Review article

The prevalence of prolonged grief disorder in bereaved individuals following unnatural losses: Systematic review and meta regression analysis

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A B S T R A C T

Background: Previous research has indicated that one out of ten naturally bereaved individuals develops prolonged grief disorder (PGD). Less is known about the prevalence of PGD following unnatural deaths, such as accidents, disasters, suicides, or homicides. The aim of this study was to compute the pooled prevalence of PGD and to determine possible causes of its varied estimates.
Methods: A literature search was conducted in PsycINFO, Ovid Medline, PILOTS, Embase, Web of Science, and CINAHL. A meta-analysis using random effects models was performed to calculate the pooled prevalence rate of PGD. Multivariate meta-regression was used to explore heterogeneity among the studies.
Results: Twenty-five articles met eligibility criteria. The random-effects pooled prevalence was 49%, 95% CI [33.6, 65.4]. Death of only child, violent killings and non-western study location were associated with a higher PGD prevalence. A longer time since loss and a loss in a natural disaster were associated with a lower PGD prevalence.
Limitations: These findings should be interpreted with caution, because of the heterogeneity in study methodology.
Conclusions: This first meta-analysis of PGD following unnatural losses indicated that nearly half of the bereaved adults experienced PGD. This illustrates the importance of assessing PGD in individuals affected by loss and trauma.

1. Introduction

Unnatural deaths include sudden and violent deaths, caused by accidents, suicides, homicides, as well as disasters, terror and war (Kristensen et al., 2012b). In 2016, these causes of death approximately resulted in 2.7 million losses globally (Ritchie and Roser, 2018), leaving behind millions of bereaved family members and close friends. Several studies have indicated that unnatural deaths yield a larger risk for mental disorders in bereaved individuals than non-violent losses, such as prolonged grief disorder (PGD), posttraumatic stress disorder (PTSD), and major depressive disorder (MDD), e.g., Djelantik et al. (2017b); Farberow et al. (1992); Figley et al. (1997); Kaltman and Bonanno (2003); Murphy et al. (1999).

Theoretically, unnatural losses may cause more mental health difficulties because of the disruption of positive and self-evident assumptions about the world (i.e., 'the world is a safe place') and due to more intrusive and negative memories. Both explanations indicate that unnatural deaths lead to difficulties in integrating the loss in the autobiographical memory of the bereaved individual (Boelen et al., 2016, 2006). PGD is known to be more strongly associated with impairment in daily life than bereavement-related PTSD or MDD (Boelen and Prigerson, 2007; Silverman et al., 2000). Furthermore, in comparison with existing treatments for MDD and PTSD, PGD appears uniquely responsive to grief-specific treatments (Shear et al., 2005; Zisook and Shear, 2009). These findings underline even more the need to establish the prevalence of the condition in at-risk circumstances such as unnatural death. A recent meta-analysis by Lundorff et al. (2017) showed that one out of ten bereaved adults following a non-violent loss is at risk of developing PGD. Importantly, a meta-analysis of the prevalence of PGD following an unnatural loss has not yet been performed.

The core underlying characteristic of PGD is 'yearning for the deceased', whereas 'anxiousness' and 'depressed mood' are central to PTSD or MDD (Maercker and Znoj, 2010; Shear, 2015). Other core symptoms of PGD include 'avoidance of reminders of the death',

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'difficulties accepting the loss', and 'significant impairment of daily functioning' (Boelen and Prigerson, 2007; Silverman et al., 2000). There is some debate about the time duration of symptoms before grief turns into disturbed grief. Recently, PGD has been included in the International Classification of Diseases, 11th revision (ICD-11) (Killikelly and Maercker, 2017; Prigerson et al., 2009; World Health Organization, 2018) with a time criterium of 6 months following bereavement. Similar concepts with slightly different criteria are called persistent complex bereavement disorder (PCBD), included in the Fifth Edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5), (American Psychiatric Association, 2013) and complicated grief (Shear, 2015), and include time criteria of 12 and 6 months following bereavement, respectively. There is also an ongoing discussion about which criteria best describe the phenomenon of pathological grief (Boelen et al., 2018; Cozza et al., 2019; Maciejewski et al., 2016; Mauro et al., 2017). For consistency, we will use the term PGD for all conceptualizations of disordered grief in this paper.

Investigating the heterogeneity of the studies in a meta-analysis, such as the degree to which the prevalence is influenced by the precise cause of death, can help identify the most vulnerable group of bereaved individuals. Studies investigating PGD prevalence after unnatural losses have mostly been conducted in homogeneous samples, such as individuals who lost loved ones in a large accident, e.g., Cardoso et al. (2017), a natural disaster, e.g., Hu et al. (2015), or a specific terrorism incident, e.g., Neria et al. (2007). However, substantial heterogeneity in PGD prevalence rates can be expected across studies due to differences in study methodology, sample demographic features, and loss-related characteristics.

Several reviews have discussed predictors and correlates of PGD in both non-violent and unnatural bereaved populations (Boelen et al., 2018; Hibberd et al., 2010; Kristensen et al., 2012b; Lobb et al., 2010). In one of the most recent reviews about correlates of PGD following a violent loss, a meta-analysis was conducted in which significant effect sizes were found with regard to the following correlates: comorbid psychopathology, suicidality, rumination, the relationship with the deceased, exposure to earlier traumatic events, age and prior history of counseling (Heeke et al., 2019). However, because each reviewed study assessed different sets of correlates, only univariate analyses were possible. More knowledge about predictors for PGD in unnatural bereaved populations may improve decision-making regarding the development of appropriate strategies for identifying and improving mental health problems after unnatural loss.

The purpose of this meta-analysis was to estimate a pooled-prevalence of PGD in individuals bereaved due to unnatural causes. Therefore, we performed a literature search to summarize all published scientific evidence. Additionally, we explained heterogeneity with a multivariate meta-regression analysis.

2. Methods

2.1. Literature search strategy

The meta-analysis was performed and reported in accordance with the PRISMA guidelines for systematic reviews and meta-analyses (Moher et al., 2010). The protocol was registered in PROSPERO – an international prospective register for review protocols – in January 2018 (registration number CRD42018084631).

The literature search was conducted using the following databases: PsycINFO, Ovid Medline, PILOTS, Embase, Web of Science, and CINAHL, and was completed on December 5, 2017. The search strategy was designed in accordance with the Population, Intervention/Interest, and Outcome (PIO) principles (WHO, 2014). The following PIO was formulated: population: *adults*; intervention/interest: *bereavement due to unnatural loss*; outcome: *prevalence of PGD*. Subsequently, synonyms for the keywords *PGD, unnatural loss*, and *prevalence* were combined in the full search strategy (Supplementary Material A).

The abstracts of all the articles were imported to the online software Covidence (Covidence, 2016). Then, the eligibility of both the abstracts and full-text articles were independently reviewed by MD and AM using this software. After the records were screened based on the titles and abstracts, the abstracts of potentially eligible articles were reviewed by both reviewers and were excluded if both reviewers agreed that the eligibility criteria were not met. Thereafter, potentially eligible articles were full-text screened and reviewed to determine final eligibility. Subsequently, the 'related articles' function in PubMed was used to further explore the literature. In addition, all the articles that cited the included articles were screened using Scopus, and all reference lists were searched by hand in order to ascertain that no relevant articles were missed. Duplicates were excluded using Covidence.

Following both the title and abstract screening, as well as the full-text screening, the interrater agreement was evaluated using Cohen's Kappa statistic, κ (McHugh, 2012). During the title and abstract screening, the researchers disagreed on 326 (9%) papers yielding an interrater reliability of κ = 0.64 (p < .001), indicating substantial agreement. During the full-text screening, the researchers disagreed on 18 (5%) papers yielding an interrater reliability of κ = 0.89 (p < .001), indicating almost perfect agreement. All disagreements were discussed until both researchers agreed about the in- or exclusion.

2.2. Selection criteria

All original articles written in English examining the prevalence of PGD in bereaved individuals following unnatural losses were retrieved. To be eligible for inclusion, published studies had to be conducted on humans, and the participants' age had to be 18 years or older. As we were exclusively interested in bereaved individuals following unnatural losses, participants had to have suffered the loss of a loved one due to unnatural death causes. The PGD diagnosis had to be established using a standardized psychometric instrument or clinical interview based on the criteria of a PGD conceptualization (see Table 1 for the assessment tools we have found for PGD in the included studies of this meta-analysis). Finally, because we were specifically interested in the prevalence of the disorder, the studies had to provide a prevalence estimate of PGD or other data that could be used to calculate prevalence estimates.

We excluded the following studies: studies with participants below 18 years of age, studies including patients/mental health service users, and studies focusing on other forms of loss than bereavement due to unnatural loss. Studies including both natural and unnatural deaths or other mixed groups were only included if data about the outcome of interest could be retrieved. All intervention studies and non-empirical

Table 1
Assessment tools used in the included articles.

Assessment instrument	Author (year)	Number of items	Recommended cut-off score	Time criterion	Outcome
PG-13	Prigerson et al. (2008)	13	Criteria-based	6 months	Prolonged grief disorder
ICG	Prigerson et al. (1995)	19	>25	6 months	Complicated grief
ICG-R	Prigerson et al. (2001)	34	Criteria-based	6 months	Prolonged grief disorder
Criteria set for diagnosing complicated grief	Prigerson et al. (2004)	9	Criteria-based	6 months	Complicated grief
BGQ	Ito et al. (2012)	5	>8	12 months	Complicated grief

Note. BGQ: Brief Grief Questionnaire; ICG: Inventory of Complicated Grief; ICG-R: Inventory of Complicated Grief Revised; PG-13: Prolonged Grief-13.

studies (e.g., reviews and commentaries) were also excluded.

2.3. Quality assessment and risk of bias

To assess the included studies for possible risk of bias (RoB) and to rate the quality of evidence and study limitations, we used the risk of bias tool specifically developed for population-based prevalence studies (Hoy et al., 2012; Lundorff et al., 2017). Using this tool helped us determine the overall strength of the evidence included in the meta-analysis. Each study was scored for potential risk of bias based on the number of criteria from the tool that were met. Studies with scores of 9–10 were considered to have low RoB, studies with scores of 7–8 were considered to have moderate RoB, and studies with scores of 6 or less were considered to have high RoB (Hoy et al., 2012). The risk of bias was evaluated and double-checked independently by MD and AM.

2.4. Data extraction

Information extracted from each eligible article included: the year of publication, country in which the study was conducted, sample size, number of bereaved individuals following unnatural losses eligible for the analysis, the diagnostic instrument used and the mean score, cut-off values used by the authors, terminology of the grief investigated, the age range and mean age of participants, cause of death, relationship to the deceased, years of education, time since loss, number of female participants, recruitment method, response rate, and the RoB score. The data-extraction was performed and double-checked independently by MD and AM.

2.5. Statistical analysis

First, the raw data were converted into event rates (ER), which are defined as the proportion of the occurrence of PGD in a certain study. The ERs were calculated with 95% confidence interval (CI) for each study. A random effects model was used, because of expected heterogeneity between the studies (Borenstein et al., 2011; Egger et al., 2008). When the included studies showed no events of PGD, thus when the outcome proportions were 0, 0.5 was added to both the ER and the sample size cell before applying the logit transformation (Harrer et al., 2019; Smid et al., 2009).

Heterogeneity was assessed with the Cochran Q statistics for each analysis, where higher values suggest larger heterogeneity. In addition, the between-study heterogeneity was assessed by I2 statistics (Higgins et al., 2003). To gain insight into potential causes of heterogeneity, we originally planned to conduct a subgroup analysis in the pre-registration. A subgroup analysis is a theory-driven approach in which correlates are assessed that are not necessarily present in all studies. However, the primary aim of our study was to gain insight into potential causes of heterogeneity of the prevalence rates and not to perform a comprehensive review about effect sizes of all possible correlates. Therefore, shortly after the pre-registration we decided to change our additional analysis into a more data-driven multivariate meta-regression analysis. This approach would give us the possibility to explore the heterogeneity of the studies, while taking into account shared explained variance of the predictors (Smid et al., 2009). Only variables that were evaluated in all studies could be included using this approach. These variables were: months since loss, use of the Inventory of Complicated Grief (ICG; vs. other instruments), interview (vs. self-report questionnaire), female (vs. male) gender, sample mean age, high-income country (as defined as membership of the Organization for Economic Co-operation and Development (OECD) vs non-members), cause of death in 4 categories (suicide, natural disaster, or intentional killing vs. accident), and death of only child (vs. all kinds of other relationships). Highly correlated predictors (i.e., $r \geq 0.8$) may cause over fitting of our meta regression model to our data. Therefore, before we included these predictors in the meta-regression analysis, we assessed the multicollinearity with an inter correlation matrix (Harrer et al., 2019).

There is a chance that studies might exist that were not published because of the inadequate size and/or the lack of significance of the prevalence estimates. If this is the case, a meta-analysis of the published literature might be misleading. Therefore, to examine the publication bias, Egger's t statistics was used (Egger et al., 1997).

Finally, we tested the robustness of our findings with a permutation test in 1000 iterations. In this test, resampling methods are used to assess our meta regression model in slightly different datasets each time. After this, it recalculates the p-values of the predictors taking into account all iterations (Higgins and Thompson, 2004).

All analyses were conducted using Rstudio (version 1.1.463) (RStudio Team, 2015) with the packages meta (Schwarzer, 2007), metafor (Viechtbauer, 2010) and dmetar (Harrer et al., 2019).

3. Results

3.1. Search results

The initial search identified 643 records in PsycINFO, 744 records in Ovid Medline, 425 records in PILOTS, 2078 records in Embase, 687 records in Web of Science, and 321 records in CINAHL. After exclusion of duplicates, 3556 records were screened based on the titles and abstracts. Thereafter, 375 studies were reviewed full-text for the inclusion or exclusion criteria. The screening process and the number of studies at each stage of the search are detailed in Fig. 1. Eighty-five studies were excluded after the full-text review and 32 studies were finally considered for inclusion in the meta-analysis.

Subsequently, two articles were excluded because it was not possible to estimate the prevalence of PGD from the presented data (Boelen et al., 2016; Keesee et al., 2008). Another five articles were excluded from the main analysis because of sharing the same sample population with other articles (Huh et al., 2017; Kristensen et al., 2016, 2009, 2010; Lenferink et al., 2017; Seirmarco et al., 2012). In such cases, the studies with the highest number of bereaved individuals or studies providing the most relevant data were included. The characteristics of all 25 included studies can be found in Table 2 (Bartik et al., 2013; Cardoso et al., 2017; Dyregrov et al., 2015, 2003; Harms et al., 2015; Hu et al., 2015; Kristensen et al., 2012a, 2015; Li et al., 2015; McDevitt-Murphy et al., 2012; Mitchell et al., 2004; Morina and Emmelkamp, 2012; Morina et al., 2010, 2011; Neria et al., 2007; Prigerson et al., 1999; Schaal et al., 2009; Shear et al., 2006; Spooren et al., 2001; Stammel et al., 2013; Tsutsui et al., 2014; van Denderen et al., 2016; Williams and Rheingold, 2015; Xu et al., 2014; Yun et al., 2018).

3.2. Study characteristics

The included studies were published between 1999 and December 2017. These studies comprised a total of 4774 participants (range $n = 10$ to 803) of whom 2296 (48%) were identified as PGD cases. Of the study participants 68% was female. The mean age of the included samples ranged from 20 (Morina et al., 2011) to 72 years (Kristensen et al., 2012a). Seventeen studies were performed in OECD membership countries: six in North America (USA), four in Norway, one in the Netherlands, one in Belgium, one in Portugal, one in Korea, one in Japan, and two in Australia. Eight studies were performed in non-OECD membership countries: three in Kosovo, three in China, one in Cambodia, and one in Rwanda. The studies assessed PGD using either the Prolonged Grief-13 (PG-13), the Inventory of Complicated Grief (ICG) or the Inventory of Complicated Grief-Revised (ICG-R) (Tables 1 and 2). Two studies (Neria et al., 2007; Shear et al., 2006) used self-constructed questionnaires based on criteria of PGD. The mean time post-loss in the studies varied from less than 1 month (Mitchell et al., 2004) to 30 years (Stammel et al., 2013) (Table 2). According to the
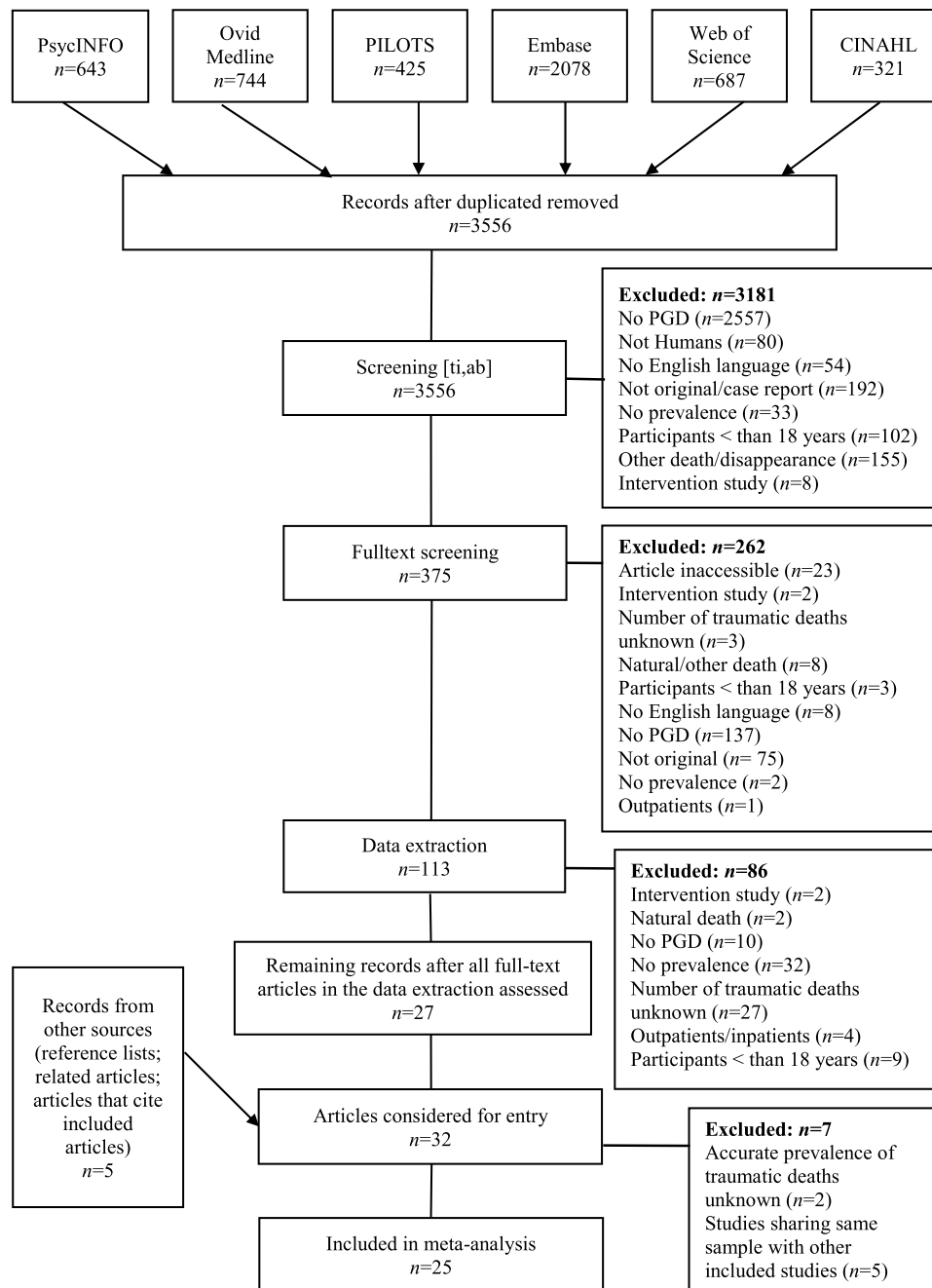
Fig. 1. PRISMA flowchart for the selection of studies. .

quality assessment scores, four studies scored low (score = 9–10), twelve studies scored moderate (score = 7–8), and nine studies scored high (score = 6 or less), on Risk of Bias (See Supplementary Material B for the detailed scores for the RoB of each study).

3.3. Meta-analysis

In total, twenty-five articles with thirty prevalence estimates were included in the overall meta-analysis. The pooled prevalence rate of PGD was 49%, 95% CI [33.6, 65.4]. Effects of the individual studies showed a high level of heterogeneity ($Q = 1090.2$, df = 29, $I^2 = 97.3\%$, $p < .001$). Egger's t-test did not indicate publication bias (intercept = -0.104, SE = 1.93, 95% CI [-362, 3.82], $t = 0.054$, df = 28, $p = .95$) (Fig. 2).

3.4. Multivariate meta-regression analysis

In the correlation matrix we did not find correlations of more than 0.8 and all predictors could be included in the meta-regression analysis (Supplementary Materials C). The meta-regression, shown in Table 3, explained 56% of the between study variance in PGD prevalence. Use of the ICG as diagnostic instrument and death of only child were associated with a higher PGD prevalence. Conversely, longer time since the loss, high-income country, and a natural disaster causing the death (vs. accidental death) were associated with a lower PGD prevalence. The meta-regression model itself was significant (Q_m (10) = 34.6, $p = .001$).

In the permutation test, the predictors longer time since loss and a natural disaster were considered as robust predictors (i.e., $p < .05$), the use of the ICG and high-income country were considered robust trends

Table 2
Characteristics of the included studies.

Author (year)	Study setting	Assessment instrument	Cutoff value	Interview	Assessment score, M (SD)	Sample recruitment and characteristics	Sample size	Bereaved individuals eligible for the analysis (n)	Cause of death	% female	Mean age (SD)	Mean time post-loss (months)	Relation to the deceased	Risk of bias
Bartik et al. (2013)	Australia	PG-13	N/A	self-report	N/A	Advertisement in local and regional media	10	10	Suicide	80%	24	56.4	Friends and family members	High
Cardoso et al. (2017)	Portugal	ICG	≥30	self report	36.1 (11.5)	Recruitment via Associação dos Familiares das Vítimas das Entre-os-Rios	40	20 (road accident victims)	Road accident	80%	46.3 (18.1)	120[b]	Immediate family members	Moderate
Dyregrov et al. (2015);	Norway	ICG	≥25	self-report	36.0 (11.1)	National Population Register	103	67 (parents)	Terrorism (Utoya)	55%	Mothers: 51.0 (6.9), Fathers: 52.0 (6.3)	18	Friends and family members	Moderate
Dyregrov et al. (2003)	Norway	ICG	>25	self-report	Suicide: 35.3 (13.5), accident 38.0 (SD: 15.0)	National police register	232	196 (suicide: 128, accident: 68)	Suicide, Accident	65%	47.2	Suicide: 15, accident: 14	Parents	Moderate
Harms et al. (2015)	Australia	ICG	N/A	self-report	N/A	Part of the Beyond Bushfires study	294	278	Natural Disaster (Black Saturday disaster)	40%	52.5	25.5	Immediate family; extended family; family connections; friends; community members	Moderate
Hu et al. (2015)	China	ICG	≥25	self-report	52.77 (10.00).	Recruitment from three of the hardest hit areas by convenience sampling	271	271	Natural disaster (Earthquake)	55%	44.9	18	Lineal relatives within three generations.	Moderate
Kristensen et al. (2012)	Norway	ICG	>25	self-report	N/A	Participants were contacted by the Brigade psychiatrist as part of the early intervention and research program	32	16 (assessed with ICG)	Natural disaster (snow avalanche)	50%	Mothers: 70.6, fathers: 73.4	276[b]	Parents	Moderate
Kristensen et al. (2015)	Norway	ICG	>25	self-report	N/A	A list of the deceased was obtained from the Norwegian Police Directorate and the next of kin identified through the Norwegian National Population Register	94	94	Natural disaster (tsunami)	40%	49.2	24	Adult children; parents; siblings; spouses /cohabitants	Moderate
Li et al. (2015)	China	ICG	≥26	self report	N/A	The survey was conducted in a	803	803	Natural disaster (Earthquake)	63%	46.7 (15.5)	12.2[b]	Child; spouse; other family; friend	Moderate

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Table 2 (*continued*)

Author (year)	Study setting	Assessment instrument	Cutoff value	Interview	Assessment score, M (SD)	Sample recruitment and characteristics	Sample size	Bereaved individuals eligible for the analysis (n)	Cause of death	% female	Mean age (SD)	Mean time post-loss (months)	Relation to the deceased	Risk of bias
McDevitt-Murphy (2010)	USA	ICG-R	≥30	self-report	32.46 (15.34)	temporary shelter community	137	54	Homocide	89%	48.61	21.58	Family members	High
Mitchell et al. (2004)	USA	ICG	>25	self-report	37.44 (14.16)	Participants were recruited with the victim advocate organization Victims to Victory (VTV)	60	60	Suicide	72%	43.3 (13.7)	0.5[b]	Spouses; parents; children; siblings; in-laws, aunts/uncles; nieces/nephews; friends or coworkers	High
Morina et al. (2012)	Kosovo	PG-13	N/A	interview	N/A	Lists of families with war-related loss were provided by communal authorities	206	100 (lone mothers)	War	100%	50.1 (7.9)	120[b]	Spouses	Low
Morina et al. (2010)	Kosovo	ICG-R	N/A	self-report	28.7 (7.2)	List of people killed during the war in 1998/1999 was provided by communal services	60	60	War	33%	40.6 (10.9)	90[b]	Children; siblings, parents, spouses	Low
Morina et al. (2011)	Kosovo	PG-13	N/A	interview	N/A	Lists of all families who had lost relatives during the war were provided by communal authorities	179	179	War	58%	20.3 (3.7)	120[b]	Young adults who lost their father during war	Low
Neria et al. (2007)	USA	Criteria set for diagnosing complicated grief	N/A	self-report	N/A	A convenience sample was recruited using an online invitation through Web sites of 9/11 family organizations or sent to the members of such organizations	704	704	Terrorism (9/11)	79%	45.1 (11.5)	36[b]	Child; Spouse; Parent; Other family member; Non-family Member	High
Prigerson et al. (1999)	USA	ICG	Upper 20% on the measure	self-report	N/A	Family members nominated peers whom they considered closest to the victim	76	76	Suicide	58%	23.8 (1.8)	6.3 (0.6)	Friends	Moderate
Schaal et al. (2009)	Rwanda	PG-13	N/A	interview	14.90 (8.10)	The sample was selected by the African	40	40	Genocide	100%	49.9 (9.0)	156[b]	Wives	High

Table 2 (*continued*)

Author (year)	Study setting	Assessment instrument	Cutoff value	Interview	Assessment score, M (SD)	Sample recruitment and characteristics	Sample size	Bereaved individuals eligible for the analysis (n)	Cause of death	% female	Mean age (SD)	Mean time post-loss (months)	Relation to the deceased	Risk of bias
Shear et al. (2006)	USA	BCQ	N/A	interview	N/A	Evangellistic Enterprise, the Methodist Church, and the Baptist Church; Project Liberty counselors invited service recipients to provide feedback on services	149	70	Natural disaster (tsunami)	69%	45.8 (14.7)	18	Child; spouse; sibling; other relative; friend; acquaintance	High
Spooren et al. (2000)	Belgium	ICG	>37	self-report	44.7 (15.1)	Recruitment via the organization 'Parents of Children who died in road traffic accidents', a self-help group	85	84	Accidents (Road accident)	61%	48 (N/A)	4.0 (38.0)	Parents	High
Stammel et al. (2013)	Cambodia	ICG-R	N/A	interview	21.0 (7.4).	Recruitment with help of local legal non-governmental organizations	775	775	Genocide	64%	56.7 (10.3)	360[b]	Spouses; children; parents; siblings; distant relative	High
Tsutsui et al. (2014)	Japan	ICG	≥26	self-report	10.5 (10.1)	Questionnaires were sent to all 88 hospital employees	82	82	Natural disaster (Tsunami/earthquake)	82%	45.8 (10.3)	8	E.g., family members, relatives, friends, colleagues, or neighbors	Moderate
van Denderen et al. (2016)	Netherlands	ICG	>25	self-report	N/A	Recruitment via support organizations for homicidally bereaved individuals / case managers of a GO	312	312	Homicide	65%	53.4 (15.5)	Casemanager group 3.1; support group: 9.4	Spouses; child; parent; sibling; other family; friends	High
Williams et al. (2015)	USA	ICG	N/A	interview	N/A	Contact information for the bereaved family members was provided to project staff by local law enforcement victim advocates offices	47	47	Homicide	79%	50.8 (11.1)	25[b]	Parents; children; siblings; spouses; grandparent; grandchild	Moderate
Xu et al. (2014)	China	ICG	≥25	interview	N/A	Community based recruitment: the bureau sent invitation letters with response forms to each	226	226	Natural disaster (earthquake)	100%	Women with. subsequent child: 39.4; women with. no	32[b]	mothers	Low

Table 2 (*continued*)

Author (year)	Study setting	Assessment instrument	Cutoff value	Interview	Assessment score, M (SD)	Sample recruitment and characteristics	Sample size	Bereaved individuals eligible for the analysis (n)	Cause of death	% female	Mean age (SD)	Mean time post-loss (months)	Relation to the deceased	Risk of bias
						woman meeting the inclusion criteria					subsequent child 40.3			
Yun et al. (2018)	Korea	ICG	≥25	self-report	52.3 (14.647)	Participants were recruited with the cooperation of the Ansan Mental Health Trauma Center	56	56	Accident (Sewol ferry accident)	61%	46.0 (8.4)	18 (1)	Parents; siblings; grandparents; aunt; wife who lived with the victims	Moderate

Note. CGA = Complicated Grief Assessment; ICG = Inventory of Complicated Grief; ICG-R = Inventory of Complicated Grief Revised; N/A = Not Available; PG-13 = Prolonged Grief-13.

(i.e., $p < .10$) and the death of only child was not considered as a robust predictor (i.e., $p > .10$) (Supplementary Materials D).

4. Discussion

We performed a meta-analysis on the prevalence of PGD in individuals bereaved due to unnatural death causes. The meta-analysis data were based on twenty-five studies and a population of 4774 participants. A pooled prevalence of nearly 50% of individuals screening positive for PGD was found. This result suggests that five out of ten unnaturally bereaved individuals developed PGD. This indicates a much higher prevalence of disturbed grief than the earlier reported prevalence of 10% by Lundorff et al. (2017), who investigated PGD in individuals bereaved due to non-violent death causes. Therefore, our results support the assumption that bereavement due to unnatural loss leads to more disturbances that are indicative of a diagnosis of PGD than bereavement due to non-violent loss (Boelen et al., 2006; Djelantik et al., 2017b; Farberow et al., 1992; Figley et al., 1997; Kaltman and Bonanno, 2003; Murphy et al., 1999).

Our meta-analysis also revealed a high amount of variability ($I^2 = 97.3\%$) in the prevalence estimates among the studies. Therefore, we conducted a multivariate meta-regression analysis to investigate the influence of potential variables and study characteristics on the pooled prevalence.

Longer time since loss was robustly associated with lower PGD prevalence rates. This finding accords with previous research which showed that grief symptoms normally decrease over time, and that most bereaved individuals eventually adjust to the loss and thereby return to adaptive functioning (Bonanno, 2004; Kristensen et al., 2012b). However, as shown in the study of Stammel et al. (2013), bereaved individuals may experience symptoms of PGD even after three decades following the unnatural loss of their loved one. According to Prigerson et al. (2009), the PGD diagnosis can be set after 6 months' post-bereavement. However, several other researchers argue that the 6-month time frame is too short and may lead to over-inclusion of the recently bereaved (Rubin et al., 2008). In the present meta-analysis, only two studies included participants in the first year after loss (Mitchell et al., 2004; Tsutsui et al., 2014), therefore we were not able to compare PGD rates between 0–6, 6–12 and more than 12 months after the loss. Further research is needed to clarify this further.

With regard to *the nature of traumatic events*, we found that suicide, intentional killing, and accidents were not significant predictors for PGD. Furthermore, in the studies among bereaved survivors of natural disasters, lower prevalence rates of PGD were found compared to other causes of unnatural losses. We could think of two reasons to explain this finding. In case of a natural disaster, there are most of the time multiple victims in the same area, which could result in a collective feeling that victims need to support each other in this shared trauma. In several meta-analyses social support has been proven to be a protective factor after a traumatic event (Brewin et al., 2000; Ozer et al., 2003). In a prior study, we have found that acceptance in the first year of the loss predicted a more favorable grief trajectory across the second year (Djelantik et al., 2017a). It could be that in a natural disaster, it is easier for the bereaved individual to accept the unnatural loss of the loved one, compared to for instance, intentional killings.

Our multivariate meta-regression showed that PGD prevalence rates found in studies performed in low- and middle-income countries were significantly higher than in *high-income countries*. However, findings with regard to cross-national and cross-cultural differences should be interpreted with caution, since a diagnosis of PGD implies that the grief responses exceed the norms for behavior considered normal for a culture. To avoid inappropriate diagnosis of PGD, it is important for clinicians to examine cultural ways of dealing with bereavement and grief in a diversity of settings, in particular outside the western world (Killikelly et al., 2018; Rosenblatt, 2008; Smid et al., 2018; Tay et al., 2016). Furthermore, disasters and wars in low- and middle-income
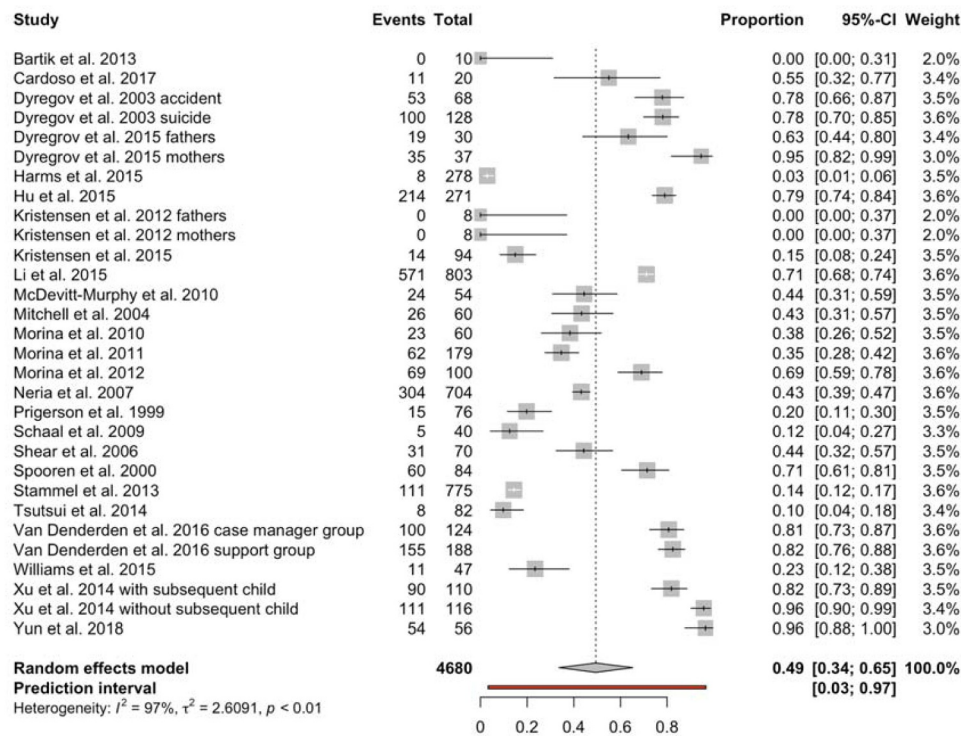
Study	Events	Total	Proportion	95%-CI	Weight
Bartik et al. 2013	0	10	0.00	[0.00; 0.31]	2.0%
Cardoso et al. 2017	11	20	0.55	[0.32; 0.77]	3.4%
Dyregov et al. 2003 accident	53	68	0.78	[0.66; 0.87]	3.5%
Dyregov et al. 2003 suicide	100	128	0.78	[0.70; 0.85]	3.6%
Dyregrov et al. 2015 fathers	19	30	0.63	[0.44; 0.80]	3.4%
Dyregrov et al. 2015 mothers	35	37	0.95	[0.82; 0.99]	3.0%
Harms et al. 2015	8	278	0.03	[0.01; 0.06]	3.5%
Hu et al. 2015	214	271	0.79	[0.74; 0.84]	3.6%
Kristensen et al. 2012 fathers	0	8	0.00	[0.00; 0.37]	2.0%
Kristensen et al. 2012 mothers	0	8	0.00	[0.00; 0.37]	2.0%
Kristensen et al. 2015	14	94	0.15	[0.08; 0.24]	3.5%
Li et al. 2015	571	803	0.71	[0.68; 0.74]	3.6%
McDevitt-Murphy et al. 2010	24	54	0.44	[0.31; 0.59]	3.5%
Mitchell et al. 2004	26	60	0.43	[0.31; 0.57]	3.5%
Morina et al. 2010	23	60	0.38	[0.26; 0.52]	3.5%
Morina et al. 2011	62	179	0.35	[0.28; 0.42]	3.6%
Morina et al. 2012	69	100	0.69	[0.59; 0.78]	3.6%
Neria et al. 2007	304	704	0.43	[0.39; 0.47]	3.6%
Prigerson et al. 1999	15	76	0.20	[0.11; 0.30]	3.5%
Schaal et al. 2009	5	40	0.12	[0.04; 0.27]	3.3%
Shear et al. 2006	31	70	0.44	[0.32; 0.57]	3.5%
Spooren et al. 2000	60	84	0.71	[0.61; 0.81]	3.5%
Stammel et al. 2013	111	775	0.14	[0.12; 0.17]	3.6%
Tsutsui et al. 2014	8	82	0.10	[0.04; 0.18]	3.4%
Van Denderden et al. 2016 case manager group	100	124	0.81	[0.73; 0.87]	3.6%
Van Denderden et al. 2016 support group	155	188	0.82	[0.76; 0.88]	3.6%
Williams et al. 2015	11	47	0.23	[0.12; 0.38]	3.5%
Xu et al. 2014 with subsequent child	90	110	0.82	[0.73; 0.89]	3.5%
Xu et al. 2014 without subsequent child	111	116	0.96	[0.90; 0.99]	3.4%
Yun et al. 2018	54	56	0.96	[0.88; 1.00]	3.0%
Random effects model		**4680**	**0.49**	**[0.34; 0.65]**	**100.0%**
Prediction interval				**[0.03; 0.97]**	

Heterogeneity: I^2 = 97%, τ^2 = 2.6091, p < 0.01

Fig. 2. Forest plot and the prevalence estimates of prolonged grief disorder following traumatic bereavement.

Table 3
Meta-regression Analysis (N = 30 Samples).

	β	SE	p	95%CI		
Study methodology						
Months since loss	−0.1	<.001	<.001	−0.02	0.00	**
Interview	−0.53	0.73	0.47	−1.95	0.89	
ICG	1.71	0.69	0.01	0.36	3.07	*
Demographic features						
Female	0.01	0.01	0.21	−0.01	0.03	
Age	0.02	0.03	0.50	−0.04	0.09	
High-income country	−1.51	0.68	0.03	−2.84	−0.19	*
Event-related characteristics						
Suicide	−1.16	0.80	0.15	−2.72	0.40	
Natural disaster	−2.24	0.72	<.001	−3.65	−0.83	**
Intentional killing	0.07	0.78	0.93	−1.46	1.60	
Death of only child	2.93	1.38	0.03	0.22	5.65	*

Note. β = regression coefficient; CI = Confidence Intervals; p = p-value;
SE = standard error ;.
* p < .05, ** p < .01, *** p < .001.

countries tend to be more devastating in terms of casualties and damage of (social) infrastructure.

Death of only child was a strong predictor for experiencing PGD in our model. However, this predictor was not considered robust in the permutation test, most likely because this predictor was only assessed in one included study (Xu et al., 2014). Previous research has clearly shown that the experience of losing a child is one of the most painful and stressful events that an adult can experience (Keesee et al., 2008; Rubin and Malkinson, 2001; Wijngaards-de Meij et al., 2005). More research is needed to validate this finding.

4.1. Limitations

Several limitations were found at review level. First, generalization of our pooled prevalence estimate needs to be conducted with caution. Although our analysis did not show any publication bias, the quality assessment revealed that most of the included studies exhibited some

methodological limitations. Many studies were not accurate representations of the target population (e.g., many studies used small sample sizes, and the recruitment was often performed through advertisement on the internet) and used other sampling methods than a random procedure. This could have resulted in a higher prevalence rate because people with disturbances were disproportionally sampled.

Furthermore, we found that the prevalence rate was higher in studies using the Inventory of Complicated Grief (ICG) than in studies using other *measurement instruments*. This could be explained by the slightly different criteria between these tools and/or calculation methods used by the authors, as well as score differences possibly due to translations to other languages or adaptations (Tables 1 and 2). As suggested by Lundorff et al. (2017), it would be highly preferable to develop one standardized assessment tool for PGD, with high specificity and sensitivity.

Thirdly, it is important to keep in mind that in most included studies the prevalence rates were based on self-report questionnaires. Scoring above a clinical cut-off of a self-report questionnaire should be recognized as an indication of disorder. A structured clinical interview is needed for a formal diagnosis.

We used a multivariate approach to explore the heterogeneity of our meta-analysis to assess predictors while taking into account their possible shared variance. A limitation of this meta-regression analysis is that we only could include predictors of which information was present in all included papers. Furthermore, our search syntax was aimed to finding studies with a reported prevalence for PGD and not to finding all possible correlates for PGD. Therefore, the amount of significant correlates is less than in the recent meta-analysis with univariate analyses by Heeke et al. (2019). Our study therefore provides preliminary insights in a multivariate assessment of risk factors for PGD. Further and more elaborate multivariate exploration of risk factors for developing PGD after unnatural losses is warranted.

5. Conclusions

This study indicates that unnatural losses are associated with a considerably higher reported rate of PGD than non-violent losses.

Globally, approximately half of the bereaved individuals might develop symptoms which meet the diagnostic criteria for PGD following unnatural losses. Our results imply that bereaved parents who lost their only child, and bereaved individuals following violent killings such as suicide, accidents, homicide, and war-related deaths are most vulnerable for developing PGD. Because of the limitations on review level due to varying sample sizes, sets of criteria and sampling bias in the included studies, future studies on the prevalence of PGD are highly recommended. They should ensure representativeness by, for example, using random sampling and an assessment tool for PGD with high sensitivity and specificity. Fundamentally, our findings strongly suggest that policy makers, public health doctors, researchers and clinicians working with people confronted with trauma and unnatural loss should be aware of bereavement and PGD symptoms.

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CRediT authorship contribution statement

A.A.A. Manik J. Djelantik: Conceptualization, Methodology, Investigation, Data curation, Supervision, Writing - original draft, Validation. **Geert E. Smid:** Data curation, Writing - review & editing, Validation. **Anna Mroz:** Investigation, Data curation, Validation. **Rolf J. Kleber:** Supervision, Writing - review & editing, Validation. **Paul A. Boelen:** Supervision, Writing - review & editing, Validation.

Declaration of Competing Interest

The authors declare that they have no conflict of interest to report.

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Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.jad.2020.01.034.

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