



## Review Article

# Co-occurrence of prolonged grief symptoms and symptoms of depression, anxiety, and posttraumatic stress in bereaved adults: A systematic review and meta-analysis



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## ABSTRACT

**Background:** ‘Complicated grief reactions’ is an umbrella term covering symptoms of prolonged grief disorder (PGS) and other post-loss complications, including symptoms of depression, anxiety, and posttraumatic stress (PTS). While PGS often co-occurs with symptoms of depression, anxiety, and PTS, no pooled prevalence estimates of their co-occurrence have yet been established.

**Methods:** The present systematic review and meta-analysis provided pooled prevalence estimates of co-occurrence of PGS and symptoms of depression, anxiety, and PTS based on the available literature, and examined possible moderators and risk of bias.

**Results:** Based on the 23 included studies, the pooled prevalence estimates indicated that 70% of adults with PGS experienced one or more other type of complicated grief reaction, and 46% experienced two or more other types of complicated grief reactions. Estimates of PGS with co-occurring depression, anxiety, and PTS were 63%, 54%, and 49%, respectively. Heterogeneity was considerable ( $I^2=92.5-95.6$ ), and subsequent moderator-analyses showed that higher estimates of co-occurrence were found in studies with longer mean time since loss, and when co-occurrence was assessed with interviews compared with questionnaires.

**Limitations:** The results should be considered preliminary due to high risk of bias of the included studies.

**Conclusions:** Co-occurring cases of PGS and other types of complicated grief reactions were more prevalent than ‘pure’ cases of PGS with no co-occurrence. More population-based studies of symptom co-occurrence in non-traumatic bereavement are needed.

## 1. Introduction

Most bereaved individuals cope adaptively with bereavement, but a significant minority experience more severe and prolonged grief reactions that cause persistent suffering (Boelen and Prigerson, 2013; Jordan and Litz, 2014). The diagnosis of *prolonged grief disorder* (PGD) was recently added to the 11th revision of the International Classification of Diseases (ICD-11) and has now also been approved for inclusion in the text revision of the 5th edition of the Diagnostic and Statistical Manual of Mental Disorder (DSM-5-TR) (Prigerson et al., 2021; World Health Organization, 2018). According to the ICD-11 diagnostic criteria, PGD is a persistent and pervasive grief reaction, in which

the bereaved individual intensely longs for or is persistently preoccupied with the deceased and experiences intense emotional pain such as sadness, guilt, anger, and has difficulty accepting the death. The grief reaction is required to persist for more than six months following the loss and clearly exceed social, cultural, or religious norms (World Health Organization, 2020). Approximately 10% of bereaved adults are estimated to experience symptoms of PGD after *non-traumatic loss* (i.e., loss due to old age or sickness; Lundorff et al., 2017) with the proportion increasing to 49% after *traumatic loss* (i.e., loss due to murder, terror, or natural disasters; Djelantik et al., 2020). Following the development of diagnostic criteria and assessment instruments, the definition of PGD has changed several times over the last decades (Boelen and

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Lenferink, 2020; Lenferink et al., 2019). Until recently, no valid self-report instruments corresponding to the ICD-11 criteria were available (Killikelly et al., 2020), and a structured clinical interview for ICD-11 PGD, which is regarded the gold standard for diagnosis, is still not available (O'Connor et al., 2020; Trembl et al., 2020). For these reasons, the findings of the majority of studies cannot directly be generalized to the diagnosis of PGD per se. However, *clinically relevant levels of symptoms of PGD (PGS)* are often used as proxy measures of PGD.

PGS is not the only type of post-loss complication that can cause persistent suffering (Rando, 2013), and theoretically, *complicated grief reactions* may be viewed as an umbrella term for a variety of post-loss complications (Larsen et al., 2018; Rando, 2013). Complicated grief reactions are proposed to cover clinically relevant levels of not only PGS, but also *symptoms of depression, anxiety, and posttraumatic stress (PTS)* (Larsen et al., 2018), which are found particularly common in the period after a loss (Jordan and Litz, 2014; Shear and Skritskaya, 2012; Stroebe et al., 2007). A meta-analysis of common mental disorders in widowhood estimated that 41% of bereaved adults showed clinically relevant levels of depressive symptoms and that 27% had clinically relevant levels of anxiety post-loss (Kristiansen et al., 2019), and another systematic review found that 12% of bereaved adults had PTS (Onrust and Cuijpers, 2006). However, all complicated grief reactions are not necessarily caused by the loss itself (Jordan and Litz, 2014; Stroebe et al., 2007). For example, symptoms of depression, anxiety, or PTS may be present before the loss of a loved one, and the loss may lead to worsening of such pre-existing symptoms (Zisook et al., 2014). While studies have evidenced the distinctiveness of PGS from these other types of complicated grief reactions (Boelen and Prigerson, 2007; Boelen et al., 2010; Bonanno et al., 2007; Prigerson et al., 1995, 1996), the common factor is that they all constitute pathological reactions following a loss that may interfere with the natural grieving process (Larsen et al., 2018; Shear and Skritskaya, 2012; Stroebe et al., 2007; Zisook et al., 2014). A broadly accepted theory, *the dual-process model of coping with bereavement*, proposes that the natural grieving process is hindered when the individual is unable to oscillate in a flexible way between confronting and avoiding both loss-oriented stressors (e.g., aspects of the loss itself) and restoration-oriented stressors (e.g., secondary consequences of the loss such as new roles) (Stroebe and Schut, 1999, 2010). In other words, a complicated grief reaction is when the individual “gets stuck” in the process of grief. In line with this theory, PGS and bereavement-related symptoms of depression, anxiety, and PTS are all believed to hinder the natural grieving process (e.g., Shear and Skritskaya, 2012; Stroebe et al., 2007; Zisook et al., 2014) and can thus be defined as complicated grief reactions.

In the bereavement literature, it is noted that PGS and other types of complicated grief reactions frequently co-occur (e.g., Jordan and Litz, 2014; Rando, 2013; Raphael et al., 2013; Shear and Skritskaya, 2012). However, the published prevalence estimates of the co-occurrence of PGS and clinically relevant levels of symptoms of depression, anxiety, and PTS vary considerably, with findings ranging from 10 to 100% (e.g., Melhem et al., 2001; Newson et al., 2011; Patel et al., 2019; Schaal et al., 2012, 2009). The wide range of these estimates may be due to between-study differences in research designs (clinical trials vs. cohort studies), recruitment contexts (clinically referred vs. community-based), sampling methods (probability vs. convenience sampling), assessment methods (clinical interview vs. self-report questionnaire), cut-off methods (criteria-based vs. cut-off value), assessment instruments (ICG vs. PG-13, etc.), and time after loss assessment of co-occurrence. Additional loss-related and sociodemographic factors, including the circumstances of the loss (e.g., traumatic, violent), age, and female gender may also influence rates of co-occurrence, as these factors have repeatedly been associated with poorer bereavement outcomes (Burke and Neimeyer, 2013; Heeke et al., 2019; Shear and Skritskaya, 2012; Stroebe et al., 2006).

Knowledge about co-occurrence is clinically important for several reasons. First, it has been shown that bereaved individuals who experi-

ence PGS concurrently with clinically relevant levels of symptoms of depression, anxiety, or PTS, generally report more severe grief symptoms, lower quality of life, and more functional impairment than individuals with clinically relevant levels of only one of these complicated grief reactions (e.g., Aoyama et al., 2018; Kersting et al., 2009; Marques et al., 2013; Simon et al., 2007). Such individuals thus constitute a patient group requiring special attention (van Loo and Romeijn, 2015). Second, knowledge about co-occurrence is important for the development and optimization of efficacious grief interventions (Melhem et al., 2001; Raphael et al., 2013; Rosner, 2015; Shear and Skritskaya, 2012). Different types of complicated grief reactions do not necessarily respond equally well to the same interventions, e.g., psychological and pharmacological treatments for bereavement-related depressive symptoms do not work optimally on PGS (Shear et al., 2005; Shear et al., 2016). Moreover, it has been suggested that individuals with PGS and co-occurring depressive symptoms might benefit from different interventions compared to those with PGS alone (e.g., combining grief therapy with antidepressant medication; Jordan and Litz, 2014; Shear et al., 2016).

An accurate, overall prevalence estimate of co-occurrence based on current empirical evidence is the first step in the direction of acquiring general evidence-based knowledge about co-occurrence of PGS and other types of complicated grief reactions. To our knowledge, only two previous meta-analyses have investigated the association between PGS and clinically relevant levels of symptoms of depression, anxiety, and PTS (Heeke et al., 2019; Kokou-Kpolou et al., 2020). Both meta-analyses revealed large statistically significant associations between PGS and these types of complicated grief reactions ( $r = 0.47\text{--}0.59$ ). However, neither of the two meta-analyses provided prevalence estimates, i.e., the proportion of individuals experienced co-occurrence. Furthermore, both meta-analyses focused on studies of individuals exposed to traumatic or violence-related loss, limiting the generalizability of results to complicated grief reactions after other types of loss.

Taken together, studies have indicated that PGS is likely to co-occur to varying degrees with different other types of complicated grief reactions, but no valid prevalence estimates of the co-occurrence of PGS and bereavement-related symptoms of depression, anxiety, and PTS are available. Our primary aim was therefore to estimate in bereaved adults with PGS the prevalence of co-occurring other types of complicated grief reactions, i.e., clinically relevant levels of symptoms of depression, anxiety, and PTS, by conducting a systematic review and meta-analysis of the currently available evidence. A secondary aim was to examine between-study differences as possible moderators of co-occurrence, including recruitment context, assessment methods, cut-off methods, and characteristics of the study participants and their loss.

## 2. Methods

A protocol for the present review was established in accordance with Preferred Reporting Items for Systematic reviews and Meta-Analysis (PRISMA) (Liberati et al., 2009) and preregistered with PROSPERO (registration number: CRD42020163188; National Institute for Health Research, 2020).

### 2.1. Eligibility criteria

Study eligibility criteria were established using the PICO approach (Sackett et al., 1996): Population (bereaved individuals with PGS), Intervention/exposure (loss of a loved one), Comparison (not applicable), and Outcome (co-occurring clinically relevant levels of PGS and symptoms of depression, anxiety, and/or PTS). Eligible studies: (i) included observational data of bereaved adults ( $\geq 18$  years) with PGS, (ii) measured clinically relevant levels of symptoms of depression, anxiety, and/or PTS simultaneously with PGS, and (iii) were published from the earliest date to August 31, 2020 in English-language journals with peer-review. Exclusion criteria were: (i) not written in English (ii) reviews, case studies, and gray literature (e.g., dissertations, conference

abstracts), (iii) studies of children and adolescents (<18 years), (iv) studies of other types of loss (e.g., losing a pet), (v) no relevant measure of PGS (i.e., not measured by validated questionnaires or clinical interviews with a mean time of at least six months since loss), (vi) clinically relevant levels of symptoms of depression, anxiety, and/or PTS were not measured by validated questionnaires or clinical interviews, (vii) no relevant assessment of co-occurrence (i.e., no reported estimate of co-occurrence or only life-time co-occurrence reported), (viii) study population not recruited on the basis of PGS, but on the presence of clinically relevant levels of symptoms of depression, anxiety, or PTS, as this does not allow for an inquiry of co-occurrence in adults with PGS, and (ix) data from randomized controlled trials (RCTs), where estimates of co-occurrence may be biased due to inclusion and/or exclusion of specific psychiatric disorders or participant characteristics (Feinstein, 1970).

## 2.2. Information sources and search strategy

Studies were identified through the electronic databases PubMed, PsycINFO, and CINAHL. The final search string consisted of the following: ("prolonged grief" OR PGD OR "complicated grief" OR CG OR "traumatic grief" OR "pathological grief" OR "persistent complex bereavement disorder" OR PCBD OR grief OR grieving OR bereave\* OR mourn\*) AND (comorbid\* OR co-morbid\* OR co-occur\* OR cooccur\* OR co-exist\* OR coexist\* OR concomit\*). When appropriate, MeSH-terms were used. Two independent reviewers (KK, LN) conducted the search. A final update of the search was conducted on August 31, 2020. Snowballing (i.e., searching reference lists of included studies) was used to identify additional relevant studies.

## 2.3. Study selection procedure

Studies were screened by two independent reviewers (KK, LN) using the Covidence systematic review software (Veritas Health Innovation, 2020). First, titles and abstracts were screened for relevance, and then full texts were screened for eligibility. The last author (MO) was consulted in case of disagreement. The level of interrater agreement was calculated for each step of the study selection process, including the risk of bias assessment, with Kappa Statistics ( $\kappa$ ) (McHugh, 2012).

## 2.4. Risk of bias assessment

Included studies were assessed for risk of bias by two independent reviewers (KK, LN) using an instrument for assessing risk of bias in prevalence studies (Hoy et al., 2012). The instrument includes ten dichotomized items (low vs. high risk of bias), with four of the items evaluating external validity and six items internal validity. If the information for an item was insufficient, the item was rated as "high risk of bias" (Hoy et al., 2012). Each item was rated with a value of 1 (low risk) or 0 (high risk). An overall risk of bias score was calculated for each study.

## 2.5. Data extraction

Data from the eligible studies were extracted by KK and double-checked independently by LN. The extracted data included: (i) Total number of participants with PGS and total number of participants with PGS and one or more other types of complicated grief reactions (i.e., clinically relevant levels of symptoms of depression, anxiety, and/or PTS), (ii) total number of participants with PGS and two or more other types of co-occurring complicated grief reactions, and (iii) total number of participants with PGS and co-occurring clinically relevant levels of symptoms of depression, anxiety, and PTS, respectively. Other data included: (iv) Circumstances of the loss (traumatic vs. non-traumatic), (v) recruitment context (clinical vs. non-clinical), (vi) response rate, (vii) sampling method (probability vs. non-probability), (viii) assessment method (survey vs. interview), (ix) mean age of the sample (years), (x) gender (percent women), (xi) mean time since loss (months), (xii)

region (Western vs. Non-Western), (xiii) grief instrument (ICG, PG-13, etc.), and (xiv) cut-off method (criteria-based vs. cut-off value). In case of missing information, the first and last authors were contacted. If discrepancies were found between the forwarded data upon request and the data reported in the original studies, authors were contacted. In case of no reply, data from the original article were used.

## 2.6. Meta-analytical strategy

The analyses were conducted with Comprehensive Meta-Analysis Version 3 (Borenstein et al., 2013). The primary outcome was an inverse-variance weighted proportion (event rate) of participants with co-occurring PGS and other types of complicated grief reactions (i.e., clinically relevant levels of symptoms of depression, anxiety, and/or PTS) (range: 0.00–1.00).

A random effects model (REM) was chosen for all analyses. REM is based on the assumption that the true effect may vary across studies and samples. If the effect sizes are heterogeneous, i.e., if the true effect sizes vary beyond random error, REM takes the between-study variance, termed  $Tau^2$ , into consideration (Littell et al., 2008).

Heterogeneity was tested for with Cochran's  $Q$  and  $I^2$ .  $Q$  is a Chi<sup>2</sup>-test where a high  $Q$  value and a small  $p$ -value are taken to indicate heterogeneity (Deeks et al., 2019). A  $p$ -value of  $\leq 0.10$  was used to determine significant heterogeneity (Higgins et al., 2003).  $I^2$  is a quantification of the proportion of the total between-study variation (0–100%) assumed to be caused by heterogeneity, rather than chance or sampling error (Higgins et al., 2003).

Possible sources of heterogeneity of the main outcome of co-occurrence of PGS and one or more other types of complicated grief reactions were explored with moderator analyses. When  $K$  (number of studies)  $\geq 3$ , this included subgroup analyses of categorical variables, e.g., clinically relevant symptom levels assessed with survey versus interview or Western versus Non-Western samples. When  $K \geq 8$ , the effects of these categorical and additional continuous moderators (e.g., mean age or percent women in the study sample) were analyzed with meta-regression.

Publication bias, i.e., that statistically significant findings in the hypothesized direction may be more likely to be published, is a widespread problem in meta-analysis (Sterne et al., 2001). While publication bias may be less likely in prevalence studies of co-occurring symptoms, which do not test a hypothesis, we explored the possibility using funnel plots and Egger's tests (Deeks et al., 2005; Egger et al., 1997) when  $K > 10$  (Sterne et al., 2011). If results were suggestive of possible publication bias, a sensitivity analysis was conducted by adjusting event rates using the Duval and Tweedie trim-and-fill method (Duval and Tweedie, 2000).

## 3. Results

### 3.1. Study selection

The study selection process is shown in Fig. 1. The level of agreement was substantial for both the title and abstract screening (94%,  $\kappa=0.73$ ; 95% CI: 0.66–0.80), and the full text screening (94%,  $\kappa=0.79$ ; 95% CI: 0.64–0.94). Snowballing resulted in eight additional eligible studies. To obtain missing data about e.g., one or more other types of complicated grief reactions, the first and last authors of 13 studies were contacted. The authors of four studies did not respond. Of these, two studies reported accurate prevalence estimates of the co-occurrence of PGS and e.g., depression and anxiety, respectively, and were thus included although no available data about co-occurrence of one or more other types complicated grief reactions. The two remaining studies had not reported any accurate prevalence estimates of co-occurrence, and were thus excluded due to missing data. This resulted in the final inclusion of 23 studies.

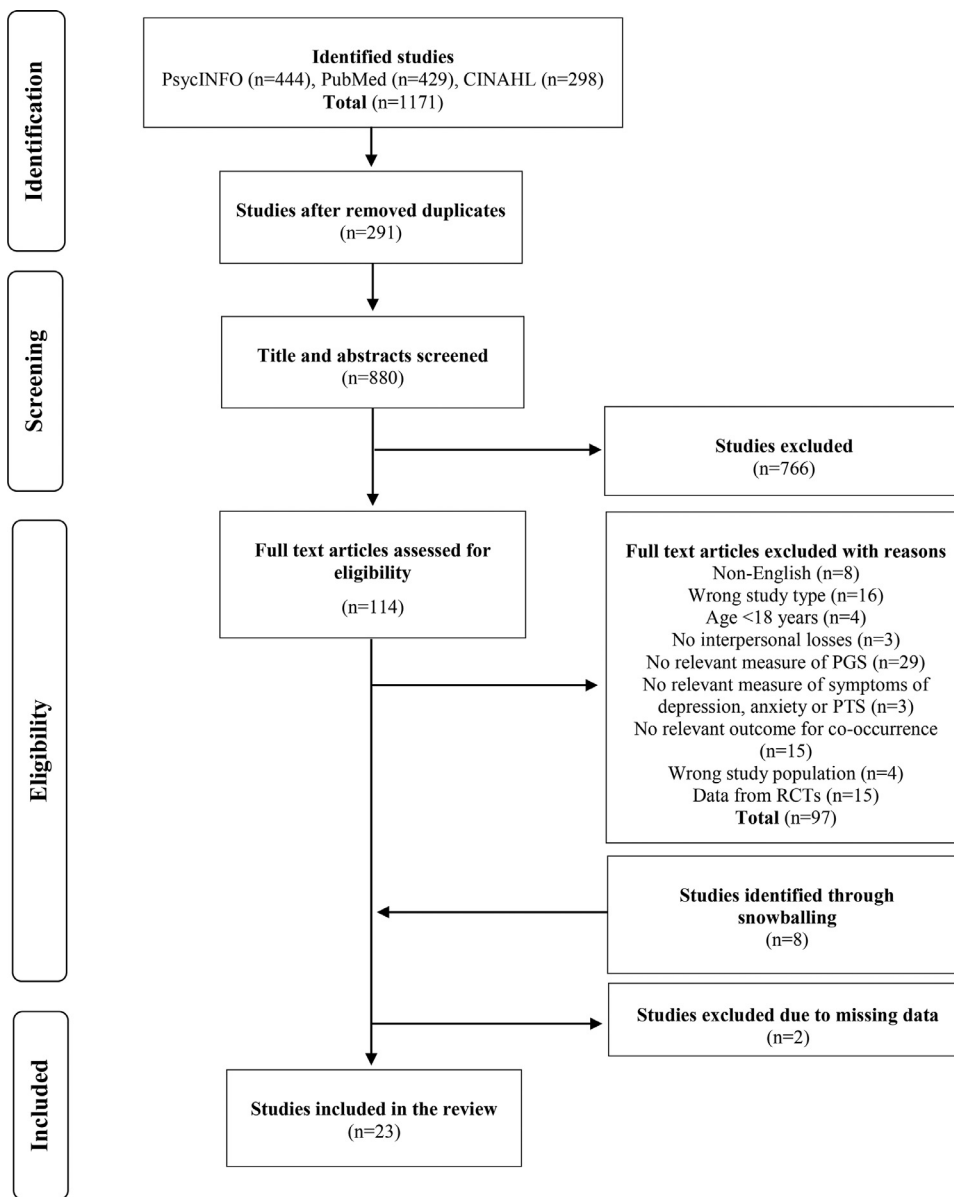


Fig. 1. Flowchart of the study selection.

### 3.2. Study characteristics

The characteristics of the included studies are summarized in Table 1. The studies were published between 1999 and 2020, and investigated a total of 19,222 participants, with a total of 3,272 (17%) participants identified as having PGS. The average sample mean age was 49 years, and the percentage of women in the study sample ranged from 29% to 100%. PGS was assessed on average 56.6 months post loss (range = 6 to 182 months). Fifteen studies (65%) examined traumatic losses and seven (30%) examined non-traumatic losses, defined as  $\geq 75\%$  of the deaths being caused by e.g., sickness or old age. The majority of studies ( $K = 19$ ; 83%) used non-probability sampling (i.e., convenience, snowball, or volunteer sampling), and the remaining four studies (17%) used probability sampling (e.g., cluster sampling, multi-stage sampling, or were census based; Hoy et al., 2012). The majority of studies recruited participants in non-clinical contexts ( $K = 16$ ; 70%), and the remaining seven studies (30%) recruited participants in clinical contexts (e.g., support groups, clinics). Twenty-one studies (91%) assessed depression, 16 assessed PTS (70%), and nine assessed anxiety (39%). Different assessment instruments were used to identify clinically relevant levels of PGS, depressive symptoms, anxiety, and PTS, respectively. Nine studies

were questionnaire-based (39%), eight used interviews (35%), and six used both questionnaires and interviews (26%). Ten studies (43%) used criteria-based definitions of PGS, whereas 11 studies (48%) used cut-off values for PGS. Furthermore, different cut-offs were used even within the same instrument (e.g.,  $ICG \geq 22, 25, 26, 30$ ). While all included studies provided estimates of co-occurrence of different types of complicated grief reactions in adults with PGS, seven studies (30%) did not provide the details needed to calculate the proportion of participants who had one or more co-occurring symptoms. For example, some studies reported the percentage of participants with co-occurring symptoms of depression, anxiety, and PTS, respectively, but gave no details regarding who had only one versus those who had more than one of these syndromes. The number of studies used for estimating the co-occurrence of PGS and one or more other types of complicated grief reactions was thus only 16 out of the 23 included studies.

### 3.3. Risk of bias assessment

The included studies were assessed for risk of bias with an interrater reliability of substantial agreement (90%;  $\kappa = 0.79$ ; 95% CI: 0.71–0.87). The total risk of bias score was high in 14 (61%) studies, eight studies

**Table 1**  
Characteristics of included studies.

Author (year)	Study setting and design	Sample characteristic and recruitment	Study population Study sample size; response rate; %PGS; %women; mean age	Mean time since loss (months)	Circumstances of the loss <sup>2</sup>	Relation to deceased	Assessment method	PGS Instrument and cut-off method	Co-occurrence Instrument and prevalence of co-occurrence with PGS
<b>Aoyama et al. (2018)</b>	Japan Cross-sectional	Bereaved family members of cancer patients were recruited from 175 institutions of Hospice Palliative Care Japan and identified through convenience sampling.	<i>n</i> = 5464 RR= 67% 14% PGS 71% women 62 years	9.5*	Non-traumatic	Family member	Questionnaire survey	BGQ Cut-off value: total score ≥ 8	Depression: PHQ-9; 58%
<b>Cozza et al. (2019)</b>	USA Cross-sectional	Bereaved individuals who lost a family member in the terror attack 9/11 were recruited via postal mail, email, advertising, and telephone calls (convenience sampling).	<i>n</i> = 454 RR= NR 35% PGS 83% women 57 years	NR	Traumatic (terror)	Family member	Questionnaire survey	ICG Cut-off value: total score ≥ 30	Depression: PHQ-9; 43% Anxiety: GAD-7; 49% PTS: PCL-5; 20%
<b>Dell'Osso et al. (2012)</b>	Italy Cross-sectional	Bereaved in- and outpatients were identified through convenience sampling at six different departments of psychiatry, at least 6 months post-loss.	<i>n</i> = 116 RR= NR 43% PGS* 66% women* 47 years*	NR	Non-traumatic	Family member or friend	Mixed (interview and questionnaire)	ICG Cut-off value: total score ≥ 25	Anxiety: ASA-27; 64% PTS: IES-r; 56%
<b>Djelantik et al. (2020)</b>	The Netherlands Cross-sectional	Bereaved individuals seeking treatment for psychological trauma, recruited from a Dutch Clinic (convenience sampling).	<i>n</i> = 458 RR= 71 28% PGS 29% women 49 years	182.4	Traumatic (NR)	NR	Questionnaire survey	TGI-SR Cut-off value: total score ≥ 61	Depression: BSI; 78% PTS: PCL-5; 92%
<b>Fisher et al. (2020)</b>	USA Longitudinal	Bereaved family members of military service members were recruited through grief support organizations, advertisements, and word-of-mouth (convenience sampling).	<i>n</i> = 581 RR= NR 32% PGS 75% women 48 years	60	Traumatic (mixed)	Family member	Questionnaire survey	ICG Cut-off value: total score ≥ 30	Depression: PHQ-9; 60% Anxiety: GAD-7; 49%
<b>Kristensen et al. (2009)</b>	Norway Cross-sectional	All Norwegians who lost a family member in the tsunami 2004 were contacted and identified through Norwegian Police Directorate and Norwegian National Population Register (census-based).	<i>n</i> = 111 RR= 58% 16% PGS* 57% women* 46 years*	26	Traumatic (natural disaster)	Family member	Mixed (Interview and questionnaire)	ICG Criteria-based cut-off	Depression: MINI; 39% PTS: MINI; 33%
<b>Latham and Prigerson (2004)<sup>1</sup></b>	USA Longitudinal	Bereaved adults were recruited through several sources: 1) New Haven Register, 2) a widowed Persons Service, 3) a community-based outreach program, and 4) advertisements, flyers, and referrals.	<i>n</i> = 309 RR= NR 11% PGS 74% women 62 years	6.3	Non-traumatic	NR	Mixed (Interview and questionnaire)	ICG-R Criteria-based cut-off	Depression: SCID; 51% PTS: SCID; 34%
<b>Li et al. (2015)</b>	China Cross-sectional	Bereaved individuals exposed to the Sichuan earthquake in 2008. One person from each household was recruited from a temporary shelter community 12–13 months after the earthquake through convenience sampling.	<i>n</i> = 803 RR= NR 71% PGS 63% women 47 years	NR	Traumatic (natural disaster)	Family member or friend	Questionnaire survey	ICG Cut-off value: total score ≥ 26	PTS: PCL-C; 39%

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Table 1 (continued)

Author (year)	Study setting and design	Sample characteristic and recruitment	Study population Study sample size; response rate; %PGS; %women; mean age	Mean time since loss (months)	Circumstances of the loss <sup>2</sup>	Relation to deceased	Assessment method	PGS Instrument and cut-off method	Co-occurrence Instrument and prevalence of co-occurrence with PGS
<b>Li and Prigerson (2016)<sup>1</sup></b>	China Cross-sectional	Bereaved individuals of first-degree relatives were recruited from two Chinese memorial websites through volunteer sampling.	$n = 1358$ RR= NR 14% PGS 51% women 42 years	26	Non-traumatic	Family member	Questionnaire survey	ICG Criteria-based cut-off	Depression: HADS; 54% Anxiety: HADS; 68%
<b>Maccallum and Bryant (2018)<sup>1</sup></b>	Australia Cross-sectional	Bereaved volunteers who had lost a family member and were interested in grief treatment or research projects were recruited through websites and advertisement.	$n = 285$ RR= NR 28% PGS* 80% women 49 years	44	Non-traumatic	Family member	Mixed (Interview and questionnaire)	PG-13 Criteria-based cut-off	Depression: BDI-II; 98% PTS: CAPS-1; 59%
<b>Maccallum and Bryant (2019)<sup>1</sup></b>	Australia Cross-sectional	Bereaved individuals who were interested in psychological treatment or grief-related research projects were recruited through advertisement in newspapers and websites (volunteer sampling).	$n = 185$ RR= NR 36% PGS 83% women 50 years	42.6	Non-traumatic	Family member	Mixed (Interview and questionnaire)	PG-13 Criteria-based cut-off	Depression: BDI-II; 97% PTS: CAPS-1; 53%
<b>McDevitt-Murphy et al. (2012)</b>	USA Cross-sectional	Bereaved individuals who experienced a murder of a loved one within the last 5 years were recruited from Victims to Victory (a help organization for homicide survivors) through convenience sampling.	$n = 44$ RR= 54% 55% PGS 89% women 49 years	20.9	Traumatic (murder)	Family member, friend, or colleague	Questionnaire survey	ICG Cut-off value: total score $\geq 30$	Depression: BDI-II; 75% PTS: PCL-C; 33%
<b>Momartin et al. (2004)</b>	Australia Cross-sectional	Bereaved Bosnian refugees were recruited from a Bosnian community residing in Sydney through snowball sampling with a mean time of 5 years since trauma/loss.	$n = 126$ RR= 86% 30% PGS* 61% women 47 years	60	Traumatic (war)	Family member, friend, or colleague	Interview-based	CBI NR	Depression: SCID; 79%
<b>Morina et al. (2010)<sup>1</sup></b>	Kosovo Cross-sectional	Bereaved individuals who lost first-degree relatives due to war-related violence 7 years earlier were identified through convenience sampling.	$n = 60$ RR= 77% 38% PGS 33% women 41 years	NR	Traumatic (war)	Family member	Interview-based	ICG-R Criteria-based cut-off	Depression: MINI, BDI-II; 65% Anxiety: BSI; 44% PTS: PDS; 65%
<b>Neria et al. (2007)<sup>1</sup></b>	USA Cross-sectional	Bereaved individuals who experienced a loss during the terror attack 9/11 were recruited 2.5–3.5 years after 9/11 through websites of 9/11 family organizations (convenience sampling).	$n = 704$ RR= 81% 43% PGS 79% women 45 years	NR	Traumatic (terror)	Family and non-family members	Questionnaire survey	ICG Criteria-based cut-off	Depression: PRIME-MD, PHQ-9; 36% Anxiety: BSI; 34% PTS: PCL-C; 43%
<b>Newson et al. (2011)<sup>1</sup></b>	The Netherlands Cross-sectional	All individuals over the age of 55 years who were living in the Ommoord district of Rotterdam were recruited and identified through cluster sampling.	$n = 5741$ RR= 75% 5% PGS 69% women* 72 years*	6.7	Non-traumatic	Family member, friend, or other	Interview-based	ICG Cut-off value: total score $\geq 22$	Depression: CESD, PSE; 10% Anxiety: CIDI; 17%

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Table 1 (continued)

Author (year)	Study setting and design	Sample characteristic and recruitment	Study population Study sample size; response rate; %PGS; %women; mean age	Mean time since loss (months)	Circumstances of the loss <sup>2</sup>	Relation to deceased	Assessment method	PGS Instrument and cut-off method	Co-occurrence Instrument and prevalence of co-occurrence with PGS
Patel et al. (2019)	USA Cross-sectional	Help-seeking individuals who lost a loved one at least 6 months ago were recruited from 6 primary care clinics through convenience sampling.	n = 1015 RR= NR 22% PGS 72% women* 41 years*	NR	NR	NR	Mixed (interview and questionnaire)	ICG Cut-off value: total score ≥ 25	Depression: PHQ-9; 39% PTS: MSE; 16%
Prigerson et al. (1999)	USA Cross-sectional	Bereaved individuals who lost a friend due to suicide were identified through snowball sampling.	n = 76 RR= 64% 20% PGS 58% women 24 years	75.6	Traumatic (suicide)	Friend	Questionnaire survey	ICG Cut-off value: total score in the upper 20%	Depression: BDI; 40%
Rheingold and Williams (2015)	USA Cross-sectional	Bereaved individuals of homicide victims murdered between 2007 and 2009 were identified through a list from the Federal Bureau of Investigation (census-based).	n = 47 RR= 30% 23% PGS 79% women 51 years	25	Traumatic (murder)	Family member	Interview-based	ICG NR	Depression: NWS Depression module; 82% PTS: NWS PTSD module; 64%
Schaal et al. (2009)	Rwanda Cross-sectional	Widows who lost their husbands during the Rwanda Genocide were recruited from nongovernment organizations and churches (convenience sampling).	n = 40 RR= NR 13% PGS 100% women 50 years	156	Traumatic (genocide)	Spouse	Interview-based	PG-13 Criteria-based cut-off	Depression: MINI; 100%
Schaal et al. (2012)	Rwanda Cross-sectional	Orphaned or widowed of the Rwanda Genocide were identified via multi-stage sampling. They were chosen randomly from subsequent households in randomly selected sectors of Butare.	n = 400 RR= 95% 8% PGS 88% women 37 years	138	Traumatic (genocide)	Family member	Interview-based	PG-13 Criteria-based cut-off	Depression: HSC-25; 88% Anxiety: HSC-25; 88% PTS: PSS-I; 84%
Shear et al. (2006)	USA Cross-sectional	Bereaved individuals who experienced a loss in the terror attack 9/11 were recruited Project Liberty, which offered free crisis counseling.	n = 70 RR= NR 44% PGS* 67% women 47 years	NR	Traumatic (Terror)	Family member, friend, or acquaintance	Interview-based	BGQ Cut-off value: total score ≥ 5	Depression: SCID; 55% PTS: NWS PTSD module; 55%
Stammel et al. (2013)	Cambodia Cross-sectional	Bereaved individuals who lost a family member during the Khmer Rouge were recruited either directly in villages or through local legal nongovernmental organizations (convenience sampling).	n = 775 RR= 49% 14% PGS 64% women 57 years	NR	Traumatic (genocide)	Family member	Interview-based	ICG-R Criteria-based cut-off	Depression: HSC-25; 83%  Anxiety: HSC-25; 77% PTS: PCL-C; 39%

Notes: Abbreviations: clinically relevant levels of prolonged grief symptoms (PGS); Clinically relevant levels of posttraumatic stress symptoms (PTS); Inventory of Complicated Grief (ICG); Inventory of Complicated Grief-Revised (ICG-R); Prolonged Grief-13 (PG-13); Core Bereavement Inventory (CBI); Brief Grief Questionnaire (BGQ); Patient Health Questionnaire 9 (PHQ-9); The Generalized Anxiety Disorder Assessment (GAD-7); The PTSD Checklist for DSM-5 (PCL-5); The Adult Separation Anxiety Symptom Questionnaire (ASA-27); The Impact of Event Scale-Revised (IES-r); The Brief Symptom Inventory Depression subscale (BSI); the MINI International Neuropsychiatric Interview (MINI); the Structured Clinical Interview for the DSM-IV (SCID); The Posttraumatic Stress Disorder Checklist (PCL-C); Hospital Anxiety and depression Scale (HADS); Beck Depression Inventory (BDI); The Clinicians Administered PTSD Scale 1 (CAPS-1); Brief Symptom Inventory (BSI); Posttraumatic stress diagnostic scale (PDS); The Primary Care Evaluation of Mental Disorders (PRIME-MD); The center for epidemiological Studies Depression Scale (CESD); Present State Examination (PSE); Composite International Diagnostic Interview (CIDI); Mental Status Examination (MSE); The National Women's Study (NWS); Hopkins Symptom Checklist-25 (HSC-25); PTSD Symptom Scale-Interview (PSS-I); Response rate (RR); Not reported (NR).

<sup>1</sup> studies not included in subgroup and moderator analyses due to missing information about co-occurrence of one or more other types of complicated grief reactions; 2) non-traumatic loss: <25% of losses in the sample were caused by traumatic events (e.g., suicide, war, disasters, terror); \*Numbers/percentages are calculated based on raw data as they were not reported in the study.

(35%) reached moderate risk of bias score, and one study (4%) had a low total risk of bias score (see Table 2). The majority of the studies had low risk of bias scores on all internal validity items ( $K = 20$ ; 87%). External validity was generally low, with 13 studies (57%) yielding high risk of bias scores on all external validity items. The low external validity was primarily related to the majority of the studies (i) used samples that differed from the general populations (i.e., trauma exposed samples, exclusively elderly people etc.), (ii) used non-probability sampling, and (iii) had high rates of non-responders (i.e., >75%,  $K = 7$ ) or failed to report non-response ( $K = 11$ ). When evaluated with meta-regression, between-study differences in estimates of co-occurrence of PGS and one or more other types of complicated grief reactions were not associated with risk of bias scores ( $\beta = -0.02$ ; 95% CI:  $-1.14$  to  $1.10$ ).

### 3.4. Meta-analysis

The pooled prevalence estimates suggested that 70% of participants with PGS have one or more co-occurring complicated grief reaction and that 46% have at least two additional co-occurring complicated grief reactions. The most prevalent co-occurring symptoms were depressive symptoms (63%), followed by anxiety (54%), and PTS (49%). As indicated by the statistically significant Q-values and the large  $I^2$  statistics, which exceeded 90% for all results, the variance of the observed prevalence estimates reflect variance in true prevalence estimates rather than sampling error. The variance of the true prevalence estimates in logit units ( $Tau^2$ ) ranged between 0.49 and 1.22, and the estimated prediction intervals were broad, e.g., showing that the true prevalence of the co-occurrence of PGS and one or more other type of complicated grief reactions for any single population will usually fall between 0.33% to 0.92%. When exploring the possibility of publication bias, there were some indications of publication bias in the direction of higher prevalence estimates of co-occurring symptoms being more likely to be published. As shown in Table 3, when imputing missing studies, the adjusted prevalence estimates were only slightly reduced.

### 3.5. Subgroup and moderator analyses

Attempting to provide a possible explanation for the considerable heterogeneity, a number of subgroup and moderator analyses were conducted. As shown in Table 3, the only between-study difference in co-occurrence reaching statistical significance was between those using interviews and those using questionnaires for assessing PGS, depression, anxiety, and PTS (84% vs. 65%,  $p = 0.029$ ). Studies using interviews appeared somewhat less heterogeneous compared to those using questionnaires ( $I^2 = 51.7$  vs.  $I^2 = 95.1$ ). None of the other subgroup analyses reached statistical significance (Western vs. Non-Western; probability vs. non-probability sampling; clinical vs. non-clinical recruitment context; criteria-based vs. cut-off based). It was not possible to conduct the subgroup analyses comparing grief instruments and circumstances of the loss (traumatic vs. non-traumatic) due to too few studies with sufficient data. The analyses of the continuous moderators revealed that longer mean time since loss in the study sample was significantly associated with higher prevalence estimates of co-occurrence ( $\beta = 0.01$ ,  $p < 0.001$ ). Response rates, mean age, and percentage of women in the study sample did not emerge as statistically significant moderators of co-occurrence.

## 4. Discussion

Only a minority of bereaved individuals experience complicated grief reactions that cause persistent suffering, but as with natural and adaptive grief, complicated grief reactions occur in different forms, including PGS, and symptoms of depression, anxiety, and PTS (Bonanno and Kaltman, 2001; Larsen et al., 2018; Rando, 2013).

The primary aim of the present systematic review and meta-analysis was to provide pooled prevalence estimates of the co-occurrence of

PGS and other types of complicated grief reactions based on the available literature. The results indicated that 70% of bereaved adults with PGS experienced co-occurrence of one or more other types of complicated grief reactions. When adjusted for the possibility of publication bias, this number was slightly reduced (64%). Taken together, our results indicate that PGS more often co-occurs with other types of complicated grief reactions than not. Furthermore, nearly half of adults with PGS (46%) experienced at least two additional complicated grief reactions. Examining the additional complicated grief reactions separately, the co-occurrence of PGS and clinically relevant levels of symptoms of depression, anxiety, and PTS were 63%, 54%, and 49%, respectively.

As high proportions of co-occurrence are often found between psychiatric disorders, the findings of high co-occurrence in the present study were expected. For example, another meta-analysis found that 52% of individuals with PTS experienced co-occurring symptoms of depression (Rytwinski et al., 2013). There could be several explanations for our findings.

First, some researchers have argued that co-occurrence of psychiatric symptoms and disorders could simply be explained as a consequence of the diagnostic systems where complex clinical conditions, such as complicated grief reactions, are categorized into separate syndromes (e.g., PGS and PTS) (e.g., Lilienfeld et al., 1994; Maj, 2005). This may result in vague diagnostic categories frequently sharing non-specific symptoms, which may artificially increase estimates of co-occurrence (van Loo and Romeijn, 2015). As considerable symptom overlap exists between PGS and depression, anxiety, and PTS (Jordan and Litz, 2014; Raphael et al., 2013; Shear and Skritskaya, 2012), symptom overlap could thus account for at least some cases of co-occurrence in the present meta-analysis. Second, transdiagnostic risk factors and mechanisms, such as negative loss-related cognitions and extreme avoidance behaviors, could offer potentially valuable explanations for the high co-occurrence (Nolen-Hoeksema and Watkins, 2011; Sauer-Zavala et al., 2017). However, to our knowledge, no coherent model of transdiagnostic risk factors and mechanisms between PGS and other types of complicated grief reactions currently exists. While the death of a loved one constitutes a particularly disruptive and stressful event that may not only trigger PGS but also increase the risk of developing symptoms of depression, anxiety, and PTS (Cole and Dendukuri, 2003; Kristiansen et al., 2019; Onrust and Cuijpers, 2006; Shear and Skritskaya, 2012), future research needs to identify transdiagnostic risk factors and mechanisms to explain the high proportion of co-occurrence in adults with PGS. Such knowledge may further contribute to more effective assessment and treatment of co-occurring cases of PGS (Nolen-Hoeksema and Watkins, 2011). Third, PGS may play a causal role in the development of symptoms of depression, anxiety, and PTS, which may contribute to high estimates of co-occurrence. Consistently, studies have shown that PGS predicted the onset or course of other types of complicated grief reactions (Djelantik et al., 2018; Lenferink et al., 2019; Melhem et al., 2004; O'Connor et al., 2015; Tsai et al., 2020). However, other researchers have emphasized that pre-existing mental illness such as depression or anxiety is a risk factor for developing PGS after a loss (Shear and Skritskaya, 2012; Simon et al., 2007). More prospective longitudinal research including pre-loss data is needed to clarify the causal relationships between different types of complicated grief reactions to better understand how co-occurring cases of PGS develop over time.

Although the present study provides the first pooled prevalence estimates of the co-occurrence of PGS and other types of complicated grief reactions, the heterogeneity was considerable ( $I^2 = 92.5$ – $95.6$ ). This suggests that the between-study variation in prevalence estimates very likely stems from true between-study differences rather than from sampling error. This was confirmed by the broad prediction intervals (95% PI: 0.33–0.92), indicating that the pooled estimates of co-occurrence do not constitute definite prevalence estimates and that estimates are likely to vary in future studies. Nevertheless, based on the available research, our results contribute with overall descriptive knowledge on



**Table 2**  
Results of risk of bias assessment.

Author (year)	External validity				Internal validity							Total risk of bias
	Representative	Sampling frame	Random selection/census	Non-response bias	Data collection	Case definition	Instrument	Mode of data collection	Prevalence period	Numerator/denominator		
Aoyama et al. (2018)	0	0	0	0	1	1	1	1	1	0	High	
Cozza et al. (2019)	0	0	0	0	1	1	1	1	1	1	High	
Dell'Osso et al. (2012)	0	0	0	0	1	1	1	1	1	1	High	
Djelantik et al. (2020)	0	0	0	0	1	1	1	1	1	1	High	
Fisher et al. (2020)	0	0	0	0	1	1	1	0	1	1	High	
Kristensen et al. (2009)	0	1	1	0	1	1	1	1	1	1	Moderate	
Latham and Prigerson (2004) <sup>1</sup>	0	1	0	0	1	1	1	1	1	1	Moderate	
Li et al. (2015)	0	0	0	0	1	1	1	1	1	1	High	
Li and Prigerson (2016) <sup>1</sup>	0	0	0	0	1	1	1	1	1	1	High	
Maccallum and Bryant (2018) <sup>1</sup>	0	0	0	0	1	1	1	1	1	1	High	
Maccallum and Bryant (2019) <sup>1</sup>	0	0	0	0	1	1	1	1	1	1	High	
McDevitt-Murphy et al. (2012)	0	0	0	0	1	1	1	1	1	1	High	
Momartin et al. (2004)	0	0	0	1	1	1	1	1	1	1	Moderate	
Morina et al. (2010) <sup>1</sup>	0	0	0	1	1	1	1	1	1	1	Moderate	
Neria et al. (2007) <sup>1</sup>	0	0	0	1	1	1	1	1	1	1	Moderate	
Newson et al. (2011) <sup>1</sup>	0	1	1	1	1	1	1	1	1	1	Low	
Patel et al. (2019)	0	1	0	0	1	1	1	1	1	1	Moderate	
Prigerson et al. (1999)	0	0	0	0	1	1	1	1	1	0	High	
Rheingold and Williams (2015)	0	0	1	0	1	1	1	1	1	1	Moderate	
Schaal et al. (2009)	0	0	0	0	1	1	1	1	1	1	High	
Schaal et al. (2012)	0	0	1	1	1	1	1	1	1	1	Moderate	
Shear et al. (2006)	0	0	0	0	1	1	1	1	1	1	High	
Stammel et al. (2013)	0	0	0	0	1	1	1	1	1	1	High	

Notes. Risk of bias assessment based on the instrument of Hoy et al. (2012). 1=low risk, 0=high risk. Total risk of bias: high risk (total score <6), moderate risk (total score=7–8), and low risk (total score=9–10).

<sup>1</sup> Studies not included in subgroup and moderator analyses due to too few studies with sufficient data.

**Table 3**  
Pooled prevalence of co-occurrence in individuals with PGS: results of the meta-analysis and subgroup analyses.

Co-occurrence of different types of complicated grief reactions	Studies	K <sup>a</sup>	Event/total N <sup>b</sup>	Proportion <sup>c</sup> (percent)	95% CI <sup>d</sup>	95% PI <sup>e</sup>	Q	Heterogeneity <i>p</i>	<i>I</i> <sup>2</sup> <sup>f</sup>	<i>Tau</i> <sup>2</sup>
One or more	All	16	1353/2334	0.70	0.61–0.78	0.33–0.92	204.3	<0.001	92.7	0.49
<i>Adjusted for possible publication bias<sup>g</sup></i>		(19)	–	0.64	0.55–0.73	–	–	–	–	–
Two or more		11	391/940	0.46	0.30–0.63	0.06–0.92	176.2	<0.001	94.3	1.22
Depression		21	1393/2651	0.63	0.53–0.72	0.21–0.91	343.0	<0.001	94.2	0.73
<i>Adjusted for possible publication bias<sup>g</sup></i>		(25)	–	0.55	0.45–0.65	–	–	–	–	–
Anxiety		9	561/1259	0.54	0.39–0.69	0.11–0.92	183.1	<0.001	95.6	0.81
PTS <sup>h</sup>		16	778/1853	0.49	0.39–0.59	0.15–0.84	199.1	<0.001	92.5	0.59
One or more	Probability sampling	3	48/61	0.75	0.49–0.91	0.00–1.00	12.1	=0.002	83.4	2.70
	Non-probability samp.	13	1305/2273	0.69	0.59–0.78	0.31–0.92	190.6	<0.001	93.7	0.47
	<i>Between-groups</i>	16					0.25	=0.617		
One or more	Clinical context	5	303/459	0.75	0.60–0.86	0.05–0.99	65.4	<0.001	93.9	1.49
	Non-clinical context	11	1050/1875	0.67	0.55–0.78	0.28–0.91	136.8	<0.001	92.7	0.44
	<i>Between-groups</i>	16					0.74	=0.391		
One or more	Criteria-based criteria	4	141/166	0.81	0.64–0.92	0.01–0.99	19.5	<0.001	84.6	1.90
	Cut-off-based criteria	10	1173/2119	0.65	0.54–0.74	0.29–0.89	141.8	<0.001	93.7	0.38
	<i>Between-groups</i>	14					2.85	=0.091		
One or more	Interviews	6	193/228	0.84	0.71–0.91	0.49–0.97	10.3	=0.066	51.7	0.27
	Survey	7	1008/1810	0.65	0.52–0.76	0.23–0.92	123.5	<0.001	95.1	0.44
	<i>Between-groups</i>	13					4.80	=0.029		
One or more	Non-Western samples	5	795/1484	0.72	0.54–0.85	0.15–0.97	103.2	<0.001	96.1	0.56
	Western samples	11	558/850	0.69	0.58–0.79	0.28–0.93	81.9	<0.001	87.8	0.54
	<i>Between-groups</i>	16					0.07	0.789		

**Notes.**

<sup>a</sup> K (number of studies) may differ (e.g., one or more < depression) due to missing information in studies about ‘one or more’ or the fact that not all types of complicated grief reactions were measured in each study.

<sup>b</sup> Total N (participants with PGS).

<sup>c</sup> Random effects model, weighted by inverse variance, calculated for  $K \geq 3$ .

<sup>d</sup> 95% confidence interval (precision).

<sup>e</sup> 95% prediction interval, calculated when heterogeneity ( $I^2$ ) > 0.

<sup>f</sup> Heterogeneity (%).

<sup>g</sup> Adjusted for publication bias with the Duval and Tweedie Trim and Fill method if  $K > 10$ . (K) = adjusted number of studies (actual + imputed “missing” studies).

<sup>h</sup> Clinically relevant levels of posttraumatic stress symptoms.

the proportion of co-occurrence of PGS and other types of complicated grief reactions, which is a starting point for uncovering the true risks of individuals with PGS experiencing other types of complicated grief reactions.

#### 4.1. Moderators of co-occurrence

A secondary aim of the present study was to examine possible moderators of co-occurrence. The included studies had several methodological differences, e.g., different assessment methods and instruments, cut-off methods, sampling methods, and recruitment context, which all could all be theorized to influence the estimates of co-occurrence (van Loo and Romeijn, 2015; Wittchen, 1996) and thus possibly explain the found heterogeneity of the co-occurrence estimates. However, only one between-study methodological difference reached statistical significance, with higher estimates of co-occurrence in studies using clinical interviews (84%) compared with studies using questionnaires as assessment method (65%). One possible explanation for our finding could be that while clinical interviews generally report lower prevalence rates compared to questionnaire-based assessment (see e.g., Georgi et al., 2019), they may be more likely to identify more severe cases of PGS, which – in turn – may have higher co-occurrence of other symptoms (cf. Simon et al., 2007).

None of the remaining methodological differences examined, e.g., sampling method, recruitment context, and cut-off method, were associated with differences in proportions of co-occurrence, a result which perhaps could be due to insufficient statistical power. Still, it should be noted that the included studies used different assessment instruments, not only for PGS, but also for the other types of complicated grief reactions, and cut-offs for PGS varied even within the same assessment instruments. While the limited number of studies did not allow us to explore the possible influence of these factors, they could be important sources of heterogeneity, as previously found in the field of bereavement (e.g., Lundorff et al., 2017).

Another explanation for the heterogenous estimates of co-occurrence may be that the included studies investigated different study populations characterized by different socio-demographic characteristics, e.g., percentage of women and mean age in the study sample, and loss-related characteristics, e.g., traumatic versus non-traumatic and different mean time since loss. However, only mean time since loss emerged as a statistically significant moderator, with longer time since loss associated with higher estimates of co-occurring symptoms. Studies have shown that cases with co-occurring PGS are more severely ill with higher levels of grief symptoms, greater functional impairment, and thus possibly longer duration of grief symptoms than those without co-occurrence (e.g., Simon et al., 2007). Higher proportions of co-occurrence could therefore be expected in individuals who suffer from PGS at a longer time after the loss. Another possible explanation for the moderating effect of mean time since loss could be that some early cases of PGS (i.e., measured around six months post-loss) may represent fluctuations in the natural grieving process (cf. the dual-process model; Stroebe and Schut, 1999) rather than pathological grief reactions per se (Lundorff et al., 2021). As a consequence, co-occurring symptoms may be less prevalent.

Although it was not possible to analyze the moderating effect of traumatic versus non-traumatic loss in the present study, higher proportions of co-occurrence may be found in bereaved individuals who experienced traumatic loss. For example, Kristensen et al. (2009) found that proportions of co-occurrence were higher among bereaved individuals who were directly exposed to a natural disaster compared with those who were not (21.9% vs. 5.2%), thus suggesting that traumatic versus non-traumatic loss may be a possible moderator of co-occurrence. In the present systematic review and meta-analysis, the majority of studies included participants who experienced traumatic loss, which could partially explain the relatively large proportion of co-occurrence found in the present study.

#### 4.2. Clinical implications

While the findings of the present meta-analysis should be considered preliminary implicating that no clinical guidelines can be established at this time, the results have potential clinical relevance. First, cases of PGS alone were less prevalent (30%) than co-occurring cases of PGS and other types of complicated grief reactions (70%). This points to the importance of screening for both depression, anxiety, and PTS as well as for PGS in bereaved patients. In the present study, we identified risk factors, which may be of relevance in the screening of bereaved patients, namely longer time since loss. Clinicians could be suggested to be particular aware of possible co-occurring complicated grief reactions in patients who continue to suffer from PGS in the long term. Second, patients who have experienced traumatic loss may be at particular risk of developing not only PGS but also other types of complicated grief reactions (Kristensen et al., 2009; Momartin et al., 2004; Raphael et al., 2013; Schaal et al., 2012), and may thereby potentially be at greater risk of developing co-occurring complicated grief reactions.

Furthermore, the high prevalence of co-occurrence in bereaved individuals with PGS may be relevant in terms of psychological treatment, as the efficacy of interventions may vary with respect to different types of complicated grief reactions (Jordan and Litz, 2014; Shear et al., 2005, 2016). If a patient suffers from co-occurring PGS and depressive symptoms, a blend of grief therapy with a particularly focus on altering negative loss-related cognitions combined with antidepressant medication could be effective (cf. Boelen et al., 2006; Shear et al., 2016). On the other hand, if a patient suffers from co-occurring PGS and PTS, trauma-focused cognitive-behavioral therapies, which include exposure, may be an effective choice (cf. Raphael et al., 2013). Likewise, the most efficacious interventions of co-occurring PGS and anxiety symptoms may also focus on exposure with the aim of reducing avoidance behaviors. Alternatively, transdiagnostic psychological interventions that target shared underlying psychopathological mechanisms may be particularly efficacious and cost-effective treatments of PGS with co-occurring complicated grief reactions (Barlow et al., 2017; Sauer-Zavala et al., 2017). However, the theoretical and empirical basis for understanding transdiagnostic psychopathological mechanisms of complicated grief reactions has yet to be established.

#### 4.3. Recommendations for future research

The findings of the present study suggest a number of recommendations for future research. First, most of the existing studies were either not representative of the general population of bereaved adults or focused specifically on traumatic losses. To achieve more robust and representative estimates, there is a need for population-based studies with a representative proportion of non-traumatic losses investigating co-occurrence in bereaved with PGS. Second, the considerable variation in assessment methods, instruments, cut-offs, and diagnostic criteria for PGS is a problem in bereavement research (cf. Lenferink et al., 2019). Accordingly, there is clear need for research focusing on developing uniform assessment methods corresponding to the ICD-11 PGD, including structured clinical interviews as gold standard for diagnosis and valid clinical cut-offs. Finally, in bereavement research, the terms ‘complicated grief’ and ‘prolonged grief’ have been used interchangeably to refer to the same grief-specific pathological condition (e.g., Boelen and Prigerson, 2013; Shear et al., 2005; Zisook et al., 2014). We would argue that it is essential to use a clear terminology, in which ‘prolonged grief disorder’ refers to a grief-specific pathological condition, as described in ICD-11 and now also planned to be included in DSM-5-TR, that can be distinguished from bereavement-related depression, anxiety, and PTS. ‘Complicated grief reactions’ on the other hand, may be reserved for a broad umbrella term that includes several different types of post-loss complications associated with the process of grief. A clearer terminology would help guide the attention of researchers as well as clinicians

to different types of post-loss complications and that PGS and symptoms of depression, anxiety, and PTS frequently co-occur.

#### 4.4. Strengths and limitations

The present systematic review and meta-analysis is the first to provide prevalence estimates of co-occurrence in adults with PGS, and has several strengths. First, the systematic review and meta-analysis is based on *a priori* developed and pre-registered research protocol and a systematic literature search in three different electronic databases supplemented with searches in reference lists of the included studies to identify additional relevant studies. Second, we explored possible sources of heterogeneity, thereby contributing with clinically and methodologically relevant insights in possible moderators of co-occurrence. Third, literature search, study selection, and risk of bias assessment were conducted independently by two reviewers, thereby reducing the risk of selection bias and assessment of study quality (Lefebvre et al., 2019).

Despite these strengths, there are also a number of limitations to be considered. First, the included studies did not assess diagnoses of mental disorders per se but assessed clinically relevant levels of PGS, depressive symptoms, anxiety, and PTS with different assessment instruments and varying cut-offs. The results of the meta-analysis can therefore not be generalized to diagnoses of mental disorders, including PGD, per se. Second, almost one third of the included studies did not provide the details needed to calculate the proportion of participants with PGS who had one or more additional co-occurring type of complicated grief reaction. Consequently, important information about estimates of co-occurrence was missing in some of the studies, which resulted in fewer studies in the meta-analysis, which could possibly challenge the robustness of the results. A third limitation is the moderate to high total risk of bias identified in the majority of the included studies, which may question the degree to which prevalence estimates represent the 'true' prevalence of co-occurrence (Hoy et al., 2012). However, the total risk of bias classification may be too conservative. For example, even well-conducted studies with relatively low risk of bias (8/10 items) were categorized as moderate risk of bias (e.g., Kristensen et al., 2009; Schaal et al., 2012). Nevertheless, low external validity seemed to be a general problem across the included studies. For example, in none of the studies, target populations were close representations of a national population. Also, the majority of studies investigated traumatic losses, and only four studies used probability sampling. As such, this challenges our ability to generalize the prevalence estimates of co-occurrence to a general population of bereaved people with PGS.

#### 5. Conclusions

The present systematic review and meta-analysis is the first to provide prevalence estimates of the co-occurrence of PGS and other types of complicated grief reactions, i.e., clinical levels of depression, anxiety, and PTS, in bereaved adults. The results indicated a pooled prevalence of 70% for co-occurrence of one or more other types of complicated grief reactions in adults with PGS and a prevalence of 46% for two or more co-occurring complicated grief reactions. The prevalence of co-occurring symptoms of depression, anxiety, and PTS were 63%, 54%, and 49%, respectively, with 30% being 'pure' cases of PGS with no co-occurrence. The results suggested considerable heterogeneity, and estimates of co-occurrence differed depending on the assessment method used, with higher estimates of co-occurrence found in studies where co-occurrence was assessed with clinical interviews compared with studies where co-occurrence was assessed with questionnaires. A second moderator of prevalence estimates was time since loss, with longer time since loss being associated with higher prevalence of co-occurrence. However, the estimates of co-occurrence should be considered preliminary, in part due to the majority of the included studies having examined traumatic losses and the relatively high risk of bias characterizing the included studies. To establish a valid estimate of co-occurrence, future

research needs to include externally valid population-based studies of co-occurrence in bereaved individuals with PGS who are representative with respect to the types of loss found in the general population.

#### Declaration of Competing Interest

The authors have none to declare.

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#### Supplementary materials

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