BMJ Open

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Journal:	*BMJ Open*
Manuscript ID	bmjopen-2018-024278
Article Type:	Protocol
Date Submitted by the Author:	18-May-2018
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Keywords:	Perinatal Death, Grief, Cohort Study, Life Change Events, Quality of health care

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Protocol for a Danish longitudinal follow-up study unfolding life and grief after the death of a child during pregnancy from gestational week 14, during birth or in the first 4 weeks of life

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Word count: 3,429

Abstract

Introduction: After the death of a child during pregnancy, birth or in the neonatal period, parents often experience feelings of guilt, disenfranchisement, feelings of betrayal by one's own body and envy of others. Such bereavement results in high rates of distress: psychologically, emotionally, physiologically and existentially. We collect data in a national, longitudinal cohort study to assess grief in mothers and their partners after the death of a child during pregnancy, birth, or in the neonatal period. Our aim is to achieve a general description of grief, emotional health, and existential values after pregnancy or perinatal death in a Danish population.

Methods and analysis: The cohort comprises mothers and their partners in Denmark who lose a child during pregnancy from 14 weeks gestation, during birth or in the neonatal period (4 weeks post partum). We started data collection in 2015 and plan to continue until 2024. The aim is to include 5,000 participants by 2024, generating the largest cohort in the field till date. Parents are invited to participate at time of hospital discharge or via the Patient Associations homepage. Socio-demographic and obstetric variables are collected. Validated psychometric measures covering attachment, continuing bonds, posttraumatic stress, prolonged grief, perinatal grief and existential values were chosen to reach our aim.

Ethics and dissemination: We use web-based questionnaires distributed at three specific time points during the first 13 months after the loss. The study was approved by The Danish National Data Protection Agency (No. 18/15684, October 7, 2014).

Strengths and limitations

- Population based longitudinal study targeting at 5,000 participants by 2024 (by January 2018, 300 completed questionnaires and ongoing)
- Unfolding grief and emotional experiences in the first 13 months after the loss by multiple validated, self-administered questionnaires
- Multi-professional approach including psychologist, midwives, perinatal epidemiologists and anthropologists
- A response rate around 50%

Introduction

When we lose a person we love, we grieve. Grief is a simultaneously universal phenomenon and an entirely individual experience. Yet, grief is also a cultural phenomenon, influenced by alternating normativity and beliefs over time (Kofod, 2017). In contemporary Western countries, grief and suffering are increasingly embedded in medical and psychiatric paradigms (Cacciatore & Ruby, 2015; Lacasse & Cacciatore, 2014). For example, the World Health Organization (WHO, 2016) is preparing criteria for a new diagnosis termed Prolonged Grief Disorder (PGD), anticipated to be introduced in the diagnostic manuals for mental disorders in 2018 (WHO, 2016). Discussions about how to define pathological grief are actualized both in professional settings and the broader population (Maccallum, Malgaroli, & Bonanno, 2017; Maciejewski, Maercker, Boelen, & Prigerson, 2016; Politikken, 2016; Thieleman & Cacciatore, 2013). There is a general agreement that the majority of bereaved individuals eventually, and without professional interventions, will arrive at a new emotional equilibrium after loss. According to Litz et al only a minority will experience PGD, suffering significant impairment in important areas of daily life functioning to a disabling degree more than six months after loss (Litz, 2014). A recent meta-analysis found a prevalence of PGD in approximately 10% in bereaved adults, however only a small fraction of the bereaved in these 14 studies included bereaved parents (Lundorff, Holmgren, Zachariae, Farver-Vestergaard, & O'Connor, 2017).

Thus, it is not clear how well these findings apply to the grieving process among parents after the death of a baby. Their grieving process might differ from grief processes in general, and a larger proportion may experience the symptoms of PGD. Put differently, a longer period of intense grief may be the normal response for parents grieving after the death of a baby. In this longitudinal, follow-up study we aim to assess grief among mothers and partners after the

loss of a child during pregnancy from gestational week 14, during birth or in the neonatal period (4 weeks post partum).

The nature of perinatal grief

The death of an unborn or newborn child can be a life-changing and devastating experience (Krosch & Shakespeare-Finch, 2017). A growing body of literature has assessed the nature of grief among parents who lose a child during pregnancy, birth, or in the neonatal period. A paper from 2016 presenting best practice points based on published research including 144 studies shows that the majority of studies on parental loss are from North America, followed by Great Britain, Sweden and Australia (Burden et al., 2016). These studies demonstrate that loss from miscarriage, intrauterine fetal death, termination of pregnancy due to fetal anomaly (TOPFA) or neonatal death often involves feelings of guilt, disenfranchisement, feelings of betrayal by ones body and envy of others (Burden et al., 2016; Toedter, Lasker, & Alhadeff, 1988). Parents lose the prospect of an entire life with the child and all the moments they dreamt of sharing (Michon, Balkou, Hivon, & Cyr, 2003b). Furthermore, bereaved parents of young babies who die have few mementoes of the child, none or few pictures and a very short narrative (Cacciatore & Flint, 2011). The loss has been called “invisible” and especially if the child died before or during birth family and friends might not regard the child’s identity as real. They may also be reluctant to talk about the dead child, leading to emotional isolation complicating the grief process (Burden et al., 2016; Hendrickson, 2009; Umphrey & Cacciatore, 2011). Some parents, mostly mothers, describe a loss of self-esteem (Hendrickson, 2009; Meert, Thurston, & Briller, 2005; Michon et al., 2003b; Wonch Hill, Cacciatore, Shreffler, & Pritchard, 2017). For the mother, the bodily unity with the dead child might be still another stressful element (Cacciatore, 2013) and for parents choosing TOPFA

feelings of guilt and doubt can further thwart the grief process (Korenromp et al., 2007; Maguire et al., 2015).

Outcomes of pregnancy and perinatal bereavement

The outcomes of pregnancy and perinatal bereavement are also assessed in international studies, finding high rates of psychological and emotional distress and diagnoses including; depression, general anxiety disorder, post traumatic stress disorder (PTSD), sense of failure, long-term guilt and intense grief for more than 2 years (Badenhorst & Hughes, 2007; Burden et al., 2016; Cacciatore, 2013; Michon, Balkou, Hivon, & Cyr, 2003a). Generally, measured by similar instruments mothers appear to be more afflicted than fathers (Michon et al., 2003a; Song, Floyd, Seltzer, Greenberg, & Hong, 2010; Stroebe, Schut, & Stroebe, 2007). One review of 11 studies assessed the association between type of loss and mental health and found lower or comparable levels of depression, anxiety and PTSD among mothers losing a child from TOPFA and mothers losing a child from stillbirth (Steinberg, 2011). The risk for PTSD was assessed in a systematic review of 48 studies. They found an increasing risk of PTSD related to higher gestational age at time of loss and certain socio-demographic and psychosocial characteristics predicting PTSD (Daugirdaite, van den Akker, & Purewal, 2015). In one study including 344 bereaved mothers, the PTSD risk was 7 times higher in mothers with a perinatal death than in the comparison group of mothers with a live birth (Gold, Leon, Boggs, & Sen, 2016). A review of 18 studies on self-blame, guilt, and shame among bereaved parents (including stillbirths and the loss of older children) reported a high prevalence of all three states and an association with grief intensity (Duncan & Cacciatore, 2015). Higher mortality rates from natural causes among mothers who experienced a perinatal death were found in two large population based studies established on register data (Calderon-Margalit et al., 2007; Hvidtjorn, Wu, Schendel, Thorlund Parner, & Brink Henriksen,

2015). Explorations of subsequent pregnancy following the loss suggest that some mothers are at increased risk of depression and anxiety (Blackmore et al., 2011; DeBackere, Hill, & Kavanaugh, 2008). Patient-centered compassionate care is valued by the parents (Cacciatore, 2013) but we identified no studies assessing the long term effect of the type of care provided at the hospital.

Despite the above-mentioned outcomes, some bereaved parents describe the loss as a pivotal event in a broader and more life-changing sense; grief can make an existential imprint in the bereaved leading potentially to both posttraumatic growth as well as posttraumatic stress, which is often mentioned in the literature (Cacciatore & Bushfield, 2007; Guldin, 2014; Krosch & Shakespeare-Finch, 2017; Oginska-Bulik & Kobylarczyk, 2017).

The theoretical framework

In this section we explicate the theoretical framework, which piloted the preparation of the questionnaires and the forthcoming data analyses.

In general, unexpected and traumatic loss increases the risk of impaired physical and emotional health in the bereaved, and the loss of a child in the perinatal period will most often be unexpected and traumatic (Stroebe et al., 2007).

Attachment style is shown to be related to adaption to the loss with more intense and enduring symptoms of grief and depression, complicated grief reactions, and decreased resilience in parents with insecure attachment style (on both avoidance and anxiety attachment) (Jaaniste, Coombs, Donnelly, Kelk, & Beston, 2017; Wijngaards-de Meij et al., 2007). The attachment theory was introduced by John Bowlby in the 1970s and provides a unique way to characterize individual differences in reactions to loss because it illuminates the nature of a person's relationships and adjustment in situations of separation (Wijngaards-de Meij et al.,

2007). The theory suggests different styles of attachment, developed through the early parent-child relationship, which will form the basis for responses to emotionally distressing situations such as bereavement (Bowlby, 1997).

The Dual Process Model (DPM) has become a widespread model in understanding grief in contemporary Western countries (Stroebe & Schut, 1999). The DPM of coping with bereavement emphasizes two concurrent types of stressors and coping processes: loss-orientation and restoration-orientation. It underlines that bereaved individuals often oscillate between these two processes throughout the course of bereavement, and a standstill in one of the two processes might be associated with prolonged grief (Stroebe & Schut, 1999).

In Freud's classic grief work theory, detachment from the decedent is emphasized as fundamental for adaption to the loss and this idea has influenced the attitude of bereaved individuals for nearly 100 years. This philosophy is now challenged by the continuing bonds theory (Klass, 1997). Continuing bonds has been defined as ''*the presence of an on-going inner relationship with the deceased person by the bereaved individual*'' representing diverse behaviors (Stroebe & Schut, 2005). The literature reveals contradictory findings of the role of continuing bonds in bereavement, with certain types of continuing bonds associated with both good and poor adjustment across different studies and moreover influenced by the social and cultural acceptance of the survivors continued relationship with the deceased (Root & Exline, 2014).

When a child dies at birth the natural order of life is disturbed and assumptive worldviews shatter, challenging three primary core beliefs relating to benevolence, meaningfulness of the world and worthiness of the self, and requiring a reorganization of worldviews (Janoff-Bulman, 1992; Krosch & Shakespeare-Finch, 2017). The disruption of core belief might lead to changes in philosophy of life or spiritual beliefs (Krosch & Shakespeare-Finch,

2017). This change can go in many directions and literature shows incongruent findings in how religiousness and spirituality relate to bereavement outcomes (Jaaniste et al., 2017).

The Danish setting

There are huge dissimilarities between the health care systems in Denmark and North America from where most of the studies originate. Danish health care is public and free of charge. Furthermore, there are different approaches in the way health care professionals support bereaved parents in creating a relationship with their dead child and acknowledge their grief (Burden et al., 2016; Møller, 2015). Additionally, patient centered psychosocial care is standard in Danish hospitals while prescription of psychiatric medication appears to be much more common in the U.S. (Lacasse & Cacciatore, 2014; Møller, 2015). Specifically, when we explore existential values and spiritual believes, findings from more religious countries as the U.S. have poor external validity in a secularized country as Denmark (Hvidt, Hvidtjorn, Christensen, Nielsen, & Sondergaard, 2017; Prinds, Hvidtjorn, Mogensen, Skytthe, & Hvidt, 2014; Prinds, Hvidtjorn, Skytthe, Mogensen, & Hvidt, 2016). Hence studies in a Danish context can expand our knowledge on grief after perinatal death.

Aim

In this longitudinal national follow-up study, we aim to assess grief symptoms among mothers and partners after the loss of a child during pregnancy, birth, or in the neonatal period. We aim to achieve a general description of grief, emotional health, and existential values after pregnancy or neonatal loss in a Danish population.

To achieve our purpose, we based the study on the following overall research questions:

1. How does the process of grief change in the first 13 months in bereaved parents?
2. What gender differences exist in the grief process?
3. How is attachment style associated with continuing bonds and grief?
4. Does gestational age at time of death influence grief?
5. Does the loss change existential or spiritual values or practices?

Methods and analysis

This nationwide population based cohort study comprises mothers and partners who lost a child during pregnancy after gestational week 14, during or after birth or in the neonatal period. We include miscarriages, termination of pregnancy due to fetal anomaly (TOPFA), stillbirths and neonatal deaths. In Denmark, a regional counsel can grant permission to perform TOPFA until GA week 22; stillbirth is defined as intrauterine fetal death from GA week 22. We use web-based questionnaires distributed at three specific time points in the first 13 months after the loss. Study data were collected and managed using REDCap electronic data capture tools hosted at University of Southern Denmark (Harris et al., 2009). The mother and her partner are asked to reply questionnaires individually. The data collection started in the Region of Southern Denmark in January 2016, in the Region of Central Jutland in January 2017, and in Summer 2018 we expanded the study to cover nationally and included the remaining three regions in Denmark. Data collection was permitted by The Danish Data Protection Agency until January 2025.

In the Regions of Southern Denmark and Central Jutland, the parents receive short written and verbal information from health care professionals about the study before leaving

hospital. Subsequently they receive the first e-mail from the project manager 4 to 8 weeks after the loss with comprehensive information about the study and a link to the questionnaire. In the rest of Denmark, parents are invited to participate through announcements on the homepage for the national patient organization "Landsforeningen Spædbarnsdød"; a patient organization offering free counseling to perinatally bereaved families. Via a link at the homepage, parents sign up with an e-mail address and receive comprehensive information and a questionnaire. Access to the study questionnaire is given only when the parent have consented participation. The questionnaire is sent to the parents at three time points: 4 to 8 weeks, 7, and 13 months after the loss. If not returned, each questionnaire is followed by reminders, the first one after 3 weeks and the second one after 6 weeks of granting access to the questionnaire. Due to an initial low response rate, we further introduced a verbal reminder in January 2018 via a telephone call made by a research assistant with experience in grief counselling. In the region of Southern Denmark basic information (age, date of birth, date of death, gestational age at birth, parity and type of loss) on all potential participants are registered, allowing us to conduct a dropout analysis.

The questionnaires

The survey was constructed with a combination of basic information in relation to socio-demographics and obstetric variables, state-of-the-art psychometrical testing by validated questionnaires and ad hoc questions specifically prepared for this study. We included seven psychometric scales addressing the different parts of our research questions (Table 1).

Socio-demographic variables

We included the following socio-demographic variables: age, sex of partner (to identify female partners), marital status (married, co-habiting, single), educational level (basic school (9–10 years

of education), intermediate length education (11–16 years of education) and university education (17 or more years of education), present occupation and occupation before the loss (on maternity leave, on sick leave, unemployed, at work or studying).

Obstetric and organizational variables

The following obstetrical variables were included: previous perinatal loss, parity, assisted reproduction, single- or multiple pregnancies, type of loss (missed abortion, miscarriages, TOPFA, intrauterine fetal death, death after birth), mode of delivery (vaginal vs. cesarean section), gestational age at the time of loss, admission to the neonatal intensive care unit (NICU) and age of child if death occurred after birth, and seeing and holding the dead child. Organizational variables were: hospital and type of department to which the couple were admitted, experienced quality of professional support (midwives, doctors, nurses, social worker, undertaker, religious person (chaplain, imam or other) and patient organization).

Psychometric scales

To measure attachment style we included a version of "The Experience in Close Relationships Scale – revised, short form (ECR-R)" (Fraley, Heffernan, Vicary, & Brumbaugh, 2011) modified to bereaved samples. The participants were asked to express how much they agreed or disagreed with 12 statements concerning how they feel in emotionally intimate relationships on a seven-point scale ranging from highly disagree to strongly agree. Scores for attachment related anxiety and attachment related avoidance were obtained by averaging a person's scores (0 to 6) on each of the 12 items.

We used the Perinatal Grief Scale (PGS), developed in 1988 to construct a comprehensive measure of perinatal grief to facilitate comparison among findings in the field

(Ritsher & Neugebauer, 2002; Toedter et al., 1988). The scale was constructed to address the potential disparities between grief in general and perinatal grief (Toedter et al., 1988). The PGS contains 33 statements covering dimensions as e.g. guilt, loneliness, and jealousy with an option of answering on a five-point scale ranging from highly disagree to strongly agree. The PGS has good internal consistency (alpha 0.95) (Toedter, Lasker, & Janssen, 2001). A clinical cut-off of 91 has been established for the PGS, where greater scores indicate a high level of perinatal grief (Toedter et al., 2001).

To assess the process of bereavement within the dual process model paradigm, the Inventory of Daily Widowed Life (IDWL) was used (Caserta & Lund, 2007). The IDWL was developed studying a group of widows in 2007. However, items in the scale could be adaptable to other losses and relationships with some modifications (Caserta & Lund, 2007). The inventory comprises 15 items on doings, thoughts, or feelings and the participants state how often within the last week they have been preoccupied by each task by choosing one of four categories; seldom or never, sometimes, quite often and nearly all the time.

We also included the Post Traumatic Stress Disorder Checklist (PCL-PTSD) (Blevins, Weathers, Davis, Witte, & Domino, 2015). The PCL was developed in 1990 and comprises 17 items corresponding to the PTSD symptom criteria in the Diagnostic and Statistical Manual of Mental Disorders (5th ed.) (Blevins et al., 2015). Respondents indicate how much they have been bothered by each PTSD symptom over the past month, using a 5-point scale ranging from not at all to extremely much (scores 1 to 5).

To address the question of continuing bonds, we included “The Two Track Bereavement Questionnaire on Life Following Loss” (Rubin et al., 2009). This model aims to devote

balanced attention to two domains of the bereavement experience: the nature of biopsychosocial functioning and the nature of the ongoing relationship to the deceased.

In order to assess the likelihood of symptoms related to the diagnosis of PGD within this cohort, we incorporated Prolonged Grief Disorder-13 (Prigerson et al., 2009). The scale includes 13 items related to feelings, thoughts and doings. High scores within the specific items associated with severe functional impairment fulfill the criterion for prolonged grief disorder.

We also included questions from "The European Value Survey" (Survey, 2006), supplemented by questions on worldviews, existential values, and spiritual beliefs developed for a study on existential meaning and motherhood (Prinds et al., 2014). The questions regarded alterations in meaning and purpose in life, religious faith, belief in afterlife, church attendance, prayer, and meditation and whether there was a need (met or unmet) for discussing these existential matters with others.

Table 1. Overview of domains and psychometric scales included in the three questionnaires, showing the number of questions within the specific scales at specific times.

Scales	Questionnaire 1	Questionnaire 2	Questionnaire 3
Time since the loss	**24-60 days**	**7 months**	**13 months**
Socio-demographic and obstetric variables	22		
The Inventory of Daily Widowed Life (IDWL) (Caserta & Lund, 2007)	17	17	17
The Two Track Bereavement Questionnaire on Life Following Loss (TTBQ) (Rubin et al., 2009)	60	60	60
The Experience in Close Relationships Scale – revised,			

short form (ECR-R) (Fraley, Heffernan, Vicary, & Brumbaugh, 2011)	12		
Selected questions from The European Value Survey (Survey, 2006)	23		23
Prolonged Grief Disorder (PG-13) (Prigerson et al., 2009)	13	13	13
Post Traumatic Stress Disorder Checklist (PCL-PTSD) (Blevins, Weathers, Davis, Witte, & Domino, 2015)	18	18	18
Perinatal Grief Scale (PGS), Toedter et al., 1988)	33	33	33
Total number of questions	**198**	**141**	**164**

Preparation and pilot test

The first author and a research assistant (L Bilenberg Pedersen) translated the psychometric scales in English language: PGS, TTBQ, and PCL from English to Danish and the last author back translated to English after which consensus was reached based on the original and back translated versions of the scales. The survey was tested for comprehensibility by seven health care professionals with experience in the field of bereavement, and 18 parents, mostly mothers, with previous loss of a small child, and the final survey was adjusted according to their evaluation and comments.

Data analyses plan

By January 1, 2018 we had received 300 completed first questionnaires. We estimate that 800 parents in Denmark every year will experience a loss during pregnancy from GA week 14 and till four weeks after birth. With the data collection now on-going in all of Denmark, we expect to include 400 mothers and 240 partners every year, with an estimated response rate of 50% among

mothers and 30% among partners. We anticipate to have included approximately 5,000 mothers and partners by January 2024, comprising the largest cohort in this field to date.

We expect to enroll the first PhD student in September 2018 in a study focusing on attachment style and continuing bonds. Data will be assessed using STATA version 15.0 (StatCorp, Texas, USA).

Ethics and dissemination

The project will be performed according to the recommendations for good scientific practice (UVVU, 2009). The Danish National Data Protection Agency has approved the project (permit number 2008-58-0035, October 7, 2014) with a data collection till 2024. Bereaved parents are in a particular vulnerable situation and inviting them to partake in research requires specific ethical considerations. However, studies show that bereaved parents find partaking in research projects to be an encouraging experience (Dyregrov, 2004), motivated by an aspiration to help other parents (Breeze, Statham, Hackett, Jessop, & Lees, 2011; Dyregrov, 2004). Participation was voluntary, anonymous and confidential. No incentives or compensation were offered. The parents gave their consent by ticking a box stating confirmation to participate in the study and afterwards access to the questionnaire itself was given.

Author contributions

Study conception and design: Hvidtjørn, Prinds, Brink Henriksen, Cacciatore and O'Connor

Acquisition of data: Hvidtjørn, O'Connor

Analysis and interpretation of data: Hvidtjørn, Prinds, Bliddal, Brink Henriksen, Cacciatore and O'Connor

Drafting of manuscript and critical revision: Hvidtjørn, Prinds, Bliddal, Brink Henriksen, Cacciatore and O'Connor

Funding statement

The study was funded by Aase and Ejnar Danielsen's Fund.

Competing interests statement

No competing interests.

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BMJ Open

Life after loss
Protocol for a Danish longitudinal follow-up study unfolding life and grief after the death of a child during pregnancy from gestational week 14, during birth or in the first 4 weeks of life

Journal:	*BMJ Open*
Manuscript ID	bmjopen-2018-024278.R1
Article Type:	Protocol
Date Submitted by the Author:	30-Aug-2018
Complete List of Authors:	Hvidtjørn, Dorte; Syddansk Universitet Det Sundhedsvidenskabelige Fakultet, Prinds, Christina; Syddansk Universitet Det Sundhedsvidenskabelige Fakultet Bliddal, Mette; University of Southern Denmark, Obestrics and Gynaelology Henriksen, Tine; Aarhus Universitet Health Cacciatore, Joanne; Arizona State University O'Connor, Maja; AArhus University
Primary Subject Heading:	Evidence based practice
Secondary Subject Heading:	Obstetrics and gynaecology, Health services research, Mental health, Patient-centred medicine
Keywords:	Perinatal Death, Grief, Cohort Study, Life Change Events, Quality of health care

Life after loss

Protocol for a Danish longitudinal follow-up study unfolding life and grief after the death of a child during pregnancy from gestational week 14, during birth or in the first 4 weeks of life

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Keywords: Perinatal Death, Grief, Cohort Study, Life Change Events, Quality of Health Care

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Word count:

Abstract

Introduction: After the death of a child during pregnancy, birth or in the neonatal period, parents often experience feelings of guilt, disenfranchisement, feelings of betrayal by one's own body and envy of others. Such bereavement results in high rates of distress: psychologically, emotionally, physiologically and existentially. These data are collected using a national, longitudinal cohort to assess grief in mothers and their partners after the death of a child during pregnancy, birth, or in the neonatal period. Our aim is to achieve a general description of grief, emotional health, and existential values after pregnancy or perinatal death in a Danish population.

Methods and analysis: The cohort comprises mothers and their partners in Denmark who lose a child during pregnancy from gestational week 14, during birth or in the neonatal period (4 weeks post partum). We began data collection in 2015 and plan to continue until 2024. The aim is to include 5,000 participants by 2024, generating the largest cohort in the field to date. Parents are invited to participate at the time of hospital discharge or via the Patient Associations homepage. Socio-demographic and obstetric variables are collected. Validated psychometric measures covering attachment, continuing bonds, posttraumatic stress, prolonged grief, perinatal grief and existential values were chosen to reach our aim.

Ethics and dissemination: Data are collected using web-based questionnaires distributed at 1-2, 7 and 13 months after the loss. The study was approved by The Danish National Data Protection Agency (No. 18/15684, October 7, 2014). The results will be disseminated in peer-review and professional journals as well as in layman magazines, lectures and radio broadcasts.

Strengths and limitations

- A comprehensive population based longitudinal study targeting at 5,000 participants by 2024 (by January 2018, 300 completed questionnaires and ongoing), and expected to become the largest cohort in this field.
- Using multiple validated, self-administered questionnaires enabling studies within attachment, continuing bonds, posttraumatic stress, prolonged grief, perinatal grief, existential values and the quality of health services.
- Multi-professional approach including psychologist, midwives, perinatal epidemiologists and anthropologists ensuring a resourceful approach.

- A robust response rate around 50%, however with 75% of the participants being mothers. Potential participants receive written and verbal reminders to improve the response rate and information addressing the importance of the fathers' participation specifically has been added.
- Non-responders can be described partly, but information on educational level and employment is not available for non-responders. A selection analysis will be performed using the available variables (e.g. age, parity, gestational age at birth and type of loss) and also comparing participants to the background population.
- Recruitment differs between the five regions of Denmark, as all potential participants are invited in two of the regions and in the remaining three regions participants are only invited via homepages, Facebook and patient organizations. Most likely, this will lead to an uneven participation rate and selection between regions; however, it will allow us to assess the efficiency of the various recruitment types.

Introduction

When we lose a person we love, we grieve. Grief is a simultaneously universal phenomenon and yet an entirely individual experience. Grief is also a cultural phenomenon, influenced by alternating normativity and beliefs over time.[1] In contemporary Western countries, grief and suffering are increasingly embedded in medical and psychiatric paradigms.[2, 3] For example, the World Health Organization is preparing criteria for a new diagnosis termed Prolonged Grief Disorder (PGD), anticipated to be introduced in the diagnostic manuals for mental disorders in 2018.[4] Discussions about how to define pathological grief are actualized both in professional settings and the broader population.[5-8] There is general agreement that the majority of

bereaved individuals eventually, and without professional interventions, will arrive at a new emotional equilibrium after loss. According to Litz et al only a minority will experience PGD, suffering significant impairment in important areas of daily life to a disabling degree more than six months after loss.[9] A recent meta-analysis found a prevalence of PGD in approximately 10% in bereaved adults, however only a small fraction of the bereaved in these 14 studies included bereaved parents.[10]

Thus, it is not clear how well these findings apply to the grieving process among parents after the death of a baby. Their grieving process might differ from grief processes in general, and a larger proportion may experience the symptoms of PGD. Put differently, a longer period of intense grief may be the normal response for parents grieving after the death of a baby. This hypothesis forms the basis of this longitudinal, follow-up study where we aim to assess grief among mothers and partners after the loss of a child during pregnancy from gestational week 14, during birth or in the neonatal period (4 weeks post partum). We include miscarriages, termination of pregnancy due to fetal anomaly (TOPFA), and the death of babies due to stillbirths and neonatal deaths.

The nature of perinatal grief

The death of a baby can be a life-changing and devastating experience.[11] A growing body of literature has assessed the nature of grief among parents who lose a child during pregnancy, birth, or in the neonatal period. One meta-analysis analyzing 144 studies about parental grief demonstrates that the majority of studies originate in North America, followed by Great Britain, Sweden and Australia.[12] Findings conclude that loss from miscarriage, stillbirth, TOPFA or neonatal death often involves feelings of guilt, disenfranchisement, feelings of betrayal by ones body and envy of others.[12, 13] Parents lose the prospect of an entire life with the child and all

the moments they dreamt of sharing.[14] Furthermore, bereaved parents of young babies who die have few mementoes of the child, none or few pictures and a very short narrative.[15] The loss has been called “invisible” and especially if the child died before or during birth family and friends might not regard the child as real. They may also be reluctant to talk about the dead child, leading to emotional isolation complicating the grief process.[12, 16, 17] Some parents, mostly mothers, describe a loss of self-esteem.[14, 16, 18, 19] For the mother, the bodily unity with the dead child might be still another stressful element.[20] For parents choosing TOPFA feelings of guilt and doubt can further thwart the grief process.[21, 22]

Outcomes of pregnancy and perinatal bereavement

The outcomes of pregnancy and perinatal bereavement are also assessed in international studies, finding high rates of psychological and emotional distress and diagnoses including; major depressive disorder, general anxiety disorder, post traumatic stress disorder (PTSD), sense of failure, long-term guilt and intense grief for more than 2 years.[12, 20, 23, 24]

Generally, measured by similar instruments mothers appear to be more afflicted than fathers.[23, 25, 26] A review of 11 studies assessed the association between type of loss and mental health and found lower or comparable levels of depression, anxiety and PTSD among mothers losing a child from TOPFA and mothers losing a child from stillbirth.[27] The risk for PTSD was assessed in a systematic review of 48 studies. They found an increasing risk of PTSD related to higher gestational age at time of loss and certain socio-demographic and psychosocial characteristics predicting PTSD.[28] Unsurprisingly, the PTSD risk appears to be 7 times higher in mothers after a perinatal death compared to mothers with a live birth.[29] A review of 18 studies on self-blame, guilt, and shame among bereaved parents (including stillbirths and the loss of older children) showed a high prevalence of all three states and an association with grief intensity.[30]

Higher mortality rates from natural causes among mothers who experienced a perinatal death were found in two large population based studies established on register data.[31, 32]

Explorations of subsequent pregnancy following the loss suggest that some mothers are at an increased risk of depression and anxiety.[33, 34] Patient-centered compassionate care is valued by the parents,[20] but we identified no studies assessing the long term effect of the type of care provided at the hospital.

Despite the above-mentioned outcomes, some bereaved parents describe the loss as a pivotal event in a broader and more life-changing sense. Grief can make an existential imprint on the bereaved parents potentially leading to both posttraumatic growth as well as posttraumatic stress, which is often mentioned in the literature.[11, 35-37]

The theoretical framework

In this section we explicate the theoretical framework, which piloted the preparation of the questionnaires and the forthcoming data analyses.

In general, unexpected and traumatic loss increases the risk of impaired physical and emotional health in the bereaved, and the loss of a child in the perinatal period will most often be unexpected and traumatic.[25]

Attachment style is shown to be related to adaption to the loss with more intense and enduring symptoms of grief and depression, complicated grief reactions, and decreased resilience in parents with an insecure attachment style (on both avoidance and anxiety attachment).[38, 39] Attachment theory, first introduced by John Bowlby in the 1970s, provides a unique way to characterize individual differences in reactions to loss because it illuminates the nature of a person's relationships and adjustment in situations of separation.[38] Different styles

of attachment, developed through the early parent-child relationship, will form the basis for responses to emotionally distressing situations such as bereavement.[40]

The Dual Process Model (DPM) has become a widespread model in understanding grief in contemporary Western countries.[41] The DPM emphasizes two concurrent types of stressors and coping processes: loss-oriented and restoration-oriented. It underscores that bereaved individuals often oscillate between these two processes throughout the course of bereavement, and a standstill in one of the two processes might be associated with prolonged grief.[41]

In Freud's classic grief work theory, detachment from the person who died is emphasized as fundamental for adaption to the loss and this idea has influenced the attitude of society and bereaved individuals for nearly 100 years. This philosophy is now challenged by the continuing bonds theory.[42] Continuing bonds has been defined as ''*the presence of an on-going inner relationship with the deceased person by the bereaved individual*'' representing diverse behaviors.[43] The literature reveals contradictory findings of the role of continuing bonds in bereavement, with certain types of continuing bonds associated with both adaptive and maladaptive adjustment in various studies. Moreover, outcomes are influenced by the social and cultural acceptance of grieving individuals and their continued relationship with the deceased.[44]

When a child dies at birth the natural order of life is disturbed and assumptive worldviews shatter, challenging three primary core beliefs relating to benevolence, meaningfulness of the world and worthiness of the self, and requiring a reorganization of worldviews.[11, 45] This disruption of core belief might lead to changes in philosophy of life or spiritual beliefs.[11] These changes may be perceived as helpful or unhelpful, as the literature shows incongruent findings in how religiosity and spirituality relate to bereavement outcomes.[39]

The Danish setting

There are huge dissimilarities between the health care systems in Denmark and North America from where most of the studies originate. Danish health care is publicly available and free. Furthermore, there are different approaches in the way healthcare professionals support bereaved parents in creating a relationship with their dead child and acknowledging their grief.[12, 46] Additionally, patient centered psychosocial care is a basic standard of care in Danish hospitals, while the prescription of psychiatric medication appears to be much more common in the U.S.[3, 46] Specifically, when we explore existential values and spiritual beliefs, findings from more religious countries, such as the U.S., have poor external validity when compared to a secularized country such as Denmark.[47-49] Hence, studies in a Danish context can expand our knowledge on grief after perinatal death.

Aim

In this longitudinal national follow-up study, we aim to assess grief symptoms among mothers and partners after the loss of a child during pregnancy, birth, or in the neonatal period. We aim to achieve a general description of grief, emotional health, and existential values after pregnancy or neonatal loss in a Danish population.

To achieve our objectives, we based the study on the following overall research questions:

1. How does the process of grief change for bereaved parents in the first 13 months after the loss?
2. What, if any, gender differences exist in the grief process?
3. How is attachment style associated with continuing bonds and grief?
4. Does gestational age at the time of death influence grief?

5. Does the loss change existential or spiritual values or practices?

Methods and analysis

This nationwide population based cohort study comprises mothers and partners who lost a child during pregnancy after gestational week 14, during or after birth or in the neonatal period. We include miscarriage, TOPFA, stillbirth and neonatal death. In Denmark, a regional counsel can grant permission to perform TOPFA until GA week 22; stillbirth is defined as intrauterine fetal death from GA week 22. We use web-based questionnaires distributed at three specific time points in the first 13 months after the loss. Study data were collected and managed using REDCap electronic data capture tools hosted at University of Southern Denmark.[50] Mothers and their partners are asked to reply to the questionnaires individually.

Data collection started in the Region of Southern Denmark in January 2016 and in the Region of Central Jutland in January 2017. In the summer of 2018 we expanded the study nationally and included the remaining three regions in Denmark. Data collection was permitted by The Danish Data Protection Agency until January 2025.

In the Regions of Southern Denmark and Central Jutland, parents receive short written and verbal information from healthcare professionals about the study before leaving hospital. Subsequently they receive the first e-mail from the project manager 4 to 8 weeks after the loss with comprehensive information about the study and a link to the questionnaire. In the rest of Denmark, parents are invited to participate through announcements on the homepage for the national patient organization “Landsforeningen Spædbarnsdød”. This agency offers free counseling to perinatally bereaved families. Via a link at the homepage, parents sign up with an e-

mail address and receive comprehensive information and a questionnaire. Access to the study questionnaire is given only when the parents have consented to participation.

The questionnaire is sent to parents at three time points: 4 to 8 weeks, 7, and 13 months after the loss. If not returned, each questionnaire is followed by reminders, the first one after 3 weeks and the second one after 6 weeks. Due to an initial low response rate, we further introduced a verbal reminder in January 2018 via a telephone call made by a research assistant with experience in grief counseling. In the Region of Southern Denmark, basic information (age, date of birth, date of death, gestational age at birth, parity and type of loss) on all potential participants are registered, allowing us to conduct a dropout analysis.

The questionnaires

The survey was constructed with a combination of basic information in relation to socio-demographics and obstetric variables, state-of-the-art psychometrical testing by validated questionnaires and ad hoc questions specifically prepared for this study. We included seven psychometric scales addressing the different aspects of our research questions (Table 1).

Socio-demographic variables

We included the following socio-demographic variables: age, sex of partner (to identify female partners), marital status (married, co-habiting, single), educational level (basic school (9–10 years of education), intermediate length education (11–16 years of education) and university education (17 or more years of education), present occupation and occupation before the loss (on maternity leave, on sick leave, unemployed, at work or studying).

Obstetric and organizational variables

The following obstetrical variables were included: previous perinatal loss, parity, assisted reproduction, single- or multiple pregnancies, type of loss (missed abortion, miscarriage, TOPFA, stillbirth, death after birth), mode of birth (vaginal vs. cesarean section), gestational age at the time of loss, admission to the neonatal intensive care unit (NICU), and age of child if death occurred after birth, and seeing and holding the dead child. Organizational variables were: hospital and type of department to which the couple was admitted, and experienced quality of psychosocial support (midwives, doctors, nurses, social worker, undertaker, religious person (chaplain, imam or other) and patient organization).

Psychometric scales

To measure attachment style we included a version of "The Experience in Close Relationships Scale – revised, short form (ECR-R)" modified to bereaved samples.[51] Participants were asked to express how much they agreed or disagreed with 12 statements concerning how they feel in emotionally intimate relationships on a seven-point scale ranging from highly disagree to strongly agree. Scores for attachment related anxiety and attachment related avoidance were obtained by averaging a person's scores (0 to 6) on each of the 12 items and the composite scores for anxiety and avoidance were highly reliable (alpha score >.80) despite based on a small number of items.[51]

We used the Perinatal Grief Scale (PGS) developed in 1988 to construct a comprehensive measure of perinatal grief to facilitate comparison among findings in the field.[13, 52] The scale was constructed to address the potential disparities between nonspecific grief and perinatal grief.[13] The PGS contains 33 statements covering dimensions as e.g. guilt, loneliness, and jealousy with an option of answering on a five-point scale ranging from highly disagree to

strongly agree. The PGS has good internal consistency (alpha 0.95).[53] A clinical cut-off of 91 has been established for the PGS, where greater scores indicate a high level of perinatal grief.[53]

To assess the process of bereavement within the DPM paradigm, the Inventory of Daily Widowed Life (IDWL) was used.[54] The IDWL was developed studying a group of widows in 2007. However, items in the scale could be adaptable to other losses and relationships with some modifications.[54] The inventory comprises 15 items on doings, thoughts, or feelings and the participants answer how often within the last week they have been preoccupied by each task by choosing one of four categories; seldom or never, sometimes, quite often and nearly all the time. IDWL was developed to measure the processes of loss-orientation (LO) and restoration-orientation (RO) and the oscillation between. The LO and RO subscales produced alpha coefficients of .90 and .79, respectively.[54]

We also included the Post Traumatic Stress Disorder Checklist (PCL-PTSD).[55] The PCL was developed in 1990 and comprises 17 items corresponding to the PTSD symptom criteria in the Diagnostic and Statistical Manual of Mental Disorders (5th ed.).[55] Respondents indicate how much they have been bothered by each PTSD symptom over the past month, using a 5-point scale ranging from not at all to extremely (scores 1 to 5). PCL scores exhibited strong internal consistency ($\alpha = .94$), and test-retest reliability ($r = .82$).[55]

To address the question of continuing bonds, we included “The Two Track Bereavement Questionnaire on Life Following Loss”.[56] This model aims to devote balanced attention to two domains of the bereavement experience: the nature of biopsychosocial functioning and the nature of the ongoing relationship to the deceased. Construct and concurrent validity were examined and were found satisfactory.[56]

In order to assess the likelihood of symptoms related to the diagnosis of PGD within this cohort, we incorporated Prolonged Grief Disorder-13.[57] The scale includes 13 items related to feelings, thoughts and behaviors. High scores within the specific items associated with severe functional impairment fulfill the criterion for PGD. Item response theory analyses derived the most informative, unbiased PGD symptoms, combination analyses identified the most sensitive and specific PGD algorithm and the scale was then tested finding high psychometric validity.

We also included questions from "The European Value Survey",[58] supplemented by questions on worldviews, existential values, and spiritual beliefs developed for a study on existential meaning and motherhood.[48] The questions addressed alterations in meaning and purpose in life, religious faith, belief in afterlife, church attendance, prayer, and meditation and whether there was a need (met or unmet) for discussing these existential matters with others.

Table 1. Overview of domains and psychometric scales included in the three questionnaires, showing the number of questions within the specific scales at specific times.

Scales	Questionnaire 1	Questionnaire 2	Questionnaire 3
Time since the loss	**24-60 days**	**7 months**	**13 months**
Socio-demographic and obstetric variables	22		
The Inventory of Daily Widowed Life (IDWL) (Caserta & Lund, 2007)	15	15	15
The Two Track Bereavement Questionnaire on Life Following Loss (TTBQ) (Rubin et al., 2009)	60	60	60
The Experience in Close Relationships Scale – revised, short form (ECR-R) (Fraley, Heffernan, Vicary, &	12		

Brumbaugh, 2011)			
Selected questions from The European Value Survey (Survey, 2006)	23		23
Prolonged Grief Disorder (PG-13) [57]	13	13	13
Post Traumatic Stress Disorder Checklist (PCL-PTSD) (Blevins, Weathers, Davis, Witte, & Domino, 2015)	17	17	17
Perinatal Grief Scale (PGS), Toedter et al., 1988)	33	33	33
Total number of questions	**195**	**138**	**161**

Preparation and pilot test

The first author and a research assistant (L Bilenberg Pedersen) translated the psychometric scales into English: PGS, TTBQ, and PCL from English to Danish and the last author back translated to English after which consensus was reached based on the original and back translated versions of the scales. The survey was tested for comprehensibility by seven health care professionals with experience in the field of bereavement, and 18 parents, mostly mothers, with a previous loss of a young child. The final survey was adjusted according to their evaluation and comments.

Data analyses plan

By January 1, 2018 we had received 300 completed first questionnaires. We estimate that 800 parents in Denmark every year will experience a loss from GA week 14 up to four weeks after birth. With the data collection now on-going in all of Denmark, we expect to include 400 mothers and 240 partners every year, with an estimated response rate of 50% among mothers and 30%

among partners. We anticipate approximately 5,000 participants by January 2024, comprising the largest cohort in this field to date.

In the region of Southern Denmark, we have the following information on non-responders: mother/partner, age, date contacted, parity, gestational age at birth and age at death, whether the loss was TOPFA, miscarriage, stillbirth or death post partum, and whether the participant wanted to take part in a bereavement support group. We will describe the non-responders/responders according to these variables.

We expect to enroll the first PhD student in September 2018 in a study focusing on attachment style and continuing bonds.

Statistical analysis plan

The characteristics of the cohort will be described by means and interquartile ranges for continuous data and by frequencies with 95% confidence interval for categorized and dichotomized data. Hypotheses will be modeled and tested using logistic regression models for dichotomous outcomes and ordinal logistic regression models where the dependent variable is based on ordinal data according to e.g. type of attachment, gender of the bereaved, or type of loss. We will use mixed effect models to examine time trends when examining changes over time in answering the same questions up to three times. All analyses will be adjusted for relevant socio-demographic and medical covariates depending on the underlying hypothesis. Data will be analyzed using STATA version 15.0 (StatCorp, Texas, USA).

Patient and Public Involvement

Eighteen parents who had previously lost a baby tested the questionnaire and their priorities, experience, and preferences were taken into account in the final version of the survey. The patient organization "Landsforeningen Spædbarnsdød" played a very active part in the design of the study. The study has a homepage where published papers will be presented to ensure that participants have assess to the results:

https://www.sdu.dk/da/om_sdu/institutter_centre/klinisk_institut/forskning/forskningsenheder/gynaekologiobstetrik/forskningsprojekter/liveteftertabet

Ethics and dissemination

The project will be enacted according to the recommendations for good scientific practice.[59] The Danish National Data Protection Agency has approved the project (permit number 2008-58-0035, October 7, 2014) with a data collection till 2024. Bereaved parents are a particularly vulnerable population and inviting them to partake in research requires specific ethical considerations. However, studies show that bereaved parents find partaking in research projects to be an positive experience,[60] motivated by an aspiration to help other parents,[60, 61] Participation was voluntary, anonymous and confidential. No incentives or compensation were offered. Participants gave their consent by ticking a box stating confirmation to participate in the study and afterwards access to the questionnaire itself was given.

The results will be disseminated in peer-reviewed and professional papers, as well as in more public layman magazines, lectures and radio broadcasts.

Author contributions

Study conception and design: Hvidtjørn, Prinds, Brink Henriksen, Cacciatore and O'Connor

Acquisition of data: Hvidtjørn, O'Connor

Analysis and interpretation of data: Hvidtjørn, Prinds, Bliddal, Brink Henriksen, Cacciatore and O'Connor

Drafting of manuscript and critical revision: Hvidtjørn, Prinds, Bliddal, Brink Henriksen, Cacciatore and O'Connor

We wish to thank "Landsforeningen Spædbarnsdød" and the parents who participated in the design of the study and the development of the survey.

Funding statement

The study was funded by Aase and Ejnar Danielsen's Fund.

Competing interests statement

No competing interests.

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BMJ Open

Life after loss
Protocol for a Danish longitudinal follow-up study unfolding life and grief after the death of a child during pregnancy from gestational week 14, during birth or in the first 4 weeks of life

Journal:	*BMJ Open*
Manuscript ID	bmjopen-2018-024278.R2
Article Type:	Protocol
Date Submitted by the Author:	06-Nov-2018
Complete List of Authors:	Hvidtjørn, Dorte; Syddansk Universitet Det Sundhedsvidenskabelige Fakultet, Department of Clinical Research; Odense Universitetshospital, Gynaecology And Obstetrics Prinds, Christina; Syddansk Universitet Det Sundhedsvidenskabelige Fakultet, Department of Clinical Research Bliddal, Mette; University of Southern Denmark, OPEN Henriksen, Tine; Aarhus Universitetshospital, Department of Paediatrics Cacciatore, Joanne; Arizona State University, School of Social Work O'Connor, Maja; AArhus University, Department of Psychology and Behavioural Sciences
Primary Subject Heading:	Evidence based practice
Secondary Subject Heading:	Obstetrics and gynaecology, Health services research, Mental health, Patient-centred medicine
Keywords:	Perinatal Death, Grief, Cohort Study, Life Change Events, Quality of health care

Life after loss

Protocol for a Danish longitudinal follow-up study unfolding life and grief after the death of a child during pregnancy from gestational week 14, during birth or in the first 4 weeks of life

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Keywords: Perinatal Death, Grief, Cohort Study, Life Change Events, Quality of Health Care

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Word count:

Abstract

Introduction: After the death of a child during pregnancy, birth or in the neonatal period, parents often experience feelings of guilt, disenfranchisement, feelings of betrayal by one's own body and envy of others. Such bereavement results in high rates of distress: psychologically, emotionally, physiologically and existentially. These data are collected using a national, longitudinal cohort to assess grief in mothers and their partners after the death of a child during pregnancy, birth, or in the neonatal period. Our aim is to achieve a general description of grief, emotional health, and existential values after pregnancy or perinatal death in a Danish population.

Methods and analysis: The cohort comprises mothers and their partners in Denmark who lose a child during pregnancy from gestational week 14, during birth or in the neonatal period (4 weeks post partum). We began data collection in 2015 and plan to continue until 2024. The aim is to include 5,000 participants by 2024, generating the largest cohort in the field to date. Parents are invited to participate at the time of hospital discharge or via the Patient Associations homepage. Data are collected using web-based questionnaires distributed at 1-2, 7 and 13 months after the loss. Socio-demographic and obstetric variables are collected. Validated psychometric measures covering attachment, continuing bonds, posttraumatic stress, prolonged grief, perinatal grief and existential values were chosen to reach our aim.

Ethics and dissemination: The study was approved by The Danish National Data Protection Agency (No. 18/15684, October 7, 2014). The results will be disseminated in peer-review and professional journals as well as in layman magazines, lectures and radio broadcasts.

Strengths and limitations

- A comprehensive population based longitudinal study targeting at 5,000 participants
- Using multiple validated, self-administered questionnaires enabling studies within attachment, continuing bonds, posttraumatic stress, prolonged grief, perinatal grief, existential values and the quality of health services
- Multi-professional approach including psychologist, midwives, perinatal epidemiologists and anthropologists ensuring a resourceful approach
- A robust response rate around 50%

- Representativeness and nonparticipation will be assessed according to age, parity, gestational age at birth and type of loss

Introduction

When we lose a person we love, we grieve. Grief is a simultaneously universal phenomenon and yet an entirely individual experience. Grief is also a cultural phenomenon, influenced by alternating normativity and beliefs over time.[1] In contemporary Western countries, grief and suffering are increasingly embedded in medical and psychiatric paradigms.[2, 3] For example, the World Health Organization is preparing criteria for a new diagnosis termed Prolonged Grief Disorder (PGD), anticipated to be introduced in the diagnostic manuals for mental disorders in 2018.[4] Discussions about how to define pathological grief are actualized both in professional settings and the broader population.[5-8] There is general agreement that the majority of bereaved individuals eventually, and without professional interventions, will arrive at a new emotional equilibrium after loss. According to Litz et al only a minority will experience PGD, suffering significant impairment in important areas of daily life to a disabling degree more than six months after loss.[9] A recent meta-analysis found a prevalence of PGD in approximately 10% in bereaved adults, however only a small fraction of the bereaved in these 14 studies included bereaved parents.[10]

Thus, it is not clear how well these findings apply to the grieving process among parents after the death of a baby. Their grieving process might differ from grief processes in general, and a larger proportion may experience the symptoms of PGD. Put differently, a longer period of intense grief may be the normal response for parents grieving after the death of a baby. This hypothesis forms the basis of this longitudinal, follow-up study where we aim to assess grief among mothers and partners after the loss of a child during pregnancy from gestational week 14, during

birth or in the neonatal period (4 weeks post partum). We include miscarriages, termination of pregnancy due to fetal anomaly (TOPFA), and the death of babies due to stillbirths and neonatal deaths.

The nature of perinatal grief

The death of a baby can be a life-changing and devastating experience.[11] A growing body of literature has assessed the nature of grief among parents who lose a child during pregnancy, birth, or in the neonatal period. One meta-analysis analyzing 144 studies about parental grief demonstrates that the majority of studies originate in North America, followed by Great Britain, Sweden and Australia.[12] Findings conclude that loss from miscarriage, stillbirth, TOPFA or neonatal death often involves feelings of guilt, disenfranchisement, feelings of betrayal by ones body and envy of others.[12, 13] Parents lose the prospect of an entire life with the child and all the moments they dreamt of sharing.[14] Furthermore, bereaved parents of young babies who die have few mementoes of the child, none or few pictures and a very short narrative.[15] The loss has been called “invisible” and especially if the child died before or during birth family and friends might not regard the child as real. They may also be reluctant to talk about the dead child, leading to emotional isolation complicating the grief process.[12, 16, 17] Some parents, mostly mothers, describe a loss of self-esteem.[14, 16, 18, 19] For the mother, the bodily unity with the dead child might be still another stressful element.[20] For parents choosing TOPFA feelings of guilt and doubt can further thwart the grief process.[21, 22]

Outcomes of pregnancy and perinatal bereavement

The outcomes of pregnancy and perinatal bereavement are also assessed in international studies, finding high rates of psychological and emotional distress and diagnoses including; major depressive

disorder, general anxiety disorder, post traumatic stress disorder (PTSD), sense of failure, long-term guilt and intense grief for more than 2 years.[12, 20, 23, 24]

Generally, measured by similar instruments mothers appear to be more afflicted than fathers.[23, 25, 26] A review of 11 studies assessed the association between type of loss and mental health and found lower or comparable levels of depression, anxiety and PTSD among mothers losing a child from TOPFA and mothers losing a child from stillbirth.[27] The risk for PTSD was assessed in a systematic review of 48 studies. They found an increasing risk of PTSD related to higher gestational age at time of loss and certain socio-demographic and psychosocial characteristics predicting PTSD.[28] Unsurprisingly, the PTSD risk appears to be 7 times higher in mothers after a perinatal death compared to mothers with a live birth.[29] A review of 18 studies on self-blame, guilt, and shame among bereaved parents (including stillbirths and the loss of older children) showed a high prevalence of all three states and an association with grief intensity.[30] Higher mortality rates from natural causes among mothers who experienced a perinatal death were found in two large population based studies established on register data.[31, 32]

Explorations of subsequent pregnancy following the loss suggest that some mothers are at an increased risk of depression and anxiety.[33, 34] Patient-centered compassionate care is valued by the parents,[20] but we identified no studies assessing the long term effect of the type of care provided at the hospital.

Despite the above-mentioned outcomes, some bereaved parents describe the loss as a pivotal event in a broader and more life-changing sense. Grief can make an existential imprint on the bereaved parents potentially leading to both posttraumatic growth as well as posttraumatic stress, which is often mentioned in the literature.[11, 35-37]

The theoretical framework

In this section we explicate the theoretical framework, which piloted the preparation of the questionnaires and the forthcoming data analyses.

In general, unexpected and traumatic loss increases the risk of impaired physical and emotional health in the bereaved, and the loss of a child in the perinatal period will most often be unexpected and traumatic.[25]

Attachment style is shown to be related to adaption to the loss with more intense and enduring symptoms of grief and depression, complicated grief reactions, and decreased resilience in parents with an insecure attachment style (on both avoidance and anxiety attachment).[38, 39] Attachment theory, first introduced by John Bowlby in the 1970s, provides a unique way to characterize individual differences in reactions to loss because it illuminates the nature of a person's relationships and adjustment in situations of separation.[38] Different styles of attachment, developed through the early parent-child relationship, will form the basis for responses to emotionally distressing situations such as bereavement.[40]

The Dual Process Model (DPM) has become a widespread model in understanding grief in contemporary Western countries.[41] The DPM emphasizes two concurrent types of stressors and coping processes: loss-oriented and restoration-oriented. It underscores that bereaved individuals often oscillate between these two processes throughout the course of bereavement, and a standstill in one of the two processes might be associated with prolonged grief.[41]

In Freud's classic grief work theory, detachment from the person who died is emphasized as fundamental for adaption to the loss and this idea has influenced the attitude of society and bereaved individuals for nearly 100 years. This philosophy is now challenged by the continuing bonds theory.[42] Continuing bonds has been defined as ''*the presence of an on-going*

inner relationship with the deceased person by the bereaved individual'' representing diverse behaviors.[43] The literature reveals contradictory findings of the role of continuing bonds in bereavement, with certain types of continuing bonds associated with both adaptive and maladaptive adjustment in various studies. Moreover, outcomes are influenced by the social and cultural acceptance of grieving individuals and their continued relationship with the deceased.[44]

When a child dies at birth the natural order of life is disturbed and assumptive worldviews shatter, challenging three primary core beliefs relating to benevolence, meaningfulness of the world and worthiness of the self, and requiring a reorganization of worldviews.[11, 45] This disruption of core belief might lead to changes in philosophy of life or spiritual beliefs.[11] These changes may be perceived as helpful or unhelpful, as the literature shows incongruent findings in how religiosity and spirituality relate to bereavement outcomes.[39]

The Danish setting

There are huge dissimilarities between the health care systems in Denmark and North America from where most of the studies originate. Danish health care is publicly available and free. Furthermore, there are different approaches in the way healthcare professionals support bereaved parents in creating a relationship with their dead child and acknowledging their grief.[12, 46] Additionally, patient centered psychosocial care is a basic standard of care in Danish hospitals, while the prescription of psychiatric medication appears to be much more common in the U.S.[3, 46] Specifically, when we explore existential values and spiritual beliefs, findings from more religious countries, such as the U.S., have poor external validity when compared to a secularized country such as Denmark.[47-49] Hence, studies in a Danish context can expand our knowledge on grief after perinatal death.

Aim

In this longitudinal national follow-up study, we aim to assess grief symptoms among mothers and partners after the loss of a child during pregnancy, birth, or in the neonatal period. We aim to achieve a general description of grief, emotional health, and existential values after pregnancy or neonatal loss in a Danish population.

To achieve our objectives, we based the study on the following overall research questions:

1. How does the process of grief change for bereaved parents in the first 13 months after the loss?
2. What, if any, gender differences exist in the grief process?
3. How is attachment style associated with continuing bonds and grief?
4. Does gestational age at the time of death influence grief?
5. Does the loss change existential or spiritual values or practices?

Methods and analysis

This nationwide population based cohort study comprises mothers and partners who lost a child during pregnancy after gestational week 14, during or after birth or in the neonatal period. We include miscarriage, TOPFA, stillbirth and neonatal death. In Denmark, a regional counsel can grant permission to perform TOPFA until GA week 22; stillbirth is defined as intrauterine fetal death from GA week 22. We use web-based questionnaires distributed at three specific time points in the first 13 months after the loss. Study data were collected and managed using REDCap electronic data capture tools hosted at University of Southern Denmark.[50] Mothers and their partners are asked to reply to the questionnaires individually.

Data collection started in the Region of Southern Denmark in January 2016 and in the Region of Central Jutland in January 2017. In the summer of 2018 we expanded the study nationally and included the remaining three regions in Denmark. Data collection was permitted by The Danish Data Protection Agency until January 2025.

In the Regions of Southern Denmark and Central Jutland, parents receive short written and verbal information from healthcare professionals about the study before leaving hospital. Subsequently they receive the first e-mail from the project manager 4 to 8 weeks after the loss with comprehensive information about the study and a link to the questionnaire. In the rest of Denmark, parents are invited to participate through announcements on the homepage for the national patient organization "Landsforeningen Spædbarnsdød". This agency offers free counseling to perinatally bereaved families. Via a link at the homepage, parents sign up with an e-mail address and receive comprehensive information and a questionnaire. Access to the study questionnaire is given only when the parents have consented to participation.

The questionnaire is sent to parents at three time points: 4 to 8 weeks, 7, and 13 months after the loss. If not returned, each questionnaire is followed by reminders, the first one after 3 weeks and the second one after 6 weeks. Due to an initial low response rate, we further introduced a verbal reminder in January 2018 via a telephone call made by a research assistant with experience in grief counseling. In the Region of Southern Denmark, basic information (age, date of birth, date of death, gestational age at birth, parity and type of loss) on all potential participants are registered, allowing us to conduct a dropout analysis.

The questionnaires

The survey was constructed with a combination of basic information in relation to socio-demographics and obstetric variables, state-of-the-art psychometrical testing by validated

questionnaires and ad hoc questions specifically prepared for this study. We included seven psychometric scales addressing the different aspects of our research questions (Table 1).

Socio-demographic variables

We included the following socio-demographic variables: age, sex of partner (to identify female partners), marital status (married, co-habiting, single), educational level (basic school (9–10 years of education), intermediate length education (11–16 years of education) and university education (17 or more years of education), present occupation and occupation before the loss (on maternity leave, on sick leave, unemployed, at work or studying).

Obstetric and organizational variables

The following obstetrical variables were included: previous perinatal loss, parity, assisted reproduction, single- or multiple pregnancies, type of loss (missed abortion, miscarriage, TOPFA, stillbirth, death after birth), mode of birth (vaginal vs. cesarean section), gestational age at the time of loss, admission to the neonatal intensive care unit (NICU), and age of child if death occurred after birth, and seeing and holding the dead child. Organizational variables were: hospital and type of department to which the couple was admitted, and experienced quality of psychosocial support (midwives, doctors, nurses, social worker, undertaker, religious person (chaplain, imam or other) and patient organization).

Psychometric scales

To measure attachment style we included a version of ”The Experience in Close Relationships Scale – revised, short form (ECR-R)” modified to bereaved samples.[51] Participants were asked to express how much they agreed or disagreed with 12 statements concerning how they feel in emotionally intimate relationships on a seven-point scale ranging from highly disagree to strongly agree. Scores

for attachment related anxiety and attachment related avoidance were obtained by averaging a person's scores (0 to 6) on each of the 12 items and the composite scores for anxiety and avoidance were highly reliable (alpha score >.80) despite based on a small number of items.[51]

We used the Perinatal Grief Scale (PGS) developed in 1988 to construct a comprehensive measure of perinatal grief to facilitate comparison among findings in the field.[13, 52] The scale was constructed to address the potential disparities between nonspecific grief and perinatal grief.[13] The PGS contains 33 statements covering dimensions as e.g. guilt, loneliness, and jealousy with an option of answering on a five-point scale ranging from highly disagree to strongly agree. The PGS has good internal consistency (alpha 0.95).[53] A clinical cut-off of 91 has been established for the PGS, where greater scores indicate a high level of perinatal grief.[53]

To assess the process of bereavement within the DPM paradigm, the Inventory of Daily Widowed Life (IDWL) was used.[54] The IDWL was developed studying a group of widows in 2007. However, items in the scale could be adaptable to other losses and relationships with some modifications.[54] The inventory comprises 15 items on doings, thoughts, or feelings and the participants answer how often within the last week they have been preoccupied by each task by choosing one of four categories; seldom or never, sometimes, quite often and nearly all the time. IDWL was developed to measure the processes of loss-orientation (LO) and restoration-orientation (RO) and the oscillation between. The LO and RO subscales produced alpha coefficients of .90 and .79, respectively.[54]

We also included the Post Traumatic Stress Disorder Checklist (PCL-PTSD).[55] The PCL was developed in 1990 and comprises 17 items corresponding to the PTSD symptom criteria in the Diagnostic and Statistical Manual of Mental Disorders (5th ed.).[55] Respondents indicate how much they have been bothered by each PTSD symptom over the past month, using a 5-point scale

ranging from not at all to extremely (scores 1 to 5). PCL scores exhibited strong internal consistency (α = .94), and test-retest reliability (r = .82).[55]

To address the question of continuing bonds, we included "The Two Track Bereavement Questionnaire on Life Following Loss".[56] This model aims to devote balanced attention to two domains of the bereavement experience: the nature of biopsychosocial functioning and the nature of the ongoing relationship to the deceased. Construct and concurrent validity were examined and were found satisfactory.[56]

In order to assess the likelihood of symptoms related to the diagnosis of PGD within this cohort, we incorporated Prolonged Grief Disorder-13.[57] The scale includes 13 items related to feelings, thoughts and behaviors. High scores within the specific items associated with severe functional impairment fulfill the criterion for PGD. Item response theory analyses derived the most informative, unbiased PGD symptoms, combination analyses identified the most sensitive and specific PGD algorithm and the scale was then tested finding high psychometric validity.

We also included questions from "The European Value Survey",[58] supplemented by questions on worldviews, existential values, and spiritual beliefs developed for a study on existential meaning and motherhood.[48] The questions addressed alterations in meaning and purpose in life, religious faith, belief in afterlife, church attendance, prayer, and meditation and whether there was a need (met or unmet) for discussing these existential matters with others.

Table 1. Overview of domains and psychometric scales included in the three questionnaires, showing the number of questions within the specific scales at specific times.

Scales	Questionnaire 1	Questionnaire 2	Questionnaire 3
Time since the loss	**24-60 days**	**7 months**	**13 months**

Socio-demographic and obstetric variables	22		
The Inventory of Daily Widowed Life (IDWL) (Caserta & Lund, 2007)	15	15	15
The Two Track Bereavement Questionnaire on Life Following Loss (TTBQ) (Rubin et al., 2009)	60	60	60
The Experience in Close Relationships Scale – revised, short form (ECR-R) (Fraley, Heffernan, Vicary, & Brumbaugh, 2011)	12		
Selected questions from The European Value Survey (Survey, 2006)	23		23
Prolonged Grief Disorder (PG-13) [57]	13	13	13
Post Traumatic Stress Disorder Checklist (PCL-PTSD) (Blevins, Weathers, Davis, Witte, & Domino, 2015)	17	17	17
Perinatal Grief Scale (PGS), Toedter et al., 1988)	33	33	33
Total number of questions	**195**	**138**	**161**

Preparation and pilot test

The first author and a research assistant (L Bilenberg Pedersen) translated the psychometric scales into English: PGS, TTBQ, and PCL from English to Danish and the last author back translated to English after which consensus was reached based on the original and back translated versions of the scales. The survey was tested for comprehensibility by seven health care professionals with experience in the field of bereavement, and 18 parents, mostly mothers, with a previous loss of a young child. The final survey was adjusted according to their evaluation and comments.

Data analyses plan

By January 1, 2018 we had received 300 completed first questionnaires. We estimate that 800 parents in Denmark every year will experience a loss from GA week 14 up to four weeks after birth. With the data collection now on-going in all of Denmark, we expect to include 400 mothers and 240 partners every year, with an estimated response rate of 50% among mothers and 30% among partners. We anticipate approximately 5,000 participants by January 2024, comprising the largest cohort in this field to date.

In the region of Southern Denmark, we have the following information on non-responders: mother/partner, age, date contacted, parity, gestational age at birth and age at death, whether the loss was TOPFA, miscarriage, stillbirth or death post partum, and whether the participant wanted to take part in a bereavement support group. We will describe the non-responders/responders according to these variables.

We expect to enroll the first PhD student in September 2018 in a study focusing on attachment style and continuing bonds.

Statistical analysis plan

The characteristics of the cohort will be described by means and interquartile ranges for continuous data and by frequencies with 95% confidence interval for categorized and dichotomized data. Hypotheses will be modeled and tested using logistic regression models for dichotomous outcomes and ordinal logistic regression models where the dependent variable is based on ordinal data according to e.g. type of attachment, gender of the bereaved, or type of loss. We will use mixed effect models to examine time trends when examining changes over time in

answering the same questions up to three times. All analyses will be adjusted for relevant socio-demographic and medical covariates depending on the underlying hypothesis. Data will be analyzed using STATA version 15.0 (StatCorp, Texas, USA).

Patient and Public Involvement

Eighteen parents who had previously lost a baby tested the questionnaire and their priorities, experience, and preferences were taken into account in the final version of the survey. The patient organization "Landsforeningen Spædbarnsdød" played a very active part in the design of the study. The study has a homepage where published papers will be presented to ensure that participants have assess to the results:

https://www.sdu.dk/da/om_sdu/institutter_centre/klinisk_institut/forskning/forskningsenheder/gynaekologiobstetrik/forskningsprojekter/liveteftertabet

Ethics and dissemination

The project will be enacted according to the recommendations for good scientific practice.[59] The Danish National Data Protection Agency has approved the project (permit number 2008-58-0035, October 7, 2014) with a data collection till 2024. Bereaved parents are a particularly vulnerable population and inviting them to partake in research requires specific ethical considerations. However, studies show that bereaved parents find partaking in research projects to be an positive experience,[60] motivated by an aspiration to help other parents,[60, 61] Participation was voluntary, anonymous and confidential. No incentives or compensation were offered. Participants gave their consent by ticking a box stating confirmation to participate in the study and afterwards access to the questionnaire itself was given.

The results will be disseminated in peer-reviewed and professional papers, as well as in more public layman magazines, lectures and radio broadcasts.

Author contributions

Study conception and design: Hvidtjørn, Prinds, Brink Henriksen, Cacciatore and O'Connor

Acquisition of data: Hvidtjørn, O'Connor

Analysis and interpretation of data: Hvidtjørn, Prinds, Bliddal, Brink Henriksen, Cacciatore and O'Connor

Drafting of manuscript and critical revision: Hvidtjørn, Prinds, Bliddal, Brink Henriksen, Cacciatore and O'Connor

Funding statement

The study was funded by Aase and Ejnar Danielsen's Fund.

Competing interests statement

No competing interests.

Acknowledgement

We wish to thank "Landsforeningen Spædbarnsdød" and the parents who participated in the design of the study and the development of the survey.

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