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## LOSING A PARENT TO CANCER AS A TEENAGER - Family cohesion, grief, long-term health and wellbeing and the development of a comprehensive care guide for personalized palliative care

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# Losing a parent to cancer as a teenager

Family cohesion, grief, long-term health and wellbeing  
and the development of a comprehensive care guide  
for personalized palliative care

DRÖFN BIRGISDÓTTIR

THE INSTITUTE FOR PALLIATIVE CARE | FACULTY OF MEDICINE | LUND UNIVERSITY





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Family cohesion, grief, long-term health and wellbeing  
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for personalized palliative care

Dröfn Birgisdóttir



**LUND**  
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Family cohesion, grief, long-term health and wellbeing  
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**MADE IN SWEDEN** 

*To all those who have lost a parent during childhood  
and have had to learn to live with the pain.*

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# List of abbreviations

|              |   |
|--------------|---|
| <b>ADL</b>   | activities of daily living  |
| <b>CI</b>    | confidence interval   |
| <b>DAG</b>   | direct analytical diagram   |
| <b>MRC</b>   | Medical Research Council (of the United Kingdom)                                  |
| <b>NVP</b>   | Nationell Vårdplan för Palliativ vård<br>[ <i>Swedish Palliative Care Guide</i> ] |
| <b>OR</b>    | odds ratio  |
| <b>PDSA</b>  | Plan-Do-Study-Act (cycle)   |
| <b>PHQ-9</b> | Patient Health Questionnaire 9  |
| <b>RR</b>    | risk ratio  |
| <b>S-PCG</b> | Swedish Palliative Care Guide<br>[ <i>Nationell Vårdplan för Palliativ vård</i> ] |



# Abstract

Around 3,500 children in Sweden experience the pain of losing a parent to death every year. Not all bereaved children face negative consequences such as depression, anxiety or even premature death, but some do. Limited evidence is available on the impact bereavement has on the family as a unit and the effect it has on teenagers' long-term health and wellbeing. Support to the family is included in national and international guidelines for good palliative care, although studies have shown that the translation of the knowledge into clinical practice can be challenging and is often insufficient.

The aim of this PhD project was to describe acute and long-term grief and family cohesion, as perceived by young adults (aged 18–26) who as teenagers (aged 13–16) have faced the death of a parent, and its association with long-term health and wellbeing. Also, to describe the development of a comprehensive clinical guidance for the care of patients with palliative care needs, including support that can help to identify children as family, and attend to their needs for information and support.

In a nationwide population-based study, the results showed that losing a parent to cancer as a teenager increases the risk of poor family cohesion during teenage years, compared with non-bereaved youths, and that the perception of poor family cohesion can last into young adulthood, but only among the maternally bereaved youths. Also, those reporting poor family cohesion in the first year after losing a parent to cancer had a greater risk for negative psychological health-related outcomes, 6–9 years after the loss. In addition, more than half of the cancer-bereaved teenagers in the study did not find a way to grieve that “felt okay” during the first 6 months after the loss, which was found to be associated with long-term unresolved grief. Furthermore, the results, of the exploratory intervention development study, showed that identifying individual care needs through a systematic and structured care guide can provide a comprehensive overview of the patients' needs; facilitate interprofessional assessment and care of the patient; and highlight the needs and possibilities beneficial for the patient and their family, throughout the whole palliative trajectory.

This PhD project provides knowledge that can be used to better understand and respond to the unmet needs of parentally bereaved children, as well as providing a support and guide that can be included in the clinical routines for patients with palliative care needs and their families.

# List of publications

This thesis is based on the following studies, which are referred to in the text by their Roman numerals:

## **Paper I**

Birgisdóttir, Dröfn; Bylund Grenklo, Tove; Nyberg, Tommy; Kreicbergs, Ulrika; Steineck, Gunnar; Fürst, Carl Johan (2019). Losing a parent to cancer as a teenager: Family cohesion in childhood, teenage, and young adulthood as perceived by bereaved and non-bereaved youths. *Psycho-Oncology*, 28(9), 1845–1853. <https://doi.org/10.1002/pon.5163>

## **Paper II**

Bylund Grenklo, Tove/Birgisdóttir, Dröfn; Beernaert, Kim; Nyberg, Tommy; Skokic, Viktor; Kristensson, Jimmie; Steineck, Gunnar; Fürst, Carl Johan; Kreicbergs, Ulrika (2021). Acute and long-term grief reactions and experiences in parentally cancer-bereaved teenagers. *BMC Palliat Care* 20, 75. <https://doi.org/10.1186/s12904-021-00758-7>

## **Paper III**

Birgisdóttir, Dröfn.; Bylund Grenklo, Tove; Kreicbergs, Ulrika; Steineck, Gunnar; Fürst, Carl Johan.; Kristensson, Jimmie (2022). Family cohesion predicts long-term health and well-being after losing a parent to cancer as a teenager: a nationwide population-based study. Manuscript submitted.

## **Paper IV**

Birgisdóttir, Dröfn; Duarte, Anette; Dahlman, Anna; Sallerfors, Bengt; Rasmussen, Birgit H.; Fürst, Carl Johan (2021). A novel care guide for personalised palliative care – a national initiative for improved quality of care. *BMC Palliat Care* 20, 176. <https://doi.org/10.1186/s12904-021-00874-4>

# Thesis at a glance

**Table 1.** Overview of the different parts of the PhD thesis.

|                                | Part 1  |   |  | Part 2   |
|--------------------------------|---|---|--|--|
| Study                          | I   | II  | III  | IV   |
| <b>Aim</b>                     | To investigate the level of perceived family cohesion during childhood, the teenage years, and young adulthood in cancer-bereaved youth compared with their non-bereaved peers.   | To investigate grief experiences and reactions in the acute bereavement phase, and their associations with long-term unresolved grief, as well as factors associated with having had an okay way to grieve in the first 6 months post-loss.                         | To investigate self-reported family cohesion in the first year after the death of a parent to cancer, and its association with long-term psychological health and wellbeing.                             | To describe the development and feasibility testing of the Swedish Palliative Care Guide (S-PCG).  |
| <b>Design</b>                  | Nationwide population-based study, with a cross sectional design.   |   |  | An exploratory intervention development study guided by the approach of interpretive description for applied practice.   |
| <b>Data collection methods</b> | Study specific questionnaire.   |   |  | Collection of written feedback.<br>Focus-group/small group/semi-structured interviews.<br>Review of patient records.   |
| <b>Main results</b>            | The cancer-bereaved participants were more likely than non-bereaved youth to report poor family cohesion during teenage years. This was also seen in young adulthood but only among the maternally bereaved participants. | Fifty-seven per cent of the bereaved teenagers had not found an okay way to grieve during the first 6 months post-loss. The acute grief experiences and reaction were found to be associated with their grief resolution 6–9 years after the death of their parent. | Reporting poor family cohesion in the first year after losing a parent to cancer was found to be strongly associated with long-term negative psychological health-related outcomes among bereaved youth. | Although further research is needed, the broad testing of the S-PCG showed its potential to provide support in identifying and addressing patients' palliative care needs, and highlighting the needs of the family, throughout the palliative trajectory. |

# Preface

Personal experiences influence the way a researcher conducts a study and should be described as part of the research scaffolding<sup>1</sup>. I will therefore here describe the personal context that is the basis for my entrance into this PhD project.

Back in Iceland, while studying physiotherapy at the university, I spent my summers working at a geriatric hospital, where I first became involved in rehabilitation of patients admitted to the palliative care unit and receiving end-of-life care. I found this work to be both rewarding and meaningful. So many of the dying persons seemed to have the urge to share with me their life stories, the lessons they had learned and their thoughts while being in the last stage of their life. I learned a lot from these moments and their wisdom shaped me. I believe one of the reasons why I have never found it hard to engage in discussions about life and death and why I am in fact able to enjoy these conversations, is related to having been raised on my parents' farm where the birth and the death of the animals were not hidden from us as children, but instead openly discussed and highlighted as part of life; and the same applied regarding the death of a relative or friend.

Later as a licensed physiotherapist I worked at a big rehabilitation centre in Iceland, in different units where people were admitted for chronic pain and psychiatric, cancer or pulmonary rehabilitation. I enjoyed working in rehabilitation and being a part of a well-functioning interdisciplinary teamwork, providing support underpinned by a holistic approach based on the person's needs, and guided by the motto "*Help to self-help*". During those years, I increasingly felt, however, that our support was offered too late, and that the health care system and society had failed to support these persons earlier on in life. I felt driven by the urge that more focus in health care should be on health prevention. This conviction ultimately led me, together with my husband and three children, to Sweden, where I studied for a Master of Science with a major in public health at Lund University.

Nearing the end of my Master's studies I met with a friend in Stockholm and joined her to attend Tove Bylund Grenklos (now my co-supervisor) PhD dissertation about the experiences of teenagers who lose a parent to cancer. I got very interested in the topic and when she explained that 30% of the participants in her study stated that no-one had told them their parent was dying, when there were only a matter of hours or days, not weeks, left in life, I thought to myself that here is a room for improvement.

Through my supervisor Carl Johan Fürst (who was also Tove's supervisor), I had the opportunity to become part of his team at the Institute for Palliative Care at Lund University and Region Skåne . Here, I was able to engage in further research on the comprehensive dataset that had already been collected for Tove's PhD project<sup>2</sup>, which also makes up the basis for my PhD project.

Furthermore, under the guidance of Carl Johan Fürst at the Institute for Palliative care in Lund, the development of the Swedish Palliative Care Guide (S-PCG) was initiated. Shortly after the onset of the project I became part of the project group and together with the project manager, Anette Duarte, was involved in all steps of the development process and the feasibility testing, a process that later was incorporated as part of my PhD project.

During my PhD project I have realized more and more that many people in my surroundings, both friends and family, have actually gone through the experience of losing a parent during their teenage years. Many of them have welcomed the chance to be able to discuss with me their own personal experience when I told them about the topic of this PhD project. I believe that we need to have the courage to address and discuss the life and death and to provide information and support to those that are facing bereavement, especially when there are children in the family. This will contribute to making death part of the life again in societies that have made it a taboo subject. As one of the great influencers of my childhood once said:

*“You have to live in such a way that you become friends with the death.”*

Astrid Lindgren (TV interview in 1987 (1:07:13))<sup>3</sup>

# Context of this thesis

When a parent with a minor child gets severely ill, it creates a huge amount of stress not only on the individual members of the family but on the family as a whole<sup>4, 5</sup>. Elevated levels of stress have been seen among adolescent and young adult individuals when their parent has been diagnosed with cancer<sup>6</sup>. Living with a dying parent has been shown to be emotionally demanding for a child<sup>7</sup> and experiencing the death of a parent has been considered one of the most challenging experiences that can occur in the life of a child<sup>8</sup>. At the same time as the child or teenager is in need of support while coping with the death of a parent, the surviving parent is also dealing with the loss, trying to adjust to the new situation and is not always capable of providing the support that is needed<sup>9</sup>. To be able to provide adequate support to bereaved children and teenagers, there is a need for more knowledge about their support needs. It is also important to gain more understanding of factors in their social environment that affect their long-term health and wellbeing.

Families who are facing the death of a parent often find themselves within the context of the health care services, and both parents and teenagers from such families have raised the need for more support from the health care<sup>10, 11</sup>. According to the Swedish Health Care laws, the health care personnel are obliged to pay special attention to children's needs for information and support when a parent, caretaker or other adult living with the children is severely ill or unexpectedly dies. This includes children who are losing a parent to terminal disease<sup>12, 13</sup>. Based on a systematic review of psychosocial outcomes in cancer-bereaved children and adolescents<sup>14</sup>, these children and adolescents and their families need support not only after the death of a parent, but also before the loss, during the illness period.

According to widely accepted definitions of the term, palliative care should not only attend to the dying person but should also address the needs of the family, both during the illness period and after the loss<sup>15, 16</sup>. This also includes the children in the family. The Swedish health care system is predominantly public, funded by taxes and decentralized. It is based on both regional and local or municipal governance, overseen by the national authorities, and includes both private and public actors<sup>17</sup>. Palliative care can be described as holistic care aiming to improve the quality of life and decrease suffering among patients who are threatened by a serious illness or facing the end of life, and their families<sup>16</sup>. Good care at the end of life should be available to everyone<sup>18, 19</sup>.

Within the Swedish health care system, palliative care is divided into general palliative care and specialized palliative care. General palliative care should be integrated into all care for people with possible palliative care needs or people who have limited time left in life. Specialized palliative care is aimed for those with difficult or complex needs at the end of life and can be provided at a specialized care unit or as part of general care with the support of a specialized palliative care consult team<sup>20</sup>.

This PhD project was conducted in two parts. Part 1 was a part of a nationwide population-based study on youth who had lost a parent to cancer as teenagers, 6–9 years previously and a matched random sample of non-bereaved controls. The study was initiated and conducted by a research group of the Division of Clinical Cancer Epidemiology at the Karolinska Institute and the Department of Research and Development at Stockholm's Sjukhem Foundation. A very comprehensive dataset had been collected prior to this PhD project and had been analysed in other studies<sup>2, 21-24</sup>, but the data in question here had yet to be studied and analysed.

Part 2 of this PhD project revolves around the development and feasibility testing of the Swedish Palliative Care Guide (S-PCG), initiated by the Institute for Palliative Care at Lund University and Region Skåne. The development of the S-PCG arose from the need to build a bridge between current evidenced-based guidelines and recommendations for the best care of the dying person and what is actually being applied in clinical practice. A perspective on the situation for the family, including children and teenagers, was embodied throughout the process. The project was conducted as a national health care quality improvement project, but used the UK Medical Research Council framework as a basis to provide a structure for the development with the aspiration to follow standards equivalent to academic requirements. The project was guided by the interpretive description approach for applied practice.

These two parts make up the basis for the PhD project that is described in this thesis.



# Background

Around 3,500 children in Sweden experience the loss of a parent to death every year<sup>25, 26</sup> and around 1,800 of them are teenagers at the time of loss<sup>26</sup>. This means that right now, and through the years, around 23,000 children under the age of 18 are living in Sweden, who have lost a parent to death<sup>27</sup>. Based on available data<sup>28</sup> it is estimated that 40% of the parental deaths are due to cancer<sup>29</sup>, which makes cancer the biggest single contributor to these premature deaths, while the rest are distributed between other causes of death, i.e. other diseases, suicide, accidents or homicide<sup>28, 29</sup>.

The terms “child”, “teenager”, “adolescent”, “youth” and “young adult” do not have a clear universal definition but in this thesis, “children” will be referred to as all people under the age of 18 as defined by the Swedish laws<sup>30</sup>. The term “teenager” is usually used to describe children from the age of 13 to 19, although the participants in this study were at the age of 13–16 years old when they lost a parent. This is equivalent to lower secondary school (or junior high school) age. Adolescents, on the other hand, are not defined by a specific age span. Rather the term “adolescence” refers to the period from the beginning of puberty to adulthood and is defined by a child’s physical, psychological and cultural transition into adulthood. It can therefore refer to broader age span, usually ranging somewhere from 10 to 19 years of age<sup>31</sup>. During the teenage years when a teenager is in adolescence they undergo huge developments. These include biological, psychological and social development<sup>32</sup>. The adolescent has a need to express independence and separation from their parents and their family, while at the same time still being dependent on them. Big life changes occurring during this period can have a great impact on the teenager that can last into adulthood<sup>33</sup>.

“Young adulthood” does not have a universal definition, and is often defined by various age spans between the age of 18 and 40<sup>34</sup> and the Swedish National Board of Health and Welfare does not give a clear definition for what constitutes as young adults. The term “young adults” in this thesis refers to participants aged 18–26, as defined by the Society for Adolescent Health and Medicine<sup>35</sup>, and the term “youth” will be used here to describe people spanning the ages from the teenage years to young adulthood, or the ages of 13 to 26.

Losing a parent is among the most stressful events that can occur in children’s and adolescents’ lives<sup>8, 36</sup> and although most children learn to cope with the situation and

establish a well-functioning life<sup>37</sup>, several studies have shown that some of these children are at increased risk of negative consequences, such as depression<sup>38-40</sup>, anxiety<sup>39-44</sup>, suicide attempts<sup>45, 46</sup> and self-injurious behaviour<sup>22, 47</sup> when compared with their non-bereaved peers. Some factors have been pointed out that might contribute to this increased risk of negative consequences, such as when the death comes after a prolonged illness<sup>48</sup> or is sudden or traumatic<sup>40, 49</sup>. Other contributing factors include not being aware that their parents' death is near<sup>21</sup>, as well as lack of support<sup>40, 50</sup>, distrust towards health care<sup>24</sup>, or further exposure to stressful events after the loss<sup>8, 51</sup>.

On the other hand, there are also protective factors that may prevent some of the negative impact that bereavement has on health and wellbeing. Family function, including cohesion, open communication and warmth and connection between the surviving parent and the bereaved children, together with good mental health of the surviving parent and their capability to provide positive parenting, are factors that seem to have the greatest protective impact on the wellbeing of bereaved children<sup>42, 44, 52-54</sup> after the loss. However, more studies are needed to look into the impact bereavement has on the family unit<sup>4</sup> and supportive factors in their wellbeing when a parent is dying<sup>55</sup>.

## Family function and family cohesion

Family cohesion has long been recognized as one of the strongest protective factors for the wellbeing of children and adolescents, overall<sup>56, 57</sup>, i.e. regardless of type of stressors. According to Bowen's family system theory<sup>58</sup>, families make up a complex social unit of members who interact with, and influence, each other's behaviours. The theory recognizes that the family as a whole, and its function, plays an important role in the wellbeing of the individuals in the family, influence their daily living and can be both disruptive and helpful<sup>59</sup>. This is related to the concept of family function, which has been defined as the capability of families to interact, work as a whole and cope with stressors. According to the widely used Circumplex Model of Marital and Family Systems, there are three dimensions that define family function: flexibility, family cohesion and communication<sup>60, 61</sup>. "Flexibility" refers to how well families are able to adapt to changes without losing the necessary stability of the family structure and roles. "Family cohesion" is a broad concept that describes the sense of emotional bonding between family members, but it also includes other factors such as support, the feeling of togetherness, intimacy and time spent together. According to Olson et al<sup>61</sup>, a balance between too much or too little cohesion is needed for a good family cohesion. Family cohesion changes over time and is influenced by stressors in the environment as well as within the family<sup>61</sup>. Good communication between the family members is the third dimension that is crucial for families to be able to adapt well to changes and stressful events, and for

good family cohesion. This includes the family members being able to share feelings, speak for themselves with clarity and conduct active listening, while showing respect and empathy<sup>60, 61</sup>.

Several scales are available that assess the family environment and its function<sup>62</sup>. In a systematic review examining the theoretical underpinnings and psychometric properties of self-reported family assessments scales, the authors found that five had been empirically validated for clinical use<sup>62</sup>. However, the authors also concluded that none of the available family measurements is suitable for clinical practice, since they all have an extensive number of items, ranging from 36 to 60. Furthermore, none of the available scales had been studied for their responsiveness to capture changes in family functioning over time<sup>62</sup>.

## Bereavement and grief

After losing a loved one, a period of grief and bereavement begins. “Bereavement” refers to the state of loss, meaning that bereaved individuals are those who have lost someone of a great value to them<sup>54</sup>. “Grief” is a term referring to the reactions people experience after a loss and grieving is considered a normal process after a loss<sup>63</sup>. Grief is a very unique experience and is greatly influenced by various factors in an individual’s life, such as personal traits, social and cultural circumstances and the relationship with the deceased<sup>64</sup>. The characteristics of the grieving process often include strong emotions such as anger, numbness, depression, guilt and anxiety<sup>63, 65, 66</sup>; cognitive reactions such as intrusive rumination or self-reproach; behavioural reactions such as crying, restlessness or social withdrawal; and psycho-somatic reactions such as loss of appetite<sup>63</sup> or sleep disturbance<sup>63, 67</sup>. These, often intense, reactions to the loss are considered to be a normal part of the acute bereavement phase and although the duration of the acute bereavement phase has not been defined, it has been suggested to include at least the first 6 months after the loss<sup>64</sup>.

To help us understand the grieving process several theories have been put forward. Some of them have been based on tasks that the grieving person has to address in order to progress through the grief<sup>68</sup> while others have used stages (i.e. as denial, anger, depression, acceptance)<sup>69</sup>. Many theories in the past have stressed that in order to heal from the pain caused by the loss the person must “work through the grief”<sup>68-70</sup> to be able to adjust to the bereavement. Although widely known, the stage theory of grief has been criticized for oversimplifying the very complex and individual process of grief, misleading people into thinking that going through the described stages is the “normal grieving process” that everyone should strive for, while the empirical evidence is showing that the majority of people do not grieve in stages<sup>71, 72</sup>. A more recent, but widely accepted theory, the dual process model, highlights that “grief work” is not only a simple linear cognitive process of facing

the loss. Its authors, Stroebe and Schut, state that it is normal during the grieving process to oscillate between the loss- and the restoration- oriented stressors that the bereaved individual has to cope with<sup>73</sup>.

Most of these theories, including the dual process model<sup>74</sup>, were initially designed around the grieving processes of adults. Although some of them have been used as a framework for understanding children's grieving processes studies have found that grief reactions of children and teenagers can often be different from those of adults<sup>75</sup>. More research is needed to build up the empirical evidence on children's and teenagers' grieving and reaction to loss<sup>64</sup>.

For most people, the intense grief reactions will usually diminish over time until the loss has become a part of the life of the bereaved one<sup>63</sup>. However, several studies have shown that some bereaved adults will continue to experience persisting problems related to grief that can continue for several years after the loss<sup>76-80</sup>. This is sometimes referred to as "pathological grief"<sup>64</sup> and several different terms have been used to describe this phenomenon of disturbed grief, such as "prolonged grief disorder"<sup>81, 82</sup>, "complicated grief"<sup>83</sup> and "persistent and complex bereavement disorder"<sup>84</sup>. Although these terms differ in the combination of symptoms that are used to define the phenomenon, they greatly overlap<sup>85</sup>. In a systematic review, Lundorff et al<sup>79</sup> found the pooled prevalence of prolonged grief disorder in bereaved adults to be around 10%, but emphasized that a large variation was seen between studies and different subgroups<sup>79</sup>. Similar to adults, it has been reported that around 10 % of bereaved children and adolescents can have sustained prolonged grief almost 3 years after a sudden parental death<sup>86</sup>, however, more studies are needed.

## Facing parental cancer and coping with loss

Children and teenagers who are faced with parental cancer have been shown to be at increased risk for elevated psychological stress<sup>87</sup> and emotional and behavioural problems<sup>87-89</sup>. In a longitudinal study, young adults who had had a parent diagnosed with cancer during their childhood were shown to be more likely to need to seek the help of psychiatric services<sup>90</sup>. As described by Faulkner and Davey<sup>91</sup>, in a literature review on the impact of cancer on children and adolescents, teenagers are often faced with the dilemma of wanting to seek more independence from the family while at the same time wanting to spend time with the family and the parent with cancer<sup>91</sup>. Problematic family functioning<sup>89, 92</sup>, severity of the cancer of the parent<sup>93, 94</sup>, and poor coping skills are factors that have been shown to lead to poor outcomes for the children<sup>89</sup>.

According to Folkman et al<sup>95</sup>, "coping" is a term for the cognitive and behavioural efforts that a person uses to manage both internal and external demands when facing a stressful situation perceived to exceed their personal resources. People have

different ways of coping with the loss of a loved one and how individuals cope has been acknowledged to be of importance for their future wellbeing. Most of the research in the past have revolved around the adult's way of coping with loss and little research has looked at these processes in children although children often express their grief differently from the way many adults do<sup>75</sup>.

Teenagers' brains are under extensive neurodevelopment<sup>96</sup>, and they usually cannot cope with the severe pain or withstand the strong emotional reactions brought on by loss, except for short moments at a time<sup>75, 97, 98</sup>. In an empirical case study on adolescents grief, Christ et al<sup>98</sup>, highlight that it is not uncommon for conflicts to arise between family members during bereavement, when they are grieving "out of sync". The teenager may experience frustration over the surviving parent's need to constantly express their grief. Similarly, when a teenager chooses to hang out with their friends immediately after the death of a loved one, the surviving adult may sometimes think they are being insensitive or disrespectful<sup>98</sup>. However, going out to "hang with friends" as if nothing has happened can be an important way for children and young adults to cope with the loss of a loved one<sup>10</sup>.

Knowledge about the variety of different grief reactions and experiences that may occur after the loss of a loved one can be helpful for the bereaved individuals when coping with loss<sup>55, 99</sup> and knowing that family members can grieve in different ways can increase the understanding within the family<sup>97, 98</sup>. However, as mentioned, not all knowledge from the adult bereavement research can be applied directly to children and teenagers<sup>100</sup> and more studies are needed to build up a strong empirical evidence to better understand the "normative grieving processes" of children and teenagers<sup>64</sup> and how they experience the loss of a parent<sup>101</sup>.

## Need for support

Parents with dependent children, who have been bereaved of a spouse, have expressed a need for more support, both before and after the loss<sup>10, 102</sup>. This includes a need for emotional support and practical help with the household and the children<sup>10</sup> from family and friends<sup>10, 55</sup>, which has been described as helpful<sup>103</sup>. Similar to adults, children and teenagers have also been shown to be in need of more support from family and friends<sup>10, 36</sup>. The surviving parent may be in a state of crisis and researchers have implied that a parent in this situation may not always be able to provide the support the child needs<sup>104, 105</sup>. Bereaved teenagers often take on greater responsibility than is usually expected at their age, in a process of so-called "child parentification", where they feel obliged to support their surviving parent and take on a greater share of household tasks and the responsibility for their minor siblings' wellbeing<sup>106</sup>. Apart from expressing the need for more support and understanding from family and friends, bereaved youth in an Australian study also expressed a

need for more information, more support in coping with the loss and more help with household tasks, as well as a desire to share the experience with peers in similar situations<sup>36</sup>. Adolescents who lost a parent to cancer have also expressed the importance of continuing life as normal, such as going to school, as a useful coping strategy<sup>10</sup>. This has similarly been reported by children after loss of a sibling<sup>107, 108</sup>.

Evidence is mounting, based on reports by both surviving parents and children, of the importance of being able to spend quality time with the ill parent before the death, which has been perceived as helpful in the adjustments following the loss<sup>11, 109-113</sup>. It is, however, not always easy to find time during the stressful illness period to spend a quality time together as a family<sup>114, 115</sup>. Open communication with parents during the end of life of a dying parent is another factor that has been identified as an important facilitator for children's adjustment after the loss<sup>55</sup>. However, children often do not want to add to the parent's burden by voicing their own needs or concerns<sup>55, 116</sup> and parents often experience difficulties telling their children when the death is inevitably near<sup>55</sup>.

Both parents and teenagers have expressed the need for more support from the health care personnel. This includes the opportunity to discuss the illness<sup>117</sup> and reflect on their feelings and grief<sup>10</sup>, and the need for clear information from the health care personnel about the diagnosis and prognosis<sup>11, 99, 118</sup> and the forthcoming death<sup>99</sup>. It also includes information about what to expect after the loss, and (for parents) how to talk to the children and support them<sup>119, 120</sup>. The need for information and support from the health care professionals on how to talk to the children is most pronounced at the time of diagnosis<sup>117</sup> and close to death, as well as for some time after the loss, and has especially been highlighted for those who lack a supportive social network<sup>121</sup>.

According to one systematic review comprising of five qualitative studies with focus on communication and support from health care professionals to families with children facing the life-threatening disease of a parent, the children wish for more information from the health care personnel about their parent's disease and prognosis<sup>117</sup>. The parents wish for guidance on how to talk to their children<sup>117</sup>. Children want to be included and informed when facing the death of a parent, but often feel left out by the adults, according to a recent integrative review and thematic synthesis<sup>7</sup>. It has previously been reported that 30% of young adults who lost a parent as teenagers stated that no one had told them that their parent's death was imminent, when there were only hours to days left before the death<sup>23</sup>. As many as 98% of them felt that teenage children should be told when the death of their parent is near<sup>23</sup> and not being told was associated with lower levels of trust in the health care<sup>24</sup>. Moreover, according to a systematic review on health care professionals' communication with and support of families with dependent children, there is a reluctance among the health care professionals to have these conversations with the children, or with the parents about how to talk to the children. More support to the health care personnel to take on this task is needed in clinical practice.<sup>117</sup>

## Need for better integration of palliative care within the health care system

Like the children and families who are facing bereavement and are in need of support, health care personnel also are in need of support and guidance regarding providing support for the children in the family<sup>117</sup>. Although definitions of “palliative care” state that it should provide the best end-of-life care possible for the dying person, as well as bereavement support for the family of the dying person<sup>15</sup>, studies are showing that bereavement needs are not being met, especially not the children’s needs<sup>122, 123</sup>. Clinical guidelines and recommendations for best-practice care have been developed to help health care professionals in attending to the needs of patients that are facing the end of life<sup>124-127</sup>. Despite this, studies have shown that it is challenging to integrate this knowledge into the clinical practice<sup>128-130</sup>. New structures are needed to assist the integration of high-quality palliative care at all levels of the health care system<sup>130</sup>.



# Rationale

Every year many children and adolescents lose a parent, exposing them to a high risk of negative consequences. Family cohesion appears to be an important factor supporting their wellbeing after the loss. Previous studies, based on the same research dataset as used in this PhD project have shown that poor family cohesion was the strongest predictor for the risk of self-injury<sup>21</sup>, unresolved grief<sup>67</sup> and distrust of the health care provided to the dying parent<sup>24</sup>. However, how family cohesion is affected over time by the death from cancer of one parent, and what other factors may be associated with poor family cohesion, remain to be studied.

As concluded in a systematic review, there is a need for more studies on the grief processes of bereaved children and adolescents, including factors related to the family system<sup>14</sup>. In another literature review, the authors concluded that more studies are needed to better understand the effect on teenagers' psychological health and wellbeing after parental death<sup>101</sup>. There is a great need to put more focus on health prevention in palliative care services<sup>131, 132</sup> but this has been a neglected area in the research literature<sup>132, 133</sup>, especially when it comes to children and adolescents<sup>131, 134, 135</sup>. It is evident that, to be able to resourcefully support bereaved children and adolescents, it is important to better understand their experience after losing a parent, and the factors in their environment that may affect their long-term health and wellbeing, as well as to get more knowledge about their need for support.

However, gaining an evidenced-based knowledge will not be of much help to patients and their families if the knowledge is not applied in clinical practice. It happens too often that evidenced-based knowledge is not translated to and used in real-life situations<sup>128, 129, 136-138</sup>, and that the access to good palliative care is not equally available according to needs<sup>130</sup>. The need for quality improvement within palliative care has been highlighted by the Swedish health authorities<sup>126, 136</sup>. According to the National Board of Health and Welfare in Sweden, there is a shortage of formal support to children and their families who have lost a parent to a sudden death<sup>139</sup>. In view of a lack of routines regarding children as family, there is a great risk that no one even asks whether there are children among the ones closest to a dying person<sup>139, 140</sup>. This is the case despite a new law that was approved by the Swedish health authorities in 2010, stating that every child has the right to be recognized as next of kin and to be provided with the information and support needed, when an important adult in their life is seriously ill, dealing with addiction or dying<sup>12, 13</sup>.

For health care to be of good quality, it is necessary that it is evidence-based and in harmony with the needs of the users<sup>141</sup>. Furthermore, in order to provide holistic care for the dying patient, with a family focus and capturing the needs of the children in the family, health care services need to adapt clinical routines that are based on what is recommended as best practice in palliative care. Not everyone has the opportunity to receive specialized palliative care<sup>130</sup> and the holistic approach that defines palliative care should not only be limited to specialized services but should be integrated into the whole health care system. All health care personnel should be able to provide care to people with palliative care needs, when faced with them, regardless of whether this is in the general practice, at a care home, or in a hospital ward. This includes integrating the needs of the family, and of the children in the family, into clinical practice.

# Aims

## Overall aim

The overall aim was to describe acute and long-term grief and family cohesion, as perceived by young adults (aged 18–26) who as teenagers (aged 13–16) faced the death of a parent to cancer, and its association with long-term health and wellbeing.

A further aim was to describe the development of a comprehensive clinical guidance for holistic care of patients with palliative care needs, including structured support for health care personnel to attend to the needs of children in the family.

## Specific aims

*Study I* – The aim of Study I was to investigate the levels of perceived family cohesion during childhood, the teenage years, and young adulthood in youths who had lost a parent to cancer in their teenage years, compared with their non-bereaved peers.

*Study II* – The aim of Study II was to investigate grief experiences and reactions in the acute bereavement phase, and their possible associations with long-term unresolved grief, as self-assessed by cancer-bereaved youths. Also, to explore associations between demographic, family, and health care-related factors and the experience of having had an okay way to grieve in the first 6 months post-loss.

*Study III* – The aim of Study III was to investigate the level of self-reported family cohesion during the first year after the death of a parent to cancer and its association with long-term psychological health and wellbeing among young adults bereaved during their teenage years.

*Study IV* – The aim of Study IV was to describe the development and feasibility testing of the Swedish Palliative Care Guide (S-PCG), which entails a comprehensive support for palliative care in clinical practice, including structured support for health care personnel to identify children in the family and further identify their need for information and support.

# Methodology

This PhD project was divided into two parts based on the method and time of data collection. The overall design and methods are summarized in Table 2.

**Table 2.** Overview of the methods used in Studies I–IV.

|                                   | Part 1  |                                      |   | Part 2   |
|-----------------------------------|---|--------------------------------------|---|--|
| Study                             | I   | II                                   | III   | IV   |
| <b>Study design</b>               | Nationwide population-based study, cross-sectional design                                     |                                      |   | Exploratory intervention development study guided by the interpretive description for applied practice approach  |
| <b>Sample</b>                     | Cancer-bereaved participants (n=622)<br>Non-bereaved participants (n=330)                     | Cancer-bereaved participants (n=622) | Cancer-bereaved participants (n=622)                      | National interdisciplinary advisory committee (n=95 members, 32 of whom participated in a workshop)<br>Health care personnel from 40 care units participating in the pilot tests (n=345), 147 of whom participated in focus group interviews<br>Patients involved in the piloting of the Swedish Palliative Care Guide (n=300)<br>Patients (n=4) and family members (n=5) participating in semi-structured interviews<br>Patient, family, and public representatives participating in focus group/small group interviews (n=13)<br>Other health care professionals/experts participating in focus group/small group/individual interviews (n=49) |
| <b>Data collection methods</b>    | Study-specific questionnaire  |                                      |   | Collection of written feedback<br>Focus group/small group/semi-structured interviews<br>Review of the patient records  |
| <b>Methodological models used</b> | A hierarchical step-model for causation of bias   |                                      |   | The Medical Research Council (MRC) framework<br>The 10/40 model developed by the International Collaborative for the Best Care for the Dying Person<br>Ten-Step Implementation Model from the International Collaborative for the Best Care for the Dying Person   |
| <b>Data analysis</b>              | Bivariable and multivariable logistic regression<br>Forward selection (likelihood ratio test) |                                      | Modified Poisson regression<br>Direct analytical diagrams | Thematic analysis inspired by Braun and Clarke   |

## Method, Part 1 (Studies I–III)

### Design

Part 1 of this PhD study had a cross-sectional design and is a part of a quantitative nationwide population-based research project on youth who as teenagers, 6–9 years previously, had lost a parent to cancer, and a matched random sample of non-bereaved controls<sup>2</sup>. The study was initiated and performed by a research group at the Division of Clinical Cancer Epidemiology at Karolinska Institute in Stockholm and the Department of Research and Development at Stockholm's Sjukhem Foundation. The comprehensive dataset used in this PhD project had already been collected prior to the onset of this study, in the years 2009–2010, but the data under discussion here had not been previously analysed.

#### *A hierarchical step-model for causation of bias*

A hierarchical step-model for causation of bias was used in the design of the study<sup>142</sup>. When dealing with real-life people in complex systems such as existing in the family and the health care system, there are many factors that need to be taken into consideration that can affect the research. The hierarchical step-model for causation of bias has been developed to help researchers when performing clinical epidemiological studies<sup>142</sup>. It was used in Part 1 of this PhD project as a structure to carefully think through and design every step of the study while having possible biases in mind, with the aim to try to minimize as much as possible the likelihood of them occurring.

### Participants

#### *Cancer-bereaved participants (Studies I–III)*

To be considered for participation in Part 1 of this research, the individual had to have lost a parent to cancer in the years of 2000–2003 when they were at the age of 13–16. The parent had to have been younger than 65 years old and to have been diagnosed with cancer at least 2 weeks before their death. Participants had to be born in Sweden or one of the Nordic countries and to have the same registered address as both parents at the time of death, and the surviving parent needed to be alive at the time of follow-up. All participants needed to have an identifiable telephone number and to be able to read, write and understand Swedish and live in Sweden at the time of the study. Using information about the lost parents from the Swedish National Causes of Death Register, the participants were identified through the Multi-Generation Register at Statistics of Sweden<sup>2</sup>.

### *Non-bereaved participants (Study I)*

A random sample from the general population was identified by the Statistic of Sweden in the proportion of 1:2 (non-bereaved:cancer-bereaved). The non-bereaved comparison group was matched by age, gender and place of residency to the cancer-bereaved group. They had to have been born in a Nordic country and have an identifiable telephone number. Additionally, to be eligible both parents had to be alive and live together<sup>2</sup>.

## **Questionnaire development**

Data were collected through a questionnaire asking about the participants' background and experiences before and after losing a parent as well as about their current situation, health and wellbeing. The questions were based on topics that had been brought up in semi-structured interviews with cancer-bereaved adolescents and youth (13–26 years old), as part of the questionnaire development process. Further questions were inspired by previous questionnaires from the research group, and by the bereavement literature and interviews with professionals and experts. Validity tests were performed face to face with volunteers after responding to an advertisement in a newspaper<sup>2</sup>.

The questionnaire contained 153 questions on 44 pages with mostly categorized response options but also some possibilities for open responses. The questionnaire for non-bereaved participants was 20 pages long and was identical to the questionnaire for cancer-bereaved participants, apart from the exclusion of questions related to cancer disease, experience of the health care provided, and bereavement<sup>2</sup>. The order of the questions was carefully decided, i.e. most of the questions on health and wellbeing preceded the questions about the period of parental illness, death and bereavement, to minimize the risk of painful memories affecting the answers<sup>2</sup>.

## **Measurements**

Since the aim was to study the participants' subjective experiences after losing a parent to cancer, the questionnaire formulation was mostly designed applying the well-established approach described in previous articles from the research group<sup>143-145</sup>, using one direct question per phenomenon. This involves using single-item questions to directly ask about the real-life issue at hand instead of using a multiple-item instrument to indicate a certain phenomenon. However, for some of the phenomena under investigation, if suitable well-established measurements were available and had been validated for the target group, they were included in the questionnaire such as the Patient Health Questionnaire 9 (PHQ-9).

Table 3 gives an overview of the main questions used in Part 1 of this PhD study, together with the response options.

**Table 3.** Overview of the main questions used in the studies making up Part 1 of the PhD project

| QUESTIONS  | RESPONSE OPTIONS   | USED IN:                       |
|--|--|--------------------------------|
| <b>FAMILY COHESION</b>   |  |                                |
| <p>Did you as a family have good cohesion:</p> <p>a) During your childhood?</p> <p>b) During your teenage years (until the death of your parent)?*</p> <p>c) 0–6 months after the death of your parent?*</p> <p>d) 7–12 months after the death of your parent?*</p> <p>e) And do you have good cohesion today?</p> <p>*For the non-bereaved participants, alternatives b, c and d were replaced by the alternative: During your teenage years?</p>   | <p>No, not at all</p> <p>Yes, a little</p> <p>Yes, moderately</p> <p>Yes, very much.</p>         | <p>Studies I, II &amp; III</p> |
| <b>ACUTE GRIEF EXPERIENCES AND REACTIONS</b>   |  |                                |
| <p><b>An okay way to grieve</b><br/>I had a way to grieve that felt okay</p> <p><b>Numbing and postponing</b><br/>I clenched my teeth, built a wall around me and lived on as if nothing had happened</p> <p><b>Concealed grief</b><br/>I withheld my grief to protect my other parent</p> <p><b>Overwhelmed by grief</b><br/>The grief was so strong it felt as if I would not survive, as if I was going crazy or was not normal</p> <p><b>Discouraged from grieving</b><br/>People stopped me from grieving by drawing away when I was sad or praising me when I was being strong</p> <p><b>Pressured to grieve</b><br/>There was pressure from others that I should be more sad than I was showing</p> | <p>Do not agree at all</p> <p>Slightly agree</p> <p>Moderately agree</p> <p>Completely agree</p> | <p>Study II</p>                |
| <b>LONG-TERM GRIEF RESOLUTION</b>  |  |                                |
| <p>Have you worked through your grief?</p>   | <p>No, not at all</p> <p>Yes, a little</p> <p>Yes, moderately</p> <p>Yes, completely.</p>        | <p>Study II</p>                |
| <b>HEALTH RELATED MEASUREMENTS</b>   |  |                                |
| <p><b>Wellbeing</b><br/>Have you experienced high levels of wellbeing in the last month?</p> <p><b>Quality of life</b><br/>Have you had a good quality of life in the last month?</p>  | <p>No, not at all</p> <p>Yes, a little</p> <p>Yes, moderate</p> <p>Yes, very high/good</p>       | <p>Study III</p>               |

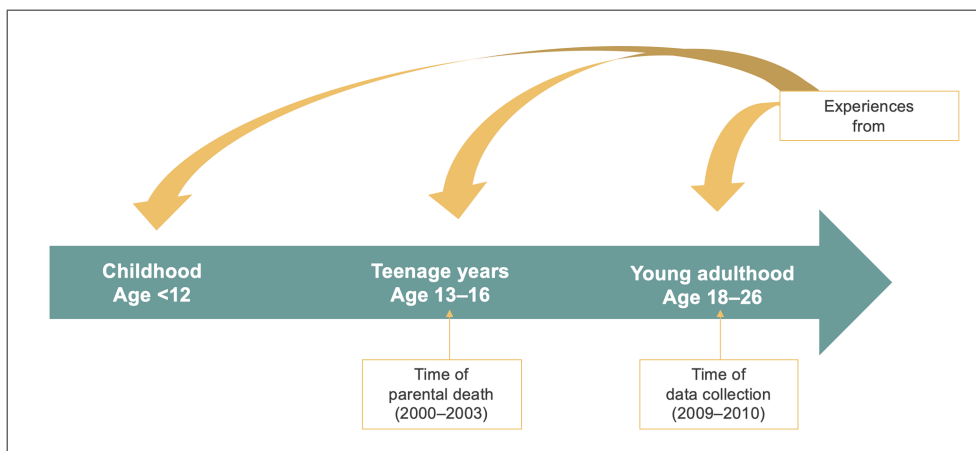


Table 3. *Continued.*

| QUESTIONS USED   | RESPONSE OPTIONS  | USED IN          |
|--|---|------------------|
| <b>HEALTH RELATED MEASUREMENTS</b>   |   |                  |
| <p><b>Depression (Patient Health Questionnaire 9 (PHQ-9) scale)</b><br/>           Over the last 2 weeks, how often have you been bothered by any of the following problems?</p> <ol style="list-style-type: none"> <li>1) Little interest or pleasure in doing things?</li> <li>2) Feeling down, depressed or hopeless?</li> <li>3) Trouble falling or staying asleep, or sleeping too much?</li> <li>4) Feeling tired or having little energy?</li> <li>5) Poor appetite or overeating?</li> <li>6) Feeling bad about yourself – or that you are a failure or have let yourself or your family down</li> <li>7) Trouble concentrating on things, such as reading the newspaper or watching television</li> <li>8) Moving or speaking so slowly that other people could have noticed? Or the opposite – being so fidgety or restless that you have been moving around a lot more than usual?</li> <li>9) Thoughts that you would be better off dead, or of hurting yourself in some way?</li> </ol> | <p>Not at all (0 points)</p> <p>Several days (about 1–3 days a week) (1 point)</p> <p>More than half the days (about 4–5 days a week) (2 points)</p> <p>Nearly every day (6–7 days a week) (3 points)</p> <p>(Moderate to severe depression: total score <math>\geq 10</math> points)</p> | <p>Study III</p> |
| <p><b>Symptoms of anxiety</b><br/>           Have you felt persistent worries (fear, anxiety) in the last month?</p> <p>Have you been unable to stop worrying or to control your worries in the last month?</p> <p>In the last month, have you felt like something terrible is about to happen?</p> <p>Have you had sudden attacks of anxiety (fear) in the last month?</p> <p><b>Problematic sleeping</b><br/>           Have you had trouble falling asleep at night in the last month?<br/>           Have you woken up during the night with anxiety or unpleasant feelings in the last month?</p> <p><b>Emotional numbness</b><br/>           Have you felt emotionally numb (cut off, like you were in a bubble or had a wall around you) in the last month?</p>   | <p>No</p> <p>Yes, occasionally</p> <p>Yes, approximately 1–3 days a week</p> <p>Yes, approximately 4–5 days a week</p> <p>Yes, approximately 6–7 days a week</p>  | <p>Study III</p> |

## Data collection

The data collection for Part 1 was conducted from February 2009 to March 2010. First, an information letter was sent to all participants who met the inclusion criteria. This was followed by an information call from a research assistant. If during the information phone call the person gave their oral consent to participate, the questionnaire and ethics information sheet was sent. A separate envelope for the reply card was sent with the questionnaire to keep the answers of the participants anonymous. Afterwards a thank you card was sent, or a reminder card following a reminder phone call to those who had not returned their questionnaire<sup>2</sup>. Figure 1 visualizes the time of parental death, as well as the periods childhood, the teenage years, and young adulthood in the participants' life, which they were asked about in the study questionnaire, and also the time of data collection in Part 1 of the PhD project.



**Figure 1.** Overview of the periods in the participants' life asked about in the questionnaire for Part 1 of the PhD project.

## Data analysis

In Part 1, associations were assessed using univariable and multivariable logistic regression (*Studies I and II*) or with modified Poisson regression (*Study III*). The results are presented as prevalence numbers and odds ratios (ORs) (*Studies I and II*) or relative risk/risk ratios (RRs) (*Study III*) with corresponding confidence intervals (CIs) set at 95%.

As previously described, the study design followed Steineck's hierarchical step-model for causation of bias<sup>142</sup> to minimize the risk of systematic errors (bias) and confounding. Confounding is caused by a third variable that is unevenly distributed between the groups and is associated with both the exposure variable as well as the outcome variable<sup>146</sup>. Based on the literature, previous research and discussions with bereavement experts, possible confounding variables were selected for each study (*Studies I–III*) after a thorough discussion within the research group. In Studies I and II, forward selection (likelihood ratio test) was performed on the pre-selected variables, while direct analytical diagrams (DAGs) were used in Study III, to build the final adjustment models. Adjustments were then made to control for possible confounding through regression models that are described in more details in Studies I–III. Table 4 provides an overview of the dependent and independent variables that were used in Studies I–III of Part 1, as well as possible confounding variables that were considered.

**Table 4.** Overview of the dependent, independent and possible confounding variables used in Studies I–III

| Study                                 | I   | II   | III   |
|---------------------------------------|---|--|---|
| <b>Independent variables</b>          | <ul style="list-style-type: none"> <li>▪ Cancer-bereaved participants (whole group)</li> <li>▪ Paternally bereaved participants</li> <li>▪ Maternally bereaved participants</li> <li>▪ Non-bereaved participants</li> </ul>   | <ul style="list-style-type: none"> <li>▪ Having had an okay way to grieve</li> <li>▪ Numbing and postponing</li> <li>▪ Concealed grief</li> <li>▪ Overwhelmed by grief</li> <li>▪ Discouraged from grieving</li> <li>▪ Pressured to grieve</li> </ul>  | <ul style="list-style-type: none"> <li>▪ Family cohesion in the first year (0–12 months) after the death of a parent.</li> </ul>  |
| <b>Dependent variables</b>            | <ul style="list-style-type: none"> <li>▪ Family cohesion during childhood</li> <li>▪ Family cohesion during the teenage years before the death of a parent*</li> <li>▪ Family cohesion during the teenage years 0–6 months after the death of a parent*</li> <li>▪ Family cohesion during the teenage years 7–12 months after the death of a parent*</li> <li>▪ Family cohesion in young adulthood</li> </ul> <p>*Or for non-bereaved participants: Family cohesion during the teenage years</p>  | <ul style="list-style-type: none"> <li>▪ Long-term grief resolution</li> </ul>   | <ul style="list-style-type: none"> <li>▪ Low wellbeing in the last month</li> <li>▪ Low quality of life in the last month</li> <li>▪ Moderate to severe depression in the last two weeks</li> <li>▪ Symptoms of anxiety once a week or more in the last month</li> <li>▪ Problematic sleeping once a week or more in the last month</li> <li>▪ Emotional numbness once a week or more in the last month</li> </ul>  |
| <b>Possible confounding variables</b> | <p><b>Background variables:</b></p> <ul style="list-style-type: none"> <li>▪ Gender of the participant</li> <li>▪ Year of birth</li> <li>▪ Residential region</li> </ul> <p><b>Family-related variables:</b></p> <ul style="list-style-type: none"> <li>▪ Number of siblings</li> <li>▪ Birth order</li> <li>▪ Mother's year of birth</li> <li>▪ Father's year of birth</li> <li>▪ Educational level of mother</li> <li>▪ Educational level of father</li> <li>▪ Ever been bereaved of a sibling</li> <li>▪ Depression in at least one parent</li> <li>▪ Alcohol/drug misuse by at least one parent</li> </ul> <p><b>Adverse events:</b></p> <ul style="list-style-type: none"> <li>▪ Having been bullied</li> <li>▪ Having been physically assaulted or sexually violated</li> <li>▪ Having ever been diagnosed with depression</li> </ul> | <p><b>Demographic variables:</b></p> <ul style="list-style-type: none"> <li>▪ Gender of the participant</li> <li>▪ Age at loss</li> </ul> <p><b>Family-related variables:</b></p> <ul style="list-style-type: none"> <li>▪ Worry about the surviving parent</li> </ul> <p><b>Health care-related variables:</b></p> <ul style="list-style-type: none"> <li>▪ The teenager's perception of the health care professionals' efforts to cure the parent</li> <li>▪ The teenager's perception of the health care professionals' efforts to prolong the parent's life</li> <li>▪ Whether the family had been given end-of-life information about the disease, treatment and death by a physician</li> <li>▪ Whether the teenager had talked with their dying parent about what was important</li> <li>▪ Awareness time when the teenager had realized that the parent would die from the disease</li> <li>▪ Awareness time when the teenager had realized that death was imminent (hours or days)</li> </ul> | <p><b>Background variables:</b></p> <ul style="list-style-type: none"> <li>▪ Gender of the participant</li> <li>▪ Age at loss</li> <li>▪ Year of birth</li> </ul> <p><b>Family-related variables:</b></p> <ul style="list-style-type: none"> <li>▪ Depression in at least one parent</li> <li>▪ Number of siblings</li> <li>▪ Educational level of the surviving parent</li> <li>▪ Alcohol/drug misuse by at least one parent</li> </ul> <p><b>Adverse events and awareness time of parent's imminent death:</b></p> <ul style="list-style-type: none"> <li>▪ Having been physically assaulted or sexually violated</li> <li>▪ Having been bullied</li> <li>▪ Awareness time when the teenager had realized that the parent would die from the disease</li> </ul> |

## Method, Part 2 (Study IV)

### Design

Part 2 of the PhD study revolved around the development of the Swedish Palliative Care Guide (S-PCG) and its extensive feasibility testing in various health care settings, during three pilot test periods. It is an exploratory intervention development study and was built on the approach of interpretive description for applied practice. The Medical Research Council (MRC) framework was used to provide a robust structure for the development of the S-PCG and the content is in coherence with the 10/40 model developed by the International Collaborative for Best Care for the Dying Person.

#### *Exploratory intervention development study*

The purpose of an intervention development study is to describe what happens from the birth of an idea until an intervention is ready for full trial or evaluation<sup>147</sup>. In other words, it is to describe the development process of an intervention or a tool, the rationale that lies behind it, how decisions were made and what methods were used, together with the findings that were made along the way<sup>147</sup>.

Exploratory studies can be defined as studies that aim to generate evidence that is needed for making informed decisions on whether to proceed with a full-scale intervention or implementation of a tool or working approach that is being developed or to continue with full-scale research that is being planned<sup>148</sup>. These types of exploratory studies are also often named, “feasibility” or “pilot studies” and can also be used to refine an intervention that is being developed<sup>148</sup>.

Similarly to suggestion of Eldrige et al<sup>149</sup>, we considered a pilot test to be a subset of a feasibility study; however, the use of the term “pilot test” in this PhD study, should not be confused with the definition of “pilot study” which is often narrowed to describing a small-scale version of a randomized controlled trials<sup>148</sup>. In their guidelines for non-randomized pilot and feasibility studies reporting, Lancaster and Thabane<sup>150</sup> highlight that pilot studies can also be used to test the feasibility of a non-randomized study or they can be used in different types of feasibility studies such as in intervention development studies or when the aim is to implement research findings into clinical practice<sup>150</sup>. Study IV describes the development and feasibility testing of the S-PCG, which was conducted during three pilot test periods and can be described as having an exploratory intervention development study design.

### *Interpretive description for applied practice*

Interpretive description is a methodological approach that was used to guide the design and the process for Part 2 of this PhD study. This approach can be used as a framework for decisions about the design and execution of high-quality studies in the field of applied practices<sup>1, 151</sup>. Studies based on the interpretive description approach are performed in a naturalistic context and acknowledge that the human knowledge is socially constructed and cannot be estranged from the essential nature within which it is located. Interpretive description acknowledges that the human experience is based on multiple constructed realities and it is in the interaction between the researcher and the participants where the knowledge is produced in partnership<sup>1</sup>. It is positioned within the epistemology of clinical discipline, highlighting that it is the health care discipline that forms what questions are asked and why, and how these questions are relevant to the study. It is based on pattern recognition and accepts an infinite variation of patterns<sup>1</sup>. In interpretive description the complexity of conducting a research in the field of health and health care is acknowledged<sup>1, 151</sup>. The researcher is encouraged to use reflection and critical thinking in every step of their research. According to the approach, researchers need to be willing to adjust their research to best serve the aim of the study rather than strictly follow a theoretical method that might not be suitable to answer the clinically relevant research question. Thus they can create knowledge that addresses “real life” challenges of the applied practices<sup>1</sup>. Interpretive description highlights that one a priori theory cannot encompass the complex realities of the real-life experiences, although the researchers are allowed to use established theories, research methods and models if they are perceived to be helpful for achieving their research aim. At the same time the researcher is encouraged throughout the research process to actively reflect on the chosen method/model and make appropriate adjustments to best serve the aim of the study<sup>1</sup>.

### *The Medical Research Council framework*

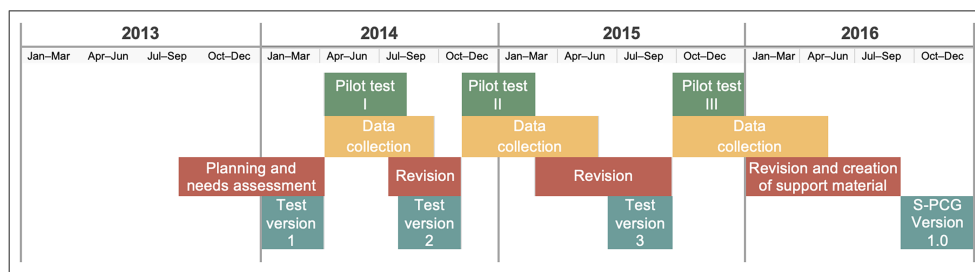
The Medical Research Council (MRC) framework was used to provide a basic structure for the development of the S-PCG. The MRC provides a framework in several steps for consideration during the development of an intervention in a complex setting<sup>152, 153</sup>. Table 5 describes the MRC framework actions that were used as an inspiration for the steps taken during the development of the S-PCG.

**Table 5.** Overview of the Medical Research Council (MRC) framework actions in relation to the steps taken during the development of the Swedish Palliative Care Guide (S-PCG).

| <b>MRC framework actions that inspired the development of the S-PCG<sup>152</sup></b>        | <b>Steps taken during the development of the S-PCG in relation to the MRC framework actions</b>   |
|--|---|
| <b>Plan the development process and identify the need</b>                                    | To identify the need for the S-PCG, several clinical field observations were performed at different units caring for patients with palliative care needs, where the need for support in clinical practice was expressed by the health care professionals. Additionally, patient record audits were made, supporting the need for actions to improve the quality of palliative care.   |
| <b>Involve stakeholders, bring together a team and establish the decision-making process</b> | Stakeholder analysis was made and a steering committee and a project group were formed including people with both academic and clinical expertise. Patient and public representatives were assigned to the project and a national interdisciplinary advisory team was formed, consisting of individuals with expertise and experience relevant to the project. A structure for the work and the decision-making process was established.  |
| <b>Review published research evidence and draw on existing theories.</b>                     | A recent literature review had recently been conducted by the health authorities and two reports with national recommendations and guidelines for palliative care had been published at the start of the project <sup>126, 154</sup> . They were used as a basis for the design of the S-PCG, together with relevant national quality indicators <sup>155, 156</sup> and an international care guide <sup>157</sup> . "The "10/40 model", developed by the International Collaborative for Best Care for the Dying Person <sup>158</sup> , was also used to guide the development of the S-PCG.   |
| <b>Draw on existing theories and articulate a programme theory</b>                           | Apart from basing the S-PCG on the 10/40 model, the four phases and the ten steps of the "Ten-step implementation model" of the International Collaborative for Best Care for the Dying Person <sup>159</sup> were used to guide the design of the implementation and the feasibility testing of the S-PCG. Langley's model for improvement including the "Plan-Do-Study-Act" (PDSA)" cycle <sup>160</sup> was used to plan, and reviewed and adjust the S-PCG project continuously throughout the development process. An initial model for the S-PCG was drawn up by the steering committee and the project group and was continuously reflected on, refined and redesigned throughout the project. |
| <b>Undertake primary data collection</b>   | An extensive data collection was performed during the development of the S-PCG, which took over 3 years and included three pilot test periods. The result of each pilot test period generated knowledge that informed the next steps that needed to be taken.   |
| <b>Understand the context and design and refine the intervention</b>                         | The national interdisciplinary team, patient- and public representatives and a broad variety of experts were included in the development process and gave feedback throughout the development process. The S-PCG was reviewed and refined after each pilot test based on the feedback given, to make sure that it was applicable to different health care settings. In total, the S-PCG had been tested in 40 different care units, involving 300 patients.   |
| <b>End the development phase</b>   | After broad and extensive testing, the S-PCG was launched in the autumn of 2016. The S-PCG documents are, however, continuously evolving. Based on new evidence and feedback from the users they will be reviewed regularly and new versions will be launched when needed. Further research has been planned and initiated with the aim to create a strong evidence base for the use of the S-PCG in the future.  |

## Setting

Part 2 of this PhD project arrives from a comprehensive national health care quality improvement project. The Institute for Palliative Care in Lund initiated the development of the S-PCG in 2013 by establishing a steering committee which included academic professors, experienced in the palliative care field (n=4) and a registered nurse with the role of quality improvement coordinator at the institute who was the project manager (n=1). The steering committee supervised the project and provided strategies for design and planning. Based on a stakeholder analysis, a project group (n=10) was formed, including both clinical and academic expertise and competence considered relevant to the project. The role of the project group was to model the content of the new care guide and lead its testing. Figure 2 illustrates the timeline of the project, from the project start until the first version of the S-PCG was launched in 2016.



**Figure 2.** Timeline overview of the development of the Swedish Palliative Care Guide (S-PCG).

The S-PCG is aimed for use in the Swedish health care sector. The content of the S-PCG is based on national<sup>126, 154</sup> and international recommendations and guidelines. As mentioned the content was inspired by and mapped to the 10/40 model from the International Collaborative for the Best Care for the Dying Person. The 10/40 model provides a description of the ten key principles and the 40 core elements, that together encompass good palliative care and should be included in the care for a dying person<sup>158, 161</sup>. They are based on the holistic approach that should characterize palliative care and include principles such as recognizing that the person is in the last few days and hours of life and that the dying person and those of importance to that person should have the opportunity to discuss, with the caregivers, their wishes, feelings and what is important to them<sup>162</sup>. The 40 elements underpin the ten principles of good palliative care and can be used as quality indicators for good palliative care. The S-PCG was tested for concurrency by the International Collaborative for the Best Care for the Dying Person.

The S-PCG was designed to put the palliative care needs of the individual in the forefront, irrespective of diagnosis, and should be feasible to use throughout the palliative trajectory, in all relevant care forms, where individuals with potential palliative care needs are being cared for.

## **Participants**

Purposive sampling was used to collect data from a broad group of participants with experiences representing different aspects of the Swedish health care settings. Participants were recruited as the project evolved, when needed.

### *National interdisciplinary advisory committee*

A national interdisciplinary advisory committee was established, starting with a group of 40 individuals but successively increasing membership to a total of 95 individuals. The committee were given the opportunity to review each version of the S-PCG during the development process.

### *Patient, family and public involvement*

A total of 22 patient, family and public representatives participated in the project during the development of the S-PCG. They were recruited through a patient and family organization, two senior organizations, and participating health care units. Furthermore, a total of 300 patients were involved in the clinical testing.

### *Health care personnel during the feasibility testing*

A total of 345 health care personnel, from a total of 40 health care units that were involved in the clinical testing of the S-PCG, participated in a whole-day educational programme prior to the pilot testing of the S-PCG. Feedback, both in the form of written comments during the pilot testing and through focus group interviews (n=147) at each unit after each pilot test period, was gathered from the health care personnel. Each unit was assigned to find participants among the personnel, willing to take part in the focus group interviews, that had been involved in the pilot test. To get a broader perspective on the feasibility of the S-PCG, they were encouraged, if possible, to include different professions in the focus group interviews.

### *Other experts who were identified as relevant to the development of the Swedish Palliative care Guide*

Throughout the project, other experts (n=49) were invited to review the content of the S-PCG documents. Some provided feedback that was gathered through focus groups meetings, workshops or small group interviews, while others were asked to give written feedback on the content. They were selected through purposive sampling when a need for their input had been identified. For example, after pilot test II it was clear that assistant nurses were underrepresented in the focus group interviews. This led to the decision to invite assistant nurses to a specific focus group discussion, to gain their perspective on the content of the S-PCG.



## Data collection

Data were collected in several ways throughout the S-PCG development process, during three pilot test periods involving 40 care units, in municipal care, hospital care and specialized palliative care in different areas of Sweden. An overview of the process of the development of the S-PCG is presented in Figure 3. The Figure also illustrates the sources of data collection. The data included written feedback, and data collected during focus group/small group/individual interviews, and workshops with an interdisciplinary team from each test unit, the national interdisciplinary advisory committee, and patient, family and public representatives, as well as professionals and experts from other clinical settings. Data were also collected through semi-structured interviews with patients and their family members; and a review of patient records used during the pilot tests of the S-PCG. The collection of the feedback was categorized under the following items:

- Relevance of the content
- Usefulness/user-friendliness
- Missing issues
- Redundancy
- Teamwork/communication
- Patient and family involvement
- Implementation

An overview with examples of the items checked/questions for each of these categories during the feasibility test of the S-PCG is presented in *Paper IV, Supplementary Table B*.

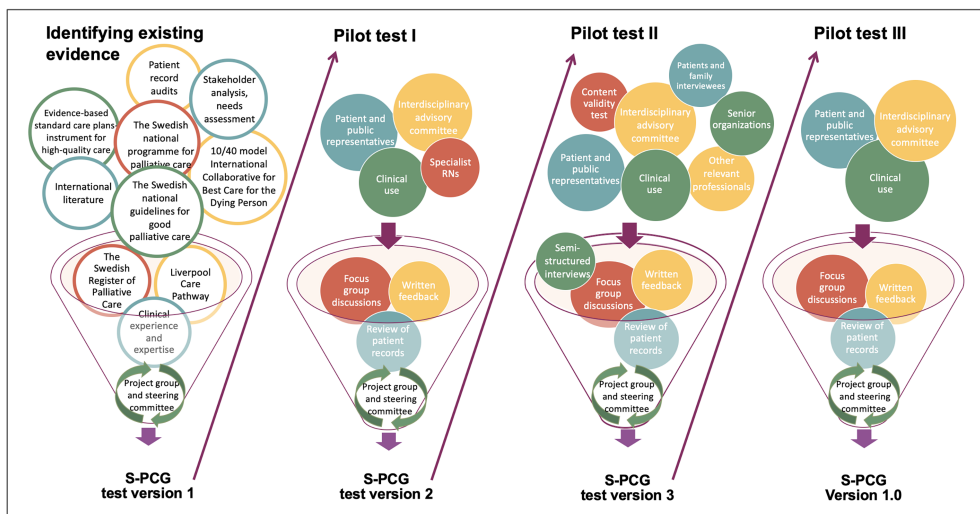


Figure 3. Overview of the development process of the Swedish Palliative Care Guide (S-PCG) (Study IV<sup>163</sup>, p. 3).

### *Written feedback*

Written feedback was collected throughout the development process. The clinical test users were provided with a special feedback sheets, available throughout each pilot test period, where they could write their comments based on the categories above. Special copies of the S-PCG documents were also printed out and made available to the test users to write feedback directly on the documents during the test period. These comments were collected from the whole team anonymously but the personnel also had the opportunity to leave their own feedback, on paper or through e-mail if preferred.

Written feedback was also collected from the national interdisciplinary advisory committee, patient, family and public representatives and other experts in the field. They were provided with special feedback sheets in Excel format, to give their feedback on each section of the S-PCG based on the abovementioned categories. They also had the option to write comments directly on a copy of the S-PCG documents or to send us their comments through the mail or via e-mail.

### *Semi-structured interviews*

A semi structured interview guide was created by the members of the project group, including the PhD student, under the guidance of an experienced researcher in the steering committee. Questions in the interview guide were formulated with focus on gathering the patients' and their family members' opinions about the content of the S-PCG. This included questions such as "*Would you like to tell us what you think about us asking how daily life works for you?*" followed by questions such as "*Do you think that this topic is relevant/important to you?*" or "*Have any of the issues we've discussed today felt inappropriate or offensive?*" and "*Is there anything else that you think is important for health care professionals to ask for or attend to, that we have not discussed?*". Follow-up questions were asked to encourage further description.

A total of nine semi-structured interviews were performed, four with patients and five with a family member the patient had nominated. One patient did not have the energy to participate in the interview, but gave consent for the family member interview to take place. The patients were recruited from care units that participated in the pilot project. Two nurses from the project group, who worked at these care units performed the interviews together. They both had long experience in palliative care. The nurses took field notes and summarized the interviews.

### *Focus group interviews*

Focus group interviewing is a qualitative research method that can be used to collect data on how a certain group make sense of a certain topic that is under investigation. It derives from the field of social science and has been used to investigate interaction within a group setting<sup>164</sup>, it can also be used to gain insight into a range of opinions about the issue of focus<sup>165</sup>. Focus groups are also commonly used to gain an insight into how a new service or product is perceived by the group and to identify potential problems, which can help with further design of the service or product<sup>164</sup>.

Focus group interviews were performed with the clinical test users, patient, family and public representatives, and other experts relevant to the field of palliative care. A focus group interview guide was designed and formulated by the project manager and the PhD student, under the guidance of the more experienced researchers in the steering committee, inspired by Krueger and Casey<sup>166</sup>. At least two moderators were present at each focus group session, and the PhD student was involved as a moderator in many of the focus groups with the different types of participants. During the first focus groups and in all, except one, of the focus groups with the patient, family and the public representatives, a more experienced researcher was also present to assist with the moderation of the session. In pilot test III, involving 35 care units at a nationwide basis, a group of moderators (n= 10, of which four were from the project group) were trained under the lead of one of the more experienced researchers in the steering committee. Their role was to assist with the implementation of test-version 3 and to help with conducting some of the focus group interviews with the clinical test users. In some cases, persons from care units working under the same management or that were situated close to each other, participated in a joint focus group interview.

Field notes were taken during all focus groups, complemented by reflection notes afterwards. For some of the focus groups, the sessions were audiotaped. The focus group interviews took place in facilities set aside for this purpose at the different care units, or in meeting rooms at the Institute for Palliative Care. Pragmatic variations in the size of the groups were allowed. Irrespective of size they all followed the same structure, as described above for focus group interviews. Interviews with two or three participants will here be referred to as “small group interviews” and those with only one participant as “individual interviews”.

A total of 40 focus group/small group interviews, with two to 16 participants per session, and three individual interviews were performed. Each interview session lasted 60–120 minutes. A total of 198 individuals participated in the focus group/small group/individual interviews. Table 6 gives an overview of the focus group/small group/individual interviews, together with the semi-structured interviews and the workshops that were held during the three pilot test periods.

**Table 6.** Overview of the focus group, small group and individual interviews and workshops performed during the development of the Swedish Palliative Care guide (S-PCG).

| Participants  | Number of sessions and type of data collection    | Number of participants in each session | Total number of participants | Number of participants by occupation  |
|---|---|--|------------------------------|---|
| Clinical test users   | Twenty-seven focus group/small group interviews   | 2–16                                   | n=147                        | Registered nurses (n=81)<br>Physicians (n=20)<br>Assistant nurses (n=23)<br>Unit managers (n=14)<br>Health care quality-development lead (n=1)<br>Physiotherapists (n=3)<br>Occupational therapist (n=3)<br>Social worker (n=1)<br>Dietitians (n=1) |
| National interdisciplinary advisory committee                                       | One workshop with two rounds of group discussions | 6–7 participants in each group         | n=32                         | Registered nurses (n=17)<br>Physicians (n=6)<br>Assistant nurses (n=2)<br>Health care quality and practice development leads (n=2)<br>Physiotherapists (n=2)<br>Occupational therapist (n=1)<br>Dietitians (n=2)                                    |
| Patient, family and public representatives  | Four focus group/small group interviews           | 2–11                                   | n=13                         | not applicable  |
| Physiotherapists in palliative care   | One focus group interview                         | 12                                     | n=12                         | Physiotherapists (n=12)   |
| Assistant nurses  | Two focus group interviews                        | 3–8                                    | n=11                         | Assistant nurses (n=11)   |
| Registered nurses from specialist care units  | Two workshops with focus group discussions        | 5–6 participants in each group         | n=11                         | Registered nurses (n=11)<br>(From specialist care units in cardiology, haematology, home-based palliative care, nephrology, pulmonology, and surgery)   |
| Patients, at a care unit participating in the pilot tests, and their family members | Nine semi-structured interviews                   | One                                    | n=9                          | not applicable  |
| Regional team for strategic development of children as next of kin                  | One focus group interview                         | Seven                                  | n=7                          | Psychologist (n=1)<br>Strategic regional developer (n=1)<br>Social workers (n=2)<br>Sociomists/Psychotherapists (n=3)   |
| Dietitians  | One small group interview                         | Three                                  | n=3                          | Dietitians (n=3)  |
| Municipal care managers   | One small group interview                         | Two                                    | n=2                          | Municipality care-managers (n=2)  |
| Religious/spiritual representative  | One individual interview                          | One                                    | n=1                          | not applicable  |
| International researcher  | One individual interview                          | One                                    | n=1                          | Physician and professor in clinical and palliative medicine (n=1)   |
| General practitioner  | One individual interview                          | One                                    | n=1                          | General practitioner (n=1)  |
|   |   |  | <b>N=250</b>                 |   |

### *Workshops*

Three workshops were held with a total of 43 participants. One whole day workshop was held with the national interdisciplinary advisory committee, involving 32 participants (registered nurses (n=17), medical physicians (n=6), assistant nurses (n=2), physiotherapists (n=2), occupational therapists (n=1), dietitians (n=2) and health care quality and practice development leads (n=2)). The participants were divided into five groups, with six to seven persons per group, and participated in two group discussion sessions of 60–90 minutes each, followed by a 45–90 minute whole-group discussion. The two sessions were separated by a lunch break in a different environment. Each group was given discussion-topics on paper. At the end of the session they were asked to write down as a group the main points from the session. Five researchers/project group members, including the PhD student, were each assigned to one of the groups to observe and take notes during the discussion.

Two half-day workshops were held with nurses (n=11) representing different specialist care units, for example in cardiology, haematology and specialized palliative care. The goal was to collect specific feed-back on the care interventions for part 2 of the S-PCG. The participants were divided into two groups, one group of five and the other of six persons. Two project group members, including the PhD student, were assigned to each group to moderate and to take field notes. Each session lasted for 90–120 minutes and was followed by a 60-minute whole-group discussion. The workshops took place at the Institute for Palliative Care in Lund.

### *Audit of patient records*

After the three pilot tests, a total of 656 S-PCG documents on 300 patients were audited. The used S-PCG documents were copied and anonymized by the health care professionals before they were handed over to the project lead. The documents were reviewed in a structured way by the project manager and the PhD student, with the help of an audit sheet that had been designed for this project. After reviewing the patient records, a summary was made for each care unit, with an overview of:

- the number of S-PCG documents that had been included and the number of patients who had been allocated to each part of the S-PCG
- what questions had been answered/what assessments had been performed and which questions/assessments had been skipped
- any obvious misunderstandings in the use of the S-PCG
- items that had been added that should possibly be included in the S-PCG
- what care-interventions had been used and if new care-interventions had been added
- which profession had documented each item and if there had been a team collaboration

## Creating structure for “children in the family”

Sections in the S-PCG were created to highlight children as members of the family. These sections are based on national and regional guidelines<sup>29, 139, 167-169</sup> and the Swedish laws on health and social services, and patient security, for children as family<sup>12, 13</sup>. The aim of these sections in the S-PCG was to create a structure the health care personnel could use as a support to identify and focus on children as family, to acquire what these children know about the prognosis and diagnosis of their ill family member, and whether they need any support.

Apart from collecting feedback from those involved in the S-PCG project, as described above, the PhD student participated in a project initiated by the Swedish National Board of Health and Welfare to involve a team of experts from each region in Sweden in developing national and regional strategies for children as next of kin. The PhD student used this opportunity to collect feedback on the content of the S-PCG regarding children as family from the other team members from her region (n=7), during one of their meetings. She also sought out other experts working with children as next of kin (n=6) to invite them to review and give written feedback on the content of the S-PCG. They included researchers (n=3) with focus on children’s bereavement, and interventions and support for children and youth; a project leader and researcher working in a national strategy for children as next of kin (n=1); a project leader at the National Board of Health and Welfare for children as next of kin (n=1); and a social worker with long clinical experience in working with children and families facing bereavement (n=1). Couple of social workers involved in the national interdisciplinary advisory committee also gave specific feedback on the sections in the S-PCG concerning children as family (n=2).

## Data analysis

Thematic analysis as described by Braun and Clarke<sup>170</sup> inspired the data analysis in Part 2 of this PhD project. Thematic analysis is part of qualitative research methodology and the term “thematic analysis” can be used as an overarching term to describe the approach to identify patterns of shared meaning or themes in qualitative data<sup>171</sup>.

Inspired by Braun and Clarke’s six-phase process of thematic analysis<sup>172</sup>, the data analysis started with familiarization of the data from the different data sources, where the project manager and the PhD student, each separately read the written feedback and listened to the audio-taped interviews, while taking notes based on their initial thoughts and thereafter creating initial codes. Two more experienced researchers also did the same, after dividing the material between them. Afterwards meetings were held where the research group discussed their take on the material and reflected on the initial coding which had been grouped together in a big matrix, under the categories *Relevance of the content*; *Usefulness/user-friendliness*; *Missing*

*issues; Redundancy; Teamwork/communication; Patient and family involvement; and Implementation*, used to provide a structure for the data analysis.

Thereafter, initial themes were generated from the matrix. Additionally, for the analysis of the comprehensive data from pilot test III, two more experienced researchers from the steering committee helped with the familiarization, the initial coding and the discussion of the basic themes. The basic themes were not seen as fixed but were continuously discussed and reviewed and new themes were developed throughout the analysis process. After the themes had been written out they were discussed in the steering committee and the project group for conceptualization, that is, as a basis for making decisions on what changes needed to be made to improve/further develop the S-PCG and create the next test-version.

# Ethical considerations

When performing a study or a quality improvement project in the health care there are always ethical aspects that need to be evaluated and taken into consideration<sup>173</sup>, especially in palliative care, which can deal with sensitive issues. The research used in Part 1 of this PhD project, involving bereaved young adults who had lost a parent to cancer and a matched sample of non-bereaved peers, was carefully planned and ethical considerations were applied throughout the whole study, following an ethical protocol that had been established by the research group and has been described elsewhere<sup>174, 175</sup>. The study was approved by the Regional Ethical Review Board of Karolinska Institute, Stockholm, Sweden (2007/836-31).

Part 2 of this PhD project involved the development and the feasibility testing of the S-PCG. The S-PCG was modelled in a comprehensive quality improvement project aiming to change clinical practice to better meet patients' palliative care needs<sup>163</sup>, but at the same time to collect knowledge during the process that could be valuable for future implementation and use of the S-PCG. The project design and implementation followed regional and institutional ethical recommendations and guidelines, as well as being guided by the Declaration of Helsinki. Informed consent was gathered from the manager of each care unit participating prior to the feasibility testing of the S-PCG, and the rules and regulations that apply to quality improvements and clinical audits within the health care, were followed, in accordance with the Swedish laws.

The ethical aspects, considerations and procedures in both parts of this PhD project were carefully evaluated, designed and performed with guidance from experienced academic researchers and followed academic standards. The European Code of Conduct for Research Integrity<sup>176</sup>, regarding reliability, honesty, respect and accountability, was reflected in the design and performance of the whole project. Below are some of the ethical considerations that were taken into account regarding this PhD project in relation to some of the basic principles of clinical ethics.



## The principles of beneficence and non-maleficence

The principle of beneficence is about the researcher's/health care personnel's obligation to work on behalf of the participant/patient and to act for the benefit and welfare of the participant/patient. It includes several ethical/moral rules such as to protect/defend the rights of others, prevent harm, and help persons in need<sup>177</sup>. Related to this, the principle of non-maleficence is the obligation to not harm the participant/patient. This includes several moral rules, such as: do not kill, do not cause pain or suffering and do not deprive others of the goods of life. This means that the benefits should always be weighed against the burden or possible risks<sup>177</sup>. This is extremely important in end-of-life care decisions where the goal is to attain the highest possible quality of life and to relieve suffering<sup>177, 178</sup>, not only for the patient themselves but also for their family.

There is a risk of causing distress or harm to participants when asking about sensitive issues such as the experience of losing a parent to death, as in Part 1 of this PhD project. In addition to phrasing questions carefully, the data collection was avoided during holiday seasons or during the month of the anniversary of the parental loss with the purpose of minimizing the risk of causing distress to the participants<sup>2</sup>. Furthermore, the information letter included contact information of the researcher/research assistant and in case the participants needed to talk to someone after answering the questions, a referral to an experienced social worker in the field was possible.

However, it is also necessary to look at the potential benefits of the study and ask whether it is ethical not to collect the present data when the information gathered may result in an improved situation for children and adolescents losing their parent to serious illness in the future. It can also be assumed that those who agreed to participate in the project might possibly even value the researcher's interest in their experiences, as was reported in a study looking into adolescents' and young adults' perception of surveys focusing on sensitive issues, such as surviving cancer<sup>175</sup>. The project was designed to be ethically sound and it was assessed that the potential benefits weighed against any potential harm. In a previous report based on the same data as used in the current study, where free text comments were analysed, the results showed that the cancer-bereaved youth felt it was both valuable to be able to help others and therapeutic to participate in the study about their experience of losing a parent to cancer. This highlights the importance of inviting young adults to participate in research on bereavement<sup>179</sup>. This is supported by the results of two other studies following the same ethical protocol, where the benefits of conducting survey studies on sensitive or trauma-related issues outweighed the risks, when care had gone into the design and the study had been performed with a sensitive approach towards the participants<sup>174, 175</sup>.

The S-PCG, which is the subject in Part 2 of this PhD study, is based on national and international recommendations on the best care for patients with palliative care needs, with the purpose of obtaining the best quality of life and minimizing suffering as much as possible for the patient. Studies have shown that early access to palliative care can both increase quality of life and prolonged life<sup>180</sup>. Since the S-PCG is aimed to support the health care personnel in identifying palliative care needs from early on and throughout the palliative care trajectory and support evidenced-based palliative care, the possible benefits that the implementation of the S-PCG can bring to the patients can be seen. The piloting took place in a well-controlled manner after the health care personnel had received training according to a training programme from the Institute of Palliative Care in Lund. However, as previously mentioned, bringing up palliative care needs, and topics related to end-of-life care, can bring up difficult thoughts and feelings that needs to be processed. These can include thoughts about death, dying and suffering and feelings of vulnerability.

The risk of doing harm by using the S-PCG was assessed to be very small compared with the potential gain, and no bigger than the risk already existing in clinical practice today. The risk was considered to be well outweighed by the potential benefits of developing the S-PCG, which was hoped to lead to greater recognition of palliative care needs among patients and their families, and to put more focus on addressing those needs. Ultimately it was hoped that the project would benefit both the participating patients, in their continuing care, and future patients and relatives.

## The principle of justice

The principle of justice is that all persons have the rights to be treated fairly, and equitably. This could mean, for example, that the distribution of health care resources should be used fair, equitable and appropriate. That does not necessarily mean that every patient should have the same care, but it could mean that every patient should get care according to their needs<sup>177</sup>.

According to the principle of human dignity and the discrimination legislation, access to health care or treatment must not be influenced by factors such as a person's gender, chronological age, social and economic status, previous lifestyle, education or ability to look after their own interests. Anti-discrimination legislation also includes transgender identity, ethnicity, religion or belief, disability and sexual orientation.

The existing bereavement literature has been criticized for having too little focus on other cultures or minority groups of the Western societies<sup>63, 181</sup>. The exclusion of children of single parents and first-generation immigrants from the sample that Part 1 of this PhD project is based on disregards the voices of a vulnerable group of the society, which means that important information may be undisclosed. However, this

exclusion criterion was set to avoid problems that might result from language barriers or the need to assess an even larger set of stressors (e.g. war trauma, refugee experiences, divorce) and other potentially confounding factors. In addition, by ensuring that the children had been living with both parents at the time of death, the possible harm that could arise if some participants did not know about the death of their biological parent before receiving an invitation to the study was avoided. This inclusion criterion was set to address the concerns expressed by the register holders during the planning of the study.

The S-PCG was designed to provide support and structure for the health care personnel when meeting adult patients with palliative care needs, regardless of their age, diagnosis or where the care is provided. Throughout the development of the guide, it has been paramount to support integration of the core values of good palliative care, which highlight the importance of always preserving human dignity. The S-PCG therefore does not go against the principle of human dignity and equal or fair access to health care. However, it is worth mentioning that the S-PCG is not intended to be used for children with palliative care needs. This could be interpreted to mean that we discriminate against children based on their age. However, since the evidence that the S-PCG is based on, applies to adult patients and their families, it would not be ethical to use this evidence as a basis for care for children facing death. On the other hand, if the S-PCG is found to provide good support for high-quality palliative care this might inspire further development of the S-PCG for minor children, based on relevant evidence.

## The principle of autonomy and confidentiality

The principle of autonomy refers to the participants' right to have a saying, make their own choices and take actions based on their own personal beliefs and values. It requires that the researcher/health care personnel should demonstrate good research ethics<sup>173</sup> and respect the participant's/patient's autonomy by providing sufficient information, which is needed to make informed decisions. This includes informed consent, telling the truth and providing confidentiality<sup>177</sup>.

The participants involved in Part 1 of this project (*Studies I–III*) were provided with both oral and written information about the research and informed about their right to withdraw from the study at any time. They were provided with an ethical information sheet and the reply card was collected separately from the completed questionnaires<sup>2</sup>.

In Part 2 of this project, apart from the managers of each participating site giving their written informed consent, the participants in the focus group interviews and the semi-structured interviews also gave informed consent to participate, as promoted under the principle of autonomy. Information emphasizing that

participation was voluntary with the option to decline participation or opt out at any time without having to give an explanation was also provided to the participants, in writing and orally. They were given the opportunity to ask further questions about the project, orally or by writing down questions on a specific information letter they were provided with. Moreover, the participants were provided with the researchers' contact information in case they had any further comments or questions they wanted to discuss with the project leader.

The patients who were receiving care according to the S-PCG were not required to give written informed consent as this was part of the health care quality improvement measures. However, both written and oral information about the project and that the care unit was participating in the project was provided, with information that a copy of the completed S-PCG documents would be used for patient record audits after they had been de-identified. If any patient would have asked for their information to be excluded from the project, this would of course have been respected.

In both parts of this PhD project, all data were handled confidentially. All results are reported at group level only; therefore, no individuals can be identified or singled out.

# Results

## Summary of the main results

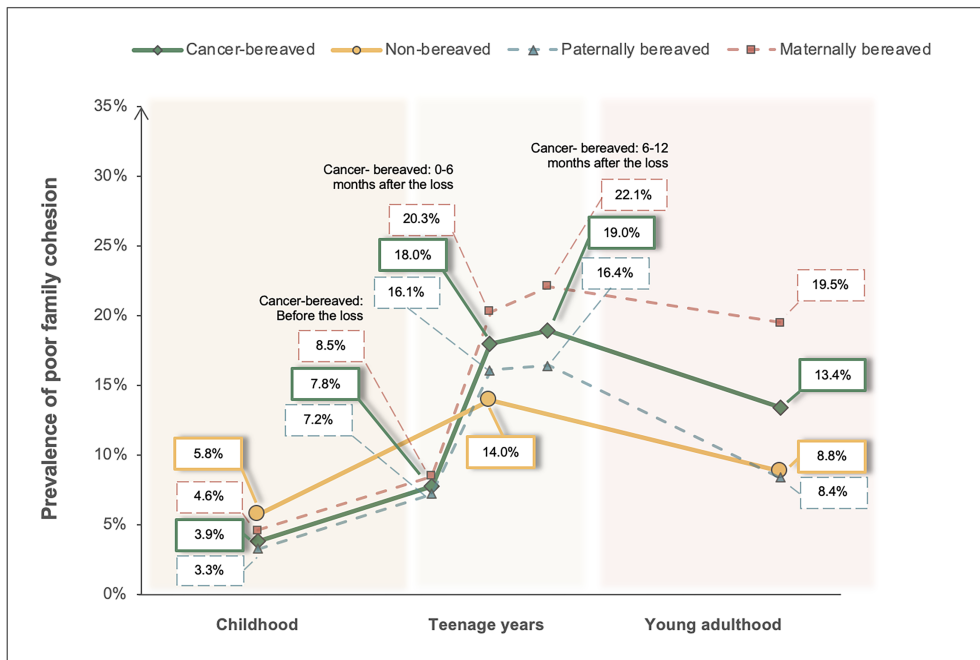
In Part 1 of this PhD project, 622 parentally bereaved young adults (73% response rate) and 330 non-bereaved participants (78% response rate) returned the study-specific questionnaire. Fifty per cent of the bereaved participants and 51% of the non-bereaved participants were male, and respectively 50% and 49%, respectively, were female. Among the bereaved participants, 337 (54%) had lost a father and 284 (46%) had lost a mother (one did not state the gender of the deceased parent) (*Studies I-III*).

Family cohesion was in focus in the first part of this PhD project (*Studies I-III*). Losing a parent to cancer as a teenager was found to be associated with increased risk of poor family cohesion during the teenage years, compared with non-bereaved youth. The perception of poor family cohesion was also found to continue into young adulthood, but only among the maternally bereaved participants (*Study I*). Poor family cohesion in the first year after losing a parent to cancer was found to be strongly associated with long-term negative psychological health-related outcomes among the bereaved youth, 6–9 years after the loss (*Study III*). Furthermore, having had good family cohesion during the first 6 months after the loss was found to be a predictor for having had an okay way to grieve in the acute bereavement phase, together with having had the opportunity to have a last conversation with the dying parent, and male gender (*Study II*). More than half of the bereaved teenagers in our study did not find a way to grieve that felt okay during the first 6 months after the loss and this in turn was found to be associated with long-term unresolved grief (*Study II*).

Part 2 of this PhD project, concerns the development and feasibility testing of the Swedish Palliative care guide (S-PCG), which, apart from identifying individuals' palliative care needs, also provides support for assessing the needs of the family. This includes determining whether there are children in the family and identifying their information and support needs (*Study IV*). After comprehensive feasibility testing conducted in 40 different health care settings, we found that the S-PCG has the potential to help provide a holistic overview of the patient's needs; it also supports team assessment and care of the patient with the aim to enhance each patient's quality of life and provide support to the family throughout the palliative trajectory (*Study IV*).

## Bereavement status and the level of self-reported family cohesion

Few participants (3.9% of the bereaved participants (paternally bereaved: 3.3%, maternally bereaved: 4.6%) and 5.8% of the non-bereaved participants) reported poor family cohesion during childhood. In total 23.5% of the bereaved participants compared with 14.0% of the non-bereaved participants reported poor family cohesion at some point during the teenage years. Looking closer at the prevalence at different time points during the teenage years, 7.8% of the bereaved participants (paternally bereaved: 7.2 %, maternally bereaved: 8.5%) reported poor family cohesion before the loss of a parent (*Study I*). A higher prevalence of poor family cohesion was reported during the first year after the loss of a parent, with a total of 21.7% of the bereaved participants (paternally bereaved: 18.8%, maternally bereaved: 25.3%) reporting poor family cohesion at some point during that period (*Study III*). In young adulthood, 13.4% of the bereaved participants (paternally bereaved: 8.4%, maternally bereaved: 19.5 %) and 8.4% of the non-bereaved participants reported poor family cohesion (*Study I*), as can be seen in Figure 4.

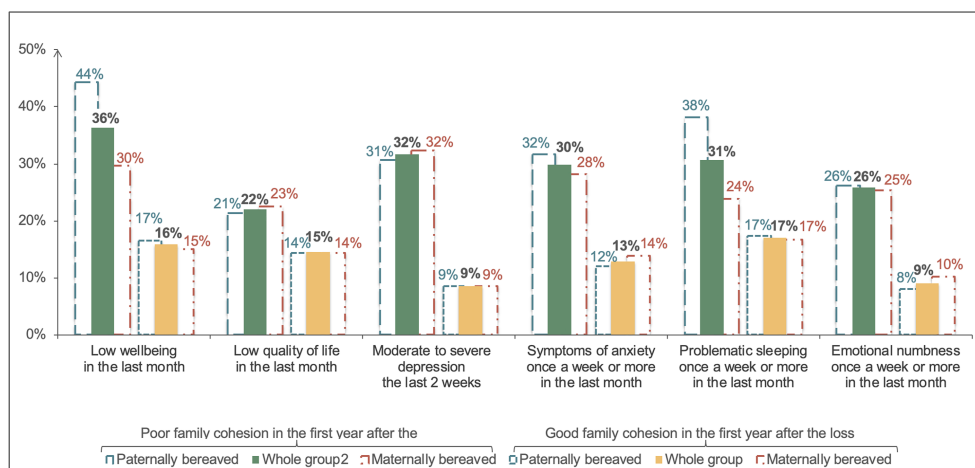


**Figure 4.** Prevalence of poor family cohesion in childhood, during the teenage years and in young adulthood among the bereaved and non-bereaved participants (based on Figure 1 in Study I).

When compared with non-bereaved youths, the bereaved participants were more likely to report poor family cohesion during the teenage years, with a crude odds ratio (OR) of 1.9 (95% CI 1.3–2.7). After controlling for several covariates, such as number of siblings and depression, the final adjusted OR ( $OR_{Adj.3}$ ) for poor family cohesion remained statistically significant for the bereaved (whole group) ( $OR_{Adj.3}$  2.0; 95% CI 1.3–3.0), paternally bereaved ( $OR_{Adj.3}$  1.7; 95% CI 1.1–2.7) and maternally bereaved ( $OR_{Adj.3}$  2.4; 95% CI 1.5–3.8) participants. This was also seen in young adulthood among the whole group of bereaved ( $OR_{Adj.3}$  1.6; 95% CI 1.0–2.6) and maternally bereaved participants ( $OR_{Adj.3}$  2.5; 95% CI 1.6–4.1) when stratified by the gender of the deceased parent, while there was no difference between the paternally bereaved and non-bereaved participants in young adulthood (*Study I*).

## Level of self-reported family cohesion in the first year after the loss, and long-term health and wellbeing

Among the bereaved participants reporting poor family cohesion in the first year after the loss of a parent, 31.6% reported moderate to severe depression in the last two weeks (i.e. in young adulthood) and 8.6% of those who had reported good family cohesion in the first year after the death of a parent (*Study III*). Similarly, participants who had reported poor family cohesion in the first year after the loss of a parent reported higher prevalence numbers in all of the long-term negative psychological health-related outcomes under investigation, compared with those who had reported good family cohesion in the first year after the loss (Figure 5).



**Figure 5.** Prevalence of health-related outcomes among young adults, 6–9 years after the loss of a parent to cancer, in relation to the level of self-reported family cohesion in the first year after the death of a parent during the teenage years

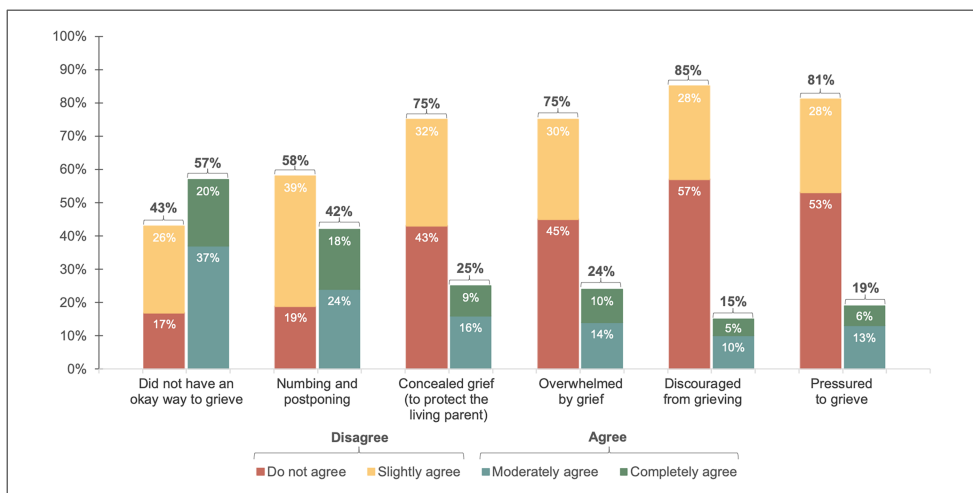
When looking at the associations between reported levels of family cohesion and long-term health and wellbeing, bereaved youth who reported poor family cohesion in the first year after the death of a parent had a higher risk of reporting symptoms of moderate to severe depression in the last two weeks, compared with those reporting good family cohesion, with a crude risk ratio (RR) of 3.67 (95% CI 2.50–5.40). After adjusting for a variety of possible confounding factors in the three-step modified Poisson regression model, we found that this difference remained statistically significant for moderate to severe depression, with an adjusted RR at the final step ( $RR_{Adj,3}$ ) of 2.63 (95%CI 1.67–4.15). Bereaved youth who reported poor family cohesion in the first year after the loss of a parent were also more likely to report lower levels of wellbeing ( $RR_{Adj,3}$  1.89; 95% CI 1.32–2.71), symptoms of anxiety ( $RR_{Adj,3}$  1.69; 95% CI 1.14–2.51), problematic sleeping ( $RR_{Adj,3}$  1.65; 95% CI 1.14–2.38) and emotional numbness ( $RR_{Adj,3}$  1.98; 95% CI 1.20–3.27) once a week or more at the time of the survey. Those reporting poor family cohesion in the first year after the loss were also more likely to have low quality of life at the time of the survey, with a crude RR of 1.50 (95% CI 1.02 – 2.22), but this difference did not remain statistically significant in the adjustments for possible confounding factors.

## Acute and long-term grief reactions and experiences

More than half of the bereaved participants, 57%, reported that they did not have a way to grieve that felt okay during the first 6 months after the loss (*Study II*). Figure 6 illustrates the prevalence of the different acute grief experiences and reactions.

A total of 45% of the parentally bereaved participants reported unresolved grief in the long term or 6–9 years after the death of a parent. Not having had a way to grieve that felt okay during the first 6 months after the death of a parent was found to be associated with long-term unresolved grief, with a crude OR of 4.32 (95% CI 2.99–6.28). After adjusting for background, family and health care-related variables the difference remained statistically significant ( $OR_{Adj,3}$  4.14; 95% CI 2.77–6.23). Long-term unresolved grief was also found to be associated with those that had been numbing and postponing (42%) ( $OR_{Adj,3}$  1.73; 95% CI: 1.22–2.47), overwhelmed by grief (24%) ( $OR_{Adj,3}$  2.02; 95% CI 1.35–3.04) and discouraged from grieving (15%) ( $OR_{Adj,3}$  2.68; 95% CI 1.62–4.56) or those who had concealed their grief to protect the living parent (24%) ( $OR_{Adj,3}$  1.83; 95% CI 1.23–2.73) (*Study II*).





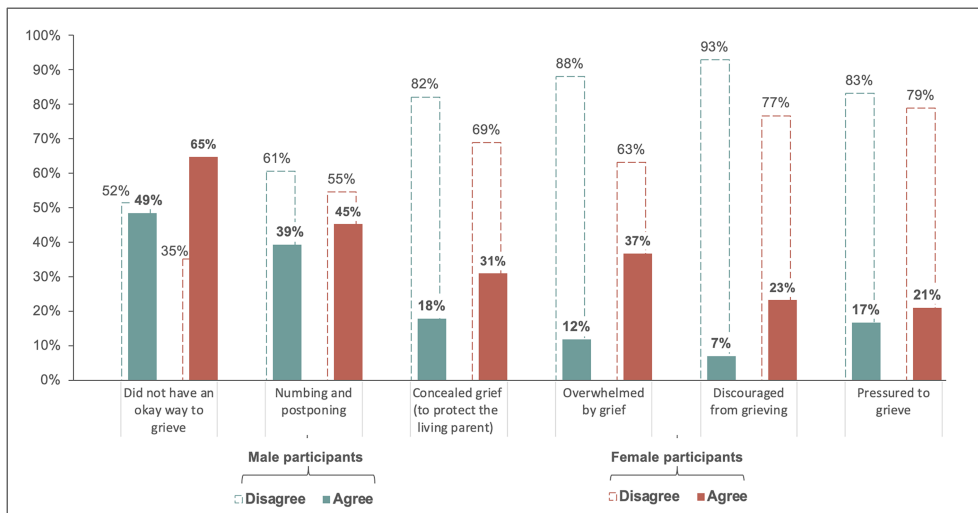
**Figure 6.** Prevalence of six different grief experiences and reactions in the first 6 months post-loss

When looking at possible predictors for *having had an okay way to grieve* in the first 6 months after the loss of a parent, associations were found with being male ( $p = 0.0020$ ), having had good family cohesion ( $p = 0.0046$ ), and having talked about what was important with the dying parent ( $p = 0.0015$ ) (*Study II*).

## Gender differences

The bereaved female participants were more likely to report poor family cohesion during their teenage years compared with the non-bereaved female participants (paternally bereaved female participants: OR: 2.7; 95% CI 1.3–3.89, maternally bereaved female participants: OR: 3.2; 95% CI 1.8–5.5) (*Study I*). This was also seen in young adulthood but only among the maternally bereaved female participants (OR 3.5; 95% CI 1.8–7.1). At the same time, no statistically significant difference was found between the bereaved and the non-bereaved male participants at any of the three periods under investigation (childhood, the teenage years and young adulthood) (*Study I*). A total of 13% of the bereaved male participants reported poor family cohesion at some point during the first year after the loss of a parent while 30.5% of the female participants did the same (*Study III*).

When looking at the acute grief reactions and experiences, 65% of the bereaved female participants, and 49% of the bereaved male participants, reported not having had an okay way to grieve. The prevalence of the different acute grief reactions and experiences among the bereaved male and female participants is shown in Figure 7.



**Figure 7.** Prevalence of acute grief experiences and reactions (in the first 6 months after the loss of a parent) among the bereaved male and female participants.

A total of 52% of the female participants and 37% of the male participants, reported unresolved grief at the time of the survey. An association between not having had an okay way to grieve and long-term unresolved grief was found both in male ( $OR_{Adj.3} 6.72$ ; 95% CI 3.65–12.84) and female participants ( $OR_{Adj.3} 2.73$ ; 95% CI 1.54–4.89). When looking at the association between long-term unresolved grief and the other acute grief experiences, some of the variables were found to be statistically significant among male participants (e.g. concealed grief:  $OR_{Adj.3} 3.41$ ; 95% CI 1.55–7.77) and other variables among the female participants (e.g. discouraged from grieving:  $OR_{Adj.3} 2.74$ ; 95% CI 1.41–4.32) (*Study II*).

# The structure and main content of the Swedish Palliative Care Guide

The S-PCG includes three parts that can each be used to identify and assess individual care needs during the whole palliative care trajectory from early identification to end-of-life care and care after the death of the patient, including bereavement care for family members. It provides structure for addressing the different individual care needs and for following up on them with care interventions. The S-PCG also provides structure for identifying the information and support needs of the children in the family, both before the death of their family member and in bereavement (*Study IV*). Figure 8 provides an overview of the different parts of the S-PCG and Figure 9 gives an overview of the main content of the S-PCG presented in the form of a circular table of contents.

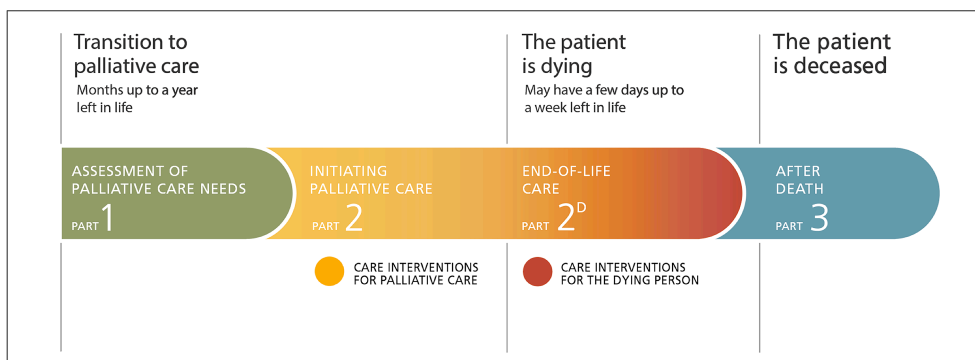


Figure 8. Overview of the Swedish Palliative Care Guide(S-PCG) and its different parts<sup>163, p.7</sup>.

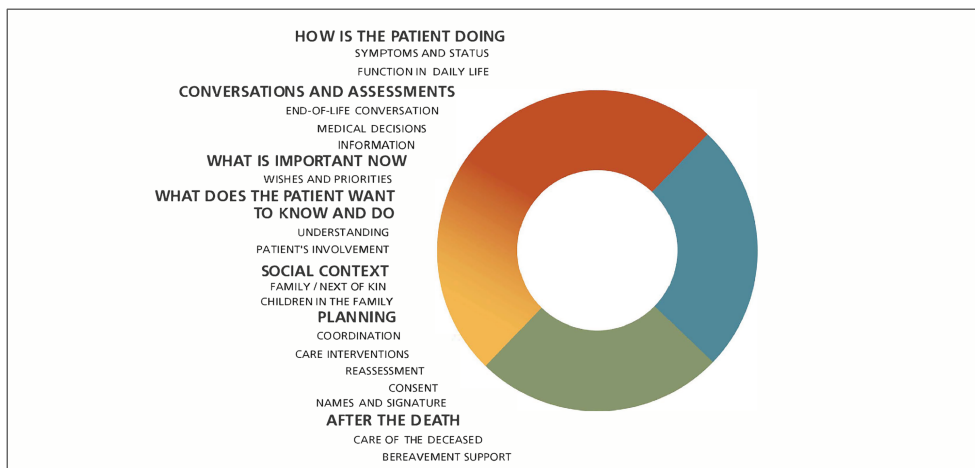


Figure 9. Overview of the main content of the Swedish Palliative Care Guide (S-PCG).

# Children in the family, and the Swedish Palliative Care Guide

There are sections in the S-PCG that focus on support to the children in the family. In part 1 of the S-PCG, which is a shorter version of the S-PCG, the questions about children in the family are included in the section on Family/persons of importance (Figure 10). Under this section the health care personnel are guided to ask about the patient’s social situation, whether there are worries in the family and if there is a need for support to a family member. The health care personnel are also guided to ask whether there is a child or children in the family and what the child knows/the children know about the patient’s diagnosis and prognosis.

**■ 1.4 FAMILY / PERSONS OF IMPORTANCE**

Does the patient live alone?  yes  no

Is there any severe distress in the family?  yes  no

Is anyone in the family in need of support?  yes  no

Are there any children (0–18 years old) in the family/of importance to the patient?  yes  no

Conversation conducted with guardian, regarding what the child/children know/s about the diagnosis/prognosis  yes  no

**Figure 10.** Family/Persons of importance in part 1 of the Swedish Palliative Care Guide (S-PCG).

Part 2 of the S-PCG includes a section specifically for children in the family, with more detailed guidance provided (Figure 11). For a child who needs, or children who need, information or supportive conversation, there is also an appendix dedicated to children in the family.

**■ 2.9 CHILDREN IN THE FAMILY (0–18 YEARS OLD)**

Are there any children in the family/of importance to the patient?  yes  no

Number of children \_\_\_\_\_

Year(s) of birth \_\_\_\_\_

Does the patient have sole custody? If yes, contact social worker  yes  no

Conversation conducted with guardian regarding what the child/children know/s about the diagnosis/prognosis  yes  no

Information brochure given to guardian  yes  no

Conversation with the child/children is needed? If yes, open the appendix on children of importance  yes  no  not applicable

Has a social worker been informed that there is a child/children involved?  yes  no

**Figure 11.** Children in the family in part 2 of the Swedish Palliative Care Guide (S-PCG).

Part 3 of the S-PCG also contains a section about children in the family, similar to the one in part 2, only with some adjustments, to give more focus on bereavement and grief.

In the test versions 1 and 2, the support for children in the family was imbedded within the actual S-PCG documents. Many comments were received, especially from elderly care units where it was felt that this content took up a lot of space when very few children in the family were among their clients.

“Most of our patients [in the nursing home] do not have minor children; that section takes up a lot of space” (Clinical test user)

This led to the solution that the basic questions about children in the family, required according to health care law in Sweden, were kept in the main documents. This included questions to determine whether there are children in the family, what they know about the diagnosis and prognosis and whether they are in need of a conversation with health care personnel or need further support. Further guidance regarding children as family was then appended to S-PCG part 2, for use before the patient’s death, and part 3, for bereavement support (Figure 12).

**■ BEREAVEMENT SUPPORT FOR CHILDREN OF THE FAMILY/OF IMPORTANCE TO THE DECEASED PERSON**

Bereavement support carried out date \_\_\_\_\_ date \_\_\_\_\_  not applicable

with child/children  with child/children and guardian

with child/children and other relative \_\_\_\_\_

Health care professionals present

social worker  doctor  nurse  assistant nurse  other

Content of the conversation \_\_\_\_\_  
 \_\_\_\_\_  
 \_\_\_\_\_

Or see electronic record entry Date \_\_\_\_\_ sign

**■ CHILD/CHILDREN IN NEED OF SUPPORT**

Support needed\*: \_\_\_\_\_  
 \_\_\_\_\_  
 \_\_\_\_\_

\* can be e.g. social, practical or counselling

Or see electronic record entry Date \_\_\_\_\_

**■ NEED TO ARRANGE CONTACT WITH:**

student health  social worker  psychologist  school/preschool

social services  grief support group  primary caregiver  other \_\_\_\_\_

not applicable

Comments \_\_\_\_\_

**Figure 12.** Children and bereavement support in part 3 (appendix) of the Swedish Palliative Care Guide (S-PCG).

# Feasibility testing and main adjustments to the Swedish Palliative Care Guide

## Relevance of the content

The content of the S-PCG was considered important and feedback was that it is relevant and provides a good overall picture of the patient's needs. It was considered appropriate for a broad number of patients although some participants giving feedback wanted to know whether the same documents were suitable for use in all care forms (Textbox 1).

**Textbox 1.** Example of the feedback regarding the relevance of the content of the Swedish Palliative Care Guide

- "The S-PCG is a good document that addresses important issues." (Patient, family and public representative)
- "You get an "overall look of things" with the S-PCG." (Clinical test user)
- "Good. It is important to ask the family about their need for support." (Patient, family and public representative)
- "Would it maybe be better to have different documents...for example, one for home care and one for hospital care?." (Interdisciplinary advisory committee)

## Usefulness – user friendliness

Overall, the S-PCG was perceived to give a good support for the care of the patient, and for gathering all the information needed. According to the feedback, the S-PCG is clear and usable. The patient, family and public representatives commented that the S-PCG came across as professional. However, it was also seen as very extensive and time-consuming to complete by the personnel. Some who gave feedback said that it was difficult for the user to know when to use part 2 and when to use part 3 of the S-PCG (early and late stages of palliative care). In addition, it was felt that filling in parts 2 and 3 meant a lot of duplication (Textbox 2).

**Textbox 2.** Example of the feedback regarding the usefulness/user-friendliness of the Swedish Palliative Care Guide

- "Yes, the documents are clear and seem to be usable." (Interdisciplinary advisory committee)
- "It's good that the S-PCG first finds the problem and then guides us to take action to solve/alleviate it" (Clinical test user)
- "Some words are maybe technical, like more medical terms, but this [the S-PCG] is also aimed for the professionals and that feels safe." (Patient, family and public representative)
- "A standing reflection is that the S-PCG documents are too comprehensive and take a very long time to complete." (Interdisciplinary advisory committee)
- "Comprehensive material but good that you can choose what is relevant for each patient." (Clinical test user)
- "Take away 'early and late stages' [of palliative care] from the title, it doesn't help and is just confusing" (Interdisciplinary advisory committee)
- "When a patient is admitted at S-PCG part 2 stage and then shortly after he is dying and needs to move on to part 3, we need to fill in all the same information again, that's a lot of work." (Clinical test user)

## Missing issues

Many who tested the S-PCG felt that nothing was missing; others asked for more space for text and for clearer instructions on how to use the S-PCG and a digital version of the S-PCG. The patients highlighted that health care personnel need to ask about pets or whether there is anything else that is worrying for the patient (Textbox 3).

**Textbox 3.** Example of the feedback regarding missing issues in the Swedish Palliative Care Guide (S-PCG).

- "The S-PCG has a good content; all the important elements are included." (Clinical test user)
- "...better instructions before the use, I am not sure how to use the different parts of the S-PCG." (Clinical test user)
- "It should be made clearer that you can skip items that are not relevant to the patient you're caring for" (Clinical test user)
- "...more space for writing what ADL-aids\* the patient has" (Interdisciplinary advisory committee)
- "They need to ask if there is something else I am worried about, such as who is taking care of my cat." (Patient, family and public representative)

\*ADL = activities of daily living]

## Redundancy

As mentioned, we got feedback that the S-PCG is very extensive; and at the same time that there was nothing that could be removed, that all contents were important as they could be relevant to some patient at some time. An exception was the medication list, which all care units document elsewhere in the patient record system (Textbox 4).

**Textbox 4.** Example of the feedback regarding redundancy in the Swedish Palliative Care Guide (S-PCG).

- "Nothing. The S-PCG is comprehensive and extensive but everything in it is important." (Clinical test user)
- "Everything in the material is essential and nothing needs to be added or removed." (Patient, family and public representative)
- "Nothing. Everything could be relevant to some patient at some point." (Interdisciplinary advisory committee)
- "It would be double documentation to have status updates and a medication list in the S-PCG because they are already in our digital patient records." (Clinical test user)

## Teamwork – communication

The S-PCG prompted discussions about the current practical routines; it highlighted the teamwork and helped the team to gain an overview of a patient's problems and needs and to discuss them in a timely manner. Some members of the teams were sceptical and it was difficult to get all on board. The patients and family representatives highlighted the importance of being given clear information about whom to contact and where to get support (Textbox 5).



**Textbox 5.** Example of the feedback regarding teamwork and communication using the Swedish Palliative Care Guide (S-PCG).

- "It's good that the S-PCG makes the end-of-life conversations visible to everyone in the team." (Clinical test user)
- "The S-PCG requires teamwork, which is good." (Interdisciplinary advisory committee)
- "The S-PCG clarifies what the assistant nurses do in the team – GOOD!" (Clinical test user)
- "As a doctor I can say that the S-PCG has to do more with the nurse's work, but it isn't overwhelming and there is much gained once part 2 has been done." (clinical test user)
- "Difficult to get employees on board in the beginning but it got better later." (Clinical test user).
- "Difficult to get doctors on board." (Clinical test user)
- "The health care provider must help the patient to find out know where to go, who to talk to, or how to get a referral to others, if the patient's needs cannot be met." (Patient, family and public representative)
- "The personnel need to make sure that the relatives are guided towards the right person/appropriate support when needed." (Patient, family and public representative)

## Patient and family involvement

Patient involvement was not felt to be a barrier for most of the clinical test users, although a few felt reluctant to involve the patients and their family. The users felt that the S-PCG clarifies the needs of the patient and their family and brings forward the work that is being done for them. The families felt reassured, that the support was improved and said that the S-PCG creates feelings of security (Textbox 6).

**Textbox 6.** Example of the feedback regarding patient and family involvement in relation to the usage of the Swedish Palliative Care Guide (S-PCG).

- "Information about the S-PCG to patients and relatives is not perceived as a barrier in our team." (Clinical test user)
- "It can be difficult with the needs of the family; they can have many needs and we want to focus our time on the patient." (Clinical test user)
- "It is good to go through the S-PCG; [it helps] you [to get to] know what the patient and the family want." (Clinical test user)
- "The S-PCG clarifies the palliative care needs." (Clinical test user)
- "The family see what we work with in the S-PCG and feel that what we do is important." (Clinical test user)
- "The S-PCG creates security for the family; you check if the document is filled in." (Patient, family and public representative)
- "The S-PCG provides reassurance to the family members, once they have been informed about the care plan." (Clinical test user)

## Implementation

Information and training in using the S-PCG is needed and this highlights the importance of basic education in palliative care. The feedback also highlighted that, in order to succeed with the implementation of the S-PCG, managers need to be on board and there needs to be adequate time to plan the use of the S-PCG. It was mentioned by the family representatives that the instructions need to include that it is important to offer a conversation with the family without the patient present. Another important comment, made by a patient representative, was to provide guidance and support for the health care personnel in performing end-of-life conversations and to include patients' stories in the education and training (Textbox 7).

**Textbox 7.** Example of the feedback regarding implementation of the Swedish Palliative Care Guide (S-PCG).

- "The S-PCG is good and useful but you need training in order to understand how to use the documents." (Clinical test user)
- "With a lot of new staff, there is a need for education in basic palliative care." (Clinical test user)
- "[There must be] more involvement of the manager." (Clinical test users)
- "The nurses wanted to test the S-PCG on all of the patients but the management said no." (Clinical test user)
- "To go through and decide on how the documentation procedures and routines should be, before using the S-PCG is important." (Clinical test users)
- "There should have been more time for planning." (Clinical test users)
- "Put in the user guide that it is important to sometimes have a conversation with the family members without the patient around. When you ask questions about how the family member is holding up or if he or she needs support it is sometimes difficult to answer honestly if the patient is close by." (Family representative)
- "Before implementing the S-PCG on a new ward, the clinic should ensure that they involve someone within the practice who is experienced in dealing with the difficult conversations, such as telling someone that they are dying. So that this person can guide and be supportive to the others." (Patient representative)
- "I suggest that you record stories from patients, asking the question 'What is important to you?' and include them in the training/introduction of the S-PCG to healthcare professionals. So, they don't forget why they're doing this work." (Patient representative)

## Main adjustments made to the Swedish Palliative Care Guide

Several changes were made to the S-PCG documents after the final testing of the S-PCG, including the merging of parts 2 and 3. Education programme and support documents were finalized and inspirational and instructional videos were made. Textbox 8 gives an overview of the most significant updates that were made.

**Textbox 8.** Examples of some of the most significant updates made before launching the Swedish Palliative Care Guide (S-PCG).

### **Content**

- The content itself did not change much in the final adjustments except for small details and adjustments to the wording.
- Only the basic questions for children in the family were retained in the main documents. More specific guidance for support was moved to an appendix, to be used when needed.

### **Usefulness – user-friendliness**

- Parts 2 and 3, of the S-PCG, were merged to form parts 2 and 2D (D for the “dying phase”), to address the problem of repetition and double documentation, and the terms “early and late palliative care” were omitted from the titles of the parts.
- A circular table of contents was added at the beginning of each document, together with a short explanation of the aim and when to use each part.
- Checkboxes for “not applicable at this moment” were added in several places in the S-PCG documents.

### **Missing issues and redundancy**

- The support questions, about the patient’s wishes and priorities, were moved from the appendix into the main document and a question about pets was added to these “support questions”.
- The medication list was removed from the S-PCG and a note to refer to the digital medication list was added to the documents.

### **Teamwork – communication**

- A brochure with information for patients and their family was created in collaboration with patient and family representatives, including a space for written information about whom to contact.
- A video about the S-PCG was created, which includes topics such as the importance and benefits of teamwork and tips on how to include the team in the implementation of the S-PCG.

### **Patient- and family involvement**

- The importance of patient involvement was raised both in an online video about S-PCG users’ experience and in the S-PCG education programme.
- Brochures were created to facilitate the implementation of the S-PCG and complement the online information videos.
- A whole-day educational programme about the S-PCG was created, including, among other things, a video with stories from a patient who was filmed.

### **Implementation**

- Support material was created, such as online videos and brochures with short, practical users-instructions as well as recommendations highlighting the importance of good planning before implementation. A video and a brochure especially targeting managers were also created.
- As mentioned above the whole-day educational programme about the S-PCG also included a video of stories from a patient.

# Discussion

## General discussion of the results

In accordance with the overall aim of this PhD project, this thesis describes family cohesion, as perceived by young adults who lost a parent to cancer in their teenage years, and its association with long-term health and wellbeing. It also describes their acute and long-term grief reactions and experiences. Furthermore, the thesis describes the development of the Swedish Palliative Care Guide (S-PCG), including support for health care personnel in attending to the needs of children as family members of the dying patient. Here, in the General discussion of the results, some of the main “take-home-messages” based on the results of the study will be discussed briefly, in relation to possible clinical implications.

### **Increased risk of poor family cohesion after losing a parent to cancer as a teenager**

Losing a parent to cancer as a teenager was found to be associated with increased levels of poor family cohesion during the teenage years, compared with non-bereaved participants, and this lasted into young adulthood among the maternally bereaved participants (*Study I*). There is a lack of research on how family cohesion changes over time from the perspective of bereaved youth. However, in a study on non-bereaved adolescents, a small decrease in cohesion within the family was seen at the beginning of adolescence, which declined to pre-adolescent’s levels at the beginning of adulthood<sup>182</sup>. A similar decline has also been seen regarding perceived parental warmth during adolescence<sup>183,184</sup>. This is considered to be due to the nature of adolescence, which drives teenagers to seek independence from their family<sup>183</sup>.

An association between impaired family cohesion and negative outcomes for bereaved adolescents, such as self-injurious behaviours<sup>21</sup> and anxiety and depression<sup>185</sup>, has been reported in some studies. In a longitudinal cohort study on non-bereaved adolescents, Rattay et al<sup>186</sup> found family cohesion to be the strongest mediating factor for emotional and behavioural problems and health-related quality of life in both nuclear families and stepfamilies, and single-parent households. Lack of family cohesion was even shown to be a stronger predictor for these problems than divorce between parents or a low socio-economic status of the family<sup>186</sup>. The

results of this thesis should therefore raise awareness that bereaved families are at risk of having poor cohesion, and highlight the need to strengthen the family cohesion in bereaved families.

According to the conclusion of a systematic review on self-reported assessments of family function<sup>62</sup>, no validated screening instruments for identifying family cohesion are available that are concise enough for application in day-to-day clinical practice. However, one of the crucial components that enables good family cohesion is communication between the family members<sup>61</sup>. In this regard, health care professionals have a major role in guiding and supporting parents to communicate with their children when a parent is facing life-threatening illness<sup>99, 187</sup>. The health care personnel could assist by, in the first hand, routinely asking whether there are any children in the family, followed by conversations with the parents, and the teenagers themselves, about what they have discussed between them, and what the children in the family know about their parent's diagnosis and prognosis, and whether the children are aware of the life-threatening condition of their parent<sup>99</sup>. Guidance for this is included in the S-PCG (*Study IV*), with the intention to encourage the health care personnel to put children as family on the agenda when using the S-PCG in clinical practice. These discussions, which should be individually adapted and conducted in a sensitive manner<sup>99, 118</sup>, may enable the health care personnel to identify those families that have not had these conversations with the children in the family.

In a study performed in a Swedish oncology setting, parents expressed that they found it difficult to talk to their children and wished for more support and guidance from the health care personnel on how to talk to their children<sup>118</sup>. As recommended in a recent practical guidance from *The Lancet*, which was based on a literature review and expert workshop, it can sometimes be helpful just to provide the parents with reassurance that it is okay to find these conversations difficult and that it is not harmful to show difficult emotions in front of their children; rather, this allows the children to also express their own feelings<sup>99</sup>. This can encourage parents to take the step and start a conversation with their children, which in turn can help the children to cope with the loss, since open communication within families has been shown to be beneficial to children's adaptation after the death of a parent<sup>188</sup>. In addition to this, it is recommended that bereaved family members should have the possibility to have a bereavement discussion with some of the health care personnel who was involved in their deceased family member's care<sup>127, 162</sup>. In line with this, a guide for bereavement support for children in the family was included in the S-PCG (*Study IV*). However, direct question about family cohesion is not included and in future revisions of the S-PCG it may be worth considering adding a question asking about their own perception of the cohesion in the family.

## **Not having had an “okay way to grieve” during the first 6 months after the loss of a parent, and long-term unresolved grief**

It is notable that the majority of the participants (57%) stated that they had not had an okay way to grieve during the first 6 months after the loss of their parent, and this was also found to be associated with unresolved grief 6–9 years after the death of the parent. As mentioned before, good family cohesion seems to play a major role in this regard as it was found to be one of the predictive factors for having had an okay way to grieve during the first 6 months after the death of a parent (*Study II*).

The main take from these results regarding grief reactions and experiences is that instead of prescribing how the teenagers should or should not grieve it is more important to encourage them to find a way to grieve that feels okay to them. This is in line with a statement made by Hanie et al<sup>8</sup> who, after empirical evaluation, highlighted that it is important not to pressure bereaved children to express their feelings if they do not find a need to do so<sup>8</sup>. This is strengthened up by a recent recommendation by Kentor and Kaplow<sup>189</sup>, that health care professionals should help children and their families to acknowledge that there is no one “right” way to grieve<sup>189</sup>. As previously discussed, open communication has been shown to benefit bereaved children<sup>188</sup> and one way to facilitate open communication after the death of a parent is to give the family and the children in the family the opportunity to address the forthcoming death before it occurs<sup>8</sup>. In relation to this, the results of this PhD study showed that having had the opportunity to talk about what is important with the dying parent was one of the significant predictors for being able to find an okay way to grieve after the loss (*Study II*).

The health care personnel can assist the bereaved-to-be families to open up conversations with the children in the family. However, health care personnel have expressed challenges in doing so, such as lack of time and high workload<sup>99, 119</sup>, lack of training, and fear of not having the competence to talk to the children or being able to provide the support that is needed<sup>119</sup>. For some of them, the challenge comes from the belief that this is not their responsibility<sup>190</sup>.

Based on these findings and supported by the results from testing the S-PCG, it is important for the health care personnel to find security in their practice. This means there must be clear routines in place. The health care personnel must know where they can turn to if the children or the family have needs that are not within their or their health care unit’s capacity to attend to. Likewise, the use of consultation, peer support and specific guidelines has also been suggested for health care personnel who meet families with children facing bereavement<sup>99</sup>. The S-PCG provides support to the health care personnel when discussion with the children in the patient’s family is needed, including structure for establishing further contact if needed, such as contact with the student health care or bereavement support groups. It is also encouraged in the S-PCG manual to discuss and clarify clinical routines with the

team before implementing the S-PCG, so that the health care personnel know what support the team can provide for the children in the family and where to turn to if other support is needed (*Study IV*).

Another aspect that has been highlighted in several literature reviews is that of the importance of the knowledge about the variety of grief reactions and coping mechanisms among children and teenagers who are grieving a parent<sup>55, 99, 189</sup>. Knowledge about teenagers' different ways of grieving may thus help bereaved youth to find a way that feels okay to them. Furthermore, research has shown that bereaved teenagers want their parents to be provided with knowledge about teenagers' different grief reactions and coping styles<sup>113</sup>. The bereavement literature has largely focused on the pathological aspects of grief, more studies are needed to build up the empirical evidence on the normative way of grieving among children and teenagers<sup>64, 113</sup>. Publishing the results of this study is one way to address this need, by contributing to filling in the knowledge gap. The development of the S-PCG, is another way to address this, as its "Children as family" sections include materials that encourage the health care professional to provide the children and their parent with information about children's grief reactions (*Study IV*). Exactly what information the health care personnel should provide has, however, not been specified in the S-PCG, nor what information is available for the health care personnel to give to the families. This might be a suggestion for further development of support materials for the S-PCG as well as the education programme.

### **Family cohesion in the first year after the loss, and long-term health and wellbeing**

Poor family cohesion, as perceived by the bereaved participants, was found to be strongly associated with negative psychological health and wellbeing, long-term (*Study III*). This is in line with other studies that have highlighted the family environment as one of the biggest mediating factors for the health and wellbeing of children of cancer patients<sup>89</sup> and bereaved children and adolescents<sup>44, 52, 191</sup>. In a study on dual-parent families with adolescent offspring, conducted in the general public, higher levels of family cohesion were found to be associated with adolescents' overall wellbeing, as well as contributing to their emotional wellbeing<sup>192</sup>. In agreement with evidence showing the importance family cohesion has for the wellbeing of teenagers, the results of this PhD study highlight the importance of supporting families that have poor cohesion or that are at risk of having poor family cohesion. Support may thus be considered an important preventive measure for their long-term health and wellbeing, although more studies are needed to define what kind of support should be provided that can strengthen the family cohesion of bereaved families.

Abel et al<sup>193</sup> have called for more emphasis on health-promoting palliative care services, including co-operation between the general health care and specialized palliative care services, as well as involvement of people from the local community and civic society<sup>193</sup>. Other studies have highlighted the need to put more focus on the public health perspective in palliative care<sup>194, 195</sup>. This includes decreasing stigma around death and dying, engaging communities in supporting bereaved individuals, including bereavement in national policies<sup>194, 195</sup> and ensuring that both professionals and families will feel enabled to discuss the issues around death and dying with children<sup>195</sup>. Also, it includes engaging in interventions that can normalize bereavement and grief in children<sup>195</sup>.

Although, there is much focus on shifting the responsibility to include society as a whole, the health care service still has an important role to play<sup>193</sup>. However, Kellehear and his fellow-researchers argue that specialized palliative care should not be provided in the same way, and universally, to all, but should be provided according to need<sup>193</sup>. Similarly, Aoun et al<sup>196</sup> highlight the importance of providing bereavement support according to need. The results of this PhD study may support these suggestions, although family cohesion was found to be a predictor for long-term health and wellbeing, the majority of the participants, in our study, reported good family cohesion. Yet, although it has been stated that bereavement support to adults should only be provided to those in need, it has been suggested that being a child and losing a parent is enough of a risk factor to benefit from bereavement support<sup>197</sup>. In a systematic review on the effects of support programmes for parentally bereaved children and their caregivers, Bergman et al<sup>198</sup> emphasized that although some children and their families are at higher risk and need more support than others, the evidence base indicates that a brief, low-cost, interventions should be available to all parentally bereaved children as this may help prevent more severe psychological health problems and problematic grieving after the loss<sup>198</sup>. However, according to several systematic and literature reviews on support programmes and interventions for bereaved children and adolescents, there is a lack of well-designed interventions studies and evidence for what kind of support or bereavement interventions should be provided to whom<sup>189, 191, 198-200</sup>. Although empirically supported interventions have been shown to be helpful for children and adolescents<sup>189</sup>, there is a call for more studies and interventions tailored especially to meet the different needs of children<sup>198</sup> and adolescents<sup>101, 199, 200</sup>, based on their developmental stage and other social factors<sup>189, 191</sup>, as well as studies on how to screen for high-risk families in need of advanced support<sup>191</sup>. To enable support from the health care to bereaved-to-be children and families, it is necessary first and foremost that assessment of the needs of family members is incorporated into clinical practice, where terminally ill patients are cared for. As previously stated, the development and implementation of the S-PCG in clinical practice is an action to address this (*Study IV*).



## Gender differences

Throughout Part 1 of this PhD project, gender was a factor that was found to influence the results of our studies. We found that female participants reported higher levels of poor family cohesion than male participants, at all time periods (*Study I*). This is in line with the results of a prospective study on young university students, showing that family relationships are of greater importance to female individuals, who seem to be more affected by changes in the family than their male peers<sup>201</sup>. Female participants were also found to report higher prevalence of all of the different acute grief experiences and reactions than male participants, and also reported higher levels of unresolved grief (*Study II*). Other studies have shown bereaved girls to be more prone to have internalizing problems<sup>8</sup> and more vulnerable to bereavement stressors than boys<sup>8, 202</sup>.

Differences in levels of reported family cohesion based on the gender of the dying parent were also found. Maternally bereaved participants reported higher levels of poor family cohesion than the paternally bereaved ones, and female participants reported higher levels of poor family cohesion, both among the paternally bereaved and among the maternally bereaved participants, compared with male participants (*Study I*). Women have been found to be more likely to have better coping strategies<sup>203</sup> and mothers are more likely than fathers to better adapt to life after the death of their partner<sup>204</sup>. Mothers have also been shown to be more capable of attending to their children's needs<sup>9</sup> and more likely to engage in positive parenting<sup>205</sup> after the loss. In a study from 2005, Ericson et al<sup>206</sup> found that providing emotional support to the family members and supporting their emotional well-being is mostly performed by the mothers of the family. However, it should be highlighted that the parental roles are culturally dependent, and that they change over time as the norms in society change. As Werner-Lin and Biank point out, differences in adaptation after the loss may have more to do with the role of the parent within the family, than with the actual gender of the parent<sup>207</sup>.

The results of this PhD project suggest the need for awareness about gender and gender roles when families are facing bereavement. However, more studies are needed to further investigate bereavement and family relationships in relation to gender roles.

## **Implications for the Swedish Palliative Care Guide to support clinical practice**

Part 2 of this PhD project describes the development of structured support for the health care personnel, to guide their clinical practice when caring for people with palliative care needs, highlighting the needs of the patient, family and children in the family throughout the palliative care trajectory.

The results show that the S-PCG has the potential to provide support for palliative care in different care settings. (*Study IV*). However, to drive a change in the complexity of a health care setting is challenging<sup>128, 208</sup> and it would be naïve to think that the S-PCG presents a simple solution to all the challenges faced by health care professionals in their everyday life. Based on comprehensive feasibility testing the results showed that there are several components that need to be considered before implementation of the care guide. Firstly, the S-PCG should not be seen as a simple screening tool, but as a way of working with a palliative care approach, and its implementation should be initiated with careful and thorough planning.

It is also worth mentioning that it was frequently highlighted that the managers of the care units need to be on board for implementation to be successful (*Study IV*). The health care personnel and their managers should look into current standards within their clinical practice; to find out: What is working well? Which are potential areas for improvement? What are the current working routines regarding the content of the S-PCG, and how will the S-PCG be incorporated into the clinical practice in a way that is most beneficial for the patients and their family, as well as for the personnel? Avoiding duplication of the same information, in the patient records, is another factor that is crucial to think about before implementing the S-PCG.

The interdisciplinary team should also discuss what aspects of their current competence needs further enhancement. This could mean discussing questions such as: Is there a need for education in; basic palliative care, communication skills, symptom assessment, and identifying and attending to the needs of children in the family? The team will also need to plan who will lead the implementation and set aside time to establish that.

Finally, it is important to highlight that the S-PCG does not aim to provide standardized care. Rather than striving for that every patient should get the same care, the aim with the S-PCG is to give every patient the opportunity to receive care according to their needs. Furthermore, the care structure provided by the S-PCG is to be used to ensure that important questions will be asked in a timely manner as relevant to each individual. In this way it will help to bridge the gap between what is recommended as a best care practice and what is being provided in the clinical practice, also with regard to the needs of the children in the family.

# Methodological considerations

## Validity

The validity of a study can be threatened by errors, which can be either random (by chance) or systematic (biases). To ensure the validity of the study and to minimize the chance of systematic errors, the hierarchical step model for causation of bias was used to guide the design of Part 1 of this PhD project. The hierarchical step model includes four steps representing the main sources of systematic errors that can threaten the validity of a study, all of which should be considered when designing and performing a study<sup>142</sup>.

### *Confounding (Step I)*

One of the main sources of systematic errors derives from confounding factors that can distort the estimation of the association between the variables under investigation. This becomes a problem when the confounding factors are unevenly distributed between the two groups that are being compared, which threatens the internal validity of the study. Randomization is a one way to minimize the risk of confounding. For ethical and practical reasons, a randomized controlled trial was not an option for this study. Despite this, the possibility of bias due to confounding should be considered and acted on. Some measures were therefore taken, such as that the control group of non-bereaved participants were matched by age, gender and type of residence. Also, to account for the error of unmeasured confounding, a comprehensive questionnaire of 40 pages, including a total of 271 items, was carefully designed, to measure, and thus enable control for, a broad variety of possible confounding factors.

### *Misrepresentation (Step II)*

In the second step of the hierarchical step model the concept of misrepresentation should be taken into consideration. This could include biases deriving from factors such as non-participation, loss of subjects to follow-up or errors during the sampling process, which might result in incomplete information gathered<sup>142</sup>. The study that Part 1 of this PhD study is based on was carefully designed to minimize the risk of misrepresentation. Firstly, the study was performed at a nationwide level, where all participants who had lost one parent to cancer during the reference period were invited to participate. Secondly, an effort was made to minimize the loss to follow-up by designing the questionnaire based on topics that were brought up in interviews with people from the target group, topics that were highlighted as important to them. Also, time and resources were allocated to a research assistant, who was experienced in respectfully talking to bereaved youth. The research assistant contacted all potential participants personally by telephone, after an information letter had been sent, to give them the opportunity to ask questions about the study before the

questionnaire and reply card were sent out. The participants were further provided with a telephone number so that they could ask any questions that might emerge during the data collection period. A reminder call was also made to those who had not returned the questionnaire. These measures are believed to have contributed to a response rate of over 70%, which is one of the biggest strengths of the study. However, since we do not have any information about the characteristics of those who declined participation we cannot be sure that there has been no attrition bias.

### *Misclassification (Step III)*

The risk of introducing bias due to misclassification or measuring errors is always present in every study. The questionnaire was carefully designed to minimize this risk. Where well-established, suitable measurements, that had been validated for the target group were available for some of the phenomena under investigation, they were included in the questionnaire. However, since the intention was to study the subjective experiences of the participants the study mostly followed the well-established method of “One direct question for one phenomenon” that asks directly about the real-life phenomenon in question. It was made sure that the questions were understood as intended, through face-validity interviews with persons from the target group. This gave the opportunity to collect a very broad set of data from the participants. Because of the cross-sectional design of the study we do not know if the data were affected by recall induced bias.

### *Analytical errors (Step IV)*

In the data analysis phase of the study there is always the risk of analytical errors. The comprehensive questionnaire included a broad range of various possible confounding variables. These had been carefully discussed and selected during the questionnaire development. Furthermore, the choice of statistical analytical methods was carefully discussed in the research group, which included both epidemiological and statistical expertise. It is, however not possible to know whether the results were influenced by unknown factors that were not included in the survey.

## **External validity**

As several years had passed since the participants had lost their parent and several more years have passed since the data used in Part 1 of this PhD project were collected, it is uncertain if their experiences are generalizable to teenagers who are facing the death of a parent today. This could be a threat to the external validity of the study, affecting the generalizability of the results to the population today. However, there is no reason to believe that the impact bereavement has on family cohesion and its association with health and wellbeing in youth has changed drastically during this time, or that the nature of grief reactions to loss has changed.

Also, not using standardized, psychometrically developed instruments limits the possibility of direct comparison between studies. However, the use of single-item questions allowed for a very comprehensive collection of data based on teenagers' own experience when losing a parent to cancer, which would have been impossible without this design. The study-specific questionnaire included 271 items, asking about their experience at different times and included a broad variety of topics. The questions and response options were understood as intended in the face validity test and in the pilot study, conducted during the development of the questionnaire.

Furthermore, the restrictions of the inclusion criterion, i.e. not including first-generation immigrants or children of single parents, limit the generalizability of the results.

## **Credibility**

“Credibility” relates to the trustworthiness of a study that is performed using qualitative research methods. According to the interpretive description approach there are four major criteria that can be used to evaluate and reflect on the quality of a study. They are: epistemological integrity, representative credibility, analytic logic and interpretive authority<sup>1</sup>. Below, they are described briefly and reflected on in relation to Study IV, together with other issues pertaining to the credibility of the study.

### *Epistemological integrity*

In the interpretive description approach, epistemological integrity is highlighted as one of the concepts enhancing the credibility of a study<sup>1</sup>. This means that the research process must evolve around a research question that is consistent with the epistemological standpoint of the research, and both the performance of the study and the analysis are able to logically adhere to the research aim. Study IV of this PhD project had a clear clinical starting point, being a health care quality improvement project. It was based on the epistemology of clinical discipline, meaning that it was based on relevant literature and best care guidelines and aimed to create knowledge that can be used to solve a problem in a real-life setting<sup>209</sup>. The aim for developing the S-PCG, was to bridge the gap between what is known to be best care practice for patients facing the death and what is applied in clinical practice. The S-PCG is a partnership production by many persons with relevant expertise in the field of palliative care and is highly grounded in clinical practice, staying true to the epistemology of clinical discipline. Interpretive description acknowledges that the disciplinary knowledge of the researcher, as a person with clinical experience and health care discipline as an educational background, will always affect the interpretation of the results<sup>1</sup>. This is not seen as a disadvantage, rather, it can serve the aim of the research to provide knowledge or develop tools that can actually be used in a real-life setting.

### *Representative credibility*

In studies following the interpretive description approach, it should be possible to demonstrate representative credibility<sup>1</sup>. To do this, it is an advantage to collect data from more than one perspective and to include methodological triangulation. This study's comprehensive study process, as described in the Methodology section, can surely be said to strengthen the representative credibility of the results. We collected data from different professions and in 40 different care units. We included both health care professionals and people with an academical background, and patients, family and public representatives. By collecting data in several ways, through written feedback, focus groups, semi structured interviews, workshops and audit of patient records, an effort was made to gain feedback from different perspectives and increase the possibility to gain insight into a broad variety of different patterns.

It can, however, be pointed out that the health care perspective had an overwhelmingly bigger representation, in terms of number of participants, than did the patient, family and public representation, and this may have affected our results. It might have benefitted the project to involve more patient, family and public representatives and to include them in the steering committee and the project group. Another criticism that could be raised is that no children were included in the creation of the sections for children as family and that their perspective was included by proxy.

Thematic analysis was chosen as a suitable way to inspire the analysis of the data as it is considered a useful method when investigating factors affecting certain processes, and to identify views regarding these processes<sup>171, 172</sup>. Also, since the thematic analysis allows for flexibility in how data are collected it suited the interpretive description approach and was considered appropriate for the study, based on the fact that the data collection during the S-PCG development process included several different type of data collection methods.

### *Analytic logic*

Another way of providing credibility to a qualitative study is to describe the analytic logic followed in the study<sup>1</sup>. This could be through an audit trail, that describes the research process from defining the problem, and describing the literature to warrant the study, to describing the methods, analysis, and interpretation of the findings<sup>210</sup>. To enhance the credibility of Study IV some of the recommended techniques were applied, such as taking field notes, taking analytical memos and reflecting on our sensemaking of the data with others in the research group, bringing in other views. The interpretations were also presented to the interdisciplinary advisory committee in a workshop, and to the project group and the steering committee, and through recurring meetings with patient and family representatives who could review, reflect and discuss the changes that had been made.

With the development of the S-PCG we wanted to address a problem that has been stated by the health authorities, that the execution of high-quality palliative care is substandard<sup>136</sup> and that it is hard to implement what is known today as “the best care practices”<sup>130</sup>. It could be seen as an disadvantage that we did not conduct our own systematic literature review at the beginning of the project, but our development project was based on two extensive reports for guiding best care practices, which had just been published, after a thorough literature review<sup>126, 154</sup>, and this provided a sound scientific basis for the study.

Using a variety of methods for data collection was one of the biggest strengths of Study IV, however, at the same time it also contributed to the biggest challenges as it generated a very big data matrix. To collect this large amount of data for interpretation did of course not give us the opportunity to dig deep into each sentence; the analysis needed to be more focused on the practical value of the feedback. For example, the audiotaped interviews were not transcribed word for word, but instead listened to while taking notes. To ensure not missing important feedback every audiotape or written comment was read or listened to by at least two persons. Notes were made, compared and discussed to ensure that all opinions were included in the big matrix, which was then used to interpret the results within the research group and later in the project group as well to guide the next steps in the development of the S-PCG. This was of course a time-consuming process but to be able to create a care guide that is applicable in different complex settings a broad variety of feedback was needed.

### *Interpretive authority*

Another important point is to always reflect on the interpretation that has been made. The interpretive description approach highlights that there is an infinitive number of ways to interpret data and we can never blindly assume that our results are the only “truth” out there<sup>211</sup>. Also, we should be aware of that the researchers’ background will influence the interpretation. To ensure that no important interpretation of the data would be missed, we first made sense of the data individually and then compared each other’s take on them, reflecting on and discussing the interpretation in a group throughout the study process. Included in the research group were people with different backgrounds and from different health care professions. An experienced researcher guided the whole process from the design of the study, through the data collection to the analysis and interpretation. This provided important opportunities to reflect and discuss the results of the study.

Since the written feedback was anonymous it was not possible to identify the individuals making the comments. We also did not, in the notes from the focus groups, state who said what. The S-PCG is aimed to be used by the whole team surrounding the patient and it should be usable for every person in the team, regardless of whether the person asking for a change is an assistant nurse, a physiotherapist or a physician. Our focus would always need to come back to

questions such as: Is this beneficial for the patient? Will this revision help the health care personnel to identify or attend to the patient's individual needs? Some might argue that using different data collection methods and gathering all the data into a one big matrix for analyses may not be appropriate. However, the initial analysis of the focus groups and interviews was made prior to the creation of the matrix, and the feedback from the interdisciplinary advisory committee was likewise analysed and gathered into a report before it was added to the matrix. All written feedback was reduced to meaning sentences, grouped based on content, and categorized before being added to the matrix.

### *Other credibility considerations*

According to the interpretive description approach, there are more factors that should be considered when examining the credibility of a study. Firstly, all research in the applied health science should be *morally defensible*. This means that it should be possible to see the need for the knowledge being produced and a potential benefit for those it should serve<sup>1</sup>.

*Disciplinary relevance* is another factor that should be considered when evaluating the credibility of a study. In other words, we must ask, Can the knowledge be beneficial to disciplinary science? Regarding Study IV, there is a clear need for improved and more equitable palliative care<sup>130</sup> and it has been highlighted that there are barriers to applying the recommended best care practices to everyday clinical practice<sup>130, 155</sup>. This provides arguments for the need to develop the S-PCG and for why this study is relevant to the health care professionals as it has the potential to provide a structure for integrating the core principles of palliative care into clinical practice.

This leads to the third and fourth factor that should also be reviewed in applied science: *pragmatic obligation* and *contextual awareness*. The former means that although it is always possible to learn more about the topic under investigation, through the eyes of others or in other settings, the results should also be able to generate usable general knowledge. Regarding contextual awareness, the feasibility of the S-PCG was tested in a variety of settings, as it was aimed to be suitable for all adult patients regardless of their age, background or place of care. The care units that tested the S-PCG were purposefully selected to represent a variety of care settings. Of course, it cannot say whether the S-PCG is appropriate for health care settings in other countries, although it was judged to be congruent with the international standards for the best care of the dying person. By carefully describing the purpose, process and context of the study, the hope is to have enabled the reader to evaluate the credibility of the study. More studies are needed to evaluate whether use of the S-PCG results in better care for people with palliative care needs and their families.



# Further research

During the course of this PhD project several considerations regarding further research were raised.

Firstly, since the question that was used to measure family cohesion was a single-item question, we do not know exactly what “family cohesion” means for the participants. In the think-out-loud face validity interviews that were performed during the questionnaire development, no participant made any comments regarding the concept of family cohesion; to them the meaning seemed to be clear. However, it would be interesting to deepen the knowledge for example through interviews with teenagers and young adults, about what family cohesion means to them, how and why the family cohesion changes after the death of a parent, what mechanisms underlie it and what factors contributes towards supporting a good family cohesion when facing the death of a parent. It would also be interesting to further investigate whether the single-item question asking directly about family cohesion is valid as a screening tool in clinical practice, and during bereavement support, to identify families in need of support.

Secondly, it would be interesting to gain a deeper knowledge on what “*an okay way to grieve*” means to teenagers and young adults and to probe deeper into what elements comprise an okay way to grieve. Likewise, it would be of interest to further research how family cohesion influences the grief experiences and grief reactions of bereaved teenagers and young adults. Also, to further investigate other grief experiences and reactions of bereaved teenagers and young adults that were not captured in our study.

Thirdly, as mentioned before, we found some gender differences in the participants’ answers and it would be of interest to further study the comprehensive and complex phenomena of family cohesion, bereavement and long-term health, while also accounting for the gender roles of both the youth and their dying parent.

Finally, since the S-PCG is a new and novel care guide, further research to evaluate the clinical impact and also the effect of the S-PCG in different care settings is needed. This involves further evaluation of patient and family outcomes, including children in the family, and the experiences of the health care personnel. Also, further studies should be conducted on the implementation of the S-PCG.

# Conclusion

Losing a parent to cancer as a teenager increases the risk of experiencing poor family cohesion after the loss, compared with non-bereaved peers. Poor family cohesion in the first year after the teenage loss of a parent increases the risk of long-term psychological health-related problems and low wellbeing in young adulthood. Furthermore, in this study the majority of participants reported that they had not, during the acute bereavement phase, found a way to grieve that had felt okay to them. Having *an okay way to grieve* was mediated by good family cohesion, male gender and having had the opportunity to have a last conversation with the dying parent. Not having had an *okay way to grieve* was in turn associated with long-term unresolved grief, 6–9 years after the death of a parent.

These results should encourage an increased awareness of family cohesion among families with teenage children who are facing the death of a parent. Helping families to strengthen their cohesion and supporting bereaved teenagers in finding a way to grieve that feels okay to them, may be a preventive health action worth considering, possibly lowering the risk of long-term unresolved grief, psychological health-related problems and low well-being among parentally bereaved teenagers. By thoughtfully implementing the S-PCG, health care personnel can take the first steps to integrate, into their clinical routines, a way of working that puts focus on children in the family and their needs, when they are facing the death of a parent or another family member.

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# Svensk sammanfattning

## Att förlora en förälder i cancer som tonåring – familjesammanhållning, sorgereaktioner och långsiktig hälsa och välbefinnande

I Sverige förlorar varje år cirka 3500 barn (under 18 år) en förälder och runt 1800 av dem är i tonåren. Bland dem finns vissa som löper ökad risk för negativa följder såsom depression, ångest och självskadebeteende. För barnens hälsa och välmående efter förlusten har familjens funktion och sammanhållning en betydelsefull roll. Kunskapen är dock begränsad, speciellt när det gäller ungdomarnas egen uppfattning om familjesammanhållning, hur den förändras över tid och dess långsiktiga effekter på deras hälsa och välbefinnande. Det saknas också kunskap när det gäller barns och ungdomars sorgupplevelser och reaktioner efter att ha förlorat en förälder. Bättre kunskap om varierande sorgereaktioner kan hjälpa kvarlevande familjemedlemmarna att hantera sorgen.

Både nationella och internationella riktlinjer framhäver att i god palliativ vård ingår stöd till familjen. Enligt svensk lagstiftning ska all personal inom hälso- och sjukvården särskilt beakta barns rätt till information, råd och stöd när en förälder är svårt sjuk eller döende. Trots detta visar forskning att barn och ungdomar efterfrågar mer stöd när en förälder avlider. Enligt Socialstyrelsen finns det en brist på rutiner och formellt stöd till barn och deras familjer som har förlorat en förälder. Aktuell forskning visar att det kan vara svårt för hälso- och sjukvårdspersonal att omsätta rekommendationerna till klinisk praxis.

För att kunna stödja ungdomar som har mist en förälder behövs en tydligare bild av deras erfarenheter före och efter dödsfallet, samt mer kunskap om faktorer som kan påverka deras hälsa och välmående. Dessutom, för att kunna erbjuda en bra vård för den döende människan, måste hälso- och sjukvården anpassa kliniska rutiner och integrera familjens, inklusive barnens, behov i den kliniska verksamheten.

Syftet med denna doktorsavhandling var att beskriva akuta och långvariga sorgereaktioner och familjesammanhållning hos unga vuxna som under tonåren mist en förälder i cancer. Vidare att beskriva samband mellan familjesammanhållning och långsiktig hälsa och välmående. Dessutom att redogöra för utvecklingen av en

nationell vårdplan för palliativ vård som också innehåller en struktur och systematik för att ge stöd till barn som närstående.

Doktorandprojektet består av två delar och innehåller fyra studier. Första delen (*Studier I–III*) utgår från en nationell populationsbaserad studie, där 622 unga vuxna (i åldern 18–26) besvarade en enkät sex till nio år efter att ha förlorat en förälder i cancer under tonåren (13–16 år) samt 330 jämnåriga från ett matchat slumpmässigt urval av befolkningen. Sambanden analyserades med univariabel och multivariabel logistisk och Poisson regression.

Andra delen (*Studie IV*) är en explorativ intervention- och utvecklings studie, baserad på tolkande beskrivning (Interpretive Description) för tillämpad vetenskap. Den beskriver utvecklingen av den Nationella Vårdplanen för Palliativ vård (NVP) som under tre perioder pilottestades i olika vårdmiljöer på 40 vårdenheter i Sverige. Utvecklingen involverade hälso- och sjukvårdspersonal, patienter, närstående och en nationell tvärprofessionell referensgrupp. Data samlades in under utvecklingsprocessen med kvalitativa metoder och analyserades med tematisk analys.

Resultaten från Studier I–III visade att ungdomar som förlorat en förälder i cancer under tonåren, hade ökad risk för låg familjesammanhållning under tonåren jämfört med kontrollgruppen, och att uppfattningen om låg familjesammanhållning kvarstod in i ung vuxenålder, men enbart bland de som hade förlorat en mamma. Bland de som hade rapporterat låg familjesammanhållning det första året efter dödsfallet av en förälder fanns ökad risk för symptom relaterade till psykisk ohälsa och lågt välmående, 6–9 år efter förlusten, jämfört med de som hade rapporterat en hög familjesammanhållning under samma period. Resultaten visade även att mer än hälften av de förlustdrabbade ungdomarna inte tyckte sig ha hittat ett sätt att sörja som ”kändes okej” för dem, under de första 6 månaderna efter föräldrarnas dödsfall. Detta visade sig också vara förknippat med långvarig obearbetad sorg.

Genom att identifiera individuella vårdbehov kan den Nationella Vårdplanen för Palliativ vård underlätta för det tvärprofessionella teamet att göra bedömningar och få en överblick över individuella palliativa vårdbehov hos patienter och deras familjer under hela den palliativa vårdprocessen (*Studie IV*).

Sammanfattningsvis genererar avhandlingen kunskap som kan användas för att bättre förstå och tillgodose de behov som finns hos barn och unga som har förlorat en förälder i cancer. Samt att beskriva stöd som kan användas för att på ett strukturerat sätt integrera personcentrerad vård som en del av kliniska rutiner för patienter med palliativa vårdbehov och deras familjer, inklusive barn som närstående.

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Paper I





# Losing a parent to cancer as a teenager: Family cohesion in childhood, teenage, and young adulthood as perceived by bereaved and non-bereaved youths

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## Abstract

**Objective:** The aim of this study was to investigate levels of perceived family cohesion during childhood, teenage years, and young adulthood in cancer-bereaved youths compared with non-bereaved peers.

**Methods:** In this nationwide, population-based study, 622 (73%) young adults (aged 18–26) who had lost a parent to cancer 6 to 9 years previously, when they were teenagers (aged 13–16), and 330 (78%) non-bereaved peers from a matched random sample answered a study-specific questionnaire. Associations were assessed using multivariable logistic regression.

**Results:** Compared with non-bereaved youths, the cancer-bereaved participants were more likely to report poor family cohesion during teenage years (odds ratio [OR] 1.6, 95% CI, 1.0–2.4, and 2.3, 95% CI, 1.5–3.5, for paternally and maternally bereaved youths, respectively). This was also seen in young adulthood among maternally bereaved participants (OR 2.5; 95% CI, 1.6–4.1), while there was no difference between paternally bereaved and non-bereaved youths. After controlling for a number of covariates (eg, year of birth, number of siblings, and depression), the adjusted ORs for poor family cohesion remained statistically significant. In a further analysis stratified for gender, this difference in perceived poor family cohesion was only noted in females.

**Conclusion:** Teenage loss of a parent to cancer was associated with perceived poor family cohesion during teenage years. This was also noted in young adulthood among the maternally bereaved. Females were more likely to report poor family cohesion. Our results indicate a need for increased awareness of family cohesion in bereaved-to-be families with teenage offspring, with special attention to gender roles.

## KEYWORDS

adolescents, bereavement, cancer, family cohesion, oncology, parental death, teenagers, young adults

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## 1 | INTRODUCTION

Losing a parent is one of the most tragic experiences that can occur in the life of a child or adolescent.<sup>1</sup> Bereaved children and youths have been shown to be at higher risk of negative consequences, such as anxiety, depression,<sup>2</sup> self-injury,<sup>3,4</sup> premature death,<sup>5</sup> and suicide attempts<sup>6</sup> compared with their non-bereaved peers.

In the literature, the most constant factors that can counteract the negative impact of bereavement are warmth and connection between the surviving parent and the bereaved child, the mental health of the surviving parent, and family functioning.<sup>7,8</sup> One of the core elements of family function is family cohesion, which is a broad concept intended to grasp the sense of emotional bonding between family members but also includes other factors, such as support and feeling of togetherness.<sup>9</sup> Poor family cohesion has been shown to be associated with anxiety and depression<sup>10</sup> and to predict higher stress responses in adolescent children of cancer-patients.<sup>11</sup> It has similarly been associated with increased mental health problems in parentally bereaved children.<sup>12,13</sup> Furthermore, family cohesion mediates the effects of parental bereavement on adolescents.<sup>7,14</sup> Also, in previous reports from this project, poor family cohesion has been strongly associated with adverse outcome.<sup>15</sup>

Family cohesion changes with time and is affected by situational stressors and changes in developmental needs as the children matures.<sup>9</sup> To be able to support bereaved children and adolescents in an efficient way, there is a need for more knowledge about which contextual family- and health care-related factors impact their well-being.<sup>16</sup> Only limited evidence exists on the impact bereavement has on the family as a unit and its function.<sup>17</sup> Further, there is a dearth of knowledge on the relationship between bereavement and family cohesion, as perceived by youths themselves.

The aim of this study was to investigate the levels of perceived family cohesion during childhood, teenage years, and young adulthood in youths who had lost a parent to cancer in their teenage years, 6 to 9 years prior to the study, compared with their non-bereaved peers.

## 2 | METHODS

For inclusion in this nationwide, population-based study, the bereaved participants needed to have lost a parent from cancer during their teenage years (at 13-16 years of age). The participants were identified through the Multi-Generation Register at Statistics Sweden by using information about the lost parents from the Swedish National Cause of Death Register. For inclusion, the lost parents had to have died before the age of 65 in the years 2000 to 2003 and been diagnosed with cancer at least 2 weeks before the death. The participant had to have been registered at the same address as both parents, and the other parent needed to be alive at the time of follow-up.

A random sample of non-bereaved participants was identified by Statistic Sweden at a ratio of 1:2 (non-bereaved:cancer-bereaved). Participants in the non-bereaved group were matched by age, gender, and place of residency. All parents were non-divorced. All participants

needed to be born in one of the Nordic countries, to understand Swedish, to have an identifiable telephone number, and live in Sweden at the time of the study.

### 2.1 | Data collection

Data collection started with an invitation letter to all participants who met the inclusion criteria, followed by an information call from a research assistant. If oral consent was given, the anonymous questionnaire, an ethics information sheet, and a reply card was sent. Information about participants' right to withdraw from the study at any time was given both orally and in writing. All participants gave oral and written consent. The reply card was returned separately in order to keep the questionnaires anonymous. Afterwards, a thank you/reminder card was sent followed by reminder phone calls to those who had not returned their reply card.

### 2.2 | Measurements

The data was collected through a study-specific questionnaire that was developed according to well-established routines.<sup>18,19</sup> This included developing the items in the questionnaire based on the literature, expert recommendations, previous questionnaires from the research group, and foremost on the topics brought up in semi-structured interviews with bereaved youths ( $n = 16$ ). The single-item questions and response alternatives were tested for face validity with 15 cancer-bereaved and two non-bereaved young adults. The concept of "family cohesion," which in the Swedish language is straightforward, was well understood. None of the participants made any remarks regarding this question throughout the process. The feasibility of the study was then tested in a pilot study. The questionnaire included a total of 271 items, of which 21 were considered relevant for this study ( $n = 5$  family cohesion,  $n = 16$  potential confounding variables).

The perception of family cohesion was evaluated with five single items, with the question:

*Did you as a family have good cohesion during:*

- your childhood (until you were approximately 11-12 years old)?
- your teenage years (until the death of your parent)?
- 0-6 months after your loss?
- 7-12 months after your loss?
- today?

There were four response alternatives: "No, not at all" and "Yes, little" (labelled poor family cohesion), "Yes, moderate" and "Yes, very good" (labelled good family cohesion).

The question for the non-bereaved participants, for whom there was no loss, had only one time-frame for the teenage years. To enable comparison of the perceived family cohesion during teenage years, between the groups, the non-bereaved participants got subquestions (b), (c), and (d) combined into one variable. Reporting poor family cohesion at one or more of these three teenage time-frames in question,

was labelled as poor family cohesion during teenage years for the cancer-bereaved participants.

### 2.3 | Data analysis

The IBM SPSS Statistics 23.0 software (IBM Corp., Armonk, N.Y., USA) was used for statistical analyses. Crude odds ratios (ORs) with 95% CIs were calculated with bivariable logistic regression. To control for possible confounding factors, 16 possible confounding variables that were considered relevant to family cohesion or bereavement were preselected on the basis of literature review and previous analysis within the research project.<sup>4,15</sup> A forward selection (likelihood ratio test) was performed on the preselected variables. All variables that met the entry criterion of  $P < .25$  at one or more of the time periods under investigation were then included in a multivariable logistic regression model used to calculate adjusted ORs with 95% CIs (Table A1). The adjusted ORs and 95% CIs for each time period were calculated with the model in three steps, every step adding more variables to the model. Further, analysis was made on the data stratified by the matching variables and also by gender of the deceased parent. Our comparisons were two-tailed and performed at the .05 significance level, apart from the forward selection (likelihood ratio test) which had the entry criterion at .25 significance level.

### 2.4 | Ethical considerations

The study was approved by the Regional Ethical Review Board of Karolinska Institute, Stockholm, Sweden (2007/836-31). To minimize the risk of causing distress to the participants, the data was not collected during holidays or during the anniversary month of participants' parental loss. The overwhelming majority of the participants perceived their participation in the study as meaningful and positive.<sup>20</sup>

## 3 | RESULTS

A total of 1272 young adults (18-26 years old) met the criteria for inclusion and were asked to participate in the study. Of these, 622 (73%) cancer-bereaved individuals, 337 of whom had lost their father and 284 their mother, and 330 (78%) non-bereaved individuals returned the questionnaire. Participants' characteristics are displayed in Table 1.

The responses of the vast majority of both cancer-bereaved and non-bereaved participants indicated good (moderate to very good) family cohesion during childhood, while 3% to 6% of the participants self-assessed the family cohesion in this period as poor (no or little) (Figure 1). Higher prevalence of perceived poor family cohesion was reported in all groups during the teenage years. In total, 20.3% of the paternally bereaved and 27.3% of the maternally bereaved participants reported poor family cohesion at one or more of the time periods during the teenage years, while 14.0% of the non-bereaved reported poor family cohesion during the teenage years. When asked about family cohesion today, ie, at the time of the survey in young

adulthood, 8.4% of the paternally bereaved participants reported poor family cohesion, while the prevalence was at 19.5% among those who had lost their mother, in comparison with 8.8% of the non-bereaved youths (Figure 1).

Table 2 shows crude ORs as well as adjusted ORs, with corresponding 95% CIs, for the reported poor family cohesion during childhood, teenage years and young adulthood. There was no statistically significant difference in reported perception of family cohesion between the groups during childhood. However, during the teenage years, the cancer-bereaved youths were more likely to report poor family cohesion compared with their non-bereaved peers: for the paternally bereaved youths, the crude OR was 1.6 (95% CI, 1.0-2.4) and for maternally bereaved youths, 2.3 (95% CI, 1.5-3.5). In young adulthood (6-9 years after the loss), the difference in perceived poor family cohesion was statistically significant for those who had lost their mother, with OR 2.5 (95% CI, 1.6-4.1), in comparison with the non-bereaved participants. The difference was not statistically significant for the paternally bereaved participants for this time period.

After the step-wise adjustments for the teenage time period, all adjusted ORs for poor family cohesion remained statistically significantly higher for the bereaved compared with the non-bereaved group, and varied between 1.5 (95% CI, 1.0-2.4) and 1.7 (95% CI, 1.1-2.7) among the paternally bereaved and between 2.2 (95% CI, 1.5-3.4) and 2.3 (95% CI, 1.5-3.8) among the maternally bereaved youths. In young adulthood, the reported perception of poor family cohesion among those who had lost a mother was statistically significantly higher compared with that among the non-bereaved participants, resulting in an adjusted OR of 2.3 (95% CI, 1.3-3.9) after the final adjustments (Table 2).

The analysis stratified by age or place of residency showed no substantial changes to the main results (data not shown). However, the cancer-bereaved females had a significantly higher risk of reporting poor family cohesion during teenage years, compared with the non-bereaved females, (OR: paternally bereaved: 2.7 [95% CI, 1.3-3.8], maternally bereaved: 3.2 [95% CI, 1.8-5.5]); and in young adulthood for the maternally bereaved females (OR: 3.5 [95% CI, 1.8-7.1]). No statistically significant difference was found between the cancer-bereaved and non-bereaved male participants.

## 4 | DISCUSSION

In this nationwide, population-based study, we found an association between the loss of a parent to cancer and poor family cohesion during the teenage years. Moreover, those who had lost their mother were more likely to report poor family cohesion also in young adulthood, 6 to 9 years after the loss. These results remained statistically significant even after adjustments for several possible confounding factors. A gender specific analyses showed that these results were statistically significant only for the female participants.

To the best of our knowledge, no previous studies have reported on family cohesion changes over time, as perceived by parentally-bereaved offspring. Factors involved in family cohesion, such as

**TABLE 1** Characteristics of the participants

|  | Cancer-Bereaved <sup>b</sup> |                     | Non-Bereaved <sup>c</sup><br>n (%) |
|--|------------------------------|---------------------|------------------------------------|
|  | n (%)                        |                     |                                    |
| Confirmed eligible <sup>a</sup>        | 851                          |                     | 421                                |
| Not reachable                          | 55 (6.5)                     |                     | 24 (5.7)                           |
| Declined participation                 | 66 (7.8)                     |                     | 28 (6.6)                           |
| Did not return the questionnaire       | 108 (12.7)                   |                     | 39 (9.3)                           |
| Participated (response rate)           | 622 (73.1)                   |                     | 330 (78.4)                         |
|  | Paternally bereaved          | Maternally bereaved |                                    |
| Gender of the deceased parent          |                              |                     |                                    |
| Male (father)                          | 337 (54.3)                   |                     | -                                  |
| Female (mother)                        |                              |                     | 284 (45.7)                         |
| Not stated <sup>d</sup>                | 1                            |                     | -                                  |
| Gender                                 |                              |                     |                                    |
| Male                                   | 170 (50.4)                   | 139 (48.9)          | 169 (51.2)                         |
| Female                                 | 167 (49.6)                   | 145 (51.1)          | 161 (48.8)                         |
| Year of birth                          |                              |                     |                                    |
| 1988-1990                              | 123 (36.7)                   | 87 (30.6)           | 119 (36.2)                         |
| 1986-1987                              | 149 (44.5)                   | 137 (48.2)          | 146 (44.4)                         |
| 1984-1985                              | 63 (18.8)                    | 60 (21.1)           | 64 (19.4)                          |
| Not stated <sup>d</sup>                | 2                            |                     | 1                                  |
| Birth order                            |                              |                     |                                    |
| Firstborn                              | 75 (22.3)                    | 69 (24.3)           | 104 (31.7)                         |
| Middle                                 | 88 (26.2)                    | 60 (21.1)           | 87 (26.5)                          |
| Youngest                               | 155 (46.1)                   | 146 (51.4)          | 127 (38.7)                         |
| No siblings                            | 18 (5.4)                     | 9 (3.2)             | 10 (3.1)                           |
| Not stated <sup>d</sup>                | 1                            |                     | 2                                  |
| Current employment status <sup>e</sup> |                              |                     |                                    |
| Studying at high school level          | 16/332 (4.8)                 | 8/281 (2.8)         | 13/325 (4.0)                       |
| Adult education at high school level   | 19/332 (5.7)                 | 12/280 (4.3)        | 18/325 (5.5)                       |
| Studying at university level           | 88/332 (26.5)                | 99/280 (35.4)       | 112/327 (34.2)                     |
| Employed or self-employed              | 199/335 (59.4)               | 155/280 (55.4)      | 182/326 (55.8)                     |
| Unemployed                             | 44/334 (13.2)                | 47/281 (16.7)       | 53/323 (16.4)                      |
| On parental leave                      | 3/332 (0.9)                  | 6/280 (2.1)         | 2/324 (0.6)                        |
| On sick leave                          | 3/332 (0.9)                  | 4/280 (1.4)         | 4/324 (1.2)                        |
| Residential region                     |                              |                     |                                    |
| Rural                                  | 23 (6.9)                     | 31 (11.0)           | 30 (9.1)                           |
| Small village or town                  | 72 (21.6)                    | 41 (14.5)           | 60 (18.3)                          |
| Mid-sized town                         | 146 (43.7)                   | 137 (48.6)          | 156 (47.6)                         |
| City of more than 500 000              | 93 (27.8)                    | 73 (25.9)           | 82 (25.0)                          |
| Not stated <sup>d</sup>                | 3                            | 2                   | 2                                  |
| Father's year of birth                 |                              |                     |                                    |
| 1960-                                  | 27 (8.3)                     | 33 (12.0)           | 63 (19.4)                          |
| 1955-1959                              | 11 (34.3)                    | 93 (34.8)           | 111 (34.3)                         |
| 1950-1954                              | 109 (33.6)                   | 75 (28.1)           | 93 (28.7)                          |
| -1949                                  | 32 (9.9)                     | 46 (17.2)           | 57 (17.6)                          |

(Continues)

TABLE 1 (Continued)

|                         | Cancer-Bereaved <sup>b</sup> |           | Non-Bereaved <sup>c</sup><br>n (%) |
|-------------------------|------------------------------|-----------|------------------------------------|
|                         | n (%)                        |           |                                    |
| Not stated <sup>d</sup> | 12                           | 9         | 6                                  |
| Mother's year of birth  |                              |           |                                    |
| 1960-                   | 72 (22.2)                    | 53 (19.9) | 112 (35.2)                         |
| 1955-1959               | 111 (34.3)                   | 93 (34.8) | 118 (37.1)                         |
| 1950-1954               | 109 (33.6)                   | 75 (28.1) | 64 (20.1)                          |
| -1949                   | 32 (9.9)                     | 46 (17.2) | 24 (7.6)                           |
| Not stated <sup>d</sup> | 13                           | 17        | 12                                 |

<sup>a</sup>All those identified by the registers who met the inclusion criteria.

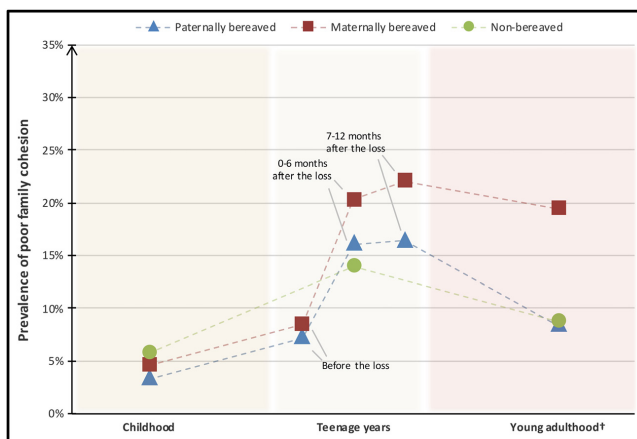
<sup>b</sup>Young adults who lost a parent to cancer between the ages of 13 and 16 years in Sweden, 2000-2003.

<sup>c</sup>A random sample from the Swedish population, matched for age, sex, and residency to the cancer-bereaved young adults.

<sup>d</sup>The group "not stated" is not included in calculations of prevalence.

<sup>e</sup>Participants were allowed to report more than one alternative.

**FIGURE 1** Prevalence of perceived poor (no/little) family cohesion among non-bereaved and cancer-bereaved youths at different time periods <sup>†</sup>At the time of the survey (aged 18–26). Note. For graphical reasons, only the frequencies between 0% and 35% are included in the Figure.



communication, emotional connection, perceived support, and relationships within the family, might possibly explain the increased prevalence of poor family cohesion among the bereaved participants.

Previous research has showed that family function is based on the interaction between individuals in the family, and when one dies, the others need to adapt to a new constellation,<sup>21</sup> affecting the whole family system. The relationship dynamics between the surviving parent and child change after the death of a parent.<sup>22</sup> This is supported in a long-term follow-up study, which showed that parentally bereaved youths had less harmonious relations with their surviving mother or father, including lack of communication, compared with their peers in non-bereaved families.<sup>23</sup> The relationship with the surviving parent has been shown to be a major factor influencing the children's coping skills and well-being.<sup>7,22,24</sup> The surviving parent is him or herself going through bereavement and emotional difficulties that may affect the capability of giving emotional support to their children or conducting positive parenting.<sup>24,25</sup>

Our results also show that among the maternally bereaved youths, the perception of poor family cohesion appeared to continue into young adulthood, years after the loss of the mother. Among the paternally bereaved participants, however, the level of perceived family cohesion in young adulthood did not differ from that in the non-bereaved controls. Studies have shown that widowed fathers have more difficulties in adapting to life after a partner's death,<sup>26</sup> while women have better coping strategies when adjusting to bereavement.<sup>27</sup> Communication, emotional bonding, and support are some of the core components of family cohesion,<sup>9</sup> and in comparison with mothers, widowed fathers have been shown to be less likely to communicate about emotions,<sup>25,28</sup> provide positive parenting,<sup>28</sup> or react to the children's loss-related needs.<sup>25</sup> However, Werner-Lin and Blank argue that the difference seen in the family adaptation to loss of a parent may be based, not on the gender of the surviving parent, but, rather, on the role the surviving parent played in the family's life preceding the illness and the death.<sup>29</sup>

**TABLE 2** Perceived family cohesion in cancer-bereaved and non-bereaved youths at different time periods

|                                    | Family Cohesion                 |                                     | Missing n (%) | Unadjusted      |                                    | Adjusted <sup>a</sup>              |                                    |
|------------------------------------|---------------------------------|-------------------------------------|---------------|-----------------|------------------------------------|------------------------------------|------------------------------------|
|                                    | Poor (no or little) n/total (%) | Good (moderate or good) n/total (%) |               | OR (95% CI)     | OR <sub>Adjustment1</sub> (95% CI) | OR <sub>Adjustment2</sub> (95% CI) | OR <sub>Adjustment3</sub> (95% CI) |
| <b>Childhood</b>                   |                                 |                                     |               |                 |                                    |                                    |                                    |
| Cancer-bereaved                    | 24/619 (3.9)                    | 595/619 (96.1)                      | 3 (0.5)       | 0.7 (0.4-1.2)   | 0.7 (0.4-1.2)                      | 0.6 (0.3-1.1)                      | 0.7 (0.3-1.3)                      |
| -Paternally bereaved               | 11/336 (3.3)                    | 325/336 (96.7)                      | 1 (0.3)       | 0.6 (0.3-1.2)   | 0.6 (0.3-1.2)                      | 0.5 (0.2-1.1)                      | 0.6 (0.2-1.2)                      |
| -Maternally bereaved               | 13/282 (4.6)                    | 269/282 (95.4)                      | 2 (0.7)       | 0.8 (0.4-1.6)   | 0.8 (0.4-1.6)                      | 0.7 (0.3-1.5)                      | 0.8 (0.4-1.7)                      |
| Non-bereaved                       | 19/329 (5.8)                    | 310/329 (94.2)                      | 1 (0.3)       | 1.0 [reference] | 1.0 [reference]                    | 1.0 [reference]                    | 1.0 [reference]                    |
| <b>Teenage years</b>               |                                 |                                     |               |                 |                                    |                                    |                                    |
| Cancer-bereaved                    | 145/618 (23.5)                  | 473/618 (76.5)                      | 4 (0.6)       | 1.9 (1.3-2.7)   | 1.9 (1.3-2.7)                      | 1.8 (1.2-2.7)                      | 2.0 (1.3-3.0)                      |
| -Paternally bereaved               | 68/335 (20.3)                   | 267/335 (79.7)                      | 2 (0.6)       | 1.6 (1.0-2.4)   | 1.6 (1.0-2.4)                      | 1.5 (1.0-2.4)                      | 1.7 (1.1-2.7)                      |
| -Maternally bereaved               | 77/282 (27.3)                   | 205/282 (72.7)                      | 2 (0.7)       | 2.3 (1.5-3.5)   | 2.2 (1.5-3.4)                      | 2.2 (1.4-3.4)                      | 2.4 (1.5-3.8)                      |
| Non-bereaved                       | 46/329 (14.0)                   | 283/329 (86.0)                      | 1 (0.3)       | 1.0 [reference] | 1.0 [reference]                    | 1.0 [reference]                    | 1.0 [reference]                    |
| <b>Young adulthood<sup>b</sup></b> |                                 |                                     |               |                 |                                    |                                    |                                    |
| Cancer-bereaved                    | 82/611 (13.42)                  | 529/611 (86.6)                      | 11 (1.8)      | 1.6 (1.0-2.5)   | 1.6 (1.0-2.5)                      | 1.6 (1.0-2.6)                      | 1.6 (1.0-2.6)                      |
| -Paternally bereaved               | 28/333 (8.4)                    | 305/333 (91.6)                      | 4 (1.2)       | 1.0 (0.6-1.6)   | 1.0 (0.6-1.7)                      | 1.0 (0.6-1.7)                      | 1.0 (0.5-1.7)                      |
| -Maternally bereaved               | 54/277 (19.5)                   | 223/277 (80.5)                      | 7 (2.5)       | 2.5 (1.6-4.1)   | 2.4 (1.5-4.0)                      | 2.5 (1.5-4.1)                      | 2.5 (1.5-4.2)                      |
| Non-bereaved                       | 29/330 (8.8)                    | 301/330 (91.2)                      | 0 (0.0)       | 1.0 [reference] | 1.0 [reference]                    | 1.0 [reference]                    | 1.0 [reference]                    |

Abbreviations: CI, confidence interval; OR, odds ratio.

<sup>a</sup>Variables added to the logistic regression model and used to calculate the adjusted ORs: OR<sub>Adjustment 1</sub>: gender, year of birth, residential region. OR<sub>Adjustment 2</sub>: variables from OR<sub>Adjustment 1</sub> + birth order, number of siblings, mother's year of birth, father's year of birth, educational level of mother, educational level of father, ever been bereaved of a sibling, depression in at least one parent, alcohol/drug misuse in at least one parent. OR<sub>Adjustment 3</sub>: variables from OR<sub>Adjustment 1, 2</sub> + have been bullied, have been physically assaulted or sexually violated, have ever been diagnosed with depression.<sup>b</sup>Young adulthood: At the time of the survey (aged 18-26).

Further analysis on the basis of the gender of the participant showed higher levels of perceived poor family cohesion among the bereaved female participants compared with the non-bereaved females, while no significant difference was found between the male participants. Family relationships have been shown to be especially prominent to female adolescents' well-being,<sup>30</sup> and they experience more emotional distress as a reaction to poor family cohesion compared with boys.<sup>31</sup> Bereaved girls have also been shown to have greater likelihood to internalize problems<sup>1</sup> and greater vulnerability than boys<sup>1,14</sup> as well as a stronger likelihood to take on more responsibility for the family life.<sup>14</sup> Our results indicate that an awareness may be needed for bereaved-to-be families with teenagers according to their role in the family and gender.

The large sample size and high participation rate (73%–78%) are the main strengths of this nationwide, population-based study. Another strength was the well-prepared and comprehensive questionnaire that was based on qualitative interviews with both bereaved and non-bereaved young adults.

Throughout the study process, an epidemiological framework adapted to this field of research was followed.<sup>32</sup> To enable adjustments, we assessed numerous possible confounding factors. When examining possible confounding factors during the data analysis phase, we performed an initial sorting by examining them one by one in relation to the outcome with a generous cut-off level (0.25) to maximize the possibility of finding factors that would explain our findings.

The questionnaire was designed using one direct question per phenomenon, where all questions were directly related to the real-life phenomena under investigation. This, enabled a comprehensive collection of data on teenagers' experience when losing a parent to cancer.

The comprehensive concept of family cohesion was self-assessed through a subjective global measurement. All of the existing validated instruments for family cohesion included a large number of items and none of them was validated for our target group at the time of data collection. In line with that, a recent systematic review of self-report family assessment measures stated that all of the validated instruments use a large number of items and no evidence exists of their responsiveness to changes in family functioning over time.<sup>33</sup>

Using a global-single-item question can sometimes be more preferable when measuring a complex phenomenon than using answers from a multiple-item scale that have been computed into one single rating.<sup>34</sup> This allows the participants to weigh into their own assessment those aspects of the phenomenon that are relevant to them.<sup>34</sup>

Since the comprehensive concept of family cohesion was self-assessed through a subjective global measurement, we cannot exactly define what family cohesion means for each participant. However, we assume that at the moment of answering the questionnaire, the feeling is real to the participant. Furthermore, none of the participants, made any remarks regarding the concept of family cohesion during the face-validity interviews. They all seemed to have a clear picture of what family cohesion meant to them.

## 4.1 | Study limitations

Our study design implies the possibility of recall-induced bias regarding data from the childhood and teenage time periods. On the other hand, to collect the data prospectively was not considered as an option because of practical, economical, and ethical reasons. Furthermore, a recent study investigating the accuracy of retrospective reports on family environment as experienced by adolescence found that retrospective and prospective reports agreed well regarding the emotional dimensions of the family life (such as family cohesion), that can be well captured with retrospective reports.<sup>35</sup>

We also have no knowledge about whether the level of family cohesion differed between our participants and the young adults who declined participation in our study, and the generalizability of our findings may not be applicable outside our setting and population.

## 4.2 | Clinical implications

Our findings showed that losing a parent to cancer as a teenager increases the risk of poor family cohesion as perceived by parentally bereaved youth. Impaired family cohesion has been shown to be associated with a number of negative outcomes for adolescents.<sup>15,36</sup> Hopefully, our findings will encourage clinicians caring for dying parents with teenage offspring to pay attention to the family cohesion, to identify those at increased risk of poor family cohesion in bereavement, and to provide support as needed. According to the results of two systematic reviews, supportive interventions can benefit bereaved-to-be families with minor children, although further research is still needed.<sup>37,38</sup> It has been shown that an intervention such as "The Family Bereavement Program" can strengthen the relationship between the surviving parent and the bereaved child or adolescent, which can have a positive effect on both the parent's and the child's health and well-being.<sup>24,39,40</sup>

## 5 | CONCLUSIONS

In this nationwide, population-based study, we found that for teenagers, losing a parent to cancer increases the risk of poor family cohesion during the teenage years, when compared with non-bereaved peers. The perception of poor family cohesion lasted into young adulthood among the maternally bereaved youths. However, these findings were only noted among females. These results warrant further investigations of family cohesion among youths facing bereavement, including influencing factors within the family, as well as bereavement support.

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## CONFLICT OF INTEREST STATEMENT

The authors have no conflict of interest to report.

## DATA AVAILABILITY STATEMENT

The data that support the findings of this study are not publicly available because of legal and ethical restrictions as described by the Swedish law and ethical boards regarding data of sensitive nature. This is in order to assure data confidentiality and to protect the privacy of the research participants.

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## APPENDIX A

### Overview of the preselected variables and variables associated with reported family cohesion at different time periods

| Childhood  | Forward Selection P <sup>a</sup> | Teenage Years   | Forward Selection P <sup>a</sup> | Young adulthood <sup>b</sup>                                      | Forward Selection P <sup>a</sup> |
|--|----------------------------------|---|----------------------------------|---|----------------------------------|
| Background variables of the participant, added at step one (adjustment 1)                      |                                  |   |                                  |   |                                  |
| Gender   |                                  | ✓Gender   | .001                             | Gender  |                                  |
| Year of birth  |                                  | ✓Year of birth  | .102                             | Year of birth   |                                  |
| Residential region   |                                  | ✓Residential region   | .151                             | ✓Residential region   | .040                             |
| Religious or spiritual   |                                  | Religious or spiritual  |                                  | Religious or spiritual  |                                  |
| Background variables of parents and family-related variables, added at step two (Adjustment 2) |                                  |   |                                  |   |                                  |
| Number of siblings   |                                  | ✓Number of siblings   | .006                             | Number of siblings  |                                  |
| ✓Birth order   | .250                             | ✓Birth order  | .211                             | Birth order   |                                  |
| ✓Mother's year of birth  | .128                             | Mother's year of birth  |                                  | ✓Mother's year of birth   | .020                             |
| Father's year of birth   |                                  | Father's year of birth  |                                  | ✓Father's year of birth   | .001                             |
| Educational level of mother  |                                  | ✓Educational level of mother                                      | .090                             | Educational level of mother                                       |                                  |
| Educational level of father  |                                  | ✓Educational level of father                                      | .180                             | Educational level of father                                       |                                  |
| Ever been bereaved of a sibling  |                                  | ✓Ever been bereaved of a sibling                                  | .140                             | Ever been bereaved of a sibling                                   |                                  |
| Depression in at least one parent  |                                  | ✓Depression in at least one parent                                | .005                             | Depression in at least one parent                                 |                                  |
| ✓Alcohol/drug misuse in at least one parent  | <.001                            | ✓Alcohol/drug misuse in at least one parent                       | <.001                            | ✓Alcohol/drug misuse in at least one parent                       | .003                             |
| Adverse events added at step three (Adjustment 3)  |                                  |   |                                  |   |                                  |
| ✓Have experienced being bullied  | .025                             | ✓Have experienced being bullied                                   | .002                             | ✓Have experienced being bullied                                   | .001                             |
| ✓Have experienced being physically assaulted or sexually violated                              | .008                             | ✓Have experienced being physically assaulted or sexually violated | <.001                            | ✓Have experienced being physically assaulted or sexually violated | .129                             |
| Have ever been diagnosed with depression   |                                  | ✓Have ever been diagnosed with depression                         | .250                             | ✓Have ever been diagnosed with depression                         | .035                             |

<sup>a</sup>The P values are based on forward selection (likelihood ratio test) with the entry criterion of P < .25.

<sup>b</sup>At the time of the survey (today), when participants were aged 18 to 26 years.

✓Variables included in the multivariable logistic regression model after meeting the entry criterion of the forward selection (likelihood ratio test).





Paper II





RESEARCH

Open Access

# Acute and long-term grief reactions and experiences in parentally cancer-bereaved teenagers



Tove Bylund-Grenklo<sup>1\*†</sup>, Dröfn Birgisdóttir<sup>2\*†</sup>, Kim Beernaert<sup>3,4</sup>, Tommy Nyberg<sup>5,6</sup>, Viktor Skokic<sup>6</sup>, Jimmie Kristensson<sup>2,7</sup>, Gunnar Steineck<sup>6,8</sup>, Carl Johan Fürst<sup>2</sup> and Ulrika Kreicbergs<sup>9,10</sup>

## Abstract

**Background:** Previous research shows that many cancer-bereaved youths report unresolved grief several years after the death of a parent. Grief work hypothesis suggests that, in order to heal, the bereaved needs to process the pain of grief in some way. This study explored acute grief experiences and reactions in the first 6 months post-loss among cancer-bereaved teenagers. We further explored long-term grief resolution and potential predictors of having had “an okay way to grieve” in the first months post-loss.

**Methods:** We used a population-based nationwide, study-specific survey to investigate acute and long-term grief experiences in 622 (73% response rate) bereaved young adults (age > 18) who, 6–9 years earlier, at ages 13–16 years, had lost a parent to cancer. Associations were assessed using bivariable and multivariable logistic regression.

**Results:** Fifty-seven per cent of the participants reported that they did not have a way to grieve that felt okay during the first 6 months after the death of their parent. This was associated with increased risk for long-term unresolved grief (odds ratio (OR): 4.32, 95% confidence interval (CI): 2.99–6.28). An association with long-term unresolved grief was also found for those who reported to have been numbing and postponing (42%, OR: 1.73, 95% CI: 1.22–2.47), overwhelmed by grief (24%, OR: 2.02, 95% CI: 1.35–3.04) and discouraged from grieving (15%, OR: 2.68, 95% CI: 1.62–4.56) or to have concealed their grief to protect the other parent (24%, OR: 1.83, 95% CI: 1.23–2.73). Predictors of having had an okay way to grieve included being male, having had good family cohesion, and having talked about what was important with the dying parent.

(Continued on next page)

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**Conclusion:** More than half of the cancer-bereaved teenagers did not find a way to grieve that felt okay during the first 6 months after the death of their parent and the acute grief experiences and reaction were associated with their grief resolution long-term, i.e. 6–9 years post-loss. Facilitating a last conversation with their dying parent, good family cohesion, and providing teenagers with knowledge about common grief experiences may help to prevent long-term unresolved grief.

**Keywords:** Adolescents, Bereavement, Cancer, Grief, Loss, Mourning, Oncology, Parental death, Teenagers, Unresolved grief, Young adults

*"I remember once waking up in the middle of the night with an excruciating pain in my heart. It was stabbing, aching and burning. If I had not been told that psychological pain can manifest itself in physical pain I would have thought that I was dying that night. The pain was so intense; I think my heart broke in thousands of pieces that night.*

*Today, eight years later, my heart is no longer in thousands of pieces – at least not for any longer period of time."*

Quote from one of the participants, a daughter who at age 14 lost her mother [1](p.31).

## Introduction

Undoubtedly, for a child, one of the most devastating experiences is the early death of a parent, which can seriously affect their health and wellbeing [2–7] in the short and long term. Bereavement in children and adolescents has been shown to be associated with increased risk of suicide attempts [8, 9] and increased mortality [10, 11], and previous research based on the same study sample as the current study showed almost a doubled risk of self-injury in the first 6–9 years following teenage bereavement [12, 13]. While not all bereaved children and adolescents will face these negative outcomes of bereavement [14, 15], risk factors such as sudden, unexpected or traumatic [5] loss, parental depression [16] and poor family cohesion [17, 18] have been identified. Complicated or prolonged grief is another factor that has been shown to be associated with those negative health-related outcomes in bereaved children and adolescents [19, 20]. This includes symptoms such as separation distress, pre-occupation with thoughts about the deceased person, and difficulties in accepting the loss or in returning to normal functioning after the loss [21–23]. It should be noted that the categorization of complicated or prolonged grief is still debated, particularly in children and adolescents [24, 25].

In bereaved adults, the characteristics of the grieving process are considered to be of importance for their wellbeing after bereavement [26–28]. In the immediate

phase after bereavement, grief may often include powerful emotions such as shock, numbness, crying, anxiety and anger [29, 30] and many theories concerning coping with or recovering from loss, regardless of whether they focus on stages [11] or tasks [12], include the notion that, in order for the person to heal, they must deal with the pain in so-called "grief work". Since Freud first came forward with this notion, the understanding of what "working through the grief" entails has changed over time [31], challenging the assumption that "grief work" is only a cognitive process of confronting the loss [31]. This can be seen in one of today's relevant grief-theories, the Dual Process Model, stating that it is part of the normal grieving process for people to shift in and out of the intense emotional reaction to loss, described as oscillation between loss- and restoration-oriented grief reactions [32]. The Dual Process model was initially designed to understand conjugal bereavement [31] and yet more research is needed to build up the empirical evidence among bereaved children and teenagers. Grief is a unique experience and is highly influenced by individual traits, the relationship with the deceased and the circumstances surrounding the death, as well as social and cultural factors [25], and grief reactions among children and teenagers can differ from adults' reactions [33]. Children and teenagers can often only tolerate the emotional pain for a short period of time compared with adults, shifting between intense feelings such as yearning, sadness or anger to rapidly returning to normal activities [33–35]. It has been highlighted that more knowledge is needed about the grieving process of children and teenagers [36–38] and many experts in the field seem to agree that not all knowledge from the adult bereavement research field can be transferred directly to children and teenagers [24]. Knowledge on various grief reactions of children and teenagers can be helpful for both bereaved children and their parents while dealing with the loss [39–41]. Nevertheless, for the last decades, the focus within the bereavement literature has mostly been on what is sometimes referred to as "pathological grief responses" among children and teenagers, while more research is also needed to better understand "normative grieving processes" [25]. While knowledge

regarding e.g. the needs of, and the meaning of grief in bereaved children, adolescents and young adults are increasingly being documented [42–44], we still need more knowledge about the youths' reactions and experiences of grief, in the immediate phase and long-term [24, 37].

To be able to provide more knowledge and to reduce suffering among parentally bereaved teenagers, more research based on teenagers' own experience is needed to describe their normative and pathological grieving processes [25] both during the acute bereavement phase and long-term.

In the preparatory interviews with cancer-bereaved youths that were performed for this research project, the parentally bereaved informants described a range of different grief reactions in the immediate post-loss phase. Some concluded that they had not found “an okay way to grieve” (data not published).

The aim of this exploratory population-based study was to investigate 1) the prevalence of a set of grief experiences and reactions in the acute bereavement phase, i.e. the first 6 months post-loss, and 2) their possible associations with unresolved grief long-term, 6–9 years after the loss of a parent to cancer, as self-assessed by cancer-bereaved youths. Further, we explored the associations between demographic, family, and health care-related factors, and the experience of having had an okay way to grieve in the first 6 months post-loss.

## Method

### Study design and study population

We conducted a population-based nationwide survey in 2009–2010 in young adults who, during their teenage years, had lost a parent to cancer. The Swedish Cause of Death Register identified the individuals who had died from cancer at an age younger than 65 (based on International Classification of Diseases, 10th revision (ICD-10), codes C00–C96) in 2000–2003. This information was then used by the Multi-Generation Register to identify children who were bereaved of a parent between the ages of 13 and 16 and who had been living with both parents at the time of the loss. Because of the great variation in maturity levels during the teenage years [45], we decided to restrict this study to the youngest group of teenagers that would match grades 7 to 9 in the Swedish middle school.

For inclusion, the participants had to be living in Sweden at the time of the survey, be fluent in Swedish, and have an identifiable telephone number; also, their other parent still had to be alive. Altogether, 851 bereaved former teenagers were confirmed eligible for the study. All participants were between 18 and 26 years old at the time of the data collection. More details on the study protocol have been published elsewhere [46].

### Data collection

At the beginning of the data collection, each participant first received an introductory letter explaining the study objective. A questionnaire was sent only to those who, during a subsequent informative telephone call, consented to participate. Participants were informed both orally and in writing about their right to withdraw from the study at any time. The questionnaires were returned in pre-stamped envelopes, separately from the response cards in order to ensure anonymity. After a few weeks, a combined thank you and reminder card was posted, followed by reminder telephone calls to those whose responses were missing.

### Questionnaire development

A study-specific questionnaire was developed based on semi-structured interviews with 15 cancer-bereaved youths, and interviews with three health care professionals specialized in grief and palliative care, as well as the bereavement literature. To ensure that the questions we constructed were understood as intended, we tested the face validity of the questionnaire and response options with 15 cancer-bereaved former teenagers (six previously interviewed and nine newly invited individuals) in think-aloud interviews. Questionnaire development followed well-established routines that have been previously described [47, 48]. The final questionnaire contained 271 question items, set in different time frames, i.e. childhood, teenage years (before and after the loss), and young adulthood (at the time of the survey). A total of 37 items were considered relevant for this study.

### Measurements

We used six single-item questions [49] to assess coping styles, grief experiences, expressions and behaviours in the acute bereavement phase, i.e. during the first 6 months after the loss of a parent (hereafter referred to as “*Acute grief experiences and reactions*”). These questions all started with “For the first half-year after your loss, would you agree with the statement: ...”, followed by:

- “I had a way to grieve that felt okay.” (hereafter labelled: *Had an okay way to grieve*, or as its negative counterpart; *Did not have an okay way to grieve (R)* for its reversed form)
- “I clenched my teeth, built a wall around me and lived on as if nothing had happened.” (*Numbing and postponing*)
- “I withheld my grief to protect my other parent.” (*Concealed grief*)
- “The grief was so strong it felt as if I would not survive, as if I was going crazy or was not normal.” (*Overwhelmed by grief*)

- “People stopped me from grieving by drawing away when I was sad or praising me when I was being strong.” (*Discouraged from grieving*)
- “There was pressure from others that I should be more sad than I was showing.” (*Pressured to grieve*)

The response options for all abovementioned questions were: “Completely agree”, “Moderately agree”, “Slightly agree” and “Do not agree at all”.

Long-term grief resolution, i.e. at the time of the survey (6–9 years after the loss), was measured with the single-item question:

- “Have you worked through your grief?”, with the response options “No, not at all”, “Yes, a little”, “Yes, moderately” and “Yes, completely”.

This single-item question was well understood by bereaved participants in the face-validity interviews and has been used in previous studies [17, 50–53]. In a study on young adults, cancer-bereaved of a sibling, this question was validated against three questions from the Inventory of Complicated Grief (ICG), and found to be strongly correlated to them [53].

Additionally, we used ten demographic variables (e.g. gender of the child), three family-related variables (e.g. family cohesion), and 15 health care-related variables (e.g. teenagers’ level of trust in the health care provided to the dying parent in the final week of life) in our data analysis.

#### Data analysis

The responses to all of the items measuring the grief experiences and reactions during the acute bereavement phase (the first 6 months post-loss) were dichotomized into “Agree” (moderately, and completely agree) and “Disagree” (slightly agree, and do not agree at all). The responses “No” and “Yes, a little” to the question of having worked through grief were labelled as “Unresolved grief” while “Yes, moderately” and “Yes, completely” were labelled as *having worked through grief*.

The relationship between the six acute grief experiences and reactions in the first 6 months following the loss, and perceived *unresolved grief* at follow-up was evaluated in terms of odds ratios (ORs). The unadjusted estimates were calculated using logistic regression which was then subsequently adjusted for three groups of possible confounders. The adjustment scheme applied decomposes into two steps. In the first step, all of the available possible confounders were classified as belonging to one of the classes “background variables”, “family-related variables” and “health care-related variables”. Within each group a logistic regression with a forward selection procedure was performed, using the variables

as predictors of “unresolved grief at follow-up”. Selection was based on likelihood ratio  $p$ -values, with a  $p$ -value of 0.25 used as a stopping criterion. This means that the selection procedure was aborted if none of the remaining candidate variables were associated with a  $p$ -value of 0.25 or less when included in the model. Prior to each selection all individuals with missing values on any of the variables within a particular group of variables were excluded. In the second step, the groups of variables selected by the forward selection procedures were sequentially used to calculate the adjusted ORs with 95% confidence intervals (CIs).

A further analysis of the data was performed, where both crude and adjusted ORs were calculated again with the same three groups of possible confounders as before, but now with the data stratified by the gender of the participants.

In order to assess what variables might be associated with *Having had an okay way to grieve* in the 6 months following the loss, all variables considered in the previous analysis were treated as potential predictors of this outcome in bivariable logistic regression models. Once again likelihood ratio  $p$ -values were used to evaluate their predictive performance. The significant variables ( $p$ -value < 0.05) were subsequently used in conjunction as predictors in a multivariable logistic regression model, in order to investigate the effect of correlations among them on their significance as predictors.

#### Results

A total of 851 cancer-bereaved youths (teenagers at the time of their loss) were confirmed eligible, 622 (73%) of whom returned the questionnaire. Fifty-four per cent of participants had lost their father and 46% had lost their mother. The characteristics of the participants are presented in Table 1.

#### Prevalence of the different acute grief experiences and reactions

Among the participants, 57% reported that they *had not had an okay way to grieve (R)*, as can be seen in Table 2. The most often agreed with out of the remaining five acute grief reactions were *numbing and postponing* (42%), *concealed grief* to protect the other parent (25%) and *being overwhelmed by grief* (24%). A total of 79 participants, or 13%, disagreed with all of the statements regarding grief during the acute bereavement phase.

#### Associations between the acute grief reactions and long-term unresolved grief

Forty-five per cent of the participants reported not having worked through their grief at the time of the survey 6–9 years post-loss. Table 3 shows the associations between the six acute grief experiences and reactions in

**Table 1** Characteristics of the study population

|  | N (%)            |
|--|------------------|
| <b>Confirmed eligible<sup>1</sup></b>                                  | <b>851 (100)</b> |
| Unreachable  | 55 (6)           |
| Declined to participate  | 66 (8)           |
| Agreed initially but did not return the questionnaire                  | 108 (13)         |
| <b>Provided information</b>  | <b>622 (73)</b>  |
| <b>Gender of the participants</b>                                      |                  |
| Male   | 309 (50)         |
| Female   | 312 (50)         |
| Not stated   | 1                |
| <b>Year of birth (age, in years, at the time of the survey)</b>        |                  |
| 1988–1990 (19–21)  | 210 (34)         |
| 1986–1987 (22, 23)   | 286 (46)         |
| 1984–1985 (24–26)  | 123 (20)         |
| Not stated   | 3                |
| <b>Birth order</b>   |                  |
| Oldest child   | 144 (23)         |
| Middle child   | 148 (24)         |
| Youngest child   | 302 (49)         |
| Only child   | 27 (4)           |
| Not stated   | 1                |
| <b>Living arrangement and marital status</b>                           |                  |
| Lives with parent, is single   | 134 (22)         |
| Lives with parent, has a partner (living apart)                        | 70 (11)          |
| Has moved away from parent, is single                                  | 153 (25)         |
| Has moved away from parent, has a partner (living apart)               | 86 (14)          |
| Lives with partner or spouse   | 176 (28)         |
| Not stated   | 3                |
| <b>Highest level of education attained (at the time of the survey)</b> |                  |
| Not applicable, never graduated  | 6 (1)            |
| Middle school ( $\leq$ 9th grade)                                      | 49 (8)           |
| High school ( $\geq$ 10th grade)                                       | 501 (81)         |
| College/university   | 54 (9)           |
| Other type of studies  | 11 (2)           |
| Not stated   | 1                |
| <b>Current employment status<sup>2</sup></b>                           |                  |
| Studying at high school level  | 24/614 (4)       |
| Adult education at high school level                                   | 31/613 (5)       |
| Studying at university level   | 187/613 (30)     |
| Employed or self-employed  | 355/616 (58)     |
| Unemployed   | 91/616 (15)      |
| On parental leave  | 9/613 (2)        |
| On sick leave  | 7/613 (1)        |

**Table 1** Characteristics of the study population (*Continued*)

|                                      | N (%)    |
|--------------------------------------|----------|
| <b>Residential area</b>              |          |
| Rural                                | 54 (9)   |
| Small village or town                | 113 (18) |
| Medium-sized town                    | 283 (46) |
| City of more than 500,000            | 166 (27) |
| Not stated                           | 6        |
| <b>Gender of the deceased parent</b> |          |
| Male                                 | 337 (54) |
| Female                               | 284 (46) |
| Not stated                           | 1        |

<sup>1</sup> Confirmed eligible = all those identified in registers who met the inclusion criteria

<sup>2</sup> More than one response alternative could be selected for this question. Number of responses per answer is provided

the first 6 months post-loss and reported unresolved grief 6–9 years later (i.e. at the time of the survey). The participants reporting not *having had an okay way to grieve (R)* were statistically significantly more likely to report *long-term unresolved grief* (OR: 4.32, 95% CI: 2.99–6.28). Statistically significant associations with unresolved grief long-term were also found in those who reported to have been *numbing and postponing* (OR: 1.73, 95% CI: 1.22–2.47), to have been *overwhelmed by grief* (OR: 2.02, 95% CI: 1.35–3.04), to have been *discouraged from grieving* (OR: 2.68, 95% CI: 1.62–4.56) and to have *concealed their grief* to protect the surviving parent (OR: 1.83, 95% CI: 1.23–2.73). All these associations remained statistically significant after controlling for the selected possible confounding demographic variables, family-related variables and health care-related variables (Table 3).

#### Gender-stratified analysis

Forty-nine per cent of the parentally cancer-bereaved male participants and 65% of the female participants reported *not having had a way to grieve that felt okay (R)* to them during the acute bereavement phase. Figure 1 illustrates the reported prevalence of the different grief experiences and reactions during the acute bereavement phase, subdivided by gender. Figure 2 shows the reported prevalence of grief resolution 6–9 years after the loss of a parent, where 37% of the male and 52% of the female participants reported long-term unresolved grief.

In further analysis of the data stratified by gender, *not having had an okay way to grieve (R)* was found to be statistically significantly associated with long-term unresolved grief in cancer-bereaved youths, both male (OR: 5.9, 95% CI: 3.4–10.3) and female (OR: 2.9, 95% CI: 1.8–5.0). These associations remained significant for both



**Table 2** Prevalence of acute grief experiences and reactions (in the first 6 months post-loss) ( $N = 622$ )

| For the first half-year after your loss, would you agree with the statement (see phrasing in italics below):  | Do not agree<br>N (%) | Slightly agree<br>N (%) | Moderately agree<br>N (%) | Completely agree<br>N (%) | Missing<br>N <sup>1</sup> |
|---|-----------------------|-------------------------|---------------------------|---------------------------|---------------------------|
| <b>DID NOT HAVE AN OKAY WAY TO GRIEVE<sup>2</sup></b><br><i>"I did not have a way to grieve that felt okay."</i>                                    | 107/614 (17)          | 158/614 (26)            | 227/614 (37)              | 122/614 (20)              | 8                         |
| <b>NUMBING AND POSTPONING</b><br><i>"I clenched my teeth, built a wall around me and lived on as if nothing had happened."</i>                      | 117/616 (19)          | 239/616 (39)            | 148/616 (24)              | 113/616 (18)              | 6                         |
| <b>CONCEALED GRIEF</b><br><i>"I withheld my grief to protect my other parent."</i>  | 266/615 (43)          | 199/615 (32)            | 97/615 (16)               | 53/615 (9)                | 7                         |
| <b>OVERWHELMED BY GRIEF</b><br><i>"The grief was so strong it felt as if I would not survive, as if I was going crazy or was not normal."</i>       | 280/616 (45)          | 186/616 (30)            | 89/616 (14)               | 61/616 (10)               | 6                         |
| <b>DISCOURAGED FROM GRIEVING</b><br><i>"People stopped me from grieving by drawing away when I was sad or praising me when I was being strong."</i> | 349/613 (57)          | 171/613 (28)            | 60/613 (10)               | 33/613 (5)                | 9                         |
| <b>PRESSURED TO GRIEVE</b><br><i>"There was pressure from others that I should be more sad than I was showing."</i>                                 | 328/616 (53)          | 172/616 (28)            | 78/616 (13)               | 38/616 (6)                | 6                         |

<sup>1</sup> Individuals with missing data are excluded from the prevalence calculations

<sup>2</sup> To facilitate comparisons and avoid double negations, we here present the variable "I had a way to grieve that felt okay" as its negative counterpart, "I did not have a way to grieve that felt okay"

genders throughout the adjustments. However, only among the male participants were *numbing and postponing* (OR: 1.73, 95% CI: 1.22–2.47), *overwhelmed by grief* (OR: 2.02, 95% CI: 1.35–3.04) and *concealed grief* (OR: 2.19, 95% CI: 1.13–4.31) statistically significantly associated with long-term unresolved grief. These associations either remained or were strengthened after adjustments (Table 3). For female participants, the association with *long-term unresolved grief* was found for those reporting having been *discouraged from grieving* (OR: 2.76, 95% CI: 1.49–5.32). The association remained statistically significant and more or less unchanged after adjustments for background, and family and health care-related variables.

#### Possible predictive factors for having had an okay way to grieve in the first 6 months post-loss

Nine out of 28 background, family, and health care-related variables were found to be statistically significantly associated with *having had an okay way to grieve*, based on the results of a univariate logistic regression (see [Supplementary Table](#)). These nine variables were then used in a multivariable logistic regression model (Table 4), where three of them were found to be statistically significantly associated with *having had an okay way to grieve*. Male participants were more likely to *have had an okay way to grieve* (OR: 1.77, 95% CI: 1.23–2.54). Those who stated that there was a *good family cohesion* during the first 6 months after the loss (OR: 2.17, 95% CI: 1.27–3.84), and those who reported that they *had talked with the dying parent about what was important* were more likely to *have had an okay way to*

*grieve* than those who did not and wished they had (OR: 2.00, 95% CI: 1.35–2.97).

#### Discussion

This exploratory nationwide population-based study of 622 parentally bereaved former teenagers shows that more than half of the participants *had not found an okay way to grieve* in the first 6 months after the loss. *Not having had an okay way to grieve*, and four out of the five other acute grief experiences and reactions, including *numbing and postponing*, *concealing the grief* to protect the surviving parent, and being *overwhelmed by grief*, were associated with long-term unresolved grief. Differences were found between male and female participants in their reported grief experiences and reactions during the acute bereavement phase. Male participants, those who had talked with the dying parent about what they perceived as important, and those who had good family cohesion after the loss were more likely to *have had an okay way to grieve* in the immediate post-loss phase.

To the best of our knowledge, this is the first documentation of the prevalence or even existence of a number of different acute grief experiences and reactions post-loss in bereaved teenagers and the association of these experiences with long-term grief resolution. While some of them (e.g. *numbing and postponing*, *concealed grief*, or being *overwhelmed by grief*) are known reactions to loss and have been mentioned, in some form, in other studies [26, 27, 29, 30, 54–56], this is, as far as we know, the first study where teenagers were asked if they had found a way to grieve that felt okay to them. These

**Table 3** Acute grief experiences and reactions, and the association with long-term unresolved grief

|  | RATIOS  | ODDS RATIOS (ORs)        | ODDS RATIOS ADJUSTED FOR BACKGROUND VARIABLES <sup>1</sup> | ODDS RATIOS ADJUSTED FOR BACKGROUND AND FAMILY-RELATED VARIABLES <sup>2</sup> | ODDS RATIOS ADJUSTED FOR BACKGROUND, FAMILY AND HEALTH CARE-RELATED VARIABLES <sup>3</sup> |
|--|---|--------------------------|--|---|--|
|  | N unresolved grief <sup>4</sup> /<br>N grieving style (%) | OR (95% CI)              | OR (95% CI)  | OR (95% CI)   | OR (95% CI)  |
| <b>DID NOT HAVE AN OKAY WAY TO GRIEVE</b>            |   |                          |  |   |  |
| Entire group Agree                                   | 187/290 (64)  | <b>4.32 (2.99–6.28)</b>  | <b>4.23 (2.91–6.22)</b>                                    | <b>4.19 (2.88–6.16)</b>   | <b>4.14 (2.77–6.23)</b>  |
| Entire group Disagree                                | 69/233 (30)   | 1.0 (ref)                | 1.0 (ref)  | 1.0 (ref)   | 1.0 (ref)  |
| Male participants Agree                              | 75/118 (64)   | <b>5.85 (3.41–10.25)</b> | <b>6.23 (3.59–11.07)</b>                                   | <b>6.64 (3.79–11.98)</b>  | <b>6.72 (3.65–12.84)</b>   |
| Male participants Disagree                           | 31/135 (23)   | 1.0 (ref)                | 1.0 (ref)  | 1.0 (ref)   | 1.0 (ref)  |
| Female participants Agree                            | 112/172 (65)  | <b>2.95 (1.77–4.96)</b>  | <b>2.94 (1.75–4.98)</b>                                    | <b>2.84 (1.68–4.85)</b>   | <b>2.73 (1.54–4.89)</b>  |
| Female participants Disagree                         | 38/98 (39)  | 1.0 (ref)                | 1.0 (ref)  | 1.0 (ref)   | 1.0 (ref)  |
| <b>NUMBING AND POSTPONING</b>                        |   |                          |  |   |  |
| Entire group Agree                                   | 120/210 (57)  | <b>1.73 (1.22–2.47)</b>  | <b>1.68 (1.17–2.40)</b>                                    | <b>1.66 (1.16–2.38)</b>   | <b>1.57 (1.07–2.30)</b>  |
| Entire group Disagree                                | 137/315 (43)  | 1.0 (ref)                | 1.0 (ref)  | 1.0 (ref)   | 1.0 (ref)  |
| Male participants Agree                              | 48/94 (51)  | <b>1.8 (1.08–3.03)</b>   | <b>1.83 (1.09–3.09)</b>                                    | <b>1.91 (1.13–3.25)</b>   | <b>2.08 (1.16–3.79)</b>  |
| Male participants Disagree                           | 59/161 (37)   | 1.0 (ref)                | 1.0 (ref)  | 1.0 (ref)   | 1.0 (ref)  |
| Female participants Agree                            | 72/116 (62)   | 1.59 (0.98–2.61)         | 1.51 (0.92–2.50)   | 1.58 (0.95–2.63)  | 1.43 (0.82–2.48)   |
| Female participants Disagree                         | 78/154 (51)   | 1.0 (ref)                | 1.0 (ref)  | 1.0 (ref)   | 1.0 (ref)  |
| <b>CONCEALED GRIEF (TO PROTECT MY LIVING PARENT)</b> |   |                          |  |   |  |
| Entire group Agree                                   | 80/133 (60)   | <b>1.83 (1.23–2.73)</b>  | <b>1.71 (1.14–2.60)</b>                                    | <b>1.64 (1.08–2.53)</b>   | <b>1.56 (1.00–2.45)</b>  |
| Entire group Disagree                                | 177/391 (45)  | 1.0 (ref)                | 1.0 (ref)  | 1.0 (ref)   | 1.0 (ref)  |
| Male participants Agree                              | 25/43 (58)  | <b>2.19 (1.13–4.31)</b>  | <b>2.27 (1.16–4.50)</b>                                    | <b>2.55 (1.27–5.19)</b>   | <b>3.41 (1.55–7.77)</b>  |
| Male participants Disagree                           | 82/211 (39)   | 1.0 (ref)                | 1.0 (ref)  | 1.0 (ref)   | 1.0 (ref)  |
| Female participants Agree                            | 55/90 (61)  | 1.41 (0.84–2.37)         | 1.45 (0.86–2.47)   | 1.20 (0.69–2.09)  | 1.04 (0.57–1.88)   |
| Female participants Disagree                         | 95/180 (53)   | 1.0 (ref)                | 1.0 (ref)  | 1.0 (ref)   | 1.0 (ref)  |
| <b>OVERWHELMED BY GRIEF</b>                          |   |                          |  |   |  |
| Entire group Agree                                   | 81/131 (62)   | <b>2.02 (1.35–3.04)</b>  | <b>1.85 (1.21–2.86)</b>                                    | <b>1.81 (1.18–2.80)</b>   | <b>1.88 (1.19–2.98)</b>  |
| Entire group Disagree                                | 175/393 (45)  | 1.0 (ref)                | 1.0 (ref)  | 1.0 (ref)   | 1.0 (ref)  |
| Male participants Agree                              | 17/28 (61)  | <b>2.35 (1.06–5.40)</b>  | <b>2.54 (1.13–5.94)</b>                                    | <b>2.73 (1.20–6.45)</b>   | <b>3.22 (1.29–8.34)</b>  |
| Male participants Disagree                           | 90/227 (40)   | 1.0 (ref)                | 1.0 (ref)  | 1.0 (ref)   | 1.0 (ref)  |
| Female participants Agree                            | 64/103 (62)   | 1.56 (0.95–2.59)         | 1.59 (0.96–2.67)   | 1.53 (0.91–2.59)  | 1.64 (0.95–2.88)   |
| Female participants Disagree                         | 85/166 (51)   | 1.0 (ref)                | 1.0 (ref)  | 1.0 (ref)   | 1.0 (ref)  |
| <b>DISCOURAGED FROM GRIEVING</b>                     |   |                          |  |   |  |
| Entire group Agree                                   | 54/78 (69)  | <b>2.68 (1.62–4.56)</b>  | <b>2.37 (1.41–4.08)</b>                                    | <b>2.31 (1.37–3.99)</b>   | <b>2.45 (1.42–4.32)</b>  |
| Entire group Disagree                                | 203/445 (46)  | 1.0 (ref)                | 1.0 (ref)  | 1.0 (ref)   | 1.0 (ref)  |
| Male participants Agree                              | 9/17 (53)   | 1.60 (0.59–4.39)         | 1.67 (0.61–4.64)   | 1.75 (0.64–4.92)  | 2.1472 (0.72–6.54)   |
| Male participants Disagree                           | 98/237 (41)   | 1.0 (ref)                | 1.0 (ref)  | 1.0 (ref)   | 1.0 (ref)  |
| Female participants Agree                            | 45/61 (74)  | <b>2.76 (1.49–5.32)</b>  | <b>2.79 (1.50–5.41)</b>                                    | <b>2.65 (1.41–5.19)</b>   | <b>2.74 (1.41–5.53)</b>  |
| Female participants Disagree                         | 105/208 (50)  | 1.0 (ref)                | 1.0 (ref)  | 1.0 (ref)   | 1.0 (ref)  |
| <b>PRESSURED TO GRIEVE</b>                           |   |                          |  |   |  |
| Entire group Agree                                   | 46/91 (51)  | 1.08 (0.69–1.69)         | 1.07 (0.68–1.71)   | 1.04 (0.65–1.66)  | 1.10 (0.67–1.80)   |
| Entire group Disagree                                | 211/433 (49)  | 1.0 (ref)                | 1.0 (ref)  | 1.0 (ref)   | 1.0 (ref)  |
| Male participants Agree                              | 18/39 (46)  | 1.22 (0.61–2.43)         | 1.25 (0.62–2.51)   | 1.30 (0.64–2.62)  | 1.76 (0.79–3.92)   |

**Table 3** Acute grief experiences and reactions, and the association with long-term unresolved grief (Continued)

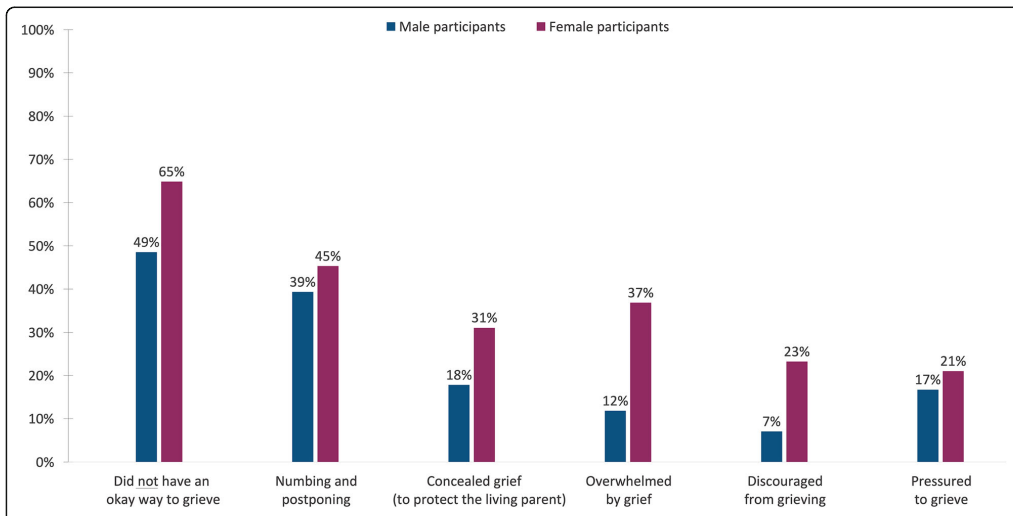
|                              | RATIOS  | ODDS RATIOS (ORs) | ODDS RATIOS ADJUSTED FOR BACKGROUND VARIABLES <sup>1</sup> | ODDS RATIOS ADJUSTED FOR BACKGROUND AND FAMILY-RELATED VARIABLES <sup>2</sup> | ODDS RATIOS ADJUSTED FOR BACKGROUND, FAMILY AND HEALTH CARE-RELATED VARIABLES <sup>3</sup> |
|------------------------------|---|-------------------|--|---|--|
|                              | N unresolved grief <sup>4</sup> /<br>N grieving style (%) | OR (95% CI)       | OR (95% CI)  | OR (95% CI)   | OR (95% CI)  |
| Male participants Disagree   | 89/216 (41)   | 1.0 (ref)         | 1.0 (ref)  | 1.0 (ref)   | 1.0 (ref)  |
| Female participants Agree    | 28/52 (54)  | 0.91 (0.50–1.68)  | 0.93 (0.50–1.73)   | 0.87 (0.47–1.64)  | 0.85 (0.44–1.68)   |
| Female participants Disagree | 122/217 (56)  | 1.0 (ref)         | 1.0 (ref)  | 1.0 (ref)   | 1.0 (ref)  |

Acute grief experiences and reactions: first 6 months post-loss. Long-term unresolved grief: 6–9 years post-loss. Agree: moderately agree and completely agree; Disagree: do not agree and slightly agree  
 Variables retained after the logistic regression in the forward selection procedure, using the variables as predictors of unresolved grief, with selection being based on likelihood ratio *p*-values and the entry criterion of *P* < 0.25: <sup>1</sup>Odds ratio adjusted for background variables: gender (in the entire group, not used in the gender-stratified data analysis), age at loss. <sup>2</sup>Odds ratio adjusted for family-related variables: worried about the surviving parent. <sup>3</sup>Odds ratio adjusted for health care-related variables: the teenager’s perception of the health care professionals’ efforts to cure the parent; the teenager’s perception of the health care professionals’ efforts to prolong the parent’s life; whether the family had been given end-of-life information about the disease, treatment and death by a physician; whether the teenager had talked with their dying parent about what was important; awareness time at which the teenager realized that the parent would die from the disease; awareness time at which the teenager realized that death was imminent (hours or days)  
<sup>4</sup>Missing values for unresolved grief (not included in the analyses): *n* = 63; demographic variables: *n* = 89; family-related variables: *n* = 13; health care-related variables: *n* = 115. Missing values are due to participants’ response of “I don’t know or remember” to selected variables. *CI* Confidence interval. *OR* Odds ratio

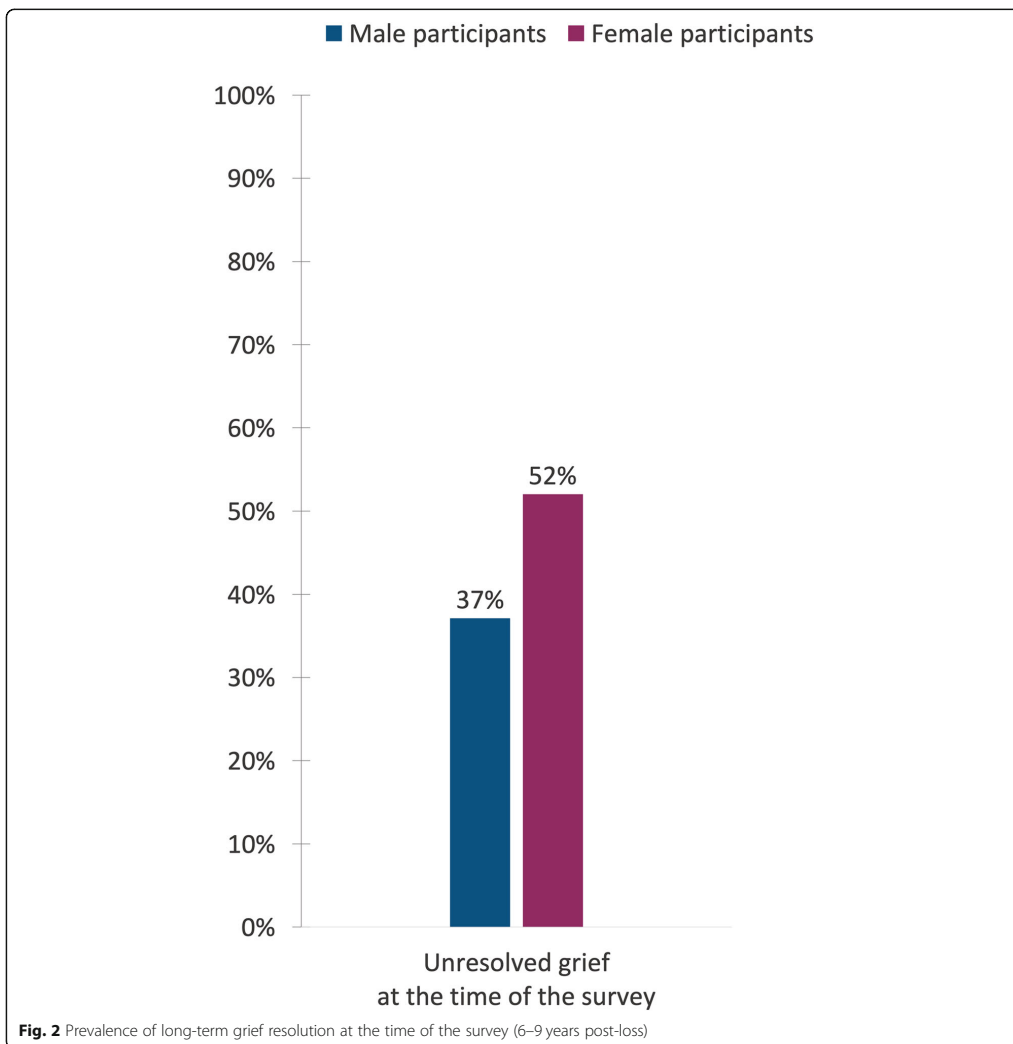
thoughts were expressed by the bereaved teenagers themselves in the preparatory interviews and therefore included in the study-specific questionnaire.

Why so many of the parentally bereaved teenagers in our study seem to have been struggling with finding a way to grieve that felt okay to them during the acute bereavement phase is unclear to us. One possible explanation could be linked to the quality of their relationship with the surviving parent, where warmth and connection as well as positive parenting skills have been shown to

benefit the children [2, 16, 25, 57–60]. Also, how the surviving parent is coping with their own grief has been shown to have an impact on their children’s grief reactions and ability to cope with the loss [59, 61–64]. Cancer, as the cause of death, has been found to significantly impact the risk of complicated grief among the bereaved [65], and may also be an explanation. Another possible explanation to consider, might be related to lack of experience and knowledge about common grief reactions in young people. More knowledge about what to expect



**Fig. 1** Prevalence of the six acute grief experiences and reactions in the first 6 months post-loss



after the death of a parent has been requested by bereaved teenagers and their surviving parents [39–41] and identified as helpful in their grieving process. It is also possible that more support is required according to individual needs after the death of a parent [66].

*Numbing and postponing* the grief was prevalent among the participants of our study. Although coping strategies that involve avoiding or suppressing emotions have been linked to psychological problems in bereaved children and teenagers and an open expression of grief is encouraged [67], it has also been argued that numbing

and postponing grief can be an important part of their way to handle the grief [67]. Teenagers' developmental stage can make them especially vulnerable to emotional stressors [68] and they are often only capable of dealing with the emotional pain for a short period of time [33–35]. The Dual Process Model describes an oscillation which is viewed as a normal part of the grieving process, allowing the person to move in and out of intense grief, and thus enabling them to deal with the loss in small doses at a time [31, 32]. We do not know whether the reported *numbing and postponing*

**Table 4** Associations between possible predictive variables and having had an okay way to grieve in the first 6 months post-loss

|  | N who had had an okay way to grieve/<br>N of individuals in the category (%) | OR (95% CI) of having had an okay way to grieve <sup>1</sup> | P-value <sup>1</sup> |
|--|--|--|----------------------|
| <b>Gender of participants</b>  |  |  | <b>0.0020</b>        |
| Male   | 156/303 (51)   | <b>1.77 (1.23–2.54)</b>                                      |                      |
| Female   | 109/310 (35)   | 1.0 (ref)  |                      |
| <b>Family cohesion during the teenage years, until the loss</b>  |  |  | 0.1459               |
| Good (moderate, or very much cohesion)   | 254/563 (45)   | 1.86 (0.81–4.68)   |                      |
| Poor (no, or a little cohesion)  | 9/48 (19)  | 1.0 (ref)  |                      |
| <b>Family cohesion during the first 6 months after the loss</b>  |  |  | <b>0.0046</b>        |
| Good (moderate, or very much cohesion)   | 239/502 (48)   | <b>2.17 (1.27–3.84)</b>                                      |                      |
| Poor (no, or a little cohesion)  | 23/109 (21)  | 1.0 (ref)  |                      |
| <b>Worried about the surviving parent the first 6 months after the loss</b>                                  |  |  | 0.0817               |
| No (no, or a little worry)   | 104/206 (50)   | 1.40 (0.96–2.05)   |                      |
| Yes (moderate, or very much worry)   | 161/407 (40)   | 1.0 (ref)  |                      |
| <b>The teenager's level of trust in the care provided to the dying parent in the final week of life</b>      |  |  | 0.5463               |
| Trust (moderate, or very much trust)   | 218/485 (45)   | 1.19 (0.68–2.08)   |                      |
| Distrust (no, or a little trust)   | 34/103 (33)  | 1.0 (ref)  |                      |
| <b>The teenager's perception of the health care professionals' efforts to cure their parent</b>              |  |  | 0.3045               |
| Good efforts (moderate, or very much)  | 212/451 (47)   | 1.37 (0.75–2.54)   |                      |
| Poor efforts (no, or a little)   | 52/160 (32)  | 1.0 (ref)  |                      |
| <b>The teenager's perception of the health care professionals' efforts to prolong the parent's life</b>      |  |  | 0.6002               |
| Good efforts (moderate, or very much)  | 211/459 (46)   | 0.84 (0.43–1.62)   |                      |
| Poor efforts (no, or a little)   | 53/152 (35)  | 1.0 (ref)  |                      |
| <b>The teenager's perception of the health care professionals' efforts to prevent the parent's suffering</b> |  |  | 0.1760               |
| Good efforts (moderate, or very much)  | 240/524 (46)   | 1.54 (0.82–2.94)   |                      |
| Poor efforts (no, or a little)   | 24/86 (28)   | 1.0 (ref)  |                      |
| <b>The teenager had talked with their dying parent about what was important</b>                              |  |  | <b>0.0015</b>        |
| Yes  | 118/225 (52)   | <b>2.00 (1.35–2.97)</b>                                      |                      |
| No, but I didn't feel a need to  | 52/100 (52)  | <b>1.79 (1.08–2.97)</b>                                      |                      |
| No, and I wish I had   | 92/280 (33)  | 1.0 (ref)  |                      |

<sup>1</sup>Multivariable model of background, family and health care-related variables that were statistically significantly associated ( $p < 0.05$ ) in the bivariable analysis with having had an okay way to grieve

Missing values: 53 individuals were excluded because of missing values for any of the variables included in the model

CI Confidence interval; OR Odds ratio

among our participants was part of oscillating coping as described by the Dual Process Model, enabling them to handle their emotional pain from the grief, or whether they were putting their grief reactions on hold for a longer period of time.

Out of the six different acute grief reactions and experiences in our study, *not having had an okay way to*

*grieve* was the factor that had the strongest association with long-term unresolved grief. A study of bereaved adults [29] found that having negative interpretations of one's own grief reactions had a strong association with bereavement distress and symptoms of traumatic grief, even when those grief reactions are generally considered to be part of a normal grieving process [29]. This

highlights the importance of encouraging or supporting bereaved teenagers to find a way of coming to terms with their own grief reactions.

Further analysis, based on the gender of the participants, showed that the female participants had a higher prevalence of all the different acute grief experiences and reactions compared to the male participants, and the female participants were more likely to report unresolved grief 6–9 years after the loss. In addition, we also found different acute grief reactions to be associated with unresolved grief in the cancer-bereaved male and female participants. However, *not having had an okay way to grieve* was found to be statistically significantly associated with unresolved grief in both genders.

There could be many reasons behind the identified gender differences. Although the literature on teenagers' grief reactions is still limited, previous research has shown differences in grief reactions between the genders, where girls have been shown to report more persistent grief responses than boys and to be more likely to have prolonged grief disorder than boys [69]. It has also been reported that both normative and problematic grief responses decline more slowly in girls than in boys [69]. Regardless of why these differences in experience between the genders occur, we could assume, based on our findings, that there might be a need to approach teenage boys and girls differently during the acute bereavement phase.

Apart from the association between being male and *having had an okay way to grieve* during the acute bereavement phase, we also found an association between good family cohesion after the loss and *having had an okay way to grieve* during the acute bereavement phase. This is in line with previous research where family function, including family cohesion, was shown to impact children's way of coping with loss [2, 4] and where bereavement support, with focus on improving the family function after the loss of a parent, was shown to be beneficial for children and teenagers [69–71].

We also found that those who had talked with the dying parent about what they perceived as important were more likely to *have had an okay way to grieve* in the acute post-loss phase. The vast majority of cancer-bereaved teenagers want to be told about the ill parent's impending death [72] and being prepared for the loss of a parent has been shown to be of importance for children's adjustments after the loss [41]. In families where children are able to openly communicate about their parent's death, the children tend to adapt better in bereavement [73]. Children and teenagers have highlighted the importance of having the opportunity to say goodbye [39, 66, 74, 75] and those who were unable to have their final talk with their dying parent have reported resentment and sadness during their grief [76, 77]. However,

for them to be able to have this opportunity, it is important for the health care personnel to communicate to the family, including the teenage offspring, when the death of a parent is near.

### Strengths and limitations

This population-based survey was conducted with a large sample, using study-specific questions based on preparatory interviews with, and tested for face validity in, the target group. It also included measurement of a number of potential confounders. This, together with a high response rate (73%) and the data collection method (with self-reported data collected directly from the former teenagers themselves, thus providing direct insight into the grief experiences and reactions of our target group) are the major strengths of our study.

Among the limitations, which should be considered when interpreting our results, are that we have no knowledge about the possible impact that unknown confounders or the responses from non-participants could have had on the results. That is, we do not know if those who declined participation in the study had more or less difficulties with grief than those who participated (i.e. potential selection bias). Not using standardized grief-measurements can be seen as a limitation. However, our intention was to study the participants' subjective grief experiences and we believe using global single-item questions, directly asking about the real-life phenomena under investigation, can also be considered a strength. The questions were well understood by all of the bereaved participants in the face-validity interviews in this and other studies [50–52] and in this case it allowed us to collect a comprehensive data on teenagers' own subjective experience when losing a parent to cancer.

Because of our study design, i.e. cross-sectional, we cannot rule out the possibility of recall-induced bias and that current grief resolution may have partly influenced some participants' self-assessment of past events and circumstances. However, for ethical and practical reasons, collecting this data prospectively in a cohort study design was not possible. Instead, we had to mimic a longitudinal study design by anchoring the questions in childhood, teenage years, pre and post loss and today (at the time of the survey). In addition, we cannot know for what length of time our participants experienced the reported grief reactions and experiences, i.e. whether their answers reflected the whole first 6 months post-loss or whether the reactions occurred for a shorter part of that time. It is also noteworthy that in our exploratory study we found that 13% of the participants disagreed with all six of the statements regarding grief experiences, indicating a need to further explore other possible grief experiences and reactions that were not captured here.

Also, the eligibility criteria limit the generalizability of our findings to other groups such as teenagers from single-parent households, newly arrived immigrants or children outside the age range of this study.

### Implications

To be able to adjust to life after loss, both teenagers and their parents may benefit from knowledge of what to expect and the variety of grief reactions [39, 40]. The findings from our study indicate that it is important not to impose specific expectations on how the teenager's grief should or should not be expressed or dealt with. Rather, we should try to gain a deeper understanding of how the young person experiences their own reactions and if they are okay with that or not. The findings that more than half of the participants did *not have a way to grieve that felt okay to them* during the acute bereavement phase and that many of them felt the need to suppress or conceal their grief to protect others, highlight the importance of attending to the needs of bereaved teenagers and encouraging them to find a way to grieve that feels okay to them. Further research probing deeper into what constitutes an okay way to grieve and what does not would be useful.

Regarding clinical implications, pre-loss communication between health care professionals and the family might facilitate the possibility to say goodbye. Health care professionals should be aware of the impact of good family cohesion and communication [37, 40, 41, 78] and facilitate it when a parent is seriously ill or dying, as this may potentially prevent long-term unresolved grief in bereaved youth. This could e.g. mean providing information about various grief reactions, screening for families and teenagers at risk for complicated grief and provide support as needed, such as through the Family bereavement program [69] or its Swedish adaptation; The Grief and Communication Family Support Intervention [79]. Apart from the role health care professionals can have in bereavement support it is also important to take more of a public health approach [80, 81]. Public awareness about the impact of social support, not only from the family but also from e.g. school professionals and peers [82, 83] may improve the wellbeing of bereaved youth.

### Conclusion

More than half of the parentally bereaved participants had not found a way to grieve that felt okay to them during the acute bereavement phase. This, as well as several of the acute grief experiences and reactions measured, was associated with unresolved long-term grief. Having had an okay way to grieve in the immediate post-loss phase was predicted by male gender, good family cohesion and having had a last conversation with the dying parent. Pre- and post-loss communication

between health care professionals and the family, including the teenage children, about the imminent death, and about common acute grief experiences and reactions, normalizing the sometimes abysmal emotions that may be experienced, could facilitate coping with grief in the acute phase of bereavement, thus possibly reducing the risk of unresolved long-term grief.

### Supplementary Information

The online version contains supplementary material available at <https://doi.org/10.1186/s12904-021-00758-7>

**Additional file 1: Supplementary Table.** Associations between background, and family and health care-related variables and having had an okay way to grieve in the first 6 months post-loss.

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### Authors' contributions

All authors meet the requirements for authorship. Contributors to the conception or design of the study or the acquisition of data: TBG, UK, GS, TN, C.J.F. Contributors to the design, analysis and interpretation of data for the work: DB, TBG, KB, TN, VS, GS, UK, JK. Contributors to drafting of the article: TBG, DB, UK, KB. Contributors to writing the main manuscript text and creating the figures and tables: DB, TBG, UK, KB. All authors contributed to revising the study critically for important intellectual content. All authors read and approved the final manuscript and agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. Tove Bylund Grenklo and Dröfn Birgisdóttir contributed equally to this paper and share co-first authorship.

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### Availability of data and materials

The datasets generated and/or analysed during the current study are not publicly available due to legal and ethical restrictions as described by the Swedish law and ethical boards regarding data of sensitive nature, but are available from the corresponding authors on reasonable request. This is in order to assure data confidentiality and to protect the privacy of the research participants.

### Declarations

#### Ethics approval and consent to participate

The research was conducted in accordance with the Helsinki Declaration and was reviewed by the Regional Ethics Review Board at the Karolinska Institute (2007/836–31), that gave approval for the research to be carried out. Each participant was informed both orally and in writing about the study objective, as well as their right to withdraw from the study at any time. Each participant gave informed consent for their own participation in accordance with the Swedish law.

#### Consent for publication

Not applicable.



**Competing interests**

The authors have no conflict of interest to report.

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**Supplementary Table. Associations between background, and family and health care-related variables and having had an okay way to grieve in the first 6 months post-loss**

|   | N of having had an okay way to grieve/N of individuals in the category (%) | OR (95% CI) of having had an okay way to grieve <sup>1</sup> | P-value <sup>1</sup> |
|---|--|--|----------------------|
| <b>BACKGROUND VARIABLES</b>                 |  |  |                      |
| <b>Gender of participants</b>               |  |  | <b>&lt;0.0001</b>    |
| Male  | 156/303 (51)   | 1.96 (1.42–2.71)   |                      |
| Female                                      | 109/310 (35)   | 1.0 (ref)  |                      |
| <b>Birth year of the participant</b>        |  |  | 0.8966               |
| 1984–1985                                   | 53/120 (44)  | 1.02 (0.65–1.60)   |                      |
| 1986–1987                                   | 119/283 (42)   | 0.93 (0.65–1.34)   |                      |
| 1988–1990                                   | 91/208 (44)  | 1.0 (ref)  |                      |
| <b>Year of loss</b>                         |  |  | 0.5566               |
| 2000  | 73/163 (45)  | 0.97 (0.63–1.50)   |                      |
| 2001  | 63/141 (45)  | 0.97 (0.62–1.52)   |                      |
| 2002  | 51/134 (38)  | 0.74(0.46–1.17)  |                      |
| 2003  | 75/165 (45)  | 1.0 (ref)  |                      |
| <b>Age at loss</b>                          |  |  | 0.3005               |
| 13  | 50/120 (42)  | 1.0 (ref)  |                      |
| 14  | 66/157 (42)  | 1.02 (0.63–1.65)   |                      |
| 15  | 82/166 (49)  | 1.37 (0.85–2.20)   |                      |
| 16  | 62/157 (39)  | 0.91 (0.56–1.48)   |                      |
| <b>Birth order</b>                          |  |  | 0.8996               |
| Oldest child                                | 64/143 (45)  | 1.0 (ref)  |                      |
| Middle child                                | 64/143 (45)  | 1 (0.63–1.59)  |                      |
| Youngest child                              | 125/300 (42)   | 0.88 (0.59–1.32)   |                      |
| Only child                                  | 12/27 (44)   | 0.99 (0.42–2.26)   |                      |
| <b>Gender of the dead parent</b>            |  |  | 0.2582               |
| Male  | 150/331 (45)   | 1.20 (0.87–1.66)   |                      |
| Female                                      | 115/282 (41)   | 1.0 (ref)  |                      |
| <b>Birth year of the dead parent</b>        |  |  | 0.6021               |
| 1960–1969                                   | 33/79 (42)   | 1.0 (ref)  |                      |
| 1955–1959                                   | 69/173 (40)  | 0.92 (0.54–1.59)   |                      |
| 1950–1954                                   | 74/158 (47)  | 1.2 (0.71–2.13)  |                      |
| 1949 or before                              | 78/174 (45)  | 1.13 (0.66–1.95)   |                      |
| <b>Birth year of the living parent</b>      |  |  | 0.4758               |
| 1960–1969                                   | 42/104 (40)  | 1.0 (ref)  |                      |
| 1955–1959                                   | 81/188 (43)  | 1.12 (0.69–1.82)   |                      |
| 1950–1954                                   | 81/195 (42)  | 1.05 (0.65–1.71)   |                      |
| 1949 or before                              | 52/104 (50)  | 1.48 (0.85–2.56)   |                      |
| <b>Education level of the living parent</b> |  |  | 0.8998               |
| Middle school                               | 47/113 (42)  | 1.0 (ref)  |                      |
| High school                                 | 108/247 (44)   | 1.09 (0.70–1.72)   |                      |
| College/university                          | 94/224 (42)  | 1.02 (0.64–1.61)   |                      |
| <b>Education level of the dead parent</b>   |  |  | 0.3735               |
| Middle school                               | 53/112 (47)  | 1.0 (ref)  |                      |
| High school                                 | 06/235 (45)  | 0.91 (0.58–1.44)   |                      |
| College/university                          | 94/234 (40)  | 0.75 (0.47–1.18)   |                      |

**Supplementary table continued**

|   | <b>N of having had an okay way to grieve/N of individuals in the category (%)</b> | <b>OR (95% CI) of having had an okay way to grieve<sup>1</sup></b> | <b>P-value<sup>1</sup></b> |
|---|---|--|----------------------------|
| <b>FAMILY-RELATED VARIABLES</b>   |   |  |                            |
| <b>Family cohesion during the teenage years, until the loss</b>   |   |  | <b>0.0002</b>              |
| Good (moderate, or very much cohesion)  | 254/563 (45)  | 3.56 (1.77–7.97)   |                            |
| Poor (no, or a little cohesion)   | 9/48 (19)   | 1.0 (ref)  |                            |
| <b>Family cohesion during the 6 months after the loss</b>   |   |  | <b>&lt;0.0001</b>          |
| Good (moderate, or very much cohesion)  | 239/502 (48)  | 3.40 (2.11–5.67)   |                            |
| Poor (no, or a little cohesion)   | 23/109 (21)   | 1.0 (ref)  |                            |
| <b>Worried about the surviving parent, first 6 months after the loss</b>                                |   |  | <b>0.01</b>                |
| No (no, or a little worry)  | 104/206 (50)  | 1.56 (1.11–2.19)   |                            |
| Yes (moderate, or very much worry)  | 161/407 (40)  | 1.0 (ref)  |                            |
| <b>HEALTH CARE-RELATED VARIABLES</b>  |   |  |                            |
| <b>Cancer type</b>  |   |  | 0.9923                     |
| Central nervous system  | 26/66 (39)  | 1.0 (ref)  |                            |
| Breast, gynaecological  | 49/116 (42)   | 1.13 (0.61–2.10)   |                            |
| Gastro-intestinal   | 57/127 (45)   | 1.25 (0.69–2.31)   |                            |
| Urinary tract, prostate   | 13/29 (45)  | 1.25 (0.51–3.03)   |                            |
| Skin, sarcoma   | 17/43 (40)  | 1.01 (0.45–2.20)   |                            |
| Haematological  | 18/39 (46)  | 1.32 (0.59–2.94)   |                            |
| Lung, head and neck, thyroid  | 36/85 (42)  | 1.13 (0.59–2.19)   |                            |
| Unknown primary   | 4/12 (33)   | 0.77 (0.19–2.71)   |                            |
| Don't know, don't remember  | 26/58 (45)  | 1.25 (0.61–2.57)   |                            |
| <b>Disease recurrence</b>   |   |  | 0.5306                     |
| Several times   | 14/39 (36)  | 1.0 (ref)  |                            |
| Once  | 91/217 (42)   | 1.29 (0.64–2.68)   |                            |
| No  | 156/350 (45)  | 1.44 (0.73–2.92)   |                            |
| <b>The teenager's level of trust in the care provided to the dying parent in the final week of life</b> |   |  | <b>0.0246</b>              |
| Trust (moderate, or very much trust)  | 218/485 (45)  | 1.66 (1.07–2.62)   |                            |
| Distrust (no, or a little trust)  | 34/103 (33)   | 1.0 (ref)  |                            |
| <b>The teenager's perception of whether mistakes had been made in the parent's care</b>                 |   |  | 0.26                       |
| No  | 173/384 (45)  | 1.21 (0.87–1.70)   |                            |
| Yes   | 90/223 (40)   | 1.0 (ref)  |                            |
| <b>The teenager's perception of the health care professionals' efforts to cure the parent</b>           |   |  | <b>0.0013</b>              |
| Good efforts (moderate, or very much)   | 212/451 (47)  | 1.84 (1.27–2.71)   |                            |
| Poor efforts (no, or a little)  | 52/160 (32)   | 1.0 (ref)  |                            |
| <b>The teenager's perception of the health care professionals' efforts to prolong the parent's life</b> |   |  | <b>0.0159</b>              |
| Good efforts (moderate, or very much)   | 211/459 (46)  | 1.59 (1.09–2.34)   |                            |
| Poor efforts (no, or a little)  | 53/152 (35)   | 1.0 (ref)  |                            |
| <b>The teenager's perception of the health care professionals' efforts to prevent suffering</b>         |   |  | <b>0.0015</b>              |
| Good efforts (moderate, or very much)   | 240/524 (46)  | 2.18 (1.34–3.67)   |                            |
| Poor efforts (no, or a little)  | 24/86 (28)  | 1.0 (ref)  |                            |

**Supplementary table continued**

|  | <b>N of having had an okay way to grieve/N of individuals in the category (%)</b> | <b>OR (95% CI) of having had an okay way to grieve<sup>1</sup></b> | <b>P-value<sup>1</sup></b> |
|--|---|--|----------------------------|
| <b>Were the family given end-of-life information about the disease, treatment and death by a physician</b> |   |  | 0.1322                     |
| Yes, before, or yes, both before and after the loss  | 138/292 (47)  | 1.55 (0.91–2.67)   |                            |
| Yes, after the loss  | 13/25 (52)  | 1.88 (0.75–4.77)   |                            |
| Don't know, don't remember   | 87/223 (39)   | 1.12 (0.64–1.94)   |                            |
| No   | 26/71 (37)  | 1.0 (ref)  |                            |
| <b>The teenager had talked with the dying parent about what was important</b>                              |   |  | <0.0001                    |
| Yes  | 118/225 (52)  | 2.25 (1.57–3.24)   |                            |
| No, but I didn't feel a need to  | 52/100 (52)   | 2.21 (1.39–3.53)   |                            |
| No, and I wish I had   | 92/280 (33)   | 1.0 (ref)  |                            |
| <b>The teenager had the opportunity to say farewell to the deceased parent</b>                             |   |  | 0.0975                     |
| Yes, on several occasions  | 16/39 (41)  | 1.14 (0.48–2.68)   |                            |
| Yes, at the place of death   | 193/457 (42)  | 1.20 (0.66–2.21)   |                            |
| Yes, at another place  | 36/62 (58)  | 2.26 (1.06–4.90)   |                            |
| No   | 19/50 (38)  | 1.0 (ref)  |                            |
| <b>The teenager's perception of the seriousness of the situation 3 days before the loss</b>                |   |  | 0.3248                     |
| Already very serious and the end was near  | 152/356 (43)  | 0.59 (0.25–1.32)   |                            |
| Serious, incurable disease, but the end was not near   | 52/112 (46)   | 0.68 (0.28–1.63)   |                            |
| Serious, but treatment would probably cure the parent  | 42/111 (38)   | 0.48 (0.20–1.15)   |                            |
| Not so serious, treatment would cure the parent  | 14/25 (56)  | 1.0 (ref)  |                            |
| <b>The teenager's location at the time of loss</b>   |   |  | 0.757                      |
| With the dying parent  | 79/189 (42)   | 0.90 (0.62–1.3)  |                            |
| In a room or corridor next to the dying parent's room  | 37/92 (40)  | 0.84 (0.52–1.35)   |                            |
| On their way to the dying parent   | 20/41 (49)  | 1.19 (0.62–2.30)   |                            |
| In school or some other place  | 128/288 (44)  | 1.0 (ref)  |                            |
| <b>Awareness time at which the teenager realized that the parent's disease was incurable</b>               |   |  | 0.2208                     |
| At the time of death   | 42/102 (41)   | 0.92 (0.58–1.45)   |                            |
| Hours – days before the death  | 61/154 (40)   | 0.86 (0.58–1.28)   |                            |
| Weeks – months before the death  | 125/289 (43)  | 1.0 (ref)  |                            |
| 6 months or longer before the death  | 36/66 (55)  | 1.57 (0.92–2.71)   |                            |
| <b>Awareness time at which the teenager realized that the parent would die from the disease</b>            |   |  | 0.0917                     |
| At the time of death   | 51/123 (41)   | 0.84 (0.54–1.29)   |                            |
| Hours – days before the death  | 73/193 (38)   | 0.72 (0.49–1.05)   |                            |
| Weeks – months before the death  | 111/242 (46)  | 1.0 (ref)  |                            |
| 6 months or longer before the death  | 28/50 (56)  | 1.50 (0.82–2.80)   |                            |
| <b>Awareness time at which the teenager realized that death was imminent (hours or days)</b>               |   |  | 0.3953                     |
| At the time of death, or never   | 77/191 (40)   | 1.0 (ref)  |                            |
| Hours to 2 days before the death   | 126/293 (43)  | 1.13 (0.77–1.62)   |                            |
| 3 days or longer before the death  | 61/127 (48)   | 1.37 (0.87–2.15)   |                            |

<sup>1</sup>Associations between having had an okay way to grieve and various background, family or health care-related variables, assessed with bivariable logistic regression.

CI = confidence interval; OR = odds ratio.

Paper III





# **Family cohesion predicts long-term health and well-being after losing a parent to cancer as a teenager: A nationwide population-based study. *Manuscript submitted.***

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Paper IV





RESEARCH ARTICLE

Open Access



# A novel care guide for personalised palliative care – a national initiative for improved quality of care

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## Abstract

**Background:** Even when palliative care is an integrated part of the healthcare system, the quality is still substandard for many patients and often initiated too late. There is a lack of structured guidelines for identifying and caring for patients; in particular for those with early palliative care needs. A care guide can act as a compass for best practice and support the care of patients throughout their palliative trajectory. Such a guide should both meet the needs of health care professionals and patients and families, facilitating discussion around end-of-life decision-making and enabling them to plan for the remaining time in life. The aim of this article is to describe the development and pilot testing of a novel Swedish palliative care guide.

**Methods:** The Swedish Palliative Care Guide (S-PCG) was developed according to the Medical Research Council framework and based on national and international guidelines for good palliative care. An interdisciplinary national advisory committee of over 90 health care professionals together with patient, family and public representatives were engaged in the process. The feasibility was tested in three pilot studies in different care settings.

**Results:** After extensive multi-unit and interprofessional testing and evaluation, the S-PCG contains three parts that can be used independently to identify, assess, address, follow up, and document the individual symptoms and care-needs throughout the whole palliative care trajectory. The S-PCG can provide a comprehensive overview and shared understanding of the patients' needs and possibilities for ensuring optimal quality of life, the family included.

**Conclusions:** Based on broad professional cooperation, patients and family participation and clinical testing, the S-PCG provides unique interprofessional guidance for assessment and holistic care of patients with palliative care needs, promotes support to the family, and when properly used supports high-quality personalised palliative care throughout the palliative trajectory. Future steps for the S-PCG, entails scientific evaluation of the clinical impact and

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effect of S-PCG in different care settings – including implementation, patient and family outcomes, and experiences of patient, family and personnel.

**Keywords:** Quality improvement, Clinical practice guidelines, Decision support, Palliative care, Dying, Clinical pathway, Patient-centred care, Personalised care, Early identification, Early Palliative Care

## Background

For many patients with terminal illness, access to and quality of palliative care is substandard and random [1–3]. This leads to unnecessary suffering for patients and families left without adequate interventions and support. Evidence-based palliative care as well as patient involvement in decisions and the caring process are promoted by international [4–8] and Swedish national recommendations and the Swedish law [9, 10]. One of the major challenges for improved palliative care is the operationalization of such recommendations [11, 12]. It is well known that in spite of the general and legal aims for evidence-based care, it is a challenge to transform evidence-based guidelines, whether national, regional or local, into clinical practice [11, 13–16]. In order to improve the outcome of care for patients and families there is a need to change the behaviour among health care workers [17, 18]. The most common approach is education aimed at augmenting knowledge, attitudes and skills. Interventions that are most likely to attain behavioural change in health care often combines: restructuring of practice, altering of norms and attitudes (e.g. through education), together with external audits and feedback [19] and sensemaking [20].

Several tools have been developed to support the process of screening for palliative care needs and to guide the team to take necessary action [21–25]. The tools include overarching guidance for future care planning based on a number of prompts supporting a comprehensive assessment and care involving patient, family and team members. Clinical guidelines and pathways have also been designed to help health care professionals make relevant decisions and guide best-practice care [11, 19, 21, 26, 27]. One example from end-of-life care is the Liverpool Care Pathway (LCP) [28], which has been embraced as a useful guide for the care of the dying patient but also encountered strong critique [15, 29–32].

An unmet demand for early identification of palliative care needs is evident, but finding the patient with palliative care needs, and systematically assess and address such needs, is a challenge for professionals in most health care settings [33–38]. This calls for a systematic approach even for those working in specialized palliative care. The Swedish health care professionals working with palliative care at the end-of-life have called for a more supportive structured around care for patients earlier on in

the palliative trajectory. A more proactive approach to palliative care is also encouraged by the World Health Organization [39], and several initiatives, including new development of clinical guides to promote care of the dying, have already moved in this direction [5, 25, 40–43]. Early integration of palliative care competency and early identification of patient needs have been shown to be effective in reducing suffering, increasing quality of life, and even prolonging survival [44–47].

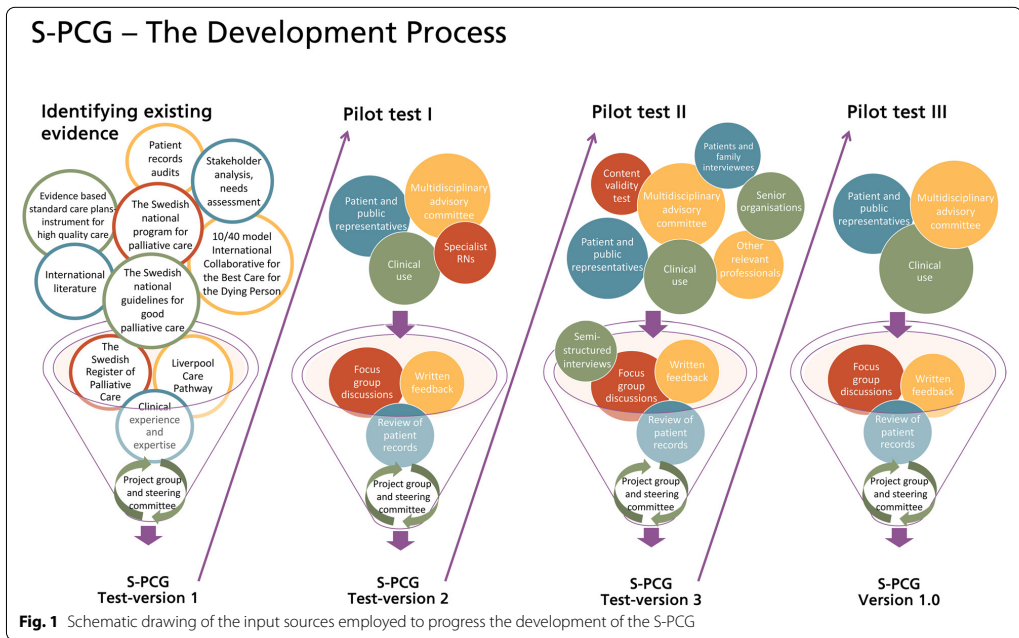
With the ambition to meet the challenges of transforming knowledge into clinical palliative care practice we have developed a guide, named “The Swedish Palliative Care Guide” (S-PCG), to inform best practice and to meet the palliative care needs of patients and families throughout the palliative trajectory. The guide aims to provide support for a timely initiation of evidence-based personalised palliative care and is designed to meet palliative care needs on an individual basis. The guide should support that the quality of care is adequate for every adult patient and family with palliative care needs regardless of diagnosis or place of care (at home, or in a residential care home, hospice, or hospitals) and cover the whole palliative care trajectory. Throughout the development of the guide, it has been in the forefront to support integration of the principles of good palliative care into clinical practice rather than just provide strict instructions for implementation.

Our purpose in developing the S-PCG was to provide support to any given interdisciplinary team at a health care facility, helping them to provide the best possible personalised palliative care. The S-PCG aim is to help identify patients, assess palliative care needs, give decision support and help choose relevant care interventions, in order to enhance the greatest possible well-being of patients with limited time left in life. The aim of this article is to describe the comprehensive development process and the resulting “product” of the S-PCG.

## Method and process of development

### Study design

The work of compiling and testing the guide was carried out in 2013–2016. We used the Medical Research Council (MRC) framework to provide a robust structure for the process [48]. This article describes the phases of developing as well as feasibility and piloting. The study followed the ethical guidelines stated in the Declaration



of Helsinki [49] and was performed in accordance to the Swedish laws and the local and national ethical review authority considerations concerning quality improvements and clinical audit within the health care.

**Developing**

**Reviewing the current standards for palliative care and defining the need for guidance**

A steering committee oversaw the project, provided strategy and performed stakeholder analysis. A project group modelled the new care guide and led the testing of it. A national interdisciplinary advisory committee was established to review the content of the S-PCG. In order to cover the full range of the palliative care team and represent the different fields of health care, it included 95 health care professionals, researchers and others relevant for patients with palliative care needs. (Supplementary table A).

The S-PCG was designed based on current national and international evidence as described in regulatory documents issued by health care authorities, specifically the 2013 National Guidelines for Good Palliative Care at the End of Life [9], the 2012–2014 National Program for Palliative Care [50], together with quality indicators in the Swedish Palliative Registry and other relevant

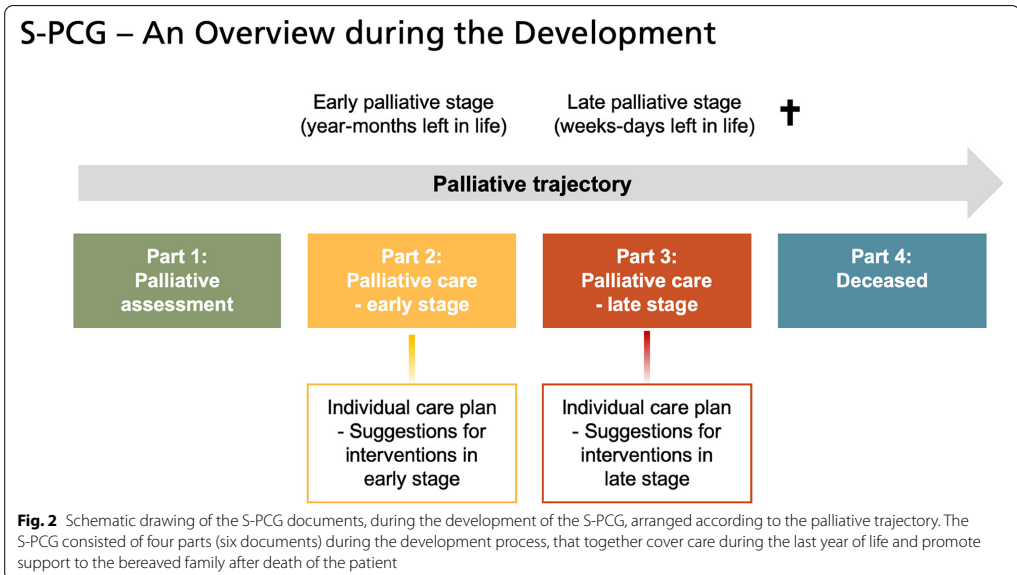
national indicators [51, 52]. The sections in the S-PCG on care for the dying person and care of the deceased person were inspired by the Liverpool Care Pathway (LCP) [28] and included the key principles and core elements from the 10/40 model set up by the International Collaborative for the Best Care of the Dying Person [53, 54]. The 10/40 model includes description of the ten principles together with the 40 core elements, used as quality indicators for good palliative and demonstrate good palliative care [54].

To evaluate the current standards of care and the needs for improvements, clinical field observations were performed at different units caring for patients with palliative care needs, as well as patient records audits. An overview of S-PCGs development process is shown in Fig. 1.

**Modelling a new palliative care guide**

The S-PCG was designed to include five different elements:

- Guidance on how to identify the patient that may have palliative care needs.
- A systematic approach for a comprehensive assessment of palliative care needs including recommendations for specific validated assessment tools to identify specific symptoms, problems and needs.



- c. Guidance to negotiate goals of care, care planning and care coordination.
- d. Symptom and needs-oriented care plans to give concrete, evidence- and experience-based suggestions for personalised evidence-based care activities.
- e. Guidance for taking care of the deceased and bereavement support.

The first test-version of the S-PCG was drafted by the project group in 2013, initially consisting of four parts covering the palliative trajectory (Fig. 2).

**Patient, family and public involvement**

Patient-, family- and public representatives were assigned to the project, to critically review the S-PCG and provide written as well as oral feedback to each version of the S-PCG, during recurrent meetings. Also, to co-create an S-PCG patient information brochure that was produced. To gain more insight into the priorities and wishes of patients and their family, semi-structured interviews were performed with a total of 11 patients and family members, used to shape the content of the S-PCG. Focus-groups interviews were also performed with representatives from two senior organisations. A total of 300

**Table 1** Number of patients participating, and S-PCG documents tested in pilot tests I – III

| Clinical Test       | Number of care units  | Number of Patients | Number of S-PCG documents tested |           |                  |            |                  | Total          |            |
|---------------------|-----------------------|--------------------|----------------------------------|-----------|------------------|------------|------------------|----------------|------------|
|                     |                       |                    | Part 1                           | Part 2    | Part 2 care plan | Part 3     | Part 3 care plan |                |            |
| Pilot test I        | 7                     | 28                 | 6                                | 11        | -                | 16         | 16               | 13             | 62         |
| Pilot test II       | 6                     | 22                 | 13                               | 1         | 1                | 6          | 9 <sup>b</sup>   | 0 <sup>c</sup> | 30         |
| Pilot test III      | 34                    | 250                | 16                               | 62        | 46               | 148        | 144              | 148            | 564        |
| <b>Total number</b> | <b>40<sup>a</sup></b> | <b>300</b>         | <b>35</b>                        | <b>74</b> | <b>47</b>        | <b>170</b> | <b>169</b>       | <b>161</b>     | <b>656</b> |

<sup>a</sup> Seven of the care units participated in more than one pilot test

<sup>b</sup> More patients were enrolled into the care plan for Part 3 than into the decision support of Part 3 itself

<sup>c</sup> The test units at the hospital had a well-functioning care plan for after the death in their digital hospital records, that prior to the start of the test had been revised to ensure that all of the content from S-PCG part 4 was included

patients were involved during the feasibility testing of the S-PCG.

### Feasibility and pilot testing

In agreement with the MRC framework and as recommended by the Ten Step Implementation Model from the International Collaborative for the Best Care of the Dying Person [54], pilot tests were performed to test the feasibility and usefulness of the newly designed S-PCG. The S-PCG was tested in a total of three pilot tests to ensure that it was feasible to use in broad clinical settings. Since the S-PCG is aimed towards patients with palliative care needs throughout the palliative trajectory, irrespective of diagnosis or the place of care, the only recruitment criteria for the care units were that they were based within the Swedish health care setting and serving adult patients that had or might have palliative care needs. The number of care units, patients and S-PCG documents used in the pilot tests are outlined in Table 1. The number of pilot tests were not pre-decided but constantly evaluated throughout the process. The need for further pilot-testing of the S-PCG was evaluated by the project group and the steering committee after each pilot test.

After each Pilot test period the experiences of staff were evaluated via focus groups interviews, written feedback and review of the documentation in the S-PCG used during each test-period. Feedback was also collected from other health care professionals, patients- and public representatives as well as the national interdisciplinary advisory committee who reviewed the content of the S-PCG parallel to the feasibility testing. An example of the items checked during the evaluation can be seen in Supplementary table B.

The collected feedback, from each pilot period, was then gathered in a large matrix. A thematic analysis inspired by Braun and Clarke [55, 56] was used and the feedback categorized based on: The relevance of the content; usability/user-friendliness; if anything was missing; or redundant; and other comments such as teamwork, implementation needs and patient involvement. The results of the feedback were then thoroughly discussed in the project group and the steering committee and used to improve the next test-version of the S-PCG (Fig. 1).

#### Pilot test I

For the first feasibility study, units from different health care services were recruited, including five nursing homes (one of which specialized in dementia care), a specialized palliative home care service, and a general home care service. The selection of care units for Pilot test I, was based on their own initiative, i.e. they contacted us

for quality improvement support and showed interest in testing the care plan that was under development.

Prior to initiation of the feasibility study, the personnel (n=166) underwent two days of training and the S-PCG documents together with written tutorial were handed out. The study ran for four consecutive months (May–August 2014).

During the study, 28 patients received care according to the S-PCG. As their diseases progressed, the majority of patients required care according to more than one part of the S-PCG, and thus in total over 60 documents from S-PCG Part 1-4 were used in *Pilot test I* (Table 1).

In addition to the patient and public representatives and the interdisciplinary advisory committee, nurses (n=11) from different specialist care units (i.e. surgery, nephrology, cardiology, haematology, pulmonology and home-based palliative care) also critically reviewed S-PCG Part 2 documents, including the care plan. This was due to the novelty of recommending interventions specifically for patients early in the palliative trajectory.

The collected feedback from *Pilot test I* was compiled, analysed, condensed and then categorised in relation to; the content itself, the usability, functionality and the relevance, and evaluated by the project group. The first test version of S-PCG was considered to be relevant and gave clear and structured support throughout the palliative care trajectory. Apart from comments about the layout and wording, the users requested some adjustments aimed for the care of the elderly. They also raised questions about what was needed for a successful implementation of the S-PCG in the team, such as information and knowledge. The most significant updates after *Pilot test I* included: adaption to better meet the needs of elderly patients with multiple chronic diseases, enhanced focus on the wishes and priorities of the patient, and adaptations to facilitate the working procedures of the team. The updated version of the S-PCG was denoted *test-version 2* (Fig. 1).

#### Pilot test II

Since *Pilot test I* only included care units from municipalities and specialized palliative care there was a need to include units from hospital care in *Pilot test II*. Therefore, the *S-PCG test-version 2* was subsequently tested at a nephrology department at a central hospital and five associated dialysis units in surrounding local hospitals. These units had all taken the initiative to contact us and volunteered to participate in the testing of the care plan. Prior to initiation of *Pilot test II*, a training session was arranged with the personnel (n=90), and an instruction manual was handed out. A designated contact person from each unit received additional training in order to be

able to provide on-site support. *Pilot test II* ran for three consecutive months (December 2014–February 2015), after which the experiences of the staff were evaluated as described above. During the second pilot test, 22 patients received care according to the S-PCG (Table 1), with a total of 30 S-PCG documents being used.

In addition to the review from the interdisciplinary advisory committee and the patient and public representatives, semi-structured interviews with patients and next of kin and focus group discussions with representatives from senior organisations were carried out. The patients, their next of kin and the senior public representatives generally considered the S-PCG to be a clear and professional support for the staff, highlighting important issues, and placing their needs in focus.

Additional comments were collected from other professions, that had been underrepresented during the evaluation but highly relevant to the development of the S-PCG. This included assistant nurses, dieticians, occupational- and physiotherapists, municipality care-managers, spiritual representatives, and delegates from the Swedish Registry of Palliative Care.

Furthermore, a content validity test was performed together with five care units that had not participated in *Pilot test I* or *II*: two specialized palliative care-unit, one oncology unit at a hospital, and two geriatric nursing homes. Each unit used the S-PCG for a minimum of 10 patients before giving feedback.

All the collected input from *Pilot test II* together with the content validity test, was compiled and categorized as described before, and used to further develop the S-PCG into *test-version 3* (Fig. 1). The feedback from the content validity test was very similar to the feedback from *Pilot test II*. The results showed that the S-PCG was, for the most parts, easy to understand and fill in — although some found it minorly confusing. Comments were made on a lack of clarity in the layout and in determining when to use the different parts of the material. The S-PCG was considered very comprehensive but at the same time everything was considered relevant. All units wished for the S-PCG to be made available in digital form, within their own patient records system. The most significant updates made to the S-PCG after *Pilot test II* were layout adjustments to give a clearer overall overview of patient needs; adaption to better facilitate cooperation between different users; and the addition of the S-PCG logotype. Further adjustments were also made to the user-manual, clarifying how to use the different parts of the S-PCG.

#### **Pilot test III**

The *S-PCG test-version 3* was tested between October and December 2015. To ensure variation and broad

testing of the S-PCG, *Pilot test III* included 34 care units in various settings within specialized palliative care, municipalities and hospitals.

As before, a training program was provided to the personnel (n=89), particularly to the new units (n=27 units), concerning the structure, content and usage of the S-PCG. Furthermore, selected representatives from the units got in-depth training and were given the task to support the implementation and evaluation processes on site. During *Pilot test III*, 250 patients received care according to the S-PCG with a total of 564 S-PCG documents being used (Table 1).

The evaluation of *Pilot test III* followed the previous described structure. The results highlighted the importance of education, of the managers' involvement and the need of cooperation and communication, between different professions and different healthcare providers. The content of the *S-PCG test-version 3* was considered useful for the care of an enlarged number of patients, increased the opportunity to discuss the patient's problems in real time and became a support for the staff's shared overall view of patient needs and facilitated the planning of the care. However, the content was also perceived as lengthy and, layout was in various need of simplification. Comments, also reflected an overall expressed preference for a digital format. Also, as the focus was more on care needs rather than prognosis, users experienced difficulty in differentiating between early and late phase, i.e. between S-PCG part 2 and 3 (see Fig. 2). Furthermore, unnecessary re-documentation of the same information was also experienced, if shortly after initiation of S-PCG part 2, the patient was identified as dying and needed care according to part 3.

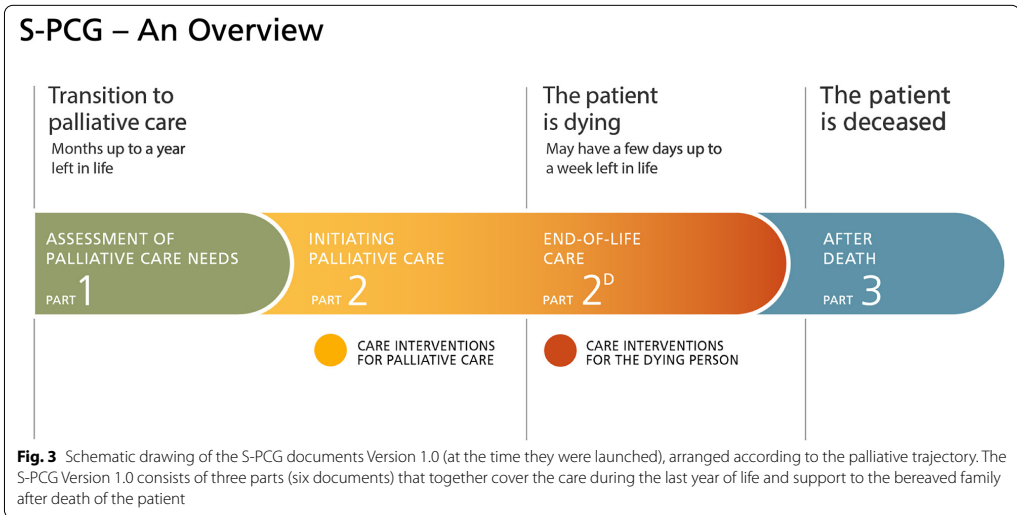
To address this, one of the most substantial changes after the evaluation during *Pilot test III* was merging parts 2 and 3. A circular table of contents was added at the front of each of the three parts to facilitate and clarify that the use of the S-PCG is always based on the patient needs, and specification of support for the children as next-of-kin was moved to an appendix. Due to the extensive number of different digital medical records systems in Sweden it was decided not to provide S-PCG as a digital medical record at this stage of development, but rather encourage thorough imbedding of the S-PCG into the existing medical records already in use.

Supplementary table C gives an example of the general feedback provided during *Pilot test III*, together with some of the main changes made to the S-PCG.

#### **Results**

This article outlines the development of a novel *Swedish palliative care guide* (S-PCG) intending to improve the end of life care for adult patients irrespective of diagnosis.





The extensive and expansive stepwise multi-unit and interprofessional testing and evaluation procedures resulted in *S-PCG Version 1.0*, which was launched in September 2016. It consists of three parts and includes, in total, six documents (Fig. 3). The S-PCG version 1.0 was reviewed and assessed by the International Collaborative for the Best Care of the Dying Person, which stated that the S-PCG was an excellent care plan, detailed and comprehensive. It was approved by the International Collaborative to be congruent with the principles and core elements for the best care for the dying person [54].

The S-PCG was designed to give support and structure to health care professionals when meeting adult patients with potential palliative care needs, irrespective of diagnosis. The S-PCG can be initiated at any stage of the palliative trajectory by choosing the relevant part that is best suited to meet the current needs of the patient. It provides a structure to identify the patients' status and needs through assessing symptoms, function, social situation as well as to highlight the importance of capturing the patient priorities and wishes. We promote the use of validated assessment instruments such as the Integrated Palliative Outcome Scale (IPOS) [57, 58], Edmonton Symptoms Assessment Scale (ESAS) [59] and the Abbey Pain Scale [60] within the S-PCG. By capturing the individual care-needs, the S-PCG can help create an overall picture and shared understanding of the needs and possibilities beneficial for each patient's quality of life and of his next of kin. Table 2 gives an overview of the main topics and sections that are included in the S-PCG.

Table 2. Overview of the key topics and sections in the S-PCG, with examples of issues/tools included in the care guide.

**The different parts of S-PCG**

S-PCG Part 1 is a two-page concise tool that provides simplified support for the identification of the patients' palliative care needs and initiate care planning. It can be used wherever patients with palliative care needs are encountered, for example in general practice, nursing homes and in- and outpatient hospital care. It can be used during consultations or as an assessment tool for multi-professional team rounds.

S-PCG Part 2 is an in-depth assessment of the same topics as in the Part 1, and is intended to support the provision of care for patients with palliative care needs regardless of time left in life. Part 2 consists of a guideline for initiating palliative care, assessment tools, and an associated care plan for recommended interventions for common symptoms and problems, which can be individually initiated according to the identified care needs of the patient. It focuses on defining common goals for care, and may support decisions and palliative care in the time range of months or up to a year left in life. Part 2 also has an appendix regarding children as next of kin.

S-PCG Part 2<sup>D</sup> can be initiated when a patient is assessed as likely dying. Part 2<sup>D</sup> adds on to Part 2, but focuses on the issues and symptoms that are frequent in the last few days of life. It includes guidance to recognise the dying phase in and hence initiate discussions on

**Table 2** Overview of S-PCGs key topics, sections and example of the issues included in the S-PCG

| Key topic                       | Section/item             | Example of issues or tools   |
|---------------------------------|--------------------------|--|
| <b>SYMPTOM AND STATUS</b>       | Symptoms and status      | Assessment of symptoms and status with validated tools such as IPOS <sup>a</sup> , Abbey Pain scale or ESAS            |
|                                 | Communication skills     | The patient's ability to communicate or need for assistance (e.g. interpreter)   |
|                                 | Function in daily life   | Assessment of level of function (ECOG <sup>c</sup> ) and activities of daily living (ADL)                              |
| <b>COMMUNICATION/ DECISIONS</b> | End-of-life conversation | Regarding prognosis and focus of care; Treatment interventions and life-sustaining treatments preferences              |
|                                 | Medical decisions        | Regarding current medical interventions, treatments and DNR <sup>d</sup> ; Prescription for anticipatory medication    |
|                                 | Information              | Practical information for the patient and/or the family (e.g. brochures, available benefits, support groups)           |
|                                 | Understanding            | Insight about current prognosis and focus of care  |
| <b>PREFERENCES</b>              | Wishes and priorities    | What is important right now; Spiritual and cultural needs; Involvement in care and treatment                           |
| <b>SOCIAL CONTEXT</b>           | Family <sup>e</sup>      | Family members distress/worries; Involvement in care; and Need for support   |
|                                 | Children                 | Minor children in the family and assessment of their need for information and support                                  |
| <b>PLANNING</b>                 | Coordination of care     | Contact information and need for referral (e.g. to specialized palliative care, dietician, religious/spiritual leader) |
|                                 | Care interventions       | Individual care interventions together with suggestions of possible interventions for each symptom/condition           |
|                                 | Reassessment and consent | Plan for new assessment of palliative care needs; Consent to share information with other care providers               |
| <b>LAST DAYS OF LIFE</b>        | Signs of dying           | Signs that the patient might be dying (e.g. the patient is bedridden; deteriorating level of consciousness)            |
|                                 | Recognition of dying     | Recognition by the physician that the patient may be entering the last days of life                                    |
|                                 | Special requests/needs   | Special requests and needs of the patient and/or family before and/or after the death (e.g. rituals, symbols)          |
|                                 | Care of the dying        | Continuous assessment of symptoms and status, and care interventions during the last days of life                      |
| <b>AFTER THE DEATH</b>          | Care of the deceased     | Practical, spiritual and cultural procedures and routines after death  |
|                                 | Bereavement support      | Information to the family (e.g. about practical issues, grief and support groups) and bereavement support              |

<sup>a</sup> IPOS = Integrated Palliative care Outcome Scale (58\*). <sup>b</sup>ESAS = The Edmonton Symptom Assessment System (59\*). <sup>c</sup>ECOG = The Eastern Cooperative Oncology Group performance status

<sup>d</sup> DNR = Do-not-resuscitate order. <sup>e</sup>The concept family is used here in its broadest sense and includes all persons of significance to the patient. \* Refers to the reference-number in the reference list

shifting the goals of care in the awareness of a most likely soon approaching death. Dying patients require frequent attention and symptom assessment, which is now thoroughly supported, including frequent reassessment in the care plan of Part 2<sup>D</sup>.

S-PCG Part 3 comprises a clear and condensed guide and thorough plan for care after death, in accordance with Swedish national care standards [50, 61, 62]. It supports relevant routines after a patient has died, including recommendations on how to care for the deceased person and promotes bereavement support for the family, including children in the family.

#### **Implementation and dissemination of the S-PCG**

The development of the S-PCG started as a local initiative based on national recommendations. It has been well received by regional and national palliative care

authorities and organisations and has been given support by the Swedish National Board of Health and Welfare. It is now included in the Swedish National Palliative Care Guidelines [63].

Lessons were learned from the Liverpool Care Pathway (LCP) [28] which was phased out in 2014 as a consequence of a critical governmental report entitled "More Care, Less Pathway" [64]. This statement and the possible risk that guidelines develop into checklists, supported our effort in operationalizing not only knowledge but also the palliative care approach into the novel care guide.

To facilitate a robust implementation, all parts of the S-PCG, information and support materials are openly available at the website of The Institute for Palliative Care [65]. The documents for clinical use are accessible after registration. The managers of the registered units

are responsible for the local application of the S-PCG including securing staff training and quality monitoring. Regular follow-up of results from the Swedish Registry of Palliative Care as well as audits of patient records are recommended. An audit tool has been designed to assist this procedure.

Brochures and instructional films of the S-PCG have been made available online [65]. An educational program has been developed for units aiming to implement the S-PCG and, to make it accessible to more users, an online S-PCG educational program is under development. Theme days/workshops for registered S-PCG units, aimed for education, inspiration and networking have also been arranged and the S-PCG has been presented at several conferences both in Sweden and internationally.

The S-PCG has been well received by the health care personal and at the beginning of the year 2021 a total of 305 care units were registered as S-PCG users in Sweden. Some regions have made the use of S-PCG compulsory within their district. The S-PCG has now been incorporated within several digital patient record systems in Sweden and research programs evaluating the clinical impact and effect of S-PCG in different care settings have been initiated.

## Discussion

We have now developed a care guide (S-PCG) that helps to identify adult patients with palliative care needs early and right through to end-of-life. It provides assessment tools and structured plans for documentation and guidance to support continued personalized palliative care. We have described the initial development of S-PCG, aimed to provide a link between evidence based best-practice care according to the core principles of palliative care, and professional behavior in everyday clinical practice.

Methodological challenges included the processing of the extensive information and feedback from the various care settings, health care professionals as well as patients and families. However, the collected expertise of the participants is unique and has contributed substantially to the development of S-PCG throughout the palliative care trajectory.

A majority of those who gave feedback on the S-PCG during the final pilot testing confirmed that the content was relevant to a broad group of patients and gave a good overall understanding of patients' needs. It was perceived as a good support to clinical practice, although it is worth mentioning that the majority of the participating care units contacted us expressing a need for a care guide and on their own initiative volunteered to participate in the testing of the S-PCG. This might predispose respondents to a more positive attitude towards the care guide,

thus affecting the result of how the guide was received. However, it can also be noted that in many of the testing units the decision was made by the managers and not all personnel that gave feedback were positive towards the S-PCG from the beginning.

Although the majority of the users were positive towards the use of S-PCG, it was at the same time seen as very comprehensive, time consuming and it was confusing to the users when to use the different parts of the S-PCG. It is essential to routine screening for palliative care needs within clinical practice [22] and for that to happen it is important not only to pilot test the instrument during the developmental stage but also to take into account the users' feedback into the final product. Based on the feedback, we made some significant changes to the design of the S-PCG, such as merging part 2 and 3 to make the documentation more efficient and user-friendly, without compromising the content. We also clarified the instructions on when to use the different parts of the S-PCG and emphasized a thorough planning of the use before implementation.

The S-PCG includes a brief guidance to screen for patients with potential palliative care needs. Apart from the "surprise question" regarding prognosis [66], the items covering disease stage, functional decline, disease progression and symptom burden are formulated to be fully transparent to the patient and family. The widely used surprise question gives a prognostic perspective, can be used as a reflective tool for team members, and together with other tools such as the PCST (palliative care screening tool) may help clinicians to identify patients with palliative care needs [67, 68]. As our intention was not to use a scoring system but rather to merely support the clinical assessment, the surprise question was not included in the main S-PCG documents as a criterion for potential palliative care needs. Instead it was highlighted in the user manual.

To promote transparency, we made it a priority for the content of the S-PCG to be understandable and non-offensive to patients and family members who may want to read these documents. We therefore included patient-, family- and public representatives in the discussion of the content of the S-PCG. In the planning and execution of the next MRC phases [48] (evaluating the implementation and the use of the S-PCG) we will intensify our partnered work with patients and families – strengthening user involvement from the level of consultation, to eventually, reach collaboration and equal partnership [69].

The potential limitation of not performing our own systematic review of the relevant scientific literature is, in fact, overshadowed by our access to ongoing updates in national recommendations and relevant evidence-based documents that were used [9, 13, 50, 70]. Further, the

large group of health care professionals and patient representatives ensured clinical experience and gave relevant guidance when other sources did not contribute the substantial knowledge than one could wish for.

The strengths of the S-PCG is that regardless of medical diagnosis and whether the patient is being treated in a hospital, at home, or in a nursing home or hospice, the S-PCG can provide structure and guidance for the care. It puts the patients' needs in focus and is designed to promote communication between different caregivers and encourage collaboration between health care professionals and the patient and their family. However more research is needed and the S-PCG will be updated continually based on new scientific evidence as well as clinical experience, the users input and patients and their families experience.

## Conclusions

After extensive development work and broad testing, the S-PCG has the potential to provide meaningful support in identifying palliative care needs; facilitates inter-professional assessment and care of these patients; and emphasizes the needs of the family throughout the palliative trajectory. It supports high-quality personalised palliative care, and when properly used may help patient and families express their too-often-neglected needs, support individual negotiation of goals of care, and subsequently promote relevant care. Choosing to implement S-PCG includes responsibility for its use in concordance with the principles of good palliative care. The next step entails scientific evaluation of the clinical impact and effect of S-PCG in different care settings – including implementation, patient and family outcomes, and experiences of patient, family and staff.

## Abbreviations

**S-PCG:** The Swedish Palliative Care Guide; **MRC:** The Medical Research Council; **LCP:** Liverpool Care Pathway; **IPOS:** Integrated Palliative Outcome Scale; **ESAS:** Edmonton Symptoms Assessment Scale.

## Supplementary Information

The online version contains supplementary material available at <https://doi.org/10.1186/s12904-021-00874-4>.

Additional file 1.

Additional file 2.

Additional file 3.

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## Authors' information

Not applicable.

## Authors' contributions

ADU coordinated the development and the testing of S-PCG in collaboration with BR and CJF. ADU, DB, BR, BS, and CJF were involved in the modelling phase. The preliminary documents were developed and refined by ADU, DB, BR, BS, and CJF. ADU was responsible for the training and supervision during pilot studies and together with DB, performed the focus groups interviews and analysed the feedback from the pilot units, patients and public representatives and the advisory committee. CJF, BR and BS independently analysed some of the focus groups discussions to check the reliability of ADUs and DBs analysis. DB and ADA compiled and systematized the data, and were responsible for writing the manuscript together with CJF. All authors commented critically on several drafts of the manuscript and have approved the final version.

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## Availability of data and materials

The datasets generated and/or analysed during the current study are not publicly available due to legal restriction as described by the Swedish law regarding data of sensitive nature and data protection, but can be available from the Institute for Palliative Care in Lund, Sweden on reasonable request.

## Declarations

### Ethics approval and consent to participate

This study describes the development of a structured clinical decision support tool, aiming to improve patient care outcomes and the delivery of health care for patients with palliative care needs. Therefore (in accordance to the Swedish laws) it did not require an official ethical approval by the Swedish National Ethical Review Authority. However, due to the sensitive nature of the topic the whole design and implementation was guided by the ethical guidelines stated in the Declaration of Helsinki and overseen in accordance to regional and institutional ethical recommendations. An informed consent was collected from the manager of each participating site prior to the clinical feasibility testing of the S-PCG.

### Consent for publication

Not applicable.

### Competing interests

The authors declare that they have no competing interests.

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**Supplementary table A.** Professions represented in the steering committee, project group and in the interdisciplinary advisory committee.

|   | <b>N</b>  | <b>Areas of responsibility/speciality</b>  |
|---|-----------|--|
| <b>Steering committee</b>                         | <b>5</b>  | Professor in palliative medicine (n=1), professor in palliative care nursing (n=1), associate professor in hematology (n=1), associate professor in palliative care nursing (n=1), project manager and quality improvement nurse (n=1) |
| <b>Project group</b>                              | <b>10</b> | Palliative care nursing (n=4), quality improvement nursing (n=2), palliative care research (n=2), palliative medicine (n=1), physiotherapy and public health (n=1)   |
| <b>Interdisciplinary advisory committee</b>       | <b>95</b> |  |
| <b>Professions:</b>                               |           |  |
| Registered nurses                                 | 41        | Palliative care (n=18), community nursing (n=11), oncology (n=4), cardiology (n=2), surgery (n=2), geriatric (n=1), lung (n=1), neurology (n=1), dementia care (n=1)   |
| Innovation improvement leads                      | 12        | Palliative care (n=6), oncology (n=2), surgery (n=1), geriatric (n=1), community care (n=1), hospital care (n=1)   |
| Physicians  | 11        | Palliative medicine (n=7), oncology (n=2), cardiology (n=1), GP (n=1)  |
| Assistant nurses                                  | 8         | Palliative care (n=3), community care (n=3), oncology (n=1), geriatric/neurology (n=1),  |
| Community matrons                                 | 5         | Community care (n=5)   |
| Unit managers                                     | 5         | Community care (n=3), palliative care (n=1), geriatric (n=1)   |
| Dieticians  | 3         | Palliative care (n=2), geriatric (n=1)   |
| Occupational therapists                           | 2         | Palliative care (n=1), quality improvements within occupational therapy (n=1)  |
| Social workers                                    | 2         | Palliative care (n=2)  |
| The Swedish Register of Palliative Care personnel | 2         | National quality register (n=2)  |
| Researchers                                       | 2         | Palliative care (n=2)  |
| Physiotherapist                                   | 2         | Palliative care (n=2)  |

**Supplementary table B: Example of items checked to evaluate the feasibility and usefulness of the S-PCG, during the pilot testing.**

| Sources and methods for collection of information  | Themes/items checked   | Clarification   |
|--|--|---|
| <p><b>CLINICAL TESTS (PILOT I-III)</b><br/>Focus groups interviews with clinical users<br/><br/>Written feedback from clinical users<br/><br/>Content validity test, through focus groups interviews and a questionnaire</p>   | <p><b>Relevance of the content</b><br/><br/><b>Usefulness / User-friendliness</b><br/><br/><b>Missing issues</b><br/><br/><b>Redundancy</b><br/><br/><b>Teamwork – Communication</b></p>   | <p>Is the content relevant and adequate for the care of people with palliative care needs and for your workplace?<br/>Is the content of the S-PCG easy/difficult to understand and use?<br/>Can you describe your experience of participating in the testing of the S-PCG? What has been good and what needs to change? Can you describe what it has been like to work with each part of the S-PCG? Advantages and disadvantages of the S-PCG?<br/><br/>Is there anything missing from the S-PCG documents? Anything that you would like us to add to the S-PCG?<br/><br/>Is the S-PCG too detailed? Is there anything that you think is unnecessary to include in the S-PCG and could be removed?<br/><br/>How has the cooperation within the team during the test of the S-PCG? Have you been able to involve all team members? What has worked well and what has not worked well? Has the S-PCG test influenced or initiated discussions about your current palliative care practices? If so, what discussions have you had?</p>   |
| <p><b>INTERDISCIPLINARY ADVISORY COMMITTEE</b><br/>Written feedback, on each part of the S- PCG, section by section<br/><br/>Whole day workshop<sup>1</sup> with the group</p>   | <p><b>Patient- and family<sup>2</sup> involvement</b><br/><br/><b>Implementation</b></p>   | <p>Do you think the S-PCG has added any value for patients and their family? Can you describe how it has been to involve patients and families in the S-PCG? In what way did you involve patients and families? If not, what prevented you from doing so? How do the patient, family and public representatives perceive the concepts and the vocabulary of the S-PCG? Was there anything in the S-PCG that felt inappropriate or offensive to the patient and family?<br/><br/>What training/information do you think is needed for the introduction/start of the S-PCG? Or the implementation of S-PCG? What do you think about the S-PCG user manual? Is it usable? Is it too comprehensive or is it missing some information? Do you think you could implement the S-PCG at your workplace with the information given?<br/><br/>What do you think of the S-PCG in general? Is it adequate? Is the content important? Is there anything that you think is important that is missing from the S-PCG? Is there anything that you think is unnecessary to include in the S-PCG and should be removed.<br/><br/>How do you perceive the concepts and the vocabulary of the S-PCG? Was there anything that was difficult to understand? Was there anything in the S-PCG that felt inappropriate or offensive? Is there anything that you think should be changed/rephrased/removed?</p> |
| <p><b>PATIENT-, FAMILY- AND PUBLIC REPRESENTATIVES</b><br/>Focus-groups meetings<br/><br/>Written feedback on each part of the S-PCG, section by section<br/><br/>Semi-structured interviews with patients that were admitted to the S-PCG during Pilot II and their family<sup>2</sup> member</p> | <p><b>Perception of the S-PCG based on the themes mentioned above</b><br/><br/><b>Perception of concepts and vocabulary used in the S-PCG documents</b><br/><br/><b>Perceptions about the different topics in the S-PCG, and if and what complementary written information is needed</b></p> | <p>Would you like to tell us what you think about us asking about:<br/>Symptoms? How daily life works for you?; if you have had an informative conversation with your doctor about your diagnosis and prognosis?; Your understanding of the situation with your disease and the prognosis?; Your wishes and preferences for the care?; Your social network and what you/your family needs help with?; If there are children in the family<sup>2</sup> and about their needs for support?; The possible need for other health care contacts (e.g. dietician, counsellor, district nurse?); Your consent for the relevant staff to access the information in this material?; Your wishes and preferences regarding your participation in decisions about care and treatment?<br/><br/>Do you think that this topic is relevant/ important to you? or do you think it should not be included in the S-PCG? Do any of the issues we've discussed today felt inappropriate or offensive? Is there anything else that you think is important for health care professionals to ask for or attend to that we have not discussed? Is there any information that you think is important to get in writing, e.g. in a brochure?</p>  |
| <p><b>REVIEW OF THE PATIENT RECORDS USED IN THE PILOT TESTS I - III</b></p>  | <p><b>How the S-PCG documents were used during the pilot tests</b></p>   | <p>We reviewed: The number of S-PCG documents included; number of patients allocated to each part of the S-PCG; if the question was answered/the assessment was made; any obvious misunderstandings in using S-PCG; what items had been added that possibly should be included in S-PCG; what care-interventions had been used and added; which questions had not been answered; which profession documented each question; if there had been a team collaboration.</p>   |

1) Workshop with the interdisciplinary advisory committee was only held once, during Pilot test II. 2) The concept family is used here in its broadest sense and includes all persons of significance to the patient.



**Supplementary table C:** Summary of the general feedback<sup>1</sup> from the evaluation of the S-PCG during Pilot test III, with an example of comments from the clinical test users; patient-, family<sup>2</sup>- and public representatives; the interdisciplinary advisory committee and the issues addressed/ raised after the evaluation.

| Items checked                         | Comments from the clinical test users  | Patient-, family <sup>2</sup> - and public representatives  | The interdisciplinary advisory committee   | Summary of issues addressed/raised after the evaluation   |
|---------------------------------------|--|---|--|---|
| <b>RELEVANCE OF THE CONTENT</b>       | <p><b>The content is relevant</b></p> <p>"S-PCG has a good content, all the important elements are included"</p> <p>"S-PCG is suitable for all our patients"</p> <p>"Relevant content and we almost didn't need to add anything"</p> <p><b>Gives a good overall picture of the patient needs</b></p> <p>"You get the "overall look of things" with S-PCG"</p> <p>"S-PCG provides a good overview, gives consensus and encourages holistic approach for the whole person"</p> <p><b>Is S-PCG suitable for all care forms?</b></p> <p>"Should there be different versions for different types of care?"</p> <p>"There is no time to have those long conversations in the hospitals"</p> <p>"Most of our patients (in the nursing home) do not have minor children, that section takes up a lot of space"</p> | <p><b>Important content</b></p> <p>"S-PCG is a good document that addresses important issues"</p> <p>"Good, it is important to ask the family for their need for support"</p> <p>"It is detailed but I think detailed questions are better than big overall ones"</p>   | <p><b>Relevant content</b></p> <p>"Good and clear content"</p> <p>"Yes, the content is relevant and I think it is good that you have the care activities listed as well, you can choose those that fit the patients' needs"</p> <p><b>Is the same document suitable for all care forms?</b></p> <p>"Would it maybe be better with different documents...for example, one for home care and one for hospital care"</p>              | <p><b>The content</b></p> <p>The content of the S-PCG did not change much, small adjustments were made in word phrasing. A decision was made to hold on to one version of the S-PCG, suitable for all care-settings. This, was done because the fundamental concept of the S-PCG is centered around the individual patient needs. This was highlighted in the creation of a user's manual, an educational program and in S-PCG brochures.</p> <p>Only the basic questions for children as family<sup>2</sup> were kept within the main documents and the more specific support was moved to an appendix, to be use when needed.</p> |
| <b>USEFULNESS – USER-FRIENDLINESS</b> | <p><b>S-PCG gather all information and is easy to use</b></p> <p>"It was an advantage that all information is gathered in one place"</p> <p>"Easy to use – since not everything has to be filled in for every patient"</p> <p>"It is good that S-PCG first find the problem and then guide us to take action to solve/alleviate it"</p> <p><b>Very comprehensive and time consuming</b></p> <p>"Time-consuming"</p> <p>"Very comprehensive"</p> <p>"Comprehensive material but good that you can choose what is relevant for each patient"</p>   | <p><b>Feels clear and professional</b></p> <p>"S-PCG feels professional"</p> <p>"S-PCG is clear/straightforward"</p> <p>"Nothing in the S-PCG feels inappropriate or offensive"</p> <p>"Didn't think concepts and wording were inappropriate or needed to be changed, it seemed professional"</p> <p>"Some words are maybe more medical terms but this is also aimed for the professionals and that feels safe"</p> | <p><b>Clear and usable</b></p> <p>"Yes, the documents are clear and seem to be usable"</p> <p>"S-PCG part 3 and part 4 are integrating, well in my Palliative Care Unit"</p> <p>"The overview paper in the Part 2 care measurements is very helpful"</p> <p><b>Too comprehensive and time consuming</b></p> <p>"A standing reflection is that the S-PCG documents are too comprehensive and take a very long time to complete"</p> | <p><b>Changes of the structure and layout</b></p> <p>A circular table of content was added to clarify that the use of the S-PCG does not require documentation in the order they are presented and that it is always based on the patient needs. To highlight this, checkboxes for "not applicable at this moment" were also added to several sections of the S-PCG documents.</p>  |

| Items checked  | Comments from the clinical test users  | Patient-, family <sup>2</sup> - and public representatives   | The interdisciplinary advisory committee  | Summary of issues addressed/raised after the evaluation  |
|--|--|--|---|--|
| <p><b>USEFULNESS – USER-FRIENDLINESS (continued)</b></p> | <p><b>S-PCG is a good support for the care of the patient</b><br/>           “S-PCG puts palliative care in focus and clarifies what you can do for the patient”<br/>           “S-PCG gives good support to new staff and new graduates so they will know what to do”<br/>           “Exceptional support to have during conversations with the patient and the family”<br/> <b>Difficult to decide when to use part 2 and 3 of the S-PCG</b><br/>           “The care plan for part 3 is good and shows clearly what we are doing for the patient, but it starts too late”<br/>           “If a patient is admitted to S-PCG Part 2 and then shortly after he is dying and needs to move onto part 3, then we need to fill in all the same information again, that is a lot of work”<br/>           “Same information in Part 2 and part 3, feels like a repetition”</p> |  | <p><b>Keep the support-questions visible</b><br/>           “I think that the support questions for the patient wishes and priorities should be visible in the S-PCG documents. These are good questions”</p> | <p><b>Changes of the structure and layout (continued)</b><br/>           The support-questions, for the patient wishes and priorities, were moved from the appendix into the main document.<br/>           Parts 2 and 3, of the S-PCG, were merged together, into Part 2 and 2D (D for the dying phase), to address the problem with repetition and double documentation. The titles of part 2 and former part 3 (now 2D) were changed to give a clearer description.<br/>           Also, the aim and a short description of when to use each specific part was added to the front page of each part of the S-PCG.</p>   |
| <p><b>MISSING ISSUES</b></p>                             | <p><b>Clearer instructions on how to use the S-PCG</b><br/>           “It should be made clearer that you can skip items that are not relevant for the patient you are caring for”<br/>           “Better instructions before the use, I am not sure how to use the different parts of the S-PCG”<br/> <b>“Clearer layout and more space for text”</b><br/>           “More space for the answer’s text/comments”<br/>           “Can you expand the comment field under wishes and priorities”<br/>           “Better structure for the care interventions in part 3”<br/> <b>S-PCG in digital form</b><br/>           “S-PCG in digital form so you can type in the computer and then print it out for the patient records”<br/>           “The S-PCG needs to be included in the digital patient records system”</p>  | <p><b>Ask about pets or something else that can be worrying for the patient</b><br/>           “They need to ask if there is something else I am worried about, such as who is taking care of my cat”<br/>           “Information about who is responsible for what”</p> | <p><b>More space for ADL and care interventions for edema</b><br/>           “More space for writing what ADL-aids the patient has”</p>   | <p><b>Layout and support for implementation</b><br/>           Clearer layout was created, with more space for text as suggested. The instructions on how to use the S-PCG were clarified in the user-manual.<br/> <b>Patients wishes</b><br/>           A question about pets was added to the “support questions” under the section about patient’s wishes and priorities.<br/> <b>S-PCG in digital form</b><br/>           Due to the extensive number of different digital medical records systems in Sweden it was decided not to provide S-PCG as a digital medical record at this stage of the development, but rather encourage thorough imbedding of the S-PCG into the existing medical records. Digital interactive PDF documents of the S-PCG were however created for download.</p> |

| Items checked                   | Comments from the clinical test users   | Patient-, family <sup>2</sup> - and public representatives   | The interdisciplinary advisory committee  | Summary of issues addressed/raised after the evaluation   |
|---------------------------------|---|--|---|---|
| <b>REDUNDANCY</b>               | <p><b>Nothing is redundant</b><br/>           “Nothing, S-PCG is comprehensive but everything is important!”<br/>           “Nothing is redundant”</p> <p><b>Double documentation</b><br/>           “It would be double documentation to have status updates and a medication list in the S-PCG since it is in our digital patient records already”<br/>           “Risk for double documentation”<br/>           “Part 4 is good but we already have a good checklist for after the death in the computer. No need for both”</p>  | <p><b>Nothing needs to be removed</b><br/>           “Everything in the material is essential and nothing needs to be added or removed.”<br/>           “Can’t think of anything that you can remove from the care guide”</p>  | <p><b>Everything can be relevant at some point</b><br/>           “Nothing, everything can be relevant to some patient at some point”</p> <p><b>Other comments</b><br/>           “You need to sign very often in the documents, in different places. Is that necessary?”</p> | <p><b>Not much could be removed</b><br/>           Overall very few could point out anything to remove from the S-PCG, so apart from small changes in paraphrasing, the content stayed the same.<br/>           The medication list was removed from the S-PCG and an opportunity to refer to the digital medication list was added to the documents.</p>   |
| <b>TEAMWORK – COMMUNICATION</b> | <p><b>S-PCG leads to discussion within the team about important issues</b><br/>           “S-PCG is a well drafted document that leads to good discussions on the important issues”<br/>           “Good that S-PCG makes the end-of-life conversations visible to everyone in the team”<br/>           “The S-PCG has started discussions about our current way of working”</p> <p><b>Resistance in the beginning and difficulties to get all team members on board</b><br/>           “Difficult to get employees on board in the beginning but it got better later”<br/>           “Difficult to get doctors on board”</p> <p><b>S-PCG can highlight the teamwork</b><br/>           “S-PCG can have a positive impact on the teamwork”<br/>           “As a doctor I can say that S-PCG lands more on the nurses’ side, but it is not overwhelming and there is much gained once part 2 has been done”<br/>           “Testing the S-PCG confirms that we (in our team) have very good procedures for the care”<br/>           “The S-PCG makes the assistant nurses feel more responsible for, and more involved in the palliative care”<br/>           “S-PCG clarifies what the assistant nurses do in the team – GOOD!”</p> | <p><b>Give patients and family<sup>2</sup> a clear information about who to contact and where to get support</b><br/>           “Contact information, we meet so many caretakers and I don’t know who I can call”<br/>           “The health care must help the patient to know where to go, who to talk to, or refer to others if the patient’s needs cannot be met.”<br/>           “The personnel need to make sure that the relatives are guided towards the right person/appropriate support when needed”</p> | <p><b>S-PCG demands teamwork</b><br/>           “S-PCG demands teamwork which is good”</p>  | <p><b>Highlighting the importance of teamwork</b><br/>           The online film about the S-PCG and what to think about before implementation includes topics about the importance of teamwork and tips on how to include the team in the implementation of the S-PCG.</p> <p><b>Guide the patient towards those that are needed</b><br/>           Space for written information about who to contact and contact information was added to a patient and family brochure.</p> |

| Items checked                                      | Comments from the clinical test users  | Patient-, family <sup>2</sup> - and public representatives  | The interdisciplinary advisory committee | Summary of issues addressed/raised after the evaluation  |
|--|--|---|--|--|
| <b>PATIENT- AND FAMILY<sup>2</sup> INVOLVEMENT</b> | <p><b>Patient involvement was not at barrier</b><br/>           "Information about S-PCG to patients and relatives is not perceived as a barrier"</p> <p><b>Family felt reassured and the support was improved</b><br/>           "S-PCG provides reassurance to the family members, once they have been informed about the care plan"<br/>           "Before the family were asking questions such as: has he/she gotten any pain-medicine recently? But now they can just see what we have done in the S-PCG, and all of a sudden there is time for other type of conversations"</p> <p><b>S-PCG clarifies the palliative care needs and the work that is being done</b><br/>           "S-PCG clarifies the palliative care needs"<br/>           "The family sees what we work with in the S-PCG and feel that it is important what we do"<br/>           "It is good to go through the S-PCG, you know what the patient and the family want"</p> <p><b>Some experienced barriers to involve the family</b><br/>           "It can be difficult with the needs of the family, they can have many needs and we want to focus our time on the patient"</p> | <p><b>Creates security for the family</b><br/>           "S-PCG creates security for the family, you check if the document is filled in"</p>  |  | <p><b>Patient involvement</b><br/>           A brochure with information for patients and their family was created together with patient- and family representatives.<br/>           The importance of patient involvement was raised in an online film about S-PCG users-experience as well as in the S-PCG education program.</p>  |
| <b>IMPLEMENTATION</b>                              | <p><b>Information and training in the S-PCG is needed</b><br/>           "More training is needed on S-PCG"<br/>           "You need to go through it together in the team to feel comfortable before you start using the S-PCG"<br/>           "Important to have knowledge of the S-PCG if it is to work"<br/>           "The S-PCG is good and useful but you need training in order to understand how to use the documents"</p> <p><b>Education in palliative care is needed</b><br/>           "With a lot of new staff, education in basic palliative care is needed"<br/>           "Doctors need more knowledge of palliative care in the hospitals"<br/>           "Training in basic palliative care for everyone"</p>   | <p><b>Highlight the importance of having a conversation with the family without the patient around</b><br/>           "To put in the user guide that it is important to sometimes have conversation with the family members without the patient around. When you ask questions about how the family member is holding up or if he/she needs support it is sometimes difficult to answer honestly if the patient is close by."</p> |  | <p><b>Brochures</b><br/>           A brochure containing short introduction on what S-PCG is and how it is intended to be used was created and made available both in print and online.<br/>           Another brochure was created with recommendations before implementation highlighting the importance of good planning before implementation.<br/>           The third brochure includes short practical users-instructions, aimed to be carried in the clinical work if needed.<br/>           The fourth brochure is aimed for patients and their families.</p> |

| Items checked                     | Comments from the clinical test users   | Patient-, family <sup>2</sup> - and public representatives' stories in the education  | The interdisciplinary advisory committee | Summary of issues addressed/raised after the evaluation  |
|-----------------------------------|---|---|--|--|
| <b>IMPLEMENTATION (continued)</b> | <p><b>Time to plan the use and the documentation-routines of the S-PCG</b></p> <p>"Time for planning the implementation"</p> <p>"To go through and decide on how the documentation procedures and routines should be, before the use is important"</p> <p>"We missed more time for planning"</p> <p><b>Support and understanding from managers</b></p> <p>"More involvement of the manager"</p> <p>"More support and assistance to the engage staff"</p> <p>"The nurses wanted to test the S-PCG on all of the patients but the management said no"</p> | <p><b>Include patients' stories in the education</b></p> <p>"I suggest that you record stories from patients with the question 'What is important to me?' and include it in the training/introduction of NVP to healthcare professionals. So, they don't forget why they are doing this work"</p> <p><b>Provide guidance and support for the health care personal to perform end-of life conversations.</b></p> <p>"Before implementing S-PCG on a new ward each clinic should ensure that they involve someone within the practice who is experienced in dealing with the difficult conversations, such as telling someone that they are dying. So that this person can guide and be supportive to the others"</p> |  | <p><b>Four films were created</b></p> <p>A film about the S-PCG including, among other things, interviews with patient, experts in palliative care, managers and health-care personnel as well as staged material.</p> <p>Another film aimed to inspire managers and leaders.</p> <p>A third film about how to use and what to think about before implementing the S-PCG.</p> <p>A fourth film about the users experience of using the S-PCG in clinical practice.</p> <p>All films are openly accessible online (65)<sup>3</sup>.</p> <p><b>Education</b></p> <p>The need for more training and preparation before implementation, as well as clearer instructions on how to use the S-PCG, was highlighted in the user-manual, in our films and in our brochures. A whole day educational program about S-PCG was also created, including, among other things, stories from a patient that was filmed.</p> <p>To address the need for education in basic palliative care, links to on-line education in palliative care were provided.</p> |

1) The more detailed feedback that was given for each section of every part of the S-PCG was very comprehensive and is not included in the table. This could include comments such as "Move section 2.8 further ahead" or "Under section 3.10, change: *contact with religious leader to contact with religious/spiritual leader*" or "add a checkbox for not applicable under section 4.5". All comments were however taken into consideration and discussed within the project group during the evaluation of the S-PCG.

2) The concept family is used here in its broadest sense and includes all persons of significance to the patient.

3) See reference number 65 in the reference list.





“Just think, there are no experiences that can compare to those of the childhood, imagine then if people could understand how important it is precisely with everything that concerns children...  
...because it shapes them for life”

(Astrid Lindgren in a letter to a friend, February 1963)



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