which suggest that it might be fruitful to evaluate the effectiveness of CBT for individuals who suffer from elevated psychopathology levels following the disappearance of a significant other.

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ACKNOWLEDGEMENTS

We would like to thank the “Vereniging Achterblijvers na Vermissing”, “AVROTROS Vermist”, Victim Support the Netherlands, and Child Focus for their support with the recruitment of relatives of missing persons. In addition, we would like to thank the Dutch police, especially Carlo Schippers and Irma Schijf, for their kind collaboration throughout this research project. Also thanks to M. E. Timmerman for statistical consultation.

5

Exploration of the associations between responses to affective states and psychopathology in two samples of people confronted with the loss of a loved one

Lenferink, L.I.M., Wessel, I., & Boelen, P.A. (2018). Exploration of the associations between responses to affective states and psychopathology in two samples of people confronted with the loss of a loved one. *The Journal of Nervous and Mental Disease*, *206*(2), 108-115.

ABSTRACT

Adaptive regulation of positive and negative affect following the loss of a loved one may foster recovery. In two studies, using similar methods but different samples, we explored the association between positive (i.e., dampening and enhancing) and negative (i.e., rumination) affect regulation strategies and symptoms levels of post-loss psychopathology.

Study 1 used data from 187 people confronted with the death of a loved one. In Study 2, the sample consisted of 134 relatives of long-term missing persons. Participants completed self-reports tapping prolonged grief, depression, posttraumatic stress symptoms, and affect regulation strategies. Hierarchical regression analyses showed that both negative and positive affect regulation strategies explained significant amounts of variance symptom-levels in both samples. In line with previous work, our results suggest that negative and positive affect regulation strategies relate to post-loss psychopathology. Future research should explore how both affect regulation strategies may adequately be addressed in treatment.

Keywords: bereavement; affect regulation; missing persons; rumination; trauma.

The death of a significant other is a universal experience. Some people develop psychological complaints, including depression, posttraumatic stress disorder (PTSD), and persistent and disabling grief reactions, also referred to as prolonged grief disorder (PGD; Prigerson et al., 2009), although the vast majority does not (see for overviews Lundorff, Holmgren, Zachariae, Farver-Vestergaard, & O'Connor, 2017; Onrust & Cuijpers, 2006).

There is evidence that negative affect regulation strategies, including rumination, are related to elevated symptom-levels of PGD, depression, and PTSD following loss (Eisma et al., 2015; Morina, 2011; Nolen-Hoeksema, Parker, & Larson, 1994). Rumination refers to repetitive thinking about the nature, causes, and consequences of negative affect (Nolen-Hoeksema, Wisco, & Lyubomirsky, 2008). Nolen-Hoeksema et al. (1994) found stronger tendencies to ruminate about one's depressed mood one month after losing a family member to be associated with elevated depression at 6 months post-loss. Another study showed a longitudinal link between grief-related rumination and both levels of depression and PGD in a community sample of bereaved individuals (Eisma et al., 2015).

Traditional views suggested that experiencing positive emotions post-loss is an indication of psychopathology (e.g., repression or denial; Bowlby, 1980; Freud, 1957; Kübler-Ross, 1973). However, it has been repeatedly shown that positive emotions following loss promote resilient outcomes and recovery. For example, one study among conjugally bereaved individuals, using ecological momentary assessment indicated that experiencing daily positive emotions mediated the effect of trait resilience on emotional recovery (Ong, Bergeman, Bisconti, & Wallace, 2006). Similarly, a longitudinal study showed that experiencing positive emotions following spousal bereavement was associated with less depression and PGD (Tweed & Tweed, 2011). Another study showed that positive facial expressions while talking about the deceased spouse six months post-loss was inversely related to PGD levels 14 and 25 months post-loss (Bonanno & Keltner, 1997).

The adaptive effects of positive affect following a stressful event, including loss, have been emphasized in the broaden-and-build theory of Fredrickson (1998, 2001). According to this theory positive emotions broaden a person's scope of attention and thought and action tendencies, which results in building, among others, social (e.g., social support networks) and psychological recourses (e.g., resilience; Fredrickson, 1998; Fredrickson, 2001; Fredrickson & Branigan, 2005; Fredrickson & Levenson, 1998; Garland et al., 2010). These resources may serve as a buffer in times of adversity. For instance, in the face of the loss of a relative, the experience of positive emotions (e.g., love, gratitude) may encourage the individual to engage in social activities that foster adjustment, which, in turn, may lead to the maintenance or enhancement of positive affect (i.e., referred to as the "upward spiral of positive emotions" by Fredrickson, 2001). In addition, this may counter the pain and sadness associated with the loss (referred to as the "undoing hypothesis" by Fredrickson and Levenson, 1998). Suppression of positive emotions may block their effects on the

recovery processes, whereas adaptive positive affect regulation strategies may help to maintain these emotions.

Interest in the role of positive affect regulation has increased with the advent of the Response to Positive Affect questionnaire (RPA; Feldman, Joormann, & Johnson, 2008). The RPA is proposed to assess three strategies of how people respond to positive affect. The first strategy ("dampening") involves devaluating, suppressing, or downgrading positive affect (e.g., "When I feel happy, I remind myself these feelings won't last"). The other two strategies are coined "enhancing" strategies (also referred to as "positive rumination" in prior research, e.g., Nelis et al., 2016) and include self-focused positive rumination (e.g., "When I feel happy, I feel I can achieve everything") and emotion-focused positive rumination (e.g., "When I feel happy, I savour that moment"). More dampening has been associated with increased levels of depression (Raes et al., 2012). Stronger endorsement of enhancing strategies has been found to be associated with lower depression levels (Nelis, Holmes, & Raes, 2015).

Several studies using the RPA in non-clinical samples found that positive affect regulation strategies are related to depression scores above and beyond brooding, concurrently (Raes, Daems, Feldman, Johnson, & Van Gucht, 2009) as well as longitudinally (Raes et al., 2012, 2014). Although it has been suggested that strategies to regulate positive emotions are likely involved in recovery from loss (Folkman, 2001; Stroebe & Schut, 2001), to the best of our knowledge, this notion has never been empirically tested.

We explored to what extent negative and positive affect regulation strategies are related to psychopathology following the loss of a loved one. Therefore, we studied two different samples. The first sample constituted of people confronted with the recent death of a loved one. The second sample consisted of people confronted with the long-term disappearance of a loved one, a type of loss that is also referred to as "ambiguous loss" (Boss, 2006). Studies on the psychological consequences of disappearances of loved ones are scarce and predominantly focus on disappearances in armed conflicts (i.e., war and state terrorism; Lenferink, de Keijser, Wessel, de Vries, & Boelen, in press). In both samples, we explored to what extent positive affect regulation strategies (i.e., dampening and enhancing) explained variance in PGD, depression, and PTSD above and beyond negative affect regulation strategies (i.e., rumination).

STUDY 1

Methods

Participants and procedures

We used the data of an on-going study of 187 adults whose significant other died in the past year. Participants were recruited via announcements on websites providing information about grief and loss. Most of the participants were women (64.7%), aged 59.9 (SD = 12.7) years on average, and had a primary to moderate educational level (50.3%). The majority experienced the death of a spouse or child (56.1%). On average, the death took place 3.9 (SD = 1.6) months earlier, in most cases (92.5%) due to a natural cause (e.g., disease).

All participants signed informed consent. Ethics approval for conducting the study was obtained from a local ethics committee. Because Study 1 was part of a larger research project (the Utrecht Longitudinal Study on Adjustment To Loss [ULSATL study], see, e.g., Boelen, 2017) only the measures used in the current study are described.

Measures

Prolonged grief

The 11-item PGD scale (Boelen, Keijsers, & van den Hout, 2012) was administered to assess PGD symptom as put forth by Prigerson et al. (2009). This measure is based upon items included in the revised Inventory of Complicated Grief. Accordingly, items represent one separation distress symptom, nine cognitive and emotional symptoms, and one functional impairment symptom. Participants were instructed to rate how frequently they experienced each grief reaction during the preceding month on 5-point scales ranging from 1 (“never”) to 5 (“always”). The item scores were summed and represented an overall PGD severity score. The PGD scale was developed and validated in the context of prior research (Boelen et al., 2012). Cronbach’s alpha in the current sample was .92.

Depression

The 7-item depression subscale of the Hospital Anxiety and Depression Scale (HADS-D) was administered to assess depression levels (Zigmond & Snaith, 1983). Participants chose one out of four answers that described how frequently they experienced the symptom during the past week (e.g., “I feel as if I am slowed down”). Item-scales range from 0 to 3, with higher scores representing more severe depression. The item scores were summed to form an overall depression severity

score. The HADS-D has good psychometric properties (Bjelland, Dahl, Haug, & Neckelmann, 2002). Cronbach's alpha in the current sample was .92.

Posttraumatic stress

The Posttraumatic Diagnostic Scale (PDS) was administered to assess 17 PTSD symptoms according to the DSM-IV criteria (Foa, Cashman, Jaycox, & Perry, 1997). Participants rated how frequently they experienced each symptom during the past month on a 4-point scale ranging from 0 ("Not at all/only one time") to 3 ("5 or more times a week/almost always"). The wording that referred to "the stressful event" in the instruction and items were replaced by "the death of your loved one" (e.g., "Having upsetting thoughts or images about the death of your loved one that came into your head when you didn't want them to"). The item scores were summed to form an overall PTSD severity score. The PDS showed adequate psychometric properties (Foa et al., 1997). Cronbach's alpha in the current sample was .89.

Strategies to regulate positive affect

The RPA assesses strategies to regulate positive affect (Feldman et al., 2008; Raes et al., 2009). Participants rated what they generally do when they feel happy, excited, or enthused on a 4-point scale ranging from 1 ("almost never") to 4 ("almost always"). The items refer to three strategies coined "dampening" (8 items, e.g., "Remind yourself these feelings won't last"), "self-focused positive rumination" (4 items, e.g., "Think 'I am achieving everything'"), and "emotion-focused positive rumination" (5 items, e.g., "Think about how happy you feel"). One item of the dampening subscale ("Think about how hard it is to concentrate") was removed from the Dutch translation of the RPA (Raes et al., 2009). Following the example of Nelis et al. (2016) we omitted another item of the "dampening" subscale (i.e., "This is too good to be true") and the two positive rumination scales were combined into one subscale (i.e., "enhancing"). The results of a principal component analysis in both of our samples confirmed Nelis et al.'s (2016) findings. The item scores of both subscales were summed to form an overall dampening or enhancing score. The Dutch RPA showed adequate psychometric properties (Raes et al., 2009). Cronbach's alpha for the dampening subscale and enhancing subscale in the current sample was .69 and .88, respectively.

Brooding

The 5-item Brooding subscale of the Ruminative Response Scale (RRS) was used to assess the tendency to ruminate (Treynor, Gonzalez, & Nolen-Hoeksema, 2003). Participants rated what they generally think or do when they feel sad (e.g., "I think 'Why do I always react this way?'") on 4-point scales ranging from 1 ("almost never") to 4 ("almost always"). The item scores were summed to

form an overall brooding severity score. The RRS has been found to have adequate psychometric properties (Treynor et al., 2003). Cronbach's alpha in the current sample was .71.

Statistical analyses

First, zero-order Spearman's rho correlations were calculated to examine the association between all independent and dependent variables. Second, three separate hierarchical regression analyses were performed with symptom levels of PGD, depression, or PTSD as consecutive dependent variable. Step 1 of each regression model consisted of the sociodemographic variables that showed associations with the dependent variables in univariate testing. We used Mann-Whitney tests or Spearman's rho correlations for univariate testing. Step 2 consisted of brooding. Step 3 consisted of the two subscales of the RPA (i.e., dampening and enhancing). A Bonferroni-corrected alpha level of <.02 (i.e., .05/3 because we conducted three main analyses in each sample) was considered statistically significant for the hierarchical regression analyses. Less than 5% of responses on the items of the dependent and independent variables were missing. Missing data were therefore imputed with the mean.

Sample size calculation

Because the current data were obtained as part of an ongoing study, there was no a priori sample size calculation based on our particular research question. However, based on a sensitivity analysis for a multiple regression analysis to examine the R^2 increase of two predictors with seven predictors in total, 80% power, and alpha .02, our sample size of 187 was sufficient to detect a small to medium effect size ($f^2 = 0.07$).

RESULTS

Preliminary Analyses

Table 1 displays the results of univariate testing for the relatives of deceased persons. Gender, educational level, kinship to the deceased, and cause of death (but not time since loss) were significantly related to symptom-levels of PGD, depression, and/or PTSD.

The affect regulation strategies were all significantly related to PGD, depression, and PTSD. Enhancing was negatively associated with all variables. The other correlations were all positively directed. Brooding was positively related to dampening ($r_s = .44$, $p < .001$) and enhancing was negatively related to brooding ($r_s = -.31$, $p < .001$) and dampening ($r_s = -.16$, $p = .03$).

Table 1. Association between independent and dependent variables in relatives of deceased persons (n = 187)

	Prolonged grief	Depression	Posttraumatic stress
Gender, *U*	2774.00**	2598.00***	2222.00***
Men, *Mdn, IQR*	19.0 (14.3)	1.0 (3.3)	6.0 (8.4)
Women, *Mdn, IQR*	26.0 (16.0)	5.0 (9.0)	13.0 (13.3)
Educational level, *U*	7879.00*	3758.50	3615.50*
Primary to moderate, *Mdn, IQR*	25.0 (16.25)	3.5 (8.0)	10.5 (13.0)
High, *Mdn, IQR*	20.0 (16.5)	2.0 (7.3)	9.0 (13.7)
Time since death (in months), r_s	.09	.07	.03
Kinship, *U*	2396.50***	2791.50***	2679.50***
Deceased is child/spouse, *Mdn, IQR*	28.0 (14.0)	5.0 (7.2)	12.0 (12.0)
Deceased is other, *Mdn, IQR*	18.0 (12.1)	1.0 (6.5)	5.0 (12.1)
Cause of death, *U*	682.50**	806.00*	812.00*
Natural, *Mdn, IQR*	23.0 (15.0)	3.0 (7.0)	9.0 (13.4)
Suicide/accident/homicide, *Mdn, IQR*	33.0 (16.0)	8.0 (8.0)	14.0 (11.0)
Brooding, r_s	.47***	.41***	.47***
Dampening, r_s	.16*	.26***	.20**
Enhancing, r_s	-.28***	-.36***	-.26***
Prolonged grief, r_s		.77***	.87***
Depression, r_s			.80***

Note. *** = p <.001; ** = p < .01; * = p < .05; Mdn = median; IQR = interquartile range.

Regression analyses

Table 2 shows the results of the hierarchical regression analyses for relatives of deceased persons. The individual variance inflation factor of each independent variable was < 2 indicating no cause for concern about multicollinearity.

The sociodemographic variables explained 22.6% of the variance in PGD, 16.6% in depression, and 18.7% in PTSD (all ps < .001). In the second step of the analyses, brooding explained an additional 27.3% of the variance in PGD, 22.8% in depression, and 26.3% in PTSD (all ps < .001). In the third step, the enhancing and dampening subscale of the RPA explained an additional 6.6% of the variance in depression (p < .001), and 2.6% in PTSD (p = .01), but the additional variance explained in PGD did not reach statistical significance (ΔR^2 = 2.1, p = .02). In the final regression models, brooding was significantly and positively related to depression and PTSD. Enhancing was significantly and inversely related to depression. Dampening was significantly and positively related to depression.

Table 2. Results of hierarchical regression analyses for relatives of deceased persons (n = 187)

	Prolonged grief				Depression				Posttraumatic stress			
	B (SE)	β	ΔR^2	ΔF (df)	B (SE)	β	ΔR^2	ΔF (df)	B (SE)	β	ΔR^2	ΔF (df)
Step 1: Sociodem.	-	-	.23***	13.28 (4, 182)	-	-	.17***	12.18 (3, 183)	-	-	.19***	10.49 (4, 182)
Step 2: Brooding	1.98 (0.20)	.54***	.27***	98.44 (1, 181)	0.88 (0.11)	.49***	.23***	68.60 (1, 182)	1.55 (0.19)	.47***	.26***	86.41 (1, 181)
Step 3:			.02	4.00 (2, 179)			.07***	10.98 (2, 180)			.03*	4.39 (2, 179)
Brooding	1.70 (0.24)	.46***			0.60 (0.12)	.33***			1.73 (0.19)	.53***		
Dampening	0.20 (0.23)	.05			0.29 (0.12)	.16*			0.35 (0.21)	.10		
Enhancing	-0.26 (0.10)	-.15**			-0.19 (0.05)	-.23***			-0.22 (0.09)	-.14		

Note. *** = p <.001; ** = p < .01; * = p < .02 (.05/3); RPA = Response to Positive Affect Questionnaire; Sociodem. = sociodemographic characteristics; - = not displayed; df = degrees of freedom.

STUDY 2

Methods

Participants and procedures

Data were available from 134 participants included in an on-going study examining correlates and treatment of psychopathology in relatives of missing persons (Lenferink, van Denderen, de Keijser, Wessel, & Boelen, 2017; Lenferink, Wessel, de Keijser, & Boelen, 2016). People whose significant other disappeared at least three months earlier were eligible to participate. Data were collected through invitation letters sent by the editorial office of a Dutch television show about missing persons (26.9%), a Dutch peer support organization (22.4%), and a non-governmental organization for victim support (15.7%). Some participants were recruited via snowball-sampling (26.1%) or other ways of recruitment (e.g., media-attention) (9.0%). Most participants were women (66.4%) and participants were 57.8 (SD = 14.2) years old on average. Most participants had a primary to moderate educational level (56.7%). Forty-four per cent experienced the disappearance of spouse or child. On average, the disappearance took place 15.5 (SD = 17.0) years earlier. About one-third of the participants (32.8%) thought the disappearance was due to criminal act (e.g., homicide or kidnapping). The 134 participants represented 89 unique cases of missing persons. A missing person was defined as "Anyone whose whereabouts is unknown whatever the circumstances of disappearance. They will be considered missing until located and their well-being or otherwise established" (Association of Chief Police Officers, 2010, p. 15).

All participants signed informed consent. Ethics approval for conducting the study was obtained from a local ethics committee. Because Study 2 was part of a larger research project, only the measures used in the current study were described. See Lenferink et al. (2016) for a description of other measures used in this larger project.

Measures

Prolonged grief

The Dutch translation of the 29-item revised Inventory of Complicated Grief (ICG-r) was administered to assess symptom levels of PGD and other putative markers of disturbed grief (Boelen, van den Bout, de Keijser, & Hoijtink, 2003). Participants rated how frequently they experienced each grief reaction during the preceding month on 5-point scales ranging from 1 ("never") to 5 ("always"). The item scores were summed to form an overall PGD severity score. The wording that referred to "death" in the instruction and items were replaced by referring to the disappearance (e.g., "Ever since he/she has been missing it is hard for me to trust people"). It is

noteworthy that the original ICG-r (which was originally introduced as the Inventory of Traumatic Grief) has 30 items with 5-point scales with varying answer options, e.g., from 1 (“almost never”) to 5 (“always”) and from 1 (“No sense of numbness”) to 5 (“An overwhelming sense”; Prigerson & Jacobs, 2001). The Dutch ICG-r demonstrates adequate psychometric properties (Boelen et al., 2003). Cronbach’s alpha in the current sample was .96.

Depression

The 30-item Inventory of Depressive Symptomatology-Self-Report (IDS-SR) was used (Rush, Gullion, Basco, Jarrett, & Trivedi, 1996). For each item a description is given of a depressive symptom (e.g., “Feeling sad”). Participants were instructed to choose one out of four answers (range 0 to 3) that described how frequently they experienced the symptom during the past week (e.g., “I feel sad nearly all of the time”). The item scores were summed to form an overall depression severity score. The IDS-SR showed good psychometric properties (Rush et al., 1996). Cronbach’s alpha in the current sample was .92.

Posttraumatic stress

The 20-item PTSD Checklist for DSM-5 (PCL-5) was administered to assess symptoms of PTSD according to DSM-5 criteria (Blevins, Weathers, Davis, Witte, & Domino, 2015). Participants were instructed to rate to what extent they experienced each symptom during the past month on a 5-point scale ranging from 0 (“Not at all”) to 4 (“Extremely”). The wording that referred to ‘the stressful experience’ in the instruction and items were replaced by ‘the events that are associated with the disappearance’ (e.g., “In the past month, how much were you bothered by repeated, disturbing, and unwanted memories of the events that are associated with the disappearance?”). The item scores were summed to form an overall PTSD severity score. The PCL-5 showed adequate psychometric properties (Blevins et al., 2015). Cronbach’s alpha in the current sample was .95.

Affect regulation strategies

Similar to Study 1, the RPA and the brooding subscale of the RRS were administered. Cronbach’s alpha’s for the dampening subscale and the enhancing scale were .82 and .83, respectively. Cronbach’s alpha of the brooding subscale was .77.

Statistical analyses

The same statistical analyses were used as in Study 1.

Sample size calculation

Based on a sensitivity analysis for a multiple regression analysis to examine the R^2 increase of two predictors with six predictors in total, 80% power, and alpha .02, our sample size of 134 was sufficient to detect a small to medium effect size ($f^2 = 0.09$)

RESULTS

Preliminary analyses

Duration of the disappearance, kinship to the missing person, and/or presumed cause of disappearance were significantly related to the indices of psychopathology. Gender and educational level were not significantly related to the indices of psychopathology (see Table 3).

All affect regulation strategies were significantly related to PGD, depression, and PTSD (see Table 3). Enhancing was negatively and dampening and brooding were positively associated with all indices of psychopathology. Brooding was positively associated with dampening ($r_s = .45$, $p < .001$). Enhancing was negatively associated with dampening ($r_s = -.23$, $p = .01$) and not significantly related to brooding ($r_s = -.13$, $p = .13$).

Table 3. Association between independent and dependent variables in relatives of missing persons (n = 134)

	Prolonged grief	Depression	Posttraumatic stress
Gender, *U*	1919.00	1647.50	1774.50
Men, *Mdn, IQR*	63.0 (39.0)	9.0 (16.3)	13.0 (23.0)
Women, *Mdn, IQR*	63.0 (39.0)	14.0 (23.5)	19.0 (30.0)
Educational level, *U*	2064.00	1910.00	2045.00
Primary to moderate, *Mdn, IQR*	63.5 (34.0)	9.7 (18.4)	18.5 (28.0)
High, *Mdn, IQR*	61.2 (42.8)	14.0 (17.0)	14.5 (29.8)
Duration of disappearance (in years), r_s	-.35***	-.21*	-.22*
Kinship, *U*	1250.50***	1693.00*	1474.50**
Missing is child/spouse, *Mdn, IQR*	76.0 (39.0)	15.0 (22.0)	23.0 (29.0)
Missing is other, *Mdn, IQR*	52.0 (30.0)	10.0 (17.0)	12.0 (26.0)
Presumed cause of disappearance, *U*	1524.00*	1799.50	1603.00
Accident/voluntarily/no presumption, *Mdn, IQR* Criminal act, *Mdn, IQR*	60.2 (36.3) 69.5 (40.25)	12.0 (18.3) 12.5 (22.8)	13.0 (27.3) 21.5 (27.0)
Brooding, r_s	.66***	.69***	.69***
Dampening, r_s	.34***	.28**	.40***
Enhancing, r_s	-.24**	-.23**	-.28**
Prolonged grief, r_s		.76***	.83***
Depression, r_s			.83***

Note. *** = $p < .001$; ** = $p < .01$; * = $p < .05$. ; Mdn = median; IQR = interquartile range.

Regression analyses

Table 4 shows the results of the hierarchical regression analyses. The individual variance inflation factor of each independent variable was < 2 indicating no cause for concern about multicollinearity.

The sociodemographic variables explained 23.2%, 7.1%, and 8.9% of the variance in symptom-levels of PGD, depression, and PTSD, respectively (all *p*s < .01). In the second step of the analyses, brooding explained an additional 33.4% of the variance in PGD, 37.6% in depression, and 46.6% in PTSD (all *p*s < .001). Adding the enhancing and dampening subscale of the RPA to the model explained and additional 2.8% of the variance in PGD ($p = .01$) and 3.4% ($p = .01$) in PTSD, but did not significantly explain additional variance in depression ($p = .05$). In the final regression models, brooding was significantly and positively related to PGD and PTSD. Enhancing was significantly and inversely related to PGD and PTSD levels. We also ran multilevel regression analyses in order to account for the hierarchical structure of the data (i.e., the fact that 134 participants were associated with 89 unique missing persons). These analyses yielded similar patterns of results.

Table 4. Results of hierarchical regression analyses for relatives of missing persons (n = 134).

	Prolonged grief				Depression				Posttraumatic stress			
	B (SE)	β	ΔR^2	ΔF (df)	B (SE)	β	ΔR^2	ΔF (df)	B (SE)	β	ΔR^2	ΔF (df)
Step 1: Sociodem.	-	-	.23***	13.00 (3, 129)	-	-	.07**	4.96 (2, 130)	-	-	.09**	6.37 (2, 130)
Step 2: Brooding	4.88 (0.49)	.61***	.33***	98.55 (1, 128)	2.85 (0.30)	.63***	.38***	87.85 (1, 129)	4.00 (0.34)	.70***	.47***	135.32 (1, 129)
Step 3:			.03*	4.28 (2, 126)			.03	3.19 (2, 127)			.03**	5.19 (2, 127)
Brooding	4.68 (0.54)	.58***			2.92 (0.33)	.64***			3.76 (0.37)	.66***		
Dampening	0.10 (0.46)	.01			-0.29 (0.29)	-.07			0.26 (0.33)	.05		
Enhancing	-0.90 (0.31)	-.17**			-0.49 (0.20)	-.16			-0.68 (0.23)	-.18**		

Note. *** = p <.001; ** = p < .01; * = p < .02 (.05/3); RPA = Response to Positive Affect Questionnaire; Sociodem. = sociodemographic characteristics; - = not displayed; df = degrees of freedom.

DISCUSSION

The current study, using two different samples of people confronted with the loss of a loved one, explored to what extent strategies to regulate positive and negative affect are related to emotional distress associated with the loss. Positive affect regulation strategies (i.e., dampening and enhancing) explained significant amounts of variance in symptom-levels of depression and PTSD (and not PGD) above and beyond negative affect regulation strategies (i.e., rumination) in relatives of deceased persons (i.e., Study 1). Study 2, among relatives of missing persons, yielded similar results, except that positive affect regulation strategies explained significant amounts of variance in symptom-levels of PGD and PTSD (and not depression levels) above and beyond rumination. These findings were partly in line with previous research indicating the associations between affect regulation strategies with depression levels in non-clinical samples (Raes et al., 2009, 2012, 2014). We extended prior work by also exploring the association between affect regulation strategies and PGD and PTSD levels.

Because the current study relied on different samples (i.e., relatives of recently deceased persons versus relatives of long-term missing person) and measures, we analysed the samples separately. However, the differences in measured used may explain the differences in findings. The examined response styles represent cognitions in response to affects, which may explain why the positive affect regulation strategies explained a significant amount of variance above and beyond rumination in outcome measures containing more cognitive and affective symptoms (Raes et al., 2012). More specifically, the depression measure used in Study 1 contained solely cognitive and affective symptoms and in Study 2 also somatic symptoms; the PGD measure used in Study 2 contained a greater variety of cognitive and affective grief reactions compared with the PGD measure used in Study 1. Nevertheless, the finding that we obtained similar patterns of results across the two samples supports the generalizability of the associations across people confronted with different types of losses.

Rumination has been frequently identified as a maladaptive strategy to regulate negative affect post-loss (Eisma et al., 2015; Morina, 2011; Nolen-Hoeksema et al. 1994). Our findings contrast with traditional grief theories suggesting that positive emotions following loss may signal denial and avoidance (Bowlby, 1980; Freud, 1957; Kübler-Ross, 1973) and accord with more recent theoretical and empirical work stressing the beneficial role of experiencing positive emotions post-loss (Bonanno & Keltner, 1997; Fredrickson, 2001; Garland et al., 2010; Ong et al., 2006; Tweed & Tweed, 2011).

Our results indicate, among others, that increase use of positive thoughts about affective experience, one's own qualities, and favourable life circumstances to regulate positive affects (i.e., enhancing rather than dampening of positive affect) is uniquely associated with lower levels

of emotional problems after loss. This accords with different grief theories that emphasize that engagement in activities that are potentially pleasurable and the ability to experience and maintain positive affect during bereavement are important for coming to terms with loss (Folkman, 2001; Stroebe & Schut, 2001).

Some psychotherapeutic approaches, such as complicated grief treatment (Shear & Gribbin Bloom, 2017) and cognitive behavioural therapy (Bryant et al., 2014) have been used to effectively target disturbed grief. Complicated grief treatment, even more explicitly than cognitive behavioural therapy, includes elements intended to strengthen positive affect, for instance retrieving positive memories and pursuing pleasurable and satisfying social relationships. Nevertheless, many grief interventions predominantly focus on alleviating negative affect with relatively little attention for savouring of positive affect (see Doering & Eisma, 2016 and Boelen & Smid, 2017 for overviews). Development of additional interventions that address both negative *and* positive affect regulation strategies might yield greater treatment effects (Boelen, 2016; Carl, Soskin, Kerns, & Barlow, 2013). Examples of potential effective interventions are mindfulness-based interventions. Key to mindfulness-based interventions is the development of decentering awareness (Germer, Siegel, & Fulton, 2013). Decentering includes the metacognitive ability to disengage from negative thoughts or feelings, by observing them as mental events in a wider context (Teasdale et al., 2002). From the perspective of the broaden-and-build theory (Fredrickson, 1998, 2001) decentering awareness may broaden one's attention, which in turn may lead to more flexible thinking styles and positive emotions (e.g., compassion; Garland et al., 2010). Results from pilot studies showed the potential effectiveness of mindfulness-based treatment for reducing depression and PTSD levels among bereaved people (O'Connor, Piet, & Hougaard, 2014; Thieleman, Cacciatore, & Hill, 2014). Future studies are needed to further explore the potential beneficial effect of this type of grief treatment.

Limitations

Several limitations of the current study should be noted. Firstly, because of the explorative nature of our study and the small sample sizes, our findings should be interpreted with caution. Future studies should replicate our findings before firm conclusions can be drawn. Second, our cross-sectional research design precludes any causal inferences about the associations between the variables. Thirdly, different measures were used to assess psychopathology levels in the current study, which limits the comparability of our findings between the two samples. In addition, throughout this article we use the term PGD to refer to persistent and disabling grief reactions, whereas in previous studies persistent complex bereavement disorder (PCBD; APA, 2013) and complicated grief (CG; Shear et al., 2011) have also been used to denote persistent and disabling grief responses. We used the PGD scale (which has not yet been thoroughly validated and has only been used in Dutch research) and ICG-r to assess PGD severity levels. This limits the

comparability between the findings obtained in the two samples, and with studies using different conceptualizations of persistent and disabling grief reactions, for instance, studies using PCBD criteria according to DSM-5 (APA, 2013) or more recently proposed guidelines (i.e., diagnostic prototypes (First, 2012) for PGD in the ICD-11 (Maercker et al. (2013); see also Mauro et al. (2017)). Furthermore, although we did not intend to identify clinical cases of PGD (which would require expert clinical interviewing), participants were not all bereaved for more than 6 months and, therefore, could not meet the proposed time criterion for PGD (Maercker et al., 2013; Prigerson et al., 2009). Caution is therefore warranted for generalizing our findings to clinical samples.

Fourthly, our study relied solely on self-reports, which may lead to an overestimation of psychopathology levels (Engelhard et al., 2007). Fifthly, dependent variables (PGD, depression, and PTSD levels) were strongly correlated, which may not seem to support the use of separate statistical models. However, previous factor analytic studies showed similar high correlations among these constructs, but also emphasized that these constructs are distinguishable (Boelen & van den Bout, 2005; O'Connor, Lasgaard, Shevlin, & Guldin, 2010).

Conclusion

The current study explored positive affect regulation strategies in people exposed to the recent death or long-term disappearance of a significant other. Our findings suggest that elevated tendencies to dampen positive affect, reduced tendencies to savour positive affect, and dwelling on negative affect (i.e., brooding) are associated with increased symptom-levels of PGD, depression, and PTSD in two samples confronted with the loss of a loved one. Future research among clinical samples is needed to further explore adaptive and maladaptive regulation of positive affect in the onset, maintenance, and treatment of psychopathology levels post-loss.

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ACKNOWLEDGEMENTS

We would like to thank all participants and collaborators (i.e., the “Vereniging Achterblijvers na Vermissing”, “AVROTROS Vermist”, Victim Support the Netherlands, Child Focus, and the Dutch police).

6

Grief rumination mediates the association between self-compassion and psychopathology in relatives of missing persons

Lenferink, L.I.M., Eisma, M.C., de Keijser, J., & Boelen, P.A. (2017). Grief rumination mediates the association between self-compassion and psychopathology in relatives of missing persons. *European Journal of Psychotraumatology*, *8*(sup6), 1378052.

ABSTRACT

Background

The disappearance of a loved one is a unique type of loss, also termed "ambiguous loss", which may heighten the risk for developing prolonged grief (PG), depression, and posttraumatic stress (PTS) symptoms. Little is known about protective and risk factors for psychopathology among relatives of missing persons. A potential protective factor is self-compassion, referring to openness toward, and acceptance of one's own pain, failures, and inadequacies. One could reason that self-compassion is associated with lower levels of emotional distress following ambiguous loss, because it might serve as a buffer for getting entangled in ruminative thinking about the causes and consequences of the disappearance (coined "grief rumination").

Objective

In a sample of relatives of missing persons we aimed to examine (1) the prediction that greater self-compassion is related to lower symptom-levels of PG, depression, and PTS and (2) to what extent these associations are mediated by grief rumination.

Method

Dutch and Belgian relatives of long-term missing persons ($N = 137$) completed self-report measures tapping self-compassion, grief rumination, PG, depression, and PTS. Mediation analyses were conducted.

Results

Self-compassion was significantly, negatively, and moderately associated with PG, depression, and PTS levels. Grief rumination significantly mediated the associations of higher levels of self-compassion with lower levels of PG ($a*b = -0.11$), depression ($a*b = -0.07$), and PTS ($a*b = -0.11$). Specifically, 50%, 32%, and 32% of the effect of self-compassion on PG, depression, and PTS levels respectively, was accounted for by grief rumination.

Conclusions

Findings suggest that people with more self-compassion experience less severe psychopathology, in part, because these people are less strongly inclined to engage in ruminative thinking related to the disappearance. Strengthening a self-compassionate attitude using, for instance, mindfulness-based interventions, may therefore be a useful intervention to reduce emotional distress associated with the disappearance of a loved one.

Keywords: bereavement, loss, trauma, missing person, compassion, repetitive thinking

Highlights

- This is the first study that examined the associations between self-compassion and emotional distress in the context of grief and loss.
- Relatives of missing persons with more self-compassion experience less emotional distress.
- The buffering effect of self-compassion on emotional distress may be explained by its dampening effect on ruminative thoughts related to the disappearance.

The death of a significant other is one of the most painful life events one can experience and may give rise to psychopathology, including posttraumatic stress (PTS; 11.8%; Onrust & Cuijpers, 2006), depression (21.9%; Onrust & Cuijpers, 2006), and symptoms of prolonged grief (PG; 9.8%; Lundorff, Holmgren, Zachariae, Farver-Vestergaard, & O'Connor, 2017). People confronted with a potential traumatic loss (e.g., homicide or suicide) are particularly susceptible to elevated post-loss psychopathology levels (Kristensen, Weisæth, & Heir, 2012). The long-term disappearance of a close relative is a unique type of potentially traumatizing loss (also coined an "ambiguous loss"; Boss, 2006). The psychological consequences of this loss for those left behind have barely been researched (see for an overview Lenferink, de Keijser, Wessel, de Vries, & Boelen, 2017a).

There is some evidence that relatives of missing persons are at heightened risk to develop psychopathology. For instance, prevalence rates of clinically significant symptom levels of PG (23.3%), depression (68.5%), and PTS (67.1%) were high among people whose relative disappeared in the context of political repression on average 13 years earlier (Heeke, Stammel, & Knaevelsrud, 2015). Another study also showed high prevalence rates of PG (47.0%) and PTS (23.1%) among people whose relative disappeared due to various reasons (e.g., voluntarily missing or presumed homicide without a body) on average 16 years earlier (Lenferink, van Denderen, de Keijser, Wessel, & Boelen, 2017b). To the best of our knowledge, no studies have yet examined variables associated with psychopathology among relatives of missing persons that could potentially be modified in treatment. Gaining insights in these variables is important for the development and refinement of treatment options.

A growing body of research suggests that self-compassion is positively associated with well-being (for an overview: Zessin, Dickhäuser, & Garbade, 2015) and negatively associated with depression (e.g., Costa & Pinto Gouveia, 2011; Gilbert, McEwan, Matos, & Rivis, 2011; Kuyken et al., 2010; Neff, Pisitsungkagarn, & Hseih, 2008; Raes, 2010, 2011; van Dam, Sheppard, Forsyth, & Earleywine, 2011, Roemer et al., 2009), anxiety (e.g., Costa & Pinto Gouveia, 2011; Gilbert et al., 2011; Neff 2003; Neff et al., 2007; Raes, 2010; Roemer et al., 2009; van Dam et al., 2011), and (posttraumatic) stress (e.g., Dahm et al., 2015; Costa & Pinto Gouveia, 2011; Gilbert et al., 2011; Hiraoka et al., 2015; Raque-Bogdan, Ericson, Jackson, Martin, & Bryan, 2011; Thompson & Waltz, 2008). Self-compassion is defined as the tendency to be open to one's own pain and suffering, to experience feelings of kindness toward oneself, to recognize that one's experience is part of a common human experience, and to take an understanding, non-judgmental attitude toward one's failures and inadequacies (Neff, 2003).

Being self-compassionate seems particularly helpful for people who have faced potentially traumatic events, such as the death of a significant other. Studies have shown that people exposed to traumatic events who showed higher levels of self-compassion reported lower levels of PTSD both concurrently (Dahm et al., 2015; Hiraoka et al., 2015; Thompson & Waltz, 2008) and

longitudinally (Hiraoka et al., 2015; Zeller, Yuval, Nitzan-Assayag, & Bernstein, 2015). Furthermore, a laboratory study showed that a self-compassionate attitude toward a previous adverse life event can be induced and, once induced, leads to less emotional distress during the retrieval of memories of this event (Leary, Tate, Adams, Allen, & Hancock, 2007). Finally, clinical trials with PTSD patients have shown that self-compassion can be increased in treatment and promotes recovery from trauma (Beaumont, Galpin, & Jenkins, 2012; Hoffart, Øktedalen, & Langkaas, 2015; Kearney et al., 2013).

Strengthening of a self-compassionate attitude is an important aspect of mindfulness-based treatments (MacBeth & Gumley, 2012). Mindfulness and self-compassion are both rooted in Buddhist traditions and conceptually related. However, the targets of mindfulness and self-compassion differ. In general, mindfulness refers to present moment awareness to any inner experiences, whereas self-compassion is targeted at embracing one's own suffering (Neff & Dahm, 2015). Preliminary findings of small clinical trials among bereaved people indicate that mindfulness-based approaches might be equally beneficial for targeting psychopathology levels in bereaved people as in non-bereaved people who suffer from similar complaints, such as depression (O'Connor, Piet, & Hougaard, 2014; Thieleman, Cacciatore, & Hill, 2014). However, the link between self-compassion and psychopathology after bereavement, including PG symptoms, has, to the best of our knowledge, never been studied.

Although there is evidence that self-compassion is related to positive outcomes in people confronted with adverse life events, and it is conceivable that self-compassion has similar beneficial effects for bereaved people, less is known about mechanisms that may underlie the association between self-compassion and psychopathology (Raes, 2010). Exploring these mechanisms could further our understanding about the role of self-compassion in recovery from loss and trauma and could help to improve interventions fostering self-compassion in the treatment of trauma- and loss-related distress.

One possible explanation for the beneficial role of self-compassion in dealing with traumatic events is that self-compassion is associated with engagement with, rather than avoidance of painful thoughts, memories, and feelings (Leary et al., 2007; Thompson & Waltz, 2008; Zeller et al., 2015). Accordingly, some researchers have argued that self-compassionate people "may be more likely to experience a natural process of exposure to trauma-related stimuli" (Thompson & Waltz, 2008, pp. 558). Exposure to loss-related stimuli is a critical ingredient of effective grief treatment (Bryant et al., 2014). Similarly, theoretical (Boelen, van den Hout, & van den Bout, 2006; Maccallum & Bryant, 2013; Stroebe & Schut, 1999) and empirical work (Boelen & van den Bout, 2010; Boelen, de Keijser, & Smid, 2015; Eisma et al., 2013; Schnider, Elhai, & Gray, 2007) emphasized that strategies to avoid and minimize engagement with painful feelings and thoughts associated

with the loss are key to the onset and maintenance of psychopathology following the loss of a loved one.

One such avoidance strategy is repetitive negative thinking about the causes and consequences of the loss, also coined "grief rumination" (Eisma et al., 2014a). In contrast to Nolen-Hoeksema's (2001) view, rumination may not be a maladaptive confrontational coping style, but could instead serve to refrain from admitting the loss and adjusting to it (Boelen, 2006; Stroebe et al., 2007). For example, ruminative thinking about why the loss occurred, how it could be prevented, and how best to respond to it could suppress more painful loss-related thoughts (e.g., about the true consequences of the loved one never coming back; Boelen, 2006; Eisma et al., 2013; Stroebe et al., 2007).

Grief rumination may concern different issues, including (1) counterfactuals about the loss (i.e., imagining alternative realities in which the loss could have been prevented), (2) reactions of others to the loss, (3) the unfairness of the loss, (4) the meaning and consequences of the loss, and (5) thoughts about one's (emotional) reactions to the loss (Eisma et al., 2014b). Research demonstrated that the first four types of grief rumination were concurrently and/or longitudinally related to elevated symptom levels of PG and/or depression levels. Interestingly, ruminative thinking about one's emotional reactions to the loss was unrelated to PG and depression levels concurrently, but predicted less PG and depression levels over time (Eisma et al., 2015).

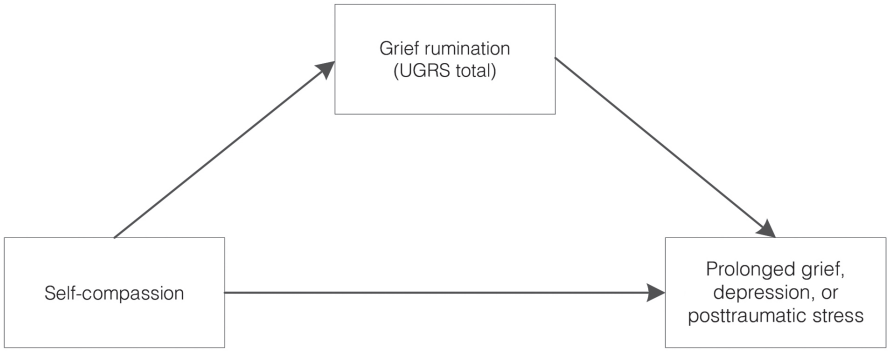
It has been repeatedly shown that rumination is concurrently and longitudinally linked to elevated PG, depression, and PTS levels following loss (Eisma et al., 2012, 2013, 2014b, 2015; Eisma & Stroebe, in press; Ito et al., 2003; Morina, 2011; Nolen-Hoeksema, Parker, & Larson, 1994). It has been proposed that rumination might also be an important cognitive strategy that causes and/or maintains psychopathological symptomatology in relatives of missing persons (Boss, 2006; Heeke et al., 2015; Lenferink, Wessel, de Keijser, & Boelen, 2016). To our knowledge, this notion has never been studied. According to the goal-discrepancy theory, ruminative thoughts reflect concerns and goals that have not yet been attained. Furthermore, those who have more extreme or unattainable goals may be more inclined to ruminate (Ehring & Watkins, 2008; Martin & Tesser, 1989). Given that the disappearance of a loved one is inherently linked to uncertainties (e.g., not knowing whether the person suffered or is alive or dead) that are uncontrollable (Boss, 2006), more than natural losses (e.g., caused by illness) disappearances may give rise to pervasive ruminative thinking.

It has been argued that elevated self-compassion might serve as a buffer for getting entangled in rumination, thereby preventing the exacerbation of emotional distress (Leary et al., 2007; Thompson & Waltz, 2008; Zeller et al., 2015). Accordingly, cross-sectional studies have shown that people with higher levels of self-compassion are less inclined to ruminate (Neff, 2003; Svendsen, Kvernenes, Wiker, & Dundas, 2016). Two other studies supported the mediating effect of rumination

in the relation between self-compassion and depression and anxiety (Krieger, Altenstein, Baettig, Doerig, & Holtforth, 2013; Raes, 2010).

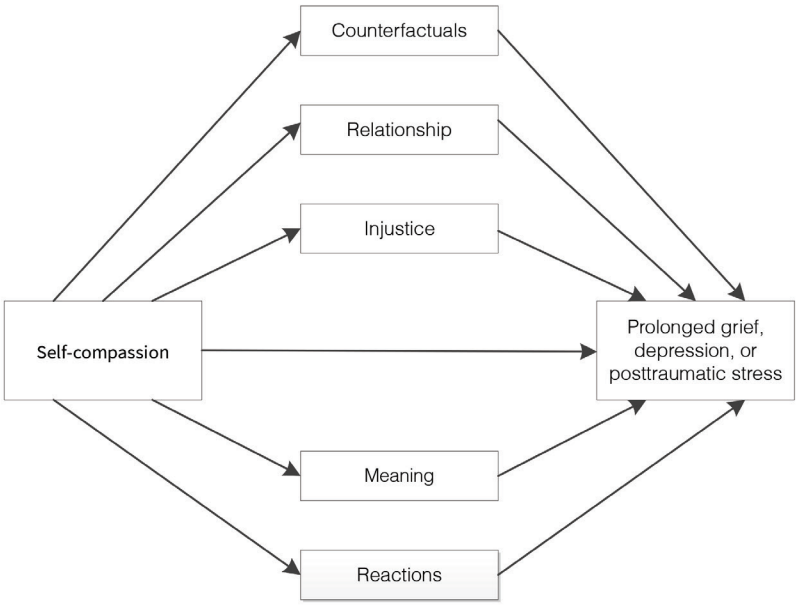
The current study was concerned with self-compassion, grief rumination, and psychopathology among relatives of missing persons. Specifically, first, we tested the prediction that higher levels of self-compassion were related to lower PG, depression, and PTS levels (Hypothesis 1). Because there is evidence that self-compassion is equally related to different types of symptoms (e.g., depression, anxiety, and stress; MacBeth & Gumley, 2012) we had no hypotheses with regard to the relative strength of the associations of self-compassion with PG, depression, and PTS levels. Second, we tested the prediction that the associations between self-compassion and post-loss psychopathology would be mediated by grief rumination (Hypothesis 2; see Figure 1). An additional aim of our study was to explore to what extent different subtypes of grief rumination mediate the association between self-compassion and psychopathology. Based on prior work (Eisma et al., 2015), we expected that rumination concerning the counterfactuals about the disappearance, reactions of others to the disappearance, the unfairness of the disappearance, and the meaning and consequences of the disappearance, partially mediated the association between self-compassion and post-loss psychopathology levels (Hypothesis 3; see Figure 2).

Figure 1. Single-mediation models



Note. We examined the potential mediating effect of grief rumination in the association between self-compassion and levels of prolonged grief (model 1), depression (model 2), and posttraumatic stress (model 3).

Figure 2. Multiple-mediation models



Note. We examined the potential mediating effect of subtypes of grief rumination in the association between self-compassion and levels of prolonged grief (model 1), depression (model 2), and posttraumatic stress (model 3).

METHODS

Procedures

Data were used from an ongoing study about correlates and treatment of psychopathology in relatives of missing persons (Lenferink et al., 2016, 2017b). Adults fluent in Dutch whose spouse, family member, or friend was missing for at least three months were eligible to participate. Data for the current study were collected between July 2014 and July 2016. The following definition of a missing person was used in the current study: "Anyone whose whereabouts is unknown whatever the circumstances of disappearance. They will be considered missing until located and their well-being or otherwise established" (Association of Chief Police Officers, 2010, pp.15). Participants were recruited via invitation letters sent by different collaboration partners (i.e., (peer-) support organizations and the editorial office of a Dutch television show about missing persons), a website of the research project, and media-coverage. In addition, people signing up for the study were asked to invite others. The ethical board of the University of Groningen approved this study. All participants gave written informed consent.

Measures

PG levels were assessed with the Inventory of Complicated Grief (ICG), which measures 19 grief reactions (Boelen, van den Bout, de Keijser, & Hoijtink, 2003; Prigerson et al., 1995). The ICG is together with the PG-13 a frequently used measure to assess grief reactions (Bui et al., 2015). We used the ICG, because a validated Dutch translation of the PG-13 was not available at the time that the current study took place. Items were reformulated such that they referred to the disappearance (e.g., "I feel I have trouble accepting the disappearance"). Participants rated the presence of symptoms during the preceding month on 5-point scales (0 = "never" to 4 = "always"). Cronbach's alpha in the current study was .92.

Depression severity was assessed with the 30-item Inventory of Depressive Symptomatology-Self-Report (IDS-SR; Rush, Gullion, Basco, Jarrett, & Trivedi, 1996). Each item represents a depressive symptom (e.g., "Falling asleep"), with four answer options ranging from 0 to 3 (e.g., answer option 0 = "I never take longer than 30 minutes to fall asleep"). Participants were instructed to select one option that best described how frequently they experienced the symptom during the past 7 days. Cronbach's alpha in the current study was .93.

PTS was assessed with the 20-item PTSD Checklist for DSM-5 (PCL-5; Blevins, Weathers, Davis, Witte, & Domino, 2015; Boeschoten, Bakker, Jongedijk, & Olff, 2014). The disappearance was the anchor-event; participants rated to what extent they experienced each symptom (e.g., "In the past month, how much were you bothered by feeling very upset when something reminded you of the events that are associated with the disappearance?") on 5-point scales ranging from 0 = "not at all" to 4 = "extremely". Cronbach's alpha in the current study was .95.

Self-compassion was assessed with the 24-item Dutch Self-Compassion Scale (SCS; Neff, 2003; Neff & Vonk, 2009). Participants rated how often they behave in the stated manner on 7-point scales ranging from 1 = "almost never" to 7 = "almost always" (e.g., "When I'm feeling down I tend to obsess and fixate on everything that's wrong" reverse scored). Cronbach's alpha in the current study was .88.

Grief rumination was assessed with the Utrecht Grief Rumination Scale (UGRS; Eisma et al., 2014b). Participants rated how often they experienced 15 ruminative thoughts (i.e., "How frequently in the past month did you… [ruminative thought]") on 5-points scales ranging from 1 ="never" to 5 = "very often". The total score was used in the current study as well as the scores on the five three-item subscales. The subscale "Counterfactuals" assesses counterfactual thinking about alternative past realities, related to the disappearance (e.g., "… analyze whether you could have prevented his/her disappearance"; α = .78), the "Relationship" subscale includes thoughts related to social responses to the disappearance (e.g., "…query whether you receive the right support from family members"; α = .77), the "Injustice" subscale includes thoughts about the unfairness of the disappearance (e.g., "…think about the unfairness of this disappearance"; α

= .73), the "Meaning" subscale taps into thoughts about the consequences and meaning of the disappearance (e.g., "…think how your life has been changed through his/her disappearance"; α = .83), and the "Reactions" subscale assesses thoughts related to negative (emotional) reactions following the disappearance (e.g., "…did you try to understand your feelings about the disappearance"; α = .76). Cronbach's alpha for the total UGRS score was .91.

Higher scores on each measure were indicative of higher levels of psychopathology, self-compassion, and grief-related ruminative thinking. The wording that referred to "death" or "stressful event" in the above mentioned measures were adapted to refer to the "disappearance". The psychometric properties of all measures used in the current study are (at least) adequate (Blevins et al., 2015; Boelen et al., 2003; Eisma et al., 2012, 2014b; Neff, 2003; Rush et al., 1996).

Statistical analyses

Because the sum scores on the PG, depression, and PTS measures did not meet the assumption of normality (based on histograms and skewness and kurtosis values) non-parametric (Spearman's rho) correlations were calculated between all variables. Mediation analyses were performed using the PROCESS plug-in for SPSS (Hayes, 2013). First, three single-mediation models were estimated with self-compassion as independent variable and PG, depression, or PTS levels as dependent variables, and the UGRS total score (denoting grief rumination) as mediator. Second, three multiple mediation models were tested using the same independent and dependent variables, and total scores of the 5 UGRS scales as mediator variables. In a prior study using the same data, time since disappearance (in years) and dichotomized kinship to the missing person (0 = missing person is child or spouse, 1 = other) were found to be significantly associated with levels of PG, depression, and PTSD (Lenferink, Wessel, & Boelen, submitted). Therefore, these two variables were included as covariates in the mediation models.

Unstandardized regression coefficients were estimated for each path of the mediation model. Path *a* reflects the effect of the independent variable (i.e., self-compassion) on the mediator (i.e., grief rumination total or subscale scores) while controlling for the covariates, path *b* represents the effect of the mediator on the dependent variable (i.e., PG, depression, or PTS levels) while controlling for the independent variable (and covariates), path *c* is the total effect of the independent variable on the dependent variable, and path *c'* is the direct effect of the independent variable on the dependent variable while controlling for the effect of the mediator(s) and covariates. Bias-corrected 95% bootstrap confidence intervals (BC 95% CI) for the indirect effect (*a*b*) of the independent variable on the dependent variable through the mediator(s) were computed based on 5000 bootstrap resamples. These indirect effects were considered statistically significant when zero was not included in the BC 95% CI. The proportion of the effect of the independent variable on the dependent variable explained by the mediator(s) was

calculated by using the following formula: 1 – *c'/c* (MacKinnon, Fairchild, & Fritz, 2007). Less than 5% of the data was missing per item. Missing data were therefore imputed with the mean.

RESULTS

Preliminary analyses

Socio-demographic and loss-related characteristics of the 137 participants are displayed in Table 1.

Table 1. Characteristics of the participants (*n* = 137)

Gender, *N (%)*	
Men	45 (32.8)
Women	92 (67.2)
Age (years), *M (SD)*	57.9 (14.1)
Educational level, *N (%)*	
Low	32 (23.4)
Middle	45 (32.9)
High	60 (43.8)
Lost relative is, *N (%)*	
Partner/spouse	18 (13.1)
Child	44 (30.7)
Parent	14 (10.2)
Sibling	31 (22.6)
Other family member	29 (21.2)
Other	3 (2.2)
Number of years since loss, *M (SD)*	15.2 (16.9)
Type of disappearance, *N (%)*	
Criminal act	44 (32.1)
Voluntarily	33 (24.1)
Accident	33 (24.0)
No specific suspicion	27 (19.7)
Unique victims	90 (65.7)
Recruitment via	
Editorial office of T.V.-show about missing persons	36 (26.3)
Peer support organizations	31 (22.6)
Non-governmental support organization	21 (15.3)
Family or friends	37 (27.0)
Other	12 (8.8)

Table 2. Zero-order correlations between all variables (n = 137)

	2	3	4	5	6	7	8	9	10
1. Prolonged grief	.70***	.77***	-.35***	.79***	.62***	.63***	.69***	.68***	.60***
2. Depression		.83***	-.41***	.64***	.51***	.57***	.41***	.60***	.52***
3. Posttraumatic stress			-.46***	.70***	.55***	.57***	.54***	.63***	.58***
4. Self-compassion				-.29**	-.21*	-.25**	-.24**	-.32***	-.18*
5. UGRS total					.85***	.76***	.80***	.81***	.84***
6. UGRS Counterfactuals						.49***	.68***	.56***	.67***
7. UGRS Relationship							.44***	.60***	.60***
8. UGRS Injustice								.56***	.56***
9. UGRS Meaning									.62***
10. UGRS Reactions									

Note. UGRS = Utrecht Grief Rumination Scale; * $p < .05$; ** $p < .01$; *** $p < .001$.

Correlations between variables are presented in Table 2. Self-compassion was inversely related to scores on the measures of psychopathology and grief rumination. Grief rumination scores were all positively related to the indices of psychopathology.

Single-mediation analyses

Table 3 shows the results of the mediation analyses with relevant covariates (i.e., kinship to the missing person and time since disappearance) being taken into account. It was found that higher levels of self-compassion were significantly associated with lower tendencies to ruminate about the disappearance (a = -0.14) and lower symptom levels of PG (c = -0.22), depression (c = -0.25), and PTS (c = -0.34). Furthermore, higher tendencies to ruminate were significantly related to increased symptom levels of PG (b = 0.79), depression (b = 0.51), and PTS (b = 0.75) when taking self-compassion into account. Zero was not included in the BC 95% CI's of the indirect effects indicating that grief rumination significantly mediated the associations of higher levels of self-compassion with lower levels of PG (a^*b = -0.11), depression (a^*b = -0.07), and PTS (a^*b = -0.11). In total, 50%, 32%, and 32% of the effect of self-compassion on PG, depression, and PTS levels, respectively, was accounted for by grief rumination.

Multiple-mediation analyses

Table 3 also shows the results of the multiple-mediation models. The three UGRS subscales "Relationship", "Injustice", and "Meaning" emerged as unique mediators of the association between self-compassion and symptom levels of PG and PTS. For depression, two mediators were significant, namely UGRS "Relationship" and UGRS "Meaning". All mediators combined, accounted for 59%, 36%, and 35% of the effects of self-compassion on symptom levels of PG, depression, and PTS, respectively.

Table 3. Mediation analyses (n = 136[1])

Model	Mediator	*a*	*b*	Total effect *c*	Direct effect *c'*	Unique indirect effect *a*b* (BC 95% CI)	MacKinnon effect size
Prolonged grief							
Single mediation	UGRS total	-0.14**	0.79***	-0.22***	-0.11**	-0.11* (-0.20, -0.03)	.50
Multiple mediation	UGRS counterfactuals	-0.02	0.16		-0.09**	<-0.01 (-0.03, 0.01)	.59
	UGRS relationship	-0.03*	1.18***			-0.03* (-0.07, -0.01)	
	UGRS injustice	-0.03*	1.73***			-0.05* (-0.11, -0.01)	
	UGRS meaning	-0.04***	0.90**			-0.04* (-0.08, -0.01)	
	UGRS reactions	-0.02	0.15			<-0.01 (-0.03, 0.01)	
Depression							
Single mediation	UGRS total	-0.14**	0.51***	-0.25***	-0.17***	-0.07* (-0.13, -0.03)	.32
Multiple mediation	UGRS counterfactuals	-0.02	0.39		-0.16***	-0.01 (-0.05, 0.01)	.36
	UGRS relationship	-0.03*	0.85*			-0.02* (-0.07, <-0.01)	
	UGRS injustice	-0.03*	0.05			<-0.01 (-0.04, 0.03)	
	UGRS meaning	-0.04***	1.02*			-0.04* (-0.10, -0.01)	
	UGRS reactions	-0.02	0.30			-0.01 (-0.04, 0.01)	
Posttraumatic stress							
Single mediation	UGRS total	-0.14**	0.75***	-0.34***	-0.23***	-0.11* (-0.19, -0.03)	.32
Multiple mediation	UGRS counterfactuals	-0.02	0.34		-0.22***	-0.01 (-0.05, 0.01)	.35
	UGRS relationship	-0.03*	1.04*			-0.03* (-0.09, <-0.01)	
	UGRS injustice	-0.03*	1.02*			-0.03* (-0.10, <-0.01)	
	UGRS meaning	-0.04***	0.94			-0.04* (-0.10, <-0.01)	
	UGRS reactions	-0.02	0.51			-0.01 (-0.05, 0.01)	

Note. [1]= one participant did not fill in the date of the disappearance of his/her loved one and was therefore excluded from the mediation analyses; UGRS = Utrecht Grief Rumination Scale; *a* = the effect of X on M while controlling for the covariates; *b* = the effect of the mediator on Y, while controlling for X, other mediators, and covariates; *c* = the effect of X on Y; *c'* is the direct of X on Y while controlling for the mediator(s) and covariates; BC 95% CI = Bias corrected bootstrap confidence intervals (5000 resamples); * $p < .05$; ** $p < .01$; *** $p < .001$.

DISCUSSION

The aim of the current study was to investigate the associations between self-compassion and psychopathology levels among people confronted with the long-term disappearance of a loved one. Moreover, we examined whether these associations were mediated by grief rumination.

In support of our first hypothesis, self-compassion was significantly and negatively associated with PG, depression, and PTS levels, even when taking grief rumination and relevant background variables into account. We found moderate correlations (r_s = -.35 to -.46) between self-compassion and psychopathology levels; correlations appear to be lower than the overall large correlation between psychopathology and self-compassion (r = -.54) found in a meta-analysis (MacBeth & Gumley, 2012). These previous large effects were predominantly based on non-clinical (student) samples. Correlations between self-compassion and PTS ranged from small to large in trauma exposed samples (Dahm et al., 2015; Hiraoka et al., 2015; Thompson & Waltz, 2008).

In line with our second hypothesis, the single-mediation models showed that the associations between self-compassion and PG, depression, and PTS levels were mediated by grief rumination. In other words, people who have stronger tendencies to approach their emotional pain in an open and understanding way (i.e., more self-compassion), are less likely to get entangled in ruminative thoughts related to the disappearance which, in turn, attenuates psychopathology levels. This accords with and extends previous research indicating that rumination mediates the linkage between self-compassion and depression and anxiety (Krieger et al., 2013; Raes, 2010). Although we did not test it directly, these findings also seem to support previous research denoting that self-compassion may be viewed as a way of exposure to internal threats (Krieger et al., 2013; Thompson & Waltz, 2008) and rumination as a way of avoidance of painful aspects of the loss (Eisma et al., 2014a; Stroebe et al., 2007).

Partly in accordance with our third hypothesis, the multiple-mediation analyses indicated that ruminative thoughts about the meaning of the disappearance (i.e., UGRS meaning) and reactions of others to the disappearance (i.e., UGRS relationship) were significant mediators of the associations between self-compassion and PG, depression, and PTS levels. Thoughts about the unfairness of the disappearance (i.e., UGRS injustice) only significantly mediated the associations between self-compassion and PG and PTS levels. Contrary to our expectations, ruminative thinking about alternative past realities in which the person did not disappear (i.e., UGRS Counterfactuals) was not a significant mediator. Although zero order correlations between counterfactual thinking and self-compassion and psychopathology were statistically significant, these associations disappeared once other variables (e.g., other rumination subtypes, time since disappearance) were taken into account. Previous studies also have shown that some types of rumination uniquely mediate the effect of self-compassion on depression and/or anxiety while

others do not. For instance, Raes (2010) found that brooding but not reflection mediated the association between self-compassion and depression; Krieger et al. (2013) found that symptom-focused rumination but not self-focused rumination mediated the linkage of self-compassion with depression. Taken together, our findings suggest once more that some forms of ruminative thinking are more maladaptive than others when it comes to dealing with the loss of a loved one (Eisma et al., 2015; Stroebe et al., 2007).

It is important to note that grief rumination was a partial mediator in the current study. This indicates that other phenomena may also explain the associations between self-compassion and post-loss psychopathology levels including, for instance, adaptive emotion-regulation skills (e.g., the ability to tolerate unpleasant emotion; Diedrich, Burger, Kirchner, & Berking, 2016). Of course, many other factors may potentially be involved in the association between self-compassion and post-loss psychopathology (see for an overview Barnard & Curry, 2011). Following previous studies (cf. Krieger et al., 2013; Raes, 2010) we examined the mediating role of grief rumination in the association between self-compassion and psychopathology levels. However, we cannot preclude the possibility that self-compassion mediates the association between rumination and psychopathology. Future studies, preferably using a longitudinal design and clinical samples, may further study the temporal relationships between these constructs, for example by using cross-lagged analyses (cf. Krieger, Berger, & Holtforth, 2016).

A number of limitations need to be taken into account while interpreting our findings. First and foremost, the cross-sectional design precludes drawing conclusions about temporal precedence and causality. Second, due to composition of our convenience sample (i.e., a Dutch and Belgian community sample of relatives of long-term missing persons), the generalizability of our findings to relatives of missing persons in general, but also people confronted with other loss-experiences, with clinical levels of psychopathology, who are more recently bereaved, and have other cultural backgrounds may be limited. Third, self-report measures instead of diagnostic interviews were used, which may have led to overestimation of psychopathology levels (Engelhard et al., 2007). In addition, we did not take the nested structure of our data (i.e., the 137 participants were related to 90 unique missing persons) into account in the analyses. Because our sample included only a relatively small number of relatives of the same missing person, it is unlikely that this has increased the chance of Type I error. Lastly, our sample size was relatively small (see Fritz & MacKinnon, 2007), which may have increased the risk of Type II error.

Despite these limitations, the current study is the first that examined the associations between self-compassion and psychopathology levels in the context of grief and loss. Although more research is needed to draw firm conclusions about these associations among people confronted with the loss of a loved one, the results of our study suggest that fostering self-compassion in treatment might reduce post-loss psychopathology levels by reducing ruminative tendencies.

There is evidence that enhancement of self-compassion and reduction of ruminative tendencies are mechanisms of change in mindfulness-based treatments for recurrent depression (van der Velden et al., 2015). Moreover, other third-wave cognitive behavioural treatments, such as compassion-focused therapy and acceptance and commitment therapy, are increasingly being used to target these phenomena (Beaumont & Hollins Martin, 2015; Dindo, Van Liew, & Arch, 2017). The first results of small trials among bereaved persons showed that mindfulness-based interventions might be effective in reducing symptoms of depression (O'Connor et al., 2014; Thieleman et al., 2014), anxiety, and PTS (Thieleman et al., 2014). Although non-significant reductions were found in PG and PTS levels in O'Connor et al.'s (2014) study and some participants in Thieleman et al.'s study (2014) reported increased symptomatology following the mindfulness-based treatment, it may be valuable to further study the potential effectiveness of these interventions, because the current treatment-of-choice, cognitive behavioural therapy, results in clinically relevant change in PG levels in at most 50% of people with PG (Doering & Eisma, 2016). Consequently, based on the current findings, we recommend further exploration of the value of self-compassion in recovery from loss.

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ACKNOWLEDGEMENTS

We would like to thank all participants and collaborators (i.e., the “Vereniging Achterblijvers na Vermissing”, “AVROTROS Vermist”, Victim Support the Netherlands, Child Focus, and the Dutch police). Also thanks to the funding agencies The Victim Fund, Foundation for the stimulation of bereavement research, and the University of Groningen who supported this work.

6

7

I've changed, but I'm not less happy: Interview study among nonclinical relatives of long-term missing persons

Lenferink, L.I.M., de Keijser, J., Piersma, E., & Boelen, P.A. (in press). I've changed, but I'm not less happy: Interview study among nonclinical relatives of long-term missing persons. *Death Studies*. doi: 10.1080/07481187.2017.1347213

ABSTRACT

Twenty-three nonclinical relatives of long-term missing persons were interviewed. Patterns of functioning over time were studied retrospectively by instructing participants to draw a graph that best described their pattern. Patterns most frequently drawn were a recovery and resilient/stable pattern. Participants were also asked to select five out of fifteen cards referring to coping strategies, which they considered most helpful in dealing with the disappearance. Acceptance, emotional social support, mental disengagement, and venting emotions were most frequently chosen. This study provided some indication of coping strategies that could be strengthened in treatment for those in need of support.

Keywords: bereavement, complicated grief, coping, disenfranchised grief, resilience

The disappearance of a significant other, also described as an ambiguous loss (Boss, 2006), is a unique type of loss to deal with due to the intersection of grief and holding on to hope (Wayland, Maple, McKay, & Glassock, 2016). Many researchers have suggested that a prolonged and debilitating grief process following the disappearance of a loved one is a normal response to an abnormal situation (Betz & Thorngren, 2006; Boss, 2006; Hollander, 2016). Interestingly though, several studies have shown that a considerable number of relatives of long-term missing persons do not suffer from long-lasting psychological complaints, such as prolonged grief, posttraumatic stress, or depression (see for an overview Lenferink, de Keijser, Wessel, de Vries, & Boelen, 2017).

Historically, experiencing little distress following the death of a significant other has been considered as a dysfunctional grief response. For example, Freud (1957) emphasized the importance of acknowledging and expressing painful emotions as part of the 'grief work'. Kübler-Ross (1973) proposed that people who failed to go through 'the stages of grief', including anger and depression, experienced a distorted grief process. Bowlby (1980) considered the absence of overt grief reactions post-loss as denial, which eventually could lead to a delayed grief response (Horowitz, 1976; Worden, 1991).

These traditional views on grief processes have been challenged by, among others, two longitudinal studies using latent class growth modeling (Galatzer-Levy & Bonanno, 2012; Melhem, Porta, Shamseddeen, Walker Payne, & Brent, 2011). These studies showed that the most common response to the loss of a significant other is characterized by a consistent pattern of little distress over time ("*resilient/stable pattern*"). Another common pattern emerging in these longitudinal studies is characterized by an increase in distress immediately post-loss followed by a gradually decrease ("*recovery pattern*"). Interestingly, Galatzer-Levy and Bonanno (2012) also examined depression levels from pre-loss to several years post-loss and identified an additional pattern characterized by high depression levels pre-loss followed by a significant decrease of depression levels post-loss (i.e., "*improved pattern*"). Only a small minority displayed a maladaptive pattern characterized by prolonged severe distress following loss (Galatzer-Levy & Bonanno, 2012; Melhem et al., 2011). Noteworthy, these studies did not support a delayed grief pattern.

Although the interest in different grief trajectories has increased, literature regarding different response patterns following the long-term disappearance of a significant other is lacking. Consequently, in the current study we sought to explore whether the same *adaptive* response patterns (i.e., *"resilient/stable", "recovery",* and *"improved"* pattern) can be identified among relatives of missing person. To this end we focused on nonclinical relatives of missing persons only (i.e., people with minimal psychological complaints). We used a pragmatic approach to study response patterns, by asking participants retrospectively about their patterns of functioning from one year prior to the disappearance until now (cf. Burr & Klein, 1994; Mancini, Sinan, & Bonanno, 2015a).

We also aimed to enhance our understanding about coping strategies involved in adaptive responses to the disappearance of a significant other, in terms of the theory of stress, appraisal, and coping developed by Lazarus and Folkman (1984). This theory is widely used to describe different coping strategies used by people to deal with varying stressful situations. Two categories of coping strategies are distinguished in this theory, namely problem-focused (i.e., directed at managing and changing the stressor) and emotion-focused coping strategies (i.e., directed at managing the emotional consequences of the stressor). A survey study among Pakistani relatives of missing persons showed that problem-focused coping strategies are associated with less psychological distress, while emotion-focused coping strategies are linked to increased psychological distress (Basharat, Zubair, & Mujeeb, 2014).

Although the categorization of emotion- and problem-focused coping strategies has been frequently used, researchers have argued that whether a coping strategy is adaptive or maladaptive depends on the specific situation (Folkman, 1984). Furthermore, reviews evaluating the factor structure of measures to assess coping strategies (Litman, 2006; Skinner, Edge, Altman, & Sherwood, 2003) propose a multidimensional factor structure instead of a two-factor structure. Taking all this into consideration, we decided to study individual coping strategies, rather than categories of coping strategies.

Specifically, in the current interview-study we aimed to explore what types of coping strategies were deemed helpful by nonclinical people confronted with the disappearance of a significant other. Furthermore, we were interested in the way in which particular coping strategies helped in dealing with the long-term disappearance of their loved one. In doing so, we were able to examine how relatives of missing persons explain the usefulness of particular coping strategies, which results in more in-depth information about the role of coping strategies.

By exploring patterns of functioning and helpful coping strategies of nonclinical relatives of missing persons, we aimed to gain understanding in how people adaptively deal with this potential stressor. In addition, these insights were considered useful input for developing interventions to prevent as well as reduce chronic complaints.

METHOD

Participants

Table 1 displays the characteristics of the participants. Twenty-three persons, related to 15 unique long-term missing person cases, participated in the interview-study. Three persons experienced the disappearance of a spouse, five the disappearance of a child, two the disappearance of a parent, and thirteen the disappearance of a sibling. Fifteen (65.2%) participants were women.

The age of the participants ranged between 26 and 81 years (M = 59.7; SD = 11.6 years). The disappearance took place between 1 and 70 years earlier (M = 23.0; SD = 20.0 years). On average, the duration of the interviews was 39 minutes.

Procedure

Participants were recruited as part of an on-going research project examining correlates and treatment of psychopathology in relatives of missing persons (Lenferink, Wessel, de Keijser, & Boelen, 2016; Lenferink, van Denderen, de Keijser, Wessel, & Boelen, 2017). Inclusion criteria for participation in the present interview-study were: 1) confronted with the disappearance of a spouse, sibling, child, or parent since at least three months earlier, 2) score below thresholds for clinical levels of PGD, PTSD, and depression (see Lenferink et al. (2016) for details about the thresholds and the respective questionnaires), 3) 18 years or older, 4) residing in the Netherlands, 5) fluent in Dutch language, and 6) gave consent in a previous survey-study to be contacted for future research.

At the time of the start of this interview-study, 95 participants were included in the survey study, 25 of whom fulfilled the inclusion criteria for the interview-study. These 25 participants were approached by telephone and briefly informed about the procedure and aims of the interview-study. Except for two participants (i.e., they declined because they did not want to relive the disappearance), all participants were interested and therefore received an information letter via regular mail. After 10 working days the participants were again approached by telephone and all participants were still willing to participate. The individual face-to-face interviews could take place at the participants' home, a public place, or at the university. All participants chose to undergo the individual interview at their home. A trained interviewer conducted the interviews in May, June, and July 2015. Each participant gave written consent. Approval from a local ethical review board was obtained for conducting the current study.

Table 1. Characteristics of the participants

Response ID	Gender	Age in years	Time since disappearance in years	The missing person is a ... of the participant	Case number	Presumed reason of disappearance
103000	Woman	52	30	Sibling	103	Victim of crime
103004	Man	46	30	Sibling	103	Victim of crime
108000	Man	49	16	Sibling	108	Accident
109000	Woman	57	3	Child	109	Accident
109001	Man	59	3	Child	109	Accident
109002	Woman	26	3	Sibling	109	Accident
114000	Woman	51	28	Sibling	114	Victim of crime
131002	Woman	67	43	Sibling	131	Left voluntarily
131003	Woman	61	43	Spouse	131	Left voluntarily
140001	Woman	70	69	Parent	140	Accident
147000	Man	59	3	Sibling	147	Left voluntarily
147006	Woman	58	3	Sibling	147	Left voluntarily
147010	Woman	55	3	Sibling	147	Left voluntarily
150002	Woman	58	1	Spouse	150	Left voluntarily
156000	Man	79	14	Child	156	Victim of crime
156002	Woman	81	14	Child	156	Victim of crime
157001	Man	57	32	Sibling	157	Left voluntarily

Table 1 (continued). Characteristics of the participants

157002	Man	59	32	Sibling	157	Left voluntarily
159002	Woman	66	36	Sibling	159	Victim of crime
160000	Man	73	70	Parent	160	Victim of war/disaster
177000	Woman	59	16	Spouse	177	Accident
190000	Woman	58	18	Sibling	190	Left voluntarily
191001	Woman	72	20	Child	191	Victim of crime

Note. Participants with the same casenumber are related to the same missing person

Interview

The semi-structured interviews were conducted following a pre-developed interview scheme (see Supplemental Material A). The interview scheme was pilot tested with one volunteer (not included in this study) who experienced the long-term disappearance of a child. No major issues were raised during this pilot interview. The interview scheme consisted of several parts. In the first part the participant was asked to draw a graph of the discourse of their functioning from one year prior to the disappearance up to the day of the interview following the example of Burr and Klein (1994). Time since disappearance (ranging from "one year prior to the disappearance" to "the current moment") was presented on the x-axis and level of functioning (ranging from 0 to 100%) was presented on the y-axis. The interviewer introduced this graph task as follows: *"Please indicate in the graph what the progress of the impact of the disappearance has been on your psychological, social, occupational, and physical functioning, starting from one year before the disappearance until now."* Prior to this question the interviewer gave an example of this task. After drawing the graph, the interviewee was asked to elaborate on the graph he/she has drawn (e.g., "*We see that on, (indicate time point of raise/drop of functioning) this time point your functioning [raised/dropped], what would you consider the reason for that?").* As described in the interview scheme (see Supplemental Material A) the interviewer used specific prompts to encourage the participant to elaborate more, including "*Could you tell me a little bit more about that?" or "Could you give an example of that?"*

The second part of the interview was based on a procedure previously described by Paap et al. (2014) and consisted of a card-sorting task. Fifteen cards that represented all coping strategies distinguished in the 15 subscales of the modified Dutch version of the COPE (Carver, Scheier, & Weintraub, 1989; i.e., the COPE easy; Kleijn, Heck, & Waning, 2000) were presented to the participants. The following 15 cards were presented: *Planning*, *Denial*, *Acceptance*, *Positive reinterpretation*, *Restraint-coping*, *Humor*, *Instrumental social support*, *Turning to religion*, *Active-coping*, *Behavioural disengagement*, *Suppression of competing activities*, *Venting emotions*, *Emotional social support*, *Mental disengagement*, and *Substance use*.

Each coping strategy was illustrated by two examples. The examples were two randomly chosen items of the respective subscale of the COPE easy. Because ranking all 15 coping strategies was considered to be too demanding, the participants were asked to select five coping strategies that, in their opinion, had been most helpful in coping with the disappearance ever since it occurred (cf. Paap et al., 2014). After having selected five cards, they were asked to rank order the five cards from most helpful to least helpful. Finally, they were asked to explain for each chosen coping strategy why this strategy was helpful to them (i.e., "Please indicate for each chosen card the reasons why you considered this coping strategy as helpful."). In case the participant was unable to select five cards, he/she was instructed to choose as many cards as he/she wanted.

Data analysis

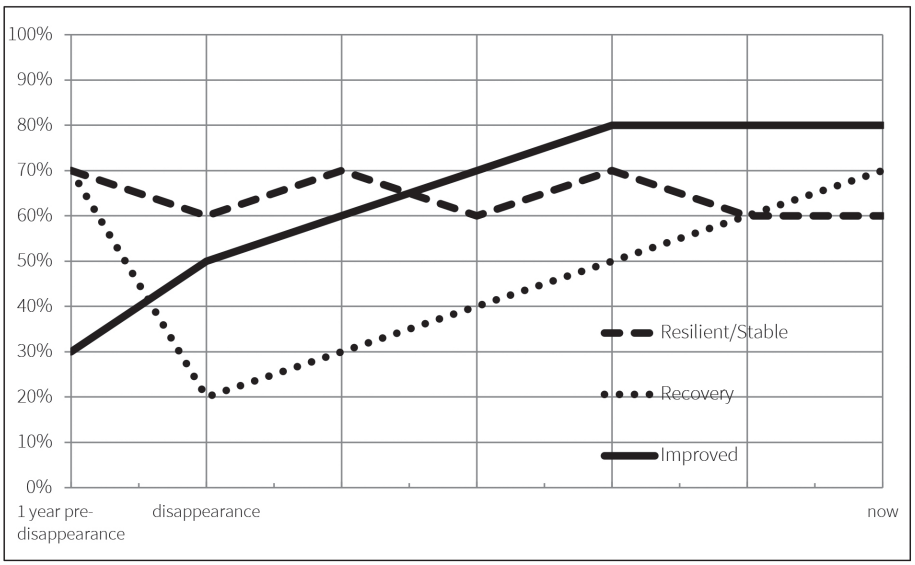
All interviews were recorded and transcribed verbatim. Both parts of the interview (i.e., graph task and card-sorting task) yielded quantitative (in terms of frequencies of patterns drawn and frequency of selected cards) and qualitative data (in terms of texts explaining why the participant drew a pattern and selected the cards).

Quantitative data analysis

Graph task

A pattern that was characterized by stable high functioning (≥ 60% over time) was labelled as a *resilient/stable* pattern. A pattern that was characterized by an initial drop (below 60%) in functioning immediately post-disappearance, followed by an gradual increase in functioning over time to healthy levels (≥ 60%) was labelled as a *recovery* pattern. Graphs that showed an increase (≥ 60 %) in functioning post-disappearance compared with functioning levels of < 60 one year prior to the disappearance were labelled as an *improved* pattern. The threshold of 60 was based on the Global Assessment of Functioning (GAF) scale, in the fourth edition of the Diagnostic Statistical Manual of Mental Disorders (DSM-IV) that defines scores of ≥ 60 as moderate to minimal impairment (American Psychiatric Association, 2000). See Figure 1 for an example of each pattern drawn by participants. After each pattern was labelled, the frequency of each pattern was summed.

Figure 1. Examples of patterns drawn by participants.



Card-sorting task

The frequency of each chosen coping strategy in the card-sorting task was summed (i.e., the unweighted sumscore). The weighted sumscore was based on the rank order of each chosen coping strategy. Following Paap et al. (2014) the coping strategy with the highest ranking was scored as '5', the second ranking as '4', the third ranking as '3', the fourth ranking as '2', and the fifth ranking as '1'.

Qualitative data analysis

The qualitative data of the graph task and card-sorting task were analysed by two independent raters. Inconsistencies between the raters were resolved by discussion. Methods from grounded theory were used to analyse the data (Corbin & Strauss, 2008). The following consecutive steps were performed for each part (i.e., graph task and card-sorting task) of the interview. First, irrelevant texts (i.e., texts that were not related to the question asked by the interviewer) were removed. Second, the remaining text was divided into meaningful and coherent smaller pieces of texts, referred to as "units". Third, the units were interpreted and then labelled with a theme that reflected the content of the unit (also referred to as 'open coding'; Corbin & Strauss, 2008). Then these themes were reanalysed to check for similarities between the labels. Adjustments of labels were made if desirable. In the final step the so-called 'axial coding' took place. During this phase, overarching major themes across the subthemes were identified. See Table 2 for an example of the analytic process.

Table 2. Examples showing how units were coded into main themes and subthemes for the graph task

Unit (participant's characteristics)	Subtheme	Main theme
"And then I went to live here, already in '74, so I've been here for quite some years, so that's very stable in my life. Maybe that had something to do with it as well, that I look for some stability, look for peace in nature and that that works well for me. Not for everybody, I think, but it does, for me." (ID 131003, woman, 61 years old, and spouse went missing 43 years earlier)	Stability in life/relationships has a protective function	Intrapersonal consequences
"And at this point here [at the time of disappearance] I've rated it at 40%. It had a lot of impact on me then… a lot of disquiet, in the form of alertness. When I would bike through the city, or past the station, you'd have a lot of people there. He didn't have a roof over his head anymore, so he slept outside a lot. He slept in nature, he liked that. But because of that, when I'd see people sleeping, who looked a little like him, I'd wonder: 'Could that be my brother?'. So I was constantly very alert about running into him somewhere." (ID 147010, woman, 55 years old, and sibling went missing 3 years earlier)	To think you are recognizing the missing person leads to psychological distress	Whereabouts of the missing person
"The realization only came when my mother passed away. My mother passed away quite young. I was 20 so you went from one thing into the other. And really, only after that moment it turns out that you had quite a tumultuous childhood. Where there was little joy…I was 15 when my mother was diagnosed with a terminal illness and was 20 when she passed away. And only after that you realize all that's happened." (ID 140001, woman, 70 years old, and parent went missing 69 years earlier)	Deaths of significant others trigger reminders of the disappearance	Life events
"And I just kept working [circa one year after the disappearance] and I found it very pleasant to just keep on working. Flip the switch." (ID 147006, woman, 58 years old, and sibling went missing 3 years earlier)	Work offers distraction/ reward	Occupational consequences
"Then [the moment of the disappearance] you have a breakdown, but after that you do go like: "I have to be there…" then you're like: "This can't be" but you immediately pick back up where you left off like, "I have to be there for the children."" (ID 103000, woman, 52 years old, and sibling went missing 30 years earlier)	Taking care of others is satisfying	Social support

RESULTS

Graph task

The majority of the participants (*n* = 15) drew the 'recovery' pattern, followed by the 'stable/resilient' pattern (*n* = 7). One participant drew an 'improved' pattern.

In what follows, the most frequently mentioned main themes and subthemes are presented that derived from the qualitative analyses of the data regarding the reasons why the participants drew the graph the way they did. Examples of questions asked by the interviewer are: *"What would you consider the reason for starting the graph at this point and not higher or lower?"* and *"What would you consider the reason for drawing a peak/drop at this point?"*. Table 2 shows examples of the coded units for the graph task.

"Intrapersonal consequences", "Whereabouts of the missing person", "Life events", "Social support", and *"Occupational consequences"* were the most frequently main themes that arose from explanations for the way the participants drew the graph (see Table 3 for an overview of the main themes and subthemes derived from the qualitative analysis). The subtheme experiencing stability in life and in relationships with others was mostly mentioned as protective factor against negative intrapersonal consequences. Self-blame (e.g., thought about how the participant could have prevented the disappearance) was the most frequently mentioned subtheme that had negative intrapersonal consequences. The second main theme included statements related to the *Whereabouts of the missing person*. A common subtheme was the emotional distress experienced by participants once they realized that their spouse, child, parent, or sibling may never return. In addition, thinking to recognize the missing person gave rise to psychological distress. With respect to *Life events*, formation or expansion of family life (e.g., getting married and/or having children) was frequently mentioned by participants as reason for increase in functioning. However, experiencing the death of a significant other triggered reminders of the missing person. *Social support* was described in several ways; receiving social support or offering social support to others was described as helpful, while some mentioned family issues related to the disappearance, lack of social support from spouse, or having difficulty discussing the disappearance with others as barriers. *Occupational consequences* post-disappearance included absence from work. Notably, others stated that they continued working, because they experienced it as a helpful distraction.

Table 3. Overview of main themes and subthemes of the qualitative analysis of the graph task

Main themes (number of occurrences/number of participants referring to main theme)	Subthemes (number of participants referring to subtheme)
Intrapersonal consequences (26/14)	Stability in life/relationships has a protective function (6)
	Thoughts of self-blame are not helpful (3)
	Thinking about the missing person dominates daily life (2)
	Posttraumatic growth (2)
	Breaking the negative thought cycle by resuming daily activity (2)
	Surreal sensation (2)
	Sleep problems (2)
Whereabouts of the missing person (21/14)	Realizing the missing person may never return can lead to psychological distress (7)
	To think you are recognizing the missing person leads to psychological distress (3)
	To think that psychological symptoms of the missing person caused the disappearance fosters acceptance (3)
	To abandon the urge to find a reason or explanation for the disappearance is experienced as helpful (2)
	Looking/searching for the missing person gives a sense of satisfaction (2)
	To realize that the loved one has likely passed away gives a sense of peace (2)
Life events (18/13)	Family formation/expansion is experienced as positive (9)
	Deaths of significant others trigger reminders of the disappearance (5)
	Other adverse life events trigger reminders of the disappearance (2)

Table 3 (continued). Overview of main themes and subthemes of the qualitative analysis of the graph task

Social support (18/12)	To have an effective support network (3)
	The disappearance dominated family life (3)
	Taking care of others is satisfying (2)
	Tension within the family is experienced as an obstacle (2)
	Lack of support from the partner (2)
	To find it difficult to discuss the disappearance with acquaintances (2)
Occupational impact (15/11)	Absence from work (5)
	Work offers distraction (5)
	To experience difficulties during work (3)
Practical / judicial matters (11/10)	It is a burden to arrange practical/judicial matters (4)
	It is a burden to arrange presumptive proof of death (2)
	Media attention is experienced as burdensome (2)
To seek professional support (10/6)	The use of psycho-pharmaceuticals is effective (4)
	Psychological treatment is effective (3)
General description of the impact of the disappearance (7/7)	The disappearance of a loved one is a life-changing experience (6)
Learning to cope with the disappearance (7/5)	Learning to tolerate negative emotions (5)
To conduct rituals (5/4)	Having a memorial service is experienced as helpful (2)

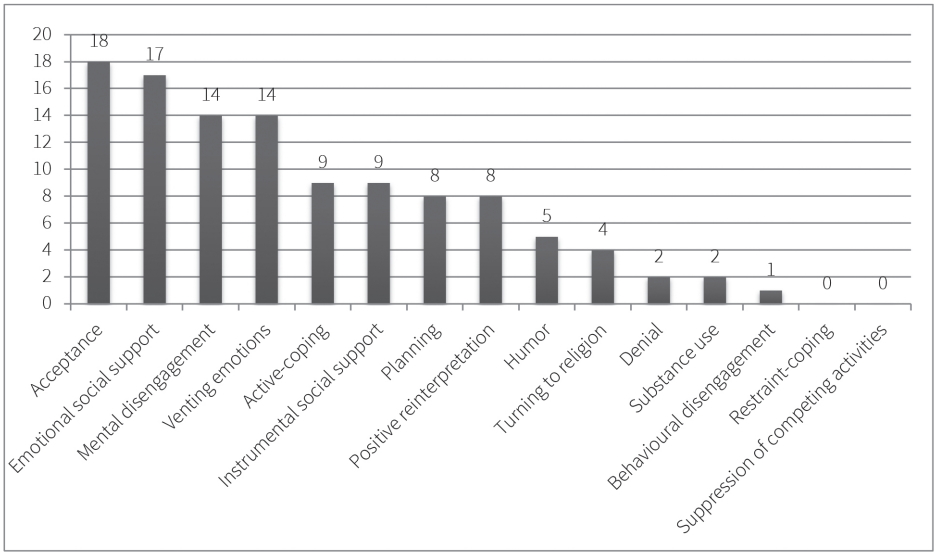
Note. Subthemes that were mentioned once are not displayed in this Table.

Card-sorting task

All participants selected five coping strategies that they perceived as most helpful in dealing with the disappearance, except for two participants (i.e., they only found two or four coping strategies helpful).

See Figure 2 for the frequency of chosen coping strategies. The following four coping strategies were most frequently chosen: *Acceptance* (*n* =18), *Emotional social support* (*n* = 17), *Mental disengagement* (*n* = 14), and *Venting emotions* (*n* = 14).

Figure 2. Frequency of chosen coping strategies



When taking the rank order of the chosen coping strategies into account, Acceptance was considered as most helpful, followed by Emotional social support, Venting emotions, and Mental disengagement respectively. Because the rank order did not reveal meaningful differences in the interpretation of the results, details are not shown.

Most participants described *Acceptance* as dealing with the fact that the disappearance is out of their control and that it is impossible to influence it. Others described *Acceptance* as a process of adjustment that develops over time.

"But that it's always ahead of me, that it completely dictates my life, that is not the case anymore. It's just going past me. It's become a part of my life and I can handle that reasonably well, now. So I've learned to live with it in that way." (ID 103000, woman, 52 years old, and sibling went missing 30 years earlier)

Some participants also mentioned barriers towards acceptance, such as thinking about the reason of disappearance, feeling sorry for yourself, and thinking about the presumed perpetrator who made their loved one disappear.

Talking with friends and family members was the most commonly mentioned source of *Emotional social support*. Receiving emotion social support was perceived as helpful because it eased the emotional burden.

"To find moral support is a way for me to vent. It's useful to me in the sense that it bothers me less, then." (ID 147000, man, 59 years old, and sibling went missing 3 years earlier)

Mental disengagement was described in various ways, including engaging in activities that were perceived as joyful prior to the disappearance, going back to work, doing household tasks, and performing physical exercise. The most prominent reason for disengagement was preventing to get stuck in repetitive negative thinking.

"Then you're working on other things and you don't have to think, that's also something. And that isn't because you can't handle it in that moment, but it lets you get on with things. Especially in the beginning." (ID 191001, woman, 72 years old, and child went missing 20 years earlier)

Sadness (i.e., crying) and anger were the most reported emotions when describing *Venting emotions*. Expressing these negative emotions with others felt comforting. Some stated that it was inevitable to express their emotions; others had more difficulties with venting their emotions.

"Only later, with seeking help… I had to really learn that. To learn to have an eye for the emotional side of things." (ID 114000, woman, 51 years old, and sibling went missing 28 years earlier)

The next four most helpful coping strategies were: *Active coping*, *Instrumental social support*, *Planning*, and *Positive reinterpretation*. *Active coping* was predominantly described as taking care of practical and legal issues associated with the disappearance and as taking control over one's own life. *Instrumental social support* was provided by mental healthcare professionals and alternative medical practitioners, in the form of mental support, and by the police and justice system in the form of practical support in the process of searching for the missing person. *Planning* included taking action to search for the missing person, which comforted the participants. *Positive reinterpretation* was described in terms of finding meaning or focusing on positive things since the disappearance, including more intense social contact, more self-awareness, and more interest in others. See Table 1 in the Supplemental material B for the main themes and subthemes that derived from the qualitative analyses of the card-sorting task.

Other notable results

During the interviews several notable recurrent themes emerged that were not specifically linked to the reasons why the participants drew the graph the way they did or to one specific card of the card-sorting task. Firstly, none of the participants explicitly stated that the missing person was dead. Instead they stated, for example: *"You really know concretely that he's no longer with us." (ID 109000, woman, 57 years old, and child went missing 3 years earlier)* and *"Above that, I was more and more starting to think, like: "He is completely gone.""(ID 131002, woman, 67 years old, and sibling went missing 43 years earlier).*

A second notable recurrent theme was searching for the missing person. In some cases active searches were perceived as helpful, because 'doing everything in your power' gave them peace. Others were passively searching for the missing person, for example, by paying attention to places/situations that were linked to the missing person in the hope to find the missing person, or by searching for the missing person in their dreams:

"I did find myself very restless, also an inner restlessness. And that definitely stayed like that for a while, just after the disappearance, and that plummeted for me (from 40% at the time of disappearance to 20% within one year of the disappearance), because, in my sleep, I dreamed about my brother. Then I went to look for him. Then, while asleep, I fell down the stairs, which caused brain damage." (ID 147010, woman, 55 years old, and sibling went missing 3 years earlier)

Another recurrent theme was performing rituals to honour the missing person, for example by organizing a memorial service, visiting the place where the missing person was last seen, or dedicating activities to the missing person, to illustrate this: *"But I've run a few marathons, since then, and I do those while wearing a shirt with a picture of my sister printed on it. You know, in that way, I am also doing it a little bit for her." (ID 103000, woman, 52 years old, and sibling went missing 30 years earlier)*

Finally, some participants noticed that they found a new sense of personal strength following the disappearance. As a result of the disappearance they gained new experiences and insights and learned how to appreciate the little things in life. Multiple participants mentioned that the disappearance of a significant other is a life-changing experience, but not necessarily a devastating experience:

"This sort of thing hits you in the core of your being, it cannot not affect you… the outcome is, I often think, that another person comes out of it. But not a less happy person." (ID 10800, man, 49 years old, and sibling went missing 16 years earlier).

DISCUSSION

By conducting semi-structured interviews with 23 nonclinical relatives of long-term missing persons we aimed to gain insights into a) patterns of functioning over time and b) helpful coping strategies to deal with the disappearance of a close family member or spouse. In line with previous studies among people confronted with the death of a significant other (Galatzer-Levy & Bonanno, 2012; Melhem et al., 2011), we identified three adaptive patterns as a result of the graph task: a 'recovery' pattern, 'stable/resilient' pattern, and 'improved' pattern. Fifteen participants drew the recovery pattern that is characterized by an initial drop in functioning immediately post-disappearance followed by a significant stable increase in functioning. The most common pattern following the death of a loved one (Galatzer-Levy & Bonanno, 2012; Melhem et al., 2011), namely the stable/resilient pattern, was drawn by seven participants. This pattern is characterized by high level of functioning with no significant peaks or drops. One participant drew the 'improved' pattern, characterized by an increase in functioning post-disappearance compared with pre-disappearance.

We extended prior research about self-identified trajectories (cf. Mancini, Bonanno, & Sinan, 2015b; Mancini et al., 2015a), by examining exploratory why the participants drew the graph the way they did. Based on the explanations of the participants, a high or increased level of functioning was predominantly explained as a result of experiencing stability in life and relationships, formation or expansion of family life, continuing occupational tasks, and receiving or offering social support. These findings are consistent with previous research showing that interpersonal resources may serve as buffer for emotional distress following the loss of a significant other (Mancini et al., 2015a).

Taken together, the results of the graph task indicated that adaptive response patterns among relatives of missing persons show similarities with adaptive response patterns among bereaved individuals. This contrasts with statements made in the literature claiming that the disappearance of a significant other is "inherently traumatic" (Boss, 2006, p. 4) and is "a never-ending roller-coaster…and takes its toll on family members physically, cognitively, behaviorally, and emotionally" (Betz & Thorngren, 2006, p. 361). However, future studies with larger sample sizes, including nonclinical and clinical relatives of missing persons, need to further examine response patterns of relatives of missing persons, before conclusions can be drawn about prototypical response pattern for relatives of missing persons. Preferably longitudinal analyses of latent classes could be used (cf. Forbes et al., 2016), but also cross-sectional studies whereby participants need to choose one graph that best describe their pattern retrospectively (cf. Mancini et al., 2015a; 2015b) and study the association between factors and distinct trajectories may be a fruitful avenue to pursue.

The card-sorting task provided insights into potentially helpful ways of dealing with the disappearance of a loved one. Acceptance, described as dealing with the fact that the disappearance is out of their control, was considered as the most helpful coping strategy. This accords with the perspective of Boss (2006) who stated that 'tempering mastery', in terms of "learning to live with not knowing", is one of the main goals of relatives of missing person in maintaining a meaningful life. Indeed, learning to accept that some life experiences are uncontrollable and unpredictable may be one of the major challenges faced by relatives of missing persons. Wayland et al. (2016) emphasized that maintaining hope, in a way that goes beyond merely hoping that the missing person would return (e.g., hoping for a better future) is helpful in adjusting to a life where a relative is missing. These perspectives are reminiscent of cognitive behavioural (Boelen, van den Hout, & van den Bout, 2006) and constructivist and meaning-making based approaches (Janoff-Bulman, 1992; Neimeyer, 1998) toward coping with loss and trauma, in which the ability to maintain a positive outlook of the self, life, and the future in the face of adversity, is deemed critical in dealing with this adversity. Accordingly, interventions that help to tolerate uncertainty, maintain hope, and retain positive views are potentially fruitful in the treatment of relatives of missing persons suffering from persistent distress (Boss, 2006; Lenferink et al., 2016; Wayland et al., 2016).

Previous research indicated that a lack of social support might be a risk factor for development of psychopathology post-disappearance (Quirk & Casco, 1994; Robins, 2010). Accordingly, our findings showed that emotional social support, provided by family members and friends, and venting emotions with others were among the most helpful coping strategies. In an extension of the Dual Process Model of coping with bereavement, Stroebe and Schut (1999; 2015) encourage integration of interpersonal factors in research and treatment of grief-related distress. For instance, they stated that differences between family members in their continuing bonds with the deceased and acceptance of the loss may hinder the adaptation process of individual family members. In a similar vein, differences in for instance views on the search for the missing person, the cause of the disappearance, and the fate of the missing person may hinder the adaptation process. Paying attention to interpersonal factors in research and treatment may therefore also be important for relatives of missing persons.

Mental disengagement, described as engaging in social or occupational activities to prevent to get stuck in repetitive negative thinking, was also one of the most chosen helpful coping strategies. The latter contrasts previous studies considering mental disengagement (together with behavioural disengagement and substance use) as a maladaptive avoidant coping style (Carver et al., 1989; O'Connor & O'Connor, 2003). This finding highlights the importance of studying coping strategies situation-specific instead of generic (Folkman, 1984).

The design of the current study did not allow us to explore which factors (e.g., time since disappearance, type of disappearance, intra- and interpersonal factors) were related to specific

patterns of functioning and how the coping strategies were related to the patterns of functioning. This could be examined by using a quantitative design in order to identify protective and risk factors for developing psychopathology post-disappearance. It should also be noted that our sample predominantly consisted of participants who experienced the disappearance of a significant other many years ago. Thus, our findings not necessarily generalize to people recently confronted with a disappearance. For instance, themes as media-attention, searching for the missing person, and organizing practical/judicial matters might have been of more importance to people who experienced the disappearance more recently than for people of whom a relative disappeared decades earlier. Our sample was small and consisted of nonclinical relatives of missing persons, which therefore also limits the generalizability to all nonclinical relatives of missing persons, but also people with clinically relevant psychopathology levels.

To conclude, by exploring patterns of functioning and helpful coping strategies of nonclinical relatives of long-term missing persons, we aimed to gain understanding in how people adaptively deal with this potential stressor. These insights may offer guidelines for more research into this under-researched field and are potentially useful for developing interventions to prevent as well as reduce chronic complaints in relatives of missing persons.

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ACKNOWLEDGEMENT

We would like to thank Melanie Lemmers for conducting the interviews and her assistance with the data-analyses as second rater. Also a special thanks to Steven de Jong and Anna van der Gaast-Witkowska for translating the quotes and interview scheme.

SUPPLEMENTAL MATERIAL A

Interview scheme

Graph task

In a moment I will ask you to draw a graph, which indicates your functioning starting from one year before the disappearance until now. Before we start I would like you to look at an example first, so that you know what you are expected to do.

(Interviewer gives the example graph to the participant)

This is an example of a woman who lost her job in 2001. She was asked to draw in a diagram how the degree of her happiness was influenced by the loss of her job, starting from one year before the loss of her job until now. As you can see, she set the level of her happiness one year before the loss of her job at 90%. She explains that in the year before she lost her job she had a good life and enjoyed her work and her family. You can see that after she lost her job she has been feeling unhappy for three years. The woman explains that she could not find any other job in the first year. After being unemployed for a year, she got a job. She had to get used to this new job at first and she did not feel that it was right for her. She felt lonely and unhappy. After three years, we can see in the diagram that her happiness slowly increased till 50%. The woman stated that she kept bonding with her colleagues, which made her work more pleasant. Then the level of happiness decreased at the time when the company was being reorganized. She noticed that because of that she started suffering from stress. In 2009 she started taking a course and she became a volunteer. Due to that, her self-confidence and her joy of life increased. In 2011 she started her own company, which she still enjoys every day.

You are expected to draw a graph as well.

(Interviewer gives the diagram to the participant)

This is the diagram you are supposed to work with. On the left you see the vertical line ranging from 0 – 100% and at the bottom of the diagram there is a horizontal line indicating time. The diagram starts one year before the disappearance. At this point your loved one went missing. The end of the line is now.

(Interviewer indicates the point where the disappearance took place and writes down the year of the disappearance)

I would like to ask you to indicate in the diagram how big the impact of the disappearance was with respect to your functioning starting from one year before the disappearance until now. The term 'functioning' concerns psychological functioning (such as the presence and the severity of psychological problems), social functioning (such as the degree to which you take part in social activities with friends and family), occupational functioning (such as functioning with respect to work or education), and physical functioning (such as experiencing physical problems).

One hundred percent functioning means that you did not experience any limitations in your functioning related to the disappearance in all the above-mentioned domains, 0% functioning means that you constantly experienced severe limitations in your functioning which were related to the disappearance of your loved one.

Please indicate in the graph what the progress of the impact of the disappearance has been on your psychological, social, occupational, and physical functioning, starting from one year before the disappearance until now

(Let the participant draw the graph)

I would like to discuss the graph with you and to talk about your reasons for drawing the graph in the way you did.

7

(Interviewer discusses the course of the graph with the participant and asks for explanations regarding the course, discussing one or more of the questions described below)

I. We see that you started the graph at this point *(indicate the starting point of the degree of functioning).* What are the reasons that the graph started there and not lower or higher?
II. We see that on, *(indicate time point of drop of functioning)* this time point your functioning dropped, what would you consider the reason for that? *(Prompts: Could you tell me a little bit more about that? Could you give an example of that?)*
III. We see that on, *(indicate time point of raise of functioning)* this time point your functioning raised, what would you consider the reason for that? *(Prompts: Could you tell me a little bit more about that? Could you give an example of that?)*
IV. We see that on, *(indicate the time point at which functioning is stable)* this time point your functioning remained stable, what would you consider the reason for that? *(Prompts: Could you tell me a little bit more about that? Could you give an example of that?)*
V. We see that at this moment your functioning corresponds to this point *(indicate the points that corresponds)* what would you consider the reason for that? *(Prompts: Could you tell me a little bit more about that? Could you give an example of that?)*

Card-sorting task

During the last part of this interview we are going to look into various ways of coping with a stressful experience. You can see fifteen cards in front of you, with a description of a strategy people can use while coping with a stressful event on each card. We call them coping strategies. On each card you will find two examples which give an indication of what one can understand by this coping strategy. Try not to pay too much attention to the example on the cards while answering the following questions. Pay attention to the general description of the strategy in particular.

Please choose 5 cards with the coping strategies that you have been applying while coping with the disappearance of your loved one and which you experienced as the most helpful.

(Let the participant choose 5 cards)

You have chosen the following five cards *(Interviewer reads the five chosen cards out loud).* Please arrange the chosen cards from most helpful to least helpful.

(Let the participant arrange 5 cards from most helpful to least helpful)

You chose the following top 5 *(Interviewer fills in the following top 5)*:

1.
2.
3.
4.
5.

Please indicate for each chosen card the reasons why you considered this coping strategy as helpful.

SUPPLEMENTAL MATERIAL B

Table 1. Overview of main themes of the qualitative analysis of the card-sorting task for the four most frequently chosen coping strategies

Chosen coping strategy	Main themes (number of occurrences/number of participants referring to main theme)	Subthemes (number of participants referring to subtheme)
Acceptance	Description of acceptance (20/15)	Acceptance that there is no possibility of influencing the disappearance (6)
		Learning to live with the disappearance (3)
		Acceptance of the fact that the loved one will not return (2)
		Putting the disappearance into perspective (2)
		Acceptance comes with time (2)
		Acceptance is paradoxical (2)
	Barrier to acceptance (5/5)	n/a
	Presumed reason of the disappearance is related to acceptance (5/5)	Acceptance of voluntary disappearance (4)
	Description of living with a disappearance (4/4)	Taking charge of one's own life (4)
Emotional social support	Description of emotional support from social network (17/13)	Family, friends and/or acquaintances gave moral support (8)
		Understanding from social network (4)
		Discussing the disappearance with others (3)
	Importance of emotional social support (7/7)	Moral support as a way of coping with emotions (3)
	Lack of emotional social support (2/2)	Lack of moral support from parents (2)
	Description of venting emotions (2/1)	n/a
	Description of emotional support from professionals (2/2)	n/a

Table 1 (continued). Overview of main themes of the qualitative analysis of the card-sorting task for the four most frequently chosen coping strategies

Mental disengagement	Description of mental disengagement (13/9)	Participating in enjoyable activities (4)
		Taking care of household chores (2)
		Taking part in individual activities (2)
		Resuming general daily activities (2)
	Reasons for mental disengagement (6/6)	Preventing repetitive (negative) thoughts (4)
Venting emotions	Description of venting emotions (15/9)	Sadness (4)
		Anger (3)
		Expressing emotions through discussing the disappearance with others (2)
		Expressing feelings (2)
	Importance of venting emotions (2/2)	Not expressing emotions has negative consequences (2)
	Reasons for venting emotions (2/2)	Expressing emotions as a personality trait (2)

Note. Main themes and subthemes that were mentioned once are not displayed in this Table. n/a = not applicable because this main theme consisted of subthemes that were only mentioned once.

7

8

Cognitive behavioural therapy for psychopathology in relatives of missing persons: Study protocol for a pilot randomised controlled trial

Lenferink, L.I.M., Wessel, I., de Keijser, J., & Boelen, P.A. (2016). Cognitive behavioural therapy for psychopathology in relatives of missing persons: Study protocol for a pilot randomised controlled trial. *Pilot and Feasibility Studies*, *2*(1), 19.

ABSTRACT

Background It is hypothesized that the grieving process of relatives of missing persons is complicated by having to deal with uncertainty about the fate of their loved one. We developed a Cognitive Behavioural Therapy (CBT) with mindfulness that focuses on dealing with this uncertainty. In this article we elucidate the rationale of a pilot randomised controlled trial (RCT) for testing the feasibility and potential effectiveness of this CBT for reducing symptoms of psychopathology in relatives of missing persons.

Methods A pilot RCT comparing participants of the CBT condition (*n* = 15) with waiting list controls (*n* = 15) will be executed. Individuals suffering from psychopathology related to the long-term disappearance of a loved one are eligible to participate. The treatment consists of eight individual sessions. Questionnaires tapping psychological constructs will be administered before, during, and after the treatment. The feasibility of the treatment will be evaluated using descriptive statistics (e.g., attrition rate). The primary analysis consists of a within-group analysis of changes in mean scores of persistent complex bereavement disorder from baseline to immediately post-treatment and follow-up (12 weeks and 24 weeks post-treatment).

Discussion A significant number of people experience the disappearance of a loved one. Surprisingly, an RCT to evaluate a treatment for psychopathology among relatives of missing persons has never been conducted. Knowledge about treatment effects is needed to improve treatment options for those in need of help. The strengths of this study are the development of a tailored treatment for relatives of missing persons, and the use of a pilot design before exposing a large sample to a treatment that has yet to be evaluated. Future research could benefit from the results of this study.

Trial registration: NTR4732 (The Netherlands National Trial Register (NTR))

Keywords: missing persons, psychopathology, grief, cognitive behavioural therapy

The disappearance of a significant other is a potentially devastating loss, due to the lack of knowledge whether the disappeared is dead or alive. Research on the psychological consequences for relatives of the disappeared is scarce and has mainly been conducted in the context of armed conflicts (Baraković, Avdibegović, & Sinanović, 2013; Campbell & Demi, 2000; Heeke, Stammel, & Knaevelsrud, 2015; Pérez-Sales, Durán-Pérez, & Herzfeld, 2000; Powell, Butollo, & Hagl, 2010; Quirk & Casco, 1994; Robins, 2010; Shalev & Ben-Asher, 2011). These studies indicate that the disappearance of a significant other is associated with elevated levels of posttraumatic stress disorder (PTSD), major depressive disorder (MDD), and/or disturbed grief.

Grief following the death or disappearance of a loved one

Whereas little is known about emotional consequences of disappearance, there is a large body of knowledge about both uncomplicated and disturbed grief after the death of a loved one. Grief after the death of a loved one is typically characterized by transient sadness, preoccupation with the circumstances surrounding the loss, and longing for the deceased (Hall, 2014). People mostly adapt well to the death of a significant other. Nonetheless, about 10% of the bereaved experience disturbed grief (Middleton, Raphael, Burnett, & Martinek, 1998; Prigerson et al., 2009). When grief complaints persist or increase at least 6 months post loss and are associated with distress and impairments in daily functioning, it can be defined as complicated grief (also referred to as prolonged grief disorder; Shear et al., 2011). Although complicated grief partly overlaps with PTSD and MDD, research has shown that these disorders are distinguishable (for overview see Bryant (2014)). Persistent Complex Bereavement Disorder (PCBD) was included in the fifth edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5) as condition for further study (APA, 2013). PCBD encompasses persistent separation distress, preoccupation with the loss and the circumstances of the loss beyond 12 months after the loss (APA, 2013).

There are at least three reasons to assume that grieving the loss of a disappeared relative is more complex and longer lasting than grieving the loss of a deceased relative. To begin with, the on-going uncertainty about the fate of the missing person may lead to preoccupations with his/her potential whereabouts (Blaauw, 2002; Campbell & Demi, 2000). Constantly thinking about the missing person may exacerbate negative emotions, interfere with daily life tasks and lead to exhaustion (Blaauw, 2002; Robins, 2010; Clark, Warburton, & Tilse, 2009). In addition, families of the disappeared are often confronted with financial, emotional, and practical issues for which they receive little professional support (Blaauw, 2002; Campbell & Demi, 2000). Finally, family conflicts, social marginalization and a lack of social support from the community have been considered to increase the psychological burden (Quirk & Casco, 1994; Robins, 2010).

Taken together, compared to individuals bereaved by the death of a loved one, relatives of missing persons might experience more severe PTSD, MDD, and disturbed grief (henceforth

referred to as PCBD operationalized as complicated grief). To the best of our knowledge, this hypothesis was tested in only four quantitative studies (Heeke et al., 2015; Powell et al., 2010; Quirk & Casco, 1994; Zvizdić & Butollo, 2001) and was confirmed in two of them (Powell et al., 2010; Quirk & Casco, 1994). For example, a study among women with unconfirmed and confirmed loss of their husband in a war-related context showed that the former group was more at risk to experience severe MDD symptoms compared to the latter group (Powell et al., 2010). In contrast, two other studies did not show significant differences in the severity of symptoms of psychopathology (Heeke et al., 2015; Zvizdić & Butollo, 2001). For example, relatives of Colombians who had disappeared or died in an armed conflict reported similar levels of PCBD, MDD, and PTSD symptoms (Heeke et al., 2015).

There are reasons for doubting that the results of these studies are applicable to relatives of missing persons in general. Firstly, all four studies were conducted in the context of armed conflicts. The participants were highly traumatized by different war-related stressors (e.g., torture). It might therefore be difficult to distinguish the effect of the disappearance from the effects of other traumatic events. Secondly, the studies were conducted in non-Western samples. In general, cultures may differ in how to deal with loss, but more specifically in how to deal with relatives of missing persons (Robins, 2010). Finally, there is preliminary evidence that relatives who have more hope that their loved one is still alive also experience more severe PCBD complaints (Heeke et al., 2015). Within the context of war-related disappearances the majority of the relatives reported being convinced that their loved one was dead (Heeke et al., 2013; Quirk & Casco, 1994). Therefore, relatives who assume that the disappeared person is dead might suffer less from psychological distress than those who assume that the disappeared person might still be alive. Nevertheless, these comparative studies and other studies (Campbell & Demi, 2000; Pérez-Sales et al., 2010; Robins, 2010; Shalev & Ben-Asher, 2011) show that relatives of missing persons might be susceptible for developing symptoms of PCBD, MDD, and PTSD.

One important step would be to design and evaluate an intervention to target these psychopathological symptoms. To the best of our knowledge, this was only done once; a controlled trial compared the effectiveness of two cognitive behavioural therapy (CBT)-based interventions among women who lost a husband due to death or to disappearance during the Srebrenica massacre (Hagl, Powell, Rosner, & Butollo, 2014). Small to medium pre- to post-treatment effect sizes for both condition were found for disturbed grief and PTSD. However, the methodological drawbacks of this study, including non-random allocation of participants and lack of a threshold of severity of psychopathology as inclusion criterion, need to be taken into account when interpreting the results. Nevertheless, CBT is the treatment of choice for bereavement-related psychopathology (Currier, Holland, & Neimeyer, 2010). CBT might therefore also be effective in the treatment of psychopathology among relatives of missing persons.

CBT for relatives of missing persons

A cognitive behavioural theory of PCBD offers a framework for determining variables that should be targeted in CBT among persons who developed PCBD following the death of a significant other (Boelen, van den Hout, & van den Bout, 2006). This and several other theories about PCBD highlight the important role of negative cognitions and maladaptive behavioural strategies in the development and persistence of PCBD (Boelen et al., 2006; Maccallum & Bryant, 2013; Shear & Shair, 2005). Cognitive variables include negative views on the self ("I am worthless since he/she died") and life ("My life has no purpose since he/she died"), a pessimistic view on the future ("I don't have confidence in the future"), and catastrophic meanings assigned to one's own reactions to the loss ("If I would elaborate on my feelings, I would lose control"). Maladaptive behavioural strategies include anxious avoidance and depressive avoidance (Boelen et al., 2006). The former refers to the avoidance of loss-related stimuli out of fear that confrontation with these stimuli will be unbearable. The latter refers to withdrawal from social, recreational, educational, and/or occupational activities fuelled by the belief that these activities are pointless and/or unfulfilling. Problems with integration of the loss into the autobiographical memory are also associated with PCBD. This results in easily triggered intrusive thoughts, images and memories upon confrontation with loss-related stimuli (Boelen et al., 2006). Studies among bereaved individuals suffering from PCBD showed the beneficial effect of targeting these cognitive and behavioural variables using CBT (Boelen, de Keijser, van den Hout, & van den Bout, 2007; Rosner, Bartl, Pfoh, Kotoučová , & Hagl, 2015).

In addition, symptoms of PCBD have been associated with rumination (Eisma et al., 2015; van der Houwen, Stroebe, Schut, Stroebe, & van den Bout, 2010). Rumination encompasses repetitive thinking about one's negative feelings, their consequences and/or antecedents (Nolen-Hoeksema, 1991). As the disappearance of a person is surrounded by uncertainties, all kinds of repetitive thoughts (e.g., about the whereabouts of the missing person), including ruminative thoughts (e.g., "What am I doing to deserve this?") might add to the exacerbation and maintenance of symptoms of psychopathology among relatives of missing persons (Blaauw, 2002; Clark et al., 2009; Heeke et al., 2015; Robins, 2010). Rumination can be regarded as a form of "maladaptive coping", i.e. an unproductive way to master the consequences of the disappearance. Other forms of maladaptive coping that may be pertinent to recovery from the disappearance of a loved one include substance use and suppression of unwanted thoughts and memories.

Although never studied systematically, it is conceivable that these cognitive and behavioural variables are also involved in the maintenance of psychopathology among relatives of missing persons. For instance, relatives of missing persons might no longer perceive the world as a safe place. As a result, they may experience a reduced sense of control and elevated vulnerability. This poses threats to the view of themselves, life and the future (Campbell & Demi, 2000; Clark

et al., 2009). Relatives may tend to get preoccupied with the missing person and get entangled in repetitive negative thinking and intrusive memories (Campbell & Demi, 2000; Clark et al., 2009; Heeke et al., 2015; Powell et al., 2010). Consequently they could withdraw from previously fulfilling activities. Hoping to find the missing person and at the same time coming to terms with the disappearance can result in conflicting feelings (Holmes, 2008). Holding on to hope that the loved one will return might be used as avoidance strategy to cope with emotions associated with the thought that the separation is permanent (Clark et al., 2009). Some avoid to discuss what might have happened to the missing loved one, fearing that this may be perceived as giving up hope (Families and Friends of Missing Persons Unit, 2010). Active searching may therefore provide distraction from dwelling on the worst-case scenarios (Holmes, 2008).

CBT could give relatives of missing persons insights into how cognitive processes (e.g., thoughts about how they should have prevented the disappearance) affect their emotions (e.g. sadness) and behaviour (e.g., withdrawing from social activities). Unlike treatment for bereavement-related psychopathology, an intervention for relatives of missing persons should not be primarily focused on closure or coming to terms with the irreversibility of the loss. Instead, this treatment ought to be focused on tolerating the ambiguity surrounding the loss and maladaptive repetitive thinking, including thoughts about the whereabouts of the missing person (Boss, 2007; Robins, 2010). Adding elements of mindfulness to CBT might serve this treatment aim. In contrast to focusing on external events (e.g., finding out what happened to the missing person), mindfulness is focused on inner psychological experiences (e.g., one's thoughts, sensations and feelings). Furthermore, mindfulness is not focused on the past or future, but on the present (Segal, Williams, & Teasdale, 2013). Mindfulness-based therapy aims to increase the patient's awareness of his/her inner thoughts, feelings, and bodily sensations in a non-judgemental way through the practice of training in mindfulness meditation (Segal et al., 2013). It has frequently shown to reduce levels of psychopathology (Khoury et al., 2013). A systematic review showed that the beneficial effect of CBT with mindfulness might be, among other variables, due to the reductions of repetitive negative thinking and enhancement of self-compassion (van der Velden et al., 2015). Self-compassion can be viewed as an emotion-regulation strategy (Diedrich, Grant, Hofmann, Hiller, & Berking, 2014; Neff, 2003a) that could play a protective role by preventing to become entangled in negative thoughts and emotions (MacBeth & Gumley, 2012). Although these mindfulness-based cognitive therapies have been tested mainly among persons with recurrent depression, preliminary results showed that this approach could also be effective in treatment of grief-related psychopathology (O'Connor, Piet, & Hougaard, 2014; Thieleman, Cacciatore, & Hill, 2014).

Study objectives

The aim of this pilot study is to evaluate the feasibility and the potential effectiveness of CBT with elements of mindfulness in reducing PCBD, MDD, and PTSD symptoms and enhancing the extent of mindfulness among relatives of missing persons. Based on previous studies, we expect that the treatment effect will be mediated by changes in negative cognitive and behavioural variables plus enhancement of self-compassion.

The feasibility of the pilot RCT will be explored by the evaluation of the: a) specifics of potential participation bias, b) attrition rate, c) methods used and study design, d) treatment fidelity and, e) strengths and suggestions for improvements of the treatment from the perspective of the participant. The primary objective of the analysis of the pilot RCT is to assess changes in mean scores on a measure of PCBD from baseline to one week, 12 weeks, and 24 weeks post-treatment. A within group instead of a between groups analysis will be executed because this will give the most rigorous information about the potential effects of this treatment given the small sample size.

Secondary objectives of the analyses of the pilot RCT are: a) to evaluate whether the mean scores on measures tapping PCBD, MDD, PTSD, and mindfulness of the treatment group differ from the waiting list control group at the post-treatment/post-waiting period assessment when adjusting for the baseline scores, and b) to test whether the treatment effect is mediated by changes in negative cognitive variables (i.e. reductions in negative grief cognitions, intrusive memories, rumination and repetitive negative thinking) and behavioural variables (i.e. avoidance behaviours) and enhancement of self-compassion. In addition, the extent of change in repetitive negative thinking, intrusive memories, and self-compassion in response to the treatment will be explored at micro-level by means of tracking measures tapping these constructs during the treatment.

METHOD

Design

This is a multi-centre two-arm pilot RCT exploring the feasibility and potential effectiveness of CBT for relatives of missing persons suffering from psychopathology, in comparison with a waiting list control group. The intervention group will start with the treatment within one week after randomisation. The waiting list control group will receive the treatment 12 weeks after randomisation. An allocation ratio of 1:1 will be used. The participants will be asked to fill in questionnaires prior to the treatment and one week, 12 weeks, and 24 weeks post-treatment. Participants in the waiting list control group will complete an additional questionnaire one week prior to their actual start of the treatment. In addition, the participant will be asked to complete

a brief questionnaire at the start of each session to assess the potential change in repetitive negative thinking, intrusive memories and self-compassion during the treatment.

Ethics approval

Ethics approval for performing this study has been obtained from the Ethical Committee Psychology at the University of Groningen in the Netherlands (ppo-014-087).

Participants

First, second and third degree (adoption- or step-) family members, spouses and friends of missing persons, who are missing for more than three months, are fluent in written and spoken Dutch, and are 18 years of age or older are eligible to sign up for the study. A missing person is defined as: "Anyone whose whereabouts is unknown whatever the circumstances of disappearance. They will be considered missing until located and their well-being or otherwise established" (Association of Chief Police Officers, 2010, p. 15). The three months criterion was used in a previous study (Tarling & Burrows, 2004) and is also chosen for this study in consultation with representatives of a peer support group and a non-governmental organisation for relatives of missing persons in the Netherlands. Additional inclusion criteria are: a) meeting criteria for PCBD, MDD, and/or PTSD, b) written informed consent, c) absence of mental retardation, d) absence of substance abuse or dependence, e) absence of psychotic disorder, f) no high risk of suicide, and g) no concurrent psychological treatment. In case the missing loved one will be found dead or alive during the period of participation, the participant is offered the opportunity to finish the treatment but will be excluded from further analyses.

Recruitment of participants

All recruitment procedures aim to enrol individuals of whom a loved one has been missing for participation in a survey-study. The survey study aims to explore the psychological consequences for relatives of missing persons. The survey-study and pilot RCT are both part of the same research project. We will recruit participants through several pathways. Firstly, representatives from a television show focused on the search of missing persons and a peer support group will distribute invitation letters to relatives of missing persons. Secondly, Victim Support the Netherlands, a governmental organization in which professionals, mostly social workers, offer practical and legal support to victims, will inform potential participants about the research project. Thirdly, other participants will be recruited via announcements in the media, presentations at meetings for relatives of missing persons and snowball sampling (each participant is asked to invite others). After signing up for the survey-study, the participants will be sent a questionnaire including an information letter and informed consent form for participation in the survey study. The letter

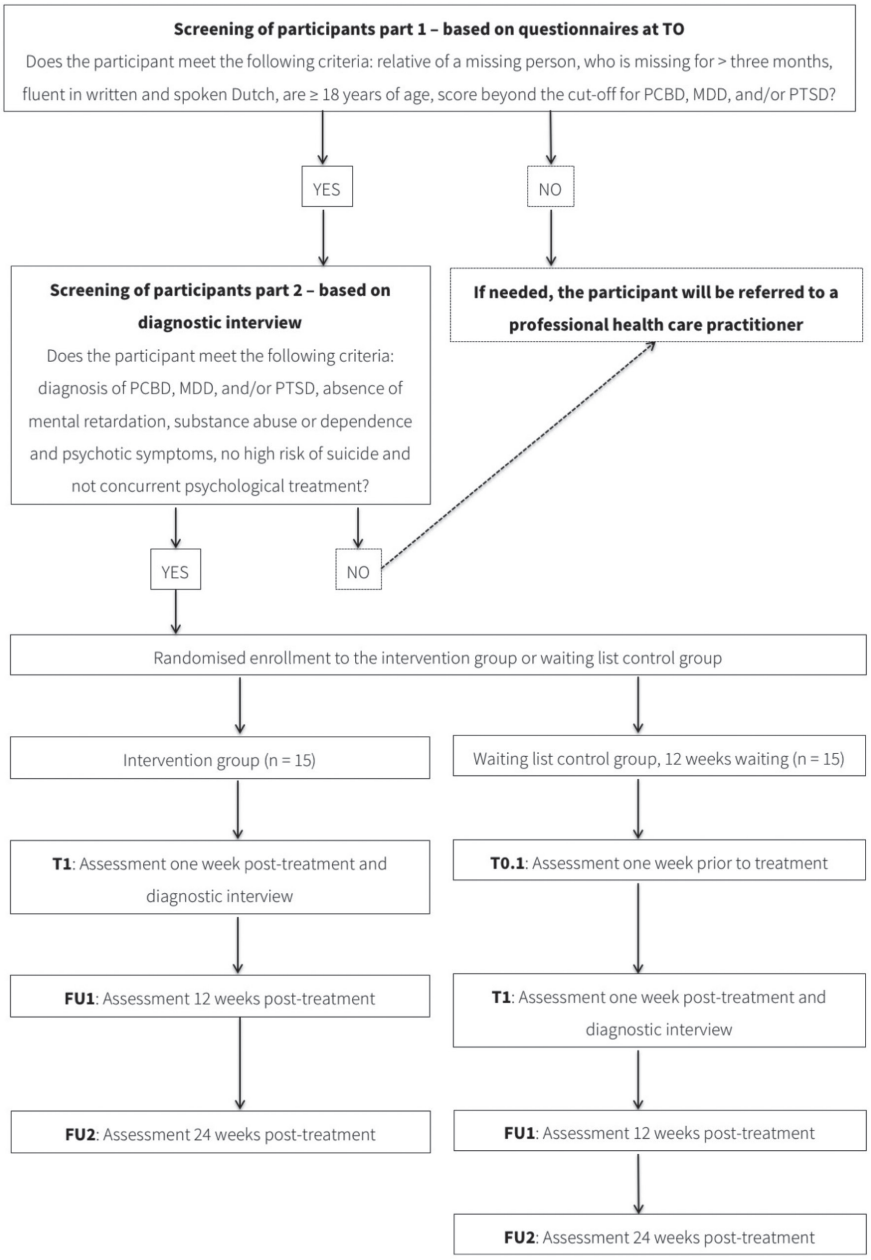
informs participants about the aims of the survey-study and the possibility to participate in a subsequent study designed to evaluate a psychological intervention specifically developed for relatives of missing persons.

Procedure and randomisation

After receiving the completed questionnaires and the signed informed consent form for participation in the survey study, the participants will be screened for in- and exclusion criteria for the intervention study. The screening procedure comprises two parts. The first part consists of screening participants for in- and exclusion criteria based on questionnaires (questionnaires and cut-off criteria are described below). Participants who score above the threshold for PCBD, MDD, and/or PTSD will be offered written information about the intervention study together with an informed consent form for participation in the intervention study. The second part of the screening procedure consists of a clinical interview composed of the M.I.N.I. Plus version 5.0.0. and the Traumatic Grief Interview. A trained independent psychologist will conduct the interview by telephone. The M.I.N.I. Plus is developed to diagnose axis I DSM-IV psychiatric disorders and has good psychometric properties (Sheehan et al., 1998; van Vliet, Leroy, & van Megen, 2000). The following modules of the M.I.N.I. Plus will be used: major depressive episode, dysthymia, suicidality, PTSD, alcoholic dependence, substance dependence, psychotic disorders. In addition, the Traumatic Grief Interview will be administered to assess symptoms of PCBD (Boelen, de la Rie, & Smid, 2000); based on this interview participants meet criteria for PCBD when they score a 2 ("sometimes") or higher on at least 1 B-cluster symptom, at least 6 C-cluster symptoms and a score of 1 ("seldom") or higher on the D-cluster symptom in accord with the proposed criteria of PCBD in the DSM-5 with exclusion of the criterion that at least 12 months must have been elapsed since the loved one has gone missing (APA, 2013).

Randomisation will take place after the participant is screened for eligibility based on the M.I.N.I. Plus and the TGI. A random number generator will be used to perform the blocking randomisation procedure. Relatives of the same missing person will be allocated to the same study arm (i.e. intervention or waiting list condition), in order to prevent transfer of information. An independent researcher will conduct the randomisation procedure. Neither the participants nor the researchers will be blinded. The M.I.N.I. Plus and the Traumatic Grief Interview will be conducted again after the treatment, to examine whether numbers of cases of PCBD, MDD, and PTSD have decreased. The researchers will reimburse costs related to the treatment that are not covered by the respondents' health insurance. Travel expenses that are related to the visits to the therapist will also be reimbursed. See Figure 1 for a flowchart of the procedures.

Figure 1. Schematic display of the study procedures



Note. T0 = baseline measure; T1 = post-treatment assessment; T0.1 = post-waiting period measure; FU1 = follow-up measure 1; FU2 = follow-up measure 2.

Sample size

To find a within-subjects difference in PCBD symptom across 4 time points (baseline and one week, 12 weeks, and 24 weeks post-treatment) of medium effect size (Currier et al., 2010) with a power of 80%, an α of 0.05, assuming the correlation between the measures to be .50, a sample size of in total 24 participants is sufficient. By taking into account a dropout rate of 19%, based on a review that reported a mean attrition rate of 19% among studies that evaluated the effectiveness of CBT for bereavement-related psychopathology (Currier et al., 2010), a total sample size of 29 is required.

Measures

Primary outcome measure

Inventory of Complicated Grief. Self-rated symptoms of complicated grief as underlying concept of PCBD will be assessed with the 19-item Inventory of Complicated Grief (Boelen, van den Bout, de Keijser, & Hoijtink, 2003; Prigerson & Jacobs, 2001). ”Respondents are asked to rate how frequently they experienced 19 grief reactions during the last month, on a 5-point scale ranging from 0 (“never”) to 4 (“always”). The Inventory of Complicated Grief is together with the PG-13 (Prigerson, Vanderwerker, & Maciejewski, 2008) a frequently used instrument to administer grief reactions. Opposed to the former instrument a validated Dutch translation of the PG-13 is not available. The Dutch translation of the Inventory of Complicated Grief, that demonstrated adequate psychometric properties (Boelen et al., 2003), is therefore chosen as primary outcome measure. We adapted the items of the Inventory of Complicated Grief by referring to the disappearance instead of death (e.g., *“Ever since he/she has been missing it is hard for me to trust people”*). The Inventory of Complicated Grief is also used as screening instrument in the first part of the screening procedure (see Figure 1) to assess whether the participant is eligible for the treatment. Participants meet the criteria for PCBD when they score above the cut-off of > 25 (Prigerson & Jacobs, 2001).

Secondary outcome measures

PTSD Checklist for DSM-5. The severity of PTSD complaints will be assessed with the 20-item PTSD Checklist for DSM-5 (Boeschoten, Bakker, Jongedijk, & Olff, 2014; Weathers et al., 2013). This measure is adapted from the PTSD Checklist for DSM-IV and is in accord with the criteria of PTSD of the DSM-5. Respondents are asked to rate to what extent they experienced PTSD symptoms during the last month on a 5-point scale ranging from 0 (“not at all”) to 4 (“extremely”). A total score (range 0-80) will be obtained by summing item-scores. The initial psychometric properties of the PTSD Checklist for DSM-5 are good (Blevins, Weathers, Davis, Witte, & Domino, 2015). We

adapted the wording 'the stressful experience' in the instruction and the items to 'the events that are associated with the disappearance' (e.g., *"In the past month, how much were you bothered by repeated, disturbing, and unwanted memories of the events that are associated with the disappearance?"*). The PTSD Checklist for DSM-5 will also be used to screen for eligibility in the first part of the screening procedure (see Figure 1). The provisional cut-off score of > 38 or the diagnostic rule of scoring at least a 2 ("moderately") on at least 1 cluster B item, 1 cluster C item, 2 cluster D items, and 2 cluster E items will be used as inclusion criteria (Weathers et al., 2013).

Inventory of Depressive Symptomatology – Self-report. The severity of depressive symptoms will be assessed with the 30-item Inventory of Depressive Symptomatology – Self-report (Rush et al., 1986; Rush, Gullion, Basco, Jarrett, & Trivedi, 1996). Descriptions of depressive symptoms are provided (e.g., *"Feeling sad"*) and respondents are asked to choose an answer that best describes how they felt during the last week (e.g., *"I feel sad nearly all of the time"*). The items are presented as multiple-choice items, with four options. Total score ranges from 0-84 and will be obtained by summing up 28 of the 30 items. This widely used measure has good psychometric properties (Rush et al., 1996). The Inventory of Depressive Symptomatology – Self-report will also be used to screen for eligibility in the first part of the screening procedure (see Figure 1), whereby a score of > 13 is defined as an indication of mild depression (Rush et al., 2003; Trivedi et al., 2004).

Southampton Mindfulness Questionnaire. Extent of mindfulness will be assessed with the 16-item Southampton Mindfulness Questionnaire (Chadwick et al., 2008; van der Valk, van de Waerdt, Meijer, van den Hout, & de Haan, 2013). This instrument is specifically developed to assess changes in the ability to respond mindfully to distressing thoughts and images (Chadwick et al., 2008), which are key elements of the treatment (e.g., *"Usually when I experience distressing thoughts or images I am able just to notice them without reacting"*). Respondents are asked to rate their agreement with each item on a 7-point scale ranging from 0 ("totally agree") to 6 ("totally disagree"). After reverse coding of some items, a total score (range 0-96) will be obtained by summing the scores for each item. The instrument showed adequate psychometric properties (Chadwick et al., 2008).

Potential Mediators

Self-Compassion Scale. The extent of self-compassion will be assessed with the Self-Compassion Scale (Neff, 2003b). The Dutch version of the Self-Compassion Scale consists of 24 items (Neff & Vonk, 2009) instead of the 26 items in the original version. Respondents are asked to rate how often they behave in the stated manner on a 7-point scale (instead of a 5-point scale in the English version) with anchors "almost never" and "almost always" (e.g., *"When I'm feeling down I tend to obsess and fixate on everything that's wrong"*). After reverse coding of some items, the total

score (range 24-168) will be obtained by summing all items. The instrument showed adequate psychometric qualities (Neff, 2003b).

Trauma Memory Questionnaire. The Trauma Memory Questionnaire consists of 13 items divided over two subscales, namely intrusion and disorganization (Boelen, 2012; Halligan, Michael, Clark, & Ehlers, 2003). Only the 8-item intrusion subscale will be administered in this study. We adapted the words that refer to "death" to "disappearance" (e.g., *"My memories of the disappearance consist of vivid images"*). The items represent different characteristics of intrusive memories associated with the disappearance. Respondents are asked to rate their agreement with each item on a scale ranging from 0 ("not at all") to 4 ("very strongly"). The original and translated version showed both adequate psychometric properties (Boelen, 2012; Halligan et al., 2003).

Grief Cognition Questionnaire. Negative cognitions associated with the disappearance will be assessed with four subscales of the Grief Cognition Questionnaire (Boelen & Lensvelt-Mulders, 2005). We adapted the wording of the items by referring to the disappearance instead of death. The subscales represent negative beliefs about the self (6 items, e.g., *"Since he/she has been missing, I feel less worthy"*), life (4 items, *"My life is meaningless since he/she has been missing"*), the future (5 items, *"I don't have confidence in the future"*) and one's own grief-reactions (4 items, *"Once I would start crying, I would lose control"*). Respondents rate their agreement with each item on 6-point scales with anchors "disagree strongly" and "agree strongly". Psychometric properties of this measure are adequate (Boelen & Lensvelt-Mulders, 2005).

Depressive and Anxious Avoidance in Prolonged Grief Questionnaire. The extent of avoidance behaviour will be assessed with the 9-item Depressive and Anxious Avoidance in Prolonged Grief Questionnaire (Boelen & van den Bout, 2010). As with the other measures, we adapted the words that refer to "death" to "disappearance". Five items represent depressive avoidance (*"I avoid doing activities that used to bring me pleasure, because I feel unable to carry out these activities"*) and four items represent anxious avoidance (*"I avoid situations and places that confront me with the fact that he/she has been missing and possibly may never return"*). Participants rate their agreement with each item on 6-point scale with anchors "not at all true for me" to "completely true for me". Psychometric properties of the subscales are adequate (Boelen & van den Bout, 2010).

Perseverative Thinking Questionnaire. The Perseverative Thinking Questionnaire is a 15-item measure to assess the severity of content-independent repetitive negative thinking (Ehring et al., 2011; Ehring, Weidacker, Emmelkamp, & Raes, 2012). This measure represents the key features of repetitive negative thinking, the perceived unproductiveness of repetitive negative thinking and the mental capacity that is captured by repetitive negative thinking. Respondents will be asked to rate each item on a 5-point scale ranging from 0 ("never") to 4 ("almost always") (e.g., *"My*

thoughts repeat themselves"). The Dutch translation yielded adequate psychometric properties similar to those of the original German and English versions (Ehring et al., 2011, 2012).

Ruminative Response Scale. The brooding subscale of the Ruminative Response Scale will be administered to measure the tendency to ruminate (Raes & Hermans, 2007; Treynor, Gonzalez, & Nolen-Hoeksema, 2003). Brooding represents a dysfunctional style of depressive rumination (Treynor et al., 2003). The respondents are instructed to rate what they generally do or think when they feel sad. Respondents will be asked to rate each of the 5 items on a 4 point-scale ranging from 1 ("almost never") to 4 ("almost always") (e.g., *I think "Why do I always react this way?"*). Psychometric properties of this measure are adequate (Schoofs, Hermans, & Raes, 2010; Treynor et al., 2003).

Measures used for monitoring treatment progress per session

Self-Compassion Scale - Short Form. The 12-item Self-Compassion Scale - Short Form (Raes, Pommier, Neff, & van Gucht, 2011) is a shortened version of the Self-Compassion Scale (Neff, 2003b). The Self-Compassion Scale - Short Form consists of two items of each of the six subscales of the extended version. A specific time frame ("the past 7 days") was added to the instructions in order to monitor the changes in current self-compassion session by session. Psychometric properties of this measure are good (Raes et al., 2011).

Repetitive negative thinking visual analogue scale. We developed the following item to assess the frequency of repetitive negative thinking: *"In the past 7 days, to what extent have you been bothered by repeated negative thoughts related to the disappearance of your loved one?"*. The item is rated on a visual analogue scale.

Characteristics of intrusive memories and images. Six items were developed to assess characteristics of intrusive memories or images during the past 7 days, based on the Intrusive Memory Interview (Hackmann, Ehlers, Speckens, & Clark, 2004). The first two items assess the content of a recurrent and unwanted memory or image related to the disappearance that was most bothering during the preceding seven days. First, the participant is asked to write down the content of this memory or image in five key words. Second, the participant is asked to indicate whether the memory or image represents something that "actually", "possibly" or "did not happen". The other four items consist of visual analogue scales that assess the following characteristics of the intrusive memories or images: the extent of distress associated with it, the degree to which it appear to happen in the present, frequency, and vividness (e.g., *"How vivid were the intrusions?"*).

Other measures

Sociodemographic variables (e.g., gender, date of birth, and educational level) of the participants and missing persons will be registered at the first measurement occasion. The number of sessions

the participants attended will be registered at the one week post-treatment assessment. In case the participant attended less than 8 sessions the reason(s) for drop-out will be registered. The following two open-ended questions regarding the evaluation of the treatment will also be administered at the one week post-treatment assessment: "About which aspects of the treatment are you satisfied?" and "About which aspects of the treatment are you less satisfied?". The questionnaires and time points are summarised in Table 1.

Table 1. Overview of variables, concepts, measures, and time points.

Variable	Concept	Measure	Time points
Primary outcome measures	Severity of PCBD symptoms	ICG	T0, T0.1, T1, FU1, FU2
Secondary outcome measures	Severity of PTSD symptoms	PCL-5	T0, T0.1, T1, FU1, FU2
	Severity of depressive symptoms	IDS-SR	T0, T0.1, T1, FU1, FU2
	Extent of mindfulness	SMQ	T0, T0.1, T1, FU1, FU2
Mediator	Severity of grief cognitions	GCQ	T0, T0.1, T1, FU1, FU2
	Severity of rumination	RRS	T0, T0.1, T1, FU1, FU2
	Severity of repetitive negative thinking	PTQ	T0, T0.1, T1, FU1, FU2
	Ability to be self-compassionate	SCS	T0, T0.1, T1, FU1, FU2
	Severity of intrusive memories	TMQ	T0, T0.1, T1, FU1, FU2
	Severity of avoidance behaviour	DAAPGQ	T0, T0.1, T1, FU1, FU2
Process of change session by session	Ability to be self-compassionate	SCS-SF	Start of each treatment session
	Severity of intrusive memories and images	Intrusive memories and images vas	Start of each treatment session
	Frequency of repetitive negative thinking	RNT vas	Start of each treatment session
Other variables	Sociodemographic information		T0
	Evaluation of the treatment and reason(s) for drop-out		T1

Note. ICG = Inventory of Complicated Grief; PCL-5 = Posttraumatic Stress Disorder Checklist for DSM-5; IDS-SR = Inventory of Depressive Symptomatology – Self Report; SMQ = Southampton Mindfulness Questionnaire; GCQ = Grief Cognition Questionnaire; RRS = Ruminative Response Scale; PTQ = Perseverative Thinking Questionnaire; SCS = Self-Compassion Scale; TMQ = Trauma Memory Questionnaire; DAAPGQ = Depressive and Anxious Avoidance in Prolonged Grief Questionnaire; SCS-SF = Self-Compassion Scale – Short Form; RNT vas = Repetitive Negative Thinking visual analogue scale; T0 = baseline measure; T1 = post-treatment assessment; T0.1 = post-waiting period measure; FU1 = follow-up measure 1; FU2 = follow-up measure 2.

Treatment

The treatment is manualized and consists of eight 45-minutes individual face-to-face therapy sessions offered in a period of maximally 12 weeks. The treatment protocol draws from CBT for treatment of PCBD (Boelen, 2006; Boelen et al., 2007). Participants learn cognitive-behavioural skills and mindfulness practices that enable them to disengage from dysfunctional cognitive patterns related to their missing loved one, both during the sessions and through homework assignments.

In the first session therapist and client introduce themselves, exchange expectations regarding the treatment and the participant is invited to share his/her story about the disappearance of the loved one. The participant receives a manual (containing psycho-education, writing assignments and exercises for how to handle maladaptive thoughts). Social support is the theme of the second session. The client is asked to invite a relative to join the client in the second session. Mindfulness exercises are introduced in session 3. The therapist explains that mindfulness training consists of sustained attention focused on the body and breath, which integrates a decentred view of thoughts as passing mental events (i.e. "rather than simply being their emotions, or identifying personally with negative thoughts and feelings, patients relate to negative experiences as mental events in a wider context" (Teasdale et al., 2002, p. 276). A mindfulness exercise is practiced and the participant will be encouraged to perform mindfulness-exercises at home at least five times a week from therapy session 3 to 8. The mindfulness exercises (Schurink, 2009) that stem from mindfulness based cognitive therapy (Segal et al., 2013) are offered on the website of our research program (www.levenmetvermissing.nl) and via a CD-ROM. The participant is asked to report aspects of their mindfulness-experiences in a mindfulness-diary (e.g., which exercise, duration of the exercise, experiences with the exercise). During session 4 to 8, CBT will be applied that consists of cognitive restructuring and exposure interventions. Cognitive restructuring aims to change dysfunctional thought patterns and exposure techniques aim to reduce anxiety and avoidance behaviour. Four structured writing assignments are added in a fixed order to the protocol for three reasons. Firstly, the assignments can serve as exposure assignment (e.g., in one of the writing assignments participants are instructed to write about their deepest thoughts and feelings associated with the most stressful memory or image related to the disappearance that keeps popping into their mind; Pennebaker & Beall, 1986). Secondly, the assignments can increase awareness of inner thoughts and feelings (Pennebaker, 1993; Pennebaker, Colder, & Sharp, 1990). Lastly, writing about traumatic experiences helps to keep the therapeutic process flowing between the sessions (Cummings, Saint, Hayes, & Park, 2014). Two of the writing assignments are imaginary exposure-assignments encouraging participants to tolerate thoughts and feelings related to the disappearance. The first assignment (between session 1 and 2) addresses the impact of the disappearance. The participant is instructed to write about the most intense thoughts

and feelings during the first days of the disappearance, followed by a description of how his/her life was before the disappearance and the most painful consequences of the disappearance on his/her life. The second assignment (between session 2 and 3) addresses the description of an image or memory related to the disappearance. The participant is invited to write about the most painful image or memory related to the disappearance and to describe all experiences (thoughts, feelings, and sensations) that come to his/her mind while thinking about that image or memory in as much detail as possible. The third writing assignment (between session 4 and 5) consists of writing a supportive letter to a hypothetical friend that is facing the same situation. This assignment aims to promote the development of new perspectives on the disappearance and its circumstances and to challenge dysfunctional thoughts and behaviour patterns. In the fourth writing assignment (between session 7 and 8) the participant is instructed to write about what he/she has learned during the treatment and how he/she dealt with the disappearance prior to treatment, how he/she deals with it at the moment and how he/she wishes to deal with in in the future. This final assignment aims to foster empowerment of the participant, in order to deal with possible difficulties in the future. The same sort of writing assignments has been successfully used in treatments of PTSD and PCBD (Lange et al., 2003; Wagner & Maercker, 2007).

A group of qualified therapists has received an 8-hour training to apply the treatment protocol. The first, third, and last author offered the training. The therapists are all governmentally licensed as psychotherapists or mental health care psychologists. They are all part of a network of trained therapists divided over the Netherlands and are experienced in the treatment of PCBD, MDD, and PTSD as a result of extraordinary, highly impact loss-related situations.

Treatment fidelity

Treatment fidelity will be monitored by asking the therapists to report all compliances and deviations from the treatment protocol in a journal. In addition, supervision takes place every month. Participants are asked to send the writing assignments and mindfulness-diary to the first author at the end of each treatment in order to monitor the compliance.

Analyses

Feasibility analyses

Analyses regarding the feasibility of this study will primarily be descriptive in terms of attrition rate, reasons for dropout, compliance with and deviations from the treatment protocol as well as a summary of the data regarding the strengths and suggestions for improvements of the treatment from the perspective of the participant.

Potential participation bias will be explored by comparing relatives who volunteered to participate in the study to those who refused to be in the study with respect to sociodemographic variables, but also by comparing the mean scores of PCBD, MDD, and PTSD.

Statistical analyses

Within-subjects repeated measures ANOVA with time (baseline, one week, 12 weeks, and 24 weeks post-treatment) as the repeated measure will be performed to evaluate the effects of the treatment in terms of changes in mean scores of PCBD.

Between-subjects multivariate analysis of covariance (MANCOVA) will be performed to compare changes in PCBD, MDD, PTSDD, and mindfulness scores between the treatment and waiting list condition. Specifically, scores at post-treatment (in the treatment condition) and post-waiting period (in the waiting list condition) on measures tapping these phenomena will be compared, including baseline scores as covariates.

Within-subjects MANOVA with time (baseline, one week, 12 weeks, and 24 weeks post-treatment) as the repeated measure will be performed to evaluate the short- and long-term effects of the treatment in terms of change in mean scores of PCBD, MDD, PTSD, and mindfulness. To test whether the treatment effect is mediated by changes in cognitive variables (i.e. grief cognitions, intrusive memories, rumination, and repetitive negative thinking), behavioural variables (i.e. avoidance behaviour), and self-compassion, only the scores of measures tapping these constructs that show the strongest association (based on univariate analyses) will be added to a multiple regression model. Finally, to assess the potential change at micro-level in repetitive negative thinking, intrusive memories and self-compassion throughout the course of the treatment a within-subjects repeated measures ANOVA with time (session 1 to session 8) as the repeated measure will be performed with mean scores of measures tapping repetitive negative thinking, intrusive memories and self-compassion as outcome measures.

Data will be analysed according to the intention-to-treat principle. Missing data will be handled using multiple imputations. The Statistical Package for the Social Sciences will be used for the analysis. The small sample size of the pilot RCT increases the risk of type II error; the preliminary results regarding the potential effectiveness will therefore be reported with caution following the recommendations for conducting a pilot study (Arain, Campbell, Cooper, & Lancaster, 2010).

DISCUSSION

Worldwide a significant number of people experience the disappearance of a loved one. Surprisingly, no RCT has ever been conducted to systematically evaluate the effectiveness of a psychological treatment for persistent psychopathology among relatives of missing persons. Knowledge about

treatment effects is needed to improve treatment options for those in need of help. This article describes the rationale and methods of a multi-centre pilot RCT focused on the evaluation of the feasibility and preliminary effectiveness of CBT with elements of mindfulness for relatives of missing persons. The treatment is based on CBT for bereavement-related psychopathology and aims to reduce dysfunctional cognitions and avoidance behaviour. Elements of mindfulness are added to the therapy to enhance skills to tolerate the ambiguity surrounding the disappearance and to prevent participants becoming entangled in negative thoughts.

Strengths of this study are a) the development of a treatment that is targeted to a specific population, b) the evaluation of the possible mechanisms of change of the treatment, c) the inclusion of both an intervention and waiting list control group, d) the use of a pilot design before exposing a large sample to a treatment that has yet to be evaluated, e) the assessment of eligibility not only by self-report questionnaires but also with a clinical interview, and f) the monitoring at micro level of potential change mechanisms of the treatment. One of the limitations of this study is the lack of power to conduct multilevel analysis to take into account the nested structure of the data (i.e. repeated measures, nested in individuals, who are, in turn, nested in families or other social systems associated with the same missing person). In addition, the recruitment procedures are focused on a non-treatment seeking population. The percentages of individual who are willing to participate might therefore be relatively low compared to other studies and the recruitment of sufficient numbers of participants might therefore be a challenge. Moreover, treatment fidelity will be monitored by methods that rely on self-report which might be prone to bias. Finally, participants whose loved one disappeared less than 12 months earlier can formally not fulfil the criteria of PCBD, because one of the proposed criteria of PCBD is that at least 12 months has elapsed since the loss. The reason for this time frame is to distinguish normal from pathological grief (APA, 2013). Because little is known about the grief reactions following the disappearance of a loved one, a shorter time frame is chosen to offer support for those in need of help.

As a result of this pilot study we may have some evidence whether CBT is effective for relatives of missing persons. Since this is a pilot study the future results of this study need to be interpreted with caution. The recommendations for the execution of a large-scale RCT based on the results of this pilot study are expected to be relevant for future research. Future intervention studies should verify the results of this study and other possibly effective treatments in order to offer an evidence-based intervention for this population. Taken together, this study will provide the first results of a pilot RCT that evaluates the potential effectiveness of CBT for relatives of missing persons suffering from psychopathology.

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9

Feasibility and potential effectiveness of cognitive behavioural therapy and mindfulness for relatives of missing persons: A pilot study

Lenferink, L.I.M., de Keijser, J., Wessel, I., & Boelen, P.A. (Submitted). Feasibility and potential effectiveness of cognitive behavioural therapy and mindfulness for relatives of missing persons: A pilot study

ABSTRACT

Relatives of long-term missing persons need to deal with uncertainties related to the disappearance. These uncertainties may give rise to ruminative thinking about the causes and consequences of the loss. Focusing on tolerating uncertainties in treatment of relatives of missing persons might be crucial for recovery. Adding elements of mindfulness to cognitive behavioural therapy (CBT+M) might serve this aim. The current pilot randomised controlled trial (RCT) evaluated the feasibility and potential effectiveness of CBT+M (immediate intervention versus waiting list controls) for reducing persistent complex bereavement disorder (PCBD), major depressive disorder (MDD), and posttraumatic stress disorder (PTSD), and enhancing mindfulness. Regarding the potential effectiveness of CBT+M, our primary aims were to detect within-group changes in symptom levels and mindfulness from pre-treatment to one week, 12 weeks, and 24 weeks post-treatment. Data from self-report measures (tapping psychopathology and mindfulness) as well as clinical diagnostic interviews (assessing PCBD, MDD, and PTSD) were gathered among relatives of missing persons with clinically relevant psychopathology levels. Nine of seventeen people completed the treatment. The limited response-rate (31.7%) and high dropout rate (47.1%) raises questions about the feasibility of the protocol. Participants completing the treatment were satisfied with treatment's quality and reported high treatment compliance. CBT+M coincided with moderate to large reductions in psychopathology levels (Hedges' g varied from 0.35 - 1.09) and small to moderate changes in mindfulness (Hedges' g varied from -0.10 - 0.41). CBT+M appears promising enough to warrant further examination in relatives of missing persons. Recommendations are provided to increase the feasibility of future trials.

Most people will face the death of someone significant at some point in their lives. Sadness and longing for the deceased are common grief responses. When grief reactions endure and are so intense that they cause significant impairment in daily life, a diagnosis of persistent complex bereavement disorder (PCBD) may be considered[1]. PCBD is included as condition for further study in the fifth Diagnostic and Statistical Manual of Mental Disorders (DSM-5; APA, 2013). PCBD shows similarities with, yet is distinguishable from major depressive disorder (MDD) and posttraumatic stress disorder (PTSD; Boelen, van de Schoot, van den Hout, de Keijser, & van den Bout, 2010; O'Connor, Lasgaard, Shevlin, & Guldin, 2010; Prigerson, Bierhals, Kasl, & Reynolds, 1996). About 10% of people exposed to a non-violent loss develop PCBD (Lundorff, Holmgren, Zachariae, Farver-Vestergaard, & O'Connor, 2017).

Although cognitive behavioural therapy (CBT) is the treatment of choice for loss-related psychopathology (Currier, Holland, & Neimeyer, 2010), only about half of the bereaved people show clinically relevant reductions in PCBD following CBT (Doering & Eisma, 2016). Two trials indicate that mindfulness is a useful complementary intervention for bereaved people (O'Connor, Piet, & Hougaard, 2014; Thieleman, Cacciatore, & Hill, 2014). For instance, elderly bereaved people with clinically relevant psychopathology levels receiving mindfulness-based CBT (n = 12) reported significantly larger reductions in MDD severity from pre-treatment to 5 months post-treatment compared with 18 people in a waiting list control condition (O'Connor et al., 2014). In addition, in an uncontrolled trial among a treatment-seeking bereaved sample (n = 42), mindfulness-based treatment coincided with significant declines in MDD and PTSD levels from pre- to post-treatment (Thieleman et al., 2014).

Compared with literature on emotional distress in bereaved people (for overviews see Burke & Neimeyer, 2013; Lobb et al., 2010; Wittouck, Van Autreve, De Jaegere, Portzky, & Van Heeringen, 2011), literature on distress in relatives of missing persons is limited (Heeke & Knaevelsurd, 2015; Lenferink, de Keijser, Wessel, de Vries, & Boelen, in press). The scant research in this area suggests that PCBD, MDD, and PTSD are more common following the disappearance of a loved one than after the non-violent death of a loved one (Lenferink et al., in press). The disappearance of a significant other may be more challenging than separation caused by death, due to the uncertainty about the permanence of the separation (Boss, 2007; Hollander, 2016). This uncertainty may give rise to ruminative thinking about the whereabouts of the missing person and the circumstances related to the disappearance (Campbell & Demi, 2000; Lenferink, Eisma, de Keijser, & Boelen, 2017a; Robins, 2010). At first, perseverative thinking about the disappearance may be helpful in the search of the missing person (Lenferink, de Keijser, Piersma, & Boelen, 2017b). As time goes

1. Also referred to as prolonged grief disorder in the forthcoming 11th edition of the International Classification of Diseases (ICD-11; Maercker et al., 2013). PCBD and prolonged grief disorder tap the same diagnostic entity (Maciejewski, Maercker, Boelen, & Prigerson, 2016).

by, perseverative thinking may grow into a maladaptive coping strategy leading to exhaustion, concentration, and sleep problems (Clark, Warburton, & Tilse, 2009; Robins, 2010).

Focusing on tolerating uncertainties by adding mindfulness to CBT (henceforth referred to as CBT+M) might be beneficial for relatives of long-term missing persons. According to Chadwick et al. (2008) training mindfulness skills teaches people to 1) decentre awareness (i.e., to view inner experience such as thoughts and feelings as temporary and not related to the self), 2) divert attention toward (rather than away from) negative inner experiences, 3) accept these inner experiences in a non-judgemental manner, and 4) let inner experiences pass without reacting. Several trials, in predominantly people with depressive symptoms, have shown that ruminative thinking is an important mechanism of change in mindfulness-based interventions (Gu, Strauss, Bond, & Cavanagh, 2015).

To the best of our knowledge, only one treatment study among relatives of missing persons has been conducted; this trial included women whose husbands went missing or were killed during the war in Bosnia-Herzegovina. That trial indicated that dialogical exposure group therapy (based on a CBT framework) and supportive group therapy both reduced PTSD and grief (i.e., yielding small to moderate effect sizes; Hagl, Rosner, Butollo, & Powell, 2015). Yet, the generalizability of the findings to people confronted with a disappearance not related to the war in Bosnia-Herzegovina is limited due to the unique features of this sample (e.g., severe exposure to other war-related stressors, low levels of literacy, Islamic background). More research is needed to enhance knowledge about the treatment of psychopathology in relatives of missing persons.

We aimed to evaluate the feasibility and potential effectiveness CBT+M for reducing PCBD, MDD, and PTSD symptoms, and enhancing mindfulness among relatives of missing persons with clinically significant psychopathology, using a pilot randomised controlled trial (RCT), comparing CBT+M with a waiting list control condition. A study-protocol of this study was published previously (Lenferink, Wessel, de Keijser, & Boelen, 2016). In line with that study-protocol, the feasibility of the treatment was examined by reporting (1) participation bias, (2) attrition rate, (3) treatment fidelity, and (4) participants' evaluations of the treatment. Regarding the preliminary effectiveness of CBT+M we expected within-group reductions in PCBD, MDD, and PTSD levels and an increase in state mindfulness from pre-treatment to one week, 12 weeks, and 24 weeks post-treatment.

In our study-protocol (Lenferink et al., 2016), we planned to examine three secondary objectives. However, we did not proceed with these analyses, because the final sample size of 17 randomised participants was too small. Firstly, we displayed reductions in percentages in the outcome measures for the treatment and waiting list control group, instead of testing whether changes in symptom and mindfulness levels differed between the groups. Secondly, we visually inspected the patterns of changes and calculated reliable change indices (RCI), instead of statistically testing associations between presumed mechanisms of change (including changes in

negative grief cognitions, intrusive memories, rumination, repetitive negative thinking, avoidance behaviours, and self-compassion) and the outcome measures. Thirdly, we were not able to explore session-to-session changes in repetitive negative thinking, intrusive memories, and self-compassion, because too few participants completed measures needed to do so.

METHOD

Participants and procedures

The pilot study is part of a larger Dutch project investigating the impact of the long-term disappearance of a significant other (Lenferink et al., 2017a, 2017b). Following the definition of the Association of Chief Police Officers (2010) a missing person is "Anyone whose whereabouts is unknown whatever the circumstances of disappearance. They will be considered missing until located and their well-being or otherwise established" (p. 15).

Adults who experienced the disappearance of a spouse, family member, or friend more than three months earlier were invited to take part in a survey between July 2014 and July 2016 (see Lenferink et al., 2017a, 2017c, 2018). Participants were recruited via (peer) support organizations, a Dutch television show for relatives of missing persons, a website of the research project (www.levenmetvermissing.nl), and other media-attention. Moreover, participants were asked to invite other relatives. The survey was accompanied by a letter that informed participants about a subsequent study designed to evaluate a tailored intervention for relatives of missing persons. Participants who scored above clinical thresholds for PCBD, MDD, and/or PTSD (described below) were potentially eligible for participation in the pilot RCT and received an information letter with details about the treatment and the study.

People who gave written consent for participation in the pilot RCT were interviewed by telephone using the M.I.N.I. Plus, version 5.0.0. (Sheehan et al., 1998) and the Traumatic Grief Inventory (TGI; Boelen & Smid, 2017a). A trained psychologist performed these semi-structured diagnostic interviews aimed at screening for the following inclusion criteria: 1) presence of PCBD, MDD, and/or PTSD, 2) absence of mental retardation, 3) absence of substance abuse, 4) absence of psychotic symptoms, 5) no high risk of suicide, and 6) not concurrently receiving support from a psychologist or psychiatrist. Subsequently, another researcher carried out a blocking randomisation procedure. This procedure increases the chance that each condition contains an equal number of participants (for more information see Efird, 2011). Eligible participants were randomly allocated to the immediate treatment group or waiting list control group. Participants allocated to the immediate treatment group started the treatment, whereas the participants of the waiting list control group started the treatment after 12 weeks of waiting. Inclusion in the pilot RCT was possible between January 2015 and July 2016.

Participants completed questionnaires before treatment (referred to as T0) and at three time points post-treatment i.e., after: one week (referred to as T1), 12 weeks (referred to as FU1), and 24 weeks (referred to as FU2). Participants in the waiting list control group completed an additional questionnaire in the last week of the waiting period (referred to as T0.1) in order to examine between-group effects (treatment vs. waiting). Furthermore, relevant modules of the M.I.N.I (including MDD and PTSD) and the TGI were also administrered by an independent psychologist one week post-treatment. See Figure 1 for schematic display of the design.

CBT with elements of mindfulness

The manualised treatment consisted of eight individual face-to-face sessions. Drawing from CBT for bereaved individuals (Boelen, 2006; Boelen, de Keijser, van den Hout, & van den Bout, 2007) the primary aim was to help relatives to change maladaptive cognitions and avoidance behaviours related to the disappearance in session and through homework exercises. Mindfulness and writing exercises were added to CBT as homework assignments. Mindfulness exercises were based on mindfulness-based cognitive therapy (Segal, Williams, & Teasdale, 2013) and were offered on CD-ROM and online (Schurink, 2009). Participants were instructed to practice these exercises at home at least five times a week from session 3 through 8. The aim of mindfulness was to teach participants how to tolerate ambiguity related to the disappearance. Four structured writing exercises served to encourage imaginary exposure, to alter negative cognitions and behaviours, and to empower participants. These were derived from internet-based interventions for PCBD (Wagner & Maercker, 2007). Figure 1 schematically depicts the treatment. The content of the treatment is discussed in more details in our study-protocol (Lenferink et al., 2016).

Power analysis

An a priori power analysis showed that 24 participants would be sufficient to find a within-subjects difference of a medium effect size in PCBD levels across four measurement occasions (pre-treatment measure, T1, FU1, and FU2) with 80% power and an α of .05. By taking into account a dropout rate of 19% (cf. Currier et al., 2010), we aimed to include 29 participants in total.

Measures

Primary outcome measure

The 19-item *Inventory of Complicated Grief* (ICG) assessed disturbed grief reactions (Boelen, van den Bout, de Keijser, & Hoijtink, 2003; Prigerson et al., 1995), referred to as PCBD in the current study. Participants were instructed to rate how frequently they experienced each grief reaction (e.g., “Ever since he/she has been missing it is hard for me to trust people”) during the preceding month on 5-point scales (0 = “never” to 4 = “always”). The ICG has demonstrated adequate

psychometric properties. Scores > 25 are indicative of clinically significant grief (Prigerson et al., 1995). Cronbach's alpha in the current study was .83 at T0.

Secondary outcome measures

The 20-item *PTSD Checklist for DSM-5* (PCL-5) assessed PTSD levels in accord with the DSM-5 criteria (Blevins, Weathers, Davis, Witte, & Domino, 2015; Boeschoten, Bakker, Jongedijk, & Olff, 2014). Participants rated to what extent they experienced each PTSD symptom (e.g., "In the past month, how much were you bothered by repeated, disturbing, and unwanted memories of the events that are associated with the disappearance?") during the preceding month on 5-point scales ranging from (0 = "not at all" to 4 = "extremely"). The PCL-5 showed adequate psychometric properties (Blevins et al., 2015). The provisional cut-off of >38 (Marx et al., 2014) or the diagnostic rule of scoring at least a 2 ("moderately") on at least 1 cluster B item, 1 cluster C item, 2 cluster D items, and 2 cluster E items are indicative of clinically relevant PTSD (APA, 2013). Cronbach's alpha in the current study was .86 at T0.

The 30-item *Inventory of Depressive Symptomatology – Self-Report* (IDS-SR) assessed MDD levels (Rush, Gullion, Basco, Jarrett, & Trivedi, 1996). Each item consists of a description of a depressive symptom (e.g., "Feeling sad"). Participants were instructed to choose one out of four answers (range 0 - 3) that best described how frequently they experienced the symptom during the preceding week (e.g., "I feel sad nearly all of the time"). The IDS-SR showed good psychometric properties (Rush et al., 1996). Scores >13 were indicative of mild depression (Rush et al., 2003). Cronbach's alpha in the current study was .82 at T0.

The 16-item *Southampton Mindfulness Questionnaire* (SMQ) assessed the ability to respond mindfully to distressing thoughts and images (Chadwick et al., 2008; van der Valk, van de Waerdt, Meijer, van den Hout, & de Haan, 2013). Participants were instructed to rate their agreement with each item (e.g., "Usually when I experience distressing thoughts or images I am able just to notice them without reacting") on 7-point scales (0 = "totally agree" to 6 = "totally disagree"). After reverse coding of some items, higher total scores indicated lower mindfulness in response to distressing thoughts and images related to the disappearance. The SMQ showed good psychometric properties (Chadwick et al., 2008). Cronbach's alpha in the current study was .73 at T0.

The instructions and/or items of the ICG, PCL-5, and SMQ were adapted to refer to the disappearance. Other measures were used for exploring potential mechanisms of change of treatment. Because we adapted our initial analytic-plan we removed the details and data regarding these measures to Appendix A.

Other measures

In the pre-treatment survey we asked about the presumed cause of disappearance and belief about the whereabouts of the missing loved one. The presumed cause of disappearance was categorised as: voluntary, victim of criminal act, victim of accident, and no (specific) suspicion. Belief about the whereabouts of the missing loved one was categorized as: I think (s)he is alive, I doubt whether (s)he is alive, and I think (s)he is not alive. In addition, we asked whether participants had previously sought professional support for dealing with the disappearance. This variable originally consisted of 5 answer categories (1 = yes, I searched for support, but did not find it, 2 = yes, I receive support at the moment, 3 = yes, I received support and I think it was helpful, 4 = yes, I received support, but I think it was unhelpful, and 5 = no, I did not seek support). We dichotomised (i.e., 1 and 5 = no, and 2 to 4 = yes) this variable for the feasibility analyses to avoid small sample sizes in some cells. We also asked "Do you have experience with performing mindfulness-exercises?" with answer options: 1 = yes, I practice mindfulness more than once each week, 2 = yes, I practice mindfulness more than once each month, 3 = Yes, I practice mindfulness less than once each months, 4 = No, I don't practice mindfulness. In the T1 assessment participants' perspective on the quality of the treatment was assessed by the following two open-ended questions: 1) What aspects of the treatment are you satisfied with, and 2) What aspects of the treatment are you less satisfied with?

During the administration of the M.I.N.I. and TGI pre- and post-treatment we asked the participants to rate to what extent they experienced hope that their loved one was still alive on a scale from 1 ("no hope") to 10 ("a lot of hope"; cf. Heeke, Stammel, & Knaevelsrud, 2015). In addition, we asked participants during the pre-treatment interviews whether they were diagnosed by a psychologist, psychotherapist, or psychiatrist with a mental disorder prior to the disappearance of their loved one with answer options yes or no.

Participants were asked to keep a diary about their experiences with the mindfulness exercises, including questions such as which exercise they conducted at what day and time (henceforth referred to as "mindfulness diary"). The therapists were asked to write about the compliance and deviations of the protocol in a diary after each session (henceforth referred to as "therapist diary"). This therapist diary included specific items for each session. For instance, did the participant (1) invite a significant other for session two and (2) conduct the homework exercises (e.g., writing exercises)?

Analyses

Feasibility

Series of logistic regression analyses, with one predictor at a time, were performed to examine which background and sociodemographic characteristics and psychopathology levels (i.e., levels

of PCBD, MDD, and PTSD) distinguished relatives of missing persons who were willing to receive compared to those who declined professional support. Less than 5% of the data per item was missing, and missing data were therefore imputed with the mean item scores.

With respect to attrition rate, we reported the reasons why participants dropped out of the study, but we were not able to statistically test differences between dropouts (n = 8) and completers (n = 9) due to the small sample sizes. Treatment fidelity was monitored by screening the therapist diaries, mindfulness diaries, and writing assignments. In addition, during the treatment, adherence to the protocol was monitored by discussing the progress of the treatment with the therapist each month (by telephone or email).

The strengths and improvements of the treatment were described based on the participants' answers to the two open-ended questions asked in the T1 assessment. Data of the completers were analysed, using methods from grounded theory (Corbin & Strauss, 2008). Accordingly, answers were divided into meaningful units and then labelled with meaningful labels that reflected the content of these units (called subthemes). Overarching major themes across the subthemes were identified (called main themes).

In addition to our study-protocol, we added two case descriptions to our trial illustrating one successful and one less successful case of CBT+M, respectively. Both case descriptions were based on information gathered from the therapists. The participants gave written consent for gathering this information. Names and other identifying information were altered in the case descriptions to protect confidentiality.

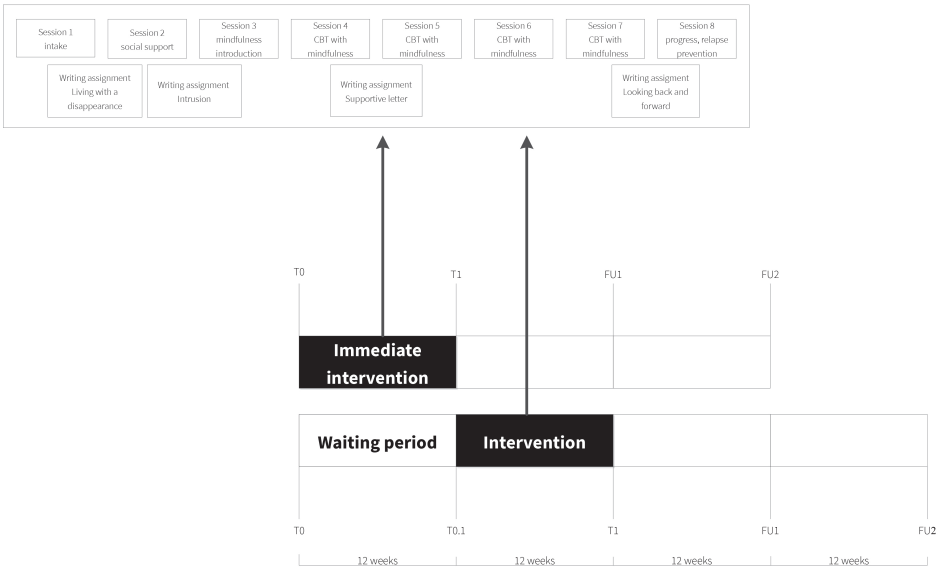
Effectiveness

Repeated measures analyses of variance (RM-ANOVAs) were used to test for significant differences over time (i.e., pre-treatment, T1, FU1, and FU2) in mean scores of PCBD, MDD, PTSD, and mindfulness, using data of all participants who completed the treatment. If the omnibus-test showed a significant main effect of time, the least significant difference (LSD) test was performed to examine differences between measurement occasions. When the assumption of sphericity was violated (i.e., $p < .05$ on the Mauchly's test) the Huynh-Feldt corrected F-value is reported. Because of the small sample size, Hedges' g effect sizes were calculated for the LSD comparisons, whereby effect sizes of 0.2 are considered small, 0.5 as moderate, and 0.8 as large (Cohen, 1988; cf. O'Connor et al., 2014). Reliable Change Indices (RCI) were calculated for each participant using the following formula from Jacobson and Truax (1991, p. 14): RC = $\frac{X2-X1}{\sqrt{2(SE)^2}}$, with X2 representing a participant's score at T1, FU1, or FU2, X1 representing scores at pre-treatment, and SE is the standard error of the pre-treatment mean scores. RCI >1.96 indicates that the change in scores is unlikely due to chance ($p < .05$). Prevalence rates of PCBD, MDD, and PTSD based on the clinical interviews (including the M.I.N.I. and TGI) prior and post-treatment were summarised. If the participant did

not meet diagnostic criteria for PCBD, MDD, and PTSD at post-treatment, this was labelled as "in full remission". Meeting diagnostic criteria for one or two disorders, but fewer disorders post-treatment compared with pre-treatment was labelled as "partly recovery". No change or increase in number of disorders was labelled as "not recovered".

Contrary to our initial analytic plan (Lenferink et al., 2016), we did not perform between-subjects statistical analyses (immediate intervention versus waiting list controls) and multiple regression analyses (to test possible mechanisms of change in the treatment), due to the small sample size of the current study. We were also not able to perform the planned analyses with the data that were to be collected each treatment session, because only one participant completed all these measures. Lastly, we did not conduct an intention-to-treat analysis for the within-group comparisons, because of all 8 participants dropping out from the study, 3 did not start the treatment and 5 received only one or two sessions. We did not include available data from these individuals in the analyses, because that was not considered to yield meaningful insights into the preliminary effectiveness of CBT+M (Gupta, 2011).

Figure 1. Design of pilot RCT



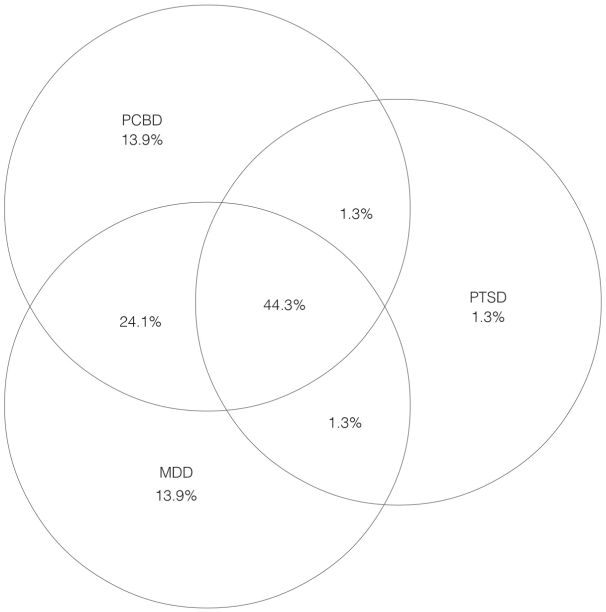
Note. T0 = pre-treatment assessment of the immediate intervention group or pre-waiting assessment of the waiting list control group; T0.1 = pre-treatment assessment of the waiting list control group; T1 = one week post-treatment assessment; FU1 = 12 weeks post-treatment assessment; FU2 = 24 weeks post-treatment assessment.

RESULTS

Participants

In total, 137 relatives of long-term missing persons participated in the survey (see Lenferink et al., 2017a, 2017c, 2018). Of them, 66 (48.2%) scored above the threshold of self-rated PCBD, 66 (48.2%) above the threshold for mild MDD, and 38 (27.7%) met provisional criteria for PTSD. In total, 79 (57.7%) passed at least one threshold. Figure 2 depicts comorbidity between clinically relevant levels of self-rated PCBD, MDD, and PTSD among these 79 participants.

Figure 2. Schematic display of comorbidity between self-rated PCBD, MDD, and PTSD (*n* = 79)

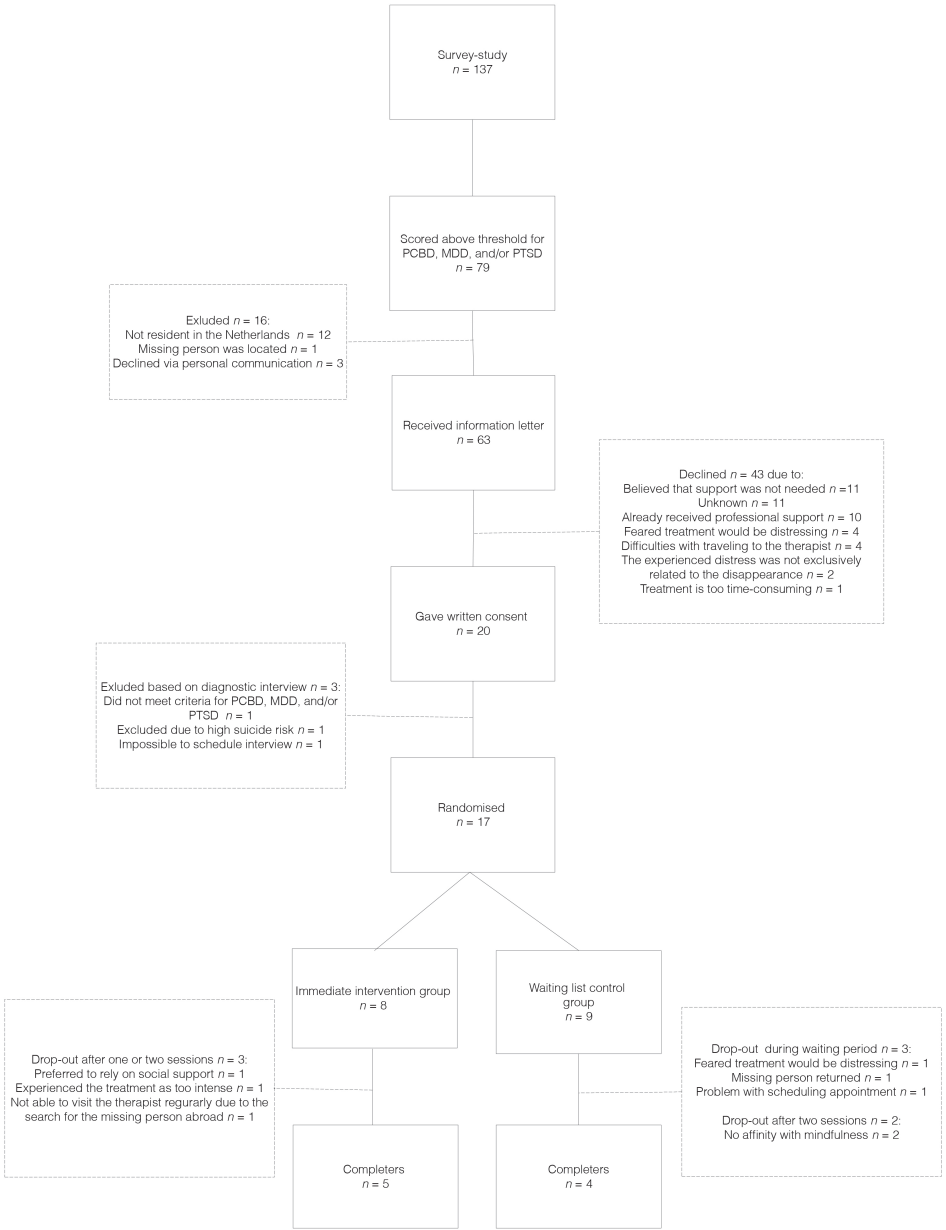


Note. Threshold for self-rated PCBD was a score of > 25, for MDD a score of > 13, and for PTSD a score of > 38.

Sixty-three of these 79 participants were send an invitation letter to participate in the current study (see Figure 3 for more details). Forty-three potential participants declined. The two primary reasons to decline participation were: 1) I believe that professional support is not needed (25.6%) and 2) I already received professional support (23.3%). Twenty participants signed up for the study, of whom 17 were eligible to participate based on results from the clinical diagnostic interviews (i.e., the M.I.N.I. and TGI; see Figure 3 for reasons for exclusion of three potential participants). Eight participants were randomly allocated to the immediate intervention group and nine to the

waiting list control condition. Five participants of the immediate intervention group and four participants of the waiting list control group completed the treatment (see Figure 3).

Figure 3. Flowchart of pilot RCT



Feasibility analyses

Participation bias

Table 1 shows the characteristics of the people who were eligible to participate in the study but declined (*n* = 43) and people who were eligible and willing to participate in the study (*n* = 20). The logistic regression analyses showed that the latter participants scored significantly higher on MDD and PTSD levels than persons who declined to participate. The two groups did not differ on the other variables.

Background characteristics of the participants

Table 2 shows the characteristics of the participants who were randomised. Of all 17 participants included in the pilot RCT, twelve participants were female (70.6%) and 8 participants (47.1%) had a high educational level. The mean age of the participants was 54.65 (SD = 12.50, range 22 to 71) years. The disappearance took place 11.71 (SD = 16.39) years earlier (range 3 months to 47 years). Four (23.5%) participants had a missing child, four (23.5%) a missing spouse, two (11.8%) a missing parent, six (35.3%) a missing sibling, and one (5.9%) a missing foster child. The presumed reason of the disappearance was in four cases (23.5%) a criminal act (e.g., presumed homicide), four cases (23.5%) a voluntarily disappearance (e.g., run away), three cases (17.6%) an accidental disappearance (skiing accident), and six persons (35.3%) had no (specific) presumption about the reasons of disappearance.

Table 1. Characteristics of people who declined and approved to participate

	People who declined to participate in the study (*n* = 43)	Participants who signed up for the study (*n* = 20)	Exp. (B) (95% CI)
Gender (0 = male), *N, %*	11 (25.6)	6 (30.0)	0.80 (0.25 - 2.60)
Age, *M (SD)*	60.62 (13.12)	54.40 (12.79)	0.97 (0.93 - 1.01)
Educational level (0 = low to moderate), *N, %*	24 (55.8)	10 (50.0)	1.26 (0.44 - 3.66)
Kinship (0 = missing person is child/spouse), *N, %*	22 (51.2)	9 (45.0)	1.28 (0.44 - 3.71)
Time since disappearance in years, *M (SD)*	12.68 (14.60)	11.35 (15.78)	0.99 (0.96 - 1.03)
Fate missing person (0 = criminal act) vs, *N, %*	13 (30.2)	4 (20.0)	
voluntarily	12 (27.9)	5 (25.0)	1.35 (0.29 - 6.26)
accident	9 (20.9)	5 (25.0)	1.81 (0.38 - 8.64)
no (specific) presumption	9 (20.9)	6 (30.0)	2.17 (0.47 - 9.95)
Believe about whereabouts (0 = he/she is dead) vs, *N, %*	26 (60.5)	9 (45.0)	
doubt whether he/she is alive	11 (25.6)	5 (25.0)	1.31 (0.36 - 4.82)
he/she is alive	6 (14.0)	6 (30.0)	2.89 (0.74 - 11.28)
Received previous professional support due to the disappearance (0 = no)	20 (46.5)	10 (50.0)	0.87 (0.30 - 2.51)
PCBD level, *M (SD)*	33.53 (11.70)	34.96 (12.01)	1.01 (0.97 - 1.06)
MDD level, *M (SD)*	21.81 (11.89)	33.05 (12.46)	1.08 (1.02 - 1.13)**
PTSD level, *M (SD)*	27.27 (15.97)	38.19 (13.62)	1.05 (1.01 - 1.09)*

Note. PCBD = persistent complex bereavement disorder; MDD = major depressive disorder; PTSD = posttraumatic stress disorder; Exp. (B) = odds ratio; 95% CI = 95% confidence interval; * $p < .05$; ** $p < .01$.

Attrition rate and reasons for dropout

In total 8 out of 17 participants dropped out (47.1%). Three participants dropped out of the immediate treatment group after receiving one or two sessions. One participant reported that he preferred to rely on social support rather than professional support as the disappearance of his spouse occurred about 5 months before treatment (ID138001; i.e., representing participant's ID number). The second participant reported that she experienced the first session of the treatment as too stressful since the disappearance of her spouse took place about 5 months earlier (ID202000). The third participant was unable to visit the therapist because she travelled regularly to search for her missing sibling who disappeared abroad less than six months earlier (ID207000).

Five participants from the waiting list condition dropped out. Three participants dropped out during the waiting period; one because the missing person was located (ID166000) and one because she worried that the therapy would be too intense (ID168000). A third participant repeatedly had difficulties with scheduling appointments with the therapist (ID112000). Consequently, she was unable to start treatment within the timeframe of the current study and was therefore considered a dropout. One couple whose child disappeared about 6 months earlier, received only two sessions (ID139000 and ID139001) once they eventually started treatment. They were reluctant to receive mindfulness and preferred to continue treatment without mindfulness, as a result they could not be included in further analyses.

Table 2 shows the characteristics of the participants who were randomised. The participants who completed the study all represented a unique missing person case. Due to the small group sizes we did not statistically test differences between dropouts (n = 8) and completers (n = 9) in terms of baseline characteristics.

Table 2. Characteristics of the participants who were randomised ($n = 17$)

Participant ID	Gender	Age in years	Times since disappearance in years	The missing person is the participant's…	Presumed reason of disappearance	Received previous professional support due to the disappearance?	Diagnosed with a mental disorder prior to the disappearance?	Previous experience with practicing mindfulness	Condition (0 = immediate intervention, 1 = waiting list)
Participants who completed the treatment ($n = 9$)									
ID127000	Female	72	32	Sibling	No specific presumption	Yes	No	No	0
ID129001	Female	60	19	Sibling	Left volutarily	Yes	No	No	1
ID133000	Female	59	1	Sibling	Accident	Yes	No	No	0
ID167002	Female	51	4	Foster child	Criminal act	No	No	No	1
ID169000	Female	64	45	Sibling	No specific presumption	Yes	No	No	0
ID205001	Male	48	47	Parent	Accident	Yes	No	Yes, >1 each week	0
ID209000	Female	69	1	Child	Left volutarily	Yes	No	No	1
ID214000	Male	58	1	Spouse	Left volutarily	Yes	No	No	0
ID230000	Female	62	1	Child	No specific presumption	Yes	No	No	1

Table 2 (continued). Characteristics of the participants who were randomised (n = 17)

Participants who dropped out of treatment (n = 8)									
ID112000	Female	51	27	Sibling	Criminal act	No	No	No	1
ID138001	Male	47	< 6 months	Spouse	No specific presumption	No	No	No	0
ID139000	Male	65	< 6 months	Child	No specific presumption	No	No	No	1
ID139001	Female	65	< 6 months	Child	Criminal act	Yes	No	No	1
ID166000	Male	22	8	Parent	No specific presumption	No	No	Missing	1
ID168000	Female	55	14	Spouse	Criminal act	No	No	Yes, <1 each month	1
ID202000	Female	52	< 6 months	Spouse	Left volutarily	No	No	Yes, <1 each month	0
ID207000	Female	33	< 6 months	Sibling	Accident	No	No	No	0

Treatment fidelity

Based on the therapist diaries, all nine participants received eight treatment sessions, except for one participant (ID230000) who received six sessions. That participant stated that despite missing her disappeared child, she no longer experienced repetitive thoughts and intrusive memories about her child, because of the practice of mindfulness. All nine participants conducted the writing assignments. The participants were asked to invite a significant other to discuss social support in session 2; only four participants did so. No other major deviations from the protocol took place, based on monthly communication (by telephone or email) with the therapists. Seven participants gave their consent to collect the mindfulness diaries. Based on these diaries, the participants performed the mindfulness exercises during an average of 25 days (range 11 to 49 days). None of the participants received additional support from a psychologist, psychotherapist, or psychiatrist after completion of the treatment (assessed at 12 weeks and 24 weeks post-treatment), except for one participant (ID167002).

Two case descriptions

Successful treatment

Eva (ID127000) was almost 40 years old when her brother was travelling around the world for over two years. One day she lost contact with her brother who was still abroad. After repeated searches, they only found his bike. Due to her brother's disappearance her family of origin was disrupted. Eva's mother was so torn apart by the disappearance that she died of a broken heart, according to Eva. Eva struggled with her emotions regarding the loss of her brother and mother and was unable to find emotional support from her family of origin, but also from her own husband and children. She became severely depressed and was institutionalised for her depression. Thirty-three years later when she signed up for the study, her brother was still missing. She experienced weekly intrusions regarding her brother's disappearance.

Based on the diagnostic interview before treatment, Eva met criteria for PCBD, MDD, and PTSD. One week post-treatment, Eva no longer met any of the diagnostic criteria. At the start of the treatment Eva felt lonely and cried when she talked about her missing brother. During her childhood, Eva's brother played an important role in her life. Their parents were traditional in terms of that Eva was expected to become a good wife and mom instead of going to school and work. Eva's brother was expected to become a priest. Eva and her brother supported each other to make their own choices in life. Eva's brother fled from the life that was planned for him by his parents by travelling the world. In treatment and by conducting the writing assignments Eva realised how important her brother was to her and how important it was for Eva to speak out freely about her thoughts and feelings. She had not only lost her brother, but also her support to stand up for herself. The therapist emphasised that it was his choice to leave, which reduced

Eva's guilt feelings. Her intrusions were replaced by positive memories regarding her brother. The mindfulness exercises were helpful to Eva; they calmed her down, helped her to confront and tolerate the sadness when thinking about her brother, and she was able to enjoy the little things in life more. At the end of treatment, she felt more capable of tolerating the sadness surrounded by the disappearance and was determined to continue to compensate the sadness by focusing more on what is important to her.

Less successful treatment

About four years earlier, Lucy (ID167002) was a single mother who took her six children on holiday to South Africa. Her oldest child Mary was 16 years old at the time, born in South Africa, but raised by her foster mother Lucy in the Netherlands. During their stay in a hotel, Mary disappeared at nighttime. After days of searching, Lucy received a phone call by Mary's biological mother who told her that she took Mary and that Lucy would never see her again.

Lucy expressed that she was hesitant to start treatment, because she stated that she could cope quite well with the disappearance. When she started to talk and write about the disappearance in treatment, strong feelings of guilt arose from thoughts as "If Mary had stayed in my room, it might would not have happened". These thoughts coincided with intrusive images about the night of Mary's disappearance. Although Mary conducted assignments on challenging her unhelpful thoughts (e.g., "I failed as a mother"), she was not convinced that this was beneficial. She argued that she was already aware of her own cognitive pitfalls. She thought that the writing and mindfulness exercises suited her better, because it helped her to get in touch with her emotions. These exercises were emotionally intense for Lucy, because she was afraid that she would loose control over her emotions. From when she was little, she taught herself to control her emotions, because she did not want to turn out like her mother. Her mother has always been emotionally unstable and was therefore not able to take care of her and her brothers. Similar to when she was younger, Lucy always felt the urge to take care of others. Mary's disappearance did not only trigger Lucy's anxiety to fail, it also fueled her strong sense of responsibility. For instance, she was worried that her other children were traumatised by the disappearance. Imaginary exposure assignments were conducted to expose Lucy to challenging emotional situations. Lucy found this helpful but also stressful. Overall, the treatment was perceived as insightful, but it "cut open previous wounds" as Lucy said, and therefore she received more therapy sessions afterwards.

Notably, after treatment, Lucy no longer met the criteria for PCBD and MDD. However, her scores on the questionnaires at T1, FU1, and FU2 indicated that her psychopathology levels of increased compared with pre-treatment[2].

Strengths and improvements of the treatment from the participants' perspective

Based on the qualitative analysis of the answers to the first open-ended question (i.e., *"What aspects of the treatment are you satisfied with?"*), all participants mentioned at least one aspect of the treatment that they appreciated. Six participants were satisfied with the client-therapist relationship (ID127000, ID129001, ID169000, ID205001, ID209000, ID230000). They reported that they felt connected with the therapist and described the therapeutic atmosphere as save and supportive (*"I felt safe and supported during the treatment. There was all the attention for the grief."* ID209000). Five participants wrote that the mindfulness-exercises were a strong element of the treatment (e.g., ID129001, ID167002, ID169000, ID205001, *"Mindfulness is a pleasant method for me to keep myself balanced. I will continue it at fixed times"* ID230000), three participants were satisfied with the writing exercises (ID127000, ID167002, ID205001), and two participants mentioned the CBT part as beneficial (ID129001, ID209000).

Based on the qualitative analysis of the answers to the second open-ended question (i.e., *"What aspects of the treatment are you less satisfied with?"*) four participants wrote that they did not have suggestions for improvement. Five participants gave the following suggestions for improvement. Four participants mentioned aspects of the content of the treatment they did not appreciate. Three participants were less satisfied with the treatment-protocol: one mentioned that she would like to attend to more than eight session (ID129001), another participant (ID205001) suggested to use fewer assessments (not clinical interview and surveys together), and one participant reported that the protocol was too strict *("It was too much according the protocol, it therefore felt impersonal."* ID133000). Two participants were not optimistic about the use of mindfulness (ID209000, ID214000) of which one mentioned that trauma-focused therapy would be more suitable (*"I wonder if the traumatic character of a disappearance is sufficiently tackled with mindfulness. I think something like "trauma-treatment" is needed."* ID214000). Another participant felt uncomfortable about the amount of homework (ID133000), and one participant

2. Lucy received additional support from another therapist between T1 and FU2, which may explain the increase in psychopathology levels post-treatment. An explanation for the deviation between survey and interview scores one week post-treatment is that Lucy realised after treatment that her complaints were more attributable to non-disappearance related issues; she therefore may reported similar PCBD levels at the pre-treatment survey and T1, but during the interview post-treatment she emphasized that her primary complaints were not grief-specific (resulting in absence of PCBD). In contrast to the MDD questionnaire, we specifically asked in the interview if MDD symptoms were attributable to the disappearance. This may explain why her MDD levels in the survey increased, but MDD related to the disappearance was absent during the interview.

(ID167002) mentioned that she would prefer to focus more on other issues, not solely related to the disappearance.

Within-subjects treatment effects

Table 3 shows the main findings of the within-subjects effects for all nine completers. All participants reported a decline in PCBD, MDD, and/or PTSD levels post-treatment except for one participant (ID167002) who reported an increase in psychopathology levels and one participant (ID209000) who reported somewhat stable psychopathology levels over time.

The overall RM-ANOVA showed a significant main effect of time over all four measurement occasions on PCBD symptoms ($F(3, 24) = 3.87$, $p = .022$). Based on the LSD comparisons, there was a significant decline in PCBD symptoms from pre-treatment to FU2 ($t(8) = 2.89$, $p = .020$, Hedges'$g = 0.57$). No other statistically significant changes in PCBD symptoms were found. Based on the individual RCI, two participants (22.2%), four participants (44.4%), and four participants (44.4%) reported clinically significant reductions in PCBD levels from pre-treatment to T1, FU1, and FU2, respectively.

Because the assumption of sphericity was violated ($\chi^2(5) = 17.59$, $p = .004$) the Huynh-Feldt corrected *F*-value was reported for changes in MDD levels. There was also a significant main effect of time on MDD symptoms ($F(1.89, 15.15) = 6.30$, $p = .011$). The MDD symptoms significantly decreased from pre-treatment to T1 ($t(8) = 3.08$, $p = .015$, Hedges'$g = 0.97$), to FU1 ($t(8) = 2.78$, $p = .024$, Hedges'$g = 1.09$), and to FU2 ($t(8) = 2.69$, $p = .028$, Hedges'$g = 1.07$). Based on the individual RCI, four participants (44.4%), five participants (55.6%), and four participants (44.4%) reported clinically significant reductions in MDD levels from pre-treatment to T1, FU1, and FU2, respectively. One participant (11.1%) reported clinically significant increase in MDD levels from pre-treatment to FU2.

Because the assumption of sphericity was violated ($\chi^2(5) = 15.04$, $p = .011$) the Huynh-Feldt corrected *F*-value was reported for changes in PTSD levels. No significant main effect of time on PTSD levels was found ($F(1.61, 12.91) = 4.04$, $p = .050$). Based on the individual RCI, two participants (22.2%), four participants (44.4%), and four participants (44.4%) reported clinically significant reductions in PTSD levels from pre-treatment to T1, FU1, and FU2, respectively. One participant (11.1%) reported clinically significant increase in PTSD levels from pre-treatment to FU2.

Compared with pre-treatment mindfulness levels, the mindfulness levels increased at T1, but decreased at FU1, and FU2 on average. No significant main effect of time on mindfulness levels was found ($F(3, 24) = 1.09$, $p = .372$). Based on the individual RCI, one participant (11.1%), one participant (11.1%), and two participants (44.4%) reported clinically significant improvements in mindfulness (indicated by lower mindfulness levels) from pre-treatment to T1, FU1, and FU2, respectively. Two participants (22.2%) reported clinically significant decrease (indication for less

mindfulness) from pre-treatment to T1, one participant (11.1%) clinically significant decrease in mindfulness levels from pre-treatment to FU1 and FU2.

Results of diagnostic interviews

Based on the diagnostic interview, all participants met criteria for PCBD and MDD before treatment. Five out of nine participants (55.6%) met criteria for PTSD before treatment. Four participants (ID127000, ID133000, ID167002, 205001) were in full remission post-treatment, three partly recovered (ID129001, ID214000, ID230000), and two did not recover (ID169000, ID209000). See Table 3 for more details.

Before and after treatment we asked the participants about their hope that the missing relative was still alive. The extent of hope seemed to remain quite stable prior and post-treatment (see the last two rows in Table 3). Those who had the least hope that their missing relative was still alive before treatment (ID127000, ID133000, ID205001) were those who were in full remission post-treatment.

Between-subjects treatment effects

The sample sizes of the two conditions (immediate intervention and waiting list control condition) were too small to statistically test between-subject treatment effects. Instead, we displayed the reductions (in percentages) in the outcome measures in Appendix B. In short, the participants in the immediate intervention group ($n = 5$) had at least twice as large reduction in PCBD, MDD, and PTSD levels on average from baseline to post-treatment compared with difference in scores from baseline to post-waiting period of the waiting list controls ($n = 4$).

Table 3. Self-report individual and mean scores PCBD, MDD, PTSD, and mindfulness before and after treatment and interview-based prevalence rates for the completers ($n = 9$)

	Measurement occasion	ID numbers										Effect size compared with pre-treatment
Self-report		127000	129001	133000	167002	169000	205001	209000	214000	230000	Mean (SD)	Hedges' *g*
PCBD scores	Pre-treatment	26	19	22	17	33	32	38	55	25	29.67 (11.70)	
	T1	13˙	21	19	21	19*	27	38	47	25	25.56 (10.62)	0.35
	FU1	6˙	26	11*	25	23*	16*	37	55	16	23.89 (14.82)	0.41
	FU2	7*	19	11*	23	19*	22	35	51	11*	22.00 (13.67)	0.57*
MDD scores	Pre-treatment	40	36	37	12	44	28	22	48	12	31.00 (13.29)	
	T1	19*	24	18*	22	25*	24	15	29*	8	20.47 (6.26)	0.97*
	FU1	5*	28	16*	17	17*	11*	26	32*	11	18.11 (8.89)	1.09*
	FU2	12*	26	13*	28˙	19*	19	18	28*	5	18.74 (7.79)	1.07*
PTSD scores	Pre-treatment	33	22	32	15	58	52	21	55	12	33.33 (17.68)	
	T1	18	21	23	14	30*	26*	17	45	9	22.56 (10.50)	0.71*
	FU1	15*	17	12*	22	17*	10*	31	46	4	19.33 (12.55)	0.87
	FU2	5*	18	11*	39˙	23*	17*	24	42	2	20.11 (13.75)	0.80
Mindfulness scores	Pre-treatment	43	59	25	23	48	63	46	53	43	44.78 (13.63)	
	T1	34	50	57˙	48˙	48	58	25*	57	38	46.11 (11.50)	-0.10
	FU1	32	61	29	37˙	48	51	51	56	26*	43.44 (12.68)	0.10
	FU2	4*	51	26	43˙	47	56	43	54	18*	38.00 (17.96)	0.41

Table 3 (continued). Self-report individual and mean scores PCBD, MDD, PTSD, and mindfulness before and after treatment and interview-based prevalence rates for the completers ($n = 9$)

Diagnostic interviews		127000	129001	133000	167002	169000	205001	209000	214000	230000	% met criteria
PCBD diagnosis	Pre-treatment	yes	yes	yes	yes	yes	yes	yes	yes	yes	100
	Post-treatment	no	no	no	no	yes	no	yes	yes	yes	44.4
MDD diagnosis	Pre-treatment	yes	yes	yes	yes	yes	yes	yes	yes	yes	100
	Post-treatment	no	yes	no	no	yes	no	yes	no	no	33.3
PTSD diagnosis	Pre-treatment	yes	no	no	no	yes	yes	no	yes	yes	55.6
	Post-treatment	no	no	no	no	yes	no	no	no	no	11.1
Extent of hope	Pre-treatment	1	8	2	8	5	1	10	10	10	
	Post-treatment	1	5	1	5	5	1	9	10	8	

Note. The one week pre-treatment assessment consists of T0 data of the immediate intervention group and T0.1 data of the waiting list control condition; T1 is 1 week post-treatment assessment; FU1 = 12 weeks post-treatment assessment; FU2 = 24 weeks post-treatment assessment; PCBD = persistent complex bereavement disorder; MDD = major depressive disorder; PTSD = posttraumatic stress disorder. Column 3 to 11 represent individual scores and in case the score at T1, FU1, and FU2 significantly ($p < .05$) reliable differed from the pre-treatment score, based on the reliable change index it was marked with *; * $p < .05$.

DISCUSSION

This study evaluated the feasibility and potential effectiveness of CBT+M in terms of reductions of PCBD, MDD, and PTSD, and enhancement of mindfulness among relatives of missing persons. Given that the current study is, to the best of our knowledge, the first trial examining the effects of a treatment solely for relatives of missing persons, we examined the feasibility and potential effectiveness of a treatment specifically tailored to this unique population. We adapted a grief-specific CBT protocol (Boelen, 2006; Boelen et al., 2007) by adding elements of mindfulness (derived from mindfulness-based cognitive therapy (Segal et al., 2013)) and writing assignments (derived from internet-based grief therapy (Wagner & Maercker, 2007)).

The relative high rates of people scoring above clinical thresholds for psychopathology found in our sample of 137 relatives of missing persons suggests that there is a need for professional support for this unique population. To illustrate this, the rates of clinically relevant self-rated levels of PCBD (48.2%), MDD (48.2%), and PTSD (27.7%) are higher in the sample of people confronted with the disappearance of a loved one, on average 15 years earlier, than rates found in people confronted with a non-violent loss in the past 6 months using comparable instruments and cut-offs (Newson, Boelen, Hek, Hofman, & Tiemeier, 2011; O'Connor, 2010). While the rates found in the current study may not be representative, because of our self-selected sample, previous studies also showed high rates of clinically relevant psychopathology levels among people confronted with the disappearance of a loved one (see for an overview Lenferink et al., in press). Remarkable is that about half of these relatives of missing persons with elevated psychopathology levels received previous professional support related to the disappearance, pointing to the need of optimizing treatment for relatives of missing persons.

Those who scored above the threshold for PCBD, MDD, and/or PTSD were invited to take part in this pilot study, but 68.3% declined. They thought it was unnecessary or reported that they already received professional support. Furthermore, those who declined reported lower MDD and PTSD levels than those who signed up for the study. These findings indicate that our inclusion criteria may have been too liberal (e.g., mild depression levels instead of severe levels). In general, it is difficult to include participants in trials examining loss-related psychopathology (considering the sample sizes of conditions in grief trials vary from 11 to 101 (see Boelen & Smid, 2017b for an overview)). Obtaining a large sample of relatives of missing persons, in a small country such as the Netherlands, in which the occurrence of a disappearance is rare (Schouten, van den Eshof, Schijf, & Schippers, 2016), would take many years. The limited response rate could also partly be explained by the use of an outreach recruitment strategy. Recruitment of hard-to-reach or rare populations, such as relatives of missing persons, is challenging and we therefore actively recruited participants who did not initially seek treatment (Shaghaghi, Bhopal, & Sheikh, 2011).

Our dropout rate from treatment of 43.8% (i.e., when not taking into account the participant whose missing loved one returned) was considerably higher than the anticipated 19% based on previous studies evaluating CBT for bereaved people (Currier et al., 2010). It should be noted that most people who discontinued treatment experienced the disappearance in the preceding year and were still actively searching for the missing person or thought that the therapy was too intense. It therefore seems recommendable to offer treatment at least one year post-disappearance, which is also in line with the time criterion for PCBD in the DSM-5 (APA, 2013) and previous trials among people confronted with a loss (e.g., Bryant et al., 2014). One couple discontinued treatment after two sessions, because they expected that mindfulness was not helpful to them. This could have been prevented by providing more detailed information about the content of the treatment before signing up for the treatment. For instance, in our information letter we did not explicitly refer to the use of mindfulness in treatment.

With regard to the feasibility of the treatment protocol, no major deviations were reported, except that not all participants were able to invite a significant other to the treatment. Only one participant reported that he/she preferred more therapy sessions, indicating that the other participants thought eight sessions were sufficient, although eight sessions is relatively few compared with other grief treatments (Boelen & Smid, 2017b). All participants conducted the writing and mindfulness exercises. Overall, participants were satisfied with the content and implementation of the treatment, but some were less satisfied with the amount of homework (including mindfulness exercises), number of assessments, and the strictness of the protocol.

Concerning the potential effectiveness of the treatment, our primary aim was to examine whether participants could benefit from the treatment. On average, the expected patterns of reductions in PCBD, MDD, and PTSD from pre-treatment to one week, 12 weeks, and 24 weeks post-treatment were observed. More specifically, for PCBD small to moderate effect sizes were found, for MDD large effect sizes, and for PTSD moderate to large effect sizes at one week, 12 weeks, and 24 weeks post-treatment compared with pre-treatment. Six out nine participants reported significant reliable reductions in PCBD, MDD, and/or PTSD levels. One participant reported increases in psychopathology from pre- to post-treatment. Because this participant is the only participant who received additional support following the treatment, it is unknown whether this increase is due to CBT+M. Changes in PCBD, MDD, and PTSD levels were summarized for the immediate intervention and waiting list control condition to give an indication of the potential effectives of CBT+M compared with natural remission. These findings suggest that the intervention contributed to alleviation of psychopathology levels.

The clinical interviews, including the M.I.N.I. and TGI, showed similar results. Overall prevalence rates of psychopathology post-treatment substantially declined compared with pre-treatment prevalence rates. We also assessed the experienced extent of hope that the missing relative was

still alive pre- and post-treatment during the interviews. Because the treatment was focused on tolerating ambiguity instead of adapting it, it is not surprising that the levels of hope seem to remain stable in treatment. Noteworthy, those who had no hope that their loved one was still alive seem to benefit most from the treatment. This finding is in line with previous research indicating that more hope among relatives of missing persons is related to elevated psychopathology levels (Heeke et al., 2015; Lenferink, de Keijser, Wessel, & Boelen, submitted).

Unexpectedly, on average the mindfulness levels seem to increase (representing less mindfulness) from pre-treatment to one week post-treatment. This increase on average is due to two people (ID133000 and ID167002) reporting reliable increase in mindfulness levels post-treatment, whereas for the other people mindfulness remained stable or decreased (representing improvement in mindfulness). Participant ID133000 was also the one who stated that she found the protocol too strict and she was not satisfied with the amount of homework. This dissatisfaction may have lead to less practice of mindfulness, which has been related to lower mindfulness levels in previous research (Carmody & Baer, 2008). Participant ID167002 reported that CBT+M cut open old wounds and she continued treatment after eight sessions. This could be interpreted as if the treatment gave rise to negative thoughts, which she found difficult to disengage from, which may explain the increase in mindfulness levels. Previous research has found that mindfulness is one of the most important mechanisms of change in mindfulness-based interventions (Gu et al., 2015). To enhance our understanding of how mindfulness-based grief treatments work, it would be worthwhile for future research to examine to what extent mindfulness, but also other potential mediators, such as ruminative thinking and self-compassion (Gu et al., 2015), mediate the therapeutic effects.

Limitations and recommendations

Several limitations should be taken into account. First and foremost, the sample size was too small to draw any firm conclusions about the effectiveness of CBT+M. One way of overcoming recruitment difficulties is to collaborate internationally and/or extend the duration of the recruitment phase. The small sample size necessitated us to remove our secondary objectives from our initial analytic-plan (Lenferink et al., 2016). For instance, we were unable to test statistical differences between post-treatment/post-waiting psychopathology levels of the immediate intervention group and waiting list control condition. The two previous trials that examined the effects of mindfulness-based treatment for people confronted with a loss did not include a control group (Thieleman et al., 2014) or used a waiting list control group (O'Connor et al., 2014). Consequently, the additional effect of integrating mindfulness in treatment of loss-related distress remains to be studied. Studies comparing the effects of CBT with CBT+M might enhance our knowledge about the efficacy of mindfulness for treatment of people confronted with a loss.

We were also not able to collect sufficient data at each treatment session for examining potential mechanisms of change, because if collected it contained too much missing data or these data were not collected because it was too time-consuming according to the therapists. Instead of collecting these data at the start of each treatment session, using the therapist as test instructor, it might be more successful to collect these data before the start of the treatment session, preferably by the researcher.

Due to the small sample size and high dropout rate we only reported the scores of the completers, which may overestimate the preliminary effectiveness (Gupta, 2011). Future studies with sufficient sample sizes should include participants in the analyses who dropped out of the treatment. This might yield a more accurate estimate of the efficacy of a treatment in clinical practice, because discontinuing treatment is also likely to occur in daily practice (Gupta, 2011).

Because we focused on relatives of missing persons in the Netherlands and consequently did not include relatives of people who went missing in war or due to political repression abroad, it is unknown to what extent these recommendations apply to people exposed to the disappearance of a significant other in armed conflict. Given the growing number of refugees and people living in conflict areas who are confronted with the disappearance of a significant other (Chen et al., 2017; UNHCR, 2017), it might be fruitful to explore to what extent (parts of) our protocol could be effectively implemented in this much larger group of relatives of missing persons. People exposed to the disappearance of a significant other in armed conflict are likely also exposed to trauma and (multiple) loss (Chen et al., 2017). Current treatment approaches for refugees, such as narrative exposure treatment, are predominantly focused on reducing PTSD levels (Silove, Ventevogel, & Rees, 2017), whereas it is unknown to what extent these treatments are effective for reducing PCBD levels (Gwozdziewycz & Mehl-Madrona, 2013). Adding modules to existing treatments for refugees, for instance CBT+M to target grief-related distress, might give first insights into the effectiveness of such treatments.

Notwithstanding these limitations, the results of this study are encouraging. Adding elements of mindfulness to CBT seems feasible and potentially effective in reducing psychopathology levels in relatives of missing persons. Because of the 1) limited research about effective treatments for relatives of missing persons, 2) elevated risk for psychopathology in relatives of missing persons, and 3) promising results of small and/or uncontrolled trials examining the effect of mindfulness-based treatment to target grief-related complaints, it seems valuable to continue investigating the effects of CBT+M on reducing post-loss psychopathology in future research.

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ACKNOWLEDGEMENTS

We would like to thank all participants. We would also like to thank the "Vereniging Achterblijvers na Vermissing", "AVROTROS Vermist", Victim Support the Netherlands, and Child Focus for their support with the recruitment of relatives of missing persons. In addition, we would like to thank all therapists for making this study possible.

APPENDIX A

Measures

The 24-item Dutch version of the Self-Compassion Scale (SCS) assessed levels of self-compassion (Neff & Vonk, 2009). The participants rated how often they behave in the stated manner on 7-point scales with anchors 1 = "almost never" and 7 = "almost always" (e.g., "I try to see my failings as part of the human condition"). Higher scores indicated higher levels of self-compassion. The SCS showed adequate psychometric properties (Neff, 2003). Cronbach's alpha in the current study was .89 at T0.

The 8-item subscale "Intrusion and disorganization" of the Dutch version of the Trauma Memory Questionnaire (TMQ; Boelen, 2012), assessed levels of intrusive memories related to the disappearance (e.g., "I experience strong emotions when remembering the disappearance"). The participants rated their agreement with each item on 5-point scales ranging from 1("not at all") to 5 ("very strongly"). The TMQ showed adequate psychometric properties (Boelen, 2012; Halligan Michael, Clark, & Ehlers, 2003). Cronbach's alpha in the current study was .82 at T0.

Nineteen items, representing four subscales, of the Grief Cognitions Questionnaire (GCQ; Boelen & Lensvelt-Mulders, 2005), assessed levels of negative cognitions related to the disappearance. These items included negative beliefs about the self (six items, e.g., "I see myself as a weak person since he/she disappeared"), life (four items, e.g., "My life is useless since he/she has been missing"), the future (five items, e.g., "I don't expect that I will feel better in the future"), and one's own grief reactions (four items, e.g., "If I let go of my emotions, I will go crazy"). Participants rated their agreement with each item on 6-point scales with anchors 0 = "disagree strongly" and 5 = "agree strongly". The GCQ showed adequate psychometric properties (Boelen & Lensvelt-Mulders, 2005). Cronbach's alpha in the current study was .92 at T0.

The 9-item Depressive and Anxious Avoidance in Prolonged Grief Questionnaire (DAAPGQ) assessed avoidance behaviour (Boelen & van den Bout, 2010). The DAAPGQ represents depressive avoidance (five items; e.g., "I develop very few new activities since he/she has been missing, because I'm unable to do so.") and anxious avoidance (four items; e.g., "I avoid to dwell on painful thoughts and memories connected to his/her disappearance.") Participants rated each item on 6-point scales with anchors 0 = "not at all true for me" to 5 = "completely true for me". The DAAPGQ has adequate psychometric properties (Boelen & van den Bout, 2010). Cronbach's alpha in the current study was .77 at T0.

The 15-item Perseverative Thinking Questionnaire (PTQ) assessed levels of disappearance-related repetitive negative thinking (e.g., "I keep asking myself questions without finding an answer"; Ehring et al., 2011; Ehring, Weidacker, Emmelkamp, & Raes, 2012). Participants rated each item on 5-point scales ranging from 1 = "never" to 5 = "almost always". The PTQ has adequate

psychometric properties (Ehring et al., 2011, 2012). Cronbach's alpha in the current study was .95 at T0.

The 5-item brooding subscale of the Ruminative Response Scale (RRS) assessed levels of ruminative thinking about a depressed mood (e.g., "I think "What am I doing to deserve this?"; Schoofs, Hermans, & Raes, 2010; Treynor, Gonzalez, & Nolen-Hoeksema, 2003). Participants rated each item on 4-point scales ranging from 1 = "almost never" to 4 "almost always". The RRS has adequate psychometric qualities (Schoofs et al., 2010; Treynor et al., 2003). Cronbach's alpha in the current study was .23 at T0.

We adapted the wording referring to "death" to "disappearance" in the TMQ, GCQ, and DAAPGQ.

Results

Table 1 shows the individual and mean scores on the SCS, TMQ, GCQ, DAAPGQ, PTQ, and RRS. Reliable change indices were calculated by comparing the scores at T1, FU1, or FU2 to pretreatment scores. Hedges' *g* effect sizes were calculated for the changes in mean scores from T1, FU1, or FU2 to pretreatment scores.

On average, levels of the SCS at T1, FU1, and FU2 were higher than pretreatment levels. The levels of TMQ, GCQ, DAAPGQ, PTQ, and RRS at T1, FU1, and FU2 were lower than pretreatment levels.

Table 1. Self-report individual and mean scores of potential mechanisms of change before and after treatment ($n = 9$)

	Measurement occasion	ID numbers										Effect size compared with pre-treatment
Self-report		127000	129001	133000	167002	169000	205001	209000	214000	230000	Mean (SD)	Hedges' *g*
SCS	Pre-treatment	93	87	76	111	98	90	139	111	127	103.53 (20.24)	
	T1	162 *	96	69	100	96	76	140	108	138	109.44 (31.06)	-0.22
	FU1	158 *	91	103 *	105	113	104	127	110	137	116.44 (20.70)	-0.60
	FU2	145 *	86	116 *	missing	92	96	140	115	140	166.25 (23.52)	-2.73
TMQ	Pre-treatment	22	22	24	17	24	30	32	37	21	25.44 (6.29)	
	T1	14 *	17	20	17	24	25	35	35	23	23.33 (7.53)	0.29
	FU1	18	16 *	20	17	15 *	19 *	35	37	18	21.67 (8.28)	0.49
	FU2	17	20	16 *	29*	24	27	33	35	19	24.44 (6.97)	0.14
GCQ	Pre-treatment	23	20	38	7	28	35	25	65	22	29.22 (16.12)	
	T1	0*	22	19*	11	19	42	27	56	3*	22.11 (17.91)	0.40
	FU1	0*	27	18 *	8	19	20 *	32	56	0*	20.00 (17.46)	0.52
	FU2	0*	18	14 *	25 *	4 *	31	30	61	1*	20.44 (19.31)	0.47
DAAPGQ	Pre-treatment	10	17	10	9	27	19	6	28	33	17.67 (9.75)	
	T1	0*	4 *	5	15	9 *	20	9	34	20 *	12.89 (10.54)	0.45
	FU1	0*	12	4	11	9 *	9 *	5	33	17 *	11.11 (9.58)	0.65
	FU2	0*	14	5	15	9 *	18	10	31	4 *	11.78 (9.22)	0.59

Table 1 (continued). Self-report individual and mean scores of potential mechanisms of change before and after treatment ($n = 9$)

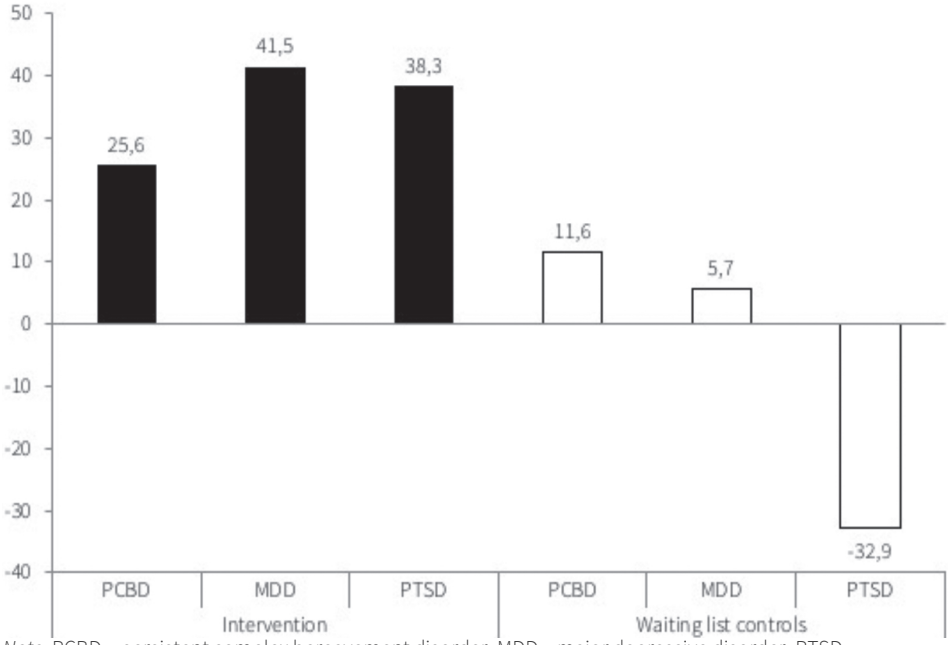
PTQ	Pre-treatment	28	54	33	29	45	51	54	64	39	44.11 (12.63)	
	T1	18	47	35	39	43	51	43	57	33	40.67 (11.36)	0.27
	FU1	19	56	21 *	37	15 *	34 *	47	55	20 *	33.78 (16.02)	0.68
	FU2	17	46	28	50 *	33 *	42	41 *	59	20 *	37.33 (13.96)	0.49
RRS	Pre-treatment	8	12	11	12	9	9	8	12	10	10.11 (1.69)	
	T1	6 *	11	11	9 *	10	9	7	11	7 *	9.00 (1.94)	0.58
	FU1	5 *	10 *	9 *	12	6 *	6 *	8	13	7 *	8.44 (2.79)	0.69
	FU2	5 *	11	14 *	12	8	6 *	8	10 *	9	9.22 (2.86)	0.36

Note. SCS = Self-Compassion Scale; TMQ = Trauma Memory Questionnaire; GCQ = Grief Cognitions Questionnaire; DAAPGQ = Depressive and Anxious Avoidance in Prolonged Grief Questionnaire; PTQ = Perseverative Thinking Questionnaire; RRS = Ruminative Response Scale; * $p < .05$.

APPENDIX B

Figure 1 displays the average reductions (in percentages) in PCBD, MDD, and PTSD for completers in both conditions. These rates were computed by 100-((Average post-treatment score/average pre-treatment score)*100). In black, the reductions from pre-treatment to post-treatment for the immediate intervention group are displayed. In white, the reductions from pre-waiting to post-waiting for the waiting list controls are displayed. On average, the participants in the immediate intervention group report 25.6% to 41.5% reduction in PCBD, MDD, and PTSD levels, whereas the waiting list controls report 5.7% to 11.6% reduction in PCBD and MDD levels, respectively, and 132.9% increase in PTSD levels.

Figure 1. Percentages of reductions in mean PCBD, MDD, and PTSD levels for the immediate intervention (*n* = 5) and waiting list control condition (*n* = 4).



Note. PCBD = persistent complex bereavement disorder; MDD = major depressive disorder; PTSD = posttraumatic stress disorder.

10

General discussion

INTRODUCTION

The overarching aim of this dissertation was to enhance knowledge about consequences of, and care after the disappearance of a significant other. In this concluding chapter we summarize and reflect on the main findings of the studies included in this dissertation. In doing so, we follow the order of the three parts of this dissertation: phenomenology of ambiguous loss (Part I), emotion regulation strategies in relatives of missing persons (Part II), and treatment of distress among relatives of missing persons (Part III). Next, we elaborate upon the limitations of this dissertation that were not sufficiently addressed in the preceding chapters. Furthermore, based on the findings presented in this dissertation, implications for research and practice are considered.

1. Part I: Phenomenology of ambiguous loss

Boss (2006) labelled the disappearance of a significant other as an ambiguous loss, because the missing person is physically absent, but psychologically present. She claimed that this type of loss is "the most stressful kind of loss due to the ambiguity" (p. 7). In Chapter 2 and Chapter 3 the empirical evidence related to this claim was synthesized, and a study was conducted to further knowledge on the mental health consequences of a disappearance vs. homicidal loss of a significant other.

1.1. Summary of findings of Chapter 2 - 3

In **Chapter 2** we provided a systematic overview of scientific literature on psychological symptoms in people confronted with the disappearance of a significant other. Findings of 11 quantitative peer-reviewed studies on the prevalence rates and correlates of psychological symptoms in relatives of missing persons were summarized. In addition, findings of studies comparing psychopathology levels between relatives of missing persons and deceased persons were described. The small number of studies and heterogeneity between the studies (e.g., methodological quality, difference in measures used, and sample composition) preclude drawing firm conclusions about prevalence rates and correlates of psychological symptoms in relatives of missing persons. Overall, findings of six comparative studies did not support the assumption that relatives of missing persons reported more severe psychological symptoms than their bereaved counterparts. It is noteworthy that all comparative studies concerned comparisons of relatives of persons who disappeared against relatives of persons who were killed in the context of armed conflict (i.e., war or state terrorism). Most of these relatives had also been exposed to other potential traumatic events potentially affecting the nature and severity of their symptoms. Consequently, in these studies it was difficult, if not impossible, to distinguish the effects of the loss from the effects of other potential traumatic stressors.

Based on these findings, we conducted a comparative study outside the context of armed conflict. In **Chapter 3**, data from two studies were used to compare prolonged grief (PG) and posttraumatic stress (PTS) symptom levels between 134 relatives of long-term missing persons and 331 homicidally bereaved people. In the sample of relatives of missing persons, 47.0% and 23.1% reported self-rated PG and PTS levels above clinically relevant thresholds, respectively (for more information on thresholds, see Brewin, Andrews, & Rose, 2000; Hoge, Riviere, Wilk, Herrell, & Weathers, 2014; Prigerson et al., 1995). In the sample of homicidally bereaved people, 83.1% and 31.4% reported clinically relevant self-rated PG and PTS levels, respectively. Contrary to previous assumptions (Boss, 2006; Heeke & Knaevelsrud, 2015) PG and PTS levels were significantly higher in homicidally bereaved individuals than in relatives of long-term missing persons (d = .86 and .28, respectively).

2. Part II: Emotion regulation strategies in relatives of missing persons

The systematic review in Chapter 2 showed that research on the psychological impact of the disappearance of a significant other is scarce, exclusively focused on disappearances in the context of armed conflict, and does not shed light on factors associated with psychopathology that are amendable to change in treatment. In Chapter 4 - 7 we aimed to fill this gap by enhancing knowledge about why some people may have less difficulty in coping with the disappearance of a significant other than others. To do so, we examined cross-sectional survey-data collected in a sample of over 130 Dutch and Belgian people confronted with the disappearance of a family member, spouse, or friend at least three months earlier (in Chapter 3 - 6). Furthermore, during the data-collection phase of the survey-study, a subsample was selected of first-degree family members or spouses of missing people who scored below clinically significant levels of PG, PTS, and depression (Chapter 7). Interview-data were collected among this subsample of 23 people.

2.1 Summary of findings of Chapter 4 -7

In line with a cognitive-behavioural model of PG (Boelen, van den Hout, & van den Bout, 2006), prior findings indicated that negative cognitions and avoidance behaviours are related to distress among bereaved individuals (Boelen, de Keijser, & Smid, 2015; Boelen & Eisma, 2015; Boelen, van Denderen, & de Keijser, 2016). We examined in **Chapter 4** to what extent these findings generalize to relatives of missing persons. In line with our hypotheses, negative cognitions and avoidance behaviours explained 40% to 60% of the variance in PG, PTS, and depression levels over and above sociodemographic variables. Thus, relatives of missing persons who held more negative cognitions and engaged in more avoidance behaviours were more likely to experience elevated psychopathology levels. Based on these findings, we concluded that it might be beneficial to address these cognitive-behavioural variables in treatment.

Previous studies indicated that experiencing/expressing positive mood fostered recovery from loss (Bonnano & Keltner, 1997; Keltner & Bonanno, 1997; Ong, Bergeman, Bisconti, & Wallace, 2006; Tweed & Tweed, 2011). However, research examining associations between individual differences in the *regulation* of positive affect and post-loss psychopathology is lacking. In **Chapter 5**, we aimed to fill this gap in knowledge by examining to what extent negative and positive affect regulation strategies are related to psychopathology following the death (Sample 1) and long-term disappearance (Sample 2) of a significant other. Negative affect regulation strategies included depressive rumination, which refers to repeatedly pondering on the nature, causes, and consequences of a sad/depressed mood. Positive affect regulation strategies included enhancing and dampening of positive affect. Enhancing of positive affect (which has also been referred to as "positive rumination") concerns "the tendency to respond to positive affective states with recurrent thoughts about positive self-qualities, positive affective experience, and one's favourable life circumstances" (Feldman Joormann, & Johnson, 2008, p. 509). Dampening of positive affect refers to "the tendency to respond to positive moods states with mental strategies to reduce the intensity and duration of the positive mood state" (Feldman et al. 2008, p. 509). Based on depression research (Raes et al., 2014; Raes, Daems, Feldman, Johnson, & Van Gucht, 2009; Raes, Smets, Nelis, & Schoofs, 2012), we hypothesized that positive affect regulation strategies would explain variance in psychopathology levels over and above negative affect regulation strategies. This hypothesis was tested in two separate samples. As expected, positive affect regulation strategies explained variance in PG, PTS, and/or depression levels over and above negative affect regulation strategies. Pending replication of our findings in longitudinal research, these findings support the usefulness of future explorations of how these two affect regulation strategies impact post-loss mental health, and how they might be effectively targeted in treatment.

The disappearance of a loved one is inherently linked to uncertainties (e.g., not knowing whether the person suffered or is alive or dead) that are uncontrollable (Boss, 2006). Disappearances may, therefore, more than natural losses (e.g., caused by illness) give rise to ruminative thinking about the causes and consequences of the loss (Boss, 2006; Heeke, Stammel, & Knaevelsrud, 2015). It has been argued that a self-compassionate attitude (i.e., recognizing and embracing one's own suffering and distress) might serve as a buffer for getting entangled in ruminative thinking, which in turn inhibits exacerbation of psychological distress (Krieger, Altenstein, Baettig, Doerig, & Holtforth, 2013; Raes, 2010; Thompson & Waltz, 2008). In **Chapter 6**, we examined whether greater self-compassion is related to lower psychopathology levels in relatives of long-term missing persons. Furthermore, we tested to what extent these associations were mediated by repeatedly thinking about the causes and consequences of the loss (i.e., grief rumination). We concluded, with caution because of our cross-sectional design, that relatives of missing persons who have

stronger tendencies to approach their emotional pain in an open and understanding way (i.e., more self-compassion), are less likely to get entangled in ruminative thinking which, in turn, attenuates psychological symptoms. Strengthening a self-compassionate attitude to counter ruminative thinking in treatment, by means of for instance mindfulness training, may therefore be useful in alleviating emotional distress following the disappearance of a loved one.

The two main aims of the interview study, presented in **Chapter 7**, were 1) to examine retrospectively patterns of functioning over time, and 2) to explore what coping strategies people with relatively low levels of PG, PTS, and depression found most helpful in dealing with the disappearance. In the first part of the interview participants were asked to draw a graph of the discourse of their functioning from one year prior to the disappearance up to the day of the interview (cf. Burr & Klein 1994). The most frequently identified pattern of functioning over time was the recovery pattern. Fifteen out of 23 people reported this pattern that is characterized by an initial decrease in functioning immediately following the disappearance followed by a significant stable increase in functioning. Seven out of 23 people reported a stable/resilient pattern that is characterized by a high level of functioning with no significant increases or decreases. The second part of the interview consisted of a card-sorting task (cf. Paap et al., 2014). We presented 15 cards that represented all 15 subscales of a measure to assess coping strategies (i.e., the COPE easy; Kleijn, Heck, & Waning, 2000). We instructed the participant to select five out of fifteen cards that, in his/her opinion, had been most helpful in dealing with the disappearance ever since it occurred. Subsequently, the participant was asked to explain why he/she considered the chosen coping strategy as helpful. Acceptance, interpreted as learning to live with not knowing (Boss, 2006), was most often chosen as helpful coping strategy. This indicated that, according to relatives with little to no symptoms looking back on responses to the disappearance, learning to tolerate uncertainty is of utmost importance for relatives of missing persons. Venting emotions with and receiving emotional support from family members and friends were also frequently chosen as helpful coping strategies. This highlights the importance of integrating the social context of a client, such as interpersonal relationships, in professional support of relatives of missing persons. Lastly, mental disengagement, described as engaging in social or occupational activities to avoid repetitive negative thinking, was also chosen as helpful coping strategy.

3. Part III: Treatment of distress among relatives of missing persons

Altogether, the findings from these three correlational studies (Chapter 4 - 6) indicated that, similar to bereaved individuals, relatives of missing persons who experience more negative cognitions, and engage in more avoidance behaviors, and ruminative thinking are more likely to experience elevated psychopathology, including PG, PTS, and depression. We expanded prior work on maladaptive strategies of coping with the loss of a significant other, by exploring the role

of adaptive coping strategies. Our findings suggest that enhancing positive affect by optimistic thoughts about oneself and inner positive emotions (i.e., enhancing of positive affect) and being more self-compassionate are potential protective factors for experiencing psychological consequences following the disappearance of someone significant (Chapter 5 – 6). These findings seem to offer tentative support for efforts to develop and evaluate the potential effectiveness of cognitive behavioural therapy with elements of mindfulness (CBT+M) for relatives of missing persons with elevated psychopathology levels.

3.1 Treatment: Summary of findings of Chapter 8 - 9

CBT aimed at confronting the reality of the loss and its' irreversibility, is the treatment of choice for bereaved people (Boelen & Smid, 2017; Currier, Holland, & Neimeyer, 2010; Doering & Eisma, 2016). Clinicians have argued that exerting pressure on relatives of missing persons to move on or achieve closure in treatment is counterproductive and may provoke resistance (Boelen & Smid, 2017; Boss, 2006; Glassock, 2006). Instead of focusing on unmanageable external factors related to the disappearance, fuelled by the uncertainty surrounded by the disappearance, learning how to manage persistent negative thoughts and feelings, for instance with mindfulness training, might be promising (Boss, 2006). In **Chapter 8**, we offered a rationale for a pilot randomized controlled trial (RCT) for evaluating the feasibility and potential effectiveness of CBT+M versus waiting list controls for reducing PG, PTS, and depression levels and enhancing mindfulness in relatives of missing persons in need of professional support. Based on the preliminary findings of the pilot RCT, described in **Chapter 9**, we concluded that, except for one out of nine participants, CBT+M coincided with reductions in psychopathology severity. This indicates that the treatment is promising enough to warrant further examination. However, in order to increase the feasibility of future trials among relatives of missing persons, we recommend collaborating internationally and/or extending duration of recruitment phase, to maximize the sample size.

4. Reflections on Part I: Phenomenology of ambiguous loss

Findings from Chapter 2 and Chapter 3 do not support claims that the disappearance of a significant other is "the" most stressful type of loss (Boss, 2006; Heeke & Knaevelsrud, 2015). It seemed important to examine this claim, because statements such as "grief is frozen, life is put on hold, and people are traumatized" (Boss, 2010, p. 137) may undermine hope in relatives of missing persons that it is possible to live a meaningful life when someone significant is still missing. Furthermore, without evidence, such claims may reinforce stigma. It has been argued and empirically supported that people exposed to a potentially traumatic loss (e.g., suicide and homicide), and people experiencing heightened loss-related distress, are at heightened risk to receive and perceive stigmatizing reactions from others (Chapple, Ziebland, & Hawton, 2015;

Eisma, 2018; Pitman, Osborn, Rantell, & King, 2016). Stigmatizing reactions may include social embarrassment, social avoidance, and stereotyping (Cvinar, 2005). Perceived stigmatization by others has also been observed in relatives of missing persons (Robins, 2010). Elevated stigmatization is related to a host of negative outcomes, including decreased help-seeking behavior, shame, guilt, and maintenance of distress (Carpiniello & Pinna, 2017; Clement et al., 2015; Pitman et al., 2016).

Chapter 3 indicates that homicidally bereaved people are at higher risk to develop elevated PG and PTS symptoms compared with relatives of missing persons, generated some confusion because it ran counter to expectations. Pending replications, we can only speculate about explanations for these differences. One explanation for the differences in psychopathology levels between the samples, might be related to third variables that were not examined in Chapter 3. For instance, homicidally bereaved people need to deal with the fact that their significant other has been killed by someone, whereas in the sample of relatives of missing persons only 33% presumed that someone else is accountable for the disappearance (i.e., the missing person was presumed to be victim of kidnapping or homicide). The idea that a third party is accountable for one of the most devastating experiences that a person could face may give rise to a host of negative cognitions, e.g., shattered worldview, thinking life is meaningless, diminished self-worth, and vengeful thoughts. These negative cognitions have been identified as mediators of elevated psychopathology levels following violent loss (Boelen et al., 2015, 2016; Mancini, Prati, & Black, 2011). Findings from Chapter 5 partially support the assumption that people who thought their missing loved one was a victim of a crime reported higher psychopathology levels than those who presumed their missing loved one went missing due to another cause (i.e., voluntarily or accidently missing or had no specific assumption about the cause). Furthermore, this hypothesis might also explain why comparative studies in the context of armed conflict (as described in Chapter 2) overall did not find differences in psychopathology levels between people with unconfirmed and confirmed loss of a significant other.

5. Reflections on Part II: Emotion regulation strategies in relatives of missing persons

In part II of this dissertation, correlates of psychopathology post-disappearance that can be targeted in treatment were examined. Because empirical knowledge about these possible correlates was absent in the literature on ambiguous loss (see Chapter 2), we drew from theoretical and empirical work from the fields of PG, PTS, and depression. For the reasons set out in Chapter 8, we looked at possible correlates of psychopathology post-disappearance from the perspective of cognitive-behavioural and mindfulness-based theorizing.

5.1 Maladaptive emotion regulation strategies

In this dissertation, we zoomed in on several intrapersonal thinking processes, because we argued in Chapter 1 that the disappearance, more than the death of a significant other gives rise to repetitive thinking. Although this hypothesis rests on an untested assumption, the findings from three correlational studies (Chapter 4-6) indicated that perseverative negative thinking processes are important related factors of PG, PTS, and depression symptom-levels in relatives of missing persons.

For instance, the findings from Chapter 5 and 6 indicate that rumination about a depressed mood (i.e., depressive rumination) and grief-specific ruminative thinking (i.e., grief rumination) are strongly related to emotional distress in relatives of missing persons. While we did not statistically test the differences in associations (e.g., with Steiger's Z tests cf. Nelis, Holmes, & Raes, 2015), the correlations of maladaptive strategies (i.e., depressive and grief rumination) with psychopathology levels appears to be higher than the associations between adaptive emotion regulation strategies (i.e., self-compassion and enhancing of positive affect) and psychopathology levels. Together with findings supporting a role for rumination in the development and/or maintenance of a variety of mental health problems (see for example overviews in grief, Eisma & Stroebe, 2017; depression, Papageorgiou & Wells, 2004; PTSD, Szabo, Warnecke, Newton, & Valentine, 2017), our finding underlines the importance of targeting this transdiagnostic phenomenon in treatment for relatives of missing persons.

5.2 Adaptive emotion regulation strategies

Prior work was extended on the role of maladaptive emotion regulation strategies in dealing with the loss of a significant other, by exploring the role of adaptive emotion regulation strategies (Chapter 5 - 7). The importance of this extension can be understood from the two continua model, stating that mental health is more than merely the absence of psychopathology (Keyes, 2005). According to this model, mental health and mental illness are related, but are also distinct dimensions; one continuum indicates the absence or presence of mental health, whereas the other represents the absence or presence of mental illness. Factor analytic studies across a variety of samples have supported that mental health and mental illness are related but distinct construct (Keyes, 2005; Keyes et al., 2008; Westerhof & Keyes, 2008). This implies that solely focusing on risk factors of psychopathology in research and clinical practice does not reflect the full picture of mental health, and research into factors promoting positive mental health is important too (Bohlmeijer, Bolier, Steeneveld, Westerhof, & Walburg, 2013).

Contrary to earlier theories of grief (Bowlby, 1980; Freud, 1957; Kübler-Ross, 1973; Worden, 1991), since the 1990s theoretical work put more emphasis on the importance of expanding the focus from reducing maladaptive emotion regulation to enhancing adaptive emotion regulation

strategies following loss (Bonnano & Kaltman, 1999; Stroebe & Schut, 1999). For instance, the dual process model of coping with bereavement states that positive affect and conducting restoration-oriented tasks (e.g., starting new relationships) are necessary for optimal adjustment to loss (Stroebe & Schut, 1999). This has been supported by the interview-study in Chapter 7, in which formation or expansion of family life, continuing occupational tasks, and receiving or offering social support, were identified as factors promoting adaptation to the long-term disappearance of a significant other.

5.2.1 Enhancing positive affect

In Chapter 5 we used the broaden-and-build theory of Fredrickson (1998, 2001) as a theoretical framework for examining positive affect regulation strategies following loss. In contrary to negative emotions, experiencing positive emotions is assumed to broaden one's attention, enabling flexible thinking and augmenting people's coping resources (Fredrickson, 2001; Fredrickson & Branigan, 2005). Previous research has shown that positive affect can be stimulated by mindfulness training (Garland, Geschwind, Peeters, & Wichers, 2015) and mediates the effect of mindfulness-based cognitive therapy on depression levels in partially remitted depressed people (Batink, Peeters, Geschwind, van Os, & Wichers, 2013). While there is an ongoing debate about how and why attention is broadened during positive affective states (see for an overview Vanlessen, De Raedt, Koster, & Pourtois, 2016), experiencing positive affect, for instance gratitude, seems to buffer for detrimental effects of exposure to potential traumatic events (van Dusen, Tiamiyu, Kashdan, & Elhai, 2015), including the death of a loved one (Ong et al., 2006; Tweed & Tweed, 2011).

Raes et al. (2012) concluded that, in the context of depression, how people respond to positive affect is at least as important as how people respond to negative affect. More specifically, in a non-clinical sample the effect of rumination on depression levels three months later disappeared when dampening of positive affect and baseline depression levels were taking into account, whereas the effect of dampening remained significant. Contrary to Raes et al.'s (2009, 2012, 2014) findings, we found in our cross-sectional study, in Chapter 5, that rather than dampening of positive affect, enhancing of positive affect was uniquely associated with psychopathology following the disappearance and death of a significant other.

These deviations between the findings from Raes et al.'s and our findings might be explained by differences in, among others, sample composition. For instance, Raes et al. (2009, 2012, 2014) used samples mostly consisting of students and/or people experiencing below threshold depression levels, while we studied people exposed to a significant loss with low to clinically relevant symptom levels. While the finding that we obtained similar patterns of results across two samples confronted with a loss supports the generalizability of the findings across people confronted with different types of losses, future research in clinical samples who have experienced other types

of losses is needed to further examine the relative importance of positive and negative affect regulation strategies and test the generalizability of these findings. It would be interesting to examine whether longitudinal studies replicate our findings in bereaved people. A longitudinal study in a disaster-bereaved sample that we are currently conducting may shed light on this issue.

5.2.2 Self-compassion

Two proposed key pathways in explaining the effectiveness of mindfulness-based interventions are enhancing self-compassion and reducing depressive ruminative thinking (Gu, Strauss, Bond, & Cavanagh, 2015; Svendsen, Kvernenes, Wiker, & Dundas, 2017; van der Velden et al., 2015). Instead of blaming and judging oneself – fuel for ruminative thinking – a more self-compassionate attitude has assumed to enable people to give less authority to self-critical thinking leaving room for the development of more helpful thinking patterns (Kuyken et al., 2010). Several treatment trials have shown that self-compassion and depressive rumination mediate the impact of mindfulness-based interventions on depression and anxiety symptoms (see for overviews Gu et al., 2015; van der Velden et al., 2015).

In theory, embracing and accepting sadness associated with the loss (i.e., self-compassion) might be viewed as a way of natural exposure to internal threats (Thompson & Waltz, 2008). Whereas ruminating about how the loss could have been prevented, has been argued to be a way of avoiding or suppressing more painful aspects of the loss, and thereby hampering the grieving process (Boelen, 2006; Eisma et al., 2013; Stroebe et al., 2007). Thus, people who naturally expose themselves to negative internal experiences may be less inclined to ruminate, which reduces symptom-levels.

The notion that ruminative thinking mediates the association between self-compassion and psychopathology has, to our knowledge, only been studied cross-sectionally in the context of depression and anxiety (Johnson & O'Brien, 2013; Krieger et al., 2013; Raes, 2010). In Chapter 6, we showed that these previous findings generalize to relatives of missing persons. Because results of cross-sectional mediation analyses can be misleading (as has been illustrated by Maxwell & Cole, 2007), longitudinal studies, preferably including at least three waves of data, are needed to further assess this potential mediation effect. Furthermore, alternative mediation models should be tested. For instance, other mediational analyses, also using cross-sectional designs, indicated that both self-compassion and rumination mediate the association between mindfulness and depression (Svendsen et al., 2017).

6. Reflections on Part III: Treatment of distress among relatives of missing persons

Reviews of literature on grief treatment indicate that CBT is the most promising treatment of grief-related distress (Boelen & Smid, 2017; Currier et al., 2010; Doering & Eisma, 2016). To our

knowledge, only one trial has evaluated treatment effects for relatives of missing persons (Hagl, Rosner, Butollo, & Powell, 2015). For the reasons set out in Chapter 8, the feasibility and potential effectiveness of CBT+M versus waiting list controls was evaluated in a pilot study in Chapter 9.

Systematic reviews and meta-analyses have shown that mindfulness-based interventions are effective in reducing current depressive symptoms and preventing recurrent depressive symptoms (Chiessa & Serretti, 2011; Galante, Iribarren, & Pearce, 2013). The effects of mindfulness in treating bereavement-related distress have previously been evaluated in two small studies among bereaved people (O'Connor, Piet, & Hougaard, 2014; Thieleman, Cacciatore, & Hill, 2014). In a controlled pilot study the effects of MBCT (n = 12) on symptom-levels of PG, depression, PTSD, and working memory were compared with waiting list controls (n = 18) among elderly bereaved people with clinical relevant levels of PG, depression, and/or PTS (O'Connor et al., 2014). A significantly larger reduction in symptom-levels of depression was found from pre-treatment to 5 months post-treatment for the MBCT group compared with the waiting list controls. In an uncontrolled trial the effects of a mindfulness-based intervention on symptom-levels of PTSD, general anxiety, and depression were evaluated among a treatment-seeking bereaved sample (n = 42). On average, all symptom-levels significantly reduced from pre- to post-treatment (Thieleman et al., 2014). Together with the preliminary findings from the study presented in Chapter 9, the results regarding mindfulness-based interventions to reduce grief-related distress are encouraging. Obviously, larger trials are required to draw firm conclusions about the effectiveness of mindfulness-based interventions for people exposed to deaths or disappearances.

Because of the use of a waiting list control group or the absence of a control group, the additional effect of integrating mindfulness in treatment of people confronted with a loss, has yet to be studied. Previous studies among people with clinical depression and people with heightened risk for relapse have shown that individual and group-based MBCT yield similar positive results as found with CBT (cf. Manicavasgar, Parker, & Perich, 2011; Omidi, Mohammadkhani, Mohammadi, & Zargar, 2013; Williams et al., 2014). Comparing the effects of CBT+M vs. CBT for people confronted with the death or disappearance of someone significant could shed light on whether the addition of mindfulness to CBT in treatment of PG is beneficial compared with CBT only.

We developed and used a treatment protocol of CBT+M, presented in Chapter 8, which enables replication by others. Although this protocol was based on CBT for PGD (Boelen, 2006; Boelen, de Keijser, van den Hout, & van den Bout, 2007) and MBCT for recurrent depression (Segal, Williams, & Teasdale, 2013) that have proven to be effective, it also deviated from these previous protocols. For instance, compared with CBT for PGD, our treatment consisted of eight sessions (vs. 12 sessions). Compared with MBCT, our protocol included weekly individual sessions of 45 minutes (vs. 2-hours weekly group sessions) and mindfulness-exercises were added as homework assignments (vs. in-session mindfulness exercises). Consequently, we would recommend using

evidence-based protocols for mindfulness-based interventions in future trials (cf. Segal et al., 2013), which increases the comparability of treatment effects between study samples.

In general, the findings from our pilot study were somewhat discouraging. Because of difficulties with recruiting sufficient participants for this pilot study, we adapted the initial analytic plan. For instance, we were not able to statistically test the possible change in psychopathology levels from pre-treatment to post-treatment/waiting between the immediate intervention group and waiting list controls. Therefore, the study does not allow us to draw firm conclusions about the potential effectiveness of CBT+M. Notwithstanding this limitation, this study generated some important recommendations for future research that have been discussed in detail in Chapter 9.

7. Methodological considerations

Some limitations that were not (sufficiently) addressed in the previous chapters should be noted.

Firstly, the data used in Chapter 3 through Chapter 6, and partly in Chapter 9, were based on the same dataset of 137 relatives of missing persons. The increased risk of false positive results, because of multiple testing, needs to be taken into account when considering the findings from all these studies (Streiner & Norman, 2011). Furthermore, two different subsamples (n = 23 in Chapter 7 and n = 17 in Chapter 9) were selected from this larger group to collect additional data. Because all collected data relied on the same self-selected sample, any biases present in this sample limit the generalizability of the findings to all relatives of missing persons (Stroebe, Stroebe, & Schut, 2003). For instance, people who signed up for participation in our research might have received less social support (than those who refused) and chose to participate as a way of sharing their experiences with others. Not wanting to look back on the disappearance, because they "got over it", might be an often used reason for refusing participation. Those who signed up for the study might therefore be more psychologically distressed than those who declined. Replication studies across independent representative samples of relatives of missing persons are therefore urgently needed.

Secondly, all measures used in this dissertation were validated in samples other than relatives of missing persons. More specifically, evaluation of the psychometric properties of the instruments predominantly took place in non-clinical samples of people exposed to a non-violent loss. Consequently, it is unknown to what extent previous findings regarding the psychometric properties of these instruments generalize to people exposed to a potential traumatic loss in general, and relatives of missing persons in particular. Furthermore, we adapted the wording referring to "death" to "disappearance" for some measures, which may result in different interpretation of these items. For instance, it is conceivable that the content of certain items of scales are less (e.g., "Feeling that a part of myself disappeared with the missing person" in the ICG(-r)) or more (e.g. "Seeing the missing person" or "Trouble accepting the disappearance" in the

ICG(-r)) applicable to relatives of missing persons than bereaved people. Validation studies across samples of people confronted with different types of losses can provide valuable insights into this matter.

Thirdly, the ICG(-r) was used as measure to assess symptom-levels of PG and other putative markers of disturbed grief in Chapters 2 through 6, and Chapter 9. The 19-item Inventory of Complicated Grief (ICG) is the most widely used self-report measure in empirical studies examining correlates (see for an overview Lobb et al. 2010), risk factors (see for an overview Burke & Neimeyer, 2013), and treatment of (see for an overview Boelen & Smid, 2017) disturbed grief reactions since its introduction in the mid 1990s (Prigerson et al. 1995). However, items of the ICG(-r) do not fully overlap with diagnostic criteria of Persistent Complex Bereavement Disorder (PCBD; per DSM-5; APA, 2013), PGD (per ICD-11; Maercker et al., 2013) or complicated grief (as proposed by Shear et al., 2011). Our findings may therefore not generalize to studies using PCBD, complicated grief, and/or PGD criteria.

8. **Future research directions**

There are some topics that may warrant more attention in bereavement research in general and research in relatives of missing persons in particular. Three of these topics are discussed below.

8.1 Prolonged grief following the disappearance and death of a significant other

We argue that the assessment of PG levels in relatives of missing persons is important for future research into ambiguous loss, but also more broadly for the field of bereavement research. Some people are hesitant regarding applying measures (i.e., the ICG) and diagnostic criteria of PG to relatives of missing persons. They have doubts whether people faced with ambiguous loss "..fit into the dimensions…" of the ICG-r (Glassock, 2006, p. 46). Hollander (2016) stated that it is not possible to diagnose relatives of missing persons with PG disorder, because PG is an abnormal reaction to a normal situation, whereas with ambiguous loss disturbed grief reactions are considered a normal response to an abnormal situation.

We acknowledge that pathologizing of grief reactions in people confronted with an ambiguous loss, but also an unambiguous loss, should be avoided. Yet, we plea, as illustrated by the work in this dissertation, for exploration of PG in relatives of missing persons for several reasons. To start with, assuming that *chronic* grief following the disappearance of a significant other is not pathological but rather natural is not based on empirical evidence, because (a) longitudinal studies among relatives of missing persons are lacking (see Chapter 2), (b) self-rated levels of PG among two samples of relatives of long-term missing persons (i.e., disappearance happened on average 12 and 15 years earlier) did not reach clinically relevant levels in at least 50% of the participants (see Chapter 3 and Heeke et al., 2015), and (c) findings from Chapter 7, based on

trajectories of functioning levels over time from retrospect, contradict that chronic reactions are the norm among relatives of missing persons, and even indicate that different pathways to "normal" functioning (i.e., functional level before the disappearance) could be identified following the disappearance of a significant other. Secondly, as a matter of fact, such claims assuming that grief is chronic following the disappearance of a significant other, could raise psychopathology levels through stigmatization (as was earlier described in more details in this General Discussion section).

To enhance our understanding of PG following ambiguous and unambiguous loss, it is important to conduct longitudinal studies examining how grief reactions develop over time. One interesting way to examine this is by using latent growth modeling. This method has been used previously to identify heterogeneous patterns of responses following loss (Aneshensel, Botticello, & Yamamoto-Mitani, 2004; Galatzer-Levy & Bonanno, 2012; Zhang, Mitchell, Bambauer, Jones, & Prigerson, 2008). These studies indicated that three to four trajectories are typical for people confronted with the death of a significant other due to an illness: 1) stable low levels of complaints (i.e., resilient pattern), 2) elevated levels of complaints followed by a significant decline (i.e., recovery pattern), 3) stable slightly elevated levels of complaints (i.e., repeatedly symptomatic pattern), and 4) chronic elevated levels of complaints (i.e., chronic pattern). However, it remains unclear to what extent these findings generalize to people confronted with the disappearance of a significant other.

Furthermore, this method may also be used to examine what time criterion should be applied in order to differentiate normal from PG reactions. As stated before, PG reactions do not differ from normal grief reactions with respect to the nature of the complaints, but rather the duration of the complaints (Bryant, 2014). According to the proposed diagnostic criteria for PCBD in DSM-5 (APA, 2013), grief reactions can be considered pathological following at least 12 months post-loss, whereas the ICD-11 (Maercker et al., 2013) state that PG disorder can be diagnosed 6 months post-loss. Because current studies using latent class growth modeling (Aneshensel et al., 2004; Galatzer-Levy & Bonanno, 2012; Zhang et al., 2008), examined predominantly depression levels over time following bereavement and/or examined grief-related complaints at least 6 months post-loss they are not able to answer this question about when grief reactions could be considered pathological. Future studies examining grief reactions over time, using different time intervals shortly to longer following the loss of a significant other may therefore be useful to enhance knowledge about when and for whom grief can be considered pathological.

Although we plea for more research into PG following the disappearance of a significant other, it is formally not (yet) possible to diagnose PCBD (as per DSM-5) or PG disorder (as per ICD-11) in relatives of missing persons. Not only because these diagnostic criteria are under consideration for inclusion in these diagnostic handbooks, but also because the loss event exclusively concerns

the *death* of a significant other (APA, 2013; Maercker et al., 2013). Extending this criterion to the disappearance of a significant other is, in our view, desirable to facilitate screening and treatment of, but also research on, disturbed grief reactions following the disappearance of a significant other.

8.2 Mechanisms of prolonged grief

According to cognitive-behavioural models (Beck, 1987; Boelen et al., 2006; Ehlers & Clark, 2000; Shear & Shair, 2005), negative cognitions and avoidance behaviours *underlie* disorders. In this dissertation we used similar frameworks in our cross-sectional studies in Chapter 4 – 6, which preclude to draw causal inferences. Longitudinal designs are of course needed to draw conclusions about reciprocal associations Although there is a growing body of longitudinal research examining the effects of negative cognitions and avoidance behaviours over time by controlling for baseline grief-related symptom levels (cf. Boelen & Eisma 2015; Eisma et al., 2015), these studies do not allow for drawing conclusions about how the "underlying" processes are reciprocally related to each other and to grief-related symptoms. More recently, cross-lagged models have been used, for instance in PTSD research, to explore possibly reciprocal association between cognitions, behaviours, and symptoms (Dekel, Peleg, & Solomon, 2013; Palosaari, Punamäki, Diab, & Qouta, 2013; Shahar, Noyman, Schnidel-Allon, & Gilboa-Schechtman, 2013). Exploring possible reciprocal associations between cognitions, behaviours, and grief-related symptomatology is not only of relevance for our understanding of the etiology of grief-related disorders, but has also important clinical implications. For instance, in case support will be found for negative cognitions preceding symptoms, psychoeducation about how negative cognitions may be related to later onset of symptomatology, may prevent people to develop symptoms. Or the other way around (in case support will be found for symptoms preceding negative cognitions), early interventions should focus less on challenging negative cognitions, but could for instance focus more on enhancing resilience, for instance by providing information on helpful coping strategies or bolstering social support in family support meetings (see for an overview Horn, Charney, & Feder, 2016).

8.3 Research among people exposed to forced displacement

Worldwide, we face one of the biggest forced displacement movements in history (UNHCR, 2017). Forced displacement is often accompanied with leaving family members and friends behind without knowing whether they are still alive (Chen, Hall, Ling, & Renzaho, 2017). This type of loss could be considered an ambiguous loss (Biehal, Mitchell, & Wade 2003). Disappearances due to forced displacement take place in a context with continued exposure to potential traumatic stressors pre-migration (e.g., deaths of family members, physical and sexual violence), but also

post-migration (e.g., unstable financial status, language barrier), resulting in heightened risk of experiencing comorbid symptoms (cf. Chen et al., 2017).

Given the magnitude and complexity of this phenomenon, more research on mental health issues in people exposed to forced displacement is needed to further our understanding about adequate screening and treating of psychological symptoms in this population. We identified at least three consecutive steps that could move the field forward: 1) systematic assessment of exposure to the (forced) disappearance of a significant other, 2) examination of trauma, depression, *and* grief responses, and 3) evaluation of treatment of complex trauma and multiple/ambiguous loss.

The first step is to systematically assess exposure to (forced) disappearances of significant others in the context of forced displacement. To date, not all checklists to assess trauma exposure include an item to assess exposure to the disappearance of a loved one. For instance, a large epidemiological study among over 3,000 Georgian (formerly) internally displaced people affected by armed conflict did not include an item on disappearance of a significant other in their trauma exposure checklist (Makhashvili et al., 2014). Studies that assessed exposure to disappearance of a significant other in asylum seekers and refugees, indicate that this is a frequently reported potentially traumatic event and an important risk factor of poor mental health (see for overviews Alemi, James, Cruz, Zepeda, Racadio, 2014; Fazel, Reed, Panter-Brick, & Stein, 2012).

The second step includes widening the scope of research to examination of grief reactions among people exposed to forced displacement. The vast majority of the current studies examining mental health issues among people who fled from armed conflict is focused on PTSD, depression, and general anxiety (see for an overview of 13 systematic reviews about mental health issues among refugees and asylum seekers Turrini et al., 2017). As suggested by others (Silove, Ventevogel, & Rees, 2017), examining mental health issues among refugees and asylum seekers should move beyond assessment of PTS, anxiety and depression, and should also focus on PG. The first studies that examined PG reactions in this population indicated that elevated PG levels are common and distinguishable from PTSD levels (Craig Sossou, Schnak, & Essex, 2008; Momartin, Silove, Manicavasagar, & Steel, 2004; Nickerson et al., 2014).

The third step constitutes of treatment of psychological distress in forcibly displaced people. Current evidence-based treatment of forcibly displaced people with mental disorders is predominantly focused on alleviating PTS symptoms with narrative exposure treatment (NET; Silove et al., 2017). Although this approach has shown to be effective for reducing PTS levels, it is not known to what extent this treatment is useful in reducing PG levels (Gwozdziewycz & Mehl-Madrona, 2013). Recently, brief eclectic psychotherapy for traumatic grief (BEP-TG), targeting comorbid symptoms, including PG reactions, has been developed for people exposed to trauma and (multiple/ambiguous) loss (Smid et al., 2015). Preliminary findings of a group-based day

treatment, including BEP-TG, were promising (de Heus et al., 2017). However, more research into the nature, prevalence, and treatment of PG in people exposed to complex trauma and (multiple/ambiguous) loss is needed to adequately support forcibly displaced people in need of professional help.

9. Recommendations for professionals

Resulting from the work in this dissertation several recommendations for professionals supporting relatives of missing person could be offered.

People working at support organizations (e.g., peer-support organizations, Victim Support, and International Committee of the Red Cross) and investigation services could use the knowledge derived from this dissertation to adequately inform relatives of missing persons about the risk of developing long-lasting psychological complaints, and what factors contribute to enhancing or alleviating these complaints. Based on our findings, we think it is important for people working at support organizations to stress to relatives of missing persons that the disappearance of a significant other is not inherently linked to chronic psychological complaints. Adopting strategies, such as openness and understanding toward one's own suffering, accepting that the disappearance is uncontrollable, and receiving emotional support from others, seem helpful. In addition, getting entangled in ruminative thinking about one's own sad mood and/or the causes and consequences of the disappearance is a common response in relatives of missing persons, but could be detrimental in the long-term. If needed, support organization could refer relatives of missing persons to licensed therapists divided over the Netherlands who have experience with treating people exposed to extraordinary, highly impact loss-related situations. The insights from this dissertation could also be used to stress to policy makers the need of exploring more options of how to support relatives of missing persons more effectively. For instance, by easing the financial burden for relatives of missing persons so that they can concentrate on the psychological adaptation process.

For people working therapeutically with relatives of missing persons it is recommended to use screening instruments for PGD, PTSD, and depression to gain insights into the nature of psychological complaints of the patient. In addition, it is worthwhile to examine levels of ruminative thinking, because this seems to be one of the most important factors coinciding with psychological symptoms. Due to the lack of sufficient research about the effects of treatment for relatives of missing people, it is not possible to recommend evidence-based treatment of psychological complaints post-disappearance. However, given the resemblances in nature and clinical correlates of distress between relatives of missing persons and deceased persons in combination with the findings of Chapter 9, it seems useful to target distress in relatives of missing persons with techniques from CBT. Adding mindfulness to CBT to teach relatives of missing

persons how to tolerate the uncertainty surrounding the disappearance might be worthwhile to consider.

10. Closure

This dissertation provides first insights in the severity of PG, PTS, and depression symptoms in relatives of long-term missing persons outside the context of armed conflict. Hypotheses regarding possible correlates of PG, PTS, and depression symptoms that are amendable to change in treatment of relative of missing persons are generated and tested. However, more research, preferably longitudinal studies, is urgently needed to examine to what extent our findings are replicable and generalize to all relatives of missing persons.

A treatment protocol tailored to relatives of missing persons was developed and the feasibility and potential effectiveness was evaluated. As a result of this dissertation, a nationwide network of therapists was trained to use our treatment protocol of CBT+M. Together with the treatment protocol, a treatment manual for relatives of missing persons was developed. This information is freely available at a website that was developed for relatives of missing persons and professionals (www.levenmetvermissing.nl). While, the results regarding CBT+M to reduce grief-related distress are encouraging, larger trials, preferably comparing CBT+M with an active control condition, are needed to draw firm conclusions about the effectiveness of mindfulness-based interventions for people exposed to the death or disappearance of a significant other.

Future research directions were provided, whereby we plea for further examination of 1) how grief reactions develops over time in people exposed to ambiguous and unambiguous loss 2) reciprocal associations between possible mechanisms underlying distress post-loss, and 3) grief-related distress in the large group of refugees and asylum seekers confronted with the disappearance of a significant other. Recommendation for professional working (therapeutically) with relatives of missing persons were given.

To conclude, in this dissertation we provided insights into the consequences of, and care after the long-term disappearance of a significant other outside the context of armed conflict; an event experienced by relatively few people, but for some with potentially long-lasting psychological consequences.

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Samenvatting

Eén op de tien mensen die worden geconfronteerd met een niet-gewelddadig overlijden van een dierbare, loopt risico om een langdurige rouwstoornis (LRS)1 te ontwikkelen (Lundorff, Holmgren, Zachariae, Farver-Vestergaard, & O'Connor, 2017). Het meemaken van de vermissing van een dierbare, verhoogt het risico op rouw-gerelateerd psychisch lijden (Kristensen, Weisæth, & Heir, 2012). LRS symptomen lijken op normatieve reacties van rouw, maar verschillen van deze reacties met betrekking tot de intensiteit en duur. Een diagnose van LRS kan van toepassing zijn op mensen die reacties van rouw ervaren die significant psychisch lijden en belemmeringen in het dagelijks leven veroorzaken, volgend op het overlijden van iemand die belangrijk voor hen was, waarbij het verlies ten minste zes maanden geleden plaatsvond (Prigerson et al., 2009). Verhoogde langdurige rouw (LR) klachten worden in verband gebracht met een veelheid van negatieve gevolgen voor de lichamelijke en geestelijke gezondheid (Maciejewski, Maercker, Boelen, & Prigerson, 2016; Prigerson et al., 1997). LR symptomen overlappen met, maar zijn te onderscheiden van, symptomen van posttraumatische stress (PTS) en depressie (Boelen, van den Bout, & de Keijser, 2003; O'Connor, Lasgaard, Shevlin, & Guldin, 2010).

Hoewel het niet voldoende empirisch onderzocht is, zijn er verscheidene redenen om aan te nemen dat het rouwproces volgend op de langdurige vermissing van een dierbare verschilt van het overlijden van een dierbare. Ten eerste kan het niet weten of het verlies tijdelijk of permanent is, leiden tot preoccupaties met de (omstandigheden van de) vermissing, wat op zijn beurt herstel van het verlies kan compliceren (Boss, 2006). Ten tweede kan het vasthouden aan de hoop op de terugkeer van de (lichamelijke overblijfselen van de) vermiste dierbare en het tegelijkertijd omgaan met de afwezigheid van diegene het voor mensen onmogelijk maken om verder te gaan met hun leven, wat LR reacties versterkt (Heeke, Stammel, & Knaevelsrud, 2015). Bovendien kan het uitblijven van een begrafenisceremonie of een herdenkingsplek ook bijdragen aan een verstoord rouwproces (Castle & Phillips, 2003). Ten vierde kan het ontbreken van specifieke wetgeving die families van vermiste personen in staat stelt om het wettelijk recht te verwerven om de zaken van de vermiste persoon te beheren[2], resulteren in aanzienlijke financiële gevolgen. Bijvoorbeeld, het verlies van het inkomen van een vermist persoon, terwijl de hypotheek, belastingen en verzekeringen van de vermiste blijven doorlopen, is waarschijnlijk ook een last voor degenen die achterblijven. Daarnaast kunnen familieleden van vermiste personen ermee worstelen om hun weg te vinden in complexe wettelijke en eigendomskwesties, met beperkte beschikbaarheid van professionele ondersteuning (Blaauw & Lähteenmäki, 2002; Holmes, 2008). Als laatste kunnen sociale marginalisatie, stigmatisering en gebrek aan sociale ondersteuning extra stressfactoren

1. Vrije vertaling van prolonged grief disorder
2. Met uitzondering van de verklaring van vermoedelijk overlijden die in Nederland na vijf jaar kan worden aangevraagd (Ministerie van Veiligheid en Justitie; 2017).

zijn die bijdragen aan verhoogde psychopathologie na de vermissing van een dierbare (Hollander, 2016; Quirk & Casco, 1994; Robins, 2010).

Echter wetenschappelijk onderzoek naar de psychologische gevolgen van de vermissing van een dierbare is schaars. Het overkoepelende doel van deze dissertatie was dan ook de psychische gevolgen van en de zorg na de langdurige vermissing van een dierbare in kaart te brengen. In Deel I van de dissertatie werd nader ingegaan op de psychologische impact van de vermissing van een dierbare. In Deel II werden verschillende correlaten van psychopathologie bij verwanten van vermisten onderzocht, waarbij werd ingezoomd op variabelen die vatbaar zijn voor verandering in therapie. In Deel III werd een rationale van een pilot behandelstudie gepresenteerd, waarbij cognitieve gedragstherapie met elementen van mindfulness (CGT+M) werd vergeleken met een wachtlijstcontrolegroep. Daarnaast werd de haalbaarheid en potentiële effectiviteit van CGT+M onderzocht.

1. Deel I: Fenomenologie van ambigu verlies

Boss (2006) bestempelt de vermissing van een dierbare als een ambigu verlies, omdat de vermiste persoon fysiek afwezig, maar psychologisch aanwezig is. Ze stelt dat dit soort verlies "het meest stressvolle soort verlies is vanwege de ambiguïteit" (p. 7). Echter berust deze stellingname op een klinische observatie en is onbekend in hoeverre dit empirisch wordt ondersteund. In Hoofdstuk 2 en Hoofdstuk 3 werd dit nader onderzocht.

1.1 Samenvatting van bevindingen in Hoofdstuk 2 - 3

In **Hoofdstuk 2** boden we een systematisch overzicht van de wetenschappelijke literatuur over psychische symptomen bij mensen die geconfronteerd zijn met de vermissing van een dierbare. Bevindingen uit elf kwantitatieve peer-reviewed studies naar de prevalentiecijfers en correlaten van psychische symptomen bij verwanten van vermisten werden samengevat. Daarnaast werden de resultaten beschreven van studies die de psychopathologie niveaus van verwanten van vermiste en overleden personen vergelijken. Het kleine aantal studies en heterogeniteit van de studies, bijvoorbeeld met betrekking tot de methodologische kwaliteit, gebruikte meetinstrumenten en samenstelling van samples, beletten het trekken van harde conclusies over prevalentiecijfers en correlaten van psychische symptomen bij verwanten van vermisten. In het algemeen ondersteunden de resultaten van zes vergelijkingsstudies de aanname niet dat verwanten van vermiste personen ernstigere psychologische symptomen rapporteerden dan verwanten van overleden personen. Het is belangrijk op te merken dat alle vergelijkingsstudies betrekking hadden op vergelijkingen van verwanten van personen die vermist raakten of werden gedood in een context van gewapend conflict (b.v. oorlog of staatsterrorisme). De meeste van deze verwanten waren ook blootgesteld aan andere potentieel traumatische gebeurtenissen

die mogelijk de aard en ernst van hun symptomen hebben beïnvloed. Als gevolg daarvan is het moeilijk, zo niet onmogelijk, om in deze studies de effecten van het verlies te onderscheiden van de effecten van andere potentieel traumatische stressoren.

Op basis van deze resultaten voerden we een vergelijkingsstudie uit buiten de context van gewapend conflict. In **Hoofdstuk 3** werden vragenlijstdata uit twee studies gebruikt om de niveaus van de symptomen van LR en PTS te vergelijken tussen 134 verwanten van langdurige vermisten en 331 nabestaanden van moord. In de groep van verwanten van vermisten rapporteerden respectievelijk 47.0% en 23.1% LR en PTS niveaus boven klinisch relevante ondergrenzen (voor meer informatie over ondergrenzen, zie Brewin, Andrews, & Rose, 2000; Hoge, Riviere, Wilk, Herrell, & Weathers, 2014; Prigerson et al., 1995). In de groep van nabestaanden van moord, rapporteerden respectievelijk 83.1% en 31.4% klinisch relevante LR en PTS niveaus. In tegenstelling tot eerdere aannames (Boss, 2006; Heeke & Knaevelsrud, 2015), waren LR en PTS niveaus significant hoger bij nabestaanden van moord dan bij verwanten van vermisten (respectievelijk *d* = .86 en .28).

2. Deel II: Strategieën voor het reguleren van emoties bij verwanten van vermisten

Het systematisch literatuuroverzicht in Hoofdstuk 2 toonde aan dat onderzoek naar de psychologische impact van de vermissing van een dierbare schaar is en exclusief gefocust is op vermissing in de context van gewapend conflict en geen licht werpt op factoren die verband houden met psychopathologie die vatbaar zijn voor verbetering door middel van therapie. In Hoofdstuk 4 - 7 stelden we ons ten doel deze lacune te vullen door de kennis te vergroten over waarom sommige mensen meer moeite kunnen hebben om om te gaan met de vermissing van een dierbare dan anderen. Om dat te doen, verzamelden we cross-sectionele onderzoeksgegevens in een steekproef van meer dan 130 mensen uit Nederland en België die waren geconfronteerd met de vermissing van een familielid, partner of vriend(in) ten minste drie maanden eerder (in Hoofdstuk 3 - 6). Bovendien werd er, tijdens de data-verzamelfase van de vragenlijststudie, een subgroep geselecteerd van eerstegraads familieleden of partners van vermiste personen die onder klinisch significante niveaus van LR, PTS en depressie scoorden (Hoofdstuk 7). Interviewgegevens werden verzameld onder deze subgroep van 23 mensen.

2.1 Samenvatting van de bevindingen in Hoofdstuk 4 - 7

In lijn met een cognitief-gedragsmodel van LR (Boelen, van den Hout, & van den Bout, 2006), gaven eerdere resultaten aan dat negatieve cognities en vermijdingsgedrag verband houden met psychisch lijden onder nabestaanden. In **Hoofdstuk 4** onderzochten we in welke mate deze resultaten generaliseerbaar waren naar verwanten van vermisten. In lijn met onze hypotheses, verklaarden negatieve cognities en vermijdingsgedrag 40% tot 60% van de variantie in niveaus

van LR, PTS en depressie bovenop sociodemografische variabelen. Dus, verwanten van vermisten die meer negatieve cognities hadden en meer vermijdingsgedrag vertoonden, liepen een grotere kans om verhoogde psychopathologie niveaus te ervaren. Op basis van deze bevindingen concludeerden we dat het helpend zou kunnen zijn om behandeling te richten op deze cognitieve-gedragsvariabelen.

Eerdere studies gaven aan dat het ervaren/uitdrukken van positieve emoties herstel na verlies stimuleert (Bonnano & Keltner, 1997; Keltner & Bonanno, 1997; Ong, Bergeman, Bisconti, & Wallace, 2006; Tweed & Tweed, 2011). Echter, onderzoek dat verbanden in kaart brengt tussen individuele verschillen in het *reguleren* van positief affect en psychopathologie na verlies ontbreekt. In **Hoofdstuk 5** stelden we ons ten doel deze lacune te vullen door te onderzoeken in welke mate strategieën van negatief en positief affect verband houden met psychopathologie volgend op de dood (Steekproef 1) en langdurige vermissing (Steekproef 2) van een dierbare. Strategieën voor het reguleren van negatief effect omvatten depressief rumineren, wat verwijst naar herhaaldelijk piekeren over de aard, oorzaken en gevolgen van een verdrietige/depressieve stemming. Strategieën voor het reguleren van positief affect omvatten het versterken en dempen van positief affect. Het versterken van positief affect (dat ook is aangeduid als "positief rumineren") heeft betrekking op "de neiging om op toestanden van positief affect te reageren met herhaaldelijke gedachten over positieve eigen kwaliteiten, positieve affectieve ervaring en de eigen gunstige levensomstandigheden" (Feldman Joormann, & Johnson, 2008, p. 509). Het dempen van positief affect verwijst naar "de neiging om op toestanden van positieve stemming te reageren met mentale strategieën die de intensiteit en duur van de toestand van positieve stemming verminderen" (Feldman et al. 2008, p. 509). Op basis van eerder werk op het gebied van depressie (Raes et al., 2014; Raes, Daems, Feldman, Johnson, & Van Gucht, 2009; Raes, Smets, Nelis, & Schoofs, 2012), formuleerden we de hypothese dat strategieën voor het reguleren van positief affect variantie in psychopathologie niveaus zouden verklaren bovenop strategieën voor het reguleren van negatief affect. Deze hypothese werd in twee verschillende steekproeven getoetst. Zoals verwacht, verklaarden strategieën voor het reguleren van positief affect variantie in niveaus van LR, PTS en/of depressie bovenop strategieën voor het reguleren van negatief affect. In afwachting van replicatie van onze bevindingen in longitudinaal onderzoek, ondersteunen deze bevindingen de bruikbaarheid van toekomstige verkenningen van hoe deze strategieën voor het reguleren van affect de geestelijke gezondheid na verlies beïnvloeden en hoe behandeling hier effectief op gericht kan worden.

De vermissing van een dierbare is inherent verbonden met onzekerheden (b.v. niet weten of de vermiste heeft geleden of levend of dood is) die onbeheersbaar zijn (Boss, 2006). Een vermissing kan daarom meer dan een natuurlijk overlijden (b.v. veroorzaakt door ziekte) aanleiding geven voor rumineren over de oorzaken en gevolgen van het verlies (Boss, 2006; Heeke et al., 2015).

Er is gesteld dat een houding van zelfcompassie (d.w.z. het herkennen en omarmen van eigen lijden en verdriet) als een buffer zou kunnen dienen tegen het verstrikt raken in rumineren, wat op zijn beurt de verergering van psychologisch psychisch lijden remt (Krieger, Altenstein, Baettig, Doerig, & Holtforth, 2013; Raes, 2010; Thompson & Waltz, 2008). In **Hoofdstuk 6** onderzochten we of meer zelfcompassie verband houdt met lagere psychopathologie niveaus bij verwanten van vermisten. Bovendien testten we in welke mate deze verbanden werden gemedieerd door herhaaldelijk nadenken over de oorzaken en gevolgen van het verlies (oftewel rouw rumineren). We concludeerden, onder voorbehoud vanwege onze cross-sectionele opzet, dat verwanten van vermisten die sterkere neigingen hebben om hun emotionele pijn op een open en begripvolle manier te benaderen (oftewel meer zelfcompassie) minder waarschijnlijk verstrikt raken in rumineren, wat op zijn beurt psychologische symptomen vermindert. Het tijdens behandeling versterken van een houding van zelfcompassie om rumineren tegen te gaan, bijvoorbeeld door middel van mindfulness training, kan daarom nuttig zijn voor het verlichten van emotioneel psychisch lijden volgend op de vermissing van een dierbare.

De twee hoofddoelen van de interview studie, gepresenteerd in **Hoofdstuk 7**, waren 1) het retrospectief onderzoeken van functioneringspatronen over tijd, en 2) het verkennen van welke coping strategieën mensen met relatief lage niveaus van LR, PTS en depressie het meest helpend vonden bij het omgaan met de vermissing. In het eerste deel van het interview werd de deelnemer gevraagd om een grafiek te tekenen van het beloop van zijn/haar functioneren vanaf een jaar voorafgaand aan de vermissing tot op de dag van het interview (cf. Burr and Klein 1994). Het meest frequent geïdentificeerde functioneringspatroon over tijd was het herstelpatroon. Vijftien van de 23 mensen rapporteerden dit patroon dat wordt gekenmerkt door een aanvankelijke verslechtering van het functioneren vlak na de vermissing, gevolgd door een stabiele verbetering van het functioneren. Zeven van de 23 mensen rapporteerden een stabiel/veerkrachtig patroon dat wordt gekenmerkt door een hoog niveau van functioneren zonder significante verslechtering of verbetering. Het tweede deel van het interview bestond uit een kaart-sorteertaak (cf. Paap et al., 2014). We presenteerden 15 kaarten die alle 15 subschalen vertegenwoordigden van een meetinstrument om coping strategieën te meten (the COPE easy; Kleijn, Heck, & Waning, 2000). We gaven de deelnemers de opdracht om vijf van de vijftien kaarten uit te kiezen die, naar zijn/haar mening, het meest helpend waren geweest bij het omgaan met de vermissing sinds het moment dat die plaatsvond. Vervolgens werd de deelnemer gevraagd om uit te leggen waarom hij/zij de gekozen coping strategie als helpend beschouwde. Situatie accepteren, geïnterpreteerd als leren leven met het niet weten (Boss, 2006), werd het vaakst gekozen als helpende coping strategie. Dit gaf aan dat, volgens familieleden met weinig tot geen symptomen die terugkeken op reacties op de vermissing, het leren verdragen van de onzekerheid van het allergrootste belang is. Het uiten van emoties tegen en ontvangen van morele steun van familieleden en vrienden werden ook

vaak gekozen als helpende coping strategieën. Dit benadrukt het belang van het integreren van de sociale context van een cliënt, zoals interpersoonlijke relaties, in professionele ondersteuning van familieleden van vermiste personen. Tenslotte werd afleiding zoeken, omschreven als bezig zijn met sociale of beroepsactiviteiten om herhaaldelijk negatief denken te voorkomen, ook gekozen als een helpende coping strategie.

3. Deel III: Behandeling psychische klachten bij verwanten van vermisten

Alles bij elkaar, gaven de bevindingen van deze drie correlationele studies (Hoofstuk 4 - 6) aan dat, vergelijkbaar met nabestaanden, verwanten van vermisten die meer negatieve cognities ervaren, en meer vermijdingsgedrag en rumineren vertonen, waarschijnlijk meer verhoogde psychopathologie niveaus ervaren, waaronder LR, PTS en depressie. We breidden eerder werk over inadequate coping strategieën na het verlies van een dierbare uit door de rol van adaptieve coping strategieën te exploreren. Onze bevindingen suggereren dat het vergroten van positief affect door optimistische gedachten over jezelf en innerlijke positieve emoties (b.v. het versterken van positief affect) en meer zelfcompassie potentieel beschermende factoren zijn tegen het ervaren van psychische klachten na de vermissing van een dierbare (Hoofdstuk 5 - 6). Deze bevindingen lijken ons initiatief te ondersteunen voor het evalueren van de potentiële effectiviteit van CGT+M voor verwanten van vermisten met verhoogde psychopathologie niveaus.

3.1 Behandeling: Samenvatting van bevindingen in Hoofdstuk 8 - 9

CGT gericht op het aangaan van de confrontatie met de realiteit van het verlies en de onomkeerbaarheid daarvan is de voorkeursbehandeling voor nabestaanden (Boelen & Smid, 2017; Currier, Holland, & Neimeyer, 2010; Doering & Eisma, 2016). Clinici hebben gesteld dat het uitoefenen van druk op verwanten van vermisten tijdens behandeling om “verder te gaan” of het “af te sluiten” contraproductief is en weerstand kan oproepen (Boelen & Smid, 2017; Boss, 2006; Glassock, 2006). In plaats van te focussen op onbeheersbare externe factoren gerelateerd aan de vermissing, aangewakkerd door onzekerheid die gepaard gaat met de vermissing, zou het leren tolereren van aanhoudende negatieve gedachten en gevoelens, bijvoorbeeld door middel van mindfulness training, behulpzaam kunnen zijn (Boss, 2006). In **Hoofdstuk 8** boden we een onderbouwing voor een pilot gerandomiseerd gecontroleerd onderzoek om de haalbaarheid en potentiële effectiviteit van CGT+M versus wachtlijst controlegroep te evalueren voor het verminderen van niveaus van LR, PTS en depressie en het vergroten van mindfulness bij verwanten van vermisten die in aanmerking kwamen voor professionele hulp. Op basis van de bevindingen van deze pilotstudie, beschreven in **Hoofdstuk 9**, concludeerden we dat, met uitzondering van één op de negen deelnemers, CGT+M samenviel met verminderingen van de ernst van psychopathologie. Dit geeft aan dat de behandeling veelbelovend genoeg is om

verder onderzoek te rechtvaardigen. Echter, om de haalbaarheid van toekomstige studies onder verwanten van vermisten te vergroten, bevelen we aan om internationaal samen te werken en/of de duur van de wervingsfase te verlengen, om de omvang van de steekproef te maximaliseren.

4. Aanbevelingen voor professionals

Als resultaat van het werk in deze dissertatie zouden verscheidene aanbevelingen voor professionals die verwanten van vermisten ondersteunen, kunnen worden gedaan.

Mensen die bij hulporganisaties werken (b.v. lotgenotencontact organisaties, Slachtofferhulp, het Internationale Comité van het Rode Kruis) en opsporingsdiensten zouden de kennis ontleend aan deze dissertatie kunnen gebruiken om verwanten van vermisten adequaat te informeren over het risico op het ontwikkelen van langdurige psychologische klachten, en over welke factoren bijdragen aan het vergroten of verlichten van deze klachten. Op basis van onze bevindingen zijn wij van mening dat het belangrijk is voor mensen die bij hulporganisaties werken om naar verwanten van vermisten toe te benadrukken dat de vermissing van een dierbare niet inherent verbonden is met chronische psychologische klachten. Aanpassingsstrategieën, zoals openheid en begrip ten opzichte van het eigen lijden, aanvaarden dat de vermissing onbeheersbaar is, en het ontvangen van emotionele steun van anderen, lijken te helpen. Daarnaast is verstrikt raken in herhalende gedachten over de eigen verdrietige stemming en/of de oorzaken en gevolgen van de vermissing een veelvoorkomende reactie bij verwanten van vermisten, die echter op de lange termijn schadelijk zou kunnen zijn. Indien nodig, zouden hulporganisaties verwanten van vermisten kunnen verwijzen naar bevoegde therapeuten verspreid over Nederland die ervaring hebben met het behandelen van mensen die zijn blootgesteld aan buitengewone verliessituaties. De inzichten uit deze dissertatie zouden ook gebruikt kunnen worden om richting beleidsmakers de noodzaak te benadrukken van het verkennen van meer mogelijkheden om familieleden van vermiste personen effectiever te ondersteunen. Bijvoorbeeld door de financiële last voor familieleden van vermiste personen te verlichten, zodat zij zich meer kunnen concentreren op het psychologische aanpassingsproces.

Voor mensen die therapeutisch met verwanten van vermisten werken, wordt aanbevolen om screeningsinstrumenten voor LRS, PTSS en depressie te gebruiken om de aard van de klachten in kaart te brengen. Verder is het zinvol om niveaus van rumineren te onderzoeken, omdat dit één van de belangrijkste factoren lijkt te zijn die samenhangen met psychologische symptomen. Vanwege het gebrek aan voldoende onderzoek naar de effecten van behandeling voor verwanten van vermisten, is het niet mogelijk om een evidence-based behandeling aan te bevelen voor het reduceren van psychische klachten na de vermissing van een dierbare. Echter, gezien de overeenkomsten in de aard en klinische correlaten van psychisch lijden tussen verwanten van vermiste en overleden personen, in combinatie met de bevindingen in Hoofdstuk 9, lijkt het

zinvol om psychisch lijden bij verwanten van vermisten met aan CGT ontleende technieken te behandelen. Het toevoegen van mindfulness aan CGT om verwanten van vermisten de onzekerheid te leren verdragen, zou het overwegen waard kunnen zijn.

5. Tot slot

Door middel van deze dissertatie worden eerste inzichten geboden in de ernst van symptomen van LR, PTS en depressie bij verwanten van vermisten buiten de context van gewapend conflict. Hypotheses met betrekking tot mogelijke correlaten van symptomen van LR, PTS en depressie die vatbaar zijn voor verandering in therapie werden gegenereerd en getest. Echter, meer onderzoek, bij voorkeur longitudinale studies, is dringend nodig om te onderzoeken in welke mate onze bevindingen repliceerbaar zijn en generaliseerbaar zijn naar alle verwanten van vermisten.

Een behandelprotocol afgestemd op verwanten van vermisten is ontwikkeld en de haalbaarheid en potentiële effectiviteit werd geëvalueerd. Als resultaat van deze dissertatie, werd een landelijk netwerk van therapeuten getraind om het behandelprotocol van CGT+M te gebruiken. Samen met het behandelprotocol werd een werkboek ontwikkeld voor verwanten van vermisten. Deze informatie is gratis beschikbaar op een website die werd ontwikkeld voor verwanten van vermisten en voor professionals (www.levenmetvermissing.nl). Hoewel de resultaten met betrekking tot CGT+M om rouw-gerelateerd psychisch lijden te verminderen bemoedigend zijn, zijn grotere studies, bij voorkeur studies waarbij CGT+M wordt vergeleken met een actieve controlegroep, nodig om harde conclusies te trekken over de effectiviteit van op mindfulness gebaseerde interventies bij mensen die zijn blootgesteld aan het overlijden of vermissing van een dierbare.

Er zijn onderzoeksrichtingen voor de toekomst geboden, waarmee we pleiten voor verdere verkenning van 1) hoe rouwreacties zich over tijd ontwikkelen bij mensen blootgesteld aan ambigu en niet-ambigu verlies, 2) mogelijke mechanismen die aan dit psychisch lijden ten grondslag liggen, en 3) aan rouw-gerelateerd psychisch lijden in de grote groep vluchtelingen die zijn geconfronteerd met de vermissing van een dierbare. Aanbevelingen voor professionals die (therapeutisch) werken met verwanten van vermisten zijn gegeven.

Concluderend, in deze dissertatie boden we inzichten in de gevolgen van en de zorg na de langdurige vermissing van een dierbare buiten de context van gewapend conflict; een gebeurtenis die relatief weinig mensen meemaken, maar die voor sommigen mogelijk lang aanhoudende psychologische gevolgen heeft.

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Dankwoord

Dit gedeelte van mijn proefschrift wil ik optimaal gebruiken om mensen te bedanken die hebben bijgedragen aan de totstandkoming van dit proefschrift.

In de eerste plaats wil ik alle mensen die hebben deelgenomen aan het onderzoek bedanken. Ik beschouw het als een voorrecht dat ik velen van jullie heb mogen ontmoeten. Ik blijf het bijzonder vinden dat jullie bereid waren om ervaringen met mij te delen. Alle kaarten, brieven, boeken, foto's, aangrijpende verhalen en lange e-mails die ik van jullie heb mogen ontvangen hebben een diepe indruk achtergelaten.

Jos de Keijser, Paul Boelen en Ineke Wessel, als (co-)promotoren van dit project hebben jullie mij gesteund, geïnspireerd en gemotiveerd. Jos, jij hebt mij het vertrouwen en de vrijheid geven om dit onderzoeksproject vorm te geven. Onze soms lange en enerverende gesprekken waren nooit saai. De manier waarop jij mensen, organisaties, onderzoek, klinische praktijk en onderwijs bij elkaar brengt vind ik bewonderingswaardig!

Paul, samenwerken met jou is een eer. Met veel dank aan jouw input hoop ik dat mijn missie is geslaagd; dat mijn proefschrift het kleinere zusje is van de jouwe. Feedback, gesprekken, e-mails, congresbezoeken, rook- en Joni Mitchell-momenten van/met jou hebben mij als onderzoeker en mens verrijkt! Ik hoop nog lang van jou te mogen leren.

Ineke, jouw directheid en kritische blik (bijvoorbeeld jouw feedback op een eerste versie van de systematische review: "Het lijkt wel een omgevallen boekenkast") vind ik een verademing! Jouw ethische kompas hield mij scherp. De grondige wijze waarop jij naar de manuscripten hebt gekeken is onmisbaar geweest voor dit proefschrift.

Prof. dr. Rita Rosner, Prof. dr. Miranda Olff en Prof. dr. Robbert Sanderman bedankt dat jullie de tijd hebben genomen om het proefschrift te lezen en te beoordelen.

De samenwerkingspartners, de Vereniging Achterblijvers na Vermissing (de VAV), Slachtofferhulp Nederland, de Nederlandse Politie, AvroTros Vermist, Child Focus, en Jaap Schoof wil ik bedanken voor hun steun bij de dataverzameling.

Henk Langerak, in jouw rol als voorzitter van de VAV heb ik de kans gekregen om met familieleden van vermiste personen in contact te komen. De jaarlijkse VAV-bijeenkomst op de eerste vrijdag van juni in het Moreelsepark in Utrecht heeft mij elke keer weer enorm aangegrepen en doen beseffen voor wie ik dit doe.

Chantal van Disseldorp en Franck Wagemakers, jullie betrokkenheid, als projectleider "Vermissing" bij Slachtofferhulp, heeft een belangrijke bijdrage geleverd aan het ondersteunen van achterblijvers en aan dit onderzoek. Chantal, ook bedankt voor de onvergetelijke nacht in Brussel!

Irma Schijf en Carlo Schippers, namens het Landelijk Bureau Vermiste Personen van de Nederlandse Politie waren jullie vanaf de start betrokken bij dit onderzoek. Irma, jouw wijze van presenteren en spreken heeft altijd een diepe indruk op mij gemaakt. Via jou kwam ik in contact

met Carlo Schippers en Jaap Schoof. Carlo, jouw charmes zijn van wezenlijk belang geweest voor het slagen van dit onderzoek. Jij bracht mij in contact met Loes Leeman van AvroTros Vermist. Het bezoek van jou, Jos en mij aan de studio van AvroTros Vermist in het Mediapark in Hilversum en mijn bezoek aan jou op het hoofdkantoor in Zoetermeer kijk ik met veel plezier op terug! Loes Leeman, zonder jouw hulp was het niet mogelijk geweest om zoveel achterblijvers na vermissing te benaderen; ontzettend bedankt! Jaap Schoof, als voormalig directeur van het Watersnoodrampmuseum heb jij mij een bijzondere dag bezorgd in Zeeland. Bedankt voor de privérondleiding en de lunch.

Het bestuur en de donateurs van het Fonds Slachtofferhulp, jullie hebben dit onderzoek financieel mogelijk gemaakt, waarvoor ontzettend bedankt. Carlo Contino, Sandra Scherpenisse, Nikki Vader en Dusjka Stijfhoorn bedankt voor jullie betrokkenheid en interesse tijdens de uitvoering van het onderzoek. Ook Stichting Stimuleringsfonds Rouw en de Rijksuniversiteit Groningen bedankt voor het mede-financieren van het onderzoek.

Loren O'Keeffe we had a great time in Belgium, Scotland, and Melbourne. You are incredible! Keep up the good work as founder of the *Missing Persons Advocacy Network* in Australia!

Alle mensen met wie ik heb mogen samenwerken aan de artikelen in dit proefschrift: MSc. Doety de Vries, Dr. Mariëtte van Denderen, Prof. dr. Marieke Timmerman, Dr. Maarten Eisma, MSc. Eline Piersma, BSc. Melanie Lemmers, Dr. Steven de Jong, MSc. Anna van der Gaast-Witkowska, MSc. Sander Sprik, MA. Liesbeth E. A. Rischen-Sindram, MSc. Marjolijn Dijkhuis en BSc. Hedwig de Boer bedankt voor jullie inzet en bijdragen!

Alle therapeuten uit het Netwerk Traumatisch Verlies die bereid waren achterblijvers te begeleiden tijdens de behandeling ontzettend bedankt voor jullie medewerking: Pauline Borduin, Jantien Piekstra, Nettie Herrmann, Hetty Vromen, Ursula Hendriks-van den Bos, Carin de Ridder, Geert Smid, Marije van den Wijngaard, Simone de la Rie, Welmoed Pietersen, Kim van der Heiden, Natashja van den Ameele-Mullié en Welmoed Pietersen. Jullie enthousiasme, betrokkenheid, professionaliteit en deskundigheid is inspirerend!

Collega's van Klinische Psychologie en Experimentele Psychopathologie van de Rijksuniversiteit Groningen, Klinische Psychologie van de Universiteit Utrecht en Psychology van de University of New South Wales bedankt dat jullie de afdeling tot een prettige werkplek maken! Lange kantoordagen doormaken met jullie voelt als thuiskomen. Hendry, ik word altijd blij als ik jou weer zie! Ingrid van der Werff, bedankt voor al jouw hulp en gezelligheid!

De collega's van de Wijvindenfeestjesleuk.-appgroep, Rebecca Heron, Minita Franzen, Alieke Groot-Koerkamp, Rachel de Jong en Rosa Bos, bedankt voor het dansen tot in de vroege uurtjes! Rebecca, thanks for reading and correcting parts of this dissertation and for bringing sunny Carribean-vibes into our office!

Maarten Eisma, door jouw komst naar de RUG heb ik een sparringpartner, wandel-, congres-, koffie-, en kroegmaatje! Ik zie er dan ook enorm naar uit om de komende jaren met jou te blijven werken.

Steven rookbuddy de Jong, och wat is het fijn om jouw vriend te zijn! Onze gesprekken over papers, wetenschap, muziek, liefde, unieke sneeuwvlokjes en het leven zijn voor mij van cruciaal belang. Ik vind het elke keer weer heerlijk om een kijkje te krijgen in jouw bijzondere scherpe geest!

Mariëtte van Denderen, wij hebben jarenlang met veel plezier een kantoor en onze ervaringen gedeeld. Jouw promotieproject *Grief following homicidal loss* heeft de basis gelegd voor mijn proefschrift. Jij was altijd bereid om mij te helpen, waarvoor dank! Dankzij jou is mijn verhuizing naar Groningen mogelijk en dragelijk geweest. Onze avondjes naar de film, uit eten en theedrinken zijn voor mij zeer waardevol.

Doety de Vries en Eline Piersma, onze samenwerking was een feest! Ik koester met name onze vriendschap; urenlange gesprekken voeren inclusief aanstekelijke lachsalvo's en ophalen van herinneringen. Ik hoop dat wij nooit zijn uitgepraat.

Minou Lamberink, bestie, wat ben ik blij dat wij al bijna 15 jaar in de weekenden kletsen, series kijken en de bloemetjes buiten zetten. Frank Lenferink, Willemijn ten Hoopen, Ottilie Puijk, Geke Douwsma, Niels Landmeter, Sanne Oomens, en Marnix, Sarah, Thea en Eelco Sixma bedankt voor jullie steun en gezellige momenten.

Dit werk had nooit tot stand kunnen komen zonder de mensen die mij hebben gesteund en geïnspireerd in de weg ernaartoe. Harrie, de inzichten die jij mij hebt verschaft zijn van onschatbare waarde en zal ik mijn hele leven met mij meenemen. Christina Bode, jij was diegene die mij enthousiasmeerde voor de wetenschap, als eerste mijn naam in de wetenschappelijke databases heeft vereeuwigd en als referent mij heeft aanbevolen als PhD, bedankt! Muirne Paap, jij gaf mij de kans om tijdens en na mijn afstuderen als onderzoeksassistent mee te werken aan jouw post-doc project en leerde mij hoe het uitvoeren van succesvol, gedegen onderzoek er uit kan zien.

Pap en mam, jullie hebben mij geleerd wat hard werken is. Dat is wat jullie altijd hebben gedaan. Ik ben trots dat ik jullie dochter ben!

Klaas-Jelmer, door ons samenzijn weet ik dat liefde op het eerste gezicht bestaat, weet ik hoe het is om onvoorwaardelijk van iemand te houden en weet ik hoe het voelt om begrepen en geliefd te worden; "*The greatest thing you'll ever learn is just to love and be loved in return*" (George Alexander Aberle, Nature Boy).

Curriculum Vitae

Lonneke Ingrid Maria Lenferink was born in 1988 in Zwolle and grew up in a small town, Heino, close to Zwolle. From when she was little she was fascinated by music. At the age of 10 she started to play the guitar. At the age of 14 she started to sing and perform. During the last two years of the pre-university education she took jazz vocal lessons at the conservatory. She met jazz guitar player Klaas-Jelmer Sixma at one of her jazz classes. They formed a jazz duo for several years. One of the highlights during their music career was winning the Dutch National Jazz Vocal Award. Lonneke and Klaas-Jelmer did not only share their passion for jazz music, but fell in love, started a family with Pip (a lovely Staffordshire Bullterrier) and are together ever since.

After Lonneke saw a friend recovering from a psychological trauma, by a few sessions of Eye Movement Desensitisation and Reprocessing (EMDR) treatment, she was keen to become a therapist. She started to study psychology at Twente University in Enschede in 2008. While conducting her bachelor thesis about evaluating the psychometric properties of a filial maturity measure, under the supervision of Dr. Christina Bode, she became fascinated by research. Her bachelor thesis resulted in her first academic publication as a co-author. After graduating in 2013, she started a research assistant position at Twente University under the supervision of Dr. Muirne Paap, which resulted in three co-authored academic publications about assessment of quality of life in patients with chronic obstructive pulmonary disease. In 2014 she started her PhD position at the University of Groningen and Utrecht University, which resulted in the current dissertation.

After flight MH17, departing from Amsterdam, crashed in Ukraine due to a missile impact on July 17 2014 a research project was initiated in 2015 that aims to examine risk factors and treatment of psychological distress in the bereaved of the plane crash. Lonneke is responsible for the data-collection and -analysis of this research project, under the supervision of Prof. dr. Jos de Keijser, Prof. dr. Paul Boelen, and Dr. Geert Smid. During this on-going project she collaborated with researchers from the University of New South Wales (Sydney), Tilburg University (Tilburg), University of Twente (Enschede), Arq Psychotrauma Expert Group (Diemen), and Utrecht University (Utrecht). After receiving her PhD, she will continue to work on this research project, while she will also start as post-doc researcher at a project examining the consequences of, and care after the death of a significant other due to a traffic accident.

Publications in peer-reviewed international journals

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Manuscripts submitted for publication

Boelen, P. A., Lenferink, L. I. M., Nickerson A., & Smid, G. E. (submitted). Comparative evaluation of the factor structure, prevalence, and validity of disturbed grief in DSM-5 and ICD-11.

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Front cover contains edited photos based on originals from:

Alwin Sterk, born in 1951, missing since 1972; Sicco Cuperus, born in 1978, missing since 2014; Jelle Leemans, born in 1986, missing since 2013; Ingrid van Reeuwijk, born in 1965, missing since 2000; Caspar Rive, born in 1967, missing since 1993; Ineke Fleurke, born in 1965, missing since 1994; Rina van Grinsven, born in 1961, missing since 1988; Albert Snijder, born in 1957, missing since 1995; Frank Tuin, born in 1990, missing since 2011; Herman Ploegstra, born in 1974, missing since 2010; Mieke Gulien, born in 1962, missing since 1989; Milorad Mitrovic born in 1946, missing since 1994; Rob Bouwmeester, born in 1967, missing since 2007; Paul Groen, born in 1934, missing since 2001; Loes Tevel, born in 1955, disappeared and deceased in 2014