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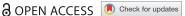
CLINICAL RESEARCH ARTICLE

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Prolonged grief disorder in Chinese Shidu parents who have lost their only child

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ABSTRACT

Background: China has the largest population of '"loss-of-only-child' parents, that are also known as Shidu parents in Chinese society; however, little is known about their unresolved grief.

Objective: This is the first study to examine the grief symptoms, prevalence, comorbidity and potential predictors of prolonged grief disorder (PGD) in such parents, taking into consideration that the new PGD diagnostic criteria ICD-11 will soon be implemented in China.

Methods: 149 Shidu parents completed assessments of PGD (PG-13), PTSD (PCL-C), depression (CES-D) and general psychiatric morbidity (GHQ-12) via in-person interviews.

Results: Of the 149 Shidu parents, 22.2% met the PGD criteria, with a mean of 7.59 years post-loss, and 62.4% experienced daily longing or yearning. Regression analysis indicated that fewer years since loss, subjective perception of poor economic situation, female gender and more hospital visits were prominent risk factors for the development of PGD. Older age of the parents at the time of child loss was also associated with PGD. Parents with PGD had higher comorbidity of PTSD or depression compared with those without PGD.

Conclusions: There is a high prevalence of PGD and a high rate of comorbidity between PGD and PTSD or depression in Shidu parents in China, which highlights the need of timely developing effective assessments and interventions to prevent PGD in this population, particularly in female, recently bereaved, low-income and aged parents who lost their only child.

Trastorno de duelo prolongado en padres chinos shidu que han perdido a su hijo único

Antecedentes: China tiene la población más grande de padres que han perdido a su hijo único, conocidos como los padres shidu en la sociedad china; sin embargo, se conoce poco respecto a su duelo no resuelto.

Objetivo: Este es el primer estudio en evaluar los síntomas de duelo, y la prevalencia, comorbilidades y predictores potenciales del trastorno de duelo prolongado (TDP) en este tipo de padres, tomando en consideración que los nuevos criterios diagnósticos para el TDP de la CIE-11 se implementarán pronto en China.

Métodos: 149 padres Shidu completaron evaluaciones para TDP (PG-13), TEPT (PCL-C), depresión (CES-D) y morbilidad psiquiátrica general (GHQ-12) mediante entrevistas en persona.

Resultados: De los 149 padres shidu, 22,2% cumplieron los criterios para TDP, con un promedio de 7,59 años pospérdida, y 62,4% experimentaron nostalgia y anhelo. Los análisis de regresión indicaron que menos años luego de la pérdida, una percepción subjetiva de una pobre situación económica, el sexo femenino, y un mayor número de consultas en hospitales eran factores de riesgo prominentes para el desarrollo de TDP. Una mayor edad de los padres al momento de la pérdida del niño también se asoció con TDP. Los padres con TDP tenían mayor comorbilidad de TEPT o depresión comparados con aquellos que no tenían TDP.

Conclusiones: Existe una alta prevalencia del TDP y una alta tasa de comorbilidad entre el TDP, el TEPT o la depresión en padres Shidu en China, lo cual resalta la necesidad de desarrollar a tiempo evaluaciones e intervenciones efectivas para prevenir el TDP en esta población, en particular en mujeres, en deudos recientes, en aquellos con bajos ingresos, y en padres de edad avanzada que perdieron a su hijo único.

中国失独父母的延长哀伤障碍研究摘要

背景: 中国有最多的'失去唯一的孩子'的父母, 在中国社会被称为失独父母, 但对于他们尚未解决的哀伤却知之甚少。

HIGHLIGHTS
• 'Loss-of-only-child' parents are known as Shidu parents in Chinese society.
• This is the first study to examine the grief symptoms, prevalence, comorbidity and potential predictors of prolonged grief disorder among Shidu parents.
• As high as 22.2% of the sample met the PGD criteria, with a mean of 7.59 years post-loss. As high as 62.4% of Shidu parents experienced daily longing or yearning.
• Fewer years since loss, subjective perception of poor economic situation, female gender and more hospital visits were prominent risk factors for the development of PGD.
• Parents with PGD had higher comorbidity of PTSD or depression compared with those without PGD.

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目的: 新的ICD-11中, PGD诊断标准将很快在中国使用, 本研究是首次对中国失独父母的哀伤症状, 患病率, 共病情况和可能的预测因素进行研究。

方法: 149名失独父母通过面谈完成PGD (PG-13), PTSD (PCL-C), 抑郁(CES-D)和一般精神疾病(GHQ-12)的评估。

结果: 149对失独父母中, 22.2%符合PGD标准, 其平均失独年限为7.59年, 62.4%的父母每天都在怀念失去的孩子。回归分析表明, 失独后年限短, 主观认为经济状况差, 女性性别和更多的就医次数是PGD发生的主要危险因素。失独时父母的年龄大, 也与PGD相关。与没有PGD的父母相比, 有PGD的父母有更高的PTSD或抑郁的共病率。

结论: 中国失独父母PGD患病率高, PGD与PTSD或抑郁的共病率高, 这提示了需要制定及时有效的评估和干预措施, 以预防该人群中PGD的发生, 尤其是女性, 最近刚丧失子女, 低收入和年纪大的失独父母。

1. Introduction

Grief disorder has been a topic of great interest for researchers and clinicians over the past two decades. Persistent complex bereavement disorder (PCBD) is defined as a disorder requiring further study in the 5th Edition of Diagnostic and Statistical Manual of Mental Disorders (DSM-5). PCBD is a code diagnostically corresponding to prolonged grief disorder (PGD), which is included in the 11th Edition of the International Classification of Diseases (ICD-11) and is defined as a new mental health disorder. PGD is a proposed diagnostic category intended to classify bereaved individuals who experience notable dysfunction for atypically long periods of time following a significant loss (Prigerson et al., 2009). Core symptoms include a pervasive yearning for the deceased or persistent preoccupation with the deceased accompanied by intense emotional pain (World Health Organization [WHO], 2016). Furthermore, PGD is characterized by difficulties in engaging in social or enjoyable activities, a reduced ability to experience positive mood, and difficulties accepting the death of the loved one (WHO, 2016). A duration criteria of 6 months is proposed to ensure that natural grief reactions in the acute state following bereavement are not confounded with the syndrome of PGD (Prigerson et al., 2009). The first systematic review and meta-analysis on the prevalence of PGD due to non-violent loss (including two Chinese studies) reports that the incidence of PGD in the adult bereaved population is 9.8% (95% CI 6.8–14.0) (Lundorff, Holmgren, Zachariae, Farver-Vestergaard, & O'Connor, 2017), and this figure is 1.8% (8/445) (He et al., 2014) to 13.9% (153/1099) within the general population of China (Li & Prigerson, 2016). The former study used Prolonged Grief Questionnaire-13 (PG-13) (Prigerson et al., 2009), in which the spouse or first-degree relative loss accounted for only 31.1% of the participants investigated. The latter study conducted by Li, J and HG Prigerson used Inventory of Complicated Grief (Prigerson et al., 1995), in which all the participants lost their spouses or first-degree relatives. The authors of this study speculate that different

assessment instruments and different relationships to the deceased may lead to disparities, highlighting the importance of investigating a homogeneous population for better understanding PGD in the context of Chinese culture. Additionally, there is limited information about differences in PGD between bereaved people in Western countries such as the Netherlands, Germany and the USA and those in Chinese populations. As PGD is a newly refined diagnostic category, epidemiological studies of the Chinese population are required to further improve our understanding about PGD and confirm its clinical utility in a unique Eastern culture.

The death of the child is one of the most traumatic bereavement events, and bereaved parents may experience persistent intense grief associated with significant distress and impairment (Morris, Fletcher, & Goldstein, 2019). Prevalence of PGD among parents bereaved of a child varies and is estimated to be 10.3% in Australian parents whose children died of cancer (McCarthy et al., 2010); 13% in German parents who cared for and lost their terminally ill children (Vollenbroich et al., 2012); 16.0% in bereaved Swedish parents whose children died of cancer in previous 1–5 years (Pohlkamp, Kreicbergs, Prigerson, & Sveen, 2018); 30% in North American and Australian parents who had lost a child to death (Keesee, Currier, & Neimeyer, 2008); 40-50% in bereaved mothers of other Western countries whose children died of sudden infant death syndrome (Goldstein, Lederman, et al., 2018; Goldstein, Petty, et al., 2018); and 94% in bereaved South Korean parents of the Sewol Ferry accident (Huh, Huh, Lee, & Chae, 2017). Due to the one-child policy, China has the largest population of 'loss-of-only-child' parents. Among them, couples above 49 years old and cannot have further children (unable to have children and unwilling to adopt children) are referred to as 'Shidu' parents (Shi et al., 2019; Yin, Sun, & Liu, 2018; Zhou et al., 2018) in Chinese society. The increasing size of this group in the population has brought a significant social impact and attracted government's attention. Domestic researchers have begun making preliminary

investigations about the mental health status of Shidu parents in recent years and revealed that these parents experienced more anxiety and depression symptoms than non-bereaved counterparts (Cao, Yang, & Wang, 2018). They were found to be less independent in their daily life or have significantly smaller social networks or fewer social supports compared with non-Shidu parents (Zheng, Lawson, & Anderson Head, 2017). However, little attention has been focused on prolonged grief. To the best of our knowledge, only one study completed a survey either via online or face-to-face interview, and reported a 16.2% occurrence of PGD in 536 Shidu parents, by using rating scale instead of clinical interview (Zhou et al., 2018). There is still a lack of empirical evidence on the prevalence of PGD in the Chinese Shidu population, particularly the data based on clinical interviews. In addition, no study has so far provided an explicit description about the full spectrum of grief symptoms experienced by Chinese Shidu parents. The present study aims to fill in the gaps.

The accumulated evidence supports that PGD is a qualitatively unique grief-related disorder distinct from bereavement-related depression, anxiety, and post-traumatic stress disorders (PTSD) (Barnes, Dickstein, Maguen, Neria, & Litz, 2012; Boelen, van de Schoot, van den Hout, de Keijser, & van den Bout, 2010; Golden & Dalgleish, 2010; Spuij et al., 2012; Tsai, Kuo, Wen, Prigerson, & Tang, 2018), which provides convincing data for the establishment of the ICD-11 PGD criteria. Although the reported comorbidity is relatively high in PGD, PTSD or depression (Boelen, Lenferink, Nickerson, & Smid, 2018; He et al., 2014; McCarthy et al., 2010; Schaal, Dusingizemungu, Jacob, Neuner, & Elbert, 2012; Schaal, Richter, & Elbert, 2014), to the best of the authors' knowledge, no study has examined the comorbidity of PGD among Chinese Shidu parents. Our previous study found that Shidu parents had a higher risk of developing PTSD and depression compared with non-bereaved counterparts (Yin, Shang, et al., 2018). To fill in this research gap, this paper for the first time examined the comorbidity of PGD in Chinese Shidu parents through in-person interviews by either a senior psychiatrist or a psychologist.

Several studies have reported risk factors associated with the development of prolonged grief in different population samples, including gender, age (Chiambretto, Moroni, Guarnerio, Bertolotti, & Prigerson, 2010), time since loss (Chiambretto et al., 2010; Coelho, Delalibera, Barbosa, & Lawlor, 2015; Heeke, Stammel, & Knaevelsrud, 2015; Keesee et al., 2008; McCarthy et al., 2010; Schaal, Jacob, Dusingizemungu, & Elbert, 2010), cause of loss (Boelen, 2015; Keesee et al., 2008; McCarthy et al., 2010), family's economic status (McCarthy et al.,

2010; Yi et al., 2018), religious beliefs (Schaal et al., 2014), emotional closeness to the deceased (Schaal et al., 2014); but produced inconsistent conclusions. For instance, some studies (Neria et al., 2007; Morina, Rudari, Bleichhardt, & Prigerson, 2010; Johnsen, Dyregrov, & Dyregrov, 2012; Schaal et al., 2014; Lai et al., 2015; Xiu et al., 2016; Yi et al., 2018, Yin, Shang, et al., 2018; Yin, Sun, et al., 2018) found that the female gender was a predictor for prolonged grief, and one study (Li, Laursen, Precht, Olsen, & Mortensen, 2005) reported that bereaved mothers had a relatively high risk of being hospitalized for any psychiatric disorder as compared with bereaved fathers, while other authors reported no significant association between grief and gender (Boelen & van den Bout, 2005; Momartin, Silove, Manicavasagar, & Steel, 2004; Schaal et al., 2010). There is limited research about potential risks of PGD in the Chinese population (He et al., 2014), much less in Shidu parents. In addition, traditional preference for sons over daughters is still dominating many Chinese parents' minds, yet associations between gender of lost child and grief severity in Shidu parents have not been researched. Previous researches in Western countries have not found any evidence that the gender of lost child predicts parental wellbeing (Espinosa & Evans, 2013; Werthmann, Smits, & Li, 2010), yet studies using data from Taiwan reported that the risk of maternal suicide or depressive symptoms increased or maternal self-rated health declined following the death of a son, but not after the death of a daughter (Chen, Kuo, Wu, & Yang, 2012; Lee, Glei, Weinstein, & Goldman, 2014). Lastly, our previous study (Yin, Shang, et al., 2018; Yin, Sun, et al., 2018) found Shidu parents had poor physical health with higher rate of chronic diseases than nonbereaved Chinese parents. Xiu et al. also reported general health may predict PGD for Chinese sample (Xiu et al., 2016). However, Robbins-Welty et al. did not find medical comorbidity was associated with chronicity and severity of complicated grief (Robbins-Welty et al., 2018). So far no study has tested whether physical conditions are associated with the grief severity of Shidu parents. In this present study, for the first time the correlates of PGD in Shidu parents were investigated, with the expectation of improving the present knowledge and developing effective and viable intervention strategies.

Chinese clinicians have limited knowledge about PGD because it is the first time to be included in ICD-11 and the translation is still in progress. This study may provide useful data and information for mental health professionals in China to raise awareness of PGD, for instance, psychiatrists or psychologists would consider the possibility of PGD, if patients report the experience of child loss. Furthermore, this study investigated the prevalence,

comorbidity and the potential risk factors of PGD within the Shidu population, which could have important clinical and therapeutic implications for this special group.

Overall, the objectives of this study were to 1) investigate the prevalence of PGD among Chinese Shidu parents, and to examine the hypothesis that these parents would report higher prevalence of PGD than the general Chinese population; 2) identify risk factors associated with PGD among Chinese Shidu parents. Specific factors hypothesized to be associated with PGD included bereavement-related characteristics (shorter time since loss, older age of deceased child, male gender of deceased child or accidental death), and parents characteristics (female, older age, no religious belief, lower family income and poor health); 3) explore the comorbidity between PGD and PTSD or depression in Chinese Shidu parents; and 4) describe the full spectrum of grief symptoms experienced by Chinese Shidu parents.

2. Methods

2.1. Participants and procedures

Data about Shidu parents were obtained from the Register of Population Statistics provided by the Health Family Planning Commission of China. The inclusion criteria were: 1) both Shidu parents were above 49 years of age, which is thought to be the age beyond female reproductivity (according to Chinese government's official definition) (Shi et al., 2019; Zhou et al., 2018, p. 2) the participants had only one biological child and this child died at least 1 year ago; and 3) the participants had not adopted a child by the time they participated in this study. Individuals with serious mental disorders (e.g. schizophrenia, Alzheimer's disease, mental retardation) were excluded. We collected altogether 800 Shidu families from 11 communities of a district in Shanghai by stratified random sampling and selected one parent from each Shidu family as the participant. Of them, 155 Shidu parents met the inclusion criteria. Local social workers who already knew and supported the Shidu families assisted the research by providing a brief description of the study during their routine support group meetings or home visits. In order to make the parents comfortable, the interview could be completed at their homes if they preferred. Local social workers introduced the interviewers to the parents in person at the beginning of the interview. Participants were provided with written informed consent before the survey, and interviewers explained the purpose and significance of the survey. Each participant got a practical gift (worth 8 US dollars) as compensation after completion of the survey. The interviewer team included a psychiatrist, a psychologist and several postgraduates majoring in psychology; they conducted the face-to-face interviews with the Shidu

parents and provided necessary support when asking about sensitive items. During the interview, only two interviewers (a psychiatrist or psychologist plus a postgraduate) and the interviewee with or without his/her partner were present. No social workers and others were around. The psychiatrist and psychologist were responsible for the clinical interview of PGD, and rating the questionnaires of PTSD and depression when face-to-face with the participants; postgraduates implemented the household survey. When the participants became emotionally agitated during the interview, the interviewers would pause to comfort them or allow them to finish the investigation at a second interview. One hundred and fifty-five Shidu parents signed the written informed consent, 149 completed the in-person interview lasting approximately 1 hour, 6 quit during the interview. Data were collected between September 2015 and January 2017. Descriptive data of the total sample are presented in Table 1.

This project was approved by the Ethics Committee of the Secondary Military Medical University (Shanghai, China) and all the research processes met ethical standards.

2.2. Instruments

2.2.1. Socio-demographic and bereavement-related information

Socio-demographic information was obtained, including gender, age, marital status, educational background, religious belief, subjective assessment of the family economic status. The loss-related variables included gender and age of the deceased child, cause of death, time since loss, and age of the parents at the time of loss, which were obtained from the Register of Population Statistics provided by the Health Family Planning Commission of China. The causes of death in the database are recorded as 'disease' or 'accident' (including traffic accidents or other accidents without intent, suicides, and homicides). We did not ask parents about the specific cause of child's death during the interview to avoid evoking further distress.

2.2.2. Questionnaires for physical health

Questionnaires for physical health outcomes included two items. One was designed for collecting information about whether the participants had underlying chronic diseases including hypertension, coronary heart disease (CHD), diabetes mellitus (DM), cerebrovascular diseases (CVD), gastrointestinal (GI) diseases, respiratory disease, rheumatism, osteoarthropathy, tumours, and mental illnesses. The other item measured the number of outpatient visits for physical health or other reasons in the past year. It was ranked in 4 levels: 1 = 0–2 times; 2 = 3–5 times; 3 = 6–9 times; 4 = ≥10 times.

Table 1. Socio-demographic and bereavement-related data of Shidu parents and comparison between PGD-positive group and PGD-negative group.

	N	%	PGD-positive group		PGD-negative group		χ2	p-value
			n	%	n	%		
Total	149	100.0	33	22.1	116	77.9		
Gender of participants								
Female	90	60.4	25	27.8	65	72.2	4.18	0.041*
Male	59	39.6	8	13.6	51	86.4		
Age of participants								
50–62	73	49.0	19	26.0	54	74.0	1.25	0.264
63–75	76	51.0	14	18.4	62	81.6		
Marital status								
Married	124	83.2	30	24.2	94	75.8	2.19	0.534
Divorced	4	2.7	0	0.0	4	100.0		
Remarried	7	4.7	1	14.3	6	85.7		
Widowed	14	9.4	2	14.3	12	85.7		
Educational Level								
Lower than high school	74	49.7	18	24.3	56	75.7	1.99	0.371
High school	55	36.9	13	23.6	42	76.4		
Over than high school	20	13.4	2	10.0	18	90.0		
Religion Belief								
None	122	81.9	27	22.1	95	77.9	0.55	0.759
Buddhism	21	14.1	4	19.0	17	81.0		
Other	6	4.0	2	33.3	4	66.7		
Subjective assessment of family economic status								
Wealthy and extremely wealthy	29	20.0	3	10.3	26	89.7	7.42	0.024*
Moderate	101	69.7	23	22.8	78	77.2		
Poor and extremely poverty	15	10.3	7	46.7	8	53.3		
Gender of deceased child								
Female	62	41.6	11	17.7	51	82.3	1.20	0.274
Male	87	58.4	22	25.3	65	74.7		
Cause of death								
Disease	107	71.8	23	21.5	84	78.5	0.09	0.760
Accident	42	28.2	10	23.8	32	76.2		
	M	SD	M	SD	M	SD	T	p-value
Age of deceased child	22.24	8.30	24.55	6.05	21.58	8.75	−1.82	0.070
Time since loss(years)	11.18	7.04	7.59	4.04	12.20	7.38	4.69	0.000***
Age of parent when losing child	51.07	8.09	53.65	6.82	50.34	8.29	−2.10	0.037*
Age of parent at present	62.25	4.88	61.24	4.74	62.53	4.90	1.35	0.180

*p-value < 0.05, ***p-value < 0.001.

2.2.3. Semi-structured clinical interview for PGD

The psychiatrist and psychologist in the research team conducted the face-to-face clinical interviews to determine the diagnosis of PGD. They used Prolonged Grief Questionnaire-13 (PG-13), a diagnostic tool of PGD, to perform the semi-structured clinical interview. Thirteen items are listed in PG-13, based on the 'PGD-2009 criteria' (Killikelly & Maercker, 2017; Prigerson et al., 2009): item #1 and 2 assess separation distress (longing or yearning for the loss, emotional pain or sorrow), item #3 to 11 are related to cognitive, emotional and behavioural symptoms (e.g. difficulties accepting the loss, bitterness/anger, and avoidance), item #12 measures duration criterion, and item #13 assesses impairment criterion. Item #1 to 11 are rated on a 5-point Likert scale (1 = not at all, 5 = several times a day or overwhelmingly), and item # 12 and 13 are answered with 'No' or 'Yes' (0 = no, 1 = yes). For an individual to be diagnosed with PGD, each of the requirements for Criteria A-E must be met. These criteria include (A) Event Criterion: the individual has experienced bereavement. (B) Separation Distress: the bereaved person must experience item #1 or 2 at least daily. (C) Duration Criterion: the symptoms of separation distress must be elevated at least 6 months after the loss, that is, item #12 must be answered as 'Yes'. (D) Cognitive, Emotional, and Behavioural symptoms: the individual must experience at least five of the items #3-11 at least 'once a day' or 'quite a bit'. (E) Impairment Criterion: the individual must have significant impairment in social, occupational, or other important areas of functioning (Prigerson et al., 2009). The present study defines 'PGD-positive' if the participant meets all of the requirements for Criteria A-E. For regression analyses, the total score of PGD symptom was obtained by summing the 11 items (items #1 to 11) capturing the frequency of grief symptoms. The translated Chinese version of PG-13 was used in the present study, knowing that it has satisfactory psychometric properties (He et al., 2014), with an internal consistency of 0.891 (Cronbach's alpha).

2.2.4. PTSD checklist-civilian version (PCL-C)

PTSD Checklist-Civilian Version (PCL-C) for DSM-IV developed by Weathers et al. (Weathers, Litz, Herman, Huska, & Keane, 1993) was used to screen for bereavement-related PTSD. The participants responded to 17 items on a 5-point scale ranging from 1 (not at all) to 5 (extremely) in three areas: intrusion, avoidance and numbing, and hyperarousal. A PTSD severity-score was computed by summing up

all symptom scores. The Chinese version of PCL-C showed excellent internal consistency and convergent validity with other measures of PTSD severity in previous studies (Yang, Yang, & Liu, 2007). A cut-off score of 50 has been proposed to suggest a PTSD diagnosis (Brewin, 2005), with an internal consistency of 0.933 (Cronbach's alpha).

2.2.5. Depressive symptoms

Depressive symptoms were measured with the 20-item Centre for Epidemiological Studies Depression Scale (CES-D) (Radloff, 1977), which is based on statements rated by the respondent (range 0–3) according to the frequency experienced in the past week. The composite scores of the CES-D items range from 0 to 60, with higher scores indicating more frequent symptoms. The measurement showed the same validity in China (Cheng & Chan, 2005) as that in Western countries (Turvey, Wallace, & Herzog, 1999). A cut-off score of 16 has been proposed to suggest a depression diagnosis (Lewinsohn, Seeley, Roberts, & Allen, 1997), with an internal consistency of 0.953 (Cronbach's alpha).

2.2.6. General mental health

The General Health Questionnaire (GHQ-12) was used to evaluate the severity of general psychiatric morbidity. As a shortened version of GHQ-60, it is well adjusted for long-term studies that require an indicator of minor psychiatric morbidity and has proved to be valid in both the clinical and general populations (Pevalin, 2000). The Chinese version of GHQ-12 has been demonstrated to have sound reliability and validity of the diagnostic performance (Pan & Goldberg, 1990). The 12 items are scored on the basis of 0-0-1-1 (the first 2 answer scores are 0 and the 2 following scores are 1). The total scores range from 0 to 12, with higher scores indicating a worse health condition. The internal consistency of GHQ-12 in the present study was 0.856 (Cronbach's alpha).

2.3. Data analysis

This study conducted 155 in-person interviews and collected 149 valid questionnaires (with 6 quitting from the investigation). Analyses were performed using IBM Statistical Package for Social Sciences version 21.0. Missing data (less than 0.5%) were dealt with by pair-wise deletion in an analysis-by-analysis approach. Descriptive data are presented as frequencies, mean scores and standard deviations (SD). Chi-square and t-tests were used to analyse between-group differences. Multiple linear regression was calculated for the association between PGD scores and independent variables. Categorical variables were firstly transformed to dummy variables and then entered into the model. A dummy variable is an artificial variable created to represent an attribute with two or more distinct categories/levels. For example, subjective assessment of the family economic status had 3 levels (wealthy and extremely wealthy, moderate, poor and extremely poor). Therefore, there are two dummy variables. If wealthy and extremely wealthy is set as a reference group, the two dummy variables are moderate vs. wealthy and extremely wealthy and poor and extremely poor vs. wealthy and extremely wealthy. Moderate vs. wealthy and extremely wealthy = '1′, if the subjective assessment of the family economic status is moderate and '0′ otherwise.

3. Results

The socio-demographic and bereavement-related data of the 149 Shidu parents are presented in Table 1. The mean age of the participants was 62.25 (50–75) years at the time of investigation. Of the 149 Shidu parents, 90 (60.4%) were female, and the mean length of time since the loss was 11.18 (1–38) years (SD = 7.04).

The mean total score of PG-13 was 28.75 (1–55) (SD = 11.95). The frequency of item responses from all the participants (n = 149) completing PG-13 is presented in Table 2. Of the 149 Shidu parents, 62.4% experienced daily longing or yearning; 40.9% experienced daily emotional pain, sorrow or grief, meeting the Separation Distress Criteria of PGD; 37.6% presented with cognitive, emotional and behavioural symptoms of avoidance behaviour daily; 36.9% had trouble in accepting the loss; 36.9% felt stunned, shocked, or dazed daily; and 22.2% (n = 33) met the criteria for PGD with a mean of 7.59 years post-loss.

Table 2. Percentage of Shidu parents experiencing symptoms of prolonged grief.

PG symptoms according to the PG-13	M	SD	Almost never	Rarely	Sometimes	Often	Always
Feelings of longing or yearning	3.62	1.35	13.4%	6.0%	18.1%	29.5%	32.9%
Intense feelings of emotional pain, sorrow, or grief	3.11	1.30	14.8%	17.4%	26.8%	23.5%	17.4%
Avoidance of reminders	2.79	1.55	33.6%	12.1%	16.8%	17.4%	20.1%
Feeling stunned, shocked, or dazed	2.82	1.49	28.9%	16.1%	18.1%	18.1%	18.8%
Confusion about role in life or a diminished sense of self	2.15	1.29	45.6%	18.1%	17.4%	12.8%	6.0%
Feelings of trouble accepting the loss	2.81	1.61	33.6%	14.1%	15.4%	12.1%	24.8%
Difficulties in trusting others	2.08	1.36	51.0%	16.8%	15.4%	6.7%	10.1%
Feelings of bitterness over the loss	2.50	1.54	41.6%	13.4%	14.8%	13.4%	16.8%
Difficulties in moving on	2.26	1.37	42.3%	20.1%	16.8%	10.7%	10.1%
Emotional numbness	2.16	1.33	45.6%	19.5%	16.1%	10.7%	8.1%
Feeling life is unfulfilling, empty, or meaningless	2.44	1.50	39.6%	20.1%	13.4%	10.1%	16.8%

Participants who met the criteria for PGD were defined as PGD-positive. The 149 participants were classified as a PGD-positive group (n = 33) and a PGD-negative group (n = 116). The socio-demographic and bereavement-related data of the two groups are compared in Table 1. Chi-square tests revealed significant differences between PGD-positive and PGD-negative groups in terms of the parent gender ($\chi 2$ = 4.178, p = 0.041), the subjective family economic status ($\chi 2$ = 7.42, p = 0.024), age of the parents at the time of child loss (t = −2.101, p = 0.037), and time since loss (t = 4.693, p < 0.001). It was found that PGD-positive group comprised significantly more females than PGD-negative group (75.6% *vs*. 56.0%, p = 0.041). The subjective family economic status was moderate or poor in 90.9% of the parents in PGD-positive group *vs*. 76.8% in PGD-negative group. The mean age of the parents at the time of Shidu in PGD-positive group was older than that in PGD-negative group (53.65 *vs*. 50.34 years, p = 0.037). The 'time since loss' in PGD-positive group was shorter than that in PGD-negative group (7.59 *vs*. 12.2 years, p < 0.001).

This study collected the morbidity of chronic diseases including hypertension, CHD and DM, and the number of hospital visits in the past year, finding that the overall morbidity of osteoarthrosis in PGD-positive group was significantly higher than that in PGD-negative group ($\chi 2$ = 7.177, p < 0.01) (Table 3). There was no significant difference in the number of hospital visits between the two groups (Table 3).

Table 4 shows the comorbidity of PGD in the Shidu parents, revealing that 30.9% (46/149) of the sample fulfilled the criteria for PTSD; 36.9%

Table 3. Chronic diseases and the number of hospital visits in the past year among Shidu parents with or without PGD.

	PGD-positive group		PGD-negative group			
	n = 33		n = 116			
	n	%	n	%	χ2	p-value
Chronic diseases						
Hypertension	12	36.4	42	36.2	0.00	0.987
Coronary heart disease	11	33.3	27	23.3	1.37	0.242
Diabetes mellitus	4	12.1	16	13.8	0.06	0.804
Cerebrovascular diseases	0	0.0	6	5.2	0.69	0.406
Respiratory diseases	2	6.1	7	6.0	0.00	0.996
Digestive diseases	3	9.1	5	4.3	0.41	0.524
Rheumatosis	3	9.1	5	4.3	0.41	0.524
Osteoarthrosis	10	30.3	13	11.2	7.18	0.007**
Tumour	2	6.1	9	7.8	0.00	1.000
Mental diseases	3	9.1	6	5.2	0.18	0.675
Other	2	6.1	13	11.2	0.29	0.590
Number of hospital visits						
0–2	4	12.1	34	29.3	6.59	0.086
3–5	7	21.2	16	13.8		
6–9	2	6.1	14	12.1		
≥10	20	60.6	52	44.8		
Total	33	100.0	116	100.0		

**p-value < 0.01.

Table 4. The comorbidity of PGD among Shidu parents.

	PGD-positive group		PGD-negative group			
	n = 33		n = 116			
	N	%	N	%	χ2	p-value
PTSD	30	90.9	16	13.8	71.59	0.000***
Depression	28	84.8	27	23.3	41.83	0.000***
Common Mental Health	25	75.8	25	21.6	33.86	0.000***
	M	SD	M	SD	T	p-value
PCL-C	64.61	9.60	36.51	12.68	11.80	0.000***
CES-D	33.79	14.44	9.71	9.20	11.56	0.000***
GHQ-12	6.76	3.87	2.06	2.31	8.73	0.000***

PCL-C = PTSD Checklist-Civilian Version; CES-D = Centre for Epidemiological Studies Depression Scale; GHQ-12 = the General Health Questionnaire; ***p-value < 0.001.

(55/149) for depression; 20.1% (30/149) participants had comorbidity of both PGD and PTSD; and 18.8% (28/149) had comorbidity of both PGD and depression. PGD-positive group had a higher co-occurrence rate of PTSD, depression and mental health morbidity than PGD-negative group (90.9% *vs*. 13.8%; 84.9% *vs*. 23.3%; 75.8% *vs*. 21.6%, p < 0.001). Three participants without PTSD and five participants without depression met the criteria for PGD.

We put parents' gender, education level, subjective economic evaluation, morbidity of osteoarthritis and hospital visits, gender of deceased child, time since loss into the regression model with PG symptom score as the dependent variable. The result showed that time since loss, subjective perception of poor economic situation, female gender of parents, and more hospital visits were significant predictor variables (Table 5). The model explained 22.2% variance in PGD symptoms (R^2 = 0.222, F = 4.991, P < 0.001). Time since loss had the most predictive utility, showing that shorter time since loss associated with more severe grief reactions. Subjective perception of poor economic situation emerged as the second potent predictor of PGD symptoms. Female participants had higher scores on PGD symptoms than male participants. More hospital visits was a significant associated factor as well. Other variables, such as parents' education level, gender of deceased child, and morbidity of osteoarthritis did not contribute significantly to the PGD symptom variance. Additionally, the cause of death was not included in the regression analysis, because the data were obtained from the Register of Population Statistics provided by the Health Family Planning Commission of China, the specific cause of death of the child was unclear, and it was inappropriate to ask Shidu parents this question during the interview. Also, religious belief was excluded in the regression analysis, as a result of most Chinese people do not have religious belief. In our sample, 81.9% reported to have no religion.

Table 5. Regression analyses with PG symptom score as the dependent variable.

Variables	PG symptom score					
	B	95% CI		t	p-value	β
Female vs. Male	4.59	0.7	8.48	2.33	0.021*	0.19
Moderate vs. Wealthy and extremely wealthy	1.67	−2.96	6.3	0.71	0.477	0.07
Poor and extremely poverty vs. Wealthy and extremely wealthy	7.64	0.84	14.45	2.22	0.028*	0.2
Lower than high school vs. High school and above	2.93	−0.69	6.55	1.6	0.112	0.12
Lost son vs. lost daughter	0.98	−2.7	4.65	0.52	0.601	0.04
Time since loss(years)	−0.47	−0.72	−0.21	−3.65	0.000***	−0.27
Morbidity of osteoarthritis	2.34	−3.01	7.69	0.87	0.388	0.07
Number of hospital visits ≥10 vs. < 10	4.11	0.24	7.99	2.1	0.038*	0.17

B = unstandardized beta; $β$ = standardized regression weight; * p-value < 0.05, *** p-value < 0.001.

4. Discussion

The results demonstrate that a significant proportion (22.2%) of the interviewed Shidu parents met the criteria for PGD with a mean duration of 7.59 years after the grief-related loss. The prevalence of PGD in these Chinese Shidu parents is much higher than that in the general Chinese population (22.2% vs. 1.8% and 13.9%) (He et al., 2014; Li & Prigerson, 2016), and twice as high as that in parents who lost their children to cancer (22.2% vs. 10.3%) according to a study abroad (McCarthy et al., 2010). These differences may be attributed to the disparity of subjects and culture context. On one hand, in the present study, all the subjects lost their only-child and unable to conceive another child. As prior studies reported, not having remaining children is one of the most significant predictors of psychosocial distress, grief and impaired health for bereaved parents (Dyregrov, Nordanger, & Dyregrov, 2003, Wijngaards-de Meij et al., 2005), parents who lost an only child seem to have higher risk for more severe grief compared to the parents who lost one child among others (Buchi et al., 2007; McSpedden, Mullan, Sharpe, Breen, & Lobb, 2017; Meert et al., 2011; Tseng, Cheng, Chen, Yang, & Cheng, 2017; Wijngaards-de Meij et al., 2005; Xu, Herrman, Bentley, Tsutsumi, & Fisher, 2014); one would predict that the death of the only-child is one of the most traumatic events for Chinese parents. On the other hand, the difference could be attributed to the relatively older age of our participants (50 years at the time of Shidu and 60 years at the time of the present investigation). Lundorff et al. reported an age-effect suggesting that older age could be associated with higher prevalence of PGD (Lundorff et al., 2017). The relatively older age of deceased children (mean age was above 20 years old) may also contribute to the high PGD rate of this study. In addition, Chinese traditional culture (e.g. family continuation) is likely to influence the grief beliefs, which, in turn, could impede Shidu parents' recovery (Shi et al., 2019; Yi et al., 2018) and lead to a high incidence of PGD. There is a Chinese saying that 'There are three unfilial acts, bearing no descendant is by far the most unforgivable'. Thus, the death of the only-child may be more devastating for Shidu parents in the context of Chinese culture. The present study has lower PGD rate than 3 studies among Western bereaved parents, i.e. 30%, 40%, and 50%, respectively (Keesee et al., 2008, Goldstein, Lederman, et al., 2018, Goldstein, Petty, et al., 2018), and the study among South Korean parents of the ferry accident (94%) (Huh et al., 2017). Their higher rates may pertain to the overrepresentation of women compared to men (4:1 in Keesee's study and samples in Goldstein's studies were all females), as some indication that mothers face greater difficulties adapting to the death of a child than fathers (Sidmore, 1999, Xiu et al., 2016), or mothers have been found to rate their grief feelings higher than fathers following the death of the infant (Lang, 1993). For the study on South Korean parents of the ferry accident, their time since loss is much shorter (18 months) than our samples (mean duration of 7.59 years), and shorter time since loss is significantly associated with more severe grief reactions as many previous studies verified.

4.1. Strengths and clinical implications

The present study provides strong evidence that Shidu parents should be exclusively screened for PGD and timely provided with effective intervention, given the high prevalence rates. However, individuals in this group are prone to isolate themselves and difficult to access, probably due to a) cultural stigma related to not passing on the family name or carrying on the family line (Shi et al., 2019; Yin, Shang, et al., 2018), b) social stigma: children appear to be the main topic of conversations among peers, Shidu parents are left out of everyday conversations (Zheng et al., 2017), c) public stigma caused by prolonged grief, i.e. stigma of having PGD (Eisma, 2018; Eisma, Te Riele, Overgaauw, & Doering, 2019). None of this study's participants had ever sought help from mental health services after the bereavement. Future studies are required to investigate public stigma of PGD and its impact on people seeking mental health services for PGD in the Chinese population. The authors of

this study agree that the advantage of including the PGD diagnosis into ICD-11 outweighs the disadvantage (Dietl, Wagner, & Fydrich, 2018), from the clinicians point of view it can at least help with accurate detection and treatment of the psychopathology resulting from grief. However, as the concept and formulation of the ICD-11 guidelines concerning PGD are based on the Western background, whether the operational classification is readily acceptable in Chinese culture needs more input from Chinese clinicians.

In addition, this study attempted to describe the full spectrum of grief symptoms experienced by Shidu parents in China. The data show that longing or yearning is the most prominent separation distress, which is consistent with other studies that the core symptom of PGD is persistent yearning and preoccupation with the deceased (He et al., 2014; Prigerson et al., 2009; Schaal et al., 2010). Avoidance behaviour, negative cognition of trouble in accepting the fact of loss and the stunned or shocked emotion were the most frequently reported phenomena among Shidu parents, and might impede successful mourning. All these findings are clinically meaningful, and the commonly reported symptoms can be the main targets for developing psychological intervention strategies for Shidu parents. Exposure therapy with cognitive behaviour therapies (CBT) by addressing the avoidance behaviour and grief pain has proved to be an effective treatment by many randomized clinical trials (Bryant et al., 2014, 2017; Rosner, Bartl, Pfoh, Kotoucova, & Hagl, 2015; Rosner, Pfoh, Kotoucova, & Hagl, 2014). For the above reasons, it should be promoted in future studies on the comprehensive support strategies for Shidu parents.

So far few studies have rigorously examined factors predicting the development of PGD among Shidu population. The results of the present study indicate that female parents, the subjective non-wealthy family economic status, older age of the parents at the time of child loss, and a short duration since child loss are associated with higher symptomatic severity of PGD. This study's regression analysis indicated that fewer years since loss, subjective perception of poor economic situation, and female gender were prominent risk factors for the development of PGD, which is compatible with the findings of many prior studies on the duration since child loss (Coelho et al., 2015; Heeke et al., 2015; Keesee et al., 2008; McCarthy et al., 2010; Schaal et al., 2010), family's economic status (McCarthy et al., 2010; Yi et al., 2018), and female gender (Johnsen et al., 2012; Lai et al., 2015; Morina et al., 2010; Neria et al., 2007; Schaal et al., 2014; Xiu et al., 2016). These risk factors are particularly valuable in screening for PGD in Shidu parents in the future. Especially the subjective

perception of poor economic situation should not be overlooked. In Chinese culture, 'Bring up children with the aim to guard against troubles in one's late years' is a traditional perception, and the proverb 'Raise children to prevent old age; accumulate grain to prevent hunger' was recorded in a book of Song Dynasty more than 1000 years ago. The Chinese elderly are mainly cared for by their children at home instead of living-in nursing homes. The present study showed that the mean age of the deceased children was above 20 years old, and by then their parents had already spent lots of savings on their children. All these factors bankrupted the Shidu parents not only spiritually and socially but economically. Other studies also reported that economic hardships could predict and aggravate grief (McCarthy et al., 2010; Yi et al., 2018).

The authors' previous study (Yin, Shang, et al., 2018) found that the physical status of Shidu parents was much worse than that of the control group, as represented by higher morbidity of chronic diseases and more hospital visits. Many studies (Prigerson et al., 2009) have shown that PGD is associated with an elevated risk of poor physical health and high level of functional impairment. The present study showed the occurrence of osteoarthrosis in PGD-positive group was significantly higher than that in PGD-negative group (7.18% vs. 30.3%, p < 0.01), knowing that osteoarthrosis would limit personal daily activities and aggravates social isolation. The regression analysis also indicated that more hospital visits is significantly related to the increased risk of post-loss grief, supports our hypothesis that poor physical health (more hospital visits indicates poor health) may have an influence on bereaved parents' grief severity. However, as a cross-sectional study, this study cannot reach a conclusion about the causal relationship between PGD and physical health (morbidity of osteoarthritis, number of hospital visits). Future longitudinal study can examine whether poor physical health impedes bereaved parents from recovery or vice versa.

The present study failed to find a significant correlation between the age of the deceased child and PGD, probably because of the relatively small sample size or because most of the deceased children in this study were already adults, and the small age difference could not produce statistical significance.

This is the first study to examine the comorbidity of PGD, PTSD and depression in the Shidu population. The findings are in line with other studies (Heeke et al., 2015) and provide additional evidence that bereaved participants with PGD were more likely to suffer from PTSD or depression than bereaved participants without PGD. This study found that Shidu parents had a high rate of PTSD and depression, but three participants without PTSD and five

participants without depression met the criteria for PGD. This raises the concern that some cases of PGD among bereaved Shidu parents would be left out if the mental health check-ups only include assessments of PTSD or depression. These findings indicate that PGD as well as PTSD and depression should carry equal weight in the assessment and treatment of bereaved Shidu parents. From another point of view, considering the high comorbidity among PGD, PTSD and depression, the vast majority of Shidu parents with PGD would be identified and treated based on other diagnoses (PTSD or depression); however, whether the treatments for PTSD and depression would relieve PGD of Shidu parents is still a question. This aspect is important to future research.

4.2. Limitations

This study had a number of limitations. First, the participants in this study were all residents in a mega-city of China, and it is not quite clear to what extent the present findings can represent the situation in rural areas in China or people with different cultural backgrounds. Also, the relatively old age of the participants may limit the generalizability of these results to other age groups. Second, PTSD and depression were evaluated by using two screening instruments (PCL-C and CES-D) rather than using clinical assessment; therefore, the common bias and subjective influences could not be avoided. Third, the cross-sectional nature of the design limits causal inferences concerning the precise relations between the predictors, and the cross-section study does not allow examination of the stability of factors over time. Fourth, the small sample size may have lacked sufficient power to observe significant associations between variables. Fifth, some highly relevant potential mediators were ignored, including the presence of grandchildren or daughters-in-laws, perceived social support, practical and financial support from communities and governments and so on. These mediators should be considered in future studies. Sixth, the present study used Prigerson's suggestion (2009) to define PGD, which is slightly different from ICD-11 diagnostic criteria and seems outdated. New ICD-11 diagnostic criteria should be used in our future studies. Finally, six parents dropped out of the study, probably because of clinically impairing grief. Given the small number of these drop-out participants, the authors of this study feel confident that the sample can represent the Shidu parents within this community sample.

5. Conclusions

Despite these limitations, this study expands previous studies by including a very unique population with a non-Western culture. In more than 20% of the Shidu parents investigated, the pathological reaction to loss appears to persist at least 7-years post bereavement, which attests that unresolved grief will endure over time if not addressed timely by effective interventions. These findings support the importance of screening Shidu parents upon the loss and 6 months after loss to assess their mental health. Ideally, Shidu parents at risk of developing PGD, especially the mother, low-income and elder parents, should be identified and provided with routine bereavement counselling before PGD becomes entrenched. This study also provides evidence of the existence and clinical significance of PGD symptoms among the Chinese population and supports the inclusion of PGD into ICD-11. Although this study is within the particular context of China, the findings suggest important implications for similar parental bereavement outside China. In the context of global ageing, quite a number of elderly people may experience the death of a child during their lifetime, and a recent study suggests that PGD is a risk factor for cognitive decline in middle-aged and older persons during a 7-year follow-up period (Perez, Ikram, Direk, & Tiemeier, 2018). Data obtained from this study show that the prevalence of PGD and the PGD comorbidity with PTSD and depression among bereaved parents are relatively high, and therefore implementing assessments and interventions for the bereaved parents is an important issue that needs to be dealt with seriously.

Disclosure statement

No potential conflict of interest was reported by the authors.

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