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Complicated Grief, Depression, Health and Attachment Style in First Degree Relatives of Individuals with a Chronic Psychotic Disorders

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Abstract

Complicated grief (CG) is a form of unrelenting grief after the death of a loved one. However, family members of individuals who suffer from Schizophrenia, Schizoaffective disorders and Bipolar disorder may experience symptoms of CG even though their loved one is still alive. The present study assessed CG and risk factors for CG in first degree relatives of individuals with severe chronic mental illness. The incidence of CG was examined in 78 parents, siblings, adult children and spouses recruited through organizations and social media that provide support services for individuals suffering from mental illness and their families. High rates of CG (39.7%) were found in this group. CG was associated with a higher prevalence of posttraumatic and depression symptoms and poorer physical health. These findings may contribute to heightening therapists' awareness of the importance of assessing, acknowledging and resolving CG in the family members of patients with chronic psychotic disorders.

Keywords Schizophrenia · Complicated grief · Caregivers · Bipolar disorder · Chronic psychotic disorders

Introduction

The term dementia praecox (premature dementia), a chronic brain disease that manifests as a psychotic disorder characterized by the rapid progressive decline of cognitive and emotional capabilities, is commonly diagnosed in late adolescence or young adulthood, and was first conceptualized in the latter part of the nineteenth century (Adityanjee et al., 1999). Over the years, the term dementia praecox has been replaced by the more global designation of Schizophrenia spectrum disorders. However, after more than a century of studying schizophrenia, the cause of the illness and its

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treatment remain unclear (Insel, 2010). Schizophrenia and bipolar disorder (BPD) are severe disabling conditions with substantial overlap in terms of risk factors and neurocognitive deficits, vocational, social and functional impairments (American Psychiatric Association, 2013; Bora, 2009; Bora et al., 2010, 2015). Due to their chronicity, early age of onset and lack of efficient treatment, many individuals with Schizophrenia, Schizoaffective disorders and BPD remain chronically ill and require daily living support (Dixon et al., 2018). Institutional care for people with chronic psychotic disorders (CPD) such as Schizophrenia, Schizoaffective disorders and BPD has increasingly shifted to communities (Dixon et al., 2018) where family members are asked to assume the role of intensive and long-term caregivers for their close relative with CPD. Family members, especially parents, must frequently take on the role of providing emotional and financial assistance as well as housing for their suffering relative. Along with a profound sense of obligation and compassion (Karp, 2000), these roles constitute an extremely multifaceted burden and have known adverse effects on caregivers' wellbeing (Angermeyer et al., 2003; Chen & Lukens, 2011). Family members whose close relative has a CPD commonly endure a form of ongoing stress that is associated with depression and an impaired quality of life (Chen & Lukens, 2011; Godress, et al., 2005; Jones, 2004; Mendenhall &



Mount, 2011). Both are linked to an elevated risk of relapse for the relative in their care (Mcfarlane, 2016).

Grief Experiences in the Context of Chronic Psychotic Disorders

Family members of individuals with CPD report experiencing grief over the loss of the person they once knew given the radical changes in this individual's emotional and cognitive abilities. They also report experiencing grief over their multiple losses in many facets of life including disruption to family routine and relationships, and lost opportunities to live a "normal life" such as engaging in fulfilling and meaningful social roles (Chen & Lukens, 2011; Godress et al., 2005; Jones, 2004). Grief in such circumstances has been associated with depression, reduced well-being and increased ill-being, and interferes with the caregivers' ability to support their close relative affected by CPD and seek support for themselves (Chen & Lukens, 2011; Godress et al., 2005; Landon et al., 2016; Sawrikar & Muir, 2018).

Complicated Grief and Risk Factors for Complicated Grief

Bereavement following the loss of a loved one is a stressful life event. It is also a universal event and a natural process (Worden, 2018). Normal grief, also referred to uncomplicated grief, encompasses a wide range of reactions that are common after a loss. Typical reactions to this loss include separation distress, yearning for the lost person, sadness, anger, disbelief and physical sensations such as lack of energy (Worden, 2018). Although most bereaved individuals eventually adjust to their loss without sustained difficulties and achieve a balance between what is lost and what life entails following it, a considerable proportion of bereaved individuals suffer from intense unrelenting grief (Lundorff et al., 2017). According to the American Psychiatric Association (2013), persistent complex bereavement disorder, or complicated grief (CG), is a form of prolonged, unrelenting grief subsequent to the decease of a loved one. One out of ten bereaved adults is at risk of developing CG (Lundorff et al., 2017), whose core symptoms include a persistent yearning for the deceased, preoccupation with the deceased and the circumstances surrounding the death, accompanied by intense emotional pain, and excessive avoidance of reminders of the loss (Prigerson et al., 2009; Shear, 2015). CG is associated with a range of negative mental and physical health outcomes including depression, anxiety, PTSD and suicidality, in addition to social, academic and work impairment and medical morbidity (Simon et al., 2007).

Studies have identified several key factors that are likely to increase the risk of CG. These include sudden loss and loss in traumatic circumstances, close kinship to the deceased, unavailability of social and emotional support, high distress at the time of death, as well as negative selfappraisals, self-blame, the comorbidity of PTSD symptoms (Nakajima et al., 2012), and depression (Nielsen et al., 2017). Attachment orientations; i.e., the patterns of relational expectations, emotions and behaviors that result from the internalization of the individual's history of attachment experiences (Bowlby, 1973), have been shown to be related to psychological adjustment after loss (Lobb et al., 2010; Maccallum & Bryant, 2018; Shear & Shair, 2005; Shear et al., 2007). The innate psychological attachment behavioral system motivates individuals to seek proximity to significant others in times of need, separation and loss as a way of protecting themselves from threats and alleviating distress. In response to the loss of a significant other, individuals with a secure attachment orientation generally perceive relationships as a source of comfort and protection and strive to maintain bonds and engage in proximity-seeking behaviors. In contrast, individuals with an anxious attachment orientation may experience exacerbated yearning and separation anxiety (Maccallum & Bryant, 2018). Individuals with an avoidant attachment orientation are more inclined to avoid emotions and to withdraw from supporting social relationships (Boelen & Klugkist, 2011). Several studies have shown that attachment orientation may be a risk factor for CG (Lobb et al., 2010). Most studies have pointed to attachment anxiety as a precursor of CG (Maccallum & Bryant, 2018; Shear & Shair, 2005). Nevertheless, attachment avoidance may contribute to grief complications such as depression (Boelen, & Klugkist, 2011; Maccallum & Bryant, 2018).

Complicated Grief and Risk Factors for Complicated Grief in Response to Chronic Psychotic Disorders

Although family members of patients suffering from CPD experience grief in response to the loss of their loved one as he or she was prior to the onset of illness, diagnostic manuals do not cover this loss, which often remains in the form of disenfranchised grief that is not openly acknowledged or socially supported (Doka, 1999). Disenfranchised grief may lead to isolation, leaving individuals with no opportunity to receive counseling for grief (Godress et al., 2005; Jones, 2004). Close family relatives of individuals suffering from CPD experience a myriad of factors that may interfere with their grief over their loss and increase their risk of developing CG. Here, we posited that this kind of loss may have five key risk pathways to CG: (1) Lack of social and professional support may hinder the processing of the pain of grief, and feelings of anxiety, shame, guilt, anger and loneliness and may increase the risk for depression and CG (Worden, 2018): the loss is disenfranchised, since the loved one is alive but different. Family members are still attached to the mental representation of the person they once knew, but the



presence of the changed personality is a constant reminder of the loss (Jones, 2004). In addition, studies have shown that despite their key role in supporting and caring for their loved one, caregivers experience inadequate collaboration, lack of professional support and evidence-based practices (Dixon et al., 2010; Landon, 2016; McAuliffe et al., 2014). (2) Prolonged traumatic bereavement and severe distress interfere with coming to terms with the loss (Worden, 2018). The course and prognosis of the illness is often uncertain, thus resulting in emotional limbo. Hope and grief oscillate between improvement and relapse time and again, thrusting caregivers back to bereavement (Karp & Tanarugsachock, 2000). (3) Self-blame interferes with coming to terms with the loss (Worden, 2018). Here, confusion and misunderstanding about the illness commonly hinder family members' ability to accommodate and modify their expectations without feelings of betrayal and guilt with respect to their relative suffering from the illness (Jones, 2004). These feelings can undermine other activities and relationships, and may make people reluctant to seek the support they need (Jones, 2004; Karp & Tanarugsachock, 2000; Landon et al., 2016). (4) The loss affects individuals' sense of self, may challenge beliefs about oneself and may shatter self-identity. Thus caregivers are subjected to loss of self-identity when facing the stressful role changes associated with care burden (e.g., siblings and children become caregivers, aging parents are challenged with unfamiliar legal and medical responsibilities) (Karp & Tanarugsachock, 2000). (5) Living in the same household with care recipient. Previous studies have suggested that living in the same household as a relative with CPD was related to greater distress, helplessness, depression and social isolation (Goossens et al., 2008; Landon et al., 2016; McAuliffe et al., 2014; Zauszniewski & Bekhet, 2014). Thus overall, social, emotional, and cognitive factors including lack of formal and social support, PTSD, depression and cognitive distortions such as self-blame are common experiences reported by caregivers (Landon et al., 2016; Mendenhall & Mount, 2011; Pejlert, 2001; Sawrikar & Muir, 2018), and may contribute to grief complication (Jones, 2004). Lack of validation and legitimation of grieve may leave individuals feeling that their distress, confusion, depression, guilt, anger, helplessness and fear are inherent to their grief. Therefore, grief remains latent, unvalidated and unresolved (Chen & Lukens, 2011; Godress et al., 2005; Jones, 2004).

To better understand the nature and unfolding of CG, the present study aimed to evaluate the prevalence of CG in relation to a family member suffering from CPD and determine the risk factors for CG. Given the research findings reviewed above on risk factors for CG (Lobb et al., 2010; Nakajima et al., 2012) and the literature on traumatic losses and the impact of caregiving for a loved one with CPD, it was predicted that (1) high rates of CG would be found in

first degree relatives of individuals with CPD, (2) CG would be associated with higher levels of depression and PTSD and reduced health, (3) self-blame, loss of self-identity, attachment anxiety orientation, PTSD, depression, living with the care-recipient, and support seeking would make unique contribution to CG in this cohort.

Method

Participants and Procedure

This study was conducted over a 14-month period from January 2018 to March 2019, and was reviewed and approved by the authors' Institutional Review Board committee and the Mental Health Services of the Ministry of Health. First-degree relatives (parents, siblings, adult children and spouses) of patients with CPD were recruited through social media posts and through organizations that provide support services for sufferers of mental illness and/or for their families. Those who consented and completed the full battery of questionnaires (n = 78) met once with the researcher (LR) and were informed of their right to confidentiality and the purpose of the study, which was defined as aiming to identify the psychological and physical reactions of family members of individuals with CPD. Written informed consent was obtained from all participants. The informed consent form included an offer to talk with the principal investigator and included contacts of professional resources oriented toward family members of individuals with CPD. All participants were first asked whether they had a close relative (sibling, child, parent or spouse) diagnosed with CPD that had caused significant changes in this relative's personality and/or behavior. Next, demographic information including contextual variables that may be associated with CG including kinship to the relative with CPD, length of time associated with the loss, and the level of impairment caused by the illness, as well as related symptoms in response to the illness of their close relative was assessed as described in the Measures below. The demographic characteristics of the sample are summarized in Table 1.

Measures

CG was assessed on the 19-item Inventory of Complicated Grief (ICG; Prigerson et al., 1995), a well-validated measure (α =0.92–0.94; test–retest reliability=0.80) (Prigerson et al., 1995). The ICG measures the severity of cognitive, emotional and behavioral symptoms of CG. Statements are rated on a Likert scale ranging from 0 to 4. An ICG score of 25 or greater is considered a positive screen for CG. An ICG score \geq 25 has been associated with greater impairment in various spheres of life and has been used in several studies



Table 1 Demographics

Variable	
Kinship to individual with CPD, % (n)	
Parent	50.0 (39)
Adult sibling	39.7 (31)
Adult child	9.0 (7
Couple	1.3(1)
Gender, % (n)	
Male	25.6 (20)
Female	74.4 (58)
Age (years), mean (SD)	51.12 (15.01)
Income level, % (n)	
Above average	38.2 (29)
Average	47.4 (36)
Below average	14.5 (11)
Type of employment, % (n)	
Full time job	51.9 (40)
Part time job	32.5 (25)
Retired	10.4 (8)
Unemployed	5.2 (4)
Marital status, % (n)	
Married	53.8 (42)
Divorced	26.9 (21)
Single	15.4 (12)
Separated	2.6 (2)
Single parent	1.3(1)
Education (years), mean (SD)	15.34 (3.07)
Level of education, % (n)	
Elementary School	1.3(1)
High school	15.4 (12)
Technical school	12.8 (10)
Academic education	70.5 (55)

CPD chronic psychotic disorder

as a positive screen for CG (Delaney et al., 2017; Mitchell et al., 2005; Shear et al., 2001; Simon et al., 2005; Sung et al., 2011). The ICG was slightly adapted by the first author to measure CG in response to those whose significant other had been diagnosed with CPD. The items were tailored to measure the family member's current severity of grieving in relation to the illness and the distress associated with the loss, as evidenced by functional debilitating preoccupation with the lost healthy person, inability to accept the loss, as well as ongoing distress, bitterness and anger (e.g., I miss him/her as he was before the illness, I cannot accept the fact that he/she will never be the same again, I feel it is unjust that I will keep on living my life while he/she cannot, I have been feeling lonely since he/she got ill). The adjusted ICG in the present study demonstrated excellent reliability (Cronbach's $\alpha = 0.914$).

Depression was measured on the Beck Depression Inventory (BDI; Beck et al., 1961), a 21-item self-report scale that measures severity of depression. This inventory is a continuous measure suitable for both psychiatric and normative populations. The scale covers affective, cognitive, motivational, and physiological symptoms of depression. Each category consists of 4 statements rated on a 0–3 point Likert scale. Scores of 0-9 are considered normal, 10-18 indicate mild depression, 19-25 moderate depression, and above 26 severe depression. In a large meta-analysis (Beck et al., 1988) the BDI exhibited high validity and reliability scores and high internal consistency ($\alpha = 0.81-0.86$). In the present study, the BDI had excellent reliability (Cronbach's α = 0.90). A BDI score of 12 or greater is considered a positive screen for depressive disorder in community surveys (Lasa et al., 2000).

Attachment orientation was assessed using the general form of the Experiences in Close Relationships scale (ECR; Brennan et al., 1998), as adapted for Hebrew speakers (Mikulincer & Florian, 2000), which taps attachment style in relationships generally rather than focusing on specific relationships. It contains 36 items with 18 items tapping attachment anxiety and 18 items tapping attachment avoidance. Items are rated from 1 (strongly disagree) to 7 (strongly agree). High scores indicate more anxiety or avoidance. In the present study, the participants were asked to think about their close relationships in general, without focusing on a specific person, and to rate the extent to which each item described themselves in these relationships. The Cronbach's alphas are reported to range from 0.86 to 0.89 for avoidant attachment and from 0.91 to 0.94 for anxious attachment (Stern et al., 2018). Studies have demonstrated the measure's high construct and predictive validity (Mikulincer & Shaver, 2007). In the present study, the ECR attachment avoidant scale had a Cronbach's of $\alpha = 0.88$, and an α of 0.94 for attachment anxiety.

PTSD was measured on the posttraumatic stress scale (PDS-5; Foa et al., 2016), a self-report measure of PTSD based on the DSM-5 criteria. Participants are asked to rate the frequency and distress associated with each PTSD symptom on a scale ranging from 0 (not at all) to 4 (6 or more times a week/severe). PTSD severity is determined by totaling the 20 PDS-5 symptom ratings (items 1–20). Scores range from 0 to 80. The PDS-5 has been found to have excellent internal consistency ($\alpha = 0.94$). The test–retest reliability for PDS was reported to be excellent at 0.88. In the present study the internal consistency for the PDS-5 was also excellent (Cronbach's $\alpha = 0.94$). A score of 28 is used as a cutoff point for possible diagnosis of PTSD, with scores between 0 and 27 indicating no diagnosis, and 28–80 indicating probable diagnosis (Foa et al., 2016). Psychological and physical health were measured on the SF-36 health measure (Ware



& Sherbourne, 1992) adapted for Hebrew speakers (Lewin-Epstein et al., 1998).

The SF-36 is a self-report, Likert scale examining a patient's perceived health, and has been shown to be a valid measure of general health in population studies (Ware, 2000). It consists of eight subscales on mental health (MH), general health (GH), role limitation due to emotional and physical problems (RP), social functioning (SF), physical functioning (PF), vitality (V) and bodily pain (BP). Two summary measures, the Physical Component Summary (PH) and the mental health component scale (MH) aggregate the scales. Scores on the SF-36 MH scale range from 0 to 100 (with high scores indicating better mental health). Both internal consistency and construct validity were reported to be strong, and the reliabilities range from 0.7 to 0.9 (Ware, 2000). In the present study the internal consistency for the SF-36 was excellent (Cronbach's α = 0.94).

Statistical Analyses

A logistic regression analysis was applied to the data to model binary CG as a dependent variable. Group differences on the categorical variables were compared using chi-square tests. Group differences on the independent continuous variables were analyzed with two-tailed t tests. All analyses were performed using SPSS version 23.0 for Windows (IBM Corp., Armonk, NY, USA, 2011).

Results

Characteristics of Relatives Suffering from CPD

Table 2 summarizes the characteristics of the recipients of care suffering from CPD. As shown in Table 2, a large proportion (76.50%) of the care recipients had moderate to severe functional impairments and had failed to achieve milestones such as independent living. In the vast majority of the participants (66.2%), the onset of CPD had occurred more than six years prior to the study, and in a large proportion (33.8%) the onset of CPD had occurred more than one year previously. More than 30% of the care recipients were living with their family members and the remainder were living in sheltered housing, or independently (3.9%) and needed long-term assistance. Most of the close family members (71.42%) received professional help in the aftermath of the illness. Of these, 26.53% participated in both individual and group treatment, 34.69% participated in individual treatment and 10.20% in group treatment. Only 27.65% thought the treatment was effective, 42.55% reported it was partially effective, and 2.12% thought treatment was not effective. About a third of the participants (28.57%) did not receive any treatment.



Table 2 Characteristics of care recipients with CPD

Variable	
Diagnosis, % (n)	'
Schizophrenia	80.8 (63)
Schizoaffective disorder	11.5 (9)
Bipolar disorder	5.1 (4)
Psychotic disorder	2.6(2)
Caregiver, % (n)	
Participant is the primary caregiver	43.6 (34)
Participant is not the primary caregiver	56.4 (44)
Residence, % (n)	
Lives with the primary caregiver	30.8 (24)
Does not live with the caregiver	69.2 (54)
Level of impairment caused by the illness, $\%$ (n)	
Completely dysfunction	2.0(1)
Very low functioning	19.6 (10)
Low functioning	19.6 (10)
Moderate functioning	35.3 (18)
Good functioning	19.6 (10)
Very good functioning	3.9 (2)
Prevalence onset of CPD, % (n)	
One year	11.6 (9)
Two years	7.7 (6)
Three years	1.2(1)
Four years	2.5 (2)
Five years	3.8 (3)
Six years	6.4 (5)
More than 6 years	66.2 (51)

CPD chronic psychotic disorder

Loss and Complicated Grief

Responses on the ICG indicated that most of the participants (87.18%) reported experiencing the loss of the person they once knew in response to major changes in personality and behavior caused by the illness. Only 12.82% of the participants reported not experiencing a sense of loss of their close relative with CPD. Acceptance over the years (n=2), lack of acquaintance with the sibling/spouse before the illness (n=5), and hope for improvement as a result of their close relative's improved functioning (n=3) were the most prevalent reasons given. Symptoms of CG were frequent. Most of the participants (80.76%) reported painful memories of the person they once knew as well as longing for him or her to come back (84.61%) and difficulty accepting the loss (74.35%). A large proportion of the family members had feelings of self-blame (60.25%), bitterness (66.66%), anger (60.25%), and difficulty trusting others since the loss (43.58%). Many (64.10%) experienced the loss as a shocking ongoing trauma. There were loss-triggered changes in roles as well as self and world schemas in 48.71% of the relatives

(e.g., the siblings became caregivers of their brother or sister). Close to half of the participants (39.74%) screened positive on the ICG using the cutoff of \geq 25. More than one third (36.23%) of the participants had higher than threshold rates of BDI (\geq 12) and 13.23% had higher than threshold rates of PDS-5.

Characteristics of Participants with Complicated Grief

Table 3 presents the mean levels of depression, PTSD, attachment style and health status of the participants with or without probable CG. As hypothesized, participants with CG had higher than threshold rates of PDS-5 (29.16% vs. 4.54%) $(\chi^2(1) = 8.19, p = 0.004)$. Participants with current probable CG had higher than threshold rates of BDI (80.00% vs. 11.36%) ($\chi^2(1) = 32.50$, p < 0.001), Self-blame and probable CG were significantly correlated ($\chi^2(1) = 20.17, p < 0.00$). Participants with current probable CG had lower scores on the SF-36 measure (M = 57.60, SD = 19.75) than participants with no CG (M=76.60, SD=14.35) (t(71)=4.75,p < 0.001). CG rates were significantly higher in participants who experienced their loss as a shocking trauma $(\chi^2(1) = 11.12, p = 0.004)$. No significant differences were found between groups in levels of kinship ($\chi^2(3) = 5.44$, p = 0.14), or in terms of level of impairment caused by the illness ($\chi^2(5) = 3.52$, p = 0.62). Significantly higher proportions of probable CG were found in primary caregivers and in caregivers who lived with the care recipient ($\chi^2(1) = 4.38$, p = 0.036; $\chi^2(1) = 7.49$, p = 0.006, respectively). Participants with higher than threshold rates of CG had higher scores on the ECR attachment anxiety (M=63.25, SD=26.01) subscale as compared to participants with lower than threshold rates of CG (M=49.07, SD=22.76) (t(64)=2.31, p=0.024). In addition, participants with higher than threshold rates of CG had higher scores on the ECR attachment avoidance (M=68.85, SD=20.34) subscale as compared to participants with lower than threshold rates of CG (M=52.26, SD=17.41) (t(64)=3.50, p=0.001). No significant differences were found between groups in terms of the amount of time elapsed since the first episode of the illness $(\chi^2(29)=31.58, p=0.33)$ and support seeking $(\chi^2(3)=1.74, p=0.43)$.

Risk Factors for Complicated Grief

A logistic regression (forward stepwise) was conducted to predict categorical CG. PTSD (PDS-5≥28), depression (BDI \geq 12), self-blame, living with the care recipient, changes in role identity, support seeking, time elapsed since the loss, and attachment orientation (anxious, avoidant) were entered into the model. We first ran the univariate analyses to determine which variables were highly correlated with CG (see Table 3). Next, we ran a multivariate model including those variables which met a preset cutoff for significance to run a multivariate model (p < 0.05); namely, probable PTSD and depression, self-blame and living with the care recipient. This model was significant ($\chi^2(3) = 40.77$, p < 0.000) and accounted for 61.10% of the variance. Self-blame and depression significantly predicted CG. Living with the care recipient approached significance (p = 0.060). The full logistic regression predicting categorical CG is presented in

Table 3 Characteristics of participants with CG and without CG

	Group		Analyses	
	CG (ICG score≥25)	No CG (ICG score < 25)	Statistic value	p
BDI, mean (SD)	17.74 (9.58)	6.45 (4.79)	t(68) = 6.57	0.000***
BDI \geq 12, % (n)	80.0 (20)	11.4 (5)	$\chi^2(1) = 32.50$	0.000***
PDS-5, mean (SD)	19.73 (15.61)	8.25 (10.01)	t(66) = 3.69	0.000***
PDS-5 ≥ 28, % (n)	29.2 (7)	4.5 (2)	$\chi^2(1) = 8.19$	0.004**
SF-36, mean (SD)	57.60 (19.75)	76.60 (14.35)	t(71) = 4.75	0.000***
ECR—avoidance items, mean (SD)	68.85 (20.35)	52.26 (17.41)	t(64) = 3.50	0.001**
			$\chi^2(43) = 42.95$	0.473
ECR—anxiety items, mean (SD)	63.25 (26.01)	49.07 (22.76)	t(64) = 2.31	0.024*
			$\chi^2(52) = 55.91$	0.330
Living with the care, % (n) recipient	62.50 (15)	37.50 (9)	$\chi^2(1) = 7.49$	0.000***
Self-blame	59.57 (28)	40.42 (19)	$\chi^2(1) = 20.17$	0.000***
Role identity	50.00 (24)	50.00 (24)	$\chi^2(4) = 8.64$	0.071

BDI beck depression inventory, PDS-5 posttraumatic stress scale, SF-36 36-item short form survey, ECR experiences in close relationships *p < 0.05



^{**}p < 0.01

^{***}p < 0.001

Table 4. These findings suggest that participants who scored positively on self-blame and participants who had probable depression were more likely to meet the criteria for CG.

Discussion

This study extends previous research on grief and the caregiving burden by examining up-to-date CG criteria (American Psychiatric Association, 2013; Shear, 2015) and the potential contributing factors of CG in first-degree relatives of individuals suffering from chronic psychotic disorders. Despite the time elapsed since the onset of CPD, the vast majority of participants had not adapted to the loss. The majority of the participants manifested symptoms of CG characterized by marked distress, yearning for the person they once knew, self-blame, bitterness and anger. About forty percent of the participants screened positive on the ICG. Complicated grief is known to be an under-recognized public health problem and is associated with depression and functional impairment (Shear et al., 2016; Sung et al., 2011). The current results thus strongly suggest that CG may be a significant problem for caregivers, and point to the importance of screening caregivers for CG. Enfranchising grief in such circumstances may help caregivers to grieve for their loss and assist them in talking about their loss and exploring their feelings, conflicts and thoughts about the loss. It is important to note that the prevalence of CG in this cohort was comparable to reported rates (12.5–78%) of CG among those bereaved by violent death (Nakajima et al., 2012). Nakajima et al. suggested that the factors affecting the high prevalence of CG in the wake of violent death include suddenness and lack of readiness, social isolation and negative self-appraisals, self-blame, the comorbidity of PTSD symptoms, as well as various social stressors such as stigma and low satisfaction with formal authorities. When social stigma exists, social support for the grievers may be less than sufficient (Doka, 1999). The results of the current study show that self-blame and depression were unique contributors to CG in the context of CPD.

Grieving is a social phenomenon, and individuals with more support, and individuals that are satisfied with it,

Table 4 Logistic regression predicting categorical CG

	В	SE	Wald	P	Exp(b)
Self-blame	2.13	0.99	4.57	0.03	8.42
Living with care recipient	1.7	0.9	3.53	0.06	5.49
Depression	3.06	0.81	14.24	0	21.45

Feelings of self-blame = 1, living in the same household as a relative with CPD = 1, depression = 1



manifest lower depression, and may allow themselves to move forward toward a better adaptation to the loss (Worden, 2018). Maladaptive self-blaming thoughts are associated with grief complications and comorbid depression (Shear, 2015). As was previously suggested, self-blame may be associated with caregivers' ability to accommodate to the loss, process their pain and guilt, and allow themselves to engage in leisure and social activities and supporting relationships (Jones, 2004; Karp & Tanarugsachock, 2000; Landon et al., 2016). Contrary to previous studies on predictors of CG in the aftermath of death (Lobb et al., 2010; Maccallum & Bryant, 2018; Wijngaards-de Meij et al., 2007), attachment orientation failed to explain the variance in CG when depression and self-blame were included. Repeated, prolonged exposure to experiences related to caregiving to a close person with CPD, helplessness and loneliness that may lead to depression may thus produce maladaptive changes in beliefs with respect to trust and intimacy, self-efficacy as well as self-esteem, resulting in oscillations in attachment orientations (Mikulincer et al., 2011). Future longitudinal studies could shed more light on the trajectories of attachment and CG in caregivers of close relatives with CPD. About a third of the participants did not receive treatment and of those participants who did, less than a third thought that the treatment was effective. It is worth noting that the Schizophrenia Patient Outcomes Research Team (PORT: Dixon et al., 2010) indicated that relatively few patients with Schizophrenia and their families have access to psychosocial evidence-based treatments. This is unfortunate since psychosocial evidence-based treatments can significantly improve family relationships and reduce caregivers' distress levels (McFarlane, 2016).

This study has several limitations. The first has to do with the use of self-reports since self-report scales may be subject to response biases. There was no clinician-based confirmatory assessment of the CG diagnosis, PTSD or depression. Thus, it was impossible to determine whether CG would have been considered the primary clinical problem. Future studies should confirm the assessment of CG using a clinician's formal diagnostic assessment for CG. Second, we did not collect data concerning the trajectory of CG at different time points starting from the initial diagnosis of CPD. Longitudinal studies could thus shed more light on factors that facilitate or hinder adjustment to such challenging circumstances, and may inform clinicians of the best practices when attending to those suffering from loss. The relatively small sample size could have inflated CG rates. Thus, future studies should assess CG rates in larger and more representative samples. Finally, the generalizability of the results may be limited, given the fact that the participants self-selected; most were recruited through organizations that provide support for families, and as such may have differed on key measurements of adjustment to loss. An additional limitation is

the cross-sectional nature of the data which precludes causal explanations about the relationship between predictors and outcomes. Despite these limitations, the findings provide information about CG and risk factors for CG in first-degree relatives of individuals with CPD. The results shed light on the importance of attending to caregivers who suffer from CG or whose grieving process can be complicated by risk factors such as self-blame and depression. Hence, the findings may contribute to better detection, prevention and specific adaptations of evidence-based practices to facilitate caregivers' natural course of grief, better clarify their roles, reduce self-blame and help prevent physical and mental health problems as well as its negative impact on various areas of life.

Implications for Treatment

Although grief persists years after close relatives are informed that their loved one is suffering from CPD, to date, diagnostic manuals fail to include the component of grief related to the loss that follows the onset of CPD. To the best of our knowledge, no specific interventions for CG in response to psychological loss have been reported. Several grief therapies models have been suggested as potential approaches to help accommodate to the loss and thereby, prevent complications of grief (Lafond, 2009; Miller, 1996). These models encompass interventions such as acknowledging and legitimizing grief, expressing emotions, and overcoming various impediments to readjustment after this loss. However, the current results draw attention to those close family relatives who have debilitating grief symptoms and may need therapy for complicated grief. One example of a specific evidence-based practice is a short-term intervention for CG (Shear, 2015) that has been demonstrated to be efficacious in several trials (Shear et al., 2014, 2016). The key components of this intervention include improving mood and developing satisfying social relationships, processing and confronting emotions, facilitating the ability to enjoy affectionate reminiscences of the lost one, and reengagement in activities. Adapting to the loss of a loved one entails accepting the finality and changed relationship with the person who died. However, unlike CG after the death of a loved one, CG in the context of psychological loss provides no closure because the loved one is both gone but also present (Boss, 2010). In addition, the loved one's behavior can sometimes be unexpected and frightening. The neurocognitive deficits in patients with CPD affect their communication abilities (American Psychiatric Association, 2013; Bora et al., 2009, 2010). Poor communication between the caregiver and the care recipient can lead to conflicts, isolation or depression in one or both of these individuals. The experiences of hospitalization, sometimes forcibly, can be traumatic and may contribute and increase feelings of self-blame and betrayal. These distinctive mediators may increase the risk of complications and enduring feelings of anger, shame and guilt, and should be considered part of the adaptations and modifications needed when providing counselling for caregivers struggling with CG associated with psychological loss. These include enfranchising the loss and understanding CPD (e.g., labeling and acknowledging the experience of grief and providing psycho-education about CG in response to a family member suffering from CPD as well as psycho-education about CPD to provide a better understanding of the illness (e.g., warning signs of relapse, course, positive and negative effects of antipsychotic medications). Interventions aimed at building an enduring connection with the loved one (Worden, 2018), in a way that can enable the caregiver to go on with his or her life include modifying expectations and improving communication, as well as accommodating to the loss by reviewing positive memories and connecting to the healthy aspects of their loved one with CPD. These interventions may facilitate the healing process of CG in response to psychological loss. Future work should concentrate on assessing the efficacy of complicated grief therapy for relatives of persons with CPD through large-scale controlled studies.

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Declarations

Conflict of interest The authors declare that they have no conflict of interest.

Ethical Approval This study was approved the Interdisciplinary Center Herzliya Institutional Review Board and the Mental Health Services of the Israel Ministry of Health Institutional Review Board.

Informed Consent Informed consent was obtained from all participants in the study.

References

Adityanjee, Y. A., Theodoridis, D., & Vieweg, V. R. (1999). Dementia praecox to Schizophrenia: The first 100 years. *Psychiatry and Clinical Neurosciences*, 53, 437–448. https://doi.org/10.1046/j.1440-1819.1999.00584.x



- American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). American Psychiatric Association.
- Angermeyer, M. C., Schulze, B., & Dietrich, S. (2003). Courtesy stigma-A focus group study of relatives of schizophrenia patients. Social Psychiatry and Psychiatric Epidemiology, 38, 593–602. https://doi.org/10.1007/s00127-003-0680-x
- Beck, A. T., Steer, R. A., & Carbin, M. G. (1988). Psychometric properties of the beck depression inventory: Twenty-five years of evaluation. *Clinical Psychology Review*, 8, 77–100. https://doi.org/10.1016/0272-7358(88)90050-5
- Beck, A. T., Ward, C. H., Mendelson, M., Mock, J., & Erbaugh, J. (1961). An inventory for measuring depression. *Archives of General Psychiatry*, 4, 561–571.
- Boelen, P. A., & Klugkist, I. (2011). Cognitive behavioral variables mediate the associations of neuroticism and attachment insecurity with prolonged grief disorder severity. *Anxiety, Stress, & Coping*, 24, 291–307. https://doi.org/10.1080/10615806.2010.527335
- Bora, E., & Pantelis, C. (2015). Meta-analysis of cognitive impairment in first-episode bipolar disorder: Comparison with first-episode schizophrenia and healthy controls. *Schizophrenia Bulletin*, 41, 1095–1104. https://doi.org/10.1093/schbul/sbu198
- Bora, E., Yucel, M., & Pantelis, C. (2009). Cognitive functioning in schizophrenia, schizoaffective disorder and affective psychoses: Meta-analytic study. *The British Journal of Psychiatry*, 195, 475–482. https://doi.org/10.1192/bjp.bp.108.055731
- Bora, E., Yucel, M., & Pantelis, C. (2010). Cognitive impairment in schizophrenia and affective psychoses. Implications for DSM-V criteria and beyond. *Schizophrenia Bulletin*, 36, 36–42. https:// doi.org/10.1093/schbul/sbp094
- Boss, P. (2010). The trauma and complicated grief of ambiguous loss. *Pastoral Psychology*, *59*, 137–145. https://doi.org/10.1007/s11089-009-0264-0
- Bowlby, J. (1973). Attachment and loss. (Vol. 2). Basic Books.
- Brennan, K. A., Clark, C. L., & Shaver, P. R. (1998). Self-report measurement of adult attachment: An integrative overview. In J. A. Simpson & W. S. Rholes (Eds.), *Attachment theory and close relationships* (pp. 46–76). Guilford Press.
- Chen, W. Y., & Lukens, E. P. (2011). Wellbeing, depressive symptoms, and burden among parent and sibling caregivers of persons with severe and persistent mental illness. *Social Work in Mental Health*, 9, 397–416. https://doi.org/10.1080/15332985.2011. 575712
- Delaney, E. M., Holloway, K. J., Miletich, D. M., Webb-Murphy, J. A., & Lanouette, N. M. (2017). Screening for complicated grief in a military mental health clinic. *Military Medicine*, 182, e1751–e1756. https://doi.org/10.7205/MILMED-D-17-00003
- Dixon, L. B., Dickerson, F., Bellack, A. S., Bennett, M., Dickinson, D., Goldberg, R. W., Lehman, A., Tenhula, W. N., Calmes, C., Pasillas, R. M., Peer, J., & Kreyenbuhl, J. (2010). The 2009 schizophrenia PORT psychosocial treatment recommendations and summary statements. *Schizophrenia Bulletin*, 36, 48–70. https://doi.org/10.1093/schbul/sbp115
- Dixon, L. B., Goldman, H. H., Srihari, V. H., & Kane, J. M. (2018). Transforming the treatment of schizophrenia in the United States: The RAISE initiative. *Annual Review of Clinical Psychology*, 14, 237–258. https://doi.org/10.1146/annurev-clinpsy-050817-084934
- Doka, K. J. (1999). Disenfranchised grief. *Bereavement Care*, 18, 37–39. https://doi.org/10.1080/02682629908657467
- Foa, E. B., McLean, C. P., Zang, Y., Zhong, J., Powers, M. B., Kauffman, B. Y., Rauch, S., Porter, K., & Knowles, K. (2016). Psychometric properties of the Posttraumatic diagnostic scale for DSM–5 (PDS–5). *Psychological Assessment*, 28, 1166–1171. https://doi.org/10.1037/pas0000258
- Godress, J., Ozgul, S., Owen, C., & Foley-Evans, L. (2005). Grief experiences of parents whose children suffer from mental

- illness. *Australian and New Zealand Journal of Psychiatry*, *39*, 88–94. https://doi.org/10.1080/j.1440-1614.2005.01518.x
- Goossens, P. J., Van Wijngaarden, B., Knoppert-van Der Klein, E. A., & Van Achterberg, T. (2008). Family caregiving in bipolar disorder: Caregiver consequences, caregiver coping styles, and caregiver distress. *The International Journal of Social Psychiatry*, 54, 303–316. https://doi.org/10.1177/0020764008090284
- Insel, T. R. (2010). Rethinking schizophrenia. *Nature*, 468, 187–193. https://doi.org/10.1038/nature09552
- Jones, D. (2004). Families and serious mental illness: Working with loss and ambivalence. *British Journal of Social Work, 34*, 961–979. https://doi.org/10.1093/bjsw/bch123
- Karp, D. A., & Tanarugsachock, V. (2000). Mental illness, caregiving, and emotion management. *Qualitative Health Research*, 10, 6–25. https://doi.org/10.1177/104973200129118219
- Lafond, V. (2009). Coming to terms with mental illness in the family—working constructively through its grief. In F. Lobban & C. Barrowclough (Eds.), A casebook of family interventions for psychosis (pp. 167–184). Wiley.
- Landon, J., Pike, B., Diesfeld, K., & Sheperd, D. (2016). The experiences of parents providing support to adult children with Schizophrenia. *International Journal of Mental Health Addiction*, 14, 385–399. https://doi.org/10.1007/s11469-016-9643-9
- Lasa, L., Ayuso-Mateos, J. L., Vazquez-Barquero, J. L., Diez-Manrique, F. J., & Dowrick, C. F. (2000). The use of the beck depression inventory to screen for depression in the general population: A preliminary analysis. *Journal of Affective Disorders*, 57, 261–265. https://doi.org/10.1016/S0165-0327(99) 00088-9
- Lewin-Epstein, N., Sagiv-Schifter, T., Shabtai, E. L., & Shmueli, A. (1998). Validation of the 36-item short-form health survey (Hebrew version) in the adult population of Israel. *Medical Care*, 36, 1361–1370. https://doi.org/10.1097/00005650-19980 9000-00008
- Lobb, E. A., Kristjanson, L. J., Aoun, S. M., Monterosso, L., Halkett, G. K. B., & Davies, A. (2010). Predictors of complicated grief: A systematic review of empirical studies. *Death Studies*, *34*, 673–698. https://doi.org/10.1080/07481187.2010.496686
- Lundorff, M., Holmgren, H., Zachariae, R., Farver-Vestergaard, I., & O'Connor, M. (2017). Prevalence of prolonged grief disorder in adult bereavement: A systematic review and meta-analysis. *Journal of Affective Disorders*, 212, 138–149. https://doi.org/10.1016/j.jad.2017.01.030
- Maccallum, F., & Bryant, R. A. (2018). Prolonged grief and attachment security: A latent class analysis. *Psychiatry Research*, 268, 297–302. https://doi.org/10.1016/j.psychres.2018.07.038
- McAuliffe, R., O'Connor, L., & Meagher, D. (2014). Parents' experience of living with and caring for an adult son or daughter with Schizophrenia at home in Ireland: A qualitative study. *Journal of Psychiatric and Mental Health Nursing*, 21, 145–153. https://doi.org/10.1111/jpm.12065
- McFarlane, W. R. (2016). Family interventions for schizophrenia and the psychoses: A review. *Family Process*, 55, 460–482. https://doi.org/10.1111/famp.12235
- Meij, W.-d, Stroebe, M., Schut, H., Stroebe, W., den Bout, J. V., van der Heijden, P., & Dijkstra, I. (2007). Neuroticism and attachment insecurity as predictors of bereavement outcome. *Journal* of Research in Personality, 41, 498–505. https://doi.org/10.1016/j. jrp.2006.06.001
- Mendenhall, A. N., & Mount, K. (2011). Parents of children with mental illness: Exploring the caregiver experience and caregiverfocused interventions. *Families in Society*, 92, 183–190. https:// doi.org/10.1606/1044-3894.4097
- Mikulincer, M., Ein-Dor, T., Solomon, Z., & Shaver, P. R. (2011). Trajectories of attachment insecurities over a 17-year period: A latent growth curve analysis of the impact of war captivity and



- posttraumatic stress disorder. *Journal of Social and Clinical Psychology*, 30, 960–984. https://doi.org/10.1521/jscp.2011.30.9.960
- Mikulincer, M., & Florian, V. (2000). Exploring individual differences in reactions to mortality salience: Does attachment style regulate terror management mechanisms? *Journal of Personality and Social Psychology*, 79, 260–273. https://doi.org/10.1037/0022-3514.79.2.260
- Mikulincer, M., & Shaver, P. R. (2007). Attachment patterns in adult-hood: Structure, dynamics, and change. Guilford Press.
- Miller, F. E. (1996). Grief therapy for relatives of persons with serious mental illness. *Psychiatric Services*, 47, 633–637. https://doi.org/ 10.1176/ps.47.6.633
- Mitchell, A. M., Kim, Y., Prigerson, H. G., & Mortimer, M. K. (2005). Complicated grief and suicidal ideation in adult survivors of suicide. Suicide & Life-Threatening Behavior, 35, 498–506. https://doi.org/10.1521/suli.2005.35.5.498
- Nakajima, S., Ito, M., Shirai, A., & Konishi, T. (2012). Complicated grief in those bereaved by violent death: the effects of post-traumatic stress disorder on complicated grief. *Dialogues in Clinical Neuroscience*, 14, 210–214. https://doi.org/10.31887/DCNS.2012. 14.2/snakajima
- Nielsen, M. K., Neergaard, M. A., Jensen, A. B., Vedsted, P., Bro, F., & Guldin, M. B. (2017). Predictors of complicated grief and depression in bereaved caregivers: A nationwide prospective cohort study. *Journal of Pain and Symptom Management*, 53, 540–550. https://doi.org/10.1016/j.jpainsymman.2016.09.013
- Pejlert, A. (2001). Being a parent of an adult son or daughter with severe mental illness receiving professional care: Parents' narratives. *Health and Social Care in the Community*, 9, 194–204. https://doi.org/10.1046/j.0966-0410.2001.00301.x
- Prigerson, H. G., Horowitz, M. J., Jacobs, S. C., Parkes, C. M., Aslan, M., Goodkin, K., Raphael, B., Marwit, S. J., Wortman, C., Neimeyer, R. A., Bonanno, G., & Maciejewski, P. K. (2009).
 Prolonged grief disorder: Psychometric validation of criteria proposed for DSM-V and ICD-11. *PLoS Medicine*, 6, e1000121. https://doi.org/10.1371/journal.pmed.1000121
- Prigerson, H. G., Maciejewski, P. K., Reynolds, C. F., III., Bierhals, A. J., Newsom, J. T., Fasiczka, A., Frank, E., Doman, J., & Miller, M. (1995). Inventory of complicated grief: A scale to measure maladaptive symptoms of loss. *Psychiatry Research*, 59, 65–79. https://doi.org/10.1016/0165-1781(95)02757-2
- Sawrikar, P., & Muir, K. (2018). Toward a family-oriented treatment approach for consumers and carers of mental illness. *International Journal of Mental Health*, 47, 158–183. https://doi.org/10.1080/ 00207411.2017.1367448
- Shear, K., Monk, T., Houck, P., Melhem, N., Frank, E., Raynold, C., & Sillowash, R. (2007). An attachment-based model of complicated grief including the role of avoidance. *European Archives of Psychiatry and Clinical Neuroscience*, 257, 453–461. https://doi. org/10.1007/s00406-007-0745-z
- Shear, K., & Shair, H. (2005). Attachment, loss, and complicated grief. Developmental Psychobiology, 47, 253–267. https://doi.org/10. 1002/dev.20091

- Shear, M. K. (2015). Clinical practice. Complicated grief. *The New England Journal of Medicine*, 372, 153–160. https://doi.org/10.1056/NEJMcp1315618
- Shear, M. K., Frank, E., Foa, E., Cherry, C., Reynolds, C. F., 3rd., Vander Bilt, J., & Masters, S. (2001). Traumatic grief treatment: A pilot study. *The American Journal of Psychiatry*, 158, 1506–1508. https://doi.org/10.1176/appi.ajp.158.9.1506
- Shear, M. K., Reynolds, C. F., 3rd., Simon, N. M., Zisook, S., Wang, Y., Mauro, C., Duan, N., Lebowitz, B., & Skritskaya, N. (2016). Optimizing treatment of Complicated grief: A randomized clinical trial. *JAMA Psychiatry*, 73, 685–694. https://doi.org/10.1001/jamapsychiatry.2016.0892
- Shear, M. K., Wang, Y., Skritskaya, N., Duan, N., Mauro, C., & Ghesquiere, A. (2014). Treatment of complicated grief in elderly persons: A randomized clinical trial. *JAMA Psychiatry*, 71, 1287–1295. https://doi.org/10.1001/jamapsychiatry.2014.1242
- Simon, N. M., Pollack, M. H., Fischmann, D., Perlman, C. A., Muriel, A. C., Moore, C. W., Nierenberg, A. A., & Shear, M. K. (2005). Complicated grief and its correlates in patients with bipolar disorder. *The Journal of Clinical Psychiatry*, 66, 1105–1110. https://doi.org/10.4088/jcp.v66n0903
- Simon, N. M., Shear, K. M., Thompson, E. H., Zalta, A. K., Perlman, C., Reynolds, C. F., Frank, E., Melhem, N. M., & Silowash, R. (2007). The prevalence and correlates of psychiatric comorbidity in individuals with Complicated grief. *Comprehensive Psychiatry*, 48, 395–399. https://doi.org/10.1016/j.comppsych.2007.05.002
- Stern, J. A., Fraley, R. C., Jones, J. D., Gross, J. T., Shaver, P. R., & Cassidy, J. (2018). Developmental processes across the first two years of parenthood: Stability and change in adult attachment style. *Developmental Psychology*, 54, 975–988. https://doi.org/10.1037/dev0000481
- Sung, S. C., Dryman, M. T., Marks, E., Shear, M. K., Ghesquiere, A., Fava, M., & Simon, N. M. (2011). Complicated grief among individuals with major depression: Prevalence, comorbidity, and associated features. *Journal of Affective Disorders*, 134, 453–458. https://doi.org/10.1016/j.jad.2011.05.017
- Ware, J. E., Jr. (2000). SF-36 health survey update. *Spine*, *25*, 3130–3139. https://doi.org/10.1097/00007632-200012150-00008
- Ware, J. E., & Sherbourne, C. D. (1992). The MOS 36-item short-form health survey (SF-36). Conceptual framework and item selection. *Medical Care*, 30, 473–483.
- Worden, J. W. (2018). Grief counselling and grief therapy: A handbook for the mental health practitioner (5th ed.). . Springer.
- Zauszniewski, J. A., & Bekhet, A. K. (2014). Factors associated with the emotional distress of women family members of adults with serious mental illness. *Archives of Psychiatric Nursing*, 28, 102– 107. https://doi.org/10.1016/j.apnu.2013.11.003

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