Perinatal Loss Manifested Through the Lens of the Extended Family Unit

Master’s Thesis

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by

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Perinatal loss secondary to a genetic disorder is a complex and multidimensional experience, often felt across the extended family unit, including parents, relatives, friends, and clinicians. Most literature on perinatal loss centers on parents, relying on quantitative methods for analysis. This highly structured perspective is limited, often leaving many questions unanswered. In contrast, this study used the qualitative methodology of narrative analysis to explore how perinatal loss and the grieving process are manifested and reported by different members of one extended family unit. It explored if and how grieving of the extended family unit impacted the narrative, future experiences, and decisions of one set of parents. This approach facilitated expansion upon the personal experiences that drive the narrative course. The goal of this endeavor was to better understand and highlight the complexities of grief following perinatal loss such that future genetic counselors and clinicians can more fully appreciate the scope of extended family grief and provide optimal care. This study addressed the grief process of three cohorts – parents, friends, and clinicians – surrounding the death of twin infants to
Spinal Muscular Atrophy, SMA. Audiotaped interview transcripts were coded for recurring themes, four of which emerged across all cohorts: reacting to diagnosis, coping with loss, finding community, and preparing for future pregnancy. While all parties expressed shock at the diagnosis, each group displayed a unique evolution of the shock response. Seeking information was reported across all groups while displaying cohort-specific patterns and all cohorts reported behavioral changes in response to the grieving process. The results of this narrative analysis suggest that greater cross-disciplinary communication is warranted. By appreciating the ways in which perinatal loss can affect the extended family unit, clinicians may provide a greater level of empathy and compassion as well as more comprehensive clinical care to families facing perinatal loss.
**TABLE OF CONTENTS**

<table>
<thead>
<tr>
<th>Section</th>
<th>Page</th>
</tr>
</thead>
<tbody>
<tr>
<td>Introduction</td>
<td>1</td>
</tr>
<tr>
<td>Methods</td>
<td>7</td>
</tr>
<tr>
<td>Results</td>
<td>9</td>
</tr>
<tr>
<td>Discussion</td>
<td>37</td>
</tr>
<tr>
<td>Conclusion</td>
<td>45</td>
</tr>
<tr>
<td>References</td>
<td>46</td>
</tr>
<tr>
<td>Appendix A: Letter of Introduction</td>
<td>48</td>
</tr>
<tr>
<td>Appendix B: Informed Consent Form</td>
<td>50</td>
</tr>
<tr>
<td>Appendix C: Interview Guide</td>
<td>54</td>
</tr>
</tbody>
</table>
LIST OF FIGURES

Figure 1 11
Figure 2 12
Figure 3 13
Figure 4 15

LIST OF TABLES

Table 1 14
Table 2 14
INTRODUCTION

Background and Significance

Perinatal loss following the onset of a genetic disorder is a complex and multidimensional experience. While many research teams have explored different aspects of the experience of losing a child, the majority of these studies do not integrate the many and varied perspectives of the network of individuals attached to the loss. Because families live with and move forward after the loss and continue to interact, and oftentimes, seek/receive continuing care from their team of healthcare providers, it may be useful to address the varied grief and coping processes of individual members. In turn, medical clinicians, including doctors, genetics professionals, and mental health professionals may be better equipped to perceive and address the needs of the family unit if they have a more complete understanding of the complex manifestations of grief and loss. This study used the technique of narrative analysis to tell the stories of perinatal loss and the experiences of one family unit, including multiple family members and the team of caregivers. The goal was to explore the commonalities, differences, and interactions of each personal narrative to better assist genetic counselors and other medical professionals in meeting the needs of the larger family unit.

Spinal muscular atrophy (SMA) refers to a group of inherited diseases with recognized genetic etiologies that cause progressive muscle degeneration and weakness, eventually leading to death (www.ncbi.nlm.nih.gov). SMA type 1 is the most severe;
children affected with this condition usually die before reaching two years of age (Cobben et al., 2008). Children with SMA type 1 typically show profound muscle weakness and lack of motor development. In later stages of the disease, low muscle tone in the diaphragm and accessory muscles of the chest can lead to ultimately fatal respiratory failure. As with any genetic and/or heritable process, genetic counseling is recommended for families affected by an SMA diagnosis. Specifically, SMA is a recognized genetic disease that follows an autosomal recessive inheritance pattern. With the exception of rare cases of a de novo mutation, both parents of an affected child are obligate carriers for this condition, and each future pregnancy faces a ¼ recurrence risk (Kolb & Kissel, 2011).

Parents experiencing the loss of an infant to SMA have catalogued their personal experience with the disease (Baldwin, 2003). Through personal anecdotes and reporting, these families paint a picture of the shock and confusion at diagnosis, challenges of caring for a baby with a medically and developmentally complex, life altering and life shortening process, pain of neonatal death, and multilayered experience of bereavement (Chad, 2008). Previous research using questionnaires completed by parents after receiving a diagnosis of SMA in their child identified aspects of the counseling process that were both helpful and hurtful to families (Meldrum, Scott, & Swoboda, 2007). While this methodology can shed light on general aspects of the family experience, individual family responses were neither explored nor expanded beyond the level of the basic questionnaire. The questionnaire approach opens the door to addressing the experience of the family unit, but is necessarily limited by the questions posed and avenues unique to each family are not explored. Each family, and each family member, has a story to tell.
that is intimate, complex, evolving and profound. This integrated story is lost through the inflexibility of a questionnaire process. In contrast, this study employed a narrative interview approach that targeted a variety of individual and group experiences shared through evolving conversations with those who experienced the perinatal loss. This study considered both the psychological as well as the sociological context of the perinatal loss experience. The family unit, as well as the entire system (individual, family, and care team), were seen as stakeholders in this process. This methodology provided a flexibility to follow the lead of individual narratives not afforded by questionnaires.

Since no cure exists for SMA, palliative care is often warranted from the time of diagnosis through the child’s inevitable death and beyond. Researchers have surveyed the experience of parents with palliative care and the subsequent availability of bereavement supports (Contro, Larson, Scofield, Sourkes, & Cohen, 2002). The death of a child is a landmark event in the life of his or her parents. Extended family and community are frequently involved in the experience of parents facing a terminal diagnosis and the imminent death of a child and often experience disenfranchised grief. Disenfranchised grief refers to the emotions experienced by individuals outside of the nuclear family, and thus not immediately affected by the loss (Selby et al., 2007). It describes a process where some forms of grief may not be outwardly acknowledged or perceived to be legitimate by the larger society. In addition, grief rights may be viewed hierarchically with respect to mothers versus fathers or to nuclear family versus extended family. This can include extended family members as well as the medical staff involved in the child’s care (Masia, 2010). The loss of a child not only affects the parents, but also the extended family and medical community as these individuals are oftentimes also involved in the
process leading up to and/or following the death. The goal of the genetic counselor in this setting can be to offer information, anticipatory guidance and support when invited into the process of a perinatal loss. By exploring the narratives of not only the couple but also the extended family unit, this study shed light on future counseling considerations.

Narrative analysis is a qualitative research methodology. Reissman describes the narrative as a personal explanation of an experience (Reissman, 1993). A narrative consists of subjective representations of a person’s memories and selective reconstruction of past events. As such, a narrative is not simply a reiteration of a timeline of past events. Narrative analysis is a means of studying the various elements of a personal story to identify themes and key qualities (Reissman, 1993). When a person shares a narrative, there are many factors that can affect what parts of the past are included, the order in which the story is told, and the emphasis a person may place on different past experiences (Reissman, 1993). Medical narrative analysis in particular is a means of building a connection with patients, or engaging in their ongoing story and experience (Charon, 2008). Narrative analysis paints a picture of the past, sheds light on the history, and helps the audience understand both the current circumstances and future direction of those telling their stories. By drawing on the narratives of multiple members of an extended family unit as well as their larger care team, we paint a broader and deeper portrait of the family experience. This study compared and contrasted individual narratives while building a larger, more comprehensive family narrative of perinatal loss.

Previous research has addressed the differences and similarities in the ways mothers and fathers describe or experience grief, specifically with respect to perinatal loss. Schwab describes the higher scores of mothers versus fathers on a Grief Experience
Inventory (Schwab, 1996). Multiple grief scales were addressed, including despair, anger, guilt, and loss of control. Another study by Vance et al compared psychological distress scales in mothers and fathers after the death of an infant with the responses of parents with a healthy child. The distress scales were significantly higher in mothers than fathers in the case of infant loss (Vance, Boyle, Najman, & Thearle, 1995). While these types of studies contribute to the understanding of grief differences and parental roles, responses to the questions were necessarily rigid and limited by a defined scale. In addition, the more open-ended nature of narrative methodology holds the potential to unmask missed opportunities and/or maternal bias in the language or framing of previous research. By exploring the personal narratives of a mother and a father along with other members of the extended family, social, and medical unit, this study adds a depth of understanding to this topic.

**Research Questions**

How does the extended family unit experience and report the grieving process surrounding perinatal loss?

Does the grief process of the extended family unit impact the narrative and future experiences and decisions of parents?

**Research Background**

Chloe and Sophia (all names have been changed to protect identity), monozygotic twin girls, passed away nearly three years ago from SMA type 1. Diagnosed at 4 months old, they died 1 ½ months later just hours apart from each other. Their parents, Helen and
Matthew, have become family advocates at their local hospital and are now deeply involved in sharing their story. I explored the story of Chloe and Sophia through a multidimensional interview strategy. In addition to interviewing Helen and her husband, I interviewed not only the immediate family, but also their extended family and medical network. As such, this study included interviews with one of Helen and Matthew’s close friends as well as Helen’s obstetrician, genetic counselor, and pre-implantation genetic diagnostics team. The transcripts of the narratives of each individual shed light on the key elements and themes that guided each personal experience and how these varied responses may influence the network as a whole and its varied members. Genetic counselors are trained to support the needs of parents dealing with perinatal loss, yet counselors are not trained to draw on the experiences of the larger family structure. The extended family unit may impact the narrative and personal experiences of parents; this study addressed the extended narrative with the aim of bringing relevance and importance to the larger genetic counseling community.
METHODS

Study Design

This study employed a semi-structured conversational interview strategy with all participants. To standardize the content of interviews as well as the manner and order in which the stories were told, the interview guide was very basic and uniform across all subjects. Participants told the story of their experiences with the lives and deaths of Chloe and Sophia. Key issues addressed in the interview guide included the emotional and psychological reaction to the diagnosis and its impact on each individual’s subsequent time and experience with the girls, both at the time and several years later.

The interview guide was structured in a chronological manner, beginning with questions addressing Helen and Matthew’s experience with the pregnancy with Chloe and Sophia, through their daughters’ births, diagnosis with SMA and subsequent deaths, and ultimately through the process of achieving another pregnancy. For each participant, the interview process began at the relevant point in time for that individual and his or her involvement with the arc of this story.

Please refer to Appendix C for the full interview guide.

Sampling and Recruitment
Study participants were recruited through an open letter of introduction (Appendix A). As the study author, I had a pre-existing relationship with Helen dating back several years.

Helen distributed the letter of introduction to members of her family, friends, and medical community prior to my visit to Helen and Matthew’s local area for the interview process. I traveled to the location in February of 2012 to meet with participants, discuss study enrollment, obtain informed consent, and to perform the narrative interview process.

This study was approved by the Brandeis University Institutional Review board under protocol #12079 as human subjects protocol by expedited review in accordance with 45 CFR §46.110 under category (7).

Data Collection Procedures

Each participant was given both the letter of introduction as well as the Statement of Informed Consent (Appendix B). Participants had a choice regarding the level of anonymity he or she wished to maintain during the interview process.

In addition, each participant had the choice to participate in audiotaping, videotaping, or both during the interview process. In all cases, participants selected audiotaping but declined videotaping.

Data Analysis

Each audiotaped interview was transcribed using a professional transcriptional service.
Transcripts of each interview were coded for key and recurring themes among all respondents as a whole, as well as by cohort (parents, friends, clinicians) using Atlas.ti version 6, software for qualitative research analysis.

Structural analysis narrative sequencing allows the listener to infer temporal order of past events from the temporal sequence of clauses in the report of those events (Labov, 1972).

After reading through interviews individually, I made some decisions regarding how to group interviews to reflect a thorough perspective on the experience of the loss and grief process for this family unit. I found that the most comprehensive stories came from the parents themselves, a close family friend, and treated all clinicians (n = 4) as a unified whole. While narratives from all participants were considered and included in the study, direct comparisons and contrasts were drawn from and across these three groupings.
RESULTS

*Interview Participants*

The interview process began with Helen and Matthew, as they serve as the focal point tying together the network of overlapping relationships. Helen and Matthew were interviewed together in their home.

A close family friend, Sara, was also interviewed. Helen and Matthew referenced Sara frequently in the interview process with respect to her recurring role and presence in the arc of their story. Sara was interviewed in a local coffee shop.

I interviewed several members of the medical care team that were involved with Helen and Matthew both during and after the loss of Chloe and Sophia. The Obstetrician, Dr. A, cared for Helen during the pregnancy and delivery of Chloe and Sophia. She was involved in the process of diagnosing Chloe and Sophia with SMA and involved with the family through the time of death. Dr. A was interviewed in her office. The Genetic Counselor (GC) did not meet with Helen and Matthew until several months after Chloe and Sophia passed away. The GC’s role began with explaining the genetics of SMA with respect to the options available in pursuing a future pregnancy unaffected with SMA. The GC was interviewed in her office. The *in vitro fertilization* with pre-implantation genetic diagnosis (IFV/PGD) team, Dr. B and Dr. C, came to be involved with Helen and Matthew when the couple was ready to take action to pursue a future pregnancy. Their role with Helen and Matthew began with preparations for the *in vitro* fertilization
procedure and included arranging proper carrier testing and biopsies of the resultant embryos. Dr. B and Dr. C were interviewed together in Dr. B’s office.

*Relationships Between Study Participants*

Figure 1 displays the overlapping and intersecting relationships between the potential and relevant pool of study participants. Helen and Matthew, as the parents of Chloe and Sophia, stand as the focal element of the narrative process. The family, friend, and medical communities then intersect and overlap with each other. Not all of these participants were able to participate in the interview process. Those who participated are shown in Figure 2.

![Figure 1: Relationships between potential pool of study participants.](image-url)
For evaluation of the common elements and recurring storytelling processes across and including all narratives, a general coding approach was used. Figure 3 shows a chart categorizing the most common elements, or themes, as well as their constituent sub-themes. In addition, the data were categorized and subsequently queried to determine thematic issues by specific cohort. This approach facilitated observance of similarities and differences. Tables 1 and 2 illustrate the breakdown of coding frequencies by research question, overarching themes, and subcodes for each theme. In each table, the most prevalent code responses for all three cohorts are highlighted for emphasis. Each of
these thematic findings is defined and discussed in greater detail and quotations by cohort are provided to exemplify findings.

| Question 1: How does the extended family unit experience and report the grieving process surrounding perinatal loss? |
| Question 2: Does the grief process of the extended family unit impact the narrative and future experiences and decisions of parents? |

<table>
<thead>
<tr>
<th>Reacting to Diagnosis</th>
<th>Coping with Loss</th>
<th>Finding Community</th>
<th>Preparing for Future Pregnancy</th>
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<tbody>
<tr>
<td>Initial Shock</td>
<td>Immediate Reaction</td>
<td>Feeling Ostracized</td>
<td>Apprehension</td>
</tr>
<tr>
<td>Seeking Information</td>
<td>Grieving Over Time</td>
<td>Contributing to Future</td>
<td>Building a Family</td>
</tr>
<tr>
<td>Preparing for Loss</td>
<td>Behavioral Changes</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Figure 3: Summary of response themes and sub-themes across participants.
**Question 1:** How does the extended family unit experience and report the grieving process surrounding perinatal loss?

<table>
<thead>
<tr>
<th></th>
<th>Parents</th>
<th>Friend</th>
<th>Clinicians</th>
<th>TOTALS</th>
</tr>
</thead>
<tbody>
<tr>
<td>Change in behavior</td>
<td>16</td>
<td>1</td>
<td>17</td>
<td>34</td>
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<tr>
<td>Contribution to future</td>
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<td>1</td>
<td>9</td>
</tr>
<tr>
<td>Feeling ostracized</td>
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<td>1</td>
<td>12</td>
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<td>7</td>
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<tr>
<td>Grief over time</td>
<td>41</td>
<td>9</td>
<td>5</td>
<td>55</td>
</tr>
<tr>
<td>Immediate grief reaction</td>
<td>16</td>
<td>6</td>
<td>0</td>
<td>22</td>
</tr>
<tr>
<td>Information seeking</td>
<td>13</td>
<td>1</td>
<td>5</td>
<td>19</td>
</tr>
<tr>
<td>Initial shock</td>
<td>21</td>
<td>4</td>
<td>5</td>
<td>30</td>
</tr>
<tr>
<td>Preparing for loss</td>
<td>13</td>
<td>5</td>
<td>2</td>
<td>20</td>
</tr>
<tr>
<td><strong>TOTALS:</strong></td>
<td>146</td>
<td>26</td>
<td>36</td>
<td>208</td>
</tr>
</tbody>
</table>

Table 1: Breakdown of coding frequencies for Question 1.

**Question 2:** Does the grief process of the extended family unit impact the narrative and future experiences and decisions of parents?

<table>
<thead>
<tr>
<th></th>
<th>Parents</th>
<th>Friend</th>
<th>Clinicians</th>
<th>TOTALS</th>
</tr>
</thead>
<tbody>
<tr>
<td>Apprehension about future pregnancy</td>
<td>20</td>
<td>5</td>
<td>26</td>
<td>51</td>
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<tr>
<td>Building a family</td>
<td>5</td>
<td>2</td>
<td>6</td>
<td>13</td>
</tr>
<tr>
<td><strong>TOTALS:</strong></td>
<td>25</td>
<td>7</td>
<td>32</td>
<td>64</td>
</tr>
</tbody>
</table>

Table 2: Breakdown of coding frequencies for Question 2.
Reacting to Diagnosis

The theme of reacting to diagnosis as a theme encompassed all those responses highlighting an individual’s initial response, the experience of searching for information, as well as the experience of preparing for the loss of Chloe and Sophia. All three cohorts (parents, friend, and the clinicians) commented heavily on the initial shock at the news of the diagnosis of SMA for the girls. The themes of initial shock, seeking information, and preparing for loss are explored both within and across cohorts in the following sections.

Theme: Initial Shock

PARENTS: For Helen and Matthew, initial shock manifested through several phases as they learned more about the future and prognosis for their daughters. When Chloe and Sophia’s health first began to decline, Helen and Matthew initially described a feeling that these complications were both temporary and able to be fixed. Over time, they came first to appreciate the life-threatening nature of these health complications and ultimately the fatal course of SMA type 1. A schematic outlining this emotional response is shown in Figure 4.

![Figure 4: Pathway of initial shock response of parents.](image)

Helen: I took her [Chloe] into our regular pediatrician and I was like, “She’s not gaining any weight. She’s not eating.”
Helen: And he was like, “This is life-threatening. You need to get down to Hospital right now. She needs surgery right now. And I’m thinking, “If this is life-threatening, why aren’t we taking an ambulance?”

Matt: And not knowing anything of it, initially I even thought, “It’s probably just a condition that’s treatable.”

Matt: All of a sudden they pull us in this room and say, “Oh, your daughters, unless you put them on machines, they’re going to die.”

FRIEND: Family friend Sara’s initial response to the diagnosis had less of an adjustment arc as her information regarding the girls’ health status was filtered through Helen and Matt. Sara quickly came to appreciate how far along Chloe and Sophia were in the disease course.

Sara: I just remember that they said anywhere from six months to two years is kind of the lifeline for what they had. And then I went home and Googled and realized that they were already at the end of all the symptoms, there wasn’t going to be a two-year. I mean, every symptom they already have.

CLINICIANS: For the obstetrician, Dr. A, the initial shock at the diagnosis was centered primarily around the novelty of the diagnosis of SMA.

Dr. A: I probably will never see another case of SMA. I mean I’ve been practicing 29 years and this is the first time I’ve seen it.

While the GC and the IVF/PGD team did not meet Matthew and Helen until after the girls had died, and as such could not comment first-hand on their initial response to the diagnosis, the GC spoke about the experience of learning about the diagnosis from Helen and Matt and commented on the gravity of the situation.

GC: I acknowledged their loss and expressed sympathy that it must have been very hard. And how can I help you now- you know, what questions do you have.
The theme of initial shock had different arcs when comparing the responses of the parents with those of friends and clinicians. While the parents showed an evolving response as their appreciation of the prognosis for their daughters gradually worsened (see Figure 4), the other cohorts expressed a strong but unchanging ‘initial shock’ response.

**Theme: Seeking Information**

Matthew and Helen remarked repeatedly on their search for information regarding the future for their daughters, how to find appropriate medical care, and how to explain what was happening to their family. The family friend similarly described a process of searching for information, mostly in a role of supporting the parents. While the clinician cohort also commented on searching for information, these comments centered primarily on information regarding the future of the family after the loss had already taken place.

**PARENTS:** Helen and Matthew both commented on searching for information within the context of defending their actions as responsible parents taking appropriate action to care for their daughters. Matt focused on one instance when Sophia lost consciousness and ultimately called 911 for emergency assistance.

**Matt:** *It definitely frightened me. I was wiggling her. I wasn’t shaking her.*

Helen commented on the lack of information given to her and her husband about the clinical and genetic testing that was performed in the hospital as well as the delivery of the diagnosis of SMA.

**Helen:** *We were getting ready to be discharged from Hospital. We were just trying to do the best we could.*

**Helen:** *I said, “What exactly is that and are you guys testing for it?” And he said, “Why don’t you Google it?” And the first thing that comes up is a*
Wikipedia article, and it says, it gives all the details of SMA-1. And it says, “Mortality is imminent” and “nobody lives past age two.” And I died inside, I’m like, “My baby is going to die.”

Helen: They said, “You have hard decisions to make, and you’ve got to think about what your next steps are.” But we weren’t really given options.

In addition to the search for information about the disease itself, the parents both commented on the struggle to find appropriate medical care for their daughters from clinicians familiar with SMA.

Helen: We did not get a lot of information. They said, “Maybe you should go see a different pediatrician.” And so we set up a pediatrician appointment for that next week, and she had never heard of it. We called my OB, and I was like, “Why didn’t we see this? I was highly monitored, it was a twin pregnancy. Why didn’t we see it?” She didn’t really know about it either at the time. So then we switched down to this other pediatrician, she didn’t know much about it either. So all of the information that we got was from our friends doing what they shouldn’t have done and Googling the shit out of it.

FRIEND: Family friend Sara similarly commented on information-searching, in response to the need for resources expressed by the parents.

Sara: I got on their [FSMA] website and filled out Helen and Matt’s information, yeah, and so they started sending them care packages and stuff. I mean basically they just said, “They’re not going to make it.”

CLINICIANS: Clinician Dr. A recalled the parents contacting her office after the diagnosis of SMA. Dr. A commented on the uncertainty she felt about how the parents were coping with the diagnosis at the time.

Dr. A: Helen was back in a couple of times dealing with a lot of depression and sadness with the diagnosis, and tried a couple of antidepressants even to try to cope with it. And as a physician you never know if people don’t come back or they’re not seeing you because they’re upset with you and angry because it wasn’t something you didn’t diagnose beforehand, or whether they’re just so overwhelmed with everything that’s going on.
Dr. A also commented on her own struggle to understand and appreciate the prognosis for Chloe and Sophia immediately following the diagnosis.

*Dr. A: I don’t know that originally when it was first diagnosed that anybody knew what the course would be. I certainly didn’t understand that the babies would probably die within that first year of life.*

The GC and the IVF/PGD team similarly were not involved with the family at the time of diagnosis, and neither commented on the process of searching for and finding relevant clinical information.

*Theme: Preparing for Loss*

‘Preparing for loss’ encompassed both the emotional and practical experiences leading up to the deaths of Chloe and Sophia. As a theme, this was largely limited to the parents and friend, and clinicians did not comment on this experience.

PARENTS: Helen and Matt remarked several times on the practical considerations regarding preparing for the loss. Practical considerations for the parents centered around legal considerations as well as home health care considerations.

*Helen: And so, within the week we had DNRs for both of them, we had living wills, we had wills for us. It’s like it was just all taken care of: And that same week afterward is when hospice came in, and that was something that the hospital had set up.*

Matt and Helen also commented on their decision to forgo life-support procedures, as well as how that decision brought them together as a couple.

*Helen: I remember sitting at the stoplight at Road and Street, and we both said at the exact same time, “Machines are not how we want our children to live.”*

*Matt: Definitely, we both agreed.*
At the same time that Matt and Helen commented on preparing both legally and medically for the loss, neither commented on the emotional process before death. Instead, both parents commented on the rushed nature of the days and weeks before death and the emotional toll it took on each.

*Matt:* So it was very quick. From diagnosis to death was one month, and there were a lot of panicked moments in there.

*Helen:* We thought Sophia would die first. She was bigger, so she was having more trouble breathing.

FRIEND: Family friend Sara remarked on her role in preparing for the loss of Chloe and Sophia primarily from the standpoint of how she could support the parents in the days and weeks before the girls’ deaths.

*Sara:* I think it was just understood that whatever she needed, whatever they needed, and we just had to hang in there and hope that they could last for longer than what we thought they were going to.

*Sara:* And I just, basically I said to her, “Whatever you need, it’s done. Morning, noon, or night, I mean I literally was at their house after that point even more.”

CLINICIANS: While Dr. A was not involved in care for the girls, she did reflect on her understanding that the parents had the resources in place for proper end-of-life care for their daughters.

*Dr. A:* And so my understanding would be that they [the parents] would be very good to help with making the end of life for the girls as easy as possible.

As with the themes of initial shock and information seeking, GC and the IVF/PGD team did not comment regarding preparations for the loss of Chloe and Sophia as they were not involved in the story at this point.
Coping With Loss

Coping with loss encompasses those reactions and narrative statements describing the experience from the time that the deaths of Chloe and Sophia took place through the months and years that followed, as well as the ways in which the loss did or did not evoke a change in behavior amongst the respondents. Those passages coded as immediate grief reaction were those describing the experience at the time of the loss and through the funeral process. Passages coded as grief over time described those statements describing how the grief process changed or evolved over time. Whenever a respondent reported changing his or her behavior, either personally or professionally, those passages were coded as changes in behavior.

Theme: Immediate Reaction

PARENTS: Helen and Matt’s immediate response was one of denial that the death was happening suddenly. Chloe’s death was sudden and unexpected for both parents. Helen commented on beginning to accept that Chloe had died when police arrived and confirmed the death for her.

Matt: I thought you were playing, I thought you were teasing me or something at first. That’s why I remember that vividly, because you said, “She’s not breathing.” And I know when I said, “No. You’re joking right?” But then I looked a little bit closer at her, and sure enough she was not breathing.

Helen: And I got there in the police car and they said, “No.” And the policeman was just like, “I’m sorry.” And I just kind of stood there.

Matt and Helen both commented on the close timing between the deaths of the two girls in combination with the starkly different circumstances surrounding the deaths.
Matt: That was one of the most difficult times, just to walk into that room and see Chloe on the gurney lifeless.

Helen: Our reaction to Chloe’s death is very different from Sophia’s. For both parents, having the ability to control some of the circumstances surrounding Sophia’s death after Chloe’s sudden death was both meaningful and a means of reducing some of the anxiety about the death. Both parents also commented on having fewer regrets surrounding Sophia’s death compared to Chloe’s death.

Helen: We didn’t want her to be a vegetable, we didn’t want that for either of them.

Helen: And so, we just took it off, and we let her die. And it was so much more peaceful.

Matt: And you held her in your arms, and she passed away.

Helen: And I don’t regret it for a minute.

The contrast between the setting and circumstances around the deaths of the two girls was a frequent and recurring element of both parents’ narratives.

FRIEND: Similarly to the parents, Sara commented on the suddenness and urgency at the time of Chloe’s death as well as the time she spent with Helen in the emergency room.

Sara: All I heard was screaming, like just screaming. So I knew it was different.

Sara: I don’t know what else you do, other than just sit there.

Sara’s recollection of Sophia’s death was similar to that of both Helen and Matt, as Sara commented on the relative peacefulness around the time of death.

Sara: I didn’t see Chloe pass away. I just saw after. And I was there. But I know Helen had said many times that she felt like Chloe’s death was so violent, like just a more violent way, where it wasn’t like that with Sophia.
The similarity in the nature of the narratives for the parents and for Sara may be a reflection of the amount of time Sara spent with the family and a shared construction of those memories.

CLINICIANS: None of the clinicians commented on the experience surrounding the time of the girls’ death. Dr. A couldn’t recall how she came to learn that the girls had died.

*Dr. A: My Office Manager came to me. I think Helen called our office and told us, or it could have been the pediatrician then that was involved with it.*

The IVF/PGD team and GC again did not come into the lives of the parents until after Chloe and Sophia passed away, and as such were not in a position to comment on an immediate grief response to the loss.

**Theme: Grieving Over Time**

PARENTS: When reflecting on how they have each experience the grief process over time, both Helen and Matt reflected on the combination of positive and negative experiences they have had.

*Helen: While we had bad experiences, we also had these amazing experiences where people sort of-- It’s sort of like everybody’s instinct just kicked in a little bit.*

Matt and Helen tended to generalize and apply their experiences with the loss of their daughters and relate it to decisions potentially faced by other families, past or present, as a way to give advice or guidance.

*Helen: And when I read people saying, “Should I ask for my kids’ medical records?” I want to say, “Don’t do it. You’re going to bawl your eyes out. Don’t do it.”*
One element of the evolution of grief over time for the parents was seeking help through grief therapy. Both Matt and Helen commented on the impact of therapy on their readiness and capacity to move through the grieving process.

Helen: I think that that’s one of the best decisions we ever made. We didn’t even let ourselves think about should we or shouldn’t we, do we need to or do we not need to. We just said, “We’ve got to go. If we want to survive, we have to go.”

Matt: We knew if there was going to be any chance that we were going to fully heal through this type of a traumatic event we needed some guidance there. Because there were definitely some dark days through there.

Helen and Matt commented on the efficacy of therapy for both of them multiple times throughout the interview, noting that the prolonged relationship with their therapist was critical in the grief progression.

FRIENDS: Sara described that her experience with grief primarily focused on concern for how Helen and Matt were coping and dealing with moving forward after the girls’ deaths and on searching to find ways to be present to their experience.

Sara: I would just say, “I know I don’t get it, but I’ll sit here and listen until the sun comes up. I mean, I know I don’t get it.” So, I think there for a while it was really tough, because we could only do so much, and we didn’t have the tools or anything to get them past that.

Sara: When I went back to work that first day and saw all the kids I started crying.

In addition to her struggle to find ways to help Helen and Matt through the grief process, Sara commented on the ways in which the experience of the girls’ deaths changed her own outlook and perspective.

Sara: I’m a totally different person, and not for the worse. I’m a totally different person, very appreciative of life.
Sara’s comments indicated that her grief over time has been a combination of striving to help Matt and Helen at the same time that she carried the grief with her in her continuing daily life.

CLINICIANS: Dr. A, the obstetrician, remarked on her struggle to decide how much support to offer to the parents following the loss. She commented that she questioned her personal practice of not attending funerals for patients in this case.

*Dr. A:* We sent flowers to her and I sent both of them a card. But I didn’t go to the funeral. I early in my practice went to the funeral of one stillbirth of one of my patients. It’s a really difficult line to cross sometimes, to figure that one out. And I have not done that since. Maybe that’s right and maybe that’s wrong, I don’t know.

In addition, Dr. A commented on how she continued to wonder about the parents as time passed with respect to how they were doing with handling and moving forward from the loss.

*Dr. A:* She was not back for quite a while to see us, I know that. And I kind of kept praying for them and hoping that if they weren’t seeing me with a pregnancy that they were seeing someone else and were having a healthy pregnancy.

Dr. A’s comments may have reflected regretting her decision not to attend the funeral, as well as her interest in seeing how the parents were adjusting over time. Like Sara, Dr. A. shared her own strong desire for the couple to conceive a subsequent healthy pregnancy. Unlike Sara, however, she did not comment on her sense of whether or not the couple had expressed a readiness or felt prepared to pursue another pregnancy. Since she had not had direct contact with the couple, this desire for Helen to be pregnant was likely independent of a knowledge of the couple’s experience, interest or readiness. Dr. B and Dr. C of the
IVF/PGD team, while not a part of the parents’ lives until months after the girls had died, did comment on their perception of the parents’ grief process.

*Dr. C: They seemed like they had grieved, and they were about as informed as anybody I’ve ever seen.*

While Dr. B and Dr. C could not speak extensively to the extended grief process, they were able to remark on the parents’ journey several months after the loss. Their comment highlighted information perhaps as a proxy for progression in the grief process.

**Theme: Behavioral Changes**

PARENTS: During the weeks and months after the girls’ deaths, Helen and Matt handled stress in different ways. Both remarked on the ways in which dealing with this stress caused them to change specific behavioral patterns. Helen commented on her heavy smoking habit at the time, and Matt reflected on his spiraling alcohol consumption.

*Helen: And I’m not proud of that at all. And sometimes we would sit in complete silence. I mean, I think I chain-smoked a little bit more than you did, like I would light one up against the next.*

*Helen: I’m not going to lie, we weren’t on the same page for everything, specifically after they died.*

*Matt: I was drinking my sorrows away more or less, and it was all coming pouring out, and the person it was getting poured on was her, and that wasn’t fair to her.*

While both parents have subsequently worked towards treating these patterns of behavior, both did reflect on their self-medicating behaviors while coping with the loss. The parents later commented on the efficacy of attending therapy together during the course of the grieving process.
FRIENDS: For Sara, changes in behavior meant finding a means of taking her drive to assist and help the parents to a larger scale through organizing a fundraising benefit.

Sara: We just wanted to raise money. We knew the guy that runs Organization through some mutual friends, and we said, “We want to put a benefit together. We want to spread the word through you guys, so we can get as much people as possible.” We started taking silent auction donations, we got signed Football Team gear, we got massage. And it turned out really well. The girls had passed by the time that it happened, but it turned out really well.

CLINICIANS: Dr. A commented on changing her standard of prenatal care after 29 years as a practitioner to include carrier screening for SMA in direct response to her experience with the family.

Dr. A: She [Helen] would have been counseled about Cystic Fibrosis, but at that point probably not SMA and Fragile X. It was later that I actually started using the pamphlet that has all three of those on it. And, really, Helen's experience with her girls is what brought me to at least mentioning that to everybody, because the carrier rate was higher than I really realized, being one in 40.

It is possible that Dr. A felt some level of guilt or regret over not having offered SMA carrier screening to Matt and Helen, although she did not elaborate further.

Drs. B and C remarked on their own apprehension regarding the genetic testing on the embryo biopsies, which prompted them to take the time to personally oversee the biopsy procedure.

Dr. B: You can imagine how anal we were in the lab during this whole process. Every embryo once it was biopsied was in a separate dish, and we didn’t touch them until we were-- And you and I must have looked at the dishes three different times before we did the transfer to confirm.

Dr. B’s commenting on his own decision to oversee the biopsy procedure, a change from his normal routine, may reflect the high degree of investment he felt in the outcome of the procedure. It might also indicate his own feelings of unexpressed grief about Helen.
and Matt’s loss or represent grief about the cumulative loss of families like Helen and Matt under his care. By definition, all cases of PGD are performed to avoid specific unwanted testable genetic outcomes. However, some proportion of cases may be based on known parental carrier status (without a previous history of pregnancy or infant death) or age associated risks (e.g., advanced maternal age).

**Finding Community**

Finding community encompassed those passages reflecting statements where the individual commented on feeling ostracized or finding community as a result of the illness and loss of the girls. Passages coded as those reflecting feeling ostracized were those where the individual reported feeling a lack of inclusion or membership in a particular community. Passages coded as finding community were those where the individual reflected on finding a way to be involved in or contribute to future progress or improvements in care for those in the broader community.

**Theme: Feeling Ostracized**

PARENTS: Matt and Helen reflected on their choice to pursue hospice care instead of using life supportive machinery and the ways in which they felt judged and without proper support from the medical community.

_Helen: If we had chosen to do a trache [tracheostomy] and CPAP [continuous positive airway pressure] on the girls, I think we would have gotten a completely different level of care. But we chose to not make them live on machines, and therefore we got, I feel like we got this kind of care, like, “Well they’re going to die anyway, so why give them anti-secretion meds or that sort of thing.”_
*Helen:* I was mad because we did very much understand what was going on, but not because somebody had taught us about it but because we had to learn on our own, and because we were making decisions that most people in the medical community do not agree with, and therefore we were not receiving the same type of care.

At the same time that the parents commented on a feeling of receiving suboptimal care and support, they both commented being forced to learn how to care for their daughters at home as they wished. Within the workplace, Matt felt ostracized for his struggle with depression and post-traumatic stress in the time following the loss.

*Matt:* I filed a discrimination suit against the company, because for them to just find out about me having post-traumatic stress disorder and to let me go seemed discriminatory to me.

Helen commented multiple times on the theme of feeling ostracized, such that several years after the loss this feeling was still palpable to her.

*Helen:* But anyway, that was my first beef. And we’ll hit the succession of beefs as we go along.

**FRIENDS:** Family friend Sara did not comment as to any sense, either personal or for the parents, of feeling ostracized. This may have been a reflection of the nature of the questions asked or a reflection of her primarily indirect involvement in health care decisions for the girls.

**CLINICIANS:** While Dr. A did not comment on a sense that she had of the parents feeling ostracized for their decisions regarding hospice and end-of-life care, she did recall the process as one without incident.

*Dr. A:* And so my understanding would be that they [the parents] would be very good to help with making the end of life for the girls as easy as possible.
Of note, Dr. A’s statement that was coded as reflective of grief over time was also coded as possibly reflecting a sense of feeling ostracized or rejected by the family herself.

Dr. A: *And as a physician you never know if people don’t come back or they’re not seeing you because they’re upset with you and angry because it wasn’t something you didn’t diagnose beforehand, or whether they’re just so overwhelmed with everything that’s going on.*

From her statement, it was unclear if Dr. A felt that the parents had not returned to her office for care after the girls’ deaths because of inwardly focused depression or because of a sense of resentment for her OB practice.

GC and the IVF/PGD team did not offer comments regarding a sense of feeling ostracized for the parents. This may have been reflective of the shorter duration of their relationships with the family.

**Theme: Contributing to Future**

PARENTS: Only the parents commented on experiences surrounding finding a sense of community or membership in the grief process. This theme was explored because of the number of times the parents commented on it but was not applicable to the other cohorts.

Helen and Matt both reflected on a duality of wanting to find community in an organization like the Families of Spinal Muscular Atrophy (FSMA) at the same time that their choice to pursue hospice instead of life-support left them feeling at times like obligate community outsiders.

*Helen:* *We don’t relate, I mean, very well at all.*

*Matt:* *I think it comes back to our decision to not keep them alive on life support. Because in the FSMA group there are not- the majority [of parents] choose to keep their children alive.*
Since Helen and Matt felt outside of the FSMA community, they were not beneficiaries of the experience, expertise and support held within this community on how to navigate the particular medical, logistical, emotional, social, and spiritual challenges faced by families affected by SMA. Helen and Matt commented on striving for a sense of belonging to the research and fundraising goals of organizations like FSMA, despite their perceived differences with many of the member families.

*Helen:* We like to do the walk and rolls, and we like to do the fundraising and that sort of thing, because we believe in the research side of things. But as far as a community goes, we don’t fit in at all.

One place where Matt and Helen were able to find a sense of belonging was through involvement in the Family Advocacy Panel at the local hospital.

*Helen:* Matt and I volunteer for Hospital, and we are on the Family Advisory Council, which is a council of parents or grandparents of patients or former patients. And we address issue that patients or families might face while there at the hospital. And we try and fix the system, like we’re constantly reviewing and fixing systems. And it’s actually a really positive group.

*Matt:* Yeah, I think we feel the best in trying to give back through our experience through being on the Family Advisory Council at Hospital.

The lack of narrative from other cohorts surrounding the quest for a sense of community may have been a reflection of the second-hand nature of their involvement with the family and their progression through the disease and associated grieving process.

**Preparing for Future Pregnancy**

Preparing for a future pregnancy encompassed those passages reflecting both the theme of apprehension or nervousness regarding the pursuit of a future pregnancy for the family as well as passages reflecting both the process and implications of building a family for
the parents. Helen and Matt underwent IVF with PGD and became pregnant with twin girls. One fetus is a non-carrier for SMA, and one fetus is an unaffected carrier.

*Theme: Apprehension*

**PARENTS:** Both Helen and Matt described their reaction to the options and decision-making process of moving forward with a pregnancy. For both, ending an affected pregnancy was not an option.

*Matt:* We were still very fragile. We were still healing and going through the grieving process. And we knew even if we were to get through that, to hit a point again where we’d have to make a decision like that, to basically pull the plug on a life like that, it wasn’t something, I think, we knew, it wasn’t something we were willing to take a gamble on.

*Helen:* It’s very ironic though, because we’re both very liberal, pro-choice, all of that, but we just knew that our mental state couldn’t handle that. That was the big deciding factor. It wasn’t, “What’s it doing to either of our bodies?” Or anything like that. It was completely based on our mental and emotional state.

At the same time that Helen and Matt commented on a unified sense of choosing IVF with PGD instead of terminating an affected spontaneous pregnancy, both parents reflected on their general sense of support for the option of termination.

**FRIENDS:** Family friend Sara described her apprehensions about a future pregnancy for Helen and Matt, as well as her hopes for the couple.

*Sara:* I was, “Let’s do it. Yay.” I mean, I wanted them to mentally be ready, I wanted them to be in a good place, and I wanted obviously them to be able to have healthy kids.

*Sara:* I was excited, because they’re good parents and they deserve to have healthy, happy babies. So, I was all for it.
CLINICIANS: Dr. A discussed future options with Helen and Matt briefly around the time that Chloe and Sophia passed away. She reflected on this conversation both as one of helping the parents through the grief process as well as one of simply providing information and options.

Dr. A: We had talked about that definitely, and I had found out supposedly that you could do pre-implantation diagnosis.

Dr. A: A lot of times you almost feel a little guilty bringing it up. I always preface it by saying, “This will never take the girls’ place, and I understand that, but I want you to know that the day that you do start thinking about your future and maybe another baby, that this is available, that this option is available.” Because otherwise there’s a hopelessness that’s involved with it.

Dr. A later reflected on her own personal experience with assisted reproductive technology. During the interview process, this aspect was not explored further but may have been relevant to Dr. A’s narrative.

GC reflected on her role of providing information about the options for a future pregnancy for the family.

So we talked about prenatal testing, we talked about PGD. And they calmly sat here and said, well, you know, we would like to meet with the IVF person, because I never want to go through this again, and I don’t want to have to make choices during a pregnancy.

GC’s comments tended to reflect her impression of the steadiness of the parents’ decisions regarding IVF with PGD and their reasoning for reaching this decision.

Drs. B and C of the IVF/PDG team both commented on the amount of information and processing that the couple had undertaken.

Dr. B: So they pretty much came in knowing what they needed and what they wanted, and we just went through the process of explaining what the logistics were.
Dr. C: They were about as well informed as any couple I’ve seen come in here. They knew exactly what was going on.

Dr. B: We’ve had some other people in similar circumstances who weren’t ready to move on, broke down. But they seemed like they had grieved, and they were about as informed as anybody I’ve ever seen.

Across the clinicians, each individual took the time to comment on his or her perception of the poise and inner strength of the parents.

Theme: Building a Family

PARENTS: Matt and Helen remarked on their feelings as a couple regarding the experience of going through IVF with PGD to achieve an unaffected pregnancy.

Matt: I feel very confident in this, in expecting for these two, and it is because I am very happy with the scientific community, what they’ve been able to do.

Helen: I’m very happy with the decisions that we’ve made up to this point.

Helen commented on how she taught herself to cope through the process of losing her daughters and pursuing the future pregnancy.

Helen: I had this motto when the girls were sick, and I just had to repeat it to myself over and over. “You’ve got this. Just take a minute, because you’ve got this.” That was my motto. If I was feeling overwhelmed with them, “Alright, you’ve got this. Let’s focus. You can do this.”

As a couple, Helen and Matt both commented on the ways in which the experience strengthened them both individually as well as together.

Helen: It’s one of those things like sometimes if we catch ourselves grumping at each other about something stupid we’re like, “Yeah. Why are we grumping at each other? This is a stupid argument. We’ve been through way worse than this.”

Matt: I won’t lie, it affects your life in a way that, because even when I go into work sometimes, it gives you perspective beyond your years.
FRIENDS: Family friend Sara commented on what the unaffected pregnancy meant to her as well as the rest of Helen and Matt’s group of close friends.

*Sara: There are no two people that deserve to have babies more, nobody. So I’m beyond excited. I can’t wait. I think it’s very sweet that they’re going to have twin girls again, and we all joke that she’s meant to carry multiples, that’s what her body is meant for, and they’re just meant to have girls. And I’m excited, very excited.*

CLINICIANS: While not involved in the action steps of achieving the future pregnancy, Dr. A commented on what this pregnancy might mean for Matt and Helen.

*Dr. A: I got the sense that they very much want to be parents, and that that is a major goal in their life, and they were going to do whatever it took to probably be parents. I tried to share personal feelings and beliefs with them in that the conception and the carrying is only a small part, a very small part of being a parent, and that it’s really about being there for the next 50 years in their life and that that’s what makes a parent, it’s not his genes and your genes.*

Drs. B and C of the IVF/PGD team commented on the sense of fulfillment each felt after being able to help achieve a pregnancy unaffected with SMA for Matt and Helen.

*Dr. B: I mean from a provider perspective, I guess it’s one of those things where you really feel like it’s the ultimate act of medicine or act of care where you’re stopping disease before it happens. There’s not too many other areas of medicine where that is possible.*

*Dr. C: We’re just delighted for-- I mean you’re delighted for everybody when they come through here.*

*Dr. B: And I think the issue is people just don’t realize. I mean, even providers don’t realize, providers who are in the field don’t even realize.*

*Dr. C: This one was- when you know you have the disease state there and you can, I mean that’s what the PGD was made for was a couple like this. Yeah, it was very rewarding for everyone, I would say.*
While not involved in the process of undergoing IVF and PGD with Helen and Matt, GC commented on her interpretation of the future pregnancy and what it meant both to the couple and to her as a provider.

GC: You don’t know, when you first meet people, if they would be willing to pursue assisted reproduction, if they were just willing to roll the dice again.

GC: This is a unique sort of situation. We do our work as genetic counselors, and we intersect people’s lives, only fleetingly, and really hardly ever do I get any feedback about was it helpful. They did go on and pursue the action steps that I threw out there. So by that measure, I say yes, it must have been valuable information. And I knew that they got it. They’re very smart people. So I felt like there wasn’t any conflict in the decisions that they were given. They were given some ways to move forward.
DISCUSSION

This study set out to determine if or how there are differences and similarities in an extended family grief response and how these responses might impact future decision-making regarding subsequent pregnancies. Thus, this section of the thesis summarizes and interprets the key findings, highlights how each of the study questions were or were not answered, discusses learning and implications/recommendations of findings, and offers commentary on the narrative process.

Interpretation and Significance

While all three cohorts necessarily responded to and reflected on their experiences surrounding the time leading up to and following the loss of Chloe and Sophia, there were both parallels and differences not only in the response themes, but also in their selective reconstruction of past events and the emphasis each cohort placed on different themes.

When faced with the initial diagnosis of SMA for the girls, all three cohorts expressed a sense of shock or disbelief. For the parents, this sense of shock was one that showed an evolving course of realizing how devastating this diagnosis would prove to be. Their reaction to the diagnosis showed a step-wise adaptation to the gravity of the medical information presented to them, as they came to terms with the reality of the fatal course of SMA type 1 (see Figure 4). The evolution of the parents’ appreciation for and
understanding of the gravity of the diagnosis could have been a reflection of the step-wise acquisition of relevant medical information they experienced over the course of their daughters’ health declines. Alternatively, this evolution could have been a reflection of a means of self-preservation, or of a level of denial or reluctance to accept the inevitably fatal disease course. The change in the parents’ emotional state in reacting to the diagnosis could also have been a combination of these two processes and/or other as yet unexplored factors.

For the family friend, her sense of shock was largely similar to the parents yet at the same time was necessarily filtered through the parents and the ways in which they chose to process and share information with their group of friends. For the cohort of clinicians, reaction to the diagnosis was similarly one of shock. This shock was not centered as much around a personal connection to the family but rather to the relative rarity of the clinical diagnosis.

Dr. A commented that in her 29 years as a practitioner in a relatively large hospital setting, she had never seen a case of SMA before. Given that the carrier rate for SMA is estimated to be 1 in 35 for Caucasians, there may be a likelihood that previous cases of SMA had gone undiagnosed. These young deaths might have been attributed to sudden infant death syndrome (SIDS) or even to cases of child abuse. While some initial and speculative research indicates that cases of infant death due to SMA may have been incorrectly attributed to SIDS, these studies are in need of further and more extensive exploration (Ogino et al 2002). Dr. A commented on the change in her own clinical practice to begin offering SMA carrier testing to all prenatal patients. As more clinicians
begin to offer SMA carrier testing to patients, it may be interesting to explore if or how the rate of clinical diagnosis for SMA with respect to SIDS may change.

The theme of seeking information was one that manifested across the three cohorts as well. The parents were largely driven to seek information on their own terms after struggling to find appropriately trained clinical care. The search for information was targeted both towards clinicians as well as to the Internet. When the parents felt that the information provided by clinicians was suboptimal, they turned primarily to Internet searches for help. While there can be valid and helpful information available electronically, there can also be a litany of sources not vetted by properly trained teams. Similarly, one may face an overwhelming wealth of information while facing a dearth of individuals available to explain that information in a meaningful and informative manner. Helen commented on her focused and negative memory of the medical resident who directed her to the Internet to search for the meaning and implications of Werdnig-Hoffman disease. This fleeting statement from a trained medical professional impacted the emotional course of the parents both immediately as well as years later.

The family friend also reflected on her search for information as a means of helping the parents through the time of diagnosis. It was largely the parents’ group of friends who found and provided the clinical and support resources that the parents sought and ultimately ended up using. Between the parents and the family friend, resource and information seeking was a recurring goal and theme of these cohorts’ narratives. I would suggest that future medical providers, be they physicians, genetic counselors, nurses, or beyond, be aware of the implications of directing patients towards Internet searches in a
setting where patients cannot ask questions or assess the validity of the plethora of information they will find.

For the cohort of clinicians available for interview, the theme of seeking information again manifested in a different course than the parent and friend cohorts. Clinicians can often be involved in the lives of families for only short periods of time, with little or no continuity or closure available for the outcome. Because the parents sought care at a large medical facility, the course of the family’s progress over time was sometimes shared by clinicians. Dr. A and GC both reflected on the almost celebrity status that this family shared in the hospital as a product of the severity, sadness, and uniqueness of the situation.

The changes in behavior evidenced across the cohort of clinicians demonstrated that providers might stand to benefit from a broader sharing of information. Dr. A changed her prenatal genetic testing practices after her experience with the family. GC for the first time coordinated parent and grandparent genetic testing and the associated prenatal options for the family. Drs. B and C deviated from their usual practice and took a personal interest in overseeing the outcome of this case. While all three of these clinician groups practiced within the same hospital system, they rarely crossed paths other than to transfer care. A more inclusive discussion of the ways in which a case like this can affect clinician practice may bring together providers, shed light on practice choices, or inform future caregiving decisions, and could potentially have a positive impact on the quality of care afforded to patients. For this case in particular, in the very least a discussion at a Morbidity and Mortality conference would be warranted to discuss the ways in which Chloe and Sophia’s diagnoses and deaths unfolded. From a larger psychosocial
perspective, this case and undoubtedly other cases may offer insight into the ways in which information-giving, decision-making, and interpersonal relationships respond to and are affected by perinatal medical crises.

This study set out to expand upon and delve deeper into the grieving process than was previously capable through the questionnaire approach. While only a limited expedition into the extended family grieving process, this study offered participants the chance to expand upon personal experiences and to personally drive the course of the narrative. While using the same interview guide across study participants served to normalize the process across cohorts, each participant’s response was highly personal and driven by self-reflection and individual reconstruction and interpretation of past events. Each cohort in this study was affected by the grieving process, regardless of whether the individual’s involvement with the family began years before Chloe and Sophia were born or years after they died.

I would recommend a greater incorporation of cross-discipline discussion of the impact of perinatal loss in the medical field. Clinicians need to be aware of the ways in which such a loss can affect not only parents but also extended family members, friends, and other members of the medical community. Such an awareness can only breed a greater level of empathy and compassion while also highlighting and normalizing the grieving process.

**Study Limitations**
This study was necessarily but unfortunately limited by a small sample size. The narrative interview process is, by its very nature, a labor-intensive foray into a microcosm of a larger picture.

If possible, it would have been ideal to incorporate more family members into the narrative process. Helen’s father was a key contributor to the family’s decision-making process and experience with the lives of Chloe and Sophia. Unfortunately, he passed away before the interview process took place. The remaining grandparents preferred not to participate in the interview process.

Individual narratives necessarily change and evolve over time. To have the opportunity to spend time with and obtain the narratives of individuals both at the time of and after the experience of perinatal loss could contribute additional valuable insight into the perinatal grief process.

Another potential study limitation was my existing personal relationship and shared personal history as a mother of multiples with Helen. I was familiar with some aspects of the parents’ story before the interview process began. As a result, I was not able to be completely objective and shared some of my experiences with study participants. However, Ellis and Berger (2001) state that researcher involvement can help subjects feel more comfortable sharing information. They refer to this type of interviewing as “collaborative interviewing” and discuss how it can enrich the research.

“*The interviewer typically shares personal experience with the topic at hand, or reflects on the communicative process of the interview. The interview is conducted more as a conversation between two equals than as a distinctly hierarchical, question and answer exchange*” (Ellis and Berger, 2001).
Overall, I believe my familiarity with the field was a positive aspect of this study rather than a negative. In addition to my willingness to share some personal stories, I found that my experience in parenting, raising multiples, and receiving the same medical care gave me a certain level of credibility and trust among the interviewees. My experience as a student of genetic counseling also helped with access to the interview sites.

**Future Directions**

Addressing the network of integrated experiences surrounding perinatal loss has the potential to contribute valuable psychosocial elements to future caregiver practices. This study approached the experiences of one family unit and associated family, social, and medical providers. At the same time, a number of limitations and questions were raised. By expanding the approach to the family narrative, future providers can integrate these insights and continue to provide patient-specific and individualized care both to the nuclear family as well as to those providing family and social support surrounding the experience of pediatric loss.

This study was limited in the number of members of the family, friend, and clinician cohorts available for interview. Future studies could benefit by an expansion of this cohort design by including more individuals in order to paint a broader and more inclusive picture of the evolving narrative.

Another possible direction for future research would be the comparison of narratives across similar cohorts in different cases. While this study addressed the grieving process in one extended psychosocial setting, it did not answer the question of
how different parents, different social networks, or different clinician groups might respond to a loss in an otherwise different setting. Some factors to consider in comparing the narrative process could include:

- How does the nature of the perinatal loss impact individual narratives? Is the narrative in response to a genetic condition different from other narratives?
- Do socioeconomic factors play a role in the way a narrative is structured or evolves?
- A variety of clinicians may or may not be involved in caring for families facing perinatal loss. How does the structure of the care team impact narrative experiences?
- What role does time play in the evolution of a narrative? Is there a difference between narrative responses over time?

This study was an overture into the possibilities that narrative analysis can bring to the field of genetic counseling and perinatal loss. The possibilities for deeper exploration of the narrative process warrant future forays into this multilayered process.
CONCLUSION

This study explored ways in which both individuals and networks of related individuals respond to the grieving process surrounding perinatal loss.

By using the approach of narrative analysis, this study expanded upon previous research employing primarily questionnaire-based methodologies to the grief process. This study addressed the loss of twin girls to SMA type 1 and the ways in which the parents, their group of friends, and their group of medical providers reported the process of experiencing perinatal loss.

This study demonstrated that an exploration into the network of interrelated personal narratives surrounding the perinatal grief process can offer insight into the ways in which individuals in different roles report the grieving process.

Although this study had a small sample size and is therefore not generalizable to the larger population, it does begin to suggest some potential changes and needed improvements in the capacity of genetic counselors to provide compassionate, informed, and relevant care through consideration of the ways in which grief can and does manifest across the family network.
REFERENCES


APPENDICES

Appendix A: Letter of Introduction

Greetings,

I am a graduate student in the Genetic Counseling Program at Brandeis University, and for my master’s thesis, I am studying perinatal loss manifested through the lens of one extended family unit. My goal is to gather and analyze the personal narratives of this extended family and their medical team in order to explore and better understand how each has experienced grief and loss following the deaths of their children. I am also interested in how these grief experiences may impact future decision making regarding childbearing for parents and the extended family unit as a whole.

Helen and Matt Pritchett have agreed to share their personal story with me for the purposes of my research project, and have given me your name as a family member, medical provider, or friend integrally involved in the care of their daughters, Sophia and Chloe. As someone personally connected to the Pritchett family, I am writing to ask for your participation in a research interview. Your participation will add a richness of information to Helen and Matt’s story and will help me to better understand how the life and death of their two daughters impacted those around them.

Specifically, I would like to conduct an in person interview with you at a time and location of your choosing. With your consent, the interview will be documented through either videotaping or audiotaping. If you are unable to participate in the interview in person, we can conduct the interview by telephone. This research protocol has been approved by the Brandeis University Institutional Review Board and provisions for protecting privacy and confidentiality have been included.

I am hoping to complete all of the interviews by March 1st, 2012. I will be in touch with you again in one week to inquire if you are interested in participating in this study. You may also contact me by email at steeves@brandeis.edu, or by phone at 617.257.0752.

I appreciate your consideration of this request and look forward to hearing from you.
Sincerely,

Marcie Steeves  
Genetic Counseling Graduate Student  
Brandeis University  
Waltham, MA
Appendix B: Informed Consent Form

BRANDEIS UNIVERSITY
DEPARTMENT OF BIOLOGY
GENETIC COUNSELING GRADUATE PROGRAM

Informed Consent to Participate in Research

Perinatal loss manifested through the lens of the extended family unit

Student Researcher: Marcie Steeves, Candidate for Master of Science in Genetic Counseling, Brandeis University
Principal Investigator: Judith Tsipis, PhD, Director, Genetic Counseling Program, Brandeis University

INTRODUCTION
We are conducting a research study to learn about the ways in which one extended family unit and their health care team has responded to and experienced the loss of a child due to a genetic disorder. This research will add a new dimension of understanding to the existing research on grief and loss in the pediatric setting. This knowledge will provide new information, future insights and perspectives for future genetic counselors to draw upon when counseling families coping with the loss of a child.

You are being invited to participate in this study because of your personal experience with the extended family unit in relation to the loss of their two daughters to spinal muscular atrophy.

Taking part in this research study is voluntary. You can decide to stop taking part in this research study at any time for any reason.

Please read all of the following information carefully. Ask any questions that you have about this research study. Sign this consent form only when you understand the information in it and have had your questions answered to your satisfaction.

If you decide to take part in this research study, you will be asked to sign this form. You will be given a copy of the signed form. You should keep your copy for your records. It has information, including important names and telephone numbers, to which you may wish to refer in the future.

PURPOSE OF STUDY
The aim of this study is to gather and analyze the personal narratives of one extended family and their medical team in order to explore and better understand how each has experienced grief and loss following the death of two sisters to spinal muscular atrophy. The study also hopes to determine if and how these varied responses impact future parental decision-making regarding additional pregnancies.

PROCEDURES TO BE FOLLOWED
This study will be conducted using an open-ended interview guide. Each participant has the choice to maintain confidentiality or to disclose some level of confidentiality through participation in videotaping. Interviews will be recorded using audiotape with the possibility of using videotape. Interviews will typically last one hour and will be conducted in a location of the participant’s choosing. Telephone interviews will be used in the event that face-to-face interviews are not possible. Participants have the option of including videotape in the interview setting. Videotape adds a valuable level of informational and contextual richness to narrative interpretation. The videotape can be performed in a manner that either masks or does not mask personal identity.

The student research plans to use the collected interview transcripts and videotapes to complete a Master’s thesis report that will summarize and highlight the narratives collected. In addition, the narrative data may be incorporated in a future conference presentation and in an educational film highlighting the videotaped narratives.

RISKS
The potential risks associated with involvement with this study include emotional reactions while relating personal experiences. We can stop the interview at any point if you become upset or uncomfortable for any reason. We have a trained counselor, Dita Obler, MS, CGC, available as a resource to you. Ms. Obler can be reached at (617)869-7567 and at dita.obler.cgc@gmail.com.

BENEFITS
There will be no direct benefit to you for your participation in this study. We hope that the personal stories and experiences shared in this study will help genetic counselors and medical professionals provide better support for patients and families in the future.

ALTERNATIVES
An alternative is to not participate in this research study.

PRIVACY AND CONFIDENTIALITY
You will have the option of keeping your identity completely confidential or revealing your identity.

If you wish to keep your identity confidential, all records containing identifying information, such as names, email addresses, telephone numbers, and home or work addresses will be kept strictly confidential during the study. Only audiotaped interviews will be performed. All study related documents and materials (including consent forms, interview transcripts, and audiotapes) will be kept in a secure location accessible only to the student researcher, and any databases containing identifiers will be password
protected using a password known only to the student researcher. Transcripts, interview notes, and audiotapes will be labeled with a coded ID number, which will be assigned to you upon enrollment into the study. If you are quoted or referred to in any written or oral reports of the study, you will be given an alternate name. You will never be referred to by your real name or any other identifying information in any written or oral reports based on the interview.

Optional videotaped interviews may be performed concurrently with audiotaped interviews. Participation in videotaping is voluntary to each participant and can be performed in a manner that either masks or does not mask personal identity.

**PAYMENT**
You will not be paid to participate in this study.

**COST**
There is no cost to participate in this study, other than the time it takes to complete the interview process.

**WHOM TO CONTACT**

If you encounter any problems related to study participation or have questions about the study, you may contact the Student Researcher, Marcie Steeves, at steeves@brandeis.edu or 617.257.0752.

You may also contact the Principal Investigator for this project, Dr. Judith Tsipis, at tsipis@brandeis.edu.

If you have questions about your rights as a research study subject, contact the Brandeis Committee for Protection of Human Subjects by email at irb@brandeis.edu, or by phone at 781-736-8136.

**PARTICIPANT’S STATEMENT**

I have read this consent form and have discussed with Marcie Steeves the procedures described above. I have been given the opportunity to ask questions, which have been answered to my satisfaction. I understand that any questions that I might have will be answered verbally or, if I prefer, with a written statement.
I understand that my participation is voluntary. I understand that I may refuse to participate in this study. I also understand that if, for any reason, I wish to discontinue participation in this study at any time, I will be free to do so.

If I have any questions concerning my rights as a research subject in this study, I may contact the Brandeis Committee for Protection of Human Subjects by email at irb@brandeis.edu, or by phone at 781-736-8136.

I have been fully informed of the above-described study with its risks and benefits, and I hereby consent to the procedures set forth above.

I understand that as a participant in the audiotaped interviews in this study, my identity and data relating to this research study will be kept confidential.

I understand that as a participant in videotaped interviews in this study, my identity may be revealed. I have the option to mask my identity in the videotape session.

By initialing, I have chosen to participate in the following levels of this project. I understand that any level of participation in this project is entirely voluntary:

_____ I agree to participate in the interview process via face to face interview.
_____ I agree to participate in the interview process via telephone.
_____ I agree to have my interview audiotaped.
_____ I agree to release my confidentiality.
_____ I agree to have my interview videotaped.
_____ I agree to have my interview videotape included in a short educational film.

____________________________________________  ____________________________________________
Date                                                Participant’s Signature

____________________________________________  ____________________________________________
Date                                                Student Researcher Signature
Appendix C: Interview Guide

Interview Guide

Tell me about the pregnancy with Chloe and Sophia.
   How did you find out about the pregnancy?
   How did you learn about having twins?
   Did you learn gender prior to delivery?

Please describe your experience with the birth of Chloe and Sophia.
   Who was there/were you there?

What was it like when Chloe and Sophia came home from the hospital?
   What were the first few days like?
   What were the first few weeks like?

Tell me about each of the girls.

How did you find out about Chloe and Sophia’s diagnosis with spinal muscular atrophy?
   How did they get referred to the hospital for assessment?

How was the diagnosis delivered?
   Who was present at the diagnosis?
   What was your reaction to the diagnosis?

What were your primary concerns after the diagnosis?
   Immediately after diagnosis and later

What were your impressions of the doctors, nurses, medical staff you encountered?
   What stood out to you?

What was the next step for you?
   Consultation with family, other clinicians?

What were you told about the future for Chloe and Sophia?
   Lifespan, quality of life?
   Changes to expect?
   Who told you?

How did you relate to your spouse, family, friends, clinicians during this time?
   What was conversation like?
   Were any topics more difficult or easier to discuss?

How did medical care for Chloe and Sophia change after their diagnosis?
   Any differences between the girls?
Did you notice any changes in the behavior or personality of Chloe and Sophia?  
If so, when?  
What kinds of changes did you notice?

How did you talk about what was happening with Chloe and Sophia with others?  
Parents?  
Siblings?  
Friends?  
Coworkers?

Did you talk about the genetics of spinal muscular atrophy?  
Who did you talk to?  
What was conversation like?

Tell me about palliative care for Chloe and Sophia.  
What options were discussed? At home or in hospital? Aggressive or palliative?  
What decision was made?  
Who contributed to this decision?

Please tell me about what medical support Chloe and Sophia needed.  
How did you choose these options?  
Who contributed to this decision?

Was there a point in time where you felt that the girls were nearing death?  
Was this time the same or different for the girls?  
How did you feel during this time?

Tell me about your recollection of Chloe’s death.  
Tell me about your recollection of Sophia’s death.  
What happened after Chloe and Sophia died?  
Tell me about the funeral/memorial services for Chloe and Sophia.  
How did you respond? Family members?

How did family members respond during this time?  
Tell me about the days and weeks after the deaths and funerals of Chloe and Sophia.  
Are you aware if the family sought any support during this time?  
If so, who?

How did the loss impact your daily life?
How did you relate to others after the loss?  

In the months and years following the loss, have there been any settings/situations that remind you of Chloe and Sophia?

Have you talked to other families affected with SMA?  
Tell me about this experience.

How did you feel about pursuing another pregnancy?  
Learning about preimplantation genetic diagnosis?  
Resources sought?

What was your experience with the PGD process?

What might a future pregnancy mean to the family?