

GRIEF DISORDERS: CLINICAL, CULTURAL, AND EPIDEMIOLOGICAL ASPECTS

EDITED BY: Clare Killikelly, Geert E. Smid and Birgit Wagner

PUBLISHED IN: Frontiers in Psychiatry and Frontiers in Psychology





frontiers

Frontiers eBook Copyright Statement

The copyright in the text of individual articles in this eBook is the property of their respective authors or their respective institutions or funders. The copyright in graphics and images within each article may be subject to copyright of other parties. In both cases this is subject to a license granted to Frontiers.

The compilation of articles constituting this eBook is the property of Frontiers.

Each article within this eBook, and the eBook itself, are published under the most recent version of the Creative Commons CC-BY licence.

The version current at the date of publication of this eBook is CC-BY 4.0. If the CC-BY licence is updated, the licence granted by Frontiers is automatically updated to the new version.

When exercising any right under the CC-BY licence, Frontiers must be attributed as the original publisher of the article or eBook, as applicable.

Authors have the responsibility of ensuring that any graphics or other materials which are the property of others may be included in the CC-BY licence, but this should be checked before relying on the CC-BY licence to reproduce those materials. Any copyright notices relating to those materials must be complied with.

Copyright and source acknowledgement notices may not be removed and must be displayed in any copy, derivative work or partial copy which includes the elements in question.

All copyright, and all rights therein, are protected by national and international copyright laws. The above represents a summary only. For further information please read Frontiers' Conditions for Website Use and Copyright Statement, and the applicable CC-BY licence.

ISSN 1664-8714

ISBN 978-2-88966-994-3

DOI 10.3389/978-2-88966-994-3

About Frontiers

Frontiers is more than just an open-access publisher of scholarly articles: it is a pioneering approach to the world of academia, radically improving the way scholarly research is managed. The grand vision of Frontiers is a world where all people have an equal opportunity to seek, share and generate knowledge. Frontiers provides immediate and permanent online open access to all its publications, but this alone is not enough to realize our grand goals.

Frontiers Journal Series

The Frontiers Journal Series is a multi-tier and interdisciplinary set of open-access, online journals, promising a paradigm shift from the current review, selection and dissemination processes in academic publishing. All Frontiers journals are driven by researchers for researchers; therefore, they constitute a service to the scholarly community. At the same time, the Frontiers Journal Series operates on a revolutionary invention, the tiered publishing system, initially addressing specific communities of scholars, and gradually climbing up to broader public understanding, thus serving the interests of the lay society, too.

Dedication to Quality

Each Frontiers article is a landmark of the highest quality, thanks to genuinely collaborative interactions between authors and review editors, who include some of the world's best academicians. Research must be certified by peers before entering a stream of knowledge that may eventually reach the public - and shape society; therefore, Frontiers only applies the most rigorous and unbiased reviews.

Frontiers revolutionizes research publishing by freely delivering the most outstanding research, evaluated with no bias from both the academic and social point of view. By applying the most advanced information technologies, Frontiers is catapulting scholarly publishing into a new generation.

What are Frontiers Research Topics?

Frontiers Research Topics are very popular trademarks of the Frontiers Journals Series: they are collections of at least ten articles, all centered on a particular subject. With their unique mix of varied contributions from Original Research to Review Articles, Frontiers Research Topics unify the most influential researchers, the latest key findings and historical advances in a hot research area! Find out more on how to host your own Frontiers Research Topic or contribute to one as an author by contacting the Frontiers Editorial Office: frontiersin.org/about/contact

GRIEF DISORDERS: CLINICAL, CULTURAL, AND EPIDEMIOLOGICAL ASPECTS

Topic Editors:

Clare Killikelly, University of Zurich, Switzerland

Geert E. Smid, ARQ National Psychotrauma Centre, Netherlands

Birgit Wagner, Medical School Berlin, Germany

Citation: Killikelly, C., Smid, G. E., Wagner, B., eds. (2021). Grief Disorders: Clinical, Cultural, and Epidemiological Aspects. Lausanne: Frontiers Media SA. doi: 10.3389/978-2-88966-994-3

Table of Contents

- 05 Editorial: Grief Disorders: Clinical, Cultural, and Epidemiological Aspects**
Geert E. Smid, Clare Killikelly and Birgit Wagner
- 09 Prolonged Grief Disorder and the Cultural Crisis**
Eva-Maria Stelzer, Ningning Zhou, Andreas Maercker, Mary-Frances O'Connor and Clare Killikelly
- 15 Mania Following Bereavement: State of the Art and Clinical Evidence**
Claudia Carmassi, Katherine M. Shear, Martina Corsi, Carlo Antonio Bertelloni, Valerio Dell'Oste and Liliana Dell'Osso
- 22 Complicated Grief: What to Expect After the Coronavirus Pandemic**
Camilla Gesi, Claudia Carmassi, Giancarlo Cerveri, Barbara Carpita, Ivan Mirko Cremone and Liliana Dell'Osso
- 27 Post-Migration Stressors and Their Association With Symptom Reduction and Non-Completion During Treatment for Traumatic Grief in Refugees**
A.A.A. Manik J. Djelantik, Annemiek de Heus, Diede Kuiper, Rolf J. Kleber, Paul A. Boelen and Geert E. Smid
- 38 Valid ICD-11 PGD Scales and Structured Clinical Interviews Needed**
Maja O'Connor, Lene Larsen, Biretha V. Joensen, Paul A. Boelen, Fiona Maccallum, Katrine Komischke-Konnerup and Richard A. Bryant
- 42 Prolonged Grief Disorder Among Refugees in Psychological Treatment— Association With Self-Efficacy and Emotion Regulation**
Oriane Lacour, Naser Morina, Julia Spaaij, Angela Nickerson, Ulrich Schnyder, Roland von Känel, Richard A. Bryant and Matthias Schick
- 52 Web-Based Bereavement Care: A Systematic Review and Meta-Analysis**
Birgit Wagner, Nicole Rosenberg, Laura Hofmann and Ulrike Maass
- 65 Association Between Prolonged Grief and Attitudes Toward Reconciliation in Bereaved Survivors of the Khmer Rouge Regime in Cambodia**
Nadine Stammel, Louisa Heinzl, Carina Heeke, Maria Böttche and Christine Knaevelsrud
- 76 Coping Strategies and Considering the Possibility of Death in Those Bereaved by Sudden and Violent Deaths: Grief Severity, Depression, and Posttraumatic Growth**
Joscelyn E. Fisher, Jing Zhou, Rafael F. Zuleta, Carol S. Fullerton, Robert J. Ursano and Stephen J. Cozza
- 86 ICD-11 Prolonged Grief Disorder Criteria: Turning Challenges Into Opportunities With Multiverse Analyses**
Maarten C. Eisma, Rita Rosner and Hannah Comtesse
- 91 The UK National Homicide Therapeutic Service: A Retrospective Naturalistic Study Among 929 Bereaved Individuals**
Suzan Soydas, Geert E. Smid, Barbara Goodfellow, Rachel Wilson and Paul A. Boelen
- 102 Traumatic Bereavements: Rebalancing the Relationship to the Deceased and the Death Story Using the Two-Track Model of Bereavement**
Simon Shimshon Rubin, Ruth Malkinson and Eliezer Witztum

- 114** *Different Trajectories of Prolonged Grief in Bereaved Family Members After Terror*
Pål Kristensen, Kari Dyregrov and Rolf Gjestad
- 122** *Psychometric Evaluation of the Swedish Version of the Prolonged Grief Disorder-13 (PG-13) in a Bereaved Mixed Trauma Sample*
Josefin Sveen, Kristina Bondjers, Julia Heinsoo and Filip K. Arnberg
- 130** *Exploring Functional Impairment in Light of Prolonged Grief Disorder: A Prospective, Population-Based Cohort Study*
Mette Kjaergaard Nielsen, Kaj Sparle Christensen, Mette Asbjørn Neergaard, Pernille Envold Bidstrup and Mai-Britt Guldin
- 143** *Feasibility of Present-Centered Therapy for Prolonged Grief Disorder: Results of a Pilot Study*
Anna Vogel, Hannah Comtesse, Agnes Nocon, Anette Kersting, Winfried Rief, Regina Steil and Rita Rosner



Editorial: Grief Disorders: Clinical, Cultural, and Epidemiological Aspects

Geert E. Smid^{1,2*}, Clare Killikelly³ and Birgit Wagner⁴

¹ ARQ National Psychotrauma Centre, Diemen, Netherlands, ² Department of Humanist Chaplaincy Studies, University of Humanistic Studies, Utrecht, Netherlands, ³ Department of Psychology, University of Zurich, Zurich, Switzerland, ⁴ Medical School Berlin, Berlin, Germany

Keywords: prolonged grief disorder, persistent complex bereavement disorder, risk, meaning attribution, epidemiology, cultural aspects

Editorial on the Research Topic

Grief Disorders: Clinical, Cultural, and Epidemiological Aspects

Grief disorders have recently been included in international diagnostic classifications, including the 5th edition of the Diagnostic and Statistical Manual of Mental Disorders [DSM-5; (1)] and the 11th edition of the International Classification of Diseases [ICD-11; (2)]. Specifically, DSM-5 now includes the category “other specified trauma- and stressor-related disorder,” with *persistent complex bereavement disorder* as one of the specific examples, and ICD-11 includes *prolonged grief disorder* (PGD). Importantly, the DSM-5 steering committee has announced an update to DSM-5 to include PGD as a separate disorder (3).

The inclusion of grief disorders marks international recognition of the clinical relevance of grief disorders and has prompted mental health professionals to diagnose and treat disordered grief. Although bereavement is known to be capable of precipitating common mental disorders such as depression and posttraumatic stress disorder, to many clinicians, the concept of a grief disorder is new and therefore unfamiliar. This Research Topic contributes toward filling the knowledge gap surrounding disordered grief by highlighting clinical, cultural, and epidemiological aspects of grief disorders.

While clinical aspects include diagnosis, prevention, and treatment of grief disorders, cultural aspects involve the provision of care in a context of multiculturalism and globalization. Epidemiological aspects encompass both clinical and cultural aspects and include risk for the development or maintenance of disordered grief as well as factors that contribute to resilience and recovery. Risk and resilience factors of grief disorders relate to meaning attribution following loss and can be divided into five categories (4). First, *loss-related factors* include non-natural or sudden causes of death and the person’s immediate grief response. Second, *cultural factors* include beliefs, rituals, and care provision. Third, *social factors* include social support, juridical, political, and economic factors, among others. Fourth, *individual factors* include gender and age, neurobiological and immunological correlates, personality and attachment, and history of trauma and loss. Fifth, *factors related to the relationship with the deceased* include a variety of factors, such as the nature and quality of the relationship, farewell rituals, and involvement in the death. **Figure 1** presents an overview of potential risk and resilience factors.

OPEN ACCESS

Edited and reviewed by:

Stefan Borgwardt,
University of Basel, Switzerland

*Correspondence:

Geert E. Smid
g.smid@centrum45.nl

Specialty section:

This article was submitted to
Mood and Anxiety Disorders,
a section of the journal
Frontiers in Psychiatry

Received: 16 March 2021

Accepted: 22 April 2021

Published: 20 May 2021

Citation:

Smid GE, Killikelly C and Wagner B
(2021) Editorial: Grief Disorders:
Clinical, Cultural, and Epidemiological
Aspects. *Front. Psychiatry* 12:681523.
doi: 10.3389/fpsy.2021.681523

The breadth of these factors highlights the profound implications the inclusion of grief disorders has for the organization and provision of mental health care, spiritual care, and end-of-life care in the prevention and treatment of grief disorders. Beyond care provision, the support from relatives, collective rituals, economic compensation and other circumstances that are shaped collectively may play a role in enhancing societal resilience and preventing disordered grief. The articles included in this collection address many of these aspects, providing essential insights while emphasizing the need for further scientific enquiry.

CLINICAL ASPECTS: DIAGNOSIS

O'Connor et al. investigated the availability of valid questionnaires, interviews, and cutoffs for ICD-11 PGD. The present systematic review reveals that no such valid tools are available for ICD-11 PGD or any of the earlier diagnostic constructs for PGD. They call for action in the field of bereavement research to start developing these tools now. Eisma et al. propose to conduct multiverse analysis on existing data, by applying different sets of criteria and diagnostic rules to determine how these affect the prevalence and other characteristics of ICD-11 PGD. After reviewing pioneering papers applying multiverse analyses to ICD-11 PGD, they recommend systematic application of multiverse analyses to help define current criteria for ICD-11 PGD, and future versions of this diagnosis.

Sveen et al. examined psychometric properties of the Swedish PG-13 as well as the latent structure of prolonged grief using the PG-13. Their findings support the use of the Swedish PG-13 to measure prolonged grief in bereaved adults. The latent structure of prolonged grief comprised two or three factors, and not a unidimensional structure as suggested previously.

Nielsen et al. used prospective data pre- and post-bereavement to explore functional impairment in PGD (assessed using the PG-13). They found that one in five bereaved relatives still reported overall functional impairment at 3 years after bereavement, although impairment in daily activities and social functioning improved from before to after bereavement. Also, functional impairment prior to bereavement was associated with the development of PGD. Finally, shedding light on the debate whether clinicians should consider mania as a possible bereavement reaction (Carmassi et al.), reviewed cases demonstrating a possible relationship between bereavement and a first manic episode.

CLINICAL ASPECTS: TREATMENT

Research on various grief-focused treatments informs clinical practice. Rubin et al. describe the two-track model of bereavement, comprising biopsychosocial functioning and relationship to the deceased. Their case illustration of

a woman who discovered her husband's dead body after his suicide illustrates how the assessment and intervention program was informed by the two tracks included in the model.

Soydas et al. use self-reported data on symptoms of posttraumatic stress, prolonged grief, depression, anxiety, and functional impairment from 929 homicidally bereaved, treatment receiving adults. They explore whether the observed effects of the intervention provided as part of the UK National Homicide Bereavement Service are associated with sociodemographic, homicide-related and clinical characteristics. Symptom reduction and improvements in functional impairment during treatment were negatively influenced by having a history of mental illness. A slow course of justice or the absence of a verdict negatively impacted treatment response on some of the observed outcomes.

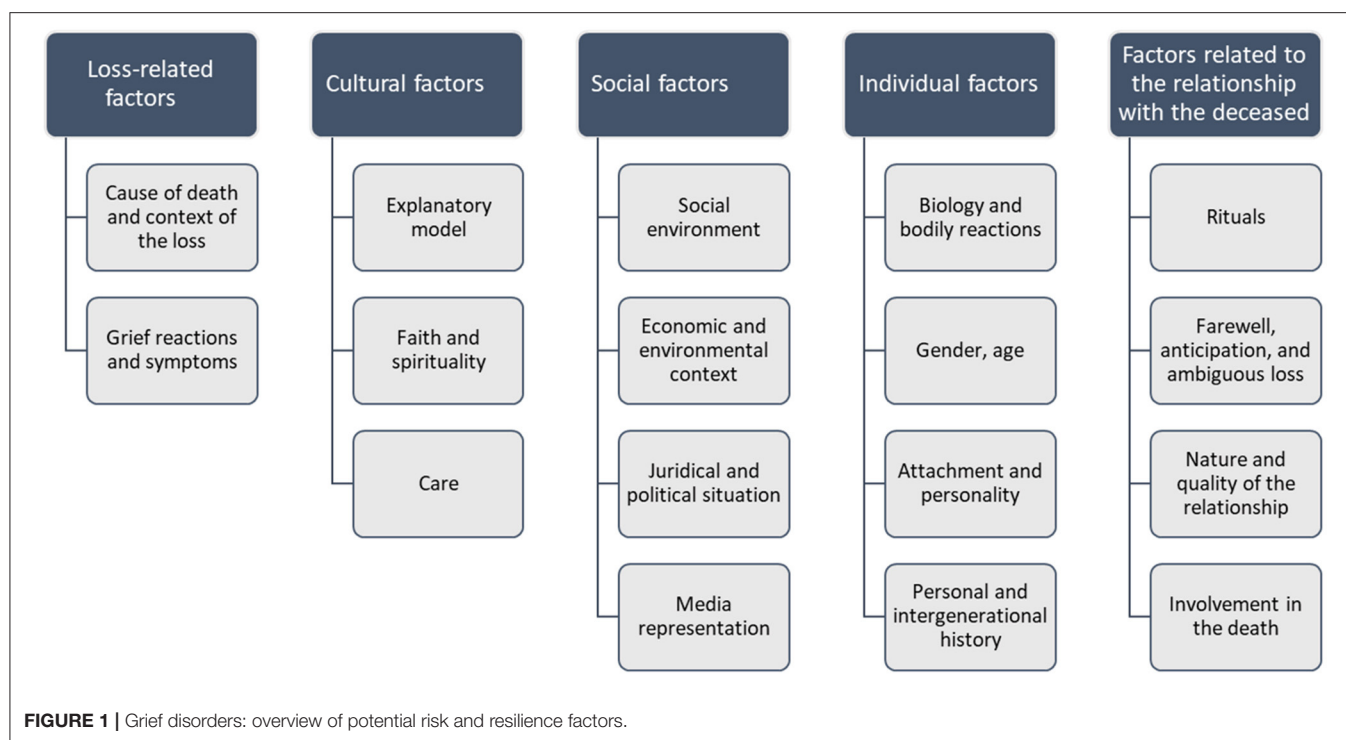
Vogel et al. evaluate the feasibility and the treatment effects of present-centered therapy (PCT) in a small sample of adults with PGD. Large decreases in PGD symptom severity at posttreatment and 3-month follow-up assessment as well as significant decreases in self-reported PGD symptoms, depression, and general psychological distress were observed. They conclude that PCT is a promising treatment for PGD and may serve as a strong comparator for future studies evaluating the efficacy of certain treatment elements in patients suffering from PGD.

Wagner et al. investigate the effectiveness of web-based bereavement interventions compared with control groups in reducing symptoms of grief in adults. The results of this meta-analysis suggest that internet-based treatments, based on CBT, can help to reduce the symptoms related to the loss of a significant person.

CULTURAL ASPECTS

Stelzer et al. summarize key findings from East-Asian research groups that challenge the applicability of North American and European definitions of PGD. This review finds that this area is currently facing a crisis in terms of the heterogenic use of different diagnostic criteria sets yielding widely different prevalence rates. It is important for researchers and clinicians to share knowledge and develop a consensus on required cultural caveats and culturally specific symptoms to improve assessment and treatment of disordered grief.

Djelantik et al. show that although refugees experience multiple post-migration stressors, they report significant symptom reductions during traumatic grief focused treatment. However, undocumented asylum seekers were more likely to drop out before completion of the treatment. Ongoing conflict in the country of origin and the total number of post-migration stressors were associated with smaller symptom reductions. They recommend that clinicians should recognize the effects of post-migration stressors on the course of symptoms and educate their refugee patients to support realistic treatment expectations.



Lacour et al. explore the relationship between the severity of PGD and cognitive and emotional factors, potentially traumatic events and living difficulties after arrival in the host country, in a clinical sample of refugees. Their findings suggest that self-efficacy and emotion regulation are determinants of PGD and constitute potential targets for treatment interventions.

Stammel et al. investigate associations between attitudes toward reconciliation and prolonged grief (PG) in bereaved survivors of the Khmer Rouge regime in Cambodia 30 years after the end of the regime. They show that a higher PG-symptom severity was related to less openness toward reconciliation. These findings are discussed in relation to cultural and religious views on death and mourning in Cambodia. The results of the study underline the importance of considering PG in regard to reconciliation efforts in post-conflict regions.

EPIDEMIOLOGICAL ASPECTS

Due to the Covid-19 pandemic, grief disorders will likely become a worldwide public health concern. Global death rates due to Covid-19 now exceed 1.5 million and the end of the pandemic is not yet in sight. Gesi et al. draw attention to people at risk of developing grief disorders due to the Covid-19 crisis, providing a useful framework for the management of bereaved people during the acute phase of the pandemic and for the implementation of tailored services in the future.

Fisher et al. investigated coping strategies in surviving relatives of U.S. military service members who died by combat, accident, or suicide. They determined that there are differential contributions of coping strategies to grief severity, depression and posttraumatic growth. Avoidant coping was a potent contributor to negative outcomes and inhibitor of posttraumatic growth. Although considering the possibility of death appeared to mitigate negative outcomes among relatives who survived combat death, avoidance of that possibility is likely protective for the majority of family members whose loved ones return home safely.

In a prospective cohort study, Kristensen et al. show that most bereaved family members following terror display a long-lasting and difficult grieving process, and that comorbid PTSD symptoms contribute to the slow recovery over the first 3 years after the incident. This has important implications for the psychosocial follow-up, which should highlight both the need for early intervention and long-term measures. Also, adequate skills among clinicians in screening for and treating both prolonged grief and posttraumatic stress is needed.

AUTHOR CONTRIBUTIONS

GS: wrote the first draft of the manuscript. All authors contributed to manuscript revision, read, and approved the submitted version.

REFERENCES

1. APA. *Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition* (DSM-5). Washington, DC: APA (2013).
2. World Health Organization. *International Classification of Diseases, 11th Revision* (ICD-11). Geneva: World Health Organization (2018).
3. APA. *Addition of a New Diagnosis, "Prolonged Grief Disorder," to the Depressive Disorders Chapter. View and Comment on Recently Proposed Changes to DSM-5.* (2020). Available online at: <https://www.psychiatry.org/psychiatrists/practice/dsm/proposed-changes> (accessed May 01, 2020).
4. Smid GE. A framework of meaning attribution following loss. *Eur J Psychotraumatol.* (2020) 11:1776563. doi: 10.1080/20008198.2020.1776563

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Copyright © 2021 Smid, Killikelly and Wagner. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.



Prolonged Grief Disorder and the Cultural Crisis

Eva-Maria Stelzer^{1,2}, Ningning Zhou^{1,3*}, Andreas Maercker¹, Mary-Frances O'Connor² and Clare Killikelly¹

¹ Division of Psychopathology and Clinical Intervention, University of Zurich, Zurich, Switzerland, ² Department of Psychology, University of Arizona, Tucson, AZ, United States, ³ Faculty of Psychology, Beijing Normal University, Beijing, China

OPEN ACCESS

Edited by:

Vivian Afi Abui Dzokoto,
Virginia Commonwealth University,
United States

Reviewed by:

Mary Alice Varga,
University of West Georgia,
United States
Amit Shira,
Bar-Ilan University, Israel

*Correspondence:

Ningning Zhou
zhouningning1990@mail.bnu.edu.cn

Specialty section:

This article was submitted to
Cultural Psychology,
a section of the journal
Frontiers in Psychology

Received: 03 September 2019

Accepted: 16 December 2019

Published: 10 January 2020

Citation:

Stelzer E-M, Zhou N, Maercker A,
O'Connor M-F and Killikelly C (2020)
Prolonged Grief Disorder
and the Cultural Crisis.
Front. Psychol. 10:2982.
doi: 10.3389/fpsyg.2019.02982

Prolonged grief disorder (PGD) is included as a new mental health disorder in the 11th edition of the International Classification of Diseases (ICD-11). Understandably, this has boosted research efforts to investigate this newcomer to psychopathology. However, the use of different diagnostic algorithms has resulted in substantially different prevalence rates both within and across cultural groups. Furthermore, global applicability of the new criteria outside of the Global North has not yet been established. This perspective presents key findings from Asian research groups and discusses the roadblocks to unified PGD research, including the heterogenic use of diagnostic algorithms and the lack of cultural compatibility of ICD-11 items. The authors discuss the key issues and address implications for practice.

Keywords: prolonged grief disorder, Asian, cross-culture, prevalence, bereavement

HIGHLIGHTS

- The new ICD-11 Prolonged Grief Disorder (PGD) criteria are conceptualized in terms of the World Health Organization's prioritization of international applicability.
- However, significant barriers exist to extrapolate prevalence rates within and across culture, and to establish global applicability.
- Barriers to unified PGD research include, among others, the use of different diagnostic algorithms and a lack of research regarding the cultural specificity of current ICD-11 items leading to an evolving cultural crisis of potential misestimation of prevalence rates.
- Emerging research from Asia confirms the worldwide heterogeneity in the use of PGD algorithms and the extent of the crisis.

INTRODUCTION

The field of grief and bereavement is at a turning point. For the first time, prolonged grief disorder (PGD) is included as a new mental health disorder in the World Health Organization's (WHO) 11th edition of the International Classification of Diseases (ICD-11) (World Health Organization [WHO], 2018). This presents the opportunity for igniting research into assessment

and treatment and providing clinicians with strong and unified groundwork for the validity of this disorder. However, currently there are some significant roadblocks to concerted PGD research. Firstly, the new formulation of PGD is preceded by several different disorder definitions that are currently in use. This has led to a wide variation in the use of diagnostic algorithms yielding substantially different prevalence rates, also across different cultural groups. This is unaided by the fact that the ICD-11 has not formally included a recommended diagnostic algorithm but rather applies a typological approach for diagnoses. Secondly, the clinical description of PGD for the ICD-11 is newly conceptualized following the WHO's prioritization of global applicability. PGD is characterized by core symptoms of longing for or preoccupation with deceased, significant symptoms of emotional distress, in addition to key cultural caveats, i.e., the duration of disorder must violate expected social and cultural norms (Killikelly and Maercker, 2017; World Health Organization [WHO], 2018). However, the global applicability of diagnostic criteria outside of the Global North has not yet been established. For example, the content of the specific items is largely derived from the Global North (Prigerson et al., 2009; Maciejewski et al., 2016). Several research groups across Asia are spearheading a new wave of research that is exploring and evaluating PGD. This perspective presents the latest research from Asia that uses the PGD criteria (as opposed to complicated grief or persistent complex bereavement disorder) and the new ICD-11 formulation of PGD. Here we present key findings from East-Asian research groups that expose the widespread difficulty with the heterogenic use of diagnostic algorithms and also challenge surface-level cultural compatibility of the specific ICD-11 items. We propose that the continued use of different diagnostic algorithms will undermine the field and lead to a crisis in terms of inaccurate prevalence rates of PGD, particularly in cross cultural research.

PREVALENCE CRISIS

Around the world, prevalence rates for PGD have been examined using several different sets of diagnostic algorithms. Most predominantly the PGDPlos criteria (Prigerson et al., 2009), ICD-11 PGD criteria (as closely as possible modeled after ICD-11; Maciejewski et al., 2016; Killikelly and Maercker, 2017), criteria from the Inventory of Complicated Grief (ICG) (Prigerson et al., 1995; Prigerson and Maciejewski, 2005; Shear et al., 2005), and variations of these criteria are used (Table 1). Not surprisingly, the use of different criteria sets has resulted in different prevalence rates of diagnosis. Using the PGDPlos criteria (Prigerson et al., 2009), prevalence rates for PGD in East Asian studies range from 1.8% for the general population (He et al., 2014) to 21.73% for those who lost their only child (Shi et al., 2019). In contrast, a recent study that applied the new ICD-11 PGD criteria following Killikelly and Maercker (2017) and Maciejewski et al. (2016) documents prevalence rates of 38.7 and 31.5% for Chinese Shidu parents (Zhou et al., under review). Finally, a range of other variations on these criteria

have revealed different prevalence rates. For instance, studies operationalizing PGD in terms of the CG formulation (Prigerson et al., 1995; Prigerson and Maciejewski, 2005; Shear et al., 2005) find prevalence rates ranging from 9.8 (Tsutsui et al., 2014) to 35% (Yu et al., 2016). And according to Yu et al. (2017) almost half of their sample (47.2) met criteria for PGD applying yet another diagnostic algorithm to dichotomize individuals in PGD vs. non-PGD clusters.

A similar variation can be found for research conducted in the Global North (Bonanno and Malgaroli, 2019; O'Connor et al., 2019). For instance, in a sample of US widow(er)s, Bonanno and Malgaroli (2019) reported prevalence rates of 4.8–12.2% when comparing different diagnostic algorithms for ICD-11 PGD criteria with varying numbers of accessory symptom items. The use of these different diagnostic algorithms makes it very difficult to extrapolate prevalence rates of PGD across Asian countries and to compare these rates with prevalence rates in the Global North, posing a significant risk of over- or underestimating the rate of disorder. For instance, a higher diagnostic threshold for assessing pathological grief has been suggested for Chinese samples (Li and Prigerson, 2016). However, this may only be the case when using the CG formulation applied by Li and Prigerson (2016). Furthermore, comparability and conclusions are limited as algorithms are derived from a number of scales, assessment procedures (self-report vs. clinical interviews), and item responses (score of at least three vs. four) to assess symptom endorsement.

BEYOND PREVALENCE

In addition, it is unclear if the prevalence rates indicate the most valid symptom expression. Although in both Chinese individuals and samples from the Global North, yearning or longing for the deceased, indicative of separation-related distress, constitute the hallmark symptom for disordered grief (Prigerson et al., 2009; He et al., 2014; Li and Prigerson, 2016; Xiu et al., 2016), there may be other culturally bound symptoms of grief that are currently not included in the ICD-11 PGD criteria (Killikelly et al., 2018). There are several examples of how mental health measurement derived from within the cultural group may inform the validity of the assessment. In a foundational series of studies, Hinton et al. (2013) explored symptoms of PGD in Cambodian refugees using a gold standard grief scale from the Global North [Prolonged Grief-13 (PG-13), Prigerson et al., 2009]. Interestingly they also developed a culturally sensitive measure of grief (CSM-G) and compared the rates of distress using these two scales. The CSM-G measure included reference to the somatic and bodily experiences of distress that are not included in the PG-13. According to the PG-13, 8% of participants experienced significant grief distress, whereas the locally derived scale revealed that over 31% of participants met criteria for severe distress. These contrasting results between etic (measures developed outside of the culture) and emic (measures developed from within the culture) scales have been consistently found across different

TABLE 1 | Common diagnostic algorithms for the assessment of PGD and their prevalence rates in East-Asian samples.

Diagnostic criteria	Algorithm	Study findings			
		Authors	Prevalence rates (%)	Sample	Sample size (N) ¹
PGDPlos (Prigerson et al., 2009)	Experience of yearning Cognitive, emotional, and behavioral symptoms: ≥ 5 Time since death: ≥ 6 months Significant impairment	He et al., 2014	1.8	Chinese sample; general population	445
		Tsai et al., 2018	5.2 (6 months post loss); 2.3 (13 months post loss)	Taiwanese caregivers	388 (6 months) 354 (13 months)
		Yi et al., 2018	8.47	Chinese earthquake survivors	1464
		Li and Prigerson, 2016	13.9	Chinese sample	1099
		Zhou et al., 2018	16.20	Chinese Shidu parents	536
		Comtesse and Rosner, 2019	20.2	Middle-Eastern asylum seekers	99
		Zhou et al., under review	20.9	Chinese Shidu parents	961
		Shi et al., 2019	21.73	Chinese Shidu parents	308
		Zhou et al., under review	38.7	Chinese Shidu parents	961
ICD-11 PGD (Killikelly and Maercker, 2017)	Experience of separation distress: yearning or preoccupation Experience of intense emotional pain symptoms: ≥ 1 Time since death: > 6 months Significant impairment	Zhou et al., under review	31.5	Chinese Shidu parents	961
ICD-11 PGD criteria with additional accessory symptoms (Maciejewski et al., 2016)	Experience of yearning or preoccupation Syndrome severity: ≥ 3 items Time since death: > 6 months Significant impairment	Zhou et al., under review	31.5	Chinese Shidu parents	961
CG formulation (e.g., (Prigerson et al., 1995; Prigerson and Maciejewski, 2005; Shear et al., 2005)	Cut-off score of > 25 (Prigerson et al., 1995) on Inventory of Complicated Grief defined as PGD-positive	Tsutsui et al., 2014	9.8	Japanese hospital workers after earthquake	82
	Prigerson and Maciejewski, 2005 Experience of yearning Cognitive, emotional, and behavioral symptoms: ≥ 4 Time since death: ≥ 6 months Significant impairment	Rajkumar et al., 2015	14.2 for tsunami survivors 25.9 for bereaved survivors	South Indian tsunami survivors	643 tsunami survivors; 351 bereaved survivors
	Cut-off score of ≥ 30 (Shear et al., 2005) on Inventory of Complicated Grief defined as PGD	Yu et al., 2016	35	Chinese widow(er)s	120
Other criteria	High PGD group: Rating of 3 or more on ≥ 3 PG-13 items with symptoms experienced at least once a week. Low PGD group: Rating of 2 or less on < 3 PG-13 items with symptoms experienced less than once a week	Yu et al., 2017	47.2 in high PGD group	Chinese sample	72

¹ If information was available, the number of participants used for prevalence estimation is presented (rather than entire sample size of a particular study).

cultural groups and for different mental health disorders (Patel et al., 1997; Fernando, 2012; Rasmussen et al., 2014; Kim et al., 2017). If the ICD-11 guidelines for PGD seek global applicability, guidance on specific adaptations for different cultural groups should be provided to improve the cross-cultural

validity (Bäarnhielm et al., 2015; Lewis-Fernández et al., 2017; Smid et al., 2018).

Additionally, features of the ICD-11 PGD criteria may be more or less common for certain cultural subgroups and thus should be weighted accordingly. We explored the

cross-cultural utility and applicability of the ICD-11 PGD criteria among Chinese and German-speaking health-care providers (Stelzer et al., in press). While health-care providers were generally aligned with the diagnostic criteria, they also identified symptoms currently missing in the ICD-11 such as the experience of somatic/physical symptoms after loss, and symptoms that should be removed from the diagnostic criteria for certain cultural groups (e.g., anger, sadness, or loss of self in China). Cultural variations also became apparent for the time-criterion. Here, Chinese health-care providers suggested to extend the criterion to match the culturally prescribed mourning period. Such differences between the Global North and Asian samples with regard to endorsement of specific accessory symptoms have also been noted in quantitative studies (Li and Prigerson, 2016; Xiu et al., 2016). In a cross-cultural comparison of bereaved parents, the Chinese sample reported a stronger sense that life is empty or meaningless whereas Swiss parents suffered from more severe grief-related preoccupation (Xiu et al., 2016). Similarly, trauma-related distress and grief hallucinations are particularly common in Chinese samples (Li and Prigerson, 2016) but less strongly endorsed in the Global North. And of course, sociocultural and logistical barriers such as a lack of culture-sensitive assessment tools and unfamiliarity with diagnostic systems further challenge the global applicability of ICD-11 PGD criteria (Stelzer et al., in press). Overall, these findings highlight the need to consider cultural factors and specific symptoms when assessing and generalizing PGD criteria across cultural groups.

DISCUSSION

Many cultural factors, working invisibly in the background, affect the way symptoms are expressed, perceived, assessed, interpreted, and documented. Religious beliefs and practices, for instance, are strongly related to culture and may account for some of the variations in grieving processes (Lobar et al., 2006; Beyers, 2017). Influenced by beliefs such as fatalism [e.g., “Life and death are decreed by fate” (Yang, 1980)], Chinese bereaved seldomly share their grief with others and when Chinese people comfort the bereaved they often do so by saying “Restrain your grief and accord with inevitable changes”. The subtle influences of cultural context are observable when someone outside the culture disrupts the invisible veil that hides them. This brings to light important cultural differences that have not been discussed in the scientific literature. As empirical research forms the basis for clinical decision-making, validation of grief reactions across cultures is essential prior to generalizing symptom clusters – especially when trying to provide adequate support and comfort to the bereaved. The initial period when guidelines are first applied by clinicians is critical – before years of practice have solidified them, making them more resistant to change. Since the *expression of grief varies by culture, despite the universal experience of grief*, it is vitally important that we detect these variations

early as we apply diagnostic criteria. In recent years, an increasing number of empirical studies (of Chinese origin in particular) have explored grief responses in samples beyond the Global North and added to a growing body of knowledge regarding prevalence rates of pathological grief. But despite these efforts from researchers and WHO stakeholders, we are far from establishing global applicability of PGD. The heterogeneity of diagnostic algorithms used by researchers and clinicians resulting in different prevalence rates within and across cultural groups constitutes one of the major challenges of cultural compatibility. If researchers keep using different diagnostic approaches to assess cases of PGD, conclusions and comparisons regarding prevalence rates and cross-cultural applicability are hampered.

We therefore recommend that expert researchers and clinicians in the field come together to decide on a consensus PGD algorithm that can be used across research groups and countries. The new wave of research emerging from Asian countries should stimulate collaboration to explore cross-cultural similarities and differences in the presentation of grief. With the novum of having disordered grief included in classification systems for mental health disorders, it is critical to no longer presume a universality of grief responses but instead move toward a culturally integrated grief framework. Further research can examine this by collecting open-ended written responses to questions or interviews about grief reactions in a culture and generating culture-specific grief items (Shoeb et al., 2007; Yin et al., 2017). Additionally, cultural clinical interviews could be employed to grasp a full understanding about grief severity. For example, recently, researchers in the Netherlands have developed the Grief and Bereavement cultural interview that aims to provide an in-depth assessment of the cultural context underlying prolonged grief distress (Smid et al., 2018). This 10-question interview could supplement a standard grief scale such as the PG-13 or the new International PGD scale (Killikelly et al., in preparation) to better guide the severity of the diagnosis and treatment planning.

Although there is no gold standard procedure for cross-cultural measurement of PGD and it is currently unknown how feasible such an assessment might be for grief, several researchers have confirmed that culturally informed mental health assessment is feasible, useful, and improves the validity of mental health diagnosis (Aggarwal et al., 2014; Kirmayer et al., 2014; Lewis-Fernández et al., 2017). Methodologically sound investigations and systematic cross-cultural comparisons can help identify in what ways grief phenomena are universal and culture specific. This knowledge, in turn, can influence existing bereavement models and clinical practice allowing researchers and clinicians to take individuals’ cultural identities into account when providing assistance and comfort to culturally diverse populations. It is important for researchers and clinicians to share knowledge and develop a consensus on required cultural caveats and culturally specific symptoms to improve assessment and treatment of disordered grief.

AUTHOR CONTRIBUTIONS

E-MS constructed the structure of the perspective and wrote most of it. NZ searched the articles conducted in

China and reviewed the results of the Chinese studies. CK proposed the idea and directed the whole process of the writing. AM and M-FO'C read and revised the manuscript in detail.

REFERENCES

- Aggarwal, N. K., Glass, A., Tirado, A., Boiler, M., Nicasio, A., Alegria, M., et al. (2014). The development of the DSM-5 cultural formulation interview-fidelity instrument (CFI-FI): a pilot study. *J. Health Care Poor Underserved* 25:1397. doi: 10.1353/hpu.2014.0132
- Bäärnhielm, S., Åberg Wistedt, A., and Rosso, M. S. (2015). Revising psychiatric diagnostic categorisation of immigrant patients after using the cultural formulation in DSM-IV. *Transcult. Psychiatry* 52, 287–310. doi: 10.1177/1363461514560657
- Beyers, J. (2017). Religion and culture: revisiting a close relative. *HTS Theo. Stud.* 73, 1–9. doi: 10.4102/hts.v73i1.3864
- Bonanno, G. A., and Malgaroli, M. (2019). Trajectories of grief: comparing symptoms from the DSM-5 and ICD-11 diagnoses. *Depress. Anxiety*. doi: 10.1002/da.22902
- Comtesse, H., and Rosner, R. (2019). Prolonged grief disorder among asylum seekers in Germany: the influence of losses and residence status. *Eur. J. Psychotraumatol.* 10:1591330. doi: 10.1080/20008198.2019.1591330
- Fernando, G. A. (2012). The roads less traveled: mapping some pathways on the global mental health research roadmap. *Transcult. Psychiatry* 49, 396–417. doi: 10.1177/1363461512447137
- He, L., Tang, S., Yu, W., Xu, W., Xie, Q., and Wang, J. (2014). The prevalence, comorbidity and risks of prolonged grief disorder among bereaved Chinese adults. *Psychiatry Res.* 219, 347–352. doi: 10.1016/j.psychres.2014.05.022
- Hinton, D. E., Nickerson, A., and Bryant, R. A. (2013). Prolonged grief in Cambodian refugees following genocide: rebirth concerns and avoidance of reminders. *J. Loss Trauma* 18, 444–460. doi: 10.1080/15325024.2012.714218
- Killikelly, C., Bauer, S., and Maercker, A. (2018). The assessment of grief in refugees and post-conflict survivors: a narrative review of etic and emic research. *Front. Psychol.* 22:1957. doi: 10.3389/fpsyg.2018.01957
- Killikelly, C., and Maercker, A. (2017). Prolonged grief disorder for ICD-11: the primacy of clinical utility and international applicability. *Eur. J. Psychotraumatol.* 8(Suppl. 6):1476441. doi: 10.1080/20008198.2018.1476441
- Kim, J., Tol, W. A., Shrestha, A., Kifle, H. M., Rayamajhi, R., Luitel, N. P., et al. (2017). Persistent complex bereavement disorder and culture: early and prolonged grief in Nepali widows. *Psychiatry* 80, 1–16. doi: 10.1080/00332747.2016.1213560
- Kirmayer, L. J., Jarvis, G. E., and Guzder, J. (2014). "The process of cultural consultation," in *Cultural Consultation: Encountering the Other in Mental Health Care*, eds L. Kirmayer, G. Jarvis, and J. Guzder, (Berlin: Springer), 47–69. doi: 10.1007/978-1-4614-7615-3-3
- Lewis-Fernández, R., Aggarwal, N. K., Lam, P. C., Galfalvy, H., Weiss, M. G., Kirmayer, L. J., et al. (2017). Feasibility, acceptability and clinical utility of the cultural formulation interview: mixed-methods results from the DSM-5 international field trial. *Br. J. Psychiatry* 210, 290–297. doi: 10.1192/bjp.bp.116.193862
- Li, J., and Prigerson, H. G. (2016). Assessment and associated features of prolonged grief disorder among Chinese bereaved individuals. *Compr. Psychiatry* 66, 9–16. doi: 10.1016/j.comppsy.2015.12.001
- Lobar, S. L., Youngblut, J. M., and Brooten, D. (2006). Cross-cultural beliefs, ceremonies, and rituals surrounding death of a loved one. *Pediatr. Nurs.* 32, 44–50.
- Maciejewski, P. K., Maercker, A., Boelen, P. A., and Prigerson, H. G. (2016). "Prolonged grief disorder" and "persistent complex bereavement disorder," but not "complicated grief," are one and the same diagnostic entity: an analysis of data from the Yale bereavement study. *World Psychiatry* 15, 266–275. doi: 10.1002/wps.20348
- O'Connor, M., Lasgaard, M., Larsen, L., Johannsen, M., Lundorff, M., Farver-Vestergaard, I., et al. (2019). Comparison of proposed diagnostic criteria for pathological grief using a sample of elderly bereaved spouses in Denmark: perspectives on future bereavement research. *J. Affect. Disord.* 251, 52–59. doi: 10.1016/j.jad.2019.01.056
- Patel, V., Simunyu, E., Gwanzura, F., Lewis, G., and Mann, A. (1997). The Shona Symptom Questionnaire: the development of an indigenous measure of common mental disorders in Harare. *Acta Psychiatr. Scand.* 95, 469–475. doi: 10.1111/j.1600-0447.1997.tb10134.x
- Prigerson, H. G., Horowitz, M. J., Jacobs, S. C., Parkes, C. M., Aslan, M., Goodkin, K., et al. (2009). Prolonged grief disorder: psychometric validation of criteria proposed for DSM-V and ICD-11. *PLoS Med.* 6:e1000121. doi: 10.1371/journal.pmed.1000121
- Prigerson, H. G., and Maciejewski, P. K. (2005). A call for sound empirical testing and evaluation of criteria for complicated grief proposed for DSM-V. *Omega* 52, 9–19. doi: 10.2190/ANKH-BB2H-D52N-X99Y
- Prigerson, H. G., Maciejewski, P. K., Reynolds, C. F. III, Bierhals, A. J., Newsom, J. T., Fasiczka, A., et al. (1995). Inventory of Complicated Grief: a scale to measure maladaptive symptoms of loss. *Psychiatry Res.* 59, 65–79. doi: 10.1016/0165-1781(95)02757-2
- Rajkumar, A. P., Mohan, T. S., and Tharyan, P. (2015). Lessons from the 2004 Asian tsunami: nature, prevalence and determinants of prolonged grief disorder among tsunami survivors in South Indian coastal villages. *Int. J. Soc. Psychiatry* 61, 645–652. doi: 10.1177/0020764015570713
- Rasmussen, A., Keatley, E., and Joscellyne, A. (2014). Posttraumatic stress in emergency settings outside North America and Europe: a review of the emic literature. *Soc. Sci. Med.* 109, 44–54. doi: 10.1016/j.socscimed.2014.03.015
- Shear, M. K., Frank, E., Houck, P. R., and Reynolds, C. F. (2005). Treatment of complicated grief: a randomized controlled trial. *JAMA* 293, 2601–2608. doi: 10.1001/jama.293.21.2601
- Shi, G., Wen, J., Xu, X., Zhou, N., Wang, J., Shi, Y., et al. (2019). Culture-related grief beliefs of Chinese Shidu parents: development and psychometric properties of a new scale. *Eur. J. Psychotraumatol.* 10:1626075. doi: 10.1080/20008198.2019.1626075
- Shoeb, M., Weinstein, H., and Mollica, R. (2007). The Harvard trauma questionnaire: adapting a cross-cultural instrument for measuring torture, trauma and posttraumatic stress disorder in Iraqi refugees. *Int. J. Soc. Psychiatry* 53, 447–463. doi: 10.1177/0020764007078362
- Smid, G. E., Groen, S., de la Rie, S. M., Kooper, S., and Boelen, P. A. (2018). Toward cultural assessment of grief and grief-related psychopathology. *Psychiatric Serv.* 69, 1050–1052. doi: 10.1176/appi.ps.201700422
- Stelzer, E.-M., Zhou, N., Maercker, A., Sun, H., Rohner, S., Merzhvynska, M., et al. (in press). Clinical utility of prolonged grief disorder in the ICD-11: the perspective of Chinese and German-speaking health care professionals (in preparation). *Psychopathology*.
- Tsai, W. I., Kuo, S. C., Wen, F. H., Prigerson, H. G., and Tang, S. T. (2018). Prolonged grief disorder and depression are distinct for caregivers across their first bereavement year. *Psychooncology* 27, 1027–1034. doi: 10.1002/pon.4629
- Tsutsui, T., Hasegawa, Y., Hiraga, M., Ishiki, M., and Asukai, N. (2014). Distinctiveness of prolonged grief disorder symptoms among survivors of the Great East Japan Earthquake and Tsunami. *Psychiatry Res.* 217, 67–71. doi: 10.1016/j.psychres.2014.03.001
- World Health Organization [WHO] (2018). *International Statistical Classification of Diseases and Related Health Problems*. Geneva: WHO.
- Xiu, D., Maercker, A., Woynar, S., Geirhofer, B., Yang, Y., and Jia, X. (2016). Features of prolonged grief symptoms in Chinese and Swiss bereaved parents. *J. Nerv. Ment. Dis.* 204, 693–701. doi: 10.1097/NMD.0000000000000539
- Yang, B. (1980). *Analects of Confucius*. Beijing: Zhonghua Book Company.
- Yi, X., Gao, J., Wu, C., Bai, D., Li, Y., Tang, N., et al. (2018). Prevalence and risk factors of prolonged grief disorder among bereaved survivors seven years after the Wenchuan earthquake in China: a cross-sectional study. *Int. J. Nurs. Sci.* 5, 157–161. doi: 10.1016/j.ijnss.2018.04.001
- Yin, F., Fang, J., Zhou, N., Shen, H., and He, Y. (2017). Development and validation of the Chinese death metaphors scale-revised. *Death Stud.* 41, 87–92. doi: 10.1080/07481187.2016.1214632

- Yu, M., Tang, S., Wang, C., Xiang, Z., Yu, W., Xu, W., et al. (2017). Avoidance of bereavement-related stimuli in Chinese individuals experiencing prolonged grief: evidence from a dot-probe task. *Front. Psychol.* 8:1201. doi: 10.3389/fpsyg.2017.01201
- Yu, N. X., Chan, C. L., Zhang, J., and Stewart, S. M. (2016). Resilience and vulnerability: prolonged grief in the bereaved spouses of marital partners who died of AIDS. *AIDS Care* 28, 441–444. doi: 10.1080/09540121.2015.1112354
- Zhou, N., Yu, W., Huang, H., Shi, G., Luo, H., Song, C., et al. (2018). Latent profiles of physical and psychological outcomes of bereaved parents in China who lost their only child. *Eur. J. Psychotraumatol.* 9:1544026. doi: 10.1080/20008198.2018.1544026

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Copyright © 2020 Stelzer, Zhou, Maercker, O'Connor and Killikelly. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.



Mania Following Bereavement: State of the Art and Clinical Evidence

Claudia Carmassi¹, Katherine M. Shear², Martina Corsi^{1*}, Carlo Antonio Bertelloni¹, Valerio Dell'Oste¹ and Liliana Dell'Osso¹

¹ Department of Clinical and Experimental Medicine, University of Pisa, Pisa, Italy, ² Columbia University School of Social Work, New York, NY, United States

OPEN ACCESS

Edited by:

Geert E. Smid,
Stichting Centrum '45, Arq
Psychotrauma Expert Group,
Netherlands

Reviewed by:

Aislinn Joanmarie Williams,
University of Iowa, United States
Paul Boelen,
Utrecht University, Netherlands

*Correspondence:

Martina Corsi
dott.martinacorsi@gmail.com

Specialty section:

This article was submitted to
Mood and Anxiety Disorders,
a section of the journal
Frontiers in Psychiatry

Received: 28 November 2019

Accepted: 14 April 2020

Published: 06 May 2020

Citation:

Carmassi C, Shear KM, Corsi M,
Bertelloni CA, Dell'Oste V and
Dell'Osso L (2020) Mania Following
Bereavement: State of the Art
and Clinical Evidence.
Front. Psychiatry 11:366.
doi: 10.3389/fpsy.2020.00366

Bereavement is the state of loss, determined in most of the cases by the death of a close person. It is probably the greatest sorrow that can occur in an individual life. Grief is a normal, healthy response to loss, evolving through stages in the process of mourning. In some cases, bereavement may lead to the outburst of manic episode: despite literature data being scarce, reports have explored this important clinical entity, variously called as “funeral mania” or “bereavement mania”. We systematically reviewed the literature exploring the possible relationships between bereavement and the onset of a manic episode, both first or recurrent pre-existing episode, besides describing a case report on a manic episode in the aftermath of a loss event, with an accurate evaluation of prior mild mood spectrum instability, supporting the role of loss-events as potential risk factor for bipolar illness progression. This article tries summarizing existing evidence on the debate whether clinicians should consider mania as a possible bereavement reaction.

Keywords: funeral mania, bereavement, complicated grief, bipolar disorder, manic episode, manic onset

BACKGROUND

Increasing interest has been recently devoted to psychopathological reactions to loss and evidences have supported the existence of a specific clinical diagnosis addressing about 7–20% of subjects who develop long-lasting symptoms of intense grief, that interfere with adaptation and re-engagement in life, differently named as Complicated Grief (CG), Traumatic Grief or Prolonged Grief Disorder (1, 2), characterized by persistent desire and pervasive yearning of the deceased person, deep pain and frequent crying or worrying (3–8). The DSM-5 introduced, for the first time, the diagnosis of “Persistent Complex Bereavement Disorder (PCBD)” in the section for conditions and criteria needing further research (9). The death of a loved one may also trigger the onset or the worsening of mental disorders, particularly Major Depression Disorder (MDD) and Post-Traumatic Stress Disorder (PTSD), however different reactions have been described in the literature in the framework of a loss event, such as mania (10, 11). This is particularly interesting, also considering the possible partial clinical and neurobiological overlap between grief reaction, mood disorder and PTSD (12–14).

The outburst of a manic episode after the death of a close one has been called “funeral mania” or “bereavement mania”. The temporal relationship between the loss and the onset of a mental disorder may vary from this later occurring immediately after the loss or later on as an anniversary reaction. “Funeral mania” refers to a typical manic episode occurring within one week from the death of a close relative or friend, while “bereavement mania” is considered to be a kind of

psychogenic mania that emerges in a short time following the death of a closed one (11, 15). Although only anecdotal cases have been reported on manic reactions after the death of close kinsmen data seem to reveal that a loss event can be either the trigger of a first manic episode in patients with a negative psychiatric history, or a predictor of manic relapse in patients with a history of bipolar disorder (BD) (16). Most of the literature, in fact, has been devoted to depression leading to important acknowledgments in most recent nosography related to the interplay between psychopathological reactions to relevant loss events and mood reactions. In this regard, one of the most important results was that the DSM-5 first adopted the “bereavement exclusion concept” in the mood depressive episode allowing to diagnose a major depressive episode independently of a specific time lapse in the aftermath of a relevant loss. Scant data, however, have focused attention on bipolar onset with a manic episode in the aftermath of a relevant loss, despite the observation of mania in response to traumatic events has been discussed in the literature (15); yet the specific mechanisms for this association remain unclear.

The purpose of the present manuscript is to summarize papers linking bereavement with bipolar onset with a manic episode or with a first manic episode in the context of a mood disorder, suggesting the need to deserve attention to this possible outcome. Secondly, we also reported a case description of a case followed in our clinic in which we examined a lifetime mood spectrum, with a mild mood instability and “mild manic states” as vulnerability, leading to consider reflecting on the role of loss-events as risk factor for illness progression.

METHODS

Search Strategy

Medline, PubMed and Scopus databases were accessed in order to research and collect English language papers published between January 1st, 1960 and November 1st, 2019. Free text terms and MeSH headings for the topics of bereavement and manic episode or bipolar disorder were combined as it follows: “(“funeral mania” OR “bereavement” OR “complicated grief”) AND (“manic episode” OR “bipolar disorder”)”.

Eligibility Criteria

We included in the present manuscript case reports published in English in indexed journals and when full manuscript was available. The following inclusion criteria were adopted: case reports or articles reporting manic episode onset in the aftermath of a significant loss in patients with no previous mood disorder or first manic episode in the context of a mood disorders after bereavement.

Study Selection

Each study was screened for eligibility by the Authors after reading the title and abstract. Any uncertainties concerning eligibility were discussed and resolved among all authors. The decisions for inclusion or exclusion are summarized in a flow

chart according to Preferred Reporting Items for Systematic reviews and Meta-Analyses (PRISMA) recommendations. Details are reported in **Figure 1**.

RESULTS

Process of Study Selection

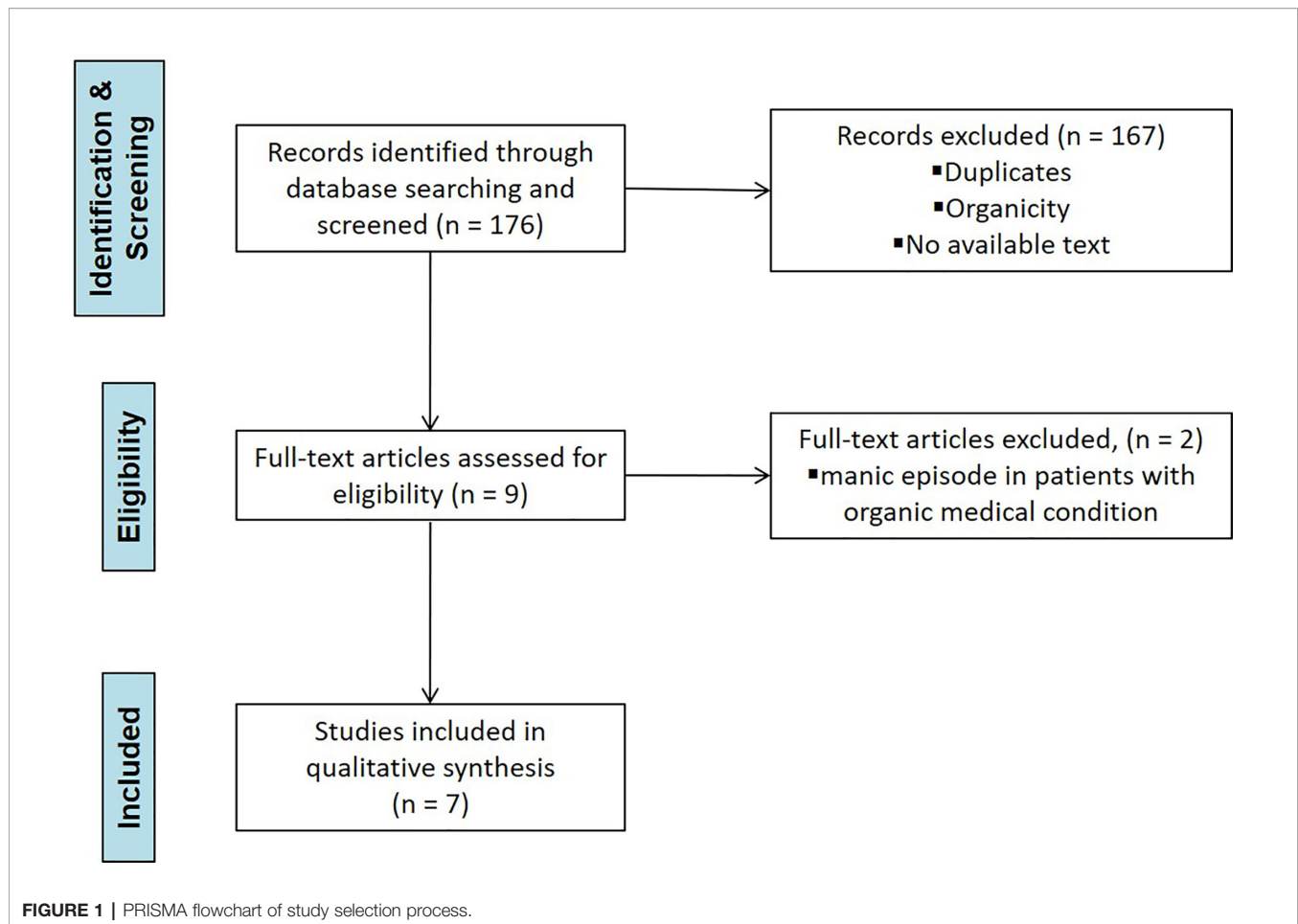
The electronic database search retrieved 176 potential results and Authors equally contributed in identifying potential information specific to this topic amongst the titles and abstracts of the publications. The first selection excluded 167 titles because: a) duplicates; b) not concerning the scope of the paper; c) not informative enough/no full manuscript available. Nine articles were identified as potentially relevant after screening of titles and abstracts. These studies were assessed for eligibility in full text. The second selection excluded two articles after being read and reviewed, as the information reported manic episode onset in the aftermath of bereavement in patients with an organic medical condition. Finally, seven papers were included in the present manuscript, reporting 15 case reports: three of these reports were excluded after being completely read and evaluated, as they describing manic relapse following bereavement in patients with a diagnosis of Bipolar Disorder (16–22). Key characteristics of included studies are summarized in **Table 1**.

Characteristics of Included Studies

Out of the 12 cases we found in literature, nine cases were women, while only three were men. The mean age was 38.25, with the youngest being a 21 years old female and the oldest a 55 years old male. Four out of the 12 case presented their onset during the second and fourth decade of their life and two during their third and fifth decade. The most represented relationships among the cases were son and husband/wife, reported in three and four cases respectively. Four patients developed the symptoms after the death of their parents, two following the mother's death and two after the deceased of the father. The remaining case concerned the death of another relative (husband's cousin). The most frequently reported cause of the death was by accident (five out of 12 cases), followed by sudden death and cancer (two cases each). In three case the cause of death was not reported. A negative psychiatric clinical history was found only in seven out of the 12 cases. The remaining five cases all reported a positive history of Major depressive disorders without previous manic episode or Bipolar history. Symptoms onset varied from a few hours/days to several years after the death. In one case, Rickarby (16) describes symptoms recurrence once every year, while someone of us published a case of a woman whose onset was on her son's birthday (19).

Case Presentation

We discuss the case of a 77-year-old patient with a diagnosis of BD hospitalized at the inpatient unit of the Psychiatric Clinic of the University of Pisa for a depressive episode, who experienced three years before a manic episode after his wife's death.



Mr. XY was a 77-year-old Italian male, widower, graduated at the high school at the age of 18 and living with the first of his two daughters. His longitudinal evaluation revealed no family loading for psychiatric illness, with no complications at birth, pregnancy and delivery. No developmental disorder or delays were referred.

The onset of the psychopathological symptomatology seems to date back to the age of 18 when, following a stressful life event (death of his father), he presented low mood levels and elevation of anxiety, resolved spontaneously in few months. In the following years the patient experienced cyclical seasonal depressive mood and energies swings, with concomitant fluctuations of the anxiety levels; however, in these years, the patient did not present interferences in the social and work functioning, with no need for psychiatric care. In January 2010 (70 years) after diagnosis of bladder carcinoma, successfully treated, the patient experienced a period characterized by slight lowering of mood level, clinophilia and increased level of anxiety. The patient decided to contact our Psychiatric Clinic, where a Major Depressive Episode was diagnosed and a psychopharmacological therapy based on Pregabalin (150 mg/day), Quetiapine (25 mg/day), Paroxetine (20 mg/die) and Delorazepam (30 mg/day) was prescribed. Afterwards, Mr XY

maintained a good psycho-affective compensation until the summer of 2014 when, subsequently an oncological disease, he lost his wife. In the following weeks he progressively showed an elevation of mood, high energy level, reduction in hours of sleep, greater search for pleasant activities (attending numerous partners, making several trips around the world), carrying out risky economic activities (dangerous real estate and commercial investments), managing by himself the psychopharmacological therapy. This period lasted about 18 months and was followed by an episode of referred low mood of about 2 months, for which he contacted again our Psychiatric Clinic; a new psychopharmacological therapy was settled, based on Pregabalin (75 mg/day) and Paroxetine (15 mg/day) with good clinical reward until 2017. In the following months Mr. XY presented depressive fluctuations of mood, energy and anxiety levels with the appearance of gastrointestinal somatizations at the epigastrium and alterations of body weight without sickness-free interval. Two hospitalizations were made in a short time frame in 2017 due to the symptomatology described above: the patient was treated by several psychopharmacological associations, reaching psychoaffective well-being with Valproic acid (300 mg/die), Aripiprazole (2.5 mg/die) and Venlafaxine (75 mg/die).

TABLE 1 | Case report studies reporting first manic episodes in the aftermath of a significant loss in patients a) without any previous lifetime mood episodes or b) with previous depressive episodes only.**A) PATIENTS WITHOUT ANY LIFETIME MOOD EPISODE**

Study	Patient (gender)	Age (years)	Relationship with the deceased	Description	Treatment
				Index episode	
Rickarby (16)	Male	55	Son (killed)	Acute Manic episode followed by recurrent manic episodes yearly in the anniversary	Hospitalization: Neuroleptics and lithium
Gill (17)	Male	25	Mother	Acute Mania with psychotic symptoms	Hospitalization: lithium
	Female	49	1. Husband	1. Acute mania	1. Hospitalization: neuroleptics and ECT
			2. Mother (4 years later)	2. Acute Mania with psychotic symptoms	2. Hospitalization: neuroleptics
	Male	46	1. Mother	1. Acute Mania	1. Hospitalization: no treatment data
			2. Father (2 years later)	2. Acute Mania	2. Hospitalization: no treatment data
	Female	24	Father (suicide)	Acute Mania	Hospitalization: no treatment data
Morgan et al. (18)	Female	37	Husband	Acute Mania with psychotic symptoms	Hospitalization: Neuroleptics
Carmassi et al. (19)	Female	52	Son	Acute Mania with psychotic symptoms	Hospitalization: Neuroleptics and mood stabilizers

B) PATIENTS WITH PREVIOUS DEPRESSIVE EPISODES ONLY

Study	Patient (gender)	Age (years)	Relationship with the deceased	Description	Treatment
				Previous history Index episode	
Rickarby (16)	Female	44	Husband	Severe depressive episodes (responsive to imipramine)	Acute Mania Hospitalization: lithium
	Female	21	Father	Severe depressive episodes with following recurrences and periods of mood instability (no mania)	Acute Mania Hospitalization: lithium
Hollender and Goldin (20)	Female	44	Son	Recurrent depressive episodes (the first occurring after the loss of her husband 3 years earlier)	Acute Mania with psychotic symptoms Hospitalization: TCA and mood stabilizers
Ranga et al. (21)	Female	34	Husband's cousin	Recurrent major depressive episodes	Acute Mania with psychotic symptoms Hospitalization: neuroleptics
Rosenman and Tayler (22)	Female	28	Husband	Post-partum depression	Acute Mania Hospitalization: no treatment data

ECT, electroconvulsive therapy; TCA, tricyclic antidepressants.

DISCUSSION

To the best of our knowledge, only four papers have reported about the onset of one's first lifetime manic episode in the aftermath of the loss of a significant other, as well as four papers described first manic episode following grief in patients with previous depressive episodes only. Several studies have reported a correlation between stressful life events and the onset or the recurrence of mood disorder. Severe negative life events, in fact, have been found to be associated with more than four times the risk of relapse and an increase up to three times in illness duration. These same events have also been found to predict the onset of mood disorders among children of people with a history of BD (23–25). In recent years, researchers have begun to consider the influence of psychosocial variables on depression and mania separately. Major losses appear to specifically predict the course of depression within BD while the effects of negative life events on mania are less clear. Nevertheless, some studies analysing severe negative life events

found a possible relationship between negative events and manic episodes, despite conducted on very small samples (11, 26–28). In one prospective study of undergraduates with bipolar spectrum disorders was found that life stressful events lead to a hypomanic symptomatology among those students with depressive cognitive styles (29).

The clinical features of mania following bereavement emerged by our review do not differ from the other types of mania. Authors describe changes in mood (euphoric), sometimes mixed with depressive feelings. Carmassi et al. (19) reported how the patient alternated elevated mood to affective instability and dysphoria. Authors often describe sudden mood switches from euphoria to hostility and aggressiveness (18). All cases reviewed are characterized by patients with an increase in energy levels, with lack of sleep, pressure to speech (often rapid and with flight of ideas), tendency to undertake many tasks and, in general, agitated and restless. Excessive spending and drinking were also reported. Despite scant literature data, a possible finding was hypersexuality with sexual disinhibition and flirting behaviors.

Several kinds of delusions were reported: persecutory (family conspiring against the patient) (18), of “grandeur” (being a millionaire) (16), having special powers to help people, thought broadcasting (18). These patients often developed delusions about the deceased: from asserting to be still in contact with him/her, to talking to him/her, to deny the reality of the death (19).

Our case presented a patient with a previous history of depressive but not (hypo)manic episodes, with a first manic episode in the aftermath of a loss event. Patient previous mood instability, characterized by exclusively depressive episodes, was carefully assessed during the hospitalization after manic episode by using a specific questionnaire investigating lifetime mood spectrum symptoms in a dimensional perspective, including atypical or not full-blown symptoms of mood, called MOODS-SR (Mood Spectrum-Self Report) (30, 31). Symptoms of (hypo) manic or mixed mood instability resulted absent or subthreshold and did not reached clinical relevance until the loss event happened. As previously reported in some other reports (16, 19), these patients appear primarily to be suffering from affective instability that does not meet criteria for a classic manic, and often seem to be display symptoms consistent with a mixed episode in the aftermath of a relevant loss. However, current DSM-5 nosography abolished the previous mixed episodes classification, replacing it with a mixed-features specifier that can be applied to episodes of major depression and (hypo)mania (9). Therefore, it could be useful reflect on the role of loss-events as risk factor for illness progression, detecting lifetime mood spectrum liability, also defined as mild mood instability and “mild manic states”, as a vulnerability to develop a full-blown manic episode.

Nevertheless, these reports do not provide an estimate of how common this phenomenon is. In a broader analysis of 1,565 Danish first psychiatric hospital admissions, authors (25) found that death of a mother or sibling was more common before an admission for mania than it was in the general population, but only for death by suicide. Given the heritability of suicide and bipolar disorder, though, people with bipolar disorder would be genetically more vulnerable to family suicide rather than people without bipolar disorder. Moreover, a USA study on 27,534 subjects found an association between unexpected death and the onset of manic episodes in a general population sample across the life course, suggesting that unexpected death of a loved one may be a substantial risk factor for the onset of a manic episode, especially among older adults, and even among those with no prior history of mood, anxiety, or alcohol disorders (32).

Although several studies found no direct effect of negative life events as a predictor of increases in manic symptoms (33, 34), it has been reported subjects may be vulnerable to increases in manic symptoms after negative life events (35–37). Independent severe negative life events have been reported to be more common in subjects who have consequently developed manic episodes rather than among healthy controls (38–40), though one study did not confirm it (41). Loss-related events, employment problems and financial difficulties have been

reported more frequently before manic episodes with respect to depressive ones (42–44).

The casual relationship between mania onset and bereavement has been discussed in the literature, but with few evidences. Psychodynamic models focused on the manic defence hypothesis (45). In these models, mania was hypothesized as a flight from painful feelings. These models predict that mania will occur after negative life events, as a defensive reaction. Newer cognitive behavioral formulations have proposed that people with bipolar disorder might avoid focusing on threatening information (46). A terror management study provided some evidence for defensive reactions among people who are vulnerable to hypomania (47). The loss of social support, particularly through bereavement, has been hypnotized to create a loss of control and can trigger mania or depression, and (hypo)manic symptoms might facilitate new social connections, whereas disinhibited and risky behavior exhibited during mania can cause the breakdown of vital relationships (48). Furthermore, grief reaction often includes sleep disruption as well as more general schedule disruption that have been hypothesized as potential mechanism through which life stressors lead to increases manic symptoms (49, 50). Sleep disruption, indeed, appears to trigger manic symptoms, as evidenced by naturalistic and experimental studies (51, 52).

Even if “Funeral mania” is a known clinical entity, it has no nosological status in DSM and ICD. The loss of a loved one is a universal experience that we are all potentially exposed to and the grief that ensues in the immediate is, however painful, a physiological reaction.

CONCLUSIONS

Bereavement could lead to several pathological reactions, whose most common, experienced by up to 7% of bereaved people, is CG (53, 54). Several authors agree on the possibility that, in a minority of cases, a pathological response to bereavement develops into CG, correlated to a significant and persistent impairment of the individual’s social-working functioning and a high suicidal risk (55, 56). As emerges from the analysis of the clinical cases mentioned above, bereavement may also be considered as a risk factor for a recurrence of a manic episode in people suffering of BD or, rarely, for the onset of a first manic episode in people without history of mental disorders. Thus, our findings should alert clinicians to the possible onset of mania in the aftermath of a significant loss either in healthy individuals or in patients affected by major depression shifting the main focus of observation from the early detection of a possible depressive onset to the eventual occurrence of a first lifetime manic episode, in order to provide adequate and prompt treatment.

DATA AVAILABILITY STATEMENT

The datasets generated for this study are available on request to the corresponding author.

ETHICS STATEMENT

Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article.

REFERENCES

- Kliem S, Lohmann A, Mößle T, Kröger C, Brähler E, Kersting A. The latent nature of prolonged grief- A taxometric analysis: results from a representative population sample. *Psychiatry Res* (2017) 260:400–5. doi: 10.1016/j.psychres.2017.11.087
- Aoyama M, Sakaguchi Y, Morita T, Ogawa A, Fujisawa D, Kizawa Y, et al. Factors associated with possible complicated grief and major depressive disorders. *Psychooncology* (2018) 27(3):915–21. doi: 10.1002/pon.4610
- Shear K, Shair H. Attachment, loss, and complicated grief. *Dev Psychobiol.* (2005) 47:253–67. doi: 10.1002/dev.20091
- Shear MK, Simon N, Wall M, Zisook S, Neimeyer R, Duan N, et al. Complicated grief and related bereavement issues for DSM-5. *Depression Anxiety* (2011) 28(2):103–17. doi: 10.1002/da.20780
- Shear MK. Grief and mourning gone awry: pathway and course of complicated grief. *Dialogues Clin Neurosci* (2012) 14(2):119–28.
- Shear MK. Getting straight about grief. *Depression Anxiety* (2012) 29(6):461–4. doi: 10.1002/da.21963
- Simon NM. Is complicated grief a post-loss stress disorder? *Depression Anxiety* (2012) 29(7):541–4. doi: 10.1002/da.21979
- Carmassi C, Shear MK, Massimetti G, Wall M, Mauro C, Gemignani S, et al. Validation of the Italian version Inventory of Complicated Grief (ICG): a study comparing CG patients versus bipolar disorder, PTSD and healthy controls. *Compr Psychiatry* (2014) 55(5):1322–9. doi: 10.1016/j.comppsy.2014.03.001
- American Psychiatric Association. *Diagnostic and Statistical Manual of Mental Disorders. 5th ed.* Arlington, VA: Author (2013).
- Kubacki A. Male and female mania. *Can J Psychiatry* (1986) 31(1):70–2. doi: 10.1177/070674378603100115
- Johnson SL, Cuellar AK, Ruggero C, Winett-Perlman C, Goodnick P, White R, et al. Life events as predictors of mania and depression in bipolar I disorder. *J Abnormal Psychol* (2008) 117(2):268–77. doi: 10.1037/0021-843X.117.2.268
- Dell'osso L, Carmassi C, Rucci P, Ciapparelli A, Paggini R, Ramacciotti CE, et al. Lifetime subthreshold mania is related to suicidality in posttraumatic stress disorder. *CNS Spectr* (2009) 14(5):262–6. doi: 10.1017/S1092852900025426
- Dell'Osso L, Da Pozzo E, Carmassi C, Trincavelli ML, Ciapparelli A, Martini C. Lifetime manic-hypomanic symptoms in post-traumatic stress disorder: relationship with the 18 kDa mitochondrial translocator protein density. *Psychiatry Res* (2010) 177(1–2):139–43. doi: 10.1016/j.psychres.2008.07.019
- Dell'Osso L, Carmassi C, Mucci F, Marazziti D. Depression, Serotonin and Tryptophan. *Curr Pharm Des* (2016) 22(8):949–54. doi: 10.2174/1381612822666151214104826
- Johnson SL. Life events in bipolar disorder: towards more specific models. *Clin Psychol Rev* (2005) 25(8):1008–27. doi: 10.1016/j.cpr.2005.06.004
- Rickarby GA. Four cases of mania associated with bereavement. *J Nervous Ment Dis* (1977) 165(4):255–62. doi: 10.1097/00005053-197710000-00005
- Gill PV. Mania following bereavement. *Br J Psychiatry* (1986) 149:123–4. doi: 10.1192/S000712500021252X
- Morgan JF, Beckett J, Zolese G. Psychogenic mania and bereavement. *Psychopathology* (2001) 34(5):265–7. doi: 10.1159/000049320
- Carmassi C, Shear MK, Socci C, Corsi M, Dell'osso L, First MB. Complicated grief and manic comorbidity in the aftermath of the loss of a son. *J Psychiatr Pract* (2013) 19(5):419–28. doi: 10.1097/01.pra.0000435042.13921.73
- Hollender MH, Goldin ML. Funeral mania. *J Nervous Ment Dis* (1978) 166(12):890–2. doi: 10.1097/00005053-197812000-00011
- Ranga K, Krishnan R, Swartz MS, Larson MJ, Santoliquido G. Funeral mania in recurrent bipolar affective disorders: reports of three cases. *J Clin Psychiatry* (1984) 45(7):310–1.
- Rosenman SJ, Tayler H. Mania following bereavement: a case report. *Br J Psychiatry* (1986) 148:468–70. doi: 10.1192/bjp.148.4.468
- Sclare P, Creed F. Life events and the onset of mania. *Psychiatry* (1990) 156:508–14. doi: 10.1192/bjp.156.4.508
- Johnson SL, Roberts JE. Life events and bipolar disorder. *psychol Bull* (1995) 117:434–49. doi: 10.1037/0033-2909.117.3.434
- Kessing LV, Agerbo E, Mortensen PB. Major stressful life events and other risk factors for first admission with mania. *Bipolar Disord* (2004) 6(2):122–9. doi: 10.1111/j.1399-5618.2004.00102.x
- Ambelas A. Life events and mania. A special relationship? *Br J Psychiatry* (1987) 150:235–40. doi: 10.1192/bjp.150.2.235
- Johnson SL, Miller I. Negative life events and recovery from episodes of bipolar disorder. *J Abnormal Psychol* (1997) 106:449–57. doi: 10.1037/0021-843X.106.3.449
- Hillegers MH, Burger H, Wals M, Reichart CG, Verhulst FC, Nolen WA, et al. Impact of stressful life events, familial loading and their interaction on the onset of mood disorder. *Br J Psychiatry* (2004) 185:97–101. doi: 10.1192/bjp.185.2.97
- Reilly-Harrington NA, Alloy LB, Fresco DM, Whitehouse WG. Cognitive styles and life events interact to predict bipolar and unipolar symptomatology. *J Abnormal Psychol* (1999) 108:567–78. doi: 10.1037/0021-843X.108.4.567
- Dell'Osso L, Armani A, Rucci P, Frank E, Fagioli A, Corretti G, et al. Measuring mood spectrum: comparison of interview (SCI-MOODS) and self-report (MOODS-SR) instruments. *Compr Psychiatry* (2002) 43(1):69–73. doi: 10.1053/comp.2002.29852
- Dell'osso L, Stratta P, Conversano C, Massimetti E, Akiskal KK, Akiskal HS, et al. Lifetime mania is related to post-traumatic stress symptoms in high school students exposed to the 2009 L'Aquila earthquake. *Compr Psychiatry* (2014) 55(2):357–62. doi: 10.1016/j.comppsy.2013.08.017
- Keyes KM, Pratt C, Galea S, McLaughlin KA, Koenen KC, Shear MK. The burden of loss: unexpected death of a loved one and psychiatric disorders across the life course in a national study. *Am J Psychiatry* (2014) 171(8):864–71. doi: 10.1176/appi.ajp.2014.13081132
- McPherson H, Herbison P, Romans S. Life events and relapse in established bipolar affective disorder. *Br J Psychiatry* (1993) 163:381–5. doi: 10.1192/bjp.163.3.381
- Johnson SL, Sandrow D, Meyer B, Winters R, Miller I, Keitner G, et al. Increases in manic symptoms following life events involving goal-attainment. *J Abnormal Psychol* (2000) 109:721–7. doi: 10.1037/0021-843X.109.4.721
- Alloy LB, Reilly-Harrington N, Fresco DM, Whitehouse WG, Zechmeister JS. Cognitive styles and life events in subsyndromal unipolar and bipolar disorders: Stability and prospective prediction of depressive and hypomanic mood swings. *J Cogn Psychother* (1999) 13:21–40. doi: 10.1891/0889-8391.13.1.21
- Reilly-Harrington NA, Alloy LB, Fresco DM, Whitehouse WG. Cognitive styles and life events interact to predict bipolar and unipolar symptomatology. *J Abnormal Psychol* (1999) 108:567–78. doi: 10.1037/0021-843X.108.4.567
- Johnson SL, Fingerhut R. Negative cognitions predict the course of bipolar depression, not mania. *J Cogn Psychother.: Int Quarterly* (2004) 18:149–62. doi: 10.1891/jcop.18.2.149.65960
- Kennedy S, Thompson R, Stancer HC, Roy A, Persad E. Life events precipitating mania. *Br J Psychiatry* (1983) 142:398–403. doi: 10.1192/bjp.142.4.398
- Hunt N, Bruce-Jones W, Silverstone T. Life events and relapse in bipolar affective disorder. *J Affect Disord* (1992) 25:13–20. doi: 10.1016/0165-0327(92)90088-N
- Bebbington P, Wilkins S, Jones PB, Foerster A, Murray R, Toone B, et al. Life events and psychosis: Initial results from the Camberwell Collaborative Psychosis Study. *Br J Psychiatry* (1993) 162:72–9. doi: 10.1192/bjp.162.1.72

AUTHOR CONTRIBUTIONS

All authors gave substantial contribution to the study and approved the final version of the manuscript and the manuscript submission to Frontiers in Psychiatry.

41. Chung RK, Langeluddecke P, Tennant C. Threatening life events in the onset of schizophrenia, schizophreniform psychosis and hypomania. *Br J Psychiatry* (1986) 148:680–5. doi: 10.1192/bjp.148.6.680
42. Silverstone T, Romans-Clarkson S. Bipolar affective disorder: causes and prevention of relapse. *Br J Psychiatry* (1989) 154:321–35. doi: 10.1192/bjp.154.3.321
43. Dell'Osso L, Carmassi C, Corsi M, Pergentini I, Socci C, Maremmi AG, et al. Adult separation anxiety in patients with complicated grief versus healthy control subjects: relationships with lifetime depressive and hypomanic symptoms. *Ann Gen Psychiatry* (2011) 10(1):29. doi: 10.1186/1744-859X-10-29
44. Dell'Osso L, Carmassi C, Musetti L, Socci C, Shear MK, Conversano C, et al. Lifetime mood symptoms and adult separation anxiety in patients with complicated grief and/or post-traumatic stress disorder: a preliminary report. *Psychiatry Res* (2012) 198(3):436–40. doi: 10.1016/j.psychres.2011.12.020
45. Adler A. *Problems of neurosis*. New York: NY Harper & Row (1964).
46. Lyon HM, Startup M, Bentall RP. Social cognition and the manic defense: Attributions, selective attention, and self-schema in bipolar affective disorder. *J Abnormal Psychol* (1999) 108:273–82. doi: 10.1037/0021-843X.108.2.273
47. Johnson SL, Ballister C, Joiner TE Jr. Hypomanic vulnerability, terror management, and materialism. *Pers Individ Dif* (2005) 38:287–96. doi: 10.1016/j.paid.2004.04.008
48. Owen R, Gooding P, Dempsey R, Jones S. The Reciprocal Relationship between Bipolar Disorder and Social Interaction: A Qualitative Investigation. *Clin Psychol Psychother* (2017) 24(4):911–8. doi: 10.1002/cpp.2055
49. Wehr TA, Sack DA, Rosenthal NE. Sleep reduction as a final common pathway in the genesis of mania. *J Psychiatry* (1987) 144:201–4. doi: 10.1176/aip.144.2.201
50. Ehlers CL, Frank E, Kupfer DJ. Social zeitgebers and biological rhythms. *Arch Gen Psychiatry* (1988) 45:948–52. doi: 10.1001/archpsyc.1988.01800340076012
51. Leibenluft E, Albert PS, Rosenthal NE, Wehr TA. Relationship between sleep and mood in patients with rapid-cycling bipolar disorder. *Psychiatry Res* (1996) 63:161–8. doi: 10.1016/0165-1781(96)02854-5
52. Colombo C, Barbini B, Campori E, Smeraldi E. Rate of switch from depression into mania after therapeutic sleep deprivation in bipolar depression. *Psychiatry Res* (1999) 86:267–70. doi: 10.1016/S0165-1781(99)00036-0
53. Kersting A, Braehler E, Glaesmer H, Wagner B. Prevalence of complicated grief in a representative population-based sample. *J Affect Disord* (2011) 131:339–43. doi: 10.1016/j.jad.2010.11.032
54. Dell'Osso B, Albert U, Atti AR, Carmassi C, Carrà G, Cosci F, et al. Bridging the gap between education and appropriate use of benzodiazepines in psychiatric clinical practice. *Neuropsychiatr Dis Treat* (2015) 11:1885–909. doi: 10.2147/NDT.S83130
55. Simon NM, Pollack MH, Fischmann D, Perlman CA, Muriel AC, Moore CW, et al. Complicated grief and its correlates in patients with bipolar disorder. *J Clin Psychiatry* (2005) 66(9):1105–10. doi: 10.4088/JCP.v66n0903
56. Dell'Osso L, Carmassi C, Rucci P, Ciapparelli A, Conversano C, Marazziti D. Complicated grief and suicidality: the impact of subthreshold mood symptoms. *CNS Spectr* (2011) 16(1):1–6. doi: 10.1017/S1092852912000090

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Copyright © 2020 Carmassi, Shear, Corsi, Bertelloni, Dell'Oste and Dell'Osso. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.



Complicated Grief: What to Expect After the Coronavirus Pandemic

Camilla Gesi^{1*}, Claudia Carmassi², Giancarlo Cerveri³, Barbara Carpita²,
Ivan Mirko Cremone² and Liliana Dell'Osso²

¹ Department of Mental Health and Addiction, ASST Fatebenefratelli-Sacco, Milan, Italy, ² Department of Clinical and Experimental Medicine, University of Pisa, Pisa, Italy, ³ Department of Mental Health and Addiction, ASST Lodi, Lodi, Italy

OPEN ACCESS

Edited by:

Birgit Wagner,
Medical School Berlin, Germany

Reviewed by:

Charles Felder Reynolds,
University of Pittsburgh, United States
Michael Grady Wheaton,
Columbia University, United States

*Correspondence:

Camilla Gesi
camillagesi@hotmail.com

Specialty section:

This article was submitted to
Psychopathology,
a section of the journal
Frontiers in Psychiatry

Received: 01 April 2020

Accepted: 13 May 2020

Published: 26 May 2020

Citation:

Gesi C, Carmassi C, Cerveri G,
Carpita B, Cremone IM and
Dell'Osso L (2020) Complicated Grief:
What to Expect After the
Coronavirus Pandemic.
Front. Psychiatry 11:489.
doi: 10.3389/fpsy.2020.00489

The COVID-19 pandemic is one of the worst public health crises in a century, with an expected amount of deaths of several million worldwide and an even bigger number of bereaved people left behind. Although the consequences of this crisis are still unknown, a significant number of bereaved people will arguably develop Complicated Grief (CG) in the aftermath of this emergency. If the current pandemic is unprecedented, the grief following the coronavirus outbreak is likely to share features with grief related to natural disasters and after Intensive Care Unit (ICU) treatment. The aim of this paper is to review the most prominent literature on CG after natural disasters, as well as after diseases requiring ICU treatment. This body of evidence may be useful for helping bereaved people during the acute phase of the COVID-19 pandemic and for drawing clinical attention to people at risk for CG.

Keywords: complicated grief, bereavement, pandemic, COVID-19, natural disaster, ICU treatment, coronavirus outbreak, SARS-CoV-2

INTRODUCTION

At the time of writing this paper, over 690,000 people around the world have been infected with COVID-19, which has already caused more than 33,000 deaths. Although pandemic projections must be treated with caution at this early stage, it has been estimated that the global death toll from COVID-19 could reach as high as 15 million even in the best-case scenario (1).

Bereavement is a common human experience and grief represents the normal reaction to the death of a loved one. The vast majority of individuals who lose someone usually adjust over a period of six to 12 months and finally develop a new sense of normalcy in their life. But for others, this process becomes troublesome and prolonged. When people get stuck indefinitely in grieving, preventing them from processing the death and moving on with life, a condition known as complicated grief (CG) may eventually arise (2). CG is a chronic, impairing form of grief, distinctive from depression and post-traumatic stress disorder and other conditions that may follow the death of a loved one (3–6).

To the best of our knowledge, no studies examined CG in the aftermath of an epidemic or a pandemic. Still, the coronavirus outbreak shares several features with natural disasters that have been previously investigated with regard to CG. Moreover, people dying from COVID-19 usually do so after being admitted to ICU and remaining isolated from their loved ones until the end. This obviously poses an even heavier burden on the loss, but also makes it useful to keep in mind what

may happen to bereaved people when the death occurs in such circumstances and what we must expect from their grieving in the aftermath of this global emergency.

Based on the above premises, the aim of this paper is to review the most prominent literature on CG after natural disasters, as well as after acute diseases requesting ICU treatment. This body of evidence may be useful for assisting bereaved people during the acute phase of the COVID-19 pandemic and for drawing clinical attention to people at risk for CG.

COMPLICATED GRIEF AFTER NATURAL DISASTERS

The loss of a loved one during a natural disaster is especially traumatic and distressing, given that the death usually happens suddenly and unexpectedly (7). Similar to epidemics, natural disasters affect bereaved people in several ways other than causing a loved one's death. They often occur as massive traumatic events, putting others' and one's own life in danger and bringing about multiple kinds of losses at the same time, including losses in human lives, properties, income, and overall wealth. Most of the recent literature on natural disasters comes from earthquakes, tsunamis, and hurricanes (8–10). Prevalence estimates of CG among bereaved survivors evaluated up to 18 months after these types of disasters range from 9% to 80%, with the loss of a child or a spouse both being associated with CG symptoms (8, 11–13). Other factors that seem to be significantly associated with CG include extensive damage to homes (11, 14), being of the female sex, having fewer years of education (8, 13, 14), and lacking social support and social competence (8, 13). Interestingly, non-bereavement losses and PTSD symptoms have both been indicated as risk factors for CG during massive disasters (8, 15). The longest longitudinal study that evaluated the evolution of grief up to six years after an earthquake found that 41% of the bereaved had been following a resilient trajectory, characterized by low levels of CG, 48% had shown a recovering trajectory, with initially high symptoms of CG and a gradual decrease thereafter, and 11% had ended in a chronic trajectory, featuring high and unremitting levels of CG symptoms for as long as six years after the event. Interestingly, compared to other long-term studies evaluating CG after non-traumatic events/non-traumatic losses, the percentage of resilient subjects was not higher, which could be an indicator of the addictive burden of exposure to a traumatic event and a traumatic loss in increasing the risk of CG (16).

COMPLICATE GRIEF AFTER DISEASES REQUIRING ICU TREATMENT

Relatives of patients admitted to the ICU are at risk of psychological morbidity during and after the hospital stay (17–19). The ICU environment may be quite a traumatic experience for both patients and relatives: people are rolled in, often unable to communicate, hooked up to machines, ventilated or intubated.

Visits are strictly controlled, and critical events, including death, may happen quickly, with no chance for relatives to be informed timely and to say goodbye. During the COVID-19 pandemic, most of the deaths have happened in the ICU after the development of an acute respiratory distress syndrome (20). In many cases, patients entered the ER, were taken into the ICU, and never came back. Meanwhile, in order to keep them isolated, no one could see them, nor could any loved one be there at the time of the death. Several news media outlets have been reporting that for those whose relatives got infected with COVID-19 and died in the ICU, the situation proved to be extremely traumatic.

Hospital and ICU mortality is especially associated with a high incidence of grief reactions, including post-traumatic stress disorder, anxiety, depression, and CG (19, 21–23). Prevalence estimates of CG in relatives of ICU decedents are quite variable, ranging from 5% to 52% as evaluated on average six months after the death (19, 22, 24, 25). In a study conducted among 50 family members of ICU inpatients, 46% of those whose relative died presented with both CG and PTSD after six months. CG was not associated with anxiety or depression at the time of the death, nor at one and six months after the event, or with sociodemographic variables, relationship to patient, or decision-making role preference (26). In a recent larger study involving 282 relatives of ICU decedents, the prevalence of CG at six months was as high as 52.1%, and CG symptoms were associated with the presence of depressive symptoms at three months. Interestingly, after 12 months, the proportion of assessable relatives with CG was unchanged, despite individual data revealing that approximately 20% of subjects developed CG between six and 12 months and a similar proportion recovered in the same period. Independent predictors of CG symptoms were either unchangeable variables, such as female sex, a relative living alone, and intensivist board certification before 2009, or potential targets for improvements, such as a patient's refusal of treatment, the death occurring while intubated, relatives present at the time of death, relatives unable to say goodbye to the patient, or poor communication between physicians and relatives (27). Another study comparing family members of patients who died in an ICU versus a non-ICU hospital setting found that uniquely distressing experiences in the ICU setting were primarily related to the patient's emotional status prior to death (anxiety, fear) and the families' perception that the patient received attention and support (19).

DISCUSSION

The COVID-19 pandemic represents one of the worst public health crises in a century, with an expected death toll of several millions of people worldwide. The prevalence of CG in the general population ranges from 2 to about 7% (10 to 20% among bereaved people, 28–31), but estimates have been shown to increase steeply in certain selected groups, such as older adults grieving the loss of a spouse, people grieving for unexpected/violent deaths, bereaved people exposed to natural or human-made disasters, or other traumatic events (7, 15, 32).

Given the lack of studies investigating grief after epidemics of infectious diseases such as COVID-19, which represents an unprecedented phenomenon, we chose to review extant literature on CG after natural disasters and after death in the ICU because we found a few similarities between both scenarios and the ongoing COVID-19 outbreak.

Together with the loss of loved ones, natural disasters imply multiple losses, the closure of schools and facilities, the stopping of productive activities, and the reduction in services and supplies, which is mirrored in the COVID-19 epidemic. Moreover, people are placed in a traumatic environment, the exposure to which is multiplied by means of news media and social networks (33). On the other hand, deaths from COVID-19 often happen in the hospital and typically in the ICU, which is known to represent a highly traumatic experience for decedents' closest relatives.

Our review underlines that female sex is a risk factor for CG, not only in the general bereaved population but also among bereaved people of ICU decedents or in the context of natural disasters. While grieving a child or a spouse has been indicated as a risk factor for CG after a natural disaster, no differences have been found with regard to the kinship to the deceased among people who lose a relative in the ICU. However, available data have hitherto shown that the vast majority of deaths due to COVID-19 is among older males (20), which may suggest the need to draw a special clinical attention to widows and, to a lesser extent, to other relatives of COVID-19 deceased in the aftermath of the emergency. Both lines of literature provide evidence that the more the death is associated with PTSD-related symptoms, the higher the risk of concurrent CG might be. If the traumatic exposure during the pandemic cannot be easily modified, the traumatic burden of the ICU should be kept as low as possible. One of the greater issues of death in the ICU is that family members may be unable to say goodbye to their loved one, which was shown to be an independent risk factor for CG (19, 27). This is crucially important among the COVID-19 bereaved, who cannot access the ICU during the whole stay and for whom the separation from the decedent may contribute to feelings of yearning, anger, and bitterness over the death, ultimately leading to CG (27). In addition, it is likely that the COVID-19 bereaved might blame themselves for not having tried harder to see their loved one while in hospital and for not taking care of their relative's comfort and dignity during the hospital stay, which has been shown to impact on families' experience of the dying process (19, 27). For the same reasons, they could feel guilty about not being present at the time of death, although witnessing the death has been shown to raise the risk of CG in one previous study on bereavement after ICU stay (27). A further important issue encountered by families of people dying in the ICU and increasing the risk of CG is the poor communication with physicians (34), that may be arguably worse in the course of the COVID-19 pandemic due to a matter of protection against contagion as well as due to hospitals' overloading and staffing shortages of some countries. Again, both lines of literature highlight that greater levels of social support are correlated with better grief outcomes (13, 27). Unfortunately, differently

from mass disasters occurring within a short period, the COVID-19 pandemic is a long-lasting condition, leading to durable changes in everyday life, and especially in social habits. This might make COVID-19 mourners particularly vulnerable, given that social distance has been claimed to be the most important action to prevent further spreading of the disease, a measure, however, that could increase the risk of unsuccessful grieving.

Lastly, it has to be noted that the ongoing COVID-19 pandemic bears the unprecedented circumstance that public funerals have become illegal in many countries due to national restrictions against gatherings and to cemeteries' overloading. Funeral and burial rituals are important for the affective adjustment of people grieving the loss of a loved one and mourners who drew comfort from planning and participating in the funeral were shown to achieve better outcomes in later grief (35). From this perspective, being prevented from holding a proper funeral for their loved ones might prevent COVID-19 mourners from gaining awareness of the reality of the death and from understanding and framing their loss, besides eliminating a significant important occasion of social support (35).

CONCLUSIVE REMARKS AND LINES OF INTERVENTION

The COVID-19 crisis has united mental health stressors that have been separately evaluated in other contexts, but which have never been seen combined in one, unprecedented disaster happening on a global scale. This makes it very difficult to infer what is going to happen to people during and in the aftermath of this global event, unless we use previous data as a provisional proxy of such a new and unknown phenomenon.

COVID-19 is expected to cause an enormous death toll worldwide. Given that each death leaves up to five people grieving behind (36), and that the prevalence of CG is estimated at approximately 10% to 20% of bereaved people (28), the number of cases of CG following COVID-19 deaths may virtually reach the number of overall COVID-19 deaths in the upcoming months. The chance of lowering the psychological impact of bereaved people during the current pandemic is small, since only a few risk factors for CG can be manipulated by intervention. One concrete option is to safeguard the connectedness between relatives and COVID-19 inpatients and to improve the communication between medical staff and relatives, ensuring the latter is updated about the clinical evolution, and eventually allowing them into the ICU as the condition gets worse. A second focus of intervention might be to apply a shared approach to end-of-life, involving family members as surrogate decision makers. Indeed, besides representing an important component of high-quality end-of-life care in the ICU (37), the involvement of the relatives in treatment-limitation decisions was shown to correlate with decreased CG symptoms among survivors (27). Third, as living alone after the death has been found to be associated with the presence of CG symptoms, it could be crucial to provide bereaved relatives with effective social support. All this may

obviously require an extra effort by the health system, that is already stressed and stretched beyond its readiness of facing the burden of the outbreak, at least in some regions (38). Still, mental health and social work departments and community organizations may share the burden of emergency and intensive care units and play a crucial role in providing emotional and practical support to the most vulnerable bereaved populations who display psychological symptoms. At the same time, mental health services could possibly be involved in surveying and alleviating critical stress levels among health workers of intensive care and emergency units, who might also be prone to developing a broad range of stress-related conditions, including CG following the death of colleagues and other staff members to coronavirus disease. Eventually, the implementation of specific services is warranted in the close future in order to convey tailored treatments to people developing CG after losing

someone because of the pandemic. Given the frequent comorbidity between CG and both PTSD and depression, as well as the high traumatic burden of COVID-19, clinical attention should also be devoted, and treatment options tailored, to address all three conditions in the aftermath of the COVID-19 global emergency.

AUTHOR CONTRIBUTIONS

CG, GC, and LD'O conceived the work. IC and BC made the literature search and revision. CG, CC, and LD'O drafted the paper. CC and GC revised the work. All authors provided approval of the version to be published.

REFERENCES

- McKibbin WJ, Fernando R. The Global Macroeconomic Impacts of COVID-19: Seven Scenarios. *CAMA* (2020) 19:1–43. doi: 10.2139/ssrn.3547729
- Shear MK. Grief and mourning gone awry: pathway and course of complicated grief. *Dialogues Clin Neurosci* (2012) 14:119–28.
- Dell'Osso L, Carmassi C, Corsi C, Pergentini I, Socci C, Maremmi AG, et al. Adult separation anxiety in patients with complicated grief versus healthy control subjects: relationships with lifetime depressive and hypomanic symptoms. *Ann Gen Psychiatry* (2011) 10:29. doi: 10.1186/1744-859X-10-29
- Dell'Osso L, Carmassi C, Musetti M, Socci C, Shear MK, Conversano C, et al. Lifetime mood symptoms and adult separation anxiety in patients with Complicated Grief and/or Post-Traumatic Stress Disorder: a preliminary report. *Psychiatry Res* (2012) 198:436–40. doi: 10.1016/j.psychres.2011.12.020
- Carmassi C, Shear MK, Massimetti G, Wall M, Mauro C, Gemignani S, et al. Validation of the Italian version inventory of Complicated Grief (ICG): a study comparing CG patients versus bipolar disorder, PTSD and healthy controls. *Compr Psychiatry* (2014) 55:1322–29. doi: 10.1016/j.comppsy.2014.03.001
- Carmassi C, Gesi C, Corsi M, Pergentini I, Cremonese IM, Conversano C, et al. Adult separation anxiety differentiates patients with Complicated Grief and/or Major Depression and is related to lifetime mood spectrum symptoms. *Compr Psychiatry* (2015) 58:45–9. doi: 10.1016/j.comppsy.2014.11.012
- Kristensen P, Weisæth L, Heir T. Bereavement and mental health after sudden and violent losses: a review. *Psychiatry* (2012) 75:76–97. doi: 10.1521/psyc.2012.75.1.76
- Shear MK, McLaughlin KA, Ghesquiere A, Gruber MJ, Sampson NA, Kessler RC. Complicated grief associated with hurricane Katrina. *Depress Anxiety* (2011) 28:648–57. doi: 10.1002/da.20865
- Dell'Osso L, Carmassi C, Massimetti G, Stratta P, Riccardi I, Capanna C, et al. age, gender and epicenter proximity effects on post-traumatic stress symptoms in L'Aquila 2009 Earthquake survivors. *J Affect Disord* (2013) 146:174–80. doi: 10.1016/j.jad.2012.08.048
- Li X, Aida J, Hikichi H, Kondo K, Kawachi I. Association of postdisaster depression and posttraumatic stress disorder with mortality among older disaster survivors of the 2011 great East Japan earthquake and tsunami. *JAMA Network Open* (2019) 12:e1917550. doi: 10.1001/jamanetworkopen.2019.17550
- Kristensen P, Lars W, Trond H. Predictors of Complicated Grief After a Natural Disaster: A population study two years after the 2004 South-East Asian tsunami. *Death Stud* (2010) 34:137–50. doi: 10.1080/07481180903492455
- Rajkumar AP, Mohan TS, Tharyan P. Lessons from the 2004 Asian tsunami: Nature, prevalence and determinants of prolonged grief disorder among tsunami survivors in South Indian coastal villages. *Int J Soc Psychiatry* (2015) 61:645–52. doi: 10.1177/0020764015570713
- Hu XL, Li XL, Dou XM, Li R. Factors related to Complicated Grief among bereaved individuals after the Wenchuan Earthquake in China. *Chin Med J (Engl)* (2015) 128:1438–43. doi: 10.4103/0366-6999.157647
- Ghaffari-Nejad A, Ahmadi-Mousavi M, Gandomkar M, Reihani-Kermani H. The prevalence of complicated grief among Bam earthquake survivors in Iran. *Arch Iran Med* (2007) 10:525–8.
- Dell'Osso L, Carmassi C, Massimetti G, Conversano C, Daneluzzo E, Riccardi E, et al. Impact of traumatic loss on post-traumatic spectrum symptoms in high school students after the L'Aquila 2009 Earthquake in Italy. *J Affect Disord* (2011) 134:59–64. doi: 10.1016/j.jad.2011.06.025
- Sveen J, Bergh Johannesson K, Cernvall M, Arnberg FK. Trajectories of prolonged grief one to six years after a natural disaster. *PloS One* (2018) 13:e0209757. doi: 10.1371/journal.pone.0209757
- Azulay E, Pochard F, Kentish-Barnes N, Chevret S, Aboab J, Adrie C, et al. Risk of post-traumatic stress symptoms in family members of intensive care unit patients. *Am J Respir Crit Care Med* (2005) 171:987–94. doi: 10.1164/rccm.200409-1295oc
- Davidson JE, Jones C, Bienvenu OJ. Family response to critical illness: postintensive care syndrome-family. *Crit Care Med* (2012) 40:618–24. doi: 10.1097/CCM.0b013e318236ebf9
- Probst DR, Gustin JL, Goodman LF, Lorenz A, Wells-Di Gregorio SM. ICU versus Non-ICU hospital death: family member complicated grief, posttraumatic stress, and depressive symptoms. *J Palliat Med* (2016) 19:387–93. doi: 10.1089/jpm.2015.0120
- Wu C, Chen X, Cai Y, Xia J, Zhou X, Xu S, et al. Risk factors associated with acute respiratory distress syndrome and death in patients with coronavirus disease 2019 pneumonia in Wuhan, China. *JAMA Intern Med* (2020) 13:e200994. doi: 10.1001/jamainternmed.2020.0994
- Lautrette A, Darmon M, Megarbane B, Joly LM, Chevret S, Adrie C, et al. A communication strategy and brochure for relatives of patients dying in the ICU. *New Engl J Med* (2007) 357:203–3. doi: 10.1056/nejmx070030
- Siegel MD, Hayes E, Vanderwerker LC, Loseth DB, Prigerson HG. Psychiatric illness in the next of kin of patients who die in the intensive care unit. *Crit Care Med* (2008) 36:1722–8. doi: 10.1097/ccm.0b013e318174da72
- McAdams JL, Puntillo K. Symptoms experienced by family members of patients in intensive care units. *Am J Crit Care* (2009) 18:200–10. doi: 10.4037/ajcc2009252
- Downar J, Barua R, Sinuff T. The Desirability of an Intensive Care Unit (ICU) clinician-led bereavement screening and support program for family members of ICU decedents (ICU Bereave). *J Crit Care* (2014) 29:311.e9–311.e16. doi: 10.1016/j.jcrc.2013.11.024
- Trevick SA, Lord AS. Post-traumatic Stress Disorder and Complicated Grief are common in caregivers of neuro-ICU patients. *Neurocrit Care* (2017) 26:436–43. doi: 10.1007/s12028-016-0372-5

26. Anderson WG, Arnold RM, Angus DC, Bryce CL. Posttraumatic stress and complicated grief in family members of patients in the intensive care unit. *J Gen Intern Med* (2008) 23(11):1871–6. doi: 10.1007/s11606-008-0770-2
27. Kentish-Barnes N, Chaize M, Seegers V, Legriel S, Cariou A, Jaber S, et al. Complicated grief after death of a relative in the intensive care unit. *Eur Respir J* (2015) 45:1341–52. doi: 10.1016/j.jad.2010.11.032
28. Middleton W, Burnett P, Raphael B, Martinek N. The bereavement response: a cluster analysis. *Br J Psychiatry* (1996) 169:167–71. doi: 10.1192/bjp.169.2.167
29. Forstmeier S, Maercker A. Comparison of two diagnostic systems for Complicated Grief. *J Affect Disord* (2007) 99:203–11. doi: 10.1016/j.jad.2006.09.013
30. Fujisawa D, Miyashita M, Nakajima S, Ito M, Kato M, Kim Y. Prevalence and determinants of Complicated Grief in general population. *J Affect Disord* (2010) 127:352–58. doi: 10.1016/j.jad.2010.06.008
31. Kersting A, Brähler E, Glaesmer H, Wagner B. Prevalence of complicated grief in a representative population-based sample. *J Affect Disord* (2011) 131:339–43. doi: 10.1016/j.jad.2010.11.032
32. Newson RS, Boelen PA, Hek K, Hofman A, Tiemeier H. The prevalence and characteristics of complicated grief in older adults. *J Affect Disord* (2011) 132:231–8. doi: 10.1016/j.jad.2011.02.021
33. Thompson RR, Holman EA, Silver RC. Media coverage, forecasted posttraumatic stress symptoms, and psychological responses before and after an approaching hurricane. *JAMA Netw Open* (2019) 2:e186228. doi: 10.1001/jamanetworkopen.2018.6228
34. Downar J, Sinuff T, Kalocsai C, Przybylak-Brouillard A, Smith O, Cook D, et al. A qualitative study of bereaved family members with complicated grief following a death in the Intensive Care Unit. *Can J Anaesth* (2020) 31:685–693. doi: 10.1007/s12630-020-01573-z
35. Gamino LA, Easterling LW, Stirman LS, Sewell KW. Grief Adjustment as Influenced by Funeral Participation and Occurrence of Adverse Funeral Events. *OMEGA - J Death Dying* (2000) 41:79–92. doi: 10.2190/QMV2-3NT5-BKD5-6AAV
36. Antonucci T, Hiroko A, Keiko T. Attachment and close relationships across the life span. *Attach Hum Dev* (2004) 6:353–70. doi: 10.1080/1461673042000303136
37. Curtis JR, White DB. Practical guidance for evidence-based ICU family conferences. *Chest* (2008) 134:835–43. doi: 10.1378/chest.08-0235
38. Armocida B, Beatrice F, Silvia U, Francesca P, Eduardo M. The Italian Health System and the COVID-19 challenge. *Lancet Public Health* (2020) 25:p11: S2468–2667(20)30074-8. doi: 10.1016/S2468-2667(20)30074-8

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Copyright © 2020 Gesi, Carmassi, Cerveri, Carpita, Cremone and Dell'Osso. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.



Post-Migration Stressors and Their Association With Symptom Reduction and Non-Completion During Treatment for Traumatic Grief in Refugees

A.A.A. Manik J. Djelantik^{1,2,3*}, Annemiek de Heus^{1,2}, Diede Kuiper³, Rolf J. Kleber^{2,3}, Paul A. Boelen^{1,2,3} and Geert E. Smid^{1,2,4}

OPEN ACCESS

Edited by:

Pedro Morgado,
University of Minho, Portugal

Reviewed by:

Tore Wentzel-Larsen,
Norwegian Centre for Violence and
Traumatic Stress Studies, Norway
Maria Gabriela Uribe Guajardo,
Sydney Local Health District, Australia
Heide Glaesmer,
University Hospital Leipzig, Germany

*Correspondence:

A.A.A. Manik J. Djelantik
A.A.A.M.J.Djelantik@uu.nl

Specialty section:

This article was submitted to
Mood and Anxiety Disorders,
a section of the journal
Frontiers in Psychiatry

Received: 18 November 2019

Accepted: 21 April 2020

Published: 27 May 2020

Citation:

Djelantik AAAMJ, de Heus A, Kuiper D,
Kleber RJ, Boelen PA and Smid GE
(2020) Post-Migration Stressors and
Their Association With Symptom
Reduction and Non-Completion
During Treatment for Traumatic
Grief in Refugees.
Front. Psychiatry 11:407.
doi: 10.3389/fpsy.2020.00407

¹ ARQ Centrum'45, Diemen, Netherlands, ² ARQ National Psychotrauma Centre, Diemen, Netherlands, ³ Department of Clinical Psychology, Faculty of Social Sciences, Utrecht University, Utrecht, Netherlands, ⁴ University of Humanistic Studies, Utrecht, Netherlands

Background: Resettled refugees exposed to trauma and loss are at risk to develop mental disorders such as posttraumatic stress disorder (PTSD) and persistent complex bereavement disorder (PCBD). Post-migration stressors have been linked to poor mental health and smaller treatment effects.

Aim: Our aim was to evaluate reductions in PTSD and PCBD symptoms and to explore the presence of post-migration stressors and their associations with symptom change and non-completion in a traumatic grief focused treatment in a cohort of refugees.

Methods: Paired sample t-tests were used to test the significance of the symptom reductions in PTSD and PCBD symptoms during treatment. The presence of post-migration stressors was derived from a qualitative analysis of the patient files. Associations between post-migration stressors and symptom reductions as well as non-completion were calculated.

Results: In this uncontrolled study, 81 files of consecutive patients were included. Significant reductions in both PCBD and PTSD symptomatology with medium effect sizes were found. Patients experienced a mean of three different post-migration stressors during the treatment. Undocumented asylum seekers were more likely to be non-completers. Ongoing conflict in the country of origin was associated with smaller PTSD symptom reductions and the total number of post-migration stressors was associated with smaller PCBD symptom reductions.

Conclusions: Treatment for resettled refugees for traumatic grief coincides with alleviations in both PCBD and PTSD symptomatology. Specific post-migration stressors were associated with reduced treatment effects and increased non-

completion. This is a first step towards well-informed improvements of mental health interventions for resettled refugees.

Keywords: traumatic loss, persistent complex bereavement disorder, posttraumatic stress disorder, day treatment program, brief eclectic psychotherapy, refugees, post-migration stressors

HIGHLIGHTS

- We evaluated reductions in PCBD and PTSD symptoms during a traumatic grief focused treatment in a treatment seeking clinical refugee sample.
- Although many post-migration stressors were present, significant symptom reductions with a medium effect sizes were observed.
- Undocumented asylum seekers were more likely to not complete the treatment.
- Ongoing conflict in the country of origin and the total number of post-migration stressors were associated with decreased symptom reductions.
- Clinicians treating mental health disorders in resettled refugees should consider the effects of post-migration stressors to prevent treatment drop-out and manage treatment expectations.

INTRODUCTION

Resettled refugees in Western countries commonly have been exposed to traumatic and loss events due to armed conflicts, persecution and/or natural disasters in their countries of origin (1, 2). It is not surprising that in this group mental disorders such as posttraumatic stress disorder (PTSD) are frequently observed with prevalence's varying between 35 to 47% (3–7). Recently, persistent complex bereavement disorder (PCBD), characterized by debilitating and prolonged grief, has been recognized as another form of psychopathology commonly seen in refugees seeking mental health support following loss and trauma with reported prevalence's varying between 6 to 10% in general refugee populations and 16% to 21% in bereaved refugee populations (7–11). PCBD has been included as a condition for further study in the 5th edition of the Diagnostic and Statistical Manual of Mental Disorders (12). An equivalent syndrome named Prolonged grief disorder (PGD) was recently included in the 11th edition of the International Classification of Diseases (ICD) (13, 14). An estimated 10% of bereaved adults confronted with natural loss will develop PCBD (15). However, unexpected and violent losses and the loss of close kin (partner or child) have been associated with substantially higher rates of psychopathology than anticipated and non-violent losses and losses other than close kin (16, 17). In a recent study, it was shown that the symptom network of PCBD is closely associated with the symptom networks of PTSD and depression (9), sometimes labeled together as “traumatic grief” (18, 19). Nevertheless, treatments of mental disorders following violent loss focusing both on PTSD and PCBD at the same time are scant and

have not been evaluated in refugee samples yet (18, 20). In a first study with a naturalistic design among 16 consecutive patients, a treatment program for traumatic grief for refugees was found feasible and coincided with significant declines in PTSD symptoms (18). While these results are promising, evaluation of the treatment in a larger cohort of patients, focusing on changes in both PTSD and PCBD symptoms, is needed. We will explain the treatment more thoroughly in the *Methods* section.

Refugees resettling in a new country often experience post-migration stressors and these stressors have been linked to poor mental health outcomes. These stressors include (a) socio-economic factors, i.e., financial and housing security and work problems; (b) social and interpersonal factors, i.e., family separation, family reunification, lack of social support, changes in social roles, discrimination, and changes in socioeconomic status, (c) process and immigration policies, i.e., detention, time of the asylum-seeking process, limited duration of residence permit, and as a consequence, living difficulties such as family conflict and unstable housing (21, 22).

The association between post-migration stressors and treatment outcome is an important research topic because these stressors may interfere with successful treatment conditions in several ways (23, 24). First, refugee patients may be too occupied by managing and arranging solutions for the post-migration stressors and this could lead to compromised treatment adherence. Second, in the treatment sessions there may be less time to conduct treatment, because the discussion of the current social stressors takes up too much time. Furthermore, sometimes the refugee may ask the clinician to provide assistance for their post-migration problems, which may result in ethical dilemmas for the clinician and slow down the treatment process. Lastly, when financial and housing problems are present, the costs for the transportation or treatment may be too high for the refugee, and he/she may withdraw from treatment. To the best of our knowledge, the association of these post-migration stressors with treatment outcome in resettled refugees has been only partially investigated in two studies (25, 26). Sonne et al. (25) investigated the post-migration stressors “employment status” and “integration” and found that not being employed was significantly, albeit weakly, correlated with poorer treatment outcome. Additionally, Schick et al. (26) found a correlation between a decrease of the amount of post-migration stressors “employment status”, “trauma exposure” and “visa status” over the time of treatment and better outcomes in anxiety and depression symptomatology. Importantly, an elaborate investigation of the presence of post-migration stressors and their association with drop-out rate or treatment outcome among refugees in a grief focused treatment has not yet been performed.

In sum, the purpose of this uncontrolled study was twofold. Firstly, our aim was to evaluate a traumatic grief focused treatment in a larger cohort of refugees than the previous feasibility study among 16 patients (18) and to include both the reductions in PTSD and PCBD symptoms. Because PTSD and PCBD are likely to co-occur in these patients (9) we were also interested in the correlation between PTSD and PCBD outcomes. We expected a medium size treatment effect for both PTSD and PCBD symptom reductions, based on the PTSD symptom change found in the feasibility study (18), and a correlated decline in PCBD and PTSD symptoms. Secondly, our aim was to explore the associations between postmigration stressors and treatment non-completion and examine associations between post-migration stressors and reductions in PCBD and PTSD symptomatology. We hypothesized that post-migration stressors would be associated with a higher drop-out rate and smaller symptom reductions during treatment.

METHODS

Patients and Procedure

This is a pre and post study design. The study used data from a convenience sample of all consecutive patients who participated in the day patient treatment for traumatic grief (DPT-TG) between October 2014 and October 2018 at ARQ National Psychotrauma Center, Netherlands, which receives nationwide referrals for specialized treatment of refugees and other traumatized groups. Patients who were referred for specialized treatment of psychotrauma were considered for inclusion in the DPT-TG if it was concluded from clinical evaluation that the death of one or more loved ones was a core traumatic event and the patient endorsed clinically relevant symptoms of grief and PTSD related to this event. In addition, patients needed to be willing to engage in group treatment. These clinical evaluations were carried out by psychiatrists and clinical psychologists as well as by psychiatric residents and clinical psychologists in training under direct supervision of a psychiatrist. Exclusion criteria for participation were acute or active suicidality (i.e., current suicide plans or suicidal behaviors or suicide attempts), severe psychotic disorder, and/or severe alcohol or substance abuse. Patients underwent standardized measurements at the start (T1) and at the end (T2) of the DPT-TG. Measures were administered by a team of independent psychologists and psychiatrists, all trained in diagnostics and receiving regular supervision from senior clinical psychologists. Measurements were part of the Routine Outcome Measurements (ROM). The medical ethics committee of Leiden University was consulted, and the study was exempted from formal review because the primary purpose of ROM is not research oriented. All patients were informed during the ROM that their answers could be anonymously used for research purposes and could object if they did not agree. None of the patients eligible for inclusion in the current study expressed objections. The data were handled confidentially, and the patient identities were only available for a small group of project managers.

Intervention

The DPT-TG is a multidisciplinary treatment program for bereaved refugees that includes exposure-based psychotherapy focused on both separation and traumatic distress. In addition, the program aims to stimulate the patients' social activity and reinforce their social networks, and offers support in relation to legal issues, work, and education. The treatment consists of a one-year weekly 5-h program divided in three phases of each 4 months. The first phase is a stabilization phase intended to increase patients' understanding of their symptoms and to become acquainted with the group and the treatment. The second phase focuses on processing the traumatic loss. During this phase, weekly sessions of individual brief eclectic psychotherapy for traumatic grief (BEP-TG) (19) are offered. BEP-TG consists of 16 sessions and was carried out by trained therapists (i.e. psychologists and psychiatrists). The third phase is focused on resocialization. Concrete individual future orientated goals are addressed in the group therapy and patients are encouraged to strengthen their social networks and assisted in applying for jobs or voluntary work (18). Patients underwent standardized measurements in the first four months of the DPT-TG, but before the start of the BEP-TG (T1), and in the last 4 months of DPT-TG, when the BEP-TG was finished (T2). DPT-TG have been found feasible in a prior study of 16 patients (18). More information about the schedule can be found in **Supplementary Materials A**. On average, patients attended a mean of 74% of the psychotherapy sessions (SD = 14.08, range 35%–100%).

Measurements and Data Collection

Traumatic Grief Inventory-Self Report (TGI-SR)

To assess the intensity of grief symptoms, the TGI-SR (27) was administered. The TGI-SR consists of two parts. The first part measures lifetime losses of loved ones and the second part assesses the intensity of the grief symptoms as experienced by the client in the past month. In case of multiple losses, the loss that is currently considered most painful is used as the anchor event for the second part. This consists of 18 items to assess the intensity of grief reactions rated on a 5-point Likert scale, ranging from "1 = never" to "5 = always", e.g., "I had intrusive thoughts and images associated with his/her death"; "I felt a strong longing or yearning for the deceased." A questionnaire diagnosis of probable PCBD according to DSM-5 was assigned to patients who scored 4 ("often") or 5 ("always") on at least one core symptom, at least six out of 12 additional symptoms, and the dysfunction criterion. The TGI sum score provides an index of severity of the grief symptoms, with a higher score associated with higher grief severity (range, 18 to 90). The TGI-SR has been validated in both a patient and a community sample (28). The internal consistency in the current study was good (Cronbach's alpha at T1 =.84 and at T2 =.92).

Clinician-Administered PTSD Scale 5 (CAPS-5)

PTSD symptom severity was assessed with the CAPS-5 (29). The CAPS-5 is a 30-item clinician rated interview, comprising 20 items assessing PTSD symptoms according to the DSM-5 (12). The intensity and frequency of symptoms during the past month are separately rated and then combined to form a single severity score for each item. Severity scores are rated on a 5-point scale ranging

from 0 (absent) to 4 (extreme/incapacitating), resulting in a total score range of 0 to 80. A diagnosis of PTSD according to DSM-5 was assigned to patients who scored ≥ 2 on at least one symptom of criterion B and C, at least two symptoms of criterion D and E, and criterion F (disturbance has lasted 1 month) and G (dysfunction criterion). An initial evaluation showed good psychometric properties for the CAPS-5 (29). The internal consistency in the current study was good (Cronbach's alpha at T1 = .86 and at T2 = .91). For one participant PTSD symptoms were assessed by the Clinician Administered PTSD Scale for DSM-IV [CAPS-4 (30)]. To be able to compare the scores of both CAPS versions, the sum scores were converted into probability scores, i.e., on a scale from 0 (lowest possible score) to 1 (highest possible score).

Socio-Demographic Information and Post-Migration Stressors

Socio-demographic information and the presence of post-migration stressors in the current patient sample were examined using information retrieved from patient files. Each file, including the admission report and notes from the therapists, was examined for the presence of post-migration stressors by AH and DK. The first three authors created a list of post-migration stressors using information from the review by Li, Liddell (21). Subsequently, additions and removals of this list of post-migration stressors were made by exploring similarities and differences across the files by AH and DK. When in doubt, the findings were discussed with the first author AD until consensus was reached. For the stressor "ongoing conflict in the country of origin", we searched the internet to find evidence of conflict at the time of treatment (**Supplementary Materials B**).

Patient Characteristics

Of the 81 patients who started, 57 patients completed the full year (completers) and 24 patients did not complete DPT-TG (non-completers). Fourteen patients dropped out completely from DPT-TG before the BEP-TG module was finished. Ten patients were not able to attend the weekly one-day treatment sessions, but continued treatment at the outpatient clinic with a customized program due to pregnancy and/or psychosocial problems. Eight patients did not complete at least one of the questionnaires due to various reasons such as psychosocial problems or illnesses in the week of the administration of the questionnaires but completed the rest of the DPT-TG. We included a flow chart in **Supplementary Materials C** with pre- and post-questionnaire numbers and reasons for drop-out. On average, patients were 42 years old ($SD = 9.24$). Most patients were male (84%) and originated from the Middle East (58%). Patients who had lost a child or partner were slightly more likely to drop-out or receive a customized treatment (**Table 1**).

Statistical Analysis

Missing Data

Missing data at item level were considered missing at random. Two items on the TGI-SR were missing for one participant at T2. For this participant, we calculated the total TGI-SR score using the mean of the valid items multiplied by the total number of items.

The missing data regarding social stressors and sum scores of PCBD and PTSD were handled using listwise deletion. The significance threshold was set at $p < .05$ for all analyses.

Power Calculation

First, we calculated the effect size from the information of the first 16 patients of the study of de Heus, Hengst (18) together with 13 consecutive patients (in total 29 patients) with the program Gpower (31). Based on this sample, we expected to find a difference between the pre and post measurements in the current study with a small to medium effect size (PTSD: $d = 0.5$, PCBD: $d = 0.4$, power = .80; $\alpha = .05$). For PTSD, a sample size of 34 completers was needed to calculate the effect size with t-test and a sample size of 63 completers was needed to calculate the effect size with analysis of variance (ANOVA). For PCBD, a sample size of 52 completers was needed to calculate an effect size with t-test and a sample size of 69 completers was needed to calculate the effect size with ANOVA. Because, we only had 57 patients who completed the DPT-TG, we used the t-test for the effect size calculations in the following sections.

Aim A: PCBD and PTSD Symptom Reductions During Treatment

We used the data of all patients who completed DPT-TG as well as the TGI-SR or CAPS on both timepoints. We performed a paired samples t-test to test the statistical significance of symptom reduction (32). Diagnostic status with regard to traumatic grief was operationalized as a categorical outcome variable that could take on the following categories: PCBD and PTSD, PTSD only, PCBD only, or no PTSD or PCBD. We used the McNemar test to test the statistical significance of the change within each of the diagnostic categories (33). To assess whether a change in diagnostic category membership had occurred within the sample as a whole across the two time points, we applied the marginal homogeneity test (33). Additionally, we calculated the Cohen's d to assess the effect size. We performed this with the TGI-SR sum score and the two subscales symptom clusters of PCBD, namely separation distress and reactive distress and social/identity disruption. We repeated this with the CAPS sum score and PTSD subscales, namely intrusive symptoms, avoidance of stimuli associated with the event, negative changes in cognitions and mood, and marked alterations in arousal and reactivity. We interpreted the effect sizes as small ($d = 0.2$), medium ($d = 0.5$), and large ($d = 0.8$) (34). Lastly, we calculated the correlation between PCBD and PTSD symptom reductions. We used the expectation-maximization (EM) algorithm as an analysis of the sensitivity of the correlation estimate to the incompleteness of our data as described in Dempster, Laird (35). This algorithm accounts for the missing data using maximum-likelihood estimates (36). For all analyses, we used SPSS version (37), except for the McNemar test which we performed in R with package "exact2x2".

Aim B: Post-Migration Stressors and the Association With Treatment Drop-Out and Symptom Reductions During Treatment

We determined whether or not the group of DPT-TG completers (both the patients with complete and missing questionnaires; $n = 57$) and DPT-TG non-completers ($n = 24$) differed

TABLE 1 | Pretreatment socio-demographic and loss-related characteristics and symptom levels of the completers and early drop-outs or customized treatment in a refugee sample.

	Completers of DPT-TG (<i>n</i> = 57)	Early drop-outs or customized treatment (<i>n</i> = 24)	Significance test for differences between groups	<i>p</i> Value
Age, M (SD)	42 (9.43)	42 (9.57)	$t(79) = 0.13$.84
Gender, <i>n</i> (%)			$\chi^2(1, n = 80) = 0.03$.86
Female	9 (16)	4 (17)		Fisher's exact test: 1.00
Male	48 (84)	19 (83)		
Marital status, <i>n</i> (%)			$\chi^2(3, n = 81) = 1.89$.56
Single	14 (25)	7 (29)		Fisher's exact test: .62
Married	32 (56)	12 (50)		
Divorced	8 (14)	2 (8)		
Widowed	3 (5)	3 (13)		
Level of education, <i>n</i> (%)			$\chi^2(2, n = 75) = 1.88$.39
Low	10 (18)	6 (25)		Fisher's exact test: .37
Middle	35 (61)	10 (42)		
High	9 (16)	5 (21)		
Region of origin, <i>n</i> (%)			$\chi^2(4, n = 81) = 2.96$.57
Dutch/Colony	2 (5)	3 (13)		Fisher's exact test: .50
Middle East	34 (60)	12 (50)		
Africa	11 (19)	4 (16)		
Bosnia Herzegovina/Serbia	9 (16)	4 (17)		
Asia	1 (2)	1 (4)		
Missing Family, <i>n</i> (%)	10 (17.5)	4 (17)	$\chi^2(1, n = 81) = 0.01$.92
Number of losses, M (SD)	5.59 (2.14)	5.65 (2.46)	$t(75) = 0.11$	Fisher's exact test: 1.00
Relationship to lost loved one ¹ , <i>n</i> (%)			$\chi^2(5, n = 60) = 10.81$.92
Partner	1 (1.8)	3 (12.5)		Fisher's exact test: 1.00
Child	4 (7.0)	2 (8.3)		.055
Parent(s)	15 (26.3)	5 (20.8)		Fisher's exact test: .038
Sibling	8 (14.0)	3 (12.5)		
Friend	15 (26.3)	1 (4.2)		
Other	1 (1.8)	2 (8.3)		
Violent loss a, <i>n</i> (%)	47 (83)	20 (83.3)	$\chi^2(1, n = 78) = 0.03$.86
PCBD according to TGI-SR, <i>n</i> (%)	30 (53)	13 (54)	$\chi^2(1, n = 81) = 0.02$	Fisher's exact test: 1.00
PTSD according to CAPS, <i>n</i> (%)	51 (90)	21 (88)	$\chi^2(1, n = 78) = 0.74$.90
Number of traumatic events, M (SD)	29 (5.24)	19 (4.48)	$t(64) = 0.75$	Fisher's exact test: 1.00
Psychiatric medication, <i>n</i> (%)	41 (72)	15 (63)	$\chi^2(1, n = 72) = 0.12$.39
Psychiatric comorbidity at during clinical intake assessment, <i>n</i> (%)				Fisher's exact test: .67
Depressive disorder	45 (79)	20 (83)	$\chi^2(1, n = 81) = 0.21$.87
Substance abuse	8 (14)	6 (25)	$\chi^2(1, n = 81) = 1.42$	Fisher's exact test: .76
Dissociative disorder	2 (4)	0 (0)	$\chi^2(1, n = 81) = 0.86$.73
				Fisher's exact test: 1.00

(Continued)

TABLE 1 | Continued

	Completers of DPT-TG (<i>n</i> = 57)	Early drop-outs or customized treatment (<i>n</i> = 24)	Significance test for differences between groups	<i>p</i> Value
Disruptive, Impulse-Control, and Conduct Disorders	2 (4)	0 (0)	$\chi^2(1, n = 81) = 0.86$.35 Fisher's exact test: 1.00
Somatic Symptom and Related Disorders	3 (5)	0 (0)	$\chi^2(1, n = 81) = 1.31$.25 Fisher's exact test: .56
Schizophrenia and other psychotic disorders	1 (2)	1 (4)	$\chi^2(1, n = 81) = 0.41$.52 Fisher's exact test: .51
Sexual Disorders	1 (2)	0 (0)	$\chi^2(1, n = 81) = 0.43$.51 Fisher's exact test: 1.00
Obsessive Compulsive Disorders	2 (4)	0 (0)	$\chi^2(1, n = 81) = 0.86$.35 Fisher's exact test: 1.00
Eating Disorder	1 (2)	0 (0)	$\chi^2(1, n = 81) = 0.43$.51 Fisher's exact test: 1.00
Indications for probable personality disorder	32 (56)	13 (54)	$\chi^2(1, n = 81) = 0.03$.87 Fisher's exact test: 1.00

[†]Concerning the loss that is considered most painful; PCBD, persistent complex bereavement disorder; PTSD, Posttraumatic Stress Disorder; DPT-TG, Day Patient Treatment for Traumatic Grief.

significantly in terms of post-migration stressors, using t-tests, chi square and Fisher exact tests. We used both chi square and Fisher exact tests because, due to our sample size, some of the expected frequencies of the post-migration stressors were smaller than 5 (38). In addition, we evaluated the associations between post-migration stressors and symptom reductions. To account for both the initial differences between scores at T1 and for measurement error inherent in the use of repeated measures on the same instrument, we first calculated the residual gain score for PCBD and PTSD (39). We regressed the sum scores at T2 on the sum scores at T1 and saved the residuals to use these as residual gain scores. Then, we examined whether these residual gain scores varied as function of the post-migration stressors using one-way ANOVA. For all analyses, we used SPSS version (37), except for the Fisher exact test which we performed in R with package “exact2x2”.

RESULTS

Aim A: PTSD and PCBD Symptom Reduction During Treatment

Table 2 shows an overview of the reductions in PCBD and PTSD symptomatology during DPT-TG. As shown in **Table 2**, patients scored significantly lower on the post-treatment grief measurement compared to the pre-treatment grief measurement [pre intervention: *M* = 66.80 (*SD* = 11.01), post intervention: *M* = 58.92 (*SD* = 14.71), *p* < .001]. With respect to the effect size we found a medium effect for PCBD and PTSD (*d* = .61 and *d* = .33, respectively) (35). PCBD diagnoses decreased from 54% (29 patients) at T1 to 41% (22 patients) at T2. PTSD diagnoses dropped from 89% (46 patients) at T1 to 70% (36 patients) at T2 (**Table 2**). Within each of the diagnostic categories (PCBD and PTSD, PTSD only, PCBD only, or no PTSD or PCBD), there was no

significant change over time. For the sample as a whole, we found a significant change in diagnostic category membership across the two assessment times: standardized MH statistic = 3.11, *p* = .002.

The symptom reduction of PCBD and PTSD was highly correlated [*r*(49) = .60, *p* < .001], also when applying expectation-maximization to account for the missing questionnaires [*r*(57) = .59].

Aim B: Post-Migration Stressors and the Association With Treatment Drop-Out and Symptom Reductions During Treatment

The following list of post-migration stressors were identified: time in Netherlands, duration of the asylum period, refugee status, language problems, work situation, housing problems, family separation of close kin, ongoing conflict in country of origin and total number of post-migration stressors. The categories of the post-migration stressors are specified in **Table 3**. Three patients experienced a change in legal status during treatment. One participant received a temporary permit, one participant lost a temporary permit and became undocumented, and one participant received, lost and received again a temporary permit during the year of treatment. Although these are interesting findings, the sample size was too small to be included in the further analyses.

On average, patients experienced three different post-migration stressors during the treatment. Undocumented asylum seekers were more likely to be non-completers (**Table 3**). Some of them completely dropped out from DPT-TG (*n* = 3) and others received a customized treatment (*n* = 2). Other post-migration stressors were not significantly associated with treatment non-completion. The post-migration stressor “ongoing conflict” was inversely associated with PTSD symptom reductions. Total number of different post-migration stressors was inversely associated with PCBD symptom reductions (**Table 4**).

TABLE 2 | Symptom reductions and diagnostic changes in the Day Patient Treatment for Traumatic Grief Completers.

	Pre-treatment score M (SD)	Post treatment score M (SD)	Significance test for differences between groups	p Value	Cohen's d
<i>Symptom reductions PCBD</i>					
<i>Patients who filled in both PCBD questionnaires (n = 54)</i>					
PCBD sum score, M (SD)	66.80 (11.01)	58.92 (14.71)	t(54) = 3.91	<.001	0.61
subdomain: separation distress. M (SD)	4.17 (0.49)	3.69 (0.89)	t(54) = 4.72	<.001	0.70
subdomain: reactive distress and social/identity disruption. M (SD)	3.56 (0.72)	3.14 (0.85)	t(54) = 3.33	.002	0.53
subdomain: functional impairment. M (SD)	3.68 (1.29)	3.19 (1.30)	t(54) = 2.63	.005	0.38
Patients endorsing separation distress, n (%)	53 (98.2)	44 (81.5)		.42	
Patients endorsing reactive distress and social/identity disruption, n (%)	49 (90.7)	47 (87.0)		.92	
Patients endorsing functional impairment, n (%)	34 (63.0)	27 (50.0)		.44	
<i>Symptom reductions PTSD</i>					
<i>Patients who completed both PTSD interviews (n = 52)</i>					
PTSD sum score, M (SD)	0.54 (0.15)	0.48 (0.19)	t(52) = 2.92	.005	0.34
Criterion B: intrusive symptoms M (SD)	0.64 (0.19)	0.54 (0.18)	t(52) = 3.44	<.001	0.55
Criterion C: avoidance of reminders of the event M (SD)	0.55 (.215)	0.41 (0.26)	t(52) = 3.49	<.001	0.62
Criterion D: negative alterations in cognitions and mood M (SD)	0.52 (0.20)	0.49 (0.24)	t(52) = 1.17	.25	0.15
Criterion E: alterations in arousal and reactivity M (SD)	0.47 (0.16)	0.44 (0.17)	t(52) = 1.10	.28	0.15
Patients meeting criterion B, n (%)	51 (98.1)	47 (90.4)		.92	
Patients meeting criterion C, n (%)	52 (100)	44 (84.6)		.48	
Patients meeting criterion D, n (%)	48 (92.3)	42 (80.8)		.60	
Patients meeting criterion E, n (%)	49 (94.2)	46 (88.5)		.84	
<i>Change in diagnostic status</i>					
<i>Participants who filled in both PCBD and PTSD questionnaires at pre-treatment (N=56) and post-treatment (N=53)</i>					
Patients endorsing the PTSD diagnosis	46 (89)	36 (70)		.32	
Patients endorsing the PCBD diagnosis	29 (54)	22 (41)		.40	
PCBD and PTSD N, (%)	27 (48)	17 (32)		.17	
PTSD only N, (%)	23 (41)	20 (38)		.76	
PCBD only N, (%)	2 (4)	6 (11)		.29	
No PTSD and PCBD	4 (7)	10 (19)		.18	
Change in diagnostic category membership within sample as a whole				.002	

DPT-TG, day patient treatment-traumatic grief; PCBD, Persistent complex bereavement disorder; PTSD, Posttraumatic stress disorder.

DISCUSSION

In this study on symptom reductions during treatment of traumatic grief in resettled refugees, a significantly correlated reduction of both PCBD and PTSD symptomatology was found with a medium effect size. Post-migration stressors were associated with both poor treatment completion and smaller symptom reductions. More specifically, not having a legal permit was associated with poor treatment completion. Ongoing conflict in the country of origin and a higher total number of post-migration stressors were associated with smaller reductions in PTSD and PCBD symptoms, respectively.

Our results showed that both PCBD and PTSD symptomatology can decrease simultaneously during treatment which is in line with the evidence available on the impact of grief focused treatment in other populations (20). However, more studies on the effectiveness of grief focused treatment in refugee populations are needed to verify our findings.

In our study, asylum seekers who were staying undocumented in Netherlands were more likely to drop out of the treatment fully or to require customized treatment. The evidence for the association between residency status and mental health has been contradictory. Although several studies have indicated that residence status is unrelated to the prevalence of mental disorders (5, 21, 40–42),

recently, in the case of PCBD, one study found that having a resident status was associated with less symptoms (7). Undocumented asylum seekers have received a negative status decision, and have only limited access to shelter, food and health care (43). Due to the combination of post-migration stressors, legal situation and uncertain future perspective, these people may be in need for special attention to prevent drop-out. Conversely, one could imagine that an improvement in legal status might be a protective factor. However, in the current study, we were unable to examine this because there was only one patient who received a temporary permit during treatment. Future studies are needed to further elucidate the association between legal status and treatment effect. Nevertheless, all other post-migration stressors were not found to be associated with treatment drop-out. It could be that clinicians' hesitance to start treatment with a refugee because of the fear for drop-out appears therefore unnecessary in most circumstances. However, because of our small sample, conclusive negative conclusions must remain tentative, pending further research. In our sample, ongoing conflict in the country of origin was significantly associated with less reductions in PTSD symptomatology. Nickerson et al. (44) evaluated the influence of fear for family remaining in the country of origin with an ongoing conflict and the mental health of Iraqi refugees in a cross-sectional survey. They found higher PTSD symptomatology and depression

TABLE 3 | Post-migration stressors pre- or during treatment in the completers and early drop-outs or customized treatment group of the refugee sample.

	Completers of DPT-TG (<i>n</i> = 57)	Early drop-outs or customized treatment (<i>n</i> = 24)	Significance test for differences between the groups	p Value
Time in the Netherlands (months), M (SD)	191.40 (104.85)	192.40 (107.56)	$t(73) = 0.29$.97
Time asylum period (months), M (SD)	66.74 (59.41)	87.40 (102.74)	$t(35) = 5.99$.45
Legal status, <i>n</i> (%)			$\chi^2(3, n = 81) = 10.0$.002 Fisher's exact test: .022
Permanent permit	46 (81)	16 (67)		
Temporary permit	7 (12)	3 (13)		
Pending	3 (5)	0 (0)		
Undocumented	1 (2)	5 (21)		
Language, <i>n</i> (%)			$\chi^2(1, n = 81) = 0.01$.92 Fisher's exact test: 1.00
Dutch	41 (72)	17 (71)		
Insufficient Dutch proficiency	16 (28)	7 (29)		
Work situation, <i>n</i> (%)			$\chi^2(3, n = 81) = 2.93$.40 Fisher's exact test: .43
Employed	2 (4)	3 (13)		
Sick leave	10 (18)	3 (13)		
Disabled	8 (14)	2 (8)		
Unemployed	37 (65)	16 (67)		
Housing problems, <i>n</i> (%)	12 (21)	8 (33)	$\chi^2(1, n = 81) = 1.37$.24 Fisher's exact test: .27
Family separation close kin, <i>n</i> (%)	18 (32)	9 (38)	$\chi^2(1, n = 81) = 0.27$.70 Fisher's exact test: .62
Ongoing conflict in country of origin, <i>n</i> (%)	30 (53)	8 (33)	$\chi^2(1, n = 81) = 2.53$.11 Fisher's exact test: .15
Total number of stressors M (SD)	2.86 (1.76)	2.83 (1.81)	$t(79) = .72$	

DPT-TG, Day patient treatment-traumatic grief.

TABLE 4 | The associations between the post-migration stressors pre- and during treatment and the TGI-SR and CAPS residual gain score.

	TGI-SR RES				CAPS RES			
	F	df1	df2	p value	F	df1	df2	p value
Legal status	1.07	3	50	.37	0.41	3	48	.75
Language	1.17	1	52	.29	0.01	1	50	.94
Work situation	0.90	3	50	.45	1.78	1	50	.19 ^a
Housing problems	1.21	1	52	.28	0.14	1	50	.72
Family separation close kin	0.68	1	52	.41	1.01	1	50	.41 ^b
Ongoing conflict in country of origin	1.36	1	52	.25 ^b	4.83	1	50	.033 ^b
Total number of stressors	4.67	1	52	.04	0.54	1	50	.47

^aDue to the violation of the assumptions regarding outliers, normality and homogeneity, we have dichotomized "work situation" for this analysis into employed versus not employed. We also performed a robust regression of the original (non-dichotomized) "work situation" variable using the package "MASS" in R. For the TGI-SR difference the t-value was 0.51, for the CAPS difference the t-value was 0.97; both values were not significant.

^bDue to the violation of the assumption regarding homogeneity we have reported the Welsh test.

CAPS RES, Clinician Administered Posttraumatic Stress Disorder residual score; TGI-SR RES, Traumatic Grief Inventory Residual score.

as well as greater mental health-related disability. It can be argued that mass media and contact with people in the home country about the ongoing conflict continues to trigger traumatic experiences and memories. Furthermore, worries about family members still in the conflict area exposed to armed conflicts and violence could play a role as to why treatment may not be effective for these individuals.

As expected, the total number of post-migration stressors was associated with smaller treatment effects. This is in line with our clinical observations of patients who sometimes appear so occupied by managing and arranging solutions for their post-migration stressors that they are not capable to adhere to the steps of a treatment protocol. Clinicians need to provide emotional and

information support for patients reporting post migration stressors, knowing that this might not have short term positive effects in mental health outcomes.

Limitations of this study must be noted. The findings of this study need to be interpreted in the light of its naturalistic (real-world) setting. It cannot be stated with certainty that the decreases in PCBD- and PTSD symptom severity can be attributed to the effects of the DPT-TG. Furthermore, we could only assess the importance of the post-migration stressors that clinicians noted in the patient files. On the one hand, one could argue that we have captured all factors that may influence the treatment process according to the clinicians and patients themselves. On the other hand, “ongoing conflict in the country of origin” was one of the few post-migration stressors found to be associated with treatment outcome and this stressor was not systematically asked by clinicians in the patient files. Therefore, there might be a lack of knowledge by clinicians about which post-migration stressors are worth assessing at the beginning of treatment and theoretically, some participants may have experienced difficulties but did not mention them in therapy. As post-migration stressors were identified at some point during the treatment and recorded in the patient files, we could not specifically determine at what point in time during the course of treatment these stressors had the largest impact. Future studies are needed to investigate the role of post-migration stressors more elaborately and standardized, by including living difficulties questionnaires in routine measures. Furthermore, our study had an uncontrolled study design and a relative small sample size, future studies need to assess the treatment effect of the DPT-TG with a control group and follow-up measurement.

Strengths of this study must also be noted. The findings of this study generate several well-informed answers to some urgent clinical questions that rise in treating resettled refugee patient populations characterized by multiple post-migration stressors. Our findings indicate that treatment for traumatic grief coincides with alleviations in both PCBD and PTSD symptoms in refugees who have faced multiple traumatic events and traumatic losses. Findings also indicate that traumatized refugees may benefit from treatment even in the presence of multiple post-migration stressors.

Implications of this study include that clinicians need to keep in mind that post-migration stressors may interfere with a smooth treatment process and that these stressors are worth documenting systematically at intake. In the case of ongoing conflict or a high number of stressors, symptom reductions may be modest, and clinicians may educate patients about these effects to manage treatment expectations and to prevent demoralization. Furthermore, special attention is needed for undocumented patients who were denied a refugee status in order to prevent them from early drop-out of treatment. Further studies are needed to investigate how to raise refugee wellbeing and assist with their resettlement in a host country can be beneficial to enhance treatment outcome in psychological interventions.

DATA AVAILABILITY STATEMENT

The ROM data of ARQ National Psychotrauma center was accessible for the authors during the time of study. The data is not publicly available. Requests to access the data should be directed to the corresponding author.

ETHICS STATEMENT

Ethical approval was not provided for this study on human participants because Measurements were part of the Routine Outcome Measurements (ROM). We consulted the medical ethics committee of Leiden University, and the study was exempted from formal review because the primary purpose of ROM is not research oriented. All patients were informed during the ROM that their answers could be anonymously used for research purposes and could object if they did not agree. None of the patients eligible for inclusion in the current study expressed objections. Written informed consent for participation was not required for this study in accordance with the national legislation and the institutional requirements.

AUTHOR CONTRIBUTIONS

AD, GS, and PB were responsible for the design of the study. AD, GS, AH, and DK were responsible for the data-collection. AD was responsible for the data-analysis and AD, GS, and PB for the interpretation of the data. AH and DK contributed to the data-analysis and paragraphs of the manuscript. PB, RK, and GS supervised AD. AD wrote the drafts of the manuscript. All authors were involved in revising the draft versions critically and all authors approved the final version of the manuscript.

ACKNOWLEDGMENTS

The authors thank the multidisciplinary team of clinicians, therapists and social workers at the DPT-TG for their co-operation in this project. Special thanks to the master students Kaje Fraaij, Marit Hoekema, Marinde van Egmond, Joyce Jager, and Tess Davidson for their help in the data collection. We also thank dr. G. Vink for his advice concerning the data-analysis.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: <https://www.frontiersin.org/articles/10.3389/fpsy.2020.00407/full#supplementary-material>

REFERENCES

- de Jong JT, Komproe IH, Van Ommeren M, El Masri M, Araya M, Khaled N, et al. Lifetime events and posttraumatic stress disorder in 4 postconflict settings. *JAMA* (2001) 286(5):555–62. doi: 10.1001/jama.286.5.555
- Knipscheer JW, Sleijpen M, Mooren TM, ter Heide FJJ, van der Aa N. Trauma exposure and refugee status as predictors of mental health outcomes in treatment-seeking refugees. *BJPsych Bull* (2015) 39(4):178–82. doi: 10.1192/pb.bp.114.047951
- Fazel M. Refugees and the post-migration environment. *BMC Med* (2018) 16(1):164. doi: 10.1186/s12916-018-1155-y
- Bogic M, Njoku A, Priebe S. Long-term mental health of war-refugees: a systematic literature review. *BMC Int Health Hum Rights* (2015) 15:29. doi: 10.1186/s12914-015-0064-9
- Steel JL, Dunlavy AC, Harding CE, Theorell T. The psychological consequences of pre-emigration trauma and post-migration stress in refugees and immigrants from Africa. *J Immigrant Minority Health* (2017) 19(3):523–32. doi: 10.1007/s10903-016-0478-z
- Nesterko Y, Jäckle D, Friedrich M, Holzapfel L, Glaesmer H. Prevalence of post-traumatic stress disorder, depression and somatisation in recently arrived refugees in Germany: an epidemiological study. *Epidemiol Psychiatr Sci* (2020) 29:E40. doi: 10.1017/S2045796019000325
- Comtesse H, Rosner R. Prolonged grief disorder among asylum seekers in Germany: the influence of losses and residence status. *Eur J Psychotraumatol* (2019) 10(1):1591330. doi: 10.1080/2008198.2019.1591330
- Nickerson A, Liddell BJ, Maccallum F, Steel Z, Silove D, Bryant RA. Posttraumatic stress disorder and prolonged grief in refugees exposed to trauma and loss. *BMC Psychiatry* (2014) 14:106–244X-14-106. doi: 10.1186/1471-244X-14-106
- Djelantik AAAMJ, Robinaugh DJ, Kleber RJ, Smid GE, Boelen PA. Symptomatology following loss and trauma: Latent class and network analyses of prolonged grief disorder, posttraumatic stress disorder, and depression in a treatment-seeking trauma-exposed sample. *Depression Anxiety* (2020) 37:26–34. doi: 10.1002/da.22880
- Bryant RA, Edwards B, Creamer M, O'Donnell M, Forbes D, Felmingham KL, et al. A population study of prolonged grief in refugees. *Epidemiol Psychiatr Sci* (2020) 29:E44. doi: 10.1017/S2045796019000386
- Tay AK, Mohsin M, Rees S, Tam N, Kareth M, Silove D. The structure and psychosocial correlates of complicated bereavement amongst refugees from West Papua. *Soc Psychiatry Psychiatr Epidemiol* (2019) 54(6):771–80. doi: 10.1007/s00127-019-01666-1
- American Psychiatric Association. *Diagnostic and Statistical Manual of Mental Disorders. 5th ed.* Arlington, VA: American Psychiatric Publishing (2013).
- Prigerson HG, Frank E, Kasl SV, Reynolds CF, Anderson B, Zubeck GS, et al. Complicated grief and bereavement-related depression as distinct disorders: preliminary empirical validation in elderly bereaved spouses. *Am J Psychiatry* (1995) 152(1):22–30. doi: 10.1176/ajp.152.1.22
- Mauro C, Reynolds CF, Maercker A, Skritskaya N, Simon N, Zisook S, et al. Prolonged grief disorder: Clinical utility of ICD-11 diagnostic guidelines. *psychol Med* (2019) 49(5):861–7. doi: 10.1017/S0033291718001563
- Lundorff M, Holmgren H, Zachariae R, Farver-Vestergaard I, O'Connor MF. Prevalence of prolonged grief disorder in adult bereavement: A systematic review and meta-analysis. *J Affect Disord* (2017) 212:138–49. doi: 10.1016/j.jad.2017.01.030
- Currier JM, Holland JM, Coleman RA, Neimeyer RA. Bereavement following violent death: An assault on life and meaning. In: Cox RGSGR, editor. *Perspectives on violence and violent death. Death, value and meaning series.* Amityville, NY: Baywood Publishing Co; US (2008). p. 177–202.
- Djelantik AAAMJ, Smid GE, Kleber RJ, Boelen PA. Symptoms of prolonged grief, post-traumatic stress, and depression after loss in a Dutch community sample: A latent class analysis. *Psychiatry Res* (2017) 247:276–81. doi: 10.1016/j.psychres.2016.11.023
- de Heus A, Hengst SMC, de la Rie SM, Djelantik AAAMJ, Boelen PA, Smid GE. Day patient treatment for traumatic grief: preliminary evaluation of a one-year treatment programme for patients with multiple and traumatic losses. *Eur J Psychotraumatol* (2017) 8(1):1375335. doi: 10.1080/2008198.2017.1375335
- Smid GE, Kleber RJ, de la Rie SM, Bos JBA, Gersons BPR, Boelen PA. Brief Eclectic Psychotherapy for Traumatic Grief (BEP-TG): toward integrated treatment of symptoms related to traumatic loss. *Eur J Psychotraumatol* (2015) 6:1. doi: 10.3402/ejpt.v6.27324
- Eddinger JR, Hardt MM, Williams JL. Concurrent Treatment for PTSD and Prolonged Grief Disorder: Review of Outcomes for Exposure-and Nonexposure-Based Treatments. *OMEGA J Death Dying* (2019), 0030222819854907. doi: 10.1177/0030222819854907
- Li SY, Liddell BJ, Nickerson A. The relationship between post-migration stress and psychological disorders in refugees and asylum seekers. *Curr Psychiatry Rep* (2016) 18(9):82. doi: 10.1007/s11920-016-0723-0
- Porter M, Haslam N. Predisplacement and postdisplacement factors associated with mental health of refugees and internally displaced persons: a meta-analysis. *JAMA* (2005) 294(5):602–12. doi: 10.1001/jama.294.5.602
- Kramer SA, Olsman E, Hoogsteder MHH, Van Willigen LHM. Sleepless Nights because of Ethical Dilemmas in Mental Health Care for Asylum Seekers. *J Refugee Stud* (2017) 31(4):466–87. doi: 10.1093/jrs/fex039
- Jacobsen K, Landau LB. The dual imperative in refugee research: some methodological and ethical considerations in social science research on forced migration. *Disasters* (2003) 27(3):185–206. doi: 10.1111/1467-7717.00228
- Sonne C, Carlsson J, Bech P, Vindbjerg E, Mortensen E, Elklit A. Psychosocial predictors of treatment outcome for trauma-affected refugees. *Eur J Psychotraumatol* (2016) 7(1):30907. doi: 10.3402/ejpt.v7.30907
- Schick M, Morina N, Mistridis P, Schnyder U, Bryant RA, Nickerson A. Changes in Post-migration Living Difficulties Predict Treatment Outcome in Traumatized Refugees. *Front Psychiatry* (2018) 9:476. doi: 10.3389/fpsy.2018.00476
- Boelen PA, Smid GE. The Traumatic Grief Inventory Self-Report Version (TGI-SR): Introduction and Preliminary Psychometric Evaluation. *J Loss Trauma* (2017) 22(3):196–212. doi: 10.1080/15325024.2017.1284488
- Boelen PA, Djelantik AAAMJ, de Keijser J, Lenferink LIM, Smid GE. Further validation of the Traumatic Grief Inventory-Self Report (TGI-SR): A measure of persistent complex bereavement disorder and prolonged grief disorder. *Death Stud* (2019) 43(6):351–64. doi: 10.1080/07481187.2018.1480546
- Weathers FW, Blake DD, Schnurr PP, Kaloupek DG, Marx BP, Keane TM. The clinician-administered PTSD scale for DSM-5 (CAPS-5). National Center for Posttraumatic Stress Disorder (2013). [Available from: www.ptsd.va.gov.
- Blake DD, Weathers FW, Nagy LM, Kaloupek DG, Gusman FD, Charney DS, et al. The development of a clinician-administered PTSD scale. *J Traumatic Stress* (1995) 8(1):75–90. doi: 10.1002/jts.2490080106
- Erdfelder E, Faul F, Buchner A. GPOWER: A general power analysis program. *Behav Res Methods Instruments Computers* (1996) 28(1):1–11. doi: 10.3758/BF03203630
- Faul F, Erdfelder E, Buchner A, Lang AG. Statistical power analyses using G* Power 3.1: Tests for correlation and regression analyses. *Behav Res Methods* (2009) 41(4):1149–60. doi: 10.3758/BRM.41.4.1149
- Agresti A. *Categorical data analysis.* Hoboken, New Jersey: John Wiley & Sons, Inc. (2003). doi: 10.1002/0471249688
- Cohen JA. *Statistical power analysis for the behavioral sciences.* New York: Lawrence Erlbaum Associates (1988).
- Dempster AP, Laird NM, Rubin DB. Maximum likelihood from incomplete data via the EM algorithm. *J R Stat Society: Ser B Methodol* (1977) 39(1):1–22. doi: 10.1111/j.2517-6161.1977.tb01600.x
- Moon TK. The expectation-maximization algorithm. *IEEE Signal processing magazine.* (1996) 13: (6):47–60. doi: 10.1109/79.543975
- Wagner WE. *Using IBM® SPSS® statistics for research methods and social science statistics. 7th ed.* Los Angeles, CA, US: Sage Publications (2019).
- Field A. *Discovering statistics using IBM SPSS statistics.* CA: Sage Publications; (2013).
- Steketee G, Chambless DL. Methodological issues in prediction of treatment outcome. *Clin Psychol Rev* (1992) 12(4):387–400. doi: 10.1016/0272-7358(92)90123-P
- Kirmayer LJ, Narasiah L, Munoz M, Rashid M, Ryder AG, Guzder J, et al. Common mental health problems in immigrants and refugees: general approach in primary care. *Can Med Assoc J* (2011) 183(12):E959–E67. doi: 10.1503/cmaj.090292
- Laban CJ, Komproe IH, Gernaat HBPE, de Jong JTV. The impact of a long asylum procedure on quality of life, disability and physical health in Iraqi asylum seekers in the Netherlands. *Soc Psychiatry Psychiatr Epidemiol* (2008) 43(7):507. doi: 10.1007/s00127-008-0333-1

42. Gerritsen AA, Bramsen I, Devillé W, van Willigen LHM, Hovens JE, Van Der Ploeg HM. Physical and mental health of Afghan, Iranian and Somali asylum seekers and refugees living in the Netherlands. *Soc Psychiatry Psychiatr Epidemiol* (2006) 41(1):18–26. doi: 10.1007/s00127-005-0003-5
43. Lahuis AM, Scholte WF, Aarts R, Kleber RJ. Undocumented asylum seekers with posttraumatic stress disorder in the Netherlands. *Eur J Psychotraumatol* (2019) 10(1):1605281. doi: 10.1080/20008198.2019.1605281
44. Nickerson A, Bryant RA, Steel Z, Silove D, Brooks R. The impact of fear for family on mental health in a resettled Iraqi refugee community. *J Psychiatr Res* (2010) 44(4):229–35. doi: 10.1016/j.jpsychires.2009.08.006

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Copyright © 2020 Djelantik, de Heus, Kuiper, Kleber, Boelen and Smid. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.



Valid ICD-11 PGD Scales and Structured Clinical Interviews Needed

Maja O'Connor^{1,2*}, Lene Larsen², Biretha V. Joensen², Paul A. Boelen^{3,4}, Fiona Maccallum⁵, Katrine Komischke-Konnerup¹ and Richard A. Bryant⁶

¹ Unit for Bereavement Research, Department of Psychology, Aarhus University, Aarhus, Denmark, ² The Danish National Center for Grief, Copenhagen, Denmark, ³ Department of Psychology, Utrecht University, Utrecht, Netherlands, ⁴ ARQ National Psychotrauma Centre, Diemen, Netherlands, ⁵ School of Psychology, University of Queensland, St Lucia, QLD, Australia, ⁶ School of Psychology, University of New South Wales, Sydney, NSW, Australia

Keywords: prolonged grief disorder, ICD-11 PGD, psychometrics, PGD scales, valid clinical cut off, structured clinical ICD-11 PGD interview, diagnostic test accuracy

OPEN ACCESS

Edited by:

Geert E. Smid,
ARQ National Psychotrauma
Centre, Netherlands

Reviewed by:

Paula Schnurr,
National Center for Posttraumatic
Stress Disorder (PTSD), United States
Rahel Bachem,
Tel Aviv University, Israel

*Correspondence:

Maja O'Connor
maja@psy.au.dk

Specialty section:

This article was submitted to
Psychopathology,
a section of the journal
Frontiers in Psychology

Received: 19 February 2020

Accepted: 30 April 2020

Published: 28 May 2020

Citation:

O'Connor M, Larsen L, Joensen BV,
Boelen PA, Maccallum F,
Komischke-Konnerup K and
Bryant RA (2020) Valid ICD-11 PGD
Scales and Structured Clinical
Interviews Needed.
Front. Psychol. 11:1120.
doi: 10.3389/fpsyg.2020.01120

INTRODUCTION

Official Diagnostic Criteria for Prolonged Grief Disorder Are Coming

In 2022, when the 11th version of the World Health Organization's International Classification of Diseases manual (ICD-11) goes into effect, it will for the first time include diagnostic criteria for Prolonged Grief Disorder (ICD-11 PGD; WHO, 2019). ICD-11 PGD refers to a debilitating grief reaction following bereavement that occurred at least 6 months earlier. It is defined as an intense, persistent, and impairing grief response, characterized by longing for and/or preoccupation with the deceased and intense emotional distress that can include sadness, guilt, anger, denial, blame, difficulty accepting the death, feeling one has lost a part of one's self, an inability to experience positive mood, emotional numbness, or difficulty in engaging with social or other activities. In addition, this response must clearly exceed the grief response expected given the person's cultural and religious context (WHO, 2019). The implementation of ICD-11 PGD will provide health professionals worldwide with a diagnosis to facilitate identification and treatment of patients with PGD. As a condition, ICD-11 PGD will likely receive increased attention in research, clinical, and teaching settings alike. The introduction of the ICD-11 diagnosis for PGD should be considered in light of the conceptualization of disordered grief within DSM 5 (American Psychiatric Association, 2013). The two diagnostic formulations have been found to have significant semantic and empirical overlap when both applied with a set number of symptoms to fulfill (Maciejewski et al., 2016; O'Connor et al., 2019). However, unlike ICD-11, the DSM-5 deemed that there was insufficient data to justify introducing a grief-related diagnosis and instead included Persistent Complex Bereavement Disorder (PCBD; Bryant, 2014) in section Discussion, as a condition for further study; the goal being that additional epidemiological and longitudinal studies may inform a formal diagnosis in future iterations of DSM. Scales have been developed to measure PCBD (Lee, 2015, 2018; Boelen et al., 2018a,b) and an adaption of PCBD (including changing the name to Prolonged Grief Disorder) based on contemporary data has very recently been proposed (see American Psychiatric Association, 2020; Boelen and Lenferink, 2020). However, to date, ICD-11 PGD is still the only official diagnosis for PGD, and it is used in most European Health Services, which is why the ICD-11 PGD will be the focus of the present review.

The State of Current Instruments Measuring Prolonged Grief Disorder

Complicated, problematic, and persistent grief responses have long been of interest to clinicians and researchers (see Killikelly and Maercker, 2018 for a brief review). Over the past two decades, a number of research groups have worked to describe and identify patients who struggle with prolonged and complicated grief responses (e.g., Prigerson et al., 2009; Shear et al., 2011). The work to develop valid scales and clinical interviews for disordered grief, however, is hampered by a lack of consensus regarding the diagnostic criteria (Lenferink et al., 2019). One of the first instruments to measure prolonged grief was the Inventory of Complicated Grief (ICG) developed in 1995 by Prigerson and colleagues to capture putative markers of what was then called Complicated Grief (Prigerson et al., 1995). The ICG has since been revised several times [Inventory of Traumatic Grief (ITG; Prigerson and Jacobs, 2001); Inventory of Complicated Grief Revised (ICG-R; He et al., 2013)], finally evolving into the PG-13 (Prigerson et al., 2009). The PG-13 is a short, easily administered self-report scale that captures most of the core characteristics, including yearning for the deceased and associated symptoms as defined in ICD-11 PGD (Prigerson et al., 2009). The ICG and its successors, particularly ICG-R and PG-13, are the instruments used in most frequently by studies supporting both ICD-11 PGD and PCBD criteria, and are the scales that most closely resembles ICD-11 PGD. Although studies have found these measures of prolonged grief to have sound psychometric properties, including internal consistency, test-retest reliability, and construct, concurrent, convergent and criterion validity (Prigerson et al., 1995; Lichtenthal et al., 2011; Carmassi et al., 2014; Papa et al., 2014; Boelen and Smid, 2017; Boelen et al., 2018a; Pohlkamp et al., 2018), none correspond directly to the ICD-11 PGD definition.

It is worth noting that a 31 item structured clinical interview for persistent grief reactions has been proposed based on the DSM-5's definition of PCBD (Bui et al., 2015). The goal of this instrument was to promote data collection on grief symptoms to inform future diagnostic criteria. However, it does not offer an assessment corresponding to the current ICD-11 PGD definition. Further, the interview was developed with treatment-seeking patients with persistent grief reactions, and thus at this stage does not provide information that discriminates between patients with and without prolonged grief (Bui et al., 2015).

ASSESSMENT OF DIAGNOSTIC TEST ACCURACY IS LACKING FOR CURRENT SCALES FOR PROLONGED GRIEF

While current self-report scales as described above may be reliable and valid in terms of capturing grief symptoms associated with functional impairment (Prigerson et al., 1995), cut off scores to distinguish between people who meet the proposed PGD ICD-11 criteria and people who do not, have apparently not been adequately validated. That is, diagnostic test accuracy (DTA; Campbell et al., 2015) may yet need to be established for these scales. DTA is typically assessed in terms of sensitivity (i.e.,

the ability of a test to detect true positive cases as defined by a reference standard, here the clinical evaluation), specificity (i.e., the ability of a test to detect those who do not have a specific condition; Trevethan, 2017), and ROC curve analysis (see Leeflang, 2014; Campbell et al., 2015). A PGD scale with established DTA would enable estimation of the prevalence of PGD, tracking of the course of the condition, and identification of risk factors for developing PGD across time, studies, and populations. A questionnaire with sound DTA could also serve as a valid screening tool for PGD. In combination with a structured clinical interview, a self-report assessment tool for ICD-11 PGD with valid DTA would facilitate clinical decision-making, including ruling out the presence of a disorder when relevant, and tracking treatment progress.

We were interested in determining the established DTA of self-report measures of prolonged grief, and therefore we conducted a systematic review of the literature. Our questionnaires of interest were the ICG and its successors, the ITG (Prigerson and Jacobs, 2001), ICG-R (He et al., 2013), and the PG 13 (Prigerson et al., 2009). As described above, these measures represent the most psychometrically sound and commonly used measures of PGD in the literature. The reference standard used in our approach was the presence of a diagnosis of PGD or significant clinical impairment related to bereavement as determined by a clinical diagnostic evaluation performed by clinicians. The review was set up in accordance with the PRISMA-DAT Statement (McInnes et al., 2018). We used the following databases: PubMed, Embase, CINAHL, PsychINFO and Web of science, and went back to January 1995, which is the year of the first publication on the ICG (Prigerson et al., 1995). For a more detailed description of the review, see the protocol submitted to PROSPERO (CRD42019138906).

Only one eligible study comparing a self-report scale and a structured clinical interview for prolonged grief was identified in this systematic review. Boelen et al. (2003) examined the psychometric properties of the Dutch version of the ITG (Prigerson and Jacobs, 2001), with groups of treatment-seeking adults who had experienced the loss of a first-degree-relative. To establish caseness of prolonged grief, participants were interviewed using the Traumatic Grief Evaluation of Response to Loss clinical interview (Prigerson et al., 1998). Unfortunately, however, the symptom-duration of this instrument for establishing caseness is 2 months (in accord with consensus criteria for what was then called "traumatic grief"; Prigerson et al., 1999), which is not compatible with the current duration requirement of 6 months in ICD-11 PGD. This means that the obtained questionnaire cut-off score (DTA) is not applicable today. As a result, we therefore conclude, based on our systematic review, that the field lacks studies that determine the DTA for the self-report scales currently used to measure the presence of disordered grief.

The Issue of Comorbidity

Disturbed grief is often comorbid with other psychological conditions, and most commonly with depression, anxiety, and PTSD (Jordan and Litz, 2014). Accordingly, the issue of differential diagnosis is crucial to determine if a bereaved

person may have ICD-11 PGD alone, or is suffering with other possible conditions such as bereavement-related depression or PTSD. This is particularly important because research indicates that whereas symptoms of prolonged grief respond well to grief-focused psychotherapy (Johannsen et al., 2019), they do not respond optimally to psychological or pharmacological treatments that target depression (Shear et al., 2005, 2016). Some of the symptoms of prolonged grief captured by the ICD-11 PGD overlap with depression, including sadness and reduced interest in activities, and so development of standardized measures of ICD-11 PGD need to be conducted along with differential assessments to ascertain the extent to which the measures can differentiate these potentially overlapping conditions.

The Need for a Structured Clinical Interview

Although self-report scales are appealing because they can be administered easily and cheaply, self-report scales are not the gold standard for assessing a psychiatric condition. Studies indicate modest concordance between self-report scales and structured clinical interviews (Eaton et al., 2000), with self-report scales typically elevating the incidence of rates of a disorder (e.g., Bui et al., 2015). For this reason, advancing the study of disturbed and prolonged grief requires the development of structured clinical interviews that correspond with the ICD-11 PGD criteria. As noted, a structured clinical interview based on PCBD has been proposed (Bui et al., 2015). However, symptoms do not correspond directly with ICD-11 PGD criteria, and the measure awaits validation with a representative sample of bereaved individuals. A structured clinical interview based on ICD-11 PGD criteria is necessary to standardize the assessment of prevalence rates of PGD with accuracy, and self-report scales alone cannot provide this.

DISCUSSION

ICD-11 PGD is a complex condition. Until now, there have been numerous definitions of disturbed grief, encompassing ICD-11, DSM-5, and other proposals (Boelen and Lenferink, 2020), which has caused confusion for the field (Lenferink et al., 2019). The introduction of the ICD-11 PGD is welcome because it, for the first time, introduces a formal definition by which disturbed grief can be described and studied. However, the advancement of our knowledge of this disorder requires standardized assessment tools that correspond to these criteria. As mentioned above, existing scales have sound psychometric properties and an ability to capture grief reactions with functional impairment (Prigerson et al., 1995; Lichtenthal et al., 2011; Carmassi et al., 2014;

Papa et al., 2014; Boelen and Smid, 2017; Boelen et al., 2018a; Pohlkamp et al., 2018), but these scales do not precisely capture ICD-11 PGD and lack evaluation of diagnostic test accuracy. There is a need for both valid structured clinical interviews for ICD-11 PGD and valid self-report scales for ICD-11 PGD with adequate diagnostic test accuracy and a valid clinical cut off. There is much to learn from earlier work on PGD scales. PGD is a young psychiatric disorder, and further revisions of symptoms and duration criteria may come for both DSM and ICD PGD. Therefore, a new scale should include items corresponding closely to ICD-11 PGD and future developments could usefully have structured scales to also index symptoms specified by DSM to allow careful and standardized comparisons of the two definitions of PGD (cf. Lenferink et al., 2019). Earlier scales (e.g., PG-13) have used multiple response formats including frequency and severity based formats. The development of a new scale would benefit from close collaboration with clinical staff working with PGD, to ensure optimal and clinically meaningful item formulations and response formats. The scale should also be tested and revised accordingly in population-based bereaved samples to ensure its representativeness for general bereavement reactions. Finally, a structured clinical interview for ICD-11 PGD must similarly be developed and validated in clinical and non-clinical samples in close collaboration with clinicians. This interview is necessary to ensure diagnostic test accuracy and to define a valid clinical cut off on the self-report scale for use in research and the clinic. Although ICD-11 will not be formally launched until 2022, this work should be commenced now. Development and use of standardized assessment tools will allow for meaningful comparisons across studies in terms of prevalence, course, and treatment outcomes for individuals with ICD-11 PGD. This will both advance our understanding of how to best conceptualize disturbed grief and foster collaborations in our effort to reduce the significant suffering associated with the condition.

AUTHOR CONTRIBUTIONS

The paper was mainly written by MO'C and LL supervised by RB. BJ and LL performed the systematic review supervised by MO'C. PB, KK-K, BJ, and FM read and commented the paper several times in the process. All authors approved the final version of the paper.

FUNDING

This opinion paper was funded by The Danish National Center for Grief. This center also covered the publication fees.

REFERENCES

- American Psychiatric Association (2013). *Diagnostic and Statistical Manual for Mental Disorders, 5th Edn.* Washington, DC: American Psychiatric Association. doi: 10.1176/appi.books.9780890425596
- American Psychiatric Association (2020). Available online at: <https://www.psychiatry.org/psychiatrists/practice/dsm/proposed-changes> (accessed May 18, 2020).
- Boelen, P. A., Djelantik, A. A. M. J., de Keijser, J., Lenferink, L. I. M., and Smid, G. E. (2018a). Further validation of the traumatic grief inventory-self report (tgi-sr): a measure of persistent complex bereavement disorder and prolonged grief disorder. *Death Stud.* 43, 351–364. doi: 10.1080/07481187.2018.1480546
- Boelen, P. A., and Lenferink, L. I. M. (2020). Comparison of six proposed diagnostic criteria sets for disturbed grief. *Psychiatr. Res.* 285:112786. doi: 10.1016/j.psychres.2020.112786

- Boelen, P. A., Lenferink, L. I. M., Nickerson, A., and Smid, G. E. (2018b). Evaluation of the factor structure, prevalence, and validity of disturbed grief in DSM-5 and ICD-11. *J. Affect. Disord.* 240, 79–87. doi: 10.1016/j.jad.2018.07.041
- Boelen, P. A., and Smid, G. E. (2017). The traumatic grief inventory self-report version (TGI-SR): introduction and preliminary psychometric evaluation. *J. Loss Trauma* 22, 196–212. doi: 10.1080/15325024.2017.1284488
- Boelen, P. A., Van Den Bout, J., De Keijser, J., and Hooijink, H. (2003). Reliability and validity of the Dutch version of the inventory of traumatic grief (ITG). *Death Stud.* 27, 227–247. doi: 10.1080/07481180302889
- Bryant, R. A. (2014). Prolonged grief: where to after diagnostic and statistical manual of mental disorders, 5th edition? *Curr. Opin. Psychiatr.* 27, 21–26. doi: 10.1097/YCO.0000000000000031
- Bui, E., Mauro, C., Robinaugh, D. J., Skritskaya, N. A., Wang, Y., Gribbin, C., et al. (2015). The structured clinical interview for complicated grief: reliability, validity, and exploratory factor analysis. *Depress. Anxiety* 32, 485–492. doi: 10.1002/da.22385
- Campbell, J. M., Klugar, M., Ding, S., Carmody, D. P., Hakonsen, S. J., Jadotte, Y. T., et al. (2015). Diagnostic test accuracy: methods for systematic review and meta-analysis. *Int. J. Evid. Based Healthc.* 13, 154–162. doi: 10.1097/XEB.0000000000000061
- Carmassi, C., Shear, M. K., Massimetti, G., Wall, M., Mauro, C., Gemignani, S., et al. (2014). Validation of the Italian version inventory of complicated grief (ICG): a study comparing CG patients versus bipolar disorder, PTSD and healthy controls. *Compr. Psychiatry* 55, 1322–1329. doi: 10.1016/j.comppsy.2014.03.001
- Eaton, W. W., Neufeld, K., Chen, L. S., and Cai, G. (2000). A comparison of self-report and clinical diagnostic interviews for depression: diagnostic interview schedule and schedules for clinical assessment in neuropsychiatry in the Baltimore epidemiologic catchment area follow-up. *Arch. Gen. Psychiatry* 57, 217–222. doi: 10.1001/archpsyc.57.3.217
- He, L., Wang, J., Tang, S., Yu, W., Qiuyuan, X. (2013). Reliability and validity of the inventory of complicated grief-revised. *Chin. Ment. Health J.* 27, 937–943. doi: 10.3969/j.issn.1000-6729.2013.12.010
- Johannsen, M., Damholdt, M. F., Zachariae, R., Lundorff, M., Farver-Vestergaard, I., and O'Connor, M. (2019). Psychological interventions for grief in adults: a systematic review and meta-analysis of randomized controlled trials. *J. Affect. Disord.* 253, 69–89. doi: 10.1016/j.jad.2019.04.065
- Jordan, A. H., and Litz, B. T. (2014). Prolonged grief disorder: diagnostic, assessment, and treatment considerations. *Prof. Psychol. Res.* 45, 180–187. doi: 10.1037/a0036836
- Killikelly, C., and Maercker, A. (2018). Prolonged grief disorder for ICD-11: the primacy of clinical utility and international applicability. *Eur. J. Psychotraumatol.* 8:1476441. doi: 10.1080/20008198.2018.1476441
- Lee, S. A. (2015). The persistent complex bereavement inventory: a measure based on the DSM-5. *Death Stud.* 39, 399–410. doi: 10.1080/07481187.2015.1029144
- Lee, S. A. (2018). Factorial structure of the persistent complex bereavement inventory: testing a hierarchical factor model. *Death Stud.* 42, 356–361. doi: 10.1080/07481187.2017.1348402
- Leeftang, M. M. G. (2014). Systematic reviews and meta-analyses of diagnostic test accuracy. *Clin. Microbiol. Infect.* 20, 105–113. doi: 10.1111/1469-0691.12474
- Lenferink, L. I. M., Boelen, P. A., Smid, G. E., and Paap, M. C. S. (2019). The importance of harmonising diagnostic criteria sets for pathological grief. *Br. J. Psychiatry* 13, 1–4. doi: 10.1192/bjp.2019.240
- Lichtenthal, W. G., Nilsson, M., Kissane, D. W., Breitbart, W., Kacel, E., Jones, E. C., et al. (2011). Underutilization of mental health services among bereaved caregivers with prolonged grief disorder. *Psychiatr. Serv.* 62, 1225–1229. doi: 10.1176/ps.62.10.pss6210_1225
- Maciejewski, P. K., Maercker, A., Boelen, P. A., and Prigerson, H. G. (2016). “Prolonged grief disorder” and “persistent complex bereavement disorder,” but not “complicated grief,” are one and the same diagnostic entity: an analysis of data from the Yale bereavement study. *World Psychiatr.* 15, 266–275. doi: 10.1002/wps.20348
- McInnes, M. D. F., Moher, D., Thombs, B. D., McGrath, T. A., Bossuyt, P. M., and The PRISMA-DTA Group (2018). Preferred reporting items for a systematic review and meta-analysis of diagnostic test accuracy studies: the PRISMA-DTA statement. *JAMA* 319, 388–396. doi: 10.1001/jama.2017.19163
- O'Connor, M., Lasgaard, M., Larsen, L., Johannsen, M., Lundorff, M., Farver-Vestergaard, I., et al. (2019). Comparison of proposed diagnostic criteria for pathological grief using a sample of elderly bereaved spouses in Denmark: perspectives on future bereavement research. *J. Affect. Disord.* 251, 52–59. doi: 10.1016/j.jad.2019.01.056
- Papa, A., Lancaster, N., and Kahler, J. (2014). Commonalities in grief responding across bereavement and non-bereavement losses. *J. Affect. Disord.* 161, 136–143. doi: 10.1016/j.jad.2014.03.018
- Pohlkamp, L., Kreicbergs, U., Prigerson, H. G., and Sveen, J. (2018). Psychometric properties of the prolonged grief disorder-13 (PG-13) in bereaved Swedish parents. *Psychiatr. Res.* 267, 560–565. doi: 10.1016/j.psychres.2018.06.004
- Prigerson, H. G., Horowitz, M. J., Jacobs, S. C., Parkes, C. M., Aslen, M., Goodkin, K., et al. (2009). Prolonged grief disorder: psychometric validation of criteria proposed for DSM-V and ICD-11. *PLoS Med.* 6:e1000121. doi: 10.1371/journal.pmed.1000121
- Prigerson, H. G., and Jacobs, S. C. (2001). “Traumatic grief as a distinct disorder: a rationale, consensus criteria, and a preliminary empirical test,” in *Handbook of Bereavement Research. Consequences, Coping and Care*, eds. M. S. Stroeve, R. O. Hansson, W. Stroebe and H. A. W. Schut (Washington, DC: American Psychological Association Press), 613–647. doi: 10.1037/10436-026
- Prigerson, H. G., Kasl, S. V., and Jacobs, S. C. (1998). *Traumatic Grief Evaluation of Response to Loss (TRGR2L)*. New York, NY: Unpublished manuscript.
- Prigerson, H. G., Maciejewski, P. K., Reynolds, C. F. III, Bierhals, A. J., Newsom, J. T., Fasiczka, A., et al. (1995). Inventory of complicated grief: a scale to measure maladaptive symptoms of loss. *Psychiatr. Res.* 59, 65–79. doi: 10.1016/0165-1781(95)02757-2
- Prigerson, H. G., Shear, M. K., Jacobs, S. C., Reynolds 3rd C. F., Maciejewski, P. K., Davidson, J. R., et al. (1999). Consensus criteria for traumatic grief. A preliminary empirical test. *Br. J. Psychiatry* 174, 67–73. doi: 10.1192/bjp.174.1.67
- Shear, K., Frank, E., Houck, P. R., and Reynolds, C. F. III. (2005). Treatment of complicated grief: a randomized controlled trial. *JAMA* 293, 2601–2608. doi: 10.1001/jama.293.21.2601
- Shear, M. K., Reynolds, C. F., Simon, N. M., Zisook, S., Wang, Y., Mauro, C., et al. (2016). Optimizing treatment of complicated grief: a randomized clinical trial. *JAMA Psychiatr.* 73, 685–694. doi: 10.1001/jamapsychiatry.2016.0892
- Shear, M. K., Simon, N., Wall, M., Zisook, S., Neimeyer, R., Duan, N., et al. (2011). Complicated grief and related bereavement issues for DSM-5. *Depress. Anxiety* 28, 103–117. doi: 10.1002/da.20780
- Trevethan, R. (2017). Sensitivity, specificity, and predictive values: foundations, plabilities, and pitfalls in research and practice. *Front. Public Health* 5:307. doi: 10.3389/fpubh.2017.00307
- WHO (2019). Available online at: <https://icd.who.int/browse11/l-m/en#/http://id.who.int/icd/entity/1183832314> (accessed May 18, 2020).

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Copyright © 2020 O'Connor, Larsen, Joensen, Boelen, Maccallum, Komischke-Konnerup and Bryant. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.



Prolonged Grief Disorder Among Refugees in Psychological Treatment – Association With Self-Efficacy and Emotion Regulation

Oriane Lacour^{1*}, Naser Morina^{1†}, Julia Spaaij¹, Angela Nickerson², Ulrich Schnyder³, Roland von Känel¹, Richard A. Bryant² and Matthis Schick¹

¹ Department of Consultation-Liaison-Psychiatry and Psychosomatic Medicine, University Hospital, Zurich, University of Zurich, Zurich, Switzerland, ² School of Psychology, University of New South Wales, Sydney, NSW, Australia, ³ Faculty of Medicine, University of Zurich, Zurich, Switzerland

OPEN ACCESS

Edited by:

Geert E. Smid,
ARQ National Psychotrauma Centre,
Netherlands

Reviewed by:

Rolf J. Kleber,
Utrecht University, Netherlands
Carina Heeke,
Freie Universität Berlin, Germany
Nadine Stammel,
Freie Universität Berlin, Germany

*Correspondence:

Oriane Lacour
orlane.lacour@usz.ch

[†]These authors share first authorship

Specialty section:

This article was submitted to
Mood and Anxiety Disorders,
a section of the journal
Frontiers in Psychiatry

Received: 13 February 2020

Accepted: 22 May 2020

Published: 05 June 2020

Citation:

Lacour O, Morina N, Spaaij J,
Nickerson A, Schnyder U, von Känel R,
Bryant RA and Schick M (2020)
Prolonged Grief Disorder Among
Refugees in Psychological Treatment
—Association With Self-Efficacy and
Emotion Regulation.
Front. Psychiatry 11:526.
doi: 10.3389/fpsy.2020.00526

Background: While Prolonged Grief Disorder (PGD) among refugees has recently started to attract scientific attention, knowledge regarding associated psychological factors remains limited. Given the multifactorial context of persecution, trauma, displacement, and exile-related difficulties, obtaining a better understanding of PGD in refugees is crucial because PGD may affect psychological well-being, level of functioning, and social integration.

Methods: In a sample of refugees receiving psychological treatment in Switzerland ($N = 88$), we examined the relationship between severity of PGD and potentially associated factors such as emotion regulation, perceived self-efficacy, as well as potentially traumatic events and post-migration living difficulties.

Results: In a regression analysis, difficulties in emotion regulation and lower perceived self-efficacy were associated with greater severity of PGD, while post-migration living difficulties and potentially traumatic events did not emerge as significant factors.

Conclusion: These findings suggest that emotion regulation and perceived self-efficacy are associated with PGD in refugees in psychological treatment and are thus potential targets for treatment interventions.

Keywords: refugees, refugee mental health, prolonged grief disorder, self-efficacy, emotion regulation, trauma, post-migration living difficulties

INTRODUCTION

There are currently 70.8 million displaced people worldwide as a consequence of violence, persecution, conflict, or human rights violation (1). Forcibly displaced people frequently experience interpersonal loss and bereavement along with potentially traumatic events (PTE), including interpersonal violence, torture, life-threatening injuries and witnessing the death of loved

ones (2). Accordingly, mental health issues such as posttraumatic stress disorder (PTSD), depression or anxiety disorders are prevalent (3).

While trauma and trauma-related disorders in forcibly displaced populations have received consistent scientific attention, research on loss and grief in this group is scarce (3–5). Prolonged Grief Disorder (PGD) has recently been recognized as a clinically relevant mental health issue which will be included as a new diagnosis in the upcoming *International Classification of Diseases, 11th Revision* (ICD-11) (6). PGD, otherwise known as complicated grief (CG), refers to a distinct set of symptoms following the loss of a loved one. Those affected generally suffer from a persistent and pervasive longing for or preoccupation with the deceased, accompanied by emotional distress and significant functional impairment persisting more than six months after the loss. In the *Fifth Edition of the Diagnostic and Statistical Manual of Mental Disorder* (DSM-5), similar symptoms were classified as Persistent Complex Bereavement Disorder (PCBD). The latter differs from the former primarily with regard to the duration of symptoms (at least twelve months) (7). Due to the heterogeneous criteria used to assess CG, PGD, and PCBD in the past three decades, reported prevalence rates differ significantly. Yet, it is estimated that one out of ten bereaved individuals is at risk for disturbed grief following a nonviolent death (8) and that this risk is even higher after an unnatural (sudden, violent) death, affecting five out of ten bereaved individuals (9). Consequently, those individuals may experience substantial psychological distress, impairments in work and social functioning (7, 10, 11), and reduced quality of life (12, 13).

In refugees and post-conflicts samples, the prevalence of PGD has been estimated to be approx. 30% with important variation depending on cultural background (14, 15). Nevertheless, as only few studies with heterogeneous methodology have investigated PGD in refugees, the reported rates in this group also substantially differ from one another. Whereas earlier studies investigating PGD in refugees have focused on the associated comorbidity, traumatic experiences, and sociodemographic factors, as well as on cross-cultural adequacy of the concept, understanding of PGD and its associated factors is incomplete. As in the general population (7, 16, 17), both traumatic loss and violent loss have emerged as important predictors of PGD in refugees (18, 19). Similarly, strong associations with murder or disappearance of a family member have been described, as well as associations with other PTE such as torture or combat exposure (20). Nevertheless, the existence of a dose–response relationship between PTE exposure and PGD is currently inconclusive. Whereas exposure to a greater number of PTE might increase the likelihood of PGD diagnosis (20), some studies have found that trauma exposure is either not a significant predictor of PGD (21, 22) or not associated with PGD symptom severity (23). Furthermore, there seems to be no association between cumulated loss and PGD diagnosis (21, 22) or symptom severity (19), although multiple loss has been associated with anxiety, depression, disability, and worse quality of life in bereaved refugees (24).

While trauma seems to be related to PGD, it is likely that other contributing factors exist. For instance, nontraumatic stressors such as difficulties arising after arrival in a safe host country—referred to as Post-Migration Living Difficulties (PMLD)—might contribute to the likelihood of PGD diagnosis (20, 25, 26) and PGD symptom level (23). In addition, in nonrefugee samples, regulatory emotional–cognitive processes have been described as conclusive underlying mechanisms of adaptive coping with bereavement (27, 28). For example, negative cognitions (*i.e.* negative beliefs about self, world, life, future, others) and distressing interpretation of grief reactions (*e.g.* “If I let go of my emotions, I will go crazy”) have been found to be strongly related to higher concurrent and prospective levels of complicated grief symptoms, and to influence symptom severity over and above sociodemographic and loss-related variables (29, 30). Furthermore, these cognitive processes seem to be important correlates to emotional problems and maladaptive reactions to loss (30). On the other hand, factors such as optimism, active coping, and help seeking, as well as a supportive social environment, appear to be associated with less severe symptoms of PGD (31). Given the particular life circumstances refugees are confronted with, including displacement, exposure to multiple PTE and PMLD, it is unclear whether the findings from the general population can be generalized to refugees.

In view of the high prevalence of PGD in refugees and its potentially detrimental effects on psychological wellbeing, level of functioning, and social integration in the host country, it is crucial to acquire a better understanding of PGD to also provide appropriate treatment. This requires not only the assessment of risk factors, but also an exploration of protective factors. Surprisingly, the latter have not been emphasized in previous studies, even though they could be highly relevant for treatment interventions. Whereas risk factors of PGD such as loss-related characteristics cannot be modified through therapy, individual-related protective and adaptive factors could constitute promising treatment targets. This follows the idea that refugees and asylum seekers can benefit from resilience-focused interventions, along with trauma-focused interventions (32). The aim of this cross-sectional study was therefore to explore the relationship between the severity of PGD in a clinical sample of refugees and associated emotional and cognitive factors, specifically perceived self-efficacy and emotion regulation, with the hypothesis that they contribute to important mechanisms of adaptation in grief.

Perceived self-efficacy refers to positive beliefs about one's abilities to influence events that affect one's life. These beliefs may alter personal goals, motivation, everyday functioning, resilience to adversity, and vulnerability to distress and depression (33). The role of self-efficacy in adaptation after both a violent (34) and nonviolent loss of a loved one (35, 36) has been established in nonrefugee samples. Whereas self-efficacy has been considered a relevant variable among refugees due to its positive association with mental health and well-being (37, 38), its relationship to PGD remains unknown. Emotion regulation can be defined as one's ability to monitor, assess, and modulate

experienced emotions in order to facilitate adaptive functioning (39). In this study, we refer to emotion regulation as conceptualized by Gratz and Roemer and involving the following features: awareness, understanding and acceptance of emotions, as well as impulse control and ability to modulate emotional responses and behaviors according to desired goals and situational demands (39). As grief is characterized by a wide range of strong, often negative emotions, difficulties in emotion regulation or active avoidance of grief-related emotions might lead to maladaptive bereavement responses (11, 40). A small number of studies among refugees suggested that emotion regulation mediates the relationship between trauma and psychological symptoms (41) and that difficulties in emotion regulation are associated with more severe symptoms of PTSD, depression, and anxiety (42). Conversely, the relationship between emotion regulation and PGD has not yet been explored in refugees.

Based on this literature, we hypothesized that perceived self-efficacy and emotion regulation would be associated with the severity of PGD in refugees in the host country Switzerland. We included reported PMLD and PTE in our exploration as relevant variables among refugees and potential further associated factors.

METHODS

Participants

Participants were 88 refugees and asylum seekers receiving psychological treatment in two outpatient clinics for victims of torture and war in Bern and Zürich, Switzerland. At the time of inclusion, participants were at different stages of treatment. They were seeking treatment for varied psychopathology. Depending on their symptom profiles and subjective needs, treatment delivered included trauma-specific therapy, but also nonspecific psychotherapy, medication, and social counseling. Participants aged 18 or older and speaking either German, English, Turkish, Farsi, Tamil, or Arabic were included. Exclusion criteria were inability to use the tablet-based software used to collect data or inability to complete self-reported questionnaires, psychotic symptoms, severe dissociative symptoms, and acute suicidality. Based on the inclusion criteria, 106 patients were invited to participate in the study and were informed through a therapist and a study team member. In total, 88 patients (87%) agreed to participate. Reasons for rejecting were mainly: no interest and no time.

Measures

If available, validated instruments in the specific language were used. All other instruments were translated and back-translated by accredited translators in accordance with gold-standard translation practices (43). Discrepancies were rectified jointly by the research team and independent bilingual individuals who were experienced in working with health-related questionnaires.

PGD was assessed using the Prolonged Grief Disorder-13 (PG-13) scale based on the PGD-2009 criteria proposed by

Prigerson and colleagues (10). This scale contains 13 items: two yes–no items on duration and impairment and eleven items assessing cognitive, behavioral, and emotional symptoms, rated on a 5-point scale (either a frequency scale ranging from “not at all” to “several times a day”, or an intensity scale ranging from “not at all” to “overwhelmingly”). To assess the likelihood of PGD diagnosis five criteria must be considered: (A) event: the respondent has experienced the loss of a loved one; (B) separation distress: grief-related yearning is experienced at least daily; (C) duration: symptoms of separation distress remain at least 6 months after the loss; (D) cognitive, emotional, and behavioral symptoms: at least five symptoms based on items 4–11 are experienced at least “once a day” or “quite often”; (E) impairment: the respondent must have significant impairment in social, occupational, or other important areas of functioning. PG-13 can be scored as a continuous variable by summing the symptom items after excluding the two yes/no items about duration and functional impairment (44). This total score (range 11 to 55) was used as a severity score of PGD in the present study. To our knowledge, there are no universally recommended cut-off scores for clinical purposes. Internal consistency for this scale was $\alpha = 0.88$ in our sample.

Overall trauma exposure was assessed combining the Harvard Trauma Questionnaire (HTQ) (45) and the Posttraumatic Diagnostic Scale (46) resulting in a total of 23 items. Items include for example: “serious accident, fire or explosion”, “being close to death”, “imprisonment”. Potentially traumatic events experienced by each participant were summed up to a total count. In addition, loss-related characteristics were derived from the items “murder of a family member or friend” and “unnatural death of a family member or friend”.

The 17-items Post-Migration Living Difficulties Checklist (47, 48) adapted to the Swiss context (49) was used to assess PMLD. Items include for example: “communication difficulties”, “separation from the family”, “difficulties obtaining financial assistance”. Each item measures the presence and importance (0 = not a problem, to 4 = very serious problem) of post-migration stressors in the past 12 months. In the present study, items with a score of 2 or above (at least moderately serious problem) were summed up to a total count of PMLD.

Emotion regulation was assessed using the 18-items short form of the Difficulties in Emotion Regulation Scale (DERS) (39), which has proved to maintain the psychometric properties of the original 36-items measure (50, 51). This self-report measure encompasses several dimensions of emotion regulation: awareness, understanding, and acceptance of emotions, as well as impulse-control and ability to engage in goal-directed behaviors regardless of the emotional state. Items include for example: “I am attentive to my feelings”, “I have difficulties making sense out of my feelings”, “When I am upset, I have difficulty getting my work done”, or “When I am upset, I have difficulty controlling my behaviors”. Each item is rated on a 5-point scale (1 = almost never, 2 = sometimes, 3 = about half the time, 4 = most of the time, and 5 = almost always). In the original version of the scale, three out of 18 items require reverse coding.

However, previous experience with the scale indicated that reverse coding might have caused language difficulties (41). Thus, for items that were originally reverse coded, the wording was modified to avoid misunderstanding. We computed a mean of reported ratings ranging from 1 (“almost never”, indicating less difficulties in emotion regulation) to 5 (“almost always”, indicating more difficulties in emotion regulation). Internal consistency for this scale was $\alpha = 0.88$ in our sample.

Perceived self-efficacy was assessed using the General Self-Efficacy (GSE) scale, a 10-items psychometric instrument measuring optimistic self-beliefs to cope with a variety of difficult demands in life (52). Items include for example: “I can remain calm when facing difficulties because I can rely on my coping abilities” or “I can solve most problems if I invest the necessary effort”. Responses are given on a 4-point scale (1 = not at all true, 2 = hardly true, 3 = moderately true, 4 = exactly true) and summed up to a final score (range 10 to 40). The GSE has been widely used and validated internationally, including in a multicultural context (53). Internal consistency for this scale was $\alpha = 0.94$ in our sample.

Procedure

Data were collected between 2015 and 2016 through a computer-based assessment tool, the Multi-adaptive Psychological Screening Software (MAPPS) (54). In MAPPS, participants accessed questionnaire items in their mother tongue either in a written or auditory form, in case of illiteracy. The duration of the assessment was approximately 60 min. If needed, participants received assistance from a psychiatrist, a clinical psychologist or a masters-level student of clinical psychology. Participants received 40 CHF (approx. 40 USD) for participation.

Data Analysis

Statistical analysis was conducted using SPSS version 25. A regression analysis was performed with PGD severity score as dependent variable. Independent variables were defined *a priori* and entered into the model in two steps: first gender, age and length of stay in Switzerland as control variables, and then PTE and PMLD counts, and GSE and DERS scores as further independent variables. Pre-conditions for regression analyses were checked in terms of normal distribution of residuals, autocorrelation of residuals (Durbin-Watson test) and homoscedasticity and were found to be satisfying for PGD severity scores. Furthermore, none of the predictors showed signs of multicollinearity.

RESULTS

Sample Characteristics

The characteristics of the sample are summarized in **Table 1**. The participants were mainly married men from different countries of origin including Turkey for the majority, but also Iran, Sri Lanka, Iraq, Bosnia, Afghanistan, and others. They were aged 46 on average and had been in Switzerland for a mean time of 13 years. Their level of education was rather high, yet the majority

TABLE 1 | Sample characteristics (N = 88).

Sociodemographic Variables	n	%
Age, mean (SD)	46.8	(9.6)
Gender (male)	71	80.7
Length of stay (years), mean (SD)	12.9	(7.2)
Country of origin		
Turkey	52	59.1
Sri Lanka	8	9.1
Iran	6	6.8
Iraq	5	5.7
Afghanistan	3	3.4
Bosnia	3	3.4
Others	11	12.5
Marital Status		
Single	16	18.2
In a relationship	7	8
Married	47	53.4
Divorced	14	15.9
Widowed	4	4.5
Education		
Primary school partially attended or completed	28	31.8
High school partially attended or completed	32	36.4
Technical college	13	14.8
Bachelor's degree	8	9.1
Post graduate degree	7	8
Employment		
Full time	11	12.5
Part time	19	21.6
Unemployed	36	40.9
Pensioned/homemaker	22	25
Visa Status		
Asylum seeker	5	5.7
Temporary visa	8	9.1
Permanent visa	23	26.1
Residency	37	42
Citizenship	15	17
Clinical Variables		
Probable PGD diagnosis	21	23.9
PGD severity score (range 11–55), mean (SD)	34.8	(9.0)
PTE count, mean (SD)	12.8	(4.1)
PMLD count, mean (SD)	8.87	(4.1)
DERS score (range 1–5), mean (SD)	3.17	(0.6)
GSE score (range 10–40), mean (SD)	23.5	(6.6)

PGD, prolonged grief disorder; PTE, potentially traumatic events; PMLD, post-migration living difficulties; DERS, difficulties in emotion regulation scale; GSE, general self-efficacy.

was either unemployed or pensioned. The majority had a secure visa status.

Reported trauma exposure was high, with participants reporting having experienced an average of 13 PTE. The most commonly reported PTE included “torture” ($n = 80$, 91%), “nonsexual assault by a stranger” ($n = 69$, 78%) and “combat situation” ($n = 68$, 77%). Importantly, sudden or violent interpersonal loss was very common. In fact, while “murder of a family member or friend” was reported by 83% ($n = 73$) and “unnatural death of a family member or friend” by 69% ($n = 61$) of the participants, 97% ($n = 85$) reported at least one of the two.

The number of reported PMLD per participant was high, with a mean of nine living difficulties considered at least as a moderately serious problem. The most commonly reported

PMLD included “worry about family back home” ($n = 75$, 85%), “loneliness, boredom or isolation” ($n = 69$, 78%) and “being unable to return to your country in case of emergency” ($n = 68$, 77%).

Almost a quarter of the participants met the criteria for probable PGD ($n = 21$, 24%). The overall severity of PGD was intermediate, with a mean PG-13 score of 34.8 ($SD = 9.0$) (range 11 to 55).

Participants showed intermediate levels of emotion regulation, reporting difficulties on average about half the time, with a mean DERS score of 3.2 ($SD = 0.6$) (range 1 to 5). They showed rather low to intermediate perceived self-efficacy, with a GSE mean score of 23.5 ($SD = 6.7$) (range 10 to 40).

Regression Analysis

The results of the regression analysis for severity of PGD adjusted for age, gender, length of stay, number of PTE, number of PMLD, emotion regulation, and self-efficacy are shown in **Table 2**. The final model proved to be significant ($p < 0.001$) and accounted for 27% of the variance. Greater PGD severity was significantly associated with both a higher DERS score and a lower GSE score. In other words, greater difficulties in emotion regulation and lower perceived self-efficacy were predictive of greater PGD severity in the regression analysis. The other factors entered in this model did not emerge as significant predictors of PGD severity.

DISCUSSION

Primary Findings

This study examined the relationship between PGD severity and potential associated factors (emotion regulation, perceived self-efficacy, PMLD, PTE) in a clinical sample of refugees. The primary finding of our study was that difficulties in emotion regulation and lower perceived self-efficacy were associated with greater severity of PGD. In contrast, PTE and PMLD counts did not emerge as significant associated factors, supporting the idea

that PGD severity is independent of overall trauma exposure and difficulties arising after arrival in a host country.

PGD and Emotion Regulation

As PGD is by definition accompanied by intense emotional pain (e.g. sadness, guilt, anger, denial, and blame), it is not surprising that the ability to regulate emotions relates to symptom severity of PGD. In fact, previous models proposed to explain PGD symptoms have integrated emotion regulation as a main feature, along with attachment style, beliefs and appraisals, autobiographical memory, and self-identity (55–58). For instance, in Boelen and colleagues' cognitive behavioral model (56), maladaptive emotion regulation strategies defined by anxious and depressive avoidance are considered maintaining core factors of PGD symptoms. In other words, for individuals suffering from PGD, the internal emotional experience related to the loss (thoughts, feelings, memories) is perceived as so intolerable that they will rather avoid the reminders of the death and adopt behavioral patterns of inactivity or withdrawal. Similarly, in Maccallum and Bryant's cognitive attachment model (58), PGD symptoms are maintained by inflexible use of emotion regulation strategies avoiding the reality of the loss, the changed identity, and the new life situation.

Following these models, the participants in our study who showed more difficulties in understanding and accepting their emotions, as well as more difficulties modulating their emotional response and behaviors, also showed higher severity of PGD. Yet, the positive pendant of these findings is worth raising: participants with better understanding and acceptance of emotions and better modulation of emotion responses showed lower severity of PGD. Following this idea, there is reason to believe that an intervention based on awareness, observation, and nonjudgmental acceptance of emotional arousal may benefit both emotion regulation and executive control, and alleviate symptoms of grief, anxiety, and depression among bereaved individuals (59).

Although evidence about emotion regulation and PGD is limited, our findings are in line with a previous study suggesting an association between both positive and negative emotion

TABLE 2 | Summary of multiple regression analysis ($N = 88$).

Outcome variable	Step	Independent variable	B	SE	Beta	t	p	F	p	R2	adj. R2
PGD Severity	1	Age	0.04	0.12	0.41	0.32	0.748	1.18	0.323	0.04	0.006
		Gender	-4.06	2.46	-0.18	-1.65	0.102				
		Length of stay	0.05	0.16	0.04	0.32	0.751				
	2	Age	0.20	0.11	0.21	1.86	0.067	5.65	<0.001	0.33	0.272
		Gender	-2.68	2.23	-0.12	-1.20	0.232				
		Length of stay	-0.11	0.15	-0.08	-0.72	0.477				
		PTE count	-0.07	0.22	-0.03	-0.33	0.741				
		PMLD count	0.24	0.22	0.11	1.10	0.273				
		DERS score	5.28	1.49	0.35	3.53	0.001				
		GSE score	-0.45	0.13	-0.33	-3.47	0.001				

PGD, prolonged grief disorder; PTE, potentially traumatic events; PMLD, post-migration living difficulties; DERS, difficulties in emotion regulation scale; GSE, general self-efficacy.

regulation strategies, and symptoms levels of PGD, depression, and PTSD in people confronted with the death of a loved one, as well as in relatives of a long-term missing person (60). Furthermore, our findings call attention to emotion regulation as a relevant factor among refugees affected by PGD.

PGD and Self-Efficacy

The association found between self-efficacy and PGD severity in our study is consistent with previous research suggesting the role of self-efficacy in adaptation after the loss of a loved one. In a study on bereavement coping among widows, self-efficacy was found to be an important predictor of emotional distress, as well as of psychological, spiritual, and physical well-being in the year following the loss of a husband (36). In another study examining successful long-term adaptation to conjugal bereavement, bereaved individuals who expressed self-efficacy had lower levels of grief over time than those who did not. Importantly, the relationship between grief symptoms and the individuals' positive evaluation of their abilities was independent of the evaluation of their actual actions or personal characteristics (61). In the context of bereavement following mass violence, some authors found that severity of persistent grief was predicted by posttraumatic stress symptoms through reduced self-efficacy and disrupted world views, illustrating a relationship between PGD, traumatic stress and self-efficacy (62). Considering the evidence about the role of perceived self-efficacy in recovery from a wide range of traumatic experiences (63–65), self-efficacy might be particularly important in traumatized refugees.

PGD and Trauma Exposure

Consistent with other studies on PGD after war and displacement, the number of experienced PTE was not associated with PGD severity (21–23). However, slightly different findings were reported in a large population-based study among refugees in Australia, which found that the likelihood of PGD diagnosis was heightened as PTE exposure increased (20).

As in the nonrefugee population, the loss of a loved one by traumatic death clearly appears to be a major risk factor for developing PGD in refugees and conflicts affected populations (5, 18–20, 25, 26). In fact, the majority of the participants in our sample had experienced the murder or unnatural death of a close one. It is important to acknowledge that refugees are almost per definition exposed to repeated violence and PTE. Yet, when exploring trauma and grief in this group, it might be worth distinguishing the traumatic experience linked to the violent nature of the loss, which clearly seems to influence PGD, from the overall traumatic experiences, which may not. In other words, it is more likely to be the type of trauma that increases the risk of PGD (e.g., “witnessing the death of a loved one”), rather than the number of traumas experienced. Trauma checklists (such as the one used in this study) measure the *diversity* of trauma experiences. When creating a count variable, each trauma type is then equally weighted (i.e., “witnessing the murder of a loved one” is weighted the same as “lack of shelter”).

This probably reduces the likelihood of finding a dose–response relationship between PTE exposure and PGD.

PGD and Post-Migration Living Difficulties

Despite the preliminary evidence for the causal role of PMLD with regard to mental health in refugees and asylum seekers (49, 66–68) and despite the high amount of reported PMLD in our sample, no association was found between the number of PMLD and PGD severity. However, evidence regarding PMLD and PGD is scarce.

According to previous studies, some specific PMLD such as adaptation difficulties, experiencing discrimination and being in receipt of government benefits, might be related to PGD diagnosis, yet there seems to be no significant relationship with the total number of PMLD (20, 26). Similarly, a study among asylum seekers in Germany showed that permanent visa status was associated with less severe PGD symptoms (23), yet the relationship between other PMLD and PGD was not explored. Finally, one study among West Papua refugees displaced in Papua New Guinea suggested an increased vulnerability for PGD due to stressful living conditions including food and water shortage, geographical isolation, and lack of services, as well as fear of hostile incursion from across the nearby border. This last example reminds us that PMLD may vary a lot from one host country to another, depending on the social, political, and economic context. For instance, the majority of refugees in our sample had a secure visa status and were *a priori* concerned by loneliness and boredom or by the situation in their country of origin and for their family rather than by fear for their immediate safety or vital needs.

Thoughts on the Prevalence of PGD

Our study further revealed that nearly one out of four participants fulfilled the diagnostic criteria for PGD. Although the time elapsed since their loss was unknown, it is likely that they had been experiencing grief-related symptoms for several years, as the participants in our sample had been in Switzerland for a mean of 13 years. This finding is important for clinical practice as it highlights that special attention should be paid to grief-related symptoms in refugees seeking psychological treatment, even several years after arrival in their host country.

Compared to the current literature, the prevalence of PGD in our sample appeared to be rather low. Nevertheless, the limited number of studies on PGD among refugees and the heterogeneity of PGD assessment make it difficult to compare results. For example, a systematic review among adult refugees exposed to bereavement and trauma reported a prevalence of PGD of 33.2% (range 15–54) (14). Yet, only four highly heterogeneous studies were included in the meta-analysis on prevalence, indicating that these results should be interpreted with caution. This issue was also highlighted in a review of 24 studies that assessed grief disorder in refugees and post-conflict samples (15). The authors reported highly diverse rates of PGD depending on the standard measures employed, with an average of 32% (range 8–69) of participants scoring in the disordered or

severe range. Furthermore, they reported that studies using culturally adapted assessment approaches revealed significant levels of culturally specific symptoms and even higher rates of disordered grief (range 31–76%).

As participants in our sample came from several countries of origin, it was not possible to adapt our PGD measure to each specific cultural background. This bias may have lowered our prevalence rate. Moreover, the PG-13 scale used in our study does not allow for clinical judgement regarding the expected social, cultural or religious norms in an individual's context. A caveat stating that the grief-related symptoms must clearly exceed sociocultural norms has recently been added to the ICD-11 criteria, which otherwise share the core features of the PGD-2009 criteria (6).

Previous studies using the PG-13 scale among refugees and conflict-affected populations have reported rates of PGD ranging from 8 to 34.6% (19, 23, 69–71). Again, a comparison is difficult as respective study samples differ substantially from ours in terms of age, gender, or setting.

Further Findings

Interestingly, the majority of the participants in our sample were male. This is in contrast with most studies on PGD in adults and in particular those on the validation of the criteria for PGD, which include a high proportion of female (10, 72). As expected, based on previous studies in refugees and conflict survivors (19, 23), PGD severity was independent from gender in our model. Yet considering that the female might be at higher risk for PGD following violent loss (73), the high amount of male in our sample might have lowered the prevalence of probable PGD diagnosis. Furthermore, gender-specific processes or social norms might influence the way that individuals express grief symptoms (74, 75). These aspects need further exploration.

Clinical Implications

Considering the high prevalence of PGD among refugees and its association with psychological comorbidity and functional impairment (20, 71), early detection and adequate treatment are essential. Left untreated, individuals suffering from PGD might remain with high levels not only of psychological (20, 76), but also of physical distress (69, 70, 73), as well as with an increased risk of suicide (70, 73, 77).

As psychological impairment may severely compromise the already challenging process of social integration in a host country, including language proficiency and financial independence (49), untreated PGD might represent not only an individual inconvenience but also an economic burden for host societies. A recent large population study, however, found that almost half of the refugees with probable PGD had not received psychological assistance (20). Explanations proposed by the authors were not only often-low help seeking among people with PGD and avoidance of confrontation to grief-related emotions, but also low mental health care utilization by refugees possibly due to stigma or lack of knowledge about referral opportunities. In the Swiss context, socio-cultural and structural barriers to accessing mental health care by refugees and asylum-seekers may include language difficulties, problems

related to medical or authority gatekeepers, lack of resources, lack of awareness about health, fear of stigma, and mismatch between the local health system and perceived needs of refugees and asylum seekers (78). Over and above addressing these barriers to psychological assessment and treatment in general, our findings underline the importance of independent grief assessment, which is often not included in screening instruments for refugees (79, 80). Despite the confounding effect of experiencing both traumatic events and interpersonal loss, refugees and conflict-affected populations seem to present distinctive symptom profiles of PGD and/or PTSD (18, 21, 26).

Another implication of our findings is that they highlight potential directions for treatment interventions. Current effective treatment of PGD relies on Cognitive Behavioral Therapy (CBT), along with exposure techniques (4, 81–83). Such techniques rely on the hypothesis that directing patients to access their emotional memories can reduce avoidance of key-memories of the deceased and facilitate adaptation by restructuring core cognitions that may complicate grief response (4). Though interventions are available, knowledge about underlying factors predicting treatment response is limited (84). Moreover, these interventions have not been tested among refugees.

Given the finding that difficulties in emotion regulation and lower perceived self-efficacy are associated with greater PGD severity, it may be useful to apply techniques focusing on improving emotion regulation and perceived self-efficacy in the treatment of PGD. Such approaches have already been examined in the treatment of trauma-related disorders. For example, the Skills Training in Affective and Interpersonal Regulation (STAIR) Narrative Therapy, an intervention focusing on emotion regulation and interpersonal skills to promote resilience, has shown promising results in alleviating trauma-related distress and improving emotional and social impairments among trauma survivors (85). Similarly, refugee torture survivors showed greater distress tolerance on trauma reminders after receiving an intervention aiming to enhance perceived self-efficacy (38). The potential effects of such interventions on PGD in refugees should be targeted by future studies.

Limitations

This study has several limitations. First, the relatively small sample size may have reduced the statistical power of the analysis. Second, the use of self-report measures may have led to several biases (e.g. interpretation of the question, introspective ability, honesty of response), whereas standardized clinical interviews could have strengthened our results. Third, as participants came from various cultural backgrounds, it was not possible to use measures validated among all cultural groups. In particular, it was not possible to use a culturally adapted approach when assessing PGD. Fourth, the cross-sectional design of the study prevents establishing any causality between perceived self-efficacy, emotion regulation and PGD. Fifth, information on loss-related characteristics was sparse, which limits the interpretation of the findings. Sixth, due to the clinical nature of our sample, interference of our findings with psychotherapeutic and pharmacological treatment cannot be ruled out. Moreover, as our findings were based on refugees in

psychological treatment, they may not be extendable to the general refugee population.

CONCLUSIONS

This study is one of the very few investigating PGD among refugees residing in western countries. Our findings highlight the relationship between both emotion regulation and perceived self-efficacy and the severity of PGD among refugees receiving psychological treatment in Switzerland. We consider the concepts of perceived self-efficacy and emotion regulation particularly relevant for clinical practice as they relate to subsequent behaviors and may constitute key targets for treatment intervention. Enhancing perceived self-efficacy and emotion regulation may be useful to improve treatment response of PGD in treatment-seeking refugees. Such intervention effects should be examined in future studies.

DATA AVAILABILITY STATEMENT

The datasets generated for this study are available on request to the corresponding author.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by KEK-ZH-Nr. 2011-0495 and KEK-BE_Nr. 152/12. The patients/participants provided their written informed consent to participate in this study.

REFERENCES

1. UNHCR. Figures at a Glance. (2019). Available from: <https://www.unhcr.org/figures-at-a-glance.html> [Accessed April 12, 2020].
2. Nickerson A, Liddell B, Asnaani A, Carlsson JM, Fazel M, Knaevelsrud C, et al. Trauma and Mental Health in Forcibly Displaced Populations: An International Society for Traumatic Stress Studies Briefing Paper. *ISTSS International Society for Traumatic Stress Studies*. (2017). Available from: <https://istss.org/public-resources/istss-briefing-papers/trauma-and-mental-health-in-forcibly-displaced-pop>.
3. Morina N, Akhtar A, Barth J, Schnyder U. Psychiatric Disorders in Refugees and Internally Displaced Persons After Forced Displacement: A Systematic Review. *Front Psychiatry* (2018) 9:433. doi: 10.3389/fpsy.2018.00433
4. Bryant RA, Kenny L, Joscelyne A, Rawson N, Maccallum F, Cahill C, et al. Treating prolonged grief disorder: a randomized clinical trial. *JAMA Psychiatry* (2014) 71(12):1332–9. doi: 10.1001/jamapsychiatry.2014.1600
5. Tay AK, Mohsin M, Rees S, Tam N, Kareth M, Silove D. The structure and psychosocial correlates of complicated bereavement amongst refugees from West Papua. *Soc Psychiatry Psychiatr Epidemiol* (2019) 54(6):771–80. doi: 10.1007/s00127-019-01666-1
6. Kilikelly C, Maercker A. Prolonged grief disorder for ICD-11: the primacy of clinical utility and international applicability. *Eur J Psychotraumatol* (2018) 8 (Suppl 6):1476441–. doi: 10.1080/2008198.2018.1476441
7. Nakajima S. Complicated grief: recent developments in diagnostic criteria and treatment. *Phil Trans R Soc B* (2018) 373:20170273. doi: 10.1098/rstb.2017.0273
8. Lundorff M, Holmgren H, Zachariae R, Farver-Vestergaard I, O'Connor M. Prevalence of prolonged grief disorder in adult bereavement: A systematic review and meta-analysis. *J Affect Disord* (2017) 212:138–49. doi: 10.1016/j.jad.2017.01.030
9. Djelantik A, Smid GE, Mroz A, Kleber RJ, Boelen PA. The prevalence of prolonged grief disorder in bereaved individuals following unnatural losses: Systematic review and meta regression analysis. *J Affect Disord* (2020) 265:146–56. doi: 10.1016/j.jad.2020.01.034
10. Prigerson HG, Horowitz MJ, Jacobs SC, Parkes CM, Aslan M, Goodkin K, et al. Prolonged grief disorder: Psychometric validation of criteria proposed for DSM-V and ICD-11. *PLoS Med* (2009) 6(8):e1000121. doi: 10.1371/journal.pmed.1000121
11. Shear MK, Simon N, Wall M, Zisook S, Neimeyer R, Duan N, et al. Complicated grief and related bereavement issues for DSM-5. *Depression Anxiety* (2011) 28(2):103–17. doi: 10.1002/da.20780
12. Boelen PA, Prigerson HG. The influence of symptoms of prolonged grief disorder, depression, and anxiety on quality of life among bereaved adults: a prospective study. *Eur Arch Psychiatry Clin Neurosci* (2007) 257(8):444–52. doi: 10.1007/s00406-007-0744-0
13. Silverman GK, Jacobs SC, Kasl SV, Shear MK, Maciejewski PK, Noaghiul FS, et al. Quality of life impairments associated with diagnostic criteria for traumatic grief. *Psychol Med* (2000) 30(4):857–62. doi: 10.1017/s0033291799002524
14. Kokou-Kpolou CK, Moukouta CS, Masson J, Bernoussi A, Cénat JM, Bacqué M-F. Correlates of grief-related disorders and mental health outcomes among adult refugees exposed to trauma and bereavement: A systematic review and

AUTHOR CONTRIBUTIONS

OL was involved in the analysis and interpretation of data, the drafting and the revision of the manuscript. MS and NM were involved in the conception and design of the study, the acquisition, analysis, and interpretation of data, and the drafting and revision of the manuscript. JS was involved in the analysis and interpretation of data and contributed to the manuscript. RK was involved in the drafting and revision of the manuscript. AN, RB, and US designed the study and contributed to the manuscript. All authors read and approved the final manuscript.

FUNDING

The study was supported by the Parrotia Foundation, the Swiss Foundation for the Promotion of Psychiatry and Psychotherapy, the Swiss Federal Office for Migration (3a-12-0495), and the Swiss Federal Office for Health (12.005187). Angela Nickerson was supported by a National Health and Medical Research Council Clinical Early Career Fellowship (1037091). The funding source played no role in the choice of the study design, in the collection, analysis, and interpretation of data, in the writing of the report, or in the decision of submitting the paper for publication.

ACKNOWLEDGMENTS

We thank all the participating patients, assessors, research assistants, and therapists involved in the project.

- future research directions. *J Affect Disord* (2020) 267:171–84. doi: 10.1016/j.jad.2020.02.026
15. Killikelly C, Bauer S, Maercker A. The Assessment of Grief in Refugees and Post-conflict Survivors: A Narrative Review of Etic and Emic Research. *Front Psychol* (2018) 9:1957. doi: 10.3389/fpsyg.2018.01957
 16. Shear MK. Clinical practice. Complicated grief. *New Engl J Med* (2015) 372(2):153–60. doi: 10.1056/NEJMcpl315618
 17. Neria Y, Gross R, Litz B, Maguen S, Insel B, Seirmarco G, et al. Prevalence and psychological correlates of complicated grief among bereaved adults 2.5–3.5 years after September 11th attacks. *J Traumatic Stress* (2007) 20(3):251–62. doi: 10.1002/jts.20223
 18. Momartin S, Silove D, Manicavasagar V, Steel Z. Complicated grief in Bosnian refugees: associations with posttraumatic stress disorder and depression. *Compr Psychiatry* (2004) 45(6):475–82. doi: 10.1016/j.comppsy.2004.07.013
 19. Schaal S, Jacob N, Dusingizemungu JP, Elbert T. Rates and risks for prolonged grief disorder in a sample of orphaned and widowed genocide survivors. *BMC Psychiatry* (2010) 10:55. doi: 10.1186/1471-244x-10-55
 20. Bryant RA, Edwards B, Creamer M, O'Donnell M, Forbes D, Felmingham KL, et al. A population study of prolonged grief in refugees. *Epidemiol Psychiatr Sci* (2019) 29:1–7. doi: 10.1017/s2045796019000386
 21. Morina N, Rudari V, Bleichhardt G, Prigerson HG. Prolonged grief disorder, depression, and posttraumatic stress disorder among bereaved Kosovar civilian war survivors: a preliminary investigation. *Int J Soc Psychiatry* (2010) 56(3):288–97. doi: 10.1177/0020764008101638
 22. Stammel N, Heeke C, Bockers E, Chhim S, Taing S, Wagner B, et al. Prolonged grief disorder three decades post loss in survivors of the Khmer Rouge regime in Cambodia. *J Affect Disord* (2013) 144(1–2):87–93. doi: 10.1016/j.jad.2012.05.063
 23. Comtesse H, Rosner R. Prolonged grief disorder among asylum seekers in Germany: the influence of losses and residence status. *Eur J Psychotraumatol* (2019) 10(1):1591330. doi: 10.1080/2008198.2019.1591330
 24. Hengst SMC, Smid GE, Laban CJ. The Effects of Traumatic and Multiple Loss on Psychopathology, Disability, and Quality of Life in Iraqi Asylum Seekers in the Netherlands. *J Nervous Ment Dis* (2018) 206(1):52–60. doi: 10.1097/nmd.0000000000000750
 25. Tay AK, Rees S, Tam N, Kareth M, Silove D. Defining a combined constellation of complicated bereavement and PTSD and the psychosocial correlates associated with the pattern amongst refugees from West Papua. *psychol Med* (2019) 49(9):1481–9. doi: 10.1017/s0033291718002027
 26. Nickerson A, Liddell BJ, Maccallum F, Steel Z, Silove D, Bryant RA. Posttraumatic stress disorder and prolonged grief in refugees exposed to trauma and loss. *BMC Psychiatry* (2014) 14:106. doi: 10.1186/1471-244x-14-106
 27. Stroebe M, Schut H, Boerner K. Models of coping with bereavement: an updated overview / Modelos de afrontamiento en duelo: un resumen actualizado. *Stud Psychol* (2017) 38(3):582–607. doi: 10.1080/02109395.2017.1340055
 28. Stroebe MS, Schut H. *Models of coping with bereavement: A review. Handbook of bereavement research: Consequences, coping, and care*. Washington, DC, US: American Psychological Association (2001) p. 375–403.
 29. Boelen PA, van den Bout J, van den Hout MA. Negative cognitions and avoidance in emotional problems after bereavement: a prospective study. *Behav Res Ther* (2006) 44(11):1657–72. doi: 10.1016/j.brat.2005.12.006
 30. Boelen PA, van den Bout J, van den Hout MA. The role of cognitive variables in psychological functioning after the death of a first degree relative. *Behav Res Ther* (2003) 41(10):1123–36. doi: 10.1016/s0005-7967(02)00259-0
 31. Lobb EA, Kristjanson LJ, Aoun SM, Monterosso L, Halkett GK, Davies A. Predictors of complicated grief: a systematic review of empirical studies. *Death Stud* (2010) 34(8):673–98. doi: 10.1080/07481187.2010.496686
 32. Laban C. Resilience-Oriented Treatment of Traumatized Asylum Seekers and Refugees. *Trauma and Migration: Cultural Factors in the Diagnosis and Treatment of Traumatized Immigrants* (2015). p. 191–208. doi: 10.1007/978-3-319-17335-1_13
 33. Bandura. Self-efficacy. *Encyclopedia of human behavior*, vol. 4 (1994). p. 71–81.
 34. Benight CC, Freyaldenhoven RW, Hughes J, Ruiz JM, Zoschke TA, Lovullo WR. Coping self-efficacy and psychological distress following the Oklahoma City bombing. *J Appl Soc Psychol* (2000) 30(7):1331–44. doi: 10.1111/j.1559-1816.2000.tb02523.x
 35. Bauer JJ, Bonanno GA. Doing and Being Well (for the Most Part): Adaptive Patterns of Narrative Self-Evaluation During Bereavement. *J Pers* (2001) 69(3):451–82. doi: 10.1111/1467-6494.00152
 36. Benight C, Flores J, Tashiro T. Bereavement coping self-efficacy in cancer widows. *Death Stud* (2001) 25:97–125. doi: 10.1080/07481180125921
 37. Sulaiman-Hill CMR, Thompson SC. Learning to Fit in: An Exploratory Study of General Perceived Self Efficacy in Selected Refugee Groups. *J Immigrant Minority Health* (2013) 15(1):125–31. doi: 10.1007/s10903-011-9547-5
 38. Morina N, Bryant RA, Doolan EL, Martin-Solch C, Plichta MM, Pfaltz MC, et al. The impact of enhancing perceived self-efficacy in torture survivors. *Depression Anxiety* (2018) 35(1):58–64. doi: 10.1002/da.22684
 39. Gratz K, Roemer L. Multidimensional Assessment of Emotion Regulation and Dysregulation: Development, Factor Structure, and Initial Validation of the Difficulties in Emotion Regulation Scale. *J Psychopathol Behav Assess* (2004) 26:41–54. doi: 10.1007/s10862-008-9102-4
 40. Simon NM. Treating complicated grief. *Jama* (2013) 310(4):416–23. doi: 10.1001/jama.2013.8614
 41. Nickerson A, Bryant RA, Schnyder U, Schick M, Mueller J, Morina N. Emotion dysregulation mediates the relationship between trauma exposure, post-migration living difficulties and psychological outcomes in traumatized refugees. *J Affect Disord* (2015) 173:185–92. doi: 10.1016/j.jad.2014.10.043
 42. Koch T, Liedl A, Ehring T. Emotion regulation as a transdiagnostic factor in Afghan refugees. *psychol Trauma : theory Res Pract Policy* (2019) 12(3):235–43. doi: 10.1037/tra0000489
 43. Bontempo R. Translation Fidelity of Psychological Scales: An Item Response Theory Analysis of an Individualism-Collectivism Scale. *J Cross-Cultural Psychol* (1993) 24(2):149–66. doi: 10.1177/0022022193242002
 44. Pohlkamp L, Kreibergs U, Prigerson HG, Sween J. Psychometric properties of the Prolonged Grief Disorder-13 (PG-13) in bereaved Swedish parents. *Psychiatry Res* (2018) 267:560–5. doi: 10.1016/j.psychres.2018.06.004
 45. Mollica RF, Caspi-Yavin Y, Bollini P, Truong T, Tor S, Lavelle J. The Harvard Trauma Questionnaire. Validating a cross-cultural instrument for measuring torture, trauma, and posttraumatic stress disorder in Indochinese refugees. *J Nervous Ment Dis* (1992) 180(2):111–6. doi: 10.1097/00005053-199202000-00008
 46. Foa EB, Cashman L, Jaycox L, Perry K. The validation of a self-report measure of posttraumatic stress disorder: The Posttraumatic Diagnostic Scale. *psychol Assess* (1997) 9(4):445–51. doi: 10.1037/1040-3590.9.4.445
 47. Silove D, Sinnerbrink I, Field A, Manicavasagar V, Steel Z. Anxiety, depression and PTSD in asylum-seekers: associations with pre-migration trauma and post-migration stressors. *Br J Psychiatry : J Ment Sci* (1997) 170:351–7. doi: 10.1192/bjp.170.4.351
 48. Steel Z, Silove D, Bird K, McGorry P, Mohan P. Pathways from war trauma to posttraumatic stress symptoms among Tamil asylum seekers, refugees, and immigrants. *J Traumatic Stress* (1999) 12(3):421–35. doi: 10.1023/a:1024710902534
 49. Schick M, Zumwald A, Knopfli B, Nickerson A, Bryant RA, Schnyder U, et al. Challenging future, challenging past: the relationship of social integration and psychological impairment in traumatized refugees. *Eur J Psychotraumatol* (2016) 7:28057. doi: 10.3402/ejpt.v7.28057
 50. Kaufman EA, Xia M, Fosco G, Yaptangco M, Skidmore CR, Crowell SE. The Difficulties in Emotion Regulation Scale Short Form (DERS-SF): Validation and replication in adolescent and adult samples. *J Psychopathol Behav Assess* (2016) 38(3):443–55. doi: 10.1007/s10862-015-9529-3
 51. Victor SE, Klonsky ED. Validation of a brief version of the Difficulties in Emotion Regulation Scale (DERS-18) in five samples. *J Psychopathol Behav Assess* (2016) 38(4):582–9. doi: 10.1007/s10862-016-9547-9
 52. Schwarzer R, Jerusalem M. “Generalized Self-Efficacy scale”. In: J. Weinman S, Wright M, Johnston, *Measures in health psychology: A user's portfolio. Causal and control beliefs*. Windsor, England: NFER-NELSON (1995). p. 35–7.
 53. Luszczynska A, Scholz U, Schwarzer R. The General Self-Efficacy Scale: Multicultural Validation Studies. *J Psychol* (2005) 139(5):439–57. doi: 10.3200/JRLP.139.5.439-457
 54. Morina N, Ewers SM, Passardi S, Schnyder U, Knaevelsrud C, Müller J, et al. Mental health assessments in refugees and asylum seekers: evaluation of a tablet-assisted screening software. *Conflict Health* (2017) 11(1):18. doi: 10.1186/s13031-017-0120-2

55. Shear K, Monk T, Houck P, Melhem N, Frank E, Reynolds C, et al. An attachment-based model of complicated grief including the role of avoidance. *Eur Arch Psychiatry Clin Neurosci* (2007) 257(8):453–61. doi: 10.1007/s00406-007-0745-z
56. Boelen PA, Van Den Hout MA, Van Den Bout J. A Cognitive-Behavioral Conceptualization of Complicated Grief. *Clin Psychol Sci* (2006) 13: (2):109–28. doi: 10.1111/j.1468-2850.2006.00013.x
57. Stroebe Ms, Schut HAW. The dual process model of coping with bereavement: Overview and update. *Death Stud* (1999) 23:197–224. doi: 10.1080/074811899201046
58. Maccallum F, Bryant RA. A Cognitive Attachment Model of prolonged grief: integrating attachments, memory, and identity. *Clin Psychol Rev* (2013) 33 (6):713–27. doi: 10.1016/j.cpr.2013.05.001
59. Huang F-Y, Hsu A-L, Hsu L-M, Tsai J-S, Huang C-M, Chao Y-P, et al. Mindfulness Improves Emotion Regulation and Executive Control on Bereaved Individuals: An fMRI Study. *Front Hum Neurosci* (2019) 12:541–. doi: 10.3389/fnhum.2018.00541
60. Lenferink LIM, Wessel I, Boelen PA. Exploration of the Associations Between Responses to Affective States and Psychopathology in Two Samples of People Confronted With the Loss of a Loved One. *J Nervous Ment Dis* (2018) 206 (2):108–15. doi: 10.1097/nmd.0000000000000781
61. Bauer JJ, Bonanno GA. I Can, I Do, I Am: The Narrative Differentiation of Self-Efficacy and Other Self-Evaluations while Adapting to Bereavement. *J Res Pers* (2001) 35(4):424–48. doi: 10.1006/jrpe.2001.2323
62. Smith A, Abeyta A, Hughes M, Jones R. Persistent Grief in the Aftermath of Mass Violence: The Predictive Roles of Posttraumatic Stress Symptoms, Self-Efficacy, and Disrupted Worldview. *psychol Trauma Theory Res Pract Policy* (2015) 7:179–86. doi: 10.1037/tra0000002
63. Benight CC, Bandura A. Social cognitive theory of posttraumatic recovery: the role of perceived self-efficacy. *Behav Res Ther* (2004) 42(10):1129–48. doi: 10.1016/j.brat.2003.08.008
64. Luszczynska A, Benight C, Cieslak R. Self-Efficacy and Health-Related Outcomes of Collective Trauma A Systematic Review. *Eur Psychol* (2009) 14:51–62. doi: 10.1027/1016-9040.14.1.51
65. Nygaard E, Johansen VA, Siqveland J, Hussain A, Heir T. Longitudinal Relationship between Self-efficacy and Posttraumatic Stress Symptoms 8 Years after a Violent Assault: An Autoregressive Cross-Lagged Model. *Front Psychol* (2017) 8:913–. doi: 10.3389/fpsyg.2017.00913
66. Laban CJ, Gernaat HB, Komproe IH, van der Tweel I, De Jong JT. Postmigration living problems and common psychiatric disorders in Iraqi asylum seekers in the Netherlands. *J Nervous Ment Dis* (2005) 193(12):825–32. doi: 10.1097/01.nmd.0000188977.44657.1d
67. Li S, Liddell B, Nickerson A. The Relationship Between Post-Migration Stress and Psychological Disorders in Refugees and Asylum Seekers. *Curr Psychiatry Rep* (2016) 18:82. doi: 10.1007/s11920-016-0723-0
68. Schick M, Morina N, Mistridis P, Schnyder U, Bryant RA, Nickerson A. Changes in Post-migration Living Difficulties Predict Treatment Outcome in Traumatized Refugees. *Front Psychiatry* (2018) 9:476. doi: 10.3389/fpsyg.2018.00476
69. Hinton DE, Nickerson A, Bryant RA. Prolonged grief in Cambodian refugees following genocide: Rebirth concerns and avoidance of reminders. *J Loss Trauma* (2013) 18(5):444–60. doi: 10.1080/15325024.2012.714218
70. Morina N, von Lersner U, Prigerson HG. War and bereavement: consequences for mental and physical distress. *PloS One* (2011) 6(7): e22140. doi: 10.1371/journal.pone.0022140
71. Steil R, Gutermann J, Harrison O, Starck A, Schwartzkopff L, Schouler-Ocak M, et al. Prevalence of prolonged grief disorder in a sample of female refugees. *BMC Psychiatry* (2019) 19(1):148. doi: 10.1186/s12888-019-2136-1
72. Boelen PA, Lenferink LIM, Nickerson A, Smid GE. Evaluation of the factor structure, prevalence, and validity of disturbed grief in DSM-5 and ICD-11. *J Affect Disord* (2018) 240:79–87. doi: 10.1016/j.jad.2018.07.041
73. Heeke C, Kampisiou C, Niemeyer H, Knaevelsrud C. A systematic review and meta-analysis of correlates of prolonged grief disorder in adults exposed to violent loss. *Eur J Psychotraumatol* (2019) 10:1583524. doi: 10.1080/20008198.2019.1583524
74. Creighton G, Oliffe JL, Butterwick S, Saewyc E. After the death of a friend: young men's grief and masculine identities. *Soc Sci Med* (1982) (2013) 84:35–43. doi: 10.1016/j.socscimed.2013.02.022
75. Stelzer E-M, Atkinson C, O'Connor M-F, Croft A. Gender differences in grief narrative construction: a myth or reality? *Eur J Psychotraumatol* (2019) 10 (1):1688130. doi: 10.1080/20008198.2019.1688130
76. Craig CD, Sossou M-A, Schnak M, Essex H. Complicated grief and its relationship to mental health and well-being among Bosnian refugees after resettlement in the United States: Implications for practice, policy, and research. *Traumatology* (2008) 14(4):103–15. doi: 10.1177/1534765608322129
77. Latham AE, Prigerson HG. Suicidality and bereavement: complicated grief as psychiatric disorder presenting greatest risk for suicidality. *Suicide Life-Threatening Behav* (2004) 34(4):350–62. doi: 10.1521/suli.34.4.350.53737
78. Kiselev N, Pfaltz M, Haas F, Schick M, Kappen M, Sijbrandij M, et al. Structural and socio-cultural barriers to accessing mental healthcare among Syrian refugees and asylum seekers in Switzerland. *Eur J Psychotraumatol* (2020) 11(1):1717825. doi: 10.1080/20008198.2020.1717825
79. Hocking DC, Mancuso SG, Sundram S. Development and validation of a mental health screening tool for asylum-seekers and refugees: the STAR-MH. *BMC Psychiatry* (2018) 18(1):69. doi: 10.1186/s12888-018-1660-8
80. Kaltenbach E, Härdtner E, Hermenau K, Schauer M, Elbert T. Efficient identification of mental health problems in refugees in Germany: the Refugee Health Screener. *Eur J Psychotraumatol* (2017) 8(sup2):1389205–. doi: 10.1080/20008198.2017.1389205
81. Bryant RA, Kenny L, Joscelyne A, Rawson N, Maccallum F, Cahill C, et al. Treating Prolonged Grief Disorder: A 2-Year Follow-Up of a Randomized Controlled Trial. *J Clin Psychiatry* (2017) 78(9):1363–8. doi: 10.4088/JCP.16m10729
82. Rosner R, Bartl H, Pfoh G, Kotoucova M, Hagl M. Efficacy of an integrative CBT for prolonged grief disorder: A long-term follow-up. *J Affect Disord* (2015) 183:106–12. doi: 10.1016/j.jad.2015.04.051
83. Boelen PA, de Keijser J, van den Hout MA, van den Bout J. Treatment of complicated grief: a comparison between cognitive-behavioral therapy and supportive counseling. *J Consult Clin Psychol* (2007) 75(2):277–84. doi: 10.1037/0022-006x.75.2.277
84. Bryant RA, Kenny L, Joscelyne A, Rawson N, Maccallum F, Cahill C, et al. Predictors of treatment response for cognitive behaviour therapy for prolonged grief disorder. *Eur J Psychotraumatol* (2017) 8(6):1556551. doi: 10.1080/20008198.2018.1556551
85. Hassija C, Cloitre M. STAIR Narrative Therapy: A Skills Focused Approach to Trauma-Related Distress. *Curr Psychiatry Rev* (2015) 11:172–9. doi: 10.2174/1573400511666150629105544

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

The handling editor declared a past collaboration with one of the authors, AN.

Copyright © 2020 Lacour, Morina, Spaaij, Nickerson, Schnyder, von Känel, Bryant and Schick. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.



Web-Based Bereavement Care: A Systematic Review and Meta-Analysis

Birgit Wagner^{1*}, Nicole Rosenberg¹, Laura Hofmann¹ and Ulrike Maass²

¹ Clinical Psychology and Psychotherapy, Medical School Berlin, Berlin, Germany, ² Clinical Psychology and Psychotherapy, University of Potsdam, Potsdam, Germany

OPEN ACCESS

Edited by:

Roumen Milev,
Queen's University, Canada

Reviewed by:

Rolf J. Kleber,
Utrecht University, Netherlands
Nazanin Alavi,
Queen's University, Canada

*Correspondence:

Birgit Wagner
birgit.wagner@medschool-berlin.de

Specialty section:

This article was submitted to
Mood and Anxiety Disorders,
a section of the journal
Frontiers in Psychiatry

Received: 24 February 2020

Accepted: 22 May 2020

Published: 24 June 2020

Citation:

Wagner B, Rosenberg N, Hofmann L
and Maass U (2020) Web-Based
Bereavement Care: A Systematic
Review and Meta-Analysis.
Front. Psychiatry 11:525.
doi: 10.3389/fpsy.2020.00525

Background: Web-based interventions have been introduced as novel and effective treatments for mental disorders and, in recent years, specifically for the bereaved. However, a systematic summary of the effectiveness of online interventions for people experiencing bereavement is still missing.

Objective: A systematic literature search was conducted by four reviewers who reviewed and meta-analytically summarized the evidence for web-based interventions for bereaved people.

Methods: Systematic searches (PubMed, Web of Science, PsycInfo, PsycArticles, Medline, and CINAHL) resulted in seven randomized controlled trials ($N = 1,257$) that addressed adults having experienced bereavement using internet-based interventions. We used random effects models to summarize treatment effects for between-group comparisons (treatment vs. control at post) and stability over time (post vs. follow-up).

Results: All web-based interventions were based on cognitive behavioral therapy (CBT). In comparison with control groups, the interventions showed moderate ($g = .54$) to large effects ($g = .86$) for symptoms of grief and posttraumatic stress disorder (PTSD), respectively. The effect for depression was small ($g = .44$). All effects were stable over time. A higher number of treatment sessions achieved higher effects for grief symptoms and more individual feedback increased effects for depression. Other moderators (*i.e.* dropout rate, time since loss, exposure) did not significantly reduce moderate degrees of heterogeneity between the studies.

Limitations: The number of includable studies was low in this review resulting to lower power for moderator analyses in particular.

Conclusions: Overall, the results of web-based bereavement interventions are promising, and its low-threshold approach might reduce barriers to bereavement care. Nonetheless, future research should further examine potential moderators and specific treatment components (*e.g.* exposure, feedback) and compare interventions with active controls.

Keywords: grief, bereavement, depression, post-traumatic stress disorder, Internet, e-health, intervention, psychotherapy

INTRODUCTION

Grief after the loss of a significant person is a natural process, and most people adapt their grief gradually to their life after the death of a loved one. However, some people experience difficulties adjusting their grief over time. These difficulties can include separation distress, avoidance behavior, yearning for the deceased or a lack of acceptance of the loss (1). The fifth edition of the Diagnostic and Statistical Manual of Mental Disorders [DSM-5; (2)] included the diagnostic criteria of the Persistent Complex Bereavement Disorder (PCBD) as diagnosis requiring further research, and the 11th edition of the International Classification of Diseases [ICD-11; (3)] included the clinical diagnosis Prolonged Grief disorder (PGD). The main difference between these two criteria sets concerns the time criterion. The PCBD criteria require a functional impairment for at least 12 months after the loss of a significant person, while the PGD criteria require only 6 months of functional impairment. Boelen and Lenferink (4) analyzed in their study four additional existing criteria sets for pathological forms of grief and compared the six diagnostic criteria regarding symptom combination, prevalence, and dimensions. The results of their study indicate a variation in dimensions and symptom combinations and prevalence rates ranging from 10 to 20%. They conclude that the existing criteria sets do not identify the same diagnostic symptom cluster. Similar findings were found in a German treatment-seeking sample (5), showing larger prevalence rates for PGD (69%) compared to PCBD (48%). Studies have shown that pathological grief differs from other disorders such as depression and PTSD and described a distinct diagnosis (4). Nonetheless, there are important associations with depressive symptoms (e.g., feelings of meaninglessness and worthlessness) as well as posttraumatic stress symptoms [e.g., distressful remembrance of the traumatic death of a loved one; (6)].

Today the research on PGD implies not only that there is a strong need for sound and valid diagnostic criteria; there is also a great need for research for treatment strategies for the bereaved.

Previous meta-analyses and systematic reviews of the treatment effects of face-to-face grief interventions have shown inconsistent results (7–11). For example, whereas it was shown that preventive interventions (i.e. to prevent a worsening of normal grief processes) were largely ineffective [$d = 0.03$ – 0.16 ; (11, 12)], interventions that were specifically aimed at prolonged grief symptoms have yielded better treatment effects [$d = 0.53$; (11); see also (10, 12–14)]. The most recent meta-analysis analyzed 31 randomized controlled studies (8). The authors found small but significant effects for psychological interventions for grief symptoms at post-intervention (Hedge's $g = 0.40$). These effects could be maintained at follow-up. In addition to these reviews, treatment approaches that were based on cognitive-behavioral components yielded good treatment efficacy (15–18).

Irrespective of the clinical evidence, manualized interventions are still not routinely used in outpatient care, nor are they easily available to clients who suffer from PGD or belong to a high-risk group. Currently, therapists specializing in the treatment of PGD are still rare. The number of bereaved people seeking psychological treatment is relatively low, too. For example,

Currow et al. (19) examined the nature of bereavement help-seeking following an expected death in a population-based survey ($N = 6,034$) in Australia, and only 1.5% reached out for help from a doctor or nurse; 2.2% contacted a grief counsellor, and 1.9% contacted a spiritual advisor. Another cross-sectional survey of young adults bereaved by suicide and other sudden deaths ($N = 3,432$) in the United Kingdom found that only 13% of the participants had received formal support from health professionals. The most common barriers to utilizing professional mental health support are the worry that it might be too painful to speak about the grief experience (20), the belief that it is too difficult to find help (21), or a fear of stigmatization, specifically for those bereaved by suicide (22).

In the past years, accumulating research has shown that internet-based interventions—particularly cognitive behavioral interventions—can be beneficial for most common mental health disorders, with treatment effects comparable to those of face-to-face treatments (23). Further, studies have shown that internet-based interventions for posttraumatic stress disorder (24, 25) yield medium to large treatment effects compared with a waitlist control group. The interventions are usually delivered in different forms, ranging from self-help treatments without a therapist's guidance to mainly text-based interventions with high therapist involvement (26–31). Web-based bereavement care has a number of advantages which can overcome typical obstacles to receiving support. For example, online interventions offer geographic independence and a widespread dissemination of treatments. In addition, they provide a user-friendly and flexible approach that is more responsive to the reality of most people living in today's digitalized societies. Finally, there is web-based support that can be relatively anonymous (e.g., self-help apps or text-based programs) and can help bereaved patients overcome their initial shame or perceived feelings of stigma and might encourage them to confront themselves with feelings of guilt or disclosure of painful feelings.

In sum, there is growing evidence that psychological interventions have a positive effect on pathological grief symptoms, and there is also research showing the efficacy of internet-based interventions for mental disorders such as anxiety and affective disorders for a variety of populations. Considering that only a minority of bereaved people actively seek traditional forms of support [e.g., (19, 32)], an additional digital dissemination of evidence-based bereavement care might reach even more mourners. With the growing interest of psychotherapy research for e-mental-health interventions, a review of web-based interventions for the bereaved is still missing.

The aim of this systematic review and meta-analysis is to investigate the effectiveness of web-based bereavement interventions compared with control groups in reducing symptoms of grief in adults. Further, we examine the effects of these interventions on PTSD or depression. Due to the fact that existing research has been based on different diagnostic criteria sets, this review does not limit its scope to ICD-11 defined PGD but also includes the other existing criteria sets of disturbed grief symptoms (4) that were, for example, defined as “complicated grief disorder” (33) or “prolonged grief disorder” (1).

METHODS

The Preferred Reporting Items for Systematic Reviews and Meta-Analyses statement (**Supplementary Table 1**; 34) was used as a guide for the literature search and reporting of results.

In PICOS terms (35), the current review addressed adults (≥ 18 years) who have experienced all types of losses of a significant person (Population). The main focus was the use of a web-based psychological treatment (Intervention) which was compared to a control group of any kind (Comparator). We focused on the results of measurements assessing grief symptoms as defined by the original study authors (Outcomes). Here grief did not necessarily have to be determined as the primary outcome in the original study, but the intervention should be aimed at grief. In addition, we examined PTSD and depression as outcome measurements. We included only randomized-controlled trials (Study design).

Identification, Selection of Studies, and Data Extraction

We included all studies according to the following criteria: (a) publication in a peer-reviewed journal; (b) presence of a randomized controlled design; (c) use of an internet-based intervention; (d) participants lost someone through death (*i.e.* bereaved individuals); and (e) assessment of prolonged grief symptoms using (f) validated grief measurements. The following exclusion criteria were also applied: (a) presentation of secondary data only; and (b) investigation of a heterogeneous sample (*e.g.* individuals suffering from traumas other than loss) without presenting results for bereaved individuals separately.

The following databases were used for the literature search: PubMed, Web of Science, PsycInfo/PsycArticles, Medline, and CINAHL. The search included the terms [(“grief” OR “mourning” OR “bereaved” OR “bereavement” OR “death”) AND (“online” OR “internet” OR “computer” OR “web”) AND (“intervention” OR “therapy” OR “self-help” OR “treatment” OR “program” OR “expressive writing”) AND (“randomized” OR “randomized” OR “controlled”)]. We searched the titles and abstracts of relevant articles that were written in English and were published between 1990 and January 17, 2020. In addition, we screened the reference lists of previous reviews and meta-analyses dealing with the issue of internet-based interventions and bereavement/trauma for relevant articles.

Four authors (BW, UM, LH, NR) extracted the following variables: author(s), publication year, title, country, time points of assessment, conditions, outcomes measures, sample size, characteristics of participants and interventions, type of analysis, means and standard deviations of outcome measures (*i.e.* PGD, depression, PTSD), and characteristics of dropouts.

The quality of the included studies was evaluated according to the list of Van Tulder et al. (36), which was based on recommendations by the Cochrane Collaboration Back Review Group (rating scale: yes, no, or unclear) and which the authors applied for their review on internet-based interventions. We eliminated one item because it was not suitable for this review and would have biased the results. The methodological quality of a study was rated good if it achieved a total yes-score ≥ 8 (*i.e.* met

at least two thirds of the criteria). The rating was implemented independently by two of the authors (NR, BW) and was discussed in the case of dissent.

Effect Size Calculation

The meta-analytical summary of the effect sizes was done using RStudio 1.1.456 (37) and the metafor package (38) for two comparisons: (1) a between-group (treatment *versus* control group) at the post assessment, and (2) a within-group design to estimate the stability of the effects of the treatment from post to follow-up assessments. When studies reported several follow-up assessments, we took the one that was closer to the mean time interval. The mean differences between the means of the symptom levels were determined using the results of the studies' intention to treat (ITT) or per protocol (PP) data if ITT were not available. Hedge's g was used as the standardized effect size (ES) to correct for sample size differences (39) and was classified as small ($g < 0.50$), moderate ($0.50 \leq g \leq 0.80$), or large ($g > 0.80$) (40). Multiple assessments of the same outcome were combined to control for dependencies within the data. Heterogeneity (*i.e.* variation in ESs between the studies) using the Q-statistic and the I² index, which indicates low (25%), moderate (50%), or high (75%) levels (41). In cases of at least moderate heterogeneity, we conducted separate meta-regressions to identify potential sources of this heterogeneity (42). The following moderators were considered relevant to explain potential differences in intervention characteristics: (a) dropout rate in the treatment and control group, (b) time since loss in months and number of treatment sessions, (c) the extent of personal therapeutic feedback during treatment (no feedback = 0, informative feedback = 1, personal therapeutic feedback = 2), and (d) the use of exposure, cognitive reappraisal, or behavioral activation within the treatment (each: not applied = 0, applied = 1). All analyses used random-effects models that allowed the amount of residual heterogeneity to be different in each study. A power analysis for random effects models (42) revealed that 6 studies are needed to achieve a power of .84 expecting a moderate mean ES of Hedge's $g = 0.50$ for grief in studies with at least 20 participants per group and moderate heterogeneity (11, 24).

RESULTS

Figure 1 shows the selection process. Seven studies were included in this review.

Study Characteristics and Methodological Quality

The study characteristics are presented in **Table 1**. One study (43) provided two between-group comparisons because the authors compared two conditions with each other (exposure *vs.* behavioral activation) and against a control group. The studies of Wagner (18) and Wagner and Maercker (44) were based on the same trial but focused on different time points: posttreatment symptoms versus follow-up. Consequently, there

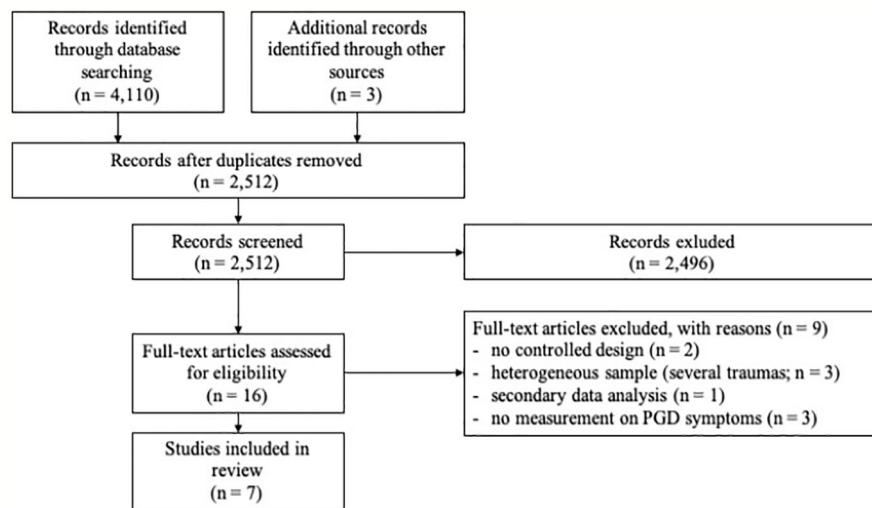


FIGURE 1 | Study flow diagram, showing the results of the literature search for this current review.

were seven studies based on six trials yielding seven comparisons at post and follow-up for grief.

All studies assessed PGD, depression, and PTSD symptoms in response to the loss (see **Table 1** for the according measurements).

The methodological quality of the included studies varied, with no study meeting all 12 criteria (see **Table 2**). Four studies (50%; 43, 45–47) were rated as having good methodological quality.

Six studies (18, 43, 45–48) reported results from ITT analyses. One study (48) applied linear mixed models using REML, two studies applied multilevel analyses (43, 47), and three studies used the last observation carried forward approach to handle missing data (18, 45, 46). To calculate the ES for our review, we referred to the corresponding sample sizes per protocol (PP) except for Kersting et al. (45), who reported means from ITT data, and Wagner et al. (18), who reported means for completers only.

Samples

The studies included a total of $N = 1,257$ randomized participants from the general population. **Table 3** presents the sample's characteristics. Women comprised between 68 and 100% of the samples. The average age of the total sample was 41.7 years. Overall, participants tended to be well-educated (i.e., college and university degrees), with five studies indicating that the education level of most of their participants was high.

Two of the included studies addressed only mothers (46) and parents (45), respectively, who had lost a child during pregnancy, whereas one study (48) included only people who had experienced the expected natural death of someone. The causes of death in the remaining studies were (unexpected) natural death, stillbirth, sudden infant death, or death by accident, suicide, or homicide. Time since loss ranged from 8.38 to 55.2 months, with a mean of 26.7 months, indicating great variability for this variable.

All studies used measurements that explicitly addressed symptoms that were labeled as “complicated” or “prolonged” grief or grief disorder [see (4) for a comparison of different criteria sets for PGD]. As **Table 1** displays, only two studies did not apply a cut-off score to indicate higher levels of PGD (46, 47).

Characteristics of the Interventions

Table 4 provides an overview of the characteristics of the different intervention programs. Three studies included minimal therapeutic support, such as explanations regarding homework and minor logistical help (43, 47, 48). Four studies involved individualized feedback after most treatment modules (18, 44–46). Further, five studies used structured writing assignments (18, 44–47) based on the Pennebaker paradigm (62). All of the included studies used an approach based on cognitive behavioral therapy (CBT).

Regarding the content of the programs, six studies used a manual and included an exposure module (18, 43–47), five studies involved cognitive reappraisal (including integration and restoration, also called social sharing) (18, 44–47), and two studies used behavioral activation modules (43, 48). Five studies were based on an adjusted protocol for the treatment of posttraumatic stress disorder through the internet by Lange et al. (63).

Dropouts

We defined dropouts as all participants who completed pre-assessments but did not complete the treatment (or waitlist period). The rates of dropouts (**Table 1**) ranged from 10.3 (18) to 58.8% (43).

All included studies provided some information about the characteristics of dropouts, except for Wagner et al. (18).

TABLE 1 | Study characteristics.

Study	Eisma et al. (43)	Kersting et al. (46)	Kersting et al. (45)	Litz et al. (48)	van der Houwen et al. (47)	Wagner et al. (18)	Wagner and Maercker (44) ^a
Condition	1. Exposure (EX) 2. Behavioral activation (BA) 3. WL control	1. Treatment 2. WL control	1. Treatment 2. WL control	1. Treatment 2. WL control	1. Treatment 2. WL control	1. Treatment 2. WL control	1. Treatment 2. WL control
Outcome	Prolonged grief (ICG-R > 25), posttraumatic stress, anxiety, grief, depressive rumination	Prolonged grief symptoms, posttraumatic stress, depression, somatization, anxiety, general mental health	Prolonged grief (score > 36 on separation and traumatic distress), Posttraumatic stress, depression, anxiety, general mental health	Prolonged grief (PG-13 > 23), depression, posttraumatic stress, anxiety	Prolonged grief symptoms, depression, positive mood, emotional loneliness	Prolonged grief (predetermined cut-off scores): failure to adapt, intrusion, avoidance, depression, anxiety, general mental, physical health	Prolonged grief (predetermined cut-off scores): failure to adapt, intrusion, avoidance, depression, anxiety, general mental, physical health
Measurement							
Grief	ICG-R	ICG	ICG	PG-13	9 items based on the criteria for complicated grief ^b	5 items from the revised symptom list for complicated grief ^c	5 items from the revised symptom list for complicated grief ^d
PTSD	PSS	IES	IES-R	PCL-b	–	IES-I / IES-A	IES-I / IES-A
Depression	HADS	BSI	BSI	BSI	CED-D	BSI	BSI
Sessions / duration^e	6 / 6-8	10 / 5	10 / 5	18 / 6	5 / 5	10 / 5	10 / 5
Assessment	Pre, Post, FU (3) ^e	Pre, Post, FU (3) ^e	Pre, Post, FU (3, 12) ^e	Pre, Post, FU (1.5, 3) ^e	Pre, Post, FU (3) ^e	Pre, Post, FU (3) ^e	FU (18) ^e
Sample size							
- Treatment (Rand/Post/FU)	EX: 18 / 15 / 12 BA: 17 / 11 / 11	48 / 33 / 29	115 / 99 / 85, 45	43 / 32 / 31, 31	460 / 201 / 190	29 / 26 / 25	29 / 26 / 22
- WL-control (Rand/Post/FU)	12 / 10 / 10	35 / 26	113 / 100	44 / 42 / 35, 35	297 / 254 / 217	26 / 25	26 / 25
Dropout rates (treatment : control)	33.3% (EX), 58.8% (BA) : 16.7%	26.7% : 21.2%	13.9% : 11.5%	22.0% : 2.3%	52.0% : 14.5%	10.3% : 3.8%	15.4% ^f

GP, general population; CBT, cognitive behavioral therapy; WL control, waitlist control; Rand, randomized; Post, post treatment; FU, completed follow-up assessment; EX, exposure; BA, behavioral activation; Pre, pre treatment; ICG, Inventory of Complicated Grief [ICG-R; (49); ICG; (50)]; PG-13, Prolonged Grief Scale (1); PSS, PTSD Symptom Scale (51); IES, Impact of Event Scale; IES-I, Intrusion; IES-A, Avoidance [IES; (52); IES-R; (53)]; PCL-C, PTSD Checklist - civilian version (54); HADS, Hospital Anxiety and Depression Scale (55); BSI, Brief Symptom Inventory (56); BDI-II, Beck Depression Inventory-II (57); CES-D, Center for Epidemiological Studies-Depression Scale (58).

^a1.5-year follow-up data based on the study by Wagner et al. (18). ^bproposed for DSM-V. ^c(59); trouble sleeping, feeling worthless, an altered sense of future, and feeling lonely or empty.

^dnumber of sessions / duration in weeks. ^ein months after the post assessment. ^fbased on participants who dropped out from the pre treatment in the study by Wagner et al. (18) to the 18-month follow-up in Wagner and Maercker (44).

Eisma et al. (43) reported no significant differences between completers and dropouts on any of the assessed variables (demographic, loss-related, symptom, and rumination). In the study by Kersting et al. (46), time since loss was significantly longer for dropouts. By contrast, time since loss was shorter for Kersting et al.'s (45) dropouts, who were also younger and lost their child significantly earlier in pregnancy (the study investigated pregnancy loss). Litz et al. (48) reported that their dropouts received more average weekly emails from the therapist during the program and that they were more often employed full-time than completers. Dropouts differed on several variables in the study by van der Houwen et al. (47): They were younger, had lower levels of education, experienced more grief and were more likely to be in the treatment condition.

Effect Sizes

The results for the between-group comparisons are displayed in **Figure 2**. The overall effect on grief symptoms was moderate and significant with moderate to high heterogeneity between the studies. The ESs for PTSD¹ and depression were almost moderate (depression) to large (PTSD) and statistically significant with low (PTSD) and high (depression) heterogeneity.

All effects remained stable from post-assessment to follow-up (3 months) with low (depression) to moderate (grief, PTSD) heterogeneity between the studies: (a) grief: $k = 7$, Hedge's $g = 0.17$, 95% CI $[-0.03, 0.36]$, $p = .094$, $I^2 = 34.1\%$, Q

¹Wagner et al. (18) report ESs for two IES subscales (*i.e.* Intrusion and Avoidance), which were combined into one ES for PTSD symptoms on the basis of a correlation of $r = .59$ (42, 53).

TABLE 2 | Methodological quality of included studies.

Study	Eisma et al. (43)	Kersting et al. (46)	Kersting et al. (45)	Litz et al. (48)	van der Houwen et al. (47)	Wagner et al. (18)	Wagner and Maercker (44) ^a
Were the eligibility criteria specified?	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Was the method of randomization described?	Yes	Yes	Yes	No	No	No	No
Was the random allocation concealed? ^b	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear
Were the groups similar at baseline regarding important prognostic indicators?	No	Yes	No	No	Yes	Yes	Yes
Were both the index and the control interventions explicitly described?	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Was the outcome assessor blinded to the interventions?	Unclear	Unclear	No	Unclear	Unclear	Unclear	Unclear
Was the dropout rate described, and were the characteristics of the dropouts compared with the completers of the study?	Yes	Yes	Yes	Yes	Yes	No	Yes
Was a long-term follow-up measurement performed? ^c	No	No	Yes	No	Yes	No	Yes
Was the timing of the outcome measurements in both groups comparable?	Yes	Yes	Yes	No	Yes	Yes	No ^d
Was the sample size for each group described by means of a power calculation?	Yes	Yes	No	No	No	No	No
Did the analysis include an intention-to-treat analysis?	Yes	Yes	Yes	Yes	Yes	Yes	No
Were the point estimates and measures of variability presented for the primary outcome measures?	Yes ^e	Yes	Yes	Yes	Yes	Yes	Yes
Total number of criteria fulfilled	8	9	8	5	8	6	6

CG, control group; FU, follow-up.

^a1.5-year follow-up data based on the study by Wagner et al. (18). ^b“Was the assignment generated by an independent person not responsible for determining the eligibility of the patients?” ^c“outcomes measured ≥ 6 months after randomization.” ^dNo control group at follow-up. ^eAvailable as a supplement.

(6) = 7.69, $p = .261$, (b) PTSD: $k = 6$, Hedge's $g = 0.17$, 95% CI $[-0.06, 0.40]$, $p = .154$, $I^2 = 27.1\%$, $Q(5) = 5.85$, $p = .321$, and (c) depression: $k = 7$, Hedge's $g = 0.07$, 95% CI $[-0.07, 0.20]$, $p = .324$, $I^2 < .01\%$, $Q(6) = 1.54$, $p = .957$.

There were only two moderators with a significant impact on the ESs ($ps < 0.05$, **Table 5**) and significantly reduced heterogeneity at post-assessment: A higher number of therapeutic sessions were associated with a higher ES for grief, and more individual feedback increased the ES for depression.

Potential outliers for all outcomes were detected using influential case diagnostics (see **Supplementary Table 2**) Viechtbauer and Cheung (64), which marked two studies: van der Houwen et al., (47) at post-assessment (grief and depression) and Kersting et al. (45) at follow-up (grief and PTSD). While the results did not substantially change after recalculating the meta-analyses without Kersting et al.'s study, the ESs increased to a moderate degree and the heterogeneities decreased to zero after recalculating the analyses without van der Houwen et al.'s study (see **Supplementary Table 3**) (65). Publication bias was not inspected because the analysis included fewer than 10 studies and heterogeneity was often greater than 10 (66).

DISCUSSION

The present review systematically evaluated the efficacy of web-based bereavement care interventions for bereaved people with higher levels of disturbed grief, based on seven RCTs using active or waitlist control groups. The effects of the web-based interventions on grief reduction were promising with moderate ESs (Hedge's $g_{between} = 0.54$), which was also stable from post to 3-months follow-up assessment. Overall, the results are in line with an earlier review

of face-to-face interventions for grief (11) which yielded an effect size of 0.53. However, the most recent review (8) found only small effects for bereavement interventions, particularly when considering publication bias ($g = 0.31$). One explanation for the diverging results might be that the present review explicitly focused on online interventions while the Johannsen et al. (8) combined face-to-face and online treatments. Another reason might lie in the slightly different definitions of grief and the assessment of its pathological levels. For example, Johannsen et al. (8) included only studies that assessed grief using one of the versions of the ICG-R or PG-13. Facing similar decisions regarding definition and assessment, this review relied on the definitions of prolonged grief that was presented by the original authors of the included studies. With the new ICD-11 criteria and the use of consistent measurements, it should be possible to reduce variance between studies resulting from variance in outcome assessments in the future. Finally, the differences might also be due to different methodological approaches. For example, Johannsen calculated their effect sizes considering pre-post differences while this review based its estimation on postscores. Nonetheless, it is noteworthy that despite these different methodological approaches, the main conclusions from the existing reviews still seem to be comparable: Psychological treatment for pathological grief in general, and based on this review delivered *via* online-based formats in particular, is effective, but it is not yet as effective as it is in treating online, for example, panic or social anxiety disorders, given the absolute numbers of ESs (67).

The largest and most robust effect, however, was found for grief-related PTSD symptoms (Hedge's $g_{between} = 0.86$) which also remained stable over time. These results are generally in line with research demonstrating the effectiveness of e-mental health approaches in the reduction of PTSD (24, 68). The result that

TABLE 3 | Sample characteristics.

Study	Eisma et al. (43)	Kersting et al. (46)	Kersting et al. (45)	Litz et al. (48)	van der Houwen et al. (47)	Wagner et al. (18)	Wagner and Maercker (44) ^a
Country	Netherlands	Germany	Germany	USA	USA, UK ^b	Germany ^c	Germany ^c
Population	GP	GP	GP	GP	GP	GP	GP
Subjects	Subjects with elevated levels of complicated grief ^d and elevated grief rumination ^e	Mothers after pregnancy loss	Parents after pregnancy loss	Bereaved caregivers with elevated levels of prolonged grief ^f	Bereaved subjects significantly distressed by the loss	Bereaved individuals with symptoms of intrusion, avoidance, and maladaptive behavior ^g	Bereaved individuals with symptoms of intrusion, avoidance, and maladaptive behavior ^g
Age^h M (SD)	45.7 (12.9)	34.3 (5.3)	34.2 (5.15)	55.4 (10.33)	43.2 (11.0)	37.6 (10.3)	36.0 (11)
Female in %	91.5	100	92.1	67.9	93.5	92.3	88
Education in %ⁱ							
- Low	40.4	9.0	4.4	1.2	16.8	15.6	
- Medium		44.9	12.7	31.1	47.0	39.2	41
- High	59.6	46.2	82.9	67.8	36.2	31.2	31
Relationship to the deceased in %							
- Spouse/Partner	40.4 ^j	—	—	82.1	30.4	10	10
- Child	—	100	100	4.7	42.5	62	61
- Parent	—	—	—	7.3	16.6	6	10
- Parent-in-law	—	—	—	—	—	—	—
- Grandparent	—	—	—	—	—	—	—
- Sibling	—	—	—	2.5	10.4	4	3
- Aunt/uncle	—	—	—	—	—	—	—
- Relative	—	—	—	2.5	—	4	16 ^k
- Friend	—	—	—	1.2	—	14	—
Type of loss in %							
- Expected natural death	Cause of death was categorized as nonviolent	—	—	100	—	—	—
- Natural death	(78.7%) or violent (22.8%)	—	—	—	65.8	36	42
- Pregnancy loss		100	100	—	—	—	—
- Stillbirth/SID		—	—	—	—	18	16
- Accident		—	—	—	22.1 ^l	26	23
- Suicide		—	—	—	12.2	20 ^m	19 ^m
Time since loss in months M (SD), [range]	31.0 (45.1)	15.4 (27.4), [1-144]	9.93 (24.11)	8.38 (2.97)	40.44 (62.88)	55.2 (78.6), [14-348]	48 (60), [14-192]

SID, sudden infant death; GP, general population.

^a1.5-year follow-up data based on the study by Wagner et al. (6).

^bIncluding English speakers living in other countries. ^cIncluding German speakers living in other countries. ^dScore on the Inventory of Complicated Grief > 25. ^eScore on the Utrecht Grief Rumination Scale > 40. ^fScore on the Prolonged Grief Scale > 22 as well as functional impairment in social, occupational, or household responsibilities. ^gCaused by the death of a significant other within predetermined cut-off scores that were not described in the study. ^hIn years. ⁱIncludes incomplete information. The labels "low", "medium", and "high" refer to the classification used by the original study authors, which are not always explained in more detail. Generally, low education relates to the early school forms (i.e., primary school, high school or vocational school) and higher education to the final stages in college or university (e.g., masters or doctoral degree). ^jRelationship of the remaining 59.6% could be children, siblings, or parents. ^kIncluding relatives and friends. ^lIncluding accident and homicide. ^mIncluding suicide and homicide.

interventions had stronger effects on posttraumatic symptoms than on prolonged grief had already been noted previously (8) but still needs clarification. One reason could be that the disturbed grief and PTSD have "less clear boundaries" (6, p. 2446). In fact, the majority of the current treatment protocols (i.e., 86% of the included studies) are largely tailored to treat distressing memories that are associated with the loss and reduce avoidance using exposure-elements. Distressing remembrance is a cluster that shows the largest overlap between PCBD and PTSD

(6). Thus, the current treatment protocols might address mutual grief and posttraumatic stress symptoms more than they address the specific symptoms of prolonged grief [i.e., role confusion, meaninglessness, and loneliness; (6)].

The weakest effect was found for depression (Hedge's $g_{between} = 0.44$), which is still comparable to the effects of general internet-based treatments for depression [(69–71); but see e.g., (72) for larger effects] and the effects of general PGD interventions for secondary depression outcomes (8). There might be two reasons for the

TABLE 4 | Characteristics of the intervention programs

Study	Eisma et al. (43)	Kersting et al. (46)	Kersting et al. (45)	Litz et al. (48)	van der Houwen et al. (47)	Wagner et al. (18)	Wagner and Maercker (44) ^a
Therapeutic approach	CBT	CBT	CBT	CBT	CBT	CBT	CBT
Content / Modules	e-mailed homework assignments; Exposure ^b : writing assignments, imaginal and/or in vivo exposure exercises; Activation ^c : 7-day activity diary, engagement in value-based activities	Writing assignments ^d : (1) 4 self-confrontation (describe the traumatic loss and its circumstances), (2) 4 x cognitive restructuring (supportive letter to a hypothetical friend, develop new perspectives on the loss), (3) 2x social sharing (symbolic farewell letter)	Writing assignments ^d : (1) 4 self-confrontation (describe the traumatic loss and its circumstances), (2) 4 x cognitive restructuring (supportive letter to a hypothetical friend, develop new perspectives on the loss), (3) 2x social sharing (symbolic farewell letter)	HEAL intervention: (1) psycho-education about loss and grief, (2) instruction in stress management and other coping skills, (3) behavioral activation (self-care + social re-engagement), (4) accommodation of loss + goal achievement, (5) relapse prevention	Writing assignments ^d : (1) 2x exposure (most-distressing aspects of the loss), (2) 2x cognitive reappraisal (identify unhelpful thoughts, develop helpful thoughts; letter to a hypothetical friend), (3) 1x integration, and restoration (letter to deceased from future)	Writing assignments ^d : (1) 4x exposure (circumstances of death, most-distressing aspects of the loss), (2) cognitive reappraisal (letter to a hypothetical friend, develop new role, rituals, positive resources), (3) integration, and restoration (letter to significant person, most important memories, coping with death, changes)	Writing assignments ^d : (1) 4x exposure (circumstances of death, most-distressing aspects of the loss), (2) cognitive reappraisal (letter to a hypothetical friend, develop new role, rituals, positive resources), (3) integration, and restoration (letter to significant person, most important memories, coping with death, changes)
Exposure	Yes	Yes	Yes	No	Yes	Yes	Yes
Cognitive restructuring	No	Yes	Yes	No	Yes	Yes	Yes
Behavioral activation	Yes	No	No	Yes	No	No	No
Sessions / Duration	6 sessions / 6-8 weeks	10 sessions à 45 min / 5 weeks	10 sessions à 45 min / 5 weeks	18 sessions à 20 min / 6 weeks	5 sessions / 5 weeks	10 sessions à 45 min / 5 weeks	10 sessions à 45 min / 5 weeks
Frequency of contact	After each assignment; intake interview by telephone	Once per week; telephone call before treatment ^e	Once per week; telephone call before treatment ^e	Periodically via email, on average 0.6 emails and 0.24 calls per week; single phone call at the beginning of treatment	Once per week for instruction	Twice per week	Twice per week
Therapeutic feedback	Informative (explaining homework and maximizing treatment adherence)	Individual written feedback, instructions for the next writing assignment	Individual written feedback, instructions for the next writing assignment	Informative (information about program)	Informative (general guidelines for writing), no feedback on writing assignments	Individual written feedback every 2 nd essay, instructions for the next writing assignment	Individual written feedback, every 2 nd essay, instructions for the next writing assignment
Manual	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Multimedia	No	No	No	No	No	No	No
Reminder	No	No	No	Yes	Yes	No	No

CBT, cognitive behavioral therapy.

^a1.5-year follow-up data based on the study by Wagner et al. (6). ^bBased on Boelen et al. (15). ^cBased on Lejuez et al. (60). ^dBased on Lange et al. (61). ^eFor applicants who scored high on any of the screening instruments to check for worsening of symptoms.

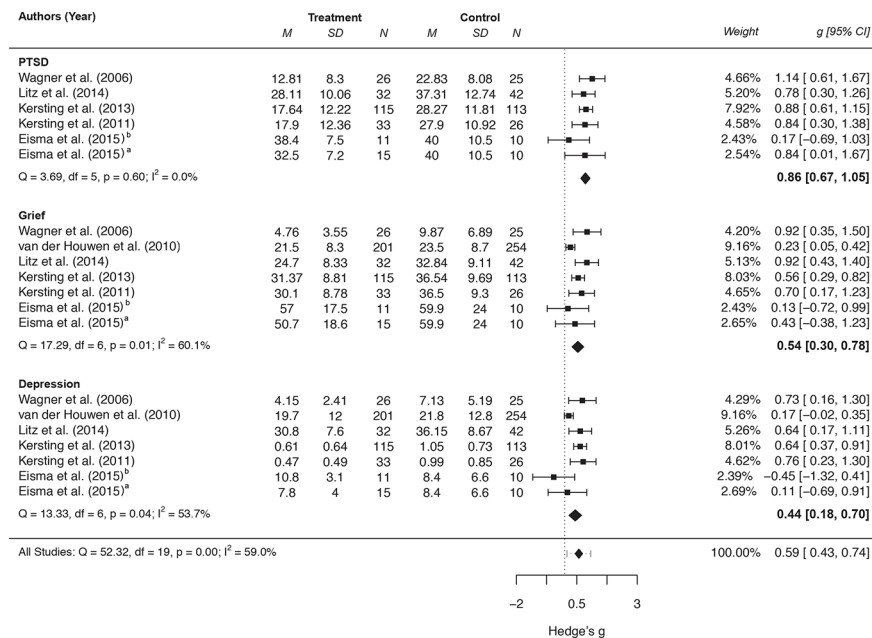


FIGURE 2 | Forest plot of between-group effect sizes of internet-based interventions for PGD, depression and PTSD symptoms. ^aComparison between Exposure-based treatment and waitlist control group. ^bComparison between Behavioral activation and waitlist control group.

TABLE 5 | Meta-regressions to test for the influence of moderators in models with moderate to high heterogeneities.

	Between-Group Comparison						Stability (Post-Follow Up)					
	Grief			Depression			Grief			PTSD		
	B	95%CI	I ²	B	95%CI	I ²	B	95%CI	I ²	B	95%CI	I ²
Dropout TG ^a	-0.005	[-0.02, 0.01]	8.38	-0.02	[-0.03, -0.002]	<0.01	<-0.001	[-0.02, 0.02]	35.5	0.01	[-0.02, 0.03]	49.1
Dropout CG ^a	-0.01	[-0.04, 0.02]		-0.01	[-0.02, 0.04]		-0.004	[-0.04, 0.03]		-0.004	[-0.05, 0.04]	
Time s. loss ^b	0.004	[-0.01, 0.02]	<.01	<-0.001	[-0.02, 0.02]	38.8	-0.01	[-0.03, 0.001]	<0.01	-0.01	[-0.02, 0.01]	14.1
No. sessions	0.07**	[0.02, 0.12]		0.05	[-0.02, 0.12]		-0.02	[-0.07, 0.03]		-0.04	[-0.11, 0.03]	
Feedback ^c	0.11	[-0.15, 0.37]	44.7	0.20*	[0.002, 0.39]	21.5	0.04	[-0.17, 0.26]	30.5	0.03	[-0.30, 0.36]	33.9
Exposure ^d	-0.16	[-0.76, 0.44]	55.1	0.21	[0.48, 0.91]	69.4	-0.09	[-0.63, 0.46]	41.7	-0.04	[-0.63, 0.55]	36.6
Reappraisal	-0.08	[-0.64, 0.48]	57.6	0.30	[-0.32, 0.92]	68.8	-0.08	[-0.56, 0.41]	41.8	-0.003	[-0.40, 0.40]	<0.01
Activation	0.08	[-0.48, 0.64]	57.6	-0.30	[-0.92, 0.32]	68.8	0.08	[-0.41, 0.56]	41.8	0.003	[0.40, -0.40]	<0.01

TG, treatment group; CG, control group. Significant effects are in bold ($p < 0.01$). ^aIn percent, ^bin months, ^cvariable was treated as continuous on a rating scale of 0 = no feedback, 1 = informative feedback, and 2 = personal therapeutic feedback, ^d0 = no exposure, 1 = feedback. I² = residual heterogeneity.

* $p < 0.05$. ** $p < 0.01$.

smaller effects on depression. First, there was high heterogeneity in our results that might partly be explained by the variability in the extent of therapeutic feedback. As our moderator analyses showed, studies with more personal feedback were more likely to report higher ESs for depression. Previous studies also reported that individualized internet interventions achieve higher outcomes compared to pure self-help or no guided tools (73). However, because our meta-regressions were somewhat underpowered, we interpret the results for depression cautiously with the outlook that more studies are needed to disentangle the associations between interventions, depression, and feedback. Second, given that major depression and PCBD are more distinct categories than PTSD and

PBCD (6), the comparably smaller ESs might reflect the narrow focus of current treatment protocols to grief and not depression. However, because there is still overlap between both diagnoses with respect to, for example, worthlessness and guilt, the applied treatment concepts might still address some parts of depressive symptoms. Again, Johannsen et al. (8) also found no significant effect of bereavement treatments on depression and hypothesized that there might be no “spill-over effect of grief interventions on other psychological morbidities” (p. 78). This raises the question what kind of treatment ingredients are currently missing in treatment protocols that would address depressive symptoms more directly. Eisma et al. (43) examined a pure behavioral

activation module but did not yield sufficient ES for depression. However, it would be interesting to combine writing assignments using exposure and cognitive reappraisal [e.g., based on the approach by (50)] with behavioral activation.

Considering the attrition rates of the reviewed treatments, our results showed that, generally, the average attrition rate of 27% is comparable with other internet-based treatment studies (74). However, one interesting finding was the wide range of dropout rates between the treatment groups (*i.e.*, 10 to 58%). One factor that might contribute to this range could be the degree of guidance and personal feedback, because the highest dropout rates were found in treatments that offered only informative feedback (43, 47). These studies also mentioned that some participants dropped out because the intervention was “too impersonal”, or that the “treatment did not help”, “was too difficult”, or that they had “no confidence” in it [(43), p. 5; 47, p. 362]. The behavioral activation group in Eisma et al.'s study, for example, had the highest dropout rate, which might further point to difficulties that are associated with implementing behavioral changes by oneself. In addition, the average dropout rate for studies providing informative feedback was 41% ($SD = 17$) and only 17% ($SD = 15$) for studies providing personal feedback. Previous research suggested the importance of guidance in internet-based interventions for their efficacy and its superiority on adherence over completely unguided interventions (75, 76). This does not necessarily mean that the treatment concepts are ineffective but that additional factors (*i.e.*, guidance) are needed to facilitate change. The results of the outlier analysis similarly point to this direction, because, after excluding van der Houwen et al.'s study, heterogeneity between the studies was reduced and the ESs for grief and depression rose to a moderate level. This study applied writing assignments including exposure and reappraisal elements. Both seemed to be effective in the other included studies, but the intervention was very short (*i.e.*, five sessions) and dispensed with personal feedback. Thus, the dropout rate was high.

Similar to the range of dropouts, there was a large range between the number of sessions across studies (*i.e.*, five to 18). The moderator analyses showed that a higher number of treatment sessions were associated with higher effect sizes for grief. It is noticeable that the interventions with the highest dropout rates (43, 47) also had the lowest number of sessions (*i.e.*, six and five) and the lowest personal support. Again, this finding might also add to the hypothesis that participants suffering from PGD need more personal support over a longer period of time.

In sum, the interpretation of the present results on web-based bereavement care are mostly in line with previous research on face-to-face treatments: While online treatment is largely effective in treating PTSD symptoms, its effect on grief symptoms themselves is comparably lower, although moderate and stable over time. The effects on depression, however, are less clear and tend to be small.

Limitations

A number of limitations of this review need to be addressed. First, only a small number of studies could be included in this review. E-mental health for the bereaved is still in its infancy, and further randomized controlled trials are needed. Although we

reached the required number of at least seven studies to achieve adequate power, the number of included studies varied depending on the specific comparison and outcome (*i.e.* for meta-regressions). Also, not all studies included 20 participants per study group (43). It remains unclear whether the nonsignificant results for study characteristics (*e.g.* influence of exposure vs. no exposure) are true effects or whether our analyses were underpowered (42, 77).

Second, because all interventions were based on CBTs and almost all used exposure modules, we cannot make specific conclusions about their effectiveness compared to other treatment approaches. Nonetheless, they seem promising, since memories of the death and its circumstances are often experienced as very distressful and are therefore avoided. In line with this assumption is the rationale and effectiveness of face-to-face exposure-based treatments for PTSD (78) and PGD symptoms (15, 79). Interestingly, Eisma et al.'s (43) behavioral activity intervention did not reveal significant effects on grief-related PTSD, but the exposure group did. However, because of the very small sample sizes in each group (11 vs. 10 participants at the post assessment) and because of the significant baseline differences, the results were not considered in the effect size calculations for this review.

Third, the fact that in most studies, different types of losses were addressed (*e.g.* parental loss, bereaved parents, suicide bereavement) complicates the process of reaching a reliable conclusion.

Finally, this group of bereaved people included in this meta-analysis might not reflect the naturalistic setting in the treatment of clients with disturbed grief symptoms who are traditionally looking for help in self-help groups and who might additionally suffer from comorbid disorders, increased suicidality, and alcohol and substance abuse. Consequently, effectiveness studies should be conducted to provide better knowledge about the generalization of previous results from randomized controlled trials.

Future Directions and Practical Implications

Since this review included all studies that measured PGD symptoms on a continuous scale, future studies might address PGD diagnosed according to one validated sound diagnostic criterion. Further, the identification of relevant moderators, such as symptom severity, type of loss, length of treatment, and amount of therapist support is needed to improve treatment outcomes from web-based interventions. Further, randomized controlled trials should involve active control groups, such as face-to-face groups or self-help interventions and compare writing assignments directly to other online-approaches.

Generally, there is a great need to understand the help-seeking patterns of bereaved people depending on gender, type of loss (*e.g.* suicide, illness), age group (*e.g.* adolescents, the elderly), and relationship to the deceased (*e.g.* child, parent, sibling). Further, dismantling studies should be conducted as well as blended care interventions, which could include internet-based interventions in existing psychosocial support (*e.g.* self-help groups). A challenging task might be the inclusion and treatment of less-educated participants in internet-based interventions because, so far, they are reaching a better-educated group of people (80).

In addition, future research studies should examine the underlying mechanisms of effective treatments. For example, it is worth mentioning that all interventions [except the behavioral activation group in (43)] included extensive structured writing assignments, which might be one promising treatment approach that is applicable to both, online and face-to-face interventions. A number of previous studies showed that expressive writing assignments on specific bereavement-related themes can be effective in reducing the symptoms of PGD (81) because it enhances deep reflection on the loss, leads to new perspectives, or supports the process of sense-making (82). Only recently, Sloan and colleagues (83) compared a five session written exposure intervention with cognitive processing therapy (CPT), a first line treatment approach for PTSD. The results indicate that the written exposure intervention was equally effective as CPT in trauma-related symptom reductions. Expressive writing might enhance a deep reflection on the loss or the traumatic experience and might help to develop a narrative of it. The emotional processing during writing might finally lead to new perspectives that are perceived as less threatening. Further, writing might support the process of sense-making and relieve the distressed mourner (82). Taken together, the positive treatment effect of the analyzed internet-based interventions could be largely due to the written tasks. Hence, written assignments that foster self-disclosure and reduce avoidance seem worth considering in future online-interventions as well.

This review offers some practical implications. First, internet-based interventions are a promising alternative to treat PGD when face-to-face encounters are not possible or wanted. Because all reviewed interventions were based on CBT and mostly used writing assignments based on the Pennebaker paradigm (49), there seems to be evidence that clinicians can implement these structured writing approaches in their clinical practice. The main elements hereby are the examination of distressing remembrance using exposure elements, and cognitive restructuring of grief-related dysfunctional thoughts, as well as the integration and restoration (*i.e.*, development of new goals for the future). Although more research is still

necessary to improve internet-based treatments for PGD, clinicians might pay special attention to very specific grief- and depression-related symptoms in addition to posttraumatic stress symptoms. Further, it seems advisable to offer online programs that are not too short (*i.e.*, >6 sessions) and to provide personal feedback and guidance to avoid frustration and early dropout.

In conclusion, the results suggest that internet-based treatments, based on CBT, can help to reduce the symptoms related to the loss of a significant person in a relatively short time. Yet, more qualitative RCTs with adequate power on that topic are needed.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are openly available in Open Science Framework at https://osf.io/7re5m/?view_only=66a8043dfe644b4699d619e03ee10c76.

AUTHOR CONTRIBUTIONS

BW: substantial contribution to the conception, drafting the work, and revising it critically for important intellectual content; final approval of the version to be published. UM: substantial contribution to every aspect of the manuscript, critical revision, interpretation of the data, and final approval and agrees to be accountable. LH: substantial contribution to interpretation of data, critical revision, and final approval and agrees to be accountable. NR: Initial substantial contribution to the conception and design of the work.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: <https://www.frontiersin.org/articles/10.3389/fpsy.2020.00525/full#supplementary-material>

REFERENCES

1. Prigerson HG, Horowitz MJ, Jacobs SC, Parkes CM, Aslan M, Goodkin K, et al. Prolonged grief disorder: Psychometric validation of criteria proposed for DSM-V and ICD-11. *PLoS Med* (2009) 6(8):e1000121. doi: 10.1371/journal.pmed.1000121
2. Association AP. *Diagnostic and statistical manual of mental disorders (DSM-5®)* Washington D.C.: American Psychiatric Pub. (2013).
3. World Health Organization. (2019). *International Statistical Classification of Diseases and Related Health Problems, 11 ed.* Retrieved from <https://icd.who.int/browse11/l-m/en>
4. Boelen PA, Lenferink LIM. Comparison of six proposed diagnostic criteria sets for disturbed grief. *Psychiatry Res* (2020) 285:112786. doi: 10.1016/j.psychres.2020.112786
5. Comtesse H, Vogel A, Kersting A, Rief W, Steil R, Rosner R. When does grief become pathological? Evaluation of the ICD-11 diagnostic proposal for prolonged grief in a treatment-seeking sample. *Eur J Psychotraumatol* (2020) 11(1):1694348. doi: 10.1080/20008198.2019.1694348
6. Malgaroli M, Maccallum F, Bonanno GA. Symptoms of persistent complex bereavement disorder, depression, and PTSD in a conjugally bereaved sample: a network analysis. *psychol Med* (2018) 48(14):2439–48. doi: 10.1017/S0033291718001769
7. Allumbaugh DL, Hoyt WT. Effectiveness of grief therapy: A meta-analysis. *J Couns Psychol* (1999) 46(3):370. doi: 10.1037/0022-0167.46.3.370
8. Johannsen M, Damholdt MF, Zachariae R, Lundorff M, Farver-Vestergaard I, O'Connor M. Psychological interventions for grief in adults: A systematic review and meta-analysis of randomized controlled trials. *J Affect Disord* (2019) 253:69–86. doi: 10.1016/j.jad.2019.04.065
9. Kato PM, Mann T. A synthesis of psychological interventions for the bereaved. *Clin Psychol Rev* (1999) 19(3):275–96. doi: 10.1016/S0272-7358(98)00064-6
10. Neimeyer RA. Searching for the meaning of meaning: grief therapy and the process of reconstruction. *Death Stud* (2000) 24(6):541–58. doi: 10.1080/07481180050121480
11. Wittouck C, Van Aultre S, De Jaegere E, Portzky G, van Heeringen K. The prevention and treatment of complicated grief: a meta-analysis. *Clin Psychol Rev* (2011) 31(1):69–78. doi: 10.1016/j.cpr.2010.09.005

12. Currier JM, Neimeyer RA, Berman JS. The effectiveness of psychotherapeutic interventions for bereaved persons: a comprehensive quantitative review. *Psychological Bull* (2008) 134(5):648–61. doi: 10.1037/0033-2909.134.5.648
13. Andriessen K, Kryszinska K, Hill NTM, Reifels L, Robinson J, Reavley N, et al. Effectiveness of interventions for people bereaved through suicide: a systematic review of controlled studies of grief, psychosocial and suicide-related outcomes. *BMC Psychiatry* (2019) 19(1):49. doi: 10.1186/s12888-019-2020-z
14. Schut H, Stroebe MS, van den Bout J, Terheggen M. The efficacy of bereavement interventions: Determining who benefits. In: . *Handbook of bereavement research: Consequences, coping, and care*. Washington, DC, US: American Psychological Association (2001). p. 705–37.
15. Boelen PA, de Keijser J, van den Hout MA, van den Bout J. Treatment of complicated grief: a comparison between cognitive-behavioral therapy and supportive counseling. *J Consult. Clin Psychol* (2007) 75(2):277. doi: 10.1037/0022-006X.75.2.277
16. Rosner R, Lumbeck G, Geissner E. Effectiveness of an inpatient group therapy for comorbid complicated grief disorder. *Psychother Res* (2011) 21(2):210–8. doi: 10.1080/10503307.2010.545839
17. Shear MK, Frank E, Houck PR, Reynolds CF. Treatment of complicated grief: a randomized controlled trial. *JAMA* (2005) 293(21):2601–8. doi: 10.1001/jama.293.21.2601
18. Wagner B, Knaevelsrud C, Maercker A. Internet-based cognitive-behavioral therapy for complicated grief: a randomized controlled trial. *Death Stud* (2006) 30(5):429–53. doi: 10.1080/07481180600614385
19. Currow DC, Allen K, Plummer J, Aoun S, Hegarty M, Abernethy AP. Bereavement help-seeking following an ‘expected’ death: a cross-sectional randomised face-to-face population survey. *BMC Palliat. Care* (2008) 7,19. doi: 10.1186/1472-684X-7-19
20. Andriessen K, Lobb E, Mowl J, Dudley M, Draper B, Mitchell PB. (2018). Help-seeking experiences of bereaved adolescents: A qualitative study. *Death studies*. pp. 1–8. doi: 10.1080/07481187.2018.1426657
21. Lichtenthal WG, Corner GW, Sweeney CR, Wiener L, Roberts KE, Baser RE, et al. Mental Health Services for Parents Who Lost a Child to Cancer: If We Build Them, Will They Come? *J Clin Oncol* (2015) 33(20):2246–53. doi: 10.1200/JCO.2014.59.0406
22. Pitman AL, Osborn D, Rantell K, King M. The stigma perceived by people bereaved by suicide and other sudden deaths: A cross-sectional UK study of 3432 bereaved adults. *J Psychosom. Res* (2016) 87:22–9. doi: 10.1016/j.jpsychores.2016.05.009
23. Andersson G, Cuijpers P, Carlbring P, Riper H, Hedman E. Guided Internet-based vs. face-to-face cognitive behavior therapy for psychiatric and somatic disorders: a systematic review and meta-analysis. *World Psychiatry* (2014) 13(3):288–95. doi: 10.1002/wps.20151
24. Kuester A, Niemeyer H, Knaevelsrud C. Internet-based interventions for posttraumatic stress: A meta-analysis of randomized controlled trials. *Clin Psychol Rev* (2016) 43:1–16. doi: 10.1016/j.cpr.2015.11.004
25. Sijbrandij M, Kunovski I, Cuijpers P. Effectiveness of Internet-Delivered Cognitive Behavioral Therapy for Posttraumatic Stress Disorder: A Systematic Review and Meta-Analysis. *Depress Anxiety* (2016) 33(9):783–91. doi: 10.1002/da.22533
26. Andersson G. Internet-based cognitive-behavioral self-help for depression. *Expert Rev Neurother* (2006) 6(11):1637–42. doi: 10.1586/14737175.6.11.1637
27. Andersson G, Topooco N, Havik O, Nordgreen T. Internet-supported versus face-to-face cognitive behavior therapy for depression. *Expert Rev Neurother* (2016) 16(1):55–60. doi: 10.1586/14737175.2015.1125783
28. Johansson R, Andersson G. Internet-based psychological treatments for depression. *Expert Rev Neurother* (2012) 12(7):861–869; quiz 870. doi: 10.1586/ern.12.63
29. Ruwaard J, Schrieken B, Schrijver M, Broeksteeg J, Dekker J, Vermeulen H, et al. Standardized web-based cognitive behavioural therapy of mild to moderate depression: a randomized controlled trial with a long-term follow-up. *Cognit Behav Ther* (2009) 38(4):206–21. doi: 10.1080/16506070802408086
30. Spek V, Cuijpers PIM, Nyklicek I, Riper H, Keyzer J, Pop V. Internet-based cognitive behaviour therapy for symptoms of depression and anxiety: a meta-analysis. *psychol Med* (2006) 37(03):319–28. doi: 10.1017/S0033291706008944
31. Wagner B, Horn AB, Maercker A. Internet-based versus face-to-face cognitive-behavioral intervention for depression: a randomized controlled non-inferiority trial. *J Affect Disord* (2014) 152-154:113–21. doi: 10.1016/j.jad.2013.06.032
32. Pitman AL, Rantell K, Moran P, Sireling L, Marston L, King M, et al. Support received after bereavement by suicide and other sudden deaths: a cross-sectional UK study of 3432 young bereaved adults. *BMJ Open* (2017) 7(5):e014487. doi: 10.1136/bmjopen-2016-014487
33. Shear MK, Simon N, Wall M, Zisook S, Neimeyer R, Duan N, et al. Complicated grief and related bereavement issues for DSM-5. *Depression Anxiety* (2011) 28(2):103–17. doi: 10.1002/da.20780
34. Moher D, Liberati A, Tetzlaff J, Altman DG, The PG. Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. *PloS Med* (2009) 6(7):e1000097. doi: 10.1371/journal.pmed.1000097
35. World Health Organization. *WHO handbook for guideline development*. Geneva: WHO (2014).
36. Van Tulder M, Furlan A, Bombardier C, Bouter LE. Editorial Board of the Cochrane Collaboration Back Review group. Updated method guidelines for systematic reviews in the cochrane collaboration back review group. *Spine* (2003) 28(12):1290–9. doi: 10.1097/01.BRS.0000065484.95996.AF
37. RStudio Team. (2016). *RStudio: Integrated Development for R: RStudio, Inc., Boston, MA URL* <http://www.rstudio.com/>.
38. Viechtbauer W. Conducting meta-analyses in R with the metafor package. *J Stat Softw* (2010) 36(3):1–48. doi: 10.18637/jss.v036.i03
39. Hedges L, Olkin I. *Statistical Methods for Meta-Analysis*. Boston: Academic Press (1985).
40. Cohen J. *Statistical power analysis for the behavioral sciences*. Hillsdale, NJ: Erlbaum (1988).
41. Crombie IK, Davies HT. What is meta-analysis. *What is* (2009), Hayward Medical Communications, a division of Hayward Group Ltd. 1–8. Retrieved from <http://www.bandolier.org.uk/painres/download/whatis/Meta-An.pdf>
42. Borenstein M, Hedges LV, Higgins JP, Rothstein HR. *Introduction to meta-analysis*. Chichester: John Wiley & Sons. (2011).
43. Eisma MC, Boelen PA, van den Bout J, Stroebe W, Schut H, Lancee J, et al. Internet-Based Exposure and Behavioral Activation for Complicated Grief and Rumination: A Randomized Controlled Trial. *Behav Ther* (2015) 46(6):729–48. doi: 10.1016/j.beth.2015.05.007
44. Wagner B, Maercker A. A 1.5-year follow-up of an Internet-based intervention for complicated grief. *J Trauma Stress* (2007) 20(4):625–9. doi: 10.1002/jts.20230
45. Kersting A, Dolemeier R, Steinig J, Walter F, Kroker K, Baust K, et al. Brief Internet-based intervention reduces posttraumatic stress and prolonged grief in parents after the loss of a child during pregnancy: a randomized controlled trial. *Psychother Psychosom.* (2013) 82(6):372–81. doi: 10.1159/000348713
46. Kersting A, Kroker K, Schlicht S, Baust K, Wagner B. Efficacy of cognitive behavioral internet-based therapy in parents after the loss of a child during pregnancy: pilot data from a randomized controlled trial. *Arch Women's Ment Health* (2011) 14(6):465–77. doi: 10.1007/s00737-011-0240-4
47. van der Houwen K, Schut H, van den Bout J, Stroebe MS, Stroebe W. The efficacy of a brief internet-based self-help intervention for the bereaved. *Behav Res Ther* (2010) 48(5):359–67. doi: 10.1016/j.brat.2009.12.009
48. Litz BT, Schorr Y, Delaney E, Au T, Papa A, Fox AB, et al. A randomized controlled trial of an internet-based therapist-assisted indicated preventive intervention for prolonged grief disorder. *Behav Res Ther* (2014) 61:23–34. doi: 10.1016/j.brat.2014.07.005
49. Prigerson HG, Jacobs SC. Traumatic grief as a distinct disorder: A rationale, consensus criteria, and a preliminary empirical test. In: Stroebe MS, Hansson RO, Stroebe W, Schut H, editors. *Handbook of bereavement research: Consequences, coping, and care*. Washington DC, US: American Psychological Association (2001). p. 613–45.
50. Prigerson HG, Maciejewski PK, Reynolds CF 3rd, AJ B, JT N, Fasiczka A, et al. Inventory of Complicated Grief: A scale to measure maladaptive symptoms of loss. *Psychiatry Res* (1995) 59(1–2):65–79.
51. Foa EB, Cashman L, Jaycox L, Perry K. The validation of a self-report measure of posttraumatic stress disorder: The Posttraumatic Diagnostic Scale. *Psychol Assess* (1997) 9(4):445. doi: 10.1037/1040-3590.9.4.445
52. Horowitz M, Wilner N, Alvarez W. Impact of Event Scale: A measure of subjective stress. *Psychosom Med* (1979) 41(3):209–18. doi: 10.1097/00006842-197905000-00004

53. Maercker A, Schützwohl M. Erfassung von psychischen Belastungsfolgen: Die Impact of Event Skala-revidierte Version (IES-R). *Diagnostica* (1998). 44: 130–41. doi: 10.1037/t55092-000
54. Weathers FW, Litz BT, Herman D, Huska J, Keane T. *The PTSD checklist-civilian version (PCL-C)*. Boston, MA: National Center for PTSD (1994).
55. Zigmond AS, Snaith RP. The hospital anxiety and depression scale. *Acta Psychiatr Scand* (1983) 67(6):361–70.
56. Derogatis LR. *Brief Symptom Inventory (BSI): Administration, scoring, and procedures manual*. Minneapolis: National Computer Systems (NCS) (1993).
57. Beck AT, Steer RA, Brown GK. *Manual for the BDI-II*. San Antonio, TX: Psychological Corporation (1996).
58. Radloff LS. The CES-D scale: A self-report depression scale for research in the general population. *Appl Psychol Meas* (1977) 1(3):385–401. doi: 10.1177/014662167700100306
59. Langner R, Maercker A. Complicated grief as a stress response disorder: Evaluating diagnostic criteria in a German sample. *J Psychosom Res* (2005) 58(3):235–42. doi: 10.1016/j.jpsychores.2004.09.012
60. Lejuez CW, Hopko DR, Acierio R, Daughters SB, Pagoto SL. Ten Year Revision of the Brief Behavioral Activation Treatment for Depression: Revised Treatment Manual. *Behav Modification* (2011) 35(2):111–61. doi: 10.1177/0145445510390929
61. Lange A, Rietdijk D, Hudcovicova M, van de Ven J-P, Schrieken B, Emmelkamp PMG. Interapy: A controlled randomized trial of the standardized treatment of posttraumatic stress through the internet. *J Consult Clin Psychol* (2003) 71(5):901–09. doi: 10.1037/0022-006X.71.5.901
62. Pennebaker JW. Writing about emotional experiences as a therapeutic process. *psychol Sci* (1997) 8(3):162–6. doi: 10.1111/j.1467-9280.1997.tb00403.x
63. Lange A, Schoutrop M, Schrieken B, van de Ven J-P. Interapy: a model for therapeutic writing through the Internet. In: Lepore SJ, Smyth JM, editors. *The writing cure: How expressive writing promotes health and emotional well-being*. Washington, DC, US: American Psychological Association (2002). p. 215–38.
64. Viechtbauer W, Cheung MW. Outlier and influence diagnostics for meta-analysis. *Res Synth. Methods* (2010) 1(2):112–25. doi: 10.1002/jrsm.11
65. Niemeyer H, Musch J, Pietrowsky R. Publication bias in meta-analyses of the efficacy of psychotherapeutic interventions for depression. *J Consult Clin Psychol* (2013) 81(1):58–74. doi: 10.1037/a0031152
66. Higgins JPT, Green S eds. *Cochrane Handbook for Systematic Reviews of Interventions Version 5.1.0*. [updated March 2011]. The Cochrane Collaboration, 2011. Available from www.handbook.cochrane.org
67. Andrews G, Basu A, Cuijpers P, Craske MG, McEvoy P, English CL, et al. Computer therapy for the anxiety and depression disorders is effective, acceptable and practical health care: an updated meta-analysis. *J Anxiety Disord* (2018) 55:70–8. doi: 10.1016/j.janxdis.2018.01.001
68. Sloan DM, Gallagher MW, Feinstein BA, Lee DJ, Pruneau GM. Efficacy of telehealth treatments for posttraumatic stress-related symptoms: a meta-analysis. *Cognit Behav Ther* (2011) 40(2):111–25. doi: 10.1080/16506073.2010.550058
69. Cowpertwait L, Clarke D. Effectiveness of web-based psychological interventions for depression: a meta-analysis. *Int J Ment Health Addict* (2013) 11(2):247–68. doi: 10.1007/s11469-012-9416-z
70. Richards D, Richardson T. Computer-based psychological treatments for depression: a systematic review and meta-analysis. *Clin Psychol Rev* (2012) 32(4):329–42. doi: 10.1016/j.cpr.2012.02.004
71. So M, Yamaguchi S, Hashimoto S, Sado M, Furukawa TA, McCrone P. Is computerised CBT really helpful for adult depression?—A meta-analytic re-evaluation of CCBT for adult depression in terms of clinical implementation and methodological validity. *BMC Psychiatry* (2013) 13(1):113. doi: 10.1186/1471-244X-13-113
72. Szein DM, Koransky CE, Fegan L, Himelhoch S. Efficacy of cognitive behavioural therapy delivered over the Internet for depressive symptoms: A systematic review and meta-analysis. *J Telemed. Telecare* (2018) 24(8):527–39. doi: 10.1177/1357633X17717402
73. Andersson G, Cuijpers P. Internet-based and other computerized psychological treatments for adult depression: a meta-analysis. *Cogn Behav Ther* (2009) 38(4):196–205. doi: 10.1080/16506070903318960
74. Melville KM, Casey LM, Kavanagh DJ. Dropout from Internet-based treatment for psychological disorders. *Br J Clin Psychol* (2010) 49(4):455–71. doi: 10.1348/014466509X472138
75. Baumeister H, Reichler L, Munzinger M, Lin J. The impact of guidance on Internet-based mental health interventions—A systematic review. *Internet Interventions* (2014) 1(4):205–15. doi: 10.1016/j.invent.2014.08.003
76. Domhardt M, Gefleim H, von Rezori RE, Baumeister H. Internet- and mobile-based interventions for anxiety disorders: A meta-analytic review of intervention components. *Depression Anxiety* (2019) 36(3):213–24. doi: 10.1002/da.22860
77. Hempel S, Miles JN, Booth MJ, Wang Z, Morton SC, Shekelle PG. Risk of bias: a simulation study of power to detect study-level moderator effects in meta-analysis. *Syst. Rev* (2013) 2(1):107. doi: 10.1186/2046-4053-2-107
78. Bradley R, Greene J, Russ E, Dutra L, Westen D. A multidimensional meta-analysis of psychotherapy for PTSD. *Am J Psychiatry* (2005) 162(2):214–27. doi: 10.1176/appi.ajp.162.2.214
79. Bryant RA, Kenny L, Joscelyne A, Rawson N, Maccallum F, Cahill C, et al. Treating prolonged grief disorder: a randomized clinical trial. *JAMA Psychiatry* (2014) 71(12):1332–9. doi: 10.1001/jamapsychiatry.2014.1600
80. Lampe LA. Internet-based therapy: too good to be true? *Aust New Z J Psychiatry* (2011) 45(4):342–3. doi: 10.3109/00048674.2011.560138
81. Kalantari M, Yule W, Dyregrov A, Neshatdoost H, Ahmadi S. Efficacy of writing for recovery on traumatic grief symptoms of Afghani refugee bereaved adolescents: A randomized control trial. *OMEGA-J. Death Dying* (2012) 65(2):139–50. doi: 10.2190/OM.65.2.d
82. Lichtenthal WG, Cruess DG. Effects of directed written disclosure on grief and distress symptoms among bereaved individuals. *Death Stud* (2010) 34(6):475–99. doi: 10.1080/07481187.2010.483332
83. Sloan DM, Marx BP, Lee DJ, Resick PA. A brief exposure-based treatment vs cognitive processing therapy for posttraumatic stress disorder: A randomized noninferiority clinical trial. *JAMA Psychiatry* (2018) 75(3):233–9. doi: 10.1001/jamapsychiatry.2017.4249

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Copyright © 2020 Wagner, Rosenberg, Hofmann and Maass. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.



Association Between Prolonged Grief and Attitudes Toward Reconciliation in Bereaved Survivors of the Khmer Rouge Regime in Cambodia

Nadine Stammel^{1,2*}, Louisa Heinzl¹, Carina Heeke¹, Maria Böttche^{1,2} and Christine Knaevelsrud¹

¹ Department of Clinical Psychological Intervention, Freie Universität Berlin, Berlin, Germany, ² Research Department, CENTER ÜBERLEBEN, Berlin, Germany

OPEN ACCESS

Edited by:

Geert E. Smid,
ARQ National Psychotrauma Centre,
Netherlands

Reviewed by:

Maarten C. Eisma,
University of Groningen, Netherlands
Maja O'Connor,
Aarhus University, Denmark

*Correspondence:

Nadine Stammel
nadine.stammel@fu-berlin.de

Specialty section:

This article was submitted to
Mood and Anxiety Disorders,
a section of the journal
Frontiers in Psychiatry

Received: 25 February 2020

Accepted: 19 June 2020

Published: 10 July 2020

Citation:

Stammel N, Heinzl L, Heeke C,
Böttche M and Knaevelsrud C (2020)
Association Between Prolonged Grief
and Attitudes Toward Reconciliation in
Bereaved Survivors of the Khmer
Rouge Regime in Cambodia.
Front. Psychiatry 11:644.
doi: 10.3389/fpsy.2020.00644

Background: During the Khmer Rouge regime in Cambodia, about a quarter of the population died, resulting in many individuals losing close relatives. Still today, many individuals are suffering from the psychological consequences of these losses, which might also affect the process of reconciliation within the Cambodian society. The aim of this paper is therefore to investigate the association between symptoms of prolonged grief and attitudes toward reconciliation.

Methods: A sample of 775 survivors of the Khmer Rouge regime who lost relatives during the conflict were interviewed about their losses and traumatic events, prolonged grief (PG; Complicated Grief Assessment Self-Report, CGA-SR), posttraumatic stress disorder (PTSD Checklist - Civilian Version) and attitudes toward reconciliation (Readiness to Reconcile Inventory, RRI).

Results: A higher symptom severity of PG was significantly associated with readiness to reconcile even when controlling for other relevant variables ($\beta = -0.22$; $p < .001$). Persons who met caseness criteria for PG were significantly less ready to reconcile, $t(773) = 5.47$, $p < .001$, than persons who did not meet caseness for PG.

Conclusion: PG seems to be a relevant mental health correlate of attitudes toward reconciliation. The results of the current study underline the importance of also considering PG with regard to the reconciliation process in Cambodia and possibly also in other post-conflict regions.

Keywords: Cambodia, bereavement, grief, reconciliation, forgiveness, post-conflict, violent death, genocide

INTRODUCTION

During the Khmer Rouge (KR) regime from 1975 to 1979, millions of Cambodians suffered grave human rights violations, including mass executions, forced labor, torture and starvation. The KR regime caused the death of about 1.7 million Cambodians—approximately one quarter of the Cambodian population at that time (1)—leaving behind a substantial number of bereaved and traumatized individuals.

Decades after the end of the KR regime, many Cambodians continue to suffer from mental health problems stemming from this time. About 30 years after the end of the KR regime, approximately 14% of KR survivors were suffering from posttraumatic stress disorder (PTSD; 2). A national representative survey conducted in 2011 found rates of 27.4% for anxiety and 16.7% for depression in the general population (3). Rates of psychiatric morbidity among survivors of the KR regime are even higher (4). Even though many survivors of war and violent conflict are confronted with the death of loved ones, the psychological consequences of these losses have only recently been addressed in war-torn societies. While the majority of survivors adjust to their loss after a certain period of grieving and continue with their lives, others show severe psychological symptoms connected to their grief over long periods of time (5). Over the last decades of research, this pathological form of grief has been given a variety of labels (e.g., “complicated grief (disorder)”, “disturbed grief”, and “pathological grief”), and was finally included as “prolonged grief disorder” (PGD) in the 11th edition of the International Classification of Diseases (ICD-11; 6) in a new category for stress-related disorders. In the Diagnostic and Statistical Manual of Mental Disorders, fifth edition (DSM-5; 7), PGD was not formally included as a distinct clinical entity and was instead included as a condition for further research, termed “persistent complex bereavement disorder”. The ICD-11 and DSM-5 definitions of the disorder differ from each other in terms of symptoms and symptom duration requirement (6 vs. 12 months, respectively). This represents a significant challenge, as the use of diverse scales to assess PG has been shown to result in different prevalence rates (8). It may thus be the case that research findings obtained using one set of criteria may not apply to other definitions of PG (9). The current study is based on the criteria provided by Prigerson and colleagues (10). These authors originally introduced the term “prolonged grief disorder”, which also represents the most widely used term in the literature. The applied criteria were the most contemporary criteria at the time of our study. The defining feature of all sets of criteria for PGD is separation distress (i.e., persistent longing or yearning for the lost person) combined with cognitive, emotional and behavioral symptoms resulting in functional impairment. However, as the applied criteria differ slightly from the current PGD ICD-11 definition, we will use the term “prolonged grief (PG)” when describing the results of our study to avoid confusion with the current ICD-11 definition.

While around 10% of bereaved persons who have lost a significant other by natural death are estimated to develop PGD (11), the prevalence of PGD is usually higher in war-affected populations. Almost 35% of a sample of young adults who had lived through the war in Kosovo met criteria for PGD (12). In a sample in Rwanda, 8% met the criteria more than ten years after the loss of a loved person (5). One reason for the higher prevalence of PGD after violent conflicts might be the high numbers of violent and unnatural deaths that people have to face during war. A violent cause of death (i.e., death by homicide, suicide, or accident) has been recognized to have a higher association with PGD than natural deaths (5, 13, 14). A study

investigating the construct of PGD in Cambodia also confirmed its validity (15).

The consideration of cultural aspects of the grieving process, such as its culturally accepted length, has been emphasized in the new ICD-11 criteria set (6). Moreover, in Cambodia, a consideration of religious and spiritual beliefs about death and grieving also seems to be of special relevance. Cambodian culture and social norms are influenced by Buddhism and folk religion, in which spiritual beings such as ghosts play an essential role (16). Many Cambodians believe that when somebody dies due to unnatural causes, this person cannot easily reincarnate, and their spirit instead remains in an “in-between” state (17, 18). Furthermore, if the person had thoughts of hate or was yearning for another person in the moment of death, this is thought to further make it difficult for that person to be reborn (16, 18). Another important aspect of mourning in Cambodia is Buddhist rituals, such as cremation and paying respect to the deceased with offerings at the temple, to ensure that the deceased can reincarnate. These rituals could not be carried out during the KR regime (16).

Due to the interplay of the historic circumstances of the genocide and cultural beliefs, it may be expected that Cambodians show a greater vulnerability to be affected by grief-related mental health problems (16). Findings indicate that the symptoms shown by Cambodian refugees are often shaped by their beliefs about the state of the person they have lost: Many individuals had frequent nightmares in which they saw their relatives who had died during the KR regime, and interpreted this as a sign that their loved ones had still not reincarnated (18, 19). Moreover, visual auras (as a symptom of migraine) were interpreted as evil spirits, which patients experienced as highly impairing and worrying (20). These cultural ideas and interpretations of specific symptoms may also shift a certain responsibility onto the bereaved persons, because they feel it is their duty to help with their loved one’s reincarnation by performing religious rituals for the deceased (16). Not being able to “make merit” for one’s ancestors can thus have negative consequences for one’s own next life (21) or one’s current life, if ancestors haunt survivors in the form of spirits (20). This can lead people to frequently perform rituals for the deceased and to be highly concerned about the lost person (17, 22), potentially rendering it more difficult to come to terms with the death and to focus on one’s own life again, even after several years.

Even after the regime of the KR ended, Cambodia remained strongly affected both by ongoing political instability and the long-term consequences of the KR (21, 23). In many villages in Cambodia, former victims and perpetrators continued to live side by side. Often, people did not know whether or what kind of crimes had been committed by former KR members in their communities, and feelings of anger and revenge still lingered under the surface (24). Although in Cambodian culture, anger is generally not shown in public (25), many survivors of the KR regime reported feelings of revenge and hatred toward the former KR (2, 26). For a long time, the atrocities committed by the KR were not publicly addressed in Cambodia. Only about 30 years after the end of the Khmer Rouge regime were the Extraordinary Chambers in the Courts of Cambodia (ECCC), also known as Khmer Rouge tribunal, established to investigate

the atrocities committed by the KR. While the primary purpose of the ECCC is to seek retributive justice, it was also expected to contribute to national reconciliation (27).

The awareness and importance of reconciliation in war-torn societies has grown in the past decades. Reconciliation between former opponents has become a key concept for sustainable peace activities in post-conflict societies. Although there is not yet a consensus definition of post-conflict reconciliation, most authors agree that reconciliation is a long-term process aiming at (re)building relationships between formerly hostile groups (e.g., 28, 29). Reconciliation also involves letting go of hatred and desire for revenge (29) as well as changing stereotypic beliefs about both the adversary group and one's own group (30, 31). This includes being able to differentiate between members of the adversary group, i.e., perceiving them as individuals and human beings, rather than generalizing them as "evil". It is assumed that coping with conflict-related traumatic events can have positive benefits for victims' openness to reconciliation with former perpetrators, for example through legal-political measures such as war tribunals or truth commissions (32). This is partly supported by empirical research showing that persons with a poorer mental health outcome are generally less open to reconciliation and forgiveness (33–35). Studies have mostly focused on symptoms of PTSD (PTSD; 33, 34) when studying attitudes toward reconciliation or forgiveness and mental health, but to a lesser extent have also investigated other mental health outcomes such as depression and anxiety (5, 36). To our knowledge, the association of PGD with attitudes toward reconciliation or forgiveness in post-conflict societies has not yet been empirically investigated. However, it seems plausible that persons who are still suffering from conflict-related loss(es) will be less open to reconcile with those whom they hold responsible for the death of their loved ones.

Generally, evidence on risk and facilitating factors of reconciliation in post-conflict settings is very limited, as study findings are inconsistent and longitudinal data are still missing. Attitudes toward reconciliation and forgiveness do not seem to be directly related to the victims' traumatic experiences, such as the number of traumatic events experienced (33, 37) but rather seem to be related to mental health consequences of traumatic events. However, research findings are not consistent. Furthermore, research on sociodemographic characteristics such as gender and educational level has yielded inconsistent results. While in some studies, a higher level of education was related to more openness to reconciliation (38, 39), other studies found the opposite to be the case (34, 40). Findings also differ with respect to whether one gender is associated with being more or less open to reconciliation (36, 41, 42).

In summary, empirical evidence on reconciliation and mental health, characteristics of PG as well as cultural aspects about loss and the grieving process in Cambodian society suggest an interplay between PG and the process of reconciliation in Cambodia. The main aim of this paper was therefore to investigate associations between attitudes toward reconciliation and PG symptoms in bereaved survivors of the KR regime. In

particular, we wanted to find out (1) whether bereaved victims of the KR with PG, who lost at least one family member during the KR regime, differ in terms of their attitudes toward reconciliation from those without PG; (2) whether readiness to reconcile is related to PG symptoms and loss- and trauma-specific variables as well as sociodemographic characteristics; and (3) whether PG symptom severity is associated with readiness to reconcile beyond PTSD symptom severity as well as loss- and trauma-specific and sociodemographic variables.

Based on research on attitudes toward reconciliation in post-conflict settings and other trauma-related mental health problems described above, we expected that persons with PG would be less ready to reconcile than those without PG and that PG symptom severity would be negatively associated with readiness to reconcile, also when controlling for other relevant variables (number of lost close family members, trauma exposure, PTSD symptom severity, and sociodemographic characteristics).

METHODS

Sample and Procedure

Participants were recruited between October 2008 and May 2009 through a convenience sampling approach in 19 out of 24 provinces in Cambodia. Potential participants were either contacted with the help of local non-governmental organizations or directly in their villages. Inclusion criteria for participants were date of birth before 1975 and considering oneself as a victim of the KR regime. For this analysis, we only included participants who had lost at least one (close or distant) family member during the KR regime. Of 1593 persons who were contacted, 796 did not wish to participate and a further 22 persons had to be excluded due to a lack of important data (missing or ambiguous information regarding the loss to which participants' answers referred). The final sample thus comprised 775 individuals. The same data set was used in a previous publication on PG in Cambodia (43); the present analysis is thus secondary in nature.

After participants had been fully informed about the study and the voluntary nature of participation, written informed consent was obtained. Given the high illiteracy rate, the self-rating questionnaires were administered in the form of face-to-face interviews. The interviews were conducted in the local Khmer language by eight local Cambodian interviewers who held a B.A. degree in psychology and had previously participated in a two-week training course on the assessment of relevant concepts and use of the questionnaire measures. To maintain the quality of the interviews, the interviewers were supervised on a regular basis. Participation in the study was voluntary; no financial compensation was offered, but participants received a small gift consisting of a traditional Cambodian scarf "Krama" and a piece of soap (worth the equivalent of USD 1.5) after the end of the interview. Psychological counseling by telephone was offered to all participants in case of need. The study protocol was approved by the University of Constance Review Board and the Cambodian Ministry of the Interior.

Measures

Symptoms of PGD were assessed using the Complicated Grief Assessment Self-Report (10). If participants reported the loss of more than one family member during the KR regime, they were asked to specify whose death was most difficult to cope with and to relate their answers to this person. The CGA-SR is a self-report questionnaire that assesses the frequency of separation distress and whether this separation distress disrupts one's everyday life (Criterion A). Criterion B contains eight cognitive, behavioral and emotional symptoms (difficulties accepting the death, trusting people, moving on, feeling bitter over the death, feeling numb or difficulty connecting with others, feeling that life is empty or meaningless, as well as future holds no meaning or purpose and feeling on edge, jumpy or easily startled), measured on 5-point Likert scales from 1 to 5. Criterion C contains the question whether the symptoms lead to impairment in functional areas, and criterion D assesses whether the symptoms have already lasted for at least six months. Caseness for PG was indicated when a person fulfilled at least one of the two criterion A symptoms, as well as the C and D criteria, and stated being affected by at least four of the eight symptoms in criterion B to an extent of at least four points. Symptom severity of PG was measured by summarizing the values of symptom items with Likert scales. Higher scores indicate a higher symptom severity, with a minimum score of 9 and a maximum score of 45. Internal consistency of the CGA-SR in the current study was $\alpha = .82$.

Readiness to reconcile was measured using the Readiness to Reconcile Inventory (RRI) (44). The RRI is a 13-item self-rating instrument assessing individual openness to reconcile with members of the former adversary group, thereby focusing exclusively on the perspective of the victims. In this study, the term "former adversary group" was substituted with "former Khmer Rouge". The RRI consists (41) of three subscales: 1. openness to positive relationships with former members of the KR (six items; example item: "I would like us and the former Khmer Rouge to live in peace together"), 2. absence of feelings of revenge toward former members of the KR (five items; example item: "I have feelings of revenge when I think of the former Khmer Rouge"), and 3. differentiation ability (two items; example item: "The majority of the former Khmer Rouge are good people"). The latter describes the ability to differentiate between individual perpetrators and the group of former members of the KR. All items of subscale 2 (absence of revenge) are negatively coded. Participants are asked to indicate their level of agreement with each item on a 5-point Likert scale from 1 (*strongly disagree*) to 5 (*strongly agree*), with higher scores indicating greater readiness to reconcile. The factor structure and the internal consistency of the RRI and its pilot version were confirmed in several studies with different samples (37, 44, 45). Cronbach's alpha for the RRI total scale in the current study was $\alpha = .87$. For the subscales, Cronbach's alphas were as follows: openness to interactions, $\alpha = .85$; absence of revenge, $\alpha = .87$; differentiation ability, $\alpha = .72$.

Symptoms of PTSD were measured using the PTSD Checklist-Civilian Version (PCL-C; 46), a widely used 17-item standardized self-report rating scale that corresponds to the DSM-IV diagnostic criteria for PTSD. Participants were asked

to rate each item on a 5-point Likert scale from 1 (*not at all*) to 5 (*extremely*) referring to the past month. The sum score was used as an indicator of PTSD symptom severity. The Cambodian version of the PCL-C has shown excellent reliability in earlier studies (2, 38). In the present study, the internal consistency was $\alpha = 0.88$.

Potentially traumatic events were assessed using an adjusted checklist based on the first part of the Harvard Trauma Questionnaire (47) and the Posttraumatic Diagnostic Scale (48). Additionally, seven traumatic events specific to the Khmer Rouge regime (e.g., forced labor and forced marriage) were included. In total, 29 types of traumatic experience were assessed, creating an index of overall trauma exposure. Event types were rated as potentially traumatic if participants indicated having experienced or witnessed them. If the event included the death of another person, also having heard about such an event was rated as potentially traumatic.

In addition to traumatic events, loss-related questions were administered, including how many losses within one's close family (i.e., spouse, parent, child, and sibling) as well as within one's distant family (i.e., grandparent, grandchild, uncle/aunt, nephew/niece, cousin, and in-law) were experienced during the KR regime.

All questionnaires were translated from English into Khmer by bilingual psychologists, and subsequently back-translated by interpreters who were unfamiliar with the original English version in order to verify their correspondence. Discrepancies between the original and back-translated versions were discussed, and wherever necessary, the Khmer version was adjusted. Recommendations for cross-cultural adaptation were considered throughout the translation process (e.g., 49).

Statistical Analysis

A first screening of the data showed that only a small amount of data was missing (0.29%). Missing values within the CGA-SR, the PCL-C, and the RRI were replaced using the expectation-maximization method (EM; 50). T-tests were calculated to determine whether people who met caseness criteria for PG according to the CGA-SR and those who did not differed in their attitudes toward reconciliation. This was done for the RRI total scale as well as for the RRI subscales (openness to interactions, absence of revenge, differentiation ability). Correlation analysis was performed to determine associations of the RRI and its subscales with the investigated variables. Hierarchical stepwise multiple linear regression was applied to investigate whether PG symptom severity (using the sum score) can explain variance in the readiness to reconcile beyond sociodemographic, loss- and trauma-specific variables, as well as PTSD symptom severity. The demographic variables gender, age, and education were entered in the first step of the regression analysis. The number of traumatic event types and the number of losses within the close family were entered in the second step, PTSD symptom severity was entered in the third step, and PG symptom severity was entered in the final step. To better understand how PG symptoms and reconciliation are associated, the regression analysis was also repeated for the subscales of the RRI as outcome variables. Relevant assumptions for multiple regression analysis were tested (linearity, multicollinearity,

independency of residuals, and homoscedasticity) and did not reveal any violation. The analysis was conducted using SPSS 25.

RESULTS

Sociodemographic Characteristics

Almost two thirds of the sample were female, and the mean age was 56 years. On average, participants had spent four years in formal education. Most of the participants identified themselves as Khmer and Buddhists. About two thirds of the sample were married and about 30% were widowed. The detailed sociodemographic characteristics of the sample are shown in **Table 1**.

Loss- and Trauma-Specific Characteristics, PG, and Readiness for Reconciliation

Participants stated that they had lost on average 6.4 relatives during the KR regime, of whom on average 3.4 were part of the close family and 3.2 were part of the distant family. When participants were asked which loss was the most difficult to cope with, and upon which their answers on the CGA-SR were thus based, 14.6% ($n = 113$) reported the loss of a spouse, 5.3% ($n = 41$) the loss of a child, 34.1% ($n = 264$) the loss of a parent; 28.6% ($n = 222$) the loss of a sibling, and 17.4% ($n = 135$) the loss of a distant relative.

Participants reported on average 14 different traumatic event types. The five most frequently reported traumatic event types were lack of food or water (95.6%; $n = 741$), forced labor (94.1%; $n = 729$), forced separation from family members (81.1%; $n = 629$), deportation/forced displacement (79.9%; $n = 619$), and life-threatening illnesses (70.5%; $n = 547$).

Of the whole sample, 111 participants (14.3%) met caseness criteria for PG as measured by the CGA-SR. The mean sum score of the CGA-SR across the whole sample was 20.99. The mean sum total score of the RRI was 32.28. The mean sum scores of the RRI subscales were 13.00 for openness to interactions, 15.68 for absence of revenge and 3.60 for differentiation ability, respectively.

The detailed descriptive data on losses, traumatic event types, PTSD, PG, and readiness for reconciliation are shown in **Table 2**.

Differences Between Participants With and Without PG Regarding Readiness for Reconciliation

To ascertain whether people who meet caseness criteria for PG are less ready to reconcile than persons who do not meet caseness criteria, t-tests for the RRI total score as well as for the RRI subscales were performed. For this purpose, the sample was divided into persons who fulfilled the criteria for PG according to the CGA-SR and those who did not.

As shown in **Table 3**, participants meeting caseness criteria for PG were significantly less ready to reconcile than participants who did not meet caseness for PG according to the CGA-SR. This was the case for the RRI total score as well as for all RRI subscales. These differences had a medium effect size for the RRI

TABLE 1 | Sociodemographic characteristics.

Characteristic	N = 775
Sex = female, n (%)	498 (64.3)
Age, M (SD)	56.7 (10.3)
Years of formal education, M (SD)	4.1 (3.6)
Self-identity	
Khmer, n (%)	678 (87.5)
Cham, n (%)	45 (5.8)
Chinese, n (%)	15 (1.9)
Other, n (%)	37 (4.8)
Religion	
Buddhist, n (%)	702 (90.6)
Muslim, n (%)	58 (7.5)
Other, n (%)	15 (1.9)
Marital status	
Married, n (%)	497 (64.1)
Widowed, n (%)	231 (29.8)
Divorced, n (%)	25 (3.2)
Single, n (%)	11 (1.4)
Other, n (%)	11 (1.4)

total scale as well as for the subscale absence of revenge. For openness to interactions as well as for differentiation ability, the effect sizes of the differences were small.

Associations of Readiness to Reconcile With PG and Sociodemographic and Loss- and Trauma-Specific Variables

To analyze associations of PG with other variables, the sum score of the CGA-SR was applied. Correlations of PG symptom severity, readiness to reconcile and the other variables included in this analysis can be found in **Table 4**. To investigate whether PG symptom severity can explain variance in the readiness to reconcile beyond PTSD symptom severity, sociodemographic, and loss- and trauma-specific variables, a stepwise hierarchical regression analysis was performed with the RRI total score as dependent variable. The same analysis was performed with the three RRI subscales, respectively. Sociodemographic characteristics (gender, age, education) were entered into the analysis first, followed

TABLE 2 | Descriptive data on losses, traumatic events, PG symptom severity, PTSD symptom severity and readiness to reconcile.

	M	SD	Answer Range	
			Min	Max
Number of losses (in total)	6.40	6.79	1	59
Number of losses (close family)	3.35	3.37	0	27
Number of losses (distant family)	3.16	6.18	0	50
Number of traumatic event types	14.01	3.84	3	25
PG symptom severity ¹	20.99	7.37	9	45
PTSD symptom severity ²	30.10	11.35	3	75
RRI total score	32.28	10.94	13	62
RRI Openness to Interactions	13.00	5.94	6	30
RRI Absence of Revenge	15.68	5.99	5	25
RRI Differentiation Ability	3.60	1.64	2	9

PG, Prolonged Grief; PTSD, Posttraumatic Stress Disorder; RRI, Readiness to Reconcile Inventory.

¹Complicated Grief Assessment Self Report (CGA-SR); ²PTSD-Checklist Civilian Version (PCL-C).

TABLE 3 | T-tests with p-values, confidence intervals, and Cohen's d.

Outcome	PG ¹ (n = 111)		No PG ¹ (n = 664)		Average difference (95% CI)	t (df)	d
	M	SD	M	SD			
Readiness to Reconcile ²	27.12	10.34	33.15	10.81	6.03 (3.86–8.19)	t (773) = 5.47***	0.57
Openness to Interactions ²	10.98	5.11	13.33	6.01	2.35 (1.29–3.41)	t (165.14) = 4.37***	0.42
Absence of Revenge ²	12.91	6.31	16.15	5.81	3.23 (2.05–4.42)	t (773) = 5.35***	0.53
Differentiation Ability ²	3.23	1.60	3.67	1.64	0.44 (0.11–0.77)	t (773) = 2.63**	0.27

PG, prolonged grief; CI, confidence interval; d, Cohen's d; ¹ as measured with CGA-SR; ² as measured with the RRI; **p < .01, ***p < .001 (two-tailed p-values).

TABLE 4 | Pearson correlation coefficients.

	Readiness to Reconcile ¹	Openness to Interactions ²	Absence of Revenge ²	Differentiation Ability ²	Male gender	Age	Education (years)	Number of losses (close family)	Number of traumatic event types	PTSD symptom severity ³	PGD symptom severity ⁴
Readiness to Reconcile ¹	1										
Openness to Interactions ²	0.85***	1									
Absence of Revenge ²	0.82***	0.42***	1								
Differentiation Ability ²	0.59***	0.52***	0.29***	1							
Male gender	0.26***	0.34***	0.09*	0.16***	1						
Age	0.01	−0.12**	0.13***	0.02	−0.01	1					
Education (in years)	0.17***	0.25***	0.03	0.11**	0.40***	−0.22***	1				
Number of losses (close family)	−0.12**	−0.06	−0.13***	−0.9*	−0.08*	0.18***	−0.05	1			
Number of different traumatic events	−0.03	0.11**	−0.17***	0.03	0.09*	−0.07	0.09*	0.13***	1		
PTSD symptom severity ³	−0.31***	−0.22***	−0.33***	−0.10**	−0.25***	−0.05	−0.12**	0.12**	0.18***	1	
PG symptom severity ⁴	−0.34***	−0.23***	−0.35***	−0.17***	−0.23***	−0.02	−0.10**	0.18***	0.11**	0.61***	1

PTSD, Posttraumatic stress disorder; PG, Prolonged grief disorder; ¹RRI; ²Subfactor of the RRI; ³PCL-C; ⁴ CGA-SR; *p < .05, **p < .01, ***p < .001 (two-tailed p-values).

by loss- and trauma-specific variables (number of losses within the close family, number of traumatic event types) in the second step, PTSD symptom severity in the third step, and PG symptom severity in the fourth step. Detailed data of the stepwise regression analysis of the RRI total score are depicted in **Table 5**.

RRI Total Score

Sociodemographic characteristics explained 7.4% of the variance in readiness to reconcile. Of the investigated sociodemographic variables, gender and education were significantly associated with readiness to reconcile, with males as well as more educated persons showing higher scores for readiness to reconcile. The inclusion of loss- and trauma-related variables in step 2 explained 1.3% of additional variance. While there was no significant association between trauma exposure and the RRI total score, the number of losses within the close family was associated with readiness to reconcile in the second step (more losses were associated with less readiness to reconcile), but this association lost its significance when the PG symptom score was entered in the fourth step. PTSD symptom severity was significantly negatively associated with readiness to reconcile. The inclusion of PG symptom severity in the final step explained significantly more variance than did the model without PG symptom severity (2.9% of incremental

variance), with a regression coefficient of $\beta = -.22$, $p < .001$. Thus, PG symptom severity was negatively associated with readiness to reconcile even after controlling for the other mentioned variables. The final model explained 17.7% of the variance in the RRI total score.

RRI Subscales

PG symptom severity was significantly negatively associated with all RRI subscales ($\beta = -.12$, $p < .01$; $\beta = -.24$, $p < .001$; $\beta = -.14$, $p < .01$). PTSD symptom severity, which was entered in step 3, accounted for significant additional variance in RRI-openness to interactions ($\Delta R^2 = 0.02$, $p < .001$) as well as in RRI-absence of revenge ($\Delta R^2 = 0.07$, $p < .001$) but not in RRI-differentiation ability ($\Delta R^2 = < 0.01$, $p = .12$). The inclusion of PG in the final step explained significantly more variance in all subscales than the models without PG symptom severity (0.9%, 3.4%, and 1.3% of additional variance), respectively. The final regression models explained between 5.4% and 17.9% of the variance in the subscales. RRI-openness to interactions was significantly associated with male gender, younger age and higher educational level as well as with a higher trauma exposure and lower PTSD and PG scores. RRI-absence of revenge was significantly related to older age, fewer losses within the close family, and less trauma exposure, as well as with lower PTSD

TABLE 5 | Hierarchical multiple regression analysis predicting readiness to reconcile (N = 774).

	Variables	B (SE B)	β	R ²	ΔR^2
Step 1	Male gender	5.21 (0.87)	0.23***	0.074	0.074***
	Age	0.03 (0.04)	0.03		
	Education (years)	0.26 (0.12)	0.08*		
Step 2	Male gender	5.09 (0.87)	0.22***	0.087	0.013**
	Age	0.05 (0.04)	0.05		
	Education (years)	0.27 (0.12)	0.09*		
	No. of losses (close family)	-0.32 (0.12)	-0.10**		
	No. of traumatic event types	-0.13 (0.10)	-0.05		
Step 3	Male gender	3.65 (0.86)	0.16***	0.144	0.057***
	Age	0.03 (0.04)	0.03		
	Education (years)	0.24 (0.11)	0.08*		
	No. of losses (close family)	-0.25 (0.11)	-0.08*		
	No. of traumatic event types	0.01 (0.10)	0.00		
	PTSD symptom severity ¹	-0.24 (0.03)	-0.25***		
Step 4	Male gender	3.33 (0.85)	0.15***	0.173	0.029***
	Age	0.03 (0.04)	0.03		
	Education (years)	0.23 (0.11)	0.08*		
	No. of losses (close family)	-0.17 (0.11)	-0.05		
	No. of traumatic event types	0.00 (0.10)	0.00		
	PTSD symptom severity ¹	-0.12 (0.04)	-0.13**		
	PG symptom severity ²	-0.32 (0.06)	-0.22***		

PTSD, Posttraumatic stress disorder; PG, prolonged grief; ¹PCL-C; ²CGA-SR; * $p < .05$, ** $p < .01$, *** $p < .001$ (two-tailed p -values).

and PG scores. RRI-differentiation ability was significantly associated with lower PG score and male gender, but not with the PTSD symptom score. The three stepwise regression analyses of the RRI subscales can be found in the supplementary material (Supplementary Table 1).

DISCUSSION

The present study examined associations of PG with attitudes toward reconciliation 30 years post loss in a sample of bereaved survivors of the KR regime who had lost at least one family member during the KR regime. Most participants had lost more than one relative: On average, the loss of approximately six family members was reported, including about three close family members. Even three decades after experiencing the loss(es), around 14% of the participants met criteria for PG according to the CGA-SR.

The results confirm our hypothesis that bereaved victims who met caseness criteria for PG were significantly less ready to reconcile compared to those who did not fulfill these criteria. This pattern was also found for all three subscales of the RRI. Thus, persons with probable PG were significantly less open to interacting with former members of the KR, had more feelings of revenge toward former members of the KR, and were less able to differentiate between individual perpetrators and members of the KR as a whole. As hypothesized, PG symptom severity was negatively associated with readiness to reconcile, also when controlling for PTSD, loss- and trauma-specific variables, as well as sociodemographic variables that were also expected to

be associated with attitudes toward reconciliation. PG symptom severity explained around 3% of unique variance in readiness to reconcile and between 0.9% and 3.4% of additional variance in the RRI subscales. PG symptom severity was more strongly related to readiness to reconcile than any other analyzed variable.

Even though PG symptom severity explained a rather small amount of additional variance in readiness to reconcile, it should be considered that PTSD symptom severity was controlled for in a prior step, and that PTSD has so far been investigated as a main mental health correlate of attitudes toward reconciliation. As PTSD and PG correlate highly in the context of violent loss (51), it is not surprising that PG symptoms did not explain a large amount of additional variance. Moreover, in the current study, the association between PG and PTSD symptoms was rather strong ($r = .59$). The fact that in our study, PG symptom severity was the strongest predictor of readiness to reconcile, beyond PTSD as well as trauma- and loss-related variables, speaks for the importance of PG in this regard. Our results thus suggest that PG should be considered as a mental health correlate of reconciliation besides PTSD and other mental health outcomes.

Interestingly, the number of losses within the close family was significantly related to readiness to reconcile, but this association lost its significance when PG symptom severity was entered into the regression analysis. Only in one subscale of the RRI, namely, absence of revenge, was the number of losses within the close family still significantly related in the final regression model, albeit to a smaller extent than PG symptom severity. This implies that it is not the losses *per se*, but rather the psychological reaction to the losses which seems to be related to attitudes toward reconciliation with the former KR. Nevertheless, it is possible that qualitative aspects of losses are particularly linked to attitudes toward reconciliation rather than quantitative aspects such as the number of losses within the close family. Trauma exposure was not related to the total score of the RRI inventory, but was associated with the RRI subscales openness to interactions and absence of revenge. As expected, a higher trauma exposure was associated with more feelings of revenge. Surprisingly, this association was reversed for openness to interactions, with a higher trauma exposure being related to more openness to interactions. Most previous studies conducted in post-conflict settings did not find a relationship between trauma exposure and attitudes toward reconciliation (33, 35, 52), which is in line with the results of the current study. However, our results also suggest a more complex interaction between trauma exposure and different aspects of reconciliation, highlighting the need for more research in this regard. As in previous studies, higher levels of PTSD symptoms were associated with less readiness to reconcile. This pattern was also found for the RRI subscales, with the exception of RRI-differentiation ability, where no significant association was found. Among the investigated sociodemographic variables, we found that males and participants with a higher level of education were more open to reconciliation, although there were some variations in the subscales.

The study results imply that PG could play a similar role to PTSD as a mental health-related correlate of reconciliation attitudes

and that PG should be considered with regard to the reconciliation process in Cambodia and possibly also in other post-conflict settings. These results are in line with previous research showing that persons with poorer mental health outcomes were generally less ready to reconcile (33, 34, 35). However, previous studies primarily focused on PTSD when investigating attitudes toward reconciliation and mental health. To our knowledge, no other empirical study has yet focused on the relationship between PG and readiness to reconcile or forgiveness in post-conflict settings. Perhaps unsurprisingly, for persons still struggling with conflict-related loss of a loved one, being open to reconciliation with the former perpetrators is very difficult. It might be that persons who are still suffering from the conflict-related loss do not feel entitled to be open to reconciliation or feel that they would betray the deceased if they did reconcile. Especially when thoughts about the deceased and grieving occur on an everyday basis, as is common in PG, moving beyond negative feelings toward those who are held responsible for the death of their loved ones is highly challenging. Persistent feelings of anger about the loss and a reluctance to socialize are possible aspects of PGD (53). Therefore, symptoms of PG might reduce willingness to interact with former perpetrators and increase negative feelings toward them. Instead, the symptoms might strengthen resentment against perpetrators.

A further binding element of PG and reconciliation after conflict might be the inability to find sense and meaning in what has happened. In particular, people bereaved by homicide have difficulties finding any sense in their bereavement (54). Being able to make sense of a loss, in terms of understanding the reasons for the death and being able to place the experience within one's own worldview and belief system, might be an important part of coping with a death in general (55, 56). In the case of genocide in particular, it might be extremely difficult for the bereaved to understand why a loved one was taken from them and to find any sense in it. The fact that a national intention to process and work through the KR period was barely existent in the first decades after the genocide (57–59) presumably adds to individual difficulties to understand how the genocide could have happened. Field and Chhim (38) also found that within a Cambodian sample, those who were not able to find something beneficial in the processing of the experience of loss had a higher wish for revenge.

Besides this aspect, various cognitive components, such as negative cognitions about the self and the future and catastrophic misinterpretations (60) have been shown to be correlates of PGD (61, 62). Boelen, van Denderen (62) showed that not only PTSD and PGD, but also feelings of revenge and anger were linked to several negative cognitions. Moreover, rumination has also been shown to be associated with maladaptive grief reactions and could further hinder the development of a conciliatory attitude (63, 64). A longitudinal study found that rumination about injustice after a loss predicted higher symptom levels of complicated grief (65). It can thus be assumed that the harrowing experience of injustice in the case of murder or even genocide shakes one's own belief system, triggers negative cognitions and coping styles, and makes adaptive reactions generally more difficult. A study by Tay, Rees (66) also

emphasized the link between the sense of injustice and severe grief reactions following human rights violations.

Another important aspect to be considered when looking at attitudes toward reconciliation and PG is the cultural context in Cambodia, especially religious and spiritual beliefs about death and mourning. Research suggests that religious beliefs have the potential to be both a risk (67) and a protective factor (5) for PGD, presumably depending on the ways of coping they offer to the bereaved. During the KR regime, many deaths were violent and unnatural, religion was forbidden and traditional ceremonies after death were not possible (16). For many Cambodians, these aspects may be reasons to believe that a family member was not reincarnated (17). The importance of the lack of Buddhist ceremonies for survivors of the KR regime also becomes apparent in a culturally adapted therapy for KR survivors in which a Buddhist ritual was added to the regular therapy. The ceremony, which was led by monks, was conducted at a mass grave site in Cambodia with the goal of helping the ancestors of the survivors to find a peaceful place (68). Bereaved persons might thus see the KR as responsible for more than the death of a loved one, namely, for having put the deceased in a state in which he or she cannot reincarnate (16). Some persons feel haunted by their relatives, which further connects grief-related symptoms of rumination or nightmares to the deeds of the KR (17–19). Therefore, coming to terms with what has happened and changing the attitude toward the perpetrators might be further complicated by religious and spiritual beliefs about death, causing people to worry about their loved ones even decades after they have died.

Several limitations of the present study should be considered when interpreting the results. First of all, the sample was not recruited through random sampling. Despite the large sample size, the results may thus not be generalizable to all victims of the Khmer Rouge regime. Our measure to assess PG, the CGA-SR, differs from the newly released classification of PGD in the ICD-11 and from the DSM-5 persistent complex bereavement disorder (PCBD) with regard to content and the number of symptoms required to meet diagnosis. The findings of the current study are therefore not generalizable to other definitions of the disorder. Furthermore, the CGA-SR has not been validated for use in Cambodia, although the construct of PGD was validated by Field, Strasser (15) using a similar questionnaire, which speaks for the general applicability of PGD in Cambodia. Moreover, the RRI is a relatively new questionnaire. Although its factor structure has been confirmed in several samples, some caution is warranted when interpreting the results. In particular, the third subscale (ability to differentiate) requires further caution, as it consists only of two items. Finally, the cross-sectional design of the present study does not allow conclusions to be drawn about causal relationships between the assessed variables.

In conclusion, so far, research on post-conflict reconciliation and mental health outcomes has primarily focused on PTSD. However, PG is also very likely to be highly prevalent in post-conflict regions, given the high number of deaths and their mainly unnatural, often violent and man-made nature. The results of the current study

suggest that PG should be considered as a mental health-related correlate of reconciliation. As such, the findings can contribute to a better understanding of reconciliation processes in Cambodia and possibly also in other post-conflict regions. The association between attitudes toward reconciliation and PG should therefore be investigated in other post-conflict populations applying contemporary criteria for PGD or PCBD.

DATA AVAILABILITY STATEMENT

The datasets collected for this study cannot be made publicly available, as we did not obtain consent from our participants for sharing of data when the study was conceived.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Universität Konstanz. The participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

The study was part of a larger research project on reconciliation and mental health in Cambodia. NS and CK designed the study and wrote the research proposal. NS coordinated the study and supervised the data collection in the field. CK supervised the study. NS, LH, and CH analyzed the data. LH, NS, CH, and MB contributed to the data interpretation and structure of the manuscript. NS and LH drafted the manuscript. All authors contributed to the article and approved the submitted version.

REFERENCES

- Kiernan B. *The Pol Pot Regime: Race, Power and Genocide in Cambodia under the Khmer Rouge 1975–1979*. New Haven and London: Yale University Press (2009).
- Sonis J, Gibson JL, de Jong JT, Field NP, Hean S, Komproue I. Probable posttraumatic stress disorder and disability in Cambodia: associations with perceived justice, desire for revenge, and attitudes toward the Khmer Rouge trials. *JAMA* (2009) 302(5):527–36. doi: 10.1001/jama.2009.1085
- Sepanski DM, Lahar CJ, Khann S, Kao S, Schunert T. Four decades following the Khmer rouge: sociodemographic factors impacting depression, anxiety and PTSD in Cambodia. *J Ment Health* (2019) 28(2):175–80. doi: 10.1080/09638237.2018.1466039
- de Jong JM, Komproue IH, Van Ommeren M, El Masri M, Araya M, Khaled N, et al. Lifetime events and posttraumatic stress disorder in 4 postconflict settings. *JAMA* (2001) 286(5):555–62. doi: 10.1001/jama.286.5.555
- Schaal S, Jacob N, Dusingizemungu J-P, Elbert T. Rates and risks for prolonged grief disorder in a sample of orphaned and widowed genocide survivors. *BMC Psychiatry* (2010) 10:55:1–9. doi: 10.1186/1471-244X-10-55
- World Health Organization. (2018). *ICD-11 for mortality and morbidity statistics (ICD-11 MMS) 2018 version*. Available from: <https://icd.who.int/browse11/l%20E%2080%90m/en>.
- American Psychiatric Association. *Diagnostic and statistical manual of mental disorders : DSM-5. 5 ed.* Washington, DC [u.a.]: American Psychiatric Publ. (2013). XLIV, 947 S. p.
- Boelen PA, Lenferink LI. Comparison of six proposed diagnostic criteria sets for disturbed grief. *Psychiatry Res* (2020) 285:112786. doi: 10.1016/j.psychres.2020.112786

FUNDING

The study was funded by the German Ministry of Foreign Affairs (VN06.-320.21/2008/24) and Psychology Beyond Borders for a research project on reconciliation and mental health in Cambodia. The funding agencies were not involved in the design, implementation, analysis, or reporting of the results.

We acknowledge support by the Open Access Publication Initiative of Freie Universität Berlin.

ACKNOWLEDGMENTS

We would like to thank Transcultural Psychosocial Organization (TPO Cambodia) for their collaboration. We are especially grateful to Dr. Chhim Sothea and Sopheap Taing (TPO Cambodia) and Judith Strasser of the German Development Service (DED) for their support and advice in the implementation and carrying out of the study in Cambodia. We would further like to thank Estelle Bockers for her help in coordinating and supervising the study in Cambodia. We are very grateful to our data collectors Solida Sun, Sokkhim Srun, Dina Suos, Longkeat Khe, Vathana Hoy, and Veasna Try. This research would not have been possible without their openness and trust of all the persons that voluntarily took part in our study.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: <https://www.frontiersin.org/articles/10.3389/fpsy.2020.00644/full#supplementary-material>

- Lenferink LIM, Boelen PA, Smid GE, Paap MCS. The importance of harmonising diagnostic criteria sets for pathological grief. *Br J Psychiatry* (2019) 13:1–4. doi: 10.1192/bjp.2019.240
- Prigerson HG, Maciejewski PK. A Call for Sound Empirical Testing and Evaluation of Criteria for Complicated Grief Proposed for DSM-V. *OMEGA — J Death Dying* (2006) 52(1):9–19. doi: 10.2190/ANKH-BB2H-D52N-X99Y
- Lundorff M, Holmgren H, Zachariae R, Farver-Vestergaard I, O'Connor M. Prevalence of prolonged grief disorder in adult bereavement: A systematic review and meta-analysis. *J Affect Disord* (2017) 212:138–49. doi: 10.1016/j.jad.2017.01.030
- Morina N, Lersner UV, Prigerson HG. War and bereavement: consequences for mental and physical distress. *PloS One* (2011) 6(7):1–10. doi: 10.1371/journal.pone.0022140
- Boelen PA. Peritraumatic distress and dissociation in prolonged grief and posttraumatic stress following violent and unexpected deaths. *J Trauma Dissociation* (2015) 16(5):541–50. doi: 10.1080/15299732.2015.1027841
- Kristensen P, Weisæth L, Heir T. Bereavement and mental health after sudden and violent losses: a review. *Psychiatry* (2012) 75(1):76–97. doi: 10.1521/psyc.2012.75.1.76
- Field NP, Strasser J, Taing S, Horiuchi S, Chhim S, Packman W. Prolonged grief following the recent death of a daughter among mothers who experienced distal losses during the Khmer Rouge era: Validity of the prolonged grief construct in Cambodia. *Psychiatry Res* (2014) 219(1):183–90. doi: 10.1016/j.psychres.2014.05.014
- Boehnlein JK. Clinical relevance of grief and mourning among Cambodian refugees. *Soc Sci Med* (1987) 25(7):765–72. doi: 10.1016/0277-9536(87)90034-7
- Hinton DE, Nickerson A, Bryant RA. Prolonged grief in Cambodian refugees following genocide: Rebirth concerns and avoidance of reminders. *J Loss Trauma* (2013) 18(5):444–60. doi: 10.1080/15325024.2012.714218

18. Hinton DE, Field NP, Nickerson A, Bryant RA, Simon N. Dreams of the dead among cambodian refugees: frequency, phenomenology, and relationship to complicated grief and posttraumatic stress disorder. *Death Stud* (2013) 37 (8):750–67. doi: 10.1080/07481187.2012.692457
19. Hinton DE, Hinton AL, Pich V, Loeum JR, Pollack MH. Nightmares among cambodian refugees: the breaching of concentric ontological security. *Cult Med Psychiatry* (2009) 33(2):219–65. doi: 10.1007/s11013-009-9131-9
20. Hinton DE, Reis R, de Jong J. Migraine-like visual auras among traumatized cambodians with ptsd: fear of ghost attack and other disasters. *Cult Med Psychiatry* (2018) 42(2):244–77. doi: 10.1007/s11013-017-9554-7
21. Van de Put WACM, Eisenbruch M. The Cambodian experience. In: de Jong JM, editor. *Trauma, war, and violence: Public mental health in socio-cultural context*. New York: Kluwer Academic / Plenum Publishers (2002). p. 93–156.
22. McGrew L. *Reconciliation in Cambodia: victims and perpetrators living together, apart [Dissertation]*. L. Conventry University: ProQuest Dissertations Publishing (2011).
23. Etcheson C. *After the killing fields: Lessons from the Cambodian genocide*. Westport, CT: Praeger Publishers (2005).
24. Etcheson C. The limits of reconciliation in Cambodia's communes. In: Skaar E, Gloppen S, Suhrke A, editors. *Roads to reconciliation*. Lanham: Lexington Books (2005). p. 201–24.
25. Murrell A. Reconciliation and healing of a nation. *Med Confl Surviv* (2001) 17 (2):146–50. doi: 10.1080/13623690108409569
26. Pham PN, Vinck P, Balthazard M, Hean S, Stover E. *So we will never forget: a population-based survey on attitudes about social reconstruction and the Extraordinary Chambers in the Courts of Cambodia*. Berkely: Human Rights Center, University of California (2009).
27. Public Affairs Section of the Extraordinary Chambers in the Courts of Cambodia. (2017). *An Introduction to the Khmer Rouge Trials*. [cited 2020 23.02.2020]. Available from: <https://www.eccc.gov.kh/sites/default/files/publications/ECCCBooklet7ed%28Eng%29.pdf>.
28. Hamber B. *Transforming Societies after Political Violence*. New York: Springer (2009).
29. Bar-Tal D, Bennink GH. The nature of reconciliation as an outcome and as a process. In: Bar-Siman-Tov Y, editor. *From conflict resolution to reconciliation*. Oxford: Oxford University Press (2004). p. 11–38.
30. Bar-On D. Empirical criteria for reconciliation in practice. *Intervention* (2005) 3(3):180–91.
31. Bar-Tal D. From Intractable Conflict Through Conflict Resolution to Reconciliation: Psychological Analysis. *Political Psychology* (2000) 21 (2):351–65.
32. Thoms O, Ron J, Paris R. State-Level Effects of Transitional Justice: What Do We Know? *Int J Of Transit Justice* (2010) 4(3):329–54. doi: 10.1093/ijtj/ijq012
33. Bayer CP, Klasen F, Adam H. Association of trauma and PTSD symptoms with openness to reconciliation and feelings of revenge among former Ugandan and Congolese child soldiers. *JAMA* (2007) 298(5):555–9. doi: 10.1001/jama.298.5.555
34. Pham PN, Weinstein HM, Longman T. Trauma and PTSD symptoms in Rwanda: Implications for attitudes toward justice and reconciliation. *JAMA* (2004) 292(5):602–12. doi: 10.1001/jama.292.5.602
35. Doran JM, Kalayjian A, Toussaint L, DeMucci J. The relationship between trauma and forgiveness in post-conflict Sierra Leone. *psychol Trauma: Theory Rese Pract Policy* (2012) 4(6):614–23. doi: 10.1037/a0025470
36. Kaminer D, Stein DJ, Mbanga I, Zungu-Dirwayi N. The Truth and Reconciliation Commission in South Africa: relation to psychiatric status and forgiveness among survivors of human rights abuses. *Br J Psychiatry* (2001) 178(4):373–7. doi: 10.1192/bjp.178.4.373
37. Stammel N, Neuner F, Böttche M, Knaevelsrud C. Construction of a questionnaire for readiness to reconcile in victims of human rights violations. *Eur J Psychotraumatol* (2012) 3(1):1–9. doi: 10.3402/ejpt.v3i0.15785
38. Field NP, Chhim S. Desire for Revenge and Attitudes Toward the Khmer Rouge Tribunal Among Cambodians. *J Loss Trauma* (2008) 13(4):352–72. doi: 10.1080/15325020701742086
39. Longman T, Pham P, Weinstein HM. Connecting justice to human experience: Attitudes toward accountability and reconciliation in Rwanda. In: Stover E, Weinstein HM, editors. *My neighbor, my enemy: Justice and community in the aftermath of mass atrocity*. Cambridge: Cambridge University Press (2004). p. 206–25.
40. Heim L, Schaal S. Rates and predictors of mental stress in Rwanda: investigating the impact of gender, persecution, readiness to reconcile and religiosity via a structural equation model. *Int J Ment Health Syst* (2014) 8 (37):1–9. doi: 10.1186/1752-4458-8-37
41. Allan A, Allan MM, Kaminer D, Stein DJ. Exploration of the association between apology and forgiveness amongst victims of human rights violations. *Behav Sci Law* (2006) 24(1):87–102. doi: 10.1002/bsl.689
42. Stein D, Seedat S, Kaminer D, Moomal H, Herman A, Sonnegg J, et al. The impact of the Truth and Reconciliation Commission on psychological distress and forgiveness in South Africa. *Soc Psychiatry Psychiatr Epidemiol* (2008) 43:462–8. doi: 10.1007/s00127-008-0350-0
43. Stammel N, Heeke C, Bockers E, Chhim S, Taing S, Wagner B, et al. Prolonged grief disorder three decades post loss in survivors of the Khmer Rouge regime in Cambodia. *J Affect Disord* (2013) 144(1):87–93. doi: 10.1016/j.jad.2012.05.063
44. Stammel N, Bockers E, Neuner F, Chhim S, Taing S, Knaevelsrud C. The Readiness to Reconcile Inventory: Assessing attitudes toward reconciliation in victims of war and conflict. *Eur J psychol Assess* (2015) 33(6):436–44. doi: 10.1027/1015-5759/a000304
45. Eichhorn S, Stammel N, Glaesmer H, Klauer T, Freyberger HJ, Knaevelsrud C, et al. Readiness to reconcile and post-traumatic distress in German survivors of wartime rapes in 1945. *Int Psychogeriatr* (2015) 27(5):857–64. doi: 10.1017/S1041610214002695
46. Weathers FW, Huska JA, Keane TM. *The PTSD Checklist-Civilian Version (PCL-C)*. Boston, MA: National Center for PTSD-Behavioral Science Division (1991).
47. Mollica RF, Caspi-Yavin Y, Bollini P, Truong T, Tor S, Lavelle J. The Harvard Trauma Questionnaire. Validating a cross-cultural instrument for measuring torture, trauma, and posttraumatic stress disorder in Indochinese refugees. *J Nerv Ment Dis* (1992) 180(2):111–6. doi: 10.1097/00005053-199202000-00008
48. Foa EB, Cashman L, Jaycox L, Perry K. The validation of a self-measure of posttraumatic stress disorder: The Posttraumatic Diagnostic Scale. *psychol Assess* (1997) 9(4):445–51. doi: 10.1037/1040-3590.9.4.445
49. Guillemin F, Bombardier C, Beaton D. Cross-cultural adaptation of health-related quality of life measures: literature review and proposed guidelines. *J Clin Epidemiol* (1993) 46(12):1417–32. doi: 10.1016/0895-4356(93)90142-N
50. Tabachnick BG, Fidell LS. *Using multivariate statistics*. 5. ed. Boston: Pearson (2007).
51. Heeke C, Kampisiou C, Niemeyer H, Knaevelsrud C. A systematic review and meta-analysis of correlates of prolonged grief disorder in adults exposed to violent loss. *Eur J Psychotraumatol* (2019) 10(1):1583524. doi: 10.1080/2008198.2019.1583524
52. Biro M, Milin P. Traumatic experience and the process of reconciliation. *Psihologija* (2005) 38(2):133–48. doi: 10.2298/PSI0502133B
53. Maercker A, Brewin CR, Bryant RA, Cloitre M, van Ommeren M, Jones LM, et al. Diagnosis and classification of disorders specifically associated with stress: proposals for ICD-11. *World Psychiatry* (2013) 12(3):198–206. doi: 10.1002/wps.20057
54. Currier JM, Holland JM, Coleman RA, Neimeyer RA. Bereavement following violent death: An assault on life and meaning. In: Stevenson RG, Cox GR, editors. *Perspectives on violence and violent death. Death, value and meaning series*. Amityville, NY, US: Baywood Publishing Co (2008). p. 177–202.
55. Currier JM, Holland JM, Neimeyer RA. Sense-making, grief, and the experience of violent loss: toward a mediational model. *Death Stud* (2006) 30(5):403–28. doi: 10.1080/07481180600614351
56. Keesee NJ, Currier JM, Neimeyer RA. Predictors of grief following the death of one's child: the contribution of finding meaning. *J Clin Psychol* (2008) 64 (10):1145–63. doi: 10.1002/jclp.20502
57. Ciorciari JD. Cambodia's trek toward reconciliation. *Peace Rev* (2011) 23 (4):438–46. doi: 10.1080/10402659.2011.625786
58. Manning P. Reconciliation and perpetrator memories in Cambodia. *Int J Transit Justice* (2015) 9(3):386–406. doi: 10.1093/ijtj/ijv015
59. Tek FL. Justice at the Extraordinary Chambers in the Court of Cambodia? *Peace Rev* (2011) 23(4):431–7. doi: 10.1080/10402659.2011.625738
60. Boelen PA, de Keijser J, Smid G. Cognitive-behavioral variables mediate the impact of violent loss on post-loss psychopathology. *psychol Trauma: Theory Rese Pract Policy* (2015) 7(4):382–90. doi: 10.1037/tra0000018

61. Boelen PA, van den Bout J, van den Hout MA. The role of cognitive variables in psychological functioning after the death of a first degree relative. *Behav Res Ther* (2003) 41(10):1123–36. doi: 10.1016/S0005-7967(02)00259-0
62. Boelen PA, van Denderen M, de Keijser J. Prolonged Grief, Posttraumatic Stress, Anger, and Revenge Phenomena Following Homicidal Loss: The Role of Negative Cognitions and Avoidance Behaviors. *Homicide Stud* (2016) 20(2):177–95. doi: 10.1177/1088767915580674
63. Milman E, Neimeyer RA, Fitzpatrick M, MacKinnon CJ, Muis KR, Cohen SR. Rumination moderates the role of meaning in the development of prolonged grief symptomatology. *J Clin Psychol* (2019) 75(6):1047–65. doi: 10.1002/jclp.22751
64. Morina N. Rumination and avoidance as predictors of prolonged grief, depression, and posttraumatic stress in female widowed survivors of war. *J Nerv Ment Dis* (2011) 199(12):921–7. doi: 10.1097/NMD.0b013e3182392aae
65. Eisma MC, Schut HA, Stroebe MS, Boelen PA, van den Bout J, Stroebe W. Adaptive and maladaptive rumination after loss: A three-wave longitudinal study. *Br J Clin Psychol* (2015) 54(2):163–80. doi: 10.1111/bjc.12067
66. Tay AK, Rees S, Chen J, Kareth M, Silove D. Factorial structure of complicated grief: associations with loss-related traumatic events and psychosocial impacts of mass conflict amongst West Papuan refugees. *Soc Psychiatry Psychiatr Epidemiol* (2016) 51(3):395–406. doi: 10.1007/s00127-015-1099-x
67. Neimeyer RA, Burke LA. Complicated grief in the aftermath of homicide: spiritual crisis and distress in an African american sample. *Religions* (2011) 2(2):145–64. doi: 10.3390/rel2020145
68. Esala JJ, Taing S. Testimony therapy with ritual: a pilot randomized controlled trial*. *J Trauma Stress* (2017) 30(1):94–8. doi: 10.1002/jts.22163

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Copyright © 2020 Stammel, Heinzl, Heeke, Böttche and Knaevelsrud. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.



Coping Strategies and Considering the Possibility of Death in Those Bereaved by Sudden and Violent Deaths: Grief Severity, Depression, and Posttraumatic Growth

Joscelyn E. Fisher^{1,2*}, Jing Zhou^{1,2}, Rafael F. Zuleta^{1,2}, Carol S. Fullerton², Robert J. Ursano² and Stephen J. Cozza²

¹ Henry M. Jackson Foundation for the Advancement of Military Medicine, Bethesda, MD, United States, ² Center for the Study of Traumatic Stress, Department of Psychiatry, Uniformed Services University of the Health Sciences, Bethesda, MD, United States

OPEN ACCESS

Edited by:

Geert E. Smid,
ARQ National Psychotrauma Centre,
Netherlands

Reviewed by:

Casimiro Cabrera Abreu,
Queens University, Canada
Tom Kingstone,
Keele University, United Kingdom

*Correspondence:

Joscelyn E. Fisher
joscelyn.fisher.ctr@usuhs.edu

Specialty section:

This article was submitted to
Mood and Anxiety Disorders,
a section of the journal
Frontiers in Psychiatry

Received: 27 April 2020

Accepted: 16 July 2020

Published: 06 August 2020

Citation:

Fisher JE, Zhou J, Zuleta RF, Fullerton CS, Ursano RJ and Cozza SJ (2020) Coping Strategies and Considering the Possibility of Death in Those Bereaved by Sudden and Violent Deaths: Grief Severity, Depression, and Posttraumatic Growth. *Front. Psychiatry* 11:749. doi: 10.3389/fpsy.2020.00749

Background: Bereavement by sudden and violent deaths can lead to increased grief severity, depression, and reduced posttraumatic growth compared to those bereaved by natural causes. These outcomes can be affected by coping strategies and whether a survivor had been “prepared” for the death. The present study examined the effect of coping and considering the possibility of death on grief severity, depression, and posttraumatic growth in those bereaved by sudden deaths.

Methods: Participants bereaved by suicide, accident, or combat deaths completed an online survey about demographics (including the cause of death), coping, grief severity, depression, and posttraumatic growth. A factor analysis of the coping measure yielded factors representing three coping strategies: avoidant coping, supportive coping, and active coping. These three strategies, the causes of death and considering the possibility of death were used as predictors of either grief severity, depression, or posttraumatic growth in multivariate linear regression models.

Results: Each coping strategy and cause of death was differentially associated with grief severity, depression, and posttraumatic growth. Specifically, supportive coping and active coping were each only associated with higher posttraumatic growth. In contrast, avoidant coping was associated with all outcomes (higher grief severity and depression and lower posttraumatic growth). In addition, accidents and suicides (compared to combat deaths) had independent effects on grief severity and posttraumatic growth. Considering the possibility of death interacted with avoidant coping and also with supportive coping to predict grief severity in combat-loss survivors.

Discussion: Findings highlight the differential contributions of coping strategies and their complex relationships with cause of death in contributing to grief severity, depression, and posttraumatic growth. Avoidant coping contributed to negative outcomes and inhibited posttraumatic growth, suggesting its importance as a target for therapeutic intervention.

Although supportive and active coping facilitated posttraumatic growth, they had less of a role in mitigating grief severity or depression in this study. Although considering the possibility of death appeared to mitigate negative outcomes among survivors of combat death, avoidance of that possibility is likely protective for the majority of family members whose loved ones return home safely.

Keywords: bereavement, coping, depression, grief, posttraumatic growth, preparedness, possibility of death

INTRODUCTION

Bereavement is a common and universal stressor that sometimes leads to complicated grief or depression (e.g., 1). These outcomes occur more frequently for those who have been bereaved by sudden and violent deaths, such as accidents, suicides, or military combat deaths (2). Although some studies identified differences in psychological health between survivors of suicide loss and other causes of death (3–5), other studies reported minimal or no differences (6–9).

These potential differences in mental health outcomes could be attributed to coping strategies used following a death. Coping has been defined and classified in various ways (10–12). Many studies have operationalized coping after bereavement by using either the COPE (10) or Brief COPE (13), a self-report measure that assesses 14 coping strategies (14–24). These coping strategies (as measured by COPE or Brief COPE subscales) were grouped by Schnider et al. (24) into three categories: *problem-focused* coping (active coping, planning, instrumental support, and religion), *active emotional coping* (venting, positive reframing, humor, acceptance, and emotional support), and *avoidant emotional coping* (self-distraction, denial, behavioral disengagement, self-blame, and substance use). This three-group structure is consistent with several factor analyses of the COPE and Brief COPE in non-bereaved samples (25–29). However, the combinations of individual subscales that defined the three factors has differed between studies, leading to variations in coping strategy labels [e.g., Engagement, Disengagement, and Help-Seeking in (25); Active Coping, Repressive Coping, and Affective Coping in (22)]. Alternate coping inventories employed in other studies of bereaved samples have similarly yielded three-dimensional coping models [e.g., task-oriented, emotion-oriented, and avoidance-oriented coping; (30)].

Despite the variety of instruments and labels employed, associations between certain types of coping strategies and distinct outcomes have been consistent in bereaved samples. Problem-focused coping (16) has been associated with posttraumatic growth in suicide loss survivors, and affective coping has been associated with lower post-traumatic stress disorder (PTSD) symptom scores (21). Avoidant (or repressive) coping has been associated with increased severity of negative outcomes, such as grief (17–19, 24), PTSD (22), mental distress (20), and depression (18), but has also been associated with lower grief severity (30), suggesting that the relationship between avoidant coping and grief

severity is complex and likely affected by additional factors related to the death.

As many studies of bereavement coping strategies either combined causes of death or looked at one cause of death alone (e.g., suicide), there is limited information about the effects of different coping strategies according to specific types of sudden and violent deaths. Use of avoidant coping (31) and decreased use of support from others (32, 33) have been reported in survivors of suicide loss, but to our knowledge, no studies have reported about coping strategies specifically in accidental deaths. Although there have been no published research studies about coping strategies used following combat deaths, anecdotal reports suggest that combat-loss survivors are more likely to receive community support, feel a sense of pride in their family member's sacrifice, and find meaning after the death compared to other sudden and violent deaths. These reports might translate to greater use of supportive and active (e.g., positive reframing) coping strategies compared to suicide-loss or accident-loss survivors.

In addition to the effect of coping strategies, the unexpected nature of sudden and violent deaths is also likely to contribute to increased risk of mental health difficulties and reduced well-being. For example, unexpected deaths and perceptions of less preparedness for the death have both been associated with complicated grief (34), increased depression (35, 36), and other mental health disorders (36). Other variables related to the nature of a death (i.e., violent, extent of suffering, prolonged dying process, opportunity to say goodbye) have been associated with "bereavement intensity", but being unprepared for a death was the strongest predictor of this association (14).

Despite the usefulness of the construct and the importance of its associations, the term "preparedness" is ambiguous. For example, even when deaths are expected, survivors may not feel "prepared" for a death. Additionally, preparedness may refer to logistical, as well as psychological readiness for the death. A more precise indication of psychological readiness may be whether the individual considered the possibility of death, as it more clearly refers to a psychological process. Different causes of sudden deaths (combat deaths vs. accidents vs. suicide) would be expected to vary in the degree to which the possibility of death was considered. For instance, death is a known risk associated with military combat operations, so surviving family members whose service member died (either due to combat, accident or suicide) while deployed on a combat mission would likely have considered such a possibility. Survivors of some suicide deaths may have considered the possibility of death, especially if attempts had been previously made or if the death followed a period of suffering from a mental illness. In contrast, survivors of

Abbreviation: PTG, Posttraumatic Growth.

accidents are the least likely to have considered such a possibility since accidents, by definition, are likely to be unpredictable. Whether a bereaved individual considered the possibility of death prior to its occurrence combined with the coping style used after the death could explain differences in mental health outcomes between those bereaved by deaths due to combat, suicide, and accidents.

The current study examined the effects of coping strategies and considering the possibility of death on grief severity, depression, and posttraumatic growth in survivors bereaved by suicide, accident, and combat. It was hypothesized that suicide-loss survivors would be more likely to use avoidant coping and less likely to use support-type strategies, resulting in higher grief severity and depression, and reduced positive outcomes compared to accident-loss and combat-loss survivors. In comparison, combat-loss survivors would use more supportive coping and/or active coping strategies, which would be associated with more posttraumatic growth and lower grief severity. Survivors of combat death were also expected to have been more likely to have considered the possibility of death compared to survivors of suicides and accidents, given that all combat deaths occurred during combat deployment compared to smaller percentages of accidents and suicides that occurred during combat deployment. This combination of coping and considering the possibility of death would contribute to less adverse and more positive outcomes.

METHODS

Participants

Surviving parents, spouses/partners, siblings, and adult children of U.S. military service members who died by combat, accident, or suicide between September 11, 2001 and January, 2014 were enrolled as part of the National Military Family Bereavement Study (NMFBS¹), a study of the impact of military service member death on their family members. Participants were recruited for the NMFBS through grief support organizations, online advertisements, and word-of-mouth and provided informed consent after receiving a description of the study. Analyses for this manuscript included: 328 survivors bereaved by suicide, 384 survivors bereaved by accidents, and 997 survivors bereaved by combat deaths (total $n = 1,709$). Time since death ranged from under 6 months to over 12 years (mean = 4.89 years; mode = 3 years), though most participants (65%) had been bereaved between 1 and 7 years at the time of assessment. One hundred thirty-one family members were bereaved less than 1 year (34 of these 131 were bereaved less than 6 months).

Design

Participants provided online consent and completed an online survey located on the NMFBS website¹. The survey assessed demographics, information about relationship to the service member, circumstances of the death, and physical and psychological reactions and was designed for participants to

complete in 30–45 min. Eighty-three percent of participants who consented to participate then completed the study. The study was conducted in accordance with ethical standards as approved by the Human Research Protection Program in the Office of Research at the Uniformed Services University of the Health Sciences and all analyses were conducted on de-identified data.

Measures

The following measures were analyzed for the present study.

Brief COPE (13). (28 items): 14 subscales: 1) Active Coping, 2) Planning, 3) Positive Reframing, 4) Acceptance, 5) Humor, 6) Religion, 7) Using Emotional Support, 8) Using Instrumental Support, 9) Self-Distraction, 10) Denial, 11) Venting, 12) Substance Use, 13) Behavioral Disengagement, and 14) Self-Blame. The overall scale has good performance characteristics. Instructions asked how the participant has been coping with “stress in your life since the death of your service member”.

Inventory of Complicated Grief (ICG; 37) is a 19-item self-report measure of clinically impairing grief symptom severity during the last month. The ICG has been widely used as a screening tool to assess grief severity (38–40).

Patient Health Questionnaire (PHQ-9; 41) is a 9-item measure that has been used as a reliable measure of depression in medical and general population settings (41). Participants were asked how often they had been bothered by symptoms during the previous 2 weeks.

Posttraumatic Growth Inventory—Short Form (PTGI-SF; 42) is a measure of positive changes or growth after traumatic events assessed on a six-point Likert scale. A 10-item short form of the PTGI was used in this study. Participants responded to each item according to whether it was true as a result of the death of their family member.

Possibility of death: One item assessed the frequency with which participants considered the possibility of their loved one's death. Participants chose one of the following five options to complete the stem: “During the month prior to my service member's death...”: (1) I never thought about the possibility of my service member dying; (2) I thought about the possibility of my service member dying 1–3 times during the month (or about once or twice every 2 weeks); (3) once each week; (4) 3–5 times each week; (5) every day. Responses to this item were dichotomized as “never thought about the possibility” (i.e., 1) and “thought about the possibility” (i.e., 2–5) for the present study analyses.

Data Analysis

Descriptive statistics (means, standard deviations, frequency distributions, crosstabs, etc.) of the study participants' demographics and other characteristics were examined. In order to determine how coping strategies clustered among bereaved family members within the sample, an exploratory factor analysis (EFA) was conducted. The EFA of the 14 Brief COPE subscales supported a three-factor structure (eigenvalue greater than 1). These factors were labeled *supportive*, *avoidant*, and *active* coping. A confirmatory factor analysis (CFA) was then conducted based on the EFA and prior literature to determine whether the coping subscales factored into three factors

¹<https://militarysurvivorstudy.org>

representing supportive, avoidant, and active coping strategies. The supportive coping factor consisted of two coping subscales: emotional support (factor loading: 0.80) and instrumental support (factor loading: 0.85). The avoidant coping factor consisted of three coping subscales: denial (factor loading: 0.60), behavioral disengagement (factor loading: 0.72), and self-blame (factor loading: 0.61), and the active coping factor consisted of three coping subscales: active coping (factor loading: 0.80), positive reframing (factor loading: 0.54), and planning (factor loading: 0.73). Indices of model fit indicated that the CFA model had a good fit [comparative fit index (CFI) = 0.92, standardized root mean square residual (SRMR) = 0.06, root mean square error of approximation (RMSEA) = 0.10]. Coping subscales in each resulting factor were identified and then summed to create a continuous score that represented each coping strategy. Each factor score was standardized by dividing the number of subscales in the factor. As these three factors are conceptually similar to other studies that derived a three-factor structure from the COPE and Brief COPE (25–29), these standardized factor scores were included in subsequent regression analyses as indices of coping strategies.

To examine the effects and interactions of coping strategies, cause of death (suicide, accidents, and combat), and possibility of death (never thought about possibility of death vs. thought about the possibility) on grief severity, depression, and posttraumatic growth, multivariate linear regression analyses were performed. Linear splines were applied to examine the relationship between time since death and outcomes when appropriate. In order to adjust for the cluster effects of participants nested within families, generalized estimating equations were used to fit linear regressions in all analyses (43). Model assumptions were checked using histograms, normal probability plots, scatter plots, smooth spline, plots, and other model diagnostics. Potential confounding variables (i.e., time since death, relationship type, participant age, gender) were also included in the models.

All statistical analyses were performed using SAS 9.4 (SAS Institute Inc., Cary, North Carolina) and statistical significance was defined as $p < 0.05$ using two-tailed tests.

RESULTS

Participant Characteristics

Demographic characteristics of the study participants, causes of service member death, relationship to the deceased, coping strategy factor summary scores, thoughts about the possibility of death, and means and standard deviations of the ICG, PHQ-9, and PTGI-SF scores are presented in Table 1.

Possibility of Death and Cause of Death

More than half (56%) of the participants thought about the possibility of their service member's death. The percentages differed between each of the causes of death ($p < 0.05$). The percentages were much higher among combat-loss survivors (70%) compared to accident-loss survivors (41%) and suicide-loss survivors (29%).

TABLE 1 | Demographics and other characteristics of study participants.

Characteristics	Total (N = 1709)	
	N or M	% or SD
Age in years (M and SD)	46.98	13.26
Gender		
Male	358	20.97%
Female	1349	79.03%
Cause of death of DSM*		
Combat related	997	58.34%
Accident	384	22.47%
Suicide	328	19.19%
Time since death in years (M and SD)	4.89	2.87
Relationship to the deceased		
Parent	965	56.57%
Spouse	351	20.57%
Sibling	345	20.22%
Adult child	45	2.64%
Possibility of death		
Never thought about possibility of death	740	44.15%
Thought about possibility of death	936	55.85%
Coping factor summary scores (M and SD, range = 2–8)		
Supportive	4.84	1.69
Avoidant	3.44	1.32
Active	4.97	1.54
ICG total score (M and SD)	25.42	14.98
PHQ total score (M and SD)	8.31	6.79
PTGI total score (M and SD)	25.15	12.80

*Deceased service member.

DSM, Deceased service member; ICG, Inventory of Complicated Grief; PHQ, Patient Health Questionnaire; PTGI, Posttraumatic Growth Inventory.

To determine whether being deployed at the time of death was associated with survivors considering the possibility of death, the above was stratified according to deployment status at the time of death. Twenty-three percent of accidental deaths and 8% of suicide deaths occurred during combat deployment. Of survivors of accidents and suicide whose loved one was combat-deployed at the time of death, 64% and 42%, respectively considered the possibility of death, compared to 34% and 28% of respective survivors of accidents and suicides whose loved one was not combat-deployed at the time of death. In contrast, 70% of combat survivors thought about the possibility of death.

Possibility of Death and Outcomes

Participants who never thought about the possibility of their service member's death had significantly higher grief severity total scores (mean = 26.1, standard deviation = 15.8) than those who had thought about the service member's death (mean = 24.6, standard deviation = 14.2, $p = 0.04$). There were no significant differences in either depression or posttraumatic growth between participants who never thought about their service member's death and those who did.

Coping Strategies and Cause of Death

Means of each coping strategy within each cause of death were compared. Suicide-loss survivors had significantly higher mean avoidant coping scores (mean = 3.7, standard deviation = 1.3) compared to combat-loss survivors (mean = 3.3, standard deviation = 1.3, $p < 0.05$), but not accident-loss survivors (mean = 3.5, standard deviation = 1.3). Supportive coping and active coping strategies did not differ according to cause of death.

Coping Strategies and Cause of Death Predicting Outcomes

To examine the effects of coping strategies and causes of death on grief severity, depression, and posttraumatic growth ($N = 1709$), three separate multivariate linear regression models were conducted in which all coping strategies and all causes of death were entered as predictors (Table 2). Avoidant coping predicted grief severity. The second model indicated that avoidant coping was also associated with depression. The third model indicated that all coping strategies predicted posttraumatic growth; avoidant coping was negatively associated with posttraumatic growth, while active coping and supportive coping were each positively associated with posttraumatic growth. In addition, accident deaths and suicide deaths were associated with lower posttraumatic scores compared to combat deaths. There were no significant interactions between any of the coping strategies and causes of death in any of the three models.

Possibility of Death, Coping Strategies, and Interactions Predicting Outcomes

To examine the effect of considering the possibility of death on grief severity, depression, and posttraumatic growth, three additional regression models were conducted. These models added the possibility of death as an additional predictor to the models that contained cause of death and the three coping strategies. As described above, all coping strategies predicted posttraumatic growth and avoidant coping predicted higher scores on grief severity and depression measures. Considering the possibility of death was not associated with any of the three outcomes (grief: $p = 0.95$; depression: $p = 0.053$; posttraumatic growth $p = 0.71$).

Interactions between the possibility of death and each coping strategy were then tested. None of the interactions between possibility of death and any of the three coping strategies predicted posttraumatic growth or depression. However, the interaction between possibility and avoidant coping predicted grief severity (Table 3 and Figure 1). The interaction suggests that in general, more avoidant coping was associated with increases in grief severity. However, for those who never considered the possibility of death, increased use of avoidant coping was associated with higher increases in grief severity than those who did consider the possibility.

Possibility of Death, Coping, and Interactions Stratified by Cause of Death

In order to further examine the interactive effect of possibility of death and avoidant coping on grief severity according to different causes of death, the regression analyses described above were repeated with the addition of stratifying by cause of death. There were no significant interactions between the possibility of death and coping strategies predicting grief severity in accident

TABLE 3 | Interactions between possibility of death and coping strategies predicting grief ($N = 1,709$)*.

Predictors	ICG Total Score	
	Estimate (SE)	p-value
Possibility of death * supportive coping		
Never thought about possibility of death	0.45 (0.37)	0.23
Thought about possibility of death	Ref	Ref
Possibility of death * avoidant coping		
Never thought about possibility of death	1.08 (0.42)	0.01
Thought about possibility of death	Ref	Ref
Possibility of death * active coping		
Never thought about possibility of death	0.56 (0.42)	0.18
Thought about possibility of death	Ref	Ref
Cause of death		
Combat related	Ref	
Accident	0.99 (0.69)	0.15
Suicide	-1.74 (0.77)	0.02

*Model adjusted for participants age, gender, relationship with service member (parent, spouse, sibling or adult child), and time since death.

ICG, Inventory of Complicated Grief.

The text in bold indicates a p-value less than .05.

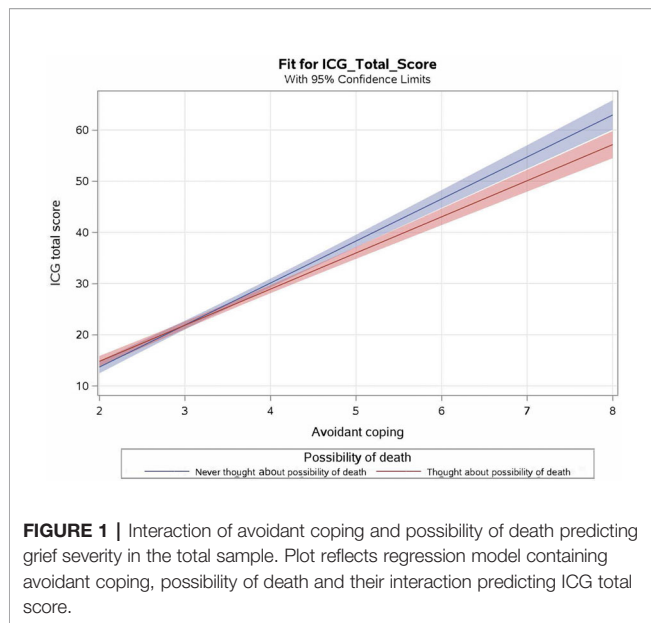
TABLE 2 | Coping strategies and cause of death predicting grief, depression and posttraumatic growth ($N = 1709$)*.

Predictors	ICG Total Score		PHQ Total Score		PTGI Total Score	
	Estimate (SE)	p-value	Estimate (SE)	p-value	Estimate (SE)	p-value
Coping strategies						
Supportive coping	0.03 (0.19)	0.87	-0.14 (0.09)	0.13	0.89 (0.20)	<0.01
Avoidant coping	7.29 (0.22)	<0.01	3.03 (0.11)	<0.01	-1.41 (0.23)	<0.01
Active coping	0.27 (0.22)	0.21	0.05	0.61	2.99 (0.21)	<0.01
Cause of death						
Combat related	Ref		Ref		Ref	
Accident	0.95 (0.68)	0.16	-0.25 (0.33)	0.43	-1.85 (0.72)	0.01
Suicide	-1.76 (0.73)	0.02	0.26 (0.41)	0.52	-1.94 (0.79)	<0.01

*Models adjusted for participants age, gender, relationship with service member (parent, spouse, sibling or adult child), and time since death.

ICG, Inventory of Complicated Grief; PHQ, Patient Health Questionnaire; PTGI, Posttraumatic Growth Inventory.

The text in bold indicates a p-value less than .05.



bereaved or suicide bereaved. However, the interaction between considering the possibility of death and supportive coping predicted grief severity in combat-bereaved individuals ($N = 997$). For those who never thought about the possibility of death, more supportive coping was associated with increased grief severity scores. However, for those who did consider the possibility of death, grief scores did not vary according to the amount of supportive coping (**Table 4** and **Figure 2A**).

The interaction between considering the possibility of death and avoidant coping also predicted grief severity for combat-bereaved participants. For those who never considered the possibility of death, more avoidant coping was associated with increases in grief severity than those who did consider the possibility (**Table 4** and **Figure 2B**).

DISCUSSION

The present study investigated the relationships between causes of death, coping strategies, whether participants considered the possibility of death, and post-bereavement outcomes, including grief severity and depression, as well as posttraumatic growth. In summary, the findings indicated that each type of coping strategy was differentially associated with these three outcomes. Specifically, avoidant coping had broad and negative effects on post-bereavement outcomes by worsening grief severity and depression, and decreasing posttraumatic growth. In contrast, neither supportive coping nor active coping were associated with either grief severity or depression, but were associated with higher levels of posttraumatic growth, suggesting unique pathways for supporting growth within the population of those who have been suddenly and violently bereaved. In addition to these effects of coping strategies on outcomes, accidents and suicides (compared to combat deaths) had independent effects

TABLE 4 | Interactions between possibility of death and coping strategies predicting grief in combat-bereaved ($N = 997$)*.

Predictors	ICG Total Score	
	Estimate (SE)	p-value
Possibility of death * supportive coping		
Never thought about possibility of death	1.47 (0.53)	0.01
Thought about possibility of death	Ref	Ref
Possibility of death * avoidant coping		
Never thought about possibility of death	2.24 (0.59)	<0.01
Thought about possibility of death	Ref	Ref
Possibility of death * active coping		
Never thought about possibility of death	0.25 (0.59)	0.67
Thought about possibility of death	Ref	Ref

*Model adjusted for participants age, gender, relationship with service member (parent, spouse, sibling or adult child), and time since death.

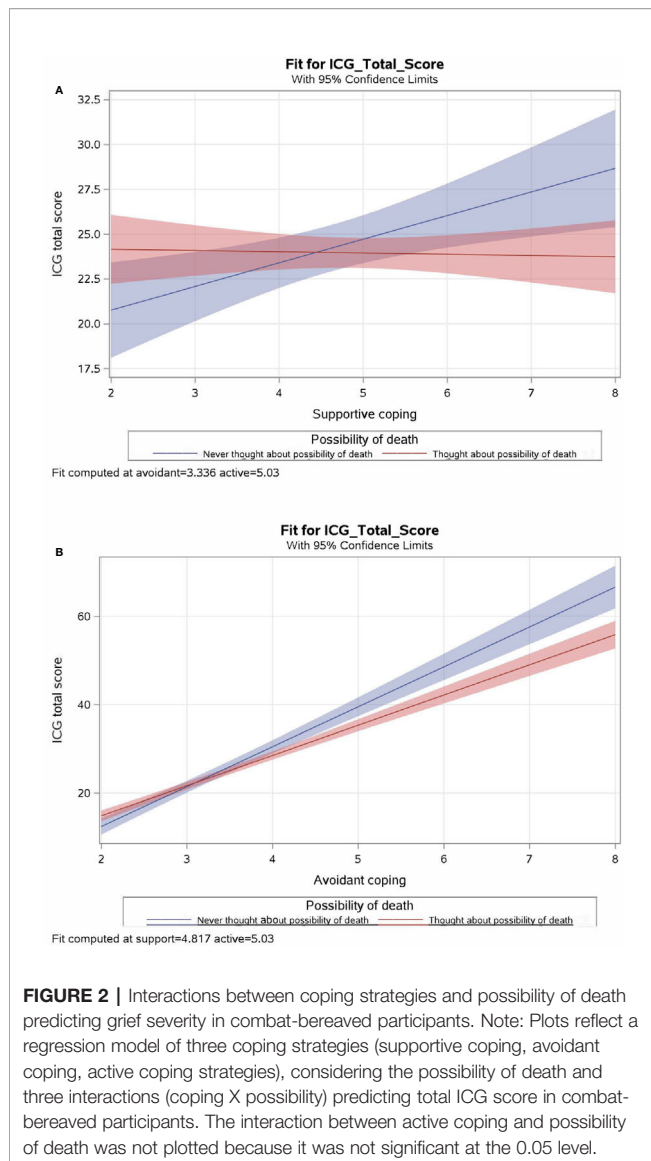
ICG, Inventory of Complicated Grief.

The text in bold indicates a p-value less than .05.

on grief severity and posttraumatic growth. These findings imply that there are circumstances unique to causes of death, which are independent of coping strategies, that contribute to these outcomes. It was anticipated that whether a survivor had considered the possibility of death might account for differences between combat, suicide, and accident-related deaths. Although a higher percentage of combat-loss survivors had considered the possibility of death compared to accident-loss survivors and suicide-loss survivors, this variable alone did not account for additional variance in outcomes when coping strategies were included. However, the interaction between considering the possibility of death and specific coping strategies did account for grief severity among combat-death survivors. Implications of these findings are discussed below.

Each type of coping strategy and cause of death was differentially associated with grief severity, depression, and posttraumatic growth. Specifically, supportive coping and active coping were each associated with higher posttraumatic growth, and avoidant coping was associated with lower posttraumatic growth. In contrast, supportive coping and active coping were not associated with either depression or grief severity, though avoidance was associated with grief severity and depression. These results are consistent with other studies that found multiple coping strategies associated with posttraumatic growth in bereaved individuals (44), especially active coping strategies (45). However, findings also raise interesting questions about therapeutic coping-focused approaches that intend to minimize grief severity and depression, and suggest that emphasis on avoidant coping, rather than focusing on supportive or active coping, would better address those outcomes.

The substantial negative effect of avoidant coping on all three post-bereavement outcomes (higher grief and depression and lower posttraumatic growth) in this study was striking and is consistent with prior studies (17–19, 24). Though avoidant coping was strongly and negatively associated with grief severity, suicide deaths also accounted for additional variance in grief severity. A direct comparison of grief severity scores by cause of death (without age, gender, time since death, relationship covariates) indicated that suicide and accident survivors had *higher* grief severity compared to combat survivors. However, when coping strategies (and covariates)



were taken into account, avoidant coping, in particular, accounted for the variance associated with higher grief severity. In fact, the variance associated with suicide-loss was unexpectedly associated with *lower* grief severity in this model. These findings suggest that negative effects of suicide on grief severity described in the literature may be accounted for by the effect of avoidance rather than any unique characteristic of suicide deaths. This result is consistent with a previous study that also found that avoidant coping fully mediated the effect between suicide bereavement and grief (31). This relationship between avoidance and grief is consistent with the clinical literature that has characterized avoidance as a symptom of a disorder of prolonged and impairing grief (46) and a target of evidence-based interventions for this grief disorder (47, 48).

In addition to the effects of coping strategies, cause of death was distinctly related to outcomes, in that accident and suicide deaths (compared to combat deaths) had independent effects on

grief severity and posttraumatic growth. This pattern of results implies that there are circumstances unique to causes of death that are independent of coping strategies which impact these outcomes and warrant further investigation. It was anticipated that one of the characteristics that differs between causes of death might be whether a survivor had considered the possibility of death. Indeed, considering the possibility of a family member's death was differentially associated with each cause of death and with each outcome when examined alone, (i.e., without accounting for coping strategies). As hypothesized, 70% of combat-loss survivors considered the possibility of death, likely due to the known risk of combat operations. In contrast, only 29% of suicide-loss survivors considered the possibility of death. This pattern may be related to stigma associated with self-disclosure among those having thoughts of suicidal ideation (49, 50), or that even when informed that someone is struggling with emotional distress or mental illness, family members may deny the possibility of suicide as a potential outcome. Never considering the possibility of death was associated with higher grief severity than those who did consider it, consistent with Barry et al. (34) who found the same relationship between grief and preparedness.

However, considering the possibility of death did not account for additional variance in outcomes when coping strategies were included. Instead, the interaction between considering the possibility of death and specific coping strategies accounted for grief severity. Specifically, considering the possibility of death interacted with avoidant coping to predict grief severity. Though this interaction was significant in the full sample, when the sample was stratified by cause of death, it was only significant for combat-loss survivors. The interaction indicated that, overall, more avoidant coping was associated with increased grief severity. However, for those who never considered the possibility of death, increased use of avoidant coping was associated with even higher increases in grief severity compared to those who did consider the possibility. In other words, those who reported not considering the possibility of death and also engaged in avoidant coping (defined in this study as denial, behavioral disengagement, and self-blame) experienced an additive effect which led to higher grief severity. In fact, not considering the possibility of death may itself be an avoidant strategy. For family members of military service members who are involved in combat, such an avoidant strategy may be a healthy way to deny the risks that their loved one is engaging in. However, in the event of death, avoidance of the thought of death while employing continued avoidance coping would explain its additive effect and is consistent with evidence that avoidance behaviors prevent grief integration and lead to prolonged and impairing grief (51).

Considering the possibility of death also interacted with supportive coping to predict grief severity in combat-loss survivors. For those who never thought about the possibility of death, more supportive coping was associated with higher grief severity. At first glance this finding may appear confusing—why would supportive coping result in higher grief severity? However, these findings are associative rather than causal. An explanation of these results is that combat-bereaved survivors who never considered the possibility of the death of their loved ones were more likely to have higher grief severity, under which circumstances

they more actively engaged supportive coping. That we identified this relationship in combat-death survivors but not survivors of suicide or accidents may be due to the larger number of combat-death survivors in our sample. However, it could also be explained by the greater availability of community and grief support among combat-death survivors, especially compared to suicide, which is often associated with stigmatization and isolation (3).

Findings of the study should be interpreted in context of its limitations and strengths. Its cross-sectional design does not allow conclusions regarding causal mechanisms between coping strategy and grief severity, depression, or posttraumatic growth. In addition, the item used to assess consideration of the possibility of death required retrospective recall and may be subject to bias or inaccuracies, especially given the wide range of time since loss within the sample. It may be more precise to interpret this variable as the participant's perception of whether the possibility of death was considered. In addition, the sample consisted of approximately 80% female participants, so findings may not be as generalizable to males. Despite these limitations and to our knowledge, this is the first study to investigate the relationship between considering the possibility of death (or preparedness) and coping strategies. It is also the first to report on coping strategies used by combat-loss survivors compared to suicide-loss and accident-loss survivors. In addition, the large sample size allowed examination of distinct effects of each of the three coping strategies in combination with considering the possibility of death on grief severity, depression, and posttraumatic growth.

In conclusion, study findings highlight the differential contributions of coping strategies and their complex relationships with cause of death in contributing to grief severity, depression, and posttraumatic growth. Consistent with the existing literature, avoidant coping, in particular, was a potent contributor to negative outcomes, as well as an inhibitor of posttraumatic growth, suggesting its importance as a target for therapeutic intervention. Although supportive and active coping facilitated posttraumatic growth, they had less of a role in mitigating grief severity or depression in this study. These findings are consistent with therapeutic targets described in evidence-based programs (48, 52), and suggest that targeting coping strategies in community support programs or therapeutic interventions can minimize grief severity and promote posttraumatic growth among bereaved military family members. Specifically, efforts to decrease avoidance (e.g., accepting the reality of the death, enhancing behavioral and social engagement) and to increase supportive and active coping (e.g., seeking and accepting assistance, developing

meaningful understanding of the death) should be recommended approaches to enhance survivors' grief adaptation. Considering the possibility of death appeared to mitigate negative outcomes among survivors of combat death. Although this finding might suggest the importance of preparing military family members for the possibility of death, avoidance of that possibility is likely protective for the vast majority of family members whose loved ones return home safely.

DATA AVAILABILITY STATEMENT

The datasets presented in this article are not readily available because they are restricted due to privacy reasons. Requests to access the datasets should be directed to the Principal Investigator, National Military Family Bereavement Study (SC, stephen.cozza@usuhs.edu). Requests to access this data will be decided at the discretion of the Principal Investigator and the Uniformed Services University based upon the scientific merits of any proposed project.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by institutional review board at the Uniformed Services University of the Health Sciences. The participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

JF conceptualized the presented ideas and developed them in consultation with JZ and SC. JZ performed the data analyses and SC provided guidance. JF wrote the manuscript with substantial contributions from JZ and SC. RZ assisted with the preparation of the final version of the manuscript. CF and RU provided leadership.

FUNDING

This paper represents research funded by the Department of Defense—Congressionally Directed Medical Research Programs (W81XWH-11-2-0119) and the American Foundation for Suicide Prevention (Grant Number SRG-1-043-17).

REFERENCES

1. Stroebe M, Schut H, Stroebe W. Health outcomes of bereavement. *Lancet* (2007) 370(9603):1960–73. doi: 10.1016/S0140-6736(07)61816-9
2. Kristensen P, Weisæth L, Heir T. Bereavement and mental health after sudden and violent losses: a review. *Psychiatry* (2012) 75(1):76–97. doi: 10.1521/psyc.2012.75.1.76
3. Jordan JR. Is suicide bereavement different? A reassessment of the literature. *Suicide Life Threat Behav* (2001) 31(1):91–102. doi: 10.1521/suli.31.1.91.21310
4. Moore MM, Cerel J, Jobes DA. Fruits of trauma? Posttraumatic growth among suicide-bereaved parents. *Crisis* (2013) 36(4):241–8. doi: 10.1027/0227-5910/a000318
5. Pitman A, Osborn D, King M, Erlangsen A. Effects of suicide bereavement on mental health and suicide risk. *Lancet Psychiatry* (2014) 1(1):86–94. doi: 10.1016/s2215-0366(14)70224-x
6. Andriessen K. Suicide bereavement and postvention in major suicidology journals: lessons learned for the future of postvention. *Crisis* (2014) 35(5):338–48. doi: 10.1027/0227-5910/a000269
7. Hung NC, Rabin LA. Comprehending childhood bereavement by parental suicide: a critical review of research on outcomes, grief processes, and

- interventions. *Death Stud* (2009) 33(9):781–814. doi: 10.1080/07481180903142357
8. Kuramoto SJ, Brent DA, Wilcox HC. The impact of parental suicide on child and adolescent offspring. *Suicide Life Threat Behav* (2009) 39(2):137–51. doi: 10.1521/suli.2009.39.2.137
 9. Murphy SA, Johnson LC, Wu L, Fan JJ, Lohan J. Bereaved parents' outcomes 4 to 60 months after their children's deaths by accident, suicide, or homicide: a comparative study demonstrating differences. *Death Stud* (2003) 27(1):39–61. doi: 10.1080/07481180302871
 10. Carver CS, Scheier MF, Weintraub JK. Assessing coping strategies: a theoretically based approach. *J Pers Soc Psychol* (1989) 56(2):267–83. doi: 10.1037//0022-3514.56.2.267
 11. Lazarus RS, Folkman S. *Stress Appraisal and Coping*. New York: Springer (1984). 460p.
 12. Parker JD, Endler NS. Coping with coping assessment: a critical review. *Eur J Pers* (1992) 6(5):321–44. doi: 10.1002/per.2410060502
 13. Carver CS. You want to measure coping but your protocol's too long: consider the brief COPE. *Int J Behav Med* (1997) 4(1):92–100. doi: 10.1027/s15327558ijbm0401_6
 14. Buckley T, Spinaze M, Bartrop R, McKinley S, Whitfield V, Havyatt J, et al. The nature of death, coping response and intensity of bereavement following death in the critical care environment. *Aust Crit Care* (2015) 28(2):64–70. doi: 10.1016/j.aucc.2015.02.003
 15. Chae JH, Huh HJ, Choi WJ. Embitterment and bereavement: the Sewol ferry accident example. *Psychol Trauma* (2018) 10(1):46–50. doi: 10.1037/tra0000308
 16. Drapeau CW, Lockman JD, Moore MM, Cerel J. Predictors of posttraumatic growth in adults bereaved by suicide. *Crisis* (2019) 40(3):196–202. doi: 10.1027/0227-5910/a000556
 17. Harper M, O'Connor RC, O'Carroll RE. Factors associated with grief and depression following the loss of a child: a multivariate analysis. *Psychol Health Med* (2014) 19(3):247–52. doi: 10.1080/13548506.2013.811274
 18. Harper M, O'Connor RC, O'Carroll RE. The relative importance of avoidance and restoration-oriented stressors for grief and depression in bereaved parents. *Psychol Health Med* (2015) 20(8):906–15. doi: 10.1080/13548506.2014.990908
 19. McDevitt-Murphy ME, Zakarian RJ, Luciano MT, Olin CC, Mazzulo NN, Neimeyer RA. Alcohol use and coping in a cross-sectional study of African American homicide survivors. *J Ethn Subst Abuse* (2019) 18:1–16. doi: 10.1080/15332640.2019.1598905
 20. Murphy SA, Chung IJ, Johnson LC. Patterns of mental distress following the violent death of a child and predictors of change over time. *Res Nurs Health* (2002) 25(6):425–37. doi: 10.1002/nur.10060
 21. Murphy SA, Johnson LC, Chung IJ, Beaton RD. The prevalence of PTSD following the violent death of a child and predictors of change 5 years later. *J Trauma Stress* (2003) 16(1):17–25. doi: 10.1023/A:1022003126168
 22. Murphy SA, Johnson LC, Lohan J. The effectiveness of coping resources and strategies used by bereaved parents 1 and 5 years after the violent deaths of their children. *Omega (Westport)* (2003) 47(1):25–44. doi: 10.2190/HDUM-6A52-9958-7V0E
 23. Murphy SA, Johnson LC, Weber NA. Coping strategies following a child's violent death: how parents differ in their responses. *Omega (Westport)* (2002) 45(2):99–118. doi: 10.2190/D9NT-WC8V-KMJN-3P8T
 24. Schnider KR, Elhai JD, Gray MJ. Coping style use predicts posttraumatic stress and complicated grief symptom severity among college students reporting a traumatic loss. *J Couns Psychol* (2007) 54(3):344–50. doi: 10.1037/0022-0167.54.3.344
 25. Gutierrez F, Peri JM, Torres X, Caseras X, Valdés M. Three dimensions of coping and a look at their evolutionary origin. *J Res Pers* (2007) 41(5):1032–53. doi: 10.1016/j.jrp.2007.01.006
 26. Hasking PA, Oei TPS. Confirmatory factor analysis of the COPE questionnaire on community drinkers and an alcohol-dependent sample. *J Stud Alcohol* (2002) 63(5):631–40. doi: 10.15288/jsa.2002.63.631
 27. Litman JA. The COPE inventory: dimensionality and relationships with approach- and avoidance-motives and positive and negative traits. *Pers Individ Dif* (2006) 41(2):273–84. doi: 10.1016/j.paid.2005.11.032
 28. Lyne K, Roger D. A psychometric re-assessment of the COPE questionnaire. *Pers Individ Dif* (2000) 29(2):321–35. doi: 10.1016/S0191-8869(99)00196-8
 29. Wang AWT, Cheng CP, Chang CS, Chen DR, Chen ST, Shieh V, et al. Does the factor structure of the Brief COPE fit different types of traumatic events? A test of measurement invariance. *Eur J Psychol Assess* (2018) 34(3):162–73. doi: 10.1027/1015-5759/a000321
 30. Anderson MJ, Marwit SJ, Vandenberg B, Chibnall JT. Psychological and religious coping strategies of mothers bereaved by the sudden death of a child. *Death Stud* (2005) 29(9):811–26. doi: 10.1080/07481180500236602
 31. Nam I. Suicide bereavement and complicated grief: experiential avoidance as a mediating mechanism. *J Loss Trauma* (2016) 21(4):325–34. doi: 10.1080/15325024.2015.1067099
 32. Bailey SE, Kral MJ, Dunham K. Survivors of suicide do grieve differently: empirical support for a common sense proposition. *Suicide Life Threat Behav* (1999) 29(3):256–71. doi: 10.1111/j.1943-278X.1999.tb00301.x
 33. Farberow NL, Gallagher-Thompson D, Gilewski M, Thompson L. The role of social supports in the bereavement process of surviving spouses of suicide and natural deaths. *Suicide Life Threat Behav* (1992) 22(1):107–24. doi: 10.1111/j.1943-278X.1992.tb00479.x
 34. Barry LC, Kasl SV, Prigerson HG. Psychiatric disorders among bereaved persons: the role of perceived circumstances of death and preparedness for death. *A J Geriatr Psychiatry* (2002) 10(4):447–57. doi: 10.1097/00019442-200207000-00011
 35. Burton AM, Haley WE, Small BJ. Bereavement after caregiving or unexpected death: effects on elderly spouses. *Aging Ment Health* (2006) 10(3):319–26. doi: 10.1080/13607860500410045
 36. Keyes KM, Pratt C, Galea S, McLaughlin KA, Koenen KC, Shear MK. The burden of loss: unexpected death of a loved one and psychiatric disorders across the life course in a national study. *Am J Psychiatry* (2014) 171(8):864–71. doi: 10.1176/appi.ajp.2014.13081132
 37. Prigerson HG, Maciejewski PK, Reynolds III CF, Bierhals AJ, Newsom JT, Fasiczka A, et al. Inventory of Complicated Grief: a scale to measure maladaptive symptoms of loss. *Psychiatry Res* (1995) 59(1):65–79. doi: 10.1016/0165-1781(95)02757-2
 38. Boelen PA, de Keijser J, van den Hout MA, van den Bout J. Treatment of complicated grief: a comparison between cognitive-behavioral therapy and supportive counseling. *J Consult Clin Psychol* (2007) 75(2):277–84. doi: 10.1037/0022-006X.75.2.277
 39. Mitchell AM, Kim Y, Prigerson HG, Mortimer-Stephens M. Complicated grief in survivors of suicide. *Crisis* (2004) 25(1):12–8. doi: 10.1027/0227-5910.25.1.12
 40. Szanto K, Shear MK, Houck PR, Reynolds III CF, Frank E, Caroff K, et al. Indirect self-destructive behavior and overt suicidality in patients with complicated grief. *J Clin Psychiatry* (2006) 67(2):233–9. doi: 10.4088/jcp.v67n0209
 41. Kroenke K, Spitzer RL, Williams JB. The PHQ-9: validity of a brief depression severity measure. *J Gen Intern Med* (2001) 16(9):606–13. doi: 10.1046/j.1525-1497.2001.016009606
 42. Cann A, Calhoun LG, Tedeschi RG, Taku K, Vishnevsky T, Triplett KN, et al. A short form of the Posttraumatic Growth Inventory. *Anxiety Stress Coping* (2010) 23(2):127–37. doi: 10.1080/10615800903094273
 43. Liang KY, Zeger SL. Longitudinal data analysis using generalized linear models. *Biometrika* (1986) 73(1):13–22. doi: 10.1093/biomet/73.1.13
 44. Wolchik SA, Cox S, Tein JY, Sandler IN, Ayers TS. Six-year longitudinal predictors of posttraumatic growth in parentally bereaved adolescents and young adults. *Omega (Westport)* (2009) 58(2):107–28. doi: 10.2190/om.58.2.b
 45. Michael C, Cooper M. Post-traumatic growth following bereavement: a systematic review of the literature. *Couns Psychol Rev* (2013) 28(4):18–33.
 46. American Psychiatric Association. *Diagnostic and statistical manual of mental disorders*. Virginia: American Psychiatric Association (2013). 947p.
 47. Shear MK. Complicated grief. *N Engl J Med* (2015) 372(2):153–60. doi: 10.1056/NEJMcip1315618
 48. Shear MK, Frank E, Houck PR, Reynolds III CF. Treatment of complicated grief: a randomized controlled trial. *JAMA* (2005) 293(21):2601–8. doi: 10.1001/jama.293.21.2601

49. Hom MA, Stanley IH, Podlogar MC, Joiner. TE. "Are you having thoughts of suicide?" Examining experiences with disclosing and denying suicidal ideation. *J Clin Psychol* (2017) 73(10):1382–92. doi: 10.1002/jclp.22440
50. Ganzini L, Denneson LM, Press N, Bair MJ, Helmer DA, Poat J, et al. Trust is the basis for effective suicide risk screening and assessment in veterans. *J Gen Intern Med* (2013) 28(9):1215–21. doi: 10.1007/s11606-013-2412-6
51. Shear MK. Exploring the role of experiential avoidance from the perspective of attachment theory and the dual process model. *Omega (Westport)* (2010) 61(4):357–69. doi: 10.2190/OM.61.4.f
52. Shear MK, Wang Y, Skritskaya N, Duan N, Mauro C, Ghesquiere A. Treatment of complicated grief in elderly persons: a randomized clinical trial. *JAMA Psychiatry* (2014) 71(11):1287–95. doi: 10.1001/jamapsychiatry.2014.1242

Disclaimer: The opinions and assertions expressed herein are those of the author (s) and do not necessarily reflect the official policy or position of the Uniformed Services University, the Department of Defense, or the American Foundation for Suicide Prevention.

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Copyright © 2020 Fisher, Zhou, Zuleta, Fullerton, Ursano and Cozza. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.



ICD-11 Prolonged Grief Disorder Criteria: Turning Challenges Into Opportunities With Multiverse Analyses

Maarten C. Eisma^{1*}, Rita Rosner² and Hannah Comtesse²

¹ Department of Clinical Psychology and Experimental Psychopathology, University of Groningen, Groningen, Netherlands,

² Department of Psychology, Catholic University Eichstaett-Ingolstadt, Ingolstadt, Germany

Keywords: prolonged grief disorder, persistent complex bereavement disorder, complicated grief, criteria, algorithm, criticism, critique, multiverse analyses

OPEN ACCESS

Edited by:

Birgit Wagner,
Medical School Berlin, Germany

Reviewed by:

Sebastian Trautmann,
Medical School Hamburg, Germany
Nadine Stammel,
Freie Universität Berlin, Germany
Carina Heeke,
Freie Universität Berlin, Germany

*Correspondence:

Maarten C. Eisma
m.c.eisma@rug.nl

Specialty section:

This article was submitted to
Mood and Anxiety Disorders,
a section of the journal
Frontiers in Psychiatry

Received: 24 February 2020

Accepted: 16 July 2020

Published: 07 August 2020

Citation:

Eisma MC, Rosner R and Comtesse H
(2020) ICD-11 Prolonged Grief
Disorder Criteria: Turning Challenges
Into Opportunities With
Multiverse Analyses.
Front. Psychiatry 11:752.
doi: 10.3389/fpsy.2020.00752

INTRODUCTION

Recently, prolonged grief disorder (PGD), a diagnosis characterized by severe, persistent and disabling grief, was formally included in the 11th revision of the International Classification of Diseases [ICD-11; (1): **Table 1**]. To meet PGD_{ICD-11} criteria one needs to experience persistent and pervasive longing for the deceased and/or persistent and pervasive cognitive preoccupation with the deceased, combined with *any* of 10 additional grief reactions assumed indicative of intense emotional pain for at least six months after bereavement. Contrary to the 5th revision of the Diagnostical and Statistical Manual of Mental Disorders [DSM-5; (11)] and the 10th revision of the International Classification of Diseases [ICD-10; (12)], the ICD-11 only uses a typological approach, implying that diagnosis descriptions are simple and there is no strict requirement for the number of symptoms one needs to experience to meet the diagnostic threshold.

Some researchers have argued that PGD_{ICD-11}'s typological approach is helpful, as it will lead to greater sensitivity in case identification in clinical practice and increased cross-cultural applicability (13). Others have highlighted that the typological approach allows for flexible diagnostic algorithms in research, so that PGD_{ICD-11} criteria can be adapted to resemble the characteristics of both stricter and more lenient precursor criteria (14). In the current contribution, we take a different, complementary position. We highlight a series of challenges in using the PGD criteria for research purposes and discuss the application of a method that employs flexibility of PGD_{ICD-11} diagnostic approach to address these challenges, which may help in working toward the unbiased, structured, and transparent identification of optimal criteria for disturbed grief.

A CRITIQUE OF PGD ICD-11 FOR RESEARCH PURPOSES

A first challenge to researchers applying PGD_{ICD-11} criteria is that they were completely new when first introduced and differed substantially from previously proposed diagnostic criteria sets (15). For example, PGD_{ICD-11} contains multiple symptoms not found in any prior proposed criteria set, such as guilt, blame and the inability to experience positive mood [for a full criteria set comparison: (16)]. Furthermore, oft-used measures to assess disturbed grief responses, such as versions of the

TABLE 1 | Overview of ICD-11 diagnostic criteria for PGD and results of multiverse analyses of PGD_{ICD-11}.

ICD-11 criteria for PGD	Study	Sample	Measure ¹	Results
A. At least one of the following: a persistent and pervasive longing for the deceased or a persistent and pervasive preoccupation with the deceased.	Boelen and Lenferink (2)	855 bereaved community members	ICG-R, SCL-90 depression scale	Prevalence rate of PGD_{ICD-11} and diagnostic agreement with PCBD, PGD₂₀₀₉, PGD_{ICD-11} BD, CG, and PGD_{DSM-5}², respectively, for various additional symptom thresholds: 1+ = 19.8%, κ = .66, .67, .98, .92, .93; 2+ = 18.1%, κ = .69, .73, .96, .89, .94; 3+ = 16.1%, κ = .75, .79, .89, .82, .89; 4+ = 13.4%, κ = .78, .82, .79, .72, .81; 5+ = 10.8%, κ = .83, .82, .67, .61, .72; 6+ = 7.8%, κ = .76, .76, .52, .47, .56; 7+ = 5.0%, κ = .59, .57, .36, .32, .39 Number of possible symptom combinations to meet PGD_{ICD-11} caseness for various additional symptom thresholds: 1+ = 3069, 2+ = 3039, 3+ = 2904, 4+ = 2544, 5+ = 1914, 6+ = 1158, 7+ = 528
B. Examples of intense emotional pain Accompanied by intense emotional pain e.g. sadness, guilt, anger, denial, blame; difficulty accepting the death; feeling one has lost a part of one's self; an inability to experience positive mood; emotional numbness; difficulty in engaging with social or other activities.	Boelen et al. (3)	551 bereaved community members	ICG-R, SCL-90 depression scale	Prevalence rate of PGD_{ICD-11} and diagnostic agreement with PCBD for various additional symptom thresholds: 1+ = 19.2%, κ = .51, 2+ = 17.6%, κ = .56; 3+ = 15.4%, κ = .62; 4+ = 11.1%, κ = .75; 5+ = 8.3%, κ = .84, 6+ = 5.3%, κ = .71
C. Time and impairment Persisted for an abnormally long period of time (more than 6 months at a minimum); following the loss, clearly exceeding expected social, cultural or religious norms for the individual's culture and context. The disturbance causes significant impairment in personal, family, social, educational, occupational or other important areas of functioning.	Bonanno and Malgaroli (4)	282 bereaved community members	"Structured clinical interview"	Prevalence rate of PGD_{ICD-11} and diagnostic agreement with PCBD for various symptom thresholds: 1+ = 11.7%, percentage match = 36.4%, 3+ = 8.8%, percentage match = 48.0%; 5+ = 4.6%, percentage match = 76.9%
	Comtesse et al. (5)	113 bereaved treatment-seekers	ICG, PG-13	Sensitivity and specificity values from ROC analysis of PGD_{ICD-11} for various additional symptoms thresholds (interview-diagnosed PGD₂₀₀₉ as criterion standard): 1+ = 100% and 6%, 2+ = 100% and 9%, 3+ = 100% and 30%, 4+ = 98% and 56%, 5+ = 88% and 75%, 6+ = 82% and 92%, 7+ = 65% and 96%, 8+ = 37% and 100%, 9+ = 15% and 100%, 10+ = 7% and 100% Prevalence rate of PGD_{ICD-11} and diagnostic agreement with PCBD for various additional symptom thresholds: 1+ = 69.1%, κ = .51; 6+ = 46.9%, κ = .77
	Cozza et al. (6)	1732 bereaved military family members	CGQ	Diagnostic agreement of PGD_{ICD-11} with PCBD, PGD₂₀₀₉, and CG, respectively, for thresholds of one or two additional symptoms for all criteria-sets: κ for PGD _{ICD-11} and PCBD = .90 or .87, κ for PGD _{ICD-11} and PGD ₂₀₀₉ = .89 or .86, κ for PGD _{ICD-11} and CG = .93 or .89
	Mauro et al. (7)	261 bereaved treatment-seekers	SCI-CG	Diagnostic agreement of PGD_{ICD-11} with ICG cut-off ≥ 30 and clinician-rated CG and functional impairment for various additional symptom thresholds: 0+ = 95.8%, 1+ = 95.8%, 2+ = 95.0%, 3+ = 91.6%, 4+ = 86.2%, 5+ = 72.8%, 6+ = 45.2% (5+ and 6+ estimates fall in between PGD ₂₀₀₉ rates of agreement: 59.0%)

PGD, prolonged grief disorder; PGD_{ICD-11}, ICD-11 diagnostic criteria for PGD; PGD_{ICD-11} BD, diagnostic criteria according to the ICD-11 beta draft [or a description see (2)]; PGD₂₀₀₉, diagnostic criteria for PGD according to Prigerson et al. (3); PCBD, DSM-5 diagnostic criteria for persistent complex bereavement disorder; CG, diagnostic criteria for complicated grief (9); CGQ, Complicated Grief Questionnaire; ICG, Inventory of Complicated Grief; ICG-R, Inventory of Complicated Grief Revised; PG-13, Interview for Prolonged Grief-13; SCI-CG, Structured Clinical Interview for Complicated Grief; SCL-90, Symptom-Checklist-90. ¹ In all cited studies, items of the respective measure(s) were used to assess (approximations of) PGD_{ICD-11} symptoms. Symptoms were regarded as present if judged as 'present' by an interviewer (4) or if an item was scored higher than a specific value (e.g., ≥ 4) on the respective scale(s) (e.g., five-point Likert scale). Each symptom score was dichotomously coded as "absent" (0) or "present" (1). PGD_{ICD-11} caseness was then determined following the ICD-11 diagnostic rule.

² PGD_{DSM-5} was interpreted here according to proposed guidelines [(10), i.e., three additional symptoms].

Inventory of Complicated Grief [e.g., ICG; (17)] do not fully assess PGD_{ICD-11} criteria [6; for a recent review illustrating this point: (18)]. Therefore, the development of new, reliable and valid instruments is critical to assess the characteristics and validity of PGD_{ICD-11} and determine for common research purposes [e.g., establishing prevalence, risk factors, treatment efficacy] who meets PGD_{ICD-11} criteria. However, the typological approach in PGD_{ICD-11} poses a substantial challenge to the development of such instruments.

First, the plain language used to formulate additional criteria makes it unclear what precisely each criterion implies. Single-

word criteria "guilt," "anger," "denial," and "blame" are particularly problematic. For example, "blame" could refer to self-blame or other-blame, blame for the death, or blame for something else. Since self-blame is much more prevalent in bereaved persons than blaming others (19) the interpretation of this criterion influences its prevalence. Moreover, blaming oneself for the death perpetuates disturbed grief, whereas blaming others for the death does not (20), so the characteristics and clinical correlates of this criterion may be very different depending on how it is interpreted [for a related discussion: (21)].

Second, even core criteria of “longing” and “preoccupation,” shared with most prior proposed grief disorders, are potentially problematic in their implementation. For example, it is unclear if a higher item-score threshold should be used to indicate that someone experiences persistent and pervasive longing, as it is the most frequently reported experience in bereavement (22), and, as a consequence, one of the least sensitive criteria in distinguishing those with and without disturbed grief (14). Preoccupation with the deceased is also already being interpreted differently by influential researchers in the grief field, with some viewing it as intrusive images about the death (23), and others as a process similar to grief rumination (24). While imagery and rumination are related processes, they are dissimilar in phenomenal characteristics, such as their duration, sensory experiences, and emotional correlates (25). The interpretation of this criterion will therefore have repercussions for what we regard as PGD.

Third, many key characteristics of PGD_{ICD-11} (e.g., prevalence, classification, symptom heterogeneity) depend heavily on the chosen diagnostic algorithm. Pioneering research on characteristics of this disorder (with measures approximating actual criteria) used the minimal criteria as specified in the ICD-11 (i.e., at least one core criterion, and at least one additional criterion) in addition to the time and disability criteria (7, 26). It soon became apparent that applying these minimal criteria led to much higher prevalence rates for PGD_{ICD-11} than for prior proposed criteria of PGD [PGD₂₀₀₉; (8)] and persistent complex bereavement disorder [PCBD; DSM-5, (10)]. This algorithm is thus relatively lenient, and applying it may lead to overdiagnosis and limited generalizability of findings on two of the most-studied grief disorder proposals (i.e., PGD₂₀₀₉; PCBD) to PGD_{ICD-11} (14). This elicits the question: If the diagnostic algorithm directly derived from the ICD-11 text is too liberal, which diagnostic rules are then optimal for research?

MULTIVERSE ANALYSES IN RESEARCH ON PGD ICD-11

In summary, a fundamental challenge for grief researchers in using the PGD_{ICD-11} diagnosis is that its criteria are open for multiple interpretations and that the only diagnostic algorithm mapping one-on-one on the diagnosis description is too lenient. While the current criteria cannot easily be amended, their systematic investigation can make them more useful to researchers, for instance by providing a basis for achieving consensus on symptom interpretation, algorithms, and future PGD criteria. We propose that multiverse analyses can be particularly helpful in achieving such goals. Multiverse analyses typically consist of a procedure wherein one performs similar analyses across multiple datasets generated by making reasonable but variable choices on excluding, transforming and coding data (27). For example, when using a reaction time task with skewed data, one may perform analyses based on the median or the mean or analyze the data using parametric or non-parametric statistical tests. By comparing outcomes of multiple analyses, one can establish the degree of uncertainty about the conclusions one arrives at and the robustness of findings to arbitrary decisions

made in data preparation and analysis. For example, one may discover that the direction and significance of effects is similar regardless of these decisions or that some decisions lead to significant effects, whereas others do not. The first scenario would allow for strong conclusions and the second scenario would signal caution is warranted in the interpretation of findings.

We advocate a similar but conceptually distinct procedure wherein empirical research examining the characteristics of PGD_{ICD-11} systematically vary certain aspects of these criteria (e.g., using a more stringent cut-off for longing) or the diagnostic algorithm (e.g., varying the number of additional symptoms). A comparison of results obtained with multiple interpretations of criteria can help illuminate how robust specific results are dependent on multiple interpretations of the PGD_{ICD-11} criteria. For example, one may be able to investigate the robustness of group differences between people with and without PGD on risk-factors and protective factors or treatment effectiveness (e.g., percentage with and without diagnosis after treatment) dependent on different interpretations of PGD_{ICD-11}. Additionally, critical information can be gathered on the influence of variations in symptom interpretations and algorithms on PGD_{ICD-11} characteristics and how these characteristics compare to other proposed criteria sets [e.g., the newly developed PGD criteria for the upcoming text revision of DSM-5, (10)]. That is, multiverse analyses can be applied to shed light on a variety of clinically relevant characteristics of PGD_{ICD-11} (e.g., retest and interrater reliability, specificity and sensitivity of classification, distinctiveness from other disorders, associations with functional impairment) when systematically modifying interpretations of its criteria.

Only a handful of studies have thus far applied such analyses, which have predominantly narrowly focused on examining characteristics of PGD_{ICD-11} and their comparability against external standards when varying the number of additional criteria (see **Table 1** for a summary). It has been observed that minimal PGD_{ICD-11} criteria yield a similar prevalence as the relatively lenient Shear et al. criteria (9) for complicated grief (7), but almost two times higher prevalence than relatively strict PCBD criteria (2, 28, 29). Multiverse analyses in community samples demonstrated that similar prevalence estimates and good diagnostic agreement with PCBD and PGD₂₀₀₉ appears to be achieved with five additional criteria for PGD_{ICD-11} [(2–4), cf. (7)]. Similarly, in treatment-seekers, minimal PGD_{ICD-11} criteria correctly classified people against a relatively lenient standard of 30 or higher on the ICG (6, 7), yet as many as six additional symptoms were necessary to yield comparable prevalence and good diagnostic agreement with PGD₂₀₀₉ and PCBD (5). Moreover, one study demonstrated the influence of the number of additional symptoms on symptom heterogeneity, theoretically demonstrating that the number of ways to meet PGD_{ICD-11} criteria ranges from 3.069 for one to 528 for seven additional symptoms (2). A less heterogeneous diagnosis is clearly preferable, as it would lead to less variability within groups of people meeting grief disorder criteria, making the distinction between these people more useful for research and practice (30). Taken together, these examples illustrate that the

properties of PGD_{ICD-11} depend on both the chosen diagnostic rule and the stringency of the comparison standard, and that the number of additional symptoms is critical in determining prevalence, clinical classification, and symptom profiles.

For future research, we recommend multiverse analyses varying not only the algorithm, but additionally symptom interpretations of single-item criteria and cognitive preoccupation and cut-offs for the presence of the longing criterion. We also advise to substantially expand the current focus of multiverse analyses of PGD_{ICD-11} to establish the robustness of clinically-relevant findings (e.g., on treatment efficacy) and the variety of other aspects relevant to the validity of a diagnosis [for reviews: (13, 24)]. The latter includes - but is not limited to: reliability of classification [e.g., (31)], the structure of symptoms [e.g., (32)], distinctiveness from related disorders [e.g., (33)], and relationships with functional impairment [e.g., (34)]. We further advocate transparency in applying multiverse analyses and recommend: open access publication, data accessibility (e.g., through availability in repositories), fully specifying the origins and formulations of items used to assess PGD_{ICD-11} and, if applicable, other disturbed grief criteria, and complete reporting of the variations of PGD_{ICD-11} and outcomes under investigation.

DISCUSSION

In the absence of clearly defined criteria and diagnostic rules for PGD_{ICD-11}, researchers should broadly apply structured methods to examine the characteristics of this disorder and compare it against past and future proposed grief disorders. Multiverse analyses can be a powerful tool to determine the validity and clinical usefulness of the PGD_{ICD-11} criteria. By systematically varying the number of core and additional symptoms, the interpretation of symptoms, and the

standards for meeting symptoms, we can evaluate how such decisions influence the characteristics of PGD_{ICD-11}, also in relation to different external standards. This will create a comprehensive research base enabling us to enhance our understanding of PGD_{ICD-11} and of disturbed grief more generally. Creating this research base is no panacea: it cannot undo the inherent weaknesses of the PGD_{ICD-11} criteria. However, the systematic evaluation of this information will help clarify under which circumstances diagnoses behave similarly or differently, providing a stepping stone to harmonize PGD_{ICD-11} criteria with other criteria sets and to develop more optimal future disturbed grief criteria.

AUTHOR CONTRIBUTIONS

All authors developed the ideas for this article. ME and HC wrote the first draft of the manuscript. RR contributed important intellectual content by providing critical revisions to the first draft.

FUNDING

ME was supported by a Netherlands Organization for Scientific Research (NWO) Veni grant [Grant ID: 016.veni195.113]. The funder did not play a role in the study design, the writing of the report or in the decision to submit the article for publication. RR was supported by a grant of Deutsche Forschungsgemeinschaft [Grant ID: RO2042/7-1]. The funder did not play a role in the study design, the writing of the report or the decision to submit the article for publication.

REFERENCES

1. World Health Organization. *ICD-11 beta draft website [Information]*. (2018). Available at: <https://icd.who.int/browse11/l-m/en> (Accessed on 24th June 2020).
2. Boelen PA, Lenferink LIM. Comparison of six proposed diagnostic criteria sets for disturbed grief. *Psychiatry Res* (2020) 285:112786. doi: 10.1016/j.psychres.2020.112786
3. Boelen PA, Lenferink LIM, Smid GE. Further evaluation of the factor structure, prevalence, and concurrent validity of DSM-5 criteria for persistent complex bereavement disorder and ICD-11 criteria for prolonged grief disorder. *Psychiatry Res* (2019a) 3:206–10. doi: 10.1016/j.psychres.2019.01.006
4. Bonanno GA, Malgaroli M. Trajectories of grief: Comparing symptoms from the DSM-5 and ICD-11 diagnoses. *Depress Anxiety* (2020) 37:17–25. doi: 10.1002/da.22902
5. Comtesse H, Vogel A, Kersting A, Rief W, Steil R, Rosner R. When does grief become pathological? Evaluation of the ICD-11 diagnostic proposal for prolonged grief in a treatment-seeking sample. *Eur J Psychotraumatol* (2020) 11:1694348. doi: 10.1080/20008198.2019.1694348
6. Cozza SJ, Shear MK, Reynolds CF, Fisher JE, Zhou J, Maercker A, et al. Optimizing the clinical utility of four proposed criteria for a persistent and impairing grief disorder by emphasizing core, rather than associated symptoms. *Psychol Med* (2020) 50:438–45. doi: 10.1017/S0033291719000254
7. Mauro C, Reynolds CF, Maercker A, Skritskaya N, Simon N, Zisook S, et al. Prolonged grief disorder: Clinical utility of ICD-11 diagnostic guidelines. *Psychol Med* (2019) 49:861–7. doi: 10.1017/S0033291718001563
8. Prigerson HG, Horowitz MJ, Jacobs SC, Parkes CM, Aslan M, Goodkin K, et al. Prolonged grief disorder: Psychometric validation of criteria proposed for DSM-V and ICD-11. *PLoS Med* (2009) 6:e1000121. doi: 10.1371/journal.pmed.1000121
9. Shear MK, Simon N, Wall M, Zisook S, Neimeyer R, Duan N, et al. Complicated grief and related bereavement issues for DSM-5. *Depress Anxiety* (2011) 28:103–17. doi: 10.1002/da.20780
10. American Psychiatric Association. *View and comment on recently proposed changes to DSM-5*. (2020). Available at: <https://www.psychiatry.org/psychiatrists/practice/dsm/proposed-changes> (Accessed on 24th of June 2020).
11. American Psychiatric Association. *Diagnostic and statistical manual of mental disorders*. 5th ed. American Psychiatric Publishing: Arlington, VA (2013).
12. World Health Organization. *ICD-10: international statistical classification of diseases and related health problems: tenth revision*. 2nd ed. Geneva, Switzerland: World Health Organization (2004).
13. Killikelly C, Maercker A. Prolonged grief disorder for ICD-11: The primacy of clinical utility and international applicability. *Eur J Psychotraumatol* (2017) 8:1476441. doi: 10.1080/20008198.2018.1476441
14. Simon NM, Wall MM, Keshaviah A, Dryman MT, LeBlanc NJ, Shear MK. Informing the symptom profile of complicated grief. *Depress Anxiety* (2011) 28:118–26. doi: 10.1002/da.20775

15. Eisma MC, Lenferink LIM. Response to: Prolonged grief disorder for ICD-11: The primacy of clinical utility and international applicability. *Eur J Psychotraumatol* (2017) 8:1512249. doi: 10.1080/20008198.2018.1512249
16. Lenferink LIM, Boelen PA, Smid G, Paap M. The importance of harmonising diagnostic criteria sets for pathological grief. *Brit J Psychiat* (in press), 1–4. doi: 10.1192/bjp.2019.240
17. Prigerson HG, Maciejewski PK, Reynolds CF, Bierhals AJ, Newsom JT, Fasiczka A, et al. Inventory of complicated grief: A scale to measure maladaptive symptoms of loss. *Psychiatry Res* (1995) 59:65–79. doi: 10.1016/0165-1781(95)02757-2
18. Trembl J, Kaiser J, Plexnies A, Kersting A. Assessing prolonged grief disorder: A systematic review of assessment instruments. *J Affect Disord* (2020) 274:420–34. doi: 10.1016/j.jad.2020.05.049
19. Davis CG, Lehman DR, Wortman CB, Silver RC, Thompson SC. The undoing of traumatic life events. *Pers Soc Psychol Bull* (1995) 21:109–24. doi: 10.1177/0146167295212002
20. Eisma MC, Epstude K, Schut HAW, Stroebe MS, Simion A, Boelen PA. Upward and downward counterfactuals after loss: A multi-wave controlled longitudinal study. *Behav Ther* (in press). doi: 10.1016/j.beth.2020.07.007
21. Greene T. Blame, PTSD and DSM-5: An urgent need for clarification. *Eur J Psychotraumatol* (2018) 9:1468709. doi: 10.1080/20008198.2018.1468709
22. Maciejewski PK, Zhang B, Block SD, Prigerson HG. An empirical examination of the stage theory of grief. *JAMA* (2007) 297:716–23. doi: 10.1001/jama.297.7.716
23. Boelen PA, Smid GE. The Traumatic Grief Inventory Self-Report Version (TGI-SR): Introduction and preliminary psychometric evaluation. *J Loss Trauma* (2017) 22:196–212. doi: 10.1080/15325024.2017.1284488
24. Simon NM, Shear MK, Reynolds CF, Cozza SJ, Mauro C, Zisook S, et al. Commentary on evidence in support of a grief-related condition as a DSM diagnosis. *Depress Anxiety* (2020) 37:9–16. doi: 10.1002/da.22985
25. Speckens AE, Ehlers A, Hackmann A, Ruths FA, Clark DM. Intrusive memories and rumination in patients with post-traumatic stress disorder: A phenomenological comparison. *Mem* (2007) 15:249–57. doi: 10.1080/09658210701256449
26. Boelen PA, Lenferink LIM, Nickerson A, Smid GE. Evaluation of the factor structure, prevalence, and validity of disturbed grief in DSM-5 and ICD-11. *J Affect Disord* (2018) 240:79–87. doi: 10.1016/j.jad.2018.07.041
27. Steegen S, Tuerlinckx F, Gelman A, Vanpaemel W. Increasing transparency through a multiverse analysis. *Perspect Psychol Sci* (2016) 11:702–12. doi: 10.1177/1745691616658637
28. Boelen PA, Spuij M, Lenferink LIM. Comparison of DSM-5 criteria for persistent complex bereavement disorder and ICD-11 criteria for prolonged grief disorder in help-seeking bereaved children. *J Affect Disord* (2019b) 250:71–8. doi: 10.1016/j.jad.2019.02.046
29. O'Connor M, Lasgaard M, Larsen L, Johannsen M, Lundorff M, Farver-Vestergaard I, et al. Comparison of proposed diagnostic criteria for pathological grief using a sample of elderly bereaved spouses in Denmark: Perspectives on future bereavement research. *J Affect Disord* (2019) 251:52–9. doi: 10.1016/j.jad.2019.01.056
30. Lenferink LIM, Eisma MC. 37.650 ways to have “persistent complex bereavement disorder” yet only 48 ways to have “prolonged grief disorder”. *Psychiatry Res* (2018) 261:88–9. doi: 10.1016/j.psychres.2017.12.050
31. Lichtenthal WG, Maciejewski PK, Demirjian CC, Roberts KE, First MB, Kissane DW, et al. Evidence of the clinical utility of a prolonged grief disorder diagnosis. *World Psychiatry* (2018) 17:364–5. doi: 10.1002/wps.20544
32. Golden A-M, Dalgleish T. Is prolonged grief distinct from bereavement-related posttraumatic stress? *Psychiatry Res* (2010) 178:336–41. doi: 10.1016/j.psychres.2009.08.021
33. Malgaroli M, Maccallum F, Bonanno G. Symptoms of persistent complex bereavement disorder, depression, and PTSD in a conjugally bereaved sample: A network analysis. *Psychol Med* (2018) 48:1–10. doi: 10.1017/S0033291718001769
34. Maccallum F, Bryant RA. A network approach to understanding quality of life impairments in prolonged grief disorder. *J Trauma Stress* (2020) 31:106–15. doi: 10.1002/jts.22383

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Copyright © 2020 Eisma, Rosner and Comtesse. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.



The UK National Homicide Therapeutic Service: A Retrospective Naturalistic Study Among 929 Bereaved Individuals

Suzan Soydas^{1,2*}, Geert E. Smid^{2,3}, Barbara Goodfellow⁴, Rachel Wilson⁴ and Paul A. Boelen^{1,2}

¹ Department of Clinical Psychology, Utrecht University, Utrecht, Netherlands, ² ARQ Centrum'45, ARQ National Psychotrauma Centre, Diemen, Netherlands, ³ University of Humanistic Studies, Utrecht, Netherlands, ⁴ ASSIST Trauma Care, Rugby, United Kingdom

OPEN ACCESS

Edited by:

Paul Stokes,
King's College London,
United Kingdom

Reviewed by:

Anik Debrot,
University of Lausanne, Switzerland
Liliana Dell'Osso,
University of Pisa, Italy
Liliane Efinger,
University of Lausanne, Switzerland

*Correspondence:

Suzan Soydas
s.soydas@uu.nl

Specialty section:

This article was submitted to
Mood and Anxiety Disorders,
a section of the journal
Frontiers in Psychiatry

Received: 13 November 2019

Accepted: 11 August 2020

Published: 28 August 2020

Citation:

Soydas S, Smid GE, Goodfellow B,
Wilson R and Boelen PA (2020) The
UK National Homicide Therapeutic
Service: A Retrospective
Naturalistic Study Among
929 Bereaved Individuals.
Front. Psychiatry 11:878.
doi: 10.3389/fpsyt.2020.00878

Homicidal bereavement puts survivors at risk of developing a broad range of lasting and severe mental health problems. Previous research has often relied on relatively small and homogenous samples. Still, little is known about what factors influence the expression of symptoms following homicidal bereavement. Preventive and curative treatments often do not consider the complex coherence between the emotional, judicial, financial, and societal challenges that likely arise following a homicide. Despite the severity of its consequences on mental health, no gold standard for the preventative and curative treatment of mental health issues in homicide survivors exists. We aimed to introduce a time-limited, traumatic grief-focused outreaching model of care designed specifically for homicide survivors, and to examine its potential effectiveness. Furthermore, we aimed to investigate what factors influence the severity of mental health problems and response to treatment. In the current study, self-reported data on five different outcome measures, namely, symptoms of posttraumatic stress, prolonged grief, depression, anxiety, and functional impairment were available from 929 homicidally bereaved treatment receiving adults. We used Latent Growth Modeling to analyze our repeated measures data and to classify individuals into distinct groups based on individual response patterns. Results showed that the current model of care is likely to be effective in reducing mental health complaints following homicidal bereavement. Having a history of mental illness, being younger of age and female, and having lost either a child or spouse consistently predicted greater symptom severity and functional impairment at baseline. For change in symptom severity and functional impairment during treatment, having a history of mental illness was the only consistent predictor across all outcomes. This study was limited by its reliance on self-reported data and cross-sectional design without a control group. Future prospective, longitudinal research across different cultures is needed in order to

replicate the current findings and enhance generalizability. That notwithstanding, findings provide a first step toward evaluating a novel service-delivery approach for homicide survivors and provide further insight in the development of mental health complaints following bereavement by homicide.

Keywords: homicide, bereavement, loss, grief, posttraumatic stress disorder, bereavement interventions, psychological interventions

INTRODUCTION

In response to the death of a loved one, many people experience acute emotional distress and symptoms of mental health disorders, which generally decrease over time. However, for a significant minority, these symptoms may persist and develop into psychopathology, such as posttraumatic stress disorder (PTSD), major depressive disorder (MDD), anxiety disorders and prolonged grief disorder (PGD) [e.g., (61, 62)]. In recent literature on pathological grief reactions, PGD (1) is also referred to as persistent complex bereavement disorder (PCBD), a condition identified for further study in the DSM-5 [American Psychiatric Association (APA) (2)]. Because PGD is formally included in the ICD-11 [World Health Organization (WHO) (4)] and is related to PCBD (3, 4), in the current study the term PGD will be used.

Violent circumstances of the death constitute an important risk factor for developing psychopathology after loss (5). A well-known yet understudied example of violent death is homicide, which is defined by the WHO (4) as a fatal injury inflicted by another person with the intention to injure or kill. The term “homicide” covers the offences of murder and manslaughter (including corporate manslaughter) and infanticide (6). Every homicide is estimated to affect the lives of approximately 7 to 10 close relatives (7, 8), as well as those of numerous friends, neighbors, and co-workers. Those affected are often referred to as “homicide survivors” (9). Homicide survivors are typically underrepresented in media coverage, research reports, and government funds, that tend to focus on perpetrators (7, 10).

Research on the mental health of homicide survivors is relatively scarce, often involving homogenous and small samples (11–13). Studies suggest that homicide survivors are at elevated risk for developing PGD and at nearly doubled risk for developing PTSD, compared to other victims of violence and compared to individuals who were bereaved due to other causes (5, 14). Additionally, homicide survivors are prone to developing depression and anxiety symptoms, as well as feelings of guilt and responsibility. A study of over 400 families of homicide survivors showed that all participants reported that their mental health was somehow affected, with 25% of the survivors no longer working, 20% struggling with alcohol addiction, and 44% experiencing marital problems resulting in divorce or separation (10). Other studies reported problems such as eating disorders, insomnia (14, 15), sleep disorders, and suicide ideation (16, 17).

The high prevalence of mental health complaints in homicide survivors may be due to a number of complexities that are specific for bereavement following a homicide. Firstly, the

shocking circumstances of the loss may induce intrusive images (18). Secondly, a homicide might shatter assumptions people have about themselves, others and the world around them, giving rise to feelings of injustice, unfairness, guilt, anger, anxiety, and revenge (14, 19). This may be especially true for those who lost someone to whom the attachment was especially strong, such as a child or spouse (5). Thirdly, in the immediate aftermath of a homicide, additional stressors associated with police investigations, trial procedures and media coverage may fuel distress (20). Although feelings of anger and revenge might not diminish after the conviction of the perpetrator (21), an ongoing trial, unsentenced trials, and dissatisfaction about the criminal justice are associated with higher PTSD scores and a complicated grieving process (19, 21, 22). Fourthly, survivors’ relationships with friends, family, and the community can become strained, as they might face stigmatization, social withdrawal, and isolation, thereby alienating them from society (10, 11, 22). In the United Kingdom (UK), Victims’ Commissioner Casey was appointed by the Ministry of Justice (MoJ) to promote the interests of victims and witnesses of crimes. In her review on the needs of families bereaved by homicide, she found that that separation, forced relocation, job loss, and financial difficulties commonly occurred (10).

Although the emotional consequences of homicidal loss evidently merit clinical attention, options for preventive and curative treatments are scarce. In existing literature on the treatment of traumatic loss, grief- and trauma-focused interventions are described, as well as interventions integrating working mechanisms of both (23, 24). For the treatment of PTSD, trauma-focused cognitive behavioral therapy (CBT) and Eye Movement Desensitization and Reprocessing (EMDR) are implemented in treatment guidelines (25). Furthermore, social work and family-focused therapy are recommended (19). Due to the aforementioned complexities surrounding a homicide, homicide survivors constitute a special group warranting a standardized treatment tailored to their specific needs. However, few well-researched options for treatment exist. One study found EMDR and CBT to be promising treatments for homicidally bereaved individuals (25), but their sample was characterized by a large treatment delay. It is therefore of importance to fortify the evidence base of existing treatments and to investigate their effectiveness over different samples.

In the UK, the attention toward the needs of homicide survivors increased strongly after publication of Casey’s report (12). It was concluded that homicide survivors constitute a vulnerable group that did not receive the support they needed. In 2010, ASSIST Trauma Care, a specialist Not-for-Profit

Organization offering specialized therapeutic services to those affected by psychological trauma, was contracted through the government to deliver this support.

ASSIST Homicide Bereavement Therapeutic Service—A Model of Care

Upon request by the homicide survivor or recommendation by Victim Support or other homicide support agencies that provide practical, judicial, and emotional support to homicide survivors, homicide survivors can be referred to ASSIST Trauma Care. Upon first contact, ASSIST Trauma Care provides information about their services and performs a first diagnostic assessment. Then, based on this assessment and the client's needs, clients are offered (a) so termed "early intervention", consisting of one to five face-to-face sessions oriented on psycho-education and coping strategies only, or b) face-to-face traumatic-grief focused CBT (TG-CBT). The current study focuses on the TG-CBT only. The TG-CBT consists of approximately 15 sessions in which psycho-education, exposure on the traumatic loss, writing exercises, giving meaning, and activation are covered. Although the TG-CBT is tailored to the client's needs, it follows a systematic approach and includes the same therapeutic ingredients for all patients. These include an initial 15 sessions that can be extended depending on individual circumstances, the duration of legal and court proceedings and higher complexity of the case; implementation of evidence-based trauma-focused CBT [National Collaborating Centre for Mental Health (NICE) (26)] and grief-focused CBT (26–28); provision of an outreach model of care with the sessions taking place at the office of ASSIST or at the homes of the clients; individual sessions with possibilities of involvement of supporting others; deployment of qualified and experienced therapists receiving regular training and supervision (from the Oxford Cognitive Therapy Centre); and close collaboration with caseworkers of Victim Support to provide practical and judicial support throughout the process.

Purpose of the Present Study

The first aim of the current retrospective study was to offer a preliminary evaluation of the potential effects of a traumatic grief-focused CBT in terms of reductions in symptoms of posttraumatic stress (PTS), prolonged grief (PG), depression and anxiety, and general functional impairment, based on data of over 900 people. We also investigated whether age, gender, being a direct witness to the homicide, closeness to the victim, recentness of the loss, status of the funeral and verdict, duration of therapy, and number of missed sessions were associated with changes in symptoms following treatment. The second aim was to examine whether baseline symptom severity was related to the aforementioned characteristics.

Concerning our first aim, we hypothesized that PTS, PG, depression, and anxiety symptom severity and general functional impairment would be significantly lower at the end of treatment compared to the start of treatment. We tentatively expected that higher symptom severity at the start of treatment would be associated with larger symptom reduction during treatment. We did not expect to find any effects for gender, age and recentness

of the loss on symptom change during treatment (26). Because of the exposure-oriented nature of the therapy, we expected clients who were direct witnesses to the homicide to show a larger reduction in PTS, PG, and anxiety symptoms than those who were not. Being a parent or partner of the victim was expected to be unrelated to the magnitude of symptom reductions (29). We expected the funeral being held or verdict being spoken during therapy or not at all rather than before therapy would result in smaller reduction of PTS and PG symptoms during treatment; and that clients with a so termed "history of mental health problems", defined as previous mental health treatment and/or a history of abuse (30), would benefit less from therapy as demonstrated by a smaller reduction of all symptoms. Finally, we expected no association for therapy duration or number of missed sessions with change in symptoms during treatment.

For our second aim, we hypothesized that female gender, younger age, being a direct witness to the homicide, having a closer relation to the victim (being a parent or partner to the victim) and the funeral or trial not yet have taken place would negatively influence symptom severity at the start of treatment (21, 31). Since clinical PTSD and PGD levels following violent loss have been shown to remain relatively stable over time without treatment, we expected that baseline symptom severity would not be associated with the time that had passed since the loss occurred (13, 30). We expected that clients with higher baseline symptoms would require more TF-CBT sessions but also miss more TF-CBT sessions.

As suggested by previous research, many factors may influence the expression of symptoms and the effects of treatment following bereavement by homicide (7, 11, 32). To the knowledge of the authors, the current study is unique in that it evaluates a systematic treatment approach specifically tailored to the needs of homicide survivors, in terms of changes in various outcomes (including symptoms of PG, PTS, depression, and anxiety and general functioning) in a sample that is large enough to explore potential moderators of treatment effects (i.e., differences in treatment effects for different subgroups). The present study adds to existing research in a number of ways. First, this study contributes to the overall understanding of the manifestation and prevalence of a variety of mental health problems following a homicide, building on previous inconclusive research in which various and smaller samples were used. Secondly, by describing the effects of a traumatic-grief focused outreaching model of care, it can inform the development of guidelines for psychosocial care tailored to the needs of homicide survivors. Finally, the current study explores whether the possible effects of the intervention differ as a function of several sociodemographic, homicide-related, and therapy-related characteristics, which may further add to the development of such guidelines.

METHOD

Study Design

The present study was designed as a retrospective, naturalistic pretest-posttest study among homicide survivors who received

and completed traumatic grief focused cognitive behavioral treatment through the services of ASSIST Trauma Care.

Participants

Between July 2010 and March 2017, 2,042 persons were referred to ASSIST following bereavement by homicide. After referral and initial contact, 607 persons (29.7%) decided not to engage. Reasons for this varied from time issues to not wanting to live through the traumatic experience, to already being engaged in a therapy elsewhere (33). The so termed “Early intervention” was offered to 330 persons (20.9%). The remaining 1,105 persons (54.1%) engaged in the full course of TG-focused therapy (CB-CBT) evaluated in the current study. For the purpose of the study, 146 children below the age of 18 years (13.2%) were excluded from analyses, leaving 941 adults. Of these, 12 (1.3%) did not complete any of the questionnaires at baseline nor at the end of therapy and were therefore also excluded from analyses. The final sample used in the current study consists of 929 adults, all of whom completed therapy by March 2017 and completed at least one baseline measure.

Ethical Standards

The current collaborative study with ASSIST Trauma Care was approved by the Ethics Committee of the Faculty of Social and Behavioral Sciences of Utrecht University and conducted in accordance with ethical guidelines from the UK for service evaluation. Written consent was obtained from all participants at the start of treatment. All information from the clients’ files was anonymized and entered into a dataset that was made accessible only to the researchers and stored on a secured server.

Procedure

Questionnaires to measure symptoms of PTSD, PGD, depression, and anxiety and functional impairment were administered at intake and at the end of treatment. Information about demographics, the homicide, judicial procedures and whether or not the client had a history of mental health problems was documented in the client’s files. In 2010, only symptoms of PTSD were measured. Later, the additional symptoms were added to the test battery to provide a more complete picture of the effects of the service.

Measures

Predictors

Sociodemographic variables included age and gender. Homicide-related information included relationship to the deceased, which was originally categorized into 22 categories. To simplify the analyses, we dichotomized this variable into having lost a partner or child rather than someone other than a partner or child (0 = No, 1 = Yes). Furthermore, dichotomized variables indicated whether or not the client witnessed the homicide (0 = No, 1 = Yes); if, as compared to the funeral taking place before therapy, the funeral took place during therapy (0 = No, the funeral took place before therapy, 1 = Yes, the funeral took place during therapy) or had not (yet) taken place (0 = No, the funeral took place before therapy, 1 = Yes, the funeral has not yet taken place); if, as compared to a verdict being spoken before therapy, a

verdict was spoken during therapy (0 = No, a verdict was spoken before therapy, 1 = Yes, a verdict was spoken during therapy), or had not (yet) been spoken (0 = No, the verdict was spoken before therapy, 1 = Yes, the verdict has not yet been spoken); and whether or not the loss had occurred less than six months ago (0 = No, 1 = Yes). For each client, clinicians indicated whether or not they had a so-termed “history of mental health problems” referring to previous mental health treatment and/or a history of abuse (0 = No, 1 = Yes). Lastly, based on the client files, we gathered information on therapy-related variables including duration of therapy in hours and number of missed sessions.

Impact of Event Scale (IES)

Symptoms of posttraumatic stress were measured by the IES (34). This self-report questionnaire includes 15 items rated on 4-point scales ranging from 0 (not at all), 1 (rarely experienced), 3 (sometimes experienced), to 5 (often experienced), the summation of which provides an index of traumatic stress. A score of 26 was used as a cut-off for clinically relevant scores (23, 34). The scale has sound psychometric properties (34). In the present study, reliability was acceptable for the total scale ($\alpha = .78$).

Inventory of Complicated Grief (ICG)

PG symptoms were measured by the 19-items self-report ICG. (35). Items are rated on 5-point scales ranging from 0 (never) to 4 (always). A cut-off score of 26 was used to indicate a probable PGD (35). The ICG has shown high internal consistency, test-retest reliability, and concurrent validity (35). In the current study, Cronbach’s alpha was good ($\alpha = .86$).

Patient Health Questionnaire (PHQ-9)

Self-reported symptoms of depression were measured using the 9-item PHQ-9 (36). Items are rated on 4-point scales ranging from 0 (not at all) to 3 (nearly every day) with higher scores indicating more severe depression. A score of 10 or higher indicates a moderate level of depression (37). The PHQ-9 is a reliable and valid measure (36). In the current study, Cronbach’s alpha was good ($\alpha = .85$).

Generalized Anxiety Disorder Scale (GAD-7)

The GAD-7 (38) is a short self-report questionnaire that measures symptoms of anxiety. The questionnaire includes seven items. Each item can be scored on 4-point scales ranging from 0 (not at all) to 3 (nearly every day). A score of 10 or higher indicates a moderate level of anxiety (38). Evidence supports the reliability and validity of the GAD-7 (39). In the current study, Cronbach’s alpha was good ($\alpha = .84$).

Work and Social Adjustment Scale (WSAS)

With the WSAS (40), the degree of impaired functioning attributable to an identified disorder can be measured. The self-report scale contains five items referring to several facets of life. Items are scored on scales ranging from 0 (not at all) to 8 (very severely), indicating how much a person’s problems affect their ability to carry out certain activities. The WSAS has good psychometric properties (40). Cronbach’s alpha in this sample was good ($\alpha = .81$).

Data Analyses

Descriptive analyses were performed and assumptions were checked using SPSS [version 25.0; (63)]. Frequencies, means and bivariate and polychoric correlations were calculated for all variables, the latter one using Mplus version 7 (41). Dropout analyses were performed by comparing clients with posttreatment scores with clients with no posttreatment scores using one-way analyses of variance for continuous variables and chi-square tests for categorical variables. We used Latent Growth Modeling, a statistical technique used in structural equation modeling (SEM), to analyze our repeated measures data and to classify individuals into distinct groups based on individual response patterns. LGM was conducted with Amos [version 19.0; (42)]. Missing data were handled using full information maximum likelihood (FIML) estimation (43). With FIML, a likelihood function for each individual is estimated based on the variables that are present so that all the available data are used. Literature indicates that the bias in FIML parameter estimates is relatively unaffected by the amount of missing data (44). In SPSS, missing data were handled using pairwise deletion. At baseline, 6.9% of scores on the IES were missing, and 44.6%, 21.3%, 20.6%, and 53.2% of scores on the ICG, PHQ, GAD, and WSAS, respectively. At the posttreatment assessment, 13.5% of scores on the IES were missing, and 41.1%, 24.2%, 22.7%, and 54.7% of scores on the ICG, PHQ, GAD, and WSAS, respectively. The large amount of missing data is explained by the gradual addition of other questionnaires to the IES to measure a broader range of mental health complaints than just PTS symptoms. Due to this reason, the values are considered to be missing at random. Despite the variation in sample size on the outcome measures,

the effect size for these analyses are expected to exceed Cohen's convention for a small effect ($d = .20$) with a power of .80 (45).

An example of a latent growth model for PTS symptoms is displayed in **Figure 1**. In total, five latent growth models were specified, each comprising (1) baseline levels of PG, PTSD, depression, and anxiety and functional impairment as well as (2) change during treatment in these symptoms and functional impairment as the two latent factors. The pre- and posttreatment scores on these measures were specified as the indicators. For the baseline level factor, paths with regression weights fixed to one were drawn to pre- and posttreatment scores, while for the change during treatment factor, only a path was drawn to posttreatment scores with regression weight fixed to one. Residual error variances of the indicators were fixed at zero to enable the model to be estimated. The model was saturated, implying a perfect fit. This was deemed appropriate in light of our aims, as we sought to deploy a model that allowed us to explore the size, direction, and statistical significance of direct and indirect path coefficients of all possible effects. Hence, no model fit indices for the model are given. This model fits prior research using a comparable sample and design (46). For the calculation of Cohen's d (47), we used the mean and standard deviation of the change in symptoms during treatment and the correlation between pre- and posttreatment symptoms for all measures (48). To each of the five models, all independent predictors were specified to both factors. Standardized coefficients were calculated to be able to evaluate the relative strength of each independent variable in predicting the dependent variable. Squared multiple correlations were calculated to estimate the percentage of explained variance by the model.

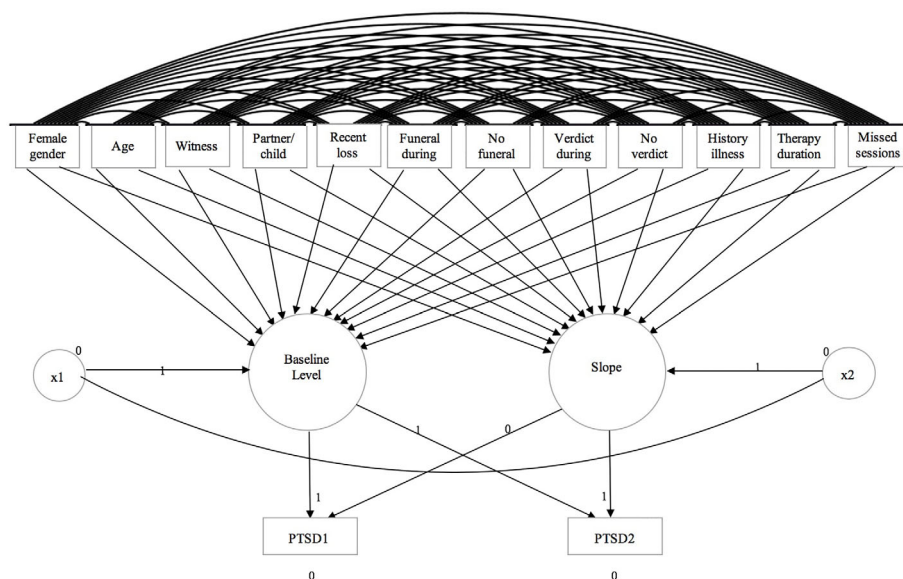


FIGURE 1 | Example of the latent growth model with PTS symptoms as outcome.

RESULTS

Descriptive Statistics

Descriptive data of sociodemographic, homicide-related, history of mental illness, and therapy-related variables of the complete sample consisting out of 929 clients are presented in **Table 1**.

Individuals for whom posttreatment scores were unavailable, for whatever reason (e.g., ended therapy preliminary, or finished therapy but were unable to fill out the end measures), were considered study-dropouts. Patients who completed posttreatment measures ($N = 859$) did not differ from study-dropouts ($N = 70$) in terms of gender, age, being a direct witness to the homicide, recentness of the loss, verdict status, history of mental health problems, or any of the therapy-related variables. However, study-dropouts were more likely than completers to have lost a child or spouse (40.0% vs. 28.6%, $p = .040$), having had a funeral during therapy instead of before therapy (13.0% vs. 5.1%, $p = .006$) and not having had a funeral yet instead of having had a funeral before therapy (4.3% vs. 1.2%, $p = .030$).

Aim 1: Treatment Effect Sizes and Predicting Change During Treatment

Table 2 shows symptom severity as measured with the IES, ICG, PHQ, and GAD and functional impairment as measured with

the WSAS; percentages of people scoring above the cutoff at the start and end of treatment; and the change in symptoms during treatment plus Cohen's d effect sizes. All symptoms and functional impairment reduced significantly and effect sizes were all high.

Table 3 shows the associations between sociodemographic, homicide-related, and therapy-related characteristics on the one hand and the change of symptoms and functioning during treatment on the other hand. The intercept and slope of all measures correlated significantly and negatively. This indicates that clients with higher baseline symptom severity showed a larger decline in symptoms during treatment than those with lower baseline symptom severity.

Age was not related to the change of any of the symptoms; having a history of mental health problems was associated with a smaller reduction of all symptoms; and the verdict being spoken during therapy compared to before therapy was associated with smaller reductions in PTS and PG symptoms.

Female gender was associated with larger improvement in general functioning but not with changes in symptoms. Being a direct witness of the homicide did not have an effect on change in symptoms of PTS or PG, but was, in fact, associated with greater reductions in depression and anxiety symptoms. No funeral being held yet was related to smaller reductions on PG

TABLE 1 | Sociodemographic characteristics, homicide characteristics, history of mental health problems and therapy characteristics at baseline.

Variable	<i>M</i>	<i>SD</i>	<i>n</i>	%	<i>nobs</i>	% <i>n</i>
Sociodemographic						
Female gender			700	75.4	929	100.0
Age	43.46	14.45			817	87.9
Homicide related						
Witness			178	19.2	928	99.9
Child/spouse			485	52.2	929	100.0
Recent loss (≤ 6 months)			470	50.6	915	98.5
Funeral before therapy			860	92.9	926	99.7
Funeral during therapy			53	5.7	926	99.8
No funeral			13	1.4	926	99.7
Verdict before therapy			304	32.7	810	87.2
Verdict during therapy			270	29.1	810	87.2
No verdict			236	25.4	810	87.2
History of mental health problems			367	39.5	927	99.8
Therapy related						
Therapy duration (hours, range: 3–59)	16.46	7.60			928	99.9
No. of missed sessions	1.74	2.26			927	99.9

nobs, number of observations; %*n*, percentage of total number of observations.

TABLE 2 | Mean symptom severity and functional impairment at start and end of treatment and change in symptoms and functional impairment during treatment.

	Start of treatment					End of treatment					Change during treatment		
	<i>M</i>	<i>SD</i>	%cut	<i>n</i>	% <i>n</i>	<i>M</i>	<i>SD</i>	%cut	<i>n</i>	% <i>n</i>	<i>M</i>	<i>SD</i>	<i>d</i>
IES	50.37	13.55	95.1	876	94.3	21.21	16.36	33.2	814	87.6	29.25***	17.62	1.94
ICG	43.45	13.93	89.0	521	56.1	23.64	15.38	40.2	554	56.6	19.57***	14.37	1.33
PHQ	15.87	6.83	58.6	741	79.8	6.50	6.57	12.9	713	76.7	9.39***	6.76	1.40
GAD	13.95	5.54	76.8	747	80.4	6.10	5.38	21.6	727	78.3	7.83***	6.12	1.43
WSAS	20.83	10.23	52.2	440	47.4	9.06	8.69	12.6	426	45.6	12.04***	8.98	1.26

IES, Impact of Events Scale; ICG, Inventory of Complicated Grief; PHQ, Patient Health Questionnaire; GAD, General Anxiety Disorder; WSAS, Work and Social Adjustment Scale; %cut, percentage of scores above cut-off score; %*n*, percentage of total number of observations. Baseline and change symptoms based on FIML and end symptoms based on available data.

*** $p < .001$.

TABLE 3 | Associations of demographic and homicide characteristics, history of mental health problems, and therapy characteristics with symptoms of PTSD, PGD, general anxiety, depression, and functional impairment at baseline and change during treatment.

Variable	IES		ICG		PHQ		GAD		WSAS	
	Baseline <i>B</i>	Slope <i>B</i>	Baseline <i>B</i>	Slope <i>B</i>	Baseline <i>B</i>	Slope <i>B</i>	Baseline <i>B</i>	Slope <i>B</i>	Baseline <i>B</i>	Slope <i>B</i>
Sociodemographics										
Female gender	0.17***	−0.06	0.17***	−0.04	0.18***	−0.05	0.18***	−0.06	0.16***	−0.10*
Age	−0.11**	0.07	−0.11*	0.09	−0.01	0.02	−0.08*	0.07	0.01	−0.05
Homicide related										
Witness	0.05	−0.03	0.01	−0.04	0.06	−0.09*	0.04	−0.09*	0.00	−0.01
Child or spouse	0.07	0.04	0.16***	0.05	0.09*	−0.01	−0.02	0.05	0.15**	−0.01
Recent loss (≤ 6 months)	0.11**	−0.06	0.09	−0.13*	0.05	−0.03	0.04	−0.05	0.04	−0.05
Funeral during therapy	−0.07*	0.04	−0.07	0.04	−0.03	0.01	−0.05	0.01	−0.01	0.02
No funeral	0.01	0.02	−0.13***	0.11*	−0.02	0.04	−0.05	0.03	−0.02	0.08
Verdict during therapy	−0.03	0.10*	0.02	0.13*	−0.04	0.08	0.02	0.04	0.12*	−0.08
No verdict	−0.05	0.11*	0.03	0.07	−0.05	0.06	−0.04	0.08	0.03	0.03
History of mental health problems	0.09**	0.22***	0.17***	0.16***	0.22***	0.15***	0.16***	0.13***	0.09	0.26***
Therapy related										
Therapy duration (hours)	0.10**	0.02	0.12**	0.04	0.11**	0.04	0.09*	0.08*	0.15**	−0.05
No. of missed sessions	0.12***	−0.04	0.08	−0.05	0.11**	−0.08*	0.12***	−0.10*	0.14**	−0.07
Baseline scores	−0.54***		−0.49***		−0.62***		−0.63***		−0.66***	
R ²	0.12**	0.08	0.18	0.08	0.17	0.04	0.14	0.06	0.16	0.08

B, Standardized regression coefficient (beta weight); IES, Impact of Events Scale; ICG, Inventory of Complicated Grief; PHQ, Patient Health Questionnaire; GAD, General Anxiety Disorder; WSAS, Work and Social Adjustment Scale; R², proportion of variance by the model. **p* < .05. ***p* < .01. ****p* < .001.

symptoms but was not associated with changes in PTS symptoms. No verdict being spoken was associated with smaller reductions on PTS symptoms but was unrelated with changes in PG symptoms. Recentness of the loss was associated with larger symptom reductions.

The funeral being held during therapy was unrelated to change in symptoms and functioning. In addition, recent loss was associated with larger PG symptom reduction and was not associated with changes of any other symptoms during treatment.

We found a larger decrease in anxiety symptoms for clients who had a shorter therapy duration. Clients who benefitted more from therapy in terms of symptoms of anxiety and depression tended to miss more sessions.

Aim 2: Predicting Symptoms at Baseline

Data on the extent to which symptoms of PTS, PG, depression, and anxiety and functional impairment varied as a function of the sociodemographic, homicide-related, and therapy-related variables are summarized in **Table 3**.

Female gender was associated with higher baseline symptom severity on all measures. Younger age was associated with higher baseline symptom severity except for depression symptoms and functional impairment; and having a history of mental health problems was associated with higher baseline symptom severity but not with higher functional impairment. Having lost a child or spouse was associated with higher baseline PG and depression symptom severity and with higher functional impairment, but not with other symptoms. Recent loss was associated only with higher baseline PTS symptom severity.

Being a direct witness to the homicide was not related to higher baseline symptom severity; the funeral being held before the start of therapy compared to during therapy was associated with higher baseline PTS symptom severity while the funeral being held before therapy compared to the funeral not being held

at all was associated with higher PG symptom severity; and the timing of the verdict showed no association with baseline symptom severity.

Clients with higher baseline symptom severity tended to have a longer therapy duration and miss more sessions.

Table 3 shows the relative strength of the association between the sociodemographic, homicide-related, history of mental illness, and therapy-related predictors; and baseline symptom severity and symptom reduction during treatment, as indicated by their standardized coefficients and their significance; and the proportion of variance explained by all the predictors on baseline symptom severity and symptom reduction during treatment.

DISCUSSION

The current study introduced and evaluated the effects of an outreach model of care following bereavement through homicide and examined whether baseline symptom severity and functional impairment and their changes during treatment varied as a function of sociodemographic- and homicide-related characteristics, having a history of mental health problems and treatment-related characteristics. In total, 929 treatment receiving homicide survivors were included. Prevalence of severe symptoms across disorders and general functional impairment at baseline was high, adding to the body of literature describing the broad range of detrimental effects of homicide on the mental health of homicide survivors [e.g., (13)]. Altogether, the decrease in symptoms across disorders and functional impairment following treatment suggests the potential effectiveness of the treatment. For change in symptom severity and functional impairment during treatment, having a history of mental illness was the only consistent predictor across all outcomes. Being a direct witness

was predictive of change in depression and anxiety symptoms and the status of the verdict was predictive of change in PG and PTS symptoms.

Having a history of mental illness, being younger of age and female and having lost either a child or spouse consistently predicted higher symptom severity and functional impairment at baseline.

The first aim of the study was to introduce a traumatic grief-focused outreaching model of care and to provide a preliminary evaluation of its potential effectiveness in terms of reduction on symptoms of PTS, PG, depression, and anxiety and functional impairment. As shown by the LGM, scores on all measures decreased significantly and with a large effect size, thereby confirming our first hypothesis. This cautiously implies that the current model of care as employed by ASSIST Trauma Care might be effective in reducing a broad range of mental health complaints in treatment receiving homicide survivors, while overcoming the geographic, financial or waiting-list barriers for care as reported by the Victims Commissioner (10). Consequently, that model of care could serve as a base to develop international guidelines for the treatment of homicide survivors. The current model includes several specific components that merit attention, namely, close collaboration between primary care (e.g., Victim Support) and specialized care (e.g., organizations offering TGF-CBT), access to timely assessment, access to TGF-CBT that is free of charge, availability of care across the whole country, and ongoing training and supervision of caretakers offering the TGF-CBT, specific to the complexities of the context and aftermath following a homicide.

Furthermore, we examined whether change in symptom severity and baseline symptoms varied as a function of sociodemographic variables, characteristics of the homicide, and having a history of mental illness. Importantly, the variable “history of mental illness” was most strongly and consistently associated with change in symptoms and baseline symptoms. Given the large sample size of the current study, this provides strong indications that previous experiences such as a history of mental health problems or abuse might be an important determinant for poorer response to psychotherapeutic care and the development of a variety of complaints after bereavement following a homicide.

Corresponding to the findings of Boelen, van Denderen, and de Keijser (5) who found that feelings of anger and revenge did not diminish according to the progress of the judicial process, baseline symptom severity did not differ as a function of verdict status in the current sample. However, additional or secondary stressors following traumatic loss may negatively influence the course of complaints and obstruct recovery. Our findings of smaller reductions of PGD and PTS symptoms implicate that a slow course of justice or the absence of a verdict might negatively impact treatment response. This underlines the impact of the judicial process on the therapeutic outcome and the importance of close collaboration between case workers and the therapists to provide additional information and support regarding the course of justice.

Partly in par with our expectations, except for PTS symptoms, baseline symptom severity and functional impairment were unrelated to recentness of the loss. This strengthens previous findings that distress following homicide does not naturally decrease over time (13, 30). We also found that homicide survivors whose loss occurred recently displayed a greater decline in PG symptoms, suggesting that offering intervention in the early months following loss may actually be beneficial for a subgroup experiencing severe distress (49, 50). In fact, these findings support the public health model of bereavement support that differentiates between low, moderate, and high risk and need for support following bereavement (51). Considering bereavement by homicide as a high-risk category, immediate referral to specialist care is desirable.

Homicide survivors witnessing the homicide did not display higher symptom severity at baseline compared to those who were not a direct witness. This accords with the findings of Hertz, Prothrow-Stith, and Chery (9), based on a literature search on homicide victims, showing that witnesses and non-witnesses display similar symptom severity; and the notion that intrusive images similar to those of direct witnesses may be evoked by reconstructive fantasy based on police and witness reports (18, 52, 53). Lastly, closer kinship to the deceased did not have a significant relation with baseline PTS symptom severity, suggesting that PTS symptoms are likely to develop following a homicide regardless of closeness to the victim. This may be explained by the violent and horrific nature of the event itself. Corroborating the finding of a high risk bereavement profile following such a loss (51), having lost a partner or spouse was actually strongly associated with higher baseline PG symptom severity in the current study. However, we did not measure quality of the relationship with the deceased [cf. (54)] and it would be of interest to do so in future research.

Finally, as expected, clients with more severe baseline symptoms eventually underwent more sessions, which is in accordance with prior findings that patients with more severe and complex symptoms receive treatments that last longer (55). Somewhat unexpectedly, however, clients who benefitted more from therapy in terms of declines in anxiety and depression also tended to miss more sessions. One possible explanation for this finding might be that when clients feel they benefit from therapy, they are less strongly motivated for the treatment and more inclined to skip the last sessions. However, an alternative explanation may be that patients who had fewer sessions engaged in less exposure, allowing them to avoid confrontation with the acceptance of the loss [cf. (56)].

The findings of the current study may have several practical implications. One key message for “stakeholders” of homicidal bereavement (e.g., mental health care workers, policy makers, and police and court officials) is that homicide survivors are likely to develop a broad range of severe mental health complaints that can be successfully alleviated by offering timely TG-CBT treatment. Early assessment, extra sessions and application of additional interventions parallel to CBT (e.g., group therapy) may prove beneficial for individuals with a history of mental health problems. The occurrence of the funeral and verdict may

temporarily increase symptoms, pointing toward the possible necessity of additional sessions to ensure therapeutic holding during those events, as well as the importance of prompt handling of the judicial process. Since the status of the funeral was unrelated to therapy outcome, it might not be necessary to postpone an intervention to after the funeral. Lastly, therapists might consider that exposure therapy may be as beneficial for those who did not witness the crime as for those who did. Concluding, it may be of importance to coordinate treatment with other parties involved so that it can be tailored to the intertwinement between the complex external circumstances and the needs of the individual at that time.

Limitations and Strengths

Some limitations of the current study should be taken into consideration. First of all, because this study did not include a control group, we were unable to evaluate the effects of TGF-CBT relative to other treatment or no treatment and natural recovery. Secondly, the use of self-questionnaires prohibits any statements about the presence of disorders to be made and may lead to an overestimation of symptom severity (57). Prevalence rates in the current study were indeed higher than those in other studies (14, 21, 58, 59). Because this study focused on help receiving individuals, this might be attributable to a selection bias as people with low level of complaints were not included, thereby limiting the generalizability. In the same line of reasoning, since help seeking has been demonstrated to discriminate between those who benefit most and those who do not (60), the decline in scores may be positively biased. Conversely, the later addition of questionnaires other than the IES to the test battery may underestimate the prevalence of symptoms other than PTS symptoms. Additionally, as only two time points were used in the LGM, no statements could be made about the shape of growth of symptoms over time. Lastly, in aiming to describe the current service-delivery approach to inform international guidelines, we did not unravel which specific elements of the therapy were effective, such as cognitive restructuring, exposure, the outreaching nature of the therapy or the practical support as provided by Victim Support. Notwithstanding these considerations, the results of this study strengthen and quantify previous findings on grief interventions using smaller sample sizes and various specific subgroups that are difficult to generalize. This study is the first to address a multitude of mental health complaints after bereavement through homicide in such a large clinical sample while considering predictors related to the homicide, having a history of mental illness, sociodemographic information, and the therapy.

Implications for Future Research

Future studies should include a control group and follow-up measurements in order to determine the effectiveness and endurance of the current model of care on PTSD, PGD, depression, and anxiety disorders. Prospective, longitudinal studies are needed to examine the causal direction between the predictors used in this study and the development of symptoms of PTS, PG, depression, and anxiety following homicide,

preferably including more than two waves so individual patterns in changes of symptoms can be explored.

Concerning generalizability, it would be of interest to explore whether the current service-delivery approach as offered by ASSIST could be implemented in other countries and cultures with similar effects and to examine whether the current model could be beneficial for other populations facing bereavement and/or traumatic experiences. Lastly, it would be of great interest to explore whether clearer patterns of predictors will emerge when examining the co-occurrence of these symptoms or symptom profiles, using latent class and latent trajectory analyses. In conclusion, the service-delivery approach described in the current study appears to be effective in reducing mental health complaints following bereavement through homicide. Having a history of mental illness was a strong predictor for reaction to treatment. The results furthermore provide insight in the severe and lasting detrimental mental health outcomes that may arise following homicidal loss and provides evidence that timely, outreaching, specialized treatment can be beneficial to counteract these effects.

DATA AVAILABILITY STATEMENT

The datasets generated for this study will not be made publicly available because the data used in the current study contains information from a patient population.

ETHICS STATEMENT

The current collaborative study with ASSIST Trauma Care was approved by the Ethics Committee of the Faculty of Social and Behavioral Sciences of Utrecht University and conducted in accordance with ethical guidelines from the UK for service evaluation. Written consent was obtained from all participants at the start of treatment.

AUTHOR CONTRIBUTIONS

BG developed the intervention, and together with RW collected, managed, and entered the data. GS and SS designed the study, analyzed the data, and together with PB did the writing. All authors contributed to the article and approved the submitted version.

ACKNOWLEDGMENTS

The authors would like to thank the staff of ASSIST Trauma Care for the data collection, data-entering and valuable suggestions, and Dr. Niels van der Aa for his assistance with the design and statistical analyses.

REFERENCES

- Prigerson HG, Horowitz MJ, Jacobs SC, Parkes CM, Aslan M, Goodkin K, et al. Prolonged grief disorder: Psychometric validation of criteria proposed for DSM-V and ICD-11. *PLoS Med* (2009) 6(8). doi: 10.1371/journal.pmed.1000121
- American Psychiatric Association. *Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5)*. American Psychiatric Association: Washington, DC (2013).
- Boelen PA, Lenferink LIM, Nickerson A, Smid GE. Evaluation of the factor structure, prevalence, and validity of disturbed grief in DSM-5 and ICD-11. *J Affect Disord* (2018) 240:79–87. doi: 10.1016/j.jad.2018.07.041
- Boelen PA, Lenferink LIM, Smid GE. Further evaluation of the factor structure, prevalence, and concurrent validity of DSM-5 criteria for Persistent Complex Bereavement Disorder and ICD-11 criteria for Prolonged Grief Disorder. *Psychiatry Res* (2019) 273:206–10. doi: 10.1016/j.psychres.2019.01.006
- Boelen PA, van Denderen M, de Keijser J. Prolonged Grief, Posttraumatic Stress, Anger, and Revenge Phenomena Following Homicidal Loss: The Role of Negative Cognitions and Avoidance Behaviors. *Homicide Stud* (2015) 20:177–95. doi: 10.1177/1088767915580674
- Office for National Statistics. Homicide. (2017). <https://www.ons.gov.uk/peoplepopulationandcommunity/crimeandjustice/compendium/focusonviolentcrimeandsexualoffences/yearendingmarch2016/homicide>.
- Zinzow HM, Rheingold AA, Hawkins AO, Saunders BE, Kilpatrick DG. Losing a loved one to homicide: Prevalence and mental health correlates in a national sample of young adults. *J Trauma Stress* (2009) 22:20–7. doi: 10.1002/jts.20377
- Redmond LM. *Surviving: When someone you know was murdered*. Clearwater, FL: Psychological Consultations and Educational Services Ltd (1989).
- Hertz MF, Prothrow-Stith D, Chery C. Homicide survivors: Research and practice implications. *Am J Prev Med* (2005) 29:288–95. doi: 10.1016/j.amepre.2005.08.027
- Casey L. *Review into the Needs of Families Bereaved by Homicide*. London: Ministry of Justice (2011).
- Mastrocinque JM, Metzger JW, Madeira J, Lang K, Pruss H, Navratil PK, et al. I'm Still Left Here With the Pain: Exploring the Health Consequences of Homicide on Families and Friends. *Homicide Stud* (2015) 19:326–49. doi: 10.1177/1088767915537494
- Stern SB. *Immediate aftercare program for survivors and families of homicide*. Georgia: Kennesaw State University (2010). Available at: <http://www.worldbank.org/en/news/press-release/2016/04/08/aging-in-thailand—addressing-unmet-health-needs-of-the-elderly-poor>.
- van Denderen M, de Keijser J, Kleen M, Boelen PA. Psychopathology Among Homicidally Bereaved Individuals: A Systematic Review. *Trauma Violence Abus* (2015) 16:70–80. doi: 10.1177/1524838013515757
- Zinzow HM, Rheingold AA, Byczkiewicz M, Saunders BE, Kilpatrick DG. Examining posttraumatic stress symptoms in a national sample of homicide survivors: Prevalence and comparison to other violence victims. *J Trauma Stress* (2011) 24:743–6. doi: 10.1002/jts.20692
- Amick-McMullan A, Kilpatrick DG, Veronen LJ, Smith S. Family survivors of homicide victims: Theoretical perspectives and an exploratory study. *J Trauma Stress* (1989) 2:21–35. doi: 10.1007/BF00975764
- King K. It Hurts So Bad: Comparing Grieving Patterns of the Families of Murder Victims with Those of Families of Death Row Inmates. *Crim Justice Policy Rev* (2004) 15:193–211. doi: 10.1177/0887403404263625
- Kovandzic TV, Vieraitis LM, Yeisley MR. The Structural Covariates of Urban Homicide: Reassessing the Impact of Income Inequality and Poverty in the Post-Reagan Era. *Criminology* (1998) 36:569–600. doi: 10.1111/j.1745-9125.1998.tb01259.x
- Kristensen P, Weisæth L, Heir T. Bereavement and Mental Health after Sudden and Violent Losses: A Review. *Psychiatry Interpers Biol Process* (2012) 75:76–97. doi: 10.1521/psyc.2012.75.1.76
- Armour M. Violent death: Understanding the context of traumatic and stigmatized grief. *J Hum Behav Soc Environ* (2006) 14:53–90. doi: 10.1300/J137v14n04_04
- Zinzow HM, Rheingold AA, Byczkiewicz M, Saunders BE, Kilpatrick DG. Examining posttraumatic stress symptoms in a national sample of homicide survivors: Prevalence and comparison to other violence victims. *J Trauma Stress* (2011) 24:743–6. doi: 10.1002/jts.20692
- van Denderen M, de Keijser J, Huisman M, Boelen PA. Prevalence and correlates of self-rated posttraumatic stress disorder and complicated grief in a community-based sample of homicidally bereaved individuals. *J Interpers Violence* (2016) 31:207–27. doi: 10.1177/0886260514555368
- Amick-McMullan A, Kilpatrick DG, Resnick HS. Homicide as a Risk Factor for PTSD Among Surviving Family Members. *Behav Modif* (1991) 15:545–59. doi: 10.1177/01454455910154005
- van Denderen M, de Keijser J, Stewart R, Boelen PA. Treating complicated grief and posttraumatic stress in homicidally bereaved individuals: A randomized controlled trial. *Clin Psychol Psychother* (2018) 25:1–12. doi: 10.1002/cpp.2183
- Smid GE, Kleber RJ, de la Rie SM, Bos JBA, Gersons BPR, Boelen PA. Brief eclectic psychotherapy for traumatic grief (BEP-TG): Toward integrated treatment of symptoms related to traumatic loss. *Eur J Psychotraumatol* (2015) 6:1–11. doi: 10.3402/ejpt.v6.27324
- National Collaborating Centre for Mental Health. *NICE Clinical Guidelines, No. 26. 2, Post-traumatic stress disorder*. London: National Institute for Health and Clinical Excellence (2005). doi: 10.1016/j.amepre.2013.01.013.
- Alves-Costa F, Hamilton-Giachritsis C, Christie H, van Denderen M, Halligan S. Psychological Interventions for Individuals Bereaved by Homicide: A Systematic Review. *Trauma Violence Abus* (2019). doi: 10.1177/1524838019881716
- Boelen PA, van den Bout J, van den Hout MA. Negative cognitions and avoidance in emotional problems after bereavement: A prospective study. *Behav Res Ther* (2006) 44:1657–72. doi: 10.1016/j.brat.2005.12.006
- Elklit A, Kurdahl S. The psychological reactions after witnessing a killing in public in a Danish high school. *Eur J Psychotraumatol* (2013) 4:1–7. doi: 10.3402/ejpt.v4i0.19826
- World Health Organization. *International Classification of Diseases, 11th Revision*. Geneva: World Health Organization (2018).
- Djelantik AAAMJ, Smid GE, Kleber RJ, Boelen PA. Early indicators of problematic grief trajectories following bereavement. *Eur J Psychotraumatol* (2018) 8:1423825. doi: 10.1080/2008198.2018.1423825
- Burke LA, Neimeyer RA, McDevitt-Murphy ME. African American Homicide Bereavement: Aspects of Social Support That Predict Complicated Grief, PTSD, and Depression. *OMEGA - J Death Dying* (2010) 61:1–24. doi: 10.2190/OM.61.1.a
- Rheingold AA, Zinzow H, Hawkins A, Saunders BE, Kilpatrick DG. Prevalence and mental health outcomes of homicide survivors in a representative US sample of adolescents: Data from the 2005 National Survey of Adolescents. *J Child Psychol Psychiatry Allied Discip* (2012) 53:687–94. doi: 10.1111/j.1469-7610.2011.02491.x
- Mack J, Goodfellow B. *Homicide Bereavement Therapeutic Service Evaluation*. Rugby: ASSIST Trauma Care (2005).
- Horowitz M, Wilner N, Alvarez W. Impact of Event Scale: A Measure of Subjective Stress. *Psychosom Med* (1979) 41:209–18. doi: 10.1097/00006842-197905000-00004
- Prigerson HG, Maciejewski PK, Reynolds CF, Bierhals AJ, Newsom JT, Fasiczka A, et al. Inventory of complicated grief: A scale to measure maladaptive symptoms of loss. *Psychiatry Res* (1995) 59:65–79. doi: 10.1016/0165-1781(95)02757-2
- Kroenke K, Spitzer RL, Williams JBW. The PHQ-9: Validity of a brief depression severity measure. *J Gen Intern Med* (2001) 16:606–13. doi: 10.1046/j.1525-1497.2001.016009606.x
- Kroenke K, Spitzer RL. The PHQ-9: A new depression diagnostic and severity measure. *Psychiatr Ann* (2002) 32:509–15. doi: 10.3928/0048-5713-20020901-06
- Spitzer R, Kroenke K, Williams J, Lowe B. A Brief Measure for Assessing Generalized Anxiety Disorder. *Arch Intern Med* (2006) 166:1092–7. doi: 10.1001/archinte.166.10.1092
- Löwe B, Decker O, Müller S, Brähler E, Schellberg D, Herzog W, et al. Validation and Standardization of the Generalized Anxiety Disorder Screener (GAD - 7) in the General Population. *Med Care* (2008) 46:266–74. doi: 10.1097/MLR.0b013e318160d093
- Mundt JC, Marks IM, Shear MK, Greist JH. The Work and Social Adjustment Scale: A simple measure of impairment in functioning. *Br J Psychiatry* (2002) 180:461–4. doi: 10.1192/bjp.180.5.461

41. Muthén LK, Muthén BO. *Mplus User's Guide. Sixth Edit.* Los Angeles, CA: Muthén & Muthén (2011).
42. Arbuckle JL. *SPSS Amos 19.0 User's Guide.* Chicago, IL: Amos Development Corp (1999). Available at: <http://ir.nmu.org.ua/handle/123456789/125632>.
43. Preacher KJ, Wichman AL, MacCallum RC, Briggs NE. *Latent growth curve modeling.* Sage: California (2008).
44. Enders CK, Bandalos DL. The relative performance of full information maximum likelihood estimation for missing data in structural equation models. *Struct Equ Model* (2001) 8:430–57. doi: 10.1207/S15328007SEM0803_5
45. Zhang Z, Wang L. Statistical power analysis for growth curve models using SAS. *Behav Res Methods* (2009) 41:1083–94. doi: 10.3758/BRM.41.4.1083
46. Smid GE, van der Meer CAI, Olff M, Nijdam MJ. Predictors of Outcome and Residual Symptoms Following Trauma-Focused Psychotherapy in Police Officers With Posttraumatic Stress Disorder. *J Trauma Stress* (2018) 31:764–74. doi: 10.1002/jts.22328
47. Cohen J. *Statistical Power Analysis for the Behavioral Sciences. 2nd edition.* Hillsdale, NJ: Lawrence Erlbaum Associates (1988).
48. Rosner R, Kruse J, Hagl M. A Meta-Analysis of Interventions for Bereaved Children and Adolescents. *Death Stud* (2010) 34:99–136. doi: 10.1080/07481180903492422
49. Kersting A, Dölemeyer R, Steinig J, Walter F, Kroker K, Baust K, et al. Brief internet-based intervention reduces posttraumatic stress and prolonged grief in parents after the loss of a child during pregnancy: A randomized controlled trial. *Psychother Psychosom* (2013) 82:372–81. doi: 10.1159/000348713
50. Litz BT, Schorr Y, Delaney E, Au T, Papa A, Fox AB, et al. A Randomized Controlled Trial of an Internet-Based Therapist-Assisted Indicated Preventive Intervention for Prolonged Grief Disorder. *Behav Res Ther* (2014) 61:23–34. doi: 10.1038/jid.2014.371
51. Aoun SM, Breen LJ, Howling DA, Rumbold B, McNamara B, Hegney D. Who needs bereavement support? A population based survey of bereavement risk and support need. *PloS One* (2015) 10:1–14. doi: 10.1371/journal.pone.0121101
52. Boelen PA, Huntjens RJC. Intrusive images in grief: An exploratory study. *Clin Psychol Psychother* (2008) 15:217–26. doi: 10.1002/cpp.568
53. Rynearson K. Bereavement After Homicide: A Descriptive Study. *Am J Psychiatry* (1984) 141:1452–4. doi: 10.1176/ajp.141.11.1452
54. Spanier GB. Measuring Dyadic Adjustment: New Scales for Assessing the Quality of Marriage and Similar Dyads. *J Marriage Fam* (1976) 38:15–28. doi: 10.1016/j.copsyc.2014.11.003
55. Boerema AM, Cuijpers P, Beekman ATF, Hellenthal A, Voorrips L, van Straten A. Is duration of psychological treatment for depression related to return into treatment? *Soc Psychiatry Psychiatr Epidemiol* (2016) 51:1495–507. doi: 10.1007/s00127-016-1267-7
56. Stroebe M, Schut H. The dual process model of coping with bereavement: Rationale and description. *Death Stud* (1999) 23:197–224. doi: 10.1080/074811899201046
57. Engelhard IM, Arntz A, Van den Hout MA. Low specificity of symptoms on the post-traumatic stress disorder (PTSD) symptom scale: A comparison of individuals with PTSD, individuals with other anxiety disorders and individuals without psychopathology. *Br J Clin Psychol* (2007) 46:449–56. doi: 10.1348/014466507X206883
58. Kristensen P, Dyregrov K, Dyregrov A, Heir T. Media exposure and prolonged grief: A study of bereaved parents and siblings after the 2011 utøya island terror attack. *Psychol Trauma Theory Res Pract Policy* (2016) 8:661–7. doi: 10.1037/tra0000131
59. Lenferink LIM, de Keijser J, Smid GE, Djelantik AAAMJ, Boelen PA. Prolonged grief, depression, and posttraumatic stress in disaster-bereaved individuals: latent class analysis. *Eur J Psychotraumatol* (2017) 8:1298311. doi: 10.1080/2008198.2017.1298311
60. Brodbeck J, Berger T, Biesold N, Rockstroh F, Znoj HJ. Evaluation of a guided internet-based self-help intervention for older adults after spousal bereavement or separation/divorce: A randomised controlled trial. *J Affect Disord* (2019) 252:440–9. doi: 10.1016/j.jad.2019.04.008
61. Djelantik AAAMJ, Smid GE, Kleber RJ, Boelen PA. Symptoms of prolonged grief, post-traumatic stress, and depression after loss in a Dutch community sample: a latent class analysis. *Psychiatry Res* (2017) 247:276–81. doi: 10.1016/j.psychres.2016.11.023
62. Lenferink LIM, van Denderen M, de Keijser J, Wessel I, Boelen PA. Prolonged grief and post-traumatic stress among relatives of missing persons and homicidally bereaved individuals: a comparative study. *J Affect Disord* (2017) 209:1–2. doi: 10.1016/j.jad.2016.11.012
63. IBM Corp. *IBM SPSS Statistics for Windows, Version 25.0.* Armonk, NY: IBM Corp (2017).

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Copyright © 2020 Soydas, Smid, Goodfellow, Wilson and Boelen. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.



Traumatic Bereavements: Rebalancing the Relationship to the Deceased and the Death Story Using the Two-Track Model of Bereavement

Simon Shimshon Rubin^{1,2,3,4*}, Ruth Malkinson^{1,5} and Eliezer Witztum¹

¹ International Laboratory for the Study of Loss, Bereavement and Human Resilience, University of Haifa, Haifa, Israel, ² School of Psychological Sciences, University of Haifa, Haifa, Israel, ³ Zramim, Postgraduate Psychotherapy Program, University of Haifa, Haifa, Israel, ⁴ Department of Psychology, Max Stern Emek Jezreel College, Emek, Israel, ⁵ Mitra-Israel Center for REBT, Rehovot, Israel

OPEN ACCESS

Edited by:

Clare Killikelly,
University of Zurich, Switzerland

Reviewed by:

Katie Moraes de Almondes,
Federal University of Rio Grande do
Norte, Brazil
Kossigan Cyrille Kokou-Kpolou,
University of Picardie Jules Verne,
France

*Correspondence:

Simon Shimshon Rubin
Rubin@psy.haifa.ac.il

Specialty section:

This article was submitted to
Psychopathology,
a section of the journal
Frontiers in Psychiatry

Received: 02 March 2020

Accepted: 17 August 2020

Published: 15 September 2020

Citation:

Rubin SS, Malkinson R and Witztum E
(2020) Traumatic Bereavements:
Rebalancing the Relationship to the
Deceased and the Death Story Using
the Two-Track Model of Bereavement.
Front. Psychiatry 11:537596.
doi: 10.3389/fpsy.2020.537596

Bereavements that occur under external traumatic circumstances increase the risk for dysfunction, trauma symptomatology, as well as disordered and prolonged grief. While the majority of individuals who have experienced traumatic bereavements do not meet formal criteria for posttraumatic stress disorder (PTSD), persistent complex bereavement disorder (PCBD), or prolonged grief disorder (PGD), the degree of distress and dysfunction for these bereaved can be quite significant. The assessment and intervention paradigms in use with traumatic bereavements often prioritize the trauma and bypass the centrality of the interpersonal loss. By using a bifocal approach in conceptualizing bereavement, the Two-Track Model of Bereavement (TTMB) rebalances the approach to the class of traumatic bereavements. Track I examines biopsychosocial functioning and symptoms of trauma, and track II focuses on the nature of the ongoing relationship with the deceased and the death story that may also have elements of traumatic response. The model and its application serve to identify both adaptive and maladaptive responses to loss along both axes to optimally focus interventions where needed. The story of the death, the psychological relationship with the deceased, and the presence of biopsychosocial difficulties each have a part to play in assessment and intervention. A case study of assessment and intervention following traumatic bereavement due to suicide illustrates how attention to each of these factors in the TTMB can facilitate change. Ultimately, the relational bond with the deceased is a major vector in grief and mourning. Assessment and intervention with traumatic bereavements require attention to dysfunction and symptoms of trauma as well as to the death story and the state of the relationship to the deceased.

Keywords: bereavement, prolonged grief disorder, traumatic bereavement, Two-Track Model of Bereavement, posttraumatic stress disorder, continuing bonds, suicide, psychotherapy

INTRODUCTION

Bereavement following the death of a loved one is universal. How one grieves and mourns that loss, and how a broad array of variables influence grief and its outcome have received increasing attention and specification. Since the beginning of the 20th century, the field of thanatology has undergone tectonic changes in the way bereavement is understood. One revolution in bereavement is the break with Freud's

(1) view of mourning as a process leading to the minimization of the emotional connection to the deceased. In place of a view that breaking the bonds (decathexis) with the deceased is adaptive, grieving is recognized as the reworking and maintenance of the attachment bond rather than its severance and has been designated the *continuing bonds* paradigm (2–4). The psychological processes in grief and mourning are related to the life-change reorganization that requires adaptation to the absence of the deceased's physical presence (5–7). As a life stressor of major proportions, the death of a significant attachment figure puts the bereaved at risk of developing a broad variety of difficulties and dysfunctions including, but not limited to, depressive disorders. At the same time, the majority of bereaved respond to loss without sustained difficulties (8).

The estimated prevalence of bereaved at risk to develop variations of prolonged grief disorder (PGD) and complications of grief generally range between 7–15% across all categories with traumatic circumstances of loss yielding significantly higher estimates (9–13).

TRAUMATIC BEREAVEMENTS

The term *traumatic bereavements* originated in response to clinical reality where bereavements occurring under traumatic circumstances resulted in the familiar mixture of symptom pattern of PTSD together with acute grief (14). Traumatic bereavements increase the risk for dysfunction and symptomatic difficulties and complicated grief (7, 15–18). Traumatic bereavement is a term that stresses the interface of traumatic circumstances with bereavement (14, 19, 20). The potential significance of the interplay between the traumatic and bereavement elements in a variety of loss has received significant attention in the literature over the years, but consensus on diagnosis, research, and intervention remain elusive (7, 20–22).

In a meta-analysis studying the efficacy of grief interventions for a variety of bereavements, both traumatic loss and child loss demonstrated benefits from intervention while “uncomplicated losses” did not show benefit (23). This linkage would support the understanding that both trauma and the death of a child pose extraordinary challenges for post-death grieving and adaptation. Child loss has itself been described as a traumatic loss (24, 25). In our own studies of heightened grief scores with the two-track bereavement questionnaire for complicated grief years after the loss, we noted that the elevated scores that characterized only 5% of adults bereaved of their parents, applied to 10% of the spousal bereaved and to fully 25% of those who had lost children (26). These results support a position that the loss of a child is a loss of traumatic proportions in comparison with other family losses.

The interplay of trauma and bereavement is complex. In research examining adults who were not physically present but were bereaved in the 9-11 attacks, approximately 3 years later 43% received a classification of complicated grief with PTSD among the major comorbid conditions (27). A recent study looked at the presence of symptoms of prolonged grief disorder, posttraumatic stress disorder, and depression for 458 persons presenting for treatment at a Dutch clinic specializing in

trauma (28). In the bereaved group, 45% reported a history of violent loss. For their bereaved sample, the authors report clinically relevant PGD scores for 28%, PTSD for 78% and depression for 28%. This overlap between symptoms of trauma and of grief in a clinical setting focusing on trauma is a repeated and robust finding. It underscores the overlap between the two classes of response.

In the framework of the revisions and discussions preceding the DSM -5 (29–31), suggestions to include complicated grief and prolonged grief were rejected. Instead, a new term was added and specified in the category of conditions for further study—persistent complex bereavement disorder (32). This term was put forth to replace terms in use such as pathological grief and complicated grief and was structured with proposed criteria ordered in the current DSM format. It is particularly significant to note that following specified criteria, the diagnostician is requested to address whether the circumstances of death are traumatic:

Specify if:

“With traumatic bereavement: Bereavement due to homicide or suicide with persistent distressing preoccupations regarding the traumatic nature of the death (often in response to loss reminders), including the deceased’s last moments, degree of suffering and mutilating injury, or the malicious or intentional nature of the death”. [(32), pp.789–792]

The parallel diagnostic formulation for complications of grief in the ICD 11 is classified for the first time under - 6B42 prolonged grief disorder (33). This emphasizes the criteria extended time and dysfunction such that “The disturbance causes significant impairment in personal, family, social, educational, occupational, or other important areas of functioning” (33, 34). In contrast to the DSM-5, the ICD-11 excludes the circumstances of traumatic bereavement from its definition thus ignoring significant component of traumatic bereavement (33). **Table 1** presents the diagnostic criteria for PCBD and PGD. It is important to note that proposed changes for a revised DSM-5 currently under consideration include creating a formal diagnosis of PGD in the DSM section II (35). We agree with those advocating for the inclusion of PGD in a DSM-5 revision in the chapter on trauma and stressor-related disorders rather than in the chapter on depressive disorders (36). Irrespective of any decision on where to place the bereavement diagnosis, we believe it valuable for any modification of the DSM regarding disordered bereavement to retain the specification “with traumatic bereavement” as an important qualifier. The significance of the intersection of trauma and bereavement are consistent findings in the clinical and research literatures (e.g., 35).

It is premature to determine the extent to which the changes in nomenclature of the DSM and the ICD will impact the ways in which practitioners and researchers approach trauma and bereavement (37, 38). In our experience in Israel and internationally, the emphasis on trauma and PTSD in the context of bereavement has the paradoxical effect of obscuring the centrality of the interpersonal relationship bond in these circumstances (39,

TABLE 1 | A comparison of the PCBD diagnosis in DSM-5 (2013) and the PGD diagnosis of ICD-11 (2019).

Category	DSM-5 persistent complex bereavement-related disorder (PCBD— conditions for further study)	ICD PGD (2019)
Event	Death of a close other	Death of a close other
Time frame and degree	12 months; “on most days to a clinically significant degree”	6 months and clearly exceeds norms for culture/context
Response focus	1 of yearning/longing; sorrow/pain in response to death; preoccupation with deceased or circumstances of death.	Longing for the deceased or persistent preoccupation with the deceased
Symptoms	6 of the following: 1) marked difficulty accepting the death; 2) disbelief or emotional numbness over the loss; 3) difficulty with positive reminiscing about deceased; 4) bitterness or anger related to the loss; 5) maladaptive appraisals about oneself in relation to deceased or death; 6) excessive avoidance of reminders of the loss; 7) desire to die to be with deceased; 8) difficulty trusting other people since the death; 9) feeling alone or detached from other people since the death; 10) feeling that life is meaningless or empty without deceased or belief that one cannot function without deceased; 11) confusion about one's role or diminished identity; 12) difficulty pursue interests or plan for future since loss	Persistent and pervasive grief response characterized by longing for the deceased or persistent preoccupation with the deceased accompanied by intense emotional pain (e.g., sadness, guilt, anger, denial, blame, difficulty accepting the death, feeling one has lost a part of one's self, an inability to experience positive mood, emotional numbness, difficulty in engaging with social, or other activities).
Traumatic death circumstance	Specify if: With traumatic bereavement with persistent distressing preoccupations regarding the traumatic nature of death	No specifier
Degree of impairment	Clinically significant in social, occupational or other important areas of functioning	Significant impairment in personal, family, social, educational, occupational, or other important functioning
Qualifier	Beyond expected norms for relevant cultural, religious, or developmental stage	Exceeds social, cultural, or religious norms

40). One might say that from the trauma perspective, bereavement under traumatic circumstances that results in the familiar symptom pattern of PTSD is PTSD. From the bereavement perspective, however, the symptom pattern of PTSD following the death of a significant relational figure is but one aspect of what may actually be predominantly dysfunctional grief and disordered mourning (41, 42). Furthermore, we believe that there are additional aspects of traumatizing elements that operate in traumatic bereavement. In some cases, the circumstances of the traumatic death events are highly intermeshed with the interpersonal relationship to the deceased that itself has strong conflictual and negative characteristics (17). Bereavements following troubled interpersonal relationships are one example of this. While the DSM-5 describes both homicide and suicide as qualifying as traumatic circumstances, in reality there are important distinctions to be made. One major difference relates to the fact that the person who dies from suicide is both the one who took life and the one whose life was taken. For many of the bereaved, this combination complicates the grief. Overall, assessment and intervention benefit from attention to greater specificity regarding the complicating and traumatizing features involved in bereavement (43).

The literature on trauma and post-trauma has changed how both clinicians and the lay public think about life-threatening events and their impact (32, 44). With the passage of time and the expanding literature base, clinical practitioners and the general public are better informed as to the incidence, prevalence, and pernicious deleterious effects of exposure to traumatic events (45, 46). Similarly, awareness of the various intervention programs and their reported efficacy has increased as well (6, 47, 48).

Alongside the generally positive effect of the expanding wellspring of knowledge and expertise that has accrued to date in the trauma field, there is a less welcome side effect to this phenomenon as it relates to bereavement. Specifically, since major life threatening events directed either at the self or at a loved one are considered events of significant magnitude as to

satisfy the criterion for a traumatic stressor, this categorization is often seen to encompass the death of a loved one. Once the death of a loved one is categorized as a major stressor of potentially traumatic proportions, the interpersonal and attachment issues involved in bereavement may be downplayed or missed (41, 42, 49). For example, combat soldiers whose comrades have fallen in battle report attention and follow-up related to the post-trauma and minimal consideration of the grief and mourning for fallen comrades (50).

We shall return to amplify these points with clinical material after the presentation of the Two-Track Model of Bereavement (TTMB).

TWO-TRACK MODEL OF BEREAVEMENT (TTMB): A MODEL FOR RESEARCH AND PRACTICE

The TTMB was developed in order to make sense of research and clinical data that demonstrated successful adaptation to life post loss while maintaining a strong connection to the relationship with the deceased in the present (51, 52). The model itself addresses response to interpersonal loss from a bifocal perspective considering both the biopsychosocial functioning of the bereaved (track I), and the nature of the ongoing relational bond to the deceased (track II), across the life cycle (51, 52). Track I's focus on biopsychosocial dysfunction reflects both the medico-psychiatric attention to human suffering and the psychological perspectives rooted in the broad domain of mental health, stress, and trauma literatures. Included here are the biological, behavioral, cognitive, emotional, intrapersonal, and interpersonal ways of one's being in the world. These can be negatively affected following loss for extended periods (5, 14). In cases where there is reason to suspect manifestations of post-trauma, inquiry into the triad of re-experiencing, avoiding, and hyper-alertness is warranted as part of the assessment (7). Viewed most broadly, the exploration of negative and positive changes in

the general domain of function span a wide range of areas. The biopsychosocial approach shares much with the general clinical evaluation of persons facing stressful challenges to their previous mode of living in the world and the way in which they made sense of its meaning (53). These challenges are not fundamentally unique to the bereavement experience and the stresses it brings with it. Thus, exposure to natural disasters, combat, sexual victimization, terror, and a variety of life-threatening situations would warrant the explorations of this first domain in many of the same ways as with the assessment of responses post-bereavement.

The second track of the model, the relationship to the deceased, traces its origins and significance to a wholly different set of assumptions and postulates. In modern terminology, we would stress the relevance of the attachment and object relations literature and the nature of the attachment bond as central to what is unique to bereavement and loss (2, 5, 54). The core insight related to track II of this model is that reworking the relationship to the deceased and the continuing bond with the deceased post-death is a critical feature for understanding response to bereavement. This is the central feature of what makes the loss process unique. The second

domain of the two-track framework prioritizes the nature of the relationship to the deceased and the current status of the emotional bond to the deceased. How the psychological experience of the relationship and its accessibility have changed following death are central pieces of this approach (4). An additional focus of track II is rooted in the story of the death and how it has been integrated and assimilated by the bereaved. Particularly in cases of traumatic deaths, the events surrounding the death may remain unassimilated with potentially serious consequences (10, 22). In some cases, this unintegrated and traumatic segment interferes with the processing of the relationship and the access to the broad range of memories, associations, and emotions concerning the life course with the deceased. **Figure 1** illustrates the domains of the TTMB as they unfold over time.

The importance of combining the two perspectives of functioning and relationship in broad ways formed the basis for the TTMB (7, 51, 52). In this model, the process of adaptation to interpersonal loss is linked to the disruption of homeostatic functioning that accompanies major life stressors on the one hand but also as a byproduct of relating and reconfiguring



FIGURE 1 | Illustration of the two-track model of bereavement (TTMB)¹.

¹The assessment over time waves represent pictorially how a series of time points would look if graphed. The amplitudes are for illustration of fluctuation and not for purposes of conveying degree or direction of the variables.

aspects of the relational bond and attachment system with the deceased on the other hand. The TTMB facilitates the assessment of both functioning and the nature of the continuing attachment to the deceased when significant others die—and this across the entire course of the bereaved person's lifetime. **Figure 2** conveys the mix of independence and interdependence of the two tracks.

The clinical implications of the model derive directly from its binocular focus. The extent to which potential psychological interventions should privilege one or both domains of the response to loss remains an important clinical question (26). The basic elements of the assessment schema of the TTMB and a rubric for clinician ratings are presented in **Table 2**. The 10 domains of track I's biopsychosocial functioning and track II's 11 domains beginning with the death story are relevant in assessing the response to loss at any point in time following the loss. **Table 2** summarizes the scoring scale for clinician assessment of bereaved individuals.

In assessing the impact of loss in cases of traumatic bereavements, the objective circumstances of the loss, with attendant symptomology characteristic of post-trauma are part of the biopsychosocial response. At the same time, however, inquiry and assessment of the death story are highly significant in assessing how the ongoing relationship to the deceased is affected by that (55, 56). Thus parallel focus is also relevant in relationships where the losses are not death but involve significant changes in requiring adjustment due to major changes in the attachment figure (25, 57). For example, in a recent publication, the rationale for a two-track model of dementia grief (TTM-DG) showed how the particular circumstances involved in caring/caretaking for a close family member suffering from dementia was best understood from the bifocal perspective of the TTMB (58). In particular, the ongoing loss

of personhood and identity for a person suffering from cognitive decline and dementia is a challenge and loss for those closest to him or her. For a fuller elaboration of the current applications of the model in clinical work and research, see *Working With the Bereaved: Multiple Lenses on Loss and Mourning* (7).

Focusing on the extent of behavioral difficulties and symptoms of various types is valuable and is sensitive to the ways that bereavement challenges the bereaved to readjust to a new external reality. Whether or not the bereavement response meets criteria for a formal diagnosis, however, many persons seeking assistance after the death of a close person can benefit from the bifocal approach advocated. Many aspects of the relationship to the deceased both pre and post loss require us to learn more about whom was lost. This includes attention to what aspects of that relationship were lost for the bereaved and what portion of that relationship remains active and available to the bereaved after the death. In the case of circumstances of traumatic bereavement, the linkage of trauma with bereavement has often had the paradoxical effect of obscuring the decidedly interpersonal and intrapersonal impact of the loss and replacing it with a focus on the trauma elements (42, 59).

WORKING CLINICALLY WITH THE TTMB IN A CASE OF TRAUMATIC BEREAVEMENT

The following brief case study of assessment and intervention illustrate the use of the TTMB in a case of traumatic



FIGURE 2 | The TTMB in traumatic bereavement².

² Reprinted with permission from Rubin et al., 2012

TABLE 2 | The TTMB clinician rating scale.

Track I: Biopsychosocial difficulties in functioning and post-trauma		Track II: Ongoing relationship to the deceased and death story		
Clinician evaluation		Clinician evaluation		
1. Traumatic responses 2. Anxious affect and cognitions 3. Depressive affect and cognitions 4. Somatic and Health 5. Familial relationships 6. General interpersonal 7. Lowered self-esteem and distress 8. Involvement in life 9. Meaning in life 10. Strengths (growth/resilience)		1. Death story difficulties/unintegrated/interferes with connection to deceased 2. Yearning and closeness 3. Imagery/memory of deceased 4. Positive affect 5. Negative/unmodulated affect 6. Preoccupation/avoidance (specify) 7. Idealization/devaluation (specify) 8. Conflict in the relationship 9. Loss trajectory (rate each: shock, searching, disorganization, and reorganization) 10. Self-system difficulties vis-à-vis deceased and/or death story (specify) 11. Memorialization		
Clinician scoring key				
Absent/mild/limited		Moderate	Severe/strong	Continue to monitor
				Insufficient information

bereavement. In particular, the clinical example illustrates how bereavement can operate as a complex series of traumatizing experiences that have roots and links to both tracks of the TTMB.

Helen, a 36 year old woman, came for treatment to SSR 2 years after the suicide of her husband David. In the initial session, Helen said she was only now ready to get help. Early in the first session, she spontaneously described how she had discovered her husband's suicide and body. Returning home from a brief business trip abroad, she had opened the door to the master bathroom where she was assaulted by "horrific sights and smells and waves of nausea." Her husband's decaying body was lying in the bathtub several days after he had killed himself. He had slit his wrists and the blood flow into the water had left everything a discolored pink after the water had drained away. Her affect as she described her experiences conveyed minimal emotion. While it was clear that the experiences were distressing and painful, her reporting was done in a factual way that was devoid of emotion.

Helen: "Years later, I close my eyes, [and sometimes still] see his body lying there and the red color of the bathtub surrounding him. I have dreams of being physically tortured by someone wearing his face. I have intrusive thoughts about him, this used to be maybe 100 times a day, but now it is much much less. I did most of my work from home and barely went out. I sat in the dark, self-medicated with alcohol and marijuana. I wanted to suffer so I did not see any mental health professionals".

In the initial meetings, Helen shared her personal history and details about her husband and their time together. She was the eldest of two daughters who grew up in an intact family. She had always been "a bit of a tomboy, interested in sports, computers, and mathematics." Previous losses included the death of her father when she was 19; the departure of her younger sister for a job overseas, and her mother's subsequent remarriage and move to the sister's city of residence. Helen had many strengths and was successful during her years of education, and eventually took her skills set to work in the computer world of high-tech which

required a fair amount of international travel. Helen had been introduced to her future husband by a mutual friend and they immediately forged a connection. Soon they were living together, and they married a year later. All told, they had been together for 12 years before David's suicide. David had been raised in a small town, had done well in school and college, eventually entering into government service. At age 30 he had begun to suffer from depressive episodes. He was seen by a mental health professional and prescribed medications. He stopped both soon thereafter. The couple had weathered several of David's depressions including one for which he was hospitalized briefly. Several weeks before her business trip abroad, David had again gone into what seemed to be a depressive mood, but he assured her that his depression was under control and there was no need to see anyone. He said he would be fine so she need not be concerned for his wellbeing. During her stay overseas, they were in contact for the first 2 days before he told her that he was going to be incommunicado for several days as he was going to hike in an area where there was no cellphone coverage. Upon her return, she found his lifeless body at home.

At the time of her request for therapy, Helen exhibited some symptoms of stress and depressive affect, but these did not significantly interfere significantly with her day to day functioning. She had seen a physician for medication for sleep difficulties the first year, but was now sleeping relatively well although there were the occasional nightmares reported. As indicated above, she continued to have intrusive thoughts and images of the death scene on a daily basis, but these were no longer interfering with her functioning. She continued to avoid places that reminded her of David's depressions and she had withdrawn from interactions with their mutual friends. She described heightened vigilance and anxiety in response to phone calls. Not infrequently, and particularly when the phone rang at odd hours, she felt a reflexive fear that the call was about bad news affecting her family overseas. These responses were much reduced from what she had experienced for many months following David's suicide.

Queried about the nature of the memories, thoughts and feelings about the relationship with David, Helen said that she felt shut

down and unable to connect with their good times together. She felt guilty for having left him to go on her business trip, denied feelings of anger vis-a-vis the suicide or his deception and the way he hid his state of mind from her. The memories of David and the recollections of the times when she had enjoyed his company eluded her along with the feelings of pleasure they once held.

Assessment and Intervention Plan With the TTMB

The diagnostic categories applicable to Helen in the DSM-5 were from the trauma and stressor related disorders (32). Helen's current functioning involved significant suffering and many symptoms associated with post-trauma although not sufficiently meeting criteria for a DSM-5 diagnosis of PTSD. The experience of her husband's suicide met the criterion for exposure to a traumatic event; there were intrusions of the death scene; avoidance of places that triggered memories; negative alterations in mood, and a degree of hyper alertness as manifest in her response to phone ringing. The circumstances and degree of clinically significant distress met conditions for "Other Specified Trauma- and Stressor-Related Disorder" with the further specification of persistent complex bereavement disorder. We would point out that this mix of trauma and bereavement in the more formal diagnosis reflects clinical, conceptual and diagnostic overlap, and interpenetration.

From the perspective of the TTMB, Helen had significant distress on many of the track I biopsychosocial categories. Feelings of anxiety, depression, a sense that her life was adrift and without meaning, reduced involvement in social interactions with family and friends, along with her lowered self-esteem reflected the extent to which her biopsychosocial functioning was impaired. At present, on the trauma dimension, there were elements of the triad of intrusions/re-experiencing, avoidance, and hyperarousal. These were significant and consistent with a residue of active post-trauma.

Examining track II's death story and relational bond with her deceased husband conveyed significant difficulties. In the quote included earlier, Helen graphically described David's suicide and the experience of finding his body. The story of the death emphasized the shocking elements, but nowhere was she able to integrate this experience into a broader narrative. There were no indications of some degree of acceptance and assimilation of this experience with the bulk of her relationship with David. Helen returned repeatedly to the question of why he had killed himself in the way that he did without caring about what it would be like for her to find him like that. The narrative of his death did not fit into her description of his depressions and struggles with suicidal thoughts. It was as if this experience began and stopped with the discovery of his body days after his death.

The story of the relationship with David and the limited degree to which memories of their lives together were accessible to her indicated that the relationship bond required therapeutic attention. Helen had difficulty accessing the wellspring of memories of their relationship. Memories of the positive elements of their relationship were few and vague. There was little in the way of perspective or the ability to reference the thick history of their years together. Negative

and conflictual elements of the connection with David were present and associated with pain. She felt guilty over having left David to go overseas and also a sense of general unworthiness and failure as a spouse. Despite the objective dislocation that David's death had caused her, she repeated stated that she was unable to be angry at him for what he had done. With access to positive memory and emotions of their relationship constricted and with a painful focus on David's suicide in the fore, the relationship domain was problematic. In this condition, progression toward adaptive grief and mourning were stymied (2, 5, 7).

Helen was having difficulties on track I and track II and the question of how to focus intervention with respect to these variables was highly relevant. On track I, the elements of trauma, depressive thoughts and emotions, lowered self-esteem, constricted connection with life tasks, connecting to meaning in life, were all rated by SSR as elevated and of moderate severity. The combination of these variables, together with limited family support and other interpersonal relationships, powerfully affected Helen's ability to engage in her life. These track I biopsychosocial difficulties convey significant dysfunction and distress and intervention would need to take them into account.

On track II, Helen had not integrated the death story nor had she been able to retell the story with any degree of flexibility or perspective. The ongoing sense of shock in the way the death event was experienced interfered strongly with her ability to fully describe her husband and the many years of their relationship. This aspect of the death-related trauma is rated on track II because of its significance in impeding grief and mourning and for its interference with access to the interpersonal relationship and the continuing bond with the deceased. The relationship with David was scored as highly significant with the variables related to the death story, negative affect, preoccupation with the deceased, conflict in the relationship, and assault to the self-system as a result of the relationship being coded as of severe proportions. Yearning was pronounced, while positive affect, memories of the good times, and progress toward memorialization of the deceased were limited. **Table 3** gives the clinician scoring of Helen at the end of the intake evaluations on the TTMB variables discussed.

The Treatment Plan Based on the TTMB

In considering a treatment plan, once again, the TTMB framework was used in formulating the intervention plan. Helen's response to the traumatic loss 2 years post-death was characterized by significant difficulties in functioning, features of traumatic stress, and relation based elements that served to reinforce each other. They limited her ability to function and her ability to grieve in ways that facilitated working through of aspects of the loss. With difficulties on both tracks, it was important to formulate an intervention plan that would take into account both domains of the TTMB.

The focus on track I's biopsychosocial functioning and trauma suggested a range of intervention possibilities. The treatment components most relevant for the biopsychosocial difficulties combined elements of supportive psychotherapy with a variety of techniques and exercises that have been associated

TABLE 3 | Helen–Clinical Assessment with the TTMB rating scale at beginning of treatment.

Track I: Biopsychosocial functioning		Track II: Relationship to the deceased and death story	
	Clinician evaluation		Clinician evaluation
1. Traumatic responses	Moderate	1. Death story	Severe
2. Anxious affect/cognitions	Mild	2. Yearning and closeness	Moderate
3. Depressive affect/cognitions	Moderate	3. Imagery/memory	Limited
4. Somatic and health	Mild	4. Positive Affect	Limited
5. Familial relationships	Mild/limited	5. Negative Affect	Severe
6. General interpersonal	Mild/limited	6. Preoccupation/Avoidance	Severe
7. Lowered self-esteem	Moderate	7. Idealization/devaluation	Mild–deval
8. Involvement in life	Moderate	8. Conflict in the relationship	Severe
9. Meaning in life	Moderate	9. Loss Trajectory (Shock, Searching, Disorganization, and Reorganization)	Sh- Severe Se – Limited Dis- Moderate ReO - Limited
10. Strengths (growth/resilience)	Limited	10. Self-system	Severe
		11. Memorialization	Limited
Clinician scoring key			
Absent/mild/limited		Severe/strong	Continue to monitor
			Insufficient information

that included techniques drawn from evidence based treatment strategies (60, 61).

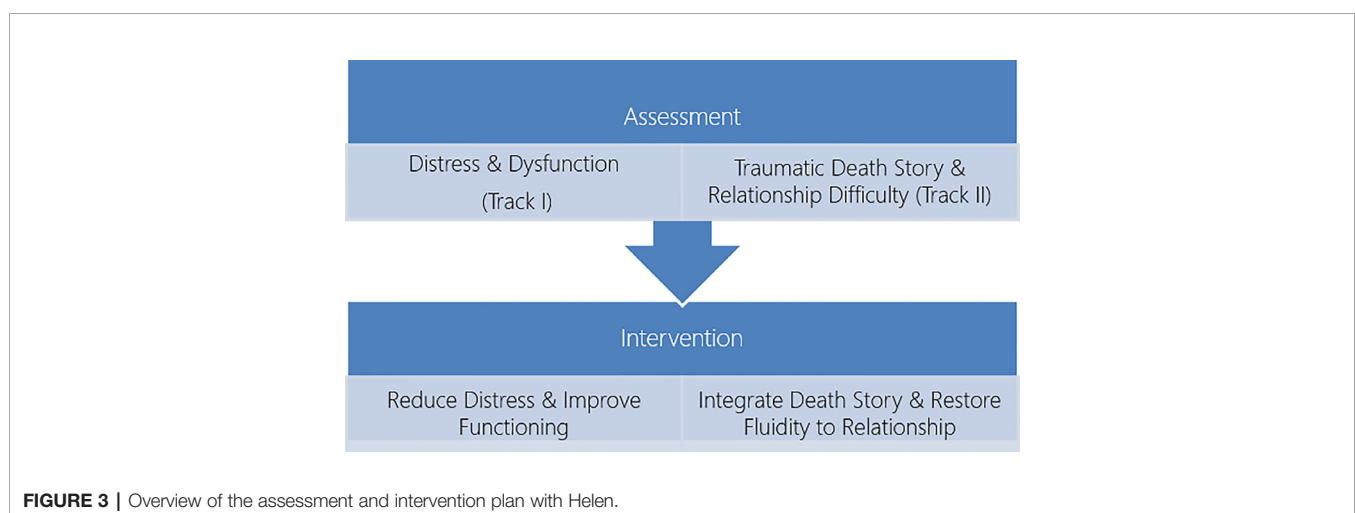
Concurrent with interventions related to track I, the focus on track II drew attention to the importance of transforming the death story. The frozen and shocking death scene effectively locked the death by suicide event into a kind of video clip that did not develop into a story. Finding ways to help Helen take the death story and continue on beyond the death scene, ultimately integrating it to the broader story of David's life, death, and memory were an overarching goal of the track II intervention. The goals of treatment here emerged squarely from the track II conceptualization of the significance of the continuing bond to, and the dynamic relationship with, the complex of memories, thoughts and emotions linked to the deceased. This connection can serve as a positive and supportive presence for the bereaved across the life span (7). **Figure 3** illustrates the double focus of the intervention plan and goals.

Whatever the foci and plans for therapy, it was important to get Helen's cooperation and to enlist her in the treatment plan. Following the initial sessions of history taking, the therapist

shared the basic outline of the TTMB as a way of thinking about responses to loss with the goal of psychoeducation and helping Helen make sense of her response to David's suicide. Following a discussion of the model, the predominant areas of difficulty and dysfunction that she experienced were shared back with her. Therapist and patient were in agreement on the need for intervention, and the areas in her life that were particularly troubling. The goals of treatment were to help her feel better, to assist her with the various domains of functioning that troubled her, to ease the pain and shock of her memory of David's death, and to help her regain a more flexible and supportive connection with his memory.

Critical Junctures in Helen's Treatment

The initial weeks of treatment were devoted to building the therapeutic alliance with Helen. In this initial stage of therapy she filled in details of her current life and the nature of the challenges she faced as well as her feelings of estrangement and longing for David. In order to assist her in reducing her anxiety, depression, and tension, one set of interventions were aimed at increasing



her ability to modulate her affect. Self-observation, journaling, and release of tension *via* physical exercise were prescribed, and she was able to follow through on all these (7). The benefits of mindfulness were explained, and practice in the sessions was introduced (62). Even though she did not continue with mindfulness, the focus on allowing emotions and thoughts to be seen as reflecting the ebb and flow of the mind rather than as thoughts to engage or avoid were helpful to her. Above all, encouraging her to share her thoughts and feelings about those encounters and areas of her life that troubled her without the requirement that they reflect the trauma or the loss were beneficial. This allowed her to explore her thoughts regarding meaning in life, her mixed feelings toward family and friends, and the nature of difficulties in her work and home that were of benefit to her. As she followed through with regular exercise, sporadic periods of mindfulness at home, sharing her thoughts in the sessions and journaling occasionally at home, her mood and sense of self began to improve. These in turn contributed to the strengthening of the therapeutic alliance.

The next stage of treatment began after the therapeutic alliance and initial benefits from treatment were in place. Building on the therapeutic alliance, the major intervention focus directed at Helen's relationship to David and the manner of his death became the center of this next phase of treatment.

The techniques used in psychotherapy for bereavement are myriad (60). In situations where the relationship is constrained or moribund finding ways to open possibilities for change and flow in the relationship can be transformative. Chair work where the bereaved and deceased "interact" and letter writing to the deceased are among the better known of these techniques (60, 63). From the TTMB perspective, in the case of Helen, because the circumstances of her husband's death were so traumatic, they effectively stymied her ability to reorganize her relationship to him. The therapeutic technique was designed to permit Helen to soften the boundaries between the living and the dead, the real and the imaginary, and to introduce the possibility of change and the evolution of the relationship. One could think of this intervention technique as a way of seeking to open "transitional space and fluidity" into what had been a frozen and "dead" relationship (64). At the same time, this intervention technique had potential to allow for the evolution in the story of David's death and the discovery of his body that had heretofore remained traumatic, stuck, and repetitious.

Some 3 months into the therapy, having made gains in emotion regulation, having taken up walking for an hour each day, and having attempted mindfulness but deferred it for later, the more direct focus on Track II's relationship began. The letter writing that involved having Helen write a letter to David with the goal of facilitating the reconnection and expansion of the current relationship. (See chapter 10, pp 151–171 in (7) for a fuller description of this type of intervention.)

The first letter she composed was straightforward:

Dear David,
There are so many things I want to say to you....
things I've thought these past several years.... I want to
be angry at you for what you did, what you promised

me you would never do. I thought you were getting better, ...

You promised you wouldn't hurt yourself, because you said you didn't want to hurt me. So why did you do it? And to do it like that, for me to find you? ... how could you never consider what it would do to me, to find you like that?

I said I want to be angry at you, and as much as I want answers to these questions the truth is I can't be angry with you I just miss you so much. All I think about is what I would do or give up to have another 5 minutes together, just to talk to you and hug you.

In this first letter, the connection is made, and the longing and wish for reconnection are present. She introduces the possibility of complaints directed toward David for his act of suicide, but anger and disappointment are mentioned though not experienced. The longing for the reconnection are very much present.

The next letter was written two weeks later

Dear David,

Starting these letters is always so difficult. It feels strange, like I'm speaking to you again back when you were alive. ... And yet this also feels more impersonal, as even though I feel I am talking to you, I feel less close to you than I did in the past. Maybe that is because I am angry over what you did. ... The truth is that I don't know what has caused it but my feelings for you have changed, and though I think of you constantly still it is not through the same rose colored glasses I once did.

I find myself feeling frustrated more and more by the fallout of your suicide. For so long I blamed you ... How could you not consider what would happen to me, how utterly your actions would damage and destroy my life. (2nd letter).

Commentary

The degree of change between letter one and two is striking. Both letters speak to the ability of Helen to enter into the letter writing exercise and to find herself opening up with her emotions and entering into a dialogue with David. This entrance into a living dialogue brings with it potential for change. The emotion of anger that is denied in letter one emerges strongly in letter 2. More importantly, she indicates that an idealized picture of David referred to in the rose colored glasses of letter two have been replaced by a more sober assessment. The shift in how David is viewed and the change it brings with it in the relationship are pivotal in allowing grief to progress as Helen has more freedom to think about and experience a broader range of emotion than she had earlier. The second letter served as a transformative experience for Helen. Following the letter, Helen spoke about remembering more of her time together with David, and he felt both nearer to her and more alive than she had experienced him for a long time. The broadened access to

memories of David, the anger focused on David instead of only on herself, and the story of the depressions that became part of the narrative around the suicide all reflected the movement toward grieving and mourning. Connecting more fully to David also allowed her to become more whole as well.

The sense of betrayal and assault that Helen experienced in response to the suicide were a traumatizing experience for her. Most importantly, the act of suicide served to shatter the person and internal working model of the David she had known. Not only were the memories and connections to him less available, they were also colored by the action that changed the way she viewed him and their relationship. So that in addition to the trauma of the discovery of the dead body and the assault on the senses involved with that, there was an additional trauma, and one that directly related to the experience of who her husband was for her. Helen in effect was dealing with a double trauma, one of external characteristics of the encounter with the death scene, and one that was rooted in the traumatic encounter with her experience of who her husband was for her, and his actions toward her. On the one hand, the Track I trauma has an objective component of exposure to David's death and his dead body. On the other hand, the Track II trauma was also centered around the relationship with David and the betrayal that his suicide brought to Helen. The death story remained one that she had been unable to integrate and weave into the story of their relationship.

A Brief Note on the Treatment Conclusion

Helen's initial letters to David were followed by more letters, by chair work where she took both roles in the dialogue with her deceased husband, and by her newly recovered ability to talk about their relationship. She continued her journaling, exercise, and self-care activities. The reconnection to the relationship with David allowed her to integrate the death story with the relationship in a way that prioritized David's struggles with depression as most significant, and the mode of death and death scene as secondary. Positive memories, humorous incidents, and discussion of the painful periods of depression came up regularly. Treatment continued for another half-year. Notable changes followed and included a deeper engaged with her work, family, and friends; increasing positive emotions; and reduction in traumatic imagery and dreams. Yearning for David was less pronounced, the death story was better integrated with the life story, and Helen found increased support in the memory of the good times in her relationship with David. In the concluding sessions, her experience of the therapist and therapeutic relationship, and how these contributed to her improvement, were part of the closing of this treatment.

CONCLUDING REMARKS

The interplay between trauma and bereavement in traumatic bereavements has received much attention but consensus remains elusive (65). Attention to the extreme stresses involved in bereavements occurring under traumatic circumstances is

critical to understanding both adaptive and maladaptive bereavement responses. At the same time, the decidedly interpersonal aspects of bereavement, grief and mourning should remain squarely in the center of the understanding of traumatic loss. It is a mark of the maturation of the bereavement field that many clinicians with a bereavement focus pay particular attention to circumstances of trauma. The traumas involved in the death circumstances can be joined by traumas related to the psychological representations of the attachment bond and interpersonal relationship. Any number of bereavement circumstances can result in the bereaved responding with a combination of trauma-related elements. These may be most appropriately understood from a combination of the biopsychosocial (including stress and PTSD), the degree of integration of the story of the death, and the degree and types of psychological engagement with the continuing bond with the deceased. We advocate for the use of the TTMB because its conceptual and applied perspective address the relevant biopsychosocial functioning, death narrative, and relationship with the deceased variables (7, 14, 56).

Clinical work is often geared to assessment and interventions seeking to restore balanced biopsychosocial functioning. In interventions following disordered and maladaptive bereavement, the rebalancing and reworking of the relationship to the deceased may proceed without any direct assistance or awareness of the therapist. In other situations, therapy may focus on the ongoing relationship to the deceased without particular attention to the biopsychosocial status of the griever. It is most efficacious, however, for clinicians to use a balanced approach such as that of the TTMB to address both function and the relationship to the deceased. Such an approach facilitates the return to more adequate function, growth, and adaptation to loss over time.

AUTHOR CONTRIBUTIONS

SSR, RM, and EW contributed to the design and organization of the article. SSR supplied the case material. RM and EW commented and expanded the discussion of the case. All three are responsible for **Table 1** and **Figures 2** and **3**. SSR provided **Figure 1** and **Tables 2** and **3**. EW spearheaded the consideration of the diagnostic issues in the DSM and the ICD, was in charge of references, and fact checked all material. SSR was the lead author who did the most of the writing and editing. RM and EW expanded and critiqued each draft leading to the final product. The authors view this as a team effort. The order of the names reflects not only the extent of contributions on the one hand but also the three names testify to the fact that "the whole is more than the sum of its parts."

ACKNOWLEDGMENTS

The authors thank Alexander Manevich for his valuable assistance at all stages of the manuscript preparation.

REFERENCES

- Freud S. Mourning and Melancholia. In: Strachey J, editor. *Standard edition of the complete psychological works of Sigmund Freud*, vol. 14. London: Hogarth (1957). p. 237–58. (Original work published 1917).
- Klass D, Silverman P, Nickman S eds. *Continuing bonds: New understanding of grief*. Taylor & Francis: Washington, DC (1996).
- Klass D, Steffen E eds. *Continuing bonds in bereavement: New directions for research and practice*. Routledge: New York (2018).
- Rubin SS. The resolution of bereavement: A clinical focus on the relationship to the deceased. *Psychotherapy* (1985) 22(2):231–5. doi: 10.1037/h0085499
- Bowlby J. "Attachment and loss". In: *Loss, sadness and depression*, vol. 3. New York: Basic Books (1980).
- Malkinson R. "Cognitive grief therapy: Constructing a rational meaning to life following loss". In: *PsychNET*. New York: W W Norton & Co (2007). Available at: <https://psycnet.apa.org/record/2006-13248-000>. [cited 2020 Aug 18]. Available from.
- Rubin SS, Malkinson R, Witzum E. *Working with the bereaved: Multiple lenses on loss and mourning*. Routledge: New York (2012).
- Bonanno GA. Loss, Trauma, and Human Resilience: Have We Underestimated the Human Capacity to Thrive after Extremely Aversive Events? *Am Psychol* (2004) 59(1):20–8. doi: 10.1037/0003-066X.59.1.20
- Kersting A, Brähler E, Glaesmer H, Wagner B. Prevalence of complicated grief in a representative population-based sample. *J Affect Disord* (2011) 131(1–3):339–43. doi: 10.1016/j.jad.2010.11.032
- Kristensen P, Weisaeth L, Heir T. Bereavement and mental health after sudden and violent losses: A Review. *Psychiatry* (2012) 75(1):76–97. doi: 10.1521/psyc.2012.75.1.76
- Lundorff M, Holmgren H, Zachariae R, Farver-Vestergaard I, O'Connor M. Prevalence of prolonged grief disorder in adult bereavement: A systematic review and meta-analysis. *J Affect Disord* (2017) 212:138–49. doi: 10.1016/j.jad.2017.01.030
- Shear MK. Grief and mourning gone awry: Pathway and course of complicated grief. *Dialogues Clin Neurosci* (2012) 14(2):119–28.
- Steil R, Gutermann J, Harrison O, Starck A, Schwartzkopff L, Schouler-Ocak M, et al. Prevalence of prolonged grief disorder in a sample of female refugees. *BMC Psychiatry* (2019) 19(1):148. doi: 10.1186/s12888-019-2136-1
- Malkinson R, Rubin SS, Witzum E eds. *Traumatic and Non-traumatic Loss and Bereavement: Clinical Theory and Practice*. International /Psychosocial Press: Madison, CT (2000).
- Barlé N, Wortman CB, Latack JA. Traumatic bereavement: Basic research and clinical implications. *J Psychother Integr* (2017) 27(2):127–39. doi: 10.1037/int0000013
- Shear MK, Smith-Caroff K. Traumatic loss and the syndrome of complicated grief. *PTSD Res Q* (2002) 13(1):1–16. doi: 10.1037/e400332008-001
- Rubin SS, Malkinson R, Witzum E. Clinical aspects of a DSM Complicated Grief Diagnosis: Challenges, dilemmas, and opportunities. In: Stroebe MS, Hansson RO, Schut H, Stroebe W, editors. *Handbook of bereavement research and practice: Advances in theory and intervention*. Washington, DC: American Psychological Association Press (2008). p. 187–206.
- Stroebe MS, Hansson RO, Schut H, Stroebe W eds. *Handbook of Bereavement Research and Practice*. In: *Advances in Theory and Intervention*. Washington, DC: American Psychological Association Press.
- Smid GE, Kleber RJ, de la Rie SM, Bos JBA, Gersons BPR, Boelen PA. Brief eclectic psychotherapy for traumatic grief (BEP-TG): Toward integrated treatment of symptoms related to traumatic loss. *Eur J Psychotraumatol* (2015) 6. doi: 10.3402/ejpt.v6.27324
- Stroebe M, Schut H, Finkenauer C. The traumatization of grief? A conceptual framework for understanding the trauma-bereavement interface - PubMed. *Isr J Psychiatry* (2001) 38(3–4):185–201.
- Neria Y, Litz BT. Bereavement by traumatic means: The complex synergy of trauma and grief. *J Loss Trauma* (2004) 9(1):73–87. doi: 10.1080/15325020490255322
- Rynearson EK. *Retelling violent death*. Routledge: New York (2001).
- Currier JM, Neimeyer RA, Berman JS. The Effectiveness of Psychotherapeutic Interventions for Bereaved Persons: A Comprehensive Quantitative Review. *Psychol Bull* (2008) 134(5):648–61. doi: 10.1037/0033-2909.134.5.648
- Christ GH, Bonanno G, Malkinson R, Rubin SS. Bereavement experiences after death of a child. Appendix E. In: Field MJ, Behrman RE, editors. *When children die: Improving palliative and end-of-life for children and their families*. Washington, DC: Institute of Medicine/National Academy Press (2003).
- Yehene E, Brezner A, Ben-Valid S, Golan S, Bar-Nadav O, Landa J. Factors associated with parental grief reaction following pediatric acquired brain injury. *Neuropsychol Rehabil* (2019) 45(1):1–24. doi: 10.1080/09602011.2019.1668280
- Rubin SS, Bar-Nadav O. The Two-Track Bereavement Questionnaire for Complicated Grief (TTBQ-CG31). In: Neimeyer RA, editor. *Techniques of Grief Therapy: Assessment and Intervention*. New York: Routledge (2016). p. 87–98.
- Neria Y, Gross R, Litz B, Maguen S, Insel B, Seiramarco G, et al. Prevalence and psychological correlates of complicated grief among bereaved adults 2.5–3.5 years after september 11th attacks. *J Trauma Stress* (2007) 20(3):251–62. doi: 10.1002/jts.20223
- Djelantik A, Robinaugh DJ, Kieber RJ, Smid GE, Boelen PA. Symptomatology following loss and trauma: Latent class and network analyses of prolonged grief disorder, posttraumatic stress disorder, and depression in a treatment seeking trauma-exposed sample. *Depress Anxiety* (2020) 37(1):26–34. doi: 10.1002/da.22880
- Prigerson HG, Horowitz MJ, Jacobs SC, Parkes CM, Aslan M, Goodkin K, et al. Prolonged Grief Disorder: Psychometric Validation of Criteria Proposed for DSM-V and ICD-11. Brayne C, editor. *PLoS Med* (2009) 6(8):e1000121. doi: 10.1371/journal.pmed.1000121
- Shear MK, Simon N, Wall M, Zisook S, Neimeyer R, Duan N, et al. Complicated grief and related bereavement issues for DSM-5. *Depress Anxiety* (2011) 28(2):103–17. doi: 10.1002/da.20780
- Wagner B, Maercker A. The Diagnosis of Complicated Grief as a Mental Disorder: A Critical Appraisal. *Psychol Belg* (2010) 50(1–2):27. doi: 10.5334/pb-50-1-2-27
- American Psychiatric Association. *Diagnostic and Statistical Manual of Mental Disorders. 5th Ed.* American Psychiatric Association Press: Arlington (2013).
- World Health Organization. *International classification of diseases for mortality and morbidity statistics (11th Revision)*. (2018).
- World Health Organization. *ICD-11 - Mortality and Morbidity Statistics - 6B42 Prolonged Grief disorder*. (2019). Available at: <https://icd.who.int/browse11/l-m/en#/http://id.who.int/icd/entity/1183832314>.
- American Psychiatric Association. *View and Comment on Recently Proposed Changes to DSM-5*. (2020). Available at: <https://www.psychiatry.org/psychiatrists/practice/dsm/proposed-changes>.
- Eisma MC, Boelen PA, Lenferink LIM. Prolonged grief disorder following the Coronavirus (COVID-19) pandemic. *Psychiatry Res* (2020) 288:113031. doi: 10.1016/j.psychres.2020.113031
- Killikelly C, Maercker A. Prolonged grief disorder for ICD-11: the primacy of clinical utility and international applicability. *Eur J Psychotraumatol* (2017) 8 (sup6):1476441. doi: 10.1080/20008198.2018.1476441
- Maciejewski PK, Prigerson HG. Prolonged, but not complicated, grief is a mental disorder. *Br J Psychiatry* (2017) 211(4):189–91. doi: 10.1192/bjp.bp.116.196238
- Maercker A, Znoj H. The younger sibling of PTSD: similarities and differences between complicated grief and posttraumatic stress disorder. *Eur J Psychotraumatol* (2010) 1(1):5558. doi: 10.3402/ejpt.v1i0.5558
- Lahav Y, Solomon Z. *From reliving to remembering: Treatments for trauma*. Reising. (Hebrew: Israel (2019).
- Witzum E, Malkinson R, Rubin SS. Traumatic grief and bereavement resulting from terrorism: Israeli and American perspectives. In: Heilman SC, editor. *Death, bereavement, and mourning*. New York: Transaction Books (2005).
- Witzum E, Malkinson R, Rubin SS. Loss, traumatic bereavement and mourning culture: The Israel example. In: Ataria Y, Gurevitch D, Pedya H, Neria Y, editors. *International Handbook of Trauma and Culture*. New York: Springer (2016).
- Layne CM, Kaplow JB, Oosterhoff B, Hill RM, S. Pynoos R. The Interplay between Posttraumatic Stress and Grief Reactions in Traumatically Bereaved Adolescents: When Trauma, Bereavement, and Adolescence Converge. *Adolesc Psychiatry (Hilversum)* (2017) 7(4):266–85. doi: 10.2174/2210676608666180306162544
- Herman J. *Trauma and recovery*. Basic Books: New York (1997).

45. Eilefson SL. *The Trauma Thesis: Medical and Literary Representations of Psychological Trauma in the Twentieth Century*. Loyola University Chicago: Chicago, IL (2015). Available at: https://ecommons.luc.edu/luc_diss/1635.
46. Kleber RJ. Trauma and Public Mental Health: A Focused Review. *Front Psychiatry* (2019) 10(JUN):451. doi: 10.3389/fpsy.2019.00451
47. Malkinson R. The ABC of rational response to loss. In: Niemeyer RA, editor. *Techniques of grief therapy: Creative principles for counseling the bereaved*. New York: Routledge (2013). p. 129–32.
48. Watkins LE, Sprang KR, Rothbaum BO. Treating PTSD: A Review of Evidence-Based Psychotherapy Interventions. *Front Behav Neurosci* (2018) 12:258. doi: 10.3389/fnbeh.2018.00258
49. Green BL. Traumatic loss: Conceptual and empirical links between trauma and bereavement. *J Pers Interpers Loss* (2000) 5(1):1–17. doi: 10.1080/10811440008407845
50. Rubin SS, Malkinson R, Koren D, Mor Yosef S, Witztum E. Military bereavement and combat trauma. In: Ringel S, Brandell JR, editors. *Trauma: Contemporary Directions in Theory, Practice, and Research*. Thousand Oaks, CA: SAGE Publications (2011).
51. Rubin S. A two-track model of bereavement: Theory and application in research. *Am J Orthopsychiatry* (1981) 51(1):101–9. doi: 10.1111/j.1939-0025.1981.tb01352.x
52. Rubin SS. The Two-Track Model of bereavement: Overview, retrospect and prospect. *Death Stud* (1999) 23(8):681–714. doi: 10.1080/074811899200731
53. Neimeyer RA ed. *Meaning Reconstruction and the Experience of Loss*. American Psychological Association: Washington, DC (2001).
54. Bar-Nadav O, Rubin SS. Love and Bereavement: Life Functioning and Relationship to Partner and Spouse in Bereaved and Nonbereaved Young Women. *OMEGA - J Death Dying* (2016) 74(1):62–79. doi: 10.1177/0030222815598035
55. Kosminsky PS, Jordan JR. *Attachment-informed grief therapy: The clinician's guide to foundations and applications*. Routledge: New York (2016).
56. Rubin SS, Malkinson E, Witztum E. The Two-Track Model of Bereavement and Continuing Bonds. In: Klass D, Steffen E, editors. *Continuing bonds in bereavement: New directions for research and practice*, 2nd Ed. New York: Routledge (2018). p. 17–30.
57. Yehene E, Zaksh Y, Davidian M, Bar-Nadav O, Elyashiv M. Locked-in your heart-shaped box: Familial-role and attachment orientation as predictors of grief in prolonged disorders of consciousness vs. death. *Death Stud* (2020) 44(8):510–20. doi: 10.1080/07481187.2019.1586795
58. Rubin SS, Manevich A, Doron II. The Two-Track Model of Dementia Grief (TTM-DG): The theoretical and clinical significance of the continuing bond in sickness and in death. *Death Stud* (2019) 1–17. doi: 10.1080/07481187.2019.1688014
59. Cloitre M. The “one size fits all” approach to trauma treatment: should we be satisfied? *Eur J Psychotraumatol* (2015) 6(1):27344. doi: 10.3402/ejpt.v6.27344
60. Neimeyer RA. *Techniques of grief therapy: Creative principles for counseling the bereaved*. Niemeyer RA, editor. Routledge: New York (2013).
61. Lambert MJ ed. *Bergin and Garfield's handbook of psychotherapy and behavior change*. 6th Ed. Wiley: New York (2013).
62. Williams M, Teasdale J, Segal Z, Kabat-Zinn J. *The Mindful Way through Depression*. Guilford: New York (2007).
63. Witztum E, van der Hart O, Friedman B. The use of metaphors in psychotherapy. *J Contemp Psychother* (1988) 18(4):270–90. doi: 10.1007/BF00946010
64. Winnicott D. *Playing and reality*. Basic Books: New York (1971).
65. Djelantik AAAMJ, Smid GE, Mroz A, Kieber RJ, Boelen PA. The prevalence of prolonged grief disorder in bereaved individuals following unnatural losses: Systematic review and meta regression analysis. *J Affect Disord* (2020) 265:146–56. doi: 10.1016/j.jad.2020.01.034

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Copyright © 2020 Rubin, Malkinson and Witztum. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.



Different Trajectories of Prolonged Grief in Bereaved Family Members After Terror

Pål Kristensen^{1*}, Kari Dyregrov² and Rolf Gjestad^{1,3}

¹ Center for Crisis Psychology, University of Bergen, Bergen, Norway, ² Faculty of Health and Social Sciences, Western Norway University of Applied Sciences, Bergen, Norway, ³ Research Department, Division of Mental Health, Haukeland University Hospital, Bergen, Norway

OPEN ACCESS

Edited by:

Birgit Wagner,
Medical School Berlin, Germany

Reviewed by:

Sebastian Trautmann,
Medical School Hamburg, Germany
Lonneke Lenferink,
University of Groningen, Netherlands
Jos De Keijser,
University of Groningen, Netherlands

*Correspondence:

Pål Kristensen
pal.kristensen@uib.no

Specialty section:

This article was submitted to
Mood and Anxiety Disorders,
a section of the journal
Frontiers in Psychiatry

Received: 24 March 2020

Accepted: 14 September 2020

Published: 14 October 2020

Citation:

Kristensen P, Dyregrov K and
Gjestad R (2020) Different Trajectories
of Prolonged Grief in Bereaved Family
Members After Terror.
Front. Psychiatry 11:545368.
doi: 10.3389/fpsy.2020.545368

Introduction: The loss of a loved one in a terror incident is associated with elevated risk for mental health disorders such as prolonged grief disorder (PGD) and posttraumatic stress disorder (PTSD), but the long-term adaptation after such losses are not well understood. This study aims to explore the trajectories of PGD among parents and siblings ($n = 129$) after the 2011 terror attack on Utøya Island, Norway.

Methods: The 19-item Inventory of Complicated grief (ICG) was used to measure PGD at 18, 28, and 40 months post-loss. Latent class growth analysis (LCGA) was used to identify trajectories of grief and a multinomial regression analysis was conducted to examine predictors of class membership.

Results: The analysis identified three grief trajectories; *moderate/decreasing* class (23%), *high/slow decreasing* class (64%), and a *high/chronic* class (13%). Predictors of high/slow recovery or chronic grief was female gender, previous depressive symptoms, and intrusion and avoidance symptoms.

Conclusion: The findings highlights the difficult grief process and slow recovery that characterizes the majority of close family members bereaved by a terror-incident. Community mental health programs should strive for both early outreach and long-term follow-up after such incidents.

Keywords: bereavement, terror, prolonged grief, trajectories, latent class growth analysis

INTRODUCTION

Prolonged grief disorder (PGD) is a chronic, unrelenting grief that was included in the revised International Classification of Diseases (ICD-11) in 2018 (1) and is now also proposed for inclusion in DSM-5. PGD is distinguishable from other mental disorders such as depression and PTSD (2, 3), and is associated with poor physical health, reduced quality of life, and functional impairment (4). The core symptoms of PGD are intense yearning or preoccupation with the deceased, combined with severe emotional pain related to the loss (e.g., difficulties accepting the death, anger or bitterness, feeling that life is meaningless). PGD affects ~10% after expected losses, but certain subgroups show higher prevalence rates. A recent meta-analysis found that 50% of bereaved after unnatural losses (e.g., suicide, homicide, accident or combat-related deaths) experience PGD (5).

Persons who lose a loved one to terror-incidents are more vulnerable for developing PGD and other mental health disorders than persons bereaved by other types of disasters (5). High prevalence of PGD (43–83%) and comorbid PTSD (43–85%) have been found after different terror-incidents such as 9/11 (6), the 2011 Utøya-killings (7), and the 2015 terror attack in Paris (8). Long-term studies are limited, but one recent study by Cozza et al. (9) examined the mental health status of bereaved family members 15 years after the 9/11 incident and concluded that the majority of family members could be considered healthy.

Both degree of exposure to the incident and closeness in relationship to the deceased affects the level of mental health difficulties. In a study of 704 adults bereaved by the 9/11 terror attacks Neria, Gross et al. (6) found elevated levels of PGD among persons that witnessed the attacks on television and among parents who had lost a child.

The co-existence of trauma reactions or PTSD symptoms and grief may create a synergy effect that delays recovery (10). A common assumption is that traumatic imagery or intrusions may hinder normal grief from progressing. However, recent studies have also suggested that increase in grief levels predict later levels of PTSD and not vice versa (11, 12). Overall, the combination of PGD and PTSD is associated with functional impairment (13).

Longitudinal bereavement studies have traditionally relied on mean scores to measure grief reactions across time. While these studies are informative, mean scores do not capture the diversity of grief reactions over time (14). More advanced quantitative methods, latent class growth analysis are able to identify subpopulations or different pathways to adjustment, also termed trajectories. Few studies have investigated trajectories of grief or PGD after disasters. In a study of 170 adults bereaved by the South East-Asian tsunami in 2004 Sveen et al. (15) examined the course of PGD on three different occasions up to 6 years after the disaster. Three distinct trajectories were identified: recovery (48% of the sample), resilient (41%), and chronic (11%). The strongest predictor of chronic grief was loss of a child. Lenferink et al. (16) examined trajectories of PGD, depression, and PTSD on four occasions up to 2½ years post-loss among 172 adults bereaved by a plane disaster in Ukraine in 2014. Two trajectories of grief were identified: mild (82%) and chronic (18%). Low education was associated with the chronic grief trajectory. These studies demonstrate that the majority of disaster-bereaved adults will adjust to their loss, but a significant minority continue to struggle with chronic grief reactions several years after the death.

While studies of disaster-bereaved populations suggest that resilience and recovery are the two most common trajectories, no study has, as far as we know, examined trajectories of PGD among close family members after a terror-incident. Subsequently, our knowledge of their long-term adaptation after terror is scarce.

AIM OF THE STUDY

The main aim of this study was to examine the trajectories of PGD among close family members bereaved by the 2011

terror attack at Utøya, Norway. A second aim was to investigate predictors of the different grief trajectories. We chose factors that has shown to predict PGD in other studies such as gender (17), type of loss (18), exposure to the incident (18), previous losses (19) and pre-loss mental health difficulties (20). In addition we chose to explore whether trauma reactions such as intrusions and avoidance would affect recovery from their loss (21). Satisfaction of social support was also included since it is regularly mentioned among the bereaved as important for recovery. These factors were all measured on timepoint 1.

THE EVENT

On July 22nd, 2011 a Norwegian born terrorist detonated a 950 kg car bomb close to several governmental buildings in the city of Oslo, killing 8 persons and severely injuring 10. Just before the bomb went off the terrorist drove to Utøya, a small island outside of Oslo, where over 500 adolescents and young adults were attending a political youth summer camp for the Norwegian Labor party. The terrorist, dressed as a police officer, chased people all over the island for 1 h and 20 min with the aim of killing as many as possible. Before he was arrested, he had killed 69 persons between 14 and 51 years of age, and physically injured 56.

MATERIALS AND METHODS

This project is derived from a longitudinal study, which examines the mental health effects of the loss of a child or sibling in the 2011 terror attack at Utøya Island, Norway. Participants completed a self-report questionnaire 18 (T1), 28 (T2), and 40 months (T3) after the attack.

PROCEDURES AND PARTICIPANTS

Biological parents and siblings, as well as step-parents and step-siblings of the adolescents and adults who were killed at Utøya were invited to participate in the study. Public records of the deceased were linked to the National Population Register (NPR) in order to obtain the names of the biological parents and siblings. The parents and siblings were first sent an informational letter regarding participation in the study. Those who wanted to participate returned their informed consent forms and received the questionnaire by ordinary mail or filled it in using Survey Monkey (a digital platform). Parents and/or siblings who were on Utøya during the terror attack were excluded from the study since they were invited to participate in a project on the direct survivors' mental health. As two of the deceased were non-Norwegian citizens and not registered in the NPR, their families were also excluded from the study. Moreover, one of the deceased had no parents alive.

A total of 208 biological parents and siblings were identified through the NPR, and 121 of them participated (58%). In addition, seven step-parents and one step-sibling participated in the study. Thus, the total sample consisted of 129 bereaved, that is, 86 parents and 43 siblings. At T1 103 persons participated, at

T2 123 persons participated (including 25 new persons), and at T3 111 persons participated.

A total of 83 (64%) persons participated at all three time points. A total of 17 persons (13.2%) dropped-out from the study either after T1 ($n = 5$) or T2 ($n = 12$). There were no significant differences between 'drop-outs' and 'completers' neither on the sumscore on the ICG ($M = 39.18$ vs. $M = 36.62$), $t = 0.75$ (96), $p = 0.46$), on the intrusion subscale ($M = 17.12$ vs. $M = 16.15$, $t = 0.51$ (96), $p = 0.61$) nor on the avoidance subscale ($M = 15.35$ vs. $M = 13.06$, $t = 1.28$ (96), $p = 0.20$). The participants came from 51 families representing 74% of the deceased, with one-to-six members belonging to the same family.

The project was approved by the Regional Committees for Medical and Health Research Ethics in Norway.

MEASURES

The survey consisted of questions mapping socio-demographics, loss-related questions, questions related to exposure, previous depressive symptoms, previous losses, psychosocial help after the terror-incident, PTSD and PGD.

Exposure was measured with the following question: "Were you in contact with your child or sibling either by telephone or SMS during the terror attack?" (yes vs. no). Previous depressive symptoms were measured with the following question: *Have you ever before July 22nd 2011 had low mood, felt depressed or had feelings of hopelessness for 4 weeks or more* (yes vs. no)? Previous losses were measured with the following question: *Have you any time before July 22nd 2011 lost someone through death whom you were closely attached to?* (yes vs. no). Received psychosocial help was measured with the following question: *Have you received help from mental health care after your loss?* (yes vs. no).

The *Inventory of Complicated Grief* (ICG) (22) was used to measure PGD. The ICG consists of 19 items; answers were ranked on a 5-point scale ranging from never (0) to always (4). Scores vary from 0 to 76 with a high score indicating a higher level of PG. The ICG has shown high internal consistency, test-retest reliability, and concurrent validity (22). The Norwegian version of ICG has shown good psychometric properties (23), and the internal reliability was found to be good across all three time points ($\alpha = 0.89$ – 0.92). A cut-off score >25 indicates probable PGD (24).

The *Impact of Event Scale-Revised* (IES-R) (24) was used to measure symptoms of post-traumatic stress. IES-R consists of 22 items divided into three subscales: intrusion (re-experiencing the traumatic event), avoidance (avoiding trauma reminders), and hyperarousal (disturbed sleep, irritability, hypervigilance). Replies were given on a 5-point scale ranging from not at all (0) to extremely (4). The Norwegian version has shown good psychometric properties in a non-clinical sample (25). The internal reliability was found to be good across the three time points for intrusion ($\alpha = 0.83$ – 0.90), for avoidance ($\alpha = 0.83$ – 0.85), and arousal ($\alpha = 0.86$ – 0.88).

One item from The *Crisis Support Scale* (26) was used to measure satisfaction with social support: "Overall, are you

satisfied with the social support you have received after the terror incident?" This question has been used in other disaster studies (27). The respondents rated their overall satisfaction on a seven-point Likert scale, ranging from 1 (never) to 7 (always).

STATISTICAL ANALYSIS

SPSS 23.0 was used for descriptive statistics, frequency, cross tabulation, student t -test, generalized mixed logistic regression, and linear mixed models (28). These two mixed (multilevel) models were used in order to account for family clustering in the data. The p -level was set to 0.05 (two-tailed). Mplus 7.4 was used for analyzing level and change in PGD as latent contrast difference score models (29). This model, which is a restricted latent growth curve model, analyzing each interval as separate pieces of change, was combined with mixture modeling in order to explore data heterogeneity and identify different sub-groups (latent classes) with different levels and change profiles of PGD (30). The mixture model was specified as a Latent Class Growth Analysis (LCGA) which constrain the variances in intercept and slope factors constrained to zero. This model may give one more class than the growth mixture model (GMM), with these variances estimated freely.

Based on an unconditional latent difference score in PGD, LCGA were analyzed (31). Evaluation of the number of classes was based on the entropy index, and the model fit indices from Akaike Information Criterion (AIC), the Bayesian Information Criterion (BIC) and sample-adjusted BIC (SA-BIC). Lower AIC, BIC, and SA-BIC values indicate a better model fit (32, 33), while entropy values should preferably be over 0.80, at least 0.70 (31). BIC and bootstrap LRT have been found to perform best out of all these indices across several simulated models (34).

Statistically significant improvement as a result of adding classes was tested by the Vuong-Lo-Mendell-Rubin Likelihood Ratio Test (LRT), and by a parametric bootstrapped likelihood ratio test for $k-1(H_0)$ vs. k classes with 1,000 bootstrap draws (35). The evaluation was in addition based on the estimated class size and relative class frequencies. A sample size of 25 classified subjects (5%) has been proposed (36).

The Maximum Likelihood Estimator with robust standard errors (MLR) was chosen in order to account for non-normality (32). Due to observation within families, the clustering option was used in order to achieve correct standard errors. The Full Information Maximization Likelihood method uses all available data under the "Missing at Random" assumption (32, 37). Thus, the effect of missing data is minimized (38), in contrast to listwise deletion methods, and ensures stronger statistical power and generalizability to the results. However, this do not rule out that missingness may be related to the values that would be observed if given, the "missing not at random" situation.

After the model with the best fit was determined, a multinomial logistic regression analysis examined potential predictors of class membership, including gender, type of loss, previous losses, previous depressive symptoms, exposure to the attack, received psychosocial help after the attack, social support

TABLE 1 | Sample characteristics and mean sum scores on ICG and IES-R ($n = 129$).

	Frequency	Percentage
Age		
12–24	27	20.9
25–40	17	13.2
40+	85	65.9
Gender		
Female	77	59.7
Male	52	40.3
Type of loss		
Child	86	66.7
Sibling	43	33.3
Exposed to incident		
Yes	22	24.2
No	107	75.8
Pre-22.7 depression		
Yes	24	24.5
No	105	75.5
Previous loss		
Yes	100	77.5
No	29	22.5
Psychosocial help after 22.7		
Yes	88	68.2
No	41	31.8
	Mean (range)	SD
SS—satisfaction with social support (at T1)	4.67 (1–7)	1.59
IES-R Intrusion (sumscore at T1)	16.38 (1–32)	6.41
IES-R Avoidance (sumscore at T1)	14.36 (0–30)	6.34
ICG (total sumscore at T1)	37.52 (4–66)	11.59
ICG T2 (total sumscore at T1)	34.49 (2–72)	13.92
ICG T3 (total sumscore at T1)	32.46 (3–70)	13.48

(measured at T1), intrusion (measured at T1), and avoidance (measured at T1). The multinomial regression compares each group with a reference category.

RESULTS

The characteristics of the sample and descriptive statistics of PG is shown in **Table 1**. A total of 67% had lost a child among the participants. About half the parents were female (52.3%). Their mean age was 51.6 years ($SD = 6.9$), and almost all parents were married or cohabitants (85.5%). The mean age of the deceased children was 18.7 years ($SD = 5.4$). In the sibling sub-group 74.4% were female. Their mean age was 22.3 years ($SD = 7.6$), and 55.8% had lost a sister. The mean age of the deceased siblings was 18.6 years ($SD = 5.2$).

Based on the fit measures and the number of subjects and frequencies, a three-class solution was decided to be the final model (**Table 2**). The six-class model represented the variability in the observed scores even better but could not be used for

TABLE 2 | Latent class growth analysis (LOGA) of the PG sum score with fit measures for evaluation ($n = 129$).

Classes	AIC	BIC	SABIC	LRT	LRTs	Entropy	Least class predicted n (%)
				p -value ^a	p -value ^b		
1	3,090	3,107	3,088	-	-	-	-
2	2,987	3,016	2,984	0.310	0.000	0.71	55 (42.6)
3	2,906	2,946	2,902	0.004	0.000	0.89	17 (13.2)
4	2,891	2,942	2,885	0.142	0.001	0.81	15 (11.6)
5	2,886	2,950	2,880	0.259	0.099	0.80	14 (10.9)
6	2,887	2,961	2,879	0.689	-	0.81	8 (6.2)

AIC, akaike information criterion; BIC, bayes information criterion, SABIC, sample-adjusted bi; LRT, likelihood ratio test. ^aVuong-Lo-Mendell-Rubin LRT for $k-1(H_0)$ vs. k classes. ^b Parametric bootstrapped LRT for $k-1(H_0)$ vs. k classes (1,000 bootstrap draws). Clustering corrections removed in order to being able to estimate the models.

further analyses due to the very low sample size in several classes. The three-class model is presented in **Figure 1**. The subjects in the *moderate/decreasing class* ($N = 29/22.5\%$) started with a PG score of 25.58, and showed a statistically significant reduction in the first period (-0.78 , $p < 0.01$), but not in the second period (0.01 , $p = 0.972$). The *high/slow decreasing class* ($N = 83/64.3\%$) started with a PG score of 39.30 and showed statistically significant reduction in both time intervals ($T1-T2 = -0.32$, $p < 0.01$; $T2-T3 = -0.25$, $p < 0.05$). The *high/chronic grief class* ($N = 17/13.2\%$) started with a PG score of 50.95, increased their scores significantly in the first interval ($T1-T2 = 0.62$, $p < 0.05$), but showed no further change ($T2-T3 = -0.12$, $p = 0.526$).

The generalized mixed multinomial regression results are presented in **Table 3**. Adjusted predictor estimates showed an increased probability for being in the *high/slow decreasing class* in contrast to the *moderate/decreasing class* (reference), if the subject was female, reported symptoms of depression before the attack, and had higher scores on intrusion (measured at T1). We found increased probability for being in the *high/chronic grief class* in contrast to the *moderate/decreasing grief class* (reference) if the subject was a female, reported symptoms of depression before the attack, and had higher scores on intrusion or avoidance (both measured at T1). The type of loss was not related to being in the *high/chronic grief class*.

DISCUSSION

This is, to our knowledge, the first study of trajectories of PGD following a terror-incident, and we found that a three-class model best represented the data. All three groups started above the recommended threshold for probable PGD (22), but had different developments across time. The classes included a high/slow recovery trajectory of (64%) with initial high level of grief which attenuated slowly from 1 to 3½ years after the incident; a moderate/decreasing trajectory (23%) with an initial moderate to high level of grief, which decreased below the threshold for PGD and a chronic trajectory (13%) which started with very high level of grief which even increased slightly from 1 to 3 1/2 years after the loss.

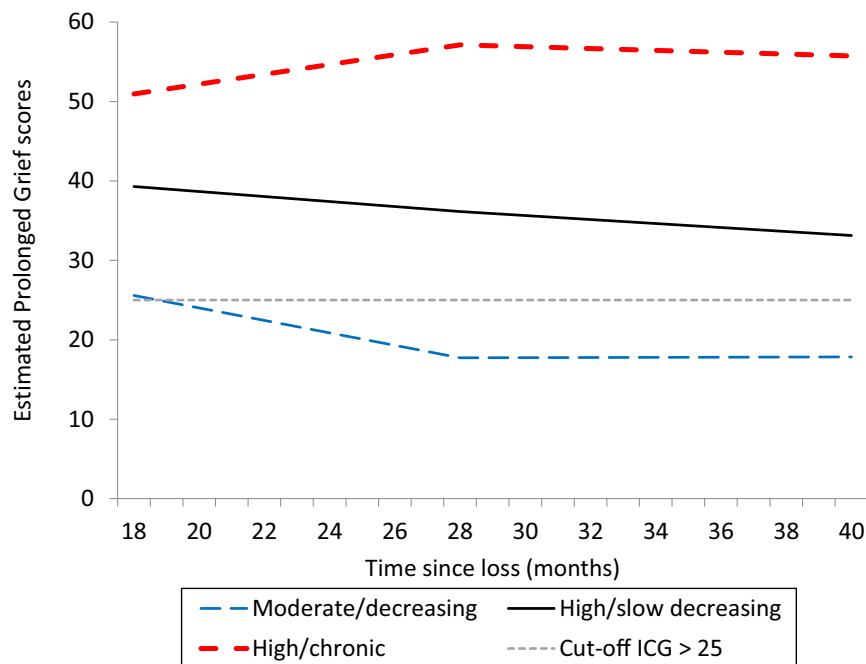


FIGURE 1 | Different grief trajectories among bereaved family members after the 2011 terror attack on Utøya Island.

The lack of a distinct resilient group is noteworthy, since most disaster-studies find resilience to be among, if not the most common trajectory (15, 16). Still, it is consistent with studies that suggest that resilience is far from normative among grieving parents (39). It is worth noting that the sample in the current study was homogenous constituting only of parents and siblings of those who were killed. Closeness in the relationship to the deceased is a stronger risk factor for PGD than both the circumstances of the death and time since the loss (40, 41). Also, almost all of those who were killed were adolescents between 14 and 19 years of age, which is an age that has been associated with more intense parental grief than loss of either a younger child or adult child (42). Also, some suggest that vulnerable persons find more meaning in participating in research (43). If this is true we may have a biased sample. On the other hand, those who struggle most with their grief may also refuse to participate in order to avoid reminders of the loss (44).

The finding that almost 80% of the participants displayed either a high grief level and a slow recovery or chronic grief is alarming, and suggest that the large majority of terror-bereaved family members may struggle for several years with adapting to their loss. Several factors may account for this. In addition to losing a child or a sibling, which is a devastating experience in itself, the terror-bereaved need to cope both with the traumatic circumstances of the loss and with secondary stressors such as media coverage and a potential trial which function as constant reminders of the incident. This affects their opportunity to grieve privately, but also to take time off from their grief (45).

Our finding that pre-loss depressive symptoms predicted chronic grief is regularly documented in studies of PGD, and should be routinely screened for after traumatic losses (4). Also, our finding that comorbid PTSD symptoms intrusion and avoidance predicted a slow recovery and chronic grief are consistent with a recent meta-analysis (21). Horowitz stated that intrusions are common after sudden traumatic events, and that avoidance is a strategy that is used to ward off the painful emotions related to these intrusions (46). While avoidance can be adaptive in the early stages of a loss, if it is used as the main coping strategy to regulate grief it may hinder the bereaved from confronting the reality of their loved one's death or processing the loss (47). Intrusive thoughts or re-enactment fantasies of how their loved one was killed can interfere with positive reminiscing of the deceased (48). This suggest that PTSD symptoms may hinder or delay the resolution of grief. Recent studies have, however, found that an increase in grief levels can predict later PTSD levels and not vice versa (11). This suggest that PGD and PTSD may influence each other reciprocally, which requests a thorough assessment in clinical practice since it may have implications for the choice of treatment.

LIMITATIONS

The current study has several limitations. Due to confidentiality, the ethical committee did not permit inquiries about non-responders beyond the information of gender and age of the deceased. However, there were no statistically significant

TABLE 3 | Trajectory group (class) prediction based on generalized mixed multinomial regression analyses.

Class predictors	B	p	Exp (B)	95% CI	95% CI
				Low	High
High/decreasing					
- Gender	1.88	0.007	6.57	1.66	25.99
- Loss (child)	0.16	0.823	1.17	0.29	4.77
- Exposure	0.35	0.662	1.42	0.29	6.88
- Previous depressive symptoms	3.03	0.012	20.67	1.96	218.33
- Previous loss	1.25	0.072	3.50	0.89	13.72
- Satisfied with social support (at T1)	−0.39	0.119	0.68	0.41	1.11
- Received psychological help (at T1)	−0.50	0.557	0.61	0.12	3.20
Intrusion (at T1)	0.16	0.010	1.17	1.04	1.33
- Avoidance (at T1)	0.08	0.162	1.09	0.97	1.22
High/chronic					
- Gender	3.31	0.005	27.48	2.79	271.17
- Loss (child)	2.02	0.082	7.53	0.77	73.26
- Exposure	1.75	0.102	5.73	0.71	46.34
- Previous depressive symptoms	3.23	0.025	25.24	1.50	425.71
- Previous loss	0.31	0.762	1.36	0.19	9.92
- Satisfied with social support (at T1)	−0.47	0.165	0.62	0.32	1.21
- Received psychological help (at T1)	0.95	0.404	2.60	0.28	24.34
- Intrusion (at T1)	0.32	0.001	1.38	1.14	1.67
- Avoidance (at T1)	0.26	0.009	1.30	1.07	1.58

The Moderate level/improved class was set as reference category. Intercept values not presented ($n = 129$).

group differences between participants and non-participants concerning these demographic variables. Furthermore, the study suffer from low sample size. In the LCGA two classes had a relatively small number of predicted persons. The estimated relationships are a result of the included predictors, but also a result of the potential predictors not included in the model. Also, we used single non-validated questions in the assessment of previous depressive symptoms and psychosocial help after the attack. These measures may be subject to reporting bias. The use of ICG to tap grief symptoms does not capture the current symptoms or criteria for of PGD as defined in either DSM-5 or ICD-11 (1). Comparative studies show that it matters which criteria are used to define disturbed grief in terms of prevalence and predictive validity (49).

CLINICAL IMPLICATIONS

There are several implications of these findings. First of all, the high level of PGD and slow recovery characterizing the majority of bereaved suggest that an early out-reach in the follow-up of family members bereaved by terror is warranted (50). Use of adequate screening measures may be helpful in

reaching those who need help most. As studies have shown, persons with PGD may not seek adequate help due to different barriers, and not all will accept help in the early stages of the loss. A plan for long-time follow-up of the bereaved is also indicated. Some bereaved will need help first several months after their loss when there is a marked erosion of support from social network. Collective gatherings are highly valued among bereaved families were a combination of psychoeducation and grief groups are organized (51). Some bereaved also need specialized help for unrelenting PGD. Clinicians need to be familiar with assessment and treatment of both trauma-related disorders and different grief disorders. Several treatment models of traumatic bereavement are promising and have shown good effects (52, 53).

CONCLUSION

Terror-bereaved family members are vulnerable for a long-lasting and difficult grief process, comprising of both trauma symptoms and PGD. This suggest that there is a need for both early interventions and interventions capturing those who need long-term help. Also, mental health personnel working with terror-bereaved families should have be trained in both assessment of and treatment of trauma or PTSD and PGD.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Regional Committees for Medical and Health Research Ethics in Norway. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

PK: lead author of introduction, materials and methods section, and discussion. KD: project leader, commented on manuscript. RG conducted analysis, written statistical analysis section, results section, commented on manuscript. All authors contributed to the article and approved the submitted version.

FUNDING

The project was funded by Center for crisis psychology, Bergen, Norway and The Danish Egmont Foundation.

REFERENCES

1. Killikelly C, Maercker A. Prolonged grief disorder for ICD-11: the primacy of clinical utility and international applicability. *Eur J Psychotraumatol.* (2017) 8(Suppl 6):1476441. doi: 10.1080/20008198.2018.1476441
2. Kristensen P, Dyregrov K, Dyregrov A. What distinguishes prolonged grief disorder from depression? *J Norwegian*

- Med Association.* (2017) 137:538–9. doi: 10.4045/tidsskr.16.0629
3. Shear K, Simon N, Wall M, Zisook S, Neimeyer R, Duan N, et al. Complicated grief and related bereavement issues for DSM-5. *Depress Anxiety.* (2011) 28:103–17. doi: 10.1002/da.20780
 4. Prigerson HG, Horowitz MJ, Jacobs SC, Parkes CM, Aslan M, Goodkin K, et al. Prolonged grief disorder: psychometric validation of criteria proposed for DSM-V and ICD-11. *PLoS Med.* (2009) 6:e1000121. doi: 10.1371/journal.pmed.1000121
 5. Djelantik A, Smid GE, Mroz A, Kleber RJ, Boelen PA. The prevalence of prolonged grief disorder in bereaved individuals following unnatural losses: systematic review and meta regression analysis. *J Affect Disord.* (2020) 265:146–56. doi: 10.1016/j.jad.2020.01.034
 6. Neria Y, Gross R, Litz B, Maguen S, Insel B, Seirmarco G, et al. Prevalence and psychological correlates of complicated grief among bereaved adults 2.5–3.5 years after September 11th attacks. *J Traumat Stress.* (2007) 20:251–62. doi: 10.1002/jts.20223
 7. Dyregrov K, Dyregrov A, Kristensen P. Traumatic bereavement and terror: the psychosocial impact on parents and siblings 1 1/2 year after the 22.07.11 terror-killing in Norway. *J Loss Trauma.* (2014) 20:556–76. doi: 10.1080/15325024.2014.957603
 8. Pirard P, Motreff Y, Lavalette C, Vandentorren S, Baubet T, Messiah A. Mental health impact and health care use of the bereaved persons 8–11 months after the November 2015 Paris terror attacks. *Eur J Public Health.* (2018) 28:635. doi: 10.1093/eurpub/cky212.635
 9. Cozza SJ, Fisher JE, Fetchet MA, Chen S, Zhou J, Fullerton CS, et al. Patterns of comorbidity among bereaved family members 14 years after the September 11th, 2001, terrorist attacks. *J Trauma Stress.* (2019) 32:526–35. doi: 10.1002/jts.22407
 10. Armour M. Violent death: understanding the context of traumatic and stigmatized grief. *J Human Behavi Social Environment.* (2007) 14:53–90. doi: 10.1300/J137v14n04_04
 11. Djelantik AMJ, Smid GE, Kleber RJ, Boelen PAJCP. Do prolonged grief disorder symptoms predict post-traumatic stress disorder symptoms following bereavement? A cross-lagged analysis. *Compr Psychiatry.* (2018) 80:65–71. doi: 10.1016/j.comppsy.2017.09.001
 12. O'Connor M, Nickerson A, Aderka IM, Bryant RA. The temporal relationship between change in symptoms of prolonged grief and posttraumatic stress following old age spousal bereavement. *Depress Anxiety.* (2015) 32:335–40. doi: 10.1002/da.22349
 13. Kristensen P, Weisæth L, Hussain A, Heir T. Prevalence of psychiatric disorders and functional impairment after loss of a family member: a longitudinal study after the 2004 Tsunami. *Depress Anxiety.* (2015) 32:49–56. doi: 10.1002/da.22269
 14. Orcutt HK, Bonanno GA, Hannan SM, Miron LR. Prospective trajectories of posttraumatic stress in college women following a campus mass shooting. *J Trauma Stress.* (2014) 27:249–56. doi: 10.1002/jts.21914
 15. Svein J, Bergh Johannesson K, Cervin M, Arnberg FK. Trajectories of prolonged grief one to six years after a natural disaster. *PLoS ONE.* (2018) 13:e0209757. doi: 10.1371/journal.pone.0209757
 16. Lenferink L, Nickerson A, de Keijser J, Smid GE, Boelen PA. Trajectories of grief, depression, and posttraumatic stress in disaster-bereaved people. *Depress Anxiety.* (2020) 37:35–44. doi: 10.1002/da.22850
 17. Kristensen P, Weisæth L, Heir T. Predictors of complicated grief after a natural disaster: a population study two years after the 2004 South-East Asian tsunami. *Death Stud.* (2010) 34:137–50. doi: 10.1080/07481180903492455
 18. Johannesson KB, Lundin T, Hultman CM, Frojd T, Michel PO. Prolonged grief among traumatically bereaved relatives exposed and not exposed to a tsunami. *J Trauma Stress.* (2011) 24:456–64. doi: 10.1002/jts.20668
 19. Goldstein RD, Petty CR, Morris SE, Human M, Odendaal H, Elliott A, et al. Pre-loss personal factors and prolonged grief disorder in bereaved mothers. *Psychol Med.* (2019) 49:2370–8. doi: 10.1017/S0033291718003264
 20. Shear K. Clinical practice. Complicated grief. *N Engl J Med.* (2015) 372:153–60. doi: 10.1056/NEJMc1315618
 21. Heeke C, Kampisiou C, Niemeyer H, Knaevelsrud C. A systematic review and meta-analysis of correlates of prolonged grief disorder in adults exposed to violent loss. *Eur J Psychotraumatol.* (2019) 10:1583524. doi: 10.1080/2008198.2019.1583524
 22. Prigerson HG, Maciejewski PK, Reynolds CE, 3rd, Bierhals AJ, Newsom JT, Fasiczka A, et al. Inventory of Complicated Grief: a scale to measure maladaptive symptoms of loss. *Psychiatry Res.* (1995) 59:65–79. doi: 10.1016/0165-1781(95)02757-2
 23. Thimm JC, Davidsen M, Elsness MM, Vara H. Reliability, factor structure, and validity of the Inventory of Complicated Grief (ICG) in a general bereaved sample in Norway. *Scand Psychol.* (2019) 6:e7. doi: 10.15714/scandpsychol.6.e7
 24. Weiss DS, Marmar CB. The impact of event scale-revised. In: Wilson JP, Keane TM, editors. *Assessing Psychological Trauma and PTSD.* New York, NY: Guilford Press (1997). p. 399–411.
 25. Eid J, Larsson G, Johnsen BH, Laberg JC, Bartone PT, Carlstedt B. Psychometric properties of the Norwegian impact of event scale-revised in a non-clinical sample. *Nordic J Psychiatry.* (2009) 63:426–32. doi: 10.1080/08039480903118190
 26. Joseph S, Andrews B, Williams R, Yule W. Crisis support and psychiatric symptomatology in adult survivors of the Jupiter cruise ship disaster. *British J Clin Psychol.* (1992) 31(Pt 1):63–73. doi: 10.1111/j.2044-8260.1992.tb00968.x
 27. Johannesson KB, Arinell H, Arnberg FK. Six years after the wave. Trajectories of posttraumatic stress following a natural disaster. *J Anxiety Disord.* (2015) 36:15–24. doi: 10.1016/j.janxdis.2015.07.007
 28. IBM Corp. *IBM SPSS Statistics for Windows.* Version 23.0. Armonk, NY: IBM Corp. (2015).
 29. Newsom JT. *Longitudinal Structural Equation Modeling.* New York, NY: Routledge, Taylor & Francis Group (2015). p. 411.
 30. Muthén LK, Muthén BO. *Mplus 7.4.* Los Angeles, CA: Muthén & Muthén (2015).
 31. Wang J, Wang X. *Structural Equation Modeling: Applications Using Mplus.* West Sussex, UK: Wiley, A John Wiley & Sons, Ltd., Publication (2012). doi: 10.1002/9781118356258
 32. Kline RB. *Principles and Practice of Structural Equation Modeling.* 3 ed. New York, NY: The Guilford Press (2010). p. 425.
 33. Muthén LK, Muthén BO. *Mplus. Statistical Analysis With Latent Variables. User's Guide.* Version 7. 7 ed. Los Angeles, CA: Muthén & Muthén (2007).
 34. Nylund KL, Asparouhov T, Muthén BO. Deciding on the number of classes in latent class analysis and growth mixture modeling: a monte carlo simulation study. *Struct Equ Model.* (2007) 14:535–69. doi: 10.1080/10705510701575396
 35. Muthén LK, Muthén BO. *Mplus User's Guide.* Version 8. 7 ed. Los Angeles, CA: Muthén & Muthén (2007).
 36. Wickrama KAS, Lee TK, O'Neal CW, Lorenz FO. *Higher-Order Growth Curves and Mixture Modeling With Mplus.* New York, NY: Routledge (2016). p. 320.
 37. Muthén LK, Muthén BO. *Mplus 7.3.* Los Angeles, CA: Muthén & Muthén (1998).
 38. Bollen KA, Curran PJ. *Latent Curve Models: A Structural Equation Perspective.* Hoboken, NJ: Wiley-Interscience (2006).
 39. Infurna FJ, Luthar SS. Parents' adjustment following the death of their child: resilience is multidimensional and differs across outcomes examined. *J Res Pers.* (2017) 68:38–53. doi: 10.1016/j.jrp.2017.04.004
 40. Lobb EA, Kristjansson LJ, Aoun SM, Monterosso L, Halkett GK, Davies A. Predictors of complicated grief: a systematic review of empirical studies. *Death Stud.* (2010) 34:673–98. doi: 10.1080/07481187.2010.496686
 41. Fernández-Alcántara M, Zech E. One or multiple complicated grief (s)? The role of kinship on grief reactions. *Clin Psychol Sci.* (2017) 5:851–7. doi: 10.1177/2167702617707291
 42. Wijngaards-de Meij L, Stroebe M, Schut H, Stroebe W, van den Bout J, van der Heijden P, et al. Couples at risk following the death of their child: predictors of grief versus depression. *J Cons Clin Psychol.* (2005) 73:617–23. doi: 10.1037/0022-006X.73.4.617
 43. Aldwin DF. How good is survey measurement? Assessing the reliability and validity of survey measures. In: Marsden PV, Wright JV, editors. *Handbook of Survey Research.* Bingley, UK: Emerald Group Publishing (2010). p. 405–37.
 44. Grievink L, van der Velden PG, Yzermans CJ, Roorda J, Stellato RK. The importance of estimating selection bias on prevalence estimates shortly after a disaster. *Ann Epidemiol.* (2006) 16:782–8. doi: 10.1016/j.annepidem.2006.04.008

45. Stroebe M, Schut H. The dual process model of coping with bereavement: rationale and description. *Death Stud.* (1999) 23:197–224. doi: 10.1080/074811899201046
46. Horowitz MJ. Psychological processes induced by illness, injury, and loss. In: Green C, Meagher R, Million T, editors. *Handbook of Clinical Health Psychology*. New York, NY: Springer US; Plenum Press (1982). p. 53–67.
47. Dyregrov K, Dyregrov A, Kristensen P. In what ways do bereaved parents after terror go on with their lives, and what seems to inhibit or promote adaptation during their grieving process? A qualitative study. *Omega-J Death Dying.* (2016) 73:374–99. doi: 10.1177/0030222816653851
48. Layne CM, Kaplow JB, Oosterhoff B, Hill RM, S Pynoos R. The interplay between posttraumatic stress and grief reactions in traumatically bereaved adolescents: when trauma, bereavement, and adolescence converge. *Adolesc Psychiatry.* (2017) 7:266–85. doi: 10.2174/2210676608666180306162544
49. Lenferink LI, Boelen PA, Smid GE, Paap MC. The importance of harmonising diagnostic criteria sets for pathological grief. *Br J Psychiatry.* (2019) 216:1–4. doi: 10.1192/bjp.2019.240
50. Allsopp K, Brewin CR, Barrett A, Williams R, Hind D, Chitsabesan P, et al. Responding to mental health needs after terror attacks. *BMJ.* (2019) 366:l4828. doi: 10.1136/bmj.l4828
51. Dyregrov A, Dyregrov K, Straume M, Grønvold Bugge R. Weekend family gatherings for bereaved after the terror killings in Norway in 2011. *Scand Psychol.* (2014) 1:e8. doi: 10.15714/scandpsychol.1.e8
52. Lenferink L, de Keijser J, Smid GE, Boelen PA. Cognitive therapy and EMDR for reducing psychopathology in bereaved people after the MH17 plane crash: findings from a randomized controlled trial. *Traumatology.* (2020). doi: 10.1037/trm0000253
53. Pearlman LA, Wortman CB, Feuer CA, Farber CH, Rando TA. *Treating Traumatic Bereavement: A Practitioner's Guide*. New York, NY: Guilford Publications (2014).

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Copyright © 2020 Kristensen, Dyregrov and Gjestad. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.



Psychometric Evaluation of the Swedish Version of the Prolonged Grief Disorder-13 (PG-13) in a Bereaved Mixed Trauma Sample

Josefin Sveen^{1,2*}, Kristina Bondjers¹, Julia Heinsoo¹ and Filip K. Arnberg^{1,3}

¹ Department of Neuroscience, National Centre for Disaster Psychiatry, Psychiatry, Uppsala University, Uppsala, Sweden,

² Department of Health Care Sciences, Palliative Research Centre, Ersta Sköndal Bräcke University, Stockholm, Sweden,

³ Stress Research Institute, Stockholm University, Stockholm, Sweden

OPEN ACCESS

Edited by:

Clare Killikelly,
University of Zurich, Switzerland

Reviewed by:

Jos De Keijser,
University of Groningen, Netherlands
S. M. Yasir Arafat,
Enam Medical College, Bangladesh

*Correspondence:

Josefin Sveen
josefin.sveen@neuro.uu.se

Specialty section:

This article was submitted to
Mood and Anxiety Disorders,
a section of the journal
Frontiers in Psychiatry

Received: 10 March 2020

Accepted: 16 November 2020

Published: 03 December 2020

Citation:

Sveen J, Bondjers K, Heinsoo J and
Arnberg FK (2020) Psychometric
Evaluation of the Swedish Version of
the Prolonged Grief Disorder-13
(PG-13) in a Bereaved Mixed Trauma
Sample. *Front. Psychiatry* 11:541789.
doi: 10.3389/fpsy.2020.541789

Background: This study aimed to examine the psychometric properties of the Swedish PG-13 in a bereaved trauma exposed sample. A second aim was to examine the latent structure of prolonged grief using the PG-13.

Methods: The participants were adults ($n = 123$) taking part in an ongoing longitudinal study regarding the effects of potentially traumatic events. Participants had experienced a potentially traumatic event in the past 5 years and had reported a death of a significant other either as their primary traumatic event or in addition to another traumatic event. Assessment included self-report of prolonged grief, posttraumatic stress, and general psychological distress. Clinical interviews were used to assess depression, posttraumatic stress disorder, and disability level. The psychometric properties of the Swedish PG-13 were examined through reliability tests and assessment of associations with symptoms of posttraumatic stress, depression, general psychological distress, and disability level. Principal component analysis (PCA) and confirmatory factor analyses (CFA) were used to assess the latent structure.

Results: The internal consistency (Cronbach's $\alpha = 0.86$) and test-retest ($r = 0.86$) reliability were good. PCA suggested a three-factor model as descriptive of the latent structure of the instrument. Therefore, the CFA used this model, as well as two models suggested in the literature. The three-factor model had the best fit to data. Support of concurrent validity of PG-13 was shown by moderate positive associations with measures of posttraumatic stress, depression, and general psychological distress.

Conclusions: The Swedish PG-13 demonstrated good psychometric properties, and its use in research and practice to assess prolonged grief was supported. The factor analyses provided stronger support for models with two or three factors, as compared with a unidimensional model of prolonged grief, with the three-factor model having the best fit.

Keywords: prolonged grief, PG-13, loss, trauma, factor analysis, latent structure

INTRODUCTION

A significant minority of bereaved individuals experience persistent and severe symptoms of grief that do not diminish with time (1, 2). Chronically disabling and distressing grief reactions have, under various names, attracted research interest—most recently as Prolonged Grief Disorder (PGD). Prigerson et al. (2) proposed diagnostic criteria for PGD to be included in the 11th edition of Internal Classification of Diseases (ICD-11) and 5th version of Diagnostic and Statistical Manual of Mental Disorders [DSM-5; (3)]. However, in the DSM-5, a disorder called persistent complex bereavement disorder was included for further study (PCBD; 3), with symptoms divided into core symptoms (e.g., yearning, sorrow, emotional pain), reactive distress to the death (e.g., difficulty accepting the loss, bitterness related to the loss, avoidance of reminders of the loss), and social/identity disruption (e.g., difficulty trusting others, feeling life is empty without the deceased, confusion about one's role in life). The more recently released ICD-11 (4) includes Prolonged Grief Disorder as a disorder specifically associated with stress, differing from the PGD proposed by Prigerson et al. (2). PGD in ICD-11 is characterized by persistent and intense longing for the deceased, preoccupation with the deceased, additional symptoms suggestive of emotional pain, and a difficulty to accept the loss (5). These responses should be associated with functional impairment and persist for at least 6 months after the loss (6).

A meta-analysis by Lundorff et al. (7) found that one out of 10 bereaved adults, following a non-violent loss, is at risk of developing PGD, whereas a recent meta-analysis by Djelantik et al. (8) showed that almost half of the bereaved adults experienced PGD following unnatural losses. Previous research suggests that while PGD shares features with both depression and post-traumatic stress disorder (PTSD), it also provides unique explanatory power (2, 9–11) and is independently related to decreased mental and physical health (12–16). However, uncertainty remains regarding PGD in people with a history of traumatic events. For example, it is uncertain if the qualities of PGD mentioned above are found also among bereaved individuals who report a traumatic index event not related to bereavement.

Prigerson et al. (2) created a diagnostic self-rating instrument for their proposed criteria for PGD, the Prolonged Grief questionnaire-13 (PG-13). However, there are few studies examining the psychometric properties of the PG-13. One study has shown that the PG-13 has good psychometric properties with high sensitivity and specificity (93.3 and 98.3%) for bereaved adults (17). The Swedish PG-13 has shown satisfactory psychometric properties in a sample of cancer-bereaved parents, with the latent structure of PG-13 being explained by a unidimensional structure (18). In this study, the Swedish PG-13 was slightly adopted to bereaved parents by changing the wording “the person you lost” to “the child you lost.”

There are several studies examining the latent structure of prolonged grief using different measures, such as the Inventory of Complicated Grief and the revised version of Inventory of Complicated Grief. Most studies have found support for a unidimensional structure of prolonged grief (2, 9, 10, 19, 20).

In a sample of older adults, O'Connor et al. (21) examined the one-factor model, as well as a theoretically derived two-factor model (i.e., separation distress and traumatic distress), and found that the two-factor model had a better fit. However, the two factors were highly correlated and the one-factor model had an acceptable fit. Newson et al. (22) found four factors in a sample of older adults.

As the psychometric properties of the Swedish PG-13 has only been examined with a slightly modified version in bereaved parents, additional validation should be conducted with more heterogeneous populations. This study aimed to examine the psychometrics of the Swedish PG-13 in trauma exposed sample, who had also experienced bereavement. Internal consistency and test-retest reliability were evaluated, as was the concurrent validity of the PG-13 in relation to posttraumatic stress disorder, depression, and general psychological distress. A second aim was to examine the latent structure of prolonged grief using the PG-13.

MATERIALS AND METHODS

Participants and Procedure

The participants in the current study were individuals taking part in an ongoing longitudinal study regarding the effects of potentially traumatic events—the Traumatic Events in a Longitudinal Survey (TRACES) study—at Uppsala University, Sweden. TRACES aims to examine psychological reactions to adverse events among health care and non-health care seeking individuals and how such reactions fluctuate over time.

Participants were self-recruited from the general public in two urban counties in Sweden via advertisements in local and social media, flyers at primary care and outpatient psychiatric care, as well as via information from health-care providers. Participants signed up for the study via health care staff, who provided the research group with contact information, or via an online form. Inclusion criteria for the longitudinal study were: being ≥ 18 years of age, having experienced a potentially traumatic event in the last 5 years, and being able to understand and speak Swedish. Exclusion criteria were ongoing traumatic event or stressors (e.g., ongoing violence, disease), and concurrent severe mental illness (e.g., ongoing psychosis). Participants were compensated with two movie tickets. For the current study, participants who reported bereavement of a significant other (i.e., a family member, close relative, or close friend) were drawn from the longitudinal study. No criteria were issued on the relationship to the deceased.

All participants were screened via telephone with questions about the nature of their potentially traumatic events and additional questions related to the inclusion and exclusion criteria. Participants then responded to a survey, including several questionnaires, either via a secure online web portal or on paper. After the survey was filled out, the participants took part in structured face-to-face interviews conducted by licensed psychologists, psychologists in training, or master's students in clinical psychology under the supervision of licensed psychologists. All interviewers were trained in the interview protocols by the researchers. The interview sessions encompassed

assessment of the participants' history of potentially traumatic events, and structured clinical interviews, as detailed below. During the interview, participants who reported having experienced a loss of a significant other filled in the Swedish version of PG-13. After their interview, each participant was given a retest questionnaire to complete the following day, at the earliest, and return by post.

Measurements

PG-13

The PG-13 is comprised of 13 items which follow the diagnostic criteria suggested by Prigerson et al. (2). The questionnaire assesses symptoms of separation distress and symptoms of cognitive, behavioral, and emotional changes, as well as the duration of these symptoms and perceived functional impairment. The first four items concern symptoms—yearning, intense grief, avoidance, and numbness—during the preceding month, rated on a five-point frequency scale (*Not at all* to *Several times a day*), with the fifth item establishing whether yearning or intense grief have been present at least daily beyond 6 months after the loss (*No/Yes*). The next seven items concern confusion with one's role in life, difficulties accepting the loss, loss of trust, bitterness, difficulties moving on in life, emotional isolation, and meaninglessness. These items are rated on a five-point intensity scale (*Not at all* to *Overwhelmingly*). Lastly, one item asks about deterioration in everyday function (*No/Yes*). A total symptom severity score is computed by giving the 11 symptom items (items 1, 2, 4–12) a score of 1 to 5 and summing up these scores. Thus, symptom severity ranges from 11 to 55. A dichotomous scoring for PGD screening is also used. For a positive screening, the respondent must endorse yearning or intense grief at least once a day (items 1–2), at least 6 months after the loss (item 3), experience at least five of the remaining symptoms at least “daily” (items 4–5) or “a lot” (items 6–12), and report functional impairment (item 13). The PG-13 has previously been translated into Swedish at the National Center of Disaster Psychiatry at Uppsala University. The original PG-13 was translated into Swedish by a professional translator and then revised by lic. clin. psychologists with extensive experience of grief. A second professional translator, a native English speaker with a PhD in psychology, did a back-translation of the final Swedish version. This person was blind to the English original. The psychometric properties of the Swedish version of PG-13 has been examined in cancer-bereaved parents (18).

Clinician-Administered PTSD Scale for DMS-5 (CAPS-5)

The CAPS-5 (23) is a structured clinical interview considered to be the gold standard for diagnostic assessment of PTSD. The interview assesses PTSD symptoms, as described in DSM-5, within the preceding month. It contains 20 symptom items, each assessed on a five-point Likert scale from 0 (*Not present*) to 4 (*Extreme*). The total symptom severity ranges from 0 to 80. CAPS-5 also assesses functional disability due to PTSD symptoms and the duration of the disorder.

The PTSD Checklist for DSM-5 (PCL-5)

The PCL-5 (24) was used as a self-report measure of PTSD symptoms during the preceding month. It contains 20 items, each symptom corresponding to a symptom in DSM-5. Items ask how much respondents have been bothered by each symptom during the preceding month, and are answered on a five-point Likert scale from 0 (*Not at all*) to 4 (*Extremely*). The total symptom score ranges from 0 to 80. The Swedish version of PCL-5 has been shown to have good psychometric properties (25).

The Symptom Checklist 27 (SCL-27)

The SCL-27 (26) was used as a measure of general psychological distress. It is a self-report scale and measures symptoms on six subscales: depressive, dysthymic, somatization, mistrust, social phobic, and agoraphobic symptoms. Each subscale includes four to six items. All items are answered on a four-point Likert scale and the total severity score ranges from 0 to 108. In this study, both the subscales and the total score were used in the analyses of the concurrent validity of the PG-13.

MINI Neuropsychiatric Interview (MINI 7.0)

MINI 7.0 (27, 28) is a screening interview for several psychiatric disorders. The data from the depression module, ongoing and previous depression, were used in the current study. The number of depression criteria fulfilled was used to assess the level of depression symptoms; it ranges from 0 to 9.

World Health Organization Disability Assessment Scale (WHODAS 2.0)

WHODAS 2.0 is a structured interview assessing a person's disability levels in several domains. Items are scored from 0 (*No difficulties*) to 4 (*Extreme difficulty/cannot do*). The current study employed the 12+24 version, where the interview is discontinued if the interviewee responds negatively to the first six to 12 items, and the remaining items are scored 0. If a respondent reports disability in any domain, additional domain-specific items are given and scored. The complex scoring method suggested by WHO was used, which provides a total score ranging from 0 (*No disability*) to 100 (*Full disability*). WHODAS 2.0 has demonstrated excellent psychometric properties (29).

Statistical Analyses

The statistical analyses were performed using IBM SPSS Statistics 26. The reliability was examined using Cronbach's alpha, mean inter-item correlation, and the test-retest correlation coefficient. The reliability analyses were calculated using the PG-13 symptom severity score.

Concurrent validity was assessed by examining associations between PG-13 and measures of PTSD (PCL-5 and CAPS-5), depression (MINI), and general psychological distress (SCL-27). Pearson's *r* was calculated for both concurrent validity and test-retest reliability.

The factor analyses included an exploratory and a confirmatory analysis. First, a principal component analysis

TABLE 1 | Mean (SD) scores on symptoms of prolonged grief, posttraumatic stress, depression and general psychological distress, disability level, and comparison between the two subsamples.

	All (<i>n</i> = 123)	Index event		T-score ^a
		Bereavement (<i>n</i> = 72)	Comparison (<i>n</i> = 51)	
PG-13	24.98 (8.47)	26.56 (8.35)	22.75 (8.23)	2.51*
PCL-5 ^b	24.68 (17.46)	22.81 (16.01)	27.53 (19.30)	−1.42
CAPS-5	18.39 (12.28)	16.18 (10.34)	21.51 (14.11)	−2.30*
	Intrusion	4.76 (3.89)	5.92 (4.56)	−2.70**
	Avoidance	2.10 (2.02)	2.31 (2.14)	−1.00
	NACM	6.23 (4.80)	7.12 (5.42)	−1.74
	Hyperarousal	5.31 (3.54)	6.16 (3.88)	−2.20*
Depression ^c	Ongoing	1.42 (2.73)	1.66 (3.14)	−0.76
	Previous	5.34 (3.21)	5.81 (3.09)	−1.30
SCL-27 ^b	27.72 (21.40)	25.62 (21.35)	30.93 (21.48)	−1.30
	Depressive	5.16 (3.60)	5.60 (3.72)	−1.06
	Dysthymic	6.48 (4.56)	6.87 (4.10)	−0.73
	Somatization	5.64 (6.03)	6.00 (6.06)	−1.12
	Agoraphobic	3.32 (4.42)	4.36 (5.36)	−1.88
	Social phobic	3.76 (3.87)	3.89 (3.99)	−0.28
	Mistrust	3.63 (3.64)	4.22 (3.85)	−1.30
WHODAS 2.0 ^d	15.98 (17.35)	13.15 (15.71)	20.23 (18.94)	−2.18*

Means and standard deviation for all scales used in the study. Means presented for all participants, participants with bereavement as index event and participants with other index events (i.e., comparison group). PG-13, Prolonged Grief questionnaire-13; PCL-5, PTSD Checklist for DSM-5; CAPS-5, Clinician-Administered PTSD Scale for DMS-5; SCL-27, Symptom Checklist 27; WHODAS, World Health Organization Disability Assessment Scale.

^aT-test of the difference of means between participants with bereavement as index event and participants with other index events.

^bMissing data, *n* = 114 (69 and 45).

^cMissing data, *n* = 118 (71 and 47).

^dMissing data, *n* = 113 (69 and 44).

p* < 0.05, *p* < 0.01.

(PCA) with oblique rotation was performed. The number of factors was determined by an evaluation of the scree plot and the eigenvalues. Confirmatory factor analyses were conducted using Mplus version 8 (30). Given the ordinal nature of the item response options, a robust weighted least-squares with a mean and variance adjustment (WLSMV) estimator was employed (31–33). Goodness of model fit was assessed with the χ^2 test, the comparative fit index (CFI), the Tucker-Lewis Index (TLI), the root mean square error of approximation (RMSEA), and weighted root-mean-square residual (WRMR). The following standards were used in assessing model fit: a non-significant test of model χ^2 , χ^2 /df between 2 and 3, CFI and TLI > 0.90, RMSEA < 0.08 (34), and WRMR < 1.0 (35, 36).

T-tests were conducted to explore whether there was a difference in symptoms between individuals who reported bereavement as the traumatic index event and bereaved individuals who reported a traumatic index event not related to bereavement. To compare correlation coefficients, Fisher's *z*-tests were conducted. For participants missing one or two items on PG-13 (*n* = 3), the missing values were imputed using the estimated mean values on PG-13. The same procedure was used for missing data on SCL-27 (*n* = 15). Participants with more than 50% missing values on PG-13 were excluded from the study (*n* = 3).

RESULTS

Sample Characteristics

In total, 123 participants were included in the study. A majority were women (80.5%) and the mean age was 37.85 years (SD = 14.97, range = 18–78). In all, 52.8% were working (*n* = 65), 23.6% (*n* = 29) were studying, and 65.8% (*n* = 81) had a high level of educational attainment (attended or finished university). Of those reporting that the loss was not the trauma index event (*n* = 51), 13 reported the event to be sexual violence, 12 reported somatic disease/injury, 10 reported accident, 10 individuals reported interpersonal violence, and 6 reported another traumatic event. The total sample mean score on PG-13 was 24.98 (SD = 8.48). Four participants reported no symptoms and only three participants screened positive for PGD. See **Table 1** for descriptive statistics on all measures.

Reliability

The internal consistency of the PG-13 was good, Cronbach's α = 0.86, and it was not improved by removal of any of the PG-13 items. The item-total correlations of the PG-13 items with the total score ranged from r_{itc} = 0.42 (item 9) to r_{itc} = 0.63 (item 11). The mean item-total correlation was r_{itc} =

TABLE 2 | PG-13 correlations with symptoms of posttraumatic stress, depression, general psychological distress, and disability level, and comparison between the two subsamples.

	All (<i>n</i> = 123)	Index event		<i>z</i> -value ^a
		Bereavement (<i>n</i> = 72)	Comparison (<i>n</i> = 51)	
PCL-5 ^b	0.52***	0.61***	0.42**	1.39**
CAPS-5	0.48***	0.79***	0.32*	3.94***
Intrusion	0.33***	0.63***	0.20	2.87**
Avoidance	0.49***	0.63***	0.39**	1.75*
NACM	0.48***	0.73***	0.31*	3.24**
Hyperarousal	0.38***	0.58***	0.29*	1.94*
Depression ^c	0.27**	0.44**	0.13	1.77*
Ongoing	0.27**	0.44**	0.13	1.77*
Previous	0.03	0.25*	−0.25	2.64**
SCL-27 ^b	0.56***	0.71***	0.46**	1.98*
Depressive	0.54***	0.65***	0.49**	1.21
Dysthymic	0.52***	0.65***	0.38*	1.90*
Somatization	0.42***	0.57***	0.29	1.77*
Agoraphobic	0.46***	0.63***	0.45**	1.30
Social	0.49***	0.60***	0.37*	1.54
Mistrust	0.40***	0.56***	0.35*	1.36
WHODAS 2.0 ^d	0.43***	0.62***	0.27	2.29*

Correlations between PG-13 mean score and mean score on the other scales for all participants, participants with bereavement as index event and participants with other index events (i.e., comparison group). PG-13, Prolonged Grief questionnaire-13; PCL-5, PTSD Checklist for DSM-5; CAPS-5, Clinician-Administered PTSD Scale for DMS-5; SCL-27, Symptom Checklist 27; WHODAS, World Health Organization Disability Assessment Scale.

^aComparison of correlations (*Z*-test) between participants with bereavement and participants with other index events.

^bMissing data, *n* = 114 (69 and 45).

^cMissing data, *n* = 113 (71 and 47).

^dMissing data, *n* = 115 (69 and 46).

**p* < 0.05.

***p* < 0.01.

****p* < 0.001.

0.55. The mean inter-item correlation was $r = 0.36$, range $r = 0.12$ – 0.75 . The mean inter-item correlation was lowest between “yearning” (item 1) and “trust in others” (item 8), $r = 0.12$, and highest between the two separation distress items (items 1 and 2), $r = 0.75$.

A total of 84 participants completed and returned the retest questionnaire. The temporal stability of PG-13 was good, with high correlations for the total symptom score $r = 0.86$ ($p < 0.001$). The correlations ranged from $r = 0.65$ ($p < 0.001$) to $r = 0.81$ ($p < 0.001$) at the item level. The mean time for retest was 6.26 days (SD = 9.41 days, range = 0–50 days).

Concurrent Validity

The correlations between PG-13 and the other measures of distress and function are shown in Table 2. In the total sample, there were low to moderate correlations between the PG-13 and the measure of related disorder. The strongest correlation was with the SCL-27 total score and the second strongest was with PCL-5.

Construct Validity

Principal Component Analysis

The factor analysis rendered a significant Bartlett’s test of sphericity ($p < 0.001$), the sampling adequacy was 0.83 (Kaiser-Meyer-Olkin), and 67.7% of the total variance was explained with three factors (Table 3). The first factor, labeled social/identity disruption, included four items and explained 42.9% of the variance. The second factor, labeled separation distress, included four items and explained 15.1% of the variance. The third factor, labeled traumatic distress, included three items and explained 9.8% of the variance (Table 3).

Confirmatory Factor Analysis

Based on the principal content analysis suggesting three factors, the theoretically derived two-factor model (37) and previous research supporting a one-factor model for prolonged grief (2, 9, 10, 18–20), three models with 1, 2, and 3 factors, respectively, were tested. The two- and three-factor models yielded acceptable model fit; however, the three-factor model performed slightly better (Table 4 presents model fit indices).

Between-Group Comparison

The bereavement group had significantly higher scores on PG-13 than the comparison group (Table 1). The total scores for PCL-5, CAPS-5, and WHODAS were significantly higher for the comparison group than for the bereavement group, whereas there were no significant differences between groups on SCL-27 or depression (Table 1). As noted in Table 2, the correlations in the bereavement group were stronger than in the comparison group for the majority of the measures, excepting only some of the subscales of SCL-27. In the comparison group, the correlations were non-significant between PG-13 and the PCL-5 intrusion subscale and the SCL-27 somatization subscale. This was also the case for both the interview-based depression score and the functional disability score. In contrast, in the bereavement group, all correlations for PG-13 were significant, except that with previous depression.

DISCUSSION

This study examined the psychometric properties of the PG-13 and the latent structure of prolonged grief in a Swedish sample. The Swedish PG-13 possessed adequate internal consistency and mean inter-item correlations as well as temporal stability at both the scale and item levels. Support of concurrent validity of PG-13 was shown by significant positive correlations with measures of posttraumatic stress, depression, and general psychological distress. This is consistent with there being overlap in symptoms between prolonged grief, depression, and posttraumatic stress, which are known to correlate to some extent (18, 38). Nevertheless, PGD, depression, and PTSD are distinct clinical entities (21).

The factor analyses provided stronger support for models with two or three factors, as compared to a unidimensional model of prolonged grief. The two-factor model yielded slightly poorer fit than the three-factor model. The two-factor model encompassed a separation distress factor, including yearning, which is one

TABLE 3 | Principal component analysis with oblique rotation of the PG-13.

Items	Factor loadings		
	Factor 1: Reorientation/identity	Factor 2: Separation distress	Factor 3: Traumatic distress
1. In the past month, how often have you felt yourself longing or yearning for the person you lost?		−0.85	
2. In the past month, how often have you had intense feelings of emotional pain, sorrow, or pangs of grief related to the lost relationship?		−0.89	
4. In the past month, how often have you tried to avoid reminders that the person you lost is gone?		−0.79	
5. In the past month, how often have you felt stunned, shocked, or dazed by your loss?		−0.73	
6. Do you feel confused about your role in life or feel like you don't know who you are?	0.83		
7. Have you had trouble accepting the loss?			0.52
8. Has it been hard for you to trust others since your loss?			0.64
9. Do you feel bitter over your loss?			0.87
10. Do you feel that moving on (e.g., making new friends, pursuing new interests) would be difficult for you now?	0.75		
11. Do you feel emotionally numb since your loss?	0.74		
12. Do you feel that life is unfulfilling, empty, or meaningless since your loss?	0.78		
Eigenvalues	4.7	1.7	1.1
Explained variance (%)	42.9	15.1	9.8

TABLE 4 | Fit indices for the confirmatory factor analyses of the PG-13.

Model	χ^2	df	CFI	TLI	RMSEA [90% CI]	WRMR
One factor	193.60***	44	0.89	0.87	0.17 [0.14; 0.19]	1.32
Two factors	81.69***	43	0.97	0.97	0.09 [0.06; 0.11]	0.75
Three factors	69.62**	41	0.98	0.97	0.08 [0.04; 0.11]	0.66

** $p < 0.01$.*** $p < 0.001$.

Df, degrees of freedom; CFI, Comparative fit index; TLI, Tucker-Lewis Index; RMSEA, Root mean square error of approximation; WRMR, Weighted root-mean-square residual.

of the core symptoms of prolonged grief, and a second factor including items that could be understood as manifestations of traumatic distress and items of social/identity disruption, such as confusion about finding a new role in life, difficulty to move on, and feeling that life is empty after the loss. In the three-factor model, the factors were social/identity disruption, separation distress, and traumatic distress. These findings may reflect the sample composition and they diverge from previous studies supporting a unidimensional structure of PG-13 (2, 18) and studies on other measures of prolonged grief examining the underlying structure of prolonged grief (2, 9, 10, 19, 20). However, the study by O'Connor et al. (37) found support for the theoretically derived two-factor model, in agreement with the present study. A recent study on a Hebrew version of Inventory of Complicated Grief among older bereaved parents found support for a three-factor structure (39). It should be noted that the fit of the three-factor model in the present study may be positively biased because the fit was assessed on the same sample that was used in the exploratory factor analysis. The possible positive bias for the fit of the three-factor model and the small differences in

model fit between the two-factor and three-factor models speak in favor of the two-factor model.

To the authors' knowledge, this is one of few studies examining differences in symptom severity of PTSD and other psychiatric illnesses between bereaved individuals with and without loss as the primary index event. Individuals who reported bereavement as their index event had higher levels of grief, and lower symptoms of PTSD and functional disability as compared to individuals that reported a different event than the loss as an index-event. In addition, there were overall lower associations between grief and the other measures in the comparison group. This suggested that prolonged grief symptoms were more strongly associated with symptoms of PTSD and depression when they arose from the same event. It also suggested that PTSD symptoms were slightly more pronounced and functional disability was markedly higher if the traumatic index event was not the same as the precipitating event for the PGD symptoms. Although these findings are tentative, they provide an interesting picture of the dynamics between prolonged grief symptoms and other forms of distress

and functional disability related to the type of traumatic index event.

Some limitations of this study warrant mention. Low overall symptom scores may have caused inflated correlations. Only three participants screened positive for PGD, a rate that is lower than in previous studies (7, 8, 22, 40). Additionally, the TRACES study was not primarily aimed toward bereavement and thus no additional questions were asked about the nature of the relationship between the participant and the deceased other than if it was a significant other. Another limitation is that convergent validity was not investigated. The small sample size limits the generalizability of the findings, which need to be replicated in larger samples with higher overall symptom burden. However, the findings are likely reflective of the wide range of reactions that may arise after experiencing loss, with the majority of those affected recovering well and a minority developing long-term distress. Finally, although a factor model should be validated through exploratory analysis on an independent sample, the sample size precluded such independent validation in this study. The model fit of the three-factor model should be viewed in light of this. This does not impact the advantage of the two-factor model over the unidimensional model.

In conclusion, the study supports the use of the Swedish PG-13 to measure prolonged grief in bereaved adults. The Swedish PG-13 was shown to have good reliability, including temporal stability, and findings supported construct and concurrent validity. The latent structure of prolonged grief suggested two or three factors, and not a unidimensional structure, which has been suggested previously. Overall, the PG-13 is a useful instrument in research, to increase the understanding of prolonged grief in adults. Although, due to the limitations of the study, including

the small sample size and not assessing convergent validity the results should be interpreted with caution. Future research is needed in clinical samples to confirm the evidence for validity of the Swedish PG-13.

DATA AVAILABILITY STATEMENT

The datasets generated for this study are available on request to the corresponding author.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Uppsala Regional Ethics committee. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

JS, KB, FA, and JH contributed to conception and design of the study. KB organized the database. JS performed the statistical analyses. JH and JS wrote the first draft of the manuscript. KB and FA wrote sections of the manuscript. All authors contributed to manuscript revision and read and approved the submitted version.

FUNDING

This study was funded by Socialstyrelsen (SE) and Uppsala University Hospital, Uppsala, Sweden.

REFERENCES

- Bonanno GA, Kaltman S. The varieties of grief experience. *Clin Psychol Rev.* (2001) 21:705–34. doi: 10.1016/S0272-7358(00)00062-3
- Prigerson HG, Horowitz MJ, Jacobs SC, Parkes CM, Aslan M, Goodkin K, et al. Prolonged grief disorder: psychometric validation of criteria proposed for DSM-V and ICD-11. *PLoS Med.* (2009) 6:e1000121. doi: 10.1371/journal.pmed.1000121
- American Psychiatric Association. *DSM-5, Diagnostic and Statistical Manual of Mental Disorders. 5th ed.* Washington, DC: American Psychiatric Association Press (2013). doi: 10.1176/appi.books.9780890425596
- World Health Organization. *The International Classification of Diseases and Related Health Problems version 11, ICD-11.* (2018). Available online at: <https://icd.who.int/browse11/l-m/en#/http://id.who.int/icd/entity/1183832314> (accessed November 23, 2020).
- Killikelly C, Maercker A. Prolonged grief disorder for ICD-11: the primacy of clinical utility and international applicability. *Eur J Psychotraumatol.* (2017) 8(Suppl. 6):1476441. doi: 10.1080/20008198.2018.1476441
- Maercker A, Brewin CR, Bryant RA, Cloitre M, Reed GM, van Ommeren M, et al. Proposals for mental disorders specifically associated with stress in the International Classification of Diseases-11. *Lancet.* (2013) 381:1683–5. doi: 10.1016/S0140-6736(12)62191-6
- Lundorff M, Holmgren H, Zachariae R, Farver-Vestergaard I, O'Connor M. Prevalence of prolonged grief disorder in adult bereavement: a systematic review and meta-analysis. *J Affect Disord.* (2017) 212:138–49. doi: 10.1016/j.jad.2017.01.030
- Djelantik AAAM, Smid G, Mroz A, Kleber RJ, Boelen P. The prevalence of Prolonged Grief Disorder in bereaved individuals following unnatural losses: Systematic review and meta regression analysis. *J Affect Disord.* (2020) 265:146–56. doi: 10.1016/j.jad.2020.01.034
- Prigerson HG, Frank E, Kasl SV, Reynolds CF III, Anderson B, Zubenko GS, et al. Complicated grief and bereavement-related depression as distinct disorders: preliminary empirical validation in elderly bereaved spouses. *Am J Psychiatry.* (1995) 152:22–30. doi: 10.1176/ajp.152.1.22
- Boelen PA, van den Bout J, de Keijser J. Traumatic grief as a disorder distinct from bereavement-related depression and anxiety: a replication study with bereaved mental health care patients. *Am J Psychiatry.* (2003) 160:1339–41. doi: 10.1176/appi.ajp.160.7.1339
- Golden AM, Dalgleish T. Is prolonged grief distinct from bereavement-related posttraumatic stress? *Psychiatry Res.* (2010) 178:336–41. doi: 10.1016/j.psychres.2009.08.021
- Prigerson HG, Bierhals AJ, Kasl SV, Reynolds CF III, Shear MK, Day N, et al. Traumatic grief as a risk factor for mental and physical morbidity. *Am J Psychiatry.* (1997) 154:616–23. doi: 10.1176/ajp.154.5.616
- Prigerson HG, Maciejewski PK. A call for sound empirical testing and evaluation of criteria for Complicated Grief proposed for DSM-V. *Omega.* (2005) 52:9–19. doi: 10.2190/ANKH-BB2H-D52N-X99Y
- Boelen PA, Prigerson HG. The influence of symptoms of prolonged grief disorder, depression, and anxiety on quality of life among bereaved adults: a prospective study. *Eur Arch Psychiatry Clin Neurosci.* (2007) 257:444–52. doi: 10.1007/s00406-007-0744-0

15. Boelen PA, van den Bout J. Complicated Grief and uncomplicated grief are distinguishable constructs. *Psychiatry Res.* (2008) 157:311–4. doi: 10.1016/j.psychres.2007.05.013
16. Lichtenthal WG, Nilsson M, Kissane DW, Breitbart W, Kacel E, Jones EC, et al. Underutilization of mental health services among bereaved caregivers with prolonged grief disorder. *Psychiatr Serv.* (2011) 62:1225–9. doi: 10.1176/ps.62.10.pss6210_1225
17. Maciejewski PK, Maercker A, Boelen PA, Prigerson HG. “Prolonged grief disorder” and “persistent complex bereavement disorder”, but not “complicated grief”, are one and the same diagnostic entity: an analysis of data from the Yale Bereavement Study. *World Psychiatry.* (2016) 15:266–75. doi: 10.1002/wps.20348
18. Pohlkamp L, Kreicbergs U, Prigerson HG, Sveen J. Psychometric properties of the Prolonged Grief Disorder-13 (PG-13) in bereaved Swedish parents. *Psychiatry Res.* (2018) 267:560–5. doi: 10.1016/j.psychres.2018.06.004
19. Prigerson HG, Maciejewski PK, Reynolds CF III, Bierhals AJ, Newsom JT, Fasiczka A, et al. (1995). Inventory of Complicated Grief: a scale to measure maladaptive symptoms of loss. *Psychiatry Res.* 59:65–79. doi: 10.1016/0165-1781(95)02757-2
20. Han DH, Lee JJ, Moon DS, Cha MJ, Kim MA, Min S, et al. Korean Version of Inventory of Complicated Grief Scale: psychometric properties in Korean adolescents. *J Korean Med Sci.* (2016) 31:114–9. doi: 10.3346/jkms.2016.31.1.114
21. Boelen PA, van de Schoot R, van den Hout MA, de Keijser J, van den Bout J. Prolonged Grief Disorder, depression, and posttraumatic stress disorder are distinguishable syndromes. *J Affect Disord.* (2010) 125: 374–8. doi: 10.1016/j.jad.2010.01.076
22. Newson RS, Boelen PA, Hek K, Hofman A, Tiemeier H. The prevalence and characteristics of complicated grief in older adults. *J Affect Disord.* (2011) 132:231–8. doi: 10.1016/j.jad.2011.02.021
23. Weathers FW, Blake DD, Schnurr PP, Kaloupek DG, Marx BP, Keane TM. *The Clinician-Administered PTSD Scale for DSM-5 (CAPS-5)* (2013). Retrieved from: www.ptsd.va.gov (accessed November 23, 2020).
24. Weathers FW, Litz BT, Keane TM, Palmieri PA, Marx BP, Schnurr PP. *The PTSD Checklist for DSM-5 (PCL-5)* (2013). Retrieved from: www.ptsd.va.gov (accessed November 23, 2020).
25. Sveen J, Bondjers K, Willebrand M. Psychometric properties of the PTSD Checklist for DSM-5: a pilot study. *Eur J Psychotraumatol.* (2016) 7:30165. doi: 10.3402/ejpt.v7.30165
26. Hardt J, Gerbershagen HU. Cross-validation of the SCL-27: a short psychometric screening instrument for chronic pain patients. *Eur J Pain.* (2001) 5:187–97. doi: 10.1053/eujp.2001.0231
27. Lecrubier Y, Sheehan DV, Weiller E, Amorim P, Bonora I, Harnett Sheehan K, et al. The Mini International Neuropsychiatric Interview (MINI). A short diagnostic structured interview: reliability and validity according to the CIDI. *Eur Psychiatry.* (1997) 12:224–31. doi: 10.1016/S0924-9338(97)83296-8
28. Sheehan DV, Lecrubier Y, Sheehan KH, Janavs J, Weiller E, Keskiner A, et al. The validity of the Mini International Neuropsychiatric Interview (MINI) according to the SCID-P and its reliability. *Eur Psychiatry.* (1997) 12:232–41. doi: 10.1016/S0924-9338(97)83297-X
29. Üstüün TB, Kostanjsek N, Chatterji S, Rehm J, World Health Organization (editors). *Measuring health and disability: manual for WHO Disability Assessment Schedule (WHODAS 2.0)* (2010). Available online at: <https://apps.who.int/iris/handle/10665/43974>
30. Muthen B, Muthen L. “Mplus: The Comprehensive Modeling Program for Applied Researchers”. Los Angeles, CA: Muthén (2002).
31. Muthen L, Muthen B. “Mplus User’s Guide.”. Los Angeles, CA: Muthén (1998–2017).
32. Beauducel A, Herzberg PY. On the performance of maximum likelihood versus means and variance adjusted weighted least squares estimation in CFA. *Struct Equ Model.* (2006) 13:186–203. doi: 10.1207/s15328007sem1302_2
33. Li CH. Confirmatory factor analysis with ordinal data: Comparing robust maximum likelihood and diagonally weighted least squares. *Behav Res Methods.* (2016) 48:936–49. doi: 10.3758/s13428-015-0619-7
34. Kline RB. *Methodology in the Social Sciences. Principles and Practice of Structural Equation Modeling.* New York, NY: Guilford Press (2016).
35. Yu CY. *Evaluating cutoff criteria of model fit indices for latent variable models with binary and continuous outcomes.* Doctoral thesis, Los Angeles, University of California Los Angeles (2002).
36. DiStefano C, Liu J, Jiang N, Shi D. Examination of the weighted root mean square residual: evidence for trustworthiness? *Struct Equ Model.* (2018) 25:453–66. doi: 10.1080/10705511.2017.1390394
37. O’Connor M, Lasgaard M, Shevlin M, Guldin MB. A confirmatory factor analysis of combined models of the harvard trauma questionnaire and the inventory of complicated grief-revised: are we measuring Complicated Grief or posttraumatic stress? *J Affect Disord.* (2010) 24:672–9. doi: 10.1016/j.janxdis.2010.04.009
38. He L, Tang S, Yu W, Xu W, Xie Q, Wang J. The prevalence, comorbidity and risks of prolonged grief disorder among bereaved Chinese adults. *Psychiatry Res.* (2014) 219:347–52. doi: 10.1016/j.psychres.2014.05.022
39. Lifshitz R, Ifrah K, Markovitz N, Bluvstein I, Shmotkin D. Inventory of complicated grief: factor structure among middle-aged and older bereaved parents. *Death Stud.* 1–8. doi: 10.1080/07481187.2020.1725931
40. Kersting A, Braehler E, Glaesmer H, Wagner B. Prevalence of complicated grief in a representative population-based sample. *J Affect Disord.* (2011) 131:339–43. doi: 10.1016/j.jad.2010.11.032

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Copyright © 2020 Sveen, Bondjers, Heinsoo and Arnberg. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.



Exploring Functional Impairment in Light of Prolonged Grief Disorder: A Prospective, Population-Based Cohort Study

Mette Kjaergaard Nielsen^{1,2*}, Kaj Sparle Christensen^{1,2}, Mette Asbjørn Neergaard³, Pernille Envold Bidstrup^{4,5} and Mai-Britt Guldin^{1,3}

¹ Research Unit for General Practice, Aarhus, Denmark, ² Department of Public Health, Aarhus University, Aarhus, Denmark, ³ Palliative Care Unit, Department of Oncology, Aarhus University Hospital, Aarhus, Denmark, ⁴ Psychological Aspects of Cancer, Danish Cancer Society Research Center, Copenhagen, Denmark, ⁵ Department of Psychology, University of Copenhagen, Copenhagen, Denmark

OPEN ACCESS

Edited by:

Geert E. Smid,
ARQ National Psychotrauma
Centre, Netherlands

Reviewed by:

Maarten C. Eisma,
University of Groningen, Netherlands
Hannah Comtesse,
Catholic University of
Eichstätt-Ingolstadt, Germany

*Correspondence:

Mette Kjaergaard Nielsen
mette.k.nielsen@ph.au.dk

Specialty section:

This article was submitted to
Mood and Anxiety Disorders,
a section of the journal
Frontiers in Psychiatry

Received: 24 February 2020

Accepted: 18 November 2020

Published: 09 December 2020

Citation:

Nielsen MK, Christensen KS,
Neergaard MA, Bidstrup PE and
Guldin M-B (2020) Exploring
Functional Impairment in Light of
Prolonged Grief Disorder: A
Prospective, Population-Based Cohort
Study. *Front. Psychiatry* 11:537674.
doi: 10.3389/fpsy.2020.537674

Background: Functional impairment is essential in the diagnostic criteria for prolonged grief disorder (PGD) in the ICD-11. It refers to the negative impact on everyday life, including inability to maintain the usual level of functioning. We aimed to assess the extent of functional impairment, emotion-related role limitation, and impaired social functioning before and after bereavement, and to explore associations with PGD, as measured by the 13-item Prolonged Grief Scale (PGD_{PG13}).

Method: Relatives of terminally ill patients ($n = 1,622$) completed a questionnaire before and after bereavement. The questionnaire assessed “overall functional impairment” (PG-13 item) and “aspects of functional impairment” measured by mean scores of the 36-item Short Form Survey (SF-36) subscales *emotional role* and *social functioning* (0: worst; 100: best). We analyzed associations between PGD_{PG13} and functional impairment prior to bereavement using logistic regression models adjusted for age, gender, personal relation, education, time interval to patient’s death, and pre-loss grief.

Results: In total, 51% reported overall functional impairment before bereavement, 27% reported functional impairment at 6 months after bereavement, and 19% reported functional impairment at 3 years after bereavement. The mean *emotional role* score was 47.5 (95%CI: 45.4–49.7) before bereavement, increasing to 77.4 (95%CI: 75.7–79.0) at 3 years after bereavement, compared to 85.1 (95%CI: 77.6–92.6) in a reference sample. Mean *social functioning* score increased gradually reaching the mean of the reference sample at 3 years after bereavement. PGD_{PG13} was present in 26% of those with overall functional impairment at 6 months after bereavement, decreasing to 11% at 3 years after bereavement. Pre-bereavement measures of *emotional role* and *social functioning* were associated with PGD_{PG13} at 6 months and 3 years after bereavement.

Discussion: Overall functional impairment was prevalent as reflected in low scores on daily activities and social functioning compared to a reference sample. Functioning

may be an important factor during caregiving and bereavement and pre-bereavement functional impairment was associated with PGD_{PG13}.

Future studies should investigate if maintaining daily activities and social functioning before bereavement could be key in early supportive care. Moreover, the role of functional impairment in bereavement interventions should be explored.

Keywords: functional impairment, bereavement, grief, family caregivers, Prolonged Grief Disorder (PGD), SF-36 = 36-item short form health survey, prolonged grief diagnostic criteria

INTRODUCTION

Severe illness, bereavement and other losses are likely to impair the ability to maintain daily activities and thereby cause functional impairment (1). The World Health Organization (WHO) defines functional impairment as an umbrella term in the International Classification of Functioning, Disability and Health (ICF) and conceptualizes a biopsychosocial approach to functioning in the context of health (2). Moreover, functional impairment may imply emotional suffering and is a common reason for healthcare seeking (3).

According to the 11th revision of the International Classification of Diseases (ICD-11), prolonged grief disorder (PGD_{ICD11}) entails longing for, or preoccupation with, the deceased person, which is accompanied by intense emotional pain, and functional impairment is described as “*impairment of personal, family, social, occupational, educational or other important areas of functioning*” (4). Thus, functional impairment needs to be present to fulfill the diagnostic criteria for PGD_{ICD11} (4).

Functional impairment has been included as a clinical significance criterion in the diagnosis of mental illness in the Diagnostic and Statistical Manual of Mental Disorders (DSM) by the American Psychiatric Association since 1980 (1). It was added as a new criterion in the ICD-11 to improve the distinction between normality and disturbance (5). Hence, including functional impairment in the diagnostic criteria of PGD_{ICD11} may support the clinical utility of the diagnosis, which is intended to help health professionals assess the needs of the bereaved person, to differentiate between PGD and natural grief or other disorders, and to plan treatment (6, 7).

Despite the central role of functional impairment, only few studies have explored aspects of functional impairment in bereaved persons. Maladaptive emotional symptoms of grief have been investigated without including functional impairment; these studies have revealed subgroups of people with different patterns of grief symptoms (8, 9). Functional impairment has been associated with PG symptoms measured on different grief symptom scales in disaster settings (10), in bereaved adolescents (11, 12), and in a small-scale study of bereaved persons recruited from a funeral service (13). A recent cross-sectional study used network analysis to explore associations between aspects of *quality of life impairments* on the WHO abbreviated multidimensional quality-of-life scale (WHOQOL-BREF) and PG symptoms measured on the Prolonged Grief-13 scale (PG-13) (14). The PG-13 includes

both grief symptoms and functional impairment. Different aspects of impairments were associated with different PGD_{PG13} symptoms. For example, poor psychological health was associated with the PGD_{PG13} symptoms of *meaninglessness* and *role confusion*, whereas poor social health was associated with the PGD_{PG13} symptom *lack of trust in others*. These results point at functional impairment as a complex concept encompassing a range of aspects that interact with symptoms of PG. However, previous research has focused on functional impairment in bereaved persons suffering from PGD, whereas the role of functional impairment as a risk factor for bereavement outcomes like PGD and depression remains unknown.

Stroebe et al. established groups of risk factors for adverse bereavement outcomes. These risk factors may interact, and they are mediated by the person's coping and emotion regulation (15). Risk factors include *intrapersonal factors*, such as low educational level and a history of poor mental health (16, 17), *interpersonal factors*, such as lack of social support and *situational factors* related to the context of caregiving and death (15). Social network and everyday life, including work are areas related to interpersonal and situational factors. These areas are highlighted in the PGD_{ICD11} criteria. According to the ICF, functional impairment includes bodily functions, activities, and participation in daily life including work and social life, which interact with personal factors and the context (2). The PG-13 scale is widely used for measuring prolonged grief and includes an item of overall functional impairment, without assessment of specific areas (18). The level of impairment in specific areas of daily activities and social functioning can be assessed by the Short Form survey-36 (SF-36) (19).

We aimed to examine the overall extent of functional impairment before bereavement and the level at 6 months after bereavement and at 3 years after bereavement, including the impact on daily activities and social functioning. Furthermore, we aimed to explore the association between emotion-related role and social functioning with PGD_{PG13}.

We hypothesized that functional impairment would more often be present in bereaved persons compared to an age-matched reference population, and we anticipated that the extent of functional impairment would decrease to the level seen in a reference sample at 3 years after bereavement when the proportion of severely impaired bereaved persons would be low. Furthermore, we hypothesized that all aspects of functional impairment before bereavement would be associated with PGD_{PG13} after bereavement.

METHODS

Setting and Design

We conducted a longitudinal, population-based study in a cohort of Danish family caregivers, which was established in 2012. The current study was a secondary analysis. The primary and previous secondary analysis focused on symptoms of grief and depression before and after death (17, 20, 21), trajectories of grief symptoms (8) and physical and mental health (22).

Participants were identified through patients registered with a drug reimbursement scheme due to terminal illness in the *Danish National Database of Reimbursed Prescriptions* (23). On a weekly basis, we sent an information letter and a questionnaire to newly registered patients and requested their closest family caregiver to complete the questionnaire (20). Enrolled family caregivers providing informed consent were asked to complete a questionnaire at inclusion (T0), at 6 months after bereavement (T1), and at 3 years after bereavement (T2).

In this study, we included caregivers who had completed a questionnaire at all three time points, filled in the functional impairment item of the PG-13 scale before bereavement (T0), and the total PG-13 at 6 months after bereavement (T1) (**Figure 1**).

Ethics

The Committee on Health Research Ethics of the Central Denmark Region confirmed that this study required no ethical clearance according to the Danish Act on Research Ethics Review of Health Research Projects (24). The study was approved by the Danish Data Protection Agency (file no. 2013-41-2603) and registered in the Record of Processing Activities at the Research Unit for General Practice, Aarhus (Id 207) in accordance with the provisions of the General Data Protection Regulation (GDPR). The participants gave written informed consent to participate in this study.

Functional Impairment

Overall functional impairment (yes, no) was measured as *social, occupational, and other* impairment in one measure by the item “Have you experienced a significant reduction in social, occupational, or other important areas of functioning (e.g., domestic responsibilities)?” of the PG-13. It was reported as proportions with impairment before bereavement, at 6 months after bereavement, and at 3 years after bereavement (18). Functional impairment related to limitations in daily role due to emotional impairment and limitations in social functioning was assessed by the SF-36 subscales *emotional role* and *social functioning* before bereavement, at 6 months after bereavement, and at 3 years after bereavement (19). Subscale mean scores were weighted with scores of the remaining SF-36 subscales according to the manual, and a sum score was derived (25). Scores were graded from zero to 100, with 100 indicating highest performance (i.e., better functioning). Thus, low scores indicate impaired functioning. For comparison, we used standardized mean scores

from the general Danish population aged 55–65 years as the mean age of the cohort was 62 years (26).

The *emotional role* subscale consists of three items exploring whether less time is spent on activities (RE1), accomplishment of tasks (RE2), and carefulness devoted to performing activities (RE3) (yes, no). As the items cover functioning in both daily activities and occupation, they are also applicable to retired individuals (19). The *social functioning* subscale consists of two items. One item (SF1) measures whether physical and emotional problems interfere with the extent of social activities with family, friends, neighbors, or others (no (not at all, little), yes (some, a lot, very much)). The other item (SF2) measures the extent of interference from physical health or emotional problems on social activities, such as visiting friends, relatives, or others (not at all, little of the time, some of the time, most of the time, all the time) (19). Raw means of individual items were calculated according to the manual (25).

Grief, Depressive Symptoms, and Sociodemographic Factors

All items of the PG-13 scale (18) were used to measure fulfillment of the PGD_{PG13} criteria (yes, no) at 6 months and at 3 years after bereavement. To fulfill the criteria, participants must have reported core symptoms of longing/yearning or intense feelings related to the loss at least daily (a score of at least 4 on a 5 point scale), at least five out of nine accessory symptoms such as trouble accepting the loss, bitterness and emotional numbness (a score of at least 4 on a 5 point scale), a duration of 6 months or longer, and functional impairment (18). In collaboration with the author of the PG-13 scale, Dr. Prigerson, the Danish version of the PG-13 was developed according to the WHO recommendation for translation of scales using forward and back ward translation (27). The Cronbach's alpha of the entire scale was 0.90. The documentation of the translation process is described in a paper in preparation.

Grief symptoms during the patient's illness trajectory were assessed by a version of the PGD_{PG13} adapted to terminal illness (20). In the pre-loss version, “the loss” was replaced by “your relative's illness,” the item concerning “moving on” was replaced with an item about concentration problems inspired by a previous study on grief before death (28), and the duration item was left out (please see details in **Supplementary Material A**).

Depressive symptoms were assessed before bereavement by the Beck Depression Inventory-II (29) and dichotomized according to severity (no-mild, moderate-severe).

Information on the family caregiver's personal relation to the terminally ill patient was obtained at T0 [partner, non-partner (adult child, other relation)]. Data on the family caregiver's age, gender, educational level, and the patient's survival time from drug reimbursement were extracted from the *Danish Civil Registration System* (30).

Statistical Analysis

Questionnaire data and registry-based data were combined at *Statistics Denmark* (31). Overall functional impairment was reported as proportions. Weighted mean scores of the *emotional role* and *social functioning* subscales and mean scores of the

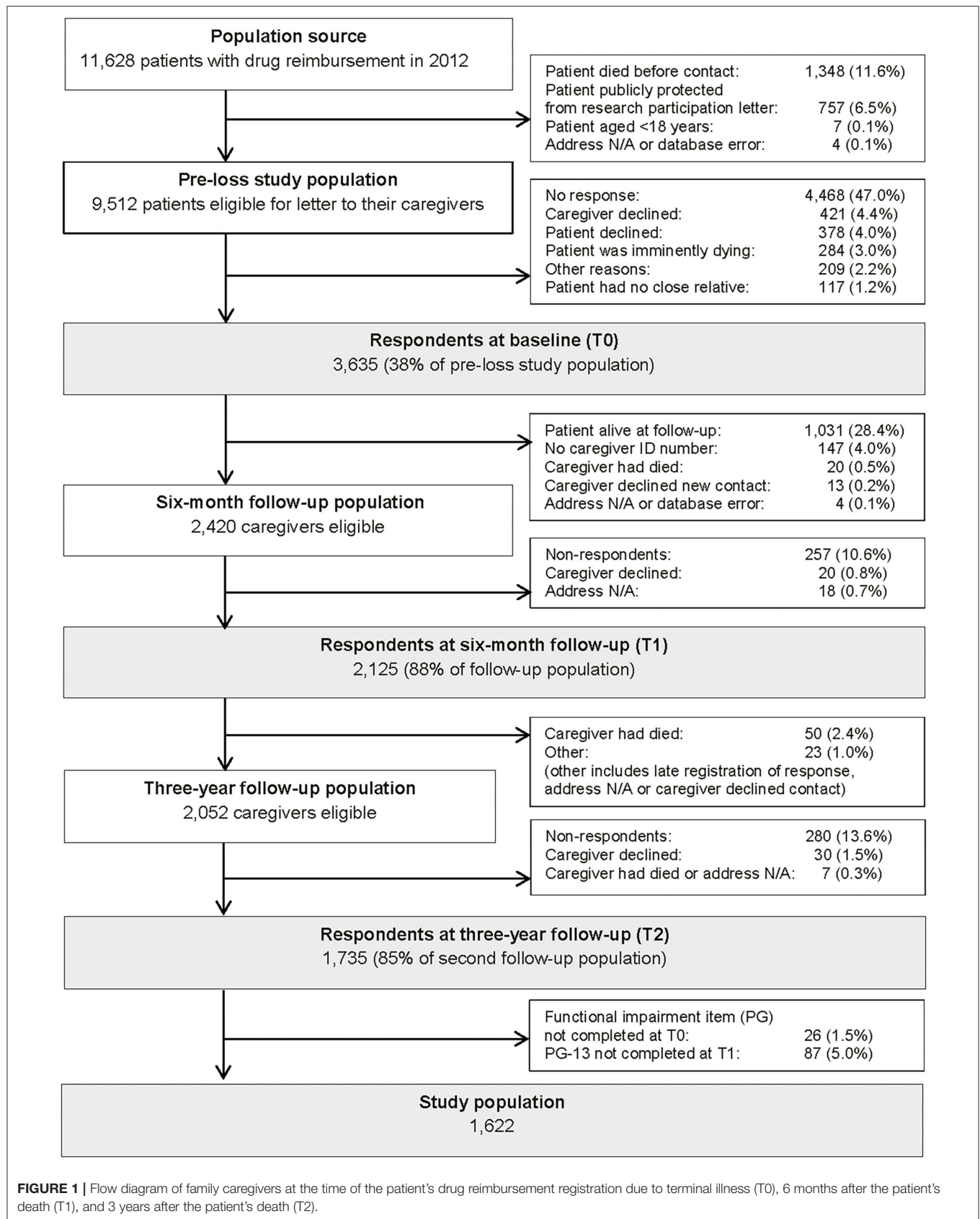


FIGURE 1 | Flow diagram of family caregivers at the time of the patient's drug reimbursement registration due to terminal illness (T0), 6 months after the patient's death (T1), and 3 years after the patient's death (T2).

TABLE 1 | Characteristics of the total study cohort ($n = 1,622$).

Caregiver characteristics	Total cohort ($n = 1,622$)			Overall functional impairment* ($n = 831$)			No overall functional impairment ($n = 791$)		
	Mean (95% CI)	n	(%)	Mean (95% CI)	n	(%)	Mean (95% CI)	n	(%)
Age, years ^a	61.8 (61.2; 62.3)			60.6 (59.8; 61.4)			63.0 (62.2; 63.7)		
Gender									
Male		480	(30)		178	(21)		302	(38)
Female		1,142	(70)		653	(79)		489	(62)
Personal relation									
Partner		1,050	(65)		552	(66)		498	(63)
Other		572	(35)		279	(34)		293	(37)
Education									
Low (≤ 10 years)		405	(25)		204	(25)		201	(25)
Intermediate (> 10 and ≤ 15 years)		780	(48)		388	(47)		392	(50)
High (> 15 years)		437	(27)		239	(29)		198	(25)
Pre-death PG criteria ^b									
Not fulfilled criteria		1,345	(83)		570	(69)		775	(98)
Fulfilled criteria		252	(16)		252	(30)		-	-
Missing		25	(2)		9	(1)		16	(2)
Depressive symptoms ^c									
No-mild symptoms		1,318	(81)		601	(72)		717	(91)
Moderate-severe symptoms		239	(15)		195	(23)		44	(6)
Missing		65	(4)		35	(4)		30	(4)
Patient characteristics	Median (IQR)			Median (IQR)			Median (IQR)		
Time from drug reimbursement** to death, days	69.4 (67.0; 71.8)			66.6 (63.4; 69.9)			72.3 (68.7; 75.8)		

*Based on the impairment item of the PG-13.

**Registration of patient with drug reimbursement due to terminal illness and invitation for study inclusion.

^aCaregivers' age at baseline.

^bProlonged grief criteria measured on an adapted pre-death version of the Prolonged Grief-13 scale.

^cDepressive symptoms measured on Beck's Depression Inventory-II.

single items (RE1-3 and SF1-2) were reported as means with 95% confidence intervals at T0, T1, and T2. *Emotional role* and *social functioning* mean scores were compared to standard scores in a Danish reference sample aged 55-64 (26), and interpreted using minimal clinical important difference (MCID) between the mean scores of the study sample and the reference sample. The MCID is the smallest change in an outcome score that the individual person will consider important and corresponds to 3-5 units on the SF-36 scale (32).

We used logistic regression with 95% confidence intervals to calculate associations between SF-36 measures of functional impairment before bereavement and PGD_{PG13}. We adjusted for age, gender, personal relation, education, time from inclusion to the patient's death, and grief symptom level before death (sum score). These covariates were socioeconomic factors and clinical factors that have been associated with in earlier studies (28, 33). Due to the number of fulfilling the criteria for PGD_{PG13} ($n = 117$ events), inclusion of these covariates did not violate the assumptions of logistic regression (34). Results were reported as odds ratios with 95% confidence intervals.

All analyses were conducted by Stata 14 (StataCorp, Texas, USA, RRID:SCR_012763).

RESULTS

Study Population

A total of 1,622 bereaved persons were included in the study (Figure 1). The mean age was 62 years (Table 1). Participants were predominantly female (70%), partner to the patient (65%), and 25% had < 10 years of education. Furthermore, pre-loss grief (fulfilling the criteria of the adapted PGD_{PG13} before bereavement) was reported in 15 and 16% had depressive symptoms before bereavement. Among those reporting overall functional impairment (PG item), the mean age was lower (60.6 years (59.8; 61.4) vs. 63.0 (62.2; 63.7)) and proportions of females (79% vs. 62%) and participants with depressive symptoms (23% vs. 6%) were higher compared to in participants without overall functional impairment (Table 1).

TABLE 2 | Functional impairment in the study sample ($n = 1,622$) before bereavement, at 6 months and 3 years after bereavement and in a standardized reference sample of Danes aged 55–64 years.

	0–6 months before bereavement		6 months after bereavement		3 years after bereavement		Reference sample [§]	
	<i>n</i>	(%)	<i>n</i>	(%)	<i>n</i>	(%)		
Overall functional impairment (PG item)^a								
Impairment criterion (single item), yes (%)	831	(51)	438	(27)	306	(19)	NA	NA
Emotional role (SF-36)^b								
	Mean (95% CI)		Mean (95% CI)		Mean (95% CI)		Mean, SD	
<i>Emotional role</i> subscale, weighted mean (95% CI)	47.5	45.4; 49.7	71.5	69.8; 73.3	77.4	75.7; 79.0	85.1	77.6; 92.6 *
RE1. Cut down on amount of time spent on work/other activities ^c	1.5	1.5; 1.5	1.78	1.8; 1.8	1.8	1.8; 1.8	1.9	0.29 #
RE2. Accomplished less than you would like ^c	1.4	1.4; 1.4	1.55	1.5; 1.6	1.7	1.6; 1.7	1.8	0.40 #
RE3. Did not do work or other activities as carefully as usual ^c	1.5	1.5; 1.6	1.8	1.8; 1.8	1.8	1.8; 1.9	1.9	0.30 #
Social functioning (SF-36)^b								
<i>Social functioning</i> subscale, weighted mean (95% CI)	77.3	76.1; 78.5	85.3	84.3; 86.3	88.9	87.9; 89.8	89.9	82.0; 97.8 *
SF1. Extent, mean (95% CI) ^d	4.3	4.2; 4.3	4.5	4.4; 4.5	4.6	4.5; 4.6	4.7	0.71 #
SF2. Time, mean (95% CI) ^e	3.9	3.9; 4.0	4.4	4.3; 4.4	4.5	4.5; 4.6	4.7	0.74 #

^aFunctional impairment item of the Prolonged Grief-13 scale (18).^bShort Form 36-item Health Outcome survey (19).^cText of the RE items: "During the past 4 weeks, have you had any of the following problems with your work or other regular daily activities as a result of any emotional problems (such as feeling depressed or anxious)".^dText of SF1 item: "During the past 4 weeks, to what extent has your physical health or emotional problems interfered with your normal social activities with family, friends, neighbors, or groups?"^eText of SF2 item: "During the past 4 weeks, how much of the time has your physical health or emotional problems interfered with your social activities (like visiting friends, relatives, etc.)?"[§]Danish standardized norm population (25).

*Mean (95% CI) in the Danish standardized norm population aged 55–64 years.

#Mean (SD) in the total standardized Danish norm population ($n = 3,602$).

NA: not applicable.

Functional Impairment Before and After Bereavement

In total, proportions of 51% reported overall functional impairment before bereavement, 27% at 6 months after bereavement, and 19% at 3 years after bereavement measured on the PG-13 impairment item (Table 2). Of those reporting overall functional impairment at 6 months, 63 (54%) also reported overall impairment before bereavement, whereas 21 (68%) of those reporting overall impairment at 3 years also had overall impairment before bereavement.

Before bereavement, the mean *emotional role* score (47.5 (95% CI: 45.4–49.7)) and the mean *social functioning* score (77.3 (95% CI: 76.1–78.5)) were significantly lower compared to the reference sample (85.1 for *emotional role* and 89.9 for *social functioning*) (Table 2). The difference between means was 37.6 for *emotional role* and 12.6 for *social functioning*. As the MCID was three to five for SF-36 subscales, the mean scores were significantly lower in the study sample (The mean scores of both subscales had increased at 6 months after bereavement and almost reached the level of the reference sample at 3 years after bereavement (*emotional role*: 77.4 (95% CI: 75.7–79.0), *social functioning*: 88.9 (95% CI: 87.9–89.8)). An MCID of five was reached for the *emotional role* subscale mean score compared to the reference sample, but no MCID was found for the *social functioning* mean score at 3 years after bereavement (Table 2).

The mean scores of all *emotional role* and *social functioning* subscale items were accordingly low in the bereaved persons,

and the social functioning item regarding *less time spent on social activity* was very low before bereavement (3.9 (3.9–4.0)) compared to a reference sample (4.7 (SD 0.74)) (Table 2).

In those with overall functional impairment before bereavement, the mean scores of *emotional role* (31.0 (95% CI: 28.3–33.7)) and *social functioning* (68.6 (95% CI: 66.8–70.4)) were lower than in the total study cohort before bereavement (Table 3a). Mean scores improved over time (Figure 2). However, an MCID of at least three was present at all measurement points in those with overall functional impairment. Women and those below 60 years of age were more likely to report overall functional impairment and limitation in *social functioning*, whereas women and those above 60 years were more likely to report limitations *emotional role* (Tables 3b,c).

Of the 438 bereaved persons reporting overall functional impairment at 6 months after bereavement, 27% fulfilled the PGD_{PG13} criteria. Accordingly, 10% of those reporting overall functional impairment at 3 years after bereavement fulfilled the PGD_{PG13} criteria (data not shown).

Functional Impairment Before Bereavement and PGD_{PG13}

Functional impairment before bereavement was associated with PGD_{PG13} at 6 months after bereavement on the *emotional role*

TABLE 3a | Functional impairment aspects (SF-36) before bereavement, 6 months after bereavement, and 3 years after bereavement in the subgroup with overall functional impairment before bereavement (PG item^a) (51% of the total population).

	Overall functional impairment ^a before bereavement (<i>n</i> = 851)					
	Before bereavement		6 months after bereavement		3 years after bereavement	
Emotional role (SF-36)^b						
<i>Emotional role</i> subscale, weighted mean (95% CI)	31.0	(28.2; 33.7)	63.2	(60.6; 65.7)	71.6	(69.1; 74.1)
RE1. Cut down on amount of time spent on work/other activities ^c	1.3	(1.3; 1.4)	1.7	(1.7; 1.8)	1.8	(1.7; 1.8)
RE2. Accomplished less than you would like ^c	1.2	(1.2; 1.3)	1.4	(1.4; 1.5)	1.6	(1.6; 1.6)
RE3. Did not do work or other activities as carefully as usual ^c	1.4	(1.3; 1.4)	1.7	(1.7; 1.7)	1.8	(1.8; 1.8)
Social functioning (SF-36)^b						
<i>Social functioning</i> subscale, weighted mean (95% CI)	68.6	(66.8; 70.4)	79.8	(78.2; 81.4)	85.1	(83.6; 86.6)
SF1. Extent, mean (95% CI) ^d	3.9	(3.9; 4.0)	4.3	(4.2; 4.3)	4.4	(4.4; 4.5)
SF2. Time, mean (95% CI) ^e	3.5	(3.5; 3.6)	4.1	(4.1; 4.2)	4.4	(4.3; 4.5)

^aFunctional impairment item of the Prolonged Grief-13 scale (18).

^bShort Form-36 Health Outcome survey (19).

^cText of the RE items: "During the past 4 weeks, have you had any of the following problems with your work or other regular daily activities as a result of any emotional problems (such as feeling depressed or anxious)".

^dText of the SF1 item: "During the past 4 weeks, to what extent has your physical health or emotional problems interfered with your normal social activities with family, friends, neighbors, or groups?"

^eText of the SF2 item: "During the past 4 weeks, how much of the time has your physical health or emotional problems interfered with your social activities (like visiting friends, relatives, etc)?"

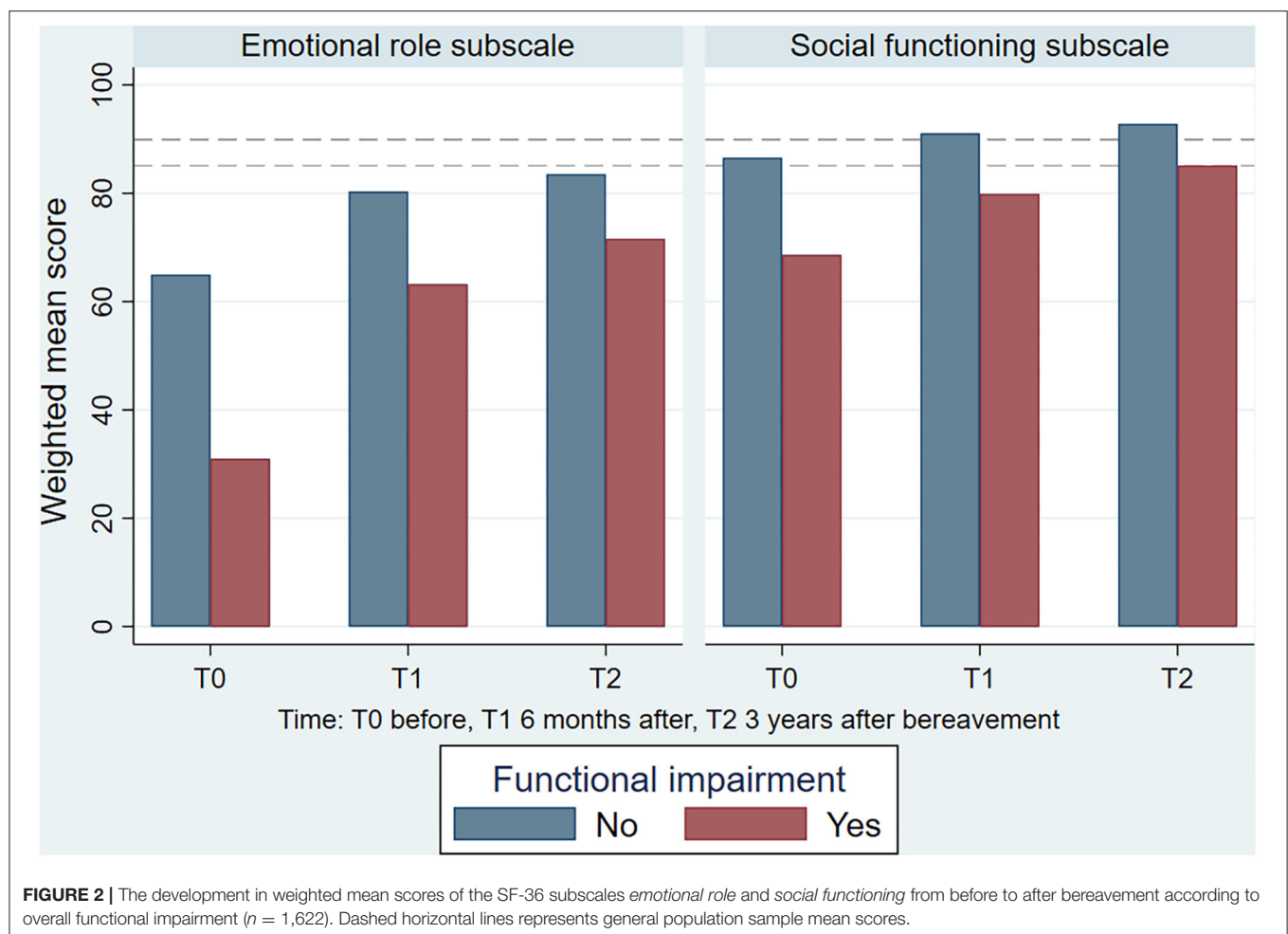


TABLE 3b | Functional impairment aspects (SF-36) before bereavement, 6 months after bereavement, and 3 years after bereavement according to gender.

	Functional impairment, males (<i>n</i> = 480 (30%))					
	Before bereavement		6 months after bereavement		3 years after bereavement	
Overall functional impairment (PG-13 scale) ^a						
Yes, <i>n</i> (%)	178	(37)	95	(20)	75	(16)
Aspects of functional impairment (SF-36 scale) ^b						
<i>Emotional role</i> subscale, weighted mean (95% CI)	53.4	(49.5; 57.4)	74.0	(70.9; 77.1)	76.6	(73.5; 79.6)
<i>Social functioning</i> subscale ² , weighted mean (95% CI)	82.8	(80.8; 84.8)	89.2	(87.5; 90.9)	89.4	(87.7; 91.2)
	Functional impairment, females (<i>n</i> = 1,142 (70%))					
	Before bereavement		6 months after bereavement		3 years after bereavement	
Overall functional impairment (PG-13 scale) ^a						
Yes, <i>n</i> (%)	653	(57)	343	(30)	213	(20)
Aspects of functional impairment (SF-36 scale) ^b						
<i>Emotional role</i> subscale, weighted mean (95% CI)	45.0	(42.5; 47.6)	70.5	(68.4; 72.6)	77.7	(75.7; 79.7)
<i>Social functioning</i> subscale ^b , weighted mean (95% CI)	75.1	(73.6; 76.5)	83.7	(82.4; 85.0)	88.6	(87.5; 89.8)

^aFunctional impairment item of the Prolonged Grief-13 scale (18).^bShort Form-36 Health Outcome survey (19).**TABLE 3c** | Functional impairment aspects (SF-36) before bereavement, 6 months after bereavement, and 3 years after bereavement according to age.

	Age <60 (<i>n</i> = 665 (41%))					
	Before bereavement		6 months after bereavement		3 years after bereavement	
Overall functional impairment (PG-13 scale)^a						
Yes, <i>n</i> (%)	384	(58)	210	(32)	137	(21)
Aspects of functional impairment (SF-36 scale)^b						
Emotional role subscale, weighted mean (95% CI)	48.4	(45.1; 51.6)	72.6	(69.9; 75.3)	79.7	(77.2; 82.3)
Social functioning subscale ^b , weighted mean (95% CI)	76.3	(74.4; 78.2)	83.1	(81.4; 84.8)	87.8	(86.2; 89.4)
	Age >60 (<i>n</i> = 957 (59%))					
	Before bereavement		6 months after bereavement		3 years after bereavement	
Overall functional impairment (PG-13 scale)^a						
Yes, <i>n</i> (%)	447	(47)	228	(24)	169	(18)
Aspects of functional impairment (SF-36 scale)^b						
Emotional role subscale, weighted mean (95% CI)	47.0	(44.1; 49.8)	70.7	(68.5; 73.0)	75.7	(73.5; 79.0)
Social functioning subscale ^b , weighted mean (95% CI)	78.1	(76.5; 79.6)	86.8	(85.5; 88.1)	89.6	(88.4; 90.8)

^aFunctional impairment item of the Prolonged Grief-13 scale (18).^bShort Form-36 Health Outcome survey (19).

(OR = 0.97 (95% CI 0.96; 0.98)) and *social functioning* (OR = 0.97 (95% CI 0.96; 0.98)) subscales of the SF-36 (Tables 4 and 5). The *emotional role* items indicating *less time spent on work or activities, accomplishment of tasks, and carefulness in performing activities*, and both *social functioning* items indicating *lower extent and less time spent on social network*, were also associated with PGD_{PG13}.

Functional impairment before bereavement on the *emotional role* and *social functioning* subscales was also associated with PGD_{PG13} at 3 years after bereavement in unadjusted analysis (Table 5).

DISCUSSION

Key Findings

The present study explored functional impairment in family caregivers and revealed novel findings. Firstly, functional impairment was present in a large proportion of bereaved persons before the bereavement, and the scores relating to specific aspects of impairment in daily activities and social functioning were much lower in this group compared to a reference sample. Secondly, the functional impairment improved over time. The mean *social functioning* scores reached the level of the reference

TABLE 4 | Functional impairment (SF-36 subscales) and the association with prolonged grief disorder (PG-13)^a at 6 months after bereavement in the total cohort (*N* = 1,622) and in the subgroup with overall functional impairment before bereavement (*n* = 831)^b.

	PG-13 Prolonged grief disorder at 6 months after bereavement ^a							
	Total cohort				Overall functional impairment ^b before bereavement			
	No PG (<i>n</i> = 1,505)		PG (<i>n</i> = 117)		No (<i>n</i> = 742 (89%))		Yes (<i>n</i> = 89 (11%))	
Emotional role (SF-36)^c								
<i>Emotional role</i> subscale, weighted mean (95% CI)	50.0	(47.7; 52.2)	16.4	(11.0; 21.8)	33.0	(30.1; 35.9)	13.8	(8.1; 19.5)
RE1. Cut down on amount of time spent on work/other activities ^d	1.5	(1.5; 1.5)	1.2	(1.2; 1.3)	1.4	(1.3; 1.4)	1.2	(1.1; 1.3)
RE2. Accomplished less than you would like ^d	1.4	(1.4; 1.4)	1.1	(1.0; 1.2)	1.3	(1.2; 1.3)	1.1	(1.0; 1.1)
RE3. Did not do work or other activities as carefully as usual ^d	1.6	(1.5; 1.6)	1.2	(1.1; 1.2)	1.4	(1.3; 1.4)	1.1	(1.1; 1.2)
Social functioning (SF-36)^c								
<i>Social functioning</i> subscale, weighted mean (95% CI)	79.0	(77.8; 80.2)	56.4	(51.8; 61.0)	70.4	(68.6; 72.3)	53.0	(48.0; 58.0)
SF1. Extent, mean (95% CI) ^e	4.3	(4.3; 4.4)	3.5	(3.3; 3.7)	4.0	(3.9; 4.1)	3.4	(3.1; 3.6)
SF2. Time, mean (95% CI) ^f	4.0	(3.9; 4.0)	3.0	(2.8; 3.2)	3.6	(3.5; 3.7)	2.9	(2.7; 3.1)

^aProlonged Grief-13 scale (18).^bFunctional impairment item of the Prolonged Grief-13 scale (18).^cShort Form 36-item Health Outcome survey (19).^dText of the RE items: "During the past 4 weeks, have you had any of the following problems with your work or other regular daily activities as a result of any emotional problems (such as feeling depressed or anxious)".^eText of SF1 item: "During the past 4 weeks, to what extent has your physical health or emotional problems interfered with your normal social activities with family, friends, neighbors, or groups?"^fText of SF2 item: "During the past 4 weeks, how much of the time has your physical health or emotional problems interfered with your social activities (like visiting friends, relatives, etc.)?"**TABLE 5 |** Functional impairment before bereavement and associations with prolonged grief disorder (PG-13)^a at 6 months and at 3 years after bereavement (*n* = 1,622).

	Prolonged grief disorder (PG-13)		
	6 months after bereavement		3 years after bereavement
	Unadjusted OR (95% CI)	Adjusted [#] OR (95% CI)	Unadjusted OR (95% CI)
Emotional role (SF-36)^b			
<i>Emotional role</i> subscale, weighted mean (95% CI)	0.98 (0.97; 0.98)	0.98 (0.98; 0.99)	0.98 (0.97; 0.99)
RE1. Cut down on amount of time spent on work/other activities ^c	0.27 (0.17; 0.43)	0.43 (0.27; 0.70)	0.347 (0.15; 0.76)
RE2. Accomplished less than you would like ^c	0.15 (0.08; 0.28)	0.24 (0.13; 0.46)	0.16 (0.05; 0.53)
RE3. Did not do work or other activities as carefully as usual ^c	0.16 (0.10; 0.26)	0.25 (0.15; 0.43)	0.13 (0.04; 0.37)
Social functioning (SF-36)^b			
<i>Social functioning</i> subscale, weighted mean (95% CI)	0.97 (0.96; 0.98)	0.98 (0.97; 0.98)	0.97 (0.96; 0.98)
SF1. Extent, mean (95% CI) ^d	0.52 (0.45; 0.61)	0.63 (0.53; 0.76)	0.53 (0.40; 0.70)
SF2. Time, mean (95% CI) ^e	0.54 (0.47; 0.63)	0.63 (0.53; 0.75)	0.52 (0.39; 0.68)

[#]Logistic regression model adjusted for age, gender, personal relation to the patient, education, time from inclusion to the patient's death and grief symptoms before death.^aProlonged Grief-13 scale (18).^bShort Form 36-item Health Outcome survey (19).^cText of the RE1-3 items: "During the past 4 weeks, have you had any of the following problems with your work or other regular daily activities as a result of any emotional problems (such as feeling depressed or anxious)".^dText of SF1 item: "During the past 4 weeks, to what extent has your physical health or emotional problems interfered with your normal social activities with family, friends, neighbors, or groups?"^eText of SF2 item: "During the past 4 weeks, how much of the time has your physical health or emotional problems interfered with your social activities (like visiting friends, relatives, etc.)?"

sample at 3 years after bereavement, whereas the mean *emotional role* scores remained significantly lower than in the reference sample at 3 years after bereavement. In the subgroup with overall functional impairment before bereavement, the *emotional role* and *social functioning* mean scores were even lower than in the total cohort.

Thirdly, low scores on the *emotional role* and *social functioning* subscales and specific aspects of reduced time,

accomplishment, carefulness in performing activities, and extent and time spent on social activities before bereavement were risk factors for PGD_{PG13}.

Comparison With Previous Finding

The extent of functional impairment was surprisingly high at all measurement points. We used the PG item of functional impairment to measure overall impairment. This broad measure

included social, occupational, and other important areas of functioning and is likely to capture functioning in general. By investigating functional impairment aspects on the SF-36 *emotional role* and *social functioning* subscales, we showed that limited “daily role functioning due to emotional state” and reduced “social functioning” were associated with overall functional impairment. This is in line with our hypothesis. The ICF interprets functional impairment in a biopsychosocial framework, emphasizing the importance of maintaining daily activities in the context of bereavement (2). Our findings of high overall functional impairment support the interpretation of functional impairment according to the ICF. Moreover, we established new knowledge as we found that a high proportion of relatives were functionally impaired; this knowledge can support the clinical assessment of bereaved persons and enhance the planning of health care services before and after bereavement.

Half of the participants reported overall functional impairment before the bereavement. End-of-life care is highly demanding for relatives, and the situation may affect their quality of life and cause psychological distress (35). Our study did not investigate the functional impairment before the end-of-life period and did not include a matched comparison group. This limits our opportunity to conclude that the functional impairment was caused by caregiving. Nevertheless, the very low mean scores of *emotional role* and *social functioning* compared to the reference sample suggest that end-of-life care considerably affects the functioning of relatives. The impact of caregiving has been explored in a comparative study ($n = 70$); this study showed that grief symptoms were more present and that general health and quality of life were significantly worse in family caregivers than in the comparison group (36). The measures of grief, general health and quality of life improved 9–10 months after bereavement. We found similar results in a large-scale sample, although functional impairment in terms of emotional impact on everyday role was still shown at 3 years after bereavement.

We showed a pattern of improved functioning in the period from before to after bereavement. This finding is in line with earlier research based on the present cohort, which showed that a higher proportion of bereaved individuals reported psychological distress (i.e., symptoms of grief and depression) before bereavement than after bereavement (17). Still, a high proportion (19%) reported overall functional impairment at 3 years after bereavement, which was unexpected as we hypothesized that only a few would still be functionally impaired for a long time after bereavement. Of the bereaved persons with overall functional impairment at 3 years after bereavement, only a small proportion fulfilled the criteria of PGD_{PG13}. Previous studies have focused on the functional impairment in bereaved persons as a consequence of PGD (10–12) and used the level of functional impairment to monitor the status of the condition. However, our findings establish that functional impairment in daily activities, social network and other areas plays an important role in the grief process in general, not only in bereaved persons with severe bereavement reactions like PGD.

In a previous study, we found that poor mental and physical health status measured by the SF-36, including the subscales of *emotional role* and *social functioning*, were risk factors for

PGD_{PG13}. Importantly, we now provide detailed knowledge on the aspects associated with functional impairment. Aspects of daily activities, including time spent, accomplishment, and carefulness in performing activities, but also time and extent of social activities were severely affected before bereavement in our cohort compared to the general population sample, and these aspects constituted risk factors of PGD_{PG13}. Daily activities prior to bereavement are related to the caregiver context and constitute situational factors in the *integrated risk factor framework for bereavement outcome*. The authors of the framework describes caregiver burden residue as a restoration-oriented stressor that may affect the adaptation to the bereavement (15). A few previous studies point to caregiver burden as a risk factor for PGD (15, 37). However, we did not find such association in a prior study, which was based on the cohort of the current study, when using the Burden Scale for Family Caregivers as a measurement tool (17). Nevertheless, we now show that impairment of specific areas of the relative's daily activities and social life during caregiving increases the risk of developing PGD_{PG13}.

Reduced extent and time spent on social activities during end-of-life care was also associated with PGD_{PG13}. Lack of social support has been associated with adverse bereavement outcome, but it is not well-established as a bereavement-specific risk factor (15). In a recent cross-sectional study based on network analysis, Maccallum et al. showed that social functioning (measured by the WHO Quality of Life scale) was associated with the PGD_{PG13} item *lack of trust in other* (14). The cross-sectional design did not allow for causal inference in the network analysis, whereas the present findings point at reduced social activities prior to bereavement as an *interpersonal* risk factor for PGD_{PG13}.

However, both a lack of social network before the patient's illness trajectory and reduced social activities due to end-of-life care may affect social life before bereavement and contribute to the development of PGD. The present study lacks information on social network before the illness trajectory as a contributing factor. Yet, the low *social functioning* scores in the study sample compared to the reference sample indicate that reduced activity because of affected physical and mental health in the end-of-life period is an important factor.

Strengths and Limitations

The strengths of the study are the population-based and prospective study design, the use of validated scales (SF-36 and PG-13), and the use of Danish registers with low levels of missing data. Additional advantages were the large sample size and the systematically collected data, although some selection bias might have been present. We contacted patients who had recently been registered under the public drug reimbursement scheme due to terminal illness, and we received completed questionnaires from relatives to 38% of these patients. At baseline, the patients of responding relatives were higher educated than were non-respondents, and relatives responding at follow-up were better educated than were non-respondents. Since high education has been associated with reduced risk of PGD_{PG13} (17), the participants included in the present study might have had better average mental health, and the extent of PGD_{PG13} could thus have been underestimated. Additionally, we expected that

functional impairment would reduce the participation in the study. As the present study sample is likely to have had better functioning than the average population of relatives, the level of functional impairment in the present study might have been underestimated. However, we believe that selection bias is unlikely to have affected the association found between functional impairment and PGD_{PG13}.

No comparison sample was included in this cohort study and this limits direct comparison with non-bereaved individuals. Hence, it was not possible to conclude on the effect of caregiving on functional impairment. Information on patients' symptom burden was not available, which may account for some of the residual confounding of the analysis as symptom burden is likely to impact the function of the participants and could also be associated with PGD. A clinical interview is necessary to diagnose PGD and the use of self-report questionnaires in this study may overestimate symptom levels (38). However, the use of self-report questionnaires enables the large-scale data collection in a general population caregiver cohort. Another shortcoming was that the measure of overall functional impairment (the PG functional impairment item) was a PG-13 item; this limited our opportunity to analyze the associations between overall functional impairment and PGD_{PG13}. Furthermore, we included no specific measure of occupational impairment. Still, the impact on daily activities may better cover the situation of all bereaved persons because loss and bereavement become increasingly common with higher age, and a large number of bereaved persons is likely to have retired. In the present study cohort, up to half of the participants had retired before the bereavement (20). Nevertheless, future studies should investigate the impact of bereavement on occupation in the group of bereaved persons in the work force.

We used the PG impairment item to assess overall functional impairment. This measure is broad and includes both occupational, social, and other impairment in a single item. Still, the PG impairment item may not fully capture ICD-11 functional impairment, which also includes impairment regarding personal, family, and educational functioning. Thus, the used measure could have underestimated the level of functional impairment. Furthermore, the PGD_{PG13} scale was developed in 2009 before the inclusion of PGD_{ICD11} as a diagnosis in 2018. Hence, the PGD_{PG13} is not a diagnostic tool and it does not fully account for the PGD_{ICD11} (38). The core symptom of intense yearning and half of the symptoms of intense emotional pain highlighted in the PGD_{ICD11} and functional impairment are covered using the PGD_{PG13}. Still, the PGD_{PG13} was found to identify 59% of clinical cases verified in a Structured Clinical Interview for Complicated Grief (SCI-CG), whereas 95.8% was identified using the PGD_{ICD11} (39). This may limit the generalizability of our results to clinical care. However, the association between functional impairment and PGD_{PG13} is independent of the differences between PGD_{PG13} and PGD_{ICD11}. Thus, our findings that impaired daily functioning due to emotional problems and impaired social functioning is associated with developing high levels of grief symptoms still shed light on areas of interest for clinical interventions during caregiving. In line with prior studies based on the study sample (17, 20), the findings may be

generalized to populations of family caregivers in similar health care settings.

Clinical Implications

Temporary functional impairment due to grief may improve over time in most persons as the grief reactions gradually lessen. Nevertheless, several bereaved persons in this sample had functional impairment for years after bereavement. Hence, healthcare interventions to restore the daily functioning may be necessary for some groups. Preventive interventions from health professionals before bereavement should focus on maintaining the daily functioning and social networks, which could also benefit the severely ill patient.

Functional impairment is a frequent reason for seeking healthcare (3), and health professionals should investigate for underlying mental illness. Prior studies have shown that PGD_{PG13} (40), persistently high grief symptom levels (8, 41), depressive symptoms (42), self-harm, suicide, and psychiatric illness (43) were long-term adverse bereavement outcomes. Since functional impairment before bereavement was associated with PGD_{PG13}, health professionals need to direct attention toward these persons at an early time point during caregiving to help them balance risk factors and prevent PGD development.

The new knowledge gained from the current study may support the clinical usefulness of functional impairment in the PGD_{ICD11} criteria. However, the PGD_{ICD11} do not include specific criteria to assess functional impairment; they rely on a thorough clinical assessment by a physician. Clinicians have recommended such flexibility to be able to consider PGD in bereaved persons maintaining a high level of functioning. Thus, assessment of both symptoms and daily functioning is crucial in the clinical care for bereaved persons.

Future Research

Future studies need to investigate the role of functional impairment after bereavement, including occupational impairment such as sick leave due to bereavement and impairment in bodily function. Studies are called for to explore whether bereaved persons would benefit from early supportive intervention to maintain their daily functioning and their social network already before bereavement. Such studies should also address the crucial question: Could development of PGD be prevented by promoting a better balance in daily life?

CONCLUSION

The present study establishes functional impairment as an important risk factor in bereavement and functional impairment was common both before and after bereavement. Although functional impairment was reduced over time, the loss may affect the daily activities of bereaved persons for a long time. Impairment in both daily work/activity aspects and social aspects before bereavement was associated with the development of PGD_{PG13}. Hence, bereaved persons may benefit from early supportive intervention to maintain their daily functioning and social network already before bereavement.

Large groups with functional impairment were identified at 6 months and 3 years after bereavement, but only small proportions had PGD_{PG13}. Hence, functional impairment call for clinical assessment of mental well-being, as it may be related to development of conditions such as depression, or it could be an independent long-term consequence of bereavement. Future research is needed to further establish the role of functional impairment in bereavement, including impairment in bereaved persons in the work force and potential associations with other adverse bereavement outcomes.

DATA AVAILABILITY STATEMENT

The datasets generated for this study will not be made publicly available. Data sharing is not possible due to protection of personal information.

ETHICS STATEMENT

The Committee on Health Research Ethics of the Central Denmark Region confirmed that this study required no ethical clearance according to the Danish Act on Research Ethics Review of Health Research Projects (24). The study was approved by the Danish Data Protection Agency (file no. 2013-41-2603) and registered in the Record of Processing Activities at the Research Unit for General Practice, Aarhus (Id 207) in accordance with the provisions of the General Data Protection Regulation (GDPR). The participants gave written informed consent to participate in this study.

REFERENCES

1. Ustün B, Kennedy C. What is “functional impairment”? Disentangling disability from clinical significance. *World Psychiatry*. (2009) 8:82–5. doi: 10.1002/j.2051-5545.2009.tb00219.x
2. World Health Organization (WHO). *How to Use the ICF. A Practical Manual for Using the International Classification of Functioning, Disability and Health (ICF)*. Geneva: WHO (2013).
3. Weiss MD, McBride NM, Craig S, Jensen P. Conceptual review of measuring functional impairment: findings from the weiss functional impairment rating scale. *Evid Based Ment Health*. (2018) 21:155–64. doi: 10.1136/ebmental-2018-300025
4. World Health Organization (WHO). *ICD-11 for Mortality and Morbidity Statistics. 6B42 Prolonged grief disorder*. (2019). Available online at: <https://icd.who.int/browse11/l-m/en#/http://id.who.int/icd/entity/1183832314> (accessed November 24, 2020).
5. First MB, Reed GM, Hyman SE, Saxena S. The development of the iCD-11 clinical descriptions and diagnostic guidelines for mental and behavioural disorders. *World Psychiatry*. (2015) 14:82–90. doi: 10.1002/wps.20189
6. Killikelly C, Maercker A. Prolonged grief disorder for iCD-11: the primacy of clinical utility and international applicability. *Eur J Psychotraumatol*. (2018) 8:1–9. doi: 10.1080/2008198.2018.1476441
7. Lichtenthal WG, Maciejewski PK, Craig Demirjian C, Roberts KE, First MB, Kissane DW, et al. Evidence of the clinical utility of a prolonged grief disorder diagnosis. *World Psychiatry*. (2018) 17:364–5. doi: 10.1002/wps.20544
8. Nielsen MK, Carlsen AH, Neergaard MA, Bidstrup PE, Guldin M. Looking beyond the mean in grief trajectories: a prospective, population- based cohort study. *Soc Sci Med*. (2019) 232:460–9. doi: 10.1016/j.socscimed.2018.10.007

AUTHOR CONTRIBUTIONS

MNi, KC, MNe, PB, and M-BG designed the study. MNi undertook the statistical analysis, wrote the first draft of the manuscript, and M-BG commented on the first draft. All authors contributed to the revised version and approved the final manuscript.

FUNDING

This work was supported by the Novo Nordisk Foundation (grant no. NNF17OC0024410) and the Danish Cancer Society (grant no. R160-A10389-16-S3).

ACKNOWLEDGMENTS

We deeply thank all participants. We also wish to thank IT supporter Hanne Beyer, data manager Kaare Flarup, statistician Henrik Schou Petersen, and language editor Lone Niedziella; all at the Research Unit for General Practice, Aarhus. Furthermore, we thank statistician Anders Helles Carlsen, Aarhus University Hospital - Psychiatry, for his work on grief trajectories.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: <https://www.frontiersin.org/articles/10.3389/fpsy.2020.537674/full#supplementary-material>

9. Boelen PA, Lenferink LIM. Symptoms of prolonged grief, posttraumatic stress, and depression in recently bereaved people: symptom profiles, predictive value, and cognitive behavioural correlates. *Soc Psychiatry Psychiatr Epidemiol*. (2020) 55:765–77. doi: 10.1007/s00127-019-01776-w
10. Kristensen P, Weisæth L, Hussain A, Heir T. Prevalence of psychiatric disorders and functional impairment after loss of a family member: a longitudinal study after the 2004 tsunami. *Depress Anxiety*. (2015) 32:49–56. doi: 10.1002/da.22269
11. Melhem NM, Porta G, Shamseddeen W, Payne MW, Brent DA. The course of grief in children bereaved by sudden parental death. *Arch Gen Psychiatry*. (2011) 68:911–9. doi: 10.1001/archgenpsychiatry.2011.101
12. Geronazzo-Alman L, Fan B, Duarte CS, Layne CM, Wicks J, Guffanti G, et al. The distinctiveness of grief, depression, and posttraumatic stress: lessons from children after 9/11. *J Am Acad Child Adolesc Psychiatry*. (2019) 58:971–82. doi: 10.1016/j.jaac.2018.12.012
13. Boelen PA, Smid GE, Mitima-Verloop HB, Keijsers J De, Lenferink LIM. Patterns, predictors, and prognostic validity of persistent complex bereavement disorder symptoms in recently bereaved adults: a latent class analysis. *J Nerv Ment Dis*. (2019) 207:913–20. doi: 10.1097/NMD.0000000000001053
14. Maccallum F, Bryant RA. A network approach to understanding quality of life impairments in prolonged grief disorder. *J Trauma Stress*. (2020) 33:106–15. doi: 10.1002/jts.22383
15. Stroebe MS, Folkman S, Hansson RO, Schut H. The prediction of bereavement outcome: development of an integrative risk factor framework. *Soc Sci Med*. (2006) 63:2440–51. doi: 10.1016/j.socscimed.2006.06.012
16. Stroebe M, Schut H, Stroebe W. Health outcomes of bereavement. *Lancet*. (2007) 370:1960–73. doi: 10.1016/S0140-6736(07)61816-9

17. Nielsen MK, Neergaard MA, Jensen AB, Vedsted P, Bro F, Guldin MB. Predictors of complicated grief and depression in bereaved caregivers: a Nationwide prospective cohort study. *J Pain Symptom Manage.* (2017) 53:540–50. doi: 10.1016/j.jpainsymman.2016.09.013
18. Prigerson HG, Horowitz MJ, Jacobs SC, Parkes CM, Aslan M, Goodkin K, et al. Prolonged grief disorder: psychometric validation of criteria proposed for dSM-V and iCD-11. *PLoS Med.* (2009) 6:e1000121. doi: 10.1371/journal.pmed.1000121
19. Ware JE, Sherbourne CD. The MOS 36-item short-form health survey (SF-36): I. Conceptual framework and item selection. *Med Care.* (1992) 30:473–83. doi: 10.1097/00005650-199206000-00002
20. Nielsen MK, Neergaard MA, Jensen AB, Bro F, Guldin MB. Psychological distress, health, and socio-economic factors in caregivers of terminally ill patients: a nationwide population-based cohort study. *Support Care Cancer.* (2016) 24:3057–67. doi: 10.1007/s00520-016-3120-7
21. Nielsen MK, Neergaard MA, Jensen AB, Vedsted P, Bro F, Guldin MB. Pre-loss grief in family caregivers during end-of-life cancer care: a nationwide population-based cohort study. *Psychooncology.* (2017) 26:2048–56. doi: 10.1002/pon.4416
22. Lenger MK, Neergaard MA, Guldin M, Nielsen MK. Poor physical and mental health predicts prolonged grief disorder: a prospective, population-based cohort study on caregivers of patients at the end of life. *Palliat Med.* (2020) 34:1416–24. doi: 10.1177/0269216320948007
23. Johannesdottir SA, Horváth-Puhó E, Ehrenstein V, Schmidt M, Pedersen L, Sørensen HT. Existing data sources for clinical epidemiology: the danish national database of reimbursed prescriptions. *Clin Epidemiol.* (2012) 4:303–13. doi: 10.2147/CLEP.S37587
24. The Danish national committee on health research Ethics. *Act on Research Ethics Review of Health Research Projects.* (2018). Available online at: <https://en.nvk.dk/rules-and-guidelines/act-on-research-ethics-review-of-health-research-projects> (accessed November 24, 2020).
25. Bjørner JB, Damsgaard MT, Watt T, Bech P, Rasmussen NK, Kristensen TS, et al. *Dansk Manual Til SF-36 [Danish manual for SF-36]*. Copenhagen: Lif [Lægemedellindustriforeningen] (1997).
26. Ware JE, Gandek B. Overview of the SF-36 health survey and the international quality of life assessment (IQOLA) project. *J Clin Epidemiol.* (1998) 51:903–12. doi: 10.1016/S0895-4356(98)00081-X
27. World Health Organisation (WHO). *Process of Translation and Adaptation of Instruments.* Available online at: https://www.who.int/substance_abuse/research_tools/translation/en/ (accessed November 24, 2020).
28. Tomarken A, Holland J, Schachter S, Vanderwerker L, Zuckerman E, Nelson C, et al. Factors of complicated grief pre-death in caregivers of cancer patients. *Psychooncology.* (2008) 17:105–11. doi: 10.1002/pon.1188
29. Beck AT, Steer RA, Brown GK. *Manual: Beck Depression Inventory.* 2nd ed. The Psycho. San Antonio, Tx: Hartcourt & Brace (1996). doi: 10.1037/t00742-000
30. Schmidt M, Pedersen L, Sørensen HT. The danish civil registration system as a tool in epidemiology. *Eur J Epidemiol.* (2014) 29:541–9. doi: 10.1007/s10654-014-9930-3
31. Schmidt M, Schmidt SAJ, Adelborg K, Sundbøll J, Laugesen K, Ehrenstein V, et al. The danish health care system and epidemiological research: from health care contacts to database records. *Clin Epidemiol.* (2019) 11:563–91. doi: 10.2147/CLEP.S179083
32. Samsa G, Edelman D, Rothman ML, Williams GR, Lipscomb J, Matchar D. Determining clinically important differences in health status measures: a general approach with illustration to the Health Utilities Index Mark II. *Pharmacoeconomics.* (1999) 15:141–55. doi: 10.2165/00019053-199915020-00003
33. Shear MK. Clinical practice. Complicated grief. *N Engl J Med.* (2015) 372:153–60. doi: 10.1056/NEJMcP1315618
34. Stoltzfus JC. Logistic regression: a Brief primer. *Acad Emerg Med.* (2011) 18:1099–104. doi: 10.1111/j.1553-2712.2011.01185.x
35. Ochoa CY, Lunsford NB, Smith JL. Impact of informal cancer caregiving across the cancer experience: a systematic literature review of quality of life. *Palliat Support Care.* (2020) 18:220–40. doi: 10.1017/S1478951519000622
36. Breen LJ, Aoun SM, O'Connor M, Johnson AR, Howting D. Effect of caregiving at end of life on grief, quality of life and general health: a prospective, longitudinal, comparative study. *Palliat Support Care.* (2020) 34:145–54. doi: 10.1177/0269216319880766
37. Alam S, Hannon B, Zimmermann C. Palliative care for family caregivers. *J Clin Oncol.* (2020) 38:926–36. doi: 10.1200/JCO.19.00018
38. Lenferink LIM, Boelen PA, Smid GE, Paap MCS. The importance of harmonising diagnostic criteria sets for pathological grief. *Br J psychiatry.* (2019) 1–4. doi: 10.1192/bjp.2019.240
39. Mauro C, Shear MK, Reynolds CF, Simon NM, Zisook S, Skritskaya N, et al. Performance characteristics and clinical utility of diagnostic criteria proposals in bereaved treatment-seeking patients. *Psychol Med.* (2017) 47:608–15. doi: 10.1017/S0033291716002749
40. Zordan RD, Bell ML, Price M, Remedios C, Lobb E, Hall C, et al. Long-term prevalence and predictors of prolonged grief disorder amongst bereaved cancer caregivers: a cohort study. *Palliat Support Care.* (2019) 17:507–14. doi: 10.1017/S1478951518001013
41. Bonanno GA, Malgaroli M. Trajectories of grief: comparing symptoms from the dSM-5 and iCD-11 diagnoses. *Depress Anxiety.* (2019) 2019:1–9. doi: 10.1002/da.22902
42. Galatzer-Levy IR, Bonanno GA. Beyond normality in the study of bereavement: heterogeneity in depression outcomes following loss in older adults. *Soc Sci Med.* (2012) 74:1987–94. doi: 10.1016/j.socscimed.2012.02.022
43. Guldin MB, Kjaersgaard MIS, Fenger-Gron M, Parner ET, Li J, Prior A, et al. Risk of suicide, deliberate self-harm and psychiatric illness after the loss of a close relative: a nationwide cohort study. *World Psychiatry.* (2017) 16:193–9. doi: 10.1002/wps.20422

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Copyright © 2020 Nielsen, Christensen, Neergaard, Bidstrup and Guldin. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.



Feasibility of Present-Centered Therapy for Prolonged Grief Disorder: Results of a Pilot Study

Anna Vogel^{1*}, Hannah Comtesse¹, Agnes Nocon², Anette Kersting³, Winfried Rief⁴, Regina Steil⁵ and Rita Rosner¹

¹ Department of Psychology, Catholic University Eichstaett-Ingolstadt, Eichstaett, Germany, ² Faculty of Applied Healthcare Science, Deggendorf Institute of Technology, Deggendorf, Germany, ³ Department of Psychosomatic Medicine and Psychotherapy, University of Leipzig, Leipzig, Germany, ⁴ Department of Clinical Psychology and Psychotherapy, Philipps-University of Marburg, Marburg, Germany, ⁵ Department of Clinical Psychology and Psychotherapy, Institute of Psychology, Goethe University, Frankfurt, Germany

OPEN ACCESS

Edited by:

Clare Killikelly,
University of Zurich, Switzerland

Reviewed by:

Rolf J. Kleber,
Utrecht University, Netherlands

Lene Larsen,
Independent Researcher,
Copenhagen, Denmark

*Correspondence:

Anna Vogel
anna.vogel@ku.de

Specialty section:

This article was submitted to
Mood and Anxiety Disorders,
a section of the journal
Frontiers in Psychiatry

Received: 13 February 2020

Accepted: 15 March 2021

Published: 15 April 2021

Citation:

Vogel A, Comtesse H, Nocon A,
Kersting A, Rief W, Steil R and
Rosner R (2021) Feasibility of
Present-Centered Therapy for
Prolonged Grief Disorder: Results of a
Pilot Study.
Front. Psychiatry 12:534664.
doi: 10.3389/fpsy.2021.534664

Present-centered therapy (PCT) was originally developed as a strong comparator for the non-specific effects of psychotherapy in the treatment of posttraumatic stress disorder. PCT qualifies as a not strictly supportive treatment as it is structured and homework is assigned between sessions. It does not focus on cognitive restructuring or exposure. A growing body of literature supports its beneficial effects. For example, it demonstrated only slightly inferior effect sizes and lower dropout rates compared to that of trauma-focused cognitive behavioral therapy in several trials with patients suffering from posttraumatic stress disorder. The current study is the first to evaluate the feasibility and the treatment effects of PCT in adults with prolonged grief disorder (PGD). Meta-analyses on psychotherapy for PGD have yielded moderate effect sizes. $N = 20$ individuals suffering from PGD were treated with PCT by novice therapists as part of a preparation phase for an upcoming RCT in an outpatient setting. Treatment consisted of 20–24 sessions á 50 min. All outcomes were assessed before treatment, at post-treatment, and at the 3-month follow-up. The primary outcome, PGD symptom severity, was assessed using the Interview for Prolonged Grief-13. Secondary outcomes were self-reported PGD severity, depression, general psychological distress, and somatic symptom severity. Furthermore, therapists evaluated their experiences with their first PCT patient and the treatment manual. In intent-to-treat analyses of all patients we found a significant decrease in interview-based PGD symptom severity at post-treatment ($d = 1.26$). Decreases were maintained up to the 3-month follow-up assessment ($d = 1.25$). There were also significant decreases in self-reported PGD symptoms, depression, and general psychological distress. No changes were observed for somatic symptoms. The completion rate was 85%. Therapists deemed PCT to be a learnable treatment program that can be adapted to the patient's individual needs. The preliminary results of PCT as a treatment for PGD demonstrate large effects and indicate good feasibility in outpatient settings. The treatment effects were larger than those reported in meta-analyses. Thus, PCT is a promising treatment for PGD. Possible future research directions are discussed.

Keywords: prolonged grief disorder, loss, psychotherapy, bereavement, present-centered therapy

INTRODUCTION

Prolonged grief disorder (PGD) has emerged as a well-defined mental disorder, distinguishable from major depression and posttraumatic stress disorder (PTSD) or other stress-related disorders (1). It has now been included in the ICD-11 (2), with slightly different criteria to those of its counterpart in the DSM-5, the “persistent complex bereavement disorder” (3). Earlier concepts of PGD encompassed complicated or traumatic grief [e.g., (4, 5)]. The core symptoms of all concepts are intense yearning and preoccupation with the deceased; reactive distress symptoms, such as feeling stunned or shocked by the loss; avoidance of reminders of the reality of the loss and emotional numbing, and finally social/identity disruption, for instance feeling detached or finding it difficult to trust others (6). The symptoms and impairment have to persist for more than 6 months after the death of a significant other.

PGD rates have varied considerably across studies due to methodological heterogeneity, sample demographic features, and loss-related characteristics. While a recent meta-analysis has found that 1 in 10 bereaved adults following non-violent death of a loved one suffer from PGD (7), representative studies report lower prevalences [e.g., 7% conditional prevalence in (8)]. Higher prevalences are associated with unnatural losses, with nearly half of the bereaved persons experiencing PGD (9). PGD has been found to be associated with both psychological and physical morbidity, such as impaired quality-of-life (10), increased risk of comorbid disorders with high rates of depression, PTSD, and anxiety disorders (11), increased suicidality (12) and deteriorated health (13). The negative consequences of PGD indicate a need for efficacious treatments. Still, there are relatively few controlled studies examining psychological treatments for PGD. Boelen and Smid (14) list three recommended psychological therapies that have been tested in at least two independent and controlled studies, including “complicated grief treatment” [including elements of exposure, cognitive restructuring, and interpersonal therapy; (15–17)], cognitive behavioral therapy [CBT; combining exposure and cognitive interventions; (18, 19)], and internet-based CBT [encompassing exposure, cognitive interventions, and behavioral activation applied using writing assignments; (20, 21)].

Meta-analyses on the treatment of complicated, traumatic or prolonged grief yielded effect sizes (ESs) for grief outcomes between 0.53 for those with clinically relevant symptoms (22), 0.53 for those undergoing psychotherapy (23), and 0.45 overall, and 0.58, when only considering those treated who were minimally 6 months post-loss (24). The respective ESs for depressive symptoms ranged between 0.16 (22) and 0.35 (24). ESs for general mental distress were reported as small [$d = 0.26$ in (24)]. In recent reviews (14, 24) on PGD treatment research, it was found that most interventions were grief-specific approaches and used exposure, cognitive restructuring, behavioral activation, and elements of interpersonal therapy. The authors also state that there are too few randomized controlled trials (RCTs) with active controls and that dismantling studies are missing. Consequently, no conclusions can be drawn about the active ingredients of successful treatment manuals.

Present-Centered Therapy [PCT; (25)] was originally developed as a strong comparator for the non-specific effects of psychotherapy in the treatment of PTSD. The goals of PCT are to enhance interpersonal connectedness, improve patients’ insight into their current symptoms, and promote a greater sense of mastery via use of effective approaches to solving problems. Therefore, it involves empathic listening and support from the therapist as well as the basic components of behavioral therapy, namely education about the links between symptoms and daily problems, and the fostering of problem-solving skills, including homework exercises. The treatment is provided within the context of intentionally compassionate and helpful acts of the therapist based on client-centered principles like offering empathy, unconditional positive regard and congruence and is led by the individual daily stressors and problems the patient presents. PCT excludes specific trauma-focused components (i.e., exposure, cognitive restructuring of dysfunctional beliefs, stress inoculation training), and therefore seems to choose an alternative approach to address avoidance: by actively dealing with the current problems that may have arisen as a result of the traumatic event, but without engaging emotionally with the traumatic event itself.

In an older meta-analysis based on five RCTs, PCT showed good results in PTSD treatment and lower dropout rates than trauma-focused approaches (26). In a recent Cochrane review (27) based on 12 RCTs, PCT was found to be superior to waitlist ($SMD = -0.84$). A comparison of PCT to trauma-focused CBT did not support PCT non-inferiority. ESs differed for PCT and trauma-focused CBT with 0.32 in favor of trauma-focused CBT. PCT resulted in 16% lower dropout rates than trauma-focused CBT. Current treatment guidelines suggest that PCT may be offered as a treatment for PTSD when trauma-focused CBT is either not available or not preferred by the patient (28).

Taken together, PCT can be deemed to be what is known as a bona fide therapy, namely a therapy based on psychological principles containing specific factors (i.e., specific techniques like fostering of problem-solving skills and homework exercises, or promoting a theory of the therapeutic change, namely by client-centered principles of offering empathy, unconditional positive regard and congruence) and delivered by trained professionals [cf. (29, 30)]. It is, therefore, a credible intervention for both patients and therapists. Unlike almost all other interventions that have been examined in patients suffering from PGD so far, PCT does not include exposure or cognitive interventions. Given that patients with PGD, as well as those with PTSD, suffer from avoidance (albeit to slightly different degrees), an intervention that takes a different approach to addressing avoidance seems to represent an interesting alternative to previously studied interventions. Furthermore, on a theoretical basis, by focusing on the active mastery of daily problems and functional coping, one might speculate that PCT resembles restoration-orientation according to the Dual Process Model of coping with bereavement (31). This is why we decided to adapt PCT to the needs of patients suffering from PGD. If PCT for PGD would prove feasible and clinically impactful, it might not only serve as an active bona fide treatment with a different treatment focus most PGD interventions had so far, but it also promises to be an ideal

active control condition in future PGD trials. Therefore, the aim of this study was to evaluate the feasibility and the treatment effects of PCT in adults with PGD, and to explore therapists' and supervisor's experiences with this new treatment.

METHODS

Participants

Participants were treatment-seeking adults aged 18 to 75, whose losses had occurred at least 6 months previously. A primary diagnosis of PGD, as assessed in the Interview for Prolonged Grief-13 [PG-13; (6, 32), see below], was required for inclusion. Because of the ongoing discussion about a multiplicity of different criteria sets for PGD (2, 3, 6, 33, 34), and because the final ICD-11 criteria were not available when we started this trial in 2017, we decided on a compromise between criteria according to Prigerson et al. (6) and the not yet finalized ICD-11 (2). To meet the criteria for PGD in the current study, it was necessary for participants to report at least (a) one separation distress symptom (rated as ≥ 4 on a 5-point-scale: 1 = never/not at all, 5 = several times a day/extremely), (b) four out of nine cognitive, emotional, and behavioral symptoms (each symptom rated as ≥ 4), and (c) significant impairment in social, occupational or other important domains, for 6 months or longer, after the loss according to the PG-13, see below. Patients had to have sufficient cognitive and German language skills, and give their written informed consent. If patients were on antidepressant medication, the treatment regime needed to be stable for at least 4 weeks prior to joining the trial. The exclusion criteria were: (1) current psychotic or severe substance use disorder, or acute suicidality; (2) ongoing psychotherapy; (3) participation in another treatment trial; and (4) continuous treatment with benzodiazepines, antipsychotics, or opioids. Any change in psychotropic medication during the course of the study was continuously monitored.

Procedure

The current trial was an integral part of a preparation phase for an RCT [(35), German Clinical Trials Register, ID: DRKS00012317]. Treatment was offered at four University outpatient mental health clinics in Germany. The study was approved by the Institutional Review Board of the Catholic University Eichstaett-Ingolstadt (2016/21), and by three Institutional Review Boards of the other study centers (Ethics Committee of the Department of Psychology and Sports of the Goethe University Frankfurt, Ethical Committee at the Medical Faculty of Leipzig University, Local Ethics Committee of the Department of Psychology of the University of Marburg). Recruitment efforts included a study website, advertisements in public and social media, newspaper and radio interviews, flyers in family practices, health and community centers, or churches, and informing general and mental health practitioners via mailings, as well as via talks and publications in the specialized press. The first patient started therapy in June 2017, the last patient finished therapy in May 2019.

The trial included assessments at baseline, post-treatment, and at the 3-month follow-up, each comprising the same clinical interviews and self-ratings described below. All assessments

were conducted by trained clinical raters who were blind to the participants' baseline assessment results and treatment progression. Study safety was ensured by monitoring for the incidence of serious adverse events (e.g., suicide attempts, death, occurrence of life-threatening conditions, events that lead to physical disability) using a therapist/rater-administered checklist every treatment session and at post-treatment and follow-up. Participants received a small financial compensation for taking part in the post-treatment and follow-up assessments (€20 for each assessment).

Treatment was administered by 20 study therapists, who were predominantly female (94%), master's level psychologists in advanced postgraduate clinical training (71%) and specialized in CBT (100%). All therapists were novices in PCT and were interested in taking part in an upcoming RCT. They were free in choosing training in PCT or an alternative grief-focused CBT training. All included patients represented each therapist's first training case. All PCT therapists attended a 2-day personal training course in PCT delivered by one of PCT's original authors, Dr. Shea. During the trial, therapists were supervised bi-weekly at the respective study center. In addition, they participated in centralized bi-weekly telephone case consultations to maintain treatment adherence. Therapeutic adherence and competence are currently being evaluated with independent ratings by two raters based on video-documented sessions selected at random.

Outcomes and Measures

The primary outcome was the PGD severity score assessed by the PG-13 in an interview format [(6, 33), German version (36), as published in (37)], which was obtained by calculating the sum of the 11 symptom item scores (range: 11–55). Cutoff scores of 34 and 35 have been suggested for the PG-13 (37, 38). Psychometric evaluation showed good internal consistency [e.g., Cronbach's alphas from 0.83 to 0.93 in (39)]. The PG-13 was also used to assess PGD diagnostic status as defined for the current study (see above) as well as a reliable change in PGD symptoms. In the current sample, the internal consistency of the total severity score (11 items) was 0.71.

Secondary outcomes were assessed that targeted self-reported symptoms of prolonged grief, depression, somatoform symptoms, and general mental distress.

Prolonged grief symptoms were also measured using the self-report measure Inventory of Complicated Grief [ICG; (40), German version ICG-D; (41)]. Participants were asked to rate the extent to which they had experienced 19 grief symptoms during the previous month on a 5-point scale ranging from 0 = never to 4 = all the time. A prolonged grief score was computed (range: 0–76), Cronbach's alpha was 0.70. Prigerson et al. (40) suggested an ICG score >25 as the threshold for distinguishing syndromal from subsyndromal levels of PGD. However, later studies [e.g., (42, 43)] used a cutoff score of ≥ 30 as a more conservative threshold to identify clinically significant cases.

Self-reported depressive symptoms were measured using the German version of the Beck Depression Inventory II [BDI-II; (44)]. The 21 items refer to symptoms of depression during the previous 2 weeks and are rated on a 4-point scale, resulting in a

total depression score ranging from 0–63. Cronbach's alpha in the current sample was 0.85.

As we had found a high level of somatoform symptoms in two earlier studies (45, 46), we decided to include a measure to specifically address somatoform complaints. The Screening for Somatoform Disorders [SOMS-7D; (47)] was used to assess self-reported somatoform symptoms. Participants were asked to rate the extent to which they had suffered from 53 somatoform symptoms during the previous 7 days on a 5-point scale (0 = not at all, 4 = very much). A somatization severity index was calculated ranging from 0–208. Cronbach's alpha was 0.89 in the current sample.

Self-reported general mental distress was measured using the Global Severity Index (GSI) from the German version of the Brief Symptom Inventory [BSI; (48)]. The BSI is a widely used 53-item measure of subjective distress caused by psychological and somatic symptoms over the previous seven days. Responses are scored on a 5-point scale (0 = not at all, 4 = extremely). The GSI is calculated using the sums for the nine subscales plus the four additional items, and divided by the total number of items to which the individual responded (score range: 0–4). In the current sample, Cronbach's alpha of the BSI-GSI was 0.94.

To obtain information about the study therapists' evaluations of PCT, we asked all study therapists to fill in an online questionnaire after having completed their first treatment case. We obtained ratings on four subscales: beliefs and attitudes about the intervention (i.e., individuals' attitudes toward and value placed on the intervention); design quality (i.e., perceived excellence in how the intervention is bundled, presented, and assembled); adaptability and trialability (i.e., degree to which an intervention can be adapted, tailored, refined, or reinvented to meet local needs and ability to test the intervention on a small scale in the organization); and resources and access to knowledge (i.e., level of resources dedicated for implementation and on-going operations and ease of access to digestible information and knowledge about the intervention and how to incorporate it into work tasks). For this questionnaire, we used items by Cook et al. (49) or generated items based on the framework model of Damschroder et al. (50). The Consolidated Framework for Implementation Research by Damschroder et al. (50) includes the most common concepts from published implementation theories. The framework is designed to allow researchers to select the concepts most relevant for their particular setting and use them to evaluate the implementation process. As we were primarily concerned with implementing a new intervention, PCT, within the well-established structures of four University outpatient clinics, we selected concepts relating to the characteristics of the intervention, individual characteristics and the inner setting of our clinics to evaluate the implementation process of PCT. Therefore, for the purpose of this study, we created four subscales to evaluate the implementation process of PCT from the viewpoint of therapists. Overall, the questionnaire consisted of 23 items to be rated on a 5-point scale (1 = I do not agree at all, 5 = I agree fully). Mean scores were calculated for each of the four subscales (mean score range: 1–5). One item on barriers to the implementation of PCT also included the option of giving an answer in an open format, if applicable. In the current

sample, the internal consistencies of the beliefs and attitudes about the intervention scale (9 items; $\alpha = 0.78$), the design quality scale (3 items; $\alpha = 0.80$), and the resources and access to knowledge scale (5 items; $\alpha = 0.78$) were good. The internal consistency of the adaptability and trialability scale (4 items; $\alpha = 0.70$) was acceptable. See **Supplementary Material 1** for the questionnaire.

The presence of comorbid mental disorders according to DSM-IV criteria at baseline was determined using the Structured Clinical Interview for DSM-IV Axis I [(51), German version: (52)]. DSM-IV criteria were used, because no validated German version for the Structured Clinical Interview for DSM-5 was available in 2017 yet. Interviewer-rated acute suicidality was assessed using the Columbia-Suicide Severity Rating Scale [C-SSRS; (53)]. The five items in the intensity of suicide ideation subscale were used to rate the intensity of current suicide ideation on 5- and 6-point scales (score range: 2–25).

Intervention

The PCT manual for PTSD (25) was adapted in cooperation with Dr. Shea for its use with PGD patients (54). To adapt PCT for treating PGD in Germany, we made the following modifications: (1). In order to make PCT a credible intervention for both patients and therapists, session length and number were adapted to the standard health insurance coverage for outpatient CBT treatments in Germany, that is, 20 50-min sessions (as compared to 10 100-min sessions). (2). Educating the patient at the beginning of treatment focused on grief-specific topics including grief symptoms and their relation to problems in day-to-day life. Furthermore, therapists were allowed to collect information relevant to the loss in the first sessions in order to establish a therapeutic relationship. (3). Up to four additional optional sessions were possible to handle special occasions or needs (e.g., suicidality, dealing with anniversaries). Altogether, a maximum of 24 therapy sessions was possible according to the protocol, see **Table 1** for an overview. Besides educating the patients on grief symptoms, PCT did not include any grief-specific cognitive-behavioral components (e.g., exposure, cognitive restructuring of dysfunctional beliefs, instructions to do specific homework). Its focus was on the daily monitoring of stressors and problems in their relation to PGD and on their active mastery. The therapists provided support and established an empathic relationship. This encouraged the expression of thoughts or feelings and explicitly focused on factors of client-centered therapy. Therefore, PCT has elements of supportive therapy, but is a more structured approach that follows a manual and includes the use of a diary to record problems throughout the week. It did not, however, use any active interventions except for giving information, pointing out themes, or other ways of fostering functional coping and the patient's problem-solving skills. If the patient brought up problems regarding the loss, discussions on the loss itself were avoided in favor of focusing on how to better cope with symptoms in daily life resulting from the loss. The aim was to achieve greater insight and support regarding the consequences of the loss. When patients were emotionally distressed during sessions, therapists acted in an

TABLE 1 | Treatment protocol of PCT adapted for PGD.

Session number	Treatment strategies
1–2	Present an overview of the program, discuss the general rationale for the treatment approach (aims of the study, session frequency, etc.), answer client's questions and identify areas of concern, collect information relevant to the loss using a PGD interview guideline. Homework: filling-in relevant forms.
2–3	Ask about the reaction to the first session, present agenda for the current session, educate the patient about frequent reactions to loss. Homework: reading the handout on frequent reactions to loss.
3–4	Present the agenda for the current session, clarify the background and the methods of the treatment, explain the use of daily monitoring in the diary card (recording of events or problems). Homework: filling-in the diary card, reading the handout on the treatment.
4–17	Revise the diary card, establish the agenda for the current session, problem-solving focused on difficulties identified by the patient. Homework: filling-in the diary-card.
18	Follow format for session 4, prepare the patient for impending termination.
19	Follow format for session 4.
20	Review the past week, review the progress in treatment, terminate therapy saying goodbye.

PCT, present-centered therapy; PGD, prolonged grief disorder.

empathic, compassionate and helpful manner and promoted functional problem-solving when appropriate.

Data Analyses

All primary and secondary outcome analyses were performed as intent-to-treat (ITT) analyses. We used the last observation carried forward (LOCF) procedure to replace missing values due to participants dropping out of the study. To examine the effects of PCT on primary and secondary outcomes, we conducted a repeated-measures multivariate analysis of variance (MANOVA) with measurement time points (pretreatment, post-treatment, follow-up) as a within-subject factor. To assess the effects on each of the outcome measures, repeated measures analyses of variance (ANOVAs) were performed with three measurement time points *post hoc*. The significance level for all analyses was set to $\alpha = 0.05$ (2-tailed). As analyses were considered in an exploratory manner, the significance level was not adjusted for multiple tests. Cohen's *d* ES was calculated for within group pre-post comparisons. Cochran's *Q* test, which is a generalization of the McNemar test for more than two measurement time points, was applied to investigate the change in diagnostic status with respect to PGD (55). Statistics were calculated using IBM SPSS Statistics 25 for Windows. The criterion for a clinically reliable change in the PG-13 severity score according to Jacobson and Truax (56) was calculated on the basis of Cronbach's α of the PG-13 severity score in the current study, as proposed by Martinovich

et al. (57). Thus, clinically reliable improvement was defined as a reduction of more than 6.88 points in the PG-13 severity score.

The open format answers regarding barriers to implementation of the PCT survey were analyzed and summarized by creating condensed meaning units according to Bengtsson (58).

RESULTS

Participant Flow

We screened 32 individuals for eligibility; 12 of them did not meet the study criteria. Six of them had not been clinically diagnosed as having PGD. See **Figure 1** for participant flow. One eligible candidate for treatment declined. As a result, 20 participants began treatment with PCT. Their ages ranged from 37 to 74, with $n = 3$ aged under 50.

Most participants were women (80%) and currently employed (50%). Fifteen participants had completed secondary education only (≤ 12 years), and five had been to college (> 12 years). The majority of participants had lost a partner (45%), or child (35%). The deaths were mostly natural (75%) but unexpected (50%). The mean time since loss was 48.8 months ($Mdn = 27.0$; range: 6–337). All patients met the criteria for PGD diagnostic status as defined for the current study.

At baseline, 16 participants (80%) met the criteria of at least one comorbid psychiatric disorder with an average of $M = 1.00$ ($SD = 0.65$) in addition to PGD. For further details, see **Table 2**.

Of all the participants who began treatment with PCT, three participants (15%) discontinued treatment prematurely after session 5, session 7, and session 12, respectively. In all of these cases, treatment was terminated because of a lack of treatment motivation. In two of these cases the reasons for dropout reported by the respective therapists were relatively long travel distances coupled with low motivation for change. In the third case a very low motivation to attend therapy sessions was mentioned. We were able to obtain further data from one of the participants who dropped out at post-treatment. Another participant did not complete the post-treatment self-report ratings, and data of the SOMS-7D of one further participant at the follow-up assessment was missing.

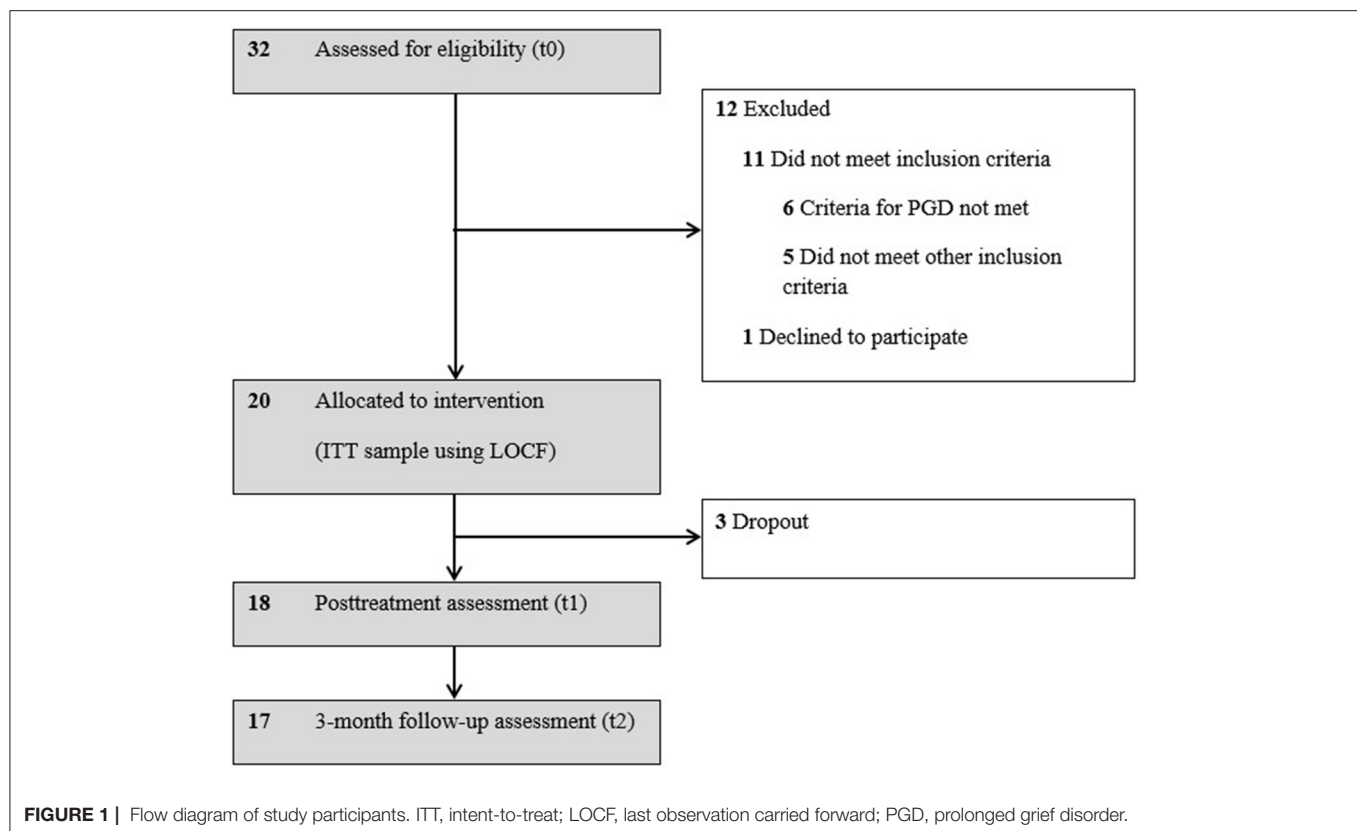
The mean duration of treatment was 23.65 weeks ($SD = 7.10$), with an average of $M = 18.70$ ($SD = 4.99$) sessions provided. Neither suicidal crises nor other serious adverse events occurred during the intervention or up to the 3-month follow-up.

Treatment Outcome

The repeated measures MANOVA of the primary (PG-13) and secondary outcome measures (ICG, BDI-II, BSI-GSI, SOMS-7D) calculated on the basis of the ITT sample ($N = 20$), demonstrated a significant effect of time, $F_{(10,108)} = 2.08$, $p = 0.032$.

Repeated measures ANOVA revealed a significant large effect for the total severity score of interview-rated PGD symptoms (PG-13) from pre- to posttreatment with $d = 1.26$. Improvements remained stable at the 3-month follow-up, $d = 1.25$. See **Table 3** for the results of respective ANOVAs and effect sizes.

At post-treatment, 15 participants (75%) no longer met the PGD criteria according to the PG-13, and 13 participants (65%)



at the 3-month follow-up. Three of the five cases who still met the PGD criteria after treatment, were the participants who had dropped out. The change in PGD diagnostic status over time was significant, Cochran's $Q(2) = 24.57$, $p < 0.001$. At post-treatment, 10 participants (50%) met the criteria for reliable change according to the PG-13 score, whereas 11 participants (55%) met the criteria for reliable change at the 3-month follow-up. No clinically relevant worsening of symptoms was observed. Throughout the trial, no serious adverse event was reported.

The results regarding self-reported PGD symptoms (ICG) revealed a significant large effect from pre- to post-treatment with $d = 1.02$, see **Table 3**. The ES remained large at the 3-month follow-up, $d = 0.90$. However, total sum scores at posttreatment as well as 3-month follow up remained at or above more conservative cutoff scores used in other studies [e.g., (43)].

Participants improved significantly from pre- to post-treatment and up to the 3-month follow-up with regard to BDI-II and BSI-GSI. ESs for improvements of depressive symptoms and general mental distress were medium to large, ranging from 0.54 to 0.86. There were no significant improvements with regard to somatoform symptoms as assessed by the SOMS-7D.

Evaluation of the Study Therapists' Perspective

Of all the PCT study therapists, 17 (85%) responded to our online questionnaire after having completed their first treatment case, including the three therapists whose patients discontinued

treatment prematurely. Their demographic characteristics are given in **Table 4**.

The mean scores of the four subscales of the evaluation of PCT can be considered as medium to high, with $M = 3.65$ ($SD = 0.54$) regarding beliefs and attitudes about the intervention, $M = 3.94$ ($SD = 0.64$) regarding design quality, $M = 3.94$ ($SD = 0.64$) regarding adaptability and trialability, and $M = 3.95$ ($SD = 0.74$) regarding resources and access to knowledge. Therefore, PCT adapted for PGD as evaluated by the study therapists seems to be an adaptable and learnable intervention of good design quality that was evaluated positively by CBT therapists.

Only nine therapists (53%) reported at least one barrier to administering PCT with their pilot cases. The reported barriers were grouped into five categories. In three cases (17.6%), therapists reported that features of a personality disorder or rigid behavior patterns complicated implementing the manual with their patient. Three categories were each mentioned by two therapists (11.8%) as a barrier: patient's lack of motivation for change or reactance, lack of permission to use typical CBT methods (like cognitive restructuring, exposition), and problems during treatment because the patient would have needed clearly defined goals. One therapist (5.9%) reported the PCT manual as being not specific enough as a further problem when administering PCT. Two of these categories (personality features and rigid behavior patterns, reactance from the patient and lack of motivation for change) seem to be associated with the individual patient's characteristics and might not, therefore,

TABLE 2 | Sociodemographic and loss-related characteristics of study participants in the feasibility trial of PCT adapted for PGD.

Characteristic	Total sample (<i>N</i> = 20)
Female, % (<i>n</i>)	80.0 (16)
Age in years, <i>M</i> (<i>SD</i>)	56.00 (8.84)
Education, % (<i>n</i>)	
<12 years	75.0 (15)
≥12 years	25.0 (5)
Employment, % (<i>n</i>)	
Employed	50.0 (10)
Unemployed	20.0 (4)
Retired	20.0 (4)
Other	10.0 (2)
Marital status, % (<i>n</i>)	
Married/in a relationship	45.0 (9)
Divorced/single	15.0 (3)
Widowed	40.0 (8)
Antidepressant medication, % (<i>n</i>)	25.0 (5)
Comorbid disorder DSM-IV^a, % (<i>n</i>)	
None	20.0 (4)
1	60.0 (12)
2	20.0 (4)
Mood disorders	80.0 (16)
Anxiety disorders	10.0 (2)
Somatoform disorders	10.0 (2)
Relation to the deceased, % (<i>n</i>)	
Child	35.0 (7)
Spouse/partner	45.0 (9)
Parent	10.0 (2)
Other	10.0 (2)
Cause of death, % (<i>n</i>)	
Natural	75.0 (15)
Unnatural	25.0 (5)
Expectation of the death, % (<i>n</i>)	
Expected	40.0 (8)
Unexpected	50.0 (10)
Time since loss in months, <i>M</i> (<i>SD</i>)	48.75 (75.56)

PCT, present-centered therapy; PGD, prolonged grief disorder.

^aAccording to the Structured Clinical Interview for DSM-IV Axis I.

be a problem specific to PCT. The other three categories (lack of target definition with the patient, unspecific manual, no permission to use typical CBT methods) seem to constitute specific problems associated with differences between the nature of the PCT program and the prior CBT-training of the therapists.

Experiences With PCT From the Supervisor's Perspective

Multicenter case consultations were put in place to monitor treatment adherence throughout the study. Study therapists became easily engaged in PCT, reported generally positive relationships with their patients and no important problems with regard to treatment adherence. They also reported that

most patients engaged very well in PCT. Some therapists expressed doubts about the effectiveness of the PCT treatment and were reminded of the body of evidence that had been published up to then. All therapists had a CBT qualification, including mastery of a minimal amount of client-centered techniques. The group repeatedly discussed appropriate and inappropriate therapist behaviors to stay within the limits of the treatment manual. Another challenging question was how important problem solving and filling in the diary card were. Some therapists, for example, reported, that patients did not fill in the diary cards or carry out specific homework assignments. During case consultations therapists were motivated to focus more on the therapeutic relationship as mode of action rather than following a CBT-typical perspective when dealing with homework assignments undone. In most cases a decision was taken to focus on behaviors that enhance a positive relationship and promote factors of client-centered therapy. With respect to problem-solving, therapists were reminded to first help the patient to identify their emotions, and only thereafter to work on problem-solving, thus avoiding the risk of talking about solutions that were trivial and not specific enough.

DISCUSSION

Outcomes and Safety of PCT for PGD

This study is the first to investigate the feasibility of PCT in patients suffering from PGD. While PCT previously had been evaluated in samples with PTSD (27), we were interested in the feasibility and treatment effects of PCT in a sample of bereaved adults. We found significant pre- to post-treatment reductions in PGD symptoms as assessed by the PG-13 and the ICG indices with large ESs. However, PCT did not yield ICG total sum scores substantially below clinical cutoffs at post-treatment or follow-up. According to the PG-13 severity score, 50% of participants showed clinically meaningful improvement, and 75% of the participants achieved remission from PGD. Improvements remained nearly stable at the 3-month follow-up. Regarding secondary outcome measures, significant improvements from pre- to post-treatment were observed with respect to depressive symptoms and general mental distress as assessed in self-report, with medium pre-post ESs and large ESs at follow-up.

The ESs regarding PGD symptoms were higher than those reported in meta-analyses for PGD treatments in adults (22–24). These results are encouraging as the self-reported baseline severity of PGD symptoms, $M_{ICG} = 41.70$, in our sample was comparable to several RCTs evaluating grief-focused treatments [e.g., $M_{ICG} = 42.6$ in (18), $M_{ICG} = 47.5$ in (19), $M_{ICG} = 45.8$ in (15), $M_{ICG} = 46.1$ in (59)]. Compared to small ESs for depressive symptoms and general mental distress reported in the literature (22, 24), our results with medium pre-post ESs and large ESs at follow-up seem to be promising. However, because of the small sample size and the uncontrolled design of the study, interpretation of these ESs should only be made with caution (60). Furthermore, compared to ICD-11 criteria, the more strict criteria set used in this study (6) might also have affected ESs, as including participants with lower symptom scores might have yielded in lower ESs.

TABLE 3 | Primary and secondary outcomes of study participants in the feasibility trial of PCT adapted for PGD adults based on intent-to-treat analyses with LOCF, $N = 20$.

Outcome	t0	t1	t2	Time effect			t0-t1	t0-t2
	<i>M (SD)</i>	<i>M (SD)</i>	<i>M (SD)</i>	<i>F</i>	<i>df</i>	<i>p</i>	<i>d</i>	<i>d</i>
PG-13	39.25 (4.61)	29.95 (9.37)	30.25 (9.08)	8.75	2	<0.001	1.26	1.25
ICG	41.70 (8.67)	29.00 (15.28)	30.70 (14.96)	5.35	2	0.007	1.02	0.90
BDI-II	26.05 (8.39)	19.75 (9.72)	18.45 (10.12)	3.71	2	0.031	0.69	0.82
BSI-GSI	1.33 (0.52)	1.02 (0.63)	0.86 (0.58)	3.48	2	0.038	0.54	0.86
SOMS-7D	30.85 (18.87)	29.20 (23.67)	24.80 (22.58)	0.41	2	0.665	0.07	0.29

PCT, present-centered therapy; PGD, prolonged grief disorder; LOCF, last observation carried forward; t0, baseline; t1, posttreatment; t2, 3-month follow-up; PG-13, interview for prolonged grief-13; ICG, inventory of complicated grief; BDI-II, beck depression inventory; BSI-GSI, brief symptom inventory global severity index; SOMS-7D, screening for somatoform disorders.

The severity of somatoform symptoms at baseline was high in the current sample, as the mean score of the SOMS-7D corresponded to percentile rank = 93 according to the German norm sample (47). This is in line with results from other bereaved samples (46, 61, 62). The non-significant differences with regard to somatoform symptoms, as assessed by the SOMS-7D, might be explained by the fact that PCT does not contain any components that are assumed to be specific mechanisms of change for the treatment of somatoform disorders (63, 64).

To adapt the treatment protocol to the standard health insurance coverage for outpatient CBT treatments in Germany, we allowed 20 to 24 50-min sessions to be administered, with a mean of 18.70 sessions conducted. Therefore, the mean number of treatment sessions was considerably higher than the number reported by meta-analyses evaluating grief-focused treatments [10 in (24), 10 to 16 in (23)]. This higher number of treatment sessions might, in part, explain the large ESs we found with respect to PTG symptoms.

Acceptability in Patients and Therapists

We did not observe any exacerbation of PGD symptoms from pretreatment to any later assessment point, nor did any adverse event occur during the trial. These results indicate that the intervention was well-received by the participants and was safe. PCT was a new treatment for all of the study therapists as PCT was previously unknown in Germany. In particular, the strong focus on client-centered techniques (following the core conditions *sensu* Rogers) while dispensing with almost all CBT methods, constituted an unusual approach for many of the therapists. They were all trained in CBT but had not undergone training with the focus on client-centered techniques prior to the current trial. In line with this, the lack of target definition with the patient, the unspecific manual, and the withholding of permission to use typical CBT methods seemed to be specific problems associated with the nature of the PCT program. Some study therapists reported them as barriers. However, these problems might decrease with an increasing number of cases treated with PCT and with growing expertise in emphasizing client-centered techniques in the therapeutic process. It is also questionable whether these problems occur at all with non-CBT therapists. Therefore, implementing PCT with therapists with

other theoretical backgrounds than CBT in future studies would be very informative.

Three participants dropped out of treatment (15%). In two of these cases, the respective patients had to travel very long distances to the treatment site combined with a reported low motivation for change, as indicated by their therapists. In the third case very low motivation to attend therapy sessions was mentioned from the start. All dropouts may also be associated with the participants' high pretreatment PG-13 severity (all above the median of 39) and ICG scores (all above the median of 40). However, in terms of completion rates, the results in our study are good, with 85% completion in PCT vs. 82% in the study by Shear et al. (59), 73% in the study by Shear et al. (15), 79% in the study by Rosner et al. (46) and 71% in the study by Boelen et al. (10).

Overall, the promising results of PCT in patients suffering from PGD as studied in this trial, but also in previous trials with PTSD patients, raise questions about possible psychological mechanisms that are responsible for change in PCT. Unlike many effective interventions for PTSD and PGD, PCT clearly does not include any exposure or cognitive restructuring. It can be assumed that exposure and cognitive restructuring cause direct symptom reductions in patients via facilitating emotional processing, minimizing avoidance, and modifying negative cognitions—mechanisms that seem to be crucial for recovering from PTSD (65) as well as PGD (66). In contrast, patients treated with PCT seem to experience enhanced psychosocial functioning through the application and practice of more effective solutions to daily stressors, and this might indirectly lead to a symptom reduction. PCT's focus on problem-solving in the present might be especially attractive for bereaved patients, as they often have to deal with new problems after the loss (e.g., inheritance issues, problems in everyday life as a result of secondary losses). Furthermore, they might be afraid of interventions that foster emotional engagement with the loss because of avoidance or unbearable emotional pain. This might be also the case in patients suffering from PTSD, for whom PCT achieved lower dropout rates than trauma-focused CBT (26, 27). Additional mechanisms underlying PCT may rely on the therapeutic benefits that emerge from a caring relationship, including mobilization of hope and optimism, and increased positive self-regard (67). These are

TABLE 4 | Demographic characteristic of study therapists in the feasibility trial of PCT adapted for PGD.

Characteristic	Total sample (N = 17)
Female, % (n)	94.1 (16)
Age in years, M (SD)	32.41 (5.85)
Education, % (n)	
Licensed psychotherapist	29.4 (5)
Master's level psychologist in advanced postgraduate clinical training	70.6 (12)
Specialization in cognitive behavioral therapy, % (n)	100 (17)
Duration of clinical work in months, M (SD)	50.88 (44.76)
Number of treated cases, M (SD)	26.75 (51.03)

PCT, present-centered therapy; PGD, prolonged grief disorder.

non-specific elements every psychotherapy contains, but they might be activated to a special degree in PCT. However, these assumptions regarding mechanisms underlying PCT and how they affect long-term effects of treatment success require precise evaluation in future process-outcome studies.

Limitations and Strengths

The generalizability of our study results is limited by the small sample size and the predominantly female (80%) sample. A more general limitation is the lack of a control group, which reduces the strength of the conclusions that can be drawn from the findings. Therefore, natural remission of symptoms or non-specific effects of supportive study attention cannot be excluded. In addition, the non-randomized design may have influenced participants' motivation for taking part in the trial and their retention other than an RCT design. Hence, an RCT with a solid sample size is necessary to test the specific efficacy of PCT for PGD. Furthermore, follow-up assessments for a longer period than 3 months after treatment would have been advisable. Since the ratings of treatment adherence by independent trained raters are not completed yet, we are unable to report on the results of a formalized adherence rating. Yet, specific focus in supervision and case consultations was given to treatment adherence and specifically to the omission of CBT interventions. Finally, using a qualitative methodological approach would have provided further insight regarding therapists' acceptability of PCT. Despite these limitations, which are primarily due to the study design, feasibility trials are of high interest, especially to foster research when there is a lack of controlled studies as for PGD (14). Feasibility trials allow to examine the safety and acceptability of a new or adapted intervention for a new target group before administering it in larger trials. They are also crucial to examine treatment and training protocols. When developing a psychological treatment, it is important to consider how easily and successfully health professionals new to grief treatment can learn to administer it. If a high amount of training is needed to teach the new treatment, dissemination and implementation might prove difficult. Finally, feasibility trials allow for developing adherence/competence measures and recruitment procedures that are crucial for subsequent RCTs,

if the treatment demonstrates clinical impact. To address these goals, the current trial served as an important first step in evaluating PCT for PGD and is in line with sample sizes of other pilot and feasibility trials with bereaved patients [e.g., (68–70)].

Implications

In conclusion, our study furnishes preliminary evidence of the feasibility, efficacy, and safety of PCT with PGD. PCT adapted for PGD was deemed to be an adaptable and easy-to-learn intervention of good design quality by our relatively young CBT therapists. Future studies are needed to address the question of the efficacy of PCT adapted to PGD compared to adequate control conditions. Based on our results, it seems reasonable to choose and evaluate PCT as an active control condition compared to a grief-focused CBT, including exposure and cognitive structuring, in a current RCT (35). Furthermore, the need for specific treatment components, such as exposure, cognitive restructuring techniques, or behavioral activation, should be addressed in dismantling studies targeting patients suffering from PGD. PCT might be a viable alternative for patients unable or unwilling to participate in grief-focused treatments. Further evaluation of PCT in RCTs may help determine which treatment components are beneficial and necessary for the individual PGD patient and might allow formulation of individualized treatment recommendations and different treatment selection [see (71)]. Therefore, further research regarding the efficacy of PCT but also regarding individual predictors of treatment success in PCT is of high interest.

DATA AVAILABILITY STATEMENT

The datasets generated for this study are available on request to the corresponding author.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Institutional Review Board of the Catholic University Eichstaett-Ingolstadt, Ethics Committee of the Department of Psychology and Sports of the Goethe University Frankfurt, Ethical Committee at the Medical Faculty of Leipzig University, Local Ethics Committee of the Department of Psychology of the University of Marburg. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

AV and RR contributed to the design of the study. RR was the principal investigator. AK, WR, and RS were leaders of the collaborating study centers. HC organized the database. AV performed the statistical analyses and wrote the first draft of the manuscript. AN and RR wrote sections of the manuscript.

All authors contributed to the manuscript revision, read and approved the submitted version.

FUNDING

Some parts of the study were funded by proFOR+, a funding program run by the Catholic University of Eichstaett-Ingolstadt. The open access publication of this article was supported by the Open Access Fund of the Catholic University Eichstaett-Ingolstadt. The associated RCT following this study was funded by a grant from the German Research Foundation (DFG RO 2042/7-1).

REFERENCES

- Jordan AH, Litz BT. Prolonged grief disorder: diagnostic, assessment, and treatment considerations. *Prof Psychol Res Pract.* (2014) 45:180–7. doi: 10.1037/a0036836
- World Health Organization (2018). *ICD-11 international classification of diseases for mortality and morbidity statistics*. 11th revision. World Health Organization (2018). Available online at: <https://icd.who.int/browse11/l-m/en>
- American Psychiatric Association, ed. *Diagnostic and Statistical Manual of Mental Disorders (DSM-5)*. 5th ed. Washington, DC: American Psychiatric Association (2013).
- Horowitz M, Siegel B, Holen A, Bonanno G, Milbrath C, Stinson C. Diagnostic criteria for complicated grief disorder. *Am J Psychiatry.* (1997) 154:904–10.
- Prigerson H, Shear MK, Jacobs SC, Reynolds CF, Maciejewski PK, Davidson JRT, et al. Consensus criteria for traumatic grief: a preliminary empirical test. *Br J Psychiatry.* (1999) 174:67–73.
- Prigerson HG, Horowitz MJ, Jacobs SC, Parkes CM, Aslan M, Goodkin K, et al. Prolonged grief disorder: psychometric validation of criteria proposed for DSM-V and ICD-11. *PLoS Med.* (2009) 6:e1000121. doi: 10.1371/journal.pmed.1000121
- Lundorff M, Holmgren H, Zachariae R, Farver-Vestergaard I, O'Connor M. Prevalence of prolonged grief disorder in adult bereavement: a systematic review and meta-analysis. *J Affect Disord.* (2017) 212:138–49. doi: 10.1016/j.jad.2017.01.030
- Kersting A, Brähler E, Glaesmer H, Wagner B. Prevalence of complicated grief in a representative population-based sample. *J Affect Disord.* (2011) 131:339–43. doi: 10.1016/j.jad.2010.11.032
- Djelantik AAAMJ, Smid GE, Mroz A, Kleber RJ, Boelen PA. The prevalence of prolonged grief disorder in bereaved individuals following unnatural losses: systematic review and meta regression analysis. *J Affect Disord.* (2020) 265:146–56. doi: 10.1016/j.jad.2020.01.034
- Boelen PA, Prigerson HG. The influence of symptoms of prolonged grief disorder, depression, and anxiety on quality of life among bereaved adults. *Eur Arch Psychiatry Clin Neurosci.* (2007) 257:444–52. doi: 10.1007/s00406-007-0744-0
- Simon NM, Shear MK, Thompson EH, Zalta AK, Perlman C, Reynolds CF, et al. The prevalence and correlates of psychiatric comorbidity in individuals with complicated grief. *Compr Psychiatry.* (2007) 48:395–9. doi: 10.1016/j.comppsy.2007.05.002
- Latham AE, Prigerson HG. Suicidality and bereavement: complicated grief as psychiatric disorder presenting greatest risk for suicidality. *Suicide Life Threat Behav.* (2004) 34:350–62. doi: 10.1521/suli.34.4.350.53737
- Stroebe M, Schut H, Stroebe W. Health outcomes of bereavement. *Lancet.* (2007) 370:1960–73. doi: 10.1016/S0140-6736(07)61816-9
- Boelen PA, Smid GE. Disturbed grief. Prolonged grief disorder and persistent complex bereavement disorder. *BMJ.* (2017) 357:j2016. doi: 10.1136/bmj.j2016
- Shear MK, Frank E, Houck PR, Reynolds CF. Treatment of complicated grief: a randomized controlled trial. *JAMA.* (2005) 293:2601–8. doi: 10.1001/jama.293.21.2601
- Shear MK, Reynolds CF, Simon NM, Zisook S, Wang Y, Mauro C, et al. Optimizing treatment of complicated grief: a randomized clinical trial. *JAMA Psychiatry.* (2016) 73:685–94. doi: 10.1001/jamapsychiatry.2016.0892
- Supiano KP, Luptak M. Complicated grief in older adults: a randomized controlled trial of complicated grief group therapy. *Gerontologist.* (2014) 54:840–56. doi: 10.1093/geront/gnt076
- Boelen PA, de Keijser J, van den Hout MA, van den Bout J. Treatment of complicated grief. A comparison between cognitive-behavioral therapy and supportive counseling. *J Consult Clin Psychol.* (2007) 75:277–84. doi: 10.1037/0022-006X.75.2.277
- Bryant RA, Kenny L, Joscelyne A, Rawson N, Maccallum F, Cahill C, et al. Treating prolonged grief disorder: a randomized clinical trial. *JAMA Psychiatry.* (2014) 71:1332–9. doi: 10.1001/jamapsychiatry.2014.1600
- Eisma MC, Boelen PA, van den Bout J, Stroebe W, Schut HAW, Lancee J, et al. Internet-based exposure and behavioral activation for complicated grief and rumination: a randomized controlled trial. *Behav Ther.* (2015) 46:729–48. doi: 10.1016/j.beth.2015.05.007
- Wagner B, Knaevelsrud C, Maercker A. Internet-based cognitive-behavioral therapy for complicated grief: a randomized controlled trial. *Death Stud.* (2006) 30:429–53. doi: 10.1080/07481180600614385
- Currier JM, Neimeyer RA, Berman JS. The effectiveness of psychotherapeutic interventions for bereaved persons: a comprehensive quantitative review. *Psychol Bull.* (2008) 134:648–61. doi: 10.1037/0033-2909.134.5.648
- Wittouck C, Van Autreve S, Jaegere E, de Portzky G, van Heeringen, K. The prevention and treatment of complicated grief: a meta-analysis. *Clin Psychol Rev.* (2011) 31:69–78. doi: 10.1016/j.cpr.2010.09.005
- Johannsen M, Damholdt MF, Zachariae R, Lundorff M, Farver-Vestergaard I, O'Connor M. Psychological interventions for grief in adults. A systematic review and meta-analysis of randomized controlled trials. *J Affect Disord.* (2019) 253:69–86. doi: 10.1016/j.jad.2019.04.065
- Shea MT, Davis N, Howard J, Key F, Lambert J. *Present-Centered Therapy (PCT) Manual*. Unpublished Manual Developed for Cooperative Studies: Treatment of Posttraumatic Stress Disorders in Women (2003).
- Frost ND, Laska KM, Wampold BE. The evidence for present-centered therapy as a treatment for posttraumatic stress disorder. *J Trauma Stress.* (2014) 27:1–8. doi: 10.1002/jts.21881
- Belsher BE, Beech E, Evatt D, Smolenski DJ, Shea MT, Otto JL, et al. Present-centered therapy (PCT) for post-traumatic stress disorder (PTSD) in adults. *Cochrane Database Syst Rev.* (2019) 2019:CD012898. doi: 10.1002/14651858.CD012898.pub2
- International Society for Traumatic Stress Studies. ISTSS prevention and treatment guidelines (2018). Available online at: <https://www.istss.org/treating-trauma/new-istss-prevention-and-treatment-guidelines.aspx> (accessed February 6, 2019).

ACKNOWLEDGMENTS

We thank all the patients who participated in this trial, and all therapists and raters who administered the interventions and assessments.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: <https://www.frontiersin.org/articles/10.3389/fpsy.2021.534664/full#supplementary-material>

29. Lambert MJ. The efficacy and effectiveness of psychotherapy. In: Lambert MJ, editor. *Bergin and Garfield's Handbook of Psychotherapy and Behaviour Change*. Hoboken, NJ: Wiley (2013). p. 169–218.
30. Wampold BE, Mondin GW, Moody M, Stich F, Benson K, Ahn H-N. A meta-analysis of outcome studies comparing bona fide psychotherapies: empirically, “all must have prizes. *Psychol Bull.* (1997) 122:203–15. doi: 10.1037/0033-2909.122.3.203
31. Stroebe M, Schut H. The dual process model of coping with bereavement: rationale and description. *Death Stud.* (1999) 23:197–224. doi: 10.1080/074811899201046
32. Prigerson HG, Maciejewski PK. *Prolonged Grief Disorder (PG-13) Scale*. Boston, MA: Dana-Farber Cancer Institute (2008).
33. Comtesse H, Vogel A, Kersting A, Rief W, Steil R, Rosner R. When does grief become pathological? Evaluation of the ICD-11 diagnostic proposal for prolonged grief in a treatment-seeking sample. *Eur J Psychotraumatol.* (2020) 11:1694348. doi: 10.1080/20008198.2019.1694348
34. Shear MK, Simon N, Wall M, Zisook S, Neimeyer R, Duan N, et al. Complicated grief and related bereavement issues for DSM-5. *Depression Anxiety.* (2011) 28:103–17. doi: 10.1002/da.20780
35. Rosner R, Rimane E, Vogel A, Rau J, Hagl M. Treating prolonged grief disorder with prolonged grief-specific cognitive behavioral therapy: study protocol for a randomized controlled trial. *Trials.* (2018) 19:241. doi: 10.1186/s13063-018-2618-3
36. Vogel A, Pfoh G, Rosner R. *PG-13+9: Interview for Prolonged Grief – Revised and Extended Translation of the PG-13*. Eichstaett: Catholic University of Eichstaett-Ingolstadt (2017).
37. Lichtenthal WG, Corner GW, Sweeney CR, Wiener L, Roberts KE, Baser RE, et al. Mental health services for parents who lost a child to cancer: if we build them, will they come? *J Clin Oncol.* (2015) 33:2246–53. doi: 10.1200/JCO.2014.59.0406
38. Pohlkamp L, Kreibergs U, Prigerson HG, Sveen J. Psychometric properties of the prolonged grief disorder-13 (PG-13) in bereaved Swedish parents. *Psychiatry Res.* (2018) 267:560–5. doi: 10.1016/j.psychres.2018.06.004
39. Litz BT, Schorr Y, Delaney E, Au T, Papa A, Fox AB, et al. A randomized controlled trial of an internet-based therapist-assisted indicated preventive intervention for prolonged grief disorder. *Behav Res Ther.* (2014) 61:23–34. doi: 10.1016/j.brat.2014.07.005
40. Prigerson HG, Maciejewski PK, Reynolds CFIII, Bierhals AJ, Newsom JT, Fasiczka A, et al. Inventory of complicated grief: a scale to measure maladaptive symptoms of loss. *Psychiatry Res.* (1995) 59:65–79. doi: 10.1016/0165-1781(95)02757-2
41. Lumbeck G, Brandstätter M, Geissner E. Erstvalidierung der deutschen Version des Inventory of Complicated Grief“ (ICG-D). *Zeitschrift für Klinische Psychologie und Psychotherapie.* (2012) 41:243–8. doi: 10.1026/1616-3443/a000172
42. Cozza SJ, Fisher JE, Mauro C, Zhou J, Ortiz CD, Skritskaya N, et al. Performance of DSM-5 persistent complex bereavement disorder criteria in a community sample of bereaved military family members. *Am J Psychiatry.* (2016) 173:919–29. doi: 10.1176/appi.ajp.2016.15111442
43. Cozza SJ, Shear MK, Reynolds CF, Fisher JE, Zhou J, Maercker A, et al. Optimizing the clinical utility of four proposed criteria for a persistent and impairing grief disorder by emphasizing core, rather than associated symptoms. *Psychol Med.* (2019) 50:1–8. doi: 10.1017/S0033291719000254
44. Hautzinger M, Keller F, Kühner C. *BDI-II. Beck Depressions-Inventar Revision*. 2nd ed. Frankfurt: Harcourt Test Services (2009).
45. Rosner R, Lumbeck G, Geissner E. Effectiveness of an inpatient group therapy for comorbid complicated grief disorder. *Psychother Res.* (2011) 21:210–8. doi: 10.1080/10503307.2010.545839
46. Rosner R, Pfoh G, Kotoučová M, Hagl M. Efficacy of an outpatient treatment for prolonged grief disorder: a randomized controlled clinical trial. *J Affect Disord.* (2014) 167:56–63. doi: 10.1016/j.jad.2014.05.035
47. Rief W, Hiller W. *Screening für somatoforme Störungen (SOMS)*. Bern: Hogrefe (2008).
48. Franke GH. *Brief symptom inventory von L. R. Derogatis (Kurzform der SCL-90-R). Deutsche Version*. Göttingen: Beltz (2000).
49. Cook JM, O'Donnell C, Dinnen S, Coyne JC, Ruzek JI, Schnurr PP. Measurement of a model of implementation for health care: toward a testable theory. *Implementation Sci.* (2012) 7:59. doi: 10.1186/1748-5908-7-59
50. Damschroder LJ, Aron DC, Keith RE, Kirsh SR, Alexander JA, Lowery JC. Fostering implementation of health services research findings into practice: a consolidated framework for advancing implementation science. *Implementation Sci.* (2009) 4:50. doi: 10.1186/1748-5908-4-50
51. First MB, Spitzer RL, Gibbon M, Williams JBW. *Structured Clinical Interview for DSM-IV Axis I Disorders (SCID-I)*. New York, NY: Biometric Research Department (1997).
52. Wittchen H-U, Zaudig M, Fydrich T. *SKID-I. Strukturiertes Klinisches Interview für DSM-IV Achse I: Psychische Störungen*. Göttingen: Hogrefe (1997).
53. Posner K, Brown GK, Stanley B, Brent DA, Yershova KV, Oquendo MA, et al. The Columbia-Suicide Severity Rating Scale: initial validity and internal consistency findings from three multisite studies with adolescents and adults. *Am J Psychiatry.* (2011) 168:1266–77. doi: 10.1176/appi.ajp.2011.10111704
54. Nocon A, Shea MT, Rosner R. *Gegenwartsakzentuierte Therapie (GAT) bei Anhaltender Trauerstörung: Manual [Present-centered therapy adapted for prolonged grief disorder]*. Eichstaett: Catholic University of Eichstaett-Ingolstadt (2017).
55. Cochran WG. The comparison of percentages in matched samples. *Biometrika.* (1950) 37:256–66.
56. Jacobson NS, Truax P. Clinical significance: a statistical approach to defining meaningful change in psychotherapy research. *J Consult Clin Psychol.* (1991) 59:12–9.
57. Martinovich Z, Saunders S, Howard K. Some comments on “Assessing clinical significance”. *Psychother Res.* (1996) 6:124–32.
58. Bengtsson M. How to plan and perform a qualitative study using content analysis. *NursPlus Open.* (2016) 2:8–14. doi: 10.1016/j.npls.2016.01.001
59. Shear MK, Wang Y, Skritskaya N, Duan N, Mauro C, Ghesquiere A. Treatment of complicated grief in elderly persons: a randomized clinical trial. *JAMA Psychiatry.* (2014) 71:1287–95. doi: 10.1001/jamapsychiatry.2014.1242
60. Lipsey MW, Wilson DB. The efficacy of psychological, educational, and behavioral treatment: confirmation from meta-analysis. *Am Psychol.* (1993) 48:1181–209. doi: 10.1037//0003-066X.48.12.1181
61. Huh HJ, Huh S, Lee SH, Chae J-H. Unresolved bereavement and other mental health problems in parents of the sewol ferry accident after 18 months. *Psychiatry Invest.* (2017) 14:231–9. doi: 10.4306/pi.2017.14.3.231
62. Rosner R, Bartl H, Pfoh G, Kotoučová M, Hagl M. Efficacy of an integrative CBT for prolonged grief disorder: a long-term follow-up. *J Affect Disord.* (2015) 183:106–12. doi: 10.1016/j.jad.2015.04.051
63. Kleinstäuber M, Witthöft M, Hiller W. Efficacy of short-term psychotherapy for multiple medically unexplained physical symptoms: a meta-analysis. *Clin Psychol Rev.* (2011) 31:146–60. doi: 10.1016/j.cpr.2010.09.001
64. Koelen JA, Houtveen JH, Abbass A, Luyten P, Eurelings-Bontekoe EHM, van Broeckhuysen-Kloth SAM, et al. Effectiveness of psychotherapy for severe somatoform disorder: meta-analysis. *Br J Psychiatry.* (2014) 204:12–9. doi: 10.1192/bjp.bp.112.121830
65. Sripada RK, Rauch Sheila AM, Liberzon I. Psychological mechanisms of PTSD and its treatment. *Curr Psychiatry Rep.* (2016) 18:99. doi: 10.1007/s11920-016-0735-9
66. Boelen PA, de Keijser J, de van den Hout MA, van den Bout J. Factors associated with outcome of cognitive-behavioural therapy for complicated grief: a preliminary study. *Clin Psychol Psychother.* (2011) 18:284–91. doi: 10.1002/cpp.720
67. Schnurr PP, Shea MT, Friedman MJ, Engel CC. Posttraumatic stress disorder and cognitive behavioral therapy—reply. *JAMA.* (2007) 297:2694–5. doi: 10.1001/jama.297.24.2694
68. Iliya YA. Music therapy as grief therapy for adults with mental illness and complicated grief: a pilot study. *Death Stud.* (2015) 39:173–84. doi: 10.1080/07481187.2014.946623

69. O'Connor M, Piet J, Hougaard E. The effects of mindfulness-based cognitive therapy on depressive symptoms in elderly bereaved people with loss-related distress: a controlled pilot study. *Mindfulness*. (2014) 5:400–9. doi: 10.1007/s12671-013-0194-x
70. Papa A, Sewell MT, Garrison-Diehn C, Rummel C. A randomized open trial assessing the feasibility of behavioral activation for pathological grief responding. *Behav Ther*. (2013) 44:639–50. doi: 10.1016/j.beth.2013.04.009
71. Barber JP, Muenz LR. The role of avoidance and obsessiveness in matching patients to cognitive and interpersonal psychotherapy: empirical findings from the treatment for depression collaborative research program. *J Consult Clin Psychol*. (1996) 64:951–8. doi: 10.1037/0022-006X64.5.951

Conflict of Interest: AN was paid fees for supervising PCT. RR was paid fees for workshops and presentations on PGD treatment.

The remaining authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Copyright © 2021 Vogel, Comtesse, Nocon, Kersting, Rief, Steil and Rosner. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

Advantages of publishing in Frontiers



OPEN ACCESS

Articles are free to read
for greatest visibility
and readership



FAST PUBLICATION

Around 90 days
from submission
to decision



HIGH QUALITY PEER-REVIEW

Rigorous, collaborative,
and constructive
peer-review



TRANSPARENT PEER-REVIEW

Editors and reviewers
acknowledged by name
on published articles

Frontiers

Avenue du Tribunal-Fédéral 34
1005 Lausanne | Switzerland

Visit us: www.frontiersin.org

Contact us: frontiersin.org/about/contact



REPRODUCIBILITY OF RESEARCH

Support open data
and methods to enhance
research reproducibility



DIGITAL PUBLISHING

Articles designed
for optimal readership
across devices



FOLLOW US

@frontiersin



IMPACT METRICS

Advanced article metrics
track visibility across
digital media



EXTENSIVE PROMOTION

Marketing
and promotion
of impactful research



LOOP RESEARCH NETWORK

Our network
increases your
article's readership