



SHORT COMMUNICATION



## Peritraumatic distress predicts prolonged grief disorder symptom severity after the death of a parent in children and adolescents

Alexis Revet<sup>a,b,c</sup>, Agnès Suc<sup>d</sup>, Françoise Auriol<sup>e</sup>, A. A. A. Manik J. Djelantik<sup>f,g</sup>, Jean-Philippe Raynaud<sup>a,b</sup> and Eric Bui<sup>h,i</sup>

<sup>a</sup>Service Universitaire de Psychiatrie de l'Enfant et de l'Adolescent, CHU de Toulouse, Toulouse, France; <sup>b</sup>CERPOP, Université de Toulouse, Inserm, UPS, Toulouse, France; <sup>c</sup>CIC 1436, Team PEPSS « Pharmacologie En Population cohorteS et biobanqueS », Toulouse University Hospital, France; <sup>d</sup>Centre de Ressource Douleur Soins Palliatifs Pédiatriques, CHU de Toulouse, Toulouse, France; <sup>e</sup>Unité de recherche clinique pédiatrique module plurithématique pédiatrique CIC Toulouse 1436, Hôpital des Enfants, CHU de Toulouse, Toulouse, France; <sup>f</sup>Department of Psychiatry, University Medical Centre Utrecht, Utrecht, The Netherlands; <sup>g</sup>Department Youth – KOOS, Altrecht GGZ, Utrecht, The Netherlands; <sup>h</sup>Department of Psychiatry, Massachusetts General Hospital, Boston, MA, USA; <sup>i</sup>Department of Psychiatry, University of Caen Normandy & Caen University Hospital, Caen, France

### ABSTRACT

**Background:** In 2015 nearly 140 million children and adolescents under 18 had experienced the death of one or both parents. Parental death is often considered the most traumatic event that a child can experience in their lifetime. While parental loss may lead to the development of prolonged grief disorder (PGD), little is known about risk factors for such negative mental health outcome in children.

**Objective:** The present study aims to examine peritraumatic reactions as predictors of PGD in children who lost a parent.

**Method:** Thirty-four children (M age = 10.9, SD = 3.2, 67.6% females) who lost a parent (time since death = 4.6 months, SD = 2.3) were assessed for peritraumatic distress and peritraumatic dissociation experienced at the time of the loss, and for PGD symptom severity at three timepoints post-loss (<6 months; 6–12 months; >12 months).

**Results:** PGD score was correlated with peritraumatic distress (.61;  $p < .01$ ) but not with peritraumatic dissociation (.24;  $p = .3$ ). Results from the mixed-model regression analysis identified peritraumatic distress as the only significant predictor of PGD symptom severity ( $B = 1.58$ ,  $SE = .31$ ;  $p < .0001$ ), with no statistically significant effect of peritraumatic dissociation ( $B = -.43$ ,  $SE = .36$ ;  $p = .2$ ), or time ( $B = -3.84$ ,  $SE = 2.99$ ;  $p = .2$ ).

**Conclusion:** Our results suggest that peritraumatic distress might be useful to identify children at risk for developing PGD, and in need of further support. The development of early preventive strategies to prevent PGD in parentally bereaved children who experienced high peritraumatic distress is warranted.

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### PALABRAS CLAVE

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### 关键词

创伤性精神痛苦; 创伤性解离; 复杂性哀伤; 儿童和青少年

### HIGHLIGHTS

- Our results suggest that peritraumatic distress might be useful to identify children at risk for developing prolonged grief disorder, and in need of further support.

## La Angustia Peritraumática predice la Severidad de Síntomas de Duelo Complicado en Niños y Adolescentes tras la muerte de un Padre o Madre

**Antecedentes:** En el 2015 cerca de 140 millones de niños y adolescentes menores a 18 años experimentaron la muerte de uno o ambos padres. La muerte parental es considerada a menudo como el evento más traumático que un niño puede experimentar en su vida. Mientras que la pérdida parental puede conducir al desarrollo de Duelo Prolongado (PGD), poco se sabe respecto a los factores de riesgo para dicha consecuencia negativa en la salud mental de los niños.

**Objetivo:** El presente estudio apunta a examinar reacciones peritraumáticas como predictores de PGD en niños que han perdido un padre o madre.

**Método:** Treinta y cuatro niños (Edad promedio = 10.9, DE = 3.2, 67,6% mujeres) quienes perdieron su padre o madre (tiempo desde la muerte = 4.6 meses, DE=2.3) fueron evaluados en relación a angustia peritraumática y disociación peritraumática experimentados al momento de la pérdida, y en relación a severidad sintomática de PGD en tres momentos diferentes tras la pérdida (< 6 meses; 6-12 meses; > 12 meses).

**Resultados:** El puntaje de PGD se correlacionó con angustia peritraumática (0.61;  $p < .01$ ) pero no con disociación peritraumática (0.24;  $p = .3$ ). Resultados del análisis de regresión de modelo mixto identificaron a la angustia peritraumática como el único predictor significativo para severidad sintomática de PGD ( $B = 1.58$ ;  $p < .0001$ ), sin efecto estadísticamente significativo de disociación peritraumática ( $B = -.43$ ;  $p = .2$ ) o según el tiempo ( $B = -3.84$ ;  $p = .2$ ).

**Conclusión:** Nuestros resultados sugieren que la angustia peritraumática puede ser útil para identificar niños en riesgo de desarrollar PGD, y en necesidad de mayor apoyo. Está justificado

el desarrollo de estrategias preventivas tempranas para prevenir PGD en niños en duelo por pérdida parental que experimentan angustia peritraumática severa.

### 儿童和青少年父母死亡后创伤性精神痛苦预测复杂性哀伤症状严重程度

**背景:** 2015年, 将近1.4亿未满18岁的儿童和青少年经历过单亲或双亲的死亡。父母死亡通常被认为是一个孩子一生中经历最大的创伤性事件。虽然父母丧失可能导致发展出延长哀伤障碍 (PGD), 但对儿童不良心理健康结果的风险因素知之甚少。

**目的:** 本研究旨在考查失去父母的儿童的创伤反应作为PGD的预测指标。

**方法:** 评估了34名丧失父母 (自死亡起时间= 4.6个月, SD = 2.3) 的儿童 (平均年龄= 10.9, SD = 3.2, 女性占67.6%) 丧失时的创伤性精神痛苦和创伤性解离, 丧失以及丧失后三个时间点 (<6个月; 6-12个月;> 12个月) 的PGD症状严重程度。

**结果:** PGD评分与创伤性精神痛苦相关 (.61;  $p < .01$ ), 但与创伤性解离无关 (.24;  $p = .3$ )。混合模型回归分析的结果表明, 创伤性精神痛苦是PGD症状严重程度的唯一显著预测因子 ( $B = 1.58$ ;  $p < .0001$ ), 而创伤前分离 ( $B = -.43$ ;  $p = .2$ ) 或时间 ( $B = -3.84$ ;  $p = .2$ ) 统计上不显著。

**结论:** 我们的结果表明, 创伤性精神痛苦可能有助于确定有发生PGD风险的儿童, 需要进一步的支持。有必要开发出早期预防策略来预防经历高创伤性精神痛苦的丧失父母的儿童的PGD。

## 1. Introduction

The United Nations International Children's Emergency Fund (UNICEF) estimated that worldwide in 2015 nearly 140 million children and adolescents under age 18 had experienced the death of one or both parents (United Nations International Children's Emergency Fund, 2017). The death of a parent is consistently considered as one of the most traumatic life events that a child or adolescent can experience (Alisic, Schoot, Ginkel, & Kleber, 2008; Yamamoto et al., 1996). The loss of one parent is a risk factor for the development of numerous psychiatric (Melhem, Porta, & Shamseddeen et al., 2011; Nickerson, Bryant, Aderka, Hinton, & Hofmann, 2013) and somatic problems (Luecken, Kraft, Appelhans, & Enders, 2009), with an increased mortality (Li et al., 2014) risk including long-term risk of suicide (Guldin et al., 2015). In addition, this type of major stressor may also have a broader negative socio-economic impact (from disruption of schooling to lower income or greater risk of unemployment in adults, for instance) (Brent, Melhem, & Masten et al., 2012).

While the intensity of the grief and life disruption after the loss usually subsides in the following months, recent meta-analyses in adults revealed a pooled prevalence of prolonged grief disorder (PGD) of 9.8% (95% CI 6.8–14.0) following natural loss (Lundorff, Holmgren, Zachariae, Farver-Vestergaard, & O'Connor, 2017) and of 49% (95% CI 33.6–65.4) following unnatural loss (Djelantik, Smid, Mroz, Kleber, & Boelen, 2020). Although more research is needed, the prevalence among children has been suggested to also range between 7% and 10% (Harrison & Harrington, 2001; Melhem et al., 2011). PGD has received increasing attention in the past couple of decades – including among children and adolescents – with its recent inclusion in the 11<sup>th</sup> edition of the WHO International Classification of Diseases, and its impending addition to the text revision of the DSM-5

(Boelen & Lenferink, 2020; Boelen, Lenferink, & Smid, 2019; Boelen, Spuij, & Lenferink, 2019; Lenferink, Boelen, Smid, & Paap, 2019). While PGD, post-traumatic stress disorder (PTSD) and major depressive disorder are often comorbid in bereaved adults (Djelantik, Robinaugh, Kleber, Smid, & Boelen, 2020) as in bereaved children (Spuij et al., 2012), recent studies (Geronazzo-Alman et al., 2019; Spuij et al., 2012) provided arguments in favour of the distinctiveness of the cluster of symptoms of prolonged grief, depression, and PTSD in bereaved children and adolescents.

To date, little is known about the risk factors for PGD among bereaved children and adolescents. Recent data suggest that losing someone to a chronic illness, female gender, feeling one could have done something to prevent the death, exposition to interpersonal conflicts, personal history of depression, and family history of anxiety disorders might be risk factors for PGD (Kaplow, Howell, & Layne, 2014; Melhem et al., 2004); however, little is known about psychological reactions that occur during or immediately after the death (Revet et al., 2020). Yet, these reactions are important as they can help to quickly identify those at risk and may lead to the development of early preventive interventions. Two types of peritraumatic reactions have been described: peritraumatic distress that indexes reactions such as fear, helplessness and horror experienced during or immediately after trauma exposure, and peritraumatic dissociation that refers to alterations in the experience of time, place and persons. In adults, peritraumatic distress has been consistently reported to be a correlate and prospective predictor of psychopathological symptoms, including PGD symptoms, after exposure to a major life event (such as the death of a loved one) (Bui et al., 2013; Hargrave, Leathem, & Long, 2012; Williams, Hardt, Henschel, & Jobe-Shields, 2020). Similarly to PTSD (Ehlers & Clark, 2000), PGD

might develop when initial (peritraumatic) distress disrupts information processing of the traumatic event or leads to poor integration of information about the death (Boelen, Hout, & Bout, 2006; Shear et al., 2007). Peritraumatic reactions have been successfully used to identify children at risk for developing PTSD symptoms after trauma (Bui et al., 2010); however, no studies have examined their ability to predict the development of PGD in the context of bereavement nor children.

The present study aimed to examine the relationship between peritraumatic reactions and PGD symptoms among parentally bereaved children and adolescents aged 6 to 17, in the first year post loss. We hypothesized that PGD symptoms will be positively associated with increased peritraumatic distress and peritraumatic dissociation.

## 2. Methods

### 2.1. Study design and ethics

This longitudinal prospective cohort study was conducted between 04/2016 and 04/2020 in Toulouse, France. The study was conducted in line with the Declaration of Helsinki and approved by the Institutional Review Board of Toulouse University Hospital and the Regional Ethics Committee for Medical and Health Research of South West France (approval number: 2015-A01132-47). Participants gave their assent, and their parents gave written informed consent.

### 2.2. Participants and procedures

Thirty-four parentally bereaved children and adolescents from 24 families, aged 6 to 17 (mean age = 10.9 years, standard deviation, SD = 3.2; 67.6% female; 70.8% lost a father; mean time since death = 4.6 months, SD = 2.3), who consulted with their surviving parent at Toulouse University Paediatrics Hospital to participate to support groups for bereaved children and adolescents, were enrolled in this study. Cancer was the main cause of parental death ( $n = 10$ ; 41.7%), followed by suicide ( $n = 6$ ; 25.0%), accident ( $n = 5$ ; 20.8%), cardiac arrest ( $n = 2$ ; 8.3%) and genetic disease ( $n = 1$ ; 4.2%). All families had a stable housing and all children or adolescents lived with their surviving parent (Table 1). Main exclusion criteria were the death of both parents, a serious medical disease, and an ongoing judiciary process related to the death.

They were then invited to complete an initial questionnaire at enrolment ( $n = 34$ ), and as well as PGD symptom severity (primary outcome) and other outcomes, at different timepoints post-loss (<6 months,

**Table 1.** Demographic and death-related characteristics of  $N = 34$  children who lost a parent, and of their surviving parent.

|   | Values            |
|---|-------------------|
| <i>Children and adolescents</i>                 |                   |
| Age, mean $\pm$ sd                              | 10.9 ( $\pm$ 3.2) |
| Female, n (%)                                   | 23 (67.6)         |
| Child or adolescent's level of education, n (%) | 23 (67.6)         |
| Primary school                                  | 11 (32.4)         |
| Secondary school                                |                   |
| <i>Parents</i>                                  |                   |
| Age, mean $\pm$ sd                              | 43.4 ( $\pm$ 7.6) |
| Surviving parent                                |                   |
| Female, n (%)                                   | 7 (29.2)          |
| Deceased parents                                |                   |
| Cause of parent's death, n (%)                  | 10 (41.7)         |
| Cancer  | 6 (25.0)          |
| Suicide   | 5 (20.8)          |
| Accident  | 2 (8.3)           |
| Cardiac arrest                                  | 1 (4.2)           |
| Genetic disease                                 |                   |
| Family life, n (%)                              | 24 (100)          |
| Stable housing                                  | 24 (100)          |
| Common life the child or adolescent             |                   |

Sd, standard deviation.

mean time since loss = 111 days [SD = 40],  $n = 30$ ; 6–12 months, mean time since loss = 235 days [SD = 46],  $n = 19$ ; >12 months post loss, mean time since loss = 390 days [SD = 19],  $n = 12$ ). The first questionnaire also included a retrospectively assessment of peritraumatic distress and dissociation experienced at the time of the loss. Because of slower recruitment than anticipated, and the relatively small final sample, we elected to only examine the primary outcome in order to minimize type 1 errors.

### 2.3. Measures

The child version of the 13-item self-report Peritraumatic Distress Inventory (PDI-C) was used to assess distress at the time of the parent's death (Bui et al., 2011). Higher total scores indicated increased distress (range, 0–52). The child version of the 10-item self-report Peritraumatic Dissociative Experiences Questionnaire (PDEQ-C) was used to assess dissociation at the time of the parent's death (Bui et al., 2011). Higher total score indicated increased dissociation (range, 10–50).

The primary outcome was PGD symptom severity measured by the 28-item Inventory Complicated Grief–Revised for Children (ICG-RC), higher total score indicating increased PGD symptoms' frequency (range, 28–140). The ICG-RC is a modified version of the adult Inventory of Complicated Grief (ICG) (Prigerson et al., 1995), used to assess grief phenomenology in children and adolescents younger than 18 years (Melhem et al., 2011; Melhem, Porta, Walker Payne, & Brent, 2013). Its psychometric properties have been examined previously and the ICG-RC showed high internal consistency (Melhem, Moritz, & Walker et al., 2007), measured by

a Cronbach's  $\alpha$  of 0.95, and evidence of convergent and discriminant validity in relation to self-reported measures of depression, anxiety, PTSD, and functional disability.

#### 2.4. Statistical analyses

The association between peritraumatic reactions and PGD symptom severity was investigated through a mixed-model regression analysis. All statistical analyses were conducted using Stata 14.2 (Statacorp, College Station, TX). The mixed-model regression ('xtmixed' command) considering time as a repeated measure, and children to be nested within their families, was conducted to allow for the inclusion of random effects at both the children and families levels. Our modelling made use of all available data. Marginal mean scores indicating the mean expected score in each condition across time points were estimated using the 'margins' command. We used a two-sided significance level of .05 with a 95% confidence interval for all analyses.

### 3. Results

The mean retrospectively assessed baseline PDI-C, PDEQ-C scores were 18.2 (SD = 9.6) and 21.1 (SD = 8.2), respectively. Mean ICG-RC scores at the three timepoints evolved as follows: <6 months, mean ICG-RC = 41.9, SD = 19.0; 6–12 months, mean ICG-RC = 35.2, SD = 18.6; >12 months, mean ICG-RC = 39.6, SD = 24.7. However, mean PGD symptom severity was not significantly associated with continuous age ( $r = 0.13$ ,  $p = .5$ ), or age group (children aged 6–10-year-olds vs. adolescents aged 11–17-year-olds) ( $t(32) = 0.35$ ,  $p = .7$ ). The last ICG score was clearly associated with PDI-C ( $r = .61$ ,  $p = .0004$ ) but the association with PDEQ-C was weaker ( $r = .36$ ,  $p = .04$ ).

Results from the mixed-model regression analysis identified PDI-C score as the only significant predictor of PGD symptom severity ( $B = 1.58$ ,  $SE = .31$ ;  $p < .0001$ ), with no statistically significant effect of PDEQ-C ( $B = -.43$ ,  $SE = .36$ ;  $p = .2$ ) or time ( $B = -3.84$ ;  $SE = 2.99$ ;  $p = .2$ ). These results remained consistent in another mixed-model regression retaining only the clearly prospective PGD outcomes. Once again, the only significant predictor of PGD symptom severity was PDI-C score ( $B = 1.29$ ,  $SE = .65$ ;  $p < .05$ ), with no statistically significant effect of PDEQ-C ( $B = .01$ ,  $SE = .69$ ;  $p = .99$ ). The ICG-RC marginal mean scores estimated by the model at the three timepoints were: <6 months, 45.0 ( $SE = 4.8$ ); 6–12 months, 41.2 ( $SE = 2.5$ ); >12 months, 37.4 ( $SE = 2.6$ ). In addition, two follow-up regression analyses focusing on single time points (one focusing on a timepoint combining all first PGD assessments ( $n = 34$ ), and the second focusing on the second time interval (6–12 months,  $n = 19$ )) showed consistent results.

### 4. Discussion

As hypothesized, we found a positive relationship between peritraumatic distress and PGD symptoms. Our PDI-C and PDEQ-C scores at baseline were quite similar to those found by Bui et al. in 2010 in a study conducted in 103 French-speaking children aged 8 to 15 presenting to an emergency department after a road traffic accident (Bui et al., 2010). However, Melhem et al. identified an ICG-RC score of 68 or higher as having the highest sensitivity and specificity in differentiating cases of PGD from noncases at 9 months (Melhem et al., 2013), which could mean our PGD score may have been sub-clinical. While the role of peritraumatic reactions in PGD has been discussed and minimized by some (2000), our results are in line with those from a study in adults which reported that peritraumatic distress may be a key mechanism in the development of both PTSD and PGD (Hargrave et al., 2012).

Interestingly, peritraumatic distress was also found to be a better predictor of acute PTSD symptoms in school-aged children than peritraumatic dissociation (Bui et al., 2010), and our results are similar for PGD symptoms in children and adolescents. The absence of significant effect between peritraumatic dissociation and the PTSD measures is in line with results from previous studies. An interesting model developed by Thomas et al. in adults (Thomas, Saumier, & Brunet, 2012) proposed that peritraumatic distress could predict PTSD's development, while peritraumatic dissociation would predict the lack of integration of the traumatic memory, leading to chronic PTSD. The fact that this model is not verified in paediatric population might be an indicator that other factors could be more prominent in the recovery process from PTSD and PGD symptoms. In particular, parents' response to the trauma and the quality of their parenting and relationships with their child or adolescent could be the main predictive factors.

PGD and PTSD are both common psychological reactions to bereavement with historical and phenomenological overlap, which poses a challenge for clinicians and researchers. Thus, it is quite common that patients' experiences are not clearly categorized as trauma vs. grief. In addition, their emotional reactions do not always fall cleanly into the categories of PGD vs. PTSD (Frumkin & Robinaugh, 2018). With this regard, Shear et al. proposed an attachment-based biobehavioral model of PGD (Shear & Shair, 2005), in which avoidance plays a key role and PGD is envisioned as a stress response syndrome that results from failure to integrate information about death of an attachment figure and/or to effectively re-engage the exploratory system in a world without the deceased. According to this model, children and adolescents with insecure attachment, who present

important peritraumatic reactions and a high level of behavioural and cognitive avoidance in the first months following the loss would be at high risk for PGD.

This study has several limitations. First, the small and predominantly female (67.6%) sample and longitudinal data that were limited to only three timepoints, both of which precluded examination of symptoms trajectories, adjustment for potential confounding variables such as parental or child's prior history of trauma, age of the children or the loss of father or mother, and the examination of the moderating effects of parental symptoms. We nevertheless found no significant association between age and PGD symptom severity. Second, we assessed the peritraumatic reactions retrospectively, and administered the PDI-C and PDEQ-C a few months after the loss. Because of this, hypothetically it could be that the measure rather corresponds to a measure of distress and dissociation in the relatively early post-loss recovery period (i.e. 2–6 months after loss) instead of peri-bereavement. However, in the questionnaire it was really clearly mentioned that the participants needed to recall the symptoms of distress at the time of the bereavement instead of the present experienced distress. Furthermore, we cannot rule out the fact that children with more severe grief reactions may remember more peritraumatic distress, which could have had an impact on the direction of our association between peritraumatic distress and PGD. Finally, the American Psychiatric Association recently approved the inclusion of PGD as a new mental disorder in the DSM-5-TR, using updated diagnostic criteria (Boelen, Eisma, Smid, & Lenferink, 2020). In this regard, it is important to note that the ICG, which is primarily composed of questions related to separation distress, may not capture all the symptoms of PGD and therefore may not accurately reflect this new diagnosis. More recent scales such as the Prolonged Grief questionnaire-13 (PG-13), introduced in the process of developing PGD diagnostic criteria proposed for inclusion in the DSM-5 and ICD-11 (Prigerson et al., 2009), and its revised version, the PG-13-R (Prigerson, Boelen, & Xu et al., 2021), are certainly more adapted to the new DSM-5-TR criteria for PGD.

In conclusion, our results suggest that peritraumatic distress might be useful to identify children in need of further support. Future research should examine the role of peritraumatic distress in an information-processing model of PGD. The development of early preventive strategies to prevent PGD in parentally bereaved children who experienced high peritraumatic distress is warranted.

### Data availability statement

The dataset supporting the findings of this study is available from the corresponding author, AR, upon reasonable request. The data are not publicly available due to legal and ethical restrictions.

### Disclosure statement

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### Contributions of authors statement

AR produced the first draft of the manuscript. EB designed the study and wrote the protocol. EB oversaw the research, drafted portions and substantively edited all drafts of the manuscript, and conducted the analyses. JPR provided additional study oversight. AR, AS and FA contributed substantively to the collection and processing of data. AS, MD, FA and JPR contributed to the interpretation of the data and critically revised the manuscript for important intellectual content. All authors contributed to and have approved the final manuscript. AR and EB take responsibility for the integrity of the data and the accuracy of the data analysis.

### Ethics statement

The study was conducted in line with the Declaration of Helsinki and approved by the Institutional Review Board of Toulouse University Hospital and the Regional Ethics Committee for Medical and Health Research of South West France (approval number: 2015-A01132-47). Participants gave their assent, and their parents gave written informed consent.

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### ORCID

Alexis Revet  <http://orcid.org/0000-0002-8051-1657>

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