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Pre-loss personal factors and prolonged grief disorder in bereaved mothers

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Original Article

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Abstract

Background. Identifying characteristics of individuals at greatest risk for prolonged grief disorder (PGD) can improve its detection and elucidate the etiology of the disorder. The Safe Passage Study, a study of women at high risk for sudden infant death syndrome (SIDS), prospectively examined the psychosocial functioning of women while monitoring their healthy pregnancies. Mothers whose infants died of SIDS were followed in bereavement.

Methods. Pre-loss data were collected from 12 000 pregnant mothers and analyzed for their associations with grief symptoms and PGD in 50 mothers whose infants died from SIDS, from 2 to 48 months after their infant's death, focusing on pre-loss risk factors of anxiety, depression, alcohol use, maternal age, the presence of other living children in the home, and previous child loss.

Results. The presence of any four risk factors significantly predicted PGD for 24 months post-loss ($p < 0.003$); 2–3 risk factors predicted PGD for 12 months ($p = 0.02$). PGD rates increased in the second post-loss year, converging in all groups to approximately 40% by 3 years. Pre-loss depressive symptoms were significantly associated with PGD. Higher alcohol intake and older maternal age were consistently positively associated with PGD. Predicted risk scores showed good discrimination between PGD and no PGD 6–24 months after loss (*C*-statistic = 0.83).

Conclusions. A combination of personal risk factors predicted PGD in 2 years of bereavement. There is a convergence of risk groups to high rates at 2–3 years, marked by increased PGD rates in mothers at low risk. The risk factors showed different effects on PGD.

Background

In the normal course of life, people experience the death of others who have been meaningful and significant to them. Most people reconcile themselves to their loss and, after a difficult period of acute grief, accommodate and regain normal thinking, emotions, and behaviors (Bowlby, 1980; Bonanno *et al*., 2004). A small yet significant proportion of bereaved individuals have more difficulty with this adaptation, continuing with severe and lasting symptoms that exceed social norms and cause impairment in daily functioning. This constellation of intrusive symptoms constitutes prolonged grief disorder (PGD) (Prigerson *et al*., 2008). PGD involves 'separation distress', characterized by significant emotional suffering and yearning, in addition to cognitive, emotional, and behavioral symptoms, more than 6 months after a significant loss (Prigerson *et al*., 2009). PGD presents as a coherent set of symptoms distinct from bereavement-related depression (Prigerson *et al*., 1995), major depressive disorder (Prigerson *et al*., 1996*b*), anxiety (Prigerson *et al*., 1996*b*), or post-traumatic stress disorder (Boelen *et al*., 2010). It has been documented worldwide (Schaal *et al*., 2010; Field *et al*., 2014; Heeke *et al*., 2015; Xiu *et al*., 2016; Coelho *et al*., 2017; Pohlkamp *et al*., 2018) and is associated with diminished health and quality of life (Boelen and Prigerson, 2007; Prigerson *et al*., 2009). Increasingly accepted as a clinical diagnosis (Maercker *et al*., 2013), PGD is included in ICD-11 (World Health Organization, 2018).

Understanding risk for more severe grief responses and PGD can help allocate scarce resources to those who most need them and maximize the efficacy of those efforts (Parkes and Weiss, 1983; Schut *et al*., 2001; Mancini *et al*., 2012; Roberts *et al*., 2017). Considerable research has examined risk factors for more extreme grief under various pathologic descriptors consisting of traumatic grief, complicated grief, prolonged grief, and persistent complex

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bereavement disorder. Several informative analytic frameworks have been proposed to organize the identified elements of the loss experience that inform adaptation (Bonanno and Kaltman, 1999; Stroebe *et al.*, 2006), including the categorization of elements as either situational factors related to the particular circumstances of a death, personal factors in the bereaved, or interpersonal factors related to the availability and nature of support for the bereaved from others (Stroebe and Schut, 2001). Some have concluded that the influence of the circumstances of death is minimal in comparison with personal features of the bereaved (Cleiren *et al.*, 1994). This research investigated personal factors and PGD.

Personal factors identified by research to enhance the risk for PGD have included demographic variables, for example, sex (Goodenough *et al.*, 2004), poverty (Goldsmith *et al.*, 2008; McCarthy *et al.*, 2010), education (Ostfeld *et al.*, 1993), or age (Wijngaards-de Meij *et al.*, 2008), and variables reflecting the kinship relationship of the bereaved to the deceased, for example, motherhood (Dyregrov and Dyregrov, 1999; Michon *et al.*, 2003), the death of a child (Kersting *et al.*, 2011), or the centrality of loss (Boelen, 2009; Papa *et al.*, 2014). In previous research, we investigated young mothers whose seemingly healthy infants died suddenly and unexpectedly from the sudden infant death syndrome (SIDS) because they concentrate many personal risk factors and, as an experimental model, have a relatively uniform exposure to their child's death without variations in illness or treatment experience. In a cross-sectional study of 356 mothers bereaved when their infants died from SIDS, we found an overall prevalence of PGD in 50% of mothers 6–48 months after their infant's death, with a rate of PGD of 57.1% at 1 year and 41.3% in the third year post-loss (Goldstein *et al.*, 2018). In comparison, a recent meta-analysis appraising 14 studies of non-psychiatric adult populations exposed to non-violent bereavement observed a pooled prevalence of PGD in 9.8% 1 year following loss (Lundorff *et al.*, 2017). The generalizability of the SIDS-related findings was supported by nearly identical prevalence rates of PGD symptoms between two studied populations of bereaved mothers living in highly disparate social circumstances, from women living in informal settlements outside of Cape Town, South Africa to middle class US populations, with the exception that mothers with more limited resources reported less persistent feelings of shock that their infant had died.

Prospective research on grief

Prospective research to identify features in those who will go on to be most strongly affected by grief have yielded complicated findings. Studies examining grief in caregivers of dementia patients had conflicting conclusions about associations with pre-loss measures of depression (Givens *et al.*, 2011; Nielsen *et al.*, 2017) while also reporting that grief symptoms were greater before the patient's death than after it (Nielsen *et al.*, 2017). Others similarly reported greater depression-related symptoms prior to death than during bereavement (Schulz *et al.*, 2003). Such prospective efforts to understand how personal factors contribute to grief may be susceptible to confounding by functional and perceptual changes that accompany the experiences of caregivers as patients near death (Lotterman *et al.*, 2014), including recall bias in pre-loss estimates and changed internal assessments of how caregivers remember themselves before their present circumstances (Toedter *et al.*, 1990). The likelihood of death following a life-limiting diagnosis or after arriving at a certain stage of life may lead to anticipatory grief and a context in which death is regarded as being in the normal course of events (Nielsen *et al.*, 2016). It is difficult to assemble cohorts that provide prospective data uninfluenced by the presence of serious illness or the likelihood of possible loss, reducing our ability to genuinely examine personal features in caregivers before the illness experience.

There are some notable exceptions where pre-loss data were collected prior to the diagnosis of illness. Bonanno *et al.* examined pre-loss variables of depressive symptoms, qualities of the marriage, coping resources, and world view in 205 bereaved life partners, using data collected when husbands living independently became 65 years old (Bonanno *et al.*, 2002). The authors identified basic bereavement patterns, while also finding that subjects with prolonged grief and chronic depression symptoms were symptomatically similar and that both were notably depressed pre-loss. Other prospective research examined pregnancy loss, finding that women with higher symptom scores for depression, anxiety, somatization and obsessive–compulsive behavior or who did not have another child at the time of the loss experienced greater grief intensity (Janssen *et al.*, 1997). Toedter *et al.* (2001) similarly found correlations between pre-loss mental health and prolonged grief at 2 years of follow-up. Neugebauer *et al.* found that miscarriage triggered depression in 54% of mothers who had previously experienced clinical depression (Neugebauer *et al.*, 1997). Although pregnancy loss research may raise questions about generalizability to deaths in life partners, livebirths, or children, this research makes rare contributions to understanding the influence of personal factors on bereavement, especially given the limited impact from intervention fidelity, anticipatory grief, or death occurring in 'the normal course of things'.

Current study

SIDS is the leading cause of post-neonatal mortality in high-income countries. A seemingly well infant is discovered dead after a sleep period. While SIDS rates have declined since the 1990s, it causes more mortality than cancer or heart disease in children aged 0–19 years in the USA (Centers for Disease Control and Prevention, 2017). The circumstances and risk factors for SIDS are many of the risk factors for prolonged grief disorder, including the situational factors of relationship with the deceased (Cleiren *et al.*, 1994), young age of the deceased (Zetumer *et al.*, 2015), lack of preparation for the death (Barry *et al.*, 2002), and suddenness of the death (Dyregrov *et al.*, 2003).

In this study, we analyzed pre-loss data collected during pregnancy in mothers whose infants ultimately died from SIDS, to identify characteristics of risk and its outcomes for PGD. We hypothesized that vulnerability factors (anxiety, depression, previous loss, alcohol use, younger age, and lack of other children in the home) in a mother when the possibility of child loss was remote would predict PGD after sudden unexpected loss. Given previously reported high rates of PGD in mothers after SIDS loss and the accompanied modest decreases of PGD over time, we hypothesized that the effects of pre-loss characteristics would become less significant over time.

Methods

Study design

The Safe Passage Study (SPS) was a prospective multi-centered study of 12 000 women at high risk for SIDS, examining aspects

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of infants and their mothers during pregnancy and their first year of life, including pre-loss vulnerability factors for PGD. Participants were recruited from informal settlements near Cape Town, South Africa and in or near Pine Ridge Indian Reservation (Northern Plains), USA, populations sharing extremely impoverished living standards, high rates of maternal alcohol drinking and approximately seven times the overall US SIDS rate (Dukes *et al.*, 2014). This ancillary study enrolled participants who were otherwise terminated from the study following their infant's death, comparing bereavement outcomes to personal and psychological data collected prospectively from pregnancy through bereavement.

Participants

Participants were mothers whose infants died from SIDS or unascertained causes of death in their first year of life after healthy discharge from birth hospitalization, with comparison data from those whose infants survived. Maternal and fetal/infant dyads were followed throughout pregnancy until 1 year post-delivery from August 2007 until January 2015. Bereaved mothers were enrolled after their infant's SIDS death from May 2013 until July 2016. This research was approved by the Institutional Review Boards at Dana-Farber Cancer Institute (protocol 13-207); The Eunice Kennedy Shriver National Institute of Child Health and Development, Bethesda, Maryland; Stellenbosch University, Stellenbosch, South Africa; Sanford Health, Sioux Falls, South Dakota; Oglala Sioux Tribe and Great Plains Institutional Review Board.

Procedures

Surveys were administered by project professional staff at each site. Pre-loss data were obtained at the earliest prenatal visit following enrollment, including the Edinburgh Depression Scale, the Spielberger State-Trait Anxiety Inventory, alcohol use, previous pregnancy loss or death of their child, and demographic measures. Post-loss data were obtained on bereaved mothers between 2 and 48 months after their infant died, with three separate grief inventories administered separated by 6-month intervals. Maternal age when the infant died, and the presence of another living child in the home at the time of the infant's death were collected post-loss.

Measures

Demographics

Basic demographic data were collected including age, race, education, and poverty. Poverty in South Africa was determined using the South African upper bound poverty line (income insufficient to purchase food and essentials meeting a daily minimum energy intake of 2100 kilocalories) (Statistics South Africa, 2015). Poverty in Northern Plains participants was defined using 2015 US poverty levels for a family of 4.

Survey instruments

Spielberger State-Trait Anxiety Inventories (STAI-T and STAI-S) were used to measure trait anxiety, the characterological predisposition to anxiety, and state anxiety, anxiety in response to a specific situation at the time of measurement (Cattell, 1966; Littleton *et al.*, 2007). Reported median reliability coefficients for the trait anxiety scale vary from 0.695 to 0.765 in samples of working adults, college students, high school students, and military recruits (Spielberger, 1983). Normative data are available but various cut-offs and criteria have been used through the literature to identify higher risk. Trait anxiety was used for the analysis as our interest was in stable pre-loss personality characteristics.

We used the Edinburgh Depression Scale, a widely used and validated scale originally designed to screen for post-partum depression (Cox *et al.*, 1996). Its reliability and validity have been demonstrated in diverse international samples (Affonso *et al.*, 2000). This research used the cut-off value of 13, the value typically used for post-partum depression screening, in efforts to more closely identify pathology with greater specificity (Su *et al.*, 2007), recognizing that a lower cut-off is sometimes used during antenatal screening (Bergink *et al.*, 2011; Kozinszky and Dudas, 2015).

A Timeline Follow-Back (TLFB) method was used at the intake prenatal interview to measure alcohol consumption (Sobell and Sobell, 1992). This methodology collected a detailed alcohol consumption history for the last drinking day and 30 days prior (Brick, 2006). We used a cut-off of drinking frequency in the upper quartile to differentiate higher alcohol intake.

The Parental Bereavement Questionnaire (PBQ) was used to measure symptoms of PGD. The PBQ is a modification of the Prolonged Grief Disorder 13-item inventory (PG-13) (Prigerson *et al.*, 2009), adapted for language specific to the loss of a young child and including other factors identified to be of particular concern to parents after the death of a pediatric-aged child. The PBQ measures diagnostic criteria for PGD in 18 items, with items addressing separation distress (yearning and emotional pain), and cognitive, emotional, and behavioral symptoms, including confusion about one's role in life or diminished sense of self, difficulty accepting the loss, avoidance of reminders about the reality of the loss, inability to trust others since the loss, difficulty moving on with life (e.g. making new friends and pursuing interests), emotional numbness since the loss, and feeling stunned, dazed, or shocked by the loss. Adaptations incorporating parent-specific language into the PG-13 were pilot tested on a sample of Massachusetts parents (Goldstein and Rimer, 2013), and input from local focus groups was used to increase reliability for local language preferences specific to Cape Town. Source PG-13 items have a sensitivity of 1.00 and specificity of 0.99 for PGD (Prigerson *et al.*, 2009). Internal consistency of the PBQ is $\alpha = 0.92$. Reliability (Kuder–Richardson) was 0.72 for PGD criteria in this sample.

Statistical analysis

Pre-loss continuous psychometric scores were compared between mothers whose child died from SIDS and a control group of mothers from the SPS whose infants did not die (1:3 case:control ratio matching age, race, and location) using two-sample *t* tests. Pre- and post-loss psychometric scores were compared using paired *t* tests.

We dichotomized continuously and ordinally distributed risk factors in order to assess the trajectories of PGD in mothers with and without risk factors across a continuous measure of time since child loss. We used a median cut-off for STAI-T (score $\geqslant$39) and maternal age at loss ($\geqslant$29 years of age). A cut-off of $\geqslant$13 was used for the Edinburgh Depression Scale to provide greater specificity (see above). High alcohol use was defined as drinking at least two times per week as measured during the intake/pregnancy interview.

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We used generalized estimating equations (GEE, logit link, binomial family) with robust standard errors and an exchangeable correlation structure to model the trajectories of PGD by the presence or absence of pre-loss risk factors over time. Time was defined as months from child loss and ranged from 2 to 48 months. Three observations that were >4 years after loss were excluded as they were considered beyond the observation period of interest. Each model predicting PGD had seven terms: risk factor (dichotomous), time (continuous), quadratic time, interaction of risk factor by time, interaction of risk factor by quadratic time, cohort, and a constant. Linear combinations of the three terms containing the risk factor were used to determine the effect of the risk factor at 6-month intervals since the loss of the child. In addition to individual risk factors, we created a count of risk factors and then trichotomized this count based on a delineation of risk for PGD that was observed between 0–1, 2–3, and >4 risk factors. We calculated a *C*-statistic (area under the curve for a receiver operating characteristic analysis) based on predicted scores from a GEE model with the six individual risk factors as independent variables, restricted to observations between 6 and 24 months from loss. All tests were two-tailed and alpha was set at 0.05.

Results

Pre-loss data were collected in 11 892 participants (59% South Africa). Cause was not determined in 55 infants who died during the enrollment period. Five mothers declined participation, and complete pre-loss psychometric data were available for 44 participants. Fewer Northern Plains deaths reflected relative enrollment, and also coincident prevention efforts leading to SIDS rates (Table 1). Mothers who ultimately lost a baby to SIDS had no significant differences in STAI composite scores, Trait Anxiety scores, or Edinburgh scores during pregnancy than controls. In the mothers whose infants died from SIDS, anxiety scores were higher during bereavement than pre-loss ($p < 0.001$), with a similar trend for depression ($p = 0.09$) (Table 2). During the period of >6 months after their infant's death, 65.3% of the bereaved mothers had at least one interview where criteria for PGD were met. Overall, symptom criteria for PGD were met in 47.3% of the observations.

Stratifying by number of pre-loss risk factors measured (anxiety, depression, alcohol use, previous loss, living child in the home, and maternal age), two or three risk factors predicted significantly greater risk for PGD for 12 months after the loss and four risk factors predicted significantly greater risk for PGD for 24 months following the infant's death (Fig. 1), in comparison to the low-risk group. No participant had more than four risk factors. A transition occurred at approximately 24–30 months post-loss, as rates of PGD increased in those with fewer risk factors with a trend toward convergence at a high rate. This trend toward an increased rate of PGD began 12 months in the low-risk group and at 22 months in the middle-risk group. The *C*-statistic using predictions from the GEE model restricted to 6–24 months after loss using the six factors (anxiety, depression, alcohol, previous loss, child in home, and age at death) was 0.83 (CI 0.71–0.95). Predictions from a simplified model of only risk factors that survived backward deletion at $p < 0.15$ (age, alcohol, and depression) still provided good discrimination (0.80, CI 0.68–0.93).

Analysis of individual risk factors in 30 months post-loss showed differences associated with each pre-loss risk factor and their trajectories for PGD, demonstrating differing contributions to PGD during that period after the infant's death (Table 3, Fig. 2). Pre-loss depression symptoms were significantly associated with PGD for 2 years into bereavement, with odds ratios (OR) over 2.8. When depression and grief were measured concurrently at visit 1, 60% of those with PGD had elevated depression scores while 40% of those without PGD had elevated depression scores ($p = 0.16$). Anxiety predicted the greatest degree of acute grief symptoms but not a significant difference in PGD, which has symptom criteria of >6 months post-loss. Mothers who had experienced prior loss of a pregnancy or child showed non-significantly *lower* rates of PGD, a constant effect over time. Higher prevalences of PGD across time were predicted in mothers who drank alcohol at least 2 days each week (OR>2.0) and older maternal age (OR>2.0), but not to statistical significance. The presence of other living children in the home was not shown to have a protective effect on PGD. Findings for cumulative risk factors, depression, and anxiety remained statistically significant after a sensitivity analysis removed Northern Plains participants.

Table 1. Demographics of participants

Number	50
Age (years)	Mean = 26.4, s.d. = 5.5, Range = 17–44
Race (*n*, %)	
White	2.8%
Cape colored (mixed ancestry)	41.82%
Native American	6.12%
Hispanic	1.2%
Education beyond high school	10%
Poverty	73%
Previous loss	
None	64.2%
Pregnancy loss	22.0%
Death of live-born child	13.8%
Trimester of pre-loss measures	
Second	36
Third	10
Number of post-loss observations (<21 months)	
1	48
2	46
3	40
Initial post-loss observation (months)	
<6	16
6 to <12	2
12 to <18	5
18 to <24	3
24 to <30	9
30 to <36	8
36–48	5

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Table 2. Pre- and post-loss anxiety and depression scores

	Controls ($n = 124$)	Pre-loss ($n = 44$)	Post-loss ($n = 50$)	p Value controls to pre-loss	p Value pre-loss to post-loss
Spielberger STAI Total	71.0 ± 20.0	73.9 ± 23.5	94.8 ± 20.9	0.43	<0.001
Spielberger Trait	40.2 ± 11.5	41.3 ± 13.2	49.1 ± 10.5	0.60	<0.001
Edinburgh	12.8 ± 5.9	13.2 ± 6.5	14.9 ± 6.3	0.71	0.09

Comparison of Spielberger and Edinburgh scores in bereaved mothers ($n = 44$) and controls, and comparison pre- and post-loss scores in bereaved mothers.

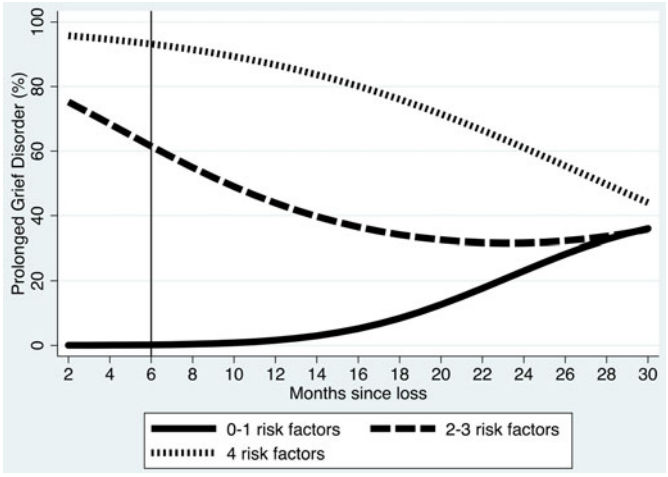

Fig. 1. Cumulative pre-loss risk factors and PGD. Symptoms criteria meeting PGD are displayed from 6 months post-loss through 30 months, until significant statistical differences are no longer seen. Symptom intensity consistent with PGD but not meeting diagnostic criteria for post-loss time interval for PGD are displayed from 2 to 6 months.

Discussion

Less than 0.1% of mortality occurs in children (Centers for Disease Control and Prevention, 2016), reflecting historic advances in medicine and prevention. The parents of pediatric-aged children who die nonetheless experience important and persistent consequences during bereavement (Rando, 1986; Rogers *et al.*, 2008), including increased mortality (Li *et al.*, 2003), mental health hospitalizations (Li *et al.*, 2005), and alterations in future parenting (Warland *et al.*, 2011). The rarity of child death in the 21st century contributes to this difficulty, while increasing the challenges of assembling prospective cohorts. This research focused on the contribution of personal factors existing prior to loss on rates of PGD in mothers following the sudden unexpected death of their infants, using standardized metrics in a cohort with limited variation in many variables influencing grief outcomes. The risk profile generated by the six prospective risk factors had a strong ability to discriminate between mothers who would ultimately suffer from PGD and those who would not. Our data show that a bereaved mother's response to loss is significantly determined before the loss occurs, and that pre-loss vulnerability factors act with cumulative, albeit time-limited, effects.

A unique aspect of these data is its genuinely prospective collection before the infant's death, when the participants were pregnant and without any expectation of their baby's death. This well-characterized sample with an elevated risk for PGD had limited variation in personal risk factors, while the collection of pre-loss psychometrics allowed us to look specifically at six pre-loss personal risk factors: anxiety, depression, alcohol use, previous loss, living child in the home, and maternal age. No <16.2% (low Edinburgh score at 12 months) of participants were predicted to have PGD when no specific risk factor was present. Personal risk factors unrelated to circumstances of death predicted PGD in the first 30 months following the infants' deaths, the time following loss when more attention is conventionally offered to the bereaved. Greater than two risk factors significantly predicted PGD in the year after the infant's death and four risk factors predicted PGD for 2 years.

Although some have argued that grief does not resolve uniformly (Stroebe *et al.*, 2017), it was notable that PGD increased in those at lower risk over time and converged at a high rate without regard for risk. Although requiring further study, the effect size estimated in the model was large and clinically significant. This may not be inconsistent with previous research on grief trajectories (Maciejewski *et al.*, 2007), because that analysis removed subjects with PGD, likely affecting symptom trajectories. Others have reported persistently high symptom levels in significant subpopulations (Maciejewski *et al.*, 2016, Prigerson *et al.*, 2009, Zisook and Shuchter, 1985) and delayed symptom peaks at approximately 2 years (Zisook *et al.*, 1982). In our participants, these later increases in PGD may in part be related to typical spacing between children, and reflect ambivalence when a mother considers a subsequent pregnancy and the birth. Mothers may scrutinize their relationship with the deceased while bonding with their next baby or when the infant reaches the age of the deceased; alternatively, they may experience heartache as peers expand families. In exit interviews, mothers identified the emotional difficulties that come with time, as their child is less remembered by others and attention to the presence of their child becomes more difficult for themselves. Mothers report high and persistent levels of role confusion following SIDS loss (Goldstein *et al.*, 2018).

Pre-loss factors have different associations with grief. Higher trait anxiety had a more significant effect on acute grief symptoms (<6 months) than PGD, which is diagnosed 6 months following loss. Higher depression scores were strongly associated with PGD but the imperfect overlap was not consistent with confounding. Alcohol use and maternal age at loss showed a consistent effect size and, arguably, the lack of statistical significance may reflect the limited power of the sample size to demonstrate the effect. While it is not uncommon for bereaved parents to report the affirmative narrative that their other children helped them cope with their grief, our data suggest a modest effect. Finally, we did not find a strong risk in mothers who experienced prior loss during pregnancy or after the death of a live-born child, but instead found a trend toward a protective effect.

Depression significantly predicted PGD for 2 years following loss but while co-occurring, was not collinear. SIDS is a loss

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Table 3. Personal risk factors and PGD over time

	6 months	12 months	18 months	24 months	30 months	36 months	42 months	48 months
4 risk factors ($n = 9$)	93.2%** $p = 0.003$	86.7** $p = 0.001$ ($p = 0.03$ *v.* 2–3 risk factors)*	76.0%** $p = 0.001$ ($p = 0.01$ *v.* 2–3 risk factors)**	61.1%* $p = 0.02$ ($p = 0.02$ *v.* 2–3 risk factors)*	44.0% $p = 0.60$	28.6% $p = 0.67$	17.2% $p = 0.74$	9.8% $p = 0.95$
2–3 risk factors ($n = 26$)	61.5%* $p = 0.02$	43.9%* $p = 0.02$	34.1% $p = 0.10$	31.6% $p = 0.58$	35.8% $p = 0.99$	47.6% $p = 0.50$	66.6% $p = 0.15$	85.6% $p = 0.09$
0–1 risk factors ($n = 9$)	0.2%	1.6%	8.4%	23.1%	36.0%	37.8%	27.4%	11.8%
High depression ($n = 26$)	75.6%* $p = 0.02$	64.8%** $p = 0.002$	54.6%** $p = 0.002$	46.3%* $p = 0.04$	40.5% $p = 0.40$	37.0% $p = 0.50$	35.8% $p = 0.22$	36.8% $p = 0.24$
Low depression ($n = 18$)	16.2%	16.2%	18.3%	23.0%	31.6%	45.2%	62.9%	80.1%
High trait anxiety ($n = 22$)	80.0%* $p = 0.01$	52.7% $p = 0.29$	34.8% $p = 0.73$	30.9% $p = 0.44$	39.5% $p = 0.95$	62.3% $p = 0.10$	87.9%* $p = 0.02$	98.3%* $p = 0.02$
Low trait anxiety ($n = 22$)	32.4%	36.8%	40.0%	40.8%	40.2%	37.9%	34.0%	28.7%
High alcohol use ($n = 10$)	81.4% $p = 0.29$	70.7% $p = 0.18$	60.9% $p = 0.18$	54.2% $p = 0.17$	51.2% $p = 0.19$	52.3% $p = 0.42$	57.4% $p = 0.72$	65.9% $p = 0.88$
Low alcohol use ($n = 36$)	64.4%	49.9%	39.7%	34.5%	33.7%	37.2%	45.3%	58.4%
Previous loss ($n = 17$)	54.6% $p = 0.19$	45.7% $p = 0.34$	39.2% $p = 0.57$	35.0% $p = 0.69$	33.0% $p = 0.63$	33.0% $p = 0.50$	35.0% $p = 0.48$	39.1% $p = 0.50$
No previous loss ($n = 29$)	76.7%	61.0%	48.1%	40.6%	38.8%	42.3%	51.4%	65.7%
⩾26 years of age at loss ($n = 26$)	73.4% $p = 0.30$	61.1% $p = 0.19$	51.4% $p = 0.18$	45.8% $p = 0.15$	44.4% $p = 0.08$	47.3% $p = 0.08$	54.5% $p = 0.26$	65.4% $p = 0.49$
<26 years of age at loss ($n = 24$)	56.4%	42.4%	32.8%	27.6%	25.8%	27.3%	32.1%	41.2%
No other children in the home ($n = 17$)	51.3% $p = 0.24$	45.8% $p = 0.59$	40.6% $p = 0.88$	36.0% $p = 0.84$	31.8% $p = 0.42$	28.2% $p = 0.06$	25.0%* $p = 0.048$	22.4% $p = 0.07$
Other children in the home ($n = 33$)	69.8%	53.7%	42.9%	38.8%	40.9%	49.6%	64.3%	81.0%

The upper section shows strata of cumulative risk factors from 6 to 48 months post-loss with prevalence of PGD and *p* values in comparison with lower risk groups. The lower section shows rates of PGD by individual risk factor over time, displaying *p* values

that threatens and devalues a bereaved mother's core role, features strongly implicated in depression (Kendler *et al.*, 1998; Kendler *et al.*, 2003), while bereavement itself increases risk for the onset of many psychiatric disorders (Keyes *et al.*, 2014). Nonetheless, PGD symptoms and diagnosis have been shown to be distinct from symptoms and diagnosis of major depressive disorder (Prigerson *et al.*, 2009), demonstrating a coherent structure with distinct components (Prigerson *et al.*, 1995; Prigerson *et al.*, 1996*a*). PGD and depression are understood to have different etiologies and risk factors, and predict different outcomes (Prigerson *et al.*, 1999). Our data are consistent with prior observations that PGD and depression demonstrate incremental validity, where, for example, grief was associated with a fivefold greater likelihood of suicidal ideation after controlling for depression, while comorbid grief and depression were not (Prigerson *et al.*, 1997).

There was a trend toward less PGD when mothers had experienced a prior loss. Other prospective studies have concluded that prior loss was associated with worsened grief during the subsequent loss (Janssen *et al.*, 1997). Conceptually, however, the ameliorating influence we observed is consistent not only with the notion of desensitization to stressors but also with the understanding of grief as an attachment reaction. In qualitative research, Warland has observed paradoxical parenting in parents after SIDS, a tendency to hover and overattend to subsequent children while maintaining a more distant attachment style (Warland *et al.*, 2011). While sample size limitations make this a matter for speculation, we suggest that parental attachment in subsequent children after a loss compensates for the vulnerability that was experienced.

This study is limited by its sample size. This small and seemingly idiosyncratic sample required following 12 000 mothers whose infants experienced a leading form of child mortality. Nonetheless, the lack of power for risk factors may fail to detect true associations (type 2 error). Power considerations aside, however, caution should be exercised about the generalizability of the findings. Prior research provided evidence that findings from this sample are generalizable to mothers after SIDS in other settings (Goldstein *et al.*, 2018). With regard to other causes of a child's death and PGD, there is little reason to conclude that the risk factors described in the sample operate differently. However, whether conclusions from this specific cohort who experienced a sudden, unexplained loss at a maternal moment of extremely strong attachment behavior can be generalizable to death in adults, notably elderly life partners, is uncertain. Yet a separation of this experience from mainstream grief research begs the question of how parental grief in young families is different and whether it thus warrants separate considerations as a psychological category. Finally, we note that the discrimination in our

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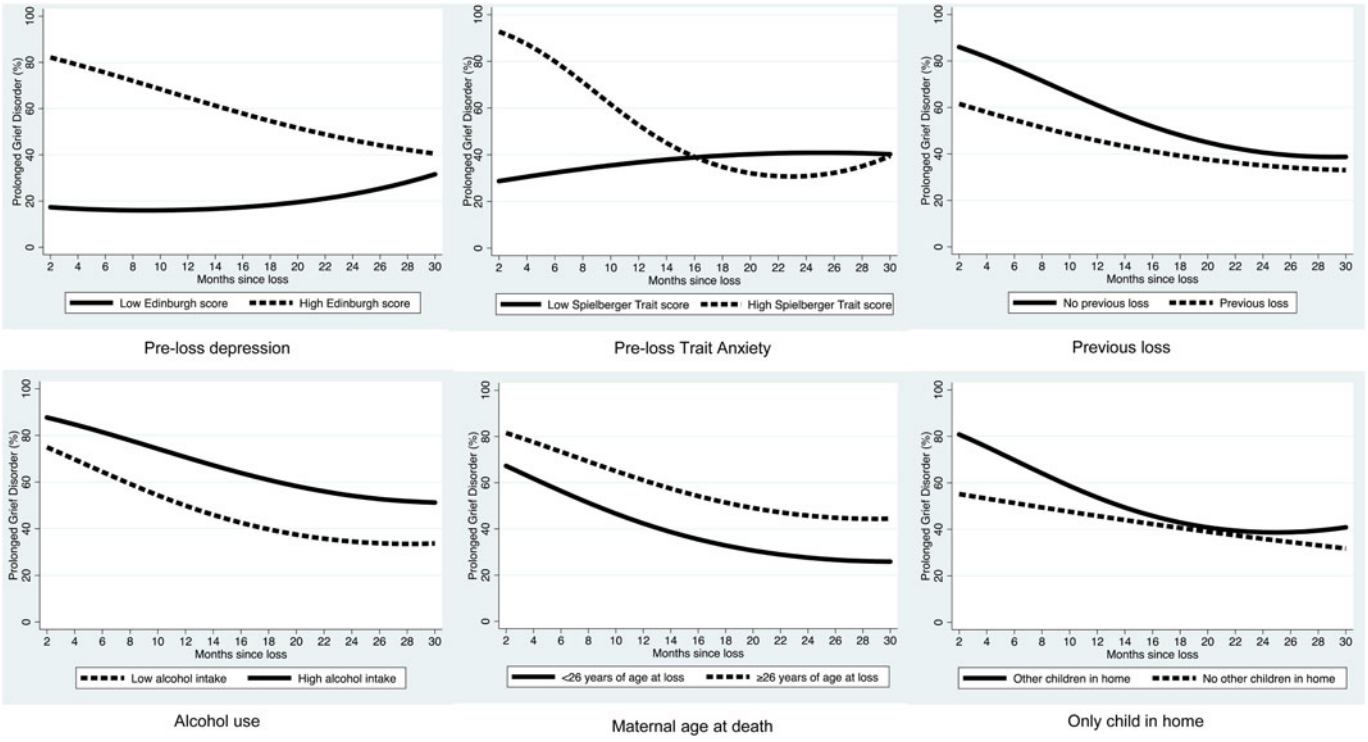

Fig. 2. PGD and individual personal factors. Trajectories of PGD for individual risk factors and their absence until 30 months post-loss. Symptom intensity consistent with PGD but not meeting diagnostic criteria for post-loss time interval for PGD are displayed from 2 to 6 months.

risk predictions may not be as strong when tested outside of the sample used to develop it.

Although lacking power to determine whether relationships vary by cohort, our finding remained significant when we conducted a sensitivity analysis removing Northern Plains participants. With ~350 births per year, Pine Ridge contributed many fewer subjects, but we chose to include its participants because they were central to the prospective design of SPS, out of respect for the fact that the research was supported by the Oglala Sioux tribe, and because of our desire to include a population not generally included in medical research by virtue of race and poverty [lowest per capita income by county in the USA (United States Census Bureau, 2017)].

This research documents that personal risk factors measured in mothers during the course of a normal pregnancy predict significantly higher risk for PGD following the sudden unexpected deaths of their infants. The risk factors had a cumulative, time-limited effect, with decreased significance after 2 years and convergence of PGD at high levels. Although limited by statistical power, we found differences in effect size and prevalence trajectories for PGD based upon pre-loss factors. Our risk prediction results showed excellent discrimination between mothers who did and did not develop PGD. Personal and psychological characteristics present in mothers before bereavement significantly influence PGD.

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