

# Components of perinatal palliative care: a systematic review protocol

**Laure Dombrecht** (✉ [laure.dombrecht@ugent.be](mailto:laure.dombrecht@ugent.be))

Vrije Universiteit Brussel <https://orcid.org/0000-0002-7174-4678>

**Kenneth Chambaere**

Ghent University and Vrije Universiteit Brussel

**Kim Beernaert**

Ghent University and Vrije Universiteit Brussel

**Ellen Roets**

University Hospital Ghent: Universitair Ziekenhuis Gent

**Mona De Vilder De Keyser**

Ghent University: Universiteit Gent

**Gaëlle De Smet**

Ghent University: Universiteit Gent

**Kristien Roelens**

University Hospital Ghent: Universitair Ziekenhuis Gent

**Filip Cools**

Free University of Brussels Hospital: Universitair Ziekenhuis Brussel

---

## Protocol

**Keywords:** perinatal palliative care, perinatal healthcare, fetuses, neonates

**Posted Date:** April 30th, 2021

**DOI:** <https://doi.org/10.21203/rs.3.rs-467354/v1>

**License:**  This work is licensed under a Creative Commons Attribution 4.0 International License.

[Read Full License](#)

---

# Abstract

**Background** When a severe diagnosis is made before or shortly after birth, perinatal palliative care can be provided to support the infant, parents and involved healthcare providers. However, since this is a relatively new and evolving field of study, a large variation exists amongst perinatal palliative care initiatives internationally. The field currently lacks a systematic overview of effectiveness and working components of such initiatives.

**Methods** A systematic search will be conducted in MEDLINE, Embase, CENTRAL, CINAHL, PsycInfo and Web of Science. We will include quantitative and qualitative study designs examining the effect of perinatal palliative care compared to regular care provided on pain and/or symptom relief, quality of care for, and quality of life of fetuses/infants, parents, family members and involved healthcare providers; and quantitative and qualitative study designs describing the components of care included in existing perinatal palliative care initiatives. Three independent authors will review titles and abstracts and screen full texts against eligibility criteria. Two standardized scales will be used to assess the quality of included studies: one for quantitative research from the NHS Centre of Reviews and Dissemination guidelines for observational studies and for quantitative research, and one for qualitative research from Hawker critical appraisal tool for evaluating qualitative and mixed research. One reviewer will carry out full data extraction and quality assessment, and a 20% random sample will be extracted and assessed by two independent reviewers. An overview table of all identified perinatal palliative care initiatives, including their components and their (preliminary) effectiveness and measured outcome variables when applicable, will be construed. Our protocol adheres to the Preferred Reporting Items for Systematic Review and Meta-Analysis Protocols (PRISMA-P) checklist for study protocols.

**Discussion** This systematic review will provide a comprehensive overview of palliative care programs in the perinatal period and their working components. This will aid the construction of a reliable and valid perinatal palliative care intervention with the aim of developing and piloting a Perinatal Palliative Care program within the existing perinatal healthcare setting, in order to provide the best possible care for fetuses and neonates with a life-limiting diagnosis, their families, and involved healthcare providers.

## Background

Despite an increase of possible medical and technical interventions for critically ill infants before and after birth<sup>1</sup>, foetal and neonatal death (i.e. from 22 weeks gestation until 28 days after birth<sup>2</sup>) occurs in 6.8 in 1000 births across Flanders<sup>3</sup>. This is comparable with death rates reported, for instance, in the United States<sup>4</sup>. A life-limiting diagnosis can be made either before the child is born or after birth<sup>5</sup>, and is associated with a high need for comfort care in fetuses or infants<sup>6</sup>, emotional distress and need for support in parents<sup>7</sup>, and a need for guidance of healthcare providers<sup>8</sup> in this stressful time. In these instances, perinatal palliative care can be provided as a multidisciplinary care approach for an infant who is diagnosed with a life-limiting disease or a complex medical disease with uncertain prognosis<sup>9</sup>, since palliative care aims to improve the quality of life of patients and their families facing such challenges

through prevention and relief of symptoms (WHO definition<sup>10</sup>). This new and emerging field of study currently lacks a systematic overview of the working components of perinatal palliative care programs and therefore, it seems that a large variation amongst initiatives occurs internationally<sup>2,6,7,9,11,12</sup>.

Palliative care programs are not routinely implemented in perinatal healthcare due to misconceptions such as fear of hastening death or due to inadequate training in providing adequate palliative care<sup>13</sup>. Earlier single center studies show proof of concept regarding the implementation of palliative care guidelines by showing improvements in reorientation of care from invasive treatments to comfort (palliative) care<sup>14</sup> and increased use of comfort medication (benzodiazepines and/or opioids)<sup>15</sup> reducing unnecessary suffering of hospitalized neonates who are dying. Additional to providing comfort care for the child, perinatal palliative care could address the complex family needs in an emotionally turbulent time by providing a family-centered approach with a focus on parental (spiritual and cultural) values, memory making, and compassionate communication between parents and providers<sup>2,16,17</sup>.

Palliative care in obstetrics and neonatology is obviously stressful for these healthcare providers causing a high risk for moral distress, burnout, and compassion fatigue<sup>18-20</sup>. Therefore, besides support for the (unborn) child and their family, perinatal palliative care could also benefit involved healthcare providers. Existing perinatal palliative care protocols include sections on staff support which could be used to counteract these problems, and improve quality of care<sup>21,22</sup>.

In contrast to palliative care research in adults, perinatal palliative care in obstetrics and neonatology is a relatively new field where the possible beneficial impact is hypothesized but has not yet been evaluated<sup>23</sup>. Several reports on perinatal palliative care protocols, teams or educational interventions exist, though the reported protocols or interventions are not evidence based and research on their feasibility and effects is lacking<sup>21</sup>. Furthermore, components mentioned in existing perinatal palliative care programs seem to show a large variation<sup>2,6,7,9,11,12</sup>. We therefore first need to systematically review the literature describing existing perinatal palliative care initiatives internationally, so that relevant working components can be summarized and used as a basis to develop such a perinatal palliative care intervention.

To our knowledge, there is currently no systematic overview to date of known perinatal palliative care initiatives and their working components. Available reviews<sup>6,7,24-27</sup> mainly focus on reporting the state of art of perinatal palliative care without discussing relevant components of care<sup>6,24</sup>, listing experiences of involved healthcare professionals or parents<sup>24,25</sup>, or they solely include either neonatal palliative care<sup>26,27</sup> or antenatal palliative care for parents who opt to continue a pregnancy after receiving a life-limiting fetal diagnosis<sup>7,23</sup>, hereby excluding families deciding to terminate the pregnancy which would also benefit from palliative care and additional support<sup>28</sup>.

### Research questions

We will conduct a comprehensive systematic literature review, focusing on the following research questions:

1. In fetuses or neonates with a potentially life-threatening or life-limiting diagnosis, what is the effect of perinatal palliative care compared to regular care provided on pain and/or symptom relief, quality of care for, and quality of life of fetuses/infants, parents, family members and involved healthcare providers?

2. Which components are incorporated in perinatal palliative care programs for fetuses or neonates with a life-threatening or life-limiting diagnosis, their parents and involved healthcare providers?

- Which actions or goals are described in each of the components?
- Which involved actor(s) is/are the main focus of the components (fetuses/infants, parents, healthcare providers...)?
- When in the care trajectory are the components situated? (from start of diagnosis/ when death is imminent/ when TOP is refused/...)
- Which population(s) is/are targeted by the components (prenatal, perinatal, neonatal, termination of pregnancy included, specific diagnoses...)?
- What are the envisioned outcomes of the program and the discussed components?

This review will be used to develop the first Belgian perinatal palliative care intervention to improve quality of care at the end of a fetus' or infant's life. Although some of the previously mentioned working components of perinatal palliative care are currently already being implemented in regular perinatal care, they are not integrated in an overall perinatal palliative care plan and they can thus be optimized.

## Methods

This study protocol has drawn upon the Preferred Reporting Items for Systematic Review and Meta-Analysis Protocols (PRISMA-P) guidelines for the reporting of systematic review protocols. The study protocol is registered through the PROSPERO registry of systematic reviews (CRD42021239005).

### Concepts and definitions

#### Perinatal palliative care:

For the purpose of this study, perinatal palliative care will be defined as a multi-component provision of care for fetuses or neonates with a serious illness in the perinatal period for which care has been redirected from life-saving to comfort-providing (22 weeks of gestation – 28 days after birth) and their parents, families and involved healthcare providers, aimed to relieve pain and control symptoms, and to improve the quality of care for, and wellbeing of, fetuses and infants, their families, and involved healthcare providers. It is holistic, family-centered, comprehensive, and multidimensional so that it addresses not only the physical aspect, but also the psychological, social, and spiritual dimensions.

#### A serious illness:

A serious illness is defined as either a lethal diagnosis in the prenatal or neonatal period, or a diagnosis for which there is little or no prospect of long-term survival without severe morbidity or extremely poor quality of life, and for which there is no cure. We will not define a limited group of diseases for which palliative care is needed, to maximize the sensitivity of our search.

## **Eligibility criteria**

### **Eligibility criteria regarding the study designs**

#### Inclusion:

For research question 1 we will include all empirical studies that examine the effect of perinatal palliative care on pain and/or symptom relief, quality of care for, and wellbeing of fetuses/infants, parents, family members and involved healthcare providers. This includes interventional, observational and survey designs (pre-post study designs, non-randomized trials, randomized controlled trials, cohorts, case-control designs, cross-sectional designs, prospective survey designs, and retrospective survey designs), and qualitative studies.

To achieve research question 2, we will expand on the previous inclusion criteria to studies that discuss the development, description and/or evaluation of a perinatal palliative care program/intervention/guideline. Additionally, protocol-papers or studies with a single case design that discuss the development, description and/or evaluation of a perinatal palliative care program/intervention/guideline will be included.

#### Exclusion:

We will exclude meta-analyses, literature reviews and searching so called 'gray literature'. Meta-analyses and reviews will be excluded because individual studies mentioned in these papers should already appear in our selection. Gray literature will be excluded from this review, as we limit our search to major databases and reference lists of selected articles.

### **Eligible study populations**

#### Inclusion:

We will include all publications that either evaluate the effect of, or describe a program, guideline or protocol concerning palliative care provided to fetuses or neonates in the perinatal period, their parents and involved healthcare providers. The study population is thus defined as fetuses or neonates from 22 weeks of gestation up until 28 days after birth with a serious illness where palliative care is being provided. Terms such as "fetus", "neonate" or "infant", or other terms related to the perinatal, prenatal or neonatal period will be used.

#### Exclusion:

Studies *solely* focusing on pediatric palliative care will be excluded, as well as studies solely focusing on adult palliative care. When it is unclear whether the age group of a study is considered eligible, the paper in question will be discussed amongst all investigators (including two gynecologists and a neonatologist) to decide whether the correct inclusion criteria apply.

## **Intervention or exposure**

### Inclusion:

We will include all publications that meet the following criteria:

- They either evaluate the effect of, or describe a program, guideline or protocol concerning perinatal palliative care, which according to the definition provided above constitutes of a multi-component program. Programs focusing for example solely on relieving pain and controlling symptoms at the end of life are thus excluded.
- The program, guideline or protocol concerns providing palliative care for fetuses and infants with a serious illness in the perinatal period, see definition of serious illness and included population above.
- For the purpose of this review, included programs need to at least identify themselves as a perinatal, prenatal and/or neonatal palliative care or perinatal, prenatal and/or neonatal hospice program.

## **Other**

### Inclusion:

We will include all publications published from 1/1/1997 onwards, because the first commentary on perinatal palliative care was published in 1997<sup>29</sup>, which was later identified as the start of perinatal palliative care<sup>30</sup>.

Additionally, we will include publications with the title and abstracts published in English. Full texts in other languages than English or Dutch will be translated by a native speaker when needed. No language restrictions will be imposed.

### Exclusion:

Studies for which a full text is unavailable or non-existent (e.g. in the case of conference participation) will be excluded from this review.

## **Systematic search**

### **Sources**

The systematic search will be conducted in MEDLINE (using the PubMed interface), Embase (using the embase.com interface), CENTRAL, CINAHL (using the EBSCO interface), PsycInfo, and Web of Science. In addition, we will conduct a manual search of the reference list of included studies.

## **Search strategy**

The search strategy will be structured around two distinct blocks: population and palliative care (see above and in the search query for PubMed in Table 1). Additionally, the block 'perinatal palliative care' will be added in case the combination of the two individual search blocks overlooks relevant research.

The PubMed database and syntax will be used as a starting point for the construction and validation of the search strategy. For construction of the search strategy, the search blocks will be based on previous search filters as well as Medical Subject Headings (MesH) terms and free text words. Relevant papers resulting from a scoping review will be consulted for MesH terms and relevant free text words to set up and/or refine the search strategy. An information specialist specialized in the development of search strategies, database searches and systematic review of literature will be consulted throughout this process.

To test the reliability and the sensitivity of this search strategy, a scoping review was done. A set of records used to validate the search strategy will be constructed by hand-searching volumes of the five journals that provided the highest number of relevant articles during this initial scoping review in the period between 2000 and 2021. This set of records will include all articles resulting from this hand-search that adhere to our eligibility criteria.

The PubMed search strategy will be translated to the other databases. The search strategy will adhere to the Peer Review of Electronic Search Strategies (PRESS) criteria for electronic search strategies as stated in the guideline statement<sup>31</sup>.

## **Publication selection**

### Abstract screening

Papers will be selected by three independent researchers (postdoctoral researcher and two master students in Medicine) following Cochrane guidelines to reduce bias with a two-step screening process using the Rayyan tool. Endnote will be used to upload all references of selected articles; to extract all titles, abstracts and pdfs; and to deduplicate search results. Any disagreement between reviewers will be resolved by discussing the eligibility criteria with a fourth reviewer, and by verifying the full-text article if necessary.

### Full-text screening

The full-text article of studies identified as potentially relevant based on their abstract (see abstract screening) will be reviewed by three independent reviewer(s) to determine whether or not the reported findings meet the inclusion criteria. Ambiguous full-text records will be examined with all investigators.

For the full-text review, we will look for the selected articles online, utilize our university library services to obtain articles requiring subscription, and/or contact colleagues and authors to obtain the full text. When no full text of the article can be obtained this way, the record will be excluded. The reference lists of all identified publications will then be screened for additional relevant publications.

## **Data extraction and quality assessment**

### **Data extraction**

From each selected full text article, the following information will be extracted (when applicable): title, author(s), publication year, journal title, study design, study (care) setting, aim/research question(s), targeted population (perinatal, prenatal and/or neonatal), components of care included in the perinatal, prenatal or neonatal palliative care program/intervention/guideline, start of the program (in time), number of participants, envisioned outcomes and effectiveness.

### **Risk of bias and quality assessment**

The quality of each included study will be assessed to inform the researchers and readers about the quality of the research and to guide the interpretation of the findings. Two standardized scales will be used namely an adapted scale for quantitative research<sup>32</sup> (from the NHS Centre of Reviews and Dissemination guidelines for observational studies and for quantitative research<sup>33</sup>, ranges from 0 or poor to 16 or good quality), and an adapted scale for qualitative research<sup>32</sup> (from Hawker critical appraisal tool for evaluating qualitative and mixed research<sup>34</sup>, ranges from 0 or very poor to 30 or good quality). One reviewer will carry out full data extraction and quality assessment, and a 20% random sample will be extracted and assessed by two independent reviewers. Discrepancies will be resolved through consensus with a third reviewer.

### **Data synthesis**

We will create an overview table of all identified perinatal, prenatal and/or neonatal palliative care programs/interventions/guidelines, their included components, the targeted population, the main actors focused on with each component, the start/timing of the program, and when applicable, their (preliminary) effectiveness and measured outcome variables.

## **Discussion**

To our knowledge, no systematic review of studies has yet been conducted that examines the existing perinatal palliative care initiatives and their working components. Additionally, existing reviews on the subject of palliative care in the perinatal period often solely focus on the relatively small group of families who receive a life-limiting fetal diagnosis before birth and decide to continue the pregnancy<sup>28,35</sup>, hereby excluding families receiving a life-limiting diagnosis either at birth or in the neonatal period. In the majority of cases, these fetuses and neonates where a life-limiting disorder is diagnosed are in essence

the same patients with similar disorders or congenital anomalies, the only difference being the occurrence of birth<sup>5</sup>. Additionally, the impact on parents and involved caregivers is very similar. Our review will not impose these unnecessary restrictions on included palliative care programs. This systematic review will thus result in a comprehensive overview of palliative care programs in the perinatal period and their working components. This will aid the construction of a reliable and valid perinatal palliative care intervention with the aim of developing and piloting a Perinatal Palliative Care program within the existing perinatal healthcare setting, in order to provide the best possible care for fetuses and neonates with a life-limiting diagnosis, their families, and involved healthcare providers. This review protocol is published to allow other researchers to compare previously established methods to the final review, and to promote quality adherence.

### **Anticipated limitations and potential problems**

An expected limitation of the review is the broad inclusion criteria and resulting broad search string, leading to a large amount of possibly irrelevant papers in the initial results. Despite the fact that most irrelevant articles will be filtered out in the abstract selection phase, we expect that the lack of specificity will lead to this phase taking a relatively long time. However, as perinatal palliative care is a new and emerging field<sup>23</sup>, variation in terminology exists and thus the broad search string is needed to ensure that no relevant information is missed. Additionally, these broad inclusion criteria will possibly result in the inclusion of studies with a large variety in etiologies of fetuses and neonates. We will ensure that a subgroup analysis is included in our synthesis based on the following hypotheses: 1) differences in perinatal palliative care approaches depending on whether the care plan starts in the prenatal versus the neonatal period; 2) differences in palliative care approaches depending on the region where care was provided; and 3) a possible evolution in time of perinatal palliative care approaches from 1997 up until 2021.

Our preliminary, non-systematic review revealed that the evidence base on perinatal palliative care programs is limited, and that research on the implementation and effects of these programs is lacking<sup>21</sup>. Outcome measures and assessments of quality of care are often not adequately examined, and when outcomes are assessed they mostly include evidence of a change of practice instead of assessing patient comfort or parents' or provider satisfaction, making it impossible to adequately examine the added value for infants, parents and healthcare providers<sup>21</sup>. We therefore expect our synthesis of effectiveness to yield limited insights. This would confirm the dire need for the rigorous evaluation of perinatal palliative care programs to provide an evidence base for the relatively new field of palliative care in obstetrics and neonatology where the possible beneficial impact is only hypothesized<sup>23</sup>.

Our inclusion criteria allow for both qualitative and quantitative studies to be included in the systematic review. This might complicate the synthesis of all available evidence. Yet by including both –systematic reviews and meta-analysis are often limited to quantitative studies – the resulting overview will be able to broaden our understanding of the current evidence base. This for example by combining existing quantitative evidence on effectiveness with qualitative evidence on the way in which quality of care was

improved for bereaved parents to hypothesize on the pathways in which working components actually improve care overall. Hereby, we will not only be able provide a broad spectrum of available perinatal palliative care programs and their components, but we also aim to provide a deeper understanding in the way these components impact on the devastating experience of losing a child in the perinatal period. Both the broad overview of components and the deeper understanding of experiences of involved persons (parents, family members, healthcare providers) are crucial in developing a new perinatal palliative care intervention.

### **Prospective registration of the review protocol**

The main components of the present review protocol will be formally registered in the PROSPERO database. Registration will be made after the search strategy has been validated (see point on search strategy), but before the first extraction of references is performed. This will allow us to demonstrate that our review was planned, had clear a priori hypotheses, and was conducted according to a set of pre-specified criteria.

## **Declarations**

Ethics approval and consent to participate: Not applicable.

Consent for publication: Not applicable.

Availability of data and material: Search strings and additional information on the search strategy are available upon written request to the corresponding author ([Laure.dombrecht@vub.be](mailto:Laure.dombrecht@vub.be)).

Competing interests: No conflicts of interest exist for the authors.

Funding: This study is funded by Research Foundation Flanders (Fonds voor Wetenschappelijk onderzoek, FWO), grant number *G023520N*.

Authors' contributions: All authors contributed to the design and implementation of the study and to the writing of the manuscript. All authors take responsibility for the integrity of this manuscript. All information reported is honest, accurate and transparent. All authors read and approved the final manuscript.

Acknowledgements: We would like to thank information specialist Nele Pauwels from the Knowledge Center for Health at Ghent University for lending her expertise in reviewing our preliminary search string.

## **References**

1. Rüegger C, Heggin M, Adams M, Bucher HU. Population based trends in mortality, morbidity and treatment for very preterm- and very low birth weight infants over 12 years. *BMC Pediatr.* 2012;12(1):17. doi:10.1186/1471-2431-12-17

2. Denney-koelsch E, Black BP, Côté-Arsenault D, Wool C, Sujeong K, Kavanaugh K. A Survey of Perinatal Palliative Care Programs in the United States: Structure, Processes, and Outcomes. *J Palliat Med.* 2016;19(10):1080-1086. doi:10.1089/jpm.2015.0536
3. Devlieger R, Martens E, Goemaes R, Cammu H. Perinatale Activiteiten in Vlaanderen 2017. *Stud voor Perinat Epidemiol.* Published online 2018.
4. Barfield WD, COMMITTEE ON FETUS AND NEWBORN. Standard Terminology for Fetal, Infant, and Perinatal Deaths. *Pediatrics.* 2016;137(5):e20160551-e20160551. doi:10.1542/peds.2016-0551
5. Dombrecht L, Beernaert K, Roets E, et al. A post-mortem population survey on foetal-infantile end-of-life decisions: a research protocol. *BMC Pediatr.* Published online 2018:1-9. <https://rdcu.be/3ZNc>
6. Balaguer A, Martín-ancel A, Ortigoza-escobar D, Escribano J, Argemi J. The model of palliative care in the perinatal setting: a review of the literature The model of palliative care in the perinatal setting: a review of the literature. 2012;25(March).
7. Wool C. State of the Science on Perinatal Palliative Care. *J Obstet Gynecol Neonatal Nurs.* 2013;42(3):372-382. doi:10.1111/1552-6909.12034
8. Haug S, Farooqi S, Wilson CG, Hopper A, Oei G, Carter B. Survey on Neonatal End-of-Life Comfort Care Guidelines Across America. *J Pain Symptom Manage.* 2018;55(3):979-984. doi:10.1016/j.jpainsymman.2017.10.023
9. Catlin A, Carter B. Creation of a Neonatal End-of-Life Palliative Care Protocol. *J Perinatol.* 2002;22:184-195. doi:10.1038/sj/jp/
10. World Health Organization. WHO Definition of Palliative Care. Published 2019. <https://www.who.int/cancer/palliative/definition/en/>
11. Wool C, Co D, Black BP, Denney-koelsch E, Kim S, Kavanaugh K. Provision of Services in Perinatal Palliative Care: A Multicenter Survey in the United States. 2016;19(3):279-285. doi:10.1089/jpm.2015.0266
12. Sidgwick P, Harrop E, Kelly B, Todorovic A, Wilkinson D. Fifteen-minute consultation: perinatal palliative care. *Arch Dis Child Educ Pract.* 2017;102:114-116. doi:10.1136/archdischild-2016-310873
13. Martin M. Missed opportunities: a case study of barriers to the delivery of palliative care on neonatal intensive care units. *Int J Palliat Nurs.* 2013;19(5):251-256.
14. Samsel C, Lechner BE. End-of-life care in a regional level IV neonatal intensive care unit after implementation of a palliative care initiative. *J Perinatol.* 2015;35(3):223-228. doi:10.1038/jp.2014.189
15. Younge N, Smith PB, Goldberg RN, et al. Impact of a palliative care program on end-of-life care in a neonatal intensive care unit. *J Perinatol.* 2015;35(3):218-222. doi:10.1038/jp.2014.193
16. McMahon DL, Twomey M, O'Reilly M, Devins M. Referrals to a perinatal specialist palliative care consult service in Ireland, 2012 – 2015. *Arch Dis Child - Fetal Neonatal Ed.* 2018;103:F573-F576. doi:10.1136/archdischild-2017-313183

17. Carter BS. Pediatric Palliative Care in Infants and Neonates. *Children*. 2018;5(21):1-9. doi:10.3390/children5020021
18. Bellieni C V., Righetti P, Ciampa R, Lacoconi F, Coviello C, Buonocore G. Assessing burnout among neonatologists. *J Matern Neonatal Med*. 2012;25(10):2130-2134.
19. Profit J, Sharek PJ, Amspoker AB, et al. Burnout in the NICU setting and its relation to safety culture. *BMJ Qual Saf*. 2014;23:806-813. doi:10.1136/bmjqs-2014-002831
20. Kenner C, Press J, Ryan D. Recommendations for palliative and bereavement care in the NICU: a family-centered integrative approach. *J Perinatol*. 2015;35(S1):S19-S23. doi:10.1038/jp.2015.145
21. Parravicini E. Neonatal palliative care. *Curr Opin Pediatr*. 2017;29(2):135-140. doi:10.1097/MOP.0000000000000464
22. Wallbank S, Robertson N. Predictors of staff distress in response to professionally experienced miscarriage , stillbirth and neonatal loss: A questionnaire survey. *Int J Nurs Stud*. 2013;50(8):1090-1097. doi:10.1016/j.ijnurstu.2012.11.022
23. Catania TR, Bernardes LS, Benute GRG, et al. When One Knows a Fetus Is Expected to Die: Palliative Care in the Context of Prenatal Diagnosis of Fetal Malformations. *J Palliat Med*. 2017;20(9):1020-1031. doi:10.1089/jpm.2016.0430
24. Wool C. Systematic review of the literature: parental outcomes after diagnosis of fetal anomaly. *Adv Neonatal Care*. 2011;11(3):182-192. doi:10.1097/ANC.0b013e31821bd92d
25. Gandino G, Bernaudo A, Fini G Di, Vanni I, Veglia F. Healthcare professionals ' experiences of perinatal loss: A systematic review. *J Health Psychol*. 2019;24(1):65-78. doi:10.1177/1359105317705981
26. Zuniga-villanueva G, Widger K, Medeiros C, Trenholm M, Streuli BJC. Specialized Pediatric Palliative Care in Neonates with Life-Limiting Illness: A Systematic Review. *Am J Perinatol*. 2020;1(212).
27. Moro T, Kavanaugh K, Okuno-Jones S, VanKleef J. Neonatal end-of-life care: A review of the research literature. *J Perinat Neonatal Nurs*. 2006;20:262-273.
28. Marc-aurele KL, Nelesen R. A Five-Year Review of Referrals for Perinatal Palliative Care. *J Palliat Med*. 2013;16(10):1232-1236. doi:10.1089/jpm.2013.0098
29. Calhoun B, Hoeldtke N, Hinson R, Judge K. Perinatal hospice: should all centers have this service? *Neonatal Netw*. 1997;16(6):101-102.
30. Limbo R, Wool C. Perinatal Palliative Care. *JOGNN - J Obstet Gynecol Neonatal Nurs*. 2016;45(5):611-613. doi:10.1016/j.jogn.2016.07.002
31. Beller EM, Glasziou PP, Altman DG, et al. PRISMA for Abstracts: Reporting Systematic Reviews in Journal and Conference Abstracts. *PLoS Med*. 2013;10(4). doi:10.1371/journal.pmed.1001419
32. Gomes B, Calanzani N, Gysels M, Hall S, Higginson IJ. Heterogeneity and changes in preferences for dying at home: a systematic review. *BMC Palliat Care*. 2013;12(7):1-13. <https://bmcpalliatcare.biomedcentral.com/articles/10.1186/1472-684X-12-7>

33. Khan KS, Riet G Ter, Glanville J, Sowden AJ, Kleijnen J. *Undertaking Systematic Reviews of Research on Effectiveness: CRD's Guidance for Carrying out or Commissioning Reviews*. Vol 4.; 2001.
34. Sheila H, Payne S, Kerr C, Hardey M, Powell J. Appraising the evidence: reviewing disparate data systematically. *Qual Health Res*. 2002;12(9):1284-1299.
35. Tosello B, Dany L, Gire C, et al. Perceptions of Lethal Fetal Abnormality among Perinatal Professionals and the Challenges of Neonatal Palliative Care. *J Palliat Med*. 2014;17(8):924-930. doi:10.1089/jpm.2014.0023

## Table

Table 1: search string PubMed

Search block	Mesh	Text
<b>Population</b>	Fetus, neonate, infant  "infant, newborn"[MESH] "Fetus"[Mesh:NoExp]	infant* [Title/Abstract] OR newborn* [Title/Abstract] OR neonat* [Title/Abstract] OR newly born [Title/Abstract] OR newly-born [Title/Abstract] OR new-born* [Title/Abstract]  Foetus [Title/Abstract] OR Fetus [Title/Abstract] OR Foetal [Title/Abstract] OR Fetal [Title/Abstract]
	Death of fetus, neonate, infant  "Infant Mortality"[Mesh] OR "Infant Death"[Mesh:NoExp]  "Fetal Death"[Mesh] OR "Fetal Mortality"[Mesh]  "Perinatal Mortality"[Mesh] OR "Perinatal Death"[Mesh]	(neonatal[tiab] OR perinatal[tiab] OR fetal[tiab] OR foetal[tiab] OR infant[tiab]) AND (death[tiab] OR mortality[tiab] OR demise[tiab] OR loss[tiab])  (Terminat*[Title/Abstract] AND pregnanc* [Title/Abstract])
<b>Palliative care</b>	"Hospice and Palliative Care Nursing"[Mesh] OR "Palliative Medicine"[Mesh] OR "Palliative Care"[Mesh] OR "Terminal Care"[Mesh]	(palliative[tiab] OR end-of-life[tiab] OR end of life[tiab] OR EOL[tiab] OR comfort[tiab] OR hospice[tiab] OR terminal[tiab]) AND (care[tiab] OR nursing[tiab] OR medicine[tiab])  palliation[tiab]
<b>Perinatal palliative care</b>		perinatal palliative care [Title/Abstract] OR perinatal hospice [Title/Abstract]
<b>Not animals</b>	NOT ("Animals"[Mesh] NOT "Humans"[Mesh])	