A Novel Narrative E-Writing Intervention (NeW-I) for Parents of Children with Chronic Life-Threatening Illnesses: Protocol for an Open-Label Randomized Controlled Trial

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Abstract

Background: Conventionally, psycho-socio-spiritual interventions for parents of children with chronic life-threatening illness begin post child loss. Pre-loss interventions addressing anticipatory grief can improve holistic well-being and grief outcomes among family caregivers of dying patients. Globally, palliative care strives to holistically support patients and their caregivers at the end-of-life. However, inadequacies exist both globally and in Singapore in providing culturally sensitive psycho-socio-spiritual support to parents whose children need pediatric palliative services. Aim: A novel evidence-based Narrative e-Writing Intervention (NeW-I) is developed to address this gap. NeW-I is a strength-focused, meaning-oriented and therapist-facilitated mobile app and web-based counseling platform that aims to enhance quality of life, spiritual well-being, hope and perceived social support, and reduce depressive symptoms, caregiver burden and risk of complicated grief among parents facing their child’s chronic life-threatening illness.

Methods: The design of NeW-I is informed by an international systematic review and a Singapore-based qualitative inquiry on the lived experience of bereaved parents of children with chronic life-threatening illness. The online NeW-I platform and the relative anonymity it offers to participants is sensitive to the unique cultural needs of Asian family caregivers who are uncomfortable with emotional expression even during times of loss and separation. Together with four local pediatric palliative care providers, NeW-I is implemented in Singapore as an open-label pilot randomized controlled trial with 72 parents. Potential effectiveness of NeW-I and accessibility and feasibility of implementing and delivering the intervention are assessed. Discussion: NeW-I aspires to improve psycho-socio-spiritual well-being of parents facing their child’s chronic life-threatening illness through a structured cyber-counseling platform, thereby enhancing holistic pediatric palliative care and parental bereavement support services. Findings from this
pilot study will inform the development of a standardized NeW-I protocol and further research to evaluate the efficacy of NeW-I in Singapore and in other Asian communities around the world.

Background
There is a global increase in the number of children living with chronic life-threatening illnesses [1, 2]. In Singapore, child deaths (age < 19) due to chronic conditions increased from 204 per year to 245 per year during the 2014-2016 period [3]. In fact, congenital anomalies as well as cardiovascular and cerebrovascular diseases account for over half of all deaths in the 0-19 age range in Singapore [4]. Due to technological development in medical care, children with chronic and life-threatening illnesses can live longer, but simultaneously face the challenge of dependency on their caregivers (usually, their parents) and disability for a prolonged period of time [1]. However, not all seriously ill children and their parents receive adequate palliative support services [3]. Being a parent-caregiver for a child with a chronic life-threatening condition is a highly stressful experience [5]. In addition to the typical challenges of parenting, parent-caregivers must navigate a complex web of stressors including the practical and financial burden of caregiving [6], strained marital and social relationships [7], and neglect of other healthy children and family members [8]. Further, parent-caregivers of children with chronic life-threatening illnesses need to frequently communicate and engage with medical professionals, but such interactions can increase parents’ anxiety and distress if they are not adequately involved in making decisions regarding their child’s treatment [9, 10]. Thus, the stressors associated with caregiving and the elevated demand on resources puts parent-caregivers at increased risk of depressive symptoms, fatigue and overall poor quality of life [11].

Parental Bereavement Trajectories of Child Loss
Our research team recently conducted a qualitative systematic review of 25 high-quality research articles published between 2000 and 2017, exploring the lived experience of parental bereavement due to a child’s chronic life-threatening illness; a four-phase parental bereavement trajectory of child loss was developed, highlighting appropriate interventions that help parents identify care needs, elicit caregiving strengths, enhance death preparedness, and foster meaning-making throughout the illness trajectory in order to reduce psycho-emotional distress during end-of-life and into bereavement [12]. Our research team conducted a second study to examine the Asian experience of parental bereavement, via meaning-oriented strength-focused interviews with 6 couples, 13 mothers, 4 fathers and 2 primary parental figures (N = 25 parental units) who lost their child to chronic life-threatening illness in Singapore [13, 14]. Grounded theory analysis revealed 7 themes and 25 sub-themes that were organized into a Trauma to Transformation Model of Parental Bereavement (See Figure 1). This culture-specific model shows the milestones of how Asian bereaved parents journeyed through their child’s life-threatening illness and eventual death, describing the ritualistic actions that aided them to better cope with their loss, to regain control over their lives, to sustain a continuing bond with their deceased child, to move forward with and ultimately transcend their grief through meaning reconstruction [13, 14].

These findings echo previous literature [15–18] in positing that parents facing potential child loss could benefit from psychosocial and therapeutic interventions as early as prognosis and throughout the illness trajectory, which could ease the transition from caregiving, through mortality and bereavement thus mitigating adverse grief outcomes (See Table 1). However, most supportive interventions for parents caring for children with chronic life-threatening illnesses only occur after bereavement [19–21], and a recent systematic review found negligible evidence to support their effectiveness [22]. As such,
there is a need to develop a pre-loss intervention to augment pediatric palliative care and parental bereavement support service - one that empowers parents to reflect on their caregiving experiences, explore and identify resources that could help them better cope with the challenges of caregiving, and support their child to live a meaningful life despite a chronic life-threatening illness.

Table 1: Clinical implications of findings from qualitative study on the lived experience of parental bereavement in Asia

| Findings from qualitative study on the lived experience of parental bereavement in Asia | Implications for clinical work |
|---|---|
| Facing their child’s impending mortality is a difficult experience which can isolate parents. Societal attitudes towards illness can make it difficult for parents to engage with their previous social networks. | Support parents in gaining a greater sense of control over their lives and strengthening resilience during the period of caregiving through empathic support and psychoeducational resources for self-care and healthy coping. |
| Parents seek to understand the medical terminology associated with their child’s illness and prognosis of the condition, so that they can evaluate potential risks and benefits of treatment procedures and make informed care decisions. | Empower parents to provide the best possible care through exploration of resources for seeking information on illness and caregiving. |
| Parents desire to give their children a chance to rise above the difficulties brought on by the illness and display strength to help their child live as fully as possible. | Facilitate meaningful family experiences that allow their children to move away from the drudgery of illness and caregiving and focus on building parent-child memories. |
| Asian parents tend to have a collaborative approach to caregiving for their sick child. They often relied on family members, relatives and other parents of sick children for support. | Explore sources of support which participants have within their close social network and how they can be harnessed in care provision. |

**Elements of a Pre-Loss Intervention for Parents Facing their Child’s Chronic Life-Threatening Illness**

In developing a pre-loss intervention that could meaningfully impact families throughout their child’s illness trajectory and leading to the final days of their child’s life, a number of important therapeutic elements need to be considered and incorporated. First, *Anticipatory Grief*, defined as the process of mourning the loss of a loved one prior to actual loss that enables caregivers to experience and adjust to various grief responses, must be central to such an intervention [17, 23]. Anticipatory grief can smoothen the
process of coping with death, since the individual has scope to come to terms with the loss in advance [24]. Studies have found that strength-based end-of-life interventions with elements that address anticipatory grief can improve adult patients’ quality of life and mitigate poor bereavement outcomes among family caregivers [25]. It is therefore possible that an anticipatory grief-based psychotherapeutic intervention for parents facing their child’s chronic life-threatening illness could aid parents in understanding and regulating emotions, enhancing death preparedness, and thereby building resilience. Second, it would be useful for a pre-loss intervention for parents facing their child’s chronic life-threatening illness to adopt a Meaning-Reconstruction Approach [26–28], with each individual actively constructing a phenomenological world of their experiences in relation to various familial and socio-cultural contexts, and supporting their sense of loss and grief accordingly. Such a meaning-reconstruction approach empowers grievers to choose whether to direct their attention to the loss and process turbulent feelings, or to focus on practical adjustments to re-engage with their everyday life. Third, a pre-loss intervention would benefit from a Narrative Approach [29, 30], which could help individuals connect with emotions that are challenging to accept, generate new meaningful stories about life and loss, and restructure negative emotional appraisal of situations such as end-of-life caregiving into more positive ones, thereby generating a sense of hope.

Two effective examples of applying the meaning-reconstruction approach and the narrative approach in supporting holistic end-of-life care are Dignity Therapy [25, 31, 32] and Family Dignity Intervention [33], both of which are evidence-based psychotherapies that address the physical, psychosocial and existential issues pertaining to one’s dignity at end-of-life. Specifically, Family dignity intervention is designed to support the collective experience of grief and loss for Asian families facing mortality, and it could add great
value to a pre-loss intervention for parents of children with chronic life-threatening illness. In practice, family dignity intervention comprises a meaning-focused interview with the patient-and-family caregiver dyad that fosters the expression of appreciation and emotional connection through the retelling of important life narratives; the interview is recorded, transcribed and edited into a legacy document, and returned to the dyad for sharing with their loved ones for healing and remembrance.

Finally, a pre-loss intervention for parents facing their child’s chronic life-threatening illness must be mindful of the caregiving responsibilities and limitations that serve as barriers for parents to engage in sit-and-talk therapy [12]. It is possible that an internet-based platform which is cost-effective and time-efficient [34] can deliver psychotherapy to such parents. Such therapist-facilitated e-platforms are increasingly used for brief and effective psychotherapy for a range of conditions including depression, anxiety, and stress [35–38]. Moreover, when internet-based platforms use writing as the modality for emotional expression and reflection, efficacy is superior, as compared to audio/video mediums [39]. Finally, the anonymity of an e-writing channel can encourage greater willingness to self-disclose [40, 41].

Present Study

Globally, pediatric palliative care interventions predominantly emphasize the stages of grief and psychological tasks that grieving parents must accomplish after their child’s death [22], and in Singapore, there is no known empirically-tested intervention to provide psycho-emotional support and psychoeducational resources to parents of children with chronic life-threatening illness. Given that Singapore is a leading nation in digital readiness, and smartphone utilization for communication is ingrained into the everyday life of its people [42], internet-based solutions can be vital in enhancing pediatric palliative care and parental bereavement support services. Further, keeping in view that
Asian family caregivers can be uncomfortable with explicit emotional expression even during times of loss and separation [43], an internet-based platform and the relative anonymity it offers to participants [41] would be appropriate for an Asian population. The present research team has integrated the aforementioned elements necessary for a pre-loss intervention for parents of children with chronic life-threatening illness and conceived Narrative e-Writing Intervention (NeW-I) to address the gap in pediatric palliative care delivery and research in the local context. Specifically, NeW-I is a novel internet-based, therapist-facilitated, strength-focused, and meaning-oriented intervention designed to provide direct service to parents facing their child’s chronic life-threatening illness.

Methodology

The NeW-I study protocol is guided by the SPIRIT 2013 checklist for reporting of clinical trial protocols [44].

Aims, Objectives and Hypothesis

The development and evaluation of NeW-I is guided by the Medical Research Council Framework for the Development and Evaluation of Complex Interventions which is widely recognized in the design and evaluation of complex interventions to improve health outcomes [45, 46]. NeW-I is also inspired by the meaning-reconstruction model [47], the narrative approach to anticipatory grief [48], dignity therapy [31, 32], family dignity intervention for holistic end-of-life care [33], and the findings of a recent investigation on Asian parental bereavement experience of child loss by our research team [13, 14]. This study has four objectives, which are to:

1. Develop a pilot study protocol for a culture-specific and meaning-oriented Narrative e-Writing Intervention (NeW-I) for anticipatory grief and bereavement support for Asian parents facing their child’s chronic life-threatening illness and impending
2. Evaluate the efficacy of NeW-I in enhancing quality of life, spiritual well-being, hope and perceived social support, and decreasing depressive symptoms, caregiver burden and risk of complicated grief among participants;

3. Assess the acceptability and feasibility of implementing NeW-I among Asian parents of children with chronic life-threatening illness in Singapore; and

4. Develop a standardized protocol for further empirical research to test the effectiveness, acceptability and feasibility of NeW-I in Singapore and in other Asian communities around the world.

It is hypothesized that intervention participants who successfully complete NeW-I will experience enhanced quality of life, spiritual well-being, sense of hope and perceived social support, and decreased depressive symptoms, subjective caregiver burden and risk of complicated grief as compared to control participants. It is also hypothesized that NeW-I is deemed an accessible and user-friendly service by participants.

**Study Design**

This study adopts an open-label randomized controlled trial design comprising two groups: (1) an intervention group (structured NeW-I protocol) and (2) a control group (journaling activity unrelated to their child’s illness).

**Sample**

The sample comprises 72 individual parents of varying ethnicity in Singapore (N=72). Where both parents of a given child participate in the study, their group allocation is randomly determined, and assessments are completed by both parents independently. To be eligible for participation in this study, the individual must be a parent whose child has been diagnosed with a chronic life-threatening illness and has a prognosis of more than 3 months at the time of enrollment. For the purpose of this study, a ‘child’ has been defined
as children and young persons between the ages of 0-19 years [49]. The individual must be able to speak, read, and write in English and provide informed consent. Individuals are excluded from this study if they are suffering from severe depressive symptoms and psychological distress as identified by two screening tools. Specifically, to protect participants’ well-being during the pilot testing of NeW-I, those who do not meet the stated cut-off scores of Patient Health Questionnaire-9 (>19) and Kessler Psychological Distress Scale (>29) are excluded as formal treatment and therapy would be more beneficial [50, 51]. Additionally, if participants cease to meet the inclusion criteria during the study (such as, due to their child’s untimely death), they are excluded from the study and provided alternative resources for psychosocial support. However, the data that has been collected until the time of their participation will be kept and analyzed so that a complete and comprehensive evaluation of the study is possible.

Sample Size Calculation

For a main trial designed with 90% power and two-sided 5% significance, a pilot sample size of 25 per arm is needed to detect a small effect size of 0.2 in the primary outcome measure [52]. A meta-analysis suggested that many high-quality psychotherapy studies (as defined by proficiently trained therapist, treatment integrity, N ≥ 50) for the treatment of depression have a mean effect size of 0.22 [53]. Allowing for an attrition rate of 30% at follow-up (a larger estimate due to end-of-life context), the sample size must be inflated by a factor of 1 / (1-0.3) = 1.43. Therefore, the minimum sample size required for this study is 72, or 36 in each group.

Study Sites and Recruitment Procedures

Purposive sampling is adopted to achieve the target sample size of 72. Potential participants are identified and contacted by the collaborating organization (leading pediatric palliative care providers in Singapore including KK Women’s and Children’s
Hospital, Club Rainbow Singapore, Muscular Dystrophy Association Singapore and Rare Disorders Society Singapore) to introduce the study to their beneficiaries. Such a sampling strategy allows recruitment of participants whose children suffer from a wide range of chronic life-threatening conditions, thus allowing for maximum variation in the sampling. If verbal consent is obtained from potential participants, their contact details are passed to the research team at Nanyang Technological University, who subsequently establish telephone contact, explain study procedures and introduce the NeW-I online platform. All personal information pertaining to potential participants is kept confidential and only the responsible researchers have access to such information.

Open recruitment is also carried out, so that all parents of children with chronic life-threatening illness have equal opportunity to participate in a potentially beneficial study, and to examine the feasibility of implementing this free and easily accessible intervention in the community. Posters are placed in strategic locations across Singapore providing information regarding the study. When interested participants contact the research team, study procedures are explained, the NeW-I platform is introduced, and registration information is provided.

**App and Intervention Procedure**

The NeW-I therapist-facilitated online platform comprises a mobile app and a website (both of which have the same content and functionality). When participants initially log on to the app or website, they are directed to a study participation and informed consent page that provides details about study procedures, rights of research participants and protection of confidentiality. After participants endorse this online informed consent form on the NeW-I platform, they are directed to a demographic information page. This is followed by a screening page where participants complete the PHQ-9 and the K-10. Those who pass the screening assessments receive confirmation of study participation and are
requested to wait for a phone call from the research team. Those who do not pass the screening assessments are thanked for their time and provided resources for psychosocial support. Individuals who pass the screening assessments receive a phone call from the NeW-I team as a means of identity-checking. Following this, participants complete baseline measures (T1 assessment). This is followed by random allocation of participants to either the intervention or the control group which is done via the NeW-I platform by using computer-generated random numbers. Participants are then directed to the first writing session. Participants may choose to begin the first writing session immediately, or delay for a maximum of 3 days. The day on which participants begin first the writing session is day 1 of week 1. Participants who do not begin their first writing session within 3 days will be dropped from the study. However, a concession period of 7 days is given to participants who have a genuine reason for their inability to stick to the protocol (such as unexpected changes in their child’s health condition) and have expressed a keen interest to participate in the study. A similar concession period of 7 days is also maintained for participants who are unable to stick to the study protocol in weeks 2, 3 and 4 (such that each participant can only be given one concession during the entire duration of their study participation). Detailed study procedures are described in detail in Figure 2.

NeW-I is delivered by trained therapists in the research team who are experts in death education and grief counselling and have the clinical competence to work with family caregivers in pediatric palliative settings. All team members have successfully completed research integrity modules under the provisions of Nanyang Technological University’s Institutional Review Board and adhere to the Board’s guidelines for safeguarding participant’s identity and confidentiality.

**Intervention and Control Group**

Both intervention and control group participants follow the procedures described in Figure
2. There are 4 weekly sessions of writing. A template is provided to ensure that participants’ writings tie in with the session objectives. To improve participants’ adherence to the study protocol, they receive an automated notification on their phone app and email each time a fresh writing session becomes available to them. Participants are assured of anonymity and confidentiality of their writing to encourage open and honest self-expression. The structured writing for each session requires 15 to 30 minutes, since exposure and time to process ideas through written disclosure over at least 3 sessions of 15 minutes each can produce effective outcomes [54].

For intervention group participants, each weekly session has a unique objective (see Table 2). Briefly, in week 1, participants reflect on the demands of caring for a child with chronic life-threatening illness, and the means to cope with these challenges. In week 2, participants consider avenues where they can seek more information about their child’s illness and resources for caregiving. In week 3, participants examine the sources of support which they have within their network of family and friends. In week 4, participants explore how they (and their children) can rise above illness-related challenges and live their lives as fully as possible.

**Table 2: Content and questions for reflective writing for intervention group**
| Objective                                                                 | Week 1                                                                 | Week 2                                                                 | Week 3                                                                 | Week 4                                                                 |
|--------------------------------------------------------------------------|------------------------------------------------------------------------|------------------------------------------------------------------------|------------------------------------------------------------------------|------------------------------------------------------------------------|
| To provide participants with a platform to reflect on the emotional,    | To explore avenues where participants can seek more information about  | To explore the sources of support which participants have within their  | To explore participant children's illness-related challenges lives as   |                                                                        |
| practical and financial demands of caring for a child with chronic      | their child's illness and resources for caregiving.                    | close network of family and friends.                                    | fully possible.                                                        |                                                                        |
| life-threatening illness, and the means to cope with these challenges.   |                                                                        |                                                                        |                                                                        |                                                                        |
| Questions for reflective writing                                         | 1) How satisfied are you with the knowledge and information that you   | 1) Tell us about some people who have been helpful or supportive in     | 1) Tell us v best about What qual you most about who he/ her.           |                                                                        |
|                                                                          | have about your child’s condition?                                      | your caregiving journey.                                               |                                                                        |                                                                        |
|                                                                          | What has been helpful in providing you with the knowledge and         | How have they helped you to cope during difficult times?               |                                                                        |                                                                        |
|                                                                          | information?                                                            |                                                                        |                                                                        |                                                                        |
|                                                                          | 2) What are some forms of support that have been helpful for you in   | 2) On a scale of 1 to 10, with 1 being “Not at All” and 10 being “Very  |                                                                        |                                                                        |
|                                                                          | providing quality care to your child?                                   | Much”, how satisfied are you with your spousal relationship? (Please    |                                                                        |                                                                        |
|                                                                          | How were these forms of support helpful?                               | omit this question if it does not apply to you.)                      |                                                                        |                                                                        |
|                                                                          | 3) What would help you to feel more competent as your child’s         | What might make that score a little higher?                            |                                                                        |                                                                        |
|                                                                          | caregiver?                                                              |                                                                        |                                                                        |                                                                        |
|                                                                          | What is one thing you could do to make that difference?               |                                                                        |                                                                        |                                                                        |
| Counselling goals                                                        | 1) To affirm the strengths that have helped participants to survive    | 1) To acknowledge participants’ efforts to seek power and control over  | 1) To assist (and their children) in building meaningful and cherished   |                                                                        |
|                                                                          | and thrive.                                                            | their seemingly uncontrollable lives through illness literacy.         | memories through reflecting on achievements and fulfillment of dreams. |                                                                        |
|                                                                          | 2) To provide psychoeducation about local social welfare organizations  | 2) To provide psychoeducation about sources for seeking more           | 2) To examine which participants’ sharing from sessions 1, 2 and 3 by   |                                                                        |
|                                                                          | that can provide them with support.                                    | information about their child’s illness, treatment options and         | taking the semantic content as it is but providing an alternative        |                                                                        |
|                                                                          |                                                                        | resources for caregiving.                                              | viewpoint of perceiving the situation.                                 |                                                                        |
|                                                                          |                                                                        |                                                                        |                                                                        |                                                                        |

For control group participants, the objective is consistent across the 4 weeks, that is, participants engage in weekly writing sessions that are unrelated to their child’s illness. This allows participants to experience the therapeutic benefits of narrative writing. (see Table 3).
| Objective                                                                 | Week 1                                                                 | Week 2                                                                 | Week 3                                                                 | Week 4                                                                 |
|--------------------------------------------------------------------------|------------------------------------------------------------------------|------------------------------------------------------------------------|------------------------------------------------------------------------|------------------------------------------------------------------------|
| To engage participants in a weekly writing session that is unrelated to   | This week, we’d love to know a little bit about you. Tell us what an    | This week, we’d like to know about what has the past week been like     | This week, we’d like to know what is the biggest challenge (e.g.        | This week, ‘know what most comf c now (e.g. fa relationship hobbies ac   |
| their child’s illness and to experience the therapeutic benefits of       | average day in your life looks like. Feel free to share with us any and| for you? Feel free to share with us in as much detail as you like!     | emotional, financial, practical etc.) that you are facing right now?   | activities et about what thing comf you?                                |
| narrative writing                                                        | every detail that you find comfortable to talk about!                  |                                                                        | Do feel free to add on anything else about this challenge that you    |                                                                        |
| Question for reflective writing                                          |                                                                        |                                                                        | would like us to know!                                                |                                                                        |
| This week, we'd love to know a little bit about you. Tell us what an     |                                                                        |                                                                        |                                                                        |                                                                        |
| average day in your life looks like. Feel free to share with us in as   |                                                                        |                                                                        |                                                                        |                                                                        |
| much detail as you like!                                                |                                                                        |                                                                        |                                                                        |                                                                        |

**Evaluation of Outcomes**

**Quantitative assessments.** Via the NeW-I platform, both intervention and control group participants fill out a socio-demographic form at baseline and are then assessed on a battery of standardized and validated measures across 5 time-points. The primary outcome measure is participants’ quality of life, as measured by the Kemp Quality of Life Scale (KQOL) [55]. Secondary outcomes are assessed using (i) a modified version of the Functional Assessment of Chronic Illness Therapy - Spiritual Well-Being scale (FACIT-Sp-12) [56], (ii) the Herth Hope Index (HHI) [57], (iii) the Patient Health Questionnaire-9 (PHQ-9) [50], (iv) the Burden Scale for Family Caregivers-Short (BSFC-s) [58], (v) the Inventory of Social Support (ISS) [59] and (vi) a modified version of the Brief Grief Questionnaire (BGQ) [60] (see Table 4).

**Table 4: Quantitative outcome measures**
| Measure                                                                 | # of items | Rating scale / Factor(s)                                      |
|------------------------------------------------------------------------|------------|--------------------------------------------------------------|
| Kemp Quality of Life Scale                                             | 1          | 7-point Likert scale; 1 factor                               |
| Functional Assessment of Chronic Illness Therapy-Spiritual Well-Being (Adapted version) | 12         | 5-point; 3 factors: meaning, peace, faith                    |
| Herth Hope Index                                                       | 12         | 4-point; 1 factor                                            |
| Patient Health Questionnaire-9 (also serves as a screening assessment) | 9          | 4-point; 1 factor                                            |
| Burden Scale for Family Caregivers - Short Version                    | 10         | 4-point; 1 factor                                            |
| Inventory of Social Support                                            | 5          | 5-point; 1 factor                                            |
| Brief Grief Questionnaire (Adapted version)                           | 5          | 3-point; 1 factor                                            |

Assessment for both intervention and control group takes place at five time-points: baseline (T1), immediately after completion of the intervention or control protocol (T2), one month after completion of the intervention or control protocol (T3), three months after completion of the intervention or control protocol (T4), and a final follow-up six months after completion of the intervention or control protocol (T5). Participants receive an automated notification on their phone app and email each time an assessment set becomes available to them. For each completed assessment, participants receive a voucher worth 30 Singapore dollars.

**Acceptability and feasibility study.** To evaluate the acceptability and effectiveness of NeW-I, all intervention participants are invited to participate in a semi-structured interview at the completion of all intervention components at T2, which explores the impact of the intervention, aspects of the intervention found to be helpful, aspects of the intervention found to be unhelpful and how they could be improved, challenges encountered in completing the intervention, and scope for enhancing intervention usability. To assess the feasibility of implementing and delivering NeW-I, the research
team maintains an audit trail of the time needed to provide feedback to participants and restructure their narrative writing, deviations from the intervention protocol (if any), uncompleted interventions and their reasons, and NeW-I therapists’ perceptions of competence, observations of participants’ experiences and responses and difficult or deviant cases. All feedback provided to participants is vetted by at least two members of the research team for data monitoring, quality and safety assurance.

Data Analysis

Quantitative data. The statistical analysis plan for the quantitative data in this study is designed as per the CONSORT reporting guidelines [61] and recommendations made by Gamble et al for clinical trials [62]. Quantitative parameters will be presented by mean ± standard deviation or median (with inter quartile range), and categorical variables will be presented with the numbers and proportions. Internal consistency of psychometric scales will be assessed using Cronbach’s alpha. To compare the primary quantitative outcome (KQOL) and secondary quantitative outcomes (FACIT-Sp-12, HHI, PHQ-9, BSFC-s, ISS, and BGQ) between the intervention and control groups over time (T1 to T5, with T2 being the primary time-point of comparison), multilevel mixed-effect regression analysis will be used with time as random effect. All demographic and other relevant covariates measured at baseline will be adjusted in the model. Mixed models allow us to include all available data in the presence of dropouts into the analysis [63]. Individual multivariable models will be constructed for each outcome to estimate effect sizes and corresponding confidence intervals. Clinically meaningful interactions will also be checked and included in the model if p<0.1. Marginal effects will be used to predict mean outcome scores for intervention and control groups. A two-sided p-value less than 0.05 will be considered statistically significant. All analyses will be conducted using SPSS (version 22.0, IBM Corp., Armonk, NY, USA) and Stata (version 15.1; StataCorp, Lakeway Drive, College Station, Texas, USA)
The intention-to-treat principle will be followed in data analysis. However, an additional sensitivity analysis will be conducted to handle missing data in analysis, where fully conditional multiple imputation with ‘n (% missing)’ imputations and 1000 iterations using Markov Chain Monte Carlo method will be used [64]. Model summary estimates will be calculated based on Rubin’s rule [65].

**Qualitative data.** All qualitative data are stored and analyzed using the QSR NVivo computer software. Phenomenological analysis is used to obtain an in-depth and comprehensive view of the dataset from an insider perspective [66]. Unique or minority voices are elicited to illuminate counterpoints to the stated views. Throughout the data analysis process, strategies to maximize research rigor and trustworthiness are prioritized. The use of such a method of data analysis has been demonstrated in previous research involving grief therapy with bereaved parents [67].

**Discussion**

NeW-I is a first-of-its-kind internet-based, therapist-facilitated, strength-focused, and meaning-oriented intervention designed to provide direct service to parents facing their child’s chronic life-threatening illness, filling in a critical service gap in local pediatric palliative care. Through an open-label randomized control trial, the efficacy of NeW-I for improving such parents’ quality of life, spiritual well-being, hope, depressive symptoms, caregiver burden, social support and risk of complicated grief is investigated across five time-points. This study further allows for: (1) a longitudinal assessment of the mental states of 36 parents (i.e. control group) to obtain a naturalistic trajectory of anticipatory grief; (2) an evaluation of the extent to which the intervention is successful in improving 36 parents’ mental well-being (i.e. intervention group); and (3) an evaluation of the extent to which these potential positive effects are sustained over time. The findings will inform
the development of a standardized NeW-I protocol which will be disseminated via research articles and presentations. This will form the foundation of further empirical research to test the effectiveness, acceptability and feasibility of NeW-I in Singapore and in other Asian communities around the world.

The internet-based narrative writing model of NeW-I and the relative anonymity it offers to participants [41] supports the unique needs of Asian family caregivers who are uncomfortable with emotional expression even during times of loss and separation [43]. It is hoped that NeW-I is perceived by parents to be a safe platform for engaging in intimate dialogue regarding their child’s caregiving, thereby enhancing parents’ experience of their child’s illness trajectory, empowering them to harness available resources to provide the best possible care to their child, while simultaneously reducing psychosocial distress and caregiver burden. Evidence from a recent systematic review [68] suggests that the guided online intervention protocol of NeW-I is likely to be cost-effective. Additionally, the online intervention protocol is convenient-to-access, allows expression of disenfranchised emotions and promotes meaning-ascription to traumatic experiences. Finally, although the current format of NeW-I is tailored for parents facing their child’s chronic life-threatening illness, the online therapeutic protocol can be adapted to deliver psychotherapy to diverse populations.

Limitations, Future Directions And Conclusion

Presently, NeW-I can only be implemented with participants who speak, read, and write English. Singapore is a multi-cultural and multi-linguistic nation [69], and future research should expand the delivery language of NeW-I and assess its acceptability and effectiveness among different linguistic and ethnic groups in Singapore and globally. Despite this limitation, NeW-I could enhance participants’ wellness by drawing attention away from their illness narrative and instead emphasize areas that research has
demonstrated to be most meaningful at the end-of-life. Expected study outcomes can generate new knowledge to inform research and practice in paediatric palliative care and parental bereavement support locally and globally.

List Of Abbreviations

NeW-I: Narrative e-Writing Intervention
KQOL: Kemp Quality of Life Scale
FACIT-Sp-12: Functional Assessment of Chronic Illness Therapy - Spiritual Well-Being scale
HHI: Herth Hope Index
PHQ-9: Patient Health Questionnaire-9
BSFC-s: Burden Scale for Family Caregivers-Short
ISS: Inventory of Social Support
BGQ: Brief Grief Questionnaire

Declarations

Ethics approval and consent to participate

This study has been approved by the Institutional Review Board of Nanyang Technological University Singapore (IRB-2018-07-009). Online endorsed informed consent is obtained from all participants before study participation. Participants’ confidentiality, safety from unintended outcomes and right to withdraw without any adverse consequences is safeguarded under the ethical provisions of HBRA studies reviewed by the Institutional Review Board of Nanyang Technological University Singapore. Any deviations from or changes to the study protocol will be promptly reported to the Institutional Review Board of Nanyang Technological University Singapore and formally implemented after seeking approval from the Board.

Consent for publication
Not Applicable

**Availability of data and material**
No additional data is available.

**Competing interests**
The authors declare that they have no competing interests.

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**Authors' contributions**
AHYH and OD conceived and designed the study, obtained funding, and drafted the manuscript. OD, GTH, THBT and CLX participated in designing the study and operationalizing procedures. GTH also contributed to training and skills development. LBA, JC, RMHH, CYM and SG helped in study planning and study execution. All authors have made substantial contribution to the development and editing of the manuscript and have approved the final version.

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**Access to data**
Information collected during this study will be kept confidential. Only the research team will have access to participants’ personal data. For any publication arising from this study, only aggregated research data without identifiable personal details will be used.

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Figures
Figure 1

Trauma to Transformation Model of Parental Bereavement in Asia
| ENROLMENT |
|-----------|
| Participants register on NeW-I platform |
| Online informed consent form |
| Screening assessment |
| Confirmation of study participation (if eligible) |
| Random group allocation (via NeW-I app) |
| Baseline assessment of outcome variables (T1) |

| INTERVENTION GROUP | CONTROL GROUP |
|--------------------|--------------|
| **Weeks 1-4** | **Weeks 1-4** |
| *Once between days 1-3:* Participants engage in a 15-30-minute structured writing session once a week for 4 consecutive weeks (to be completed in 1 seating). | *Once between days 1-3:* Participants engage in a 15-30-minute semi-structured writing session once a week for 4 consecutive weeks (to be completed in 1 seating). |
| *Once between days 4-6:* Participants receive a written empathetic response from the NeW-I therapist. | *Once between days 4-6:* Participants receive a written empathetic response from the NeW-I therapist. |

| **Week 5** | **Week 5** |
|-----------|-----------|
| Sharing of legacy document, option of revising legacy document Phone call with NeW-I therapist for closure of therapy Prompted to complete T2 assessment | Sharing of consolidated writing over the past 4 weeks Prompted to complete T2 assessment |

| POST-STUDY FOLLOW-UP |
|----------------------|
| At 1 month: Prompted to complete T3 assessment At 3 months: Prompted to complete T4 assessment At 6 months: Prompted to complete T5 assessment |

Figure 2

Description of NeW-I study procedures

Supplementary Files

This is a list of supplementary files associated with the primary manuscript. Click to
download.
SPIRIT2013-Checklist_NeW-I.pdf