

**ASSESSING PERINATAL PALLIATIVE CARE EDUCATION WITHIN GENETIC  
COUNSELING TRAINING PROGRAMS**

by

**Megan M. Hager**

BS Biological Sciences, University of Missouri, 2015

BA Spanish, University of Missouri, 2015

Submitted to the Graduate Faculty of  
The Department of Human Genetics  
of the requirements for the degree of  
Master of Public Health and Master of Science

University of Pittsburgh

2018

UNIVERSITY OF PITTSBURGH  
GRADUATE SCHOOL OF PUBLIC HEALTH

This thesis was presented

by

Megan M. Hager

It was defended on

May 14, 2018p

and approved by

Scott Maurer, MD, Associate Professor, School of Medicine, University of Pittsburgh,  
Division of Pediatric Hematology/Oncology and Blood and Marrow Transplantation,  
Children's Hospital of Pittsburgh

Candace Kammerer, PhD, Associate Professor of Human Genetics, Program Director, MPH  
in Public Health Genetics, Graduate School of Public Health, University of Pittsburgh

Meredith Jones, MS, LCGC, Genetic Counselor, Magee-Women's Hospital of University of  
Pittsburgh Medical Center

**Thesis Director:** Robin E. Grubs, MS, PhD, LCGC, Associate Professor of Human Genetics,  
Program Director, Genetic Counseling Program, Graduate School of Public Health,  
University of Pittsburgh

Copyright © by Megan M. Hager

2018

# **ASSESSING PERINATAL PALLIATIVE CARE EDUCATION WITHIN GENETIC COUNSELING PROGRAMS**

Megan M. Hager, MS, MPH

University of Pittsburgh, 2018

## **ABSTRACT**

Perinatal palliative care (PPC) is a clinical program that provides holistic care to critically ill fetuses and infants alongside psychosocial support to their family members. Genetic counselors are often consulted to explain prenatal or infantile conditions that are life-limiting or life-threatening. The function of the genetic counselor is to provide both education about a condition and supportive resources for their clients. Referral to a PPC program is one way a counselor may provide support. Therefore, being knowledgeable about the medical and psychosocial services offered by PPC programs is important. A recent study found that 11.4% of genetic counselors feel uncomfortable referring to these programs due to a lack of familiarity with PPC. This study aims to increase genetic counselor knowledge about PPC programs through the creation of educational resource materials for genetic counseling training programs accredited by the Accreditation Council for Genetic Counseling (ACGC). A Qualtrics survey was developed and administered to ACGC accredited program leadership to assess PPC education within the curricula, clinical internships, and supplementary activities. Additionally, program directors were asked if programs would benefit from creation of resource material. Survey data shows 85% of responding programs provide education about PPC, and 90% of respondents support creation of educational resource materials. Using the results of the survey to direct content and format of resource material, three

documents were created: 1) A series of case studies were created that may also be used as roleplays, 2) An annotated bibliography directing users to articles, books, videos, and websites to increase PPC knowledge, and 3) A PowerPoint covering relevant topics in PPC. Additionally, a resource map identifying locations of PPC programs within the United States was created. This study is designed to impact public health by strengthening the educational experiences of genetic counseling students, with the goal of making them more comfortable when referring clients to PPC services.

## TABLE OF CONTENTS

<b>PREFACE.....</b>	<b>XII</b>
<b>ABBREVIATIONS.....</b>	<b>XIV</b>
<b>1.0 INTRODUCTION.....</b>	<b>1</b>
<b>1.1 BACKGROUND AND SPECIFIC AIMS .....</b>	<b>1</b>
<b>1.1.1 Specific Aim 1.....</b>	<b>4</b>
<b>1.1.2 Specific Aim 2.....</b>	<b>4</b>
<b>1.1.3 Specific Aim 3.....</b>	<b>4</b>
<b>2.0 REVIEW OF THE LITERATURE.....</b>	<b>5</b>
<b>2.1 THE HISTORY OF PERINATAL PALLIATIVE CARE .....</b>	<b>5</b>
<b>2.2 CLASSIFICATION OF PERINATAL DEATH.....</b>	<b>6</b>
<b>2.3 STRUCTURE OF PERINATAL PALLIATIVE CARE PROGRAMS.....</b>	<b>11</b>
<b>2.4 REFERRING FAMILIES TO PERINATAL PALLIATIVE CARE.....</b>	<b>12</b>
<b>2.4.1 State Laws.....</b>	<b>12</b>
<b>2.4.2 Rural Outreach .....</b>	<b>13</b>
<b>2.5 CARE PROVIDED IN PERINATAL PALLIATIVE CARE PROGRAMS</b>	<b>15</b>
<b>2.5.1 Medical Care for the Fetus or Infant .....</b>	<b>16</b>
<b>2.5.2 Supportive Care for the Family.....</b>	<b>18</b>
<b>2.5.3 Facilitating the Decision-Making Process and Bridging Communication         Gaps .....</b>	<b>19</b>
<b>2.6 OUTCOMES OF PERINATAL PALLIATIVE CARE.....</b>	<b>21</b>
<b>2.6.1 Impact on Fetuses and Infants.....</b>	<b>21</b>

2.6.2	Impact on Families.....	22
2.6.3	Impact on Clinicians.....	23
2.7	<b>GENETIC COUNSELORS IN PERINATAL PALLIATIVE CARE .....</b>	<b>23</b>
2.7.1	Genetic Counselor Attitudes Toward Perinatal Palliative Care .....	24
2.7.2	Strategies for Genetic Counselors .....	25
2.8	<b>INCORPORATING EDUCATIONAL MATERIAL INTO CLINICAL TRAINING PROGRAMS .....</b>	<b>26</b>
2.8.1	Incorporating Perinatal Palliative Care Education into Physician Training.....	27
2.8.2	Incorporating Perinatal Palliative Care Education into Nurse Training.. .....	28
2.8.3	Incorporating Perinatal Palliative Care Education into Genetic Counselor Training.....	28
3.0	<b>MANUSCRIPT.....</b>	<b>30</b>
3.1	<b>BACKGROUND .....</b>	<b>30</b>
3.2	<b>MATERIALS AND METHODS.....</b>	<b>33</b>
3.2.1	Survey Design .....	33
3.2.2	Study Participants.....	34
3.2.3	Data Analysis.....	34
3.2.4	Creation of Educational Resource Material.....	35
3.3	<b>RESULTS .....</b>	<b>35</b>
3.3.1	Participants.....	36
3.3.2	PPC Content in Coursework/Curricula.....	37

3.3.3	PPC Content in Clinical and/or Supplementary Activities.....	40
3.3.4	Benefit of Educational Resources .....	42
3.4	DISCUSSION.....	45
3.4.1	Limitations.....	48
3.4.2	Future Directions .....	49
3.5	CONCLUSION .....	49
4.0	RESEARCH SIGNIFICANCE TO GENETIC COUNSELING AND PUBLIC HEALTH .....	51
5.0	EXAMINING ACCESSIBILITY OF PERINATAL PALLIATIVE CARE IN THE UNITED STATES.....	54
5.1.1	Study Aim 1 .....	57
5.1.2	Study Aim 2 .....	57
5.2	DESCRIPTION OF DATASET .....	58
5.2.1	Perinatal Palliative Care Programs .....	58
5.2.2	Genetic Counseling Training Programs .....	59
5.2.3	Infant Death Rate.....	60
5.2.4	Census data.....	60
5.3	METHODS.....	61
5.3.1	Mapping.....	61
5.3.2	Analysis Methods .....	61
5.4	RESULTS .....	62
5.4.1	PPC Program Resource Map (Aim 1).....	62
5.4.2	Results of Analysis by State (Aim 2b) .....	64



5.4.3	County-based Analyses (Aim 2c)	67
5.5	DISCUSSION	68
	APPENDIX A: SURVEY MATERIALS	71
	APPENDIX B: PERINATAL PALLIATIVE CARE EDUCATIONAL RESOURCE MATERIALS	77
	APPENDIX C: MAPS - SUPPLEMENTARY MATERIALS	114
	APPENDIX D: INSTITUTIONAL REVIEW BOARD APPROVAL	116
	BIBLIOGRAPHY	117

## LIST OF TABLES

Table 1. Fetal death reporting standards by state.....	8
Table 2. Response rate by region.....	36
Table 3. Spearman’s test applied to the correlation matrix in Figure 8.....	66
Table 4. State data.....	114
Table 5. Distribution of PPC programs within the United States.....	115

## LIST OF FIGURES

Figure 1. The percent response of what types of curricular content students consume within genetic counseling training programs.....	38
Figure 2. The percentage of responses for what topics are covered within curricular content.....	39
Figure 3. A graph of whether or not all students participate in clinical internships and/or supplementary activities.....	41
Figure 4. The percent response to what types of educational resource materials are desired by genetic counseling training programs. ....	42
Figure 5. The percent response to what educational topics should be covered in newly created educational resource materials.....	44
Figure 6. Screenshots of the custom map identifying every PPC program within the United States. ....	63
Figure 7. Number of PPC program locations in the United States by state as of December 01, 2017. ....	64
Figure 8. A correlation matrix representing strengths of relationships between the number of PPC programs, number of genetic counseling programs, the infant mortality rate, population level, and percent of the state living below the poverty line. ....	65
Figure 9. Correlation between the number of PPC and the number of GC programs after standardizing population.....	66
Figure 10. Number of PPC program locations in the United States per county. ....	67

## PREFACE

“Nothing of me is original. I am the combined effort of everyone I’ve ever known.”

-- Chuck Palahniuk, *Invisible Monsters*

I would like to start my thanks with Robin, Candy, Meredith, and Scott for serving on my thesis committee. Without your guidance, support, and editing, this thesis would not have come to fruition.

Then, I would like to thank the Department of Human Genetics. When I first interviewed for a position in the Genetic Counseling program, I was skeptical. Would I like Pittsburgh? Would I fit in? That day, several professors stopped by just to say hello and I knew I was in the right place. The support of this program is unbelievable and I’m glad to have experienced potlucks, poster sessions, and Pymatuning with this batch of ~~crazy~~ geneticists.

Additional thanks to the supervising genetic counselors who have molded me, and a big thanks to Carol May for giving me the opportunity to gain firsthand experience with a supportive care team. These clinical opportunities have given me the confidence to become a great genetic counselor.

Next, my dearest classmates, and now colleagues. I’m so glad to have gotten to know each of you. The ten of you – Julia, Julia, Joya, Jackie, Jenni, Emily, Emily, Claire, Kaitlin, and Seth – have helped shape me as a counselor by creating a space where ideas can be shared freely. And thanks for tolerating my endless barrage of emails, blog posts, and Facebook polls.

A tremendous thank you to Robin and Andrea for taking on the triplet roles of program directors, mentors, and bosses. You both have somehow managed to provide the perfect blend of

encouragement, tough love, and inspiration throughout my educational experience at Pitt. I know that you will continue to create *dynamite* genetic counselors.

I would also like to thank my parents. Without you both, the 23 pairs of chromosomes that developed into me would never have met. I also appreciate all that you've done for me as I've evolved into who I am today. Much love to the rest of my family for their continued support.

Thank you to myself for finally learning the values of self-love, reflection, and confidence. The only person standing in my way this whole time was myself.

And finally, thank you, thank you, thank you to Brennan and Sinatra for following me from Missouri to Indiana to Pennsylvania, and now to Arizona, on this meandering path of self-discovery and pursuing my dreams. I'm so lucky to have both of you in my life. I know there were many days filled with doubts and anxiety – more than I would like to admit – and I'm so glad that I got to come home to the pair of you each and every one of those days. And especially on the days where I felt unstoppable.

## **ABBREVIATIONS**

### **Frequently Used Abbreviations**

ACGC = Accreditation Council for Genetic Counseling

AGCPD = Association for Genetic Counseling Program Directors

CDC = Centers for Disease Control and Prevention

tional Center for Health Statistics

NSGC = National Society of Genetic Counselors

PPC = Perinatal Palliative Care

WHO = World Health Organization

## **1.0 INTRODUCTION**

### **1.1 BACKGROUND AND SPECIFIC AIMS**

Perinatal palliative care (PPC) is a specialized clinical program that focuses on providing holistic care to critically ill fetuses and infants.<sup>1</sup> The majority of programs also focus on psychosocial support of the family through the creation of memories and mementos, counseling services, and spiritual guidance.<sup>2</sup> In short, PPC seeks to maximize the time that families have to spend with their child by creating a plan of care that accommodates the needs of the fetus/infant, parents, additional family members, and clinical care providers.<sup>3</sup> Adult hospice care, which began in the 1960s, has reached a level of importance to be considered a global public health priority.<sup>4</sup> Perinatal services have yet to receive the same level of attention, perhaps due to the lack of standardized practices within PPC programs throughout the United States and relative newness of many programs.<sup>5,6</sup> PPC did not truly begin in earnest until the 1980s, when physicians began recognizing that infants with severe anomalies deserved the same entitlement to a “natural death” as adults.<sup>7,8</sup> This realization coincided with advances in perinatal care that both identified congenital malformations at an earlier stage in pregnancy and increased survival outcomes for infants that were preterm, low birthweight, had severe congenital malformations, or were affected by genetic conditions.<sup>8</sup> These physicians argued that aggressive neonatal care, which focused on

prolonging life,<sup>7</sup> may result in greater suffering for both the infant and family by either increasing the amount of time between birth and demise or causing a child to survive with complex needs.<sup>9</sup>

Advances in medicine have contributed greatly to an increase in diagnosis of congenital malformations and chromosome conditions during the prenatal period.<sup>10,11</sup> Additionally, medical developments have contributed to a 76.16% decrease in fetal mortality since 1942<sup>12</sup> and allowed more infants affected by preterm birth, low birthweight, severe malformation, or chromosome conditions to survive for longer periods of time after birth.<sup>9</sup> However, a subset of these fetuses and infants are not expected to survive given the severity of their conditions.<sup>13</sup> Therefore, these families should receive assistance in coordinating palliative care measures for their children. To give an approximate idea of the scope of perinatal demise, 23,595 fetal deaths and 23,446 infant deaths were reported in the United States in 2013.<sup>12</sup> There is evidence to suggest that this number is inaccurate due to complications in characterizing and reporting fetal death, leading to an underestimate of fetal demise.<sup>14,15</sup>

The majority of perinatal deaths are attributed to major malformations, chromosome conditions, and unspecified conditions relating to low birthweight or preterm birth.<sup>12,16</sup> Couples experiencing – or who have experienced – one of these pregnancy complications may be referred for genetic counseling. For those receiving a prenatal diagnosis, they will be able to discuss possible outcomes of findings indicative of a pregnancy complication (e.g. serum screening, ultrasound), further screening and testing options available to them, and pregnancy management options.<sup>17,18</sup> For those receiving a postnatal diagnosis, genetic counselors may be sought out to provide more information about the condition, its underlying cause, recurrence risk for future children, and management or treatment options.<sup>17</sup>



When a fetus or infant has a life-limiting or life-threatening condition, PPC options should be offered alongside intensive medical intervention and termination of pregnancy.<sup>13</sup> Given their training, genetic counselors are well suited for the role of coordinating palliative services for a family and also facilitating the decision-making process for affected families. This is mostly due to the fact that the tenets of the profession underscore the importance of patient autonomy and delivering medical information in an unbiased fashion.<sup>19,20</sup> Genetic counselors are trained on delivering difficult news in a way that supports the patients, supplying accurate information on conditions that may result in perinatal demise, using psychosocial skills that allow the family to emotionally process the delivered information, and on connecting patients and families to support systems within their communities.<sup>19</sup>

As more fetuses and infants with complex care needs live for longer periods of time,<sup>15,21</sup> there is a greater likelihood that genetic counselors will come in contact with these families affected by perinatal complications or loss. In general, genetic counselors are aware of palliative care options and of PPC programs in their communities. However, many counselors are uncomfortable making referrals to PPC programs due to lack of familiarity with local PPC programs or due to few years of experience in their profession.<sup>19</sup> Increased education for genetic counseling trainees may help increase the familiarity with PPC, thus increasing appropriate referrals of families who may benefit from these services. As of yet, no studies have been conducted to examine what is being taught about PPC within Accreditation Council for Genetic Counseling (ACGC) accredited genetic counseling programs.

This study will provide current information on the amount and structure of content related to PPC in ACGC Accredited genetic counseling programs, as well as any gaps in knowledge or barriers to education that may exist. The results of this study will also be used to guide the

development of educational resource material for training programs to increase the knowledge base of genetic counselors entering the field. Ultimately, increasing genetic counselors' knowledge of PPC may enhance their comfort with referring patients to PPC programs.

### **1.1.1 Specific Aim 1**

Develop a Qualtrics survey to administer to ACGC accredited genetic counseling programs to determine if PPC education is integrated into program curricula, clinical training, and other supplementary activities.

### **1.1.2 Specific Aim 2**

Use the results of the administered survey to determine if there are any gaps in knowledge of or existing barriers to educating students about PPC.

### **1.1.3 Specific Aim 3**

Develop educational resource materials to be accessed by genetic counseling programs and their students.

## **2.0 REVIEW OF THE LITERATURE**

### **2.1 THE HISTORY OF PERINATAL PALLIATIVE CARE**

PPC is a relatively new service that provides care to families experiencing fetal death (death before complete expulsion or extraction from the uterus), miscarriage (fetal death  $\leq 20$  weeks gestation), stillbirth (fetal death  $\geq 20$  weeks gestation), neonatal death (death within 28 days of life), and infant death (death within the first year of life).<sup>2</sup> PPC programs provide extensive medical care to fetuses and infants with complicated medical conditions severely limiting their life potential, in addition to providing familial support. Medical support includes pain and symptom management, improving quality of life, and increasing duration of life. Family support includes spiritual support, counseling services, and the creation of mementos.<sup>1</sup>

PPC practices began as an extension of adult hospice practices. The foundation for modern adult hospice practices originated in the 1950s and 1960s when Cicely Saunders, who became a nurse, social worker, and physician throughout her career, noticed that terminally ill patients were not receiving appropriate management for the severity of their condition.<sup>4,22</sup> Wanting to ease their suffering during end-of-life care, Saunders advocated for better pain and symptom management. She also began educating the medical community on “total pain,” which recognizes the emotional, spiritual, religious, and physical aspects of pain and suffering.<sup>22-24</sup> Saunders went on to establish a hospice center and assembled a multidisciplinary team of clinicians to address critically ill adults,<sup>22</sup> and later advocated for the use of hospice practices in cases of critically ill children.<sup>4</sup> Out of this initiative, the palliative care movement quickly spread to much of the Western world.<sup>22</sup> Since then, adult palliative care practices have reached such a level of importance in end-of-life

management that the World Health Organization (WHO) has termed it a global public health need.<sup>4,14</sup> PPC has yet to receive the same level of attention or concern.

The first mention of PPC in scientific literature was 1982, but it is thought that PPC interventions began in the early 1970s when therapeutic obstination – an extension of life with little regard to quality or suffering – was employed with premature births. It was at this time that physicians began to recognize the importance of a “natural death” for neonates born with complex congenital conditions.<sup>4</sup> Since then, the field of PPC has gained greater recognition as a viable method of care for fetuses and infants with a life-limiting or life-threatening condition,<sup>3</sup> as evidenced by the significant increase in PPC program numbers throughout the United States. Greater than 70% of existing programs in the United States are less than 10 years old.<sup>5,6</sup> Initially PPC programs merely adopted adult hospice practices.<sup>1,8</sup> These interventions have been proven inadequate at handling the specific needs of fetuses, infants, and their families. Some steps have been taken to educate clinicians on PPC practices including more training on delivery of difficult news and medical interventions that may be pursued by families.<sup>5,15</sup> But, overall, there is a lack of a formal PPC structure or training process necessitating clinicians to learn what works best through trial and error.<sup>13,25</sup>

## **2.2 CLASSIFICATION OF PERINATAL DEATH**

The National Center for Health Statistics (NCHS) collects all records of fetal and infant deaths occurring in the United States and its territories (District of Columbia, The Virgin Islands, American Samoa, Guam, the Northern Marianas, and Puerto Rico). Each state and territory collects its own annual vital statistics data and subsequently reports this to the NCHS. For statistical

purposes, the aggregate of the United States only applies to the 50 states and the District of Columbia. Guam, Puerto Rico, and the Virgin Islands are presented separately, and no data is included from the American Samoa or the Commonwealth of the Northern Mariana Islands. The NCHS compiles the data into yearly reports, which is then released as part of the Centers for Disease Control's (CDC) National Vital Statistics report.<sup>16</sup> By law, each state and territory is required to report fetal demise, issue birth certificates and death certificates for infants, and report infant demise.<sup>26</sup>

In order to understand the numbers presented in this project, it is necessary to have an understanding of how fetus, infant, and perinatal are subsequently defined. A fetus is an unborn human between the embryonic state and birth. This is more precisely defined as any pregnancy past 8 weeks gestation until term. An infant is a living human that is under 365 days of age.<sup>27</sup> The standard classification of fetal death is defined as death prior to the complete expulsion or extraction from its mother regardless of the pregnancy length, with death indicated by no evidence of life, such as breathing, a heartbeat, or voluntary movement, after he or she have been separated from the mother.<sup>12,14</sup> Fetal death encompasses miscarriage, spontaneous abortion, and stillbirth. The standard classification is used by all states and territories exempting Wisconsin and Puerto Rico, which do not stringently define fetal death.<sup>12</sup> An infant death is defined as a live birth followed by a brief survival period – a few hours to a few weeks – and then death within the first year of life. Again, life is defined as breathing, detection of a heartbeat, or voluntary movement after complete separation from the mother.<sup>27</sup>

Perinatal is a term that encompasses both fetal and infantile stages of development. Perinatal death is a statistical term used to express the number of fetal and infant deaths.<sup>27</sup> The way the definitions of fetal, infantile, and perinatal deaths are interpreted is critical to understanding

the number of perinatal deaths reported. There is little misinterpretation when it comes to defining infant death. States and territories have the ability to link data from birth certificates with data from death certificates to confirm reports of infant demise and to gather data on causes of death.<sup>12</sup> Recording fetal demise, however, is a more difficult process. The *1992 Revision of the Model State Vital Statistics Act and Regulations* recommends reporting a fetal death if products of conception are  $\geq 350$  grams or past 20 weeks gestation.<sup>28</sup> However, fetal death reporting varies greatly by state.<sup>16</sup> State standards for reporting fetal demise are generally based on gestational age and/or birthweight, which is outlined in **Table 1**.<sup>27,28</sup>

**Table 1. Fetal death reporting standards by state.**

States	Reporting Standard
AR, CO, GA, HI, NY, RI, VA	All products of human conception
AL, AK, CA, CT, FL, IL, IN, IA, ME, MD*, MN, NE, NV, NJ, NC, ND, OH, OK, OR, TX, UT, VT**, WA, WV, WY	$\geq 20$ weeks gestation
AZ, DE, ID, KY, LA, MA, MS, MO, MT, NH, SC, WI	$\geq 350$ grams or $\geq 20$ weeks gestation
MI	$\geq 400$ grams or $\geq 20$ weeks gestation
KS	$\geq 350$ grams
NM, SD, TN***	$\geq 500$ grams
PA	$\geq 16$ weeks gestation

\*MD will issue a fetal death certificate if gestation period is unknown, but fetal weight is  $\geq 500$  grams

\*\*VT will issue a fetal death certificate if gestational age is unknown but fetal weight is  $\geq 400$  grams

\*\*\*TN will file a fetal death report if no weight is recorded but gestational age is  $\geq 22$  weeks.

Understanding fetal death helps explain the strengths and weaknesses in the three most common definitions of perinatal death. Definitions are constructed by the American Academy of Pediatrics Standard Terminology for Fetal, Infant, and Perinatal Deaths.<sup>27</sup>

1. Infant deaths that occur at fewer than seven days old and fetal deaths with a stated or presumed period of gestation greater than 28 weeks.

2. Infant deaths that occur under 28 days of age and fetal deaths with a stated or presumed period of gestation greater than 20 weeks.
3. Infant deaths that occur at less than seven days of age and fetal demise with a stated or presumed period of gestation greater than 20 weeks.

The second and third definitions are the most commonly used when reporting perinatal statistics. The first definition is too restrictive because most fetal deaths occur before 28 weeks gestation. The latter two definitions technically incorporate neonatal deaths, not infant deaths. Neonates, which are a subcategory of infant, are defined as human children less than one month old. The second definition is the most inclusive, and most widely used, definition because it captures a period of time when fetal and infant deaths are more likely to occur.<sup>27</sup> These definitions have been generated primarily for statistical purposes, but it is likely that these numbers are underrepresented due to lack of a consensus definition. Additionally, infants that pass away within the first year of life are often included in perinatal demise statistics, meaning the above definitions do not capture the full extent of perinatal demise reported in the United States.<sup>12</sup>

There are other factors relating to underrepresentation of perinatal demise in the United States. Given the majority of states and territories do not report deaths prior to 20 weeks gestation, there is little data on miscarriages. The earlier the gestational age and lower the birthweight, the less likely a fetal death is to be reported.<sup>14</sup> The NCHS estimates there are greater than 1 million annual fetal losses in the United States, the majority of which occur before 20 weeks gestation.<sup>12</sup> About 500,000 women per year will be admitted to the emergency department due to vaginal bleeding during early pregnancy ( $\leq 20$  weeks gestation), and approximately half will miscarry.<sup>29</sup> In general, there is a large discrepancy between what is suspected and what is reported, which may be as a result of an expectant mother who does not realize she is pregnant, or a physician's desire

to avoid paperwork for early pregnancy loss in addition to the way fetal death is reported in the United States. However, there are other possible causes for underreporting. For example, a misunderstanding of live birth may occur, causing some to be classified as a stillbirth instead of an infant demise. Physicians may also avoid reporting premature infant death to avoid the process of issuing both a birth certificate and a death certificate.<sup>14</sup>

Per the 2013 National Vital Statistics Report on Fetal and Perinatal death, 47,041 perinatal deaths were reported. There were 23,595 fetal deaths in total and 23,446 infant deaths in total, indicating that fetal loss accounted for a slightly higher proportion (50.1%) of perinatal demise in 2013. Examining the total number of losses in greater detail, 25.2% of fetuses were lost between 20 – 27 weeks gestation, 24.9% of losses occurred at 28+ weeks gestation, 33.8% of infant losses happened before seven days of life, and 16.1% infants died after 28 days of life but before one year of life.<sup>12</sup>

Understanding the natural history of a condition and presenting medical information to families is vital to the genetic counseling process.<sup>18,20</sup> By developing a better understanding of perinatal death terminology genetic counselors will be more capable of referring families appropriately to PPC programs. For the purposes of this study, perinatal demise encompasses fetal deaths occurring after 20 weeks gestation and all infant deaths occurring before one year of life. This definition has been chosen because infants may pass away after the first month of life due to chromosome conditions, congenital malformations, and other conditions related to preterm birth and low birthweight. It should even be expected that more infants born with life-limiting and life-threatening conditions will live for longer periods of time as advances in medicine continue.<sup>21,30</sup>



## 2.3 STRUCTURE OF PERINATAL PALLIATIVE CARE PROGRAMS

Using the website [perinatalhospice.org](http://perinatalhospice.org), 268 PPC programs were identified throughout the United States. Programs are broken up into three groups: clinic-based (245), support only (22), and home-based (1). Greater than 70% of these programs are less than 10 years old.<sup>6</sup> Despite the rapid growth and development of these programs over the past decade, there is no clearly established structure that governs PPC practice.<sup>5,6</sup> This presents a problem for future establishment and development of programs, and also makes it difficult for clinicians to determine which programs offer patients the most benefit when in the distressing situation of planning care for a critically ill fetus or infant.<sup>5</sup>

Most programs offer pre- and postnatal care to the child. Several programs assist in the creation of a birth plan, maternal postpartum care, and memory making.<sup>4,6</sup> These broader categories encompass a variety of services, which tend to fall under one or more of the eight domains of palliative care identified in a recent study assessing PPC practices within the United States: 1) utilization of an interdisciplinary team, 2) physical symptom management, 3) psychosocial care, 4) family support, 5) spiritual care, 6) acknowledgement of cultural differences in provision of care, 7) end-of-life care, and 8) legal and ethical aspects of care.<sup>5</sup> While not all PPC programs have genetic counselors as part of the clinical care team, genetic counselors can still offer families support and psychosocial services by referring parents to a PPC program.<sup>19,20</sup>

## **2.4 REFERRING FAMILIES TO PERINATAL PALLIATIVE CARE**

Families may be referred to a PPC program either prenatally or postnatally. In the prenatal period, an abnormality may be identified by ultrasound, or genetic testing may identify a life-limiting or life-threatening condition.<sup>10,18</sup> In the postnatal stage, it may become apparent that the child has several health conditions that may alter his or her life course.<sup>3</sup> One study asked parents to identify how they learned of PPC services, revealing six common sources: 1) Direct referral from obstetrician or maternal fetal medicine (97%), 2) Genetic counselor (73%), 3) Internet (61%), 4) Nurse (60%), 5) Social worker (60%), and 6) other (39%).

At this time, there are no established criteria to help clinicians determine which families would most benefit from PPC services.<sup>31</sup> Services may be requested when complicated medical issues are identified prenatally or shortly after birth, or when physicians feel that the needs of the patient are beyond their scope of practice.<sup>10,18,31,32</sup> There is evidence that when a PPC team becomes involved with families of children with a life-limiting or life-threatening condition, they provide beneficial support to the family, improve clinical care team communication, and facilitate the decision making process.<sup>6,31</sup>

### **2.4.1 State Laws**

In some states, healthcare providers are legally mandated to inform women carrying a fetus with a lethal anomaly about perinatal hospice and/or PPC services. For instance, the Indiana General Assembly passed a statute in 2016 requiring the Indiana State Department of Health (ISDH) to develop materials about perinatal services for patients, and two forms certifying a provider has given them these resources.<sup>33-35</sup> The ISDH created a Perinatal Hospice Brochure and

a list of perinatal hospice providers in Indiana.<sup>35</sup> The first certification form is intended for use at the time of a lethal prenatal diagnosis and documents the provision of the aforementioned resources.<sup>33</sup> The second form is intended for use at the time a woman chooses to terminate a pregnancy of a fetus with a lethal prenatal diagnosis. It documents that the physician performing the abortion has verbally communicated with them about perinatal hospice, provided the ISDH-created resources, and that the woman is choosing to terminate rather than continue the pregnancy with perinatal hospice.<sup>34</sup> While it is important to increase PPC awareness and referrals, it should be done in a way that does not interfere with medical practice or appropriate patient-provider communication.

The American College of Obstetricians and Gynecologists published a committee opinion advocating for increased abortion access. It recommends cessation and repeal of legislation that creates barriers to access and interferes with the patient-provider relationship or medical practice. Some examples of this type of legislation are laws requiring mandatory counseling and delays, and legislation classified as TRAP (Targeted Regulations of Abortion Provider) laws. TRAP laws impose additional and unnecessary medical requirements upon abortion providers, such as mandated counseling. This type of approach may not be grounded in an appropriate ethical manner for patient-provider communication. These measures often draw out the termination process, making it more difficult for providers to perform abortions and for women to pursue an abortion.<sup>36</sup>

#### **2.4.2 Rural Outreach**

Based on location data from [www.perinatalhospice.org](http://www.perinatalhospice.org), most PPC programs are located in urban areas.<sup>37</sup> It may be difficult for families to find hospice programs willing to work with them at their home or for them to find hospital-based support depending on where they live.<sup>38,39</sup> Adult

hospice programs may not feel equipped to handle a prenatal or pediatric patient.<sup>40</sup> Due to financial distress, many rural hospitals are paring down, or completely discontinuing, inpatient services. Given the relatively low numbers of those using PPC programs in the U.S., it may not be cost effective to establish such a program in a rural community.<sup>38,40</sup> A proposed solution to reducing rural healthcare costs is telemedicine.<sup>40,41</sup> A study conducted in North Dakota found telepharmacy services were a sustainable way for rural communities to maintain contact with a pharmacist if no local practitioners were available.<sup>41</sup>

In a JAMA opinion piece, one physician, Meghann Weaver, demonstrates the utility of telehealth in pediatric hospice by sharing her experience with one family who chose to have their daughter spend her last days at home instead of in a hospital located four hours away from their home. While there were two hospice programs offering home services in their area, neither program was comfortable taking on a pediatric patient. Weaver found this response to be inadequate for her patient's needs. Over time, she was able to come into contact with a family practice doctor who had experience with geriatric hospice patients, a school nurse interested in helping the family, and the family's pastor. Using an Internet connection to establish interdisciplinary team meetings, Weaver was able to support the palliative care of her patient.<sup>39</sup> Weaver's story indicates that establishing collaborative relationships between urban palliative care programs and rural health care practitioners can have a positive impact on patients. Her experience suggests that something similar could be carried out with PPC.

## 2.5 CARE PROVIDED IN PERINATAL PALLIATIVE CARE PROGRAMS

Though the number of PPC programs in the United States has increased rapidly, there is no standardized model of care.<sup>6</sup> For those experiencing perinatal loss, the grieving process may be complicated and intense.<sup>3,15,42</sup> There is evidence to suggest that the acute grief associated with poor prenatal findings, or miscarriage, is similar to the grief experienced when a living child dies unexpectedly. Instead of an outright death of the child, though, it is instead death of a “wished-for” child.<sup>18</sup> The intensity of emotions felt by parents and family members receiving this news necessitates a plan of action that will take care of the fetus/infant’s medical needs as well as the family’s emotional needs.

Historically, intense perinatal care focused on prolonging the life of a child with complex care needs with less attention focused on their comfort.<sup>7,8,43</sup> In other cases, some parents report physicians advising them to terminate pregnancy or forgo medical interventions due to poor prognosis of their child’s condition.<sup>21</sup> As PPC programs have expanded over the past few decades, parents have become more involved in making healthcare decisions for their child. Many programs are moving toward a family-centered approach, where the parents are considered to be part of the clinical care team.<sup>32,44</sup> A family-centered approach allows the family to become collaborators<sup>44</sup> and considers the belief system of the family, their goals, and incorporates them into the decision making process. This approach may also involve educating parents about their child’s condition and teaching them how to perform aspects of their child’s caretaking while they are hospitalized.<sup>30,45</sup> These models have been shown to not only benefit the family members, but also the staff working in the programs. Families feel more competent in their roles and abilities to parent their child by providing some level of care for the infant.<sup>45</sup> Staff members feel more

confident in the family's ability to manage the situation and enjoy shifting out of the role from full-time caregiver to educator.<sup>45,46</sup>

In addition to providing medical care to the fetus or infant, and emotional support to the family, PPC programs help facilitate the decision-making process, coordinate care, advocate for the family, and improve communication between multiple care providers.

Finally, there is also a need to balance family participation and medical efficiency. Some physicians report avoiding the consultation of palliative care teams because the physicians felt the team's expectations were cumbersome, they hindered the medical decision-making process, or the team was placing the family's needs over the medical needs of the child.<sup>32</sup>

### **2.5.1 Medical Care for the Fetus or Infant**

When determining medical interventions in PPC programs, the family's understanding of their child's diagnosis and prognosis should be explored.<sup>3</sup> It is important that their belief system, culture, and religion be taken into consideration because families will attempt to process new and upsetting information in a framework that already has meaning to them.<sup>18</sup> While specific medical interventions may alter based on a family's beliefs, many parents express common goals for children in PPC programs.<sup>18,21</sup> Those include meeting their child alive, spending time as a family, and taking their child home. Many parents express a desire to spend as much time as possible with their child, but recognize that medical interventions may be burdensome or painful for the child and the rest of the family. Families may desire to change their child's care plan as his or her condition progresses.<sup>21</sup> Clinical care teams respond to these changes by regularly checking in with family members to reassess their understanding of the child's condition and how that fits into their

goals for his or her life. This allows providers to change management based on newly expressed goals.<sup>3</sup>

Physicians may have difficulty ascertaining when a PPC program should be consulted for care of a fetus or infant with a complex medical condition. Some neonatal or pediatric critical care physicians report only consulting their hospital's palliative care team when they felt the family had not chosen the "best course of action." Others avoided consulting the team if the child's condition appeared to be imminent death because they were more certain about the care path they would pursue for the child. Some of these critical care physicians decide to wait for a child's condition to become chronic (i.e. spending multiple weeks in an intensive care unit) before consulting a PPC program because they are less equipped to and more stressed by caring for patients with chronic conditions.<sup>32</sup>

Genetic testing may also be utilized by the clinical care team to provide information to the family and to guide care-making decisions. The timing of diagnosis may affect medical interventions chosen by parents and healthcare providers. In a survey of parents of children affected by either Trisomy 13 or Trisomy 18, those receiving a prenatal diagnosis were more likely to describe palliative care interventions as skin-to-skin contact – the act of placing a newborn child immediately on the mother's or father's chest to increase bonding – and medication for pain management. Parents who received a postnatal diagnosis were more likely to describe several medical interventions including use of oxygen, intravenous fluids, tube feedings, and surgeries to repair birth defects.<sup>21</sup>

## 2.5.2 Supportive Care for the Family

Leaving the hospital without a baby after a birth is an emotional experience for families.<sup>2,15</sup> The majority of PPC programs offer some form of emotional support for infants and families receiving their care, as well as follow up through phone calls and condolence cards. Families may be referred to support groups or counselors, connected with parents who have had similar experiences, or assisted in arranging religious services. Programs may also provide further resources on grief and grief management.<sup>5</sup> Many programs focus on creating memories with the child through creation of a birth plan or creation of mementos.<sup>2</sup>

Mementos allow for the creation of tangible, and meaningful, memories of parents spending time with their children. Many programs offer a wide array of mementos to parents including clothing worn by their child, pictures of their child, handprints, footprints, locks of hair, and certificates from spiritual ceremonies. These mementos help facilitate the grieving process for parents, as well as reminding them of their parental roles in their child's brief life.<sup>2</sup>

For children with complex care needs who may stay in the hospital for a longer period of time, palliative care teams are also able to offer support through gathering and dispensing information.<sup>32,44</sup> It is important to support parents through this stressful period of time. Studies show that parents with babies in the NICU are at increased risk for postpartum depression, anxiety, and post-traumatic stress disorder. Parents may also struggle to feel in control of their child's care. Both factors can impact their ability to bond with their child after birth.<sup>44,47,48</sup> Possible solutions may include referral to parent support groups, dedicated, on-unit space for parents that takes them away from the bedside, and well established relationships with clinicians.<sup>44</sup>

Intensivists report feeling ill equipped to handle children with chronic health conditions because they either do not have enough time to explain the full extent of the child's medical



problems and understand the family's needs, or they may not have received appropriate training in school to integrate the needs of the family into their care methods.<sup>25,32</sup> Some describe an intensifying relationship as a patient's condition progresses, which may negatively impact communication between clinicians and family members.<sup>25</sup> One way to establish open lines of communication is through primary nursing, a practice in which a nurse commits to caring for a particular child every time he or she is on shift. This practice can secure bonds between the family and NICU staff.<sup>44</sup> Another way to increase communication and parental engagement is by encouraging parents to assert themselves in the clinical space. This may include participating in rounds or learning how to perform a specific care task for their child.<sup>44,45</sup> For parents that struggle to voice their concerns or are unsure of how to become more involved, palliative care teams are a vital resource. PPC practitioners can increase clinician-family communication by helping families frame specific care questions, advocating for the family's needs, and organizing interdisciplinary care conferences that the family may attend.<sup>44</sup>

### **2.5.3 Facilitating the Decision-Making Process and Bridging Communication Gaps**

One way that PPC programs serve both the clinical care team and family members is improving clinician-clinician and clinician-family communication.<sup>32</sup> This may happen on an individual basis or through interdisciplinary discussions that may or may not involve the family.<sup>49</sup> When a child with complex care needs is staying at a hospital, they are usually attended to by a large interdisciplinary care team working in parallel.<sup>45</sup> It can be confusing for the family when specialists (i.e. pulmonologist, cardiologist, hematologist, etc.) do not approach the child's condition holistically because they are focused on the specific organ of their discipline.<sup>32</sup> This may result in diminished trust of the care team, particularly if one specialist is optimistic while another

is pessimistic about varying outcomes, because it gives the appearance that there is little communication about their child amongst the team.<sup>44</sup>

Palliative care teams are often able to spend extended time with families, allowing them to piece together information from a variety of specialties to better educate the family. This ultimately gives the family a better understanding of their child's prognosis and subsequent course of treatment. Some programs employ visual aids when educating families on recommended medical interventions, which helps parents make informed decisions throughout their child's care plan.<sup>6,44</sup> At times, including parents in the decision-making process is done deliberately to grant them a sense of control over the current situation. When parents are better able to participate in their child's care, they feel respected, more confident in the clinical care team, and empowered.<sup>44,45</sup>

In some cases, the care team and family may not agree about a child's care plan. Most often it manifests when a clinician wishes to withdraw support while the family wants support to continue. In these cases, it may be necessary to form a special committee to advise clinicians and family members,<sup>49,50</sup> which is something the PPC team may coordinate. The University Hospital of Zurich proposes a model that carefully selects members of this group. They suggest a committee consisting of an inner circle, outer circle, and independent moderator. The inner circle consists of clinicians directly caring for the child. This group is responsible for making the final decision, but the head of the clinic is held legally responsible for the decision made. The outer circle is comprised of medical experts, nurses, medical students, etc. not involved in care of the child but who may advise the inner circle. The discussion is to be led by a moderator uninvolved in the care of the child. While the values and wishes of the parents are brought into consideration throughout the process, this model excludes them from the initial discussion. They are fully informed in a secure setting by the clinician with whom they have the best-established relationship. Should they

disagree with the decision of the committee, a second discussion involving the parents is often scheduled. This structure was chosen to protect both clinicians and family members. While parents should be allowed to make decisions whenever possible, they may feel overwhelmed in end-of-life decision making, and clinicians directly involved in the child's care may feel stripped from their role if an outside committee were to make the decision.<sup>49</sup> An ethical paper written by Aberdeen University, Edinburgh and the Institute of Medical Ethics, Edinburgh warns against the use of committee decision making. Instead, the group recommends that these committees should only work in an advisory capacity, with families making the ultimate decision as they are the ones who bear the consequences of prolonging life or withdrawing care from their child.<sup>50</sup>

## **2.6 OUTCOMES OF PERINATAL PALLIATIVE CARE**

### **2.6.1 Impact on Fetuses and Infants**

In a recent study, one hospital compared the survival outcomes of children who received PPC after being born with a severe congenital anomaly with those who did not receive PPC services. A total of 24 fetuses and infants were included in each group. Five fetuses were lost in each group before delivery, and 16 were lost in each group in the infant period, with the majority of losses occurring by two days of age. In the group receiving PPC, three infants survived to discharge and one passed away after discharge. Similarly, three infants survived to discharge in the group that did not receive PPC, and none passed away following discharge. The outcomes in both groups were similar, but the infants receiving PPC were more likely to have an established comfort care plan and shorter time to in-hospital death than those who did not receive PPC

services.<sup>31</sup> Another study found that families who received a prenatal diagnosis were more likely to select comfort measures over intense medical intervention. Additionally, these children were more likely to pass away sooner than children diagnosed postnatally.<sup>51</sup>

## **2.6.2 Impact on Families**

While PPC intervention may not have a significant impact on length of in-hospital stay to death or discharge, it is thought to provide a benefit to families by establishing a plan of care early on – either prenatally or postnatally – and making sure a family’s goals align with the care provided at the hospital. It also helps to facilitate future decision-making processes and provides families with cohesive care throughout the duration of pregnancy and treatment.<sup>31</sup>

Parents may feel hopeless when they are faced with end-of-life care decisions for their child.<sup>52</sup> The type of stress incurred by such a situation may have an impact on both their physical and mental health.<sup>30</sup> The goal of PPC programs is to allow parents to maximize the limited amount of time they have with their child in order to facilitate bonding and loving their child.<sup>45</sup> Yet, few studies have been conducted to see if this goal is achieved or to understand the impact PPC practices have on families. This is complicated because there is no cohesive model of care, and thus no universal models to assess the impact of PPC in care.<sup>4</sup> Some studies postulate that PPC programs provide benefit to the patient’s family by reducing the amount of suffering they experience in both duration and intensity.<sup>9,45,49</sup>

### **2.6.3 Impact on Clinicians**

Several studies report that clinicians caring for a critically ill child find the work to be quite challenging, but also rewarding as many recognize the importance and impact their care has on families experiencing perinatal loss.<sup>19,25,32,45,46</sup> There are several challenging aspects that clinicians face on a regular basis, such as having a multifaceted role, operating within an interdisciplinary team, seeing little improvement in a severely ill child, and balancing life stressors outside of work.<sup>25,46</sup> In a survey of clinicians taking care of children with chronic critical illness, there were mixed results as to whether the challenges posed by these patients increased the risk for caretaker burnout. Some recognized burnout within themselves and their colleagues, but reported that they did not think it would be increased within this population of clinicians as opposed to other specialties. Interestingly, a few felt burnout would be lower in this group because helping children and families in a vulnerable situation can be very meaningful.<sup>25</sup> This is supported by another study where clinicians described rewarding aspects as making a difference for the child and their family members, determining the best courses of care, and being valued by other team members and families.<sup>46</sup> While some hospitals have established procedures to mitigate burnout, such as case debriefings, enforced time off, and colleague support, many clinicians reported struggling to engage in these self-care activities.<sup>25</sup>

## **2.7 GENETIC COUNSELORS IN PERINATAL PALLIATIVE CARE**

One of the way that genetic counselors offer support to families affected by poor perinatal outcomes is through provision of resources. This may be in the form of supplementary information

about their child's condition, contact information for support groups or families who have had similar experiences, and referrals to palliative care services.<sup>20</sup> Some groups, such as the American Academy of Nursing and the NSGC, support an increase in genetic counselor collaboration with PPC programs through referrals, coordination of care, or delivery of medical information, but there is little in the literature about which families they should refer to programs or what a collaborative role would entail.<sup>11,19</sup> With the exception of one survey about genetic counselor beliefs on PPC, the majority of genetic counseling reports related to perinatal demise consisted primarily of case studies.<sup>18</sup> At this time, no studies were identified that have been conducted to assess PPC education in ACGC accredited training programs.

### **2.7.1 Genetic Counselor Attitudes Toward Perinatal Palliative Care**

To date, only one study has been found assessing genetic counselor attitudes toward PPC. This study sought to understand and characterize the perceptions and barriers counselors encountered when referring families to these services. Overall, genetic counselors are receptive to PPC programs and its practices. The findings reported in this study supported the idea that genetic counselors have the capabilities to serve on PPC multidisciplinary teams. This was determined by demonstrating the application of the ACGC practice-based competencies to PPC practices and creating a framework for genetic counselors' roles guided by the ethical principles of autonomy, beneficence, nonmaleficence, justice, dignity, and truthfulness. There was also a discussion of genetic counselors competency with informed consent and how that is an integral part of PPC as well.

Prior to participation in the study 46.2% of participants reported being very familiar with PPC, 42.4% were somewhat familiar with PPC, and 11.4% were very-to-moderately unfamiliar

with PPC. Similar numbers were reported in response to questions about personal comfort with PPC discussion, and with referral comfort. Regarding personal comfort, ~44% were very comfortable, ~53% were somewhat comfortable, and ~2% were uncomfortable. With respect to referral comfort ~48% reported they were very comfortable, ~50% were somewhat comfortable, and ~1% were uncomfortable.<sup>19</sup> In this context, comfort refers to one's ability to operate in an "anxiety-neutral condition, using a limited set of behaviors to deliver a steady level of performance, usually without a sense of risk."<sup>53</sup>

Using the Perinatal Palliative Care Perceptions and Barriers Scale Incident created by Wool,<sup>54</sup> further analysis was done to investigate factors influencing the variability in counselor survey response. Five main variables were defined: personal comfort (24.1%), perceptions (12.4%), barriers (8.9%), years in clinical practice (5.3%), and prior familiarity (0.9%). Two recommendations were made within the study. First, more experienced counselors should act as mentors and role models for less experienced counselors in these situations, because those with more experience reported greater levels of personal and referral comfort. The second recommendation is further education of the 11.4% of genetic counselors who were very-to-moderately unfamiliar with PPC to increase their familiarity and comfort with PPC.<sup>19</sup>

### **2.7.2 Strategies for Genetic Counselors**

There is no literature that defines the role of a genetic counselor in a PPC setting. There is literature discussing genetic counselor interaction with parents after receiving an abnormal prenatal diagnosis, and surveys have been conducted that highlight what genetic counselors do to make these parents feel supported.

Interventions genetic counselors may use with families experiencing perinatal loss are derived from crisis intervention strategies. During a genetic counseling appointment, a counselor may ask questions to understand the factors influencing a family's decision-making process. Factors include support systems, handling of previous crises, beliefs about illness and disability, understanding of their child's current illness, information processing, as well as religious, cultural, educational, and intellectual frameworks.<sup>3,18</sup> After assessing, the genetic counselor forms a plan to help the family process information, facilitate grieving and/or decision-making, and discuss future planning.<sup>18</sup> One survey asked parents receiving a prenatal diagnosis of Trisomy 13/18 to indicate what genetic counselors did to make them feel supported after they decided to continue their pregnancy. Parents felt supported when genetic counselors verbalized support, delivered unbiased information, and provided.<sup>20</sup> This study supports Wool's idea that the supportive skills genetic counselors enhance during their training translates well into PPC.<sup>19</sup>

## **2.8 INCORPORATING EDUCATIONAL MATERIAL INTO CLINICAL TRAINING PROGRAMS**

Research suggests there is a need for clinicians from multiple disciplines to improve their empathetic response to families experiencing a perinatal loss, expand their knowledge of PPC practices, and increase knowledge of programs in their area so they can direct more families to PPC.<sup>15,19,20,55</sup> The literature also suggests that when clinicians are unable to provide adequate information, discharge instructions, or other forms of support to families experiencing a perinatal loss, parents may feel minimized or abandoned by their healthcare providers.<sup>15,21</sup>



### **2.8.1 Incorporating Perinatal Palliative Care Education into Physician Training**

There is recognition in the medical community that physicians caring for critically ill patients should have basic palliative care skills, which include competent pain and symptom management, end-of-life care, effective communication, the ability to facilitate decision-making, and psychological and spiritual awareness when delivering care.<sup>55</sup> The American Academy of Hospice and Palliative Medicine (AAHPM) offers physicians the opportunity to become certified in hospice and palliative medicine. To date the AAHPM has certified 7,606 physicians in hospice and palliative medicine. Since 2014, certification requires all participants to complete a 12-month, accredited hospice and palliative medicine fellowship.<sup>56</sup> Understandably, it is not possible for every physician to take on this fellowship, as it is lengthy and may not be applicable to their specialty.

Studies examining pediatric resident and fellow training shows they have few opportunities to develop palliative care skills.<sup>55</sup> There is also some evidence that neonatal and pediatric critical care physicians, i.e., intensivists, do not receive enough training to properly “hear” family members’ needs when they have a child going through intensive care for long periods of time.<sup>21</sup> One study, conducted by Brock, examined the effectiveness of didactic training and simulation training when teaching two cohorts of pediatric residents and fellows about palliative care. The cohort that was assigned to simulation-based training was shown to have more competency in palliative care skills than the cohort assigned to didactic learning.<sup>55</sup> Lack of hands-on experience often necessitates that many physicians, residents, and fellows learn through trial and error.<sup>13</sup>

## **2.8.2 Incorporating Perinatal Palliative Care Education into Nurse Training**

Nurses also report lack of palliative care training.<sup>15,45</sup> One study, which examined literature about pregnancy loss management in emergency departments, noted that nurses have an important role in providing care for women experiencing perinatal loss. The study found that there is a wealth of clinical recommendations for an array of perinatal losses – ectopic pregnancy, miscarriage, stillbirth, infant demise – but fewer recommendations related to communication of bad news and psychosocial management. Results also showed that 50% of nurses felt that bereavement counseling should begin in the emergency department, but only 10% felt competent in their bereavement counseling skills. The study's conclusion recommended that nurses should be adequately educated about psychosocial support and counseling in addition to their broad knowledge of medical management for these women.<sup>15</sup> Yet, searching the literature reveals a paucity of information regarding the content and structure of palliative care education in nursing programs. Like physicians, nurses have the opportunity to learn more about palliative care and hospice medicine. Nurses can learn about palliative care through the End-of-Life Nursing Education Consortium classes and seminar offered by the American Association of Colleges of Nursing. Similar to the previously mentioned fellowship offered to physicians, nurses choose whether or not they take these classes or specialize in palliative medicine.<sup>57</sup>

## **2.8.3 Incorporating Perinatal Palliative Care Education into Genetic Counselor Training**

Although a few studies have been conducted to evaluate simulation and didactic work experiences on residents', fellows', and nurses' preparedness to handle PPC situations, no similar studies have been conducted within genetic counseling programs.<sup>55</sup> Additionally, there is no PPC

educational resource material specific to genetic counselors described in the literature. Therefore, it is important to examine how PPC-related materials have been incorporated into genetic counseling programs to gain a better understanding of how new supplementary material may be integrated into a preexisting curriculum. In a study assessing the integration of psychiatric genetic counseling practices into programs, recent graduates reported a desire to have more instruction, with only half reporting they were comfortable counseling someone for a mental illness at the time of graduation. The most effective methods of teaching appeared to be hands-on experience in the clinic or through roleplays in class. Personal experience with mental illness also had a positive impact on the willingness and comfortability of genetic counseling students and graduates to discuss psychiatric illness in a session.<sup>58</sup> Therefore, educational resource materials could incorporate role playing scenarios and hands-on activities to help genetic counselors gain a better understanding of what families may be feeling when experiencing perinatal loss.

### **3.0 MANUSCRIPT**

#### **3.1 BACKGROUND**

Perinatal palliative care (PPC) is a specialized clinical program that seeks to maximize the time that families spend with their child after a life-limiting or life-threatening diagnosis has been given.<sup>1,3</sup> Most PPC programs pursue this goal through a plan of care that accommodates the medical and comfort needs of the critically ill child, as well as psychosocial support of family members through the creation of mementos and grief counseling.<sup>2,3</sup> Adult hospice care is important enough to be considered a global public health priority,<sup>4</sup> but perinatal services have yet to receive the same level of attention. This may be due to the relative newness of the majority of programs and lack of standardized practices within programs.<sup>5,6</sup>

PPC first appeared in the literature in the 1980s, when physicians began advocating for infant entitlement to a “natural death,”<sup>7,8</sup> which was difficult for those used to providing intensive care to critically ill infants in order to prolong life.<sup>4,3</sup> Physicians supporting infant death argued that aggressive neonatal care to extend lifespan<sup>7,43</sup> results in greater suffering for the infant and their family by increasing the length of suffering before demise or causing a child to live with complex care needs.<sup>9</sup> The majority of perinatal deaths are attributed to major malformations, chromosome conditions, and unspecified conditions relating to low birthweight or preterm birth.<sup>12,16</sup> Over the past few decades, advances in perinatal screening have increased detection of congenital malformations and chromosome conditions.<sup>8</sup> Other perinatal advances have contributed to a 76.16% decrease in fetal mortality since 1942,<sup>12</sup> and have given infants affected by preterm birth, low birthweight, severe malformation, or chromosome conditions longer periods of time to

survive.<sup>9</sup> Yet, not all such affected fetuses and infants are expected to survive given the severity of their ailments.<sup>13</sup> In 2013, 23,595 fetal deaths and 23,446 infant deaths were reported in the United States.<sup>12</sup> Therefore it is important for the ~47,000 families to learn about services provided by PPC so they are better supported.

Couples with an affected fetus or infant may be referred to genetic counseling. Genetic counselors are beneficial to families because they provide information about conditions, psychosocial support, and additional resources to help facilitate the decision-making process.<sup>17</sup> In scenarios where a fetus or infant has a life-limiting or life-threatening condition, PPC should be offered alongside intensive medical intervention and termination of pregnancy.<sup>11,13</sup> Genetic counselors possess several skills that allow them to efficiently coordinate palliative services.<sup>19,20</sup> As more fetuses and infants with critical care needs survive,<sup>15,21</sup> there is a greater likelihood that genetic counselors will come in contact with families who have suffered a perinatal loss. PPC should be something counselors are comfortable discussing with clients. While few studies have investigated the effects PPC has on parental grief following an abnormal perinatal diagnosis, there is some evidence to suggest that PPC reduces the degree of suffering they experience.<sup>45,49</sup> Given the number of people who could be referred to PPC and the potential benefits these services provide, genetic counselors should have adequate knowledge to refer families to these programs.

A survey by Wool and Dudek found that genetic counselors are generally aware of PPC services, but there is evidence that there needs to be more PPC education for genetic counselors to increase their comfort with referring families to these services. Survey results showed 11.4% of counselors were “moderately” to “very” unfamiliar with PPC practice prior to the study, 2% were personally uncomfortable with PPC knowledge, and 1% had very little referral comfort.<sup>19</sup> When compared to a similar study that assessed pediatric residents and fellows on knowledge, comfort,

and competency in pediatric palliative care, almost all participants reported being very unfamiliar with palliative care services.<sup>59</sup> This suggests a need for increased PPC training within a variety of healthcare professional training.

According to Wool, genetic counselors, personal comfort, prior perceptions of PPC, perceived barriers in referral, years of experience, and prior familiarity had a significant impact on referral ability. These five factors accounted for 24.1%, 12.4%, 8.9%, 5.3%, and 0.9% of variability in referral rates respectively. Those with fewer than 5 years of experience in the profession appeared to have a lower personal comfort rate than those who had greater than 5 years of experience. To increase genetic counselor familiarity with and referral ability to PPC, Wool and Dudek suggest trainees receive more education about PPC. Given that 46.2% reported having 5 years of experience or less in Wool and Dudek's survey,<sup>19</sup> this current study focuses on increasing the number of educational materials about PPC for Accreditation Council for Genetic Counseling (ACGC) accredited genetic counseling training programs.

Currently, no studies have investigated the educational content of PPC offered through genetic counseling training programs. This study ascertained current information on the amount and structure of content related to PPC in ACGC accredited genetic counseling programs, as well as any gaps in knowledge or barriers to education that may exist. The following were the specific aims of the study:

1. Develop a Qualtrics study to administer to ACGC accredited genetic counseling programs to determine if PPC education is integrated into program curricula, clinical training, and supplementary activities.
2. Use the results of the administered survey to determine if there are any gaps in knowledge of or existing barriers to educating students about PPC.

3. Develop educational resource materials to be accessed by genetic counseling programs and their students.

Ultimately, this study hopes to increase genetic counselors' referral ability to PPC through the creation of educational resource materials that will be shared with genetic counseling training programs.

## **3.2 MATERIALS AND METHODS**

### **3.2.1 Survey Design**

Both the study and survey (ID: PRO17090536) were approved by the Institutional Review Board of the University of Pittsburgh (**Appendix C**). The survey was created in Qualtrics. A copy of the survey is included in **Appendix A**. The survey consisted of 30 questions. It was designed to elicit responses from program leadership of ACGC accredited genetic counseling training programs regarding what is currently taught about PPC in their programs, and what, if any, educational resource materials would be beneficial to them. More specifically, the survey consisted of four main sections: PPC material in coursework (including format and content of coursework), PPC exposure in clinical rotations and/or supplementary activities, desire of PPC educational resource material (including format and content of new material), and demographics. Skip logic was used throughout the survey, so no recipient was required to answer all 30 questions in the survey. However, they were requested to answer every question presented to them. Some of the questions included a free text option so program leadership could give a more precise explanation if desired.

### **3.2.2 Study Participants**

The participant population consisted of program leadership of ACGC accredited Genetic Counseling Programs. Program leadership, as established by the ACGC, includes the following titles: Program Director, Co-Director, Assistant or Associate Director, Clinical Practicum or Fieldwork Director. Program leadership was contacted via the Association of Genetic Counseling Program Directors' (AGCPD) listserv. The AGCPD listserv includes program leadership from accredited programs as well as program leadership from programs in development. The email sent on the AGCPD listserv described the study's intent and requested that only program leadership from accredited programs respond (**Appendix A**). An anonymized survey link was included in the email. At the time the survey was sent, January 2018, there were 41 total ACGC accredited programs in the United States and Canada. Only one response was requested of each program, and a reminder email was sent to program leadership two weeks following the initial request. In total, 21 (51.2%) survey responses were received. Four partial responses were recorded, giving an overall completion rate of 85.7%.

### **3.2.3 Data Analysis**

Data collected from the survey was analyzed using descriptive statistics. Participants were required to answer every question presented to them within the survey. Because skip logic was used throughout the survey, not every respondent was required to answer each question.



### **3.2.4 Creation of Educational Resource Material**

Using the results of the survey in conjunction with the literature, the top three responses for educational resource materials were made. There were four selections indicated as the third-highest response, so in total, six educational resources were desired by respondents. A subsequent question asked respondents to select what topics they would like covered within resource material, which was taken into consideration when developing materials.

Using quotes and examples from the literature, clinical case examples and role-playing scenarios were generated to help students better understand the benefits of PPC and how to discuss palliative care with families experiencing perinatal loss. An annotated bibliography was created to encompass three of the selections in list form. Sections include: 1) peer-reviewed articles for educational purposes and to start discussions about PPC, 2) books and videos illustrating the need for PPC, and 3) websites providing resources for faculty and students to learn more about PPC. A PowerPoint presentation covering topics desired by respondents was also generated.

Additionally, a map showing all 268 PPC programs in the United States, identified from [www.perinatalhospice.org](http://www.perinatalhospice.org), was created for a related project.<sup>37</sup> The map is a resource for clinicians and patients to identify local PPC resources. This will be made available for program use along with materials generated by this study.

## **3.3 RESULTS**

The survey asked programs about PPC content in their curricular, clinical, and supplementary activities. Overall results show that 85% of respondents indicated they have PPC

content contained within regular coursework, 67% indicated students were exposed to PPC in clinical rotations, and 67% indicated students are exposed to PPC through supplementary activities. Respondents were also asked if their program would benefit from the creation of educational resource materials, and 90% of respondents indicated that new material would be beneficial to their program.

### 3.3.1 Participants

There was a 21/41 (51.2%) response rate to the survey, of which, 17/21 (81%) respondents fully completed the survey. The majority of responding programs have been in existence  $\geq 10$  years (64.7%). The other programs were in existence for 0 – 3 years (23.5%) or 4 – 9 years (11.8%). Additionally, 41.2% of programs indicated they are located in NSGC Region 4 and 35.3% are located in NSGC Region 2. These two regions cover the majority of the Midwest. No responses were received from NSGC Region 6. Response rate by region is detailed in **Table 2**. Therefore, this survey is most representative of states that have been in existence for greater than 10 years in the Midwest.

**Table 2. Response rate by region.**

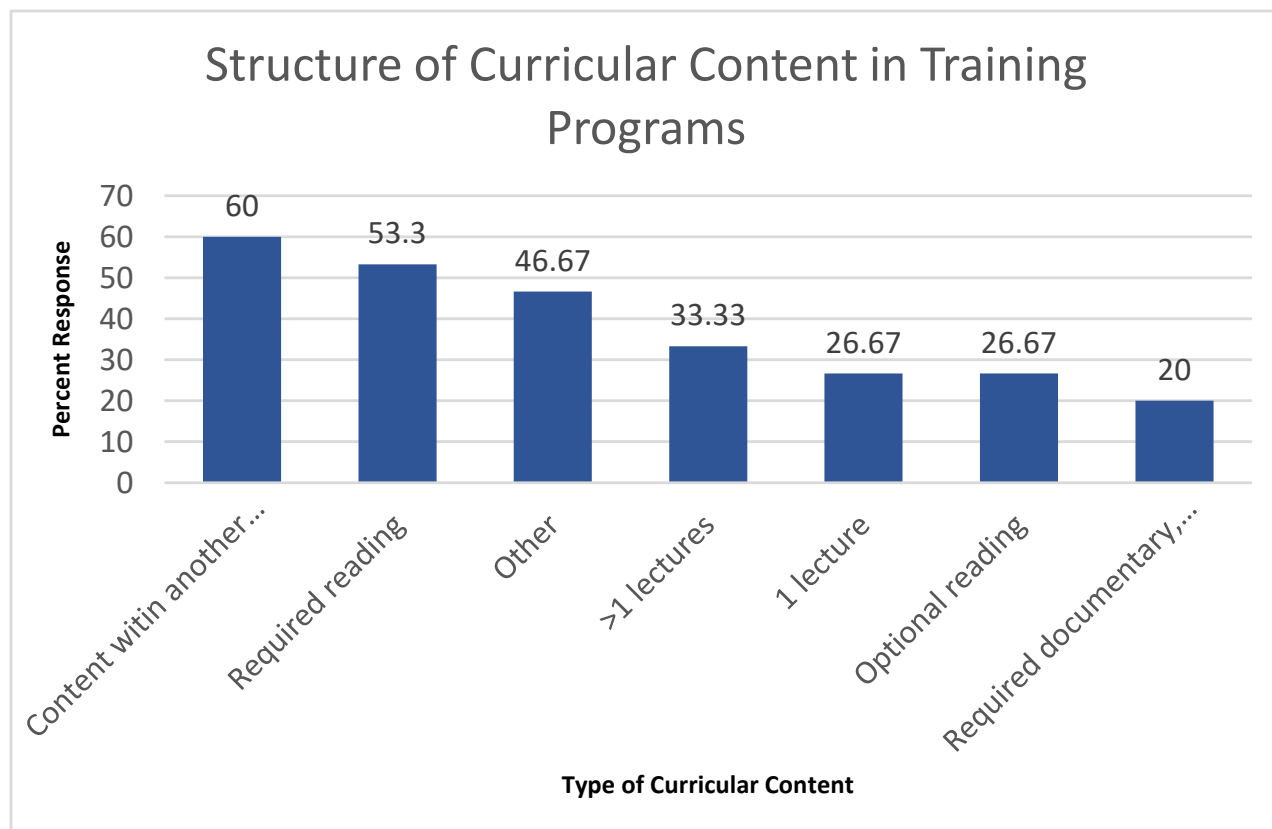
<b>NSGC Region</b>	<b>Percent Response Rate</b>
Region 1 (CT, MA, ME, NH, RI, VT, CN Maritime Provinces)	5.9%
Region 2 (DC, DE, MD, NJ, NY, PA, VA, WV, PR, VI, Quebec)	35.3%
Region 3 (AL, FL, GA, KY, LA, MS, NC, SC, TN)	11.8%
Region 4 (AR, IA, IL, IN, KS, MI, MN, MO, ND, NE, OH, OK, SD, WI, Ontario)	41.2%
Region 5 (AZ, CO, MT, NM, TX, UT, WY, Alberta, Manitoba, Sask)	5.9%
Region 6 (AK, CA, HI, ID, NV, OR, WA, British Columbia)	0%

Respondents were also asked if any members of the program leadership practiced clinically, of which, 92.6% indicated that one or more members practice clinically. Of the program

leaders that practice clinically, 35.7% work with PPC programs and make referrals within their practice. Those involved in clinical practice involving PPC were program directors, assistant/associate program directors, and clinical practicum/fieldwork directors.

### **3.3.2 PPC Content in Coursework/Curricula**

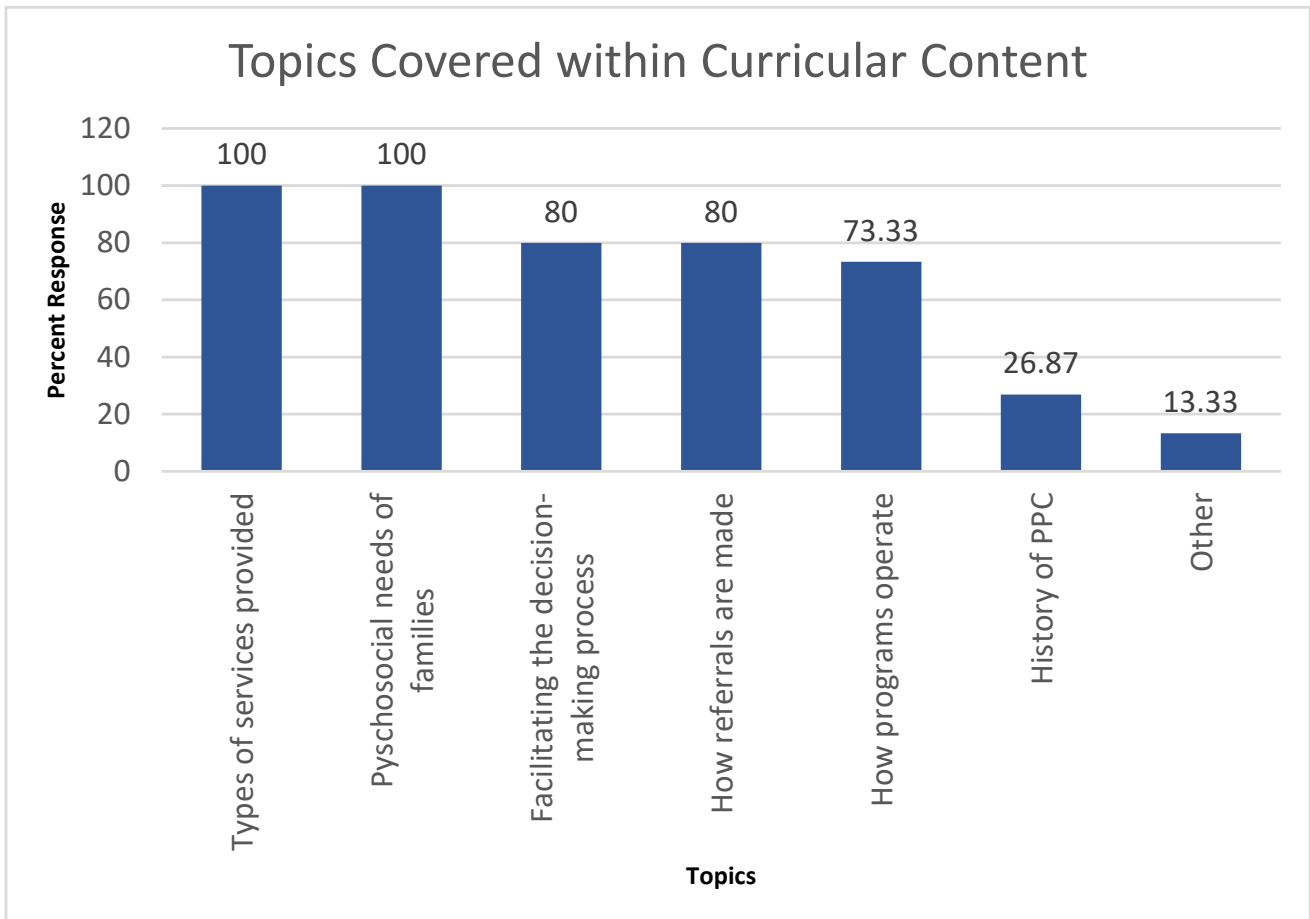
In the first section, survey questions were developed to elucidate whether PPC education was included in training programs' curricula and, if so, the format and content of the educational material. Of the respondents, 85% indicated that PPC was already incorporated into the curricular content of their program. The format of the PPC curricular material is shown in **Figure 1**. The majority of respondents indicated that information about PPC was contained within another lecture (60%) or within required reading (53.3%). Approximately one third of programs indicated there were multiple lectures with PPC content in the curricula and 26.67% had a full lecture dedicated to this material.



**Figure 1. The percent response of what types of curricular content students consume within genetic counseling training programs.**

For ‘other,’ one respondent wrote, “[There is] mention of unique challenges facing parents with perinatal loss during lecture on grief and bereavement, as well as attending Pediatric Hospice rounds/orientation and one home hospice visit with the pediatric hospice team.” Another indicated that all students must attend a clinical case conference, which may or may not have a discussion relating to PPC depending on the cases to be presented that day. Additional answers consisted of responses better classified as clinical or supplementary education. No programs indicated they have a required or optional web-based tutorial on PPC, or an optional documentary, film, or other video that addresses PPC.

The types of content offered within lectures, readings, videos, etc. is shown in **Figure 2**. All programs cover material about services provided by PPC and the psychosocial needs of families in a PPC program. Most programs also discuss facilitating the decision-making process in situations related to PPC (80%), how referrals are made to these programs (80%), and how PPC programs operate (73.3%). Only a few programs discuss the history of PPC (26.9%). Both respondents for ‘other,’ indicated students have the opportunity of an orientation and home visits through the pediatric hospice and palliative care team in their area. One respondent also described these activities as being part of an optional rotation.



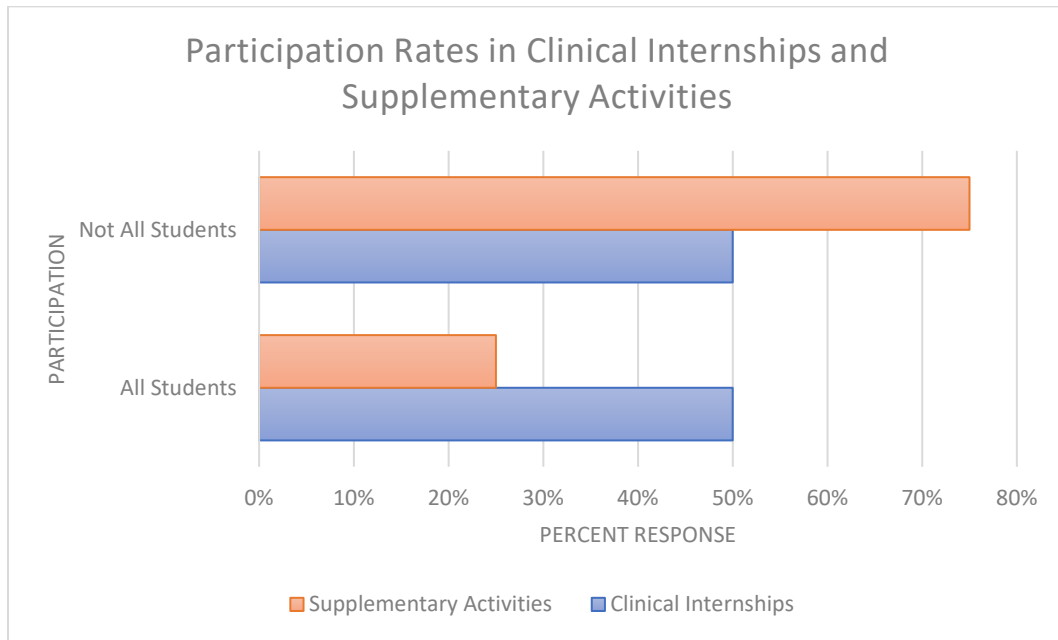
**Figure 2.** The percentage of responses for what topics are covered within curricular content.

Programs were asked about the timing of PPC educational material in the program's curricula. This was found to be quite variable, but most programs present material in both the first and second year of training (55.3%). Fewer incorporate PPC education into just the first year of training (33.3%) and fewer still incorporate educational materials into the second year of training alone (13.3%). The three programs that do not currently incorporate PPC material into their curriculum cited lack of knowledgeable faculty or staff (33.3%) and lack of time (33.3%) as barriers. One respondent indicated that students have sufficient exposure within the clinical internships provided by the program (33.3%).

### **3.3.3 PPC Content in Clinical and/or Supplementary Activities**

Most respondents (67%) indicated that PPC was available through clinical internships, of which 50% of programs require all students to participate in these activities and 50% do not (**Figure 3**). The response rate may partially be explained by patient variation in the clinical space. This may happen one of two ways. The first being that students may rotate at a site where PPC services are offered during a time when no patients need care provided by a PPC team. As one respondent wrote, "Our students are able to choose one of their rotation sites during the course of their clinical training, and a rotation with the palliative care team at our Children's Hospital is available for students to choose as a rotation site. Additionally, students may gain exposure to perinatal palliative care as a part of their prenatal or pediatric rotations, but that is not guaranteed." The second being that some programs have a structure where students do not go to every clinical site allowing student participation, so one student may be placed at a site with PPC services and their peer may be placed at a site offering the same type of services, e.g., prenatal counseling, but PPC is not provided there. One response stated, "Only two centers in the metropolitan area provide

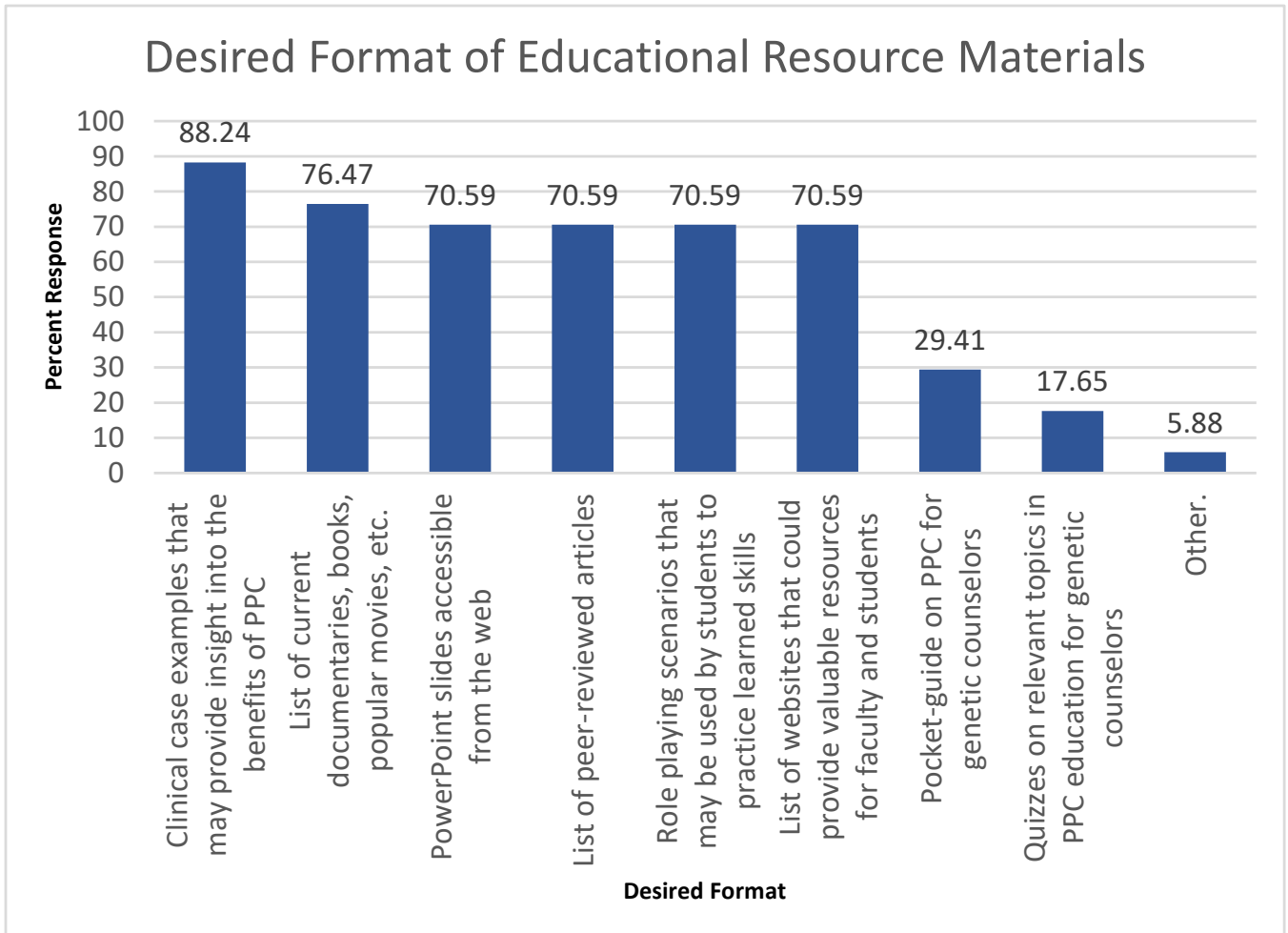
this service and allow students to participate. If the student rotates there, then they will have the experience.” So, while clinical exposure may be an important component of PPC education offered at several programs, it is not guaranteed.



**Figure 3. A graph of whether or not all students participate in clinical internships and/or supplementary activities.**

Most respondents (67%) indicated that students could gain PPC exposure through supplementary activities including conferences and optional clinical experiences (**Figure 3**). Due to the voluntary nature of these activities, 25% of programs reported that all students participated in supplementary activities.

### 3.3.4 Benefit of Educational Resources



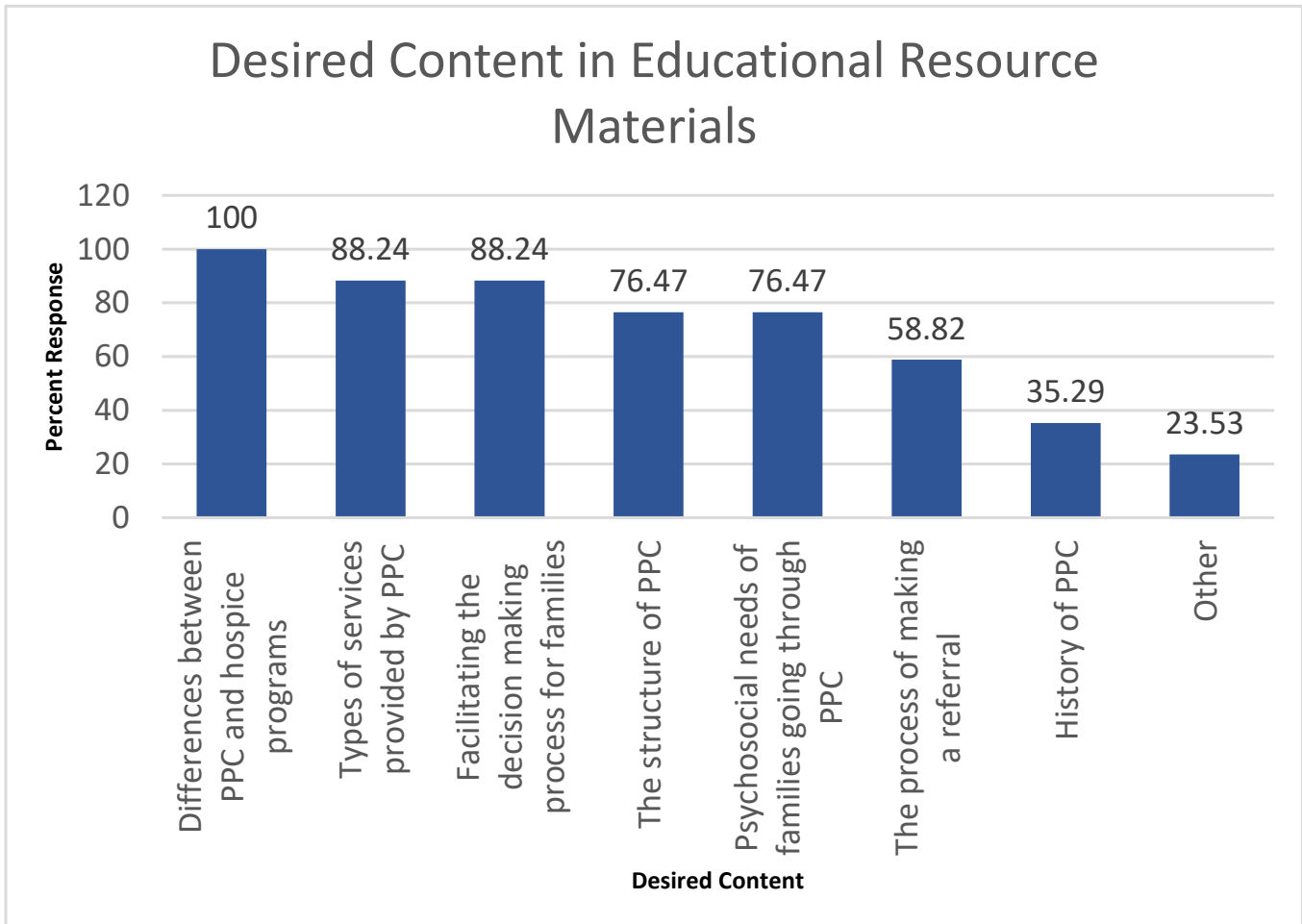
**Figure 4.** The percent response to what types of educational resource materials are desired by genetic counseling training programs.

Ninety percent of respondents indicated that creation of educational resource materials would be beneficial to their program. Survey respondents were given a list of nine formats for educational resource materials to choose from and asked to select all formats that they believed would be beneficial, shown in **Figure 4**. The study then created the top three indicated responses. Four types of materials received the same number of responses for the third most popular desired educational resource, so six total resources were created (**Appendix A**). The most-selected format



was clinical case examples (88.2%), followed by a list of current documentaries, books, etc. relating to PPC (76.5%), and then PowerPoint slides, list of peer-reviewed articles, role playing scenarios, and list of resource websites (all at 70.6%). There was less interest in a PPC pocket guide for genetic counselors (29.4%) and quizzes on PPC topics (17.7%).

Respondents were asked to indicate what subjects they would like covered in created materials. Eight choices, including the opportunity to provide a free text response, were provided to those interested in creation of new educational resource materials, which is shown in **Figure 5**. All programs wanted educational materials to cover the differences between PPC and hospice programs. The majority of programs wanted resources to include the types of services offered by PPC programs (88.24%), how to facilitate family decision-making in PPC settings (88.24%), the structure of PPC (76.5%), and psychosocial needs of families receiving PPC services (76.5%). In the 'other' category, one respondent suggested, "challenges for GCs working with families going through palliative care and reminder for use of resources to prevent compassion fatigue."



**Figure 5. The percent response to what educational topics should be covered in newly created educational resource materials.**

The 10% (2/21) that did not believe this material would be beneficial were given the opportunity to explain their reasoning. Both programs indicated they have long-standing relationships with local programs and educational content on PPC is well-integrated into the curricula. Additionally, there were concerns regarding where educational content would fit into current coursework and clinical experiences. One respondent stated, “There is likely not room to add more into the curriculum than we are already doing. If something existed and was free we could potentially incorporate it as part of the optional rotation.” Data from Qualtrics shows both

programs have been in existence for greater than 10 years. This may suggest a relationship in years of existence and strength of affiliation between training programs and PPC programs.

### 3.4 DISCUSSION

Survey response reveals information about how genetic counseling training programs educate students about PPC through curricular, clinical, and supplementary activities, an area that has not been previously studied. The study found most training programs incorporate PPC education into coursework, clinical experiences, and supplementary activities. These opportunities are often provided in both years of training programs. The majority of PPC curricular material is contained within another lecture or in assigned readings. It may be worth noting that pediatric residents and fellows report a small amount of program time being dedicated to learning about palliative care, though it is recommended that all pediatric generalists and specialists receive formal training about palliative care.<sup>55,59,60</sup> This means that many healthcare professionals, including genetic counselors,<sup>19</sup> should receive more palliative care education.

Common topics covered by programs, as identified by the survey, include types of services provided, the psychosocial needs of families receiving PPC services, facilitating the decision-making process, and how referrals are made. Reviewing the literature, there are no clearly defined guidelines for who should receive a PPC referral.<sup>32,61</sup> Some trends have been identified. Diagnoses that may warrant a consult with PPC include Trisomy 13/18 or other chromosome trisomies/problems not involving 13 and 18, anencephaly or other severe neurological conditions, renal tract abnormalities, major skeletal dysplasias, or any other diagnosis made pre- or postnatally that is life-limiting or life-threatening.<sup>31,61-63</sup>

While 90% of programs indicated a desire for new educational resource materials, 10% did not. Programs that did not indicate they wanted creation of new resources were reported to be in existence for  $\geq 10$  years. These respondents indicated a well-established relationship with PPC programs in their areas. This may indicate a relationship between these two factors. Given the small sample size, though, there is not enough information to determine the existence of such a relationship.

Further, this study uncovered some gaps in PPC education that can be addressed through creation of educational resources. The most apparent gap is that 33.3% of programs discuss the history of PPC within their training programs. Based on the results of the study, an annotated bibliography was created for programs and contains articles on topics relating to the history of PPC and other topics relating to PPC. The other area that appeared to need educational emphasis was identified by participants in free response text. Two respondents expressed a need for resources to manage compassion fatigue for counselors who may find themselves working with families experiencing perinatal loss. Previous studies suggest approximately 74% of genetic counselors are at a moderate to high risk for compassion fatigue, and approximately 25% of counselors have considered leaving the field as a result.<sup>64</sup> Other healthcare professionals have described taking care of pediatric patients with complex care needs as “relentless.”<sup>25</sup> For genetic counselors in particular, van Mol et al. theorized that certain personality traits expressed by several counselors in the study – like wanting to be in control, seeking acknowledgment, and perfectionism – may predispose genetic counselors to compassion fatigue.<sup>65</sup> A study conducted by Lee et al. also found trait anxiety, compassion satisfaction, burnout, and ethnicity other than Caucasian to be predictors of compassion fatigue.<sup>64</sup> However, healthcare providers also express that working with children in palliative care settings can be quite rewarding because they feel like they have made a

difference.<sup>46</sup> Compassion fatigue should be seen as a potential barrier for effective PPC coordination, as it makes it more difficult to form client-counselor relationships.<sup>64,65</sup> For other healthcare professionals, group dynamics and individual resilience factored into their ability to manage compassion fatigue. And while some healthcare professionals acknowledge there are ways to mitigate compassion fatigue, many do not engage in these activities on a regular basis.<sup>25,46</sup> Because of the heightened risk of compassion fatigue in genetic counselors in addition to the stress that high risk perinatal counseling may bring, articles discussing compassion fatigue and a tool to measure one's own compassion fatigue have been included in the annotated bibliography.

In addition to more traditional sources of learning, roleplaying opportunities were created to give students a chance to apply knowledge before tackling it in the clinic. In Brock's study, pediatric fellows who received simulation-based training in palliative care skills were more likely to be comfortable with PPC, more likely to engage in consults, and more likely to retain what they were being taught when compared to a cohort that only received didactic training.<sup>55</sup> In another study by Michelson, pediatric fellows reported a desire for more hands-on learning.<sup>59</sup> The roleplaying scenarios have the potential to provide similar levels of education retention, personal comfort, and referral ability.

Within the survey, respondents indicated that lack of knowledgeable faculty or staff and lack of time are barriers to PPC education within their program. As Wool's study suggests, those who are unfamiliar with PPC should be provided with additional education opportunities.<sup>19</sup> While this study focuses primarily on creation of resources for student use, other genetic counselors would also be able to use these resources to increase their knowledge about PPC. The study also addresses the barrier of time constraints by making resource materials web-accessible. Interested students may pursue topics related to PPC at their own pace. In addition to the aforementioned

concerns, respondents cited student interest as a barrier. As specialties continue to be added to the genetic counseling profession, not all students will be interested in perinatal counseling. However, the same skills used in PPC settings may also be applied to other specialties. Those with other interests may find articles discussing grief, loss, and delivering bad news cited in the annotated bibliography to be useful.

### **3.4.1 Limitations**

While this study has provided information on a topic not previously covered in the literature, it is limited by its study population. The survey was sent only to program leadership to assess what educational experiences programs provide students. While program leadership is knowledgeable about the PPC curricular, clinical, and supplementary content provided in student training, they may not possess as much information about student retention and application of this training. It also does not provide any information about genetic counseling student comfort level with PPC knowledge or their perceived referral ability. Therefore, future studies may wish to concentrate on student experiences with PPC education, or to update Wool's study from 2013.<sup>19</sup> The study is also limited by the larger proportion of respondents being from Midwestern programs that had been in existence for greater than 10 years.

This study did not develop a plan for assessment of the educational resources developed for genetic counseling training programs to determine their impact on PPC education. Future studies may wish to concentrate on assessing the benefit, if any, provided by newly created materials to see if there is an increase in PPC knowledge for current students and recent graduates.

### **3.4.2 Future Directions**

Future studies should continue to explore ways to increase genetic counselor knowledge and confidence in PPC. Student experiences with PPC education and application should be further explored. By comparing the results of this study to the results of a student-based survey, it may show additional educational areas that should be focused upon. As has been done in physician training,<sup>55</sup> research should be conducted to see if a didactic or simulation-based approach resonates more with genetic counselors when it comes to learning and applying the skills needed for PPC referral. Additionally, these studies should look at which teaching methodologies are the most time-effective as respondents indicated it is difficult to incorporate more material into the learning process.

## **3.5 CONCLUSION**

This study is the first to examine what genetic counseling training programs teach regarding PPC. Survey results show most programs provide educational experiences through curricular content, clinical internships, and supplementary activities. However, the majority of programs expressed interest in creation of further educational resource materials. Based on survey results, three resources were created for program use. Because not all programs offer PPC education, there may be two ways to apply created materials. Programs that incorporate PPC into student learning may wish to use new resources as supplementary materials. For programs without PPC education within the training structure, these materials may serve as guidance for future educational development. Regardless of how programs choose to integrate these additional

resources, the aim of this project is to provide easily accessible material that students can use to increase their knowledge of PPC, and thus comfort in referring clients to PPC services.



#### 4.0 RESEARCH SIGNIFICANCE TO GENETIC COUNSELING AND PUBLIC HEALTH

There is currently a paucity of information about PPC within the literature, and even less pertaining to genetic counselors and PPC. Yet, the public health burden posed by perinatal loss is significant. Based on the National Vital Statistics Report on Fetal and Perinatal Death, 47,041 perinatal deaths were reported in 2013, with 23,595 classified as fetal deaths and 23,446 classified as infant deaths.<sup>12</sup> There is reason to suspect the number of perinatal deaths is much higher due to underreporting caused by variation in state definitions of fetal demise, women's reluctance – or lack of recognition – to report an early miscarriage, and misunderstanding of a live birth resulting in misclassification of a stillbirth or infant death.<sup>12,27,28</sup> For comparison, the NCHS estimates there are more than 1 million annual fetal losses in the United States, most of which occur before 20 weeks gestation.<sup>12</sup> Additionally, about 500,000 women per year will be admitted to the emergency department due to vaginal bleeding during early pregnancy ( $\leq 20$  weeks gestation), and it is estimated that almost half of these women will miscarry.<sup>29</sup> Therefore, the public health burden may be higher than what current data suggest.

When someone receives an abnormal perinatal diagnosis, they are often faced with the decision to terminate or continue their pregnancy. It is difficult to assess how many of these couples are given information about PPC in the prenatal period. Palliative care may become more apparent in the case of severe anomalies identified after a birth. In the past, physicians would intensively treat severely ill infants to extend their lives as long as possible, with less attention focused on quality of life.<sup>8,43</sup> In the 1980s, some physicians began advocating for a “natural death” for infants instead of focusing on prolonging their lifespan.<sup>7,8</sup> More recently, PPC has started to

gain recognition in the medical community as a way to support families through the remainder of a pregnancy and after their child is born.<sup>55</sup> Therefore, there should be an increased awareness of PPC so families are more likely to access these services.

The majority of perinatal deaths are attributed to major malformations, chromosome conditions, and unspecified conditions relating to low birthweight or preterm birth.<sup>12,16</sup> Due to advances in prenatal screening, greater numbers of fetuses affected by birth defects, chromosome conditions, and intrauterine growth problems are being detected.<sup>8</sup> It may be possible to theorize that a greater number of families will be eligible for PPC due to the increase in detection rates. Other medical advances have increased survival rates of affected fetuses and infants.<sup>9</sup> Infants with critical conditions identified after birth may also receive PPC treatment. Whether a diagnosis is received prenatally or postnatally, these families may be referred to genetic counselors.

Genetic counseling may benefit these families. Genetic counselors provide information about conditions, emotional support, and identify resources to direct families to supportive communities and accurate information.<sup>17,19</sup> Supplying families with information about PPC would give families another resource for support and information. There is some data to suggest that PPC benefits these families by decreasing the emotional burden of perinatal loss by empowering parents to participate in their child's care and through continued contact after their child has died.<sup>13,44</sup> Due to the benefits that PPC may provide and the number of families that could be referred to these services, genetic counselors should be comfortable discussing PPC alongside intensive medical intervention and termination of pregnancy.<sup>11,13</sup> The educational resources created for this study are aimed to increase genetic counselor referral rates to PPC services by increasing familiarity and comfort with the principles of PPC.

This study adds to the genetic counseling literature as the first to examine PPC educational content in ACGC accredited genetic counseling training programs. It also contributes educational resource materials to the profession that programs can incorporate into student training, or that genetic counselors can use to expand personal knowledge about the services provided by PPC. Ultimately, the goal of this type of work is to impact public health by decreasing the burden of perinatal loss through increased referral rates. By increasing genetic counselors' knowledge of PPC, their ability to refer clients to PPC services will hopefully increase.

## 5.0 EXAMINING ACCESSIBILITY OF PERINATAL PALLIATIVE CARE IN THE UNITED STATES

At the beginning of the 20<sup>th</sup> century, the infant mortality rate in the United States was approximately 100 in 1,000 live births. Public health efforts focused on bettering living standards for mothers and babies – which included more adequate nutrition, greater access to perinatal care and healthcare for poor women, and improved surveillance and monitoring of infectious diseases – contributed to a greater than 90% decrease in infant mortality rates.<sup>66</sup> By the beginning of the 21<sup>st</sup> century, the majority of perinatal death is attributed to more innate conditions such as major congenital malformations, chromosome conditions, and unspecified conditions relating to low birthweight or preterm birth.<sup>12,16</sup> Advances in medicine have improved both the ability to prenatally diagnose these conditions and extend the survival times of affected fetuses and infants.<sup>8,10,11</sup> Yet, technological and medical advances do not ensure the survival of every critically affected fetus and infant. In 2013, the National Center for Health Statistics (NCHS) reported 23,595 fetal deaths and 23,446 infant deaths in the United States.<sup>12</sup> As advances in medicine continue, children born with a life-threatening or life-limiting condition may survive for longer periods of time<sup>13,21,30</sup>, suggesting a need for specialized practices to care for the unique needs of these children.<sup>67</sup>

Perinatal palliative care (PPC) is a clinical program comprised of an interdisciplinary team that cares for both the medical needs of a fetus or infant with a life-limiting or life-threatening disorder, and the emotional needs of his or her family members.<sup>1,3</sup> Medical efforts accommodate the treatment and comfort needs of the child, while psychosocial support is often derived through the creation of mementos, grief counseling, and/or religious support.<sup>2,3</sup> As PPC programs gain

greater recognition for its niche services, the number of programs throughout the United States has quickly grown.<sup>6,67</sup> Evidence of growth is seen in the literature through citations of the number of programs recorded on [www.perinatalhospice.org](http://www.perinatalhospice.org), a curated list of PPC programs within the United States.<sup>37</sup> In January 2013, 130 programs were listed,<sup>19</sup> and in October 2016, listings had increased to 201.<sup>6</sup> As of December 2017, 231 programs were recorded on the website, but 268 programs were identified upon further investigation because some hospital systems have multiple locations capable of offering PPC services. Because all listings are self-reported to the website, it does not provide an entirely accurate picture of the current number of programs, or total volume of growth, in the United States, but it reveals the rapidity of program development.

For decades, the traditional medical practice was to sustain a child's life for as long as possible with intensive, and often invasive, intervention.<sup>9</sup> The concept of PPC to support an infant in living and in dying is a relatively new one. Early PPC efforts are best characterized as mere adaptations of adult hospice practices. Cicely Saunders, who was a nurse, social worker and doctor during her career, is credited with starting hospice care.<sup>1,8,22</sup> Saunders recognized that terminally ill patients had unmet medical needs.<sup>4,22</sup> She defined the concept of "total pain," which recognizes the emotional, spiritual, religious, and physical aspects of pain and suffering, and began advocating for better pain and symptom management for her patients.<sup>22-24</sup> It was not until the 1980s that physicians began to advocate for the same management to be used with infants affected by life-limiting conditions.<sup>8</sup>

While PPC has grown and improved over the years, a uniform clinical care model has yet to be established.<sup>4</sup> This absence results in a lack of data on cost effectiveness and cost utilization of specific palliative treatments offered by different centers.<sup>67</sup> A survey of PPC programs in the United States revealed variable response rates when providing patient care, family care, and

administrative tasks.<sup>6</sup> There has been little investigation into how much PPC programs benefit families<sup>68</sup>, but some health care providers believe PPC has a positive impact.<sup>49</sup> In adult settings, the link is more evident. Studies have shown better symptom management, reductions in stress and anxiety, lowered caregiver burden, reduced admissions to the emergency room and/or hospital, and improvement in quality of life and/or survival as long as the palliative care program is operated by experienced professionals trained in palliative care.<sup>67</sup>

A few studies have examined inequities in palliative care, noting that people from lower socioeconomic backgrounds die younger and in poorer health. This population also has a higher likelihood of dying in a hospital compared to those from higher socioeconomic backgrounds.<sup>67</sup> Because several of these studies focus on adults, less is known about the inequities that may affect families eligible for PPC services and potential barriers to their use. The current study was done to gain a better understanding of the accessibility of PPC programs throughout the United States, as well as possible contributing factors. First, I mapped the state and county location of the 268 programs identified using the website, [www.perinatalhospice.org](http://www.perinatalhospice.org).<sup>37</sup> Next, I assessed the relationship between the PPC programs and possible contributing factors, including: number of genetic counseling programs per state, state total population size, infant death rate, and percent in poverty in each state. Finally, I estimated the total population living within the counties containing PPC programs was performed to determine what proportion of the country has ready access to palliative care. Identification of who has access to PPC is crucial to ensure the needs of the public are being adequately met.

### **5.1.1 Study Aim 1**

The first goal of this study was to develop a resource map to aid genetic counselors and other public health practitioners in referring patients to these services. This resource map would also be available to patients seeking palliative services in their area.

### **5.1.2 Study Aim 2**

The second goal of the study was to estimate accessibility of PPC programs at the state and county level, as well as assess factors that might be correlated with accessibility to PPC programs. To achieve this goal, I

- a) Obtained data on the number of PPC programs per state, as well as the infant mortality levels, population, and percent of individuals living in poverty by state
- b) Assessed possible state-level relationships between PPC program numbers and number of genetic counseling (GC) programs, infant mortality, population size, and percent population living below the poverty line.
- c) Used estimates of county-wide population numbers to identify the proportion of individuals with ready access to PPC programs.

## 5.2 DESCRIPTION OF DATASET

### 5.2.1 Perinatal Palliative Care Programs

PPC programs were identified using [www.perinatalhospice.org](http://www.perinatalhospice.org) from September to December 2017.<sup>37</sup> At the time of the analysis, 231 programs were listed. Two data points were removed because they did not appear to be in existence any longer. Twenty-four (10.5%) of the listed programs have multiple locations (range of 2 – 8) capable of providing PPC services. With these adjustments made to the data set, the total number of identified programs was 268.

As program locations were identified on the website, information was recorded about the addresses and the types of services provided.<sup>37</sup> Using information from program websites, programs two overarching categories were identified: care-based and support only. Care-based programs are ones identified as being able to offer medical care within a clinical or hospital setting. These programs may or may not offer psychosocial support, but many do.<sup>6</sup> PPC programs that were classified as support only comprised parent/family support groups, counseling services, and spiritual services. As programs were identified, additional categories were created for home-based programs and organizing networks. A home-based program is one that incorporates features of a home – personalized rooms, living rooms, kitchen, etc. – with medical care and emotional support. Such a program should not be confused with hospice programs that offer in-home services. While, in-home care is an important way to deliver PPC services to families who wish to bring their child home, none were given this specific label. This is partially a result that several care-based organizations offer in-home services, but some may do so only on an advisory basis. An organizing network is when a group of PPC programs are associated through one entity. The majority of PPC (245) programs were categorized as care-based. Only one program was described as home-based.



Twenty-two programs met criteria for support only. Five organizing networks were identified and incorporated into the mapping process of this project, but were not used in analysis of accessibility, because these did not fit into the two overarching categories of either care-based or support only.

Many of the identified care-based locations were pediatric programs or hospice centers capable of offering fetal, neonatal, and perinatal support. Almost all of these programs had an interdisciplinary team of healthcare providers who attended to patient and family needs. PPC providers included, but were not limited to, physicians, nurses, genetic counselors, chaplains, therapists, and social workers. The support groups were specific to difficulties in the perinatal period including identification of a severe fetal anomaly, prenatal diagnosis of a life-limiting or life-threatening condition, fetal loss (miscarriage or stillbirth), postnatal identification of an infant with complex care needs, and/or infant loss.

## **5.2.2 Genetic Counseling Training Programs**

Genetic counseling training programs located in the United States were identified using the website of the Accreditation Council for Genetic Counseling (ACGC).<sup>69</sup> All programs displayed on the resource map had been accredited by the ACGC. The address of each program or University housing a program was recorded. At the onset of the project, there were 37 accredited programs in existence in the United States. In February 2018, an additional two programs were added to the ACGC list of accredited programs, increasing the number to 39. The new programs were incorporated into the dataset and used in final analysis.

### 5.2.3 Infant Death Rate

Infant death rates were identified using the vital statistics report from the Center for Disease Control and Prevention (CDC).<sup>16</sup> An infant is defined as a living human under 365 days old. Infant death was chosen over perinatal death because its classification is consistent across states, whereas perinatal death is not. The CDC generates a yearly vital statistics report, on data collected by each state. Each state counts an infant death if a baby is born living (showing signs of life like breathing or voluntary movement), followed by a brief survival period, and then dies, thus causing a birth certificate and death certificate to be issued. Perinatal death, which encompasses both infant and fetal death, is harder to tabulate because fetal death report varies by state. The majority of states records a fetal demise if the fetal birthweight is greater than 350 grams, or, if birthweight is unknown, the pregnancy is beyond 20 weeks gestation. However, some states report fetal deaths occurring at any age, and other report at 500 grams or past 22 weeks gestation.<sup>27</sup>

### 5.2.4 Census data

I used Census.gov (<https://www.census.gov/>) to obtain data on state population size, percent of the population living in poverty, and per capita income.<sup>70</sup> Infant death per state in 2014 was obtained using the CDC Wonder tool.<sup>71</sup> This study also investigated whether the number of PPC programs was correlated with the number of accredited genetic counseling programs in the state.<sup>69</sup> Genetic counseling programs were assessed as a contributing factor because families in need of a PPC referral are highly likely to receive genetic counseling services,<sup>17</sup> and the assumption that training programs would offer PPC exposure to their students reasonable. For a full table of recorded variables, see **Appendix B**.

## 5.3 METHODS

### 5.3.1 Mapping

Using the aforementioned PPC website, program locations were identified and specific addresses were recorded in a Microsoft Excel document.<sup>37</sup> Program addresses were grouped by state location. Once all addresses were recorded, [www.mapchart.net](http://www.mapchart.net) was used to generate an infographic of the United States to show the distribution of PPC programs by state. This tool allows users to shade different states as desired, for example, by the number of PPC programs within the state.

Because this state-based map is a crude depiction of the distribution of PPC sites, and thus patient access, I also created an additional map of the number of PPC programs by county. To identify these counties, zip code data from previous address collection was examined in the Zip Codes To Go database, which links zip codes to county names.<sup>72</sup> The counties were recorded and mapped with [www.mapchart.net](http://www.mapchart.net).

Concomitantly, a custom map with each location of the 268 programs was created using Google MyMaps<sup>73</sup> to develop a resource to be used by public health practitioners and patients wishing to identify programs local to them.

### 5.3.2 Analysis Methods

I assessed whether the number of PPC programs per state was associated with state population size, percent of population living in poverty, or per capita income. These measures have been correlated with health. I also investigated whether the number of PPC programs was

correlated with the number of accredited genetic counseling programs in the state.<sup>69</sup> Genetic counseling programs were assessed as a contributing factor because families in need of a PPC referral are highly likely to receive genetic counseling services,<sup>17</sup> thus GC training programs are likely to offer PPC exposure to their students. All variables were plotted against each other and both parametric and non-parametric (Spearman) correlations were estimated using the statistical software program JMP PRO 13.

I also investigated access to PPC programs. For the purposes of this study, I created a variable called “ready access to PPC”. Ready access describes anyone living within a county containing  $\geq 1$  PPC program. Counties were identified by zip code. Population data (from the 2010 Census) for each identified county was collected using the Census FactFinder tool.<sup>74</sup> I estimated the proportion of the U.S. population with ready access by summing across all counties with PPC programs and then comparing this number to the total population of the United States from the 2010 Census.

## 5.4 RESULTS

### 5.4.1 PPC Program Resource Map (Aim 1)

I developed a resource tool to be used by practitioners and patients that displayed each PPC category: care-based, support only, home-based, and organizing network (**Figure 6**). Each category (care-based, support only, home-based, or organizing network) was assigned a map layer along with a distinguishing symbol. The layer feature allows users accessing the map to hide or unhide each symbol. The 245 care-based programs are represented on the map by a blue cross.

The 22 support only programs are depicted by a purple heart. The one home-based location is represented by a green house. The five organizing networks are depicted by a black office building. Additionally, genetic counseling programs, indicated by a red academic building, were included on the map.

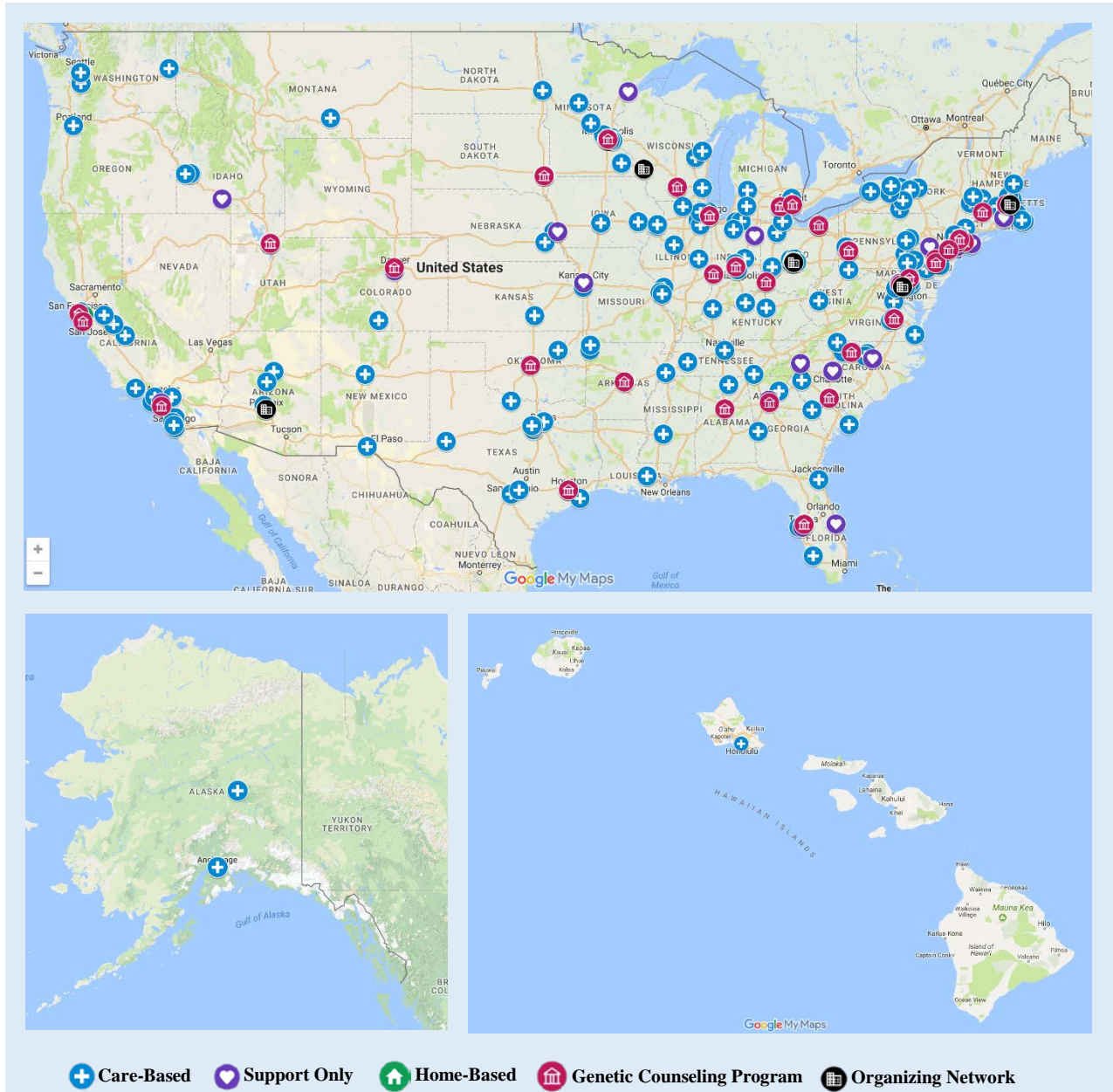
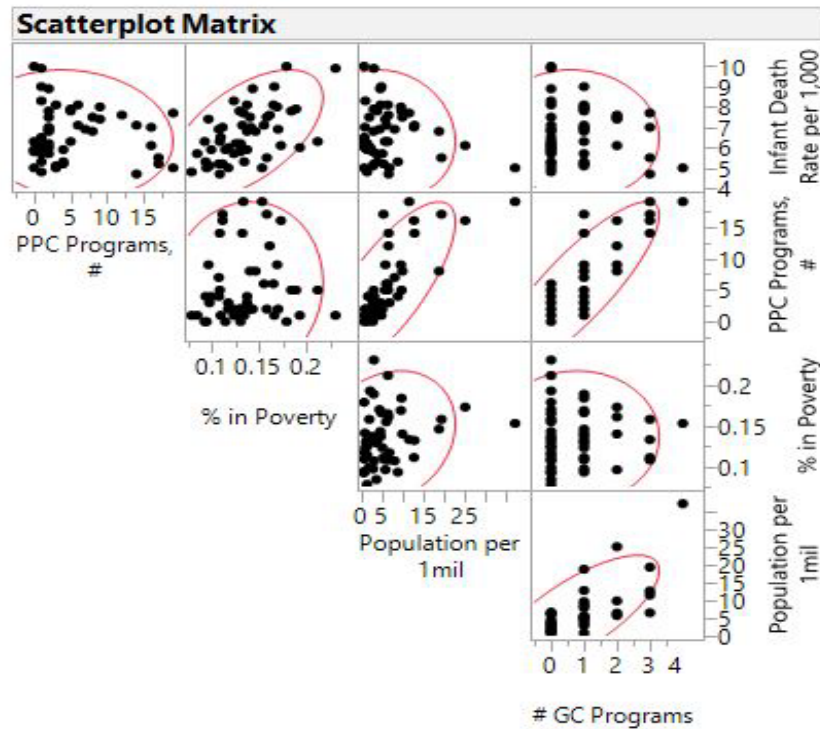


Figure 6. Screenshots of the custom map identifying every PPC program within the United States.



correlation coefficients were significant ( $p < 0.05$ ); results of the parametric correlation analyses were similar and are not presented. As expected, infant mortality rate is significantly correlated with percent of people living in poverty



**Figure 8. A correlation matrix representing strengths of relationships between the number of PPC programs, number of genetic counseling programs, the infant mortality rate, population level, and percent of the state living below the poverty line.**

( $\rho = 0.5493$ ;  $p\text{-value} = <0.0001$ ). Total state population size is correlated with the total number of PPC programs within a state ( $\rho = 0.8278$ ;  $p\text{-value} = <0.0001$ ), as well as the number of genetic counseling programs within a state ( $\rho = 0.7149$ ;  $p\text{-value} = <0.0001$ ). Finally, the number of GC programs is correlated with the number of PPC programs ( $\rho = 0.7494$ ;  $p\text{-value} = <0.0001$ ), indicating that faculty, staff, healthcare providers, and students associated with genetic counseling programs are likely to have ready access to PPC programs.

Table 3. Spearman's test applied to the correlation matrix in Figure 8.

Nonparametric: Spearman's $\rho$												
Variable	by Variable	Spearman $\rho$	Prob>   $\rho$	-0.8	-0.6	-0.4	-0.2	0	0.2	0.4	0.6	0.8
PPC Programs, #	Infant Death Rate per 1,000	0.0227	0.8743									
% in Poverty	Infant Death Rate per 1,000	0.5493	<.0001*									
% in Poverty	PPC Programs, #	0.1515	0.2886									
Population per 1mil	Infant Death Rate per 1,000	0.0562	0.6953									
Population per 1mil	PPC Programs, #	0.8278	<.0001*									
Population per 1mil	% in Poverty	0.2520	0.0744									
# GC Programs	Infant Death Rate per 1,000	0.0676	0.6372									
# GC Programs	PPC Programs, #	0.7494	<.0001*									
# GC Programs	% in Poverty	0.0558	0.6972									
# GC Programs	Population per 1mil	0.7149	<.0001*									

The relationship between the number of PPC and number of genetic counseling programs within a state may result from increased population size alone. In other words, a state with a larger population size may have more GC programs and more PPC programs, and the two are not

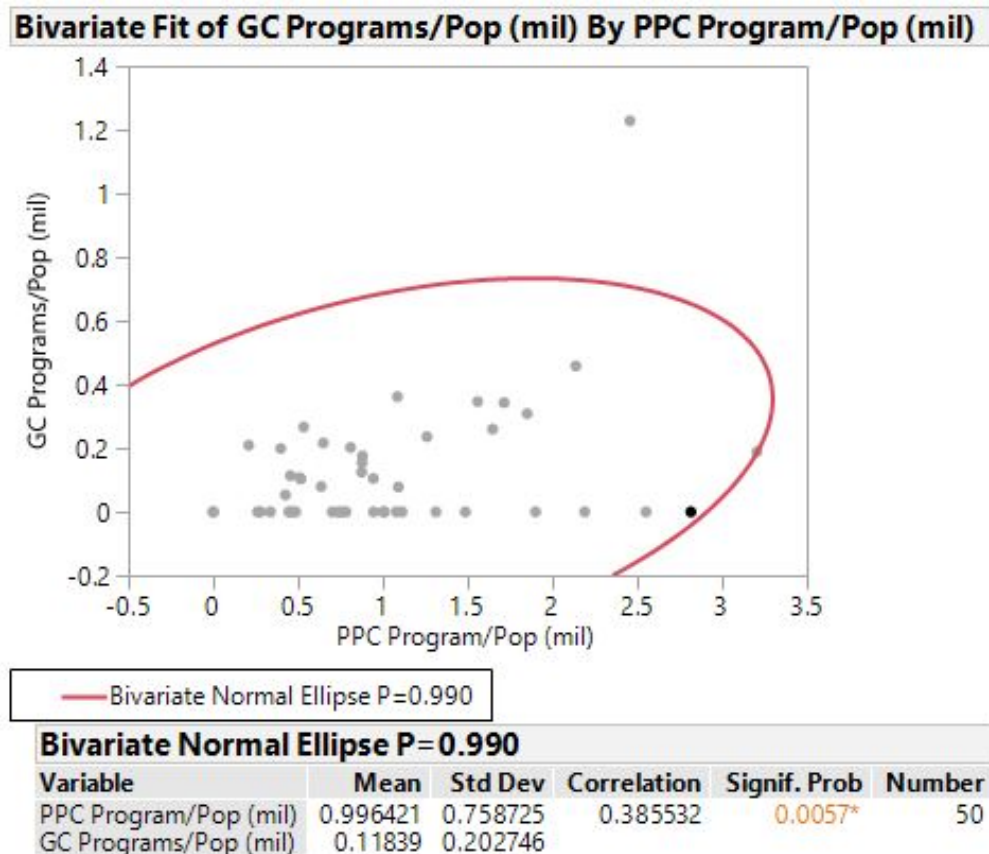


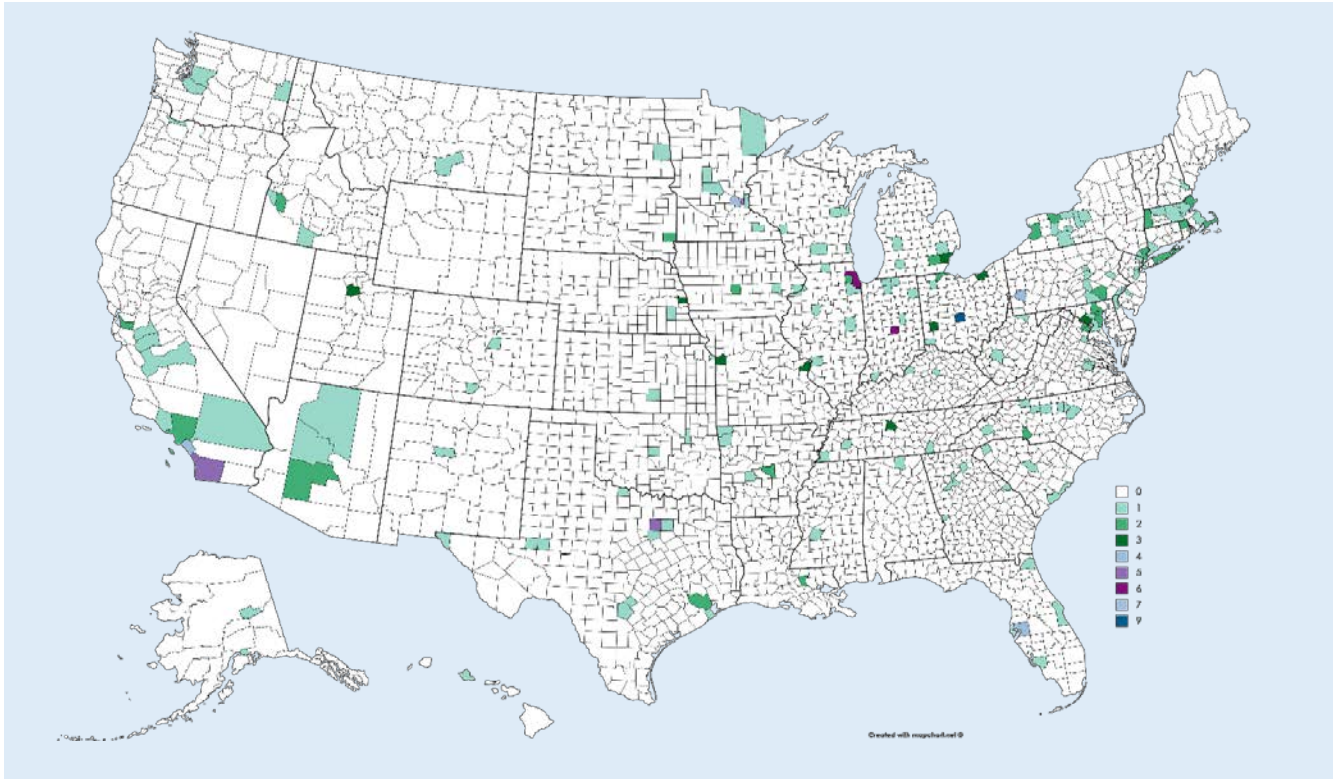
Figure 9. Correlation between the number of PPC and the number of GC programs after standardizing population.



independently correlated with one another. I performed additional analyses after accounting for state population size. The number of GC or PPC programs per person per state was calculated by dividing the number of each program by the total population of the state. These standardized variables were plotted and the correlation was estimated (**Figure 9**). The correlation between the standardized variables was 0.385, p-value = 0.0057, indicating that population size was not the only cause of the correlation between number of PPC and GC programs.

### 5.4.3 County-based Analyses (Aim 2c)

Because location of PPC programs on the state level was crude, I investigated the number of PPC programs by county. There were 175 unique counties containing the 268 PPC programs in the United States. The number of programs per county ranged from 1 – 8.



**Figure 10. Number of PPC program locations in the United States per county.**

I next mapped the number of PPC programs by county (**Figure 10**). As can be seen, large regions of the United States do not have access to a PPC program. The total population within the 175 counties with  $\geq 1$  PPC program was 119,726,345 individuals, representing 38.8% of the total United States population (308.7 million). Thus, 61.2% of the U.S. population does not have ready access to PPC services.

## 5.5 DISCUSSION

I developed a custom map to be used as a resource tool by health care practitioners and patients to identify nearby PPC program locations. While online tools identifying adult palliative care and hospice care locations throughout the U.S. exists, I did not discover another map with comprehensive location mapping such as the one created for this study. Maps displaying the number of PPC centers by state and by county were also created for this study. Inspection of both maps revealed that fewer programs exist in the center of the United States. The county map further revealed the presence of large geographic regions without ready access to PPC centers, even within states that have multiple PPC site. For example, both New York and California have  $>16$  PPC centers, but none are located in the northern regions of both states. At the county level, I estimated that 38.8% of the total U.S. population lives in a county that contains  $\geq 1$  PPC program; thus they are expected to have ready access. Given that congenital malformations, chromosomal conditions, and conditions causing preterm birth and low birth weight are equally likely to occur in areas without a PPC program, the need to increase accessibility and reduce disparities within the U.S. is critical. Many of the regions without a PPC center are rural and/or mountainous; thus, the population density is low. Developing and maintaining a PPC program in these regions may not

be cost effective for a variety of factors. Rural hospitals currently face declining profit margins and lower patient volumes, which is partly due to more patients choosing urban over rural hospitals, as well as fewer patients having sufficient coverage to seek services.<sup>38,40</sup> One potential method to lower healthcare costs is telemedicine.<sup>39-41</sup> Using an internet connection and teleconferencing, a health care practitioner, such as a general practitioner located in a rural area, who is not an expert in hospice care, would be able to obtain guidance from one or more clinicians who specialize in perinatal or pediatric palliative care. By removing the obstacle of distance, the relationships established between a PPC program and a distant site may help families maintain balance between the needs of their child and the needs of other members of their family.<sup>39</sup> These relationships could also increase PPC access in rural communities by lowering healthcare costs, which have been shown to be a sustainable health delivery model in other fields, like telepharmacy.<sup>40,41</sup>

In addition to examining geographic access to PPC centers, I assessed possible correlations between the number of PPC centers and several factors (e.g., population size) that might affect the number of PPC centers. As expected, increased infant death rate is significantly correlated with the percent of people living in poverty. Furthermore, as expected, the total population size of a state was positively correlated with both the total number of PPC programs and the number of GC programs ( $p < 0.0001$  for both comparisons). The number of GC programs was also significantly correlated with an increased number of PPC programs within a state ( $p < 0.0001$ ). This latter result may indicate states with more PPC programs per person tend to have more genetic counseling programs as well, and vice versa. That is, the relationship between number of PPC and GC programs is due to the increasing population size alone however. After controlling for population size, however, the correlation was still significant ( $p = 0.0057$ ). Another reason for the correlation

between number of GC and PPC programs is that states prioritizing one program type may also prioritize the other. An additional possibility is structural, that is, the known relationship between universities, genetic counseling programs, and hospital systems. Genetic counseling programs often rely on local resources to provide clinical training experiences to their students. If the GC program is housed within a university strongly associated with a hospital system, the hospital system may be more likely to offer PPC services.

Several limitations to this study exist. First, only county data was used to estimate who has access to PPC programs. In many instances, however, PPC programs exist near county and/or state borders, thus, some communities have ready access to centers and are not being represented in this analysis. For example, individuals residing in eastern Oregon would be within commuting distance of a center in western Idaho (**Figure 6**). Using an estimate of the population living within a specific radius (in miles) of PPC sites instead of within a single county would provide a better measure of how many people could use are located geographically closer to PPC services. Additionally, using county data does not necessarily guarantee an individual has ready access to a PPC program. Other physical and societal barriers may exist within a county or metropolitan area, including topography, access to transportation, and the knowledge of local healthcare providers. Further studies centered on the demographics of the population living around PPC programs are needed, as well as studies on other potential barriers, such as ease of transportation. By examining these factors, we may begin to gain a better understanding of barriers affecting access to PPC programs within the United States, which could then be used to develop interventions to reduce disparities in care.

## APPENDIX A: SURVEY MATERIALS

### A.1 EMAIL INVITATION AND EXPLANATION FOR SURVEY

Dear Genetic Counseling Program Directors,

My name is Meg Hager, and I am a second-year genetic counseling student at the University of Pittsburgh. Recently, I sent out a request for one member of the program leadership to participate in a survey related to my Master's thesis. As a reminder, I am conducting a research project titled "Assessing Perinatal Palliative Care Education within Genetic Counseling Programs." The purpose of this research is to ascertain curricular content regarding education on perinatal palliative care (PPC) in ACGC Accredited Programs, which will be assessed by a brief survey. The information obtained by the survey will be used to develop educational resource material for use by genetic counseling programs and their students. A recent study determined genetic counselors, while familiar with PPC practices, were uncomfortable referring patients to these services due to lack of education and experience with PPC. By creating educational resource material, the goal of this project is to increase genetic counselor knowledge of PPC.

For the purposes of this survey, only **one** response per program is needed. Please direct this survey to one person within your program leadership. The survey should take approximately 10 minutes to complete. Should you have any questions or concerns about this survey, please contact me at [meg.hager@pitt.edu](mailto:meg.hager@pitt.edu). Thank you for your participation,

Meg Hager  
University of Pittsburgh  
256-640-2559

## A.2 QUALTRICS INTRODUCTORY TEXT

This survey has been created as part of a Master’s research study entitled “Assessing Perinatal Palliative Care Education within Genetic Counseling Programs.” The purpose of this project is to ascertain curricular content regarding education on perinatal palliative care (PPC) in ACGC accredited programs. The overall goal of this project is to generate educational resource material that can be accessed by genetic counseling programs and their students.

Please answer the following questions regarding your program’s current educational practices regarding PPC for your students, as well as what materials might be valuable to you as someone within your program’s leadership. Again, only **one** response per program is needed. The survey should take approximately 10 minutes. There are no foreseeable risks associated with this project, nor are there any direct benefits to you. You will not receive any payment for participation. All answers collected from this survey will be anonymous, and will not be traced back to you. All responses are confidential, and results will be collected and stored in a private manner. Your participation is voluntary, and you may withdraw from this project at any time. If you have any questions or comments, you may contact Meg Hager at [meg.hager@pitt.edu](mailto:meg.hager@pitt.edu).

## A.3 SURVEY – UPDATE TO REFLECT ANY LANGUAGE CHANGES MADE IN QUALTRICS

### Section 1: PPC EDUCATION

1. Does your Genetic Counseling program provide education on perinatal palliative care in any courses? **If no, skip to 6.**
  - a. Yes
  - b. No

2. If yes, what education methods are provided? Please check all that apply.
  - a. One lecture on perinatal palliative care
  - b. Greater than one lecture on perinatal palliative care
  - c. Content on perinatal palliative care within another lecture
  - d. Required reading on perinatal palliative care
  - e. Optional reading on perinatal palliative care
  - f. Required web-based tutorial on perinatal palliative care
  - g. Optional web-based tutorial on perinatal palliative care
  - h. Required documentary, film or other video that addresses perinatal palliative care
  - i. Optional documentary, film or other video that addresses perinatal palliative care
  - j. Other. Please describe.
  
3. Who, or what entity, delivers this educational material?
  - a. Healthcare provider affiliated with the perinatal palliative care program
  - b. Genetic counselor
  - c. Other healthcare provider, please explain
  
4. Is this educational material presented in the first or second year of a program?
  - a. First
  - b. Second
  - c. Both
  
5. If educational content is provided, what type of content is discussed? Please check all that apply.
  - a. The history of perinatal palliative care
  - b. Types of services provided by perinatal palliative care
  - c. Psychosocial needs of families who can be served by perinatal palliative care
  - d. Facilitating the decision-making process of families in perinatal palliative care
  - e. How a perinatal palliative care program operates
  - f. How referrals are made to perinatal palliative care programs
  - g. Other. Please describe.
  
6. If no, why does your program not provide education on perinatal palliative care? Please check all that apply.
  - a. Lack of knowledgeable faculty or staff
  - b. Lack of time
  - c. Lack of educational materials
  - d. Students are disinterested
  - e. Unnecessary to genetic counselor training
  - f. Other. Please describe.
  
7. Please choose the one that is the most important factor in why your program does not provide education on perinatal palliative care:
  - a. Lack of knowledgeable faculty or staff
  - b. Lack of time

- c. Lack of educational materials
- d. Students are disinterested
- e. Unnecessary to genetic counselor training
- f. Other. Please describe.

## Section 2: PPC IN CLINICAL ROTATIONS

1. Are there any clinical rotations or internships that incorporate perinatal palliative care into its structure that students may participate in? **If no, skip to 8**
  - a. Yes
  - b. No
2. Is the clinical internship located at a local or a distant (>2 hours drive) site?
  - a. Local site
  - b. Distant site
  - c. Students have a choice between a local or a distant site
3. Do all students participate in a clinical internship that involves perinatal palliative care?
  - a. Yes
  - b. No
4. If no, please explain why: **Open Response**
5. How long do participating students spend with the perinatal palliative care team?
  - a. <1 week
  - b. 1-4 weeks
  - c. >4 weeks
6. Does a genetic counselor act as supervisor in perinatal palliative care activities?
  - a. Yes
  - b. No
7. If no, who supervises genetic counseling students in perinatal palliative care activities? **Open Response**
8. Why are there no clinical internships with a perinatal palliative care element at your program? **Open Response**
9. Are there any supplementary activities that incorporate perinatal palliative care into its structure that students may participate in?
  - a. Yes
  - b. No
10. If yes, can you describe what the supplementary activity is? **Open Response**



11. Do all students participate in this supplementary activity?

- a. Yes
- b. No

12. If no, please explain.

### Section 3: EDUCATIONAL MATERIAL

1. Would your program benefit from the creation of educational resource material concerning PPC?

- a. Yes
- b. No

2. If no, please explain.

3. What types of content would you like to be included in the educational resource material?

- a. The history of perinatal palliative care
- b. The process of making a referral to a perinatal palliative care program
- c. Differences between perinatal palliative care and other palliative/hospice programs
- d. The structure of perinatal palliative care
- e. Types of services provided by perinatal palliative care
- f. Psychosocial needs of families going through perinatal palliative care
- g. Facilitating the decision making process for families going through perinatal palliative care
- h. Other. Please describe.

4. What format would you like educational content to be in? Please select all that would be beneficial.

- a. Powerpoint slides that can be accessed from the web
- b. Pocket-guide on perinatal palliative care for genetic counselors
- c. Clinical case examples that may provide insight into the benefits of perinatal palliative care
- d. Packets of peer-reviewed articles that can be used to generate discussion
- e. List of current documentaries, books, popular movies, etc. that illustrate the need for perinatal palliative care
- f. Quizzes on relevant topics in perinatal palliative care education for genetic counselors
- g. Role playing scenarios that can be used by students to practice learned skills
- h. List of websites that could provide valuable resources for faculty and students
- i. Other. Please describe

## Section 4: DEMOGRAPHICS

1. How long has the program been in existence?
  - a. 0-3 years
  - b. 4-9 years
  - c.  $\geq 10$  years
  
2. What region of North America are you in? (NSGC regions)
  - a. Region 1 (CT, MA, ME, NH, RI, VT, CN Maritime Provinces)
  - b. Region 2 (DC, DE, MD, NJ, NY, PA, VA, WV, PR, VI, Quebec)
  - c. Region 3 (AL, FL, GA, KY, LA, MS, NC, SC, TN)
  - d. Region 4 (AR, IA, IL, IN, KS, MI, MN, MO, ND, NE, OH, OK, SD, WI, Ontario)
  - e. Region 5 (AZ, CO, MT, NM, TX, UT WY, Alberta, Manitoba, Sask)
  - f. Region 6 (AK, CA, HI, ID NV, OR, WA, British Columbia)
  
3. Do members of the program leadership currently practice clinically? Check off all that apply.
  - a. Program Director
  - b. Co-Director
  - c. Assistant/Associate Program Director
  - d. Clinical Practicum/Fieldwork Coordinator
  
4. Do members of the program leadership that practice clinically participate in perinatal palliative care activities? **If yes, skip to 5.**
  - a. Yes
  - b. No
  - c. N/A
  
5. If yes, which person in the program leadership participates in perinatal palliative care activities? Check off all that apply.
  - a. Program Director
  - b. Co-Director
  - c. Assistant/Associate Program Director
  - d. Clinical Practicum/Fieldwork Coordinator

Thank you for taking the time to participate in this survey!

## **APPENDIX B: PERINATAL PALLIATIVE CARE EDUCATIONAL RESOURCE**

### **MATERIALS**

#### **RESOURCE COVER PAGE**

The following educational resource materials were created in partial fulfillment of the thesis requirements for the MS in Genetic Counseling at the University of Pittsburgh. The thesis, entitled *Assessing Perinatal Palliative Care Education within Genetic Counseling Training Programs*, may be accessed on D-Scholarship ([d-scholarship.pitt.edu](http://d-scholarship.pitt.edu)) for review. All materials were designed by Megan M. Hager, MS, MPH, graduating class of 2018. The thesis was approved on May 14, 2018 by Scott Maurer, MD, Candace Kammerer, PhD, Meredith Jones, MS, LCGC, and Robin E. Grubs, MS, PhD, LCGC (Thesis Director).

#### **ABSTRACT**

Perinatal palliative care (PPC) is a clinical program that provides holistic care to critically ill fetuses and infants alongside psychosocial support to their family members. Genetic counselors are often consulted to explain prenatal or infantile conditions that are life-limiting or life-threatening. The function of the genetic counselor is to provide both education about a condition and supportive resources for their clients. Referral to a PPC program is one way a counselor may provide support. Therefore, being knowledgeable about the medical and psychosocial services offered by PPC programs is important. A recent study found that 11.4% of genetic counselors feel uncomfortable referring to these programs due to a lack of familiarity with PPC. This study aims to increase genetic counselor knowledge about PPC programs through the creation of educational resource materials for genetic counseling training programs accredited by the Accreditation Council for Genetic Counseling (ACGC). A Qualtrics survey was developed and administered to ACGC accredited program leadership to assess PPC education within the curricula, clinical internships, and supplementary activities. Additionally, program directors were asked if programs would benefit from creation of resource material. Survey data shows 85% of responding programs provide education about PPC, and 90% of respondents support creation of educational resource materials. Using the results of the survey to direct content and format of resource material, three documents were created: 1) A series of case studies were created that may also be used as roleplays, 2) An annotated bibliography directing users to articles, books, videos, and websites to increase PPC knowledge, and 3) A PowerPoint covering relevant topics in PPC. Additionally, a resource map identifying locations of PPC programs within the United States was created. This study is designed to impact public health by strengthening the educational experiences of genetic counseling students, with the goal of making them more comfortable when referring clients to PPC services.

## ANNOTATED BIBLIOGRAPHY

The following list of articles, books, videos, and websites is meant to be used as a reference guide for those interested in learning more about perinatal palliative care. Many of these references illustrate the psychosocial space parents may be in after learning of a life-limiting or life-threatening condition in their fetus or infant to illustrate their emotional needs at this time. These sources also emphasize the need of early, and accurate, discussions of palliative care so families have time to adjust to the news that their child may pass away quickly.

### Articles

Allen, JSF, Mulhauser, LC. Genetic Counseling After Abnormal Prenatal Diagnosis: Facilitating Coping in Families Who Continue Their Pregnancies. *Journal of Genetic Counseling*, 1995.

Allen and Mulhauser discuss genetic counseling skills that can be used to manage parental grief after continuation of a pregnancy with a fetal condition. There are a variety of clinical case scenarios identifying parental feelings and decision-making processes. The article also examines research in the field and makes suggestions for counseling interventions with families receiving an abnormal prenatal diagnosis.

Baile, W F, et al. SPIKES – A Six-Step Protocol for Delivering Bad News: Application to the Patient with Cancer. *The Oncologist*. 2000.

While this delivery model is specified for cancer patients, the methodology of SPIKES can be applied to any situation in which it is required to deliver bad news. Not only does it outline a delivery of bad news, but this text also provides examples of statements, questions, and responses that one can use in these types of situations.

Balaguer, A, et al. The Model of Palliative Care in the Perinatal Setting: A Review of the Literature. *BMC Pediatrics*, 2012.

Perinatal palliative care is a relatively new field. While it has been recognized as important since the 1980s, very little research has gone into this area of medicine. This review article examined the literature to potentially identify an empirical, best model of care in this setting. This study was unable to identify this best model of care.

Bijma, HH, et al. Decision-Making After Ultrasound Diagnosis of Fetal Abnormality. *European Clinics in Obstetrics and Gynaecology*, 2007, pp. 89 – 95.

This article discusses how advances in ultrasound technology have improved the detection rate of minor and major fetal anomalies, and how that has had an impact on the psychological well-being of parents. There is also some examination of the decision-making process following identification of an anomaly.

Broom, M, et al. Exploring Parental and Staff Perceptions of the Family-Integrated Care Model. *Advances in Neonatal Care*, 2017, pp. 1 – 8.

This is a study of parental and provider perceptions of a recent shift in NICU practices to better integrate families into the care making decisions for their children. Major components of the particular study model include bedside education and participating in cares, group education sessions, family-centered ward rounds, and psychosocial support. Overall, perceptions on both sides were positive, and healthcare providers appreciated taking on the role of educator for these families.

Caitlin, A, Carter, B. Creation of a Neonatal End-of-Life Palliative Care Protocol. *Journal of Perinatology*, 2002, pp. 184 – 195.

Caitlin and Carter were among the first to establish comprehensive guidelines for perinatal palliative care. While no formal guidelines have been established for this type of care, many of the recommendations and themes identified in this article can be seen in programs throughout the United States.

Campbell, AGM, McHaffie HE. Prolonging Life and Allowing Death: Infants. *Journal of medical ethics*, 1995, pp. 339 – 344.

This article looks at perinatal palliative care/hospice from an ethical lens. It discusses who should decide whether or not it is time to withdraw support, and provides some guidance in the decision-making process for both healthcare providers and families at the end of a child's life.

Denney-Koelsch, E, et al. A Survey of Perinatal Palliative Care Programs in the United States: Structure, Processes, and Outcomes. *Journal of Palliative Medicine*, 2016.

This survey examines what types of services are provided by perinatal palliative care programs throughout the United States. Several administrative and care tasks are identified in this survey, along with important themes and values that programs strive to uphold. Results of this study show areas of inconsistency amongst programs and points out the lack of validated assessment tools in perinatal palliative care practices.

Hinds, PS, et al. Trying to Be a Good Parent as Defined by Interviews with Parents who made Phase I, Terminal Care, and Resuscitation Decisions for their Children. *Journal of Clinical Oncology*, 2009.

This study explores the parental beliefs and perceptions of being a good parent when making end-of-life care decisions for their children. Using the results from this study allows healthcare providers deeper insight into what parents may be feeling when making decisions and how to better support them during the decision-making process.

Janvier, A, et al. Parental Hopes, Interventions, and Survival of Neonates with Trisomy 13 and 18. *American Journal of Medical Genetics*, 2016.

It is not uncommon for families' hopes and goals to change after they receive the news that their child has a life-limiting condition. This article captures an array of the beliefs felt by parents who have received either a prenatal or postnatal diagnosis of trisomy 13/18. Some common hopes identified by this study include parents hoping to meet their child alive, taking their child home, being a family, and giving their child a good life. When a better understanding of parental goals is achieved, care may be better directed to meet those needs.

LeDuff III, LD, et al. Transitional Objects to Facilitate Grieving Following Perinatal Loss. *Advances in Neonatal Care*, 2017, pp. 347 – 353.

This study illustrates the importance of physical objects and mementos to the grieving process. Parents appear to benefit from the creation of mementos – photographs, footprints, handprints, etc. – but few studies have been conducted on their effectiveness. This study shows that objects help with immediate loss and recommends further research for long-term effects of mementos.

Leuthner, SR. Palliative Care of the Infant with Lethal Anomalies. *Pediatric Clinics of North America*. 2004.

Leuthner defines perinatal palliative care and discusses multiple facets of care that should be discussed throughout the remainder of a pregnancy and after birth. He also provides guidance on which families may be the best candidates for these types of services, as well as guidelines for creation of a palliative care plan.

Riotte, CO, et al. Identifying the Types of Support Needed by Interprofessional Teams Providing Pediatric End-of-Life Care: A Thematic Analysis. *Journal of Palliative Medicine*. 2017.

While the majority of pediatric deaths occur in a hospital setting, there are few supports in place for the healthcare providers that must care for these children. This study explores the types of support needed and desired by healthcare providers involved in caring for critically ill children and identifies what types of supports are useful for clinicians to help reduce the risk for caretaker burnout.

Sebold, C, Koil, C. Genetic Library: Grief and Bereavement. *Journal of Genetic Counseling*. 2009.

This collection focuses on research related to grief, loss, and end-of-life care. This article may be used by genetic counselors to further expand their knowledge and response to grief.

Sumner, LH, et al. Extending Palliative Care Into Pregnancy and the Immediate Newborn Period. *Journal of Perinatal Neonatal Nursing*, 2006, pp. 113 – 116.

This article calls for the extension of palliative care to fetuses and infants, due to the elevated number of deaths in the perinatal period when compared to all other childhood deaths. The article both illustrates the need to help families suffering from perinatal loss and the lack of research in this particular area of medicine.

Taylor, J, Aldridge, J. Exploring the Rewards and Challenges of Paediatric Palliative Care Work – A Qualitative Study of a Multi-Disciplinary Children’s Hospice Care Team. *BMC Palliative Care*, 2017.

This study provides insight into what pediatric palliative care team members may identify as rewards and challenges within their profession. It also examines group dynamics and identifies communication avenues that appear to increase how supported providers feel. There is also acknowledgement of the increased risk of burnout in this population and how early identification of burnout is beneficial to staff.

Umberger, E, et al. Enhancing NICU Parent Engagement and Empowerment. *Seminars in Pediatric Surgery*, 2018, pp. 19 – 24.

Parents of infants with complex care needs may feel overwhelmed in the NICU setting. It is important to employ strategies that allow parents to engage in the care of their child when they are in a situation when they feel their child’s care is out of their control. This article provides insight on how parents may be feeling in the NICU and provides suggestions to provide parents the opportunity to parent their child and to make them more comfortable in the decision-making process.

Wallace, SE, et al. Parent Perspectives of Support Received from Physicians and/or Genetic Counselors Following a Decision to Continue a Pregnancy with a Prenatal Diagnosis of Trisomy 13/18. *Journal of Genetic Counseling*, 2017.

Families choosing to continue a pregnancy after a prenatal diagnosis of trisomy 13 or 18 may pose a psychosocial challenge to healthcare providers they interact with. This study was designed to assess these parents’ perceptions of physician and genetic counselor support. The study found that genetic counselors were more likely to provide unbiased information in an understandable way when compared to physicians. Parent statements were collected to illustrate what made them feel supported/unsupported, and recommendations were made to improve support to these families.

Weaver, MS. Growing Symbiotic Local Partnerships to Nurture Quality Pediatric Hospice Care in Rural Regions. Companion Planting. *JAMA Pediatrics*, 2018.

This piece describes how a group of healthcare providers made in-home hospice care possible for a child in a rural location. It was accomplished using telemedicine to allow for interdisciplinary team meetings to discuss how care should proceed. This not only shows

how to meet family needs, but also gives a nod to the future of healthcare as more e-services are provided from urban to rural settings.

Woodgate, RL, et al. Intense Parenting: A Qualitative Study Detailing the Experiences of Parenting Children with Complex Care Needs. *BMC Pediatrics*, 2015.

While this article is not specific to palliative care, it provides insight into the decision-making process of families who have severely ill children. A variety of assumed parental roles are also identified in the article, such as healthcare provider, case manager, and student.

Wool, C, Dudek, M. Exploring the Perceptions and the Role of Genetic Counselors in the Emerging Field of Perinatal Palliative Care. *Journal of Genetic Counseling*, 2013.

This is the first study examining genetic counselor perceptions about perinatal palliative care. Wool's study found that while the majority of genetic counselors were familiar with perinatal palliative care, not all of them were comfortable with making referrals to these services. Further education of genetic counselors about these programs was recommended.

Wool, C, et al. Provision of Services in Perinatal Palliative Care: A Multicenter Survey in the United States. *Journal of Palliative Medicine*, 3, 2016.

This article looks at how perinatal palliative care services are provided in a variety of program settings (e.g. academic medical centers, regional or community hospitals, local hospices, or community-based organizations) across the United States. The study found that 100% of surveyed programs provide spiritual needs and bereavement services to families and that 70% of identified programs are  $\leq 10$ -years-old.



## Books and Texts

Black, Beth, et al. *Perinatal and Pediatric Bereavement in Nursing and Other Health Professions*. Springer Publishing Company. 2015.

This text covers several aspects of facilitating the grieving process for those who have lost a child so they may continue to live after their loved one has died. The book covers these topics by providing a collection of clinical experiences, knowledge, and care models.

Blanford, Cathy. *Something Happened: A Book for Children and Parents who Have Experienced Pregnancy Loss*. Something Happened Handbook. 2012.

This story is designed to help children understand what pregnancy loss is. It addresses topics related to grief and guilt a child may experience after a baby brother or sister is miscarried. The book may also be used as a tool to help parents understand what their child is going through.

Eunpu, Deborah L. Chapter 10: Genetic Counseling Strategies for Working with Families. *Genetic Counseling Practice*. Wiley-Blackwell. 2010, pp. 235 – 251.

Within this chapter of *Genetic Counseling Practice*, Deborah Eunpu discusses how to construct a family-centered counseling session. Some strategies include using the family history as a way for families to talk through the story of a birth or death, and as a way to explore various attitudes related to illness, grief, and loss. The chapter concludes with some brief case examples, including one focusing on grieving for a baby who has recently died. Understanding family dynamics may be helpful in situations requiring palliative care.

Getting, Elizabeth. Chapter 5: Grieving: An Inevitable Journey. *Genetic Counseling Practice*. Wiley-Blackwell. 2010, pp. 95 – 124.

Grief is inevitably a part of perinatal palliative care, regardless of if it happens following prenatal diagnosis or following the birth of a child with a critical condition. In order to provide adequate healthcare as a genetic counselor, it is important to gain an understanding of what grief is. Elizabeth Getting's chapter in *Genetic Counseling Practice* is a valuable resource that places grief into the context of genetic counseling.

Gryte, Marilyn. *No New Baby: For Siblings who have a Brother or Sister Die Before Birth*. Centering Corporation. 1988.

This book discusses various feelings children may have when they discover that an expectant brother or sister has died before birth. Marilyn Gryte also seeks to answer questions that children may have about pregnancy loss through this story. More recent additions include a section for parents and grandparents.

Heustis, Jane, et al. *Companioning at a Time of Perinatal Loss: A Guide for Nurses, Physicians, Social Workers, Chaplains and Other Bedside Caregivers*. Companion Press. 2004.

This handbook covers topics relating to grief and loss that may not be covered by the standard bereavement training in one's hospital. The book encourages healthcare workers to create a compassionate bedside environment through the delivery of continual grief support.

Knight, Paula. *The Facts of Life*. Penn State University Press. 2017.

Through this semi-autobiographical graphic novel, Paula Knight tells the story of Polly, a woman who tries to become a mother but struggles to do so due to infertility, chronic illness, and miscarriage. This novel is a story of reflection on what it means to be a woman and if motherhood is a necessary part of this.

For more comics relating to fertility, miscarriage and childlessness, you may visit Knight's website at: <https://paulaknight.wordpress.com/category/the-facts-of-life/>.

Kuebelbeck, Amy. *Waiting with Gabriel: A Story of Cherishing a Baby's Brief Life*. Loyola Press. 2008.

Amy Kuebelbeck wrote this book after the loss of her son, Gabriel, who was prenatally diagnosed with hypoplastic left heart syndrome. She writes about her family's choice to pursue comfort care over termination or surgical intervention, as well as the struggle to make plans for Gabriel's birth and premature death.

Kuebelbeck, Amy. *A Gift of Time: Continuing Your Pregnancy When Your Baby's Life is Expected to be Brief*. Johns Hopkins University Press. 2011.

Amy Kuebelbeck expands upon her previous work from *Waiting with Gabriel* with this practical guide for parents wishing to continue a pregnancy for a child with a life-limiting condition. Information about perinatal hospice and palliative care is detailed throughout the book, as well as numerous parent experiences with perinatal loss.

Smith, Angie. *I Will Carry You: The Sacred Dance of Grief and Joy*. B&H Books. 2010.

Angie Smith relates her experiences of perinatal loss after she and her husband learn that their fourth child, Audrey Caroline, has several conditions incompatible with life. Instead of terminating the pregnancy, the couple decide to continue the pregnancy. Additionally, Smith discusses her faith by weaving the biblical story of Lazarus throughout the narrative.

Vredevelt, Pam. *Empty Arms: Hope and Support for Those Who Have Suffered a Miscarriage, Stillbirth, or Tubal Pregnancy*. Multnomah. 2001.

This book was written from the vantage point of a licensed professional counselor who has experienced both the loss of a baby and a 16-year-old son. Within this work, Pam Vredevelt shares her personal story while answering questions about loss, grief, and religious faith related to losing a child.

## Videos

\*Note: Videos indicated with an asterisk show images of an infant who has died.

Andrea Moore. Michael's Story: Infant Hospice Care. 23 May 2013, *YouTube*. [www.youtube.com/watch?v=gA-CmfibXZs](http://www.youtube.com/watch?v=gA-CmfibXZs).

Andrea Moore discusses her son's unexpected, postnatal diagnosis of trisomy 13 and the decision to care for him at home until he passed away. She encourages healthcare providers to discuss infant death more openly so they may help families decide on the best care options for their child at the end of his or her life.

NationalHospice. Baby Amanda. 2014, *YouTube*. [www.youtube.com/watch?time\\_continue=111&v=RBExDejLCZA](http://www.youtube.com/watch?time_continue=111&v=RBExDejLCZA).

Amanda was prenatally diagnosed with trisomy 18. Her parents discuss how perinatal hospice has allowed them to celebrate every moment they have with her because they feel well supported. The video also provides visuals of perinatal hospice caregivers working inside of Amanda's home.

Barcroft TV. DOCS: Baby Hospital Ep. 1 – 3. 2017, *YouTube*. [www.youtube.com/watch?v=Ww5D2KVp6f8&t](http://www.youtube.com/watch?v=Ww5D2KVp6f8&t).

This series follows the stories of several babies who are being cared for by the Neonatal Unit of Liverpool Women's Hospital. It gives viewers insight into how perinatal palliative care units operate, what doctors may be thinking in terms of plan of care, and even films families interacting with their babies in the unit. The decisions that some families have to make are tough, and the outcome for every child is different.

CareDimensions. A family shares their pediatric hospice experience. 06 Aug 2014, *YouTube*. [www.youtube.com/watch?v=gpQ4BFzIzM8](http://www.youtube.com/watch?v=gpQ4BFzIzM8).

Two parents discuss their son's diagnosis of Spinal Muscular Atrophy at two-months-old and the hospice care they received through CareDimensions. They discussed how supportive the healthcare providers were as they took care of their son at home.

\*The Dallas Morning News. Choosing Thomas. 1 Sep 2009, *YouTube*. [www.youtube.com/watch?v=ToNWquoXqJI&t](http://www.youtube.com/watch?v=ToNWquoXqJI&t).

Parents, T.K and Deidrea Laxus, describe their son's diagnosis of trisomy 13 at 20 weeks gestation. They decided to continue with the pregnancy and were able to bring their soon home to provide comfort care. This video helps others understand the thoughts, feelings, and decisions parents make when they have an infant with a life-limiting condition.

digifruits. Skyler Sue – Our Little Angel (Lissencephaly). 19 May 2012. *YouTube*. [www.youtube.com/watch?v=ZCuilmr\\_bEI&t](http://www.youtube.com/watch?v=ZCuilmr_bEI&t).

This video shows the 3 month and 4 day life of Skyler Sue, a child born with lissencephaly. Within minutes of her birth, she began to have seizures, which revealed her condition. The video gives viewers a better idea of what home hospice/palliative care looks like for a seriously ill infant who is both living and dying.

To learn more about Skyler Sue's life, you can read about her on her blog at [www.skylersue.com](http://www.skylersue.com).

Hospice of the Western Reserve. The Keeney Family discuss Pediatric Palliative Care. 18 Aug 2010, *YouTube*. [www.youtube.com/watch?v=6zxYoTIKt6o&t](http://www.youtube.com/watch?v=6zxYoTIKt6o&t).

The Keeney family had triplet children, two girls and one boy. At about 9-months-old, it became apparent that one of their daughters had serious health concerns. She was later diagnosed with a mitochondrial disorder. At that time, the family did not know their daughters were identical, until their other daughter became sick at 2-years-old. The Hospice of the Western Reserve stepped in to help the Keeney family by providing emotional support to the family members and medical support to their children. The Keeneys discuss the services provided and how it made them feel supported in this video.

IgniterMedia. 99 Balloons. 3 Apr 2007, *YouTube*. [www.youtube.com/watch?v=th6Njr-qkq0](http://www.youtube.com/watch?v=th6Njr-qkq0).

This video tells the story of Eliot who was prenatally diagnosed with trisomy 18 and lived for 99 days. In this video, his parents chronicle the story of his birth, life, and death through the letters read aloud by Eliot's father. Throughout the video, he discusses the positive aspects of taking care of a sick child along with some struggles.

Johns Hopkins Medicine. Holding Space: Palliative Care in the NICU at Johns Hopkins Children's Center. 30 Aug 2016, *YouTube*. [www.youtube.com/watch?v=xO8OqQkXEfQ&t](http://www.youtube.com/watch?v=xO8OqQkXEfQ&t).

In this video, four families share their experiences with perinatal loss and the care their children received at Johns Hopkins Children's Center's NICU. Healthcare providers also discuss available services within the unit and how they support families.

Karen Fauman. Principles of Pediatric Palliative Care. 04 Mar 2013, *YouTube*. [www.youtube.com/watch?v=5dnS5d\\_H fs&t=](http://www.youtube.com/watch?v=5dnS5d_H fs&t=).

While this lecture is intended for primary care physicians, it may be used by other healthcare providers to gain a better understanding of how to communicate with families of critically ill children. For instance, timing the discussion about end-of-life care earlier in a child's diagnosis, making sure families understand what their child's disease is and how it progresses, and who should first discuss palliative care with the family. Karen Fauman, MD, also discusses the differences between palliative care and hospice care.

NINRnews. Starting a Conversation about Pediatric Palliative Care. 08 Jan 2014, *YouTube*. [www.youtube.com/watch?v=hKK24Bp1elQ&t](http://www.youtube.com/watch?v=hKK24Bp1elQ&t).

This vignette provides clinicians with tips for opening conversations about palliative care and creating an ongoing dialogue with that family. The speaker not only discusses how to approach the conversation, but also defines the services provided by palliative care and what questions clinicians should ask families as they move forward. This is a useful tool for anyone who wants to sharpen their conversation about palliative care.

\*PerinatalHospice.org. Perinatal Hospice: Continuing Your Pregnancy when You Baby's Life is Expected to be Brief. 23 Mar 2016, *YouTube*. [www.youtube.com/watch?v=rTTNeqRvZ3w](http://www.youtube.com/watch?v=rTTNeqRvZ3w).

This is a slideshow of various families celebrating the birth, life, and death of their children captured by the organization Now I Lay Me Down to Sleep, a nonprofit that creates remembrance portraits for families suffering a perinatal loss. Memento making is often an important aspect of palliative care.

To learn more about family and photographer perspectives on this organization, you may view a video of their mission on their website: <https://www.nowilaymedowntosleep.org/about/mission-and-history/>.

\*Tammy Ziegler. Perinatal Hospice Video. 06 Jul 2011, *YouTube*. [www.youtube.com/watch?v=tY7mq1g9pGk&t](http://www.youtube.com/watch?v=tY7mq1g9pGk&t).

This is a video about the Warner family and their experience with perinatal hospice. It was created by their infant's nurse, Tammy. This video provides an overview palliative care for infants that are not expected to live long, how their families may react, and shows normalization of postnatal care, such as bathing and clothing the infant.

TEDx Talks. How to Manage Compassion Fatigue in Caregiving | Patricia Smith | TEDxSanJuanIsland. 07 Feb 2017, *YouTube*. [www.youtube.com/watch?v=7keppA8XRas](http://www.youtube.com/watch?v=7keppA8XRas).

Patricia Smith discusses compassion fatigue and different management strategies in this TED talk. This may be useful for any healthcare providers that are beginning to show signs of this type of stress.

\*UnityPoint Health – Meriter. Perinatal Palliative Care – The Zimmer Family Story. 15 Jul 2013. *YouTube*. [www.youtube.com/watch?v=DRYN01RGLp4&t](http://www.youtube.com/watch?v=DRYN01RGLp4&t).

The Zimmer family was expecting twins after years of fertility problems. Mother, Kelly Zimmer, recounts their experience with the perinatal palliative care team at Meriter Health Services. These services were initiated shortly after renal agenesis was discovered in their son, Judah, during the 20-week ultrasound. They were able to create a birth plan for Judah with the team in order to maximize their time as a family of four. Kelly also expanded upon the psychosocial services and how the perinatal team kept in touch in the year following Judah's death.

## Websites

Compassion Fatigue Awareness Project. *CFAP*. [www.compassionfatigue.org/](http://www.compassionfatigue.org/).

There is a greater risk for compassion fatigue in healthcare providers who work with critically ill patients. This organization is dedicated to raising awareness to compassion fatigue. It also provides caregiver resources so people may better identify and prevent compassion fatigue.

Compassion Fatigue: Could it be Compromising Your Professionalism? *Crisis Prevention Institute*. [www.crisisprevention.com/Blog/August-2015/Compassion-Fatigue](http://www.crisisprevention.com/Blog/August-2015/Compassion-Fatigue).

Because healthcare providers working with seriously ill patients are more likely to suffer from compassion fatigue and/or burnout, it is important to recognize when it is happening to you and learning what you can do to prevent it. This is a blog entry that gives an overview of compassion – its symptoms, risk factors, and strategies to alleviate it. Resources and references are also provided.

Continuing Your Pregnancy When Your Baby's Life is Expected to be Brief. *Perinatal Hospice & Palliative Care*. [www.perinatalhospice.org/](http://www.perinatalhospice.org/).

Perinatal Hospice is a website administered by Amy Kuebelbeck, author of *Waiting with Gabriel*. This website provides updated information on perinatal palliative care, parent stories, links to birth planning, and a comprehensive list of perinatal palliative care programs throughout the United States. There are also several links to resources for both parents and caregivers on the website including links to videos, websites, and journals.

E-learning Course on Perinatal Palliative Care. *International Children's Palliative Care Network (ICPCN)*. [www.icpcn.org/icpcn-introduces-new-e-learning-course-perinatal-palliative-care/](http://www.icpcn.org/icpcn-introduces-new-e-learning-course-perinatal-palliative-care/).

This is a free e-learning course for healthcare providers that was created by senior neonatal nurse, Alex Mancini. The main goal of this course is to improve the quality of perinatal care by increasing provider knowledge and awareness of services provided by palliative care, which fetuses and infants would benefit. Modules within this course include information about palliative care, the delivery or of perinatal palliative care, creating memories, case studies, resolving conflicts about end-of-life decisions, and organ donation.

Palliative Care Toolkit. *Texas Pediatric Society*. [www.txpeds.org/palliative-care-toolkit](http://www.txpeds.org/palliative-care-toolkit).

This website contains lessons on a variety of topics relating to palliative care such as Structure & Processes of Care, Ethical & Legal Aspects of Care, and Psychological and Psychiatric Aspects of Care. The website also provides sample forms and models that students may want to consider as they enter their own practice.

Prenatal Information Research. Consortium. *Prenatal Information Research Consortium*. [www.prenatalinformation.org/](http://www.prenatalinformation.org/).

The consortium brings together researchers who are interested in ethical, legal, and social issues of screening and diagnosis in the prenatal, preconception, and perinatal periods. The website provides links to current studies, publications, events, and resources. There is also a blog that covers various issues within the field.

The ProQol Measure in English and non-English Translations. *ProQOL.org*. [www.proqol.org/ProQol\\_Test.html](http://www.proqol.org/ProQol_Test.html).

The Professional Quality of Life Elements (ProQOL) Theory and Measurement tool is used to assess the amount of compassion satisfaction, burnout, and secondary traumatic stress (AKA compassion fatigue) that a professional may feel. The website provides the test and self-scoring sheet, as well as additional resources and information about the ProQOL tool.

Resolve Through Sharing: Online Education. *Resolve Through Sharing*. [www.gundersenhealth.org/resolve-through-sharing/bereavement-training/online-education/](http://www.gundersenhealth.org/resolve-through-sharing/bereavement-training/online-education/).

Resolve Through Sharing has been offering bereavement training since 1981. They provide education to healthcare providers through on-site trainings as well as webinars. These lectures and webinars cover a wide range of topics including communicating with the bereaved, frameworks for grief, and caring for families experiencing perinatal death.

## CLINICAL CASE SCENARIOS

### Scenario 1: Prenatal diagnosis

Wanda Williams is referred to genetic counseling after a first-trimester ultrasound is suspicious for anencephaly. The genetic counselor has the opportunity to discuss several different options for the pregnancy with Ms. Williams. Because second trimester detection rates of neural tube defects are higher than first trimester rates, Ms. Williams could follow up with additional ultrasounds and AFP testing until a more definitive diagnosis was made, decide to terminate the pregnancy, or consult with the perinatal palliative care team to determine what course of action would most align with her goals. After meeting with the perinatal palliative care team, Ms. Williams and her partner decide the best course of action is to terminate and create mementos of the pregnancy.

Studies show up to 40% of women will choose to continue a pregnancy after receiving a potentially lethal prenatal diagnosis.<sup>61,63</sup> Before the child is born, many programs seek to develop a birth plan, establish treatment protocol for the child once they are born (e.g. palliative care only, intensive interventions, discharge home), help the family make burial or cremation arrangements, and schedule autopsies if desired. The birth plan includes things from handling the infant (i.e., cutting the cord, bathing, dressing) to caring for the infant (i.e. pain medication, regular newborn care, resuscitation, use of a ventilator). Programs may also discuss memento making after the child is born, counseling plans, and support groups for families.<sup>6,61</sup>

One study found that most families presented with the idea of perinatal hospice took an average of 14 days to accept admission into the program, which may be related to the need to process an abnormal diagnosis. The program, a home perinatal palliative care program, conducted weekly or bimonthly check-ins with families. During these meetings, they focused on the previously described goals. On average, families were admitted to the program 45 days before giving birth, and saw the home care provider an average of 3 times. The study determined there was a 2-month gap between suspected diagnosis and referral, and recommended earlier referral for affected families. By having earlier referrals, more women would have the opportunity to receive palliative services. For example, women/families considering the termination of a pregnancy could benefit from palliative services through the creation of pregnancy memories or techniques for discussing the news of a prenatal life-limiting diagnosis to family and friends. However, most women in the study received this information shortly before they had to make a decision about termination, which was a barrier to consultation with the palliative care team.<sup>61</sup> Given the average time between referral and admission to a program, it may be beneficial to bring up palliative care as an option in cases where fetus or infant prognosis is expected to be poor or uncertain.

Note: There are several prenatal indications that may benefit from referral to perinatal palliative care including: Trisomy 13, Trisomy 18, other chromosome trisomies/problems not involving 13 or 18, anencephaly, renal tract abnormalities, major skeletal dysplasias, or an apparently non-syndromic fetus affected with several severe congenital malformations that may limit his or her potential to live after birth.<sup>61-63</sup> In contrast, fetuses with an isolated cardiac or surgical anomaly are typically referred directly to a cardiologist or surgeon instead of palliative care.<sup>31</sup> Oftentimes, it is the degree of uncertainty of a particular diagnosis that makes clinicians feel more comfortable with referring to palliative care.<sup>32</sup>



## **Scenario 2: Religious and Cultural Beliefs in Perinatal Palliative Care**

Suzy and Sam Smith are expecting their second child, when at 12 weeks Mrs. Smith receives a positive NIPT for Trisomy 13. She undergoes a CVS, which confirms the results. During an appointment with a genetic counselor, the Smiths express their hope that God will intervene and heal their son, Benedict. They also express feeling their faith is being dismissed by other members of the care team when they make statements like this. The genetic counselor begins discussing their religious beliefs to learn more about their goals for the pregnancy. When asked about the name of their son, the Smiths state that the name is a Biblical one that means ‘blessing.’ At the end of the appointment, the Smiths thank their genetic counselor for listening to them and asking questions about their religious beliefs.

When families come to a genetic counselor to discuss abnormal prenatal results, they often come to the appointment with beliefs cultivated from their religious and cultural experiences. It is important to ask questions to gain a better understanding of how their beliefs influence their goals and decisions.<sup>18</sup> Studies show that families want to be asked about their faith by their medical professionals. And while 76% of pediatricians agree that religion and faith are important to treating a patient, 51% report that they rarely or never talk about religion with their patients.<sup>75</sup> By discussing their beliefs, it is not only a way to design a care plan, but also a way in which to build rapport, such as the above example where the genetic counselor asks about the Smiths’ name choice.<sup>10,75</sup>

Often, religious clients find comfort and hope in their spirituality. These beliefs can be integrated into the care plan constructed for a family in perinatal palliative care.<sup>75</sup> For instance, families may wish to consult religious leaders when it is recommended that medical intervention be shifted away from trying to cure a disease process. In the case of a child born with severe lung disease, clinicians may advocate for removal of ventilatory support as it will not benefit the infant and not aide the healing process. Recommending that clients consult with spiritual leaders and/or family members to affirm their decision is one way to be supportive.<sup>1</sup>

Studies show that some clinicians may limit discussions about ongoing care goals and other clinical discussions with religious families to avoid being perceived as disrespectful toward family beliefs. There is also some level of discomfort between clinicians and families when the family is perceived as being unrealistic or prolonging suffering of the child. However, care goals can be reframed in a way that positively impacts both parties.<sup>75</sup> Some techniques include validating parents beliefs, acknowledging their desire for a miracle, and asking them about their religious beliefs.<sup>1,18,75</sup> Some possible phrases include, “I know this must be difficult for you, how are you managing?” or, “Given what your family is going through, what is your greatest hope at the moment?”<sup>75</sup> It is also important to maintain hope throughout the care process by adapting ‘miracles’ without detracting from the importance of truthful, clinical information. These can take shape in the form of positive clinical goals, such as getting to meet a baby diagnosed with severe congenital anomalies while he or she is still alive, or getting to breastfeed the baby, or helping the baby live long enough to meet extended family members.<sup>10,75</sup>

### **Scenario 3: Early induction**

Tina Thompson is in the second trimester of her third pregnancy. Her previous pregnancies have all ended in miscarriage. Ms. Thompson's pregnancy has been closely followed due to her previous history and because her current pregnancy is not doing well. Due to problems with the placenta, the fetus is not growing properly. The doctors have told Ms. Thompson that the pregnancy is not likely to make it to term. Ms. Thompson states that her current goal is to meet her baby alive. Her care team determines it would be appropriate to pursue expectant monitoring and schedule an early induction when the fetus begins to do more poorly, with palliative interventions to manage pain. At 23 weeks, Ms. Thompson is able to meet her daughter after being induced. She chooses to wash and clothe her daughter, and then allows other members of her family to come meet her child before she passes away. Later, she states that she is very grateful for the opportunity she had with her daughter and that she was able to live a life full of love before passing away.

In the event that prenatal screening detects a condition that threatens either the mother's or fetus' health, doctors will need to change the direction of prenatal care based on the family's wishes. It is possible to monitor the pregnancy using cardiotocography and ultrasound until labor can be induced, which results in a variety of outcomes. Some fetuses will make it to a term birth, while some pregnancies will result in miscarriage or stillbirth. In other cases, those choosing to induce a preterm labor may result in long-term survival of the child – depending on his or her condition – or death in the infantile period.<sup>76</sup>

While there is a paucity of information in the literature, there is evidence that women will choose an early induction, or C-section, to “meet their baby alive.” In one study carried out for those expecting a child with either Trisomy 13 or Trisomy 18, women who expressed a desire to meet their child were more likely to choose expectant monitoring and C-section than those who did not state that goal.<sup>77</sup>

In situations such as these, providers trained in palliative care are able to tailor their interventions to align with the patient's goals. In the case of Ms. Thompson, she is given the opportunity to meet her daughter alive, which was very important to her. When a genetic counselor is presented with a client whose fetus is facing a poor outcome, it may be to their benefit to discuss a birth plan with a perinatal palliative care team.

## ROLEPLAYS

### **Roleplay 1: Prenatal diagnosis**

Deb Daniels, a 31-year-old woman, is pregnant with her first child. She had noninvasive prenatal screening performed at 14 weeks, which came back positive for Trisomy 13. An ultrasound performed at 18 weeks revealed several congenital anomalies including a VSD, cleft lip, and spina bifida. According to Mrs. Daniels, the physician who saw her after the ultrasound told her that this confirmed a diagnosis of Trisomy 13 and strongly urged her to have an abortion so her child would not have “a life of misery.”

Soon after, Mrs. Daniels was referred for genetic counseling. She brings her husband, Dean, along to the appointment. Both are very concerned about the pregnancy. Mr. Daniels states he has been reading quite a bit about Trisomy 13 online and is worried about the quality of life their child would have if they were born. Mrs. Daniels has declined an amniocentesis because of the increased risk of having a miscarriage. The couple is very conflicted about whether terminating the pregnancy is the best option. Mrs. Daniels states, “I wish that nature would make the decision for me sometimes, you know? We are both worried about our child suffering, but we have also wanted to meet them and hold them since I found out I was pregnant. But, we’re just not sure if that’s even possible after what that physician said.”

As the counselor, what techniques can you use to help the Daniels make a decision? What options would you present to them and how?

### **Roleplay 2: Communication**

Caleb Coulter was born with hypoplastic left heart syndrome. His parents, Cassidy and Carson, elected for corrective surgery. He was immediately transferred to the CICU at the local Children’s Hospital after birth where he underwent surgery. Unfortunately, Caleb has not been doing well. He has had several cyanotic episodes and one pneumothorax, which was caused by a pulmonary bleb rupture. Several healthcare providers have consulted with the Coulters since the start of Caleb’s stay and they have started to become frustrated because it appears that the medical teams are not communicating with one another. For instance, the cardiologist came by a few days after Caleb’s initial surgery and told the family he would be by later to remove Caleb’s stitches. However, a resident had performed this task the day before. The cardiologist also told the family that Caleb would not have to undergo surgery to remove the affected part of his lung, but the pulmonology team told them he would need to have the bleb removed.

You meet with the Coulters to see how they are doing. Mrs. Coulter is obviously very angry and seems unwilling to speak. She states, “This hospital is a joke. Nobody seems to know what is going on with my baby.” Mr. Coulter adds, “I knew we should have taken him elsewhere! I am going to have to start making some phone calls.”

What can you offer to the family to alleviate these tensions? How do you get the medical team on the same page?

### **Roleplay 3: Early induction**

Millie and Matthew Martin are expecting their third child. They have recently been given the news that their child has severe gastroschisis, and surgery would most likely not have a favorable outcome. Due to their religious beliefs, they are against terminating the pregnancy, yet greatly feel that they cannot continue through the pregnancy knowing their child will die and have discussed premature induction followed by palliative intervention. They discussed the matter with a church leader, but were told that, “God would want them to carry the baby to term.” In meeting with you, the Martins ask if they are truly being selfish if they choose to end the pregnancy early so as not to draw out their own grief.

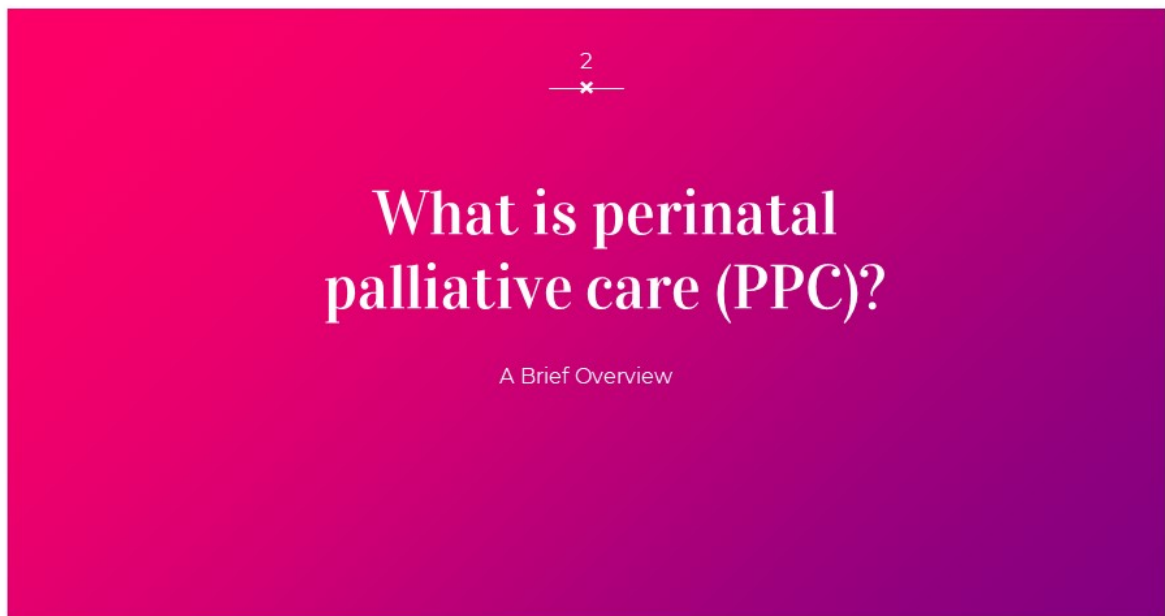
Practice discussing the matter with them and making a referral to perinatal palliative care.

Think about additional supports for the family.

## Case Studies/Roleplays Bibliography

1. Leong Marc-Aurele K, 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.
2. Breeze ACG, Lees CC, Kumar A, Missfelder-Lobos HH, Murdoch EM. Palliative care for prenatally diagnosed lethal fetal abnormality. *Arch Dis Child Fetal Neonatal Ed* 2007;92(1):F56-8. doi:10.1136/adc.2005.092122.
3. Denney-Koelsch E, Black BP, Côté-Arsenault D, Wool C, Kim S, 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.
4. 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* 2017. doi:10.1136/archdischild-2017-313183.
5. Kukora S, Gollehon N, Laventhal N. Antenatal palliative care consultation: implications for decision-making and perinatal outcomes in a single-centre experience. *Arch Dis Child Fetal Neonatal Ed* 2017;102(1):F12-F16. doi:10.1136/archdischild-2016-311027.
6. Richards CA, Starks H, O'Connor MR, et al. When and why do neonatal and pediatric critical care physicians consult palliative care? *Am J Hosp Palliat Care* 2017;1049909117739853. doi:10.1177/1049909117739853.
7. Allen JS, Mulhauser LC. Genetic counseling after abnormal prenatal diagnosis: facilitating coping in families who continue their pregnancies. *J Genet Couns* 1995;4(4):251-265.
8. Rosenbaum JL, Smith JR, Zollfrank R. Neonatal end-of-life spiritual support care. *J Perinat Neonatal Nurs* 2011;25(1):61-9; quiz 70. doi:10.1097/JPN.0b013e318209e1d2.
9. Bijma HH, van der Heide A, Wildschut HIJ. Decision-making after ultrasound diagnosis of fetal abnormality. *Eur Clinics Obstet Gynaecol* 2007;3(2):89-95. doi:10.1007/s11296-007-0070-0.
10. Catlin A, Carter B. Creation of a neonatal end-of-life palliative care protocol. *J Perinatol* 2002;22(3):184-195. doi:10.1038/sj.jp.7210687.
11. Darmstadt GL, Yakoob MY, Haws RA, Menezes EV, Soomro T, Bhutta ZA. Reducing stillbirths: interventions during labour. *BMC Pregnancy Childbirth* 2009;9 Suppl 1:S6. doi:10.1186/1471-2393-9-S1-S6.
12. Winn P, Acharya K, Peterson E, Leuthner S. Prenatal counseling and parental decision-making following a fetal diagnosis of trisomy 13 or 18. *J Perinatol* 2018. doi:10.1038/s41372-018-0107-x.

**POWERPOINT SLIDES**



3



## First, defining perinatal

- Perinatal is most often defined as any pregnancy past 8 weeks of gestation to the first 28 days of life
- The term is used mostly as a statistical term to tabulate fetus and infant death
  - Often, infants (children <365 days old) are included in perinatal statistics, so it is possible that the above definition could be expanded

4



## Defining PPC<sup>1,2,3</sup>

- PPC is a comprehensive interdisciplinary model of care for women and their families after the diagnosis of a life-limiting fetal condition
- It is holistic and extensive care for an infant who is not going to “get better”
- PPC presents a new paradigm that emphasizes living fully in the time that families have with their baby

## In Short,

- PPC is comprised of a diverse team of professionals whose focus is to provide medical care to critically ill fetuses and infants, as well as psychosocial support to their family members based on family goals.
- PPC is offered to fetuses and infants with either a life-limiting (will cause premature death) or life-threatening (may cause premature death) condition
- Services may be provided in a hospital, clinic, or in the patient's home
- It does not mean all children in these services are not receiving curative treatment

## The Distinction Between Palliative Care and Hospice Care



All hospice care is palliative care, but not all palliative care is hospice care



## History of PPC<sup>4,5</sup>

- Over the past few decades, the medical community has begun to recognize the importance of palliative and end-of-life care for fetuses and infants
- It is thought that PPC began in the 1970s with therapeutic obstination (extension of life with little attention focused on quality of life)
- The first mentions of PPC in the literature were made in 1982, when physicians began to advocate for the right to the same “natural death” that adults had
- Early methods of PPC were based on adult hospice care, which had started two decades earlier

## Total Pain

- Dame Cicely Saunders is credited with starting the hospice care movement in the 1960s after noticing her cancer patients had needs that were not being met by current standards of care
- She defined the concept of “total pain,” which recognizes the emotional, spiritual, religious, and physical aspects of pain and suffering
- Much of her work centered on adult hospice, but she later advocated for child hospice



## PPC Structure and Services

### PPC Structure<sup>6</sup>

- Although it has been almost 50 years since PPC initiated, there is no formal model or structure of care amongst programs
- One study identified eight domains of care through surveying the centers throughout the United States:
  1. Structure and processes
  2. Physical aspects
  3. Psychological and psychiatric aspects
  4. Social aspects
  5. Spiritual, religious, and existential
  6. Cultural aspects
  7. Care of the imminently dying patient
  8. Ethical and legal aspects

## 1. Structure and Processes

- Most programs provide care through the use of an interdisciplinary team, assist families in constructing care goals in a safe environment, and document patient interactions using an electronic medical record system
- When it comes to training:
  - 41% of programs report that all providers have formal training
  - 54% had some training
  - 5% had no training
- Training may come in the form of board certifications, formal training such as fellowships, or online resources/courses

## 1. Structure and Processes<sup>6</sup>

- The most common disciplines of program practitioners:

Spiritual Care	<b>89%</b>
Medicine	<b>87%</b>
Nursing	<b>84%</b>
Social Workers	<b>83%</b>
Hospice Providers	<b>73%</b>

- Most programs (83%) have care coordinators, which may be nurses, social workers, genetic counselors, physicians, and child life managers

## 2. Physical Aspects of Care<sup>6,7</sup>

### Prenatal

- Parents may opt for expectation monitoring, which monitors fetal health using cardiotocography and ultrasound
  - When the fetus begins to do poorly, families may choose early induction or C-section to meet their child alive
- Creating a birth plan helps direct physical care goals
- Prenatal genetic testing may be performed to determine if a specific diagnosis exists

### Postnatal

- Medical intervention may remain minimal, such as administering pain medication to maintain comfort and supplying oxygen with nasal canula
- Or interventions can be more invasive, such as use of a breathing tube and surgeries to correct birth defects
- The intensity of interventions chosen is influenced by when a diagnosis is made, with parents receiving a postnatal diagnosis to be more likely to choose more intense care

## 3. Psychosocial and Psychiatric Aspects of Care<sup>6,8</sup>

- Most programs provide emotional support, which may include:
  - Referral to support groups (93%), to a community of parents (80%), or counselors (89%)
  - Memorial services (80%)
  - Follow-up home visits (39%) or phone calls (45%)
  - Condolence cards (93%)
  - Assistance with memory making (100%)
  - Information about anticipatory grief (93%)
  - Provision of bereavement services (97%)
- Memory making through mementos has been shown to facilitate the grieving process
  - This may include making footprints and handprints, photography, saving a lock of hair, and recording the fetal heartbeat

## Psychosocial Needs of Families in PPC<sup>9</sup>

- Parents experiencing an abnormal perinatal diagnosis may feel conflicted
  - Often, they are simultaneously planning for the birth and death of their child
- When their child is admitted to the NICU, it can be an isolating experience for parents
  - They are not in direct control of their child's care
  - It is difficult to perform normal parenting tasks like feeding and bathing a child if they are connected to machines or fed through a feeding tube
- Supporting these families often involves engaging them in their child's care by teaching them how to perform specific tasks

## Other Aspects of Care<sup>6</sup>

### 5. Social

Many programs help families find parent support groups, either in-person or online, where they can share updates and gain support

### 6. Spiritual, Religious, and Existential

All programs report providing spiritual support, which is usually facilitated by a hospital chaplain or pastoral care provider.

It often includes using religious literature and facilitating rituals (e.g. blessings, prayers, dedications)

## Other Aspects of Care<sup>6</sup>

### 7. Care of the Imminently Dying Patient

Parents are given guidance on what to expect during the dying process.

PPC providers treat the patients with cultural and religious respect.

Parents are allowed to parent their child through bathing, holding, comforting their baby, etc. and participate in memory making.

### 8. Ethical and Legal Aspects

55% of programs have established ethical and legal guidelines

95% have an ethics committee that is consulted when providers and families face difficult end-of-life care decisions

## Establishing a Birth Plan

- This ensures that the wishes of the family are carried out in the delivery room
  - This takes into account the medical interventions the family does or does not want, their cultural and religious beliefs, and if they want their child to receive regular newborn care among other things
- Birth plans may also include information about the child's funeral
- Sample birth plans can be found at:
  - [www.txpeds.org](http://www.txpeds.org)
  - [www.perinatalhospice.org](http://www.perinatalhospice.org)

## Referring Families to PPC



**“For families experiencing an abnormal prenatal diagnosis, an array of options should be discussed with the family including pregnancy termination, fetal intervention, postnatal therapy, and palliative care.”<sup>10</sup>**

## Who Needs PPC?<sup>11,12</sup>

- There are no formalized guidelines for PPC referral
- Physicians may consult PPC teams when they feel a child's needs are beyond their scope of practice
- Clinicians may consider PPC for any diagnosis when the long-term outcome of the fetus/infant is considered poor, such as:
  - Trisomy 13/18
  - Anencephaly or other severe, neurological conditions
  - Renal tract abnormalities
  - Major skeletal dysplasias

## Who Needs PPC?<sup>13,14</sup>

- 47,041 perinatal deaths were reported in the United States in 2013, but this number is likely to be higher
  - This is mostly due to variation in state requirements for reporting pregnancy loss
- Women desiring termination of pregnancy may also benefit from PPC services through:
  - The creation of pregnancy memories
  - Leading conversations that empower parents to discuss the news of a prenatal life-limiting diagnosis with family and friends

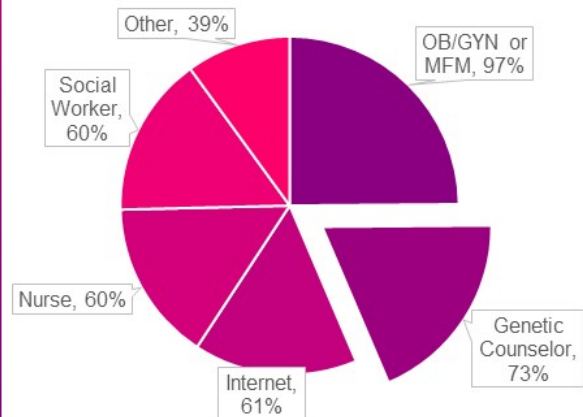


## Who Needs PPC?<sup>15</sup>

- For prenatal diagnoses, studies show that up to 40% of women choose to continue pregnancy after an abnormal diagnosis has been made
- However, families pursuing termination may also benefit from PPC services
  - The focus is shifted to bereavement counseling
  - Parents are presented with the opportunity to make pregnancy memories
- Families should be given information about PPC as soon as possible. It is estimated that there is a 2-month gap between when a diagnosis is suspected and a referral is made.
  - Due to timing issues, families wishing to terminate may not receive PPC services

## How do Families Learn about PPC?<sup>10,15</sup>

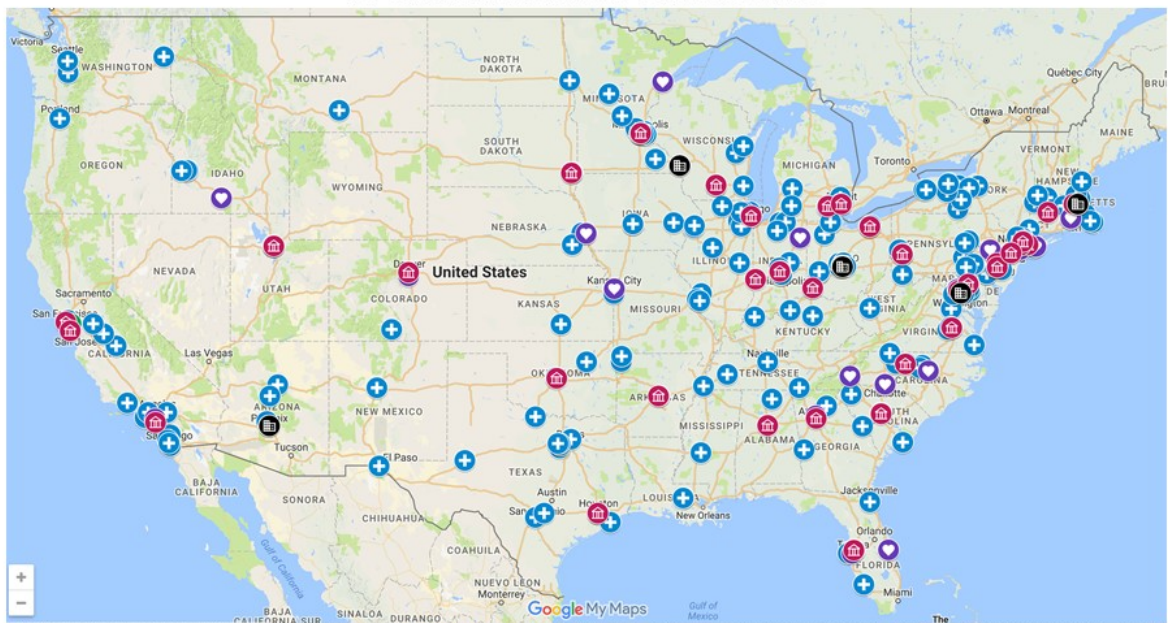
- Families may learn about PPC from a variety of health practitioners, but are most likely to hear about it from OB/GYNs, MFMS, and Genetic Counselors
- There is evidence that many clinicians are uncomfortable discussing PPC and making program referrals
- This suggests that greater awareness may benefit more families



## Making a Referral to PPC

- It is important to be aware of local PPC services when making a referral
- Establish and maintain relationships with clinicians involved in the PPC program
- Discuss prenatal options including PPC with families who have a fetus or infant that has received a life-limiting or life-threatening diagnosis
- In an inpatient setting, the PPC team can be paged to visit a family (if the hospital has a PPC team)

### PPC Locations in the United States



A screenshot of a map identifying PPC programs listed on [www.perinatalhospice.org](http://www.perinatalhospice.org). Key: Blue cross = clinical-based program, Purple heart = support-only program, Red building = genetic counseling program, and Black building = PPC consortium. Created by Meg Hager to fulfill requirements of MPH in Public Health Genetics. To be made available for practitioner use.

## Outcomes and Impact of PPC

### Outcomes of Fetuses and Infants<sup>16</sup>

- One study examined the outcomes of fetuses and infants who received PPC services and those who did not
- There were no significant differences in medical outcomes
  - 24 fetuses and infants were included in each group
  - Each group had 5 fetal losses and 16 infantile losses, with the majority occurring  $\leq 2$ -days of life
  - 3 infants survived to discharge in each group
    - 1 infant passed away after discharge from the PPC group
    - No infants passed away after discharge in the group that did not receive PPC
- This study also found no significant difference between hospital stay length before death or discharge

## Outcomes of Parents<sup>17,18,19</sup>

- Perinatal loss has an affect on both their physical and mental well-being
- Very few studies have been conducted to measure the impact of PPC on families receiving these services and families who are not
  - Few PPC centers have created formal assessment tools to measure parental outcomes
- Several studies postulate that those who receive PPC services have improved psychosocial outcomes than those who do not.

## Impact on Clinicians<sup>20,21</sup>

### **It is “Relentless”**

- It can be difficult to care for patients who inevitably pass away
- Clinicians working with critically ill children are at a higher risk of developing compassion fatigue

### **Positive Aspects**

- Many clinicians find their work to be rewarding
- Some believe they are positively impacting the family

## Compassion Fatigue and Genetic Counselors<sup>22,23</sup>

- Previous studies show that 74% of genetic counselors are at moderate to high risk for compassion fatigue
- One study postulates that certain personality traits may predispose genetic counselors to compassion fatigue including the desire to be in control, seeking acknowledgement, and perfectionism
- Additional risk factors for genetic counselors include trait anxiety, compassion satisfaction, and burnout

## Avoiding Compassion Fatigue<sup>22,23</sup>

- It is important for clinicians to be aware of their emotions in the clinical space
- Compassion fatigue can inhibit the ability to form patient-client relationships
- Techniques that may help reduce compassion fatigue include:
  - Mindfulness
  - Meditating
  - Debriefing with support staff
  - Participating in an enjoyable activity
  - Joining a support group



## References

1. Denney-Koelsch E, Black BP, Côté-Arsenault D, Wool C, Kim S, 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.
2. Catlin A, Carter B. Creation of a neonatal end-of-life palliative care protocol. *J Perinatol* 2002;22(3):184-195. doi:10.1038/sj.jp.7210687.
3. Sumner LH, Kavanaugh K, Moro T. Extending palliative care into pregnancy and the immediate newborn period. *J Perinat Neonatal Nurs* 2006;20(1):113-116. doi:10.1097/00005237-200601000-00032.
4. Silverman WA. A hospice setting for humane neonatal death. *Pediatrics* 1982;69(2):239.
5. Whitfield JM, Siegel RE, Clicken AD, Harmon RJ, Powers LK, Goldson EJ. The Application of Hospice Concepts to Neonatal Care.pdf. *Am J Dis Child* 1982;136:421-424.
6. Wool C, Côté-Arsenault D, Perry Black B, Denney-Koelsch E, Kim S, Kavanaugh K. Provision of services in perinatal palliative care: A multicenter survey in the united states. *J Palliat Med* 2016;19(3):279-285. doi:10.1089/jpm.2015.0266.
7. Janvier A, Farlow B, Barrington KJ. Parental hopes, interventions, and survival of neonates with trisomy 13 and trisomy 18. *Am J Med Genet C Semin Med Genet* 2016;172(3):279-287. doi:10.1002/ajmg.c.31526.
8. LeDuff LD, Bradshaw WT, Blake SM. Transitional objects to facilitate grieving following perinatal loss. *Adv Neonatal Care* 2017;17(5):347-353. doi:10.1097/ANC.0000000000000429.



## References

9. Umberger E, Canvasser J, Hall SL. Enhancing NICU parent engagement and empowerment. *Semin Pediatr Surg* 2018;27(1):19-24. doi:10.1053/j.sempedsurg.2017.11.004.
10. Wool C, Dudek M. Exploring the perceptions and the role of genetic counselors in the emerging field of perinatal palliative care. *J Genet Couns* 2013;22(4):533-543. doi:10.1007/s10897-013-9581-7.
11. Kukora S, Collehon N, Laventhal N. Antenatal palliative care consultation: implications for decision-making and perinatal outcomes in a single-centre experience. *Arch Dis Child Fetal Neonatal Ed* 2017;102(1):F12-F16. doi:10.1136/archdischild-2016-311027.
12. Richards CA, Starks H, O'Connor MR, et al. When and why do neonatal and pediatric critical care physicians consult palliative care? *Am J Hosp Palliat Care* 2017;1049909117739853. doi:10.1177/1049909117739853.
13. MacDorman MF, Gregory ECW. Fetal and perinatal mortality: united states, 2013. *Natl Vital Stat Rep* 2015;64(8):1-24.
14. Neonatal and Perinatal Mortality: Country, Regional and Global Estimates. *Off Rec World Health Organ* 2006.
15. Leong Marc-Aurele K, 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.
16. Courtwright AM, Laughon MM, Doron MW. Length of life and treatment intensity in infants diagnosed prenatally or postnatally with congenital anomalies considered to be lethal. *J Perinatol* 2011;31(6):387-391. doi:10.1038/jp.2010.124.

## References

17. Arlettaz R, Mieth D, Bucher H-U, Duc G, Fauchère J-C. End-of-life decisions in delivery room and neonatal intensive care unit. *Acta Paediatr* 2005;94(11):1626-1631. doi:10.1080/08035250510036543.
18. Broom M, Parsons G, Carlisle H, Kecskes Z. Exploring Parental and Staff Perceptions of the Family-Integrated Care Model: A Qualitative Focus Group Study. *Adv Neonatal Care* 2017. doi:10.1097/ANC.0000000000000443.
19. Baumann-Hölzle R, Maffezzoni M, Bucher HU. A framework for ethical decision making in neonatal intensive care. *Acta Paediatr* 2005;94(12):1777-1783. doi:10.1080/08035250510011928.
20. Taylor J, Aldridge J. Exploring the rewards and challenges of paediatric palliative care work - a qualitative study of a multi-disciplinary children's hospice care team. *BMC Palliat Care* 2017;16(1):73. doi:10.1186/s12904-017-0254-4.
21. Donohue PK, Williams EP, Wright-Sexton L, Boss RD. It's relentless: providers' experience of pediatric chronic critical illness. *J Palliat Med* 2018. doi:10.1089/jpm.2017.0397.
22. Lee W, Veach PM, MacFarlane IM, LeRoy BS. Who is at risk for compassion fatigue? An investigation of genetic counselor demographics, anxiety, compassion satisfaction, and burnout. *J Genet Couns* 2015;24(2):358-370. doi:10.1007/s10897-014-9716-5.
23. Van Mol MMC, Kompanje EJO, Benoit DD, Bakker J, Nijkamp MD. The Prevalence of Compassion Fatigue and Burnout among Healthcare Professionals in Intensive Care Units: A Systematic Review. *PLoS ONE* 2015;10(8):e0136955. doi:10.1371/journal.pone.0136955.

## Credits

- Created by Meg Hager as part of the thesis project, Assessing Perinatal Palliative Care Education in Genetic Counseling Programs, to fulfill the requirement of MS in Genetic Counseling at the University of Pittsburgh
- Approved by Robin E. Grubs, MS, PhD, LCGC, Candace Kammerer, PhD, Scott Maurer, MD, and Meredith Jones, MS, LCGC on May 14, 2018
- Design:
  - Presentation template by SlidesCarnival
  - Stock photographs from Pixabay

**APPENDIX C: MAPS - SUPPLEMENTARY MATERIALS**

**Table 4. State data.**

<b>State</b>	<b># of PPC Programs</b>	<b># of GC Programs</b>	<b>Population</b>	<b>Per Capita Income (\$)</b>	<b>Crude Infant Mortality Rate (per 1,000)</b>
AK	2	0	736,732	33,062	6.6
AL	1	1	4,849,377	23,606	8.7
AR	5	1	2,966,369	22,883	7.5
AZ	4	0	6,731,484	25,715	6.1
CA	19	3	38,802,500	30,441	4.3
CO	2	1	5,355,866	32,357	4.8
CT	1	0	3,596,677	39,373	4.9
DC	0	0	658,893	45,877	7.3
DE	1	0	935,614	30,488	6.7
FL	8	1	19,893,297	26,582	6.1
GA	5	1	10,097,343	25,615	7.5
HI	1	0	1,429,561	29,736	4.5
IA	4	0	3,107,126	28,361	4.8
ID	4	0	1,634,464	23,938	5.5
IL	14	1	12,880,580	30,417	6.6
IN	12	2	6,596,855	25,140	7.1
KS	2	0	2,904,021	27,870	6.3
KY	2	0	4,413,457	23,684	7.1
LA	2	0	4,649,676	24,800	7.5
MA	14	2	6,938,608	36,593	4.4
MD	9	2	5,976,407	36,338	6.5
ME	0	0	1,330,089	27,978	6.7
MI	8	2	9,909,877	26,613	6.5
MN	17	1	5,457,173	32,638	5.0
MO	6	0	6,063,589	26,126	6.1
MS	1	0	2,994,079	21,036	8.2
MT	1	0	1,023,579	25,989	5.5
NC	9	1	9,943,964	25,744	7.1
ND	1	0	739,482	33,071	5.0
NE	4	0	1,881,503	27,446	5.1
NH	1	0	1,326,813	34,691	4.3
NJ	4	1	8,938,175	37,288	4.4
NM	1	0	2,085,572	23,683	5.4
NV	0	0	2,839,099	25,773	5.5
NY	17	3	19,746,227	33,095	4.6



<b>Table 4 Continued.</b>					
OH	19	3	11,594,163	26,937	6.9
OK	2	1	3,878,051	25,229	8.1
OR	1	0	3,970,239	27,646	5.1
PA	16	3	12,787,209	29,220	5.9
RI	2	0	1,055,173	30,830	4.4
SC	3	1	4,832,482	24,596	6.5
SD	2	1	853,175	26,959	5.9
TN	6	0	6,549,352	24,922	6.9
TX	16	1	26,956,958	27,125	5.8
UT	3	1	2,942,902	24,877	4.9
VA	7	1	8,326,289	34,052	5.8
VT	0	0	626,562	29,178	4.6
WA	3	0	7,061,530	31,841	4.5
WI	5	1	5,757,564	28,213	5.7
WV	2	0	5,757,564	22,714	7.0
WY	0	0	584,153	26,698	6.4

**Table 5. Distribution of PPC programs within the United States.**

<b>Number of PPC Programs</b>	<b>States with Programs</b>
<b>0</b>	4
<b>1</b>	10
<b>2</b>	9
<b>3</b>	3
<b>4</b>	4
<b>5</b>	4
<b>6</b>	2
<b>7</b>	1
<b>8</b>	2
<b>9</b>	2
<b>12</b>	1
<b>14</b>	2
<b>16</b>	2
<b>17</b>	2
<b>19</b>	2

## APPENDIX D: INSTITUTIONAL REVIEW BOARD APPROVAL



### University of Pittsburgh *Institutional Review Board*

3500 Fifth Avenue  
Pittsburgh, PA 15213  
(412) 383-1480  
(412) 383-1508 (fax)  
<http://www.irb.pitt.edu>

#### **Memorandum**

To: Megan Hager  
From: IRB Office  
Date: 12/12/2017  
IRB#: [PRO17090536](#)  
Subject: Assessing Perinatal Palliative Care Education within Genetic Counseling Programs

---

The above-referenced project has been reviewed by the Institutional Review Board. Based on the information provided, this project meets all the necessary criteria for an exemption, and is hereby designated as "exempt" under section

45 CFR 46.101(b)(2)

Please note the following information:

- Investigators should consult with the IRB whenever questions arise about whether planned changes to an exempt study might alter the exempt status. Use the "**Send Comments to IRB Staff**" link displayed on study workspace to request a review to ensure it continues to meet the exempt category.
- It is important to close your study when finished by using the "**Study Completed**" link displayed on the study workspace.
- Exempt studies will be archived after 3 years unless you choose to extend the study. If your study is archived, you can continue conducting research activities as the IRB has made the determination that your project met one of the required exempt categories. The only caveat is that no changes can be made to the application. If a change is needed, you will need to submit a NEW Exempt application.

**Please be advised that your research study may be audited periodically by the University of Pittsburgh Research Conduct and Compliance Office.**

## BIBLIOGRAPHY

1. Catlin A, Carter B. Creation of a neonatal end-of-life palliative care protocol. *J Perinatol* 2002;22(3):184-195. doi:10.1038/sj.jp.7210687.
2. LeDuff LD, Bradshaw WT, Blake SM. Transitional objects to facilitate grieving following perinatal loss. *Adv Neonatal Care* 2017;17(5):347-353. doi:10.1097/ANC.0000000000000429.
3. Sumner LH, Kavanaugh K, Moro T. Extending palliative care into pregnancy and the immediate newborn period. *J Perinat Neonatal Nurs* 2006;20(1):113-116. doi:10.1097/00005237-200601000-00032.
4. 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. *BMC Pediatr* 2012;12:25. doi:10.1186/1471-2431-12-25.
5. Wool C, Côté-Arsenault D, Perry Black B, Denney-Koelsch E, Kim S, Kavanaugh K. Provision of services in perinatal palliative care: A multicenter survey in the united states. *J Palliat Med* 2016;19(3):279-285. doi:10.1089/jpm.2015.0266.
6. Denney-Koelsch E, Black BP, Côté-Arsenault D, Wool C, Kim S, 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.
7. Silverman WA. A hospice setting for humane neonatal death. *Pediatrics* 1982;69(2):239.
8. Whitfield JM, Siegel RE, Glick AD, Harmon RJ, Powers LK, Goldson EJ. The Application of Hospice Concepts to Neonatal Care.pdf. *Am J Dis Child* 1982;136:421-424.
9. Arlettaz R, Mieth D, Bucher H-U, Duc G, Fauchère J-C. End-of-life decisions in delivery room and neonatal intensive care unit. *Acta Paediatr* 2005;94(11):1626-1631. doi:10.1080/08035250510036543.
10. Bijma HH, van der Heide A, Wildschut HIJ. Decision-making after ultrasound diagnosis of fetal abnormality. *Eur Clinics Obstet Gynaecol* 2007;3(2):89-95. doi:10.1007/s11296-007-0070-0.
11. Limbo R, Brandon D, Côté-Arsenault D, Kavanaugh K, Kuebelbeck A, Wool C. Perinatal palliative care as an essential element of childbearing choices. *Nurs Outlook* 2017;65(1):123-125. doi:10.1016/j.outlook.2016.12.003.
12. MacDorman MF, Gregory ECW. Fetal and perinatal mortality: united states, 2013. *Natl Vital Stat Rep* 2015;64(8):1-24.

13. Leuthner SR. Palliative care of the infant with lethal anomalies. *Pediatr Clin North Am* 2004;51(3):747-59, xi. doi:10.1016/j.pcl.2004.01.006.
14. Neonatal and Perinatal Mortality: Country, Regional and Global Estimates. *Off Rec World Health Organ* 2006.
15. PUNCHES BE, Johnson KD, Gillespie GL, Acquavita SA, Felblinger DM. A review of the management of loss of pregnancy in the emergency department. *J Emerg Nurs* 2017. doi:10.1016/j.jen.2017.11.001.
16. Kochanek KD, Murphy SL, Xu J, Tejada-Vera B. Deaths: final data for 2014. *Natl Vital Stat Rep* 2016;65(4):1-122.
17. Uhlmann WR, Schuette JL, Yashar B. The Practice of Genetic Counseling. In: *A Guide to Genetic Counseling*. 2nd ed. John Wiley & Sons; 2009.
18. Allen JS, Mulhauser LC. Genetic counseling after abnormal prenatal diagnosis: facilitating coping in families who continue their pregnancies. *J Genet Couns* 1995;4(4):251-265.
19. Wool C, Dudek M. Exploring the perceptions and the role of genetic counselors in the emerging field of perinatal palliative care. *J Genet Couns* 2013;22(4):533-543. doi:10.1007/s10897-013-9581-7.
20. Wallace SE, Gilvary S, Smith MJ, Dolan SM. Parent Perspectives of Support Received from Physicians and/or Genetic Counselors Following a Decision to Continue a Pregnancy with a Prenatal Diagnosis of Trisomy 13/18. *J Genet Couns* 2017;27(3):1-9. doi:10.1007/s10897-017-0168-6.
21. Janvier A, Farlow B, Barrington KJ. Parental hopes, interventions, and survival of neonates with trisomy 13 and trisomy 18. *Am J Med Genet C Semin Med Genet* 2016;172(3):279-287. doi:10.1002/ajmg.c.31526.
22. Richmond C. Dame Cicely Saunders. *BMJ : British Medical Journal* 2005.
23. Saunders C. Distress in Dying. *Br Med J* 1963.
24. Saunders C. Comprehensive Approach to Therapy of Pain. *Proc R Soc Med* 1967.
25. Donohue PK, Williams EP, Wright-Sexton L, Boss RD. It's relentless': providers' experience of pediatric chronic critical illness. *J Palliat Med* 2018. doi:10.1089/jpm.2017.0397.
26. U.S. Department of Health and Human Sciences, Centers for Disease Control and Prevention, National Center for Health Statistics (NCHS). Technical Appendix - Fetal Death. *Vital Health Statistics* 2015;64(8).
27. Barfield WD, Committee on Fetus and Newborn. Standard terminology for fetal, infant, and perinatal deaths. *Pediatrics* 2016;137(5). doi:10.1542/peds.2016-0551.

28. U.S. Department of Health and Human Services, Centers for Disease Control and Prevention (C D C). State Definitions and Reporting Requirements for Live Births, Fetal Deaths, and Induced Terminations of Pregnancy.
29. Wittels KA, Pelletier AJ, Brown DFM, Camargo CA. United States emergency department visits for vaginal bleeding during early pregnancy, 1993-2003. *Am J Obstet Gynecol* 2008;198(5):523.e1-6. doi:10.1016/j.ajog.2007.11.011.
30. Woodgate RL, Edwards M, Ripat JD, Borton B, Rempel G. Intense parenting: a qualitative study detailing the experiences of parenting children with complex care needs. *BMC Pediatr* 2015;15(1):197. doi:10.1186/s12887-015-0514-5.
31. Kukora S, Gollehon N, Laventhal N. Antenatal palliative care consultation: implications for decision-making and perinatal outcomes in a single-centre experience. *Arch Dis Child Fetal Neonatal Ed* 2017;102(1):F12-F16. doi:10.1136/archdischild-2016-311027.
32. Richards CA, Starks H, O'Connor MR, et al. When and why do neonatal and pediatric critical care physicians consult palliative care? *Am J Hosp Palliat Care* 2017;35(6):1049909117739853. doi:10.1177/1049909117739853.
33. Indiana State Department of Health (ISDH). Certification of Provision of Perinatal Hospice Information (Time of Diagnosis of a Lethal Fetal Anomaly).
34. Indiana State Department of Health (ISDH). Certification of Provision of Perinatal Hospice Information (Time of Abortion Consent Decision).
35. Indiana State Department of Health (ISDH): Perinatal Hospice Information Center. Available at: <https://www.in.gov/isdh/27241.htm>. Accessed May 25, 2018.
36. Committee on Healthcare for Underserved Women. Increasing Access to Abortion. *ACOG Committee Opinion* 2014;N. 613.
37. Perinatal hospice and palliative care support. Available at: <http://www.perinatalhospice.org/list-of-programs.html>. Accessed December 13, 2017.
38. Thomas SR, Holmes GM, Pink GH. To What Extent do Community Characteristics Explain Differences in Closure among Financially Distressed Rural Hospitals? *J Health Care Poor Underserved* 2016;27(4A):194-203. doi:10.1353/hpu.2016.0176.
39. Weaver MS. Growing symbiotic local partnerships to nurture quality pediatric hospice care in rural regions: companion planting. *JAMA Pediatr* 2018. doi:10.1001/jamapediatrics.2017.3910.
40. Williams D, Holmes M. Rural Health Care Costs: Are They Higher and Why Might They Differ from Urban Health Care Costs? *N C Med J* 2018;79(1):51-55. doi:10.18043/ncm.79.1.51.

41. Scott DM, Friesner DL, Udem T, Anderson G, Sem K, Peterson CD. Perceived sustainability of community telepharmacy in North Dakota. *J Am Pharm Assoc (2003)* 2017;57(3):362-368.e5. doi:10.1016/j.japh.2017.02.005.
42. Kersting A, Wagner B. Complicated grief after perinatal loss. *Dialogues Clin Neurosci* 2012;14(2):187-194.
43. Rakel RE, Rakel DP. *Textbook of Family Medicine*. 9th ed. Elsevir Saunders; 2016.
44. Umberger E, Canvasser J, Hall SL. Enhancing NICU parent engagement and empowerment. *Semin Pediatr Surg* 2018;27(1):19-24. doi:10.1053/j.sempedsurg.2017.11.004.
45. Broom M, Parsons G, Carlisle H, Kecskes Z. Exploring Parental and Staff Perceptions of the Family-Integrated Care Model: A Qualitative Focus Group Study. *Adv Neonatal Care* 2017. doi:10.1097/ANC.0000000000000443.
46. Taylor J, Aldridge J. Exploring the rewards and challenges of paediatric palliative care work - a qualitative study of a multi-disciplinary children's hospice care team. *BMC Palliat Care* 2017;16(1):73. doi:10.1186/s12904-017-0254-4.
47. Singer LT, Salvator A, Guo S, Collin M, Lilien L, Baley J. Maternal psychological distress and parenting stress after the birth of a very low-birth-weight infant. *JAMA* 1999;281(9):799-805. doi:10.1001/jama.281.9.799.
48. DeMier RL, Hynan MT, Harris HB, Manniello RL. Perinatal stressors as predictors of symptoms of posttraumatic stress in mothers of infants at high risk. *J Perinatol* 1996;16(4):276-280.
49. Baumann-Hölzle R, Maffezzoni M, Bucher HU. A framework for ethical decision making in neonatal intensive care. *Acta Paediatr* 2005;94(12):1777-1783. doi:10.1080/08035250510011928.
50. Campbell AG, McHaffie HE. Prolonging life and allowing death: infants. *J Med Ethics* 1995;21(6):339-344.
51. Courtwright AM, Laughon MM, Doron MW. Length of life and treatment intensity in infants diagnosed prenatally or postnatally with congenital anomalies considered to be lethal. *J Perinatol* 2011;31(6):387-391. doi:10.1038/jp.2010.124.
52. Hinds PS, Oakes LL, Hicks J, et al. Trying to be a good parent" as defined by interviews with parents who made phase I, terminal care, and resuscitation decisions for their children. *J Clin Oncol* 2009;27(35):5979-5985. doi:10.1200/JCO.2008.20.0204.
53. White A. From Comfort Zone to Performance Management.pdf. 2009.
54. Wool C, Northam S. The Perinatal Palliative Care Perceptions and Barriers Scale Instrument©: development and validation. *Adv Neonatal Care* 2011;11(6):397-403. doi:10.1097/ANC.0b013e318233809a.

55. Brock KE, Cohen HJ, Sourkes BM, Good JJ, Halamek LP. Training pediatric fellows in palliative care: A pilot comparison of simulation training and didactic education. *J Palliat Med* 2017;20(10):1074-1084. doi:10.1089/jpm.2016.0556.
56. American Academy of Hospice and Palliative Medicine (AAHPM). ABMS Subspecialty Certification in Hospice and Palliative Medicine. Available at: <http://aahpm.org/certification/subspecialty-certification>. Accessed May 5, 2018.
57. American Association of Colleges of Nursing (AACN) > End-of-Life-Care (ELNEC). Available at: <http://www.aacnnursing.org/ELNEC>. Accessed May 7, 2018.
58. Low A, Dixon S, Higgs A, Joines J, Hippman C. Training to Provide Psychiatric Genetic Counseling: How Does It Impact Recent Graduates' and Current Students' Readiness to Provide Genetic Counseling for Individuals with Psychiatric Illness and Attitudes towards this Population? *J Genet Couns* 2017;27(1):1-11. doi:10.1007/s10897-017-0146-z.
59. Michelson KN, Ryan AD, Jovanovic B, Frader J. Pediatric residents' and fellows' perspectives on palliative care education. *J Palliat Med* 2009;12(5):451-457. doi:10.1089/jpm.2008.0263.
60. Sheetz MJ, Bowman M-AS. Pediatric palliative care: an assessment of physicians' confidence in skills, desire for training, and willingness to refer for end-of-life care. *Am J Hosp Palliat Care* 2008;25(2):100-105. doi:10.1177/1049909107312592.
61. Leong Marc-Aurele K, 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.
62. McMahan 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* 2017. doi:10.1136/archdischild-2017-313183.
63. Breeze ACG, Lees CC, Kumar A, Missfelder-Lobos HH, Murdoch EM. Palliative care for prenatally diagnosed lethal fetal abnormality. *Arch Dis Child Fetal Neonatal Ed* 2007;92(1):F56-8. doi:10.1136/adc.2005.092122.
64. Lee W, Veach PM, MacFarlane IM, LeRoy BS. Who is at risk for compassion fatigue? An investigation of genetic counselor demographics, anxiety, compassion satisfaction, and burnout. *J Genet Couns* 2015;24(2):358-370. doi:10.1007/s10897-014-9716-5.
65. Van Mol MMC, Kompanje EJO, Benoit DD, Bakker J, Nijkamp MD. The Prevalence of Compassion Fatigue and Burnout among Healthcare Professionals in Intensive Care Units: A Systematic Review. *PLoS ONE* 2015;10(8):e0136955. doi:10.1371/journal.pone.0136955.
66. Healthier mothers and babies. 1999:849-858. Available at: <https://www.ncbi.nlm.nih.gov/pubmed/10563522>. Accessed April 21, 2018.

67. Dzingina MD, Higginson IJ. Public health and palliative care in 2015. *Clin Geriatr Med* 2015;31(2):253-263. doi:10.1016/j.cger.2015.01.002.
68. Verberne LM, Kars MC, Schouten-van Meeteren AYN, et al. Aims and tasks in parental caregiving for children receiving palliative care at home: a qualitative study. *Eur J Pediatr* 2017;176(3):343-354. doi:10.1007/s00431-016-2842-3.
69. Accredited Programs. Available at: <http://gceducation.org/Pages/Accredited-Programs.aspx>. Accessed December 13, 2017.
70. Census.gov. Available at: <https://www.census.gov/>. Accessed December 13, 2017.
71. U.S. Department of Health and Human Services, Centers for Disease Control and Prevention (CDC). Underlying Cause of Death, 1999-2015 Request Form. Available at: <https://wonder.cdc.gov/controller/datarequest/D76>. Accessed December 13, 2017.
72. ZIP Codes by State - Find all the zip codes for your state. Available at: <http://www.zipcodestogo.com/ZIP-Codes-by-State.htm>. Accessed April 23, 2018.
73. My Maps. Available at: <https://www.google.com/maps/d/>. Accessed April 23, 2018.
74. American FactFinder. Available at: <https://factfinder.census.gov/faces/nav/jsf/pages/index.xhtml>. Accessed April 23, 2018.
75. Rosenbaum JL, Smith JR, Zollfrank R. Neonatal end-of-life spiritual support care. *J Perinat Neonatal Nurs* 2011;25(1):61-9; quiz 70. doi:10.1097/JPN.0b013e318209e1d2.
76. Darmstadt GL, Yakoob MY, Haws RA, Menezes EV, Soomro T, Bhutta ZA. Reducing stillbirths: interventions during labour. *BMC Pregnancy Childbirth* 2009;9 Suppl 1:S6. doi:10.1186/1471-2393-9-S1-S6.
77. Winn P, Acharya K, Peterson E, Leuthner S. Prenatal counseling and parental decision-making following a fetal diagnosis of trisomy 13 or 18. *J Perinatol* 2018. doi:10.1038/s41372-018-0107-x.