# The Zebra in the Room:

## An Exploration of Anticipatory Grief in Families of Children with Mitochondrial Disease and the Role of Social Work

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**CARL Research Project**
In collaboration with
Mito Families Ireland

CARL
COMMUNITY - ACADEMIC
RESEARCH LINKS
University College Cork, Ireland

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- provide civil society with knowledge and skills through research and education;
- provide their services on an affordable basis;
- promote and support public access to and influence on science and technology;
- create equitable and supportive partnerships with civil society organisations;
- enhance understanding among policymakers and education and research institutions of the research and education needs of civil society, and
- enhance the transferrable skills and knowledge of students, community representatives and researchers (www.livingknowledge.org).

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**How do I reference this report?**
Author (year) *Dissertation/Project Title*, [online], Community-Academic Research Links/University College Cork, Ireland, Available from: http://www.ucc.ie/en/scishop/completed/ [Accessed on: date].

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# Declaration of Originality

This is to certify that the dissertation titled: *'The Zebra in the Room; An Exploration of Anticipatory Grief in Families of Children with Mitochondrial Disease and the Role of Social Work'* submitted to the School of Applied Social Studies, University College Cork, in partial fulfilment of the requirements for the award of 'Masters of Social Work', is my own work.

Where the work of others has been utilised within this research, it has been cited and referenced accordingly using recognised academic conventions. This dissertation has been submitted through TurnItIn and any changes necessitated by the originality report generated, have been addressed.

Name: _____

Date: _____

Dedicated to Sophie:

*'Though she be but little, she is fierce'*

Shakespeare; A Midsummer Night's Dream

And in memory of:

Adam and Lucy, Christian and Ellen, Conor, Daithi, Evan, Jack, Katie Rose, Leah, Lily, Lollie, Oran and Sarah.

"Perhaps they are not stars,
but rather openings in heaven
where the love of our lost ones
pours through and shines down upon us
to let us know they're happy."

**(Eskimo Proverb)**

# Acknowledgements

The saying ‘it takes a village to raise a child’ has never felt more applicable to me than throughout this research process! So many people came on board to make this possible and I truly could not have done this without them. To my first year tutor Rachel McCormack for helping me to pull lots of random ideas into a cohesive project and to my second year tutor and supervisor Caroline Shore for keeping me sane and motivated through the lengthy ethics application process, thank you both.

To Kenneth Burns, Anna Kingston and the CARL team for allowing my idea to become a reality and enabling the voice of a small community be heard, I thank you and look forward to many more years of submitting research applications from the other side!

Most importantly, to all in Mito Families Ireland, especially my community liaison partner, Audrey Mc Morrow, your collaboration on this project with me is deeply appreciated and I can only hope I have done justice to you all. In particular, to those who participated in the discussion forum, thank you for sharing your experiences with me, your raw honesty blew me away and extends so much further than this research piece, it has informed who I want to be as a social worker. The title of this research ‘The Zebra in the Room’ is an acknowledgement of the experiences of the Mito community at large. Anecdotes tell us that doctors are trained to think ‘horses’ when they hear hoof beats, but the Mito community always ask for the medical profession to think outside the box and to consider that perhaps it is a zebra they hear, or in other words ‘think rare’. The remainder of the title is, of course, a play on the saying ‘the elephant in the room’, in this instance, the elephant (or zebra as the case may be) are the feelings and experiences such as anticipatory grief that may often go unvoiced or unheard but they remain present demanding attention until they are addressed- which is exactly what this research hopes to do.

To my family and partner, Paul, thank you for putting up with me throughout not only this but the many years of college that went before- I will get a real job soon… I promise!

# Abstract

This study researched the concept of anticipatory grief and used primary research to explore its affects in families of children with Mitochondrial Disease. From there it looked at what the role of social work might be in this context. In doing so, this paper considered the recommendations of the National Rare Disease Plan 2014-2018, as well as literature from foremost academics in the area of grief such as Doka, Rando and Cadell. Combining this theory with the findings of a survey completed by twenty three members of Mito Families Ireland, informed by a discussion forum, the research found that the lived experiences of these participants largely correlate to the current literature. Based on this, a number of recommendations have been made which hope to inform both policy and social work practice going forward.

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# Chapter One

# Introduction

1

IN THIS SECTION:

- Introduction
- Title
- Background
- Rationale
- Aim of Research
- Objectives
- Research Questions:
- Definitions
- My Reflexive Positioning as the Researcher
- Chapter Outline

## 1.1 Introduction

This chapter will introduce the research to be undertaken, locating it within the current available literature. It will present the aims and objectives of the research questions, providing the rationale behind them and the collaboration with the Community Academic Research Links (CARL) initiative. It will then provide a reflection on my position as the researcher and in concluding the chapter, a brief outline of the chapters contained within this project will be provided.

## 1.2 Title

'An Exploration of Anticipatory Grief in Families of Children with Mitochondrial Disease and the Role of Social Work'

## 1.3 Background

The study of grief and loss as a theoretical basis for providing care and interventions to those who have suffered bereavement is a vast area, with a multitude of academics and practitioners informing our understanding of this traumatic life event (Kubler-Ross and Kessler, 2005; McIlwraith, 1998; Worden, 1991). There is, however, a comparative dearth of literature and research surrounding the concept of 'Anticipatory Grief' (Doka, 2006; Rando, 1988; Rando, 2000).

According to The Irish Hospice Foundation (n.d.), approximately, three thousand, eight hundred children in Ireland are currently living with a life limiting illness, three hundred and fifty of which are likely to die in the coming year. Based on this, it is estimated that up to approximately three thousand, five hundred children currently have an unknown or uncertain prognosis and as such, live in families who may experience anticipatory grief. It is unknown what percentage of these children are affected by Mitochondrial Disease.

## 1.4 Location of the Research in Current Literature

The topic of Mitochondrial Disease is rarely addressed in literature in terms of its social and family impact. It has, however, featured heavily in the media over the past year in relation to the ethical approval of the scientific development of 'Mitochondrial Donation', a technique whereby the 'faulty' maternal mitochondrial DNA (mtDNA) is replaced with healthy donor mitochondrial DNA in a process similar to IVF (Human Fertilisation and Embryology Authority, 2012). This process eliminates the inheritance of problematic mtDNA and thus attempts to eliminate Mitochondrial Disease, allowing female carriers to produce children unaffected by the disease. This scientific advancement has been met with considerable criticism and has been commonly and incorrectly attributed the title of 'Three Parent IVF' by various media and journal outlets (The Telegraph, 2016; Amato, Tachibana, Sparman, and Mitalipov, 2014; The Guardian, 2016). This research aims to look behind the sensationalist headlines and seek to understand the lived experience of families affected by Mitochondrial Disease.

## 1.5 Rationale

Mitochondrial Disease is classed as a rare disease (National Rare Disease Plan, 2014) and the current information available is almost exclusively aimed at medical professionals with little emphasis on the social factors affecting the family. This dissertation hopes to highlight the social issues experienced by families affected by Mitochondrial Disease and explore the ways in which the resulting needs could be met. It is envisaged that this study will provide a basis from which further, in depth research, can be developed in order to provide a better understanding of the needs of, not only families affected by Mitochondrial Disease, but those affected by other serious rare diseases, and how these can be met or minimised through appropriate interventions.

Furthermore, in highlighting the concept of anticipatory grief, it may allow those affected to put a name on a host of experiences which they may be unable to otherwise vocalise or articulate. Olson (2014, p.559) states that having a shared understanding of the term 'anticipatory grief' may allow for both medical and support professionals to communicate better with clients about their needs and experiences as well as legitimatising that experience.

In conducting this study, it is hoped that a significant contribution can be made to the current knowledge base of both professionals and wider society in relation to both anticipatory grief and

Mitochondrial Disease and could potentially inform the practice of professionals working in this area. Most importantly, it is hoped that this study may aid the goal of Mito Families Ireland in raising awareness and understanding of an often unheard of disease and the experiences of those affected by it.

The Community Academic Research Links (CARL) initiative promotes community based research, which allows for the voice of civil society not only to be heard but to potentially inform policy and practice in a tangible way. The CARL initiative along with the civil society organisation (CSO) Mito Families Ireland and University College Cork has allowed the researcher to access this voice and develop research that may otherwise be impossible.

## 1.6 Aim of Research

To explore the effects of anticipatory grief in families of children with Mitochondrial Disease and the potential role of social work in this area.

## 1.7 Objectives:

1. To provide an understanding of anticipatory grief and how it impacts the families of children with Mitochondrial Disease.
2. To identify whether a gap exists in social work service provision to families experiencing anticipatory grief.
3. To inform best practice and potentially highlight the need for social work provision in this area.

## 1.8 Research Questions:

1. What is anticipatory grief?
2. What issues, related to anticipatory grief, are experienced by families of children with Mitochondrial Disease?
3. What social work supports have families of children with Mitochondrial Disease availed of?
4. What social work interventions could be utilised in supporting families of children with Mitochondrial Disease?

Question one is answered through secondary research, namely, a literature review, further informed by the primary research. Question two has been researched through the findings of an anonymous online survey, utilising a quantitative analysis which was developed through the use of a discussion forum consisting of members of the Community Organisation, Mito Families Ireland. In this way, the study allowed for participation and collaboration between the researcher and the members of the community organisation in the development of research.

Question three explores the supports identified by respondents, while question four examines what interventions and supports are available relating to the issues raised by respondents and an assessment as to their viability for incorporation in social work practice.

## 1.9 My Reflexive Positioning as the Researcher

According to Fox (2007), it is imperative that the researcher understands how the process and outcomes of research are affected by their own personal position in addition to the methodology.

As the founder of the community group with whom this research has been undertaken and furthermore, on a more personal level, a family member of two children with Mitochondrial Disease, it is important to address my reflexive positioning as the researcher of this study.

Both of these factors have been heavily reflected upon in how they may affect the research and it is acknowledged that there is potential for bias in interpreting the data. While I will always strive for objectivity throughout this process, I have flagged my position with the participants and supervisor in order to allow for oversight of my work.

## 1.10 Chapter Outline

**Chapter One**

Chapter one introduces the topic to be studied and explains why this research is being conducted, placing it within current literature, outlining the aim and objectives of the research as well as the overarching research questions.

**Chapter Two**

Chapter two consists of a literature review which explores the concepts which inform this study such as anticipatory grief and family systems theory. It also seeks to ascertain the position of Mitochondrial Disease in current Irish policy.

**Chapter Three**

This chapter lays out in detail the methodological approach with which this research has been conducted, including the ontology and epistemology. It explains the process of primary research and puts forth a substantial section on the ethical concerns which were considered and evaluated prior to the approval und undertaking of this research.

**Chapter Four**

Chapter four presents the findings and quantitative analysis of the primary data, linking appropriate theory where possible.

**Chapter Five**

The final chapter draws an overall conclusion based on all the previous chapters and puts forward a number of recommendations for both the community group and social work practice as well as for further research and policy. This chapter concludes with a reflective piece on the research process.

## Chapter Two

# Review of Literature

2

IN THIS SECTION:

- **Introduction**
- **Anticipatory Grief**
- **Liminality**
- **Family Systems Theory**
- **Mitochondrial Disease and Policy**
- **Conclusion**

## 2.1 Introduction

The study of grief and loss as a theoretical basis for providing care and interventions to those who have suffered bereavement is a vast area, with a multitude of academics and practitioners informing our understanding of this traumatic life event (Kubler-Ross and Kessler, 2005; McIlwraith, 1998; Worden, 1991). There is, however, a comparative dearth of literature and research surrounding the concept of 'Anticipatory Grief' (Doka, 2006; Rando, 1988; Rando, 2000). Similarly, while medical and scientific literature related to the effects of Mitochondrial Disease is ever increasing, research related to the social experience of those affected by Mitochondrial Disease is non-existent. This thesis aims to combine each of these under-researched areas in order to provide a new understanding of how anticipatory grief and mitochondrial disease affect families and how social work can intervene to limit negative experiences where possible and promote positive outcomes.

## 2.2 Anticipatory Grief

First coined by Lindemann, (1944), anticipatory grief refers to the grief one may experience when faced with a poor prognosis of a loved one's life expectancy. That is, that in anticipating the likelihood of the death of a loved one, at an unspecified time, a person may experience a grief, not unlike that suffered by the bereaved (Wheeler, 1996). For a family in which a child is diagnosed with a life-limiting illness, such as Mitochondrial Disease, the future can appear very uncertain, as prognosis can often not be estimated with any great accuracy. Fulton, Madden and Minichiello (1996, p. 1349) attribute this, in part, to developments in medical technology, which 'prolongs life and facilitates the early diagnosis of terminal illness'. In turn, the prolonging of life also increases the number of informal carers required to provide care services outside of those received from professional organisations (Olson, 2014).

Rando (1997, p.48), identifies some of the difficulties that may be experienced in anticipatory grief, including;

> powerlessness, fear, uncertainty, confusion: violations of the assumptive world; ongoing losses; personal depletion from the stress arising from demands for major re-adaptations and investments of self, time, and finances; long-term family disruption and disorganization; balancing of opposing needs, competing demands, discordant roles, clashing responsibilities, and antagonistic tasks; traumatizing physical, emotional, and social experiences during the illness; and major reactions of guilt, sorrow and depression, anger and hostility and anxiety.

According to Rando (1997, p.49), the issues arising as a result of anticipatory grief can be considered the same as those of clinical Post Traumatic Stress Disorder. There is, however, research, (Cadell, 2012), which highlights the potential for positive externalities as a result of anticipatory grief and caring for a child with a life limiting illness, namely, post traumatic growth, whereby a carer develops new skills or maybe a positive self- awareness as a result of their experience.

While our most basic understanding of grief surrounds bereavement, Rando (1988) and Sweeting and Gilhooly (1990) highlight that not only does anticipatory grief relate to the potential or forthcoming bereavement, but also to the losses which occur as a result of the life limiting illness, long before death occurs. Hopes and plans for the affected child must be reconsidered and a future which may have once seemed settled and organised may become frightening and fraught with unknown dangers, such as sudden medical crises.

## 2.3 Liminality

Little, Jordens, Paul, Montgomery and Philpson (1998), conducted a study which found that cancer survivors experienced a feeling of liminality, as a result of the uncertainty faced during serious, potentially life threatening illness. Turner (1969) describes liminality as a state of being 'in-between', or the state of transition where one is neither in one place nor another. Ferguson (2011, p.49) states that 'everyday life depends on the maintenance of order and routine and spaces where there is an absence of such order are experienced as dangerous.' It is hypothesised that a sense of liminality may occur in those caring for children with uncertain prognoses (such as in Mitochondrial Disease) and carers may feel immobilised (Olson, 2014).

## 2.4 Family Systems Theory

Cadell, Kennedy and Hemsworth (2012), highlight that the 'usual challenges' faced by parents raising children are further compounded by the stress of 'caring for a child with a life limiting illness' (p.356). In light of our understanding of systems theory and the concept of reciprocity (Payne, 2014, p.192), which maintains that 'if one part of a system changes, that change affects all the other parts, therefore they all change.', then the challenges faced in caring for a child with a life limiting illness likely effects the entire family. Cadell et. al (2012, p.358) found that families affected by a life limiting illness had to make 'significant adjustments' in their day to day lives as well as to future plans, which impinged on the life of all family members. Kerr (2000), based on Bowen's

Family Systems Theory, discusses the emotional interconnectedness of a family unit and suggests that when anxiety is present throughout the family, the emotional interconnectedness results in stress rather than comfort, further heightening levels of anxiety. This paired with the ambiguity of health and illness in a family affected by a life limiting illness (Cadell et. al., 2012) may deeply affect the wellbeing and functioning of the entire family unit. Cadell et. al.'s (2012, p.369) respondents expressed the importance of being able to 'connect with people facing similar circumstances' as they felt that friends and colleagues could not fully appreciate the various nuanced effects of having a family member with a life limiting illness. These respondents (Cadell et. al., 2012, p.371) also highlighted the lack of emotional support from professionals for both the caregivers and the siblings of a child with a life limiting illness, which implicitly highlights a focus on the medical rather than the social model.

## 2.5 Mitochondrial Disease and Policy

Launched in 2014, the National Rare Disease Plan for Ireland (NRDP) (Minister for Health, 2014) is the only national guideline for a more holistic approach to treating and managing rare disease in Ireland, a category under which Mitochondrial Disease falls. The Irish and European definition of a rare disease is one which affects '5 in 10,000' of the population (NRDP, 2014 p.7 & p.12). As there is currently no rare disease registry operational in Ireland, there are no reliable estimates of how many residents are affected by Mitochondrial Disease. Further to this, the clinical presentation of Mitochondrial Disease is so variable that many of those affected are misdiagnosed or given an unconfirmed diagnosis. The United Mitochondrial Disease Foundation (UMDF) define Mitochondrial Disease as 'the result of either inherited or spontaneous mutations in mtDNA or nDNA which lead to altered functions of the proteins or RNA molecules that normally reside in mitochondria.' A more user friendly explanation is that:

> Faulty mitochondria affect the body's ability to function and produce the energy required for organs to operate typically. How this manifests in each individual varies greatly but more common features include seizures, cognitive delay, global developmental delay and gastrointestinal issues among others (Mito Families Ireland, 2013).

Currently, there is no particular policy or legislation related to Mitochondrial Disease in Ireland and so the National Rare Disease Plan for Ireland 2014-2018 will be referred to as the primary piece of policy guidance for this study.

## 2.6 Conclusion

This study aims to take two under researched topics, anticipatory grief and Mitochondrial Disease, and explore the social relationship between them. A definition and explanation of anticipatory grief, based on the writings of academics in the area has been provided. From this, the literature review explored the concept of liminality and the family system and showed how the concept of reciprocity means that all family members may be affected by having one member diagnosed with a life limiting illness. It then focused on policy and legislation in the area of rare disease, a category under which Mitochondrial Disease falls. The aforementioned content has been used to inform and formulate a survey/questionnaire which explored the needs and experiences of families who have a child affected by Mitochondrial Disease in relation to anticipatory grief, the methodology of which will be laid out in the forthcoming chapter.

## Chapter Three

# Research Design

IN THIS SECTION:

- Introduction
- Philosophical and Theoretical Underpinnings
- Ontology
- Epistemology
- Methodology
- Research Methods
- Sampling
- Data Analysis
- Ethical Considerations
- Challenges and Limitations
- Conclusion

## 3.1 Introduction

This chapter will detail the research process undertaken for this study including the paradigms employed, the ontology and epistemology, the research sample and the approach applied in both gathering and analysing the primary data. It will also detail how the sample for both the discussion forum and survey were chosen and present the ethical considerations central to this research.

## 3.2 Philosophical & Theoretical Underpinnings

Gray & Webb (2013, p.6) state that 'theories embody frameworks of knowledge that have been developed in social work practice and the theoretical world'. This research incorporates a number of theories such as anticipatory grief and family systems. As a CARL project, which brings together community groups and academic researchers (see p.iii), it used a participatory/ transformative paradigm in that the study and research questions were designed collaboratively by the researcher and the community group, Mito Families Ireland. This is also referred to as Participatory Action Research (Alston and Bowles, 1998) whereby the participants are active in the research process and their empowerment is promoted in this process (Whittaker, 2012).

From there, a post positivist paradigm was utilised in the data collection and analysis, underpinned by critical realism (Bhasker, 1978), which ‘sees the concepts that we use to understand [reality] as a provisional way of knowing rather than a direct reflection of reality’ (Whittaker, 2012, p.8) and seeks to ‘empower [people] to change social reality by suggesting possible solutions’ (Sarantakos, 2005, p.11).

## 3.3 Ontology

According to D'Cruz & Jones (2004, p.50) ontology can be understood as 'the science or study of being' or ‘the nature of reality’ (Sarantakos, 2005, p.430).

Gathering and analysing the data from the ontological position of critical realism allowed the researcher to place space between her and the findings allowing for a more objective analysis of the data. According to Denzin and Lincoln, (2011) critical realism holds that while there is a single reality, there is no single understanding of it.

In utilising this ontological approach, the researcher aimed to analyse the commonalities in the data in order to understand how the role of social work may benefit the families of the participant, based on the lived experience of the reality they have expressed.

## 3.4 Epistemology

'Epistemology addresses the question of what counts as legitimate knowledge' (Whittaker, 2013, p.3). In analysing the data, the researcher employed an objectivist epistemology in order to produce statistical generalizable findings, which can be further explored in future research in collaboration with the CARL initiative.

## 3.5 Methodology

This study consists of both primary and secondary research, namely a literature review and a survey, informed by a discussion forum.

The primary research was conducted in two parts with members of Mito Families Ireland, for which permission was granted by both the community organisation and the Research Ethics Committee in the UCC School of Applied Social Studies. Part one of the primary research was participatory based and consisted of creating a discussion forum of five group members who worked collaboratively with the researcher in order to develop pertinent questions to be included the survey. The completed survey design consisted of a variety of question formats, such as open, closed, ranking scales and multiple choice options (Sarantakos, 2005). Where possible, most questions employed an option of 'other', to allow participants to give a response unanticipated by the researcher, although this facility went largely unused. Questions were grouped into thematic sections for coherence and logical progression (Sarantakos, 2005). Part two saw the completed survey sent to members of Mito Families Ireland, the data from which was then analysed using quantitative methods (see: 3.8 Data Analysis) and presented by the researcher.

## 3.6 Research Methods

The nature of each participant's inclusion in the sample means that he or she is a member of a family with a child with a life limiting illness. From this, it is imagined that the participants would have little time to commit and so it was deemed that an online discussion forum and survey would be the least time consuming and least intrusive method of data collection for the participants.

## 3.7 Sampling

The initial discussion forum, from which the questions for the survey were developed, consisted of five self-selected members of an online support network, Mito Families Ireland. A request for members to fill the forum was posted within the online group, along with the researcher's contact details in order to discuss what participation would involve and what topics would arise, which allowed members to make an informed decision regarding their participation, as the material could have had the potential to cause distress. This discussion was then hosted online by the researcher with the members who opted in. Upon completion of questions and design, the survey was distributed to families caring for a child with Mitochondrial Disease through Mito Families Ireland (See appendix E). The software 'Survey Monkey' was used to create the survey. Inclusion in the survey sample was limited to family members (over 18 years of age) of children with life limiting illnesses, residing in Ireland, who had not experienced bereavement in the previous six months and as such, it was a 'non-probability purposive sample' (Alder & Clark,2008, p.121). Signed informed consent was gathered from the members of the discussion forum (see sample in appendix C), while survey participants were advised that by agreeing to the conditions as set out on the information page and proceeding with the survey, informed consent was assumed. This was critical to the study as participants in the survey were guaranteed anonymity apart from their membership of a family with a child with Mitochondrial Disease. Potential participants were advised of the nature of the survey content prior to commencing, again to limit any distress.

## 3.8 Data Analysis

Utilising the software, 'Survey Monkey', the survey was designed and sent to the relevant participant pool on January 28th, 2017, where it remained open to participants until February 13th, 2017. During this period, thirty six respondents engaged with the survey, twenty three of those to the point of completion. In order to present coherent data, only the twenty three fully completed surveys were drawn upon for data analysis. The findings of this survey were analysed using quantitative methods, primarily through the use of the 'Survey Monkey' software. The data gained from open ended questions was analysed using coding where appropriate in order to group answers according to their relationship into themes (Sarantakos, 2005) and was then presented with the rest of the data predominantly in graphs, using text where appropriate. In the case of one question which asked where participants sought information after diagnosis, rather than presenting the data in a chart, the most significant answer was presented using a word cloud graphic to give readers a visual understanding of the experience of participants and the impact of that particular information source.

The findings were then applied to relevant theory which allowed the researcher to identify relationships and differences between the primary data and existing theory. This method of analysis was chosen in order to understand the scale of shared experiences in order to ascertain whether there are commonalities experienced by family members, which could be targeted by social work intervention. From this, recommendations and conclusions were made as to how social work may be able to ameliorate the negative experiences and enhance the positive experiences of families affected by mitochondrial disease.

## 3.9 Ethical Considerations

In undertaking this research, a significant number of ethical concerns had to be taken into consideration. The process began in late May 2016 through discussion of the ethical considerations with the community liaison person and a submission to the CARL team. Following revision, this was then submitted to the School of Applied Social Studies. Due to a change in the application process, a further application was drafted and submitted to the Research Ethics Committee in the UCC School of Applied Social Studies in October 2016, which permission to proceed granted in December 2016, in the understanding that minor revisions would be made. The final statement of ethical considerations are presented below.

- As previously noted the nature of the study is sensitive and could potentially be distressing for respondents. For this reason, questions were formulated to be as non-invasive as possible. The use of a survey allowed participants the opportunity to be both candid and anonymous. As this survey was hosted online, written consent could not be obtained but a notice at the beginning of the survey informed participants that continuing with the survey constituted consent (See appendices D and E). Participants were given the option to exit the survey at this time. Participants in the discussion forum were given the option to withdraw from the forum at any time up until the survey design had been completed and were notified of such**.** Signed consent was gathered from the members of the discussion forum before proceeding and all were ensured of confidentiality outside the context of the discussion forum.

- Due to the anonymous nature of the study, participants were not debriefed personally however an information sheet was drafted to inform respondents of the purpose and focus of the study, as well as to provide details as to how the data would be used, the contact information of the researcher and contact details for support agencies as well as Mito Families Ireland admin, should they wish to seek support during or after participation. Participants were also advised to contact their GP should they feel that this research had caused any distress. The researchers contact details were also provided, however, participants were informed that this could compromise their anonymity. This ‘sheet’ formed the first page of the online survey (see appendix D).

- In line with UCC data storage policy, data pertaining to the participants shall be stored on a secure server and deleted ten years after completion of the study. It will not be used for any purpose

other than stated in the information sheet and any hard copies of data will be destroyed upon completion of the study.

- Researcher positionality: as the researcher is a member of a family of children with mitochondrial disease, it was considered that there may be bias in interpreting the data. This was clearly laid out in both the research design and in the ethical approval application as well as in the participant information sheet. However, the researcher always strove to remain objective and impartial in undertaking all aspects of this research. A reflective journal was utilised throughout this process and any issues arising were raised in supervision with the researcher's tutor.

- The particular group from which participants were gathered is known to the researcher. The researcher founded this group seven years ago in response to lack of awareness and support surrounding a rare disease. It is understood that this could be considered a potential conflict of interest. In reflecting on this, however, the researcher believed that this was unlikely to affect the study or the participants. The group, numbering approximately two hundred, is member led and little power differential exists. Researcher involvement in the group is largely confined to ensuring that group rules are adhered to and organising events with members. The agenda of the group is always decided by majority vote and can be proposed by any member. Participation in this survey was voluntary and anonymous. The formulation of questions were respectful and non-invasive and it is imagined that rather than cause harm, this research provided participants with an opportunity to have their voice heard on an issue that is important to them. The issue of anticipatory grief and its effect on families is also of personal interest to the researcher. Through membership of Mito Families Ireland, this researcher began to notice the commonalities in the experiences of affected families, including the researchers own and felt that there was a gap in the research knowledge base which could be addressed by empirical research as a base for further research.

- It is understood that the personal relevance of this topic to the researcher could have had the potential to cause personal distress. For this reason, it was agreed that the administrators of Mito Families Ireland would be informed should any such issue arise for the researcher and would provide oversight throughout the research process and that the researcher would seek support from the administrators or a G.P. if necessary. The researcher also utilized ongoing supervision in order to ensure awareness of whether issues were causing any personal distress.

## 3.10Challenges & Limitations

There were a number of limitations to this study, primarily the restrictive nature of the time frame and word count. Due to these restrictions, more in depth research of the effects of caring for a child with Mitochondrial Disease could not be explored and had to be limited to anticipatory grief. Even within this filter, restraint had to be exercised as numerous unanticipated and interesting topics arose such as the effect of illness and anticipatory grief on wider family relationships but could not be included.

It is not deemed ethical to interview children under eighteen for the purpose of student research, however, it is anticipated that being able to do so would yield significant insight as to the experience of siblings of children with Mitochondrial Disease or similar.

Due to the geographical spread of members, it was deemed that hosting all aspects of the primary research online would be the most convenient option for participants; however this researcher feels that further research in this area may benefit from face to face discussions to allow for a more natural flow in conversation if a researcher could travel to participants as the constraints in participant's time are unlikely to change. If undertaking this approach, it is imagined that a researcher would need considerable relationship building skills as the topic remains a sensitive and personal one for participants.

In hosting the discussion forum, the researcher was provided with a rich and honest discussion which would have provided interesting and original data for a qualitative study. As the data was not however provided for qualitative analysis, this approach was not pursued. The researcher feels that this approach could be utilised to great benefit in future research with this community group.

## 3.11Conclusion

This chapter has discussed the methodology employed in depth, highlighting the methodological approach used in the creation of the research and the data collection and analysis. In doing so, the researcher was able to engage with the community organisation using a collaborative approach and then analyse the data in a more objective manner. This is particularly important as it is imagined that this research will provide a basis from which future research may stem with the community organisation and so bias must be limited. It then laid out the significant ethical considerations involved in undertaking this research, as well as the challenges and limitations with this study, which may be used to inform the design of further research in this area. The findings and the analysis of thus shall be discussed in the following chapter.

## Chapter Four

# Findings and Analysis

IN THIS SECTION:

- Introduction
- Personal Data
- Anticipatory Grief
- Liminality
- Diagnosis
- Family Life
- Social Work
- Conclusion

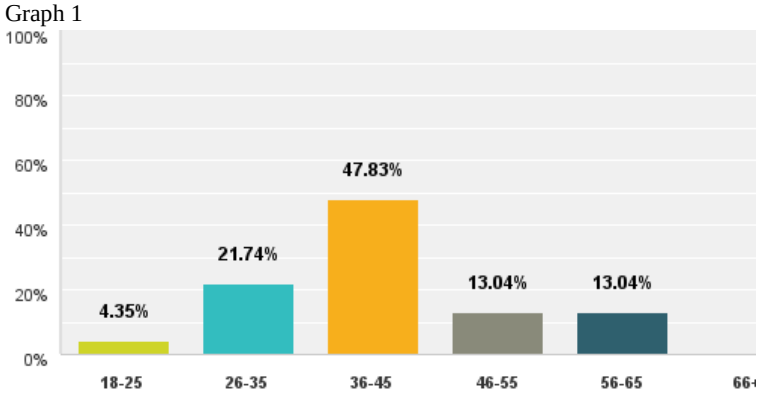
## 4.1 Introduction

This chapter presents the data as discussed in chapter three and combines it with theory and current knowledge from a variety of fields in order to provide an informed analysis. The resulting findings and analysis are grouped thematically for clarity.

## 4.2 Personal data

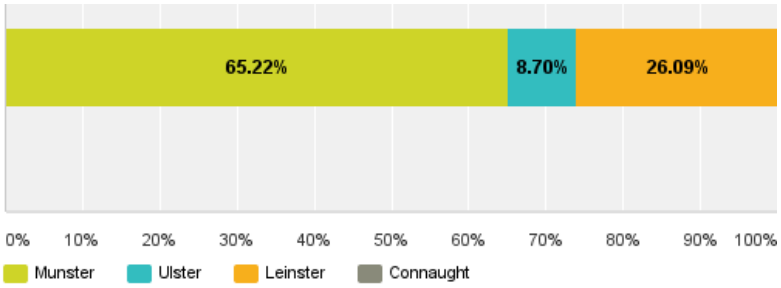
As noted in the previous chapter, only fully completed surveys were used for analysis. Due to this, there are no male participants in this analysis. In the full sample, there were three male participants, none of whom completed more than four questions. While there is no concrete explanation for this, the researcher notes that this may be an area worthy of further research. In previous research, it has been noted that many services, particularly for children, tend to be female centric in how they operate, who operates them and who they appeal to, unintentionally excluding men (Ghate, Shaw and Hazel, 2000; Carpenter, Addenbrooke, Attfield, and Conway, 2004). Martin and Doka (1998), note that masculine grief may not be expressed as openly as female grief and therefore men may not feel as comfortable in participating in research such as this.

Graph 1




As indicated, all respondents to whom this data relates are female and are aged between 18 and 65, with almost half falling into the age category of 36-45.

Graph 2



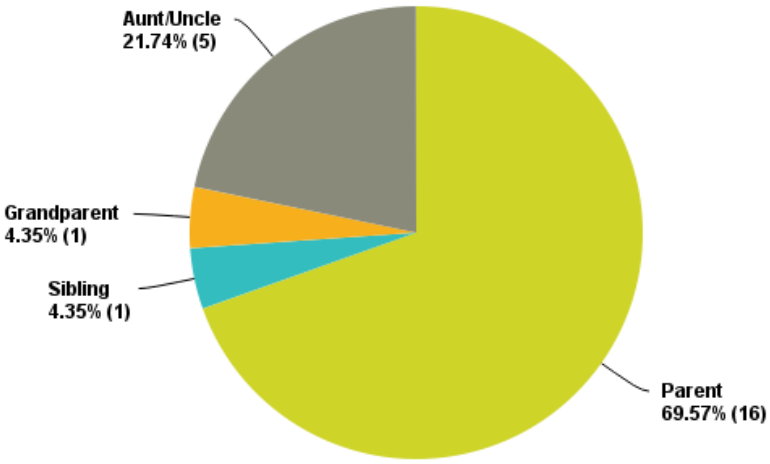

91.3 % of respondents reported residing in Munster or Leinster, while no participant indicated Connaught as their place of residence.

The larger response rates of those residing in Munster and Leinster correlate with group membership overall, as well as the density of populations in these areas, however, the significant number of members residing in Ulster is not represented in participation. A

possible explanation for this anomaly may be due to the significant number of members bereaved in the Ulster area over the past number of months. Members were asked not to participate if they had suffered bereavement in the previous six months as the researcher felt this would cause unnecessary distress.

It is unknown whether any members reside in the Connaught area, but the absence of any participant from this province may suggest that information regarding the community group has not penetrated that area. A further hypothesis might be that there may be less specialised paediatric health services in Connaught and perhaps children in the province are undiagnosed, as it is highly improbable that there are no children affected by Mitochondrial Disease in the area.

Graph 3

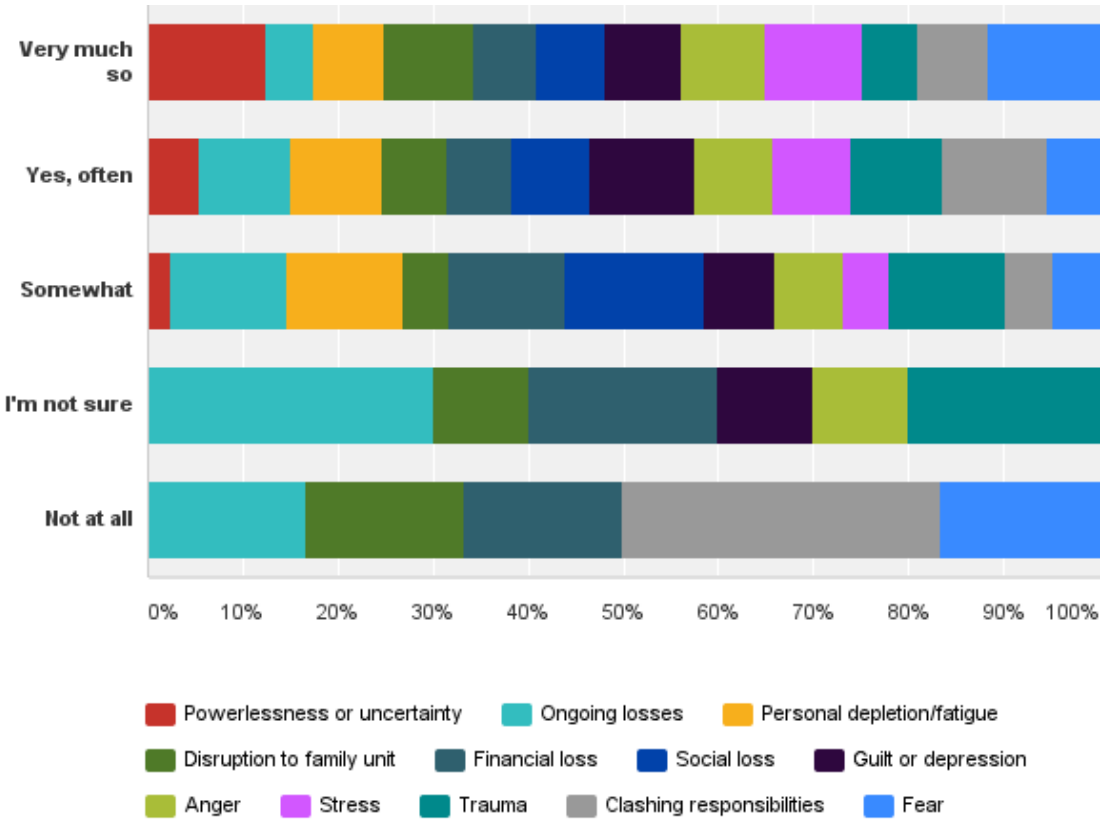



While all family members over the age of eighteen were invited to participate, the majority of those who did so indicated they were parents of a child with Mitochondrial Disease and from the data analysed earlier, it can be inferred that these respondents are mothers. Again this is largely coherent with group membership overall, as is the percentage of those who identify as an aunt or uncle and those over the age of eighteen who identify as siblings of a diagnosed child. The membership of grandparents is, however, underrepresented in participation in this research.

## 4.3 Anticipatory Grief

When presented with the description of anticipatory grief as defined in chapter two (see 2.2), 100% of survey participants felt that this accurately represented their experience since their loved one was diagnosed with Mitochondrial Disease. Many of the difficulties outlined by Rando (1997, p.48; see 2.2), in relation to the experience of anticipatory grief were agreed with by participants, in particular, powerlessness, uncertainty and fear (see graph 4). According to Mallon (2011), the period in which death is anticipated can be a time of significant emotional disturbance and change, even more so than the period following death. There are a number of factors which may attribute to this. Jewell and Blackmore (2004) note that when faced with a significant illness or disability, a family may feel separated from both the extended family and the wider community causing the family to feel more

Graph 4




vulnerable. A significant hallmark of this period can be the uncertainty with which the family live (Boss, 1999) This is further compounded by a lack of services and support, a deficit recognised by the Office for the Minister of Health (2014), which notes:

> For people with a rare disease or parents of children with rare disorders, there are often many unknowns. These unknowns can be related to the cause of their disease, the likely effects, the

availability of effective treatment and, indeed, their life expectancy and expected quality of life. (NRDP, 2014,p.17)

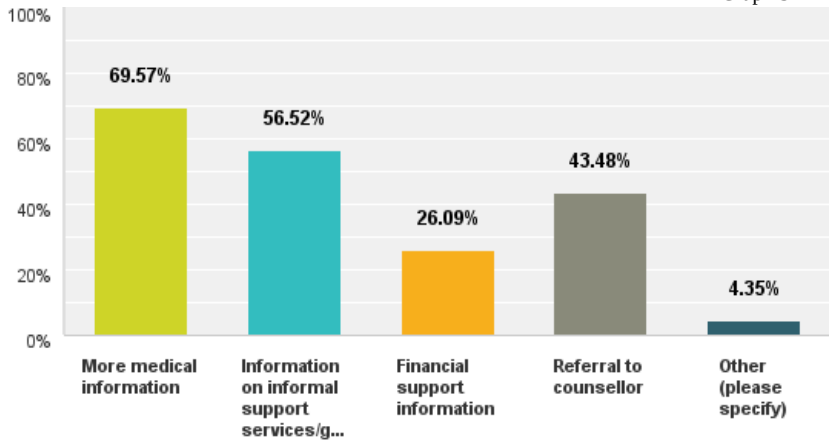
It is further recognised within the NRDP (2014) that even when the often arduous goal of securing a diagnosis is reached, the rarity of some diseases means that there 'is often no established care pathway for that disease within the Irish health and social care system' (p.61). This lack of any standardised provision may mean that families receive inadequate services, which according to Powers, Sowers and Singer disempowers carers (2006). Gregory (2005), however notes that by clarifying and identifying the concept of anticipatory grief, medical and allied health professionals can better communicate and respond to carers needs, while Frank (1993) suggests that being able to put an umbrella term on the feelings and experiences of carers facing a loss may provide them with a sense of legitimation.

Interestingly, a significant portion of participants (32%) did not feel that they had clashing responsibilities, while all participants noted feeling guilt or depression to some extent (Graph 4). Previous research has found that these concepts may be interlinked as carers struggle to be fully present in each role they may need to fulfil, as there is rarely time to devote to one single matter (Jewel et. al., 2004; Beresford, 1995)

## 4.4 Liminality

As discussed in Chapter 2, the uncertainty and lack of services faced by those caring for a child with a life limiting illness may lead to a feeling of liminality or disorder. Almost 70% of participants felt that this state strongly reflected their experience, with less than 5% feeling that this was not at all applicable to their experience. Of those who identified as experiencing a sense of liminality, they indicated that medical and support services information would have been the most helpful in reducing this (Graph 5).

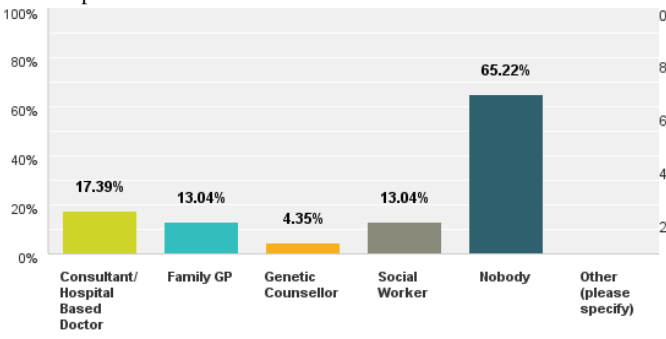
Graph 5



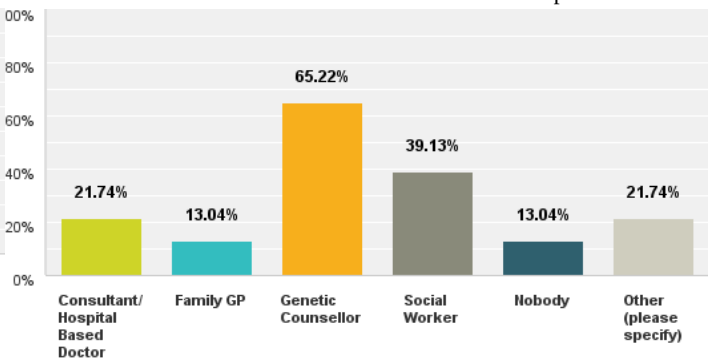
## 4.5 Diagnosis

Regarding the diagnosis of a child within the family, participants were asked what professionals they met with, within forty eight hours of diagnosis (Graph 6) versus who they would have liked to have met with (Graph 7).

Graph 6



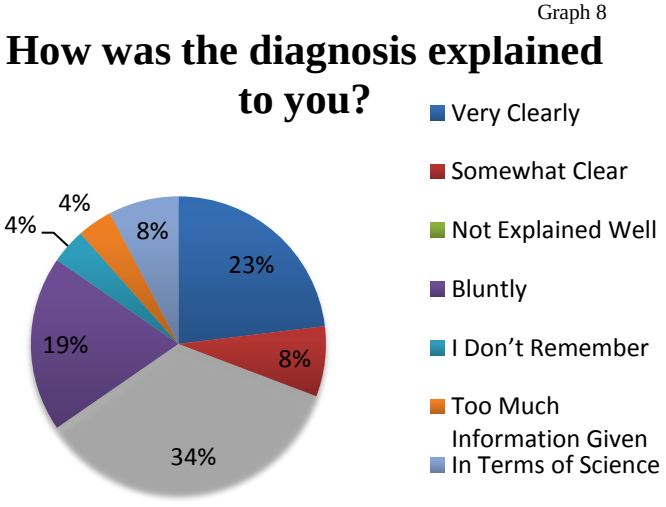

Graph 7




65% of participants indicated that they did not meet with anybody, while almost 87% indicated that they would have liked to have met with one or more professionals or someone with more knowledge or experience, including a healer, an experienced nurse or a support group. This is significant when viewed in the context of crisis theory (Thompson, 2011). Receiving a diagnosis of an incurable disease in a loved one could potentially develop into a crisis period for an individual. According to Thompson (2011), timely intervention is key to working with an individual to recognise and resolve the crisis and allow for new coping strategies and skills to grow. It is postulated that meeting with a professional, such as a social worker, within forty eight hours of diagnosis may minimise the potential for crisis or provide a good foundation for those affected to engage in further supportive work and build a positive relationship with that professional.

Furthermore, only 31% stated they had received a diagnosis that was explained either 'very' or 'somewhat' clearly. This should be considered of particular importance as Healy's (2015), research noted that when children were diagnosed with a disability, the manner in which the diagnosis was delivered significantly affected the child's

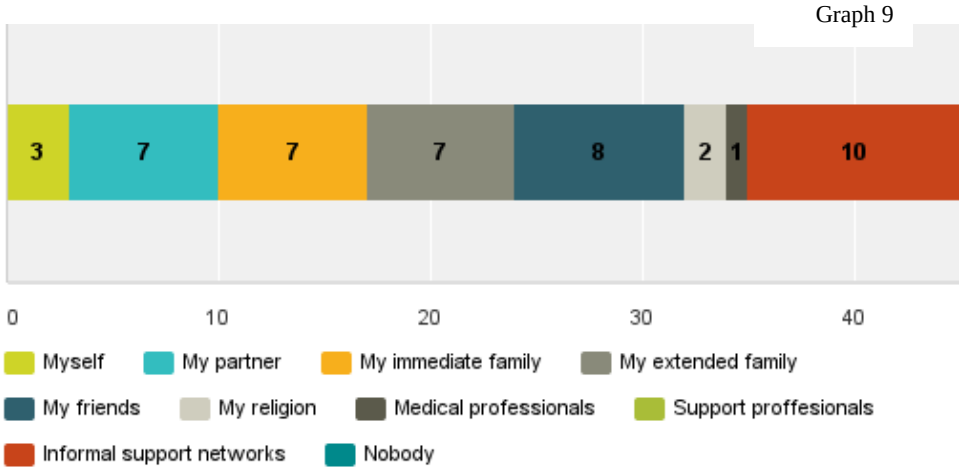
Graph 8



parents attitudes towards it and it is hypothesised that this could likely extend to parents of all children diagnosed with a serious illness. (It must be noted at this point that a flaw in the research has been recognised, as it cannot be assumed that all participants received an explanation of the diagnosis from a doctor directly and may have received the information from another family member). A 2008, Department of Children and Family Services report 'Building a Culture of Patient Safety' highlights the concept of the 'Knowledgeable Patient' (p.18) whereby patients and families would be fully informed of all aspects of their care, should be able to access a system which would 'signpost paths through the healthcare system and guide patients in accessing the care that is most appropriate to them' and have 'access to support networks'. Adopting a framework such as this would allow for a partnership style approach between professionals and families which in turn could negate or ameliorate some of the feelings of powerlessness as experienced by participants in this research (Rigall, 2012; Schippers and van Boheemen, 2009).

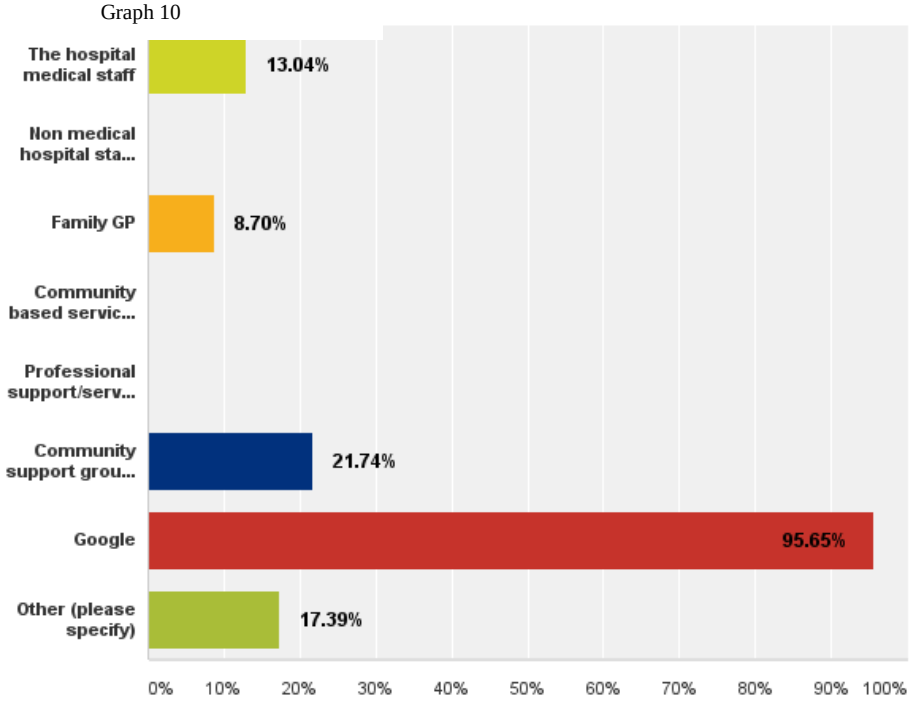
When asked where they felt they had gotten the most support from following the diagnosis, the largest option the sample indicated they received the most support from was from informal support networks, followed by friends and none chose the option of support professionals. This finding is interesting in that while a number indicated that they received support from various family members, the data is suggestive of a possible reluctance to add any further weight to the 'burden' of care already being experienced by the family. It also shows the need for informal support networks, such as Mito Families Ireland. Research by Gregory (2005) highlights the importance for people of knowing that their experience is shared by others and in essence, that they are not alone. This is recognised within the NRDP 2014 which notes the necessity of 'points of contact for others with similar conditions or experiences' and acknowledges that families living with rare disease are 'often the leading 'experts' in their diseases'(p.59),. The NRPD 2014 also acknowledges that much of the support received by those affected by rare diseases is from small patient organisations with little or no funding, run by those who are also affected and thus

Graph 9



already have considerable demands on their time and resources and therefore need statutory support and recognition.

Graph 10



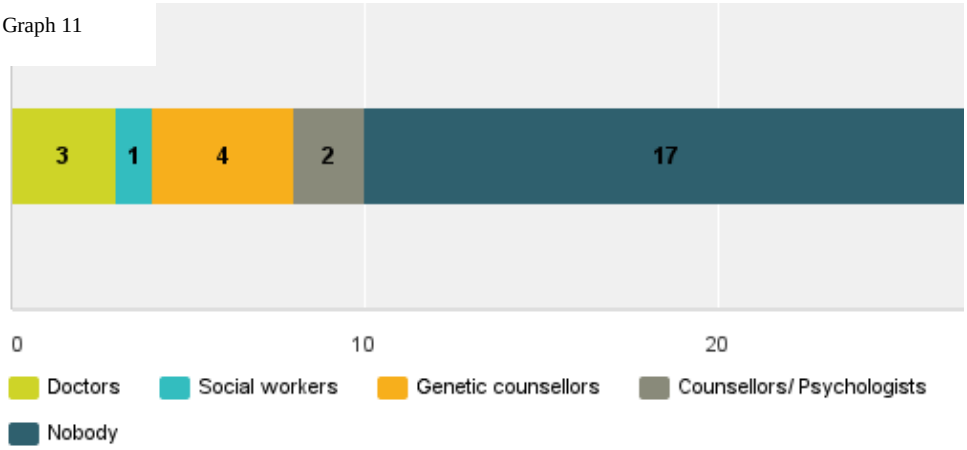

Finding support for rare diseases, be it in the form of a group or of an individual with a similar experience, could be considered significantly easier than it would have been even a decade ago due to widespread access to the internet. However, this access also means that, for many, the internet is the first place people may turn to for information, not all of which is posted by trusted sources or contextualised correctly. In this study, the research found that only 13% of participants received information on Mitochondrial Disease from professional medical staff, while almost 96% turned to 'Google' for information. While there are considerable benefits as mentioned previously, the information available could be considered overwhelming and extremely distressing to someone dealing with a recent diagnosis. The graphic presented here gives an overview of the results from 'googling' 'Mitochondrial Disease' and 'Mitochondrial Disease in Children'. The larger words are those that appear most frequently on the first two pages of each search result and gives some insight in to what a family member or affected person faces when seeking information online.

Image 1

## 4.6 Family Life

Participants in this study, according to their responses, were most often not invited as a family unit to engage with various professionals in relation to the illness. In the instances where the entire family unit was met, it was most often with a doctor or genetic counsellor.

Graph 11



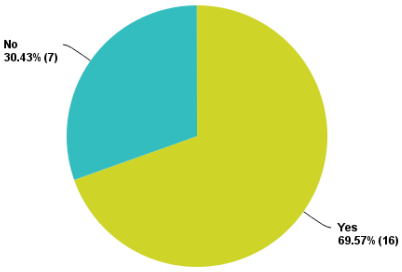

According to Leadbetter (2004), 'Family group conferencing is a process of supported mediation that allows the wider family to find solutions for themselves as opposed to solutions being found for them… '(p.204). While Leadbetter places family group conferencing in the locus of child protection, this strategy is also used within the palliative care context to positive effect. The sense of empowerment and feeling of support that can be given to a family through conferencing cannot be underestimated and could be of great benefit to families caring for a child with a life threatening illness or complex illness, as it could promote a partnership style approach. It is envisaged that this approach could empower parents to be more vocal about their needs and to problem solve more effectively and as a result build up their resilience (Weick, Rapp, Sullivan, and Kisthardt, 1989). As stated in section 2.4, the concept of reciprocity (Payne, 2014) holds that changes for one family member can affect all members (Collins, Jordan, and Coleman, 2010). This is further recognised by the NRDP (2014) which highlights that policy needs to cater for both patients and carers, based on the findings by EURORDIS which noted that:

> The whole family of a rare disease patient, whether children or adults, is affected by the disease of the loved one and can become marginalised: psychologically, socially, culturally and economically vulnerable (2005, p.57).

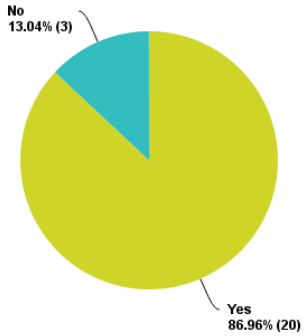
This finding is mirrored in part, by the findings of this study, whereby, when asked if the illness had impacted on their financial status, ability to socialise and employment status, participants answered yes to each, approximately 70%, 87% and 83% respectively.

Graph 12

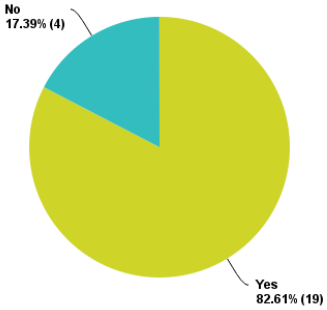
**financial status**




**ability to socialise**




**employment status**




The importance of wider supports following this finding cannot be underestimated, particularly as 50% of respondents found that their relationships with others had been negatively impacted by the diagnosis, while 19% found they had developed more positive relationships. Belle (1988) found that the way in which stress is experienced is affected by the presence or absence of a functioning support network while Monroe & Kraus (2010, p,20) highlight that resilience

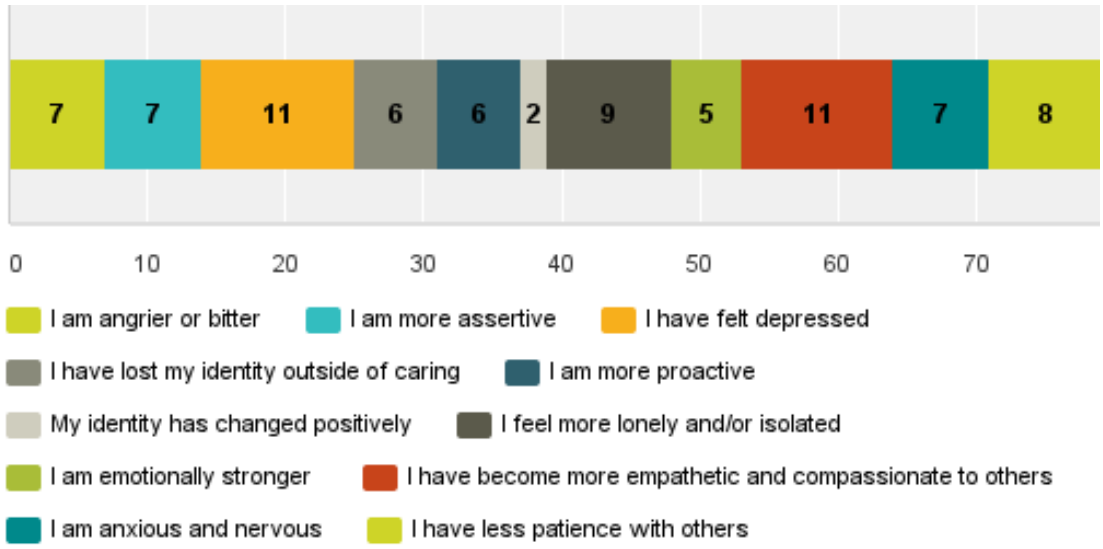
> relies on a triad of factors: genetic and constitutional, including personal attributes such as positive self-concept, a supportive family milieu, with close bonds to at least one family member and the availability to parents and children of external support systems.

When asked 'Apart from the patient and primary carers, who do you feel was most affected by the illness?' 43.5% of partcipants felt that the child's extended family including grandparents were most affected, while 30% felt it was siblings. This again highlights the far-reaching effects of illness in one family member, however, the data must be read with the understanding that the family construct of each participant is unknown and so it cannot be assumed that each affected child has siblings or grandparents etc.

Research by Clavering (2007) found that parents particularly valued when their child was seen as an individual with an illness rather than being seen solely as their illness and when they were viewed in the context of their family rather than simply as a child alone. There is often little room in the dominant medical model for the social model and this view can spill over into the wider community. In this study, only 17% of participants felt the affected child was treated no differently than they were prior to having a known illness.

When given a range of options and asked to choose which, if any, they had experienced as a direct result of the illness, participants particularly highlighted feelings of depression and loneliness or isolation.

Graph 13




The social marginalisation of rare disease families, as mentioned previously, is echoed by Jewell et al (2004) who note that this is often a hallmark of family carers. Mackey and Goddard (2006) show that this is further compounded by the lack of respite available for children with complex needs and the mental exhaustion this can cause in family members.

Participants further noted that they have become more empathetic and compassionate towards others since the time of diagnosis, however only two participants felt that their overall identity has changed positively. According to Machin (2014), using 'Meaning Reconstruction Theory' with families can allow them to construct new meaning in their lives and reach a level of acceptance of their changing lives and cope positively with it. It is not a singular process and in the case of caring for a child with a degenerative condition, both meaning and identity may need to be reconstructed continually. This may help with the loss of personal identity as identified by both the participants of this study and Woodgate (2008). The growth in empathy towards others is also echoed in research by Kriegsman and Palmer (2013).

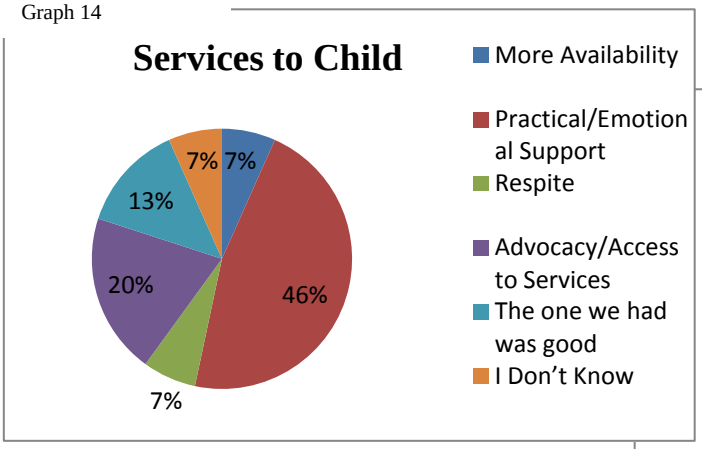
Cadell et al (2011) noted that both these positive and negative states co-exist, which could be considered comparable to the 'Dual Process Model' (Mallon, 2011) which is usually applied to grief after bereavement. This researcher, however, contends that the dual process model can also be seen in anticipatory grief whereby families manage to continue balancing most aspects and demands of family life while simultaneously grieving the ongoing losses they may experience. Further to this,

aspects of Cadell et al's 'Post Traumatic Growth' (2011) can be seen in the responses of participants where they identified becoming more assertive, proactive and compassionate towards others. This growth along with the persons experience can be used to empower others who face a similar situation, through support and advocacy (Schaefer and Moos, 2001), as can be seen in Mito Families Ireland.

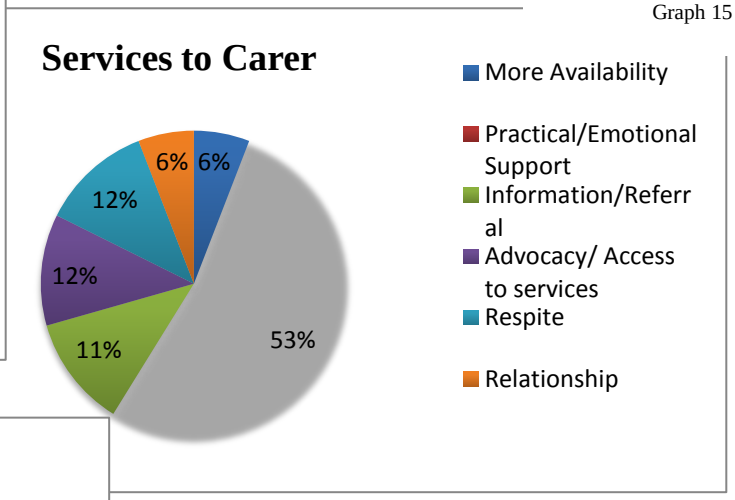
## 4.7 Social Work

Participants were asked that if they could tailor a social worker to suit their needs, what services would that social worker provide to the child with the illness, to the carer and how often would they be in contact or available to meet?

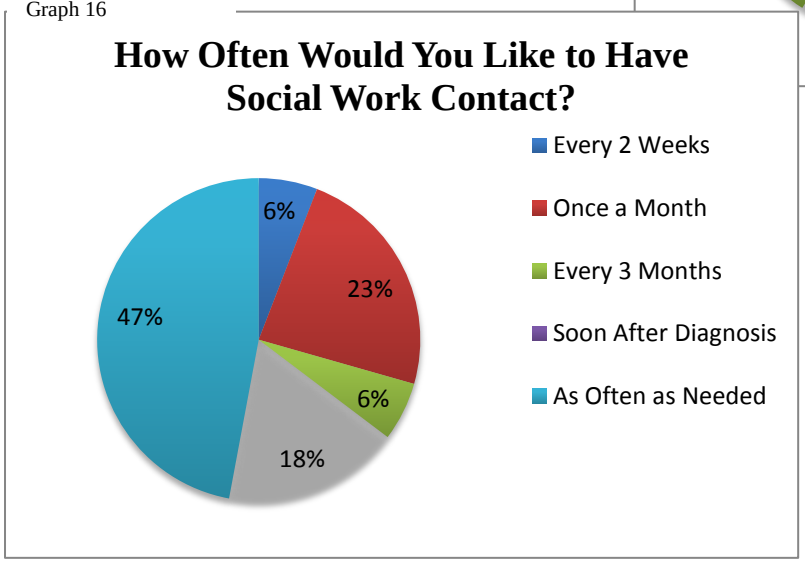
Graph 14




Graph 15




Graph 16




The NRDP (2014, p.5) recognises that 'patients and their carers require significant clinical and non-clinical support.' In particular, they note that in creating care pathways for families, appropriate information must be provided in relation to medical cards and schemes, financial allowances and grants, housing and disability services and counselling and support services (p.35-36). The NRDP 2014 highlights that 'specialised social services are instrumental for the empowerment of people living with rare conditions' (p.62) and that a lack of such support is what may result in isolation, rather than the direct effects of the disease itself. As can be seen in graphs 14, 15 and 16, this reflects the services participants in this

study want provided by a social worker. Participants also noted the need for advocacy and access to services and respite. The *Respite services for children with life-limiting conditions and their families in Ireland – A National Needs Assessment* report, 2013, found that respite services can be difficult to secure for families of children with rare disease, as they may never have a definitive diagnosis, which is often a requirement for availing of respite services. The additional stress of trying to secure basic services only adds to the difficulties already facing families (Twoy, Connolly and Novak, 2007) and should ideally be shouldered by social workers.

Participants identified that, in general, the diagnosis of Mitochondrial Disease was not explained well enough to them at the time of diagnosis. This researcher earlier postulated that having a social worker available at this time could be beneficial to families in order to provide further information or clarify the information that has been given by medical professionals. 6% of participants highlighted that the relationship with a social worker would be important to them, so it is anticipated that the presence of a social worker at the early stages of the medical journey would allow a positive and empowering relationship to develop, a proposition that is supported by 39% of participants who indicated that they would have liked to have met with a social worker within 48 hours of diagnosis. Participants also highlighted that they would like to be able to access social work support as needed. In current circumstances, this is not possible due to lack of manpower and would require significant recruitment.

The very definition of social work holds that:

> The social work profession promotes social change, problem solving in human relationships and the empowerment and liberation of people to enhance well-being. Utilising theories of human behaviour and social systems, social work intervenes at the points where people interact with their environments. Principles of human rights and social justice are fundamental to social work (*International Federation of Social Workers 2000*).

Under this definition, social workers are the ideal professionals to engage with families experiencing anticipatory grief and the issues that arise from it. It is noted, however, that 7% of participants said they did not know what services social work could provide to the affected child while Currer (2007) notes that social workers are reluctant to engage in counselling roles as they are unsure of their own remit. Social workers as professional's may need to be clearer on their role within various contexts so that they are certain of what services they can individually provide with confidence and moreover so that they can inform families of what services they can offer to them (Currer 2002; Lloyd, 2002).

It is recommended in the NRDP 2014 that rare disease modules are taught in medical and care disciplines. While the expanse of rare disease could not realistically be covered in any course, an overview of the limited services and how to access them, as well as the emotional needs of families and service users would be beneficial as a workshop or as part of an existing module social work courses. To supplement the current teachings on loss in social work degrees, which recognise that loss is often the basis of many issues social workers encounter (Gray, 2005), the addition of anticipatory and ambiguous loss would provide a fuller understanding. Further to this, learning how to work with loss in its many forms would give future social workers more confidence in providing a silent listening as well as providing practical support (Thompson 2012).

It is noted, however, that social workers can only provide services and information to clients, where services and information exist. Due to the widespread lack of information to both professionals and affected families, the NRDP recommended the creation of the National Office for Rare Disease, which would formulate a rare disease database. This office is now operational, however, information is not forthcoming. Early in this research process, this researcher requested information on the prevalence of Mitochondrial Disease among children in Ireland (see appendix F) and was informed that the database for rare diseases has not yet commenced and that the complexity of mitochondrial disorders means that one single figure is unlikely to be obtainable.

## 4.8 Conclusion

From these limited findings, it would appear that there are significant commonalities between families of children with Mitochondrial Disease, at least from a female family members perspective. All participants felt that the definition of anticipatory grief and the feelings involved described their experience well, while many noted feelings of fear, powerlessness and isolation. An overwhelming amount received the diagnosis alone and many felt it was not explained well enough. Almost half the participants sought support from informal services and over 90% turned to the internet for information on Mitochondrial Disease.

Participants highlighted that the child diagnosed with Mitochondrial Disease was treated differently by others following diagnosis and that their own work and social lives, as well as their finances, were directly affected by their caring role. They felt that the diagnosis affected the entire family unit and yet few received services directed at the entire family. Participants had little interaction with social workers but expressed that they would avail of social work support and would like frequent contact.

In considering these findings along with the knowledge gained in previous chapters, the following and final chapter will present recommendations based on these as well as an overall conclusion.

## Chapter Five

# Conclusion and Recommendations

IN THIS SECTION:

- Introduction
- Research Recommendations
- Reflective Piece
- Conclusion

## 5.1Introduction

Having conducted a literature review, undertaken primary research and presenting the methodology along with the findings and analysis, this closing chapter will make recommendations based on the information within where appropriate and present a final overall conclusion. Following this, a reflective piece on the research process shall be presented.

It is noted that these recommendations are limited to the findings of this research and so cannot be significantly generalised to the population at large, it is however imagined that these recommendations would benefit many other families affected by rare and life limiting diseases. Furthermore, the limitations of this research project means that only a very small portion of anticipatory grief and related concepts have been covered.

## 5.2Research Recommendations

### Mito Families Ireland

- As noted in chapter three, only three men engaged with the survey and none completed it. Men are also significantly under represented within the group and it is suggested that Mito Families Ireland explore ways of engaging men further, possibly conducting a small informal discussion group with the men that are currently involved in the group.

- The lack of information about services and supports available was raised a number of times during this research. There is very little nationwide support and so it is suggested that small teams of volunteers in each province come together (be it physically or online) and create a directory of services available locally or within a certain geographical distance as this will vary between provinces. This information could then be made available online and potentially within hospitals.

- Broaden awareness of the existence of Mito Families Ireland as it is possibly not reaching some areas such as Connaught, as indicated section 4.1. This could potentially be achieved as a by-product of a further recommendation, which is that the group explore the possibility of securing either charity status or statutory funding in order to extend their remit and reach. Through this, the group would have a stronger footing from which to lobby the government for services and hold them accountable for the lack of progress on the recommendations made in the National Rare Disease Plan 2014-2018.

- The group should consider continuing collaboration with the CARL initiative and UCC in order to operate from evidence based research. The discussion forum formed in this study raised significant issues that are worthy of further research and it is suggested that these are discussed with a view to submitting further research applications.

### Further Research

- The National Plan For Rare Diseases 2014-2018 was heavily relied on through this research and it is noted that little appears to have been achieved in any explicit way. It is suggested that this is an area for further research, particularly as the period covered in this plan is almost at an end. It is further recommended that Mito Families Ireland submit an application for research to CARL based on this.

- While not appropriate as a student research project due to the ethical considerations, it is suggested that research with siblings of children with life limiting diseases would allow for a better understanding of what types of services these children would benefit from.

- The researcher in this study found that participants in the discussion forum were willing to share their experiences in unexpected depth and with raw honesty, both of which would have made for a rich qualitative study. It is recommended that further study with this group is considered using a qualitative approach.

### Social Work Students/Practice

- When families receive a medical diagnosis that their child has a life limiting illness this information needs to be communicated to them in a way that is sensitive and compassionate. It also requires this information is explained without the medical jargon that often accompanies such as a diagnosis. It is suggested that social workers are ideally placed to offer this service, as with knowledge of systems theory, they understand the effect this will have on the family as well as the child. However social workers may often feel ill equipped as they may not have received adequate training in college to deliver such bad news.

- In section 4.7, some participants noted that they did not know what services a social worker could provide for the affected child. Social work interventions may not always be explained or offered to clients, particularly when operating within a medical model dominated field. It is incumbent on the social work profession/ social work practitioners to ensure that

clients/patients understand our role and services that can be offered/provided such as emotional and practical support.

- As noted in chapters 1,2 and 4, there is potential for positive growth in traumatic circumstances. Social workers should work with clients to foster this growth, such as encouraging and supporting self-advocacy which could, in turn, enhance coping skills.

- Social workers should receive practical training on the way in which the welfare system operates and ways in which it could potentially impact on clients. With this knowledge, social workers may be better equipped to advocate on behalf of their clients to secure financial payments and services They could help their clients navigate through what is sometimes a complex bureaucracy thereby reducing stress on the families they work with. It is envisaged that this may involve creating a workshop on welfare entitlements, services and grants for social work students.

- Cadell et. al. (2012) highlight that 'the role of the parent caregiver is continually changing (p.358)'. Social workers need to recognise this and be able to adapt their practice to meet the needs of the family as their roles, responsibilities, feelings and lives change at each step of their journey.

## Policy

- The Interim Report on National Rare Disease Plan for Ireland 2014-2018 shows little significant progress in achieving the goals set out in the original plan. Greater participation of patient organisations needs to be central in creating the next plan with a greater drive to create more public awareness around submissions and the ability to contribute.

- Currently, there is only one recognised treatment centre for metabolic diseases, an umbrella under which mitochondrial disorders fall, in Ireland which is based in Dublin. As noted in the NRDP, 'Ireland has a higher incidence of inherited metabolic disorders than the UK and USA' (p.30). Future policy should consider the needs of patients across Ireland and should look at the viability of metabolic clinics in local hospitals to reduce the demand and cost on families travelling to Dublin, often for minimal routine testing.

- The GRDO survey (2012) noted that 90% of respondents with rare diseases considered support organisations to be essential or important (NRDP, p.63). It is recognised that these groups are often small but may be the only thing that prevents isolation in carers and patients and need to be given state support. The National Rare Disease Office should set out the process by which patient organisations achieve state recognition.

## 5.3Conclusion

Having analysed both the primary and secondary research, the importance of understanding anticipatory grief and its effects cannot be underestimated. This researcher acknowledges that, while it cannot ameliorate all of the difficulties faced when a loved faces a poor prognosis, working with an understanding of anticipatory grief could potentially allow for better outcomes for families.

The lack of policy in this area and the failure of commencing the recommendations of the National Rare Disease Plan 2014-2018 only serve to compound the difficulties faced by families who are often left to depend on themselves or families in similar situations, for information on entitlements and services.

This research has shown that many families have not been offered social work intervention or support but many believe that may benefited from social work support. This researcher contends that social workers are best placed to work with families experiencing the difficulties of anticipatory grief and that in doing so social work practice with affected families could be informed by the Palliative Care Competence Framework in relation to social work and adapt it to suit families as appropriate (2014, pp.72-85).

At present, the resources to work in this way with all the families who would benefit from it, are not available and so the burden of support remains with small organisations such as Mito Families Ireland, which is essentially families helping each other. This situation is unlikely to change in the foreseeable future and so it is imperative that small patient organisations are given State support to continue and expand their remit in order to offer the type of services and supports families need.

## 5.4Reflective Piece

When initially considering what research I would undertake, I did not consider doing so in collaboration with CARL. I felt at the time that it would be adding additional unnecessary stress to the process. However, having found a topic I was interested in, I approached Kenneth to discuss it and he explained how it would work within the CARL framework. At that point, I still had reservations and took some time to consider my options and discuss it with the administrators of the community group I hoped to undertake research with. The group was very supportive and eager to be involved, as this type of research would have otherwise been unavailable to them. As it is a group that I have personal involvement with, my biggest fear was letting them down. This was discussed at length and with the encouragement of the group, I choose to proceed.

When I first began submitting applications to undertake the research and the application for ethical approval, I was extremely eager to get started on the research. The change in the ethical approval procedures, however, meant submitting a number of applications over a six month period and I became discouraged and began to wonder if I had made the wrong decision in pursuing primary research. It was at this time that the unanticipated benefits of working through CARL became clear to me, as I received so much encouragement and support from the CARL team, the community group and my tutor.

As my research progressed, I found that reflection became more important than I would have previously considered as some of the data I was receiving was quite emotional and personally relatable. This made me question my ability to conduct ‘grass roots’ or ‘action’ research, or whether this research would have been better served by being conducted by someone without a personal interest. My reflective journal became a benefit rather than a chore and I was glad to have the support of both my tutor and my community liaison person. After putting the raw data together, I took a step away from the research before I began the analysis as I needed the space to return to a more objective view point.

I feel I achieved this in the end but it proved to be a more difficult process than I had originally envisaged. Going forward, this is something that I feel will prepare me better to consider the emotional impact of undertaking research such as this. Reflecting back, however, I would do it again as I am passionate about my topic and that, coupled with the support I received, meant that my interest rarely wavered even when my motivation did.

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# Appendices

A: Consent from Community Organisation

B: Discussion Forum Guide

C: Discussion Forum Consent Sample

D: Information Sheet

E: Survey

F: Email from Rare Disease Office

# Appendix A


29/05/2016

To whom it may concern,

An administrator of this organisation, Tammy Meehan, has requested permission to proceed with an application to have research conducted on our behalf through the CARL program. This request has been put to the group members, who have approved it and so we the undersigned, wish to confirm that Tammy has approval to proceed in representing the organisation for this purpose.

We would also like to take this opportunity, as a group to thank you for your consideration of this research. We strive to advocate for and support those affected by serious illnesses, particularly Mitochondrial Disease and Disorders, and appreciate any awareness that highlights the issues our families are faced with.

Please do not hesitate to contact us if further information is required.

Kind regards,

Pamela Meehan

Audrey Mc Morrow

Teresa Noonan

# Appendix B

**Discussion Forum Guide: 'Anticipatory Grief in Families of Children with Mitochondrial Disease and the Role of Social Work'**

**Participants:**

The discussion forum will consist of 5-7 members of Mito Families Ireland. Members will be invited to join and as such, will be self- selected.

The invitation to join will be both emailed to group members and posted within the private Facebook group. Potential participants will be encouraged to contact me to discuss the subject matter as it may be distressing to some members and they may not want to participate. Participation will be restricted to those who are over 18, resident in Ireland and have not experienced a direct bereavement within the previous six months. At this point, I will also inform them that they may leave the discussion forum at any stage and/or contact me if they wish to discuss any matters arising.

Upon forming the group, I will welcome all members and give them an opportunity to introduce themselves to each other. As the group Mito Families Ireland (MFI) is primarily an online group and the discussion forum will be formed and conducted online, members may not have met or spoken previously. The rationale for conducting the group online is due to the fact that the MFI group members are geographically separated by a considerable distance and would have difficulty arranging childcare to attend a group in person, nor can the researcher cover any costs associated with travel. It is deemed that conducting such research via the internet will allow for greater accessibility and participation. Participants will be requested to sign an information sheet to confirm informed consent. These will be emailed to participants and a digital signature will be accepted.

**Group Rules:**

Following introductions, I will discuss group rules with participants and invite them to add to those I have outlined below:

- Confidentiality- Participation in this discussion forum is confidential. Members may individually disclose their participation if they so choose, but not the participation or details of others within the group.
- Each person is entitled to their own opinion and all opinions are valid. Please be respectful of others opinions whether you agree or disagree with them.

- Topics may be sensitive and if you feel that there is a section in which you would not like to participate in, you are under no obligation to do so. You are welcome to contact me at any point should you wish to discuss a matter further or feel you would like details of other support organisations.
- The data arising from this discussion forum will be used to create the questionnaire which will be distributed in the next phase of this research.
- This data will be kept confidential for the duration of the study, available only to me and my research supervisor. It will be securely stored on a password protected drive, to which only I will have access to. On completion of the project, the data will be retained for a further seven years and then destroyed.

**Duration:**

Each session is envisaged to take approximately one hour. However, if participants feel that they would like to discuss a topic further or indeed wish to end a session early, this can be decided by group consensus. It is estimated that two sessions will be required to explore and develop questions for the questionnaire. Each session will take place approximately one week apart and at a time which is agreed by group participants. These are conservatively estimated to begin late 2016 to allow for any disruption to the schedule as it is understood that participants may be affected by unforeseen circumstances such as hospital admissions of children. It is hoped that this will be completed prior to Christmas at the latest.

**Explanation:**

Regarding the questionnaire, it is currently envisaged that it will comprise of approximately twenty questions, with introductory questions focusing on gender, age group, and location/province. From this point, the questionnaire will be broken up into thematic sections and this is where I will be requesting input.

The themes which we will discuss are related to creating questions which will eventually provide answers to the following overarching research questions.

**5.** What issues, related to anticipatory grief, are experienced by families of children with Mitochondrial Disease?

6. What social work supports have families of children with Mitochondrial Disease availed of?

**Explanation of concepts:**

Following group rules, I will introduce the concept of anticipatory grief as 'anticipatory grief refers to the grief one may experience when faced with a poor prognosis of their loved one's life expectancy. That is, that in anticipating the likelihood of the death of their loved one, at an unspecified time, a person may experience a grief, not unlike that suffered by the bereaved'

I will also provide a list of some of the issues that may arise according to Rando (1997, p.48), such as

'powerlessness, fear, uncertainty, confusion: violations of the assumptive world; ongoing losses; personal depletion from the stress arising from demands for major re-adaptations and investments of self, time, and finances; long-term family disruption and disorganization; balancing of opposing needs, competing demands, discordant roles, clashing responsibilities, and antagonistic tasks; traumatizing physical, emotional, and social experiences during the illness; and major reactions of guilt, sorrow and depression, anger and hostility and anxiety." According to Rando (1997, p.49), the issues arising as a result of anticipatory grief can be considered the same as that of clinical Post Traumatic Stress Disorder.'

as well as Cadell's (2012), more positive 'post traumatic growth'.

I will give participants the opportunity to ask questions in order to familiarise themselves with these concepts and ensure that there is clarity on the topic.

**Themes:**

Themes will be presented to the group for discussion and members will be welcome to introduce further themes or expand as they feel relevant.

The proposed themes are as follows;

1) Anticipatory Grief- explanation, questions, and discussion.

2) Diagnosis- The process of diagnosis- discussion of needs and experiences, formulation of questions.

3) Liminality, goals, changes

4) Family systems theory- relationships, siblings

5) Stress

6) Finance, work, and the caring role

7) Medical issues, being understood and understanding- professionals and society.

8) Social life

9) Services/social work/ support

10) Growth, change

These are proposed only and flexibility will be given in that if one theme naturally follows another it will be allowed to do so. It is believed that rigidity in this schedule could stifle the emergence of ideas and questions and so this is only intended as a rough guide.

The end of the second session will be used to tie up any loose ends and close the group, thanking all for their participation.

Appendix C

# Consent Form
# Discussion Forum


**Purpose of the Study.** As part of the requirements for the Master's Degree in Social Work at UCC, I have to carry out a research study. The study is concerned with Anticipatory Grief in Families of Children with Mitochondrial Disease and the Role of Social Work.

**What will the study involve?** This first phase of the research will involve participating in a discussion panel in which the questions for the survey will be developed. This survey will then be distributed to all members of Mito Families Ireland. It is estimated that the forum will run over two sessions, each taking approximately one hour. Each session will be conducted online.

**Why have you been asked to take part?** You have been asked because this research specifically relates to the experience of families of children with Mitochondrial Disease.

**Do you have to take part?** No, you are under no obligation to take part in this research. Participation is strictly voluntary and you may withdraw from this study, if you so choose, at any time. You will not experience any favour or disfavor as a result of your participation or non-participation in this study.

**Will your participation in the study be kept confidential?** Your participation in the discussion will be confidential to all except the other members of the forum and I. You will be requested to digitally sign a consent form before participating.

**What will happen to the information which you give?** The data will be kept confidential for the duration of the study, available only to me and my research supervisor. It will be securely stored on a password protected drive, to which only I will have access to. On completion of the project, the data will be retained for a further ten years and then destroyed.

**What will happen to the results?** The results of the discussion panel will be used to create the survey, the results of which will be presented in the thesis, which will be seen by my supervisor, a second marker, the external examiner and future students on the course. The study may be published in a research journal and on the CARL website, which makes it accessible to anyone who chooses to read it. Furthermore, as a CARL project, it is hoped that Mito Families Ireland may be able to use the thesis to lobby for awareness and/or funding or other activities they deem relevant.

**What are the possible disadvantages of taking part?** Participation in the discussion forum and talking about your experience may cause some distress.

**What if there is a problem?** If you are feeling distressed as a result of your participation, please feel free to contact me at tammy.meehan@umail.ucc.ie or of course, the group, Mito Families Ireland. If you feel the need for professional support, please contact your G.P.

**Who has reviewed this study?** As a CARL project, approval must be given by the CARL panel as well as the Research Ethics Committee in the Department of Applied Social Studies, before studies like this can take place.

**Any further queries?** If you need any further information, you can contact me:
Tammy Meehan at tammy.meehan@umail.ucc.ie

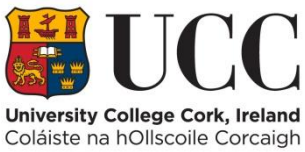
## *Disclosure: This researcher is both a founding member of Mito Families Ireland and is a family member of someone affected by Mitochondrial Disease.

**I state that I am aware of what this research involves. The purpose of the study has been explained to me. I am willing to participate voluntarily in an interview for this research with the understanding that I can withdraw at any time up until the survey design has been completed and I have been notified of such by the researcher, without repercussions**. ☐

Signed: _____ Date:_____

Appendix D

# INFORMATION SHEET




**Purpose of the Study.** As part of the requirements for the Master's Degree in Social Work at UCC, I have to carry out a research study. The study is concerned with Anticipatory Grief in Families of Children with Mitochondrial Disease and the Role of Social Work.

**What will the study involve?** The study will involve completing an anonymous online survey about your experience as a family member of someone affected by Mitochondrial Disease, which has been developed by a focus group consisting of members of Mito Families Ireland and the researcher. It is estimated that this survey will take approximately 15-20 minutes to complete.

**Why have you been asked to take part?** You have been asked because this research specifically relates to the experience of families of children with Mitochondrial Disease.

**Do you have to take part?** No, you are under no obligation to take part in this research. Participation is strictly voluntary and you may withdraw from this study, if you so choose, at any time. As this study is anonymous, you will not be asked to sign a consent form, **consent will be assumed by your participation in the study**.

**Will your participation in the study be kept confidential?** Yes, your identity will be unknown, even to me, except that you are a member of Mito Families Ireland.

**What will happen to the information which you give?** The data will be kept confidential for the duration of the study, available only to me and my research supervisor. It will be securely stored on a password protected drive, to which only I will have access to. On completion of the project, the data will be retained for a further seven years and then

destroyed.

**What will happen to the results?** The results of this research will be presented in the thesis, which will be seen by my supervisor, a second marker, the external examiner and future students on the course. The study may be published in a research journal and on the CARL website, which makes it accessible to anyone who chooses to read it. Furthermore, as a CARL project, it is hoped that Mito Families Ireland may be able to use the thesis to lobby for awareness and/or funding or other activities they deem relevant.

**What are the possible disadvantages of taking part?** While I don't envisage any negative consequences for you in taking part, it is possible that in answering the survey/questionnaire about your experience may cause some distress.

**What if there is a problem?** If you are feeling distressed as a result of your participation, please feel free to contact me at tammy.meehan@umail.ucc.ie or of course, the group, Mito Families Ireland. If you feel the need for professional support, please contact your G.P.

**Who has reviewed this study?** As a CARL project, approval must be given by the CARL panel as well the Department of Social Work, UCC, before studies like this can take place.

**Any further queries?** If you need any further information, you can contact me:

Tammy Meehan at tammy.meehan@umail.ucc.ie

***Disclosure: This researcher is both a founding member of Mito Families Ireland and is a family member of someone affected by Mitochondrial Disease.**

# Appendix E


## Anticipatory Grief

Welcome to My Survey

1 / 7 14%

**The purpose of the study:**
As part of the requirements for the Master's Degree in Social Work at UCC, I am carrying out a research study. The study is concerned with Anticipatory Grief in Families of Children with Mitochondrial Disease and the Role of Social Work.

**What will the study involve?**
The study will involve completing an anonymous online survey/questionnaire about your experience as a family member of someone affected by Mitochondrial Disease, which has been developed by a discussion forum consisting of members of Mito Families Ireland and the researcher. It is estimated that this survey/ questionnaire will take approximately 15-20 minutes to complete.

**Why have you been asked to take part?**
You have been asked because this research specifically relates to the experience of families of children with Mitochondrial Disease.

**Do you have to take part?**
No, you are under no obligation to take part in this research. Participation is strictly voluntary and you may withdraw from this study, if you so choose, at any time during the completion of the survey. As this study is anonymous, you will not be asked to sign a consent form, consent will be assumed by your participation in the study.

**Will your participation in the study be kept confidential?**
Yes, your identity will be unknown, even to me, except that you may be a member of Mito Families Ireland.

**What will happen to the information which you give?**
The data will be kept confidential for the duration of the study, available only to me and my research supervisor. It will be securely stored on a password protected drive, to which only I will have access. On completion of the project, the data will be retained for a further ten years and then destroyed.

**What will happen to the results?**
The results of this research will be presented in the thesis, which will be seen by my supervisor, a second marker, the external examiner and future students on the course. The study may be published in a research journal and on the CARL website, which makes it accessible to anyone who chooses to read it. Furthermore, as a CARL project, it is hoped that Mito Families Ireland may be able to use the thesis to lobby for awareness and/or funding or other activities they deem relevant.

**What are the possible disadvantages of taking part?**
Completing the survey and writing about your experience may cause some distress.

**What if there is a problem?**
If you are feeling distressed as a result of your participation, please feel free to contact me at tammy.meehan@umail.ucc.ie or of course, the group, Mito Families Ireland. If you feel the need for professional support, please contact your G.P.

**Who has reviewed this study?**
As a CARL project, approval has been granted by the CARL panel as well as the Research Ethics Committee in the Department of Applied Social Studies, before this study commenced.

**Any further queries? If you need any further information, you can contact me:**

Tammy Meehan at tammy.meehan@umail.ucc.ie

***Disclosure: This researcher is both a founding member of Mito Families Ireland and is a family member of someone affected by Mitochondrial Disease.**

**By clicking NEXT, you are consenting to participate in this study.**

Next

## Personal Information

2 / 7 29%

* 1. Please indicate your gender.

- ◯ Female
- ◯ Male

Other (please specify)

* 2. Which age category applies to you?

- ◯ 18-25
- ◯ 26-35
- ◯ 36-45
- ◯ 46-55
- ◯ 56-65
- ◯ 66+

* 3. Please indicate your province of usual residence.

- ◯ Munster
- ◯ Ulster
- ◯ Leinster
- ◯ Connaught

* 4. Please indicate your relationship to the person affected by Mitochondrial Disease.

- ◯ Parent
- ◯ Sibling
- ◯ Grandparent
- ◯ Aunt/Uncle

Other (please specify)

## Anticipatory Grief

3 / 7 43%

Anticipatory grief refers to the grief 'one may experience when faced with a poor prognosis of their loved one's life expectancy. That is, that in anticipating the likelihood of the death of their loved one, at an unspecified time, a person may experience a grief, not unlike that suffered by the bereaved'.

When someone is faced with this, they may experience: 'powerlessness, fear, uncertainty, confusion: violations of the assumptive world; ongoing losses; personal depletion from the stress arising from demands for major re-adaptations and investments of self, time, and finances; long-term family disruption and disorganization; balancing of opposing needs, competing demands, discordant roles, clashing responsibilities, and antagonistic tasks; traumatizing physical, emotional, and social experiences during the illness; and major reactions of guilt, sorrow and depression, anger and hostility and anxiety'.

According to Rando (1997, p.49), the issues arising as a result of anticipatory grief can be considered the same as that of 'clinical Post Traumatic Stress Disorder.'

5. Do you feel the above statement accurately reflects your experience upon/after diagnosis?

- ◯ Yes
- ◯ No

Is there anything you would like to add?

* 6. Which feelings would you say are most applicable (if any) to your experience?

| | Very much so | Yes, often | Somewhat | I'm not sure | Not at all |
|---|---|---|---|---|---|
| Powerlessness or uncertainty | ☐ | ☐ | ☐ | ☐ | ☐ |
| Ongoing losses | ☐ | ☐ | ☐ | ☐ | ☐ |
| Personal depletion/fatigue | ☐ | ☐ | ☐ | ☐ | ☐ |
| Disruption to family unit | ☐ | ☐ | ☐ | ☐ | ☐ |
| Financial loss | ☐ | ☐ | ☐ | ☐ | ☐ |
| Social loss | ☐ | ☐ | ☐ | ☐ | ☐ |
| Guilt or depression | ☐ | ☐ | ☐ | ☐ | ☐ |
| Anger | ☐ | ☐ | ☐ | ☐ | ☐ |
| Stress | ☐ | ☐ | ☐ | ☐ | ☐ |
| Trauma | ☐ | ☐ | ☐ | ☐ | ☐ |
| Clashing responsibilities | ☐ | ☐ | ☐ | ☐ | ☐ |
| Fear | ☐ | ☐ | ☐ | ☐ | ☐ |

Other (please specify)

* 7. Within 48 hours of diagnosis, after the initial meeting with the doctor, did you meet with any of the following?

- ☐ Consultant/ Hospital Based Doctor
- ☐ Family GP
- ☐ Genetic Counsellor
- ☐ Social Worker
- ☐ Nobody
- ☐ Other (please specify)

8. Who would you liked to have met with after diagnosis?

- ☐ Consultant/ Hospital Based Doctor
- ☐ Family GP
- ☐ Genetic Counsellor
- ☐ Social Worker

* 9. At the time of diagnosis, how was the diagnosis explained to you?

* 10. Who was with you when your loved one received the diagnosis? (you may tick all that apply)

- ☐ I was alone
- ☐ The patient
- ☐ My partner
- ☐ My siblings
- ☐ The patient's siblings
- ☐ My parents
- ☐ A friend/ friends
- ☐ Other (please specify)

## Liminality

4 / 7 57%

Little, Jordens, Paul, Montgomery and Philpson (1998), conducted a study which found that cancer survivors experienced a feeling of liminality, as a result of the uncertainty faced during serious, potentially life-threatening illness. Turner (1969) describes liminality as a state of being 'in- between', or the state of transition where one is neither in one place nor another. Ferguson (2011, p.49) states that 'everyday life depends on the maintenance of order and routine and spaces where there is an absence of such order are experienced as dangerous.' It is hypothesised that a sense of liminality may occur in those caring for children with uncertain prognoses (such as in Mitochondrial Disease) carers may feel immobilised (Olson, 2014).

Or in more everyday language – being in limbo

* 11. Do you feel the above statement reflects your experience?
(1 being 'yes, this strongly reflects my experience' and 5 being 'No, not at all.')

| Yes, strongly | | Unsure | | No, not at all |
|---|---|---|---|---|
| ○ | ○ | ○ | ○ | ○ |

* 12. If you felt that the above statement reflected your experience, what do you feel would have helped?

- ☐ More medical information
- ☐ Information on informal support services/groups
- ☐ Financial support information
- ☐ Referral to counsellor
- ☐ Other (please specify)

* 13. After diagnosis, where did you get information on Mitochondrial Disease from?

- ☐ The hospital medical staff
- ☐ Non medical hospital staff (Counselor, social worker, chaplaincy)
- ☐ Family GP
- ☐ Community based services (public health nurse, etc.)
- ☐ Professional support/service groups
- ☐ Community support groups (online/ in person)
- ☐ Google
- ☐ Other (please specify)

Family Systems

5 / 7 71%

Cadell, Kennedy and Hemsworth (2012), highlight that the 'usual challenges' faced by parents raising children are further compounded by the stress of 'caring for a child with a life-limiting illness' (p.356). In light of our understanding of systems theory and concept of reciprocity (Payne, 2014, p.192), which maintains that 'if one part of a system changes, that change affects all the other parts, therefore they all change.', then the challenges faced in caring for a child with a life-limiting illness likely effects the entire family. Cadell et. al (2012, p.358) found that families affected by a life-limiting illness had to make 'significant adjustments' in their day to day lives as well as to future plans, which impinged on the life of all family members. Kerr (2000), based on Bowen's Family Systems Theory, discusses the emotional interconnectedness of a family unit and suggests that when anxiety is present throughout the family, the emotional interconnectedness results in stress rather than comfort, further heightening levels of anxiety. This, paired with the ambiguity of health and illness in a family affected by a life-limiting illness (Cadell et. al., 2012) may deeply affect the well-being and functioning of the entire family unit.

* 14. Apart from the patient and primary carers, who do you feel was most affected by the illness?

- ◯ My Partner
- ◯ Me
- ◯ My Siblings
- ◯ My Parents
- ◯ The patient's siblings
- ◯ The patient's extended family
- ◯ Friends
- ◯ Other (please specify)

* 15. Has the illness affected your financial status?

- ◯ Yes
- ◯ No

* 16. Has the illness affected your ability to socialise?

- ◯ Yes
- ◯ No

* 17. Has the illness affected your employment status?

- ◯ Yes
- ◯ No

18. Has the illness affected your relationships with others in either a positive or negative manner?

- ◯ Positively
- ◯ Negativly
- ◯ Neither- my relationships remain unchanged.

Other (please specify)

* 19. Do you feel that the illness has changed how people treat the patient?

- ◯ Yes, they are given more attention by others
- ◯ Yes, others avoid/ ignore them
- ◯ No, they are treated no differently to before the illness.

Other (please specify)

20. Has your family ever been invited to meet with any professionals as a unit, to discuss the illness?

- [ ] Doctors
- [ ] Social workers
- [ ] Genetic counsellors
- [ ] Counsellors/ Psychologists
- [ ] Nobody

Other (please specify)

* 21. Illness in a family effects everyone differently. Do you feel that any of the following apply to you since diagnosis?

- [ ] I am angrier or bitter
- [ ] I am more assertive
- [ ] I have felt depressed
- [ ] I have lost my identity outside of caring
- [ ] I am more proactive
- [ ] My identity has changed positively
- [ ] I feel more lonely and/or isolated
- [ ] I am emotionally stronger
- [ ] I have become more empathetic and compassionate to others
- [ ] I am anxious and nervous
- [ ] I have less patience with others

Other (please specify)

- [ ] My partner
- [ ] My immediate family
- [ ] My extended family
- [ ] My friends
- [ ] My religion
- [ ] Medical professionals
- [ ] Support proffesionals
- [ ] Informal support networks
- [ ] Nobody

Other (please specify)

## The Role of Social Work

7 / 7 100%

Social workers may work in a variety of different areas such as hospitals, primary care centres, community organisations and nonprofit organisations among others. If you could 'tailor' a social worker to meet your needs....

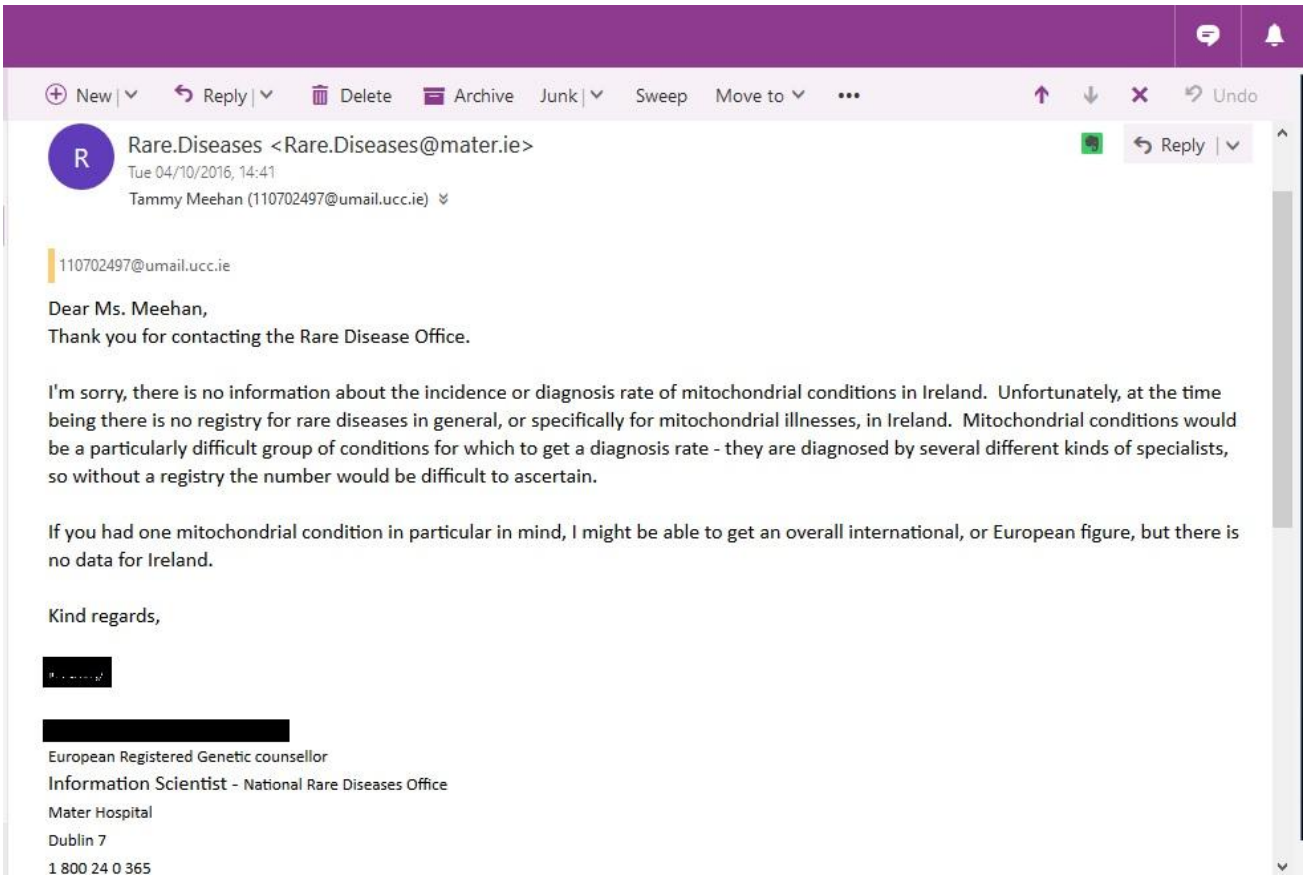
* 23. What services would they provide to the patient?

24. What services would they provide to you?

25. How often would you like to meet or contact them?

Prev Submit

# Appendix F



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R

Rare.Diseases <Rare.Diseases@mater.ie>

Tue 04/10/2016, 14:41

Tammy Meehan (110702497@umail.ucc.ie)

Reply

110702497@umail.ucc.ie

Dear Ms. Meehan,
Thank you for contacting the Rare Disease Office.

I'm sorry, there is no information about the incidence or diagnosis rate of mitochondrial conditions in Ireland. Unfortunately, at the time being there is no registry for rare diseases in general, or specifically for mitochondrial illnesses, in Ireland. Mitochondrial conditions would be a particularly difficult group of conditions for which to get a diagnosis rate - they are diagnosed by several different kinds of specialists, so without a registry the number would be difficult to ascertain.

If you had one mitochondrial condition in particular in mind, I might be able to get an overall international, or European figure, but there is no data for Ireland.

Kind regards,

[redacted]

[redacted]
European Registered Genetic counsellor
Information Scientist - National Rare Diseases Office
Mater Hospital
Dublin 7
1 800 24 0 365