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A Resiliency Perspective of the Lived Experience of Parenting Infants and Young Children with Cystic
Fibrosis in the Context of Early Lung Disease Surveillance

Cindy A Branch-Smith

This thesis is submitted in fulfilment of the requirements for the degree of
Doctor of Philosophy in Psychology

Edith Cowan University

School of Arts and Humanities

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Edith Cowan University

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Preface

The linking of hope and empowerment can be seen in a description of the virtues of hope in the book 'The Anatomy of Hope' by Jerome Groopman (2005) who stated "hope can arrive only when you realise that there are real options and that you have genuine choices. Hope can flourish only when you believe that what you do can make a real difference, that your actions can bring a future different from the present. To have hope, then, is to acquire a belief in your ability to have some control over your circumstances. You are no longer entirely at the mercy of forces outside yourself" (p. 26).

For parents dealing with their young child's chronic disease, existence of hope for their child, for future generations inflicted with the condition and hope for a cure can be generated through advancements in science and technology, and clinical agendas that allow early tracking of the course of disease progression. A Western Australian mother of a 4 year old child living with cystic fibrosis commented on how she felt about being able to track her child's condition through cutting-edge early surveillance techniques. The linking of hope and empowerment described in Jerome Groopman's book is woven through her statement - *I just think that you've got somewhere to start. If you've got nowhere to start then you can't do anything, so for me this is a starting point. I've got information and I know what the state of her lungs are like. She's 4 and I hope she has a long life and I hope we can keep her lungs as healthy as we can, and this is where we start with it. So as I said, I'm grateful that it's there, I'm grateful that I can participate in it. Firstly to get information for ourselves but secondly to help with research to help others and, ultimately to find a cure. There's lots to learn about CF I've learnt.*

Emulating transition of writing styles here in the preface, the reader will discern a change from an academic style to a more conversational tone from chapter 3 onwards from the academic setting where the literature on parenting and CF is situated, into my data collection methods and the world of my informants. That is, I'm choosing to stay true to my informants' existential experience and not to dress it with psychological terminology.

Abstract

Scientific and technological advances over the past few decades have contributed to an exponential increase in life expectancy for infants born with cystic fibrosis (CF), which can cause fatal lung disease. There has been a paradigmatic shift from reactive treatment towards early disease detection and aggressive intervention of paediatric CF. Previous research has investigated parents' experiences of the diagnosis phase and later life stages such as adolescence and transition to adulthood. Less is understood about the experience of parenting infants and young children with CF, and no research addresses this during early surveillance for CF lung disease. Early surveillance is rapidly emerging as the likely framework for future therapeutic intervention trials in young children, nationally and internationally. Thus, understanding parents' mental health is essential to providing support to families during their child's intensive, early treatment.

This research aimed to explore how parents construct their lived experiences of parenting and how they attribute meaning to these experiences. Particular attention was given to coping strategies, informed by a salutogenic model of protective factors that may contribute to family adaptation and resilience. A qualitative methodology, guided by theoretical underpinnings of phenomenology and constructivism was used to explore parents' experiences. Semi-structured interviews were conducted with 67 parents (46 mothers and 21 fathers aged between 23 and 52 years) across sites where early surveillance for CF lung disease operates in Australia. A thematic analytic approach was used to analyse the data.

Data revealed that whilst parents described adverse psychological consequences of their child's participation in early surveillance, it was also evident that beneficial outcomes eclipsed adversities. Five major themes represent the emergent structure of parents' lived experience: Redefined Expectations and Reimagined Identity, Redefined Reality; Understanding the Unknown and Understanding Uncertainty; Good Days and Bad Days, Fluctuation between Positive and Negative Outlook On Life; Early Surveillance is a Significant Event; And Early Surveillance is in the Best Interest of my Child. Collectively, these highlight that only attending to adverse experiences and outcomes would be to overlook significant beneficial psychological experiences and outcomes of this unique parenting experience. Pathways that enable adaptation to paediatric chronic disease in the context of early surveillance were also identified using a resiliency framework of family adjustment and adaption. These findings contribute to current conceptualisations of parenting children with chronic conditions undergoing modern interventions that aim to delay disease. This research may also inform policy-making and models of best paediatric clinical practice, particularly those embracing a biopsychosocial model of care that promotes parents' mental health, as well as family resilience and

adaptation. The importance of these findings underscores the need for further research to expand our understanding of the parenting experience in the unique context of early surveillance.

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Acknowledgements

While my name is alone on the front cover of this dissertation, I am by no means its sole contributor. I would like to acknowledge those who have supported me over the past 4 years (and beyond) and who, in turn, have contributed to the completion of this doctoral dissertation.

Firstly, I would like to thank the parents from Princess Margaret Hospital and The Royal Children's Hospital who volunteered their time to participate in my research. Without you this study would not have been possible. I appreciate your willingness and your trust and openness throughout the interviews. Thank you for sharing your experiences with me.

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To my friend Linda, your support and encouragement have been a shining light in dark times. Your phone calls and city visits are always appreciated. The homestead villa stays have been a wonderful way to refocus, stay fresh and push on. You've had an uncanny ability to help me see beyond the trees in the forest to include the grain of the wood. You are a true friend.

Parenting Children with Cystic Fibrosis

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Dedication

I dedicate this doctoral dissertation to my Sulli-girl. We started this journey together and you saw me through to the end. Along the way you taught me patience, stability, and responsibility. More importantly, you taught me again how to love and how to be loved. I'll miss you always, and I'm forever grateful that our paths crossed.

Publications Arising From This Thesis

- 2015 Branch-Smith, C., Pooley, J., Shields, L., Stick, S., & Douglas, T., on behalf of AREST-CF. Early surveillance of cystic fibrosis: Parents' psychosocial experiences of disease knowledge and their child's medical procedures. *Pediatric Pulmonology*, 50 (S41), 432.
- 2014 Branch-Smith, C., Douglas, T., Shields, L., Stick, S., Massie, J., & Pooley, J., on behalf of AREST-CF. Parental experiences of early pulmonary surveillance for children with cystic fibrosis: A research proposal for improved family psychosocial outcomes. *Working Papers in the Health Sciences*, 8 (1), 1-8.
- 2014 Branch-Smith, C., Pooley, J., Shields, L., Stick, S., & Douglas, T., on behalf of AREST-CF. 'Arresting' psychological issues for better health outcomes in parents of infants and young children with cystic fibrosis. *Journal of Cystic Fibrosis*, 13, (Supp. 2), S116.
- 2014 Branch-Smith, C., Pooley, J., Shields, L., Stick, S., Douglas, T., & Rosenow, T., on behalf of AREST-CF. 'Arresting' psychological issues for parents of infants and young children with cystic fibrosis. *Pediatric Pulmonology*, 49 (S38), 438.
- 2013 Branch-Smith, C., Pooley, J., Shields, L., Stick, S., & Douglas, T., on behalf of AREST-CF. What do parents experience and how do they cope with the AREST-CF early surveillance program for infants and children with cystic fibrosis? *Journal of Cystic Fibrosis*, 12, (Supp. 1), S3.

Abbreviations

AREST CF: Australian Respiratory Early Surveillance Team for Cystic Fibrosis

BAL: bronchoalveolar lavage

CIHR: Canadian Institutes of Health Research

CF: cystic fibrosis

CFA: Cystic Fibrosis Australia

CFTR: cystic fibrosis transmembrane conductance regulator

CT: chest computed tomography scan

EU: European Union

GA: general anaesthetic

HCP: healthcare professional

LMA: laryngeal mask airway

NBS: newborn screening

PMH: Princess Margaret Hospital (Perth, Western Australia)

PTSS: post-traumatic stress syndrome

RCH: Royal Children's Hospital (Melbourne, Victoria)

TIDES: The International Depression/Anxiety Epidemiological Study

Glossary

Adaptation: the process of change by which an individual becomes better-suited to their situation or environment (Kent, 1998).

Biopsychosocial: a theoretical model with the underlying premise that biological, psychological (including cognitive, emotional and behavioral), and social (socio-economical, socio-environmental, and cultural) factors, all play a significant role in human functioning in the context of disease or illness (Santrock, 2007).

Chronic condition: a disease or illness that has a prolonged course, that does not resolve spontaneously, for which a complete cure is rarely achieved (National Public Health Partnership, pp.3).

Chronic sorrow: a sense of recurring, extended and profound sadness (Olshansky, 1962)

Cognitive dissonance: an emotional state occurring when two simultaneously held attitudes or cognitions are inconsistent, or when there is conflict between belief and behaviour (Reber & Reber, 2001).

Constructivism: theoretical assumption in qualitative psychology that assumes a relativist ontology (there are multiple realities), a subjectivist epistemology (researcher and respondent co-create understandings), and a naturalistic (in the natural world) set of methodological procedures (Denzin & Lincoln, 2003).

Coping: thoughts and behaviours people use to manage internal and external demands of situations that are appraised as stressful (Lazarus and Folkman, 1984)

Covert disease progression: early disease that has advanced but goes undetected by traditional measures.

Depression: generally, an affective state characterised by a sense of inadequacy, a feeling of despondency, a decrease in activity or reactivity, pessimism, sadness and related symptoms (Reber & Reber, 2001).

Diagnosis: the identification of the nature of an illness or other problem by clinical examination of the symptoms (Kent, 1998).

Early surveillance: a set of medical procedures that allow clinicians to track the course of early, often covert, progression of lung disease long before signs and symptoms are apparent.

Family adaptation: the outcome of a family's efforts to bring a new level of balance and coherence, as well as a satisfactory level of functioning, to a family crisis situation. The process to family adaptation involves coping strategies, from an individual and family perspective (Danielson, et al., 1993).

Family resiliency: a family's potential to recover and restore itself following a crisis, change, or other stressors facing the family unit (Danielson, et al., 1993).

Hope: the feeling that what is wanted can be had or that events will turn out for the best; a desire accompanied by expectation of or belief in fulfilment (Kent, 1998).

Intervention: The act of intervening or interceding with the intent of modifying the outcome. A clinical intervention is usually undertaken to help treat or cure a condition (Kent, 1998).

Knowledge: facts, information, and skills acquired through experience or education (Kent, 1998).

Lived experience: Personal account of the world gained through direct, first-hand involvement in everyday events (Kent, 1998).

Locus of control: the extent to which individuals believe they can control events affecting them (Rotter, 1966). An internal locus of control orientation is a belief about whether the outcomes of actions are contingent on what a person does, whereas an external locus of control orientation is a belief that events are outside of personal control (Zimbardo, 1985).

Morbidity: the state of being diseased or unhealthy (Kent, 1998).

Mortality: the condition of one day having to die or the rate of failure or loss (Kent, 1998).

Normalisation: a cognitive and behavioural process of redefining one's life through which ideas and actions come to be seen as *normal* and become taken-for-granted, or natural, in everyday life (Kent, 1998).

Parent: any person/s defined by the family itself as the primary caregivers for the child/ren.

Paediatric: a branch of medicine dealing with the health and medical care of infants, children, and adolescents from birth up to the age of 18 years (Kent, 1998).

Parental: intrinsic constructs associated with being a parent

Parent identity: the role and functions of parenting that give meaning of being a parent.

Parenting: behaviours associated with, or as a result of, being a parent.

Parenting competence: a system of knowledge, skills, and abilities that allow a parent to successfully fulfil their parental responsibilities and also prevent, or deal with, crisis situations in a manner that would contribute to their child's development (Glăveanu, 2009).

Phenomenology: theoretical framework for qualitative research which focuses on people's understandings and interpretations of their experiences in their own terms, emphasising these as explanations for their actions (Liamputtong & Ezzy, 2005). In general, phenomenological psychological research aims to clarify situations lived by persons in everyday life (Giorgi & Giorgi, 2003).

Relinquishment: to give to another person or group (Kent, 1998).

Resiliency: an individual's ability to properly adapt to stress and adversity (Kent, 1998).

Resiliency Model of Family Stress, Adjustment and Adaptation: a theoretical model guiding the understanding of a family's response to a stressor and recovery from crisis (Danielson, et al., 1993).

Salutogenic: an approach focusing on factors that support human health and well-being, rather than on factors that cause disease (Antonovsky, 1987).

Self-efficacy: A person's belief about his or her ability and capacity to accomplish a task or to deal with the challenges of life (Bandura, 1994). Those with high self-efficacy believe in their capacity to influence outcome of events whereas people low on self-efficacy believe they do not have capacity to influence outcomes of events in their lives.

Therapeutic misconception: the belief that the purpose of a clinical trial is to directly benefit the individual patient rather than to gather data to develop scientific knowledge (Appelbaum, 1982).

Treatment regimen: a systematic plan designed to improve and maintain the health of a patient (Kent, 1998).

Table of Contents

Use of Thesis.....	i
Preface	ii
Abstract.....	iii
Copyright and Access Declaration	Error! Bookmark not defined.
Acknowledgements.....	v
Dedication	vii
Publications Arising From This Thesis	viii
Abbreviations.....	ix
Glossary.....	x
Table of Contents.....	xiii
List of Tables	xix
List of Figures	xx
List of Appendices	xxi
CHAPTER 1: Introduction	1
1.1 Research context.....	2
1.1.1 The psychology of parenting in paediatric chronic disease	2
1.1.2 Paediatric Cystic Fibrosis in Australia.....	10
1.1.3 Current Australian treatment management practices for infants and young children in the 21 st century	12
1.1.4 Cystic fibrosis treatment management in infants and children from diagnosis to 7 years in Western Australia and Victoria: Australian innovation for paediatric CF management	14
1.2 The current research.....	18
1.2.1 Research rationale	18
1.2.2 Exploring the lived experience of parenting children with CF in the context of early surveillance	19
1.2.2.1 Parallels drawn from medical treatment procedures.....	20
1.2.2.2 Parallels drawn from similar condition trajectories.....	21
1.2.3 Aims of the research	23
1.2.4 Research questions	23
1.2.5 Implications of the research	24
1.3 Overview of the thesis	25

CHAPTER 2: Cystic fibrosis in the 21st century	28
2.1 What is CF?.....	29
2.1.1 Prevalence, life expectancy, causes and clinical features.....	29
2.1.2 Chronology of CF into the 21 st century.....	30
2.1.3 CF diagnosis: Current Australian practice via newborn screening.....	31
2.1.4 What is it like to be a young child with CF in the 21st century?.....	32
2.1.5 Burden of early CF care on families in the 21 st century.....	34
2.1.6 Summary and conclusion.....	36
2.2 Families living with CF.....	36
2.2.1 Psychological adjustment to a CF diagnosis.....	36
2.2.2 Outcomes of parenting a child with CF.....	37
2.2.2.1 Parental stress, depression and anxiety.....	37
2.2.2.2 Guilt and chronic sorrow.....	39
2.2.2.3 Parental identity formation and parenting self-efficacy.....	41
2.2.3 Parental characteristics moderate child health outcomes.....	42
2.2.4 Summary and conclusion.....	43
2.3 Coping in families with CF.....	43
2.3.1 Coping with diagnosis.....	43
2.3.2 Coping with CF in the early years.....	45
2.3.2.1 Hope and resiliency in the face of adversity.....	47
2.3.3 Summary and conclusion.....	50
2.4 Aims and significance of this research.....	50
2.4.1 What does this study hope to achieve and why is it important?	50
2.4.2 How can this research help families?	51
2.4.3 What else can this research add to the body of knowledge?.....	52
CHAPTER 3: Substantiative Theoretical and Methodological Derivatives	54
3.1 Introduction	55
3.2 Theoretical frameworks.....	55

Parenting Children with Cystic Fibrosis

3.2.1 Transactional Model of Stress and Coping.	55
3.2.2 The Resiliency Model of Family Stress, Adjustment and Adaptation.	57
3.2.3 Summary and conclusion.	62
3.3 Study design	63
3.3.1 Conceptualising qualitative research.....	63
3.3.1.1 Methodological and theoretical perspectives and implications.....	66
3.3.2 Qualitative methods.	67
3.3.2.1 Scientific rigour.....	68
3.3.2.1.1 Operationalising rigour in qualitative research.	69
3.3.2.2 Phenomenology.....	70
3.3.2.3 Data collection technique.....	73
3.3.2.4 Sampling technique.....	75
3.3.2.5 Sample size and saturation.....	76
3.3.2.6 Thematic analysis.....	77
3.4 Summary	77
CHAPTER 4: Methods	79
4.1 Restatement of aims	80
4.2 Research questions	80
4.3 Methodology.....	80
4.3.1 Phenomenology as theoretical framework.	80
4.3.2 Constructivism as philosophical underpinnings.....	81
4.4 Methods.....	83
4.4.1 Sample.....	83
4.4.2 Participant characteristics	84
4.4.2.1 Western Australian sample.....	85
4.4.2.2 Victorian sample.....	85
4.4.3 Recruitment.	85
4.4.5 Ethics considerations.	86
4.4.5.1 Research conduct.....	86

Parenting Children with Cystic Fibrosis

4.4.5.2 Data management.....	87
4.4.5.3 Informed consent process.....	88
4.4.5.4 Consequences of the research.....	89
4.4.5.5 Role of the researcher.....	89
4.4.5.6 Additional ethical considerations.....	90
4.5 Data collection	90
4.5.1 Interview procedure.	90
4.5.2 Data collection instrument.	93
4.6 Exploratory analysis	94
4.7 Scientific rigour	99
4.7.1 Throughout the research process.....	99
4.7.2 Data generation.	100
4.7.3 Data coding and analysis.....	101
4.7.4 Presentation of findings.....	103
4.8 Summary	104
CHAPTER 5: Findings: The lived experience of parenting children with CF in the context of early surveillance	105
5.1 Identifying Themes.....	106
5.2 Interpretation of phenomena – what is the ‘lived experience’ of parenting children with CF within the context of early surveillance for lung disease?	107
5.3 Redefined expectations and reimagined identity; redefined reality	108
5.3.1 Collapsed and redefined expectations.....	108
5.3.2 New normal.....	112
5.4 Understanding the unknown and understanding uncertainty	116
5.4.1 Understanding CF.....	117
5.4.2 Understanding early surveillance for CF lung disease.	122
5.5 Good days and bad days; Fluctuation between positive and negative outlook on life	126
5.5.1 Bad days.	126
5.5.1.1 Fear and anxiety as outcomes of uncertainty in morbidity and mortality.....	127

Parenting Children with Cystic Fibrosis

5.5.1.2 Chronic sorrow.....	130
5.5.2 Good days.	132
5.5.2.1 Parenting competence supports the parental role.....	132
5.5.2.2 Gratitude, both for their child and for early surveillance.....	135
5.5.2.3 Perceived control and power through certainty of disease severity from early surveillance results.....	137
5.5.3 Fluctuation between good days and bad days.	139
5.6 Early surveillance is a significant event.....	140
5.6.1 Indicator of treatment efforts and an examination of parenting competency.	141
5.6.2 Parental outcomes directly from early surveillance.	144
5.6.2.1 Lack of control and helplessness at time of relinquishment for medical procedures.....	144
5.6.2.2 Altruism for future CF generations.....	146
5.6.3 Fear from diagnosis exacerbated at annual review.....	148
5.7 Early surveillance is in the best interest of my child.....	152
5.7.1 Perceived clinical benefit to child health – presence of the therapeutic misconception? 152	
5.7.2 Cognitive dissonance results in anxiety and/or ambivalence.....	155
5.8 Construction and conceptualisation of hope.....	160
5.9 Summary and conclusion.....	163
CHAPTER 6: Parenting a child with CF in the context of early disease surveillance: Adapting to paediatric chronic disease from a theoretical perspective.....	166
6.1 Introduction	167
6.2 Application of Resiliency Model of Family Stress, Adjustment and Adaptation.....	167
6.2.1 Newly Instituted Patterns of Functioning.....	171
6.2.2 Situational and family appraisal.....	172
6.2.2.1 Situational appraisal.....	172
6.2.2.2 Family appraisal.....	173
6.2.3 Meaning-making.	174
6.2.4 Family resources – Strengths and coping strategies as family capability.....	175

Parenting Children with Cystic Fibrosis

6.2.5 Social support.....	178
6.2.5.1 Emotional support.....	179
6.2.5.2 Network support.....	179
6.2.5.3 Altruistic support.....	180
6.2.6 Coping patterns.....	180
6.1.6.1 Family resources contribute to family coping patterns.....	181
6.1.6.2 Situational and family schema appraisals and meaning-making contribute to family coping patterns.....	182
6.2.7 The family adaptation process: Bonadaptation and maladaptation.	184
6.3 Summary and conclusion	184
CHAPTER 7: Summary and conclusions: Key research findings, implications, strengths, limitations and future directions	187
7.1 Key findings that apply to research aims and questions	188
7.2 The lived experience of parenting a child with CF in the context of early surveillance: Implications for policy and practice.....	188
7.2.1 Policy and practice implications.....	189
7.3 Parents' constructions and attributions of meaning to early surveillance.....	193
7.4 Coping with parenting children with CF in the context of early surveillance: Implications for theory, policy and practice	195
7.4.1 Theoretical implications.....	196
7.4.2 Policy and practice implications.....	199
7.5 Further considerations: Future program development and implementation, and mental health care of parents and implications for their families.....	202
7.6 Strengths and significance of the research.....	204
7.7 Limitations of my research.....	205
7.8 Directions for future research.....	206
7.9 Final considerations	208
7.10 Postscript reflection.....	210
References	212
Appendices.....	279

List of Tables

Table 1 Group descriptions and recruitment details

Table 2 Demographic information of AREST CF parents by CF clinic location and stratified child age

Table 3 Stages of thematic analysis for qualitative psychological research

Table 4 Major themes and related sub-themes

List of Figures

- Figure 1 The Resiliency Model of Family Stress, Adjustment and Adaptation
- Figure 2 Map of Australia showing the regions where AREST CF operates
- Figure 3 A conceptualisation of data management and data analysis processes
- Figure 4 Illustration outlining data collection process
- Figure 5 Overview of theoretical and procedural frameworks guiding the study
- Figure 6 Representation of how parents psychologically reach an alternate sense of normal family life
- Figure 7 How parental psychological constructs are associated with home treatment and parenting outcomes
- Figure 8 Relationships between knowledge, assurance and control
- Figure 9 How early surveillance influences perceived parenting competence and possibly self-esteem
- Figure 10 How hope is constructed by parents, and the objects of hope
- Figure 11 Adaptation phase of The Resiliency Model of Family Stress, Adjustment and Adaptation
- Figure 12 Research findings applied to The Resiliency Model of Family Stress, Adjustment and Adaptation
- Figure 13 Screening and treatment of depression and anxiety: Algorithm for parents/caregivers
-

List of Appendices

Appendix A	Invitational recruitment letter
Appendix B	Information statement and consent form – Princess Margaret Hospital
Appendix C	Information statement and consent form – Royal Children’s Hospital
Appendix D	Western Australian counselling services
Appendix E	Victorian counselling services
Appendix F	Personal reflections
Appendix G	Interview schedule
Appendix H	Initial coding of interview transcripts
Appendix I	Thematic mind-map - Trust
Appendix J	Thematic mind-map – Cognitive dissonance

CHAPTER 1

Introduction

This chapter introduces the reader to the wider context of this research by outlining the topic of parenting a child with a chronic condition, the associated experiences and the outcomes for parents. How the parenting experience has been constructed theoretically and empirically is then briefly reviewed. Next is the introduction of paediatric cystic fibrosis and current treatment endeavours, which contextualises the purpose of the research. These discussions lead to an outline of the rationale of the research. Thus, the necessity for exploring the lived experience of parenting children with CF in context of a modern and aggressive treatment regimen is presented in conjunction with the importance of exploring parenting in context. Specifically, the two participant groups are described and the research questions detailed. The chapter concludes by discussing implications of the research before outlining the structure of the thesis.

Making the decision to have a child is momentous. It is to decide forever to have your heart go walking around outside your body...Elizabeth Stone

1.1 Research context

1.1.1 The psychology of parenting in paediatric chronic disease.

Long-term conditions and chronic diseases are more commonly associated with older people and less with children. However, a significant number of children live with chronic conditions (Australian Institute of Health & Welfare, 2005). These conditions cause stress for children and their families, and demand substantial amounts of time, energy and personal resources to cope with the situation (Jessop & Stein 1989). Many different types of chronic diseases and conditions exist in the paediatric population; some are present at birth and others may develop at a later stage during infancy or childhood. While some children with chronic diseases or conditions may recover, most will not lead normal lives and will need special care or management (Jessop & Stein 1989). However, with appropriate management, many children can function well and live healthy lives (Australian Institute of Health & Welfare, 2005).

In Australia, there is a lack of an agreed general definition of what constitutes chronic disease or illness (Australian Institute of Health & Welfare, 2005). One definition from the United States Center for Disease Control states that “a chronic disease is one that, in general terms, has a prolonged course, that does not resolve spontaneously, and for which a complete cure is rarely achieved” (National Public Health Partnership, pp.3). Whilst the terms *chronic disease*, *illness*, and *condition* are used interchangeably in the literature, using the term *condition* in my thesis draws the reader away from the biomedical aspects of disease and away from the negative connotations of illness. Therefore, where appropriate the term *chronic condition* will be used in this thesis to refer to a chronic disease or illness as defined by the United States Center for Disease Control.

In Australia, a high proportion of children are diagnosed with chronic conditions each year. According to the Australian Bureau of Statistics National Health Surveys, 41% of children aged less than 15 years had a long-term health condition in 2007-2008, whilst an estimated 37% had at least one long-term condition in 2011-2012. Chronic conditions in Australian children are of primary concern for the healthcare system and community, and families alike.

The Australian healthcare system is a major advocate for children’s health and well-being with a number of national institutes caring for children with, and researching, paediatric diseases and conditions such as the Children’s Medical Research Institute, Children’s Cancer Institute Australia and The Australian Institute of Family Studies. The National Health and Medical Research Centre and The Australian Institute of Health and Welfare are also strong advocates for children’s health. They provide funding and guidance on a number of child-related health issues. The Telethon Kids Institute is a local institute with a specific focus on chronic conditions in childhood. The work of

these Institutes, and the work of others nationally and internationally, has increased children's life expectancies for various chronic conditions exponentially over the past two to three decades. For example, advances in medicine and technology have increased life expectancy of many paediatric conditions including asthma, leukaemia, sickle cell disease and cystic fibrosis (CF) (Perrin, Bloom & Gortmaker, 2007). Such advances have caused a shift from short-term management of acute and often fatal conditions to long-term management of survivable chronic conditions. Despite improvement in life expectancy, paediatric chronic conditions can impose significant burden on children, their parents, their families, and the public health system.

Integration of technology and science creating medical advancements has changed the nature of many, once fatal, diseases to chronic conditions, insofar as a child who has a chronic condition or disease in the modern era is expected to live a relatively ordinary life compared to 20 to 50 years ago. These changes in child health outcomes have significant and far-reaching impacts on parental and family psychological adaptation to paediatric chronic conditions. Medical and technological advancements have meant that more than ever, parents are finding themselves providing all or most of the care for their chronically ill children. Parents are now being medicalised (Valentine, 2010) in their efforts to keep their children healthy, by following treatment regimens and obtaining knowledge of their child's condition. Thus, in treatment of paediatric chronic conditions, parents and family become focal points, as children are living longer and requiring extensive treatment regimens to control symptoms. For these parents, their practices and their role can be significantly different from parents who have healthy children.

Improvements in diagnosis and medical treatment have meant that children are being diagnosed sooner, and as these improvements continue, their care is moving from the hospital environment into the home and community. Though paediatric healthcare is improving, there is an increased responsibility and burden on parents as primary carers of chronically ill children, which can affect them in multiple ways. The first section of this chapter outlines what is currently known about parenting children with chronic conditions, paying particular attention to theoretical understandings, research trends and empirical outcomes.

Becoming a parent is often said to be one of the most life-changing experiences that encompasses both stress and joy. The expectation is one of joy, stature and fulfilment whilst the reality includes challenge, hardships and questioning one's self as parent. Becoming a parent is considered the most significant life-cycle transition in existence, and it is the most romanticised of all the life transition stages (McGoldrick, Carter & Petkov 2011). Parenthood is common, with 299, 697 births registered in Australia in 2014 (Australian Bureau of Statistics, 2014). After the birth of an infant it is the task and goal of parents to raise, protect and support the child through their early years of life.

The transition to parenthood can be monumentally challenging when a baby is diagnosed with a chronic life threatening condition. All parents hold expectations, hopes and dreams for their child, but when a child is diagnosed with a chronic condition these aspirations are forever altered. Learning of a child's chronic condition is generally a devastating experience that usually sets in motion a myriad of negative emotional responses. Whilst symptoms between conditions vary, common factors have been identified in how parents react to their child's diagnosis. Guilt, anger, grief, shock and disbelief are common initial parental emotions to a child's diagnosis (Fallowfield & Jenkins, 2004; Goldbeck, 2006; Kars, Duijnste, Pool, van Delden & Grypdonck, 2008; Moola, 2012; Nguyen, Pertini & Kettler, 2013; Samson, et al., 2009; Willingham-Piersol, Johnson, Wetsel, Holtzer & Walker, 2008). Most research into diagnosis of paediatric chronic conditions has focussed on the mother because she has been identified as the one who generally carries the burden of care. Research in this area has shown that exhaustion and frustration, loss of freedom and support networks, and loss of ideal mother-child relationship are the main outcomes for mothers of a chronically ill child (Almasri, et al., 2011; Bruce, Lilja & Sundin, 2016; Coffee, 2006; Esdaile & Greenwood, 2003; Johnson, 2000; Lloyd & Rosman, 2005; Pelchat, Lefebvre & Perreault, 2003; Shortman, et al., 2013; Wiedebusch, Pollmann, Siegmund & Muthny, 2008).

Early conceptualisations of parenting children with chronic conditions presumed that children and their families would experience adverse psychological effects, referred to as psychopathology, which in turn, was predictably found. For example, attachment and parenting style issues (Eiser, 1993; Green and Solit, 1964; Marvin & Pianta, 1996, Venters, 1981), and depression and chronic sorrow (Bowes, Lowes, Warner & Gregory, 2009; Cashin, Small & Solberg, 2008; Johnson, 2000; Masterton, 2010; Swallow & Jacoby, 2001; Whittingham, Wee, Sanders & Boyd, 2013) have been commonly reported parental outcomes. Treatment and management of chronic conditions, and its associated burdens has been a primary focus for clinicians and researchers alike, with reports of anxiety, parenting stress and perceived parental incompetency in completing required treatment regimens (Anthony, Bromberg, Gil & Schanberg, 2011; Barakat, Patterson, Daniel & Dampier, 2008; Hall & Graff, 2011; Hassall, Rose & McDonald, 2005; Litzelman, Catrine, Gangnon & Witt, 2011; Powers, et al., 2002; Shatla, El said Sayyah, Azzam & Elsayed, 2011; Waisbren, Wee, Sanders & Boyd, 2004). These outcomes reflect the deficit-based models popular in the previous decades of understanding parents, children and their families experiencing chronic disease.

Developmental pathway models have further conceptualised trajectories that result in adaptive and maladaptive development. For example, McCubbin and McCubbin (1993) developed The Resiliency Model of Family Stress, Adjustment and Adaptation that posited how families might respond to a chronic condition within the family unit. Such developmental pathway models have identified risk and resistance factors in managing paediatric chronic conditions, and have evolved

over time to direct what is used as a basis for intervention and promotion. Mechanisms that set maladaptive trajectories have been categorised as risk factors, which are those elements within the family's environment, both inherent and indirect, that decrease likelihood of positive adaptation, whereas resistance factors increase its likelihood (Gerhardt, Walders, Rosenthal & Drotar, 2004). Researchers and clinicians alike have spent decades identifying these factors, their relationships with other mechanisms and intervening where appropriate to obtain optimal adaptation to paediatric chronic conditions.

Recent years have seen a tremendous growth and reconceptualisation in approaches to understanding the impact of chronic disease on children and their families. The traditional deficit-centred model has slowly been replaced with those models that take account of coping resources and individual competence. The emphasis has shifted from identifying psychopathology within children and their families towards an understanding of how ordinary children and their families deal with specific crises. Increasingly, models are being drawn from mainstream psychology, with the result that children and their families dealing with chronic conditions are not seen as psychopathologic, but as ordinary people in exceptional circumstances (Eiser, 1990). For example, research has shifted from identifying and intervening in maladjustment and psychopathology toward identifying individual and family coping strategies (Varni & Wallander, 1988).

Research has shifted focus from psychopathologies of caring for a chronically ill child such as depression and chronic anxiety to beneficial outcomes such as normalisation and resilience. For example, a salutogenic understanding of existence focuses on factors that support health and well-being, rather than on factors that contribute to adverse outcomes (Antonovsky, 1987). The aim of such research is to anticipate and prevent dysfunction within families before its manifestation, by strengthening parents and their families through providing education and support. Adaptive behaviours are daily living skills that assist individuals and families to become productive within society (Sparrow, 2011). Some resistance factors found to support adaptive functioning include family support and resources, and normalisation of the illness and its management into the family system. Support or resources that are currently used by the family, and additional supports accessible to the family when a crisis occurs, have been reported as protective factors (McCubbin & Patterson, 1983). Similarly, normalisation of a child's condition and its treatment regimen into family life can function as a resistance factor against practical, psychological and emotional burdens associated with the condition (Hopkins & Gallo, 2012; Knafel, Darney, Gallo & Angst, 2010; Ray, 2002). Accordingly, by its very nature, normalisation of paediatric chronic conditions into the family context may buffer and protect its members against their existence as well as against future burden, both normative and condition-specific.

Coinciding with a shift from deficit-based models to salutogenic approaches (Antonovsky, 1987) of research and clinical care, is increasing acknowledgement of the relationships between an individual's psychosocial environment and their health and well-being. Importance of family context for child health is emphasised by a biopsychosocial understanding of health, which recognises that the psychosocial environment of the child is a critical factor in the health of a child with a chronic condition (Engel, 1977; 1980). The family is the most important influence within the psychosocial environment of the child (Bronfenbrenner, 1986). Increasing both life expectancy and physical health outcomes are the primary foci of biomedical innovation, research and clinical care. However the biopsychosocial model of care (Engel, 1977; 1980) highlights importance of a holistic approach to an individual's care, inclusive of psychosocial and physical health and well-being of the child and their family.

Engel (1977; 1980) claimed that not only biological factors are important in individual functioning in the context of disease or illness. He claimed that psychological and social factors are also important considerations, both in terms of impact and outcomes of disease or illness. Engel's biopsychosocial model of disease and illness echoes a social-ecological perspective (Bronfenbrenner, 1979; 1986) to approaching human functioning in the context of disease or illness through its emphasis on interactions between biological, psychological and social factors. This contrasts the more traditional biomedical model of medicine that suggests that every disease process can be explained in terms of an underlying biological deviation from normal functioning (Stedman, 2011).

Since Engel's (1977; 1980) assertion that not only biological factors should be considered in care of sick individuals, but that psychosocial factors may impact on the biology of an illness, social-cognitive models of health behaviour have been formulated and tested. With the model's social-cognitive underpinnings, acceptance of such models varies across cultures (Santrock, 2007). Recognition of the biopsychosocial model in clinical care is evident in the United States, the United Kingdom, Australia and some European countries where integration of professional health services aims to provide care and address a patient's needs on all three levels (Gatchel & Oordt, 2003).

Over the past 20 years, qualitative studies have conceptualised family response to paediatric chronic conditions, and quantitative studies have explained the nature of family response to illness or identified individual-level variables that explained a family's response to illness. Descriptions of families' responses to illness include family life in the context of illness, impact of illness on the family system, family transitions related to illness and development of family concepts in context of the illness (Rolland, 1984, 1987). Explanations of responses to illness within families can be predicted by such factors as cohesion and conflict. For example, the responses of individual family members to

illness and interventions influence family functioning as a whole, which in turn can inform or modify functioning of each individual (Walsh, 2003).

The shift to a family focus of child health has encompassed embracing the fathers' roles in their child's care. This shift coincided with modern arrangements of family systems and child care responsibilities, resulting in fathers becoming more involved in their child's care. Whereas once, identification of mother as primary caregiver meant that aims and principles focussed on her psychological and social well-being, and her relationship with the child and other family members. The dominant or defining structure of paternal parenting has shifted in succession from an emphasis on moral guidance, to a focus on breadwinning, then to sex-role modelling, marital support, and finally, nurturance (Lamb, 2000). Traditional parental roles were gaining popularity, prompting a restricted focus on paternal nurturance and involvement in day-to-day child care, with little, if any, attention paid to the other functions or aspects of fatherhood. Consequently, academia has only recently begun to seek a broader and more inclusive understanding of fatherhood. These efforts have permitted more insightful research on the motivations and behaviour of contemporary fathers, as well as on the effects of variations in paternal behaviour on child health and development (Lamb, 2000).

Though fathers' roles and experiences are under-represented in the literature, the need to consider fathers, along with their challenges and benefits in caring for a chronically ill child are increasingly recognised (Phares, Lopez, Fields, Kamboukos & Duhig, 2005; Tiedje & Darling-Fisher, 2003). Like mothers, fathers can experience stress (Calzada, Eyberg, Rich & Querido, 2004; Weiner, Vasquez & Battels, 2001) and post-traumatic stress syndrome (Ribi, Vollrath, Sennhauser, Gnehm & Landolt, 2007). Fathers can experience difficulty discussing and expressing their family situation (Goble, 2004), which can be compounded by their own expectations that they should be strong and silent. The interaction between stress and gender can challenge the fathers' ability to cope (Arockiasamy, Holsti & Albersheim, 2008), and by remaining strong and silent, fathers are less likely to receive support (Clark & Miles, 1999). This perception of needing to portray strength within the family unit may also impede the fathers' involvement through desire to maintain control whilst at the same time feeling a loss of control (Clark & Miles, 1999; Hovey, 2005). In fact fathers can, for the most part, feel like a forgotten parent because they are often not involved in doctor's appointments or the child's treatment regimen (Sterken, 1996). Lastly, fathers' can sense incompetency in their child's care because of their perceptions of the mother as expert in care for the ill child, which can result in low self-esteem (Katz & Krulilk, 1999).

Similar to the shift from an exclusive focus on mothers as important people in the care of chronically ill children, there was a shift from an exclusive biomedical to a biopsychosocial view of care (Engel, 1977; 1980). A concurrent shift from a psychopathologic, deficit-based to a salutogenic

(Antonovsky, 1987), strengths-based perspective of care resulted in recognition of family resilience as an important buffer against future burden of a child's condition, as well against other life stressors (Benzies & Mychasiuk, 2009; Black & Lobo, 2008; Patterson, 2002; Walsh, 2003). The shift to a salutogenic approach, along with increases in life expectancy has shed new light on parenting a child with a chronic condition. These insights give hope for parents, children and their families that have not previously existed.

When a child is diagnosed with a chronic condition and life expectations are irrevocably altered, hope can take its place so that an alternative reality can become positively reframed. Hope in the context of parenting a child with a chronic condition has been referred to as a transitional refocusing from a difficult present to a positive future with dynamic possibilities within continual uncertainty (Duggleby, et al., 2010). Hope has been reported by parents of children who have various chronic conditions. Parents have hope for their child's; quality and quantity of life (Lloyd & Hastings, 2009), employment, family and other normative life transitions (Samson, et al., 2009) and they even hope for a cure to the condition for which their child suffers (Wong & Heriot, 2008). Hope can contribute to resilience for future burden of physical and mental chronic conditions (Faso, Neal-Beevers, & Carlson, 2013; Horton & Wallander, 2001; Kashdan, et al., 2002; Lloyd & Hastings, 2009). Therefore, the salutogenic (Antonovsky, 1987), along with the biopsychosocial (Engel, 1977; 1980) perspectives support the notion that hope within the family context is a highly regarded parental trait. The biopsychosocial (Engel, 1977; 1980), along with the social-ecological (Bronfenbrenner, 1979; 1986), perspective on caring for children who have chronic conditions emphasises the importance of parent and family practices and outcomes for child health outcomes.

A child's experience with their immediate environment provides the building blocks for their growth and development. Neurobiological effects of external psychosocial factors on child development, that is, psychosocial factors within the environment of the child have been shown to affect plasticity of the brain during windows of opportunity. For example, synapse formation and pruning during the early years of life are influenced by the child's environment. This has been demonstrated whereby environmental stimulation can positively and negatively affect brain development in the early years of life through effects on neurogenesis, synaptogenesis, long-term potentiation and pruning. These findings have been demonstrated at the cellular level, in vitro and in humans (Jontes, Buchanan & Smith, 2000; Mirescu, Peters & Gould, 2004; Stanwood & Levitt, 2004; Van Praag, Christie, Sejnowski & Gage, 1999). For this reason, early childhood experiences affect gene expression and therefore affect developmental and long-term health outcomes. Accordingly, the first 1000 days of a child's life, starting from conception to their age of 2 years, are important for all domains of development (Hoddinott, et al., 2013).

Social-ecology theory posits a relationship between the developing child and the settings and contexts in which the child is actively involved. Bronfenbrenner (1979) proposed a model of bi-directional influences on the child within their environment as occurring within nested settings beginning with the child, *microsystem*, and extending out to the immediate social settings of home, school and neighbourhood, *mesosystem*, and settings that do not involve the child as an active participant such as the parents' workplace, the *exosystem*, and the wider society and culture, *macrosystem*. A substantial body of literature supports utilisation of the model for families of young children, suggesting that indirect influences on child development can be traced back to parental characteristics (e.g., Armstrong, Birnie-Lefcovitch & Ungar, 2005; Brophy-Herb, Lee, Nievar & Stollak, 2007; DePanfilis & Dubowitz, 2005; Dishion & Stormshak, 2007). Consequently, based on understandings of social-ecological theories, a major line of inquiry has been to determine if and how parenting behaviours and parental characteristics influence child development.

Using this framework, parenting has been well-established as an important influence on child development, and much research has linked parenting with both beneficial and adverse child outcomes (e.g., Baumrind, Larzelere, & Owens, 2010). Research has shown parenting behaviours and parental characteristics can influence academic achievement (Cheung & Pomerantz, 2011; Englund, Luckner, Whaley, & Egeland, 2004, Pomerantz, Moorman, & Litwack, 2007), cognitive development (Ryan, Martin, & Brooks-Gunn, 2006), socio-emotional development (Bernier, Carlson, & Whippe, 2010; Dallaire, et al., 2006; El Nokali, Bachman, & Votruba-Drzal, 2010), and school dropout rates (Marcus & Sanders-Reio, 2001). Research has shown parenting can influence cognitive development (Ryan, et al., 2006), and socio-emotional development (Bernier, et al., 2010; Dallaire, et al., 2006; El Nokali, et al., 2010) in infants and young children. Moreover, family psychosocial functioning, including parenting, is well known to impact normal child development (Beauchamp & Anderson, 2010).

Based on social-ecology principles, parents play an integral role in their child's ability to adapt to living with a chronic condition, not only in terms of their child's developmental functioning but also in their ability to participate in daily activities. Principally, parent psychological distress has been identified as a risk factor for adverse outcomes in children with a variety of chronic conditions including CF (Cappelli, McGrath, MacDonald, Boland & Katsanis, 1988), cancer (Robinson, Gerhardt, Vannatta & Noll, 2007), type 1 diabetes (Whittemore, Jaser, Chao, Jang & Grey, 2012), spina bifida (Freidman, Holmbeck, Jandasek, Kukerman & Abad, 2004) and chronic pain (Palermo, Putnam, Armstrong & Daily, 2007). Therefore, parents have significant potential to beneficially or adversely affect their child's adaptation to their chronic condition (Law, Fisher, Fales, Noel & Eccleston, 2014). A child's adaptation to their condition then has both direct and indirect implications for their own

development. For example, direct implications include psychological and emotional well-being whereas indirect implications include treatment adherence.

Parenting is now well-established as an important influence on child development with extensive research linking parenting with beneficial and adverse child mental health outcomes. To that end, understanding parenting; its process, functions and outcomes, and parental characteristics and their outcomes are not only important for parents' general health and well-being per se, but it is now well-known that parents matter when it comes to their children's development and long-term health outcomes. Therefore, parenting research and intervention in the early years of a child's life will most likely beneficially impact upon the entire family system.

1.1.2 Paediatric Cystic Fibrosis in Australia

Cystic fibrosis (CF) is a complex, chronic disease for which there is currently no cure. However, life expectancy has exponentially increased over the past few decades, changing CF from a fatal childhood digestive and lung disease to a complex multi-systemic disease extending into adulthood (Davies, Alton & Bush, 2007). Advances in science, medicine and technology have resulted in life expectancy of CF patients increasing from 5 to 6 years in 1960 to a median survival age of approximately 40 years now (UK Cystic Fibrosis Registry, 2012). Improved outcomes for children born with CF are attributed to early detection and diagnosis through newborn screening (NBS) programs and improved methods of treatment and management (e.g., Farrell, et al., 2001; Farrell, et al., 2005).

Cystic fibrosis is an autosomal recessive disease most commonly affecting Caucasian people (Davies, et al., 2007; Montgomery & Howenstine, 2009) that primarily affects the gastro-intestinal system, respiratory system, and sweat glands (Davis, 2006; Rowntree & Harris, 2003). Approximately 70% of children with CF are diagnosed before the age of 1 year (Walters & Mehta, 2007), with clinical symptoms often present soon after birth (FitzSimmons, 1993). Data registry services in Europe, the United Kingdom, the United States and Australia collectively report over 10,000 children under the age of 7 years are living with CF (Cystic Fibrosis Australia, 2013; Cystic Fibrosis Foundation, 2014; European Cystic Fibrosis Society, 2014; UK Cystic Fibrosis Registry, 2013)

The generally overwhelming and demanding nature of CF on the mental health and well-being of parents likely explains why modern research still primarily focusses on psychopathology of children and their parents. A large epidemiological study currently being conducted across the United States, the United Kingdom, Europe and Australia is investigating stress, depression and anxiety for children with CF and their caregivers. Preliminary findings report that approximately a quarter of the caregivers sampled reported clinical levels of stress, depression and/or anxiety (Besier, et al., 2011; Driscoll, Montag-Leifling, Acton & Modi, 2009), an already well-known finding (Goldbeck, Quittner & Besier, 2008; Quittner, et al., 2008; Yilmaz, et al., 2008).

Two particular aspects of CF care are important in understanding how parents manage their child's CF treatment, as well as how they experience their child's condition. Firstly, treatment management primarily occurs within the home. Like some other chronic conditions that were once considered fatal (e.g., chronic kidney disease), parents whose children have CF are required to take primary responsibility for their child's treatment management, which is generally considered to be demanding and time-consuming. Therefore, treatment burden is reportedly common amongst parents, which can include parenting stress and parental role strain associated with treatment administration (Hodgkinson & Lester, 2002; Quittner, Opipari, Regoli, Jacobsen & Eigen, 1992; Quittner, et al., 1998). Sometimes treatment burden is a result of low levels of perceived competence with administration of therapies (Sawicki & Tiddens, 2012), and anxiety associated with treatment burden can occur because of increased awareness of disease trajectory (Sawicki, Hellar, Demars & Robinson, 2015). Secondly, parents find it difficult to obtain peer support from other parents experiencing the same situation because children with CF should not communicate face-to-face with each other due to issues of cross-infection. Peer support from parents whose children have the same condition is an important factor (Duffy, 2011; Eatough, et al., 2013; Kerr, Harrison, Medves & Tranmer, 2004; Shields, Young & McCann, 2008) and could be an essential component of parental adaptation to a paediatric chronic condition (Barcroft, 2015).

Traditionally in CF, the early years of life have been considered the silent years. This is because disease progression is usually asymptomatic and detection using traditional methods, such as lung function, is difficult as young children struggle to cooperate with the demands of the investigations. Parental reactions to, and experiences of, diagnosis have been thoroughly investigated (e.g., Baroni, Anderson & Mischler, 1997; De Monestrol, Brucefor, Sjoberg & Hjelte, 2011; Havermans, Tack, Vertommen, Proesmans & de Boeck, 2015; Quittner, DiGirolamo, Michel & Eigen, 1992). As too have parental responses and experiences of disease progression and treatment management later in a child's life (e.g., Dashiff, Suzuki-Crumly, Kracke, Britton & Moreland, 2013; Fidika, Herle, Herschbach and Goldbeck, 2015; Sawicki, et al., 2015), usually from approximately 10 years of age when the condition generally starts to become symptomatic. However the mental health and experiences of parents with young children who have CF diagnosed early by NBS has not generally been considered.

Following the challenge of diagnosis and responsibility of home treatment is the awareness that CF disease progression is occurring, often without symptoms and in children who look well. Due to medical and technological advancements, parents are receiving more detailed information about their child's condition and at a much earlier time than ever before. Due to the recentness of such advancements in treatment and detailed information about disease progression being generated, how parents respond to, and manage, such information remains largely unknown.

1.1.3 Current Australian treatment management practices for infants and young children in the 21st century

Much of the medical care for individuals living with CF in Australia is provided by specialist CF centres peer-reviewed by the national CF body – Cystic Fibrosis Australia (CFA). Australian national standards of CF care were developed in 2008 and promote standardised care across centres (Bell & Robinson, 2008). Cystic fibrosis centres provide a multidisciplinary allied health and medical team with facilities for relevant diagnostic testing and other subspecialist medical care (Bell 2008 Schechter & Gutierrez, 2010). A holistic approach to CF care is fundamental, and care is delivered by a multidisciplinary team of health professionals with expertise in CF (Madge & Khair, 2000). The main aims of CF care include preventing chronic infection, minimising progression of lung damage, sustaining normal nutrition and growth, maintaining independence for patients and their families, and maximising quality of life (Agent & Madge, 2007). Major CF centres provide care for local patients with CF, as well as those patients who live regionally via outreach programs (Littlewood, 2000).

The people who form a typical CF multidisciplinary team include lead clinical physician, and a specialist clinical nurse, physiotherapist, dietitian, psychologist, pharmacist, and social worker. Access to further multi-disciplinary team members including specialised laboratory services, gastroenterologist, diabetologist, and an ear, nose and throat specialist (Agent & Madge, 2007) is essential. The CF multi-disciplinary team has expert knowledge in CF and provide clinical, educational and support services for patients and their families (Agent & Madge, 2007). The specialist nurse has a unique role by being involved with the patient and their family over the lifespan: from helping them come to terms with the CF diagnosis to supporting them with end-of-life management (Agent & Madge, 2007). Currently, clinical guidelines and standards of care are generally limited to medical aspects of CF, with much less consideration of accompanying psychosocial aspects (Colombo & Littlewood, 2011; Tiddens, 2009). However, Borowitz, et al., (2009) recently developed Cystic Fibrosis Foundation evidence-based guidelines for management of infants with CF. Furthermore, a European and American collaborative consortium recently advocated for screening anxiety and depression in patients and their caregivers, making recommendations for international implementation (Abbott, et al., 2015; Quittner, et al., 2015).

As part of standard paediatric CF care in Australia, an annual review is completed for each child, which consists of a comprehensive assessment by the multidisciplinary team of all aspects of their condition and treatment (Agent & Madge, 2007). The aim of the annual review is to gain a thorough understanding of all aspects of a child's condition, identification of suboptimal treatment that can be improved, formalisation of an individual management plan; and provision of information

for centre and national databases (Agent & Madge, 2007). Important aims of the review are to inform parents of the progress of their child and provide a care plan for the next 12 months.

The annual review consists of a medical history review, assessment of CF by a specialist clinical nurse and specialist physiotherapist, nutritional assessment by a specialist dietitian, review by a psychologist and social worker, and full clinical examination (Agent & Madge, 2007). The annual review for children up to 7 years of age attending paediatric CF centres at Princess Margaret Hospital (PMH) and The Royal Children's Hospital (RCH), is unique in Australia and much of the world in that it includes pulmonary surveillance using the most detailed measures of lung disease currently available. These investigations provide information regarding the structural, microbiological and inflammatory health of the lungs. Research that is focused upon improving the physical and mental health outcomes of children with CF is embedded in the clinical program and is further supported by collaborations with teams in Chapel Hill, North Carolina, Indianapolis, Indiana, St Louis, Washington and Rotterdam, Netherlands.

As standard treatment for CF, parents must perform several hours of therapy a day in the home such as preparing a special diet, administering medications, completing airway clearance with physiotherapy, and performing nebulisation therapy so that aerosol particles can be breathed directly into the lungs (McCubbin, Bowers & Holaday, 1984). When a child with CF is in good health, treatment may take a few hours a day, but when they become ill, treatment increases substantially (Agent & Madge, 2007). Promotion of airway clearance of mucus, suppression of bacterial growth through prophylactic antibiotics and optimisation of nutritional status through pancreatic enzymes are the main pillars of CF management (Davies, et al., 2007; Montgomery & Howenstine, 2009). Due to the inability of the lungs to clear thick mucus secretions that contain a number of bacteria and other airborne particulates, airway clearance techniques are a standard part of CF care (Quittner, Barker, Marciel & Grimley, 2009). Chest physiotherapy loosens mucus in the lungs and encourages its removal through productive coughing. This process can take up to 1 hour and is typically performed twice a day (Quittner, et al., 2009).

Despite such an aggressive and proactive treatment regimen, fighting lung infection is a lifelong battle with the lungs eventually becoming chronically infected with bacteria, such as *Pseudomonas aeruginosa*. Treatment of lung infection requires oral and nebulised antibiotics in many patients: an additional 20 minutes of treatment daily (Collins, 2009). If deterioration of the patient's respiratory condition (that is, a pulmonary exacerbation) continues despite these treatments, intravenous antibiotics can be prescribed for a period of 10 to 14 days (Quittner, et al., 2009) for which a hospital admission is generally required.

The digestive system of people with CF is also affected, with most infants (85%) born with pancreatic insufficiency (Zielenski, 2000). This refers to the impediment of the pancreas to secrete

enzymes necessary to digest food resulting in its inability to effectively absorb food (Montgomery & Howenstine, 2009). This problem is addressed by consuming orally administered enzymes with every snack and meal. Malabsorption and increased resting energy expenditures are issues in CF, requiring patients to consume 100 to 200% of the recommended daily allowance of calories (Stallings, et al., 2008). Therefore, children can experience issues gaining weight if they do not maintain a high-fat diet (Ramsey, Farrell & Pencharz, 1992). Somatic growth and development of children with CF is a concern related to both the gastrointestinal and respiratory problems associated with CF.

1.1.4 Cystic fibrosis treatment management in infants and children from diagnosis to 7 years in Western Australia and Victoria: Australian innovation for paediatric CF management

The introduction of NBS to Australia over 20 years ago has resulted in early diagnosis and a paradigm shift from reactive treatment to proactive early-life disease surveillance and therapeutic intervention. The Australian Respiratory Early Surveillance Team for Cystic Fibrosis (AREST CF - www.arestcf.org) pioneered early detection and prevention of CF disease progression, and is a multi-disciplinary clinical and research collaboration between paediatric CF centres in Perth, Western Australia and Melbourne, Victoria who are dedicated to improvement of respiratory health and outcomes in children with CF. The program was initiated at Princess Margaret Hospital (PMH) in Perth in 1996 to investigate the role of inflammation in the lungs of children with CF. Since then, the program has grown into a comprehensive assessment of early life disease in CF. Bronchoalveolar lavage (BAL) was introduced in 1999, it measures inflammation and infection in the lung via a bronchoscopy. Infant and preschool lung function testing was introduced in 2003 and chest computed tomography (CT) scanning to define lung structure was introduced in 2005. All BALs and CTs for infants and young children are performed under general anaesthetic (GA). The Royal Children's Hospital (RCH) in Victoria joined the program in collaboration in 2005. The main objective of the AREST CF team is to improve the detection, prevention and treatment of early respiratory disease in young children with CF in order to improve clinical outcomes and quality of life for patients and their families.

Central to AREST CF is the early surveillance program: a unique clinical surveillance program that has the potential to modify long term outcomes for children with CF through early detection and interventions to delay the onset of lung disease, long before signs and symptoms are apparent. The program is aimed specifically at infants and preschool children from diagnosis (approximately 3 months) to 7 years who undergo annual, detailed surveillance procedures, which are relatively invasive compared to standard practices in Australia. These investigations generate detailed information about the progress of early lung inflammation, infection and structural disease that would otherwise remain largely unknown to the treating physicians and the families. Whilst early surveillance is currently unique to Perth and Melbourne, it is rapidly emerging as the likely

framework for future therapeutic intervention trials in young children nationally and internationally. Currently, the surveillance framework has been adopted by an international clinical trial (Australia, New Zealand, Netherlands), COMBAT-CF, which uses bronchoscopy and chest CT scans as outcome markers of CF disease progression. Since its inception, approximately 95% of Western Australia's and Victoria's populations of children with CF are enrolled in the program from diagnosis. Following is a detailed description of each of the procedures involved in the AREST CF early surveillance program.

Bronchoalveolar Lavage

Bronchoalveolar lavage is performed under the guidance of a bronchoscope. A bronchoscopy and BAL are performed in children who cannot yet cough up sputum. Older children can do this, which can then be tested for infections. Young children find this difficult to achieve, and as the children in the program are under the age of 7 years, this technique is appropriate. BALs consist of a GA administered and positioning of a disposable laryngeal mask airway (LMA) to facilitate passage for the bronchoscope (Stick, et al., 2009). The bronchoscope is passed via the LMA through the vocal cords into the trachea and down into the first and second main bronchi to reach the lobe where BAL is performed (Brennan, Gangell, Wainwright, & Sly, 2008). A bronchoscope is a fibre-optic tube with a camera at the end, which allows clinicians to investigate airways in the lungs. A BAL involves instilling saline into a small part of the lung and then removing it through suction. The returned fluid collects mucous located in the area of the lung where the fluid was inserted. The fluid is then assessed for bacteria or inflammation in the lungs. BAL is performed using normal saline warmed to body temperature (Brennan, et al., 2008). Both bronchoscopy and BAL are considered the gold standard for determining airway inflammation and lower respiratory tract infection (Ranganathan, 2006), and BAL is currently the only method for obtaining reliable data on lower respiratory tract infection in young children with CF (Brennan, et al., 2008).

Lung Function

Lung function assessment in young children undergoing early surveillance consists of a number of different techniques depending on the age of the child. Lung function can be assessed similarly in infants and preschool children but with slightly different techniques. There are three different lung function tests for infants up to 2 years of age. The tests take approximately 3 hours in total and are performed the week prior to the CT scan and BAL. As infants are unable to provide the cooperation required for typical lung function tests, specialised methods have been developed to assess lung function in this age group. That is, tests have been modified so that large, forced breaths are not required by infants, or the machines perform this technique for the infant. Infants are given a sedative, chloral hydrate, prior to commencement of the tests.

Multiple breath washout testing is designed to determine how evenly gas mixes in the lungs, and involves the infant breathing in gas that contains a known concentration of an inert tracer. The concentration of tracer gas in exhaled air is monitored during tidal breathing. The test provides an assessment of ventilation inhomogeneity and lung clearance index arising from mucus plugging of the small peripheral airways and airway wall thickening (Rezaee & Ren, 2006). Low-frequency forced oscillation is another technique that measures total respiratory impedance by measuring changes in pressure and flow in response to a low amplitude oscillatory pressure signal applied at the airway opening (Oostveen et al., 2003; Ranganathan, Linnane, Nolan, Gangell, & Hall, 2008).

The last test is the raised volume rapid thoraco-abdominal compression technique, which assists in determining obstruction of air flow in the lungs. Prior to starting the test procedure, the infant is wrapped in an inflatable jacket, and the infant's lungs are gently inflated with a pump. Once the lungs are filled to a set inflation pressure, the jacket is rapidly inflated, squeezing the thorax, which simulates a forced expiration (LeSouef, Turner, & Motoyama, 1996). Obstruction of airflow is measured by the amount of air that is expelled in the forced expiration.

Children over 2 years of age perform slightly different tests based on their ability to expel air from the lungs on demand, and are not sedated for lung function testing. Young children perform the multiple breath washout procedure and forced oscillation technique. However, this is not conducted under low frequency with young children and is performed using a slightly different technique. The child is required to breathe normally through a mouthpiece whilst wearing a nose clip. A low amplitude pressure oscillation is applied at the airway opening. This test is a measure of airway resistance and lung stiffness.

Young children 5 years of age and older also perform spirometry (forced expiration), which results in a measure of FEV¹ (a measure of airway obstruction). This test requires a maximal expiratory maneuver involving an inspiration to total lung capacity before a forceful exhalation to residual volume (Mayer & Allen, 2006). The test provides information about airway obstruction and is used as a standard measure of lung function in older children and adults (Kerem, Conway, Elborn & Heijerman, 2005).

Computed Chest Tomography

Lung function assessments are provided through CT. Chest scans provides imaging of the lung that shows the presence, severity and extent of lung disease. Previously, physicians have been limited to using chest x-rays, but advances in techniques now enable more comprehensive assessment of the lungs. Chest scanning is an imaging technique that is more detailed than chest x-rays in providing information on structural CF abnormalities, especially in young children. Chest scans are performed annually under GA. The CT scan is performed under sedation as precise volume control is required and infants and young children who aren't able to

cooperate (Kirks, 1998). The CT procedure has been modified to minimise exposure to ionising radiation, a common concern with imaging techniques. Currently there is international effort to develop a standardised low-dose CT protocol suitable for children with CF (Linnane, Robinson, Ranganathan, Stick, & Murray, 2008). Chest scans are considered the gold standard technique in diagnosing bronchiectasis (Chang & Redding, 2006), and has the ability to clearly determine structural components of CF lung disease (Oikonomou & Hansell, 2006; Sonnappa & Owens, 2007).

All BALs and chest CTs for those infants and young children under 7 years of age are performed under GA. Anaesthesia is induced with either sevoflurane or propofol, according to the clinical judgment of the anaesthetist (Stick, et al., 2009). Parents can stay with their infant or child for the duration of the lung function tests and whilst their infant or child is being anaesthetised. Parents must, however, leave theatre for the bronchoscopy and BAL procedures, and the CT scan due to ionising radiation. Parental participation during testing is not encouraged due to the complexity of the procedures. All procedures are generally scheduled as a day surgical case with the child discharged the same day (Brennan, et al., 2008).

A week in the life of parents involved in AREST CF during annual surveillance

Parents of infants up to 2 years of age

In the week preceding coming into the clinic for their infant's CT and BAL, parents bring their infant in for a multiple breath washout procedure, low frequency forced oscillation procedure and raised volume rapid thoraco-abdominal compression procedure. These procedures are performed on the same occasion and under sedation. With much variation within the amount of time it takes for the infant to fall asleep with sedation, for the tests to be completed and for the infant to fully recover from sedation, the average time parents are required to relinquish their infant is approximately three hours.

Parents of children aged 2-7 years

Children aged 2 years and older undergoing early surveillance perform forced oscillation technique and spirometry, as well as CT and BAL conducted under GA. This requires a full day of testing for children, and waiting for parents. The surveillance procedures for all children within the program are performed in addition to 3-monthly clinic reviews, which include height and weight measurements, social worker, physiotherapist, dietician and physician appointments.

In summary, this introduction has focused on the research context, specifically: CF in young children and treatment required in maintaining their optimal health; what parents experience and how they cope with their child's chronic condition; and what the AREST CF early surveillance program offers children with CF and their parents, along with the processes and procedures involved. How these contextual factors have informed the framework, foundation and purpose of this research has also been discussed. The following section provides a summary of the research

rationale and outlines specific aims and questions of my research. The last section concludes by highlighting the contribution of my research to contemporary understandings of children's and families' involvement in early surveillance for covert (i.e., undetected by traditional measures) CF disease progression, in particular, by highlighting the significance of the research for policy, practice, research and theory.

1.2 The current research

1.2.1 Research rationale

Despite the breadth of debate, as well as decades of clinical and theoretical awareness of the psychological needs of parents who have a child with a chronic condition, no research was found that specifically investigated the experience of parenting children with CF in the context of early surveillance for CF lung disease. The necessity to address this paucity in research was emphasised by theoretical understanding of the parenting experience that has been developed without consideration of processes and outcomes of parenting in the context of early surveillance for paediatric chronic disease. Given the current clinical and political agendas for early intervention (Jenkins, 2005; Wise, da Silva, Webster & Sanson, 2006), there has been a need for research into parental experiences of early intervention and aggressive treatments. Further emphasised by policy and program development and implementation is the need to understand parents' experiences and how they cope with early intervention and aggressive treatment for their children.

Further is the increasing recognition that if research is to become meaningful in public health systems and the provision of health care services, it must be translational (Oborn, Barrett & Racko, 2010; Ogilvie, Craig, Griffin, Macintyre & Wareham, 2009). Although no single definition of translational research has been agreed upon, it has been described as a necessity to translate knowledge created in the research setting into real-world applications that aim to improve health and well-being of individuals through provision of more effective health care systems and services (Canadian Institutes of Health Research, 2004). A fundamental rationale underpinning the view on translational research by the Canadian Institutes of Health Research (CIHR) is a sense of moral and social responsibility to ensure that research progresses such that individuals who funded and enable health research in the first place benefit from its conduct. Therefore, for research that is fundamentally explorative to become translational, it must consider aspects of human experience unreachable by positivist methods so that humanistic phenomena uncovered by the research can be addressed.

Parents of children diagnosed with CF via NBS to 7 years of age and currently under surveillance for lung disease, were targeted as research participants. The purpose of my research is to explore how parents of children undergoing early surveillance for CF lung disease construct their lived experiences of parenting, and how they attribute meaning to these experiences, drawing

attention to how they cope. A further aim of my research is to contribute to developing a modern support system. .

Accordingly, to support parents in the AREST CF program, it is important that current understandings of their experience be reviewed with reference to the lived experience of paediatric chronic disease. By asking parents not only about how they construct their experiences of parenting a child with a chronic condition but also examining how they attribute meaning to their experiences is required so that more meaningful analysis of theory, policy and clinical practice effectiveness can occur. My research aims to achieve this by exploring constructions of experiences and attributions of meanings from the perspectives of those parents involved in the AREST CF program. Information about the lived experience of parenting was explored with interview data from both sites where AREST CF operates in Australia. The sample was stratified into two groups by child age. The relevance of sampling across both sites and stratifying by child age was such that diverse experiences could be interrogated, and changes that parents experienced over time could be identified. The next section outlines what is currently known about parenting children with chronic conditions who undergo early surveillance for disease progression.

1.2.2 Exploring the lived experience of parenting children with CF in the context of early surveillance

With the successive, yet sporadic, implementation of NBS throughout the world there is now considerable research that addresses psychosocial effects of NBS and diagnosis. However, research characterising the psychological effects of early CF disease surveillance (particularly programs that use relatively invasive modalities for disease detection) following NBS does not exist, nor does literature exist on how children with CF, their parents and families experience, or cope with, early disease surveillance like that operated in the Perth and Melbourne paediatric CF centres. Therefore, little is known about what parents experience and how they cope with a previously untested program of management in young children with CF and what factors are associated with adaptive and maladaptive coping. Moreover, it is not yet clear if there are psychological sequelae of early surveillance and early (often covert) disease detection.

Anecdotal experiences and assumptions of healthcare professionals (HCPs) working within the program are that parents seem to adapt to the program over time, although clearly some parents experience, and cope with, the program better than others. Professionals conducting early disease surveillance programs following NBS diagnosis need to; understand whether or not families perceive the program to be adversely affecting their lives; and identify psychological characteristics within the family that may be associated with adjustment and maladjustment or other positive and negative experiences. Consequently, care pathways for improved mental health and well-being for families involved in early surveillance are incomplete. Therefore my research should provide

empirical support for anecdotal experiences and assumptions of HCPs working with families and may provide some understanding of psychological and emotional processes and outcomes for parents whose children undergo early surveillance for CF lung disease, which could guide future clinical interventions and facilitate further psychological research.

1.2.2.1 Parallels drawn from medical treatment procedures.

As there is no research assessing parental psychological effects of the AREST CF program, it is logical to consider research assessing similar parental experiences found within the program, as this will provide the closest idea of what parents may experience based on similarities, for example, in medical procedures. Therefore, anticipated experiences can be drawn from the literature in other diseases that require invasive procedures as part of care. These experiences include anxiety (Bauchner, Vinci, Bak, Pearson, & Corwin, 1996; Besier, et al., 2011; Chundamala, Wright, & Kemp, 2009; Menahem, Poulakis, & Prior, 2008), stress (Da Silva, Jacob, & Nascimento, 2010; Hopia, Tomlinson, Paavilainen, & Åstedt-Kurki, 2005; Hutchinson, Willard, Hardy, & Bonner, 2009), need for control over their child's care during hospitalisation (Berrios-Rivera, Rivero-Vergne, & Romero, 2008; Brosig, Mussatto, Kuhn, & Tweddell, 2007; Hallstrom & Runeson, 2001), and role strain (Quittner, et al., 1998; Quittner, et al., 1992). Repeated exposure to these experiences in the absence of supports may reflect adversely on the well-being of the family and crucially affect the child (Patterson, Budd, Goetz, & Warwick, 1993; Szyndler, Towns, Van Asperen, & McKay, 2005; Turner-Cobb & Steptoe, 1998).

As part of the early surveillance review, each child receives a GA prior to bronchoscopy and other procedures carried out in operating theatres as part of annual review, and this therefore may be regarded as a peri-operative type experience. Whilst there is a plethora of research on peri-operative experiences of children (e.g., Beringer, Segar, Pearson, Greamspet & Kilpatrick, 2014; Chieng, Chan, Klainin-Yobas & He, 2014; Fortier & Kain, 2015; Lopez, et al., 2007), much less research has been conducted on peri-operative experiences of parents whose children undergo anaesthesia.

Anxiety has been the most reported peri-operative emotional response for children, reaching a prevalence of up to 60% (Kain, Caldwell-Andrews & Wang, 2002; Thompson, 1994; Wollin et al, 2004). However, such figures do not exist for parents whose children undergo operative procedures. Several randomised controlled studies assessing anxiety of parents have compared parents who are present during the child's anaesthesia induction, compared to parents who are absent from the procedure. Kain and colleagues (2000) and Bauchner and colleagues (1996) both found that parents who were present for the child's anaesthesia were less anxious than those who did not attend. In contrast, Akinci, Kose, Ocal and Aypar (2008) found no differences between parents who were present at their child's anaesthetic induction compared to parents who were not present. Therefore, debate continues as to the effectiveness of parent presence for both children

and parents. What is not yet clear is the experience of children and parents undergoing the annual early surveillance procedures: the anaesthetic, bronchoscopy and post-anaesthetic recovery period; and the associated psychological burden on parents' ability to cope specifically with early surveillance procedures, but also as a potential additional stressor to general CF care.

1.2.2.2 Parallels drawn from similar condition trajectories.

Research into parental experiences of life-threatening illnesses with similar treatment management as CF may provide insight into how parents whose children undergo repeated early disease surveillance might experience and cope with associated events. Caution should, however, be exercised when generalising the experiences of parents of children with other chronic conditions exposed to a variety of medical procedures that differ to those experienced by children undergoing early surveillance for CF lung disease.

Cardiac

Cardiac conditions are often congenital and entail invasive procedures or surgical correction in early life and may be life threatening or life shortening. These features parallel CF, and the early surveillance program, to some extent. Considerable research of this condition has investigated parental experiences and coping whilst their child is awaiting cardiac surgery. Similar to research on parental experiences of GA, it has been reported that anxiety and emotional distress are elevated prior to surgery, and that these emotions return to normative levels post-surgery (Brosig, et al., 2007; Menahem, et al., 2008). To further understand experiences of parents whose children undergo surgery, a qualitative study explored parental experiences and coping resources and found that parents experienced a fear of their child dying that resulted in feelings of helplessness and powerlessness (Salgado et al., 2011).

Cancer

Improvements in CF prognosis and treatments have followed a similar path to that of certain paediatric cancers, such as leukaemia. It is possible to draw some similarities across surveillance and treatment processes as well as parental psychological outcomes and coping skills from this literature. Research has trended towards investigating resiliency factors in families, reporting adaptability (Rosenberg, et al., 2014; Greeff, Vansteenwegen & Geldhof, 2014), communication (Beek, Schappin, Gooskens, Huisman & Jongmans, 2014; Clarke, Sheppard & Eiser, 2008) and family support (Nicholas, et al., 2009; Shortman, et al., 2013) as important components of a family's resiliency to a paediatric cancer diagnosis. Family resilience after a paediatric cancer diagnosis is a recent construct receiving wide-spread interest (e.g., Van Schoors, Caes, Verhofstadt, Goubert & Alderfer, 2015). However, understandably, most research into parental experiences of pediatric cancer treatments has focused on negative psychological consequences, with depression, stress, anxiety and post-traumatic stress syndrome (PTSS) and their relationships to adaptation receiving

the most attention (e.g., Greening & Stoppelbein, 2007; Hoekstra-Weebers, Jaspers, Kamps, & Klip, 2001; Kazak, Boeving, Alderfer, Hwang, & Reilly, 2005; Von Essen, Sjoden, & Mattsson, 2004).

The literature examining parental outcomes following their child's diagnosis of paediatric cancer suggests mothers are more negatively affected by their child's cancer diagnosis and subsequent treatment than fathers on several psychological outcomes. Mothers demonstrate more depressive symptomatology (Von Essen, et al., 2004), more PTSS symptoms (Kazak, et al., 2005), greater caregiver demand (Klassen, et al., 2010; Steele, Long, Reddy, Luhr & Phipps, 2003) and role strain (Quittner et al., 1998), and a greater extent of perceived loss of control during required treatments (Norberg & Boman, 2011). Norberg and Boman (2011) offered an explanation for these differences. They suggested that generally mothers are in control of care and decisions about care, and the parenting role may be more central to the identification of mothers than fathers. Therefore mothers may be more affected by their child's illness and subsequent treatment because of the shift of the primary carer role from mother to HCPs.

Similar to research into parental experiences of CF, much less is known about fathers' experiences of their child's cancer diagnosis and subsequent treatment compared to the knowledge of mothers' experiences. Research has suggested that fathers with a university degree or higher show greater anxiety about their child's diagnosis than lower educated fathers (Mu, Ma, Hwang, & Chao, 2002); fathers experience guilt related to role obligations due to their incapacity to provide further care for their child because of their obligations to provide financial stability (Chesler & Parry, 2001); and fathers are more concerned about their family financial situation than family care (Hovey, 2005). It does appear that research into similar treatments, prognoses and disease trajectories may provide some insight into parental experiences, and how parents cope with early surveillance for CF lung disease. However these bodies of literature fail to take into account the exact nature of, and processes involved in, early disease surveillance for CF lung disease. As there is currently no published psychological literature examining the impact of early surveillance for young children and infants with CF on parents, the literature review has attempted to provide evidence for the types of coping resources and experiences that parents whose children undergo early surveillance may be experiencing. Therefore, caution must be exercised when considering coping and psychological outcomes of diseases that, whilst similar in terms of chronicity or severity or that have similar treatment trajectories, may have distinct and important differences to CF.

The AREST CF program should be responsive to children's, parents' and families' psychological well-being because of the potential burden of invasive surveillance procedures and subsequent aggressive treatment. Therefore, extending knowledge about parents' psychological and social experiences associated both with their child's CF and specifically with the early surveillance process is a formidable task. It is likely to lead to adequate and evidence-based intervention

targeting identified issues and enhancing beneficial experiences and positive outcomes, which may lead to better parent mental health outcomes and better child health and developmental outcomes. Moreover, urgency exists for research to explore psychological and social factors that could positively influence children's, parents and families' lives whilst maintaining the child's physical health through early surveillance of covert disease progression and subsequent treatment. Once this is understood, education for medical staff and preventative or therapeutic intervention for parents and/or children on these matters could play an important role in improving outcomes for parents, children and their families.

Consequently, in an attempt to explore parenting within the context of early surveillance for covert disease progression of a chronic condition, I recognised that the first step involved was asking parents about their experiences, whether it be about diagnosis, treatment management, the early surveillance program or other, broader, parenting experiences (Creswell, 2007). Given the paucity of information about parents' experiences within this unique context, a primary aim of my research is to explore the lived experiences of parenting in the context of early surveillance for covert disease progression. The particular aims of my research are outlined in detail in the next section.

1.2.3 Aims of the research

It is important to articulate the scope of my research. It was intended to neither advocate for or against early surveillance for CF lung disease, nor to privilege early surveillance over other clinical care pathways. The primary purpose of my research is to explore the lived experience of parenting children undergoing early CF disease surveillance by providing a comprehensive account of how parents construct their experiences and attribute meanings to early surveillance. Moreover, is the aim to describe how parents cope with their experiences. Due to the unique experience of parents in my research, the intention was not to generalise the experience of parenting within the context of early surveillance for covert disease progression but rather to offer an in-depth exploration of this phenomenon in its complexity and entirety against the wider contexts with which it occurs. Whilst parents' experiences of their child's early surveillance were explored, I also investigated the experience of parenting within the context of early surveillance.

1.2.4 Research questions

Given the research aims to explore; the lived experience and how parents construct their experiences and attribute meanings to early surveillance for CF lung disease, and how parents cope with their experiences, a qualitative inquiry paradigm is deemed methodologically appropriate. Focussing on meaning of experiences meant that my research was ontologically consistent with phenomenology (Guba & Lincoln, 1994), which emphasises the importance of investigating the lived experience from the perspective of those who have experience with it (Barkway, 2001; Creswell,

2007; Liamputtong & Ezzy, 2005; Moutsakas, 1994; Patton, 1990) and focusing on how people construct their experience in the context of their own life world (Creswell, 2007; Flood, 2010; Patton, 2002; Zippel, 2010). Therefore, phenomenological research describes humanistic phenomena as it is lived and experienced by people; rather than attempting to measure and quantify an abstracted explanatory account (Crotty, 1996). A constructivist epistemology was used to generate insights into how parents interpreted and formed meaning around their experiences. Constructivism emphasises that construction of reality occurs as a dual process between two people whereby it is constructed in the mind of the individual (Hansen, 2004) and co-constructed by researcher and participant. An interviewing approach was used to collect data from parents, as it was pragmatically and theoretically the most appropriate way to explore each for my research questions (Creswell, 2007; Denzin & Lincoln, 2005; Riessman, 2008). To this end, research questions in qualitative research paradigms are often open-ended and exploratory in nature (Elliot & Timulak, 2005). The over-arching research questions to address the aforementioned aims of my research are as follows:

- What is the lived experience of parenting in the context of early disease surveillance for CF?
- What are parents' experiences of early CF disease surveillance?
- How do parents construct their experience and attribute meanings to early surveillance for CF lung disease?
- How do parents cope with the lived experience of parenting in the context of early surveillance?

1.2.5 Implications of the research

My research contributes to a growing body of knowledge about the parenting experience in paediatric chronic disease. Uniquely, findings from my research offer insight into the lived experience of parenting a child undergoing early intervention and aggressive treatment for disease progression. This type of information can assist in providing holistic support for parents and their families. In practical terms, identification of psychological factors important in maintaining good parent and family health, and insight into experiences of parents with children undergoing early surveillance with AREST CF, may be used to inform existing and future policy decision-making and models of best clinical practice towards promoting a positive parenting experience. Accordingly, my research informs existing and future policy structures through identification of previous omissions, misconceptions and anecdotal assumptions regarding parenting children in early surveillance and aggressive treatment for their chronic condition. Moreover, my research contributes to understandings of parenting children with chronic conditions as a process of acceptance and redefinition involving positive appraisals and providing opportunities for resilience, rather than

focused solely on the adverse experiences and outcomes associated with paediatric chronic disease. Additionally, the findings from my research highlight the relevance of supporting families from a salutogenic approach rather than from the traditional deficit-focused models. Foremost, my findings contribute to establishing a more holistic understanding of the parenting experience in paediatric chronic disease and provide evidence that parenting a child with a chronic condition is a socially constructed and therefore modifiable experience.

1.3 Overview of the thesis

The structure of my thesis has been organised into seven chapters. Each chapter begins with an overview and concludes with a summary. Chapter one introduces the research topic by identifying the need to understand the current experience and construction of parenting given the current agenda of early surveillance and aggressive intervention for covert disease progression. Moreover, is the need to understand parents' attributions of meaning to their experiences and how they cope with these experiences. With a dearth of information about the lived experience of parenting in this context, I argue that to understand experiences of these parents, it needs to be explored from the perspective of parents themselves. Additionally, recognising that theoretical frameworks and clinical agendas are significant factors influencing our understanding of how parents construct their experiences of, and attribute meanings to, their child's chronic condition, as well as how they cope, will provide the foundation of discussion in my thesis. The first chapter therefore introduces the research context, development, focus and rationale of my research and outlines the specific research aims and questions relevant to the exploration. This discussion leads to the next chapter, which reviews current literature about parenting a child who has CF.

Chapter two critically examines relevant literature and context underpinning current conceptualisations of parenting children with CF. As a focus of my research is on parents' constructions of their experience, psychological perspectives of different aspects of parenting children with CF, are of primary concern in review of the literature. Specifically, this chapter aims to identify current understandings of how parents manage diagnosis and treatment within the first 7 years of life within modern approaches to CF management. This discussion then leads to the next chapter, which reviews major theoretical developments in coping with parenting a child with a chronic condition.

Chapter three outlines substantiations for theoretical and methodological choices made to conduct the research. Firstly, this chapter aims to identify current theoretical understandings of parenting a child with a chronic condition with a particular focus on theories focussing on coping, adaptation and resiliency in the face of adversity. Accordingly, dominant theories and models currently influencing contemporary debates and discussions on coping with parenting in paediatric chronic disease are reviewed. Presented in this chapter is critical review on how parenting should be

considered in the context of broader family and community conditions including the role of appraisals of the child's condition and the family's ability to cope with the demands, and their newly-instituted patterns of family functioning to cope with the demands. Next is the rationale for methodological choices of research design and paradigm. Specifically, this section aims to detail how phenomenological inquiry, data collection and analysis, and sampling techniques informed the conduct and output of the research. This leads to the next chapter, which outlines methodological actions taken to conduct the research.

Chapter four describes the methodological approach used in my research. This chapter initially discusses phenomenology and constructivist frameworks, which have informed and directed my research conduct and output. As my research adopted a qualitative methodology to explore parents' experiences, constructions, and attributions, the philosophical underpinnings of a qualitative paradigm are detailed. Participant characteristics, research procedures, ethical considerations, my role as a researcher, and data collection and analysis techniques are presented. Therefore, the aim of this chapter is to outline the research methodology and the means by which the research was conducted, and findings from the data were derived and substantiated. The next chapter presents the findings and interpretations of the data.

Chapter five introduces the research findings relevant to addressing the research questions. Specifically, this chapter presents findings on parents' experiences and constructions of parenting in the context of early surveillance for CF lung disease. Moreover, the chapter presents findings about how parents cope with their experiences, and how they attribute meaning to early surveillance for CF lung disease. Similarities and points of difference are reviewed, with findings presented in a structure of five major themes and their associated sub-themes. These major themes included: *redefined expectations and reimagined identity*; *redefined reality* (consisting of sub-themes - collapsed and redefined expectations and new normal); *understanding the unknown and understanding uncertainty* (consisting of sub-themes – understanding CF and understanding early surveillance for CF lung disease); *good days and bad days*; *fluctuation between positive and negative outlook on life* (consisting of sub-themes – bad days, good days and fluctuation between good days and bad days); *early surveillance is a significant event* (consisting of sub-themes - indicator of treatment efforts and examination of parenting competence, parental outcomes directly from early surveillance and fear from diagnosis exacerbated at each annual review can result in anxiety); and *early surveillance is in the best interest of my child* (consisting of sub-themes - perceived clinical benefit to child health and cognitive dissonance results in anxiety and/or ambivalence). The findings are critically examined with reference to current understandings and constructions of parenting in paediatric chronic disease. Theoretical application of the findings is addressed in the next chapter.

Chapter six provides further discussion of the findings by presenting a critical consideration of how relevant research findings can be applied to theoretical models presented in chapter three, with particular reference to how coping and adaptation can be achieved from a family perspective. Parental characteristics and parenting processes that contribute to family adaptation are identified, along with how some factors that may contribute to adaptation can be negatively influenced by the early surveillance experience. The chapter concludes by proposing that strength-based, salutogenic models of care and support are appropriate conceptualisations of parenting children with chronic conditions who undergo early surveillance for covert disease, and appear better approaches than the traditional deficit-models of care and support. Moreover, resiliency as an outcome for families facing adversity should be of primary consideration in current models of care for the entire family.

Chapter seven is the concluding chapter of the thesis that pulls together a summary of the salient findings and issues raised across the research. Within context of the research questions, my findings are discussed in relation to the implications they have for policy and clinical practice, and theoretical development of parenting in paediatric chronic disease. The limitations of my research are then discussed with particular reference to opportunities and recommendations for future research and practice before concluding with a post-script anecdote about my research journey.

CHAPTER 2**Cystic fibrosis in the 21st century**

This chapter describes cystic fibrosis (CF) and contextualises paediatric CF in an Australian context. The chapter is divided into four sections, each of which contributes to the rationale for conducting the research within the chosen methodology. In the first section, CF will be comprehensively explained including current diagnosis practices and treatment options. Living with CF as a child will be outlined before presenting the burden of CF management experienced by parents and their families. The second section will review parenting in paediatric CF by reviewing empirical evidence of how parents adjust to, construct meaning of, and experience, their child's CF diagnosis and parenting a child with CF in their first 7 years of life. This section will conclude with discussion about contribution of parental characteristics on child mental and physical health outcomes. The third section outlines empirical support for parents' strategies and abilities for coping with a child's CF management, before viewing how parents cope from a resiliency perspective. The fourth section of the chapter provides specific rationale for conducting the research, culminating in its aims and significance.

Out of your vulnerabilities will come your strength...Sigmund Freud

2.1 What is CF?

2.1.1 Prevalence, life expectancy, causes and clinical features.

Cystic fibrosis (CF), the most common life-shortening autosomal recessive disease of Caucasian people, is a multi-systemic and genetically inherited disease affecting 1:2,500 - 3,500 live born infants (Daly, et al., 2007; Davies, et al., 2007; Montgomery & Howenstine, 2009) though its frequency varies in specific groups (Daly, et al., 2007). An analysis of literature reviews, surveys, and registry data of European Union (EU) countries revealed mean prevalence rates of 0.737:10,000 live born infants in the 27 EU countries, which is similar to the value of 0.797:10,000 live born infants in the United States, with one outlier, namely the Republic of Ireland at 2.98:10,000 live born infants (Farrell, 2008). The high Irish prevalence rate is likely attributable to a high genetic mutation prevalence and consanguinity (Farrell, et al., 2007). Current Australian prevalence rates are approximately 1:3,000 live births (Australian Bureau of Statistics, 2009; Cystic Fibrosis Australia, 2013).

Progress in understanding CF and its impact on disease management has been rapid over the past 20 years, with CF changing from a fatal childhood digestive and lung disease to a complex multi-systemic disease extending into adulthood (Davies, et al., 2007). With advances in science, medicine and technology, life expectancy of CF patients has increased dramatically over the past four decades, increasing from less than 1 year in 1960 to a median survival age of approximately 40 years now (UK Cystic Fibrosis Registry, 2012). Availability of direct mutation analysis and the gene sequence were turning points in the history of CF and opened a new era of research into molecular and cellular studies in CF research (Zielenski, 2000). This has resulted in identification of CF gene mutations, which has allowed early diagnosis and therefore early and prophylactic treatments for CF, culminating in increasing life expectancy for people living with CF.

Cystic fibrosis is caused by mutations identified on the long arm of chromosome 7 (7q.31.2), and identification of the mutation was first isolated by positional cloning in 1989 (Kerem, et al., 2005; Riordan, et al., 1989; Rommens, et al., 1989). The mutation occurs in what is known as the cystic fibrosis transmembrane conductance regulator (CFTR) gene (Riordan, et al., 1989). Different mutations in the CFTR gene result in varying phenotypes of the disease (Rowntree & Harris, 2003). The same mutation may also result in various phenotypes, for example, two people with the same mutation may or may not be pancreatic sufficient (Davies, et al., 2007). Expositions for this anomaly are currently being proposed in the literature, with environmental (Folkesson et al., 2012) and chromosomal interactions (Drumm, Ziady, & Davis, 2012) put forth as the most plausible explanations.

Cystic fibrosis primarily affects chloride channel transport in epithelial cells in the intestine, respiratory system, pancreas, gall bladder and sweat glands (Rowntree & Harris, 2003); therefore manifesting in a number of organs, most notably in the upper and lower airways, pancreas, bowel, and reproductive tracts (Davis, 2006) leading to chronic obstructive lung disease, exocrine pancreatic insufficiency, elevated chloride concentration and infertility in males due to absence or obstruction of the vas deferens. Females also suffer from reduced fertility, possibly due to altered cervical mucus properties (Daly, et al., 2007; Zielenski, 2000). The lack of CFTR function causes gastrointestinal issues because the intestines are unable to break down fats (John Hopkins Cystic Fibrosis Center, 2015), requiring continue enzyme replacement therapy to help the intestine absorb fat-soluble vitamins. Though for most patients with CF, lung disease is the most important issue in terms of symptoms and treatment because it is the most likely cause of death (Davies, et al., 2007). The sweat gland duct is also affected; concentration of salt in sweat of CF patients is elevated (3-5 times more than normal levels) (Daly, et al., 2007). The identification of elevated salt in sweat is a clinical indication that CF is present, which is usually conducted after an abnormal Guthrie heel-prick test (Guthrie & Susi, 1963). The most recognised symptom of CF disease is chronic obstructive lung disease (Montgomery & Howenstine, 2009). Chronic, suppurative lung disease causes over 90% of the morbidity and mortality associated with CF, leading to premature death from respiratory failure in early adulthood (Rajan & Saiman, 2002).

2.1.2 Chronology of CF into the 21st century.

Based on historical medical documents, CF apparently appeared about 3,000 years ago, possibly due to migration of peoples, gene mutations, and new conditions in nourishment (Busch, 1990). The complete clinical range of CF symptoms was not recognised until the 1930s (Andersen, 1938), however aspects of CF were identified much earlier. For example, in the 18th century, Northern European folklore warned, translated to English, *woe to that child which when kissed on the forehead tastes salty. He is bewitched and soon must die* (Busch, 1990). The significance of saltiness was later demonstrated through the association between excessive salt content in sweat of affected children and the condition (Di Sant'Agnese, Darling, Perera & Shea, 1953), which resulted in the measurement of chloride and sodium in sweat as the diagnostic standard for the condition (Gibson & Cooke, 1959).

Aspects of CF were reported in the medical literature throughout the early 1900s, including a correlation between coeliac disease, CF of the pancreas and bronchiectasis (Fanconi, Uehlinger, & Knauer, 1936). In 1938 Dorothy Hansine Andersen first described CF using the term *cystic fibrosis* as a reference to the characteristic scarring, or fibrosis, and cyst formation within the pancreas. This was the result of many electrophysiological studies highly suggestive of a defect in chloride transport of CF secretory epithelial cells (Quinton, 1983). Andersen was the first to describe characteristics of

CF of the pancreas and to associate it with intestinal and lung disease prominent in CF. She also first hypothesised that CF was a recessive condition, and she led the use of pancreatic enzyme replacement to treat affected children. Subsequent research has found more than 1,000 different mutations that cause CF, for example, one study alone found more than 50 mutations in a sample of 350 German patients with CF (Dörk et al., 1994), with another study finding a further 17 mutations in a sample of 137 French CF patients (Claustres et al., 1993). Since the discovery of countless CF-causing gene mutations, many studies have investigated the correlation between CFTR genotype (that is, a person's genetically coded information) and CF disease phenotype (that is, the physical manifestation of genetically coded information) (Montgomery & Howenstine, 2009; Rowntree & Harris, 2003; Zielenski, 2000).

Therefore, early understandings of CF disease and its effect on different body organs has advanced to contemporary understandings of CF mutations, their molecular consequences, their disease mechanisms, and environmental and CF gene modifiers. This has resulted in CF being classified as a disease with a complex, multi-faceted clinical phenotype that is the sum of a number of variable clinical components that arise from the underlying genotype in a unique and organ-specific manner. Lastly, studies in CF have illustrated that CF genotype-phenotype can be confounded by secondary genetic and environmental factors and not only by different molecular mechanisms of underlying mutations (Montgomery & Howenstine, 2009; Rowntree & Harris, 2003; Zielenski, 2000).

Due to the complexity in CF gene dysfunction and variability in genotype-phenotype outcomes, hope for a cure in the foreseeable future is now understood as not as realistic as it was when the CF gene was discovered back in 1989. As an example, attempts to insert normal genes into epithelial cells have not been as therapeutically useful as anticipated (Knowles, et al., 1995). With this understanding, researchers are now focussing on gene therapies as a way to manipulate the dysfunction caused by the genetic mutations. Kalydeco (Ivacaftor) is one such treatment that has shown promising results for individuals with the G511D mutation (Ledford, 2012). Unfortunately for infants and young children with CF, Kalydeco is only currently registered for individuals with CF who are 6 years of age and older.

2.1.3 CF diagnosis: Current Australian practice via newborn screening.

The most discriminatory diagnostic test for CF is the measurement of sweat electrolyte levels (Di Sant'Agnese, et al., 1953), and it remains the gold standard for diagnosis (Montgomery & Howenstine, 2009; Rosenstein & Cutting, 1998). The median age for diagnosis with classic CF is approximately 6 months, whilst diagnosis of non-classic forms of the disease with milder symptoms can be delayed. Elevated levels of a particular protein, immunoreactive trypsinogen in the blood (Crossley, Elliott and Smith, 1979), is the most common phenotypic attribute utilised to establish a

definitive CF diagnosis, and was implemented as part of newborn screening (NBS) in New Zealand and regions of Australia in 1981 (Barben, et al., 2012). In Australia, NBS is conducted through a national public health program.

As a public health program, NBS is designed to screen infants for a number of conditions that are not clinically evident during the newborn phase, with screening usually conducted shortly after birth, usually 1-3 days (Lindner, et al., 2011). Generally conducted by state or national governing bodies, NBS programs aim to screen all infants born within their jurisdiction, with the number of conditions screened for decided by each governing body (Loeber, et al., 2012). Whilst there is some variation between states and territories as to which conditions are screened for in Australia, the following conditions are currently screened for nation-wide; phenylketonuria, galactosaemia, primary congenital hypothyroidism, some rare metabolic conditions and CF (Barlow-Stewart, Emery & Metcalfe, 2007). If left undiagnosed, many of these conditions can cause permanent physical and intellectual disability.

The implementation of NBS internationally and the development of AREST CF has resulted in prophylactic treatment for children and infants with CF in an Australian context, and a paradigm shift from reactive treatment to proactive early-life disease surveillance and therapeutic intervention. Much of the physiological improvement in long term health outcomes has been attributed to improvement in diagnosis and therapy. Therapeutic interventions administered before the onset of symptoms, such as those offered by AREST CF, may have the greatest long term benefit (Farrell, et al., 2001). This has led to the notion that asymptomatic diagnosis and early treatment may optimise early nutritional status and prevent or reduce irreversible pulmonary damage, thereby improving outcome and quality of life in people living with CF (Southern et al., 2007).

Although NBS now occurs in Australia and several countries across the world, its protocol and subsequent treatment following diagnosis is not systematically conducted, with varying treatment protocols across countries. For example, some paediatric CF centres administer treatment following positive results for inflammatory markers from bronchoscopies (e.g., Princess Margaret Hospital and Royal Children's Hospital in Australia, and the Great Ormond Street Hospital in England), whereas other CF centres do not conduct routine bronchoscopies in young children. The various treatment following diagnosis means that a systematic CF treatment approach does not currently exist. This indicates a need for a standardised, and empirically supported, treatment protocol that serves to improve clinical outcomes and quality of life for children with CF.

2.1.4 What is it like to be a young child with CF in the 21st century?

Today, children with CF live relatively conventional lifestyles, albeit with some family adjustments requiring parents to complete daily treatment regimens of physiotherapy and enzyme replacement therapies. Currently, approximately 70% of children with CF are diagnosed before the

age of 1 year (Walters & Mehta, 2007), with clinical symptoms often present at birth (FitzSimmons, 1993). Respiratory symptoms are present in more than 50% of newborns, with infections such as *Pseudomonas aeruginosa* occurring in early infancy (Walters & Mehta, 2007) and pancreatic abnormalities occurring within days of birth (Littlewood, 2007). As children get older, they may experience further CF-related symptoms and also pain associated with the disease including headaches, musculoskeletal pain and abdominal cramping (Koh, Harrison, Palermo, Turner, & McGraw, 2005; Palermo, Harrison, & Koh, 2006). These symptoms may also impact upon sleep (Quittner, Barker, Marciel & Grimley, 2009), which brings with it a multitude of both psychological and physiological issues that can be affected. For example, research has shown that sleep can adversely affect emotional responses, behaviour, attention and academic performance in children (Berger, Miller, Seifer, Cares, & Lebourgeois, 2012; Fallone, Acebo, Arnedt, Seifer, & Carskadon, 2001; Gozal, 1998; Yue, Conrad, & Dimsdale, 2008). Therefore, children's quality of life may also be detrimentally affected by the physical symptoms caused by CF (Palermo, et al., 2006).

Improvements in life expectancy of CF patients has also shifted the focus of CF research and clinical care from improving physiological outcomes toward the complex interplay among behavioural, psychosocial and health outcomes (Barker & Quittner, 2010). This change in focus has been marked by a growing number of patient-reported outcomes, such as health-related quality of life and efforts to address behavioural and psychosocial challenges, such as adherence and depression (Quittner, Modi, Lemanek, Ievers-Landis & Rapoff, 2008; Quittner, et al., 2008). However for ethical and practical reasons, this shift has not tended to focus on very young children with CF as much as adolescents and adults with CF.

As children with CF experience frequent hospital visits and repeated medical procedures (Thompson & Gustafson, 1996), it is thought that this can contribute to onset of psychological and behavioural difficulties (Boyd & Hunsberger, 1998; Pao & Bosk, 2011; Thompson, Gustafson, Hamlett & Spock, 1992a). Häglöff (1999) suggested that children of various ages were at higher risk of psychological problems following increased follow-up surgeries from early interventions. Another area of concern is children's reactions to painful or distressing medical procedures (Hedström, Haglund, Skolin, & von Essen, 2003). Children continue to find *having a needle* as one of the most distressing aspects of attending hospital (Duff & Oxley, 2007; Ellis, Sharp, Newhook, & Cohen, 2004; Kennedy, et al., 2008). One study in CF found over 30% of children felt *extremely* or *very* frightened of venepuncture, especially prior to needle insertion (Duff & Brownlee, 1999). Together, these studies highlight environmental factors which may interact with distressing aspects of CF disease. Moreover, the majority of these studies did not differentiate across all age groups, therefore failing to identify at what age such psychological and behavioural difficulties may begin to manifest. By further identifying age of onset for such difficulties, or which personality characteristics may

contribute, clinicians and other allied health professionals would be better-equipped to implement strategies to combat psychological and behavioural difficulties that occur as a result of medical procedures children with CF must undergo. There is also potential for implementation of targeted interventions for prevention rather than therapy if research can pinpoint age groups for which such difficulties may begin to manifest.

2.1.5 Burden of early CF care on families in the 21st century.

Improvements in CF management and health outcomes have meant that the landscape of parents' experiences of CF has changed. As CF is no longer classed as a fatal childhood condition, previous experiences of fatality in childhood and morbidity from the condition generally bear less relevance today for parents. However, due to advancements in treatment, CF care is now primarily the responsibility of parents. Consequently, despite improvements in life expectancy, CF can still impose significant burden on children, their parents and their families. In particular, the shift in responsibility to home treatment and daily therapy can cause treatment burden for parents (Bregnballe, Schiotz, Boisen, Pressler & Thastum, 2011; Quittner, Drotar & Ievers-Landis, 2000; Ziain, et al., 2006). Moreover, for parents whose children undergo early surveillance of lung disease, knowledge of how their child's disease is developing in their early years of life may cause a psychological burden for parents (Sawicki, et al., 2015).

Parents generally establish a family routine that is centered around CF care and management (Agent & Madge, 2007). Medical activities such as performing routine chest physiotherapy or administering oral medication to an infant may feel unnatural and awkward to parents, and the parenting role may become confused with a more medicalised role during these early years (Agent & Madge, 2007). The performance of daily treatments is also time consuming, and can interfere with parents' social functioning and work roles (Quittner, et al., 1998). Managing treatment increases parental stress and has been associated with depression, poor sleep quality and marital strain in parents of young children with CF (Glasscoe, Lancaster, Smyth, & Hill, 2007; Quittner, et al., 1992; Yilmaz et al., 2008). This means that successful management of the child with CF depends largely on the ability of the family to change its lifestyle. Families must work together to accommodate the time-consuming demands of a complicated daily treatment regimen consisting of a multitude of therapies, according to the recommendations of the healthcare team (McCubbin, et al., 1984).

Psychosocial factors that function as moderators of treatment adherence in parents have been a dominant research focus for a number of reasons. These include the responsibility of CF care within the home, the high treatment burden associated with CF management, and the recognised importance of treatment adherence for the long-term management of adolescent and adult CF. Estimates of treatment adherence by parents vary according to method of measurement, type of

treatment and age. In one of the few studies measuring adherence to several treatment components using a number of assessment methods, Modi and colleagues (2006) reported adherence rates for children ranging from 22% to 71% for objective measures and 67% to 100% for self-reported measures, suggesting inflated adherence rates for self-reported measures. The study also found significant variability between treatments, a finding that is consistent with other CF studies (Quittner, et al., 2000; Quittner, Espelage, Ievers-Landis & Drotar, 2000; Quittner, et al., 2008). The literature suggests that treatment adherence, and more so the reporting of it, is a complex matter.

Maternal depression has been associated with low adherence to airway clearance (Smith, Modi, Quittner & Wood, 2010), and identified as an important predictor of poor enzyme adherence and shows short-term effects on weight gain and growth in children (Quittner, Barker, Geller, Butt & Gondor, 2007). Another study found significant associations between parental stress, depression, eating and sleeping difficulties in children on the one hand, and poor adherence to nebulised medications during the preschool years on the other (Ward, Massie, & Glazner, 2009). Sheehan and colleagues (2014) reported correlations between parental avoidant coping and internalised behavior problems arising in children, as well as child mealtime/eating difficulties, which can result in a low nutritional status for children with CF. Together, these findings indicate that parental psychological issues such as depression and stress are likely to have an adverse effect on treatment management (e.g., attending clinic appointments) and adherence (e.g., maintaining recommended home treatments). Therefore, annual screening of depression and other potential psychological issues and adequate support for parents of children with CF during the child's early years may improve parental and child health outcomes. This underscores the notion that parents are the most important managers of treatment for young children with CF, and will require continuous family, medical and social support in their roles.

Families of older children with CF have been shown to function better than families of younger children with CF (Knafl, et al., 2013), and better family functioning has been associated with better treatment adherence (Everhart, Fiese, Smyth, Borschuk & Anbar, 2014; Patterson, et al., 1993). Collectively, studies to date highlight the importance of parent well-being and family functioning in treatment adherence. Furthermore, children in these studies would have been old enough to administer their own medication. Family functioning and treatment adherence has not been sufficiently addressed in families who have an infant or young child with CF. It is important to understand how, why, and at what point parent well-being and family functioning influences treatment adherence. An additional implication of investigating treatment adherence in families across large, undifferentiated age ranges is the difficulty in determining the relevance of findings to families with very young children. Treatment management and adherence in the older population

primarily focusses on autonomous disease management and not that of parent-led management, which is the case for the younger population.

The hidden, or asymptomatic, nature of early CF disease progression has dominated research in the young population, attention has not therefore been given to parents' experiences of, and reactions to, how a child's disease is developing. Nor has research focussed on how parents cope with such knowledge. Though it is now understood that covert disease progression can start very early in life (Ramsey, et al., 2016; Stick, et al., 2009), psychological research into parental understandings of such information are yet to be integrated.

2.1.6 Summary and conclusion.

Cystic fibrosis is the most common life-shortening disorder of Caucasians, with systemic involvement including the lungs and pancreas. Recent advances in NBS have provided opportunity for early surveillance and proactive, aggressive treatment of CF. Life expectancy of children born with CF has increased exponentially over the past 30 years, though both physiological and psychological issues remain important issues for children and their families. Moreover, parent mental health and family functioning can be affected due to home treatment and daily therapy. Currently, clinical guidelines and standards of care are generally limited to medical aspects of CF with less consideration of accompanying psychosocial aspects. Although national standards of care (Bell & Robinson, 2008) advocate for a psychologist and social worker as part of the multidisciplinary CF team, national consensus on how to assess psychological health of parents and families and how to address these issues within clinical care is urgently needed. However, a recent consensus statement from the International Committee on Mental Health in Cystic Fibrosis asserted the need to assess anxiety and depression annually in parents and caregivers of children with CF (Quittner, et al., 2015).

2.2 Families living with CF

2.2.1 Psychological adjustment to a CF diagnosis.

A CF diagnosis in a newborn is undoubtedly devastating news for parents. For most parents the diagnosis of CF in their newborn infant is unexpected (Massie, Forbes, Dusart, Bankier, & Delatycki, 2007) and often parents have little knowledge about the disease or carry out-dated misconceptions about its lethality in childhood (Jedlicka-Köhler, Götz, & Eichler, 1996) despite pre-diagnostic genetic counselling during NBS (Tluczek, Orland, Nick, & Brown, 2009). The psychological impact of a CF diagnosis is well documented in the literature with disruption of family routine, shock, denial, anger, guilt, worry and sadness ensuing (Eiser, Zoritch, Hiller, Havermans & Billig, 1995; Gayton, Friedman, Tavormina & Tucker, 1977; Marvin & Pianta, 1996; Priddis, Dunwoodie, Balding & Douglas, 2010; Quittner, et al., 1992; Turk, 1964; Venters, 1981). A crisis period is generally reported to last up to one year (Venters, 1981), with a number of studies reporting that, after a period of

initial disruption and adjustment, families generally return to relatively common and conventional levels of family functioning (Cowen, et al., 1985; Cowen, et al., 1986; Knafel & Gilliss, 2002; Pfeffer, Pfeffer, & Hodson, 2003; Sawyer, 1992; Szyndler, et al., 2005).

However, the above-mentioned research is contrasted with the notion that following diagnosis, parents continually experience negative psychological outcomes compared to controls and norms, such as more depressive symptoms, higher stress and anxiety, and greater role strain (Blair, Cull, & Freeman, 1994; Breslau, Staruch, & Mortimer, 1982; Brucefors, Hochwalder, Sjovall, & Hjelte, 2015; Goldberg, Morris, Simmons, Fowler, & Levison, 1990; Mullins et al., 1991; Quittner, et al., 1992). As a composite of these arguments for parental adjustment to diagnosis, Duff and Brownlee (2008) posited that once diagnosis is confirmed, parents typically continue to experience fluctuating states of psychological distress and normalcy as they take on the role of medical parent. Phenomenological studies into the experience of caring for a child with CF confirm this supposition. Parents appeared to be on a continuous journey of uncertainty as they struggled to adapt to their child's diagnosis, alternating between periods of adjustment, facing adversity and readjustment due to the progressive nature of the disease and changing health outcomes of their child (Glasscoe & Smith, 2008, 2011; Jessup & Parkinson, 2010). From these studies, families generally follow a pattern of initial crisis following diagnosis replaced somewhat later by adaptation and acceptance, with cyclical periods of adversity and re-adjustment possibly associated with developmental milestones and/or disease progression.

2.2.2 Outcomes of parenting a child with CF.

Similar to parenting a healthy child, parenting a child with CF can bring both hardships and joy to parents. Theoretical approaches to understand parenting a chronically ill child traditionally stem from a psychopathologic perspective. Whether parental outcomes in paediatric CF are a product of these early theoretical underpinnings is not the topic of debate here; the purpose of discussion here is to provide a substantial summary and critique of the literature, and what is and is not reported in terms of parents' experiences and outcomes. Notwithstanding the trend towards investigating psychopathologic outcomes, parental outcomes in paediatric CF include stress, depression and anxiety, guilt and chronic sorrow, with some reported effects on parental identity formation and parenting self-efficacy.

2.2.2.1 Parental stress, depression and anxiety.

The life-shortening nature of CF, time consuming treatments and ongoing morbidity (Davies, Alton, & Bush, 2007) place pressure on parents beyond normal parenting duties. With the diagnosis of CF generally occurring in the first year of life, parents must not only learn about their new baby, but also about a new and often frightening condition requiring rapid change and adjustment in parenting roles and plans for the future (Gotz & Gotz, 2000; Quittner et al., 1998; Quittner, et al.,

1992). High levels of continual parenting stress and depression are often reported (Quittner et al., 2008; 2014).

In terms of psychological effects of CF, early studies presumed that children with CF and their families would experience psychopathology, which was predictably found (Lawler, Nakielny & Wright, 1966; Pinkerton, 1969; Teicher, 1969; Tropauer, Franz, & Dilgard, 1970; Turk, 1964). Since the 1960s, most research into psychological effects of CF has focused on maternal reactions and responses. Such research has generally shown that mothers experience negative effects of CF, such as heightened depression and anxiety (Mullins, et al., 1991; Quittner, et al., 1992; Yilmaz, et al., 2008), though high rates of depression are reported for both mothers and fathers soon after diagnosis (Glasscoe, et al., 2007). A current international epidemiological study is underway, researching population-based estimates of depression and anxiety, and examining their associations with health outcomes (Quittner, et al., 2008).

This large-scale ongoing epidemiological study (The International Depression/Anxiety Epidemiological Study – TIDES), that commenced in 2007, is measuring levels of anxiety and depression in both patients with CF from ages 12 – 35 years and caregivers of children with CF ages 1 - 17 years in 18 countries (<http://www.tides-cf.org/>). An early study from this collaboration reported that almost a quarter (20%) of caregivers reported clinical levels of depressive symptomatology, and more than a quarter (28%) reported elevated levels of anxiety. What this study and other studies from Germany and America within the TIDES collaboration (Besier, et al., 2011; Driscoll, et al., 2009) indicate is that caregivers adapt surprisingly well to the demands of their infant's or young child's condition. However, these studies also indicate that a substantial proportion of caregivers report elevated levels of psychological distress associated with caregiving for a child with CF. The TIDES study will go some distance in consistently determining prevalence and severity of anxiety and depression in patients and their caregivers across countries and across cultures.

However, critiques of the TIDES study thus far have reported methodological weaknesses due to inconsistencies in psychometric measurement across countries (Webb & Bryon, 2014), bringing into question clinical significance of reported findings. Additionally, whilst the design of the study enables risk factors for depression to be identified, which will help better our understanding of the nature of psychological needs of parents with children who have CF, such knowledge of increased risk for parental depression in paediatric chronic disease is not new (Besier, et al., 2011; Glasscoe, et al., 2007; Goldbeck, et al., 2008).

Research has also shown that mothers' experiences of CF are more negative than fathers, with mothers experiencing more stress (Van Os et al., 1985) and a greater incidence of depression than fathers (Quittner, et al., 2014). However, fathers of children with CF had more depression compared with fathers of healthy children (Brucefors, et al., 2015; Goldberg, et al., 1990). To date,

there has been much less research on fathers' experiences of their child's CF, with suggestions in the literature that fathers experience greater financial strain and have less time to spend with their children and family as a result of these financial demands (Blair, et al., 1994; Goble, 2004; Hayes & Savage, 2008; Nagy & Ungerer, 1990). To determine what factors lead to parental stress and depression for both parents, and what leads to higher parental stress and depression in mothers, processes that lead to family stress in relation to their child's diagnosis have been researched. Quittner and colleagues (1992) found that mothers often took primary responsibility for their child's treatment, which led to increased stress and more depressive symptomatology.

Beyond the diagnosis phase, few studies have assessed psychological functioning of parents (Quittner, et al., 2009). Consequently, general trends of psychological functioning and adaptation for parents of infants and young children with CF have not yet been established. Psychological adaptation to a child's CF has been likened to the bereavement process, with distinct stages of shock, belief, anger, adjustment and adaptation (Cousino & Hazen, 2013). This does not, however, accurately reflect commonly reported experiences by parents. Despite hope of gene therapy and increased longevity of life, parents continue to worry they will outlive their child. What seems more appropriate is the notion of *chronic sorrow*, with dynamic phases of normalcy and routine, interspersed with intense sadness and mourning (Duff & Oxley, 2007).

2.2.2.2 Guilt and chronic sorrow.

Due to the devastating nature of paediatric chronic disease, guilt and chronic sorrow are commonly-reported outcomes for parents of children with both chronic and life-limiting conditions including type 1 diabetes, asthma, cerebral palsy, neurological disability and leukaemia (Bowes, et al., 2009; Cashin, et al., 2008; Johnson, 2000; Masterton, 2010; Weng, et al., 2012; Whittingham, et al., 2013; Zhou, Yi, Zhang & Wang, 2014). This is also the case for autosomal recessive conditions whereby hereditary is the mechanism for disease etiology including sickle cell disease, phenylketonuria, Sanfilippo syndrome and thalassemia (Grant, et al., 2013; Weng, et al., 2012). Guilt about the hereditary component of CF is common (Tluczek, Kosciak, Farrell & Rock, 2005), and is a well-established finding of parental psychological effects of a CF diagnosis in an infant (Hodgkinson & Lester, 2002; James, Hadley, Holtzman & Winkelstein, 2006; Jedlicka-Köhler, et al., 1996; McCraea, Culla, Burton & Dodge, 1973; Tropauer, et al., 1970). Feelings of guilt in parents of children diagnosed with CF have been shown to adversely affect parenting self-esteem (Courlet, Dodane & Garnier, 2014) and development of competent parental identity (Hodgkinson & Lester, 2002), which can then contribute to adverse parenting behaviours (Teicher, 1969) and hostile parent-child relationships (Tropauer, et al., 1970). Similarly, parents experiencing guilt for their child's diagnosis in other paediatric chronic conditions has been found (Grant, et al., 2013; Weng, et al., 2012; Zhou, et al., 2014).

Though understood as common amongst parents of children with chronic conditions and disabilities (Bowes, et al., 2009; Damrosch & Perry, 1989; Masterton, 2010, Neilsen, 2013; Nikfarid, Rassouli, Borimnejad & Alavimajd, 2015; Paul, Sheilini & Roach, 2014; Patrick, 2014; Whittingham, et al., 2013), prevalence rates as well as possible effects of chronic sorrow on parents of children with CF and their families have not been sufficiently studied (Moola, 2012; Priddis, et al., 2010; Shumaker, 1994). Referred to in the trauma literature as a sense of recurring, extended and profound sadness (Gravelle, 1997; Olshansky, 1962), chronic sorrow generally occurs because of the profound sense of loss parents feel as a result of their child's diagnosis including; loss of traditional parenting functions and role, loss of the ideal child and family, and loss that the child will experience (Fernández-Alcántara, et al., 2015; Cashin, et al., 2008; Whittingham, et al., 2013). Effects of chronic sorrow on parents of children with CF have not been as extensively studied as effects of guilt on parents, however Shumaker's (1994) doctoral research specifically explored outcomes of chronic sorrow for these mothers. She found high rates of depression and anxiety for mothers experiencing chronic sorrow. More recently, both Priddis et al. (2010) and Moola (2012) made reference to comparable adverse effects of chronic sorrow for parents of children with CF. Therefore, as the landscape of CF diagnosis and treatment continue to evolve, so should the focus of research follow in its footsteps. An example of such a transition could be to understand if the nature of chronic sorrow changes with the nature of earlier diagnosis, improved health outcomes and knowledge of early disease progression.

A concept analysis of the defining characteristics of chronic sorrow by Eakes, Burke and Hainsworth in 1998 resulted in a context-specific theory of chronic sorrow for parents experiencing paediatric chronic conditions. The theoretical model was developed through interviews with parents of chronically ill children. It describes critical times when parents are likely to experience episodes of chronic sorrow by identifying antecedents and trigger events. The model further describes management methods by the person experiencing chronic sorrow and by those offered for therapeutic purposes. Clements, Copeland and Loftus (1989) and Shumaker (1994) qualitatively explored these critical time points in families of children with CF, and found that chronic sorrow was present at all developmental stages and disease-related time points including exacerbations of CF, hospital admission and independence in adolescence. These times brought about recurring and cyclical periods of chronic sorrow for parents. Predicting these critical times enables the health care team to optimise and maximise the effectiveness of treatment based on the conceptual model of chronic sorrow by Eakes and colleagues.

Whilst these studies are dated, they represent the small number of studies paying particular attention to chronic sorrow in parents of children with CF. However, due to changes in CF diagnosis and management over the past 20 years, such findings may bear little relevance to parents today.

For example, current diagnosis occurs much sooner than parents in these studies, potentially commencing the cycle of chronic sorrow much sooner than previously reported. Moreover, mothers were the primary respondents in both studies, representing the research focus at the time. Therefore, it is possible that findings from mothers are not generalisable to fathers who today, are possibly much more involved with their children than previously.

Though considered normal responses to diagnosis of CF in an infant, psychological effects of guilt and chronic sorrow are worthy of close attention because persistent adverse emotions may have significant effects on both child and parent mental health, and ultimately on a child's physical health. Personality, family and other environmental factors influencing feelings of guilt and episodes of chronic sorrow are important to understand. By identifying mediators and moderators of guilt and chronic sorrow, improved support services can be offered to parents and their families at appropriate times, resulting in individualised family support.

2.2.2.3 Parental identity formation and parenting self-efficacy.

There is a dearth of research into parent identity formation, both generally and specifically in relation to paediatric chronic illness. It has primarily been reported as a psychological construct associated with adjustment to parenting (Cast, 2004; McBride & Toller, 2011; O'Connor & Barrera, 2014). Parental identity formation, or effects on parental identity, due to a CF diagnosis has not been specifically addressed; however one study reported parental identity formation in CF in a qualitative exploration of maternal stressors and coping strategies. Hodgkinson and Lester (2002) described how parental identity was negatively influenced by the child's CF diagnosis through a sense of sacrifice of the traditional parent ideology. This sacrifice took several forms; inability to return to work and becoming socially isolated; and adjusting to an identity that incorporated a genetic condition causing a substantial amount of guilt. Coming to terms with being a carrier of condition inherited by a child was part of accepting a changed identity for the mothers in the study.

The process of identity reformation after CF diagnosis may be similar to O'Connor's and Barrera's (2014) identity disintegration, conceptualised as negative perceptions of parenthood and the parental role and identity; and identity reintegration, characterised by positive reframing of parenthood and the parental role and identity in relation to the child's diagnosis. Parental characteristics that might indicate whether a parent may experience beneficial or adverse effects on parent identity are currently not clear because of the lack of clarity about effects of paediatric chronic conditions on parent identity. Moreover, at what point parents start to shift from identity reintegration to disintegration or vice versa is ambiguous. This valuable information could be utilised to determine which parents may be at risk of detrimental outcomes for their parent identity, and on their subsequent self-esteem and general well-being.

Similarly, parenting self-efficacy does not appear to be specifically addressed in the CF literature, but has been reported as a coping resource/strategy (Gibson, 1998). There has been no elaboration on this finding since. For example, relevant investigation could include parent and environmental factors that may be associated with parenting self-efficacy in CF, or what effects different levels of parenting self-efficacy may have on family functioning or on parental self-esteem. Psychological conceptualisation of parenting self-efficacy is a personal belief that one will be able to perform parenting tasks successfully (de Montigny & Lacharité, 2005) and has been shown to be a critical factor in parenting quality and self-esteem (Coleman & Karraker, 1997; Jones & Prinz, 2005). Self-esteem has recently been studied in adults with CF (Platten, Newman & Quayle, 2013), but not with parents. Nor is there investigation into how self-esteem may be tied to parental identity in paediatric CF, though psychological literature about the relationship between identity and self-esteem (Cast, 2004; Cowan & Cowan, 1992) does suggest a relationship within the parental domain of identity.

2.2.3 Parental characteristics moderate child health outcomes.

Improvements in life expectancy have shifted the focus of CF research and clinical care toward the complex interplay among behavioural, psychosocial, and health outcomes for people living with CF (Barker & Quittner, 2010). Some parental outcomes have been identified as risk factors for adverse mental health outcomes in children with CF. For example, reports have demonstrated associations between maladaptive parenting practices and children's psychosocial functioning (Cappelli, McGrath, MacDonald, Katsanis & Lascelles, 1989), and parental depression has been associated with children's quality of life (Brucefors, et al., 2015). One of the most robust findings about parental outcomes affecting child health in CF is that parent and family stress and dysfunction have an adverse effect on CF outcomes. Patterson, McCubbin and Warwick (1990) showed that parental stress, poor parental coping and maladaptive family functioning were associated with poor health outcomes (body mass index and pulmonary functioning) in children with CF. Further study by Patterson and colleagues (1993) established results which strongly suggested that a balanced family coping style, where members tend to their own needs as well as the needs of the child with CF, and low levels of stress, was positively associated with increased health outcomes (pulmonary functioning) over 10 years. Similarly, adaptive coping has been shown to influence the physical and psychological well-being of healthy and CF-affected children (Szyndler, et al., 2005) with positive effects on growth, nutrition and lung function (Patterson, et al., 1993), and respiratory infections (Turner-Cobb & Steptoe, 1998).

Parenting practices and parental factors as contributors to child health and development in paediatric chronic disease are likely to be similar for parents of children with CF. There is also a potential moderating nature of disease effects on child outcomes. For example, parental stress and

depression as a result of CF diagnosis may influence a child's health outcomes through their effects on treatment adherence (Barker & Quittner, 2016; Sheehan, Hiscock, Massie, Jaffe & Hay, 2014). A holistic view of parenting within the family context, inclusive of family functioning and illness perception as direct and indirect contributing factors to child health are recent assertions that follow the shift of focus from individual to family-level research and intervention. Given these findings, there is substantial evidence to support the idea that parenting practices and parental factors contribute to optimal child health and development in the context of CF. Recent conceptualisations of how these relationships may develop have been referred to as biological embedding (Hertzman, 1999) and intergenerational or developmental cascading (Masten & Cicchetti, 2010; Patterson, Forgatch & deGarmo, 2010).

2.2.4 Summary and conclusion.

Increasing life expectancy of children with CF is of paramount importance for clinicians and biomedical researchers alike. However, the psychosocial environment of the child is also recognised as an important factor in the health and well-being of the child, both as a moderator and mediator (Grant, et al., 2006; Robinson, et al., 2008). Therefore, parental psychological well-being is in itself extremely important, and as such, is the primary focus of my research. This section has detailed how a CF diagnosis is experienced by parents and how it becomes assimilated into family life. Most research has focussed on negative aspects of parental experiences of CF. Whilst this research remains necessary, research that investigates all experiences is essential to understand the full spectrum of parental experiences of their child's condition so that negative aspects can be minimised and positive aspects can be enhanced and taught to others. Similarly, although CF is now diagnosed early in life, there is insufficient study of the psychosocial effects of CF on children, their parents and their families in infancy and pre-school. This section has also shown how CF affects many aspects of behavioural, social and emotional development, which in turn influence health outcomes and quality of life. Therefore, it is important to consider relationships among multiple systems (for example, family, peers, health care team), as well as to identify key periods of transition that require extra monitoring and targeted interventions.

2.3 Coping in families with CF

2.3.1 Coping with diagnosis.

Further to parents' experiences of their child's CF diagnosis is the importance of how they cope. Much empirical investigation regarding parental coping with CF has focused on determining factors that influence positive and negative coping, that is, adaptive and maladaptive coping strategies that impede or facilitate adjustment to diagnosis. Research has been conducted across a number of disciplines, such as nursing, psychology and paediatrics, with influential factors revealed at societal, group and individual levels. For example, research has found that societal factors such as

access to medical services can influence coping. Thomas, Mitchell, O'Rourke and Wainwright (2006), for example, found that parents in regional areas had less access to CF-specific medical care than parents living in urban centres, often resulting in feelings of isolation and inadequate support, which negatively influenced coping strategies.

Maladaptive (or dysfunctional) coping strategies are commonly reported by parents following CF diagnosis, including strategies of avoidance and denial (e.g., Sheehan, et al., 2014; Wong & Heriot, 2008). Such maladaptive coping strategies have been associated with low levels of family supportiveness (Thompson, et al., 1992b), family conflict (Szyndler, et al., 2005), and financial strain (Goble, 2004). Though maladaptive coping strategies should be addressed by clinicians and other HCPs, it must be noted that some coping strategies applied on a short-term basis can serve adaptive purposes while the parents and/or family come to terms with the diagnosis. For example, cognitive avoidance and distraction may give parents time to adapt to behavioural aspects of diagnosis and treatment whilst maintaining their psychological well-being (Abbott, 2003). Therefore, consideration should be given to purposes of particular coping strategies at particular times.

Parental adaptive coping strategies reported by parents during their child's CF diagnosis following NBS include problem-solving, seeking social support, searching for information, or not talking with others about their situation until such time that they had accepted the diagnosis and could talk comfortably with others about it (Merelle et al, 2003, Tluczek, et al., 2005). These adaptive coping strategies have been associated with optimism, acceptance, family integration and cohesion, social support and health literacy (Abbott, Dodd, Gee & Webb, 2001; Harrop, 2007; Hodkova, Abbott, Mala & Chladova, 2008; Hymovich & Baker, 1985; Lioffi & Evans, 2005; McCubbin et al., 1983; McCubbin, et al., 1984; Pfeffer, et al., 2003; Venters, 1981).

What is evident is that the majority of research into coping with a CF diagnosis has been conducted with patients and their families prior to the introduction of NBS programs. Whilst some of the aforementioned research considers children diagnosed via NBS and their families, the number of these studies is small and research has only recently been conducted. Therefore, it is possible that psychosocial factors associated with coping identified in older studies prior to NBS no longer bear relevance to families today. Moreover, most aforementioned studies examined maternal coping because the majority of participants were mothers where both parents were involved. This may mean that findings from these studies are not generalisable to fathers who, today, can be central to a child's treatment management practice and family intervention.

Finally, there is little recent information to show whether coping styles and strategies of mothers and fathers of children with CF differ. However, mothers and fathers have been shown to cope differently with children who have other chronic conditions (Al-Yagon, 2015; Compas, et al., 2015; Doherty, et al., 2009; Eiser & Havermans, 1992; Heaman, 1995; Wiedebusch, et al., 2010).

Specifically for parents of children with CF, an early study reported fathers had lower levels of adjustment issues such as depression and anxiety than mothers (Thompson & Gustafson, 1996). However, a recent study reported similar coping strategies between mothers and fathers (Havermans, et al., 2015). To date, these findings have not been fully explored in fathers of children who have been diagnosed with CF following NBS. A local study reported fathers' perspectives of their child's CF diagnosis where coping was mentioned by a number of participants within the study. Fathers commonly reported assuming the protector role within the family, which facilitated their own, and the mother's, ability to cope (Priddis, et al., 2010). Therefore, a holistic family perspective on coping with paediatric CF requires further research into fathers' coping mechanisms, and whether they correlate with maternal coping and adjustment.

2.3.2 Coping with CF in the early years.

Research into parental coping with CF once focussed on coping later in a person's life, usually adolescence or later (e.g., Gayton, et al., 1977; Gibson, 1988; McCubbin, et al., 1983), and coping with acute episodes of illness once disease progression had commenced (e.g., Keller & Nicolls, 1990). Generally, parents of young children with CF no longer need to cope with their child dying in their early years of life. Instead, they need to cope with a demanding home treatment regimen. They also have to cope with an early diagnosis and with early knowledge of their child's covert disease progression. Therefore, generally marked by a period of parent and family stress, an initial period of change and adaptation, both psychologically and behaviourally, is required (Glasscoe & Smith, 2008; Quittner, et al., 1992). During this phase of stabilisation, parents must learn new caregiving tasks and treatment regimens (Quittner, et al., 2009). It is during this period when families will build foundations for how they will cope with the condition during childhood and adolescence (Rolland, 1987), which are important for both current and future management of the disease. Following research trends into parenting children with chronic conditions outlined in the introductory chapter, research into parental coping in the context of paediatric CF initially focussed on psychopathology with the aim of intervening and reducing maladjustment. Venters (1981) was one of the first to report on parental coping with a child's CF. She identified coping strategies that minimised condition-related hardships were statistically significantly associated with long-term adequacy of family functioning.

Over the past 30 years, there has been an abundance of research published on parental and familial coping in CF (Abbott, et al., 2001; Abbott & Gee, 2003; Cappelli, et al., 1988; Cowen, et al., 1986; Coyne, 1997; Hodgkinson & Lester, 2002; Hovey, 2005; Lewis & Khaw, 1982; McCubbin, et al., 1984; Nagy & Ungerer, 1990; Patterson, et al., 1993; Pendleton, Cavalli, Pargament, & Nasr, 2002; Quittner, et al., 1992; Quittner, et al., 1996; Spirito, Stark, Gil, & Tyc, 1995; Staab et al., 1998; Thompson, et al., 1992b; Wong & Heriot, 2008). From these studies, it has been shown that

families generally experience a period of uncertainty and disruption after diagnosis before returning to relatively common and conventional levels of family functioning and adaptability.

Many parental coping strategies have been reported in the literature. Some of the more dominant strategies include maintaining optimism (Abbott, 2009; Abbott, et al., 2001; Bauchner, et al., 1996; Hodgkinson & Lester, 2002; McCubbin, et al., 1984), cognitive restructuring (Hodgkinson & Lester, 2002; Thompson, et al., 1992b), maintaining and/or seeking social and emotional support (Hodgkinson & Lester, 2002; Hymovich & Baker, 1985; McCubbin, et al., 1984; Thompson, et al., 1992b; Wong & Heriot, 2008), seeking medical information from other parents or HCPs (Hovey, 2005; Hymovich & Baker, 1985; McCubbin, et al., 1984; Thompson, et al., 1992b), cognitive avoidance (Abbott, et al., 2001; Sheehan, et al., 2014; Staab, et al., 1998; Wong & Heriot, 2008) and distraction (Abbott, 2009; Abbott, et al., 2001).

A recent Australian study reported moderate correlations of parental depression, anxiety and stress with avoidant coping (Sheehan, et al., 2014). Avoidant coping has three aspects; focusing on venting of emotions, behavioral disengagement, and mental disengagement (Carver, Scheier & Weintraub, 1989). Avoidant coping strategies generally have little or no effect (Roth & Cohen, 1986). Furthermore, avoidant coping was significantly correlated with eating and mealtime problem behaviours in children (Sheehan, et al., 2014). What this has the potential to explain is that parental avoidant coping behaviours can contribute to children's dietary complications, which can result in low nutritional status. Therefore, it appears that parental avoidant coping can influence low nutritional status, which might be mediated by mealtime and eating complications. Collectively, studies investigating parental coping strategies identify both problem and emotion-focussed coping styles and strategies (explained in the next chapter), with emotion-focussed strategies being less effective, which is analogous to the general coping literature (e.g., Li, Cooper, Bradley, Shulman & Livingston, 2012; Regier & Parmelee, 2015; Shin, et al., 2014).

Similar to investigation of parental coping with a CF diagnosis, the majority of research into parental coping with CF in the early years of a child's life has focused on children and their families prior to the introduction of NBS programs. Therefore, it is possible that coping strategies and their effectiveness identified in studies prior to NBS are no longer appropriate reports of parenting children with CF. Moreover, most studies failed to assess the effect of development and lifespan issues on parental experiences, collapsing across child age, generally ranging from 1 to 16 years. Categories of age ranges for the youngest group generally consisted of children from birth to 6 years of age, which could result in unintentional disregard for developmental issues that may impinge on parental coping resources and other important experiences.

One coping strategy distinct to fathers of children with CF has been reported. Fathers appear to have lower self-competency about caring for their child than mothers and this was magnified by

the mother as 'expert.' This feeling of low self-competency was counteracted by mastery of the treatment regimen, which allowed fathers to be involved in the caring process and therefore feel a sense of adequacy (Goldberg, et al., 1990). So although most research is on maternal coping, fathers have been shown to cope differently, which mirrors literature in other chronic diseases (e.g., Barak-Levy & Atzaba-Poria, 2013; Compas, et al., 2015; Tway, Connolly & Novak, 2007). This is of great value to researchers, clinicians and the broader medical and psychological communities because if necessary, tailored intervention strategies for improved parental outcomes may be implemented.

2.3.2.1 Resilience and hope in the face of adversity.

In the past 10 years, research has trended towards a focus on resilience factors as mediators of adjustment and adaptation to CF (e.g., Grosseohme, Szczesniak, Dodd & Oipari-Arrigan, 2014; Mc Hugh, Mc Feeters, Boyda & O'Neill, 2016); Mitsmansgruber, et al., 2015). As such, current coping research investigates factors within a parent's capabilities that reinforce resilience, with the aim of increasing personal and family strength against the illness stressor as well as associated and future stressors. Therefore the thrust of knowledge about resilience in parents of children with CF has been conducted under the auspices of coping research. *Resilience* is a supplementary term related to coping (Fletcher & Sarkar, 2013). Whilst they are overlapping terms in the psychological trauma and loss literature, resilience generally refers to "a dynamic process encompassing positive adaptation within context of significant adversity" (Luther, Cicchetti, & Becker, 2000, pp. 543). Two critical components of resiliency are; exposure to significant threat, and achievement of positive adaptation despite major assaults on the developmental process (Luthar & Zigler, 1991; Masten, Best & Garmezy, 1990). Due to its modern interest by researchers compared to the coping construct, much less literature exists about the resilience construct.

Before discussing current literature on resilience as a psychological construct, hope despite facing adversity is a psychological construct identified as a parental coping strategy contributing to development of resilience for future burden of physical and mental chronic conditions (Barlow & Ellard, 2006; Faso, et al., 2013; Horton & Wallander, 2001; Kashdan, et al., 2002; Lloyd & Hastings, 2009; Ogston, Mackintosh & Myers, 2011; Petersen & Wilkinson, 2015; Samson, et al., 2009). Hope is a motivational factor helping to initiate and sustain action towards goals despite obstacles that might get in the way of goal attainment (Snyder, Rand, & Sigmon, 2002). In the context of parenting a child with a chronic condition, hope has been referred to as a transitional refocusing from a difficult present to a positive future with dynamic possibilities within continual uncertainty (Duggleby, et al., 2010). Therefore, hope exhibits distinct features in intentionality and agency. That is, being hopeful refers to an ability to conceptualise goals, find pathways to these goals despite obstacles and having the motivation to use those pathways (Snyder, et al., 2002). For these reasons,

hope has been associated with not only adaptive coping strategies for preventing and confronting stress, but also a sense of control over achieving one's goals (e.g., Snyder, et al., 1991).

Little research has been conducted on the lived experience of hope among parents of children with a chronic, incurable and potentially fatal condition (Samson, et al., 2009). However, Hodgkinson and Lester (2002), and Gjengedal and colleagues (2003), along with Jessup and Parkinson (2010) have reported hope as a prevalent emotion-focussed coping strategy for parents of children with CF. In these studies, hope manifested in both short and long-term form; short-term hope was for immediate good health for their child and long-term hope was for a cure to the condition. Hope has been shown to predict reduced anxiety and less emotional impact for parents of children with CF (Wong & Heriot, 2008). Hope does not necessarily come so easily for parents; at times parents felt they had to fight to experience hope (Jessup & Parkinson, 2010) or that they were going through the motions because there was no other alternative but to hope (Hodgkinson and Lester, 2002). An implication of these findings is that reduction in anxiety and emotional impact of illness resulting from hope is supportive of developing resilience.

Despite enormous treatment burden and potentially shortened mortality, parents of children with CF are generally resilient, with most reporting a high quality of life and normative levels of psychopathology (Brucefors, et al., 2015; Quittner, et al., 2014; Szyndler, et al., 2005). Resiliency research has primarily emanated from the loss and trauma literature, predominantly focusing on what makes people resilient in the face of personal, economic and environmental adversity (see discussions in Agaibi & Wilson, 2005, and Richardson, 2002). A resiliency approach to understanding outcomes of adversity entails exploring potential for personal and relational growth that can be generated from adversity (Boss, 2001). By tapping into key processes for resilience, individuals and families can emerge stronger and more resourceful to meet future challenges (Walsh, 2003). This approach to understanding the family system and its individual members is consistent with the salutogenic orientation first proposed by Antonovsky in 1987 and more recently by Seligman and Csikszentmihalyi (2000) in the positive psychology movement.

As a result of the transition from a psychopathologic view on paediatric chronic illness, resilience has become an important concept in mental health theory and research over the past two decades (Rolland & Walsh, 2006). Current foci of resilience in context of paediatric chronic conditions is the complex interplay of multiple risk and resistance factors including individual, family and broader socio-cultural influences. From a family perspective, concentrating on strength in the face of adversity has shown that the role of family is nurturance and development, and its function is management and cohesion of individual family members within a synergistic and dynamic system (Patterson, 2002; Walsh, 2003). Within the family system, protective factors have been identified

that build and foster resilience including support and communication, family meaning-making, locus of control and self-efficacy, and effective coping skills (Black & Lobo, 2008).

Support is a resiliency factor for parents managing their child's paediatric condition (Benzies & Mychasiuk, 2009; Peer & Hillman, 2014). Instrumental, social and practical forms of support assist families during times of stress and help them to maintain optimal mental and physical health (O'Leary, 1998; Walsh, 2003; Black & Ford-Gilboe, 2004). Therefore, formal and informal supports buffer parents against negative effects of stress associated with their child's condition (Peer & Hillman, 2014). Communication as a source of support also helps family members to work through and understand emotions and cognitions associated with stressful events (Armstrong, Birnie-Lefcovitch & Ungar, 2005; Finkenauer, Engles, Branje, and Meeus, 2004; Kelly & Emery, 2003). Communication within, and outside of, the family can also help families to make meaning of adverse events.

Additionally, it is well-known that how one appraises an event is a determining factor of how one will cope with the event (Lazarus & Folkman, 1984). Based on this premise, belief systems are vital for family resilience because how a family appraises, and makes meaning of, an event will help determine how they cope. Making meaning of an adverse event can change perspective of how the adversity is viewed. For example, families can develop a sense of coherence (i.e, motivation for the family to stick together) by recasting a crisis as a challenge that is manageable and meaningful to tackle (Antonovsky & Sourani, 1988). Sense of coherence refers to the extent to which families feel confident that the outcomes of a situation will be favourable for them (Hawley, 2000). Similarly, generating a positive outlook in the face of adversity functions to conserve a family's energy and efforts in managing the adversity (McCreary & Dancy, 2004; Orthner, Jones-Sanpei, & Williamson, 2004; Peer & Hillman, 2014; White, Richter, Koeckeritz, Munch & Walter, 2004). Individual factors have also been found to have an important protective function that contributes to family resiliency.

Individual protective factors that contribute to family resilience include locus of control, which refers to an individual's perceptions of where underlying responsibilities for life events resides (Rotter, 1966). A dichotomous system of internal and external loci of control exists to explain where responsibility lies. An internal locus of control orientation is a belief about whether the outcomes of actions are contingent on what a person does, whereas an external locus of control orientation is a belief that events are outside of personal control (Zimbardo, 1985). Those with an internal locus of control exhibit empowerment and agency, and are willing to put in the effort required to make positive changes in the face of adversity (Juby and Rycraft, 2004).

Related to locus of control is self-efficacy, which refers to an individual's judgment of their own ability to succeed in reaching a specific goal (Bandura, 1994), for example, parenting self-efficacy. Being able to act and think independently and confidently are important individual

protective factors (Place, Reynolds, Cousins & O'Neill, 2002). Self-efficacy has been reported as a strong predictor of family health promotion (Black & Ford-Gilboe, 2004). Lastly, effective coping skills of individual family members have a direct impact on the family resiliency process (Benzies & Mychasiuk, 2009). Coping is a response to demands appraised by a person as taxing or exceeding available resources (McCubbin & McCubbin, 1993). For example, effective coping skills have reportedly buffered against financial strain (Lloyd & Rosman, 2005) and mediated the relationship between maternal depression and child behavioural problems (Lee, 2003).

Common pathways to family resilience, such as communication through support and making meaning through appraisal, are important to understand and harness. Similarly, characterising individual protective factors important for family resilience offers a starting point for development of clinical intervention. Therefore, developing a pathway model of individual and family characteristics and processes that lead to resilience would allow characterisation of how families might respond to adversity, as well as development of targeted interventions that can be tailored to family needs. Collectively, the above-mentioned findings demonstrate clinical utility of focussing on family resilience in the context of paediatric chronic illness by; building individual and family strength, searching for commonalities among diverse pathways to resilience, and emphasising usefulness of developing effective coping strategies and positive family-level schemata.

2.3.3 Summary and conclusion.

The available psychosocial research into CF has shown how parents cope with their infant's diagnosis and how families cope with CF and associated family experiences. Various studies report that after initial disruption and adjustment, families continue to function well when a child is diagnosed with CF. These findings contrast with reports of consistent, cyclical negative effects on parents of children with CF such as anxiety, stress, depression and PTSS symptoms, particularly for mothers. Therefore, screening for potential psychosocial issues can assist clinicians to identify and understand barriers that parents and families encounter. Education and enhancing beneficial psychosocial experiences and outcomes for parents can assist them to manage CF within their family and to cope with their child's early surveillance of CF lung disease.

2.4 Aims and significance of this research

2.4.1 What does this study hope to achieve and why is it important?

With the implementation of NBS for CF in Australia, surveillance of disease progression and prophylactic treatment for children and infants with CF has been possible. This has led to the introduction of AREST CF, which conducts research in the detection, prevention and treatment of early lung disease in CF. AREST CF has now been operating in Australia for over 10 years. Whilst there is now some research into the psychological effects of a CF diagnosis via NBS, there is a dearth of research into psychological effects of early surveillance and potential consecutive prophylactic

treatment following a CF diagnosis via NBS, and no research assessing parental psychological effects of the AREST CF program. Although NBS for CF is arguably beneficial, discussion and debate needs to continue to maximise beneficial outcomes for children, their parents and their families. This should include clinicians and other HCPs coming together in subsequent treatment programs following a positive diagnosis of CF via NBS to maximise beneficial outcomes for family members who have a child diagnosed with CF.

As little is known about parents whose children undergo early surveillance for CF lung disease, the purpose of my research is to explore the lived experience of parenting in the context of early surveillance for CF lung disease and to describe how parents cope with their experiences. Moreover, using a strength-based approach to understanding parenting practices and parental characteristics in the current context will identify protective factors that can be enhanced for those possessing them and educated to others who don't. Lastly, exploring how early surveillance affects parental constructions of CF can distinguish unique parental outcomes that result from such a program, informing institutions of how such a program is interpreted by parents. Exploring how parents construct their experiences and attribute meaning to early surveillance, and to their child's condition in the context of early surveillance, will lay a foundation from which necessary clinical intervention can be developed that comprises a family perspective. Therefore this research may guide future clinical interventions aimed at improving mental health and well-being for parents managing their child's chronic condition in the context of early surveillance and facilitate further holistic research that includes all family members who are affected by a child's chronic condition.

2.4.2 How can this research help families whose children undergo early CF disease surveillance?

Although there may be some parallels between day to day experiences and coping with CF and with early surveillance, several clinical components of early surveillance may elicit different parental responses and require additional coping strategies and skills that have yet to be understood. Insights from related fields also indicate that anxiety, stress, and depression are the most commonly reported outcomes for parents with a chronically ill child. Therefore, it is important to investigate whether these negative experiences and mental health morbidities occur for parents of children engaged in early surveillance. By identifying if parents in the current context experience additional negative outcomes as a result of their child's early surveillance, it is imperative that AREST CF ensures parents are supported. As AREST CF is a unique research and clinical program, we must ensure that unique experiences and outcomes are explored and if need be, intervened. Of equal importance is ensuring that beneficial outcomes are understood so that parents' lives can be optimally enhanced. My research will have a specific focus on coping and adaptation as these are dominant topics of research in CF with plenty of published studies, but none examining coping with

CF in the context of early surveillance. Improved psychosocial outcomes for CF patients and their families can lead to improved physiological outcomes for the patient, and long-term beneficial outcomes for the entire family.

Therefore, whilst early surveillance informs parents and HCPs of the covert changes in lungs that are not detectable through conventional methods, understanding what parents experience, how they cope, and exploring psychosocial factors associated with their experiences and coping styles is essential to inform CF centres and researchers considering adopting this program. There is also a need to inform intervention strategies that aim to support the family and foster healthy coping mechanisms. As it is not yet known which psychosocial factors in a child's environment influence coping and other parental experiences during early surveillance, the findings should assist with identifying risk and protective factors for negative psychosocial outcomes and resiliency. Therefore, my research outcomes will guide clinical screening for known psychosocial and familial risk factors, and facilitate building family resiliency for future burden of CF, hence guiding the design of individualistic support for families.

2.4.3 What else can this research add to the body of knowledge?

My research contributes to a growing body of knowledge about parenting children with chronic conditions. Uniquely, findings from this research offer insight into the lived experience of parenting children with CF in the context of early surveillance; with this information assisting in understanding holistic needs of parents, particularly those in Western Australia and Victoria. In practical terms, identification of psychological factors important in maintaining optimal functioning and quality of life, and insight into constructions of experience and attributions of meaning given to CF in the context of early surveillance, may be used to inform existing and future policy decision-making models for best clinical practice towards promoting mental health and well-being. Accordingly, this research informs existing and future policy structures through identification of previously omitted information or misconceived assumptions regarding parenting a child undergoing early surveillance and the issues facing them. Moreover, my research contributes to understandings of parenting children with chronic conditions as a process of normalisation, management, resilience and reconstructing future optimistically through hope rather than focussed solely on deficits typically associated with parenting children with chronic conditions. Foremost, the findings contribute to building a holistic understanding of the parenting experience and provide evidence that parenting a child with CF in the context of early surveillance for lung disease has both adverse and beneficial outcomes that are amenable to modification. Consequently, this research; is entirely original, is based on sound theoretical and empirical concepts, contributes new knowledge relevant to the global CF community, and provides a foundation for new clinical pathways that support

patients and families. The next chapter presents theoretical underpinnings and methodological decisions for conducting the research.

CHAPTER 3**Substantiative Theoretical and Methodological Derivatives**

This chapter outlines the rationale for theoretical and methodological choices underlying the process and implementation of the research. Firstly, theoretical models chosen through which to view the findings are reviewed in reference to how they have informed the research by outlining family resiliency, adaptation, and stress and coping approaches. Secondly, chosen theoretical models are critiqued as replacement of the once dominant psychopathological view to parenting a child with CF. This leads to an introduction of the research design and the qualitative methodology used in this study. Specifically, phenomenological inquiry is reviewed in relation to how it informed the research. Methodological justifications are further described for data collection and sampling procedures. The chapter concludes by substantiating the thematic data analytic technique.

He who loves practice without theory is like the sailor who boards ship without a rudder and compass and never knows where he may cast...Leonardo da Vinci

3.1 Introduction

Viewing research findings through theoretical lenses allows a broad application of the findings beyond their current context, as well as providing an opportunity to support or refute a model's components within the findings' current context. Accordingly, this chapter initially outlines the rationale for chosen theoretical models through which to view my research findings. Following that is a description of the grounds for methodological decisions made to implement the study. Presenting substantiative evidence of the theoretical and methodological choices allows the reader to determine authenticity and appropriateness of how the study was conducted prior to learning about the operational aspects. The theoretical decisions are primarily based on literature presented in the previous chapter, whereas methodological decisions were based on the nature of the study.

Given the observation that the majority of families faced with care of a child with CF do not appear dysfunctional and are, in fact, coping successfully with a psychologically and logistically challenging situation, a contextual stress and coping conceptual framework has been developed (McCubbin & McCubbin, 1987; 1993). The framework was initially developed with families of children with CF, and provides a set of lenses through which to view these families. The shift in focus purely from parental psychopathology to beneficial psychological outcomes is exemplified by a model commonly cited to explain relationships among adaptive behaviours, family support, parenting stress and coping in the context of chronic illness. The Resiliency Model of Stress, Family Adjustment and Adaptation (McCubbin & McCubbin, 1987; 1993) arose from the notion that families not only react to stress, but they attempt to navigate their way through stresses to achieve adjustment, and eventually adaptation. Theoretical underpinnings of the Resiliency Model are grounded in Lazarus' and Folkman's (1984) Transactional Model of Stress and Coping whereby three variables are present in a situation that determines whether a crisis is created or not; the hardship of the situation; resources required to meet the situation; and appraisal of the event as either neutral or threatening. Lazarus' and Folkman's theory is a theoretical structure through which to explore and investigate cognitive appraisal of a stressful event and activation of coping resources as a response, which lays the foundation for how the Resiliency Model accounts for parental outcomes as a result of managing a chronic condition within the family environment.

3.2 Theoretical frameworks

3.2.1 Transactional Model of Stress and Coping.

Lazarus' and Folkman's Transactional Model of Stress and Coping (1984) has led to understanding and explaining stress and coping within the health sciences. Their cognitive model has defined coping as thoughts and behaviours people use to manage internal and external demands of

situations that are appraised as stressful. The coping process is initiated in response to an individual's appraisal that important goals have been harmed, threatened or lost. These appraisals are initially characterised by negative emotions and are therefore initiated in an emotional environment. One of the first coping tasks is therefore, to down-regulate (i.e., reduce) negative emotions that are stressful and may interfere with instrumental (i.e., behavioural) forms of coping. Emotions continue to be fundamental to the coping process throughout a stressful event or situation as an outcome of coping, as a response to new information, and as a result of reappraisals of the status of the stressful event or situation. If the stressful event or situation has a successful resolution, positive emotions will ensue; if the resolution is unfavourable or unclear, negative emotions will likely ensue (Folkman & Moskowitz, 2004).

According to Lazarus (1966), the individual difference of most relevance to the study of human behaviour and stress is the cognitive appraisal of stressors. His early theory placed attention on the role of cognitive appraisal in producing the quality of the individual's emotional response to a stressful encounter or event, and the ways in which a person coped with the appraised encounter or event. Lazarus believed that after appraising the stressor, an individual will use one or more coping strategies in an attempt to adjust to the situation. Similarly, which coping responses emerge is partly determined by an individual's knowledge of coping options and partly by their beliefs about the usefulness of those options. Both stress and coping then, emanate partly from the mental sets brought by the person to the event (Parker & Endler, 1996). Cognitive appraisal then became a critical aspect of stress research methodology and theory that respected individual differences in the nature and degree of stressful experiences (Crnic & Low, 2002). Cognitive appraisals refer to direct and immediate evaluations about the individual's environment with reference to their well-being. They act as evaluative frameworks that individuals utilise to make sense of events (Lazarus & Folkman, 1984). Emphasis on cognitive appraisal as a contributor to psychological outcomes has continued into the context of parenting stress (Deater-Deckard, 1998).

Two main coping dimensions studied in early literature about coping, and that have since received much research attention, were emotion-focussed coping and problem-focussed coping. In summary of the sizable literature in this area, problem-focussed coping involves strategies that attempt to solve, reconceptualise, or minimise the effects of stressful events. On the other hand, emotion-focussed coping includes strategies that involve self-preoccupation, fantasy, or other conscious activities related to emotion regulation (Parker & Endler, 1996). Avoidance-oriented coping has also been identified as another basic dimension that has received much research attention, and has been conceptualised as involving person-oriented and/or task-oriented responses. An individual can react to a stressful event by seeking out other people (i.e., social diversion) or can engage in a substitute task (i.e., distraction).

Research into coping has demonstrated some consistent findings. Well-established findings include both dispositional and situational attributes heavily influence coping behaviours (Bouchard, Guillemette & Landry-Léger, 2004; Carver & Scheier, 1994; Panayiotou, Kokkinos & Kapsou, 2014). Coping is strongly associated with the regulation of emotion (especially distress) (Webb, Miles & Sheeran, 2012). Some coping strategies (e.g, avoidance coping strategies) are consistently associated with poor mental health outcomes (Dardas & Ahmad, 2015; Meier, Carr, Currier & Neimeyer, 2013). Collectively, these findings suggest that individuals who are flexible in their choice of coping strategies should show better adaptation than individuals who have a more restricted coping repertoire.

With convergent evidence about the applicability of coping research being produced, research has considered beneficial outcomes of stress and coping. Some research has considered positive outcomes of successfully resolving a stressful encounter (e.g., Folkman & Lazarus, 1985), including outcomes such as acquisition of new coping skills and resources (Schaefer & Coleman, 1992), and the perception of growth related to the stress (Holahan & Moos, 1990). Other research has considered the frequency of positive affect during stress (e.g., Viney, 1986; Folkman, 1997), and found that positive emotion does in fact co-occur with negative emotion related to stress.

Since early conceptualisations of coping, a proliferation of research has developed across behavioural and social sciences, medicine, public health and nursing (Folkman & Moskowitz, 2004). Current understandings of coping include a multi-dimensional process that is sensitive both to the environment, and its demands and resources, and to personality dispositions that influence the appraisal of stress and resources for coping (Sommerfield & McCrae, 2000). Coping research in specialised areas, such as parenting, is the result of an attempt to try and combat some of the issues raised in general coping research and to make further generalisations about a particular population, a particular stressful encounter or event, or a particular population encountering a particular stressful situation or event.

3.2.2 The Resiliency Model of Family Stress, Adjustment and Adaptation.

The shift in focus from psychopathology to adaptive psychological outcomes of a paediatric chronic condition, particularly from a family perspective, resulted in a conceptual model to understanding relationships among adaptive behaviours, family support, parenting stress and coping. Moreover, if one accepts the basic observation that most families faced with care of a child with CF do not appear dysfunctional and are, indeed, coping successfully with a difficult situation, a resiliency framework can provide another set of lenses through which to view these families. The Family Resiliency Model of Stress, Adjustment and Adaptation (McCubbin & McCubbin, 1993) has had a number of iterations, beginning with a strong focus on psychopathology within the ABC-X model of family behaviour (Hill, 1949; 1958) and later, the Double ABC-X model of family behaviour

(McCubbin & Patterson, 1983). In the early model, *A* referred to the crisis-precipitating event, *B* referred to the family's crisis-meeting resources and *C* referred to the definition the family makes of the event via their appraisal mechanism (Hill, 1958). A shift in emphasis from crisis to transition resulted in the Resiliency Model's emphasis on adaptation as the most often needed response to a chronic condition within a family context.

The current model identifies essential interacting components of family life and explains, as well as predicts, family processes and outcomes of managing a paediatric chronic condition. The model outlines how these outcomes can vary along a continuum of extremely positive (*bonadjustment* and *bonadaptation*) to the more negative extreme of maladjustment and maladaptation. The length of time one has a chronic condition means that one's family must attempt to adjust immediately, and to adapt to the condition on a more permanent basis over time. Therefore, the model consists of two phases. The first phase is an adjustment phase, characterised by relatively minor changes in the family system, and is a predictable phase in the family's response to a stressor such as an illness. The second phase is an adaptation phase, a slower, more disruptive and resistant type of adjustment. Each phase has inter-related components combining to produce positive or negative outcomes. The model attempts to explain the processes families must negotiate and the objectives that must be accomplished to achieve *bonadaptation*. Bonadaptation refers to a family's ability to meet the needs of each individual family member, enabling them to achieve their maximum potential as well as the functioning of the family system and its transactions with the community (McCubbin & McCubbin, 1993). Accordingly, the model serves as a framework for determining whether or not a health or illness stressor becomes a crisis. See the following page for a diagram of the constructs, processes and relationships that comprise The Resiliency Model of Family Stress, Adjustment and Adaptation.

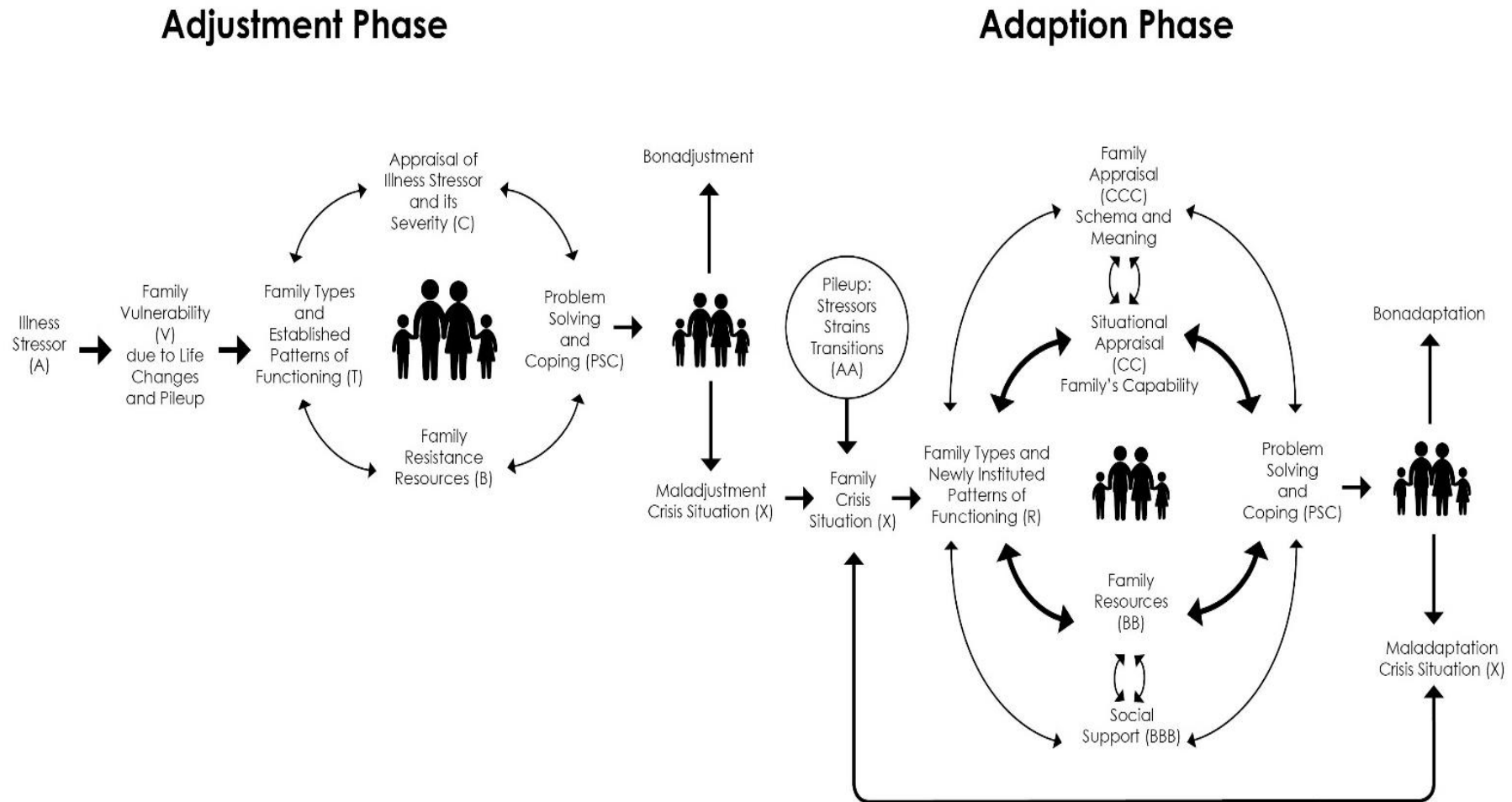


Figure 1. The Resiliency Model of Family Stress, Adjustment and Adaptation. Adapted from "Families, Health and Illness: Perspective on Coping and Intervention," by H. I. McCubbin and M. A. McCubbin, 1993, p. 23. Copyright 1993 by Mosby.

The Resiliency Model (McCubbin & McCubbin, 1993) has extracted theoretical underpinnings from a number of other models attempting to explain similar phenomena from a more generalised perspective, that is, within non-context specific events or circumstances. Firstly, a resiliency approach to family health is consistent with the broad biopsychosocial model (Engel, 1977) in that it theorises that health is the result of multiple and interacting influences, including risk in the environment and protective processes available to the family. Secondly, the impact of illness on family coping skills is explored (including successful and unsuccessful coping characteristics). It highlights the reciprocal relationship between physical and mental health and family functioning, thereby supporting Family Systems theory (Bowen, 1978) underpinning the model for understanding stress and the impact of illness on the family. Lastly, Lazarus' & Folkman's (1984) Stress and Coping theory's concept of appraisal underpins the evaluation (i.e., appraisal) of, and subsequent response to, the family stressor of paediatric chronic illness demonstrated in the Resiliency Model's individual level and family schema level of appraisal. The Resiliency Model is therefore a comprehensive and context-specific theoretical model relevant for guided investigation of familial coping and adaptation to paediatric chronic illness - and specifically for the purpose of my thesis, isolating parental coping and adaptation to paediatric CF.

Notwithstanding the model's strengths in exploring parenting and family functioning in the context of paediatric chronic illness, theoretical models nonetheless possess areas for improvement or empirical support for their justification. The Resiliency Model (McCubbin & McCubbin, 1993) posits that the ultimate key to determining the direction of a family's response to an illness stressor is the family's situational appraisal that governs, if any, of the other illness dimensions are problematic to a family. Individual-level appraisal is well-documented, however empirical investigations of family-level appraisal seem to be limited, and could therefore benefit from further investigation into the applicability of this dimension. Family-level analysis is invaluable to understand and support family functioning but it fails to identify any nuances between family members and within individual family members that function either synergistically or antagonistically within the family. Therefore, whilst the model is adept at identifying family-level factors important for developing and maintaining resilient families, individual and dyadic factors associated with resilience should be of primary consideration before embarking on a family-level analysis. Moreover, whilst the Resiliency Model was developed with data collected from families with a young child with CF, the model is primarily theoretical in its conceptualisations. Research has supported its theoretical use (DeHaan, Hawley & Deal, 2013; Hawley, 2000; Simon, Murphy & Smith, 2005), and research demonstrating its clinical application is still in its infancy (Frain, et al., 2007; Gardner, Huber, Steiner, Vazquez & Savage, 2008; Paynter, Riley, Beamish, Davies & Milford, 2013; Tak & McCubbin, 2002).

Within the scope of my thesis, concepts within the Resiliency Model (McCubbin & McCubbin, 1993) may begin to indicate how parental processes can influence, and be influenced by, systemic, family and personal factors. Parental processes affect parental coping of, and ultimately adaptation to, CF specifically, and paediatric chronic illness more generally. Whilst, according to the model, there are many factors that contribute to family adaptation (as seen in Figure 1), processes and outcomes specific to parents' ability to cope with (i.e., adapt to) their child's chronic condition will be the topic of the remainder of the current discussion.

A well-known study by Patterson (2002) investigated families of children with CF and emphasised two key features of family life that serve as protective factors under challenging conditions, both of which are inherent in the Resiliency Model. The first key feature is family-meaning making, or the family's creation of their world-view. Families vary considerably in their interpretations of health and illness, and develop strategies according to their beliefs. Therefore, the family belief system can influence how it manages daily demands and response to the accumulation of stressors over time. That is, the appraisal or meaning of an event can promote healthy adaptation to stressful conditions. The influence of Lazarus' and Folkman's (1984) work underpins this process. Confirmation of appraisal and meaning-making as contributing factors to how a family adapts to CF is work done by Baldacchino, Borg, Muscat & Sturgeon (2012) who showed that appraisal of, and meaning attributed to, a child's CF influenced the way parents chose to cope with it. They identified that the more positive the parents' appraisal of their child's condition, the more adaptive their coping mechanisms.

The other key feature of the Resiliency Model (McCubbin & McCubbin, 1993) involves establishing newly instituted patterns of family functioning. Adaptive patterns of family functioning promote healthy behaviours by providing an organisational structure and predictability to family life, and provide an element of planning ahead and role assignment. The outcomes of adaptive family functioning include family cohesion, understanding and acceptance of individual family roles, and development of self-efficacy in maintaining adaptive functioning (Ciao, Accurso, Fitzsimmons-Craft, Lock & Le Grange, 2015; Crerand, et al., 2015; Tramonti, et al., 2015).

One such adaptive pattern attributed to effective parental coping and increasing family stability is *normalisation* of the chronic condition into family life. Normalisation of the disease within the family is a prevalent coping mechanism for families of children with CF (Gibson, 1988, Glasscoe & Smith, 2011; Hodgkinson & Lester, 2002). Normalisation has also been expressed as *new normal* whereby some patients created a new sense of normality, balancing life before and after being diagnosed (Liu, Inkpen, & Pratt, 2015).

Research into parental coping with the early CF management has reported adaptive strategies such as establishing a routine that incorporates treatment into daily life, allowing the

family to achieve a sense of normality (Bluebond-Langner, 1996; Glascoe & Smith, 2011; Grosseohme, et al., 2014; Hodgkinson & Lester, 2002; Jessup & Parkinson, 2010; Pfeffer, et al., 2003). Applying the Resiliency Model (McCubbin & McCubbin, 1993) is a purposeful approach to understand how families establish CF management routines, and to identify mechanisms and outcomes of family routines. The Resiliency Model has established links between routines and other family processes, which can be used as a framework to understand outcomes of family routines.

Though establishing family routines have been associated with improved adherence and health outcomes in other disease populations (e.g., Fiese & Wamboldt, 2003), routines in families of very young children with CF have not been comprehensively examined. A recent study sought to explore parental experiences of families with young children in developing and utilising CF care routines within the family setting (Grosseohme, Filigno & Bishop, 2014). Phenomenological methodology allowed themes to emerge describing how families of children with CF aged 3 months to 13 years utilised routines to manage CF care. How families used support structures to facilitate maintenance of routines, and how families encountered, and overcame, challenges with maintaining care routines also emerged.

Along with family resources, and based on meaning-making through appraisal and newly instituted patterns of family functioning, how families problem solve and cope will then determine their level of adaptation to their child's chronic condition. Unfortunately, many studies of children with CF and their families include participants across large, undifferentiated age ranges of children. Whilst this increases generalisability of the study, it undermines both child and family developmental processes and achievements. Therefore, it would be interesting to know at what point of the child's life that families generally establish such routines so that developmentally-appropriate guidance can be offered to families that facilitate use of routines and improve CF management.

Despite the abundance of literature into parental coping with CF in the 1980s, less research has recently evolved in the literature. Movement from traditional psychopathological views of how CF is generally assimilated into the family to approaching family adaptation to CF from a resilience perspective has been well-received. However, technological and medical advancements in CF continue at a rapid pace, which may mean that techniques and interventions for effective CF management (such as those offered by AREST CF) and psychological well-being of the patient and their family may not bear relevance today.

3.2.3 Summary and conclusion.

Theoretical models provide a structural guide for exploring and identifying variables of interest and their potential associated relationships and outcomes. Commonly-cited theoretical models applicable to the empirical literature presented in the preceding chapter are The

Transactional Model of Stress and Coping (Lazarus & Folkman, 1984) and The Resiliency Model of Stress, Adjustment and Adaptation (McCubbin & McCubbin, 1993).

The Transactional Model of Stress and Coping (Lazarus & Folkman, 1984) is a commonly-cited model for understanding how stress and coping are mediated by cognitive appraisal of an event. The model emphasises how appraisal of an event will determine how the event is psychologically managed, and whether coping resources are required to manage the event. For parents experiencing paediatric chronic illness, appraisals of a child's conditions set in motion any number of emotion-focussed and problem-focussed coping strategies. How parents cope and what strategies and styles of coping they use are important explorative components of my research. Consequently, providing comprehensive discussion about the Stress and Coping Model has enabled a detailed description of specific aspects of the Resiliency Model to which my thesis pays particular attention. Within the description of the Stress and Coping Model, the framework of appraisals and coping strategies may be relevant to findings of coping mechanisms identified in my thesis.

The Resiliency Model of Stress, Adjustment and Adaptation (McCubbin & McCubbin, 1993) is a theoretical model developed with families who have a child with CF. The Resiliency Model represents the shift both from an individual and psychopathological view of parenting a child with a chronic condition to a holistic, strength-based approach to understanding how parents manage their child's chronic condition. The framework emphasises resilient properties of families, along with their strengths and capabilities to manage their challenging situation (McCubbin, et al., 1996). Exploring how parents and families manage paediatric chronic conditions from a resiliency perspective affords an opportunity to understand how concepts and their relationships might contribute to development of cognitive constructions and attributions of meaning that might influence coping and adaptation. More importantly, because of its emphasis on adaptation to chronic illness, the most desired response for families experiencing chronic illness, the model's concepts and relationships bear relevance to my research questions and objectives. Due to its focus on family processes within the context of chronic illness and its potential to isolate parental experiences and outcomes, The Resiliency Model has been chosen as the structure through which parental processes and constructs identified in my research findings are viewed and interpreted. Discussion will now shift focus to substantiating paradigmatic and methodological choices for conducting my research

3.3 Study design

3.3.1 Conceptualising qualitative research.

Methodology refers to the way issues are approached and how research is conducted according to pre-determined assumptions that shape the methods chosen to answer questions relevant to the identified issue (Gelo Braakmann & Benetka, 2008; Taylor & Bogdan, 1998). Within psychology methodology, traditionally there are two distinct approaches about which there has

been much debate regarding the criteria for knowledge and the subsequent decisions in reference to appropriate methodological decisions (Corbin & Strauss, 2008; Gelo et al., 2008; Majima & Moore, 2009). Firstly, quantitative research refers primarily to positivist claims for developing knowledge whereby strategies of inquiry include experiments and surveys using predetermined instruments, which yield statistical data. Secondly, a qualitative approach to research refers primarily to constructivist perspectives utilising strategies of inquiry such as narratives and phenomenology to collect open-ended, emerging data with the primary intent of developing themes from the data (Creswell, 2014). Therefore, despite the debate surrounding the decisions and use of quantitative or qualitative methodology, it is now widely accepted that the decision around methodological preference should be based on adopting a method appropriate to investigate the research question (Chamberlain, 2000; Corbin & Strauss, 2008; Gelo et al., 2008; Grbich, 2007; Liamputtong, 2009; Vlasiner, 2006).

A simplistic explanation of the primary difference between qualitative and quantitative research is that studies employing a qualitative form of methodology rely on linguistic rather than numerical data, and adopt meaning-based rather than statistical forms of data analysis (Elliott & Timulak, 2005). Other distinctive features of qualitative research include: an attention to understanding phenomena in their own right (as opposed to an outside perspective); open and exploratory research questions (versus closed-ended hypotheses); unlimited and emergent descriptive options (rather than predetermined choices of rating scales); use of specialised strategies for strengthening credibility of design and analyses; and definition of success in terms of discovering something new (versus confirming what was hypothesised) (Elliott, Fischer, & Rennie, 1999).

Qualitative research is inquiry aimed at describing and clarifying human experience as it appears in people's lives (Polkinghorne, 2005). Qualitative methodology provides an essential quality of data through well-grounded, rich descriptions and explanations of processes occurring within populations (Miles & Huberman, 1994). Techniques such as interviews allow researchers to acquire knowledge and an understanding of issues for a small sample of participants in much more depth (Patton, 2002). This methodology is the most appropriate when the researcher is attempting to understand unexplored complex values, emotions or ideas. Due to the exploratory nature and purpose of qualitative research, data analysis is an inductive (rather than deductive) process, and exploratory rather than confirmatory. The qualitative data analysis process is discussed in detail further in the chapter.

The qualitative research approach has its roots in both cultural anthropology and American sociology (Kirk & Miller, 1986). The intention is to understand a particular situation, event, role, group or interaction through an investigative process whereby the researcher makes sense of a social phenomenon by contrasting, comparing, replicating, cataloguing and classifying the object of

investigation (Miles & Huberman, 1994). This intense investigation requires immersion in the everyday life of the population chosen for the study. The researcher enters the participants' worlds and through ongoing interaction, seeks participants' perspectives and meanings (Marshall & Rossman, 2010).

Qualitative research has traditionally been used in research disciplines such as anthropology, sociology and psychology, but rigorous qualitative research methods over the past 20 years have resulted in a surge of this type of research in social science and increasingly in the health sciences, particularly nursing (Elliott, et al., 1999; Polit & Beck, 2014). Historically, the definition of qualitative methodology has evolved over time (Creswell, 2007; Denzin & Lincoln, 1994, 2000, 2005) to include an emphasis on the *process* of research, that is, as continuing from philosophical and theoretical assumptions (Creswell, 2007; Denzin & Lincoln, 2008).

Most qualitative research assumes that to truly and holistically understand people's experiences, researchers must attempt to understand the meanings and interpretations that people give to their experiences. That is, to "uncover the essential structure of reality" (Crotty, 1996, pp. 68). This means then that qualitative methods provide an insight into how people make sense of their experience that cannot be easily provided by other methods (Liamputtong & Ezzy, 2005).

Typically, qualitative research has a deep involvement in exploratory questions, and is suitable as the basis for inquiry when existing research in an area is contradictory or confusing, when the topic under investigation is complex (Elliot & Timulak, 2005) or when there is a lack of research in a particular area (McDonald & Daly, 1992). It has also been suggested by Holman (1993) that qualitative research is especially insightful, and can greatly contribute to the study of chronic illness because, as he suggests, patients' views are "better indicators of outcomes than common medical measures" (p. 33).

Therefore, to appropriately address these types of issues, open-ended research questions that guide participants and direct the research as data are derived from their own lived experiences are necessary (Corbin & Strauss, 2008). This means that the explorative nature of qualitative inquiry is particularly useful for gaining a holistic understanding of experiences, with emphasis placed on the understanding of relationships within systems and the on the understanding of processes involved in meaning making (Corbin & Strauss, 2008; Creswell, 2007; Janesick, 2000). Qualitative inquiry allows the researcher to create the *essence* of the experience to determine how meanings of the phenomena are formed (Denzin & Lincoln, 2008). Phenomena are described by participants and studied by the researcher (Crotty, 1996). A qualitative methodology is supportive of the philosophical position of a phenomenological and constructivist approach to research, which will be explained.

The focal point of qualitative research is gaining an in-depth and comprehensive understanding of particular individuals or groups, rather than investigating the general characteristics of a large population of individuals across particular variables (Polgar & Thomas, 2008). In contrast to the linear progression of tasks in quantitative research, qualitative research is more reiterative with the progression of the research following more of a circular motion. This refers to the flexible and reiterative construction of, and actions within, the study, inclusive of tasks such as sampling, collecting, analysing and interpreting data (Polit & Beck, 2014).

In qualitative research, no attempt is made by the researcher to control for extraneous variables, and operational definitions are not provided for the study variables. Rather, human experiences under investigation are perceived and described as a whole within their particular context (Polgar & Thomas, 2008). Having chosen the type of qualitative research to be conducted, specific strategies of inquiry exist that will guide subsequent research processes including data collection, analysis and writing (Creswell, 2003).

The collection of data usually involves speaking with those who have first-hand experience and generally consists of loose discussions between the researcher and the participant about the phenomena under investigation. Loosely structured discussions allow participants to express a range of values, beliefs, emotion and behaviours associated with the phenomena under investigation. Data analysis and interpretation are concurrent activities that consist of clustering related narrative information into a coherent format. Through a process of inductive reasoning, the researcher begins to identify themes that are used to build a rich description of the phenomena (Polit & Beck, 2014).

Therefore, qualitative research adopts an interpretative, holistic approach to the phenomena under investigation and involves making sense of the subject matter through interpreting meanings that participants give to them (Denzin & Lincoln, 2008; Patton 2002). Relevant to my research, it has been suggested that qualitative research assists to shape and give life to clinicians', nurses' and other allied health professionals' perceptions of issues or situations, their conceptualisations of potential solutions, and their understanding of patients' and families' concerns and experiences (Polit & Beck, 2014).

3.3.1.1 Methodological and theoretical perspectives and implications.

Accepted paradigms within qualitative methodology include post-positivism, critical theory and interpretivism (Denzin & Lincoln, 2003; Wills, 2007). A distinction between the two methodological approaches of quantitative and qualitative research is the interpretative nature of the qualitative research process. Consideration of interpretation is apparent throughout the entire research process, from acknowledging the researcher's own historical context at each stage of the research to defining outcomes through an interpretative process of co-construction of reality between researcher and participants.

Therefore, an interpretivist paradigm has the assumptions of a constructivist epistemology. Constructions of knowledge are conditioned by experience and culture and are therefore context and perception driven (Wills, 2007), and understanding of the social world is achieved through exploring the interpretation of that world by its participants (Bryman, 2012). Accordingly, the knowledge of reality is not available solely through direct experience, rather it is thinking and reflecting that are important facets of the process of knowing and understanding (Wills, 2007). This concept and its implications will be discussed in detail in the next chapter.

The broad notion of qualitative methodology is further broken down to specific types of methodology, for example, grounded theory, ethnography, discourse analysis, heuristic inquiry and phenomenology (Crotty, 1998; Elliott & Timulak, 2005). Concerning the decision of appropriate methodological frameworks and theoretical perspectives in view of the world as interactions between people and people, and people and meaning, there is a requirement of multiple perspectives on issues and acknowledgement of human experience in broader events situated in a social, cultural and political framework (Corbin & Strauss, 2008; Gergen 1985). To reveal the complexity and layering of the phenomenon under investigation in my research, an underlying theoretical framework informed this research, namely phenomenological inquiry. The theoretical foundation of this framework is discussed in detail in the next section with regards to how the position informed and directed my research.

3.3.2 Qualitative methods.

A qualitative approach to inquiry was used in my research as it represented the most appropriate way to investigate the overarching research questions (Corbin & Strauss, 2008; Liamputtong & Ezzy, 2005). When little or no previous knowledge has been generated about a particular topic or within a particular population, and one is trying to understand intrinsic experiences of individuals to learn how meaning is constructed, a qualitative approach is appropriate (Creswell, 2007; Liamputtong, 2009). Furthermore, a phenomenological methodological approach is supportive of the philosophical position of constructivism within the setting of interest.

Phenomenological methodology utilises thick descriptions of a lived experience to explain different phenomena (Creswell, Hanson, Plano & Morales, 2007; Martin, 2000). Data can be collected from a variety of empirical materials (for example, interviews), which describe moments and meanings in people's lives with a range of interconnected, interpretive processes used to acquire a deeper understanding of the phenomena under investigation (Denzin & Lincoln, 2008). The result of this is rich contextualised descriptions of categories that provide an in-depth understanding of the concepts of interest, within the context in which they occur (Liamputtong & Ezzy, 2005). These rich descriptions can help to explain why a particular phenomenon occurs rather than simply learning what other variables relate to the phenomena or how frequently it occurs.

Common characteristics of a qualitative methodological design, and more specifically phenomenology, include: the use of a natural setting for data collection (that is, for example, an interview conducted in the participant's home); acknowledgment of the researcher as a key instrument in the research process (that is, researcher develops the interview schedule, collects and interprets the data); and the use of inductive data analysis techniques (that is, a reiterative process to build themes). An inductive design is advantageous when little or no prior research has been conducted, or when there is a trend towards using quantitative methodology, because it creates an opportunity to develop new theory and be open to participant meanings. Utilisation of inductive design means that the researcher is not tightly prescribed to research processes, and this allows alternative ideas to emerge throughout the research that may be different from those the researcher brings to the process (Creswell, 2007; Liamputtong & Ezzy, 2005; Miles & Huberman, 1994; Silverman, 2004). By using interviews as a vehicle for data collection, the researcher can reach areas of reality that would otherwise remain inaccessible through quantitative investigation, such as people's subjective attitudes and experiences (Perakyla & Ruusuvoori, 2011). Analyses of data are then presented based on participants' perspectives as well as on the researcher's own interpretations (Creswell, 2007).

A key criticism of qualitative research is that it does not account for considerations of reliability and validity, and provides no account of how data were analysed or interpreted (Johnson & Onwuegbuzie, 2004). However, this criticism has been addressed in my research by adhering to strategies of rigour suggested by Guba and Lincoln (1994). The next section will outline Guba and Lincoln strategies used to achieve scientific rigour. The next chapter will elaborate on how these strategies were achieved in my research.

3.3.2.1 Scientific rigour.

The quality of all types of research depends on the quality of the methods used (Mays & Pope, 2000). It has been argued that evaluation of quality in qualitative research by the same measures and criteria used to assess the quality of quantitative research is fundamentally and epistemologically flawed (Hammersley, 2007; Kitto, Chesters & Grbich, 2008; Lincoln & Guba, 2000; Morrow, 2005). The research frameworks, data collection, reliability and validity processes involved in quantitative research do not fit the qualitative paradigm.

In qualitative research, rigour is the concept for quality of data and integrity of the findings; that the researcher can demonstrate the findings are worth paying attention to and worth taking into account (Lincoln & Guba, 1985). Rigour in qualitative research is relevant to the notions of validity and reliability in quantitative research, and to the reduction of bias. In qualitative research, validity does not carry with it the same meaning as it does in quantitative research, nor is it a

companion of reliability or generalisability (Creswell, 2003). Therefore, the concepts of reliability and validity need to be reconceptualised to suit qualitative methods.

Validity in qualitative research is used to determine whether the findings are an accurate representation from the view of the researcher, participant and the readers of an account (Creswell & Miller, 2000). Terminology abounds in the qualitative literature that speaks of this idea, terms such as rigour, trustworthiness, authenticity and credibility (Creswell & Miller, 2000), and is a highly debated topic (Lincoln & Guba, 2000). Lincoln and Guba (1985) proposed that constructivist inquiry required a different set of criteria for assessing quality from those of traditional social sciences; collectively the criteria are an indication of trustworthiness, a parallel to the term rigour. There are several elements of rigour within the research process that need to be achieved.

3.3.2.1.1 Operationalising rigour in qualitative research.

Viewed as the gold standard for rigorous qualitative research, Lincoln and Guba's (1985) framework of quality criteria suggests four criteria for developing rigour and trustworthiness in qualitative research; credibility, dependability, confirmability, and transferability. These criteria represent parallels to the positivists' criteria of internal validity, reliability, objectivity, and generalisability, respectively (Polit & Beck, 2014). In response to criticisms and to their own evolving views, Guba and Lincoln (1994) added a fifth criterion more distinctively within the constructivist paradigm; authenticity.

Credibility is the most important measure of rigour, and refers to establishment of confidence in the truth value of the data, and the resultant interpretative findings (Lincoln and Guba, 1985). Two aspects of qualitative research are essential to achieving credibility; conducting research in such a way that enhances believability of findings; and implementing processes that demonstrate credibility to individuals external to the research. Its quantitative equivalent is internal validity, which when adapted and applied to the qualitative paradigm ensures that the findings represent the multiple constructions of an experience adequately; that the reconstructions that have been arrived at are credible.

Dependability refers to stability, or reliability, of data over time and across contexts. This is achieved through replication of findings with the same (or similar) participants in the same (or similar) context. Credibility cannot be achieved in the absence of dependability, just as validity cannot be achieved in the absence of reliability in quantitative research (Lincoln & Guba, 1985).

Confirmability is the potential for congruence between two or more independent people about the data's accuracy, relevance and meaning. This element of rigour is concerned with establishing that the information participants provide is representative of the data, and that interpretations of data are not created by the researcher. This is achieved by displaying that the

findings reflect participants' voices and the conditions of the research, and are not the product of the researcher's motivations, perspectives, or biases. Confirmability relates to the concept of objectivity because it demonstrates where and how the findings of the research are derived from the participants and conditions of the research and not by the biases, motivations, or perspectives of the researcher (Lincoln & Guba, 1985).

Transferability, analogous to generalisability, is the extent to which the findings can be transferred, or apply to, other groups or settings, including provision of sufficient descriptive data so that others can evaluate the applicability of the data to other contexts. Generalisability is impossible to achieve and does not suit the framework or aims of qualitative research. The qualitative researcher can only provide sufficient description for consumers to reach their own conclusions as to whether findings are applicable and transferable to other contexts (Lincoln & Guba, 1985).

Authenticity is the extent to which researchers fairly and faithfully show a range of different realities. This emerges in a report when it conveys the tone of participants' lives as they are lived. A study has authenticity if it leads the reader into a vicarious experience of those who are being studied and provides readers with a heightened sensitivity to the experiences, and issues, characterised.

In summary, a qualitative methodology adopts an interpretive, naturalistic approach to the phenomenon of interest and involves making sense of a phenomenon through interpreting meanings that participants construct about them (Denzin & Lincoln, 2008; Patton, 2002). My research employed a qualitative research methodology within a phenomenological framework using in-depth exploratory interviews with a purposive sample of participants, to address the research questions of what are parents' lived experiences and which factors are important in understanding constructions of experiences and attributions of meaning for parents whose children are involved in the AREST CF early surveillance program, and how parents cope. The qualitative framework sought to explore and describe the experiences, thoughts, emotions and ideas of participants by co-constructing the different ways participants experience parenting in the context of early surveillance for lung disease, and the meanings associated with those experiences. Explanation of phenomenology as the underpinning principles of the chosen methods will now be explained before providing justification of data collection, sampling and data analysis techniques will now be outlined before detailing why, with regard to all the reviewed literature, my research addressed a pressing lack of understanding, and was therefore worthy of action.

3.3.2.1 Phenomenology.

Phenomenological theory has influenced qualitative research, in particular in the social sciences (Flood, 2010; Liamputtong & Ezzy, 2005; Zippel, 2010). It is popular in the health sciences (Borgatta & Borgatta, 1992), psychology (Giorgi, 1985, Polkinghorne, 1989), and nursing

(Nieswiadomy, 1993). Phenomenology is derived from philosophy and provides a framework for a method of research (Mapp, 2008). As the philosophical underpinnings of phenomenological inquiry direct a phenomenological study, it is imperative to understand how one relates to the other. A phenomenological approach focuses on the lived experiences of people and seeks to understand their reality from their perspective, emphasising the direct study of personal experience and understanding of the nature of human consciousness (Becker, 1992). Phenomenology is based within an interpretivist research paradigm and follows a qualitative approach (Denscombe, 2003). These concepts, fundamental to understandings of phenomenological inquiry and to qualitative research practice, have developed over time through contributions from influential phenomenological philosophers (Flood, 2010; Liamputtong & Ezzy, 2005; Patton, 2002).

Phenomenology as a philosophical method of inquiry was developed by the German philosopher, Edmund Husserl (1859-1938), who is credited with the birth of phenomenology and developed a systematic method of studying human consciousness and experience (Husserl, 1977). Husserl posited that experiences of life events in the everyday world, with theoretical underpinnings suspended, was an invaluable source of knowledge (Becker, 1992). His aim was to return to things themselves, as they appear to their perceivers, and to set aside, or *bracket*, that which we (think) already know about them (Willig, 2001). Husserl developed phenomenological inquiry to study how people describe things, with the assumption that we can only know what we experience by attending to meanings that activate conscious awareness (Flood, 2010; Patton, 2002). Husserl referred to this notion of conscious awareness of how experiential processes proceed and what is experienced through them as *intentionality* (Charmaz & McMullen, 2011). Therefore, people construct meanings as they engage with, and make sense of, the world they are interpreting. It is the researcher's task to analyse the intentional experiences of consciousness to determine how phenomena are given meaning and to arrive at its essence (Sadala & Adorno Rde, 2002). An analysis of the essence (or structure) of phenomena in context is one of the outcomes of phenomenological research (Flood, 2010).

Another German philosopher, a student of Husserl's, Martin Heidegger (1889-1976), continued Husserl's works by including elements of space and time within this broader explication of phenomenology (1962). Heidegger's work is cited as the first union of existentialism and phenomenology, resulting in existential-phenomenology; a study of human existence and experience with existential foundations within a phenomenological method (Becker, 1992). Whilst Heideggerian phenomenology proposes that researchers interpret data in terms of their own experiences and knowledge, Husserlian phenomenology advocates that researchers suspend (or bracket) their personal beliefs about the research phenomena whilst seeking to describe the participants' experiences (Mapp, 2008).

More recently, phenomenological inquirers have shifted from a pure philosophical method of inquiry to adapting and systematising the use of phenomenological methods. Led by Amedeo Giorgi (1985, 2009), methods originally developed by phenomenological philosophers, such as the intentional analyses of meaning and the analysis of the essences of phenomena have been adapted for empirically based psychological research (Wertz, et al., 2011).

Despite differences of opinion amongst qualitative researchers who use phenomenological inquiry, there are a number of commonalities in its philosophical beliefs. These include the study of lived experiences of a group of people who experience a particular phenomenon and the consideration of these experiences as being conscious ones (Flood, 2010; van Manen, 1990; Zippel, 2010), and the process of developing a description of the essence of these experiences (Flood, 2010; van Manen, 1990). At a broader level of commonality is the rejection of a subject – object dichotomy, therefore assuming that reality of an object is perceived only within the meaning of the experience of an individual (Creswell, 2007; Flood, 2010; Zippel, 2010). In considering these commonalities, phenomenological inquiry is concerned with co-construction of everyday life emphasising that people's actions are understood only when they are situated in the routines of everyday interactions in the world (Flood, 2010; Liamputtong, 2009). Therefore, a phenomenological study seeks to explore and describe people's experience of everyday life events within a definite philosophical context that produces specific assumptions about human nature and humans living within a particular nature (Becker, 1992).

Accordingly, phenomenological inquiry in philosophical terms asserts the nature of how reality comes to be experienced and understood (Flood, 2010; Grbich, 2007; Liamputtong, 2009; Patton, 1990), whereas as a research methodology, phenomenological inquiry seeks to explore how a specific aspect of lived reality is constructed by participants. This includes, on the researcher's behalf, bracketing preconceived ideas about the phenomena in question (Daly, et al., 2007; Carpenter & Suto, 2008). As either a philosophical approach to research or a methodology, its central focus is about how to investigate the lived experience of a particular phenomenon for those who experience it (Creswell, 2007; Liamputtong, 2009; Moustakas, 1994; Patton, 1990). As a research methodology for understanding the lived experiences of individuals (as in the work of Giorgi, 1985, Giorgi and Giorgi, 2003), phenomenology is a popular framework within the social and health sciences, particularly in sociology, psychology, education, nursing and health sciences (Creswell, 2007; Wills, 2007; Padgett, 2008).

When using phenomenology as a methodological framework, a study aims to understand and describe participants' experiences of their everyday lifeworld as they see it (Wills, 2007; Carpenter & Suto, 2008; Padgett, 2008). This understanding of phenomenological inquiry aligns with the purposes of my research, which seeks to explore parents' lived experiences of early surveillance

and interpret how meaning is assigned (Creswell, 2007; Flood, 2010). For this reason, the significance of phenomenological inquiry to my research is in gaining an understanding of how parents experience the phenomena of early surveillance. Of particular concern is how parents' interactions in the world influence their experiences. As such, how parents construct their experiences becomes a focal point. Accordingly, the philosophical underpinnings of a constructivist approach to research are discussed in the next chapter.

In terms of practical application, a phenomenological approach to understanding experience has led to positive health outcomes. Some examples of where phenomenological enquiry has led to positive health outcomes from knowledge gained in nursing research include: phenomenological analysis of interviews with patients who suffered from obstructive sleep apnea enabled development of a tailored intervention that promoted treatment adherence (Sawyer, Deatrick, Kuna & Weaver, 2010); a psychoeducation program was developed from phenomenological methodology that improving physical functioning, general health, anginal pain symptoms, and self-efficacy to manage pain (MCGillion, et al., 2008); and phenomenological analysis allowed elaboration of quantitative findings that showed anxiety and anger increase children's blood pressure. Phenomenological enquiry in this particular study discovered what aspects of childhood can contribute to anxiety and anger, which contributed to development of a program for teaching children how to deal with their emotions (Howell, Rice, Carmon & Hauber, 2007).

As there are no studies exploring parents' experiences of early surveillance for CF lung disease, a phenomenological study is appropriate for eliciting data that reflect parents' experiences and the ways in which they interpret, and cope with, early surveillance. Therefore, in the context of my research, I viewed experience as a valid and worthwhile source of knowledge, and everyday worlds of individuals as also valuable sources of knowledge. Accordingly, a phenomenological study exploring parents' experiences of caring for a child being treated for CF enrolled in the AREST CF early surveillance program was conducted.

3.3.2.3 Data collection technique.

There are a number of informative sources and data collection methods that can be used in qualitative research (Chwalisz, Shah & Hand, 2008; Patton, 2002). A key strategy is to obtain accounts or descriptions of the phenomena under investigation through conversations with people, and to learn about their lived experiences or their life world (Creswell, 2007; Kvale & Brinkmann, 2009). Techniques such as narrative interviewing, conversational interviewing and semi-structured interviewing attempt to understand the world from the participant's perspective and aim to uncover meanings of their experiences, rather than to provide scientific explanations (Kvale & Brinkmann, 2009, Liamputtong & Ezzy, 2005; Wengraf, 2001). Moreover, it is assumed that semi-structured interviewing techniques elicit specific knowledge, emotions, perceptions and experiences of a

phenomenon that can be articulated best by the verbal accounts of participants in their own words (Liamputtong, 2009). For this reason, a semi-structured interviewing technique is an appropriate approach to investigate the *essence* of the lived experiences of participants in my study (Creswell, 2007; Elliott & Timulak, 2005).

A semi-structured interview schedule consists of a set of broad questions about a topic that allows freedom in conversation (Liamputtong, 2009; Patton, 2002). This method was chosen as the vehicle for data collection because it allowed me to adequately explore parents' experiences whilst at the same time allowing substantial flexibility in conversation between participants. It also allowed the use of probing questions to develop more layered responses and descriptions that may be of particular interest or relevance (Chwalisz, et al., 2008; Elliot & Timulak, 2005).

The flexibility afforded by the semi-structured interview is particularly appropriate to discovering a multitude of, and complexity within and across, experiences and associated meanings. Miles and Hubermann (1994) and also Smith (1995) identified several advantages of conducting semi-structured interviews, which include assurance of flexibility in developing rapport, ensuring suitability to exploring personal and complex issues, and in providing rich data embedded in a certain context. Additionally, this style of interview suits a constructivist framework because it frees the researcher to explore how people construct meanings about their experiences, which leads to obtaining rich descriptions of the *life world* of participants (Liamputtong & Ezzy, 2005), and also applies to the consideration that one *real truth* does not exist in place of multiple realities (Guba & Lincoln, 1982).

The nature of this qualitative research project required that I have an intuitive sense of what was occurring in the data; therefore flexibility was instrumental throughout the entire research process (Corbin & Strauss, 2008). Flexibility in the semi-structured interview format allowed parents' experiences and stories to guide the interview, which meant they became active participants in the research process rather than respondents to a fixed set of questions (Elliot & Timulak, 2005; Polkinghorne, 2005). Gaining knowledge and understanding of participants' experience in such a way provides greater detail and allows for consideration of the wider context of experience that is important when dealing with complex emotions and values about a phenomenon (Miles & Hubermann, 1994; Patton, 1990).

Indicative of the dynamic flexibility required within a qualitative research design, it became apparent during data collection that telephone interviewing could increase volume of participation, therefore increasing the diversity and amount of parental voices heard. Telephone interviewing has been used extensively in quantitative research methods (Barriball, Christian, While & Bergen, 1996; Carr & Worth, 2001), mostly as a form of survey data collection. However, this particular interview mode has been much less used as a qualitative method (Sturges & Hanrahan, 2004), with debate

about its adequacy (Carr & Worth, 2001; McCoyd & Kerson, 2006; Novick, 2008; Sturges and Hanrahan, 2004; Sweet, 2002). As a method of data collection, telephone interviews possess several advantages including increased access to geographically distant participants (Sturges and Hanrahan, 2004; Sweet, 2002), the ability to take notes unobtrusively (Carr & Worth, 2001; Sturges and Hanrahan, 2004), more privacy (Sturges and Hanrahan, 2004), increased rapport (McCoyd & Kerson, 2006), convenience to participants, and elimination of any influence to respondent replies by characteristics of the interviewer, for example, class or ethnicity (Bryman, 2006). Although face-to-face interviews appear in the literature to be the 'gold standard' for qualitative research (McCoyd & Kerson, 2006), the use of telephone interviews has been reported as a versatile data collection tool (Carr & Worth, 2001), with telephone interview data judged as a source of rich, vivid, and detailed, high quality data (Chapple, 1999; Kavanaugh and Ayres, 1998; Sturges and Hanrahan, 2004; Sweet, 2002).

In my research, I was required to identify and acknowledge myself as part of the research instrument and to view the research as transitional and malleable and therefore open to modification and refinement as new knowledge was gathered (Corbin & Strauss, 2008). I engaged in a process of reflexivity, which involved continual and detailed self-reflection (see in later sections: Role of the Researcher and Rigour in Qualitative Research). The interpretative nature of this research, and my continual and intricate involvement in the research process, produced a range of strategic, ethical and personal issues in the research process (Locke & Golden-Biddle, 2002).

In summary, qualitative methodology and semi-structured interviews with parents were used to explore, understand and interpret the lived experience of parenting children with CF in the context of early surveillance. Employing a qualitative methodology allowed me to move away from quantification and causation towards discovering the essence and meaning attributed to these experiences. The style of the qualitative interview allowed me to observe experiences relative to a person's construction of phenomena and it's relation to their life world. Obtaining an inclusive account of parents' experiences was paramount to interpreting meaning of the described phenomena (Kvale & Brinkmann, 2009; Liamputtong, 2009; Polkinghorne, 2005). The next section outlines the chosen sampling technique.

3.3.2.4 Sampling technique.

A primary aim of qualitative research is to derive meaning, to uncover multiple realities, and to avoid generalisation to a target population (Creswell, 2014). Therefore, qualitative sampling completely avoids random sampling as it is not the most appropriate method for selecting individuals who will be useful informants. Non-random sampling identifies participants who have the relevant experience to meet the conceptual needs of the study and provide the information sought. Therefore, qualitative researchers use non-probability sampling techniques (Polkinghorne, 2005).

Fundamental to understanding the experience of individuals is to question those who have the relevant experience (Seidman, 1989): to understand what it is like for them and from their perspective (Dewey, 1960). Accordingly, my research applied a purposive sampling technique, which involved selecting participants, from a cohort of potential cases, who would best contribute to the research (Creswell, 2014). Maximum variation sampling (Patton, 2002) was chosen as the specific type of purposive sampling as it engages cases with demographic or other differences that have shared a common experience. This type of purposive sampling is often the sampling technique of choice in qualitative research because it is useful in exploring a broad scope of the phenomena and in identifying critical patterns that cut across variations in demographics or other differences identified through the sampling technique (Polit & Beck, 2014). Qualitative researchers often want to explore and understand the breadth of individual experience and so specifically look for individuals with demographic or other differences who have shared a common experience (Seidman, 1989).

My research recruited parents who have experience of their young child with CF enrolled in the AREST CF early surveillance program. To achieve the breadth of parental experiences, both mothers and fathers of children who attend the CF clinics at PMH, Western Australia and RCH, Victoria were invited to participate. The sample was stratified at recruitment into two groups according to the child's age (under 3 years and over 3 years). This allowed representation of maternal and paternal experiences across time in the programme, as well as sex-specific and clinic site-specific issues.

The purpose of stratified-purposive sampling (Patton, 2002) is to identify major variations in experience rather than to identify a common core experience. Stratification by child age provided some representation of the child's stage of development in relation to parents' experiences and ability to cope, and captured potential themes specific to the duration of time the child has received early surveillance since diagnosis.

3.3.2.5 Sample size and saturation.

In qualitative research, no set procedure is strictly applied to determine the sample size, as is the case for quantitative research (Patton, 2002). The purpose of sampling in qualitative research is to focus in-depth on relatively small samples selected meaningfully and strategically, rather than for the purpose of attempting to make statistical comparisons (Patton, 2002). Therefore, the sampling process is flexible and at the start of the study the number of participants is not definitively known. Data saturation, however, is a procedure used by qualitative researchers as a way of justifying the number of participants; this occurs during the data collection process (Liamputtong, 2009).

Saturation occurs when no new data are being generated from the data already collected (Padgett, 2008) and sufficient data have been obtained to account for all aspects of the

phenomenon (Morse, Barrett, Mayan, Olson, & Spiers, 2008). This refers to the idea that the sample is adequate when “the emerging themes have been efficiently and effectively saturated with optimal quality data” (Carpenter & Suto, 2008, p. 152). Mason (2010) and Morse (2000) both report adequate saturation with approximately 30 interviews, with Francis and colleagues (2009), Guest and colleagues (2006) and Burgess-Limerick and Burgess-Limerick (1998) all suggesting 12-15 in-depth interviews could be adequate.

3.3.2.6 Thematic analysis.

Engagement in exploratory qualitative research relating to parents’ constructions and experiences of parenting children with CF in the context of early surveillance relies on inductive processes such as *thematic analysis* whereby themes emerge from the data that provide rich, context-bound information (Creswell, 1994, Kvale & Brinkmann, 2009). Coding is the primary process for developing themes within the raw data by recognising important moments in the data and encoding it prior to interpretation (Boyatzis, 1998). Given thematic analysis is an analysis tool for exploring interview data, it allows identification, examination and reporting of themes within data and provides foundations for describing rich information (Braun & Clarke, 2006; Kvale & Brinkmann, 2009; Liamputtong, 2009).

Outcomes of thematic analysis are called themes, and are considered representative of a level of patterned response or meaning or construction that attempt to address the research aims and questions (Braun & Clarke, 2006). Though description is part of the analytic journey of which thick description is a valuable component, description alone is not sufficient. The data must also be challenged, extended, supported and linked to reveal their full value. Themes only attain their full significance when they are linked to form a coordinated picture or an explanatory model (Bazely, 2009). As the theoretical framework of phenomenology and philosophical assumptions of constructivism guided this research, the ways in which individuals make meaning and construct experience were of particular interest. Accordingly, thematic analysis offers a theoretically flexible approach to analysing qualitative data (Braun & Clarke, 2006; Elliot & Timulak, 2005) because it allows the researcher to organise qualitative data coherently (Miles & Huberman, 2002). Braun and Clark’s (2006) stage process for thematic analysis aligns with Giorgi’s (1985) methods of phenomenological analysis.

3.4 Summary

This section has shown how the epistemological and methodological assumptions of my research were conceptualised in response to the phenomena under investigation and the research questions being explored. This chapter has presented substantiative evidence for theoretical and methodological choices underpinning my research. Theoretical frameworks provide a structural guide, whereas methodological strategies provide an operational guide. Together, they function as a

basis through which to conduct research, view research findings, and to generate interpretations and to make recommendations. The next chapter will describe the methods used to conduct my research.

CHAPTER 4**Methods**

This chapter presents the methods used to obtain data pertaining to parents' experiences and coping strategies, and to interpret which factors are important in understanding the experience of parents who have a child with CF undergoing early surveillance for lung disease. The purpose of this chapter is to; 1) reiterate the aims of the study and outline the research questions; 2) review the theoretical framework used to conduct the study; 3) detail the participants involved; 4) describe the recruitment process; 5) explain data collection procedures; 6) outline data analysis techniques and highlight rigorous techniques within a qualitative study and finally; 8) address ethical considerations and time frame.

*What we observe is not nature itself,
but nature exposed to our method of questioning...Werner Heisenberg*

4.1 Restatement of aims

Despite growth in early surveillance over the past decade, both in terms of the amount of medical procedures performed on children as well as the recognition of the program in the CF scientific community, there was a lack of understanding the experiences of parents whose children undergo early surveillance for CF lung disease. Therefore, the overarching aim of my research was to explore and interpret the lived experience of parenting in the context of early surveillance for CF lung disease. More specifically, the exploratory nature of the research set out to identify how parents construct their experiences and attribute meanings to early surveillance for CF lung disease, and how they cope.

4.2 Research questions

To address the research questions, an in-depth, interpretive and descriptive qualitative approach was used to discover and understand the unique experiences of parents in my research. Specific research questions were explored through a phenomenological framework that sought to explore and describe the lived experiences, thoughts, emotions and ideas of parents by co-constructing the different ways they experience parenting children with CF in the context of early surveillance, and the meanings associated with those experiences. As research questions leading to qualitative data collection strategies are often open-ended and exploratory in nature (Elliot & Timulak, 2005), a qualitative methodology from a constructivist perspective was chosen to address the following research questions:

- What is the lived experience of parenting children with CF in the context of early surveillance?
- What are parents' experiences of early surveillance?
- How do parents construct their experience and attribute meanings to early surveillance for CF lung disease?
- How do parents cope with the lived experience of parenting in the context of early surveillance?

4.3 Methodology

4.3.1 Phenomenology as theoretical framework.

Based on those philosophical influences described in the previous chapter, phenomenological inquiry as a research tradition intends to describe the life worlds of individuals (Creswell, 2007; Flood, 2010). Fundamental to phenomenological inquiry is the examination and description of meaning for a number of individuals about their lived experiences of a particular

phenomenon as they are considered to be the best able to describe their experiences (Creswell, 2007; Liamputtong, 2009; Moustakas, 1994; Patton, 2002). Focussing on descriptions of people's experiences with the underlying assumption of the existence of a structure (or essence) to shared experience, central meanings mutually understood through the experience of a phenomenon can be determined (Moustakas, 1994; Patton, 2002). This description, as previously outlined as intentionality, includes *what* and *how* it is experienced (Creswell, 2007; Moustakas, 1994). This understanding of the lived experience of individuals is at the heart of phenomenology as a philosophy as well as a research method (Creswell, 2007; Liamputtong, 2009; Moustakas, 1994). Therefore, a study with a phenomenological perspective is considered different from using phenomenology to philosophically justify the methods of qualitative inquiry as legitimate in social science research (Patton, 2002).

From its philosophical inquisition into the nature of existence to its more practical application as a method of qualitative inquiry, the evolution of phenomenological inquiry can be further explained by investigating how data are interpreted. Traditionally, phenomenological inquiry was viewed as a first-person experience (Husserl, 1977). However, more recently it is common in the social sciences to observe phenomenological inquiry as a study of other people's experiences principally reported in the third person (Heidegger, 1996). Another influential philosopher in the evolution of phenomenological methodology, Giorgi (1997, 2009) set explicit, systematic and accountable procedures for analysis of phenomenological data. His methods were in contrast to the earlier informal way phenomenological research in psychology had been conducted by earlier pioneers in philosophy, psychiatry and other disciplines. Briefly, the procedures comprise four stages: reading about the experience for a sense of the whole, differentiating the description into meaning units, reflecting on the psychological meaning and significance of each meaning unit, and clarifying the psychological structure (or essence) of the phenomena (Charmaz & McMullen, 2011). Through reflecting on many manifestations of a phenomenon, I was able to identify common themes. Moreover, by articulating the inter-relationships of these themes, the essential structure of the phenomenon was revealed (Becker, 1992). Giorgi's (1997; 2009) phenomenological data analytical process is reflected in Braun and Clark's (2006) explanation of thematic analysis, of which a detailed discussion can be found further in the chapter.

4.3.2 Constructivism as philosophical underpinnings.

The philosophical underpinnings, or worldview, guiding my research is constructivism. The purpose of constructivism is to understand how the human mind actively gives meaning and order to the reality to which it is responding (Balbi, 2008). In contrast to the positivist ontology, which assumes one reality or *truth* to be discovered (Guba & Lincoln, 1994), constructivism adheres to a relativist view of knowledge (ontology), which assumes multiple and equally valid realities

(Schwandt, 2000), in that people consciously experience the world and events in diverse ways (Polgar & Thomas, 2008). This is such that no purely objective knowledge truths are said to exist - rather, the rules of knowledge are co-constructed and are therefore subject to change.

From an epistemological perspective, constructivism holds that reality is constructed in the mind of the individual (Hansen, 2004) and co-constructed by researcher and participant. The constructivist position emphasises that meaning is hidden and must be constructed through deep reflection (Schwandt, 2000). As such, the nature of this enquiry assumes that knowledge is constructed – not discovered by the mind, and reality is reflected in a contextualised way (Alvesson & Sköldbberg, 2009, Schwandt, 2000). Therefore, a distinguishing feature of constructivism is the centrality of the interaction between the researcher and the participants under investigation, which allows deep reflection to occur through the interactive researcher-participant dialogue (Ponterotto, 2005). It is through this interaction that profound meaning can be understood. This process therefore allows the joint production (through co-construction) of findings from the interactive dialogue and interpretation.

Accordingly, based on the interpretivist-constructivist understanding of knowledge, the epistemological position of knowledge is subjective and influenced by the context of the situation, including individual experience and perceptions, the social environment and the interaction between individual and researcher (Ponterotto, 2005). Duly, constructivism claims that reality is socially constructed and can be understood only within context (Wills, 2007). Thus, context is an important factor that influences how people interpret their worlds and needs to be considered in research. This is contrary to positivist philosophy, whereby context is eliminated through using controls and randomisation, and where gathered research is more representative of experimental conditions than real life (Guba & Lincoln, 1994).

Within constructive epistemology, researcher and participants are connected in the research process, findings are created through interaction, and values are acknowledged, whereas within a positivist epistemology, the researcher remains objective and separate from participants so as to keep the research unbiased and value free (Charmaz, 2000: Guba & Lincoln, 1994). Consequently, a researcher in the positivist paradigm becomes disconnected from his or her participants. As the researcher is viewed as co-constructer of knowledge from a constructivist approach, the axiological viewpoint maintains that the researcher's values and lived experience cannot be separated from the research process (Ponterotto, 2005) and must therefore be acknowledged, described and bracketed.

Bracketing refers to the way in which the intentionality of the researcher can be suppressed, thereby separating usual preconceptions that influence everyday perception so that pure constituents of conscious experience can be uncovered (Polgar & Thomas, 2008). Intentionality refers to the human capability for awareness of objects as well as their contextual features, and it

allows humans to reason about objects in the world and communicate with others (Polgar & Thomas, 2008). Bracketing of intentionality, then, is to extinguish prejudices so that the essence of experience, in relation to the phenomena under investigation, can emerge purely and without bias or distortion.

Critics of the constructivist approach have questioned its strong emphasis on relativism and subjectivity (Marshall, Kelder & Perry, 2005; Michael, 1996). The aim of constructivist inquiry is not to suggest that one absolute truth cannot exist, but rather it is subject to change via the participants' construction and the researcher's interpretation. Accordingly, the aim of research underpinned by such assumptions is to explore and understand how people's worlds and experiences are constructed (Chwalisz, et al., 2008; Gergen, 2011; Stenner, 2009), by relying as much as possible on people's views of given phenomena whilst at the same time acknowledging meanings are subjectively constructed (Creswell, 2007). The relationship and interaction between participant and researcher is significant (Ponterotto, 2005). Retrospective rigour techniques consider how criticisms and limitations of constructivism may be handled in research, which are discussed at the end of this chapter. If we are to give parents a voice (Rappaport, 1995) to discuss their experiences of their child's CF diagnosis and early surveillance, then it would seem appropriate that a constructivist perspective be utilised to inform this exploratory operation, especially considering it challenges assumptions about how meaning is formed (Alvesson & Sköldberg, 2009; Gergen, 2011). This must be a fundamental consideration within this type of inquiry.

Applying a phenomenological framework from a constructivist perspective and thematic analysis to my research permits rich description and interpretation of parents' experiences in the context of the life-world of the parent, and meaning is attained through construction and attribution rather than through empirical validity (Alvesson & Sköldberg, 2009; Flood, 2010; Liamputtong & Ezzy, 2005). Given that methodological procedures within this approach are set in the natural world (Denzin & Lincoln, 2005), the next section discusses the methods (processes and procedures) and ethical considerations associated with research set in a natural world context.

4.4 Methods

4.4.1 Sample.

All parents who participated in my research were biological parents of their children who have CF. Children were selected from AREST CF clinic data bases in Western Australia and Victoria according to age and participation in early surveillance, and parent details were obtained. Refer to Figure 2 for a map of Australia where AREST CF operates. All children in the databases were diagnosed through NBS before 6 months of age. Parents received their child's CF diagnosis within the first 3 months after birth.



Figure 2. Map of Australia showing the regions where AREST CF operates. Retrieved from www.emapsworld.com

4.4.2 Participant characteristics.

Parents of children living in Australia registered within the AREST CF clinic data bases from PMH (Western Australia) and RCH (Victoria) CF clinics aged between 6 months and 7 years of age were invited to take part in my research. Most families lived in the metropolitan area of the major city within each state, with 17 (25%) parents living rurally across both states. Parents or guardians of infants and children with an equivocal diagnosis of CF, or those who were not participating in the early surveillance program were not included. Refer to Table 1 for an overview of how the samples were recruited.

Table 1. *Group descriptions and recruitment details*

	Participant group	
	PMH parents	RCH parents
Description	Perth AREST CF parents recruited through PMH CF clinic	Melbourne AREST CF parents recruited through RCH CF clinic
Selection method	Purposively selected based on their engagement with AREST CF at PMH	Purposively selected based on their engagement with AREST CF at RCH
Number of participants	34	33

4.4.2.1 Western Australian sample.

A purposive sample of 34 parents, recruited from a population of N=60 eligible families, of children aged 6 months to 7 years engaged in the clinical and research aspects of early surveillance with AREST CF at Perth CF clinic in PMH were interviewed. A total of 24 mothers and 10 fathers participated in an interview, representing a response rate of 40% and 16% respectively. Seven interviews were conducted with both mother and father present.

4.4.2.2 Victorian sample.

A purposive sample of 33 parents was recruited from N=71 eligible families, of children aged 6 months to 7 years undergoing early surveillance with AREST CF attending the RCH CF clinic. A total of 22 mothers and 11 fathers participated in an interview, representing a response rate of 31% and 16% respectively. Another seven interviews were conducted with both mother and father present. Refer to Table 2 for demographic characteristics of the samples categorised by CF clinic location and stratified child age.

Table 2. Demographic information of AREST CF parents by CF clinic location and stratified child age

Variable	Participant group			
	PMH parents		RCH parents	
	Under 3 years	Over 3 years	Under 3 years	Over 3 years
Parent (N)	18	16	14	19
Mother	12	12	9	13
Father	6	4	5	6
Age (years)				
Range	23-52	26-42	24-39	30-43
Mean (SD)	37.13 (6.91)	34.57 (5)	31 (5.27)	33.75 (3.89)
Median	37	36	29	33
Age of child				
Range	.8-2.5	3-6.5	1-2.5	4-6
Mean (SD)	1.31 (.52)	4.53 (.99)	1.9 (.41)	5 (.65)
Median	1	4	2	5
Number of children				
Range	1	1-3	1-2	1-2
Mean	0.5	1.5	1.14	1.15
Use of child care				
Yes	8	9	8	9
No	10	7	6	10
Residential location				
Metropolitan	16	12	10	12
Rural	2	4	4	7

4.4.3 Recruitment.

The most important people in my research were the participants themselves (Buchanan & Coulson, 2005), so as part of my role within AREST CF at PMH, rapport with both the clinical staff and potential participants at PMH was established prior to commencement of my research. This was

achieved by spending time in the CF clinic with the CF nurse and also by meeting parents of children with CF during their routine clinic visits. Once the sample had been identified, I worked alongside the clinical CF nurse to secure interest in my research.

Eligible participants were identified from each site's AREST CF database, stored at The Telethon Kids Institute and RCH respectively. The CF clinical nurse at each site and I worked together to identify and approach patients' parents for inclusion in my research. Recruitment was achieved by distributing an invitation recruitment letter (refer to Appendix A) to eligible families in Western Australia, accompanied by an information statement (refer to Appendices B and C) outlining the research and what participation would require. Potential participants were required to make contact with me, at which time an interview was scheduled for a mutually convenient time and place, or telephone interviews were suggested as an alternative option when deemed necessary. I interviewed 50 parents at their homes, with 17 interviews being conducted over the telephone.

Due to differences in geographical location between RCH to PMH (where I am located), the same process of recruitment and establishing rapport and gaining access to potential participants was not possible at the Victorian site. The CF clinical nurse coordinator within the Respiratory Medicine department at RCH assisted in participant recruitment for the Victorian cohort. She established parents' interest in my research before granting me permission to contact potential participants, at which time an invitation letter and information statement were sent. I then made telephone contact with potential participants to organise an interview time and location before travelling to Victoria to conduct the interviews, either by telephone or face-to-face.

4.4.5 Ethics considerations.

Ethical issues and dilemmas in qualitative research are emergent and contextual, and therefore require situational responses. Therefore in keeping with ethics guidelines, it is worthy to note a number of processes that kept participants informed and safe for the duration of my research.

4.4.5.1 Research conduct.

All components of the research were conducted in compliance with the relevant codes - National Statement on Ethical Conduct in Human Research, developed jointly by NHMRC and Australian Research Council, and the Australian Psychological Society Ethical Guidelines, and in accordance with my research protocol. Ethics approval for this study was obtained from Edith Cowan University (8808), PMH (2023/EP) and RCH (33181-A). Research burden was a primary concern as my study has been with a sample of children with a rare condition requiring specialised care. Consequently, there has been considerable competition with clinical research groups in gaining access to the participants. At the time of recruitment, potential participants (and their children) may have been involved in many different research studies, both clinical and psychosocial in nature.

Therefore it was paramount to consider the timing of recruitment, both in Western Australia and Victoria. By working in conjunction with other AREST CF researchers, it was ensured that my research did not coincide with other psychosocial research projects.

In terms of participation in my research, I informed parents the interview was confidential, that they could stop at any time, they could withdraw from the research at any time without question, that any decision to withdraw would not affect their child's ongoing CF care, and that they could ask any questions. They were also advised the interview would be audio-recorded and transcribed and that a specific code would be recorded on their transcripts and demographic information to ensure confidentiality. After the research had been comprehensively explained, I obtained written and informed consent from participants for their involvement and for their interviews to be audio-recorded and then transcribed (refer to Appendices B and C).

4.4.5.2 Data management.

Source documents provide evidence for the existence of the participant and substantiate the integrity of the data collected (Lincoln & Guba, 2000). Once transcribed, audio-recordings were destroyed and all transcripts were de-identified by numerically labelling documents and removing all identifiable information. In my research, the data are considered partially de-identified due to the relatively small size of the CF population in Australia. As further measures of confidentiality, consent forms were kept separate from all data and all transcripts were protected in a secure environment at The Telethon Kids Institute. All external computer files were password-protected. Only I as the lead researcher, and named members of the research team had access to confidential records. I ensured all documents pertaining to this study were made available to the relevant regulatory authorities. I complied with applicable privacy and security laws for use and disclosure of information related to the research set forth in this protocol. My research was conducted under the auspices of the AREST-CF psychosocial research group and within the governance structure and research experience and expertise of AREST-CF. De-identified results and the written report were made available to all participating parents. Following the required storage period, as specified by the National Health and Medical Research Council, all data (including recordings, transcripts and demographic information) will be erased from the computer and external devices. Refer to Figure 3 for an outline of data analysis and data management processes.

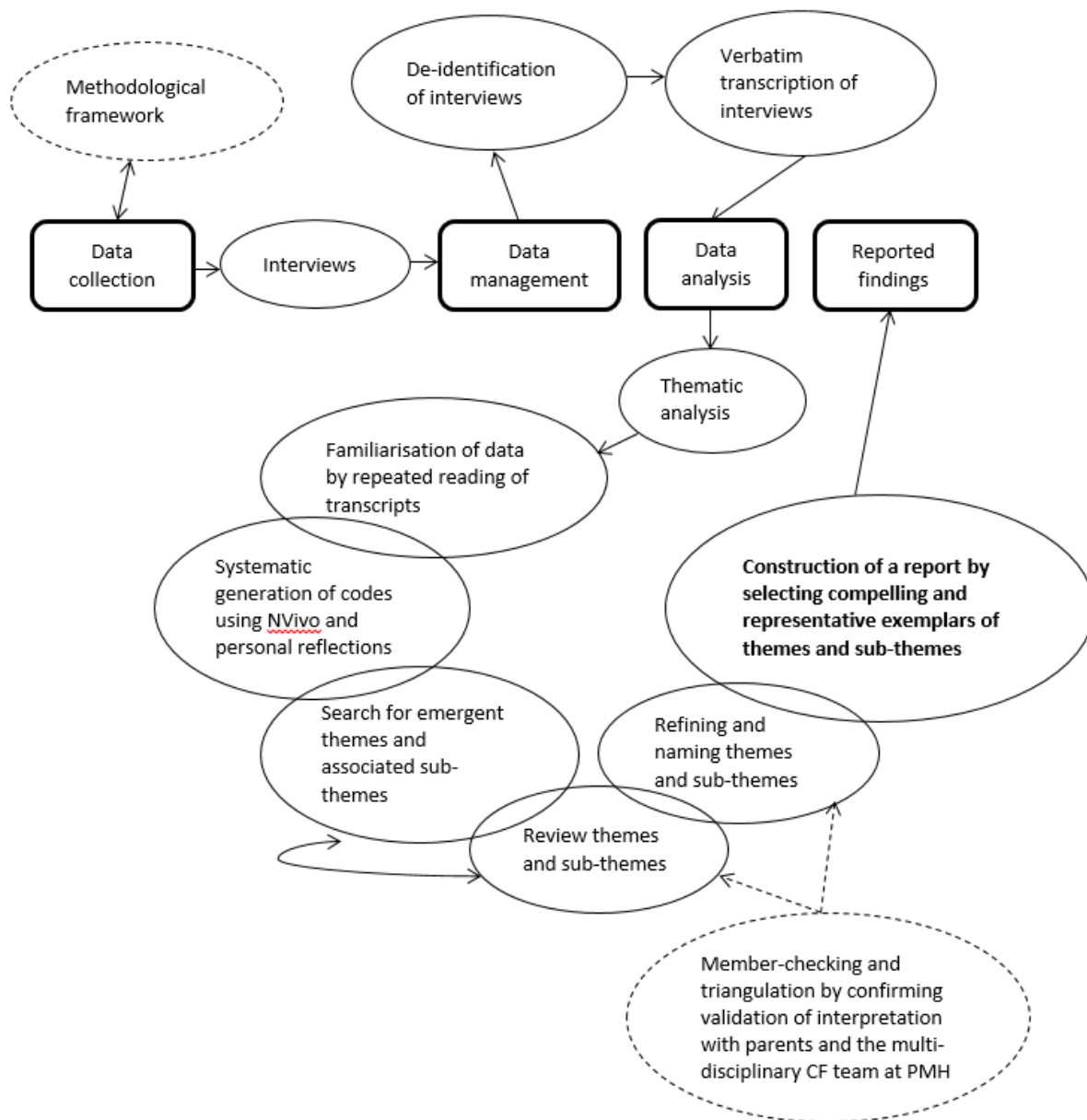


Figure 3. A conceptualisation of data management and data analysis processes

4.4.5.3 Informed consent process.

Informed consent was obtained from parents prior to their interview. This was achieved by providing them with an information statement about the nature of the research, what was expected of them as participants, how the results of the research were to be used, what information about them would be recorded, how the data would be stored and that they could withdraw from the research at any time without giving a reason. Assurances were also given that personal information would be kept secure, that any publication would not identify individual participants, and that all aspects of the Data Protection Act and other relevant legislation would be complied with. To further ensure confidentiality, parents were informed that their real names would not be used in any part of

the analysis process (transcript to report dissemination) and would be substituted with a non-identifiable number code (Corbin & Strauss, 2008). When I met with parents for their respective interviews, an information statement was given to them at which time they signed an informed consent form. Prior to telephone interviews, parents were required to sign and send their consent form back to me.

4.4.5.4 Consequences of the research.

The nature of qualitative research, and in particular the method of interviewing, lends itself to openness on behalf of participants whereby personal and/or intimate information may be disclosed. Expressing information of this nature can potentially arouse mixed emotions about sharing such information (Kvale & Brinkmann, 2009). Furthermore, it was recognised that the conversation might be uncomfortable and remind some parents about unpleasant experiences to which they may have had an adverse reaction. To manage such issues beyond debriefing and provision of available psychological support at the conclusion of each interview, I ensured that parents guided the conversation.

4.4.5.5 Role of the researcher.

Conducting research in a morally responsible manner is based on more than ethical obligations and methodological decisions and acknowledgements (Kvale & Brinkmann, 2009). Particularly in qualitative research involving interviews where the researcher is often the main instrument of data collection and thus obtaining knowledge, the moral integrity of the researcher is of utmost importance (Creswell, 2007; Kvale & Brinkmann, 2009). Given epistemological and ontological underpinnings of the research, my interpretations were explicitly informed by reflexivity, which is introspection and acknowledgement of values, history, interests and potential biases, and typifies qualitative research (Balbi, 2008). The personal-self and the researcher-self become inseparable, and acknowledgement of this concept represents honesty and openness to research by acknowledging that all inquiry is laden with values (Mertens, 2012). Reflexivity emphasises the importance of self-awareness and ownership of one's perspective (Patton, 2002). I exercised reflexivity by attempting to understand how my own experiences and background affect what I understood (Liamputtong & Ezzy, 2005), to minimise researcher effects on data collection, analysis and interpretation. Firstly, I systematically reflected on my role during inquiry (both during interviews and analyses) and continually considered my personal biography and how it potentially shaped my research. Secondly, by acknowledging potential power differentials, I was able to view the interview process as a learning opportunity (Reason & Bradbury, 2008). Lastly, I applied the practice of bracketing during data collection, analysis and interpretation (Braun & Clark, 2006).

Similar to reflexivity, bracketing is a strategy for ensuring that any and all extrinsic influences are identified and duly acknowledged. In practice, it refers to a process of identifying, acknowledging

and suspending preconceived opinions and beliefs about the phenomena under study (Patton, 2002). This allows the researcher to bracket out presuppositions in an effort to confront their data in pure form (Polit & Beck, 2014). Bracketing was achieved through application of a number of mechanisms. Firstly, I am not a parent and was consequently able to bracket preconceived notions about parenting and parenthood. Secondly, I was a novice of the AREST CF collaboration and I therefore learnt about the surveillance procedures early in the research process, and learnt about parental experiences as part of data collection, analysis and interpretation. Therefore, by being self-aware and reflexive in consciousness, and by bracketing preconceived ideas about the phenomenon under investigation, scientific rigour of interpretation was ensured. Finally, in an attempt to reflect parents' stories accurately and sensitively, retrospective rigour strategies were utilised to ensure validation of interpretation (Creswell, 2007), which was discussed in detail in the previous section of scientific rigour techniques.

4.4.5.6 Additional ethics considerations.

In the interest of both ethical treatment of participants and duty of care on my behalf as a researcher, parents were provided with a list of counselling services they could contact if, after the interview, they felt distress or concerned about anything we discussed during the interview (refer to Appendix D for Western Australian services and Appendix E for Victorian services). Lastly, it is important to note that no respondents were denied an interview, that is, all parents from the eligible sample of 131 families who responded to the research invitation were interviewed. Outcomes from the interviews are presented in the next chapter. How interviews were conducted is the topic of the next section.

4.5 Data collection

4.5.1 Interview procedure.

The selection of an appropriate data collection method is primarily dependent on the research questions (Chwalisz et al., 2008; Kvale & Brinkmann, 2009; Patton, 2002). There are two key experiences of interest in my research; parents' experiences and psychological constructions of parenting in the context of early surveillance for CF lung disease, and how parents' cope with their experiences (refer to Figure 4 for an illustration of outline of data collection). To adequately explore these, a semi-structured interviewing technique, that is, one that provides a set of questions to ask about the topics but also allows the conversation freedom to vary substantially between participants was adopted (Fylan, 2005; Wengraf, 2001).

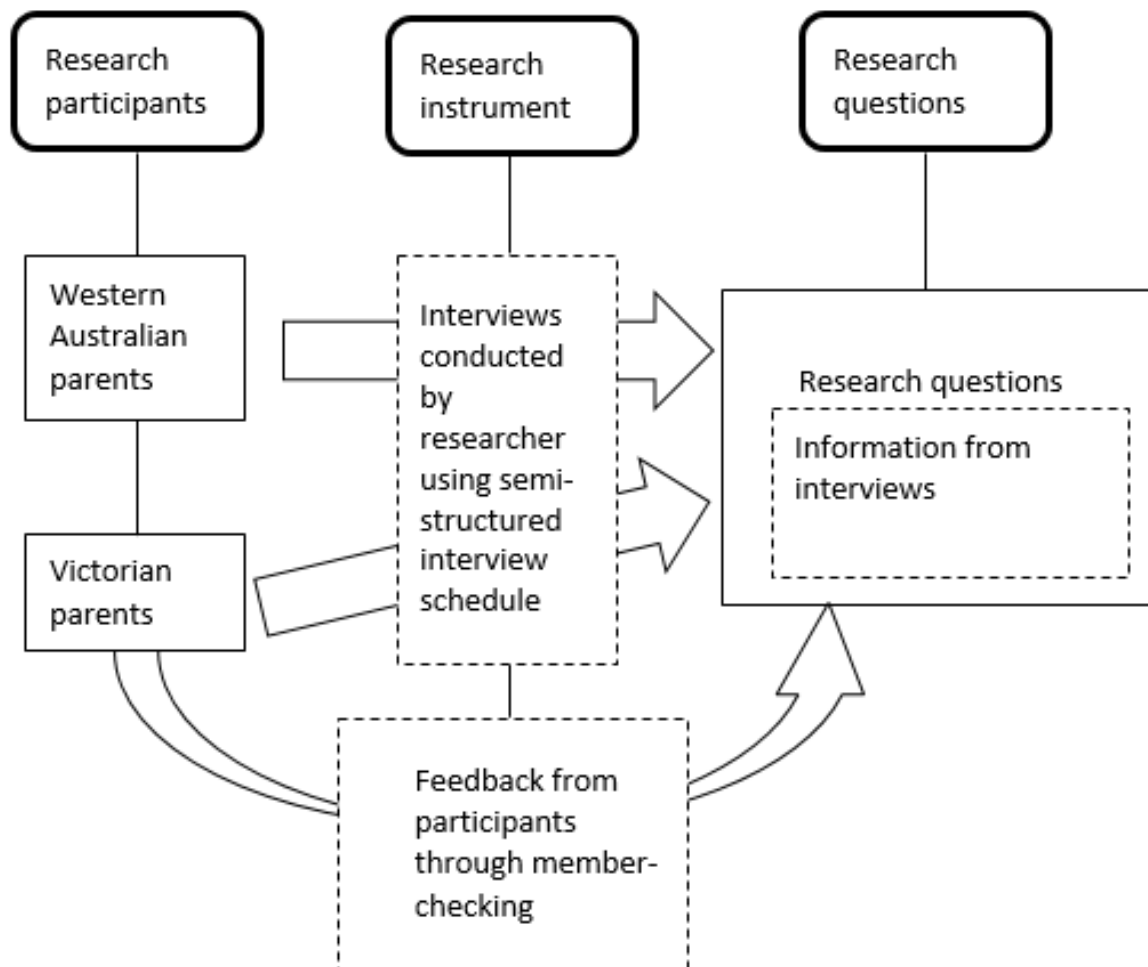


Figure 4. Illustration outlining data collection process

To develop rapport with parents (Kvale & Brinkmann, 2009), I first engaged each parent with some demographic and family questions such as age, number of children, and child care utilisation (refer to results chapter for demographic data categorised by site and stratified age). This information also provided a context for the findings emerging from interviews. Whilst all questions in the interview schedule were addressed with each parents, a flexible approach was taken to the order of questions, and further issues raised by parents were explored (Playle, 2000).

A total of 14 interviews were conducted with both mother and father present. To achieve the maximum number of participants in the most convenient way possible for the parents, interviews with either mother or father present, or both present together were accepted. A potentially positive consequence or outcome is that discussing thoughts and feelings together may have helped parents to express themselves more so than if they were interviewed alone. Limitations of this technique are explained in chapter seven.

The telephone interview became an integral data collection method in this study because it became apparent in the early stages of recruitment that for some parents, face-to-face interviews

were either challenging or inconvenient. Some parents were concerned about infection control because people with CF should not interact with other people with CF due to the likelihood of cross-infection, meaning that parents did not want to bring their child to the clinic for an extra visit. For other parents, home visits were inconvenient due to geographical location of residence or time and/or work constraints, or because their child may have been experiencing ill health. Whilst changing the environment of the interview, the content of the telephone based interview continued to be conversations about parents' experiences and I was able to probe deeply and seek clarification from parents regardless.

Parents interviewed via the telephone were willing to discuss their experiences as much as those parents who were interviewed in person. Therefore, quality and quantity of telephone interview data was not noticeably different from face-to-face interview data. Furthermore, by using telephone interviews as an alternative medium in conjunction with face-to-face interviews, increased recruitment was achieved whilst not compromising the method and philosophy of constructivism. Had telephone interviews not been offered, 17 parents' voices would not have been heard.

I stopped one interview due to apparent emotional distress of a mother expressing a particular event. She was offered the opportunity to finish the interview and I reminded of her right to withdraw from the interview and research. However, she was willing to continue with the interview. At the conclusion of this interview, the mother was reminded of the professional services available to her and encouraged to contact them if she so felt the need. The parent commented that she felt relieved about being able to express her emotions about the particular event. Other parents also described feelings of well-being and relief following the chance to express their thoughts and emotions about a difficult topic. The opportunity to share their experiences as well as their thoughts and feelings was mentioned by some parents as providing a type of therapeutic and beneficial effect, as they felt they had gained insight into their experience and relieved to express and share their feelings.

At the completion of each interview, parents were thanked for participating, reminded of my ethical obligations as a researcher and offered the chance to ask questions. To reduce risk of discomfort or distress as a result of being interviewed, regardless of how I perceived the interview, a debriefing with parents followed each interview. Parents were debriefed by allowing time to discuss any concerns they may have about the interview or any other aspect of the research. In the case of issues arising after I had left, a list of local counselling services was given to each parent they could make contact with. In the case of any issues arising for parents, I would have organised to follow up any concerns with each CF clinic coordinator who would then contact relevant support services

through their community network connections; however no parents chose to contact either CF clinic coordinators from each site about the research.

All interviews were between 30 and 90 minutes in duration, depending on how much information parents were willing to share with me. The primary reason for variation in length of interviews was how much experience parents had with AREST CF and the hospital environment. Their amount of experience was related to the age of their child and therefore exposure to early surveillance of CF. Other potential reasons for variation include amount of time available to be interviewed, level of distraction, capacity to articulate experience or level of willingness to share their experience (Morse, 2000). However, provided that data saturation is achieved, variation in length of interview is not a concern (Francis, et al., 2009).

Once I had finished each interview, I recorded personal impressions and reflections in a journal. This included notes about responses that generated interesting or relevant information for further consideration. The journal was a key element of the audit trail important for data analysis (see the last section in this chapter for further information on appropriate qualitative validation strategies). The journal was used to support data analysis by assisting with further delineation and refinement of responses to interview questions, and also by providing further context to the emergent findings. Please refer to Appendix F for a number of samples from my personal reflections written in my journal after each interview. To clarify misunderstandings in the data and also to check for verification of interpretation, follow-up telephone interviews were conducted and several e-mails were exchanged with seven parents.

4.5.2 Data collection instrument.

To obtain the interpretations of each parent's personal experiences, interviews were facilitated by an interview schedule (Smith, 2003). I developed a series of interview questions designed to answer the research questions that were piloted with the AREST CF parent representative group, a consumer reference group formed as part of a larger AREST CF psychosocial research group. The group consisted of five parents whose children had previously been part of AREST CF but who were no longer under surveillance due to their older age, and one adult with CF. The pilot was conducted to determine appropriateness and effectiveness of questioning in being able to elicit relevant information to address the research questions (Kvale & Brinkmann, 2009; Smith, 2003). A digital recorder was used to record each interview for the purpose of reviewing the interview narrative. As a result of this process, a number of questions were amended before commencement of data collection (Porter & Carter, 2000), for example, an initial question, 'are there any improvements to the service that could be made?' was further specified to 'is there anything the hospital can do to better support you or your child?' The interview schedule was then

reviewed by a staff member at the School of Psychology and Social Science, Edith Cowan University, to determine face validity and suitability of questions.

The modified interview schedule (refer to Appendix F) included a set of semi-structured questions designed to address the research questions that I then used to guide interviews. The semi-structured interviews explored parents' experiences associated with their child's involvement with early surveillance. Additionally it explored broader experiences associated with their child's CF; what coping means for caregivers involved in early surveillance, what coping strategies are employed for each of the clinical components of early surveillance, and whether coping evolves over time. Whilst all questions were addressed with each parent, a good deal of flexibility in the order of questions and exploration of further issues raised by each individual parent occurred (Playle, 2000).

The design of the interview elicited information specifically about parents' thoughts, feelings, and perceptions related to the systems and processes, practicalities, and receipt of health information involved with each annual surveillance assessment, as well as the broader impact of early surveillance of CF on individual and family life. The interview format was consistent with Smith's (2003) suggestion that an interview should begin more generally and become more specific. In addition, probing questions were used at times when I wanted to further explore issues raised by parents, for example 'can you please tell me more about that?' or 'can you please give me an example of that?'

4.6 Exploratory analysis

My research adopted and adapted Braun & Clarke's (2006) six-step guidelines to thematic analysis for qualitative psychological research, which reflect Giorgi's (1985) stages of phenomenological data analysis outlined earlier in the chapter. These phases are: familiarisation with data, generating initial codes, searching for themes among codes, reviewing themes, defining and naming themes, and producing the final report. Whilst not prescriptive (Rapley, 2011), the primary objective of following Braun and Clarke's (2006) guidelines was to ensure that an exhaustive and systematic analysis of qualitative data was conducted. Therefore, it is important that the phases are recognised and used as a guide rather than a linear or hierarchical approach so as to allow flexibility to fit the research questions and data (Elliot & Timulak, 2005; Patton, 2002). In practice, the aim was to approach analysis of raw data in a procedural manner which produced a rich description of meanings constructed by parents but which did not hinder the ad hoc, unique, evolving and unanticipated revelations often encountered through qualitative inquiry (Liamputtong, 2009). Accordingly, though there are six main phases in thematic analysis, data collection and analysis occurred simultaneously and the operations in each phase used as tools rather than prescriptive steps, thereby retaining the dynamic nature of qualitative research (Corbin & Strauss, 2008). A brief outline of the adopted and adapted guidelines suggested by Braun & Clarke (2006) is

presented in Table 3, along with a short description of requirements at each phase and outcomes from following the process within each phase.

Table 3. *Stages of thematic analysis for qualitative psychological research*

Stage	Process	Result
Stage 1 – Familiarisation with data	Read and re-read data, paying specific attention to recurring patterns.	Initial codes and detailed notes.
Stage 2 – Generating initial codes	Document where and how patterns occur by collapsing data into labels to create categories. Data complication is also completed here that involves making inferences about what the codes mean.	Comprehensive codes of how data answers research question.
Stage 3 – Searching for themes	Combine codes into over-arching themes that accurately depict the data and gather all data relevant to each potential theme.	List of candidate themes for further analysis.
Stage 4 – Reviewing themes	Determine how the themes support the data and the overarching theoretical perspective. If the analysis seems incomplete, there is a need to go back and find what is missing.	Coherent recognition of how themes are patterned to tell an accurate story about the data.
Stage 5 – Defining and naming themes	Definition of what each theme is, which aspects of data are being captured, and what is interesting about the themes.	A comprehensive analysis of what the themes contribute to understanding the data.
Stage 6 – Producing the report	Decisions must be made about which themes make meaningful contributions to understanding what is occurring within the data. Member-checking should also be completed here.	A thick description of the results.

Note. Adapted from "Using thematic analysis in psychology," by V. Braun and V. Clarke, 2006, *Qualitative Research in Psychology*, 3(2), p. 87.

Phase 1: Familiarity with the data

After interviews were conducted, I transcribed them as a way of becoming familiar with the data (Braun & Clarke, 2006; Flick, 2006). Listening to audio-recordings during this process allowed me to immerse myself in the data early on, which provided a valuable way of becoming familiar with the data (Braun & Clarke, 2006; Elliot & Timulak, 2005). To become familiar with the depth and breadth of content, all transcripts were read as a whole for deeper immersion in the data (Braun & Clarke, 2006). Reading transcripts in this way served a number of purposes. Firstly, it contextualised the data, which gave me an overall sense of parents' experiences. Secondly, it enabled me to both conserve the meaning of the data as a whole as well as start to consider structure of meanings within different interviews and across genders and age groups. Thirdly, it allowed me to begin

identifying, delineating and synthesising descriptive accounts which expressed the structure of meaning attached to the phenomenon under investigation. Lastly, I was able to raise questions about the continuity and contradictions of accounts generated by accessing multiple information sources.

Phase 2: Generating initial codes

Coding has been described as a procedure that breaks large volumes of data into smaller manageable categories (Liamputtong, 2009). At this point, the first interview was read several times for further familiarisation of the breadth and depth of the content (Braun & Clarke, 2006), and potentially salient words and phrases were developed into codes to provide meaning to the descriptive information (Miles & Hubermann, 1994) and then recorded. The data were then reduced by grouping similar categories and codes, and identifying the most salient themes. As codes were connected and clustered together, the transcript was checked for accuracy. These themes were then recorded at points of text that represented it (Liamputtong & Ezzy, 2005; Smith, 2003). This process was continued with subsequent interviews. Codes from previous cases were utilised to orient subsequent analyses, respecting convergences and divergences within the data. Constant comparative analysis was applied to develop and check for accuracy of theme boundaries, which involved comparing subsequent transcripts to previous ones, using identified codes from the first transcript to analyse the rest (Janesick, 1994; Strauss & Corbin, 1994).

The entire dataset was worked through systematically as outlined above allowing equal attention to each transcript. The coding process was conducted with the qualitative analytic computer software package NVivo 10 whereby codes were created and text collated from each interview transcript that represented them. With collation of text, much of the surrounding data were retained so as to ensure that context of the coded data was not lost (Elliot & Timulak, 2005). This process formed the basis for repeated patterns of themes to form across the dataset (Braun & Clarke, 2006, Liamputtong, 2009). Sub-themes that did not automatically fit within boundaries of major developing themes were kept as potential fragments of a major theme that may have represented a major theme not yet identified (Braun & Clarke, 2006). Storing large sections of text to a code and retaining sections of text that did not automatically fit into the emergent structure ensured that inconsistencies and contradictions were not ignored across the dataset, instead informing the overall analysis (Braun & Clarke, 2006; Grbich, 2007; Kitto, et al., 2008; Liamputtong & Ezzy, 2005; Mays & Pope, 2000; Morrow, 2005). As identified earlier, it is important in qualitative research to ensure that uniqueness of people's experiences is represented beyond generalisations of these accounts. This is important for facilitation of credibility and authenticity of the findings because condensations and interpretations of the data become more transparent, meaning established arguments from the most probable interpretations presented are made clear, therefore

allowing the reader to look for alternative interpretations (Grbich, 2007; Kitto, et al., 2008; Liamputtong & Ezzy, 2005; Mays & Pope, 2000; Morrow, 2005).

Phase 3: Theme development

Following identification of codes within the data, underlying meaning and construction of experience within codes eventually required consideration on an interpretative level (Elliot & Timulak, 2005). Underlying threads of meaning and construction from codes are called themes that aim to describe aspects within the structure of experiences that are neither objects nor things, but rather address the *how* or the *why* of experience (Liamputtong, 2009).

The process of theme development involved exporting data from the NVivo computer software package so as to manipulate data manually. From this point on, I was able to more intimately connect with the data to define and order conceptual interpretations in more nuanced ways than NVivo allows. This was made possible by cutting up sheets of paper with codes and their descriptions, and organising them across larger sheets of paper. This began development of categories (refer to Appendix H). Making the process visual in this way made it more dynamic and it allowed for movement back and forth of compelling statements and ideas across emergent themes. From this process, I was able to review the level of the coded data extracts, reading all collated data for each theme together to identify and justify a coherent pattern of thought. Essentially, this allowed analysis of the already coded data to be compared and considered in combination to form overarching themes (Braun & Clarke, 2006).

I decided to obtain a detailed thematic description of the entire data set, rather than a more detailed and nuanced account of one particular theme within the data. This enabled me to develop a sense of the predominant themes within the data, therefore reflecting an accurate representation of the entire data set (Braun & Clarke, 2006). Themes were identified using an inductive or *bottom-up* approach, which means that themes were identified without *a priori* development. This involved bracketing analytic preconceptions (*epoche*) about the phenomena based on previous literature (Crotty, 1996).

Phase 4: Reviewing themes

Several thematic mind-maps were created to consider how different codes may combine to form overarching themes (refer to Appendices I & J). These provided graphical representation of links and associations between codes and themes (Braun & Clarke, 2006). Once a candidate thematic map was decided upon, the themes were then reviewed for internal homogeneity and external heterogeneity; this meant that themes needed to cohere meaningfully whilst still retaining identifiable distinctions between themes (Patton, 2002). All collated extracts for each theme were considered for whether they appeared to form a coherent pattern representative of each theme. In considering relationships between codes, themes and different levels of themes, some codes were

found to fit into more than one topic area, which prompted generation of master themes containing sub-themes. This process further assisted in verification and review of any existing themes (Braun & Clarke, 2006; Rapley, 2011).

Once I was satisfied that the candidate themes adequately captured the contours of the coded data, consideration of the validity of individual themes in relation to the entire data set occurred through accurate reflection of the meanings evident in the data set as a whole. Established themes and related sub-themes were contrasted with the overall story told across the entire dataset. Additional coding of data within themes that had been missed in earlier coding stages also occurred at this stage (Braun & Clarke, 2006). New themes were added and some themes were dropped as they either did not fit well in the emerging structure, or were not very rich in evidence within the transcripts.

Reflective writing and member-checking became invaluable during this process. Firstly, reflective writing pointed to initial arguments to support developed conclusions, and how those conclusions were reached. The exercise of continuous writing in an ordered format also forced clarification of ideas (Bazely, 2009). Secondly, member-checking of emergent themes meant that reconstructions of a reality were confirmed as precise and truthful representations of parents' multiple realities. This was achieved by conferring with a methodological expert for confirmation of appropriate them development.

At this point, I was satisfied that the themes accurately represented the essence of the coded data. It was then that the literature was re-engaged to enhance sensitivity to subtle nuances arising in the interview data (Corbin & Strauss, 2008). This meant that where themes, associated sub-themes and concepts already existed in the literature, comparisons of their similarity and significance to experience could be noted (Braun & Clarke; Corbin & Strauss, 2008). By engaging in literature at this point, I could practice bracketing, or *epoche*, of subjectivities to maintain sensitivity to the interview data (Corbin & Strauss, 2008; Morrow, 2005). The nature of questioning during this process was how concepts were similar, or different, to those discussed in the literature (Corbin & Strauss, 2008).

Phase 5: Defining and renaming themes

By this stage of thematic analysis, central themes and their associated sub-themes had been identified and allocated working titles. However, it is through this phase that nomenclature used in the final analysis was decided. Clarification of theme and sub-theme names was undertaken and generation of a description of each theme and sub-theme developed during this phase. This was achieved by analysing data within each theme, identifying the essence of each theme and determining which aspects of that data each theme captured (Braun & Clarke, 2006). A detailed

analysis was written and consideration given to how each theme fits into the overall understanding of the data in relation to the research questions (Braun & Clarke, 2006).

The defining and naming phase involved consulting with a number of professionals from the multi-disciplinary CF care team at PMH. This served to triangulate data interpretation thereby ensuring credibility of identified themes and sub-themes (Vicary & Bishop, 2005). Compelling statements in the form of vivid quotes were carefully selected and intentionally collated to convey the *essence* of a theme and its associated sub-themes presented in the next section (Braun & Clarke, 2006). The consistency and truthfulness in this process resided with my ability to link abstracted themes and sub-themes drawn for the dataset, bringing alive for the reader an in-depth sense of how parents whose children have CF construct meaning of their child's early disease surveillance.

Phase 6: Producing a report

The goal of thematic inquiry is to illuminate the essential, structural qualities of a particular experience (Becker, 1992). This was achieved through the resultant set of themes that were derived from the parents' interviews, the research questions, theoretical frameworks and philosophical assumptions. A qualitative approach to analysis consisted of identifying an experience to explore, acknowledging and positioning my own assumptions and biases, collecting data from individuals who have experienced the phenomenon, analysing collected information by reducing it into relevant assertions and quotes, and developing a *textural* description (i.e., what parents experienced), a *structural* description (i.e., which contexts or situations have typically influenced or contributed to parents' experiences of their child's early disease surveillance) and also a combination of the two to convey an overall essence of the experience (Creswell, 2007; Moustakas, 1994). Willig (2001) suggested a format of structured themes and sub-themes with their descriptions and quotations to illustrate each theme.

The next chapter details the outcomes of this process. Whereas the previous chapter explained what scientific rigour is, its importance in qualitative research and the strategies used to achieve it, the next section details the activities I undertook to ensure rigorous techniques were adhered to.

4.7 Scientific rigour

The discussion will now turn to how rigour was achieved according to different stages of the research; data generation, coding and analysis, and presentation of findings, beginning with a short discussion of strategies that occurred throughout the entire study (Lincoln & Guba, 1985).

4.7.1 Throughout the research process.

The primary strategies applied continuously to my research were that of reflexivity (Patton, 2002) and bracketing (Polit & Beck, 2014): reflexivity during research design, data collection, analysis and interpretation; and bracketing during data collection, analysis and interpretation. Applying and

maintaining reflexivity and bracketing ensured extraneous influences and preconceived notions were minimised in the research process.

Throughout analyses, an audit trail (Charmaz, 2000) was created to enhance scientific rigour by ensuring interpretation followed a logical and conclusive path. An audit trail is documentary evidence for neutral experts or peer reviewers to review and verify the path the researcher followed from raw textual data to results. It is used to establish the rigour of a study by providing the details of data analysis and some of the decisions that led to the findings (Wolf, 2003). According to Rodgers and Cowles (1993), data for an audit trail can be classified into six categories: raw data, data reduction and analysis products, data reconstruction and synthesis products, process notes, materials relating to intentions and dispositions, and instrument development information. The audit trail then becomes part of the confirmability and dependability process.

4.7.2 Data generation.

At the point of data collection, aspects of ensuring rigour of qualitative research include representativeness of sample, sampling technique, and researcher reflexivity (Kitto et al., 2008; Mays & Pope, 2000; Morrow, 2005). I endeavoured to ensure rigour by explicitly describing the way my research was conducted including detailing methods with regard to accessing and recruiting participants, and developing rapport and trust with them. How ethical considerations were addressed also ensured rigorous research methods were employed (Kitto, et al., 2008; Liamputtong & Ezzy, 2005).

A critical moment in the qualitative research process is engagement with participants. Therefore, prolonged engagement is the investment in sufficient time to achieve the purposes of the research. I addressed this requirement by spending time learning about parents' experiences through the interview process and was directed by data saturation. Consequently, there was a point at which I was able to establish that the phenomena under investigation had been adequately explored, and was therefore confident that the data were representative of the phenomena. Prolonged engagement with parents was also achieved in Western Australia by spending time in the CF clinic prior to the interviews and introducing myself and the research to parents. Establishing rapport and trust in this way ensures credibility and authenticity. The conceivable reliability and quality of data collected and intimate familiarity with the participants and their worlds (Blumer, 1969) is the process through which the researcher can construct reality (Denzin & Lincoln, 2005).

An additional process for ensuring adherence to rigorous research methods during data collection involves creating personal reflections after completion of each interview. Field notes are a form of reflective writing (Charmaz & McMullen, 2011), which can be later examined as part of analysis, both as a reflection on the interview as well as part of reflexivity and *epoche*. By ensuring I reflected upon interviews, as a process and as an experience, credibility and dependability of

interpretation is strengthened because of additional and extraneous information to the interview that can verify its accuracy. Furthermore, when field notes are considered as part of data analysis, transferability of interpretation is achieved because other researchers can determine usefulness of findings to a population by examining how the researcher reflected on interviews and determining their effects on analysis. Therefore, possessing additional information beyond the findings themselves allows independent researchers to determine for themselves the accuracy in extrapolation of findings to other populations or contexts.

A number of other techniques described in the sampling and procedure section of this chapter also refer to strategies of rigorous research methods. Firstly, audiotaping of interviews and verbatim transcription of interview data ensure credibility and authenticity of data because of the strict adherence to accuracy in recording data. Secondly, data saturation means that credibility can be assigned to findings within that population because of the determination that no new data was being generated, therefore ensuring credibility in the findings. On a final note, previous qualitative experience ensures rigour during the interview process, specifically credibility and authenticity because the quality of information obtained is highly dependent on the interviewer (Patton, 2002). I was able to draw on my previous qualitative research experience, which gave me confidence to conduct interviews and ask questions, both as prompts and clarifiers.

4.7.3 Data coding and analysis.

To further ensure rigour of the research, how findings were generated was made explicit, affirming consistency across time and analysis techniques, making the process as repeatable as possible (Morrow, 2005; Patton, 2002). This was primarily achieved by tracking the research process from design to interpretation through use of a reflective journal detailing chronology of research activities, processes and influences (Flick, 2006; Morrow, 2005). The journal was used at this stage of the research as a critical source of interpretative understanding as concepts were dissected and explored (Bazely, 2009), which ensured credibility of interpretation because of the reflexive nature of the process. Confirmability of interpretation is also inherent in use of a self-reflective journal because of its ability to provide contextual decisions to generation of interpretation. Essentially, reflexivity refers to how data are co-constructed during collection through the influence of both researcher and participant, and offers both a methodological and interpretational log of research decisions (Corbin & Strauss, 2008). I was therefore able to maintain a level of sensitivity to ways in which I, acting as an instrument of data collection, shaped the collected data (May & Perry, 2011; Mays & Pope, 2000). Engaging in this reflexive process allowed acknowledgement that research is never completely objective, and that quality research should result in outcomes that represent as closely as possible the context being researched as opposed to the values and biases of the researcher (May & Perry, 2011; Morrow, 2005)

Assurance that research has presented adequate data and findings is largely dependent on strategies employed during data analysis including verification steps. A number of verification steps were included in the analysis process; disconfirmation and discrepancy in cases, constant immersion in data, member-checking, inter-coder checking, peer debriefing, and coherent theme development. Each of these verification strategies will be briefly described.

By discovering and examining disconfirmation and discrepancy in cases, also known as negative case analysis (Patton, 2002), I was able to make repeated comparisons and revise key assertions and categories until they accurately reflected the experiences of parents (Morrow, 2005). In qualitative research, negative cases cannot be ignored because they can provide hints that might explain what is happening for the larger sample (Bazely, 2009). Development and refinement of hypotheses occurred here because by looking at all cases, existence of ideas about the data are being tested, and refined by adjusting ideas. A conscious effort to search for disconfirmation also helped to combat any tendencies on my behalf to seek confirmation of preliminary findings and categories (Kitto, et al., 2008; Liamputtong & Ezzy, 2005; Mays & Pope, 2000; Morrow, 2005). This process was also aided through constant immersion in the data from; data collection, transcription of interviews, and continued and repeated readings of the transcripts, listening to the audio recordings, and reviewing notes and other related data (Flick, 2006).

Member-checking became important at this stage of the research because, as Lincoln and Guba (1985) highlight, this is the most critical technique for establishing credibility, which refers to determining accuracy of findings and interpretations by taking specific interpreted descriptions back to the participants for verification and clarification. Parents were consulted to ensure that reconstructions of a reality were precise and truthful representations of their multiple realities. This was achieved by conducting in-depth follow-up telephone interviews with five parents for validation of interpretation, as well as sending a brief overview of findings in the form of a newsletter to all parents providing an opportunity to respond. At this stage of the research, members from the multi-disciplinary CF care team at PMH were consulted for relevancy of findings to clinical practice and accuracy of findings based on their anecdotal experiences with families. From engagement with the multi-disciplinary care team and further engagement with parents, truthfulness of findings and uniqueness of concepts was substantiated; addressing requirements of confirmability.

Employing inter-coder checking became an important strategy in verifying data analysis. Inter-coder checking is a process of determining congruence between two or more independent people about the data's accuracy, relevance or meaning by employing an external auditor to code and interpret a portion of the data (Lincoln & Guba, 2000). To assess inter-coder reliability, a fellow research colleague was asked to code 10 interviews. Comparisons of coding techniques and

categories determined accuracy of coding and corroboration of interpretation, addressing requirements of credibility and confirmability.

Interaction with an independent person for peer debriefing during data analysis was another verification strategy. Continual interaction with a peer throughout the research process, both about the research as well as about my role within the research, gave multiple perspectives of the data, assisting me to develop and confirm ideas and interpretations. This is where my biases were questioned, meanings explored and interpretations clarified, consequently ensuring credibility in the process and therefore also in the findings. Debriefing in this way also propels discussion about initial opportunities to develop working hypotheses. My choices and subsequent decisions were subject to rational and logical explanation of their position and suppositions (Lincoln & Guba, 1985). An example of the progress made within debriefing sessions was how my interpretation of parents' knowledge of their child's CF disease progression positively influenced their everyday life worlds beyond the immediate trauma of gaining this insight.

Lastly, verifying the data through theme development included grounding categories and themes in examples and organising categories to provide coherent understanding of how they fitted together so as to provide a data-based narrative an underlying structure to organise the phenomenon for the reader (Elliot & Timulak, 2005; Morrow, 2005). Moreover, the results should exemplify symmetry of interpretation and quotation by striking a balance between the researcher's interpretations and supporting evidence by way of quotations from participants. Providing actual words of parents' accounts therefore became essential to informing the reader that the interpretations made were grounded in the lived experiences of the participants. Furthermore, providing a clear account of processes from research design and data collection to analysis through use of diagrams to aid understandings, explicitly outlining procedural information, and engaging in a number of peer auditing procedures, increased dependability of the research (Flick, 2006; Guba & Lincoln, 1994; Mays & Pope, 2000; Seale, 1999).

4.7.4 Presentation of findings.

By studying a group of people intensively so as to understand their life-world of the phenomena under investigation, there will be findings that are unique to that sample (isomorphism), but detail can be extrapolated to other similar populations and/or within similar contexts, resulting also in transferability of the findings (Patton, 2002). Provision of rich, thick descriptions as adequate evidence of any assertions, such as provision of appropriate quotes to support themes (Braun & Clark, 2006) and by providing working hypotheses together with a description of time and context of when such suppositions were found to be valid (Lincoln & Guba, 1985), achieves transferability because it gives the reader the opportunity to determine the transference of findings to similar individuals or contexts. Therefore, by providing extensive and

diverse sources of interpretation, the function of transferability occurs when the reader has adequate information to determine it. When the research report is sufficiently detailed for the reader, they can judge whether findings are applicable to similar settings and how transferable, relevant, and useful the findings are to the particular context and phenomena under investigation (Kitto, et al., 2008; Mays & Pope, 2000). By employing strategies discussed in this section, I have presented sufficient information so the research could be considered relevant, transferable, and useful to the research and clinical context of early surveillance and CF care more generally.

Accordingly, quality or validity in qualitative research is, to a certain extent, paradigm-bound with standards of trustworthiness that emerged from paradigms such as those in quantitative research (Kitto, et al., 2008; Lincoln & Guba, 2000; Mays & Pope, 2000; Morrow, 2005; Seale, 1999). However, particular qualities are also indispensable regardless of the research paradigm such as reflexivity, and issues related to adequacy of data and to interpretation and presentation of data (Alvesson & Sköldberg, 2009; May & Perry, 2011; Morrow, 2005). Accordingly, as in quantitative research, rigour and quality in qualitative research achieved through systematic, self-conscious research design, and data collection, interpretation and presentation was required (Flick, 2006; Mays & Pope, 2000). By using those strategies and techniques outlined above, I achieved a transparent and thorough account of the research process and associated findings.

4.8 Summary

The methods chapter has outlined qualitative strategies and techniques I used to achieve my research aims and address my research questions. My sample, and how I recruited them, was detailed, along with ethics considerations I made throughout the research process to ensure quality of data. More importantly, ethics considerations ensured ethical conduct of the research and appropriate consideration for my participants. Lastly, data collection procedures and data analysis techniques were outlined. The format of presenting structured themes and sub-themes is utilised in the next chapter where a composite description of parents' experiences, along with a discussion of findings and interpretations are presented. Below is an overview of the theoretical and procedural frameworks guiding the research processes that have been explained throughout the chapter.

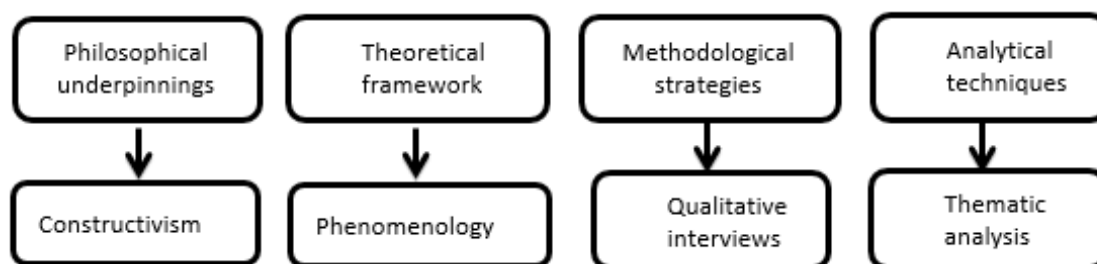


Figure 5. Overview of theoretical and procedural frameworks guiding the research

CHAPTER 5**Findings: The lived experience of parenting children with CF in the context of early surveillance**

Chapter 5 presents the findings and interpretations of this research. The aims were to explore and interpret parents' experiences and psychological constructions of parenting. Further aims were to explore how parents cope and what meanings were attributed to their experiences. Salient findings from analysis of the data are the commonalities in experiences and constructed thoughts about parenting children who undergo early surveillance for CF lung disease. Accordingly, the chapter presents the temporal experience from diagnosis through to day-to-day management of CF within context of early surveillance, through the first 7 years of life. The chapter begins by outlining the major themes and sub-themes identified from the data analysis process. Each theme within this chapter commences with an outline and summary of the identified major theme and continues with a discussion of related sub-themes and presentation of excerpts from participants' responses to demonstrate the findings and interpretations. Throughout each section, current research and literature relevant to the identified themes and sub-themes are integrated into the discussion. Interpretation of findings in light of applied literature consolidates their place within the lived experience of parenting children with CF in the context of early surveillance.

We must be willing to let go of the life we have planned, so as to have the life that is waiting for us...Joseph Campbell

5.1 Identifying Themes

A primary aim of my research was to explore how parents of children with CF residing in Perth, (Western Australia), and Melbourne (Victoria) experience and construct parenting within context of early surveillance for CF lung disease. Further aims were to explore how parents cope and what meanings were attributed to their experiences. Lastly, my research aimed to review all participant groups' experiences collectively to identify any unique commonalities and/or differences in experiences and constructed thought. Through the process of thematic analysis (as outlined in chapter 3: Methods), a substantial finding from analysis of the data was the commonality in experiences and constructed thoughts about parenting a child with CF within the context of early surveillance across the age range of children and across sites. Before I introduce you to parents in my research, it was evident that their overall experience of CF was inextricably tied to their early surveillance experience (which I will explain in due course). Therefore, to understand parents' experience of early surveillance was to also account for parents' experiences of CF. Therefore; whilst I analytically separated these two elements of consciousness, I did not attempt to experientially separate them because they are part of the same thing.

Within the process of analysis, it became apparent that themes naturally cohered in such a way that parents' experience preceded from diagnosis of their child's CF and their introduction to early surveillance through to their everyday regular and routine experience of parenting in the context of early surveillance. This was followed by distinct experiences and outcomes of early surveillance. Accordingly, presentation of themes will follow this emergent structure.

Five major themes with related sub-themes represent the emergent structure: redefined expectations and reimagined identity, redefined reality (consisting of sub-themes - collapsed and redefined expectations and *new normal*); understanding the unknown and understanding uncertainty (consisting of sub-themes – understanding CF and understanding early surveillance for CF lung disease); good days and bad days; fluctuation between positive and negative outlook on life (consisting of sub-themes – bad days, good days and fluctuation between good days and bad days); early surveillance is a significant event (consisting of sub-themes - indicator of treatment efforts and examination of parenting competence, parental outcomes directly from early surveillance and fear from diagnosis exacerbated at each annual review can result in anxiety); and early surveillance is in the best interest of my child (consisting of sub-themes - perceived clinical benefit to child health and cognitive dissonance results in anxiety and/or ambivalence). Refer to Table 4 below for an overview of themes and sub-themes. Consistent with the first three research questions, identified themes represent salient factors contributing to parents' experiences and constructions of parenting a child

with CF in context of early surveillance, as well as their attributions of meaning to early surveillance. The fourth research question is addressed throughout themes where coping with diagnosis, treatment and early surveillance is considered. Each of these themes and their related sub-themes is discussed separately in the next section.

Table 4. *Major themes and related sub-themes*

Major theme	Related sub-themes
Redefined expectations and reimagined identity; redefined reality	Collapsed and redefined expectations New normal
Understanding the unknown and understanding uncertainty	Understanding CF Understanding early surveillance for CF lung disease
Good days and bad days; fluctuation between positive and negative outlook on life	Bad days Good days Fluctuation between good days and bad days
Early surveillance is a significant event	Indicator of treatment efforts and examination of parenting competence Parental outcomes directly from early surveillance Fear from diagnosis exacerbated at each annual review can result in anxiety
Early surveillance is in the best interest of my child	Perceived clinical benefit to child health Cognitive dissonance results in anxiety and/or ambivalence

5.2 Interpretation of phenomena – what is the ‘lived experience’ of parenting children with CF within the context of early surveillance for lung disease?

This chapter is a description and interpretation of the lived experience of parenting a child diagnosed with CF within the context of early surveillance for CF lung disease. Within this chapter, I identify, describe and explain why discovered phenomena are present for parents. Where relevant, I will explain how these phenomena function, for example, how situations encountered might affect parental characteristics and outcomes. The current research landscape investigating psychological outcomes for parents in paediatric chronic disease is such that resiliency is a key beneficial concept for parents and their families due to recent focus on resistance, rather than risk factors. Therefore, I highlight the contribution of early surveillance to parenting a child with CF by identifying risk and

resistance factors that may contribute to adaptation. A process to adaptation is to build and maintain resilient parents and families for future burden of CF, which is continued into the next chapter. The first theme begins with a description of parenting a child diagnosed with CF.

5.3 Redefined expectations and reimagined identity; redefined reality

Parents whose child is diagnosed with CF, typically within the first 3 months of life, confront the reality of suffering and the premature death of their child (e.g., Carpenter & Narsavage, 2004; Priddis, et al., 2010). They can experience a multitude of negative reactions and responses including, but not limited to shock, disbelief, anxiety, guilt, fear, and depression (De Monestrol, et al., 2011; Fallowfield & Jenkins, 2004; Glasscoe, et al., 2007; Moola, 2012; Mullins, et al., 1991; Quittner, et al., 1992). Most studies have concentrated on maternal reactions to diagnosis with less consideration given to fathers. The starting point for my analysis is that new parents - both mothers and fathers - operate with expectations about what being a parent means, of what having a family entails. These expectations hold one's hopes, aspirations and fears. Once parents are informed that their child, in whom so many of their expectations are invested, suffers from a progressively fatal condition, home treatment management and clinical care regimens are swiftly explained. The initially incomprehensible diagnosis and overwhelming amount of information about treatment and care affects parents' expectations, their optimism, their hopes for the future and their perceptions of themselves as parents in such a way that causes collapse and redefinition of their expectations. This section relays these accounts through identification of two sub-themes namely; collapsed and redefined expectations and *new normal*.

5.3.1 Collapsed and redefined expectations.

Diagnosis of CF in a newborn literally dissolves initial child, parenthood and family expectations (Bluebond-Langner, 1996; Carpenter & Narsavage, 2004; Jessup & Parkinson, 2010; Priddis, et al., 2010). Upon diagnosis, emotional despair and heartache for the child, dismantling of parent identity and collapse of family expectations ensue. Parents' expectations for parenthood and family life disintegrate with the diagnosis for their infant, which means that parents were left with no expectations for their child and their family.

To illustrate, a Western Australian mother of a child aged 4 years reflected on her child's diagnosis when she stated '*you just don't see that life can have any level of normality when you're first diagnosed. You're worried about all of these things you're being told you have to do, you don't know how you're going to manage it all. You don't know what any of this means for your family or your child's future.*' The following passage from another Western Australian mother whose child was 5 years old further reflected initial despair experienced by parents about loss of expectations for her child and family, '*you just see a little baby who you're being told is sick, but you don't really know how sick, so you automatically think the worst...I think when kids are first diagnosed, it robs you a*

little bit because it really is a huge shock which shattered our lives and what we expected for our future.'

Therefore, collapse of expectations about parenthood and family life is a cognitive process that threatens parents' expectations and reality irrevocably. The threat functions to diminish parents' ability to follow their expected family life path and to believe in what they knew about parenthood. Jessup and Parkinson (2010) reported a similar process when they suggested that the future of the family was initially cancelled at diagnosis. Parents experience loss of their previously taken-for-granted world. They fear the potential loss of their child and fear failing their child through their inability to conduct the required home treatment regimen thrust upon them at diagnosis (Cohen, 1993; Jessup & Parkinson, 2010).

Parents are required to learn about the time-consuming and often difficult home treatment regimen soon after the baby's diagnosis. When parents begin to experience the intensity of home treatment and the time required to complete it, their expectations about what family life will be like are further altered, particularly what their parental role will entail (that is, they are adopting a medical role) (Glasscoe & Smith, 2011; Tong, Lowe, Sainsbury & Craig, 2010) and what their child's life will be like with these daily demands. A Western Australian mother of a 2 year old child spoke about her altered parental role, *'you know the parents have to become experts on how to deal with it and how to manage it, and that means that you have to develop quite a detailed understanding of the lungs and the disease and stuff. So you need training basically.'* Envisaging what a child's life may be like was expressed by a Western Australian father of a 4 year old child who stated *'I remember when there was about a 3-month period when you (mother) were trying to teach XXA pep (physiotherapy) and she screamed every time. I remember asking myself 'what kind-of life is our child going to have?'* After collapse of expectations surrounding diagnosis of CF, introduction of intensive home treatment management is likely to contribute to reconceptualisation of expectations of what family life will be like.

Diagnosis of CF in an infant changes the ideality of parenthood and family life. Using more conventional terminology from the CF clinical nurse coordinator at PMH (E. Balding, personal communication, February 27, 2014), "a CF diagnosis moves the goal posts of life for our families." This refers to the notion that parents' preconceived realities and expectations are shifted by knowledge of their child's condition because everything that was known and expected about parenthood and family becomes threatened (Jessup & Parkinson, 2010). The threat is extensive; it includes their baby (and their future), their ideals, their current understanding of parenthood; life as they know it and expected it to be. The *known and expected* becomes threatened because parents' expectations of the traditional (*normal*) life trajectory and what they expected of parenthood and family life are taken away.

Qualitative accounts of parenting children with leukaemia (Kars, et al., 2008), congenital heart disease (Rempel, Ravindran, Rogers & Magill-Evans, 2013), Duchenne muscular dystrophy and Rett syndrome (Gravelle, 1997) reported similar psychological experiences. Accepting and adjusting to the inconceivable reality of their child's condition was the first process for parents. This involved two sub-processes; realising the precariousness of survival and adjusting expectations. Constantly living with pressure and stress of the unknown in terms of their child's disease trajectory of longevity of life was another process parents from these studies experienced.

Part of expectations about parenthood includes the substance, and meaning, of becoming a parent. Moreover, perceptions of both parenting practices (i.e., responsibilities) and parenting functions (i.e., roles) are inextricably tied to how people perceive themselves as parents (i.e., parent identity). Parental identity refers to how an individual views him or herself in relation to their child (Maurer, Pleck & Rane, 2001). Therefore, parents' beliefs and values about their role in their child's life will contribute to how they view themselves in relation to the child. Damage to parent identity due to paediatric chronic illness is a commonly cited outcome in other chronic conditions (Bowes, et al., 2009; Cashin, et al., 2008; Marshall, Carter & Rose, 2009; Patrick-Ott, 2011; Waite-Jones & Madill, 2008), but has been insufficiently investigated in CF. Some qualitative studies briefly mentioned negative outcomes of CF on parent identity but failed to uncover any specific pathways (Hodkinson & Lester, 2002; Lioffi & Evans, 2005). With support from the literature, I have interpreted two reasons why parent identity may be damaged by a CF diagnosis.

Firstly, reports in the literature describe how parents can feel guilty about their child's condition, likely because of the genetic nature of CF (Hodkinson & Lester, 2002; Marvin & Pianta, 1996; Priddis, et al., 2010; QUITTER, et al., 1992), meaning that parents felt as though they were responsible for *giving* their child CF (Havermans, et al., 2015). By accepting responsibility of loss due to chronic illness (in this case, the child's loss), guilt can ensue (Carpenter & Narsavage, 2004; Stephenson & Murphy, 1986). Secondly, parents must come to terms with the forced reality of parenting a chronically ill child, irrevocably altering what parents envisaged parenthood would be like (Glasscoe & Smith, 2011; Patrick-Ott, 2011; Tong, et al., 2010). Contributing to loss of ideals about parenting is the requirement on parents' behalf to complete intensive home treatment, further altering ideals about the substance and function of parenting.

The latter point is particularly about the substance and function of the role in relation to parenting a chronically ill child. That is, parents are adopting a medical role that requires them to conduct medical treatment and monitor child health. It appeared that, due to demand of home treatment, care-giving formed a significant part of parenting a child beyond usual parenting tasks, which has been shown in both paediatric CF (Afonso, Gomes & Mitre, 2015; Lioffi & Evans, 2005; Slatter, Francis, Smith & Bush, 2004) and other chronic illness literature (Bjork, Wiebe & Hallstrom,

2009; Cashin, et al., 2008; Heaton, Noyes & Sloper, 2005; Kirk, Glendinning & Callery, 2005; Pelenstov, Fielder & Esterman, 2015; Tong, et al., 2010). The caring role of chronically ill children often dominates the parenting role because of the need to provide ongoing care for the child (Kirk, et al., 2005). Therefore, domination of the carer role over the parent role following diagnosis may result in a loss of the traditional parenting role and damage to self-schemata about perceptions of parenting until such time that parents can redefine - reimagine - their identity.

O'Connor's and Barrera's (2014) model of identity disintegration and reintegration in paediatric chronic illness facilitates speculation about potential mechanisms through which parent identity could be damaged and re-imagined. They conceptualised negative perceptions of parenthood and the parental role and identity as identity disintegration, and the process of identity reintegration as a positive reframing of parenthood and the parent role and identity. This framework provides a contextual layer to psychological theory about self-schemata (Piaget, 1952), and their role in identity, because it outlines what happens when parents assimilate information of CF into their parental role following their child's diagnosis.

How parent identity is repaired for parents in my research is supported by this theory and demonstrated in Slotter's and Gardner's (2014) study, where evidentiary support for the threatened self-concept generates a reimagined self-concept. Together, O'Connor's and Barrera's (2014) theory and the qualitative findings of Slotter and Gardner highlight that encoded information supporting a threatened self-concept aids in positively reframing parent role and identity. As parenting a child with CF can result in conflict between roles of carer and parent (Glasscoe & Smith, 2011), understanding mechanisms for adverse parental outcomes that may be amenable to intervention is essential to ensure that parents are fully supported.

To summarise, parents must navigate their way to understanding and subsequently accepting their child's diagnosis to preserve their own psychological well-being, and to be present for their child. Negotiation towards understanding and acceptance involves a process of resetting, rebuilding and redefining parenting and family expectations. A Western Australian mother of a 2 year old child exemplified this process when she stated '*when you are first told, when I was first told you kind-of just go "oh (deflates)," and you're whole plan collapses, and then I think you just start the plan again in some kind-of re-moulded form.*' This is achieved through psychological absorption and eventually acceptance of the diagnosis, though it is conceivable that parents come to accept their child's diagnosis over various time frames and with varying degrees of effectiveness (Priddis, et al., 2010).

New normal is the classification of the experiential and practical outcome of redefined child, parent and family expectations. It refers to the concept of parents' acceptance and embracement of a different life to what was expected before the intrusion of CF. *New normal* is the result of

knowledge acquisition and experience accumulation, that is, expectations are redefined based on understanding and experience, and then parents assess the truthfulness and achievability of those redefined expectations through further understanding and experience. *New normal* is the final outcome of redefined expectations.

5.3.2 New normal.

Through the experience of moving from collapsed to redefined expectations, parents develop a new sense of *normal* life, with CF and its management as additional components. Reconceptualising what is perceived as normal is a common theme in the chronic illness literature, as demonstrated by a metasynthesis (Nelson, 2002) of qualitative studies about mothering a child with a disability or chronic condition. Reports have demonstrated that parents of children with CF strive to maintain a degree of normality within the family domain (Liossi & Evans, 2005, Moola, 2012). Admi (1996) posited a model for living with CF that placed maintenance of a normal life at its center. Carpenter and Narsavage (2004) described how parents return to normality after the devastation of a CF diagnosis. Bluebond-Langner (1996) reported that redefinition of normal begins in the year following a CF diagnosis, and Glasscoe and Smith (2011) published a case study that detailed how a mother's awareness of CF and the treatment regimen became part of their everyday lives for her and her child.

The conceptual structure of what I have termed *new normal* is relevant for any family or person who is required to *live with* a permanent unexpected change to their everyday life world; to the essence of their existence. It refers to a new way of living that must be negotiated and eventually accepted as part of their redefined way of living. Therefore, *living with* a permanent unexpected change refers to going beyond the change simply existing in one's life, but encompasses its successful management and acceptance *as part of* one's life.

Jessup and Parkinson (2010) likened this event to parents "needing to find their bearings and reconstruct their lives in the context of a new, unanticipated scenario (pp. 355)." To that end, the conceptual basis of *new normal* for parents in my research, in the context of CF diagnosis, refers specifically to how those processes of negotiation and establishment of a reimagined sense of identity (including roles and responsibilities) and redefined expectations reach a conclusion. For example, a Western Australian mother of a 3 year old child reflected on her transition from devastation of diagnosis to the normality of the present:

it's not all bad. I think to begin with it is, but then it gets so much easier from those first days where I was filled with negative thoughts, to one year on and then 2 years on, it gets easier... it gets easy and there is a lot of normal because you just don't see it when you're first diagnosed. So just to know that all the things that you're really scared of

become a part of your day-to-day life that you don't even notice it. You manage, you just manage.

Similarly defined in the chronic disease literature, normalisation for parents is described as both a cognitive process of defining their own lives and a set of behavioural strategies for managing their child's condition (Knafl, et al., 2010; Rehm & Franck, 2000; Robinson, 1993). Furthermore, normalisation has been referred to in the CF literature as integrating CF treatment into, yet maintaining, family routines (Pfeffer, et al., 2003) including cognitions and actions that portray treatment as a necessary routine within that new sense of family life (Glasscoe & Smith, 2011; Grosssoehme, et al., 2014; Hodgkinson & Lester, 2002; Jessup & Parkinson, 2010). A landmark study focussing on psychological outcomes of CF in the family environment by Bluebond-Langner (1996) identified strategies used to manage the early stages of diagnosis that offered avenues for achieving a sense of normality including routinisation of treatment into daily life, redefinition of what is perceived as normal and reconceptualisation of the future.

Two processes simultaneously occur that function as mechanisms for negotiation of parent and family expectations. Firstly, parents are gaining knowledge about CF, the condition, its management, their role as parents of a child with CF; their individual and collective futures. Secondly, parents are beginning to have experiences of each of these aspects of their child, providing evidence of their ability to meet these new expectations and requirements. Whilst absorption of this type of knowledge, and early experiences of CF, can be initially overwhelming and devastating, it begins to give parents a framework upon which to build understanding of their child's condition. Very few parents have such a framework at diagnosis due to their lack of awareness that the condition is possible within their family, as well as the broader lack of public awareness of CF at the societal level.

Keeping in mind the supposition that early surveillance is inherently linked with CF, it too becomes part of the *new normal* for families in my research. The introduction of early surveillance for CF lung disease soon after diagnosis, along with parents' perceived hasty requirement to notify their involvement, results in the amalgamation of these two entities as part of the same experience. Whilst the disease of CF can be distinguished from the experiential encounter of early surveillance, their distinction in terms of thinking of one without the other is not possible. This is the inextricable link between the two. Therefore, accepting and embracing life with CF for parents in my research included accepting and embracing early surveillance as part of their *new normal*.

Since early surveillance is introduced so close to diagnosis, parents know no other reality of CF and so have no alternative way of thinking for comparison, and therefore seem to accept what is presented to them as a routine part of CF (Glasscoe & Smith, 2011). A Victorian father of a child aged 12 months discussed how he viewed early surveillance in relation to his child's CF; note the tense in

the quote moves from the past to the present and therefore seems to define the status quo now, *'well it's part and parcel really isn't it? For us, we've known CF with the program, never without it. We've never known any different so it's not like we can compare to not being part of the program or whatever, know what I mean. This is our experience of CF and we want to know the information we get from being part of it (early surveillance). Actually, I can't really imagine not getting this info about my kid's health, it's valuable because it can help her have a better life.'*

In early stages of diagnosis and establishment of the *new normal*, parents know what early surveillance entails pragmatically and what is expected of them and their child. However, at this point there is a lack of personal experience. In these early stages when parents are negotiating and accepting a redefined sense of family life, there is little consideration of how they will manage their personal early surveillance experience. Until such time that parents can have their first encounter with early surveillance, they cannot start to understand how to manage the personal experience. This understanding from experience will be discussed comprehensively in the next theme, because understanding from experience emerged as an interpretation of the overall parental experience of early-life CF management.

To finish outlining parents' new sense of normal, a brief reiteration is warranted of the mechanisms through which parents come to their redefined realities. A redefined sense of personal and family life emerges through understanding: an understanding that occurs over time with knowledge and experience. Understanding comes from gaining knowledge about CF and early surveillance. Accumulation of both knowledge and experience are the essence of accepting and embracing *new normal*. Therefore, a sense of *new normal* is the outcome of the transition from devastation, through negotiation, then accepting and achieving a new, redefined sense of normality. A Victorian mother of a 2 year old child described how she transitioned from devastation of her child's diagnosis to a redefined sense of normality, *'perhaps I was in shock. It took me a long time to adjust to XXX's diagnosis but now it's just kind-of normal really.'*

Accordingly, the experiential basis of *new normal* for parents in my research - the meaning of *new normal* - is that intra-psychic processes of development and negotiation have reached a point where parents feel comfortable and confident in those redefined, newly established expectations and identities, roles and responsibilities for treatment management; embracing a redefined reality and adapting to their child's diagnosis. This appears then to be a function of time and experience; time to digest and accept the diagnosis, time to develop and establish redefined expectations, time to reimagine, accept and embrace the parental self including role, responsibilities and identity, time to routinise CF treatment regimen and early surveillance for CF lung disease and time to acquire confidence and comfort in these redefined constructs through experience of them. Successful

management of these factors could represent adaptation to CF (which will be discussed in due course).

Whilst maintaining a new sense of normality appears a dynamic process, the cross-sectional nature of the research meant I was not able to discover whether maintaining a *new normal* is a dynamic process most likely because of differing treatments, varying levels of child health, hospital admissions and disease progression that need to be continuously merged into family, as suggested by Hewitt-Taylor (2009) and MacDonald and Callery (2007). Stratification of child age in the my research identified that parents of children of varying ages perceived they had achieved a redefined sense of normality, suggesting a dynamic process of redefinition due to different events and circumstances for these families needing to be incorporated into the family.

Bluebond-Langner (1996) attests to a dynamic process of redefinition to accommodate the child's condition and treatment. For example, she reported that parents were able to incorporate their child's first hospitalisation within the parameters of normal as they had redefined them by renewing their efforts to establish the situation within the realm of their *new normal*. However, it remains less understood whether, and how, further events such as child health deterioration and/or increasing hospital admissions might disrupt early conceptualisations of redefined parent and family. Parents in my research are thrust into a situation where child health deterioration and increasing hospital admissions is a product of the early surveillance experience that may influence those early conceptualisations of a redefined sense of reality.

In review, diagnosis of CF causes disruption in early parenthood by causing collapse in expectations associated with parenting because parents do not know, or have little knowledge, about CF and its management. Collapse in expectations of family life and what it means to be a parent can result in psychological distress including frustration and guilt of having an ill child, along with fear of the future and despair about the present. Collapsed expectations must be redefined with CF included in a new sense of family life – *new normal*. Negotiating a CF diagnosis and its treatment management into parent identity and expectations (parent and family) results in redefinition of these components of family life. Parents of children all ages expressed their desire to engage with other parents to gain perspective not only on clinical and genetic aspects of CF, but of the personal experiences of CF and early surveillance. As early surveillance is generally introduced to parents concurrently with diagnosis, they become part of the same thing. These are two sides of the same experience. Familiarity with a redefined reality of CF and early surveillance confirms the structure of *new normal*. Below is a diagrammatic representation of intra-psychic processes parents followed to reach the outcome of a redefined sense of reality – *new normal*. This theme identified both understanding and experience as mechanisms for negotiation of expectations and parent

identity. The next theme will explain how knowledge (per se and from experience) facilitates understanding.

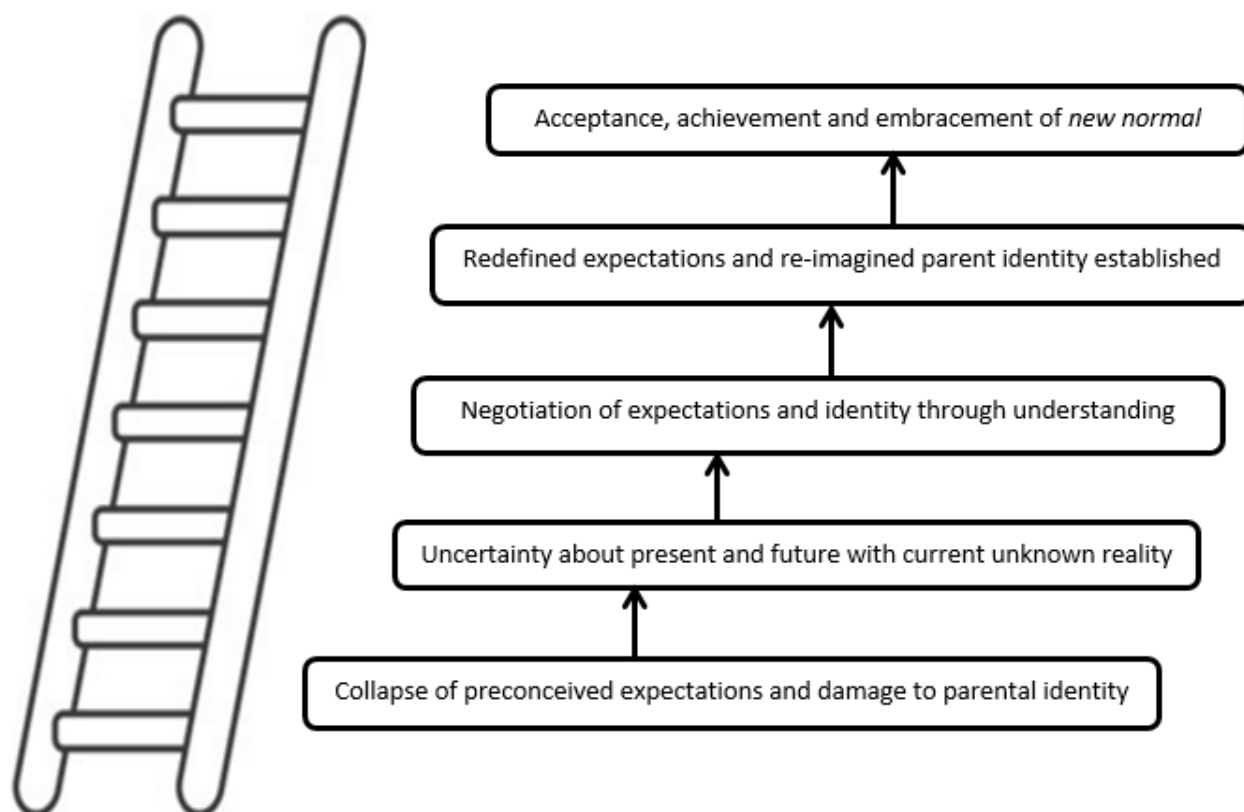


Figure 6. Representation of how parents psychologically reach an alternate sense of normal family life

5.4 Understanding the unknown and understanding uncertainty

This theme describes the journey from knowing nothing or very or little about CF and early surveillance to understanding the chronic disease and its management by AREST CF. *Understanding the unknown* refers to parents' journey from not knowing about CF and early surveillance to understanding them. Whereas *understanding uncertainty* refers to parents' understanding of the uncertain morbidity and mortality associated with CF; because part of understanding CF is understanding and accepting uncertainty in long-term health outcomes for their child.

Having little or no knowledge about CF is the dominant circumstance for parents in my research and this is consistent with other studies showing that parents of newly diagnosed infants may have heard of CF but possess no concrete understanding of this chronic condition (Massie, et al., 2007; Tluczek, et al., 2009). Lack of knowledge about CF potentiates collapse of expectations and previous knowledge about parenthood and family life. Therefore, understanding CF is of paramount importance to parents because it is understanding that enables parents to start redefining self, child and family life based on their acquired knowledge.

Essentially, it is understanding that helps parents reconstruct their collapsed life worlds. That is, *understanding* is the mechanism by which parents proceed from collapse to their *new normal*. Moreover, understanding CF and early surveillance facilitates belief and confidence in parents' ability to master their new way of life. Through the process of understanding CF and early surveillance, parents come to understand that they can achieve their *new normal*. To explore the varied aspects involved with negotiating CF and its treatment within context of early surveillance, the following sub-themes were identified: understanding CF and understanding early surveillance of CF lung disease.

5.4.1 Understanding CF.

As parents' expectations for their futures collapse, they are desperate to know they can manage their situations and their children because at the point of diagnosis, they have no sense that they are able to manage. Parents are also desperate to establish some sense of what their child's life may be like because collapse in those expectations and aspirations extends beyond their own and their families' life to the child's life. *Will they be able to play sport like other children? Will they get married and have children? Will they be able to get a job?* Projections of their children's futures such as these are questioned by parents during the early phase of diagnosis because of their lack of understanding about CF. Parents seemed desperate to claw their way back to something that they could believe and accept. This process was achieved through understanding their children's condition; how it could be managed, their prognosis and their quality of life. The following quote from a Western Australian mother of a 4 year old child exemplifies how knowledge of CF and early experiences facilitated her redefinition of expectations, *'it has got better and that comes down to experience and understanding. Like I said, those first few months after diagnosis, if you have no idea that CF is even a possibility and you're child has CF, you're hit with a sledgehammer of emotion and it's, you just, you're in a little bubble and it takes a long time for you to get out of that and I think that's what makes the difference. You suddenly start having all of these experiences, you start knowing about what's going on, that you start feeling a lot calmer about what's going on.'*

Knowledge and experience in the early stages of receiving a child's CF diagnosis allow parents to begin to come to terms with their child's diagnosis; start to make sense of it and put boundaries around it. Knowledge and experience gives parents understanding; understanding of a condition, a diagnosis that, because of lack of understanding, initially took everything about the future of parenthood and family life away. Moola (2012) described how information about the condition and experience of their child facilitated normality for the family and alleviated stress for parents. Jessup and Parkinson (2010) reported that parents redefined their family's expected future through negotiating and reconstructing their current understanding of family life after it had been threatened by a CF diagnosis by assimilating new information about CF into the expected future. That

is, knowledge about CF starts to put a limit on what a parent thinks in terms of their own, their family's and their child's futures.

Accumulation of knowledge about CF frames the devastation and gives parents a foundation to start redefining their lives and consequently giving them belief in their ability to manage their situation. It is through understanding how CF will fit into family life, and not take over family life (as first thought, or as actually happens as part of the diagnosis) that parents start to construct and harbour belief in their ability to execute and achieve what is being asked of them by medical staff. A Victorian mother of a 5 year old reflected on her transition from devastation of diagnosis to normality when she said, *'that's our life now, but you don't always feel this level-headed, sometimes you had your doubts...but that's what we've been dealt and you learn to deal with it.'* Similarly, a Western Australian mother of a 2 year old child spoke about her transition, *'I try not to make it a really big deal of it anymore...we are pretty confident now that we can handle the situation but it wasn't always like that.'* Whether parents have belief in their ability to look after their child and to master their new way of life from the beginning or whether it is a process over time, parents in my research felt they had reached some level of acceptance and achievement of their *new normal* through their understanding of CF and their belief to execute their new medicalised roles.

Accumulation of knowledge about CF and the experience of its management are tools for redefining expectations about parenthood and family life. The most salient example of this process in my research was that detailed knowledge of CF, and experience of the complex care regimen, appeared to contribute to parents' desires for treatment mastery and to their subsequent beliefs in their ability to successfully conduct treatment. Therefore, whilst parents can become overwhelmed with treatment requirements as they learn about CF care, over time they develop considerable skills (and perceived competence) in the condition's management (Bluebond-Langner, 1996; Grossoehme, et al., 2014). This process and outcome have been shown in other paediatric chronic conditions (Balling & McCubbin, 2001; Bjork, et al., 2009; Cashin, et al., 2008; Cederborg, Hultman & Magnusson, 2011; Hewitt-Taylor, 2009; Kirk, et al, 2005; Lee & Rempel. 2011; Nuutila & Salanterä, 2006; Rehm & Franck, 2000; Tong, et al., 2010). It is through gaining knowledge of the condition, and in their ability to maintain treatment, that parents develop competence and confidence in their abilities and judgements about chronically ill children (Balling & McCubbin, 2001; Cashin, et al., 2008; Cedeberg, et al., 2011; Hewitt-Taylor, 2009; Kirk, et al., 2005).

Understanding and experience of a previously unknown entity are two defining factors that provide a framework to make sense of what is unknown, so that transition to ownership and acceptance of choices in an alternative reality can take place. A similar process was reported in Bluebond-Langner's (1996) ethnographic study with parents of children with CF. The study reported that parents' ability to manage CF-related tasks increased over the first year, which in turn gave

parents confidence in their increased mastery, creating a new routine that become normalised with experience over time. By gaining knowledge, experience, confidence and competence in CF treatment management, parents are likely to manage well with the daily routine associated with CF management. From the following excerpts, it can be seen that parents progress to a point where they have a comprehensive understanding of how the CF treatment regimen fits comfortably into family life:

Western Australian mother, 5 year old child - Caring for of XXA has got a little bit easier.

No, I wouldn't say it's gotten easier but it's more manageable. I mean, I do manage which provides me with the knowledge that I can manage, know what I mean.

Western Australian mother, 18 month old child - every day you're doing 2 rounds of physio and she's on anti-biotics twice a day and vitamin supplements and saline supplements but it's just part of the routine now, so it sort-of becomes a background thing and you just do it. When we first did it we used to get achy arms and things and wonder how we were going to do this all the time, but now it's just part of the routine.

Additionally, parents who expressed they regularly received assistance from outside the home to care for their child/ren were the same parents who spoke about how they had assimilated CF management into their daily routine, suggesting it is not necessarily only time that facilitates adaptation to CF management. Identifying that family assistance outside of the home may help parents adapt to their child's CF management has been insufficiently examined (Grossoehme, et al., 2014). However, in families where grandparents provided little or no support, an increased tendency for parental depression has been observed (Hodgkinson & Lester, 2002).

Reflecting contemporary childcare arrangements, this line of enquiry could highlight to parents that sourcing family assistance could help them to successfully adapt to CF management. Due to the exploratory nature of my research, it falls short of stating causal relationships between parental factors such as use of support and adaptation to CF management. There is, however, evidence that social support is a modifiable risk or protective factor (McCubbin & McCubbin, 1993; Wallander & Varni, 1998; Horton & Wallander, 2001), suggesting social support is indeed a mechanism for adaptive coping and functioning for parents of children with CF (Szczesniak, Zou, Wetzel, Krause & Grossoehme, 2015) and other chronic conditions (e.g., Hoekstra-Weebers, et al., 2001; Kars, et al., 2008; Khurana, Katyal & Marwaha, 2006; Spiers, et al., 2011).

Determining whether family assistance and other similar support structures contribute to mal/adaptation to a CF diagnosis and to the unique CF management of AREST CF are important steps in identification of targeted approaches that can assist parents in adapting to CF management in the context of early surveillance. Moreover, implementing targeted approaches that assist adapting to CF

management may also be conducive to parents' negotiation of their *new normal* within context of early surveillance.

If we now turn to the latter part of the theme title *understanding uncertainty*, one of the fundamental aspects of understanding CF is acknowledging the characteristic variability in morbidity and mortality (i.e., life expectancy). Whilst there is an average life expectancy of 37 years for females and 40 years for males (MacKenzie, et al., 2014), long-term health outcomes for individuals with CF are variable (Rowntree & Harris, 2003; Zielenski 2000). Therefore, *understanding uncertainty* refers to the matter of understanding uncertainty in terms of morbidity and mortality as a characteristic of CF: it refers to how uncertainty in their child's life, and its associated fear, becomes a lived experience for parents of children with CF.

As part of understanding CF, parents must acknowledge and eventually accept that life expectancy and morbidity are variable; understanding the uncertainty, understanding there will be an element of unknown. Understanding uncertainty in prognosis causes fear about the present and the future (Afonso, Gomes & Mitre, 2015; Havermans, et al., 2015; Moola, 2012). Once a child is diagnosed with a chronic condition, parent's fear about their future is a salient finding in the literature (Arestedt, Persson & Benzein, 2014; Gjengedal, et al., 2003; Moola, 2012; Patrick-Ott, 2011). Fear about a child's future is supported by Jessup and Parkinson (2010) who reported parental fear at diagnosis and then ongoing fear as part of parenting a child with CF. A lived experience for parents of children with CF is living with uncertainty and fear.

Living with uncertainty is demonstrated by a Western Australian mother of a 2 year old child who said *'sometimes I feel like I'm waiting for the shit to fall because to look at her, you wouldn't know that she is a sick kid...it's like we know, I know, that it's going to happen at some point...I'm still waiting for that. It can be a really scary prospect, you know.'* The sentiment was echoed by a Victorian mother whose child was 4 years old when she stated *'she knows what CF is, she doesn't know she's going to get sicker, she doesn't know she's going to die at a young age. I'm still dealing with the fear of that happening and so I don't want that on my daughter just yet.'*

For parents in my research, the uncertainty surrounding morbidity and mortality are a source of fear that likely goes beyond general parenting levels. Descriptions are similar to those reported by parents of other chronic (Cabizuca, Marques-Portella, Mendlowicz, Coutinho & Figueira, 2009; Patrick-Ott, 2011; Zhou, et al., 2014) and life-limiting conditions (Greening & Stoppelbein, 2007; Hoekstra-Weebers, et al., 2001; Kazak, et al., 2005; Von Essen, et al., 2004). Following diagnosis, redefined expectations and indeed, *new normal* always contain a level of uncertainty around a child's morbidity and mortality, thereby inherently eliciting some level of fear.

Uncertainty in Illness theory (Mishel, 1990) presents a basis for understanding how uncertainty associated with chronic illness is experienced. There are multiple aspects of uncertainty

in illness covered by Mishel's theory, but the inability to accurately predict outcomes of the condition for the inflicted person is particularly relevant to my research. Uncertainty reflects a neutral cognitive state and is neither desired nor dreaded until the implications of uncertainty are determined. That is, individuals appraise the situation and determine the value placed on uncertainty. Parents in my research decided whether uncertainty surrounding morbidity and mortality associated with their child's CF was a positive or negative event within their redefined expectations. Uncertainty in the child's condition was primarily viewed as a negative event that induced fear.

Fear was for the unfamiliar present and uncertain future. Fear of the present was about current logistics of disease progression, including potential hospital admissions, extra treatment, care for siblings, and time off work. Fear of the future concerned fear of losing the child prematurely and fear of the child's health deterioration. Moola (2012) and Jessup and Parkinson (2010) described how parents of children with CF constantly worried about child health deterioration, conceptualised as fear by the authors of both studies. Parents of children with CF can carry a level of fear that can overcast, but may not significantly affect, parenthood and family life.

Whilst uncertainty in morbidity and mortality in most cases is initially perceived negatively by parents, and certainly influences parental fear, the potential variability in long-term health outcomes for people with CF actually provides means for hope for a child's future. To explain further, parents of a child diagnosed with a disease or disability without potential for variability in outcomes (for example, parents of a paraplegic child) could be less likely to embrace hope for positive long-term health outcomes. Whereas for parents of a child diagnosed with a chronic disease such as CF that has the potential for variable long-term health outcomes, the negativity that surrounds uncertainty in morbidity and mortality can be surpassed by hope for their child's future. Hope for a child's future is elaborated in the next chapter as an over-arching lived experience for parents in my research.

Mishel's (1990) theory explains that uncertainty in illness can be perceived as a positive event when individuals assign beneficial value to the uncertainty. That is, whilst diagnosis of CF cannot and should not be patronised into a beneficial event, when parents in my research assigned positive value to uncertainty, they were able to construct psychologically beneficial perceptions of their child's condition. Favourable perceptions from assigning positive value to uncertainty resulted in constructions of hope for the child's future in terms of quality of life (i.e., morbidity) and life expectancy (i.e., mortality). Therefore, remarkably, there appears to be an evolution, a reinterpretation (though there can be some regression), in the way uncertainty of morbidity and mortality is constructed.

This finding supports Mishel's (1990) conceptualisation of how cognitive appraisal and assignment of value influences constructions of uncertainty in illness. The finding also supports

Lazarus's and Folkman's (1984) early theory about the role of appraisal in coping with stressful events, in that appraisal of an event, and assignment of value to that event as a result of the appraisal impacts upon cognitive and emotional constructions of that event, thereby contributing to coping resources drawn upon. How cognitive appraisal of an event influences choice of coping resources and strategies will be the topic of discussion in the next chapter. The following quotes exemplify how some parents constructed perceptions of uncertainty in their child's condition in a positive manner:

Victorian father, 12 month old baby - You know, I can't control it so at the end of the day I'm not going to stress over something that I can't control...There's the other side too, some people have a fear of it getting worse. But it's like 'ok, nothing may happen.' So we'll just keep doing what we're doing and that, and away you go.

Western Australian mother, 4 year old child - in my head I always think that if you're going to think the worst then contemplate the best part as well...If you're going to sit there and say that this and this could happen, then you've also got to think that this and this could happen.

Therefore, parents' dynamic journey of learning about CF transforms them from devastation of diagnosis to understanding of CF – understanding the unknown and understanding there is uncertainty. Moreover, the uncertainty of a child's morbidity and mortality can be over-arching and all encompassing, with either positive or negative effects.

5.4.2 Understanding early surveillance for CF lung disease.

Similar to the lack of knowledge about CF at time of diagnosis, parents also lack knowledge of early surveillance. They lack understanding of the processes and procedures, and of their own experience. The procedures and their purposes are comprehensively explained to parents, but there are elements of early surveillance that are not, and cannot be, fully explained by researchers or clinicians. These elements are especially prominent for the first annual early surveillance review, but there remain elements of uncertainty in all annual reviews. Elements of early surveillance that remain uncertain during the first annual review are the substance of this sub-theme, and how they evolve to being known. Whilst explanation of early surveillance processes and procedures are greatly appreciated, parents are uncertain about their own personal experience (i.e., how their child will respond and their own psychological experience) until such time when they experience the program's components and subsequently gain further knowledge.

Around the time of diagnosis, unknown parental experiences and child responses to early surveillance procedures add to parents' feelings of uncertainty about the present and future; parents are not only negotiating redefinition of self, family and future expectations. Parents' first experience of early surveillance is generally closely paired with CF diagnosis. Consequently, the first early

surveillance review appears to continue or exacerbate similar thoughts and feelings to that of diagnosis. This is not to diminish the heart-wrenching experience of diagnosis but to demonstrate the potential follow-on effects, or likely exacerbation, of emotions at the first annual review. Qualitative studies exploring the lived experience of parenting a child with CF have reported how events such as required surgical procedures for a child can trigger thoughts about CF (Bluebond-Langner, 1996; Glassocce & Smith, 2011). Likely running concurrently with diagnosis, parents conceivably need to negotiate through their first early surveillance experience, which will eventually become part of their *new normal*. The following quote demonstrated how a Western Australian mother of a 4 year old child felt about her infant's first annual review in relation to diagnosis:

You're still getting to grips at that point with the diagnosis, because we went through the denial thing, you know "are you sure you haven't mixed up the tests?" that sort-of thing...so you still haven't really accepted it at 12 weeks, you still haven't accepted that your child has CF and that your life is changed...as I said, that first time, you are still...you know, you're still coming to grips with the diagnosis.

In terms of parents' perceptions of the received level of information about early surveillance, some parents found the level of information overwhelming. A Victorian mother of 4 year old child reflected on her experience of receiving information about early surveillance, *'it goes straight over your head because they barrel you with all this information, new procedures, new everything, and it doesn't quite sink'*. Other parents felt information they received was adequate. A Western Australian mother of a 5 year old child recalled *'I think that the level of information that we're given is good...you know, lots of information prior, so I've been really happy with all of that.'* Whereas other parents felt the information they received about early surveillance was insufficient for an informed decision, reflected by a Western Australian mother's account whose child was 4 years of age, *'I don't think we really understood what the tests were for and what the outcomes were going to mean so we felt that we could've made an uninformed decision.'*

Jessup and colleagues (2015) found that most parents' were overwhelmed with the amount and diversity of information at the time of their formal CF education, whereas Sawyer and Glazner (2004) described parents' satisfaction with the amount and content of information given during CF management training. Whilst these limited results reflect diverse parental perceptions of Australian standards of care, they do not represent information given beyond formal CF education and are therefore not generalisable to parents in receipt of information about early surveillance for CF lung disease. It is conceivable that any confusion about early surveillance may lead to perceptions of insufficient information, miscommunication or indeed, the therapeutic misconception (National Bioethics Advisory Commission, 2001), explained in detail in a later theme. Therefore, findings from

my research suggest that protocol development around information content and delivery is warranted, and could inform development of such a protocol. At the very least, medical staff can be cognisant of parent information needs and priorities, both around the time of CF diagnosis and more specifically, about early surveillance.

There is one form of communication about early surveillance that parents felt was lacking. This information is about their potential personal experiences of the day's proceedings; how they might manage with the day, how they might feel, their responses and reactions. There is no advice or illumination of the potential psychological experience that parents may encounter throughout their day. Consequently, for parents with no annual surveillance experience, there is no foundation, no boundaries to confine and control their anticipatory ideas and notions about the day's events.

No foundation or boundaries for what parents' may experience speaks to the *unknown* conceptualisation of early surveillance in multiple ways; unknown about their child's reaction, unknown of their own reaction, unknown about the staff who will disappear with their child, unknown about whether their child will be relinquished back to them, unknown about their child's prognosis and unknown about their child's longevity and quality of life. This creates a lack of expectations that could be mitigated, or at least reduced, if there was some kind of notification of what parents' personal experience may be like; a kind-of foundation or framework upon which to base and confine their anticipations.

Applicable to parents of younger children (especially first-time parents) are the psychological consequences of not possessing a frame of reference because of lack of expectations for their ensuing personal experience of annual review. No foundation or framework for expectations is the consequence of the unknown; similar to lack of expectations at the time of diagnosis. This phenomenon is demonstrated through a Victorian mother's (child aged 2 years) experience who had no assumptions of, and therefore lack of expectations about, early surveillance and how this affected her first experience, keeping in mind how closely paired to diagnosis the initial visit generally is:

When he was really small, I found it harder because you didn't really know what to expect. I didn't know how he was going to deal with it, I wondered if I was doing the right thing because he had to have things done to him that he otherwise wouldn't have to...I cried when he was a baby because it was all new and very hormonal and still unsure. You're trusting these people that you've never trusted before with your baby but now, having done it a few times you know that they do a good job.

It is worthy to note here that parents with substantial early surveillance exposure (that is, parents of children aged 3 years and older) expressed their willingness to consult with parents new to early surveillance. They also asserted their belief that such communication could promote more rapid adaptation to both diagnosis and early surveillance than their experience. A Victorian father of

a 5 year old child asserted, *'I think it would be good for people who are coming into that position to talk to other families, or if there was a network of families that you could talk to and they could tell you what's going on and what's to be expected. It wasn't until afterwards that someone spoke to me about that stuff, about what's happening, what you're feeling is normal while going through that so I think it would've really helped us at that time.'*

Parents with children under 3 years of age spoke about their desire to communicate with parents about their experiences of early surveillance - of their thoughts and emotions - with the hope of being more prepared for their first early surveillance exposure. A Western Australian mother of 2 year old child stated, *'it would also be good to get information from other parents, you know like how they get through the bronch (bronchoscopy). It would be good to get just little snap-shots about what will happen and how they dealt with it. Even just things like, from a parent's perspective watching your child go under general anaesthetic, this is what happened and this is how I dealt with it...not to dramatise it but just to bring it to people's attention. Although everyone's experience is different, at least to have some idea of what it might have been like would've been good.'*

To summarise, after collapse of parenting expectations and dismantling of parent identity, parents come to understand how to manage their child's CF through establishing expectations about CF management (including family routine) and prognosis. Knowledge of these entities gives parents a foundation to start reconstruction. An aspect of CF that becomes understood by parents is the variability in morbidity and mortality, which contributes to the lived experience of uncertainty and fear about child, family and their own future. However, the uncertainty in mortality can also result in hope for their child's future because although a clock has been set on their child's life, the variability is constructed as an avenue for hope.

As early surveillance is inextricably linked to CF, it becomes part of the lived experience of CF. The link between CF and early surveillance may continue or exacerbate psychological distress experienced at diagnosis, and could indicate psychological adversities and processes experienced by parents whose children undergo early surveillance for CF lung disease. An implication for parental outcomes as a result of their child's first early surveillance is that parents are willing to talk with other parents about their experiences so that they can have an idea of what will happen and how they might respond on a personal level. It is through experience and subsequent knowledge of their ability to manage the day's event and their child and their own responses that parents become comfortable and feel they can cope. It is through understanding, that parents come to accept their child's diagnosis and early surveillance of CF lung disease as part of the redefined expectations and reimagined parent identities. Discussion will now shift to the everyday regular routine of parenting children with CF in the context of early surveillance.

5.5 Good days and bad days; Fluctuation between positive and negative outlook on life

Most of the literature to date examining experiences of parenting children with CF has evaluated the diagnosis phase (Armoni, et al., 2008; Priddis, et al., 2010; Carpenter & Narsavage, 2004; Sawyer & Glazner, 2004; Tluczek, Kosciak, Farrell & Rock, 2005) and its resulting psychopathologies (Glasscoe, et al., 2007; Goldbeck, 2006; Merelle, et al., 2003). Fewer studies have assessed parental outcomes beyond the diagnosis period (Delelis, Christophe, Leroy, Vanneste & Wallaert, 2008; Gjengedal, et al., 2003; Jessup, et al., 2015; Tluczek, Clark, McKechnie & Brown, 2014), including examination of positive parental outcomes such as psychological growth (referred to in the literature as resilience and post-traumatic growth) (Cadell, et al., 2014, Nabors, et al., 2013; Phipps, et al., 2015).

This theme is an all-encompassing description of parental experience beyond diagnosis, that is, the day-to-day experience of parenting a child with CF within the context of early surveillance. Therefore, this theme adds to the limited knowledge about parents' experiences of CF and its management beyond diagnosis, and demonstrates how early surveillance contributes to these regular, day-to-day experiences. Taken together, this theme describes the lived experience of parenting a child with CF in the context of early surveillance.

The current theme draws upon limited literature as a contribution to how parents construct meaning around their day-to-day experience of their child's CF beyond diagnosis. The first two sub-themes detail parents' lived experience by presenting salient psychological constructs that exist on a daily basis. The final sub-theme elucidates parents' fluctuation between these two perspectives as their lived experience of CF. Overall, this theme asserts that for parents in my research, some days are good and some days are bad in terms of their perspectives on CF, and that parents fluctuate between their positive views and emotions and their negative views and emotions. This theme relays these accounts through identification of three sub-themes: bad days, good days; and fluctuation between positive and negative outlook on life.

5.5.1 Bad days.

Adverse outcomes for parents of children with CF, and for parents of children with other life-limiting and chronic conditions, have been generally expressed in the literature as caregiver burden, treatment burden and parenting stress (Aydinok, Erermis, Bukusoglu, Yilmaz & Solak, 2005; Brod, Hammer, Christensen, Lessard & Bushnell, 2009; Couzino & Hazen, 2013; Hall & Graff, 2011; Hassall, et al., 2005; Henry, et al., 2008; Hutchinson, et al., 2009; Klassen, et al., 2010; Litzelman, et al., 2011; Norberg & Steneby, 2009; Quittner, et al., 1992; Sav, et al., 2013). Similar to other studies, parents in my research described ensuing adverse psychological outcomes beyond diagnosis (Goldbeck, Besier, Hinz, Singer & Quittner, 2010; Quittner, et al., 2008; Quittner, et al., 2014). Stratification of my sample by child age identified that parents in all age groups expressed such effects, suggesting that

adverse psychological outcomes begin with the destruction of CF diagnosis (Glasscoe, et al., 2007; Goldbeck, 2006; Priddis, et al., 2010) and ensue thereafter. Adverse parental psychological outcomes comprise the first part of this theme; fear and chronic sorrow.

5.5.1.1 Fear as an outcome of uncertainty in morbidity and mortality.

Parental psychological adversity experienced on bad days primarily consisted of fear, which is a natural response to general life events (Olsson & Phelps, 2007). Whilst fear can be positive in limited levels because it can protect and motivate (Ulrich, 1983), it appeared to go beyond these limited levels. Therefore, general parental fear is exacerbated for parents of chronically ill children (Crnic & Low, 2002; Krockek & Mowder, 2012). This is not a new finding in parents of children with CF (Fidika, et al., 2015; Hodgkinson & Lester, 2008; Jedlicka-Köhler, et al., 1996; Jessup, Douglas, Priddis, Branch-Smith and Shields, 2015; Wong & Heriot, 2008). However, due to the unique nature of early surveillance, how it contributes to the presence of parents' fear has not been previously explored.

Fear is generally caused by a perception of risk; it is an affective response of unease or apprehensiveness to a situation that has been cognitively appraised as dangerous (Lerner & Keltner, 2001). Though parents do not perceive themselves in an immediately threatening, or risky, situation, it is the unknown consequences of CF that are the drivers of fear, that is, the uncertainty in morbidity and mortality identified in the previous theme. Parents experience fear for this reason intermittently, on what they expressed as their *bad days*. There are a number of dimensions to the unknown consequences of CF that generate fear. Firstly, fear of losing a child to CF was expressed by a Victorian mother of a 6 month old baby who said *'sometimes I have a constant worry in the back of my head that something might be wrong, that fear of losing my child. I think 'why us, why her.'* Secondly, fear of a child getting sick due to disease severity was described by a Western Australian mother of a 2 year old child who stated *'so far, so good sort-of thing, but there's still that element of stress. That's the other thing, because XXA has been so well, sometimes I feel like I'm waiting for the shit to fall because to look at her, you wouldn't know that she is a sick kid...It's like we know, I know, that it's going to happen at some point but we're still waiting for that, I'm still waiting for that.'* Lastly, fear of knowing a child is getting sick through knowledge of early surveillance results was expressed by a Western Australian father whose child was 18 months old when he said, *'you don't know how the results really fare compared to just being nice and clean. That's what I sort-of really worry about.'*

Self-management of psychological adversities is important for parents' well-being and that of their family members (Ackard, Neumark-Sztainer, Story & Perry, 2006; Whittemore, et al., 2013). Several strategies were identified that enabled parents to manage (or cope with) their fear that was associated with uncertainty in morbidity and mortality. Firstly, some parents spoke about their psychological efforts to eliminate or minimise any adverse thoughts or feelings (Fidika, et al., 2015;

Sheehan, et al., 2014). For example, parents tried not to think about their child's condition in such a way that it affected them psychologically. This is represented by a Western Australian mother of a 6 year old child who tries to *'push it all the way down, right down to the bottom of your feet and you don't think about it. Just, you've got to block it out, because if you do think about it, and we have at times, you get consumed by it, it just eats you up. So you've just got to, I think, block it out.'*

Secondly, parents expressed their desire and ability to release their emotions periodically during such times that warranted it (Coyne, 1997; Tong, et al., 2010; Venters, 1981; Zubrzycka, 2015). In this instance, parents would get angry or cry, which diffused their emotional burden. For example, a Victorian mother of a 5 year old child spoke about this process when she said, *'that's our life now but you don't always feel this level-headed, sometimes you have your down times...I just let myself have a good cry. I just allow myself to have those moments.'*

Thirdly, parents restructured, or reconstructed, their thoughts to reframe their situation within context of the wider society (Havermans, et al., 2015; McCubbin, et al., 1983; Venters, 1981; Zubrzycka, 2015). This refers to parents' strategy of comparing their life situation to those who have to manage worse situations, described aptly by a Western Australian father of a 2 year old child who said, *'the only thing that helps me stay sane is that you see things on TV about kids every night who don't have access to medical care and doctors and things like that. The way I stay sane is I think - how lucky are we to have access to all this.'* Whilst this does not change their situation, it changes parents' cognitive constructions of it. This has been referred to in psychological literature as social comparison (Festinger, 1964; Wills, 1981), and this concept will be elaborated upon later.

Fourthly, support was a strong resource for coping with fear (Coffee, 2006; Green & Stoppelbein, 2007; Havermans, et al., 2015; McCubbin, et al., 1983; Priddis, et al., 2010; Zubrzycka, 2015). Support was mainly used as an avenue to reduce psychological and/or emotional burden either with a partner, family or friends (Wong & Heriot, 2008); and was reported more often by mothers compared with fathers. A mother from Victoria whose child was 5 stated, *'just dealing with it together, having a support person is wonderful, it really helps me,'* and the sentiment was echoed by this Western Australian mother whose child was 6 years old when she described, *'just to kind-of talk about it with friends and family makes it easier.'* This finding is in agreement with Havermans, et al., (2015) who illustrated that mothers used social support as a coping resource more often than fathers.

Finally, staying in the present moment and not letting thoughts go too far into the future helped parents to manage their fear about uncertainty that has been built into their futures as part of that redefined *new normal* (Gjengedal, et al., 2003). To illustrate the point, a Victorian mother of a 12 month old baby asserted, *'there's no sense in lulling over what could possibly go wrong in the future, it may or may not even happen, you just deal with what is in front of you now.'* Discussion of

theoretical application and wider implications of some of these coping strategies, along with how they might apply to context of family life, occurs in the next chapter.

From the above quotes, coping with fear associated with CF is primarily represented by an emotion-focussed coping approach (Lazarus & Folkman, 1984). Recall from Chapter 2 the Transactional Model of Stress and Coping (Lazarus & Folkman) where emotion-focussed coping comprised a major component of how people choose to cope with life stressors. Emotion-focussed coping strategies are primarily utilised by parents in my research because, through their appraisal, they understand they cannot remove the stressor of CF, and so come to use strategies that enable regulation of emotions associated with it. For example, Lazarus and Folkman's psychological conceptualisation of coping with fear applies to the processes followed by parents in my research, that is, appraisal of an event as threatening induces fear, and fear induces utilisation of available coping resources dependent on the appraisal and on the availability of coping resources for the individual.

A number of parental characteristics for coping with fear were discovered. Firstly, parents whose children were 3 years of age or older often spoke about how they managed their fear, suggesting that developing skills to manage fear associated with CF and/or early surveillance may take time and experience. Secondly, parents who had access to external assistance for their children spoke about their ability to manage their fear, suggesting that resources outside of the nuclear family help parents to cope. Recall that family assistance facilitated the normalisation process as part of adjustment to diagnosis. These findings highlight importance of extended family and external networks as a continual source of support that enables parents to better function in their roles as carer, parent, and likely within themselves. Thirdly, full and part-time working mothers more often spoke about effectively coping with fear than mothers who classed themselves on full-time home duties. This particular finding is in agreement with Carver (1997), who found that working mothers coped more effectively with life stressors than mothers who did not work.

Collectively, these findings suggest there is potential for outside influences to contribute to parents' coping resources. For example, more effective coping by working mothers versus stay-at-home mothers may occur because time outside of the immediate family environment could function as a reprieve for mothers, resulting in giving them more scope to deal with personal adversity (LeMaster, Marcus-Newhall, Casad & Silverman, 2004). Parents may learn coping skills outside of the family setting either from managing work-related issues or through talking with people outside the family home (Gatrell, 2013). Finally, parents who spoke about coping with fear had older children, suggesting the possibility that generic parenting skills developed before birth of the child who has CF may buffer against fear.

Parental fear of disease progression is a recent line of inquiry examining existential outcomes of parenting a child with CF. Fidika and colleagues (2015), Jessup and Parkinson (2010) and Wong and Heriot (2008) examined fear of disease progression and/or morbidity and mortality. Together, these studies reported an ongoing process of fluctuating fear that affects parents' adjustment to their child's CF. Given that parental fear of disease progression in CF is a fairly new concept in the paediatric chronic illness literature (Fidika, et al., 2015) and that early surveillance of covert lung disease is unique to AREST CF, what is needed is investigation of how parents cope with knowledge of a child's disease progression. My research did not uncover if and how parents cope with such knowledge. Given that early diagnosis and early surveillance of paediatric disease are modern technologies, and my research uncovered the multi-dimensional nature of fear about early disease progression, a further study with focus on how parents cope with such information and their associated emotions and cognitions is warranted.

5.5.1.2 Chronic sorrow.

With parents' knowledge of paediatric chronic illness comes profound and extensive sadness. Profound and extended sadness, conceptualised in the literature as chronic sorrow (Gravelle, 1997; Olshansky, 1962), is a prevalent experience for parents during the diagnosis phase of many chronic conditions, including cerebral palsy, type 1 diabetes, Down's Syndrome, physical disability and sickle cell disease (e.g., Bowes, et al., 2009; Damrosch & Perry, 1989; Masterton, 2010, Neilsen, 2013; Nikfarid, et al., 2015; Paul, et al., 2014; Patrick, 2014; Whittingham, et al., 2013). Beyond diagnosis, a portion of parents are reported to experience chronic sorrow on a more permanent basis (Kearney & Griffin, 2001; Nikfarid, et al., 2015; Patrick-Ott, 2011; Whittingham, Wee, Sanders & Boyd, 2013).

Neither grief nor loss were explicitly discussed in relation to their child's diagnosis (as this was not the focus of interviews), though it is conceivable that parents in my research may have experienced this kind of profound sadness following their child's CF diagnosis (Moola, 2012; Priddis, et al., 2010; Shumaker, 1994). That chronic sorrow has been interpreted as an essential dimension of the lived experience for parents in my research, it is conceivable that chronic sorrow may be a continuation of grief or loss experienced at diagnosis. Though continuation of chronic sorrow from diagnosis is speculative, parents in my research expressed profound and intermittent sadness throughout their lives beyond their baby's diagnosis, which they experienced at different times and for different reasons. Therefore, it has been interpreted as chronic sorrow because the conceptualisation of profound sadness in my research is comparable to the definition of chronic sorrow described in the literature as outlined in chapter 2.

Parents' profound sadness was about their child's disease and their family situation. The recurring nature of chronic sorrow is reflected within this theme because, as will be described later, parents have *fluctuating* psychological states. Chronic sorrow is experienced intermittently

throughout parents' lives. Though understood as common amongst parents of children with CF (Shumaker, 1994), possible effects of chronic sorrow on parents and their families have not been sufficiently studied, nor have reasons why chronic sorrow may persist beyond fear of disease progression and loss of the child. My research adds to the scant body of literature by identifying reasons beyond loss and grief as objects of chronic sorrow.

Firstly, parents' expectations of a typical parental role, as well as expectations for their child and their family, could no longer be met. In the theme *redefined expectations and reimagined identity; redefined reality*, this process occurred as part of diagnosis when expectations for a typical child and family life were destroyed. These reasons for grief and loss at time of diagnosis have been reported in the CF literature (Hodgkinson & Lester, 2002; Hymovich & Baker, 1985; Jessup & Parkinson, 2010). That loss of expectations was identified at diagnosis in my research, and the literature shows both grief and loss of expectations at diagnosis can result in chronic sorrow, adds credibility to the speculation that parents in my research experienced chronic sorrow.

If the onset of chronic sorrow did indeed arise at diagnosis, then it appears the response to loss of parenting expectations continues intermittently. That is, on some days parents cognitively revisit loss of expectations and at the same time, grieve for the loss of their ideologies. It is the loss of, and grief in, losing ideologies that results in feelings of profound sadness. A Victorian mother of 12 month old baby expressed sadness about her loss of parent and family expectations when she stated, *'there's time I get upset, it's hard only because I'm used to working, socialising and being out there so it's difficult. I mean, it's not the worst thing in the world but it's just a real life-change really. A real difference in what you expected of parenting and family life, you know.'*

Secondly, parents could experience chronic sorrow when they thought about their child's loss, that is, the loss of quality of life and the potential loss of life expectancy. Parents felt sadness for their child's endurance of daily treatment, regular hospital visits, current sickness as well as their future losses. The following excerpt from a Western Australian mother of a 5 year old child aptly demonstrates parents' expressions of sadness for their child's loss, and endurance, of CF:

it's sad because it's not how you envisaged your child's life to be. You always want a healthy child and unless they had a broken arm or something, then you would never have to go to hospital. I get upset because she doesn't like it and I make her do things that she doesn't like...and the pain and stress associated with it.

Collectively, fear and chronic sorrow are adverse psychological outcomes of parents' meanings attributed to their child's CF. That is, parents learn about, and come to accept, characteristics of CF as they construct meaning around it for themselves, their child and their individual and collective futures. The result of this is fear and chronic sorrow. Parents coped with these cognitions and emotions in various ways, likely with varying degrees of effectiveness. Parents

who have; older children in early surveillance, other children, access to childcare assistance outside of the home, supportive networks and mothers who worked at least part-time expressed coping strategies they used to deal with their personal, intrinsic adversities, or that appeared as implicit coping resources. Implications of coping, along with theoretical application of coping strategies and styles will be outlined in the next chapter.

5.5.2 Good days.

Whilst parents may always carry a level of sorrow and fear about their child's life trajectory, there are good days amongst the bad days; but what does it mean to have a good day and what does it feel like for parents in relation to their child's CF in context of early surveillance? This sub-theme demonstrates what parents experience on what they expressed as *good days*, comprising parental satisfaction from perceived parenting competence, gratitude, and perceived control and power. These ensuing beneficial outcomes of managing and living with their child's CF, within context of early surveillance, are reported in the literature for parents of children with CF, and for parents of children with other life-limiting and chronic conditions, collectively expressed as resilience and adaptation (Goldbeck, et al., 2014; Van Riper, 2007; McCubbin, et al., 1996). Research has trended towards investigating such beneficial consequences of paediatric chronic illness for parents, portraying resilient parents, and whether and which parent and family characteristics may contribute or impede (e.g., Barakat, Alderfer & Kazak, 2006; Goldbeck, et al., 2001; Hullman, Fedele, Molzon, Mayes & Mullins, 2014; Peer & Hillman, 2014; Phipps, et al., 2015). It is relevant to note that parents in my research also experienced ensuing hope for their child's future, but because of its centrality to the lived experience, it is discussed as a construct that transcends both CF and early surveillance later in the chapter.

5.5.2.1 Parenting competence supports the parental role.

The parental role refers to beliefs and values about what it means to be a parent, and subsequent behaviours resulting from the belief and value system (Simon, 1992). Those parenting responsibilities and activities performed to keep a child healthy (e.g., feeding, clothing and protecting children from harm) begin to define the parental role and identity. To apply this conceptualisation of parental role and identity to the complexity of parenting a child with CF, a fundamental requirement for parents is to keep their child healthy by performing the rigorous and demanding home treatment regimen outlined in the first chapter. In this sense, treatment management becomes part of the parental role: parents' role is to care for their child beyond typical parenting responsibilities (as described in sub-theme Collapsed and Redefined Expectations).

The caring role often dominates the parenting role because of the need to provide ongoing intensive care to the child (Kirk, et al., 2005; Sullivan-Bolyai, Rosenberg & Bayard, 2006). This then

contributes to perceptions of the parent as medical carer for the child, which then becomes part of parental identity: parents become medicalised (Glasscoe & Smith, 2011, Tong, et al., 2010), and pseudo-paediatricians (Gjengedal, et al., 2003). On days when parents feel positive about their child's CF, their parental role in relation to their child's treatment management was viewed in a positive manner. A Victorian mother of a 12 month old baby expressed how she viewed herself when she said *'on the good days I feel like I'm doing the right thing with her treatment so that's the positive I get out of it as a parent, that I'm doing the best I can to protect my child'* Therefore, home treatment management becomes inherently linked to parental identity because of intensity and consistency of treatment, along with perceived ability, that is, self-efficacy, to keep the child healthy - their innate parental responsibility.

In context of my research, high parenting self-efficacy includes parents' beliefs about their ability to positively influence the health and development of their child especially in relation to their condition (Coleman & Karraker, 2000). The most plausible reason for high self-efficacy for treatment is that most CF treatment is conducted within the home, and is therefore primarily the responsibility of parents. The intensity and consistency of treatment, along with the innate responsibility to keep a child healthy meant that parents believed they could keep their child healthy through maintaining treatment. The central role of home treatment not only medicalises the parent role, but it places an additional burden on the parents to achieve a degree of efficacy in delivering the treatment. If they achieve it, the child will do better in their minds. Therefore, the child's health outcome is intrinsically linked to the parents' ability to provide home treatment. Due to the centrality of the treatment regimen to child and parent, parents in my research attributed parenting competency to achieving the required standard and level of home treatment.

Therefore, home CF management appears a primary mechanism for achieving, and assessing, parenting competence. Perceived parenting competence results in satisfaction in the parenting role (Anglely, Divney, Magriples & Kershaw, 2014; Bornstein, 2003; Johnston & Mash, 1989). Satisfaction in the parenting role from competence in parenting responsibilities supports parental identity, and support for parental identity can increase parental self-esteem (Mouton & Tuma, 1988; Ohan, Leung & Johnston, 2000). That is, playing a major role in their child's daily treatment management (responsibility) helped to support parents' belief that they were doing everything they could for their child (self-efficacy), which then gave them the satisfaction of doing so (perceived competence). This contributes to parental beliefs (parental role) and values (self-esteem), and what it means to be a parent of a child with CF (parental identity). The diagram on the following page outlines how these parental psychological constructs function together and affect each other in relation to home CF treatment and parental outcomes.

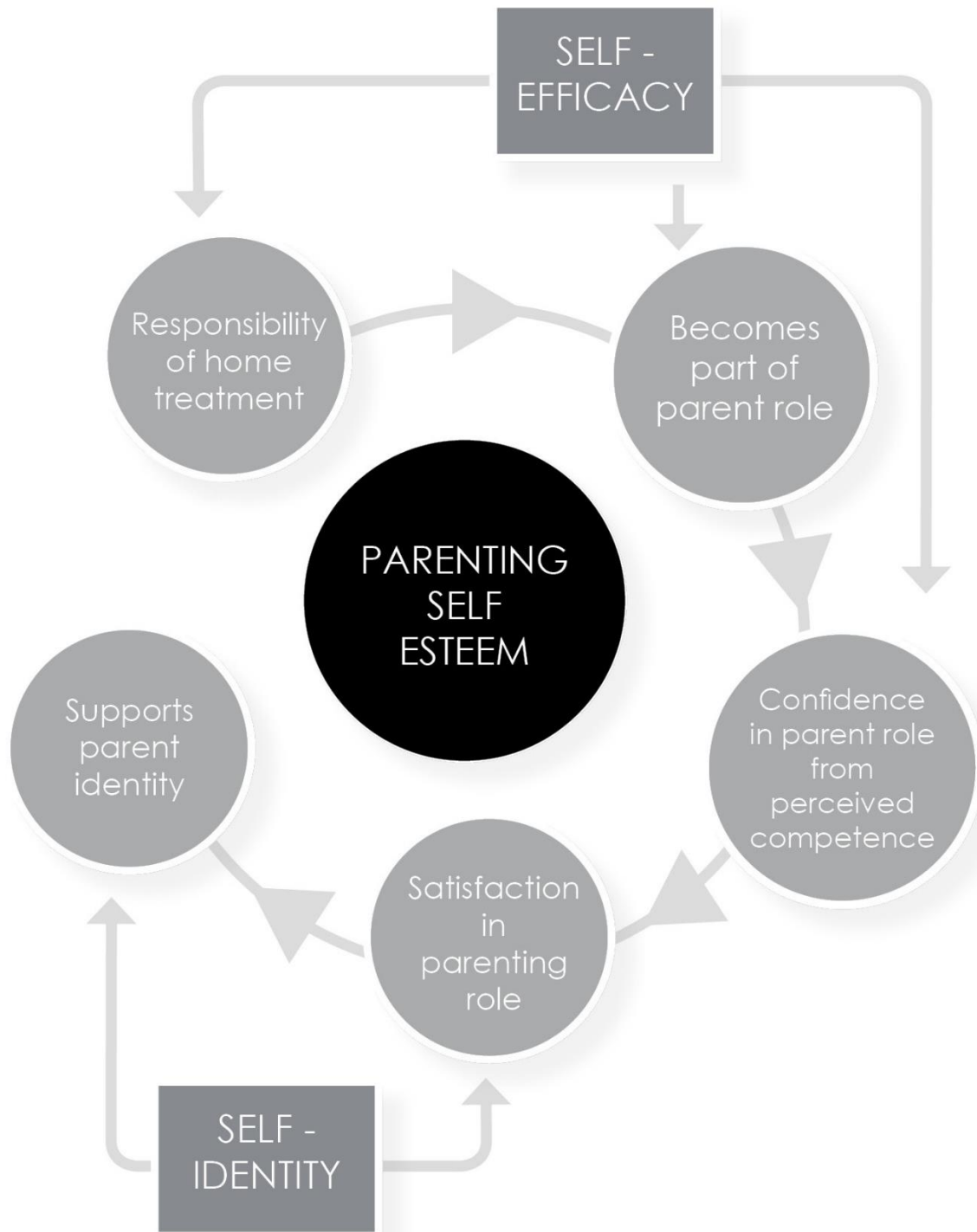


Figure 7. How parental psychological constructs are associated with home treatment and parenting outcomes

The diagram shows that it is support for, and promotion of, self-efficacy through perceived parenting competence that has an effect on parental identity and self-esteem. Promotion of self-efficacy interacts positively with self-concept, that is, support of high self-efficacy promotes positive beliefs about oneself (Fenning & May, 2013; Martin, Goldwasser & Harris, 2015). A plethora of

psychological literature supports the interaction between perceived parenting competence and self-esteem (Gilmore & Cuskelly, 2009; Ohan, Leung & Johnston, 2000; Ponomartchouk & Bouchard, 2014; Ramassini, 2000; Rogers & Matthews, 2004). The following excerpt from a Western Australian mother of a 12 month old baby highlights how her self-esteem was enhanced through beliefs in parenting competence because she was able to achieve her child's home treatment regimen, *'if we're not doing it, because her treatment is at home, it's in our hands. We're the ones giving her the antibiotics and doing her physio so we are the day-to-day treatment for her, and because it's such a large responsibility, we feel good that we can maintain it for her.'*

In conclusion, parents can experience parental satisfaction, parenting competence and increased self-esteem on days when they have a positive outlook on their child's CF, both in relation to conducting and achieving their child's home treatment as well as managing and achieving their *new normal*. Therefore, managing CF and its treatment is both difficult and rewarding for parents in their redefined roles as parents of a child requiring extensive home treatment. Consideration of parenting competence and increased self-esteem from achieving home treatment has been insufficiently investigated in the CF literature, given its importance for parenting. Recent studies by McDonald and colleagues (2013) identified self-efficacy for home treatment as a dimension of parenting a child with CF; Jessup and Parkinson (2010) reported parents' fear of failing to administer required treatment and Grossoehme and colleagues (2015) reported importance of parenting self-efficacy for treatment adherence. My research identified how and why self-efficacy for CF home treatment becomes inextricably linked to parenting competence, and discerned its effects on parental identity and self-esteem. The current finding indicates that increasing parenting self-efficacy for conducting home treatment could be a source of strength for parents to draw upon to build confidence in establishing, managing and maintaining a home CF treatment regimen.

5.5.2.2 Gratitude, both for their child and for early surveillance.

Although sadness and fear are likely to always exist on some level or in some form, parents' positive views of CF occurring on good days included a sense of gratitude for their child. Gratitude existed in two forms. Firstly, parents appreciate their child is healthy relative to other children who are inflicted with worse conditions. Secondly, parents are thankful for the unique opportunity to track their child's covert disease progression afforded to them by early surveillance. Each form of gratitude will now be explained.

Gratitude for a child with CF through comparison has been interpreted as a result of emotion-focussed coping, specifically the strategy of cognitive reappraisal. Cognitive reappraisal is an emotion regulation strategy that functions to reduce negative emotion associated with a stressor, whereby an individual reconceptualises a particular cognition so there is less burden associated with

it (Ray, McRae, Ochsner & Gross, 2010). In this instance, parents reconstruct their perceptions of their child's disease by comparing with children who have worse conditions.

This process has been labelled as social comparison (Festinger, 1964; Wills, 1981), and has been shown to be a common coping strategy when people are confronted with health problems (Van der Zee, Buunk, Sanderman, Botke & van den Bergh, 2000). For example, when individuals faced with serious health issues make comparisons with selective information from their social environment, it produces a favourable comparison situation that increases subjective well-being (Christie & Barnard, 2012). Therefore, social comparison can serve as an adaptive process to a threatening situation.

As parents in my research frequently visited their CF clinic, there was plenty of opportunity for them to compare their child's health with children who have worse health. A Victorian mother of a 5 year old child said *'I sort-of look around the hospital and think that we've got it pretty good compared to a lot of people there so I try not to complain,'* and a Western Australian father of a 3 year old child said *'yeah we kind-of think - at least we didn't get what they've got, we got off easy.'* Comparing their family situation to families who experience worse conditions enabled parents to positively reconstruct their perspective of their child's illness within context of wider society. Reconstructing perceptions of the child's condition in this light resulted in the favourable affective outcome of gratitude.

Research suggests that people who compare themselves favourably with others tend to experience better health outcomes (Buunk, et al., 2012; Kwan, Love, Ryff & Essex, 2003). Even those who receive information threatening to their self-concept, but who see themselves as better off than the average person in their situation (known as downward comparison), tend to adapt more successfully to failure or threat, and report greater life satisfaction and less psychological distress (Kedia, Mussweiler & Linden, 2014; Stewart, Chipperfield, Ruthig & Heckhausen, 2013). Accordingly, making downward social comparison has been shown to be self-protective by alleviating negative emotional consequences of stressful encounters and threats to self-esteem (Bauer, Wrosch & Jobin, 2008; Heckhausen & Brim, 1997; Wills, 1981). Findings from my research suggest that social comparisons allow parents of chronically ill children to self-evaluate in relation to others (Festinger, 1964; Kwan, et al., 2003; Moola, 2012; Tanner, 2007). Therefore, the current finding is consistent with those of other studies that support the use of social comparison as a strategy to self-enhance one's own position and experiences.

The other reason parents experience gratitude, and the principal reason that parents enrol their child in early surveillance is the opportunity to track their child's disease progression. Parents understand the unique opportunity afforded by early surveillance, thereby perceiving their situation

as lucky, resulting in feelings of gratitude (Cartwright, et al., 2011), as demonstrated by a Western Australian mother of a 6 year old child:

I'm also grateful that we can go the hospital, I'm grateful for the bronchoscopy and the CT. I'm just so grateful for what they're offering, how they do it, how they help her. Everything about it really, I'm just so grateful. Something that is so unknown, you know 'oh we'll just give this a go, we don't really know if it's going to work.' That to me would be so much more stressful. I'm just so appreciative of what's out there for CF.

Along with parents' perception of the unique opportunity afforded by early surveillance resulting in feelings of gratitude is the notion of perceived control and power over disease progression. In this case, knowledge is power. These are the first findings, to my knowledge, to describe this psychological phenomena for parents whose children undergo early surveillance for covert disease progression, and are important to communicate to other paediatric CF centres adopting the early surveillance framework (e.g., Utrecht University in The Netherlands for example).

5.5.2.3 Perceived control and power through certainty of disease severity from early surveillance results.

The primary objective of early surveillance is to detect disease for the purpose of administering proactive intervention. For parents, the primary objective of disease detection and successive intervention neutralises their primary concern of CF: that is, uncertainty in morbidity and life expectancy. To explain how parents feel a sense of control and power over their child's disease progression is to reveal how early surveillance outcomes respond to parents' perceptions of CF. To explain further, all-encompassing uncertainty becomes the lived experience for parents of children with CF, but becomes neutralised by assurance from perceived control. Power from perceived control is a reaction to assurance generated from receiving detailed information about a child's covert disease progression and subsequent early intervention.

Assurance of child health as a result of this information may be a unique construction for parents whose children undergo early surveillance. The following quote from a Western Australian mother whose child was 3 years old demonstrates how she felt assured her child was being treated as early as possible with the most sensitive measures available, *'I think it's nice to have that reassurance that everything that you're going through every day and the disruption that it causes to our family life is actually making a difference...it's nice to know that all of that is having a bearing on her health.'*

Knowledge of covert disease progression and assurance that early intervention is administered in a proactive manner creates a sense of control for parents, which gives them a sense of power. The following quote from a Western Australian mother of a 12 month old baby

demonstrates that parents feel a sense of control over their child's disease progression because of the assurance given by early surveillance that their child is being treated as early as possible with the most sensitive measures available:

it's reassuring because we're so new to what CF is, and things like that. You know, you're not sure, has she already got something? So yeah, that's what I'm looking forward to for the next one. You know, to know that she's ok. You know, using it in a positive way so that if she's not well then, bang they can get on top of it. I feel like I have some control over something that I can't see. So I think it's great, I think it's better than not knowing.

Rodriquez and colleagues (2012) suggested that the most stressful aspect of caring for a child with a chronic condition may be a lack of control, that is, parents may feel a loss of control when, for example, they are uncertain about their child's future prognosis. The quotes following the diagram demonstrate that parents felt a sense of control and power over disease progression through knowledge provided by early surveillance. The diagram below depicts how knowledge of covert disease progression from early surveillance and assurance of early treatment generates a sense of control over disease progression.

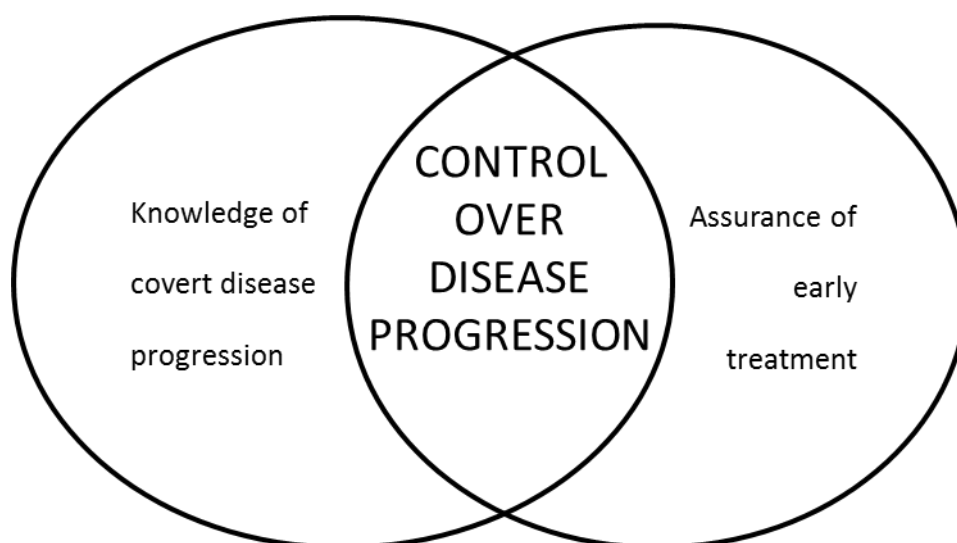


Figure 8. Relationships between knowledge, assurance and control

Victorian mother, 3 year old child - I think having more information gives me control, I like control...my goal is to be in control and information helps me to get that control.

Victorian father, 5 year old child - I can go 'ok, that's done and this is what it means,' and I can move on. I just like to know, it's a bit of control.

Victorian mother, 4 year old child - it's important to know. Knowledge is power and that's why we're doing this, we want to know.

Western Australian mother, 6 year old child - we still had a better understanding of her X-rays, so the more knowledge you have the better you feel...yeah, knowledge is power.

Therefore, on days when parents have a positive outlook on their child's life, and on CF in general, knowing they are recipients of the most detailed information about CF disease progression that is available for their child gives parents a sense of control over disease progression. Perceived power and control over disease progression are a direct consequence of early surveillance, and become of central importance to parents on a long-term basis, thereby functioning as a lived experience for parents whose children undergo early surveillance of CF lung disease.

On a final note, it is worth highlighting that whilst parents in my research felt power and control over their child's disease progression on their good days, they did not appear to experience helplessness on their bad days. Parents' feelings of helplessness as a result of their child's CF (Carpenter & Narsavage, 2004; Jessup & Parkinson, 2010) and other progressive chronic diagnoses has been supported in the literature (Hechlerl, et al., 2011; Pelentsov, Laws & Esterman, 2015), but was not a finding in my research (however feelings of helplessness as a result of early surveillance procedures will be described in a later theme). That parents in my research were not found to experience helplessness may be due to their perceptions of control and power over disease progression afforded by early surveillance neutralising the feelings of helplessness that have been reported in other studies with parents of children with CF (Coyne, 1997; Goldbeck, et al., 2010; Staab, et al., 1998).

5.5.3 Fluctuation between good days and bad days.

We now know what parents experience on particular days, but how do those days fit together to create a more global view of parents' lived experience? Parents expressed how they shifted between a positive and a negative perspective on CF. Parents appeared to fluctuate between good days and bad, those positive and negative psychological constructs. A continual cycle between good days and bad days; fear and chronic sorrow fluctuate between parental competence and satisfaction, gratitude and perceived control. Duff and Oxley (2007) described this process as oscillating phases of chronic sorrow with phases of normality and routine, punctuated with intense sadness and mourning.

A Victorian couple whose child was 2 years of age described how they fluctuated between good days and bad days when the mother stated, *'that's our life now, but you don't always feel this level-headed, sometimes you have your down times and you think 'why did this have to happen,' but it's just the way it is, that's what we've been dealt and you just have to deal with it.'* The father responded to the mother's statement by adding, *'I just stress about the condition in general. Some days you stress about it more than others. Some days are better than others.'* Others have also reported a continuously fluctuating cycle for parents of children with CF between distress and normalcy, adversity and adjustment (Duff & Brownlee, 2008; Glasscoe & Smith, 2008, 2011; Jessup & Parkinson, 2010).

It was not determined how often, or why, parents fluctuate between these two perspectives, for how long they persist with either of those outlooks, or reasons why a particular psychological state comes about. However, the substance of each of those types of days and psychological states is important information for both clinicians and other mental health professionals working with these families. With this information, HCPs can better understand parents' experiences of their child's CF that can help with clinician-parent interactions, implicating rapport and trust development.

In summary, a CF diagnosis brings much psychological distress for parents, and may continue beyond the diagnosis phase. Fear is induced by uncertainty in morbidity and mortality and chronic sorrow is induced by loss of child, parent and family expectations. Parents managed their fear through a number of mechanisms including social comparison and social support. Moreover, a number of parental characteristics were identified that may contribute to coping with fear associated with paediatric CF, for example, parents of children 3 years or older. Parents' lived experience includes a fluctuation between good days and bad days, a cyclical journey between the former and the latter (Gjengedal, et al., 2003). The good days are filled with feelings of high parental self-esteem, gratitude and perceived control and power over disease progression.

CF treatment management becomes a major component of family life and parent identity, and therefore merges into *new normal*; the redefined sense of self and family life. Psychologically, parents thrive on achieving the demanding home treatment regimen because by maintaining treatment management, perceived parenting competence supports the re-imagined parent identity as medical carer, potentially facilitating parental self-esteem. Moreover, perceived control over disease progression also becomes part of the *new normal* for parents in my research, which generates a sense of power from knowledge and gratitude from comparisons. Therefore, amongst the fear and chronic sorrow, parents experience competence and self-esteem, gratitude, control and power.

5.6 Early surveillance is a significant event

To this point, findings have described parents' lived experience of CF; the diagnosis, the unknown that, with time and experience, turns into the understood and eventually accepted. The initial experience of CF includes introduction, learning and assimilation of CF and its treatment management into family life. How introduction of CF affects parents' well-being more generally has also been outlined, with demonstration of how parents fluctuate between good days and bad days, of what each of those types of days consist, and how parents are affected by them. Introduction to, and learning about, early surveillance for CF lung disease has also been discussed. This theme shifts focus from how CF and early surveillance come to be understood, to demonstrate how early surveillance is constructed by parents as a significant event: the meanings attributed to it for perceptions of themselves as parents including factors that contribute to the process, and how early

surveillance can affect family life. This theme is about the experience of early surveillance, whereas prior discussion was about how early surveillance comes to be understood. Therefore, this theme describes parents' lived experience of early surveillance for CF lung disease.

To explore the complexity of this theme, three sub-themes were identified that detail the above varied aspects involved with early surveillance for parents in my research. The sub-themes were: indicator of treatment efforts and examination of parenting competence; parental outcomes directly from early surveillance; and fear from diagnosis exacerbated each annual review can result in anxiety.

5.6.1 Indicator of treatment efforts and an examination of parenting competency.

A common perception held by parents and society more generally is that children, and their development, outcomes and achievements are a reflection of their parents' competency to raise and care for them. The link between child outcomes and perceived parenting competency extends to early surveillance; sensitive detection of early disease progression is scrutinised against parents' treatment efforts and health awareness throughout the year. As a Western Australian father of a 5 year old child stated, *'all year you try to make sure you're doing the best thing for him so it's always almost like an exam for us. It feels like an exam to us so you sort-of just really want to know whether you've passed and whether he's passed.'*

Parents whose children received favourable annual review results expressed belief that their treatment efforts had a beneficial effect on their child's health, which created a sense of parenting competence. An excerpt from a Western Australian mother of a 5 year old child highlights accountability she felt for her child's favourable annual review results, *'it is like you are studying for a test to a certain extent and have you done well. You feel fantastic when you get a great result, you feel really good about your parenting.'*

Inversely, negative annual review results are constructed by parents as a reflection of their inability to conduct adequate levels of home CF treatment. This concerning finding of accountability for unfavourable annual review results is demonstrated by a Victorian mother whose child was 4 years old, she stated, *'you know when we get a bad result, you second-guess yourself. Could I have done something differently? Where did he pick it up? Was it our fault he got it? How can I be a better parent?'* Bluebond-Langner (1996) found that parents' sense of competence was threatened when a child was admitted to hospital for an exacerbation. Collectively, these results indicate that parents' perceived competence in relation to their child's health can be adversely affected by events attesting to a child's early disease progression.

Self-directed blame for unfavourable annual review results appears to be a consequential interaction of the model of home CF treatment and the sensitivity of measuring covert disease progression because of the link parents make to their child's home treatment. The inextricable links

between home treatment and sensitive measures of disease progression may mean that blame is unavoidable for parents whose children undergo early surveillance. Given the risk for depression, post-traumatic stress syndrome (PTSS) and anxiety from parental self-blame for paediatric chronic illness (Greening & Stoppelbein, 2007), such information could educate HCPs about parents' constructions of early surveillance outcomes and inform clinical care pathways that might target adverse parental outcomes associated with self-blame for diagnosis and/or negative test results.

High self-efficacy for treatment management resulted in parents believing that their treatment efforts will have an effect on their child's health, hence attributions of internal locus of control for child health outcomes. Parenting self-efficacy has been explained elsewhere, and locus of control refers to the extent to which individuals believe they can control events affecting them (Rotter, 1966). An internal locus of control orientation is a belief about whether the outcomes of actions are contingent on what a person does, whereas an external locus of control orientation is a belief that events are outside of personal control (Zimbardo, 1985). Extensive literature across a multitude of disciplines attests to the relationship between high self-efficacy and internal locus of control, including vocational and health behaviour (Joo, Lim & Kim, 2013; Roddenberry & Renk, 2010; Strauser, Ketz & Keim, 2002).

The finding of attributions of internal locus of control for child health does not support previous research on parents' constructions of their role in their child's health, with a number of studies showing that parents exhibited an external locus of control for their child's health. Parents believed factors such as fate, religion or medical intervention were the primary causes of child health (Lawoko & Soares 2002; Menahem, et al., 2008). The reason for this inconsistency is not clear but it may be due to the level of home treatment required by parents of children with CF. For example, parents of children with chronic kidney disease who were required to administer home treatment including dialysis and injections felt at fault if their child developed infections or other complications (Tong, et al., 2010). Attribution of internal locus of control for health outcomes and high-self-efficacy for treatment maintenance in terms of parents' constructions of their child's health and disease progression is an important finding in my research. It illuminates how parents interpret information from early surveillance, what meanings they attribute to it, and informs what effect it has on them. A passage from a Victorian father whose child was 4 years old illustrates how he felt when his child's annual review results were positive. He stated, *'I guess it's a confidence thing too. If they say that her lungs are clear then it makes you feel good as a parent.'* On the contrary, a Victorian mother of a 12 month old baby expressed how she felt when her child's results were negative. She said, *'she had quite a lot of damage in one part of her lung so that was a bit confronting for us that day. It felt like we had done something wrong as parents, even though we fully understand that not to be the case, you can't help how you feel no matter what you think, kind-*

of thing.' The diagram on the following page depicts how early surveillance results are constructed within the parental role, and how they are attributed meanings of parenting competence, which may affect parental identity and parental self-esteem. Note the contribution of early surveillance to the effects of parenting self-efficacy on parenting competence beyond the diagram outlined on page 132 showing a link between treatment management and parenting competence.

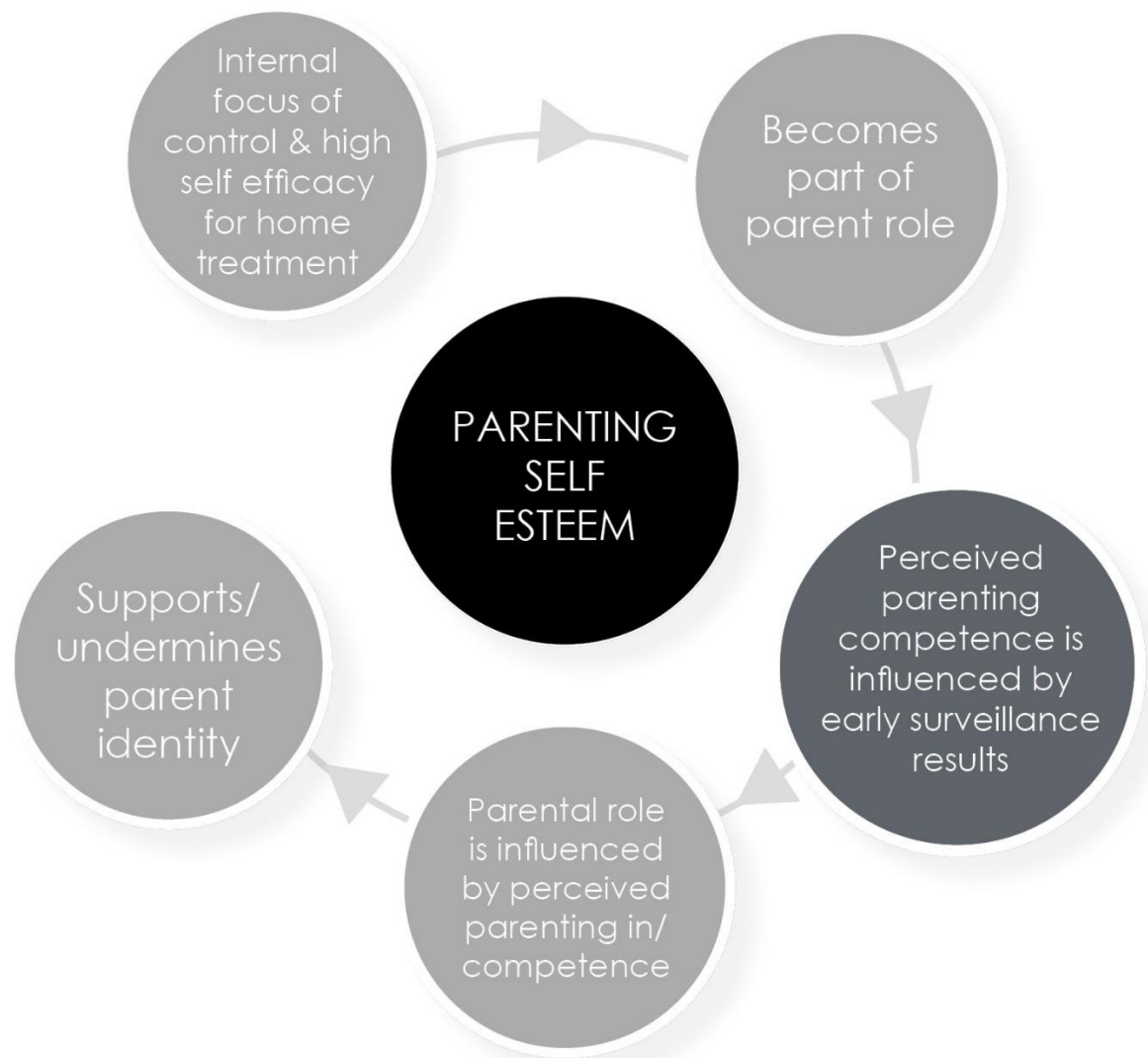


Figure 9. How early surveillance influences perceived parenting competence and possibly self-esteem

The current findings go beyond existing knowledge by highlighting how high levels of parental involvement in CF care, high importance attributed to the care and perceived accountability of child health all contribute to what it means to be a parent of a child with CF. Moreover, the current findings add to knowledge by identifying how early surveillance results may disrupt the sense of mastery parents feel about their child's treatment, which can influence perceptions of themselves as parents.

Finally, an external health locus of control for parents of chronically ill children has been shown to predict parental anxiety (Scrimin, Haynes, Altoè, Bornstein & Axia, 2009) and also has been associated with parenting stress (Hassall, Rose & McDonald, 2005). Green (2004) showed that mothers high on external locus of control felt more subjective caregiver burden than mothers who were high on internal locus of control. Moreover, studies examining personality attributes and well-being have reported that internal locus of control and high self-efficacy is associated with better psychological well-being (DeNeve & Cooper, 1998; Roddenberry & Renk, 2010; Streisand, Swift, Wickmark, Chen & Holmes, 2005). How early surveillance affects parents' psychological well-being warrants further investigation because it is plausible that such interactions exist between the early surveillance experience and parental psychological well-being. For example, attributions of internal locus of control and high self-efficacy may function as buffers against stress associated with receiving negative early surveillance results, and at the same time diminish self-esteem that could exacerbate stress (Abouserie, 1994). In summary, introduction of CF into the family system requires implementation of a new set of parenting responsibilities as part of a redefined and medicalised parental role. Parents whose children undergo early surveillance construct the sensitive measures of disease progression as an indicator – a judgement – of their ability to achieve these new parenting responsibilities and maintain child health. Therefore, this sub-theme demonstrated how early surveillance is constructed as an indicator of treatment efforts and competency throughout the year and as an examination of parenting ability, and if identified meanings attributed to those constructions affected parental identity and parental self-esteem.

5.6.2 Parental outcomes directly from early surveillance.

Significance attributed to the early surveillance event by parents meant that a number of psychological phenomena were experienced at the time of annual review. Parents experienced feelings of lack of control and helplessness when relinquishing their child for the procedures of early surveillance, that is, for the bronchoscopy and CT scan under general anaesthetic (GA). Parents also felt a sense of altruism for future CF generations as part of their decision to involve their child in early surveillance. Therefore, parents could simultaneously experience positive and negative outcomes on the day of their child's annual review.

5.6.2.1 Lack of control and helplessness at time of relinquishment for medical procedures.

Relinquishing a child to both actual harm (i.e., the procedures) and then further potential harm and threat (i.e., complications during procedures) is the most confronting component of early surveillance for parents - psychologically and behaviourally. Relinquishment of a child, the heart of the parents' life world, into a threatening situation they do not control causes feelings of helplessness. For parents of younger children, they are often relinquishing their children to people they have never met before.

Each year parents are required to relinquish their child for early surveillance procedures, and each year parents generally feel a sense of helplessness about how their child will react to the GA, as well as how they will fare during the medical procedures. A Western Australian mother of a 6 year old child expressed her feelings of helplessness when she said, *'it's very difficult because that part is out of your control and because then you walk away and you don't have sight of your child and that's extremely heart-wrenching because you have lost control and that loss of control...in your child's life when they need you the most.'* A Victorian father of a 4 year old child echoed this sentiment when he stated, *'just to know that something could potentially go wrong or you start thinking 'what if she never wakes up,' and those sorts of things... it can be, the moment that you're in the realisation of what you're doing or what the risks are, sometimes become rife and obvious. Sometimes my mind can start to get away from me and you just get to a point where you feel so helpless.'* The current finding is consistent with those of other studies (Bull & Grogan, 2010; Hughes & Callery, 2004; Salgado, et al., 2011; Vessey, Bogetz, Caserza, Liu & Cassidy, 1994) that show relinquishing children for medical procedures is generally a confronting and uncomfortable experience for parents.

The psychological consequences of relinquishment (i.e., lack of control and helplessness) generally become more manageable over time as expectations are developed based on previous experience. It is therefore assumed that as parents encounter these medical procedures more, most will cope better over time (Utens, et al., 2000). As relinquishment of their child is always a characteristic of early surveillance, as is uncertainty in child safety, so is this sense of helplessness during this time. Whilst lack of control and feelings of helplessness become easier to cope with over time and with experience, they never completely disappear.

In accordance with the finding identified in sub-theme *understanding early surveillance* - that over time, parents experience transition from no expectations to some expectations of the program, the current finding suggests that time and experience with early surveillance may facilitate parents' ability to manage and cope better with relinquishing their child for medical procedures, as well as coping with their own thoughts and emotions associated with relinquishment. The following excerpts demonstrate how expectations helped parents of children aged more than 3 years to cope with the medical procedures, whereas parents of younger children did not express this particular coping mechanism. Parents of older children expressed the evolution of their coping process on which younger parents did not comment:

Western Australian mother, 4 year old child - I would say that it's easier to handle. It all just comes back to experience and understanding again, knowing what's in store for you. Knowing how the procedures are going to go...definitely things have gotten better over time of knowing what to do and how to do it. It all just comes down to experience.

Western Australian father, 5 year old child - Yeah, it's probably got a bit easier I suppose. The first ones, you know, when he was a baby, were sort-of, you know, a little bit harder...it's probably just got a little bit easier as the years have gone on...so yeah, the way we deal with it. We know a lot more about it now. We sort-of know how to cope with it.

Literature from diverse disciplines (e.g., medicine, technology and psychology) across various situations (Borup, Brown, Konrad & Van Lente, 2006; Faasse & Petrie, 2013; Hauck, Fenwick, Downie & Butt, 2007; Liao, Gramann, Feng, Deák & Li, 2011) shows how expectations for an event help people to manage similar future events. Expectations provide a framework through which psychological boundaries created during early experiences control those psychological processes and outcomes for future experiences of similar events. In this way, and similar to creating expectations about the family and child after diagnosis, parents can reduce initially boundless thoughts and ideas of relinquishing children for procedures (and of the early surveillance event more broadly), which in turn helps parent to cope by reducing stress associated with those earlier boundless ideas and thoughts. Therefore, interventions aimed at supporting parents through their child's early surveillance procedures could encompass the need to frame expectations.

5.6.2.2 Altruism for future CF generations.

The opportunity to participate in early surveillance with the intention of contributing to future CF generations' lives through research is conceptualised in my research as altruism. As a result of their child's early surveillance involvement, parents described a sense of altruism. Parents who asserted altruism as their primary reason for early surveillance participation attributed high importance to research participation. Therefore, altruism in this instance is interpreted both as a consideration for, and as a beneficial outcome of, research participation. Parents' altruistic disposition is demonstrated by A Western Australian mother whose child was 5 years old, *'I see study participation as quite altruistic because any kind-of benefit is not necessarily going to rain down on XXX...in my eyes, we had to be altruistic because the benefits aren't going to rain down on your child but for other families, it might.'* This point of view was reflected by a Victorian father of a 2 year old child, *'the whole idea that it's going to go to contributing to people in terms of future benefits is a driving force for me.'*

Perceived direct clinical benefit for their child was another reason for participation. For these parents, altruism appeared as more of a bonus beneficial outcome than as a primary reason for research participation. These parents placed more importance on their child's direct clinical benefit than research for medical advancement, but still held importance for research participation. Cartwright, et al. (2011) found the same dominant two reasons for parents' consenting their children to a randomised controlled trial. A quote from a Western Australian mother of a 4 year old child

indicates her reasons for agreeing to early surveillance for her child, *'I hope it will help my kids hopefully more than anything, but also it's going to help future generations...I'm grateful that it's there, I'm grateful that I can participate in it. Firstly to get information for ourselves but secondly to help with research as well.'*

All parents placed some level of importance on being altruistic for research and scientific advancement purposes. They all acknowledged that early surveillance is something they wished their child did not have to do, but they certify its importance for their child's health and for future CF generations. The primary motivation behind altruism for future CF generations was that because children today have been afforded a longer and improved quality of life by parents of previous CF generations subscribing their children to such research programs, that parents wanted other children to experience the same benefit. A Western Australian mother of a 12 month old baby spoke about why she felt importance of participating in early surveillance, *'I feel like it's something little that we can do to make a difference for the kids in the future. I mean really, so much progress has been made and I feel like that's because the parents before us have sacrificed so much and gone through so many trials so I'd like to think that we can do that for parents coming through...there are families who have gone before them who have done the research and that's why our kids are doing so well now.'* However, an alternative explanation for this sense of altruism is that parents felt a sense of obligation or duty to continue the trend towards research participation, with less regard for their own innate thoughts and feelings about research participation.

From the point of damage to parental identity is the notion that an altruistic tendency may aid in its repair, and therefore contribute to effective parental coping. Feelings of altruism may form a counter-perspective of parental identity, damaged by diagnosis or receipt of unfavourable review results. Therefore, parents may draw on what positive aspects of the program they can to cope with the negative ones. To that end, negative aspects of the program (i.e., blame and guilt for negative results and lack of control and helpless due to relinquishment of a child for medical procedures) are intense and acute (i.e., experienced around the time of annual review), whereas positive aspects (i.e., perceived control and power over disease progression, parental competence and satisfaction and altruism) exist on a long-term scale. To this extent, it appears that perceived long-term outcomes aid parental coping with the intense, acute experiences around the time of annual review. It may also be that altruism experienced acutely around the time of annual review is associated with gratitude experienced on a longer-term basis on those days where parents have a positive outlook on their child's life. This idea, too, points to a relationship between acute experiences at the time of annual review for longer-term outcomes.

Literature exists on parents' initial decisions about involving their child in medical research, mostly in oncology (e.g., Chappuy, et al., 2006) and neonatology research (e.g., Jansen-van der

Weide). Altruistic motivation has been reported as a leading justification for parental consent in paediatric medical research, from polio vaccines in the 1950's (Clausen, Seidenfeld, Deasy & Clausen, 1954) and vaccine trials in the 1990s (Langley, Halperin, Mills & Eastwood, 1998), to more recent vaccine trials (Chantler, et al., 2007). The current finding is consistent with those of other studies (e.g., Chanter, et al., 2007; Chudleigh, et al., 2013; Steele, et al., 2014) and suggests that parents want to advance medical science, but primarily they want to help other children (and families) in the same situation as themselves. It is possible these results are due to participation viewed as a social obligation, particularly if their child benefited from prior medicine advancement that was a result of research participation by families previously in their situation.

5.6.3 Fear from diagnosis exacerbated at annual review.

Fear experienced around the time of annual review is distinguished from lack of control and helplessness as direct outcomes of early surveillance because fear originates at diagnosis, continues as part of the lived experience of parenting a child with CF, and is then exacerbated at annual review. It is perceived seriousness and risk of medical procedures that assigns additional reasons to previously experienced fear. The processes leading to fear differ from processes leading to lack of control and helplessness, in that medical procedures serve to increase, or intensify, fear experienced more globally, which was expressed by parents as occurring on their bad days.

The GA and the medical procedures were the components of the early surveillance program that concerned parents the most (Chudleigh, et al., 2013). Accordingly, the GA and medical procedures engender additional aspects to fear associated with CF. A Western Australian mother of a 2 year old child said, *'I'm scared of the GA more than anything else.'* Additional dimensions of fear were perceived risk of the medical procedures, potential trauma for the child, as well as lack of control and helplessness due to relinquishment. Due to the nature of early surveillance for CF lung disease, these additional reasons for fear have been reported for parents of children undergoing surgery or requiring hospital admissions (Bull & Grogan, 2010; Himes, Munyer & Henly 2003; Menahem, et al., 2008; Utens, et al., 2000).

Fear described in interviews appeared solitary and personal, that was often expressed to a partner or family member and generally not expressed within the family domain. Parents did however report being withdrawn from their family around the time of annual review. For example, a Victorian father of a 5 year old child stated, *'I just keep it to myself. When I stress, I try not to show it too much. I'm probably more reserved around the kids during that time.'* Similarly, a Victorian mother of a 2 year old child expressed her withdrawing behaviours in the time leading up to her child's annual review, *'I get a bit apprehensive around that time...it probably brings my mood down a bit, it makes me feel a bit deflated and I guess a little bit less engaged with the family for that period of*

time.' Though a limited number of parents recalled withdrawing from their families, and only for relatively short periods of time prior to their child's annual review, behavioural disengagement by parents of children with CF has been associated with child and parent maladjustment (Wong & Heriot, 2008), and should therefore be of concern to HCPs and researchers.

Parents' primary coping mechanism for fear and in the lead-up to the annual review was social support from friends, family and/or a partner. An example of how parents drew on personal support in the lead-up to annual review came from a Western Australian mother of a 2 year old child who said, *'well we talk about it, well (laughs) I'll talk to XXX (husband) about it, and I've talked to my friends about it. You know, I will talk about it.'* The current finding is consistent with CF research and other research showing that parents whose children undergo surgery commonly use social support as a primary coping mechanism (Grossoehme, et al., 2014; Hughes & Callery, 2004; Salgado, et al., 2011).

On the day of the annual review, the dominant coping strategy for relinquishing children for medical procedures was the distraction technique, an emotion-focussed coping strategy (Lazarus & Folkman, 1984) functioning to divert attention away from the stress of relinquishment until such time that the child was back with the parent. Distraction techniques included talking on a mobile phone or with the person who accompanied them, reading a magazine or visiting the café or gift shop. Examples from parents using distraction to cope include a Western Australian father of 4 year old child who said, *'we usually grab a coffee and walk around and buy a paper, go for a walk. You know, try and do tasks and things, play on my phone,'* and a Western Australian mother of a 5 year old child said, *'I usually take that time to run errands around the hospital like go and pick up some medications from pharmacy so that we're not waiting around ...so that certainly takes your mind off it...for me, if I'm not thinking about it at the time, not that it's not happening, but I can deal with it better.'*

The excerpts are from parents whose children are over 3 years of age, suggesting that coping with early surveillance may be a function of time and experience to learn effective coping techniques. Therefore, further understanding of parental factors that characterise parents who experience increased fear at the time of medical procedures, and how they cope will inform if, and what, interventions may help to alleviate fear during relinquishment of children for medical procedures.

Behavioural disengagement and distraction are considered maladaptive coping techniques (Carver, et al., 1989). They have been shown to reduce emotional and social quality of life (Kertz, Stevens, McHugh & Björgvinsson, 2015; Staab; et al., 1998) and increase depression, anxiety and stress (Sheehan, et al., 2014) both in parents of children with CF and adults with CF. Though parents described using these coping strategies for brief periods of time, due to the cross-sectional nature of

my research, I was not able to discern their temporal nature. Given my research findings, AREST CF clinicians and researchers should be cognisant of when parents *need to cope* with early surveillance, and that this period of time extends from the days prior to annual review, including the day of annual review and then ensues until receipt of early surveillance results. A Victorian mother of a 4 year old child explained this concept comprehensively when she stated what she needed to manage and when, and how she reframed the situation as leading to a perceived positive outcome:

I think at least the 48 hours before, and even the week before, we give XXX (child with CF) a present to take the hospital to have for when he wakes up after the anaesthetic, just to try and make the process a bit easier. So yeah, there's a bit of of dread...so I'm trying to, I think it's a really beneficial program so, but there's also the worry of what results we're going to get. So you kind-of think about this all before you go, and then it's about actually getting there. You know, the drive there, parking. Parking drives us up the wall. So yeah, I think there's never like, we kind-of just go 'well, we know that shit day is coming up.' You don't look forward to it obviously, I just look forward to knowing how the kids are progressing. That's probably the only positive thing as a parent.'

Currently there is only anecdotal evidence and clinicians' assumptions about points in the early surveillance process with which parents may be struggling. Beyond simple awareness of when and how parents cope, a support program or intervention strategy could be developed and implemented that is responsive to the findings in my research about parental coping strategies and techniques. Finally, if chronic sorrow was indeed experienced as a result of CF diagnosis, it too, may be exacerbated by CF coming to the forefront of parents' minds as the significant event of early surveillance looms. Perceived significance of the event may exacerbate chronic sorrow around the time of annual review as a reminder of the diagnosis and prognosis of CF itself.

Chronic sorrow theory suggests that an exacerbation can occur as a result of a medical crisis or event that magnifies the loss and disparity between reality and the life once imagined (Eakes, et al., 1998). The event can therefore trigger a return of profound sadness. Parents expressed experiences of recurring sadness throughout the year (as discussed in the theme *good days and bad; fluctuation between positive and negative*), with the annual review emerging as a trigger that exacerbates chronic sorrow. Bluebond-Langner (1996) described the content and process of triggers as information moving back and forth, that is, devastating characteristics of CF (namely morbidity and mortality) come to the forefront of parents' minds where it has been pushed to the back of the mind for the most part, resulting in those emotional and cognitive responses experienced at diagnosis "by thinking about it all over again" (Bluebond-Langner, 1996, pp. 150)

Reports in the literature portray events associated with a child's CF as intruders to a generally dormant condition in the early years of life, which can make their presence known to parents and interrupt the normality that has been redefined for the family (Bluebond-Langner, 1996; Glasscoe & Smith, 2011). It appears that for parents in my research, the early introduction of a significant event (that is, early surveillance) becomes part of the redefined sense of what is normal for the family. Not to minimise the effect this has on parents, but they appear to assimilate this event (and its associated information about their child's disease progression) into their redefined lives where family life appears to remain stable throughout (and as a result of) the event. However, it remains that fear and potentially chronic sorrow can be re-visited, and warrants further investigation.

Finally, a pertinent consideration must be that, although logically (and described in the current finding) parents cope better with feelings of lack of control and helplessness, and fear over time, it is possible parents experience PTSS or clinical levels of depression associated with their child's ongoing surgical procedures and/or subsequent treatment. For example, parental depression can be prevalent up to 1 month after a child's surgery (Ben-Amity, et al., 2006) but generally not 12 months later (Menahem, et al., 2008). The current finding and other literature (Bearden, Feinstein & Cohen, 2012; Fincher, Shaw & Ramelet, 2012; Jay & Elliot, 1990) highlights potential need for short-term intervention that supports parents' well-being prior to, and following their child's early surveillance procedures.

Whilst parents generally appear resilient to the burden of early surveillance, the results do need to be interpreted with caution. Firstly, there is the possibility that PTSS or clinical levels of depression and/or anxiety are experienced by parents who chose not to be part of the research. Secondly, as this research is based on retrospective data, parents may indeed experience withdrawal and/or PTSS symptoms annually around the time of their child's early surveillance procedures but that were not recalled during interviews.

In summary, it appears that perceived accountability for child health may be a unique psychological phenomenon for parents in my research. The accountability is constructed from the nature of CF management and its association with parents' constructions of detailed information about a child's health. The consequence of this association is that parents use their child's early surveillance results as an indicator of their treatment efforts, which influences perceived parenting competency. Parents' experience of their child's early surveillance included feelings of lack of control and helplessness from relinquishing their child for medical procedures, however parents tend to manage these feelings better over time. Parents also experienced altruism as a positive consequence of participating in medical research. Fear experienced at diagnosis can be exacerbated at the annual review due to the risk in medical procedures, potential trauma to the child and parents' feelings of

helplessness. By virtue of annual review results being unique to parents in my research, the experience of early surveillance and its outcomes produce unique parental psychological outcomes that are independent of CF.

Accordingly, the current theme demonstrated that early surveillance is constructed as a significant event that has boundaries and evokes emotion. Boundaries of early surveillance circumvent the reminder of impending annual review to receipt, and any consequences, of review results. The significance of the event appears to bring CF to the forefront of family focus. It is not that parents ever forget their child has a life-threatening condition, but potential adverse consequences of the impending annual review can aggravate psychological aspects of CF that have been somewhat suppressed or negated throughout the year. Aggravation causes exacerbation of those psychological parental outcomes of having a child with CF.

Thus far, the current theme (and its sub-themes) has begun to describe parents' lived experience of CF within the context of early surveillance and how there are confronting, and sometimes conflicting, psychological processes occurring. Moreover, both beneficial and adverse outcomes for parents of children with CF within context of early surveillance have thus far been described. Conflicting responses caused between presence of both positive and negative outcomes is the matter of the next theme *early surveillance is in the best interest of my child's health*.

5.7 Early surveillance is in the best interest of my child

This theme continues the presentation of parents' experiences and constructions of their child's early surveillance, and articulates parents' ultimate perception that early surveillance will benefit their child's long-term health because it may save, or at least extend, their child's life. The purpose of this theme is to describe the primary reason for parents' decision to enrol their child in early surveillance, concluding with discussion about what potential effects the decision to participate in early surveillance has on parents. Sub-themes identified to illustrate these constructed thoughts and experiences include; perceived clinical benefit to child health – presence of the therapeutic misconception?, and cognitive dissonance results in anxiety and/or ambivalence.

5.7.1 Perceived clinical benefit to child health – presence of the therapeutic misconception?

The primary clinical aim of early surveillance is to detect disease early so that therapeutic treatments can be introduced, with the intention to try and prevent disease progression, or at least delay it. The primary research aim is to understand the pathogenesis of lung disease in early life and identify modalities that allow clinicians to detect lung disease in its early stages. The primary aim for parents is to preserve their child's life through ensuring any disease progression is treated as early as possible. Parents' perceptions that early surveillance will directly benefit their child through improved clinical health outcomes is illustrated by quotes from a Western Australian mother of a 5

year old child who said, *'it's one of those things we know we have to do because of the benefits of it, so there's that mixed with the feelings that you don't want to do it...we prioritise what she needs as opposed to our stress,'* and from a Western Australian father of an 18 month old child who said, *'it's got to be worth it because you can give your kids a few extra years... it's the fact that it can give your child however many years of life and good quality of life.'*

Belief in improved child health outcomes is an artefact of parents' constructions that early surveillance will result in direct clinical benefit to the child. However, the clinical benefits ascribed to early disease surveillance are based on logical inference rather than evidence-based clinical outcomes. That is, that by detecting disease early in life, treatment may be instigated early that could delay or slow down disease progression. Surveillance of CF lung disease itself is not a therapeutic intervention. In absence of evidence-based outcomes, there are a number of reasons for this circumstance. As CF treatment and standards of care have changed dramatically over recent years it is difficult, if not impossible, to attribute clinical health improvements to the early surveillance processes. Also, as early surveillance is still relatively in its infancy, long-term health outcomes are yet to be characterised.

However, there are a number of indications that early surveillance leads to better health outcomes for children with CF. Evidence to date suggesting intervention as a result of early surveillance modifies clinical health outcomes is provided through comparison of early surveillance data with Australian CF registry data. This comparison shows that children with CF under 7 years of age in Western Australia and Victoria are healthier than children in other states, indicated by nutritional indices and pulmonary function (Cystic Fibrosis Australia, 2013). The issue for valid comparison is lack of control data for confounding factors that may influence these data, for example, environmental factors associated with socio-economic status may affect a child's respiratory health (Neidell, 2004; Thakur, et al., 2013). Another source of evidence suggesting early intervention leads to improved health outcomes can be inferred from NBS research, which shows that early diagnosis and subsequent early introduction of CF treatment benefits children's nutritional outcomes (Farrell, et al. 2001).

Without evidence-based information of clinical benefit, and inferences made from CF registry data and NBS data, does this mean the *therapeutic misconception* is present for parents who believe their child will benefit clinically from early surveillance of CF lung disease? Consideration of the term will enlighten the reader to its conception as well as its potential existence in the population under investigation. Therapeutic misconception is a common ethical problem encountered in human participants research. First described by Appelbaum in 1982, many people were unaware of the differences within a clinical setting between research participation and receiving treatment. Instead

the belief was that therapy and research were governed by the same goal, that is, to advance the individual patient's best interest.

Although it may be a consequence of research participation, the primary goal of medical research is to generate data that could lead to improved care for future patients (Dresser, 2002). The therapeutic misconception describes the belief that the purpose of a clinical trial is to directly benefit the individual patient rather than to gather data to develop scientific knowledge (National Bioethics Advisory Commission). Put simply, it is the belief of patients that by entering a research study, they will benefit directly from participating, regardless of whether they actually will or not.

Although the therapeutic misconception has been well described (Joffe & Miller, 2006), it has not been considered in the context where parental consent is a requirement (Woods, Hagger & McCormack, 2014). Shilling's and Young's review (2009) of parents' experiences of being asked to enter their child into a randomised controlled trial found that parents felt that experimental treatment was superior (i.e., therapeutic misconception), suggesting that parents may construct trial treatment as the answer to their child's needs. Parents in my research may construct early surveillance in this way because of the design of the program, in that children will be treated earlier than if they were not part of the research, though clinical benefit is currently unknown. The generally high recruitment rates in neonatology (Campbell, Surry & Royle, 1998), and paediatric oncology (Ablett & Pinkerton, 2003) research, and indeed in early surveillance (99%) could be indicative of parents' constructions of research participation as directly benefitting their child (Chudleigh, et al., 2013; Dixon-Woods, et al., 2008).

Lastly, evidence that attributing meaning of clinical benefit to early surveillance is a co-constructed process was highlighted when reviewing understanding of early intervention among parents. Whilst parents across both CF clinics acknowledged and made reference to keeping children healthy through performing adequate levels of standard care, they also strongly identified with the role of early intervention in their child's long-term health outcomes. The following excerpts illustrate parents' value towards the opportunity for early intervention as a result of early surveillance:

Western Australian mother of a 12 month old baby - for me, it's really important because we want to know, we want to pick things up early and get onto it. That's the whole reason we came to WA, because we know that AREST CF was here, we knew that it was a world-class program and we just want what is best for her, and I think the team can really provide that.

Victorian mother of a 4 year old child - I mean it's a weird thing because you feel like it's a tough road to be on, but then there's also the security of knowing that your child is

getting treated as soon as something comes up, so what you go through really is the best thing for your child.

In reviewing the key health improvement strategies for children with chronic conditions promoted by the Australian government, it is evident that early intervention is one of the primary foci (Commonwealth Department of Health and Aged Care, 2000; Commonwealth Department of Health and Ageing, 2009). Therefore, the current finding appears to reflect the dominant health promotion discourse within this context. Accordingly, this finding illustrates that dominant discourses may influence parents' constructions of meaning in early surveillance. Collectively, findings from my research revealed that constructs such as early intervention influence parents' thinking about their parenting behaviours and affects how they relate to their own experiences and understandings of parenting within context of their child's condition. Assumptions underlying an early intervention approach highlight how policy and programs referring to such concepts can influence parents' own constructions of meaning in health promotion and thinking about what decisions they *ought* to make. For this reason, it is important to be mindful of how certain discourses become the dominant ones and to question whose needs they serve so that informed and unbiased decisions can be made.

To conclude this sub-theme, the therapeutic misconception is detrimental to an individual's understanding of research, which is crucial for an autonomous decision because it can undermine the ability of the decision-maker to make an accurately-informed decision. If, indeed, the therapeutic misconception is present for parents in my research, it means they may make inaccurate judgements based on false beliefs as part of their process in appraising the research study. Based on the current finding there is an urgent need to identify if strategies to avoid the therapeutic misconception are required. Discussion now shifts focus to describe how cognitive dissonance can be experienced by parents once the decision to participate in early surveillance has been made. Discussion about cognitive dissonance will round out this theme by describing how early surveillance is interpreted by parents as being in the best interest of their child's health, but with some adverse psychological consequences for parents.

5.7.2 Cognitive dissonance results in anxiety and/or ambivalence.

Western society promotes autonomy for medical treatment decisions and research participation, however the consequences of such decisions are not as clear-cut as their idealities. Psychology research has shown that agency and internal locus of control associated with autonomous decision-making lead to a perceived sense of personal causality (Hashimoto & Fukuhara, 2004; Cobb-Clark, Kassenboehmer & Schurer, 2014; Pettersen, 1987). Research more specifically in paediatrics (Botti, Orfali & Iyengar, 2009, Polizzi, et al., 2015), and in keeping with psychological research on decision-making, shows that outcomes of decisions made by parents are internally

attributed to their control, thereby magnifying emotional response to an event as a result of a parental decision. This means that accountability exists in an ethical decision that emerges from parents' demand of responsibility.

Some psychological consequences of parental decision-making about children's medical research participation reported by Botti and colleagues (2009) included increased grief, distress, anxiety, guilt and blame, and ambivalence about autonomy, which resulted in decreased coping ability. Consequently, psychological confusion, or cognitive dissonance, may manifest itself in ambivalence and anxiety in decisional autonomy. For parents in my research, cognitive dissonance was experienced acutely around the time of annual review, reaching its climax at the time of relinquishing their child for medical procedures. Cognitive dissonance generally resulted in feelings of anxiety or ambivalence about the decision to participate in early surveillance (Lavine, Thomsen, Zanna & Borgida, 1998; Maio, Esses & Bell, 2000; van Harreveld, Rutjens, Rotteveel, Nordgren & van der Pligt, 2009).

Cognitive dissonance is an emotional state occurring when two simultaneously held attitudes or cognitions are inconsistent, or when there is conflict between belief and behaviour (Festinger, 1962). Festinger's (1962) cognitive dissonance theory focuses on how individuals strive for internal consistency. When inconsistency (dissonance) is experienced, individuals largely become psychologically distressed. Festinger conceptualised the existence of dissonance as psychologically uncomfortable, which will motivate an individual to try to reduce the dissonance and achieve consonance.

The presence of psychological discomfort for parents in my research as a result of cognitive dissonance may transpire through two means. Firstly, cognitive dissonance may transpire because of contradictory speculations about putting a child through a traumatic experience but for the greater good of their health. For example, a Western Australian father of a 4 year old child stated, *'we think its (early surveillance) important but there is that part of you that says "do we really need to be doing this?" So it's a lot of questions that come up. But at the end of the day, we want to do what's right for her. I suppose you just have a lot of questions that get raised in your own head.'* Secondly, cognitive dissonance may transpire because the act (behaviour) of relinquishing a child to undergo a (perceived) potential harm is contradictory to parenting responsibility of protecting children from harm. For example, a Western Australian mother of a 6 year old child said, *'I feel like my responsibility as a parent is to protect my child from harm, but then I'm putting him in a threatening and potentially harmful situation.'*

For these reasons, cognitive dissonance could become a lived experience for parents in my research. Echoed by Glasscoe and Smith (2011), they reported two frames of reference in making decisions about a child's CF treatment; self as mother, which referred to the intimate relationship

with her child and which was governed by emotional binding, and self as caregiver, which referred to her professional role that was guided by logic and reason. Therefore, cognitive dissonance can create negative psychological responses for the individual experiencing the dissonance (Elliot & Devine, 1994; Festinger, 1962; Harmon-Jones, 2000). For parents in my research, contradictory beliefs versus behaviour generated anxiety whereas contradictory beliefs about early surveillance generated ambivalence.

An example of anxiety caused by contradictory belief and behaviour came from a Victorian mother of a 6 year old child who stated, *'it just makes me feel anxious that I'm supposed to protect her, yet something bad could happen because I put her in a dangerous and possibly harmful situation.'* Contradictory beliefs can result in ambivalence towards a subject matter (Ben-Nun Bloom & Arikan, 2011; Castro, Garrido, Reis & Menezes, 2009; Moody, Galletta & Lowry, 2014). A Western Australian mother of a 3 year old child expressed ambivalence about her decision when she stated, *'yeah, I was just unsure about it. A bit apprehensive really. I didn't really want to do it but of course you want to do it at the same time to make sure he's ok. So yeah, I was just unsure about my decision to do it and then to keep doing it.'*

Ambivalence in psychological terminology refers to co-existence within an individual of both positive and negative feelings towards the same person, object or action, simultaneously drawing the individual in opposite directions (Lewin, 1939; 1943). Parents have positive feelings of power and control over their child's disease that eventuate from early surveillance results. They also have negative feelings of lack of control and helplessness due to relinquishment of their child to threat and potential harm, and possibly blame and guilt for their child's traumatic, potentially dangerous situation (Salgado, et al., 2011). The affective conflict about early surveillance leads to ambivalence about the decision to participate.

Resolution of cognitive dissonance (i.e., consonance) is explained by cognitive dissonance theory, which conceptualises attitude change from motivation to adjust attitudes to relieve cognitive dissonance (Festinger, 1962; 1964). For the contradictory behaviour against the belief about threat of harm, parents adjust their beliefs so as to align with their behaviour. In this sense, the behaviour of relinquishing their child for surveillance of their chronic, often fatal illness is perceived as better than the behaviour of not being involved in early surveillance. Another conception of this is that parents would always choose to do whatever they could for the perceived benefit of their child's health if it was deemed to be safer than not doing anything. A Victorian father of a 4 year old child showed how he adjusted his attitude to match his behaviour when he expressed that, *'our goal is keep our kid healthy. If that means putting her through some traumatic stuff with the purpose of keeping her healthy and alive for longer, then that's what we'll do... so I just have to adjust the way I think about it, which then makes me feel better about putting her through that kind of stuff.'*

For the contradictory beliefs about trauma of early surveillance versus the greater good for child and future CF generations, parents are in a continual cycle between the former and latter beliefs. However, the belief in early surveillance as the best option for a child brings a sense of cognitive resolution. For example, a Victorian mother of a 3 year old child expressed, *'there's a lot of guilt, but you know, deep down that when we decided to do the research, we know that it was for good. For her benefit and obviously for future CF kids, so we said that it's going to benefit her so that's what we try to keep always in our minds. That helps me to deal with our decision.'* If cognitive dissonance is resolved, parents can still return to the cognitive dissonant state (Wicklund & Brehm, 1976), either as part of their continual evaluation of participation at each annual review or general feelings of uncertainty.

Trust can function as a resolution to cognitive dissonance (Festinger, 1964). Partial or complete elimination of cognitive dissonance may be a function of lowering expectancy that an unfavourable or undesirable outcome will occur (Aronson, 1969). Trust functions to reduce possible outcomes for a particular event because without trust, all contingent possibilities should be always considered, leading to a paralysis of inaction (Braynov & Sandholm, 2002). A Victorian mother of a 4 year old child demonstrated how this occurs, *'I'm thinking 'is she going to die?' Then I pull my strength to thinking about 'that's irrational, what's the rational thought?'...I still feel, my heart is still beating fast but it's all fine, I trust in the doctor's abilities to look after my child.'* Therefore, it appeared that trust allowed parents to relinquish their child to the medical team by enabling parents to have confidence that their child was being taken care of. Moreover, it appeared that trust helped parents to cope with relinquishment, expressed by a Western Australian mother of 4 year old child who said:

well I think having trust in the staff helps me to cope with the procedure because I think if you sit there, you start to think about things like 'she's having the tube put down her throat, it could scratch the side of the oesophagus and then she might bleed,' so if you tend to trust that what they're doing is the right thing, then you're not worrying yourself so much.

Application of the current finding to Hall's (2002) conceptual model of trust within a healthcare setting highlights importance of perceived competence of physicians for parents in their capacity to trust medical staff. The notion of competence is central to trust within a medical setting, and is divided into interpersonal and technical competence (Hall, et al., 2002). Competence is the possession of required adequacy in a given situation (Reber & Reber, 2001), and so applied to interpersonal competence within a medical setting, is the perception parents have about the adequate nature of a physician's interpersonal skills and demeanour (Hall, et al., 2002). A Victorian mother of a 4 year old child expressed this element of trust when she said, *'trust has a lot to do with*

it...I mean, you get to know them personally, one on one. So when they come up to you and say such and such is going to happen and you look at them in the eye and you go 'yep right, ok.' Technical competence is viewed as having good practices, making correct decisions, avoiding making mistakes and producing the best achievable outcomes possible (Hall et al.), and is generally evaluated through considerations of physician experience, thoroughness and knowledge. This assessment is mainly based on patient experiences with the clinician. A Western Australian mother of a 5 year old child described this element of trust when she stated, *'I kind-of think that they do this every day so you just have to trust their judgement.'*

To conclude this sub-theme, simultaneous opposing attitudes, values, beliefs and behaviours can result in cognitive dissonance, which can result in anxiety and/or ambivalence about parents' decision to involve their child in early surveillance. Cognitive dissonance, and its resulting anxiety and/or ambivalence, appears a parental compromise for the perceived beneficial, long-term child health outcomes of early surveillance participation. Trusting in medical and research staff can help parents to manage psychological adversity associated with their child's early surveillance participation. Therefore, developing and maintaining a therapeutic relationship comprising physician competence and parental trust is of paramount importance given the reciprocal and continual nature of the therapeutic relationship for ongoing care for children with CF.

In review, the decision to participate in early surveillance for CF lung disease is primarily made on the premise that it will directly benefit a child's clinical health outcomes, resulting in transcending feelings of hope for their child's future. However, a salient parental outcome from the decision to participate in early surveillance is cognitive dissonance, which can result in anxiety and/or ambivalence about the decision. Parents are willing to deal with the such psychological adversities associated with early surveillance as a compromise for the perceived direct benefit to their child and the larger CF community; this is how early surveillance is believed to be in the best interest of the child, but my research has shown, notwithstanding psychological adversity for parents.

The final, transcending interpretation of the lived experience for parents whose children undergo early surveillance for covert CF lung disease is an assertion of hope. Hope for the future. Hope through participation in early surveillance transcends all themes, and is therefore interpreted as an over-arching and ever-lasting structure of parents' lived experience. Whilst parents did not speak explicitly about hope, its placement at the end of the findings chapter allows me to convey its transcendence in terms of its essence to the lived experience of parenting children with CF in the context of early surveillance. It allows me to leave the reader with the sense of transcendence with which it exists for parents. That is, hope for a child's future, for future CF generations, and for a cure are the primary outcomes of early surveillance for parents. The sense of hope transcends parents' experiences, emotions, cognitions, and outcomes associated specifically with early surveillance,

therefore substantiating that early surveillance is perceived by parents as being in the best interest of the child.

5.8 Construction and conceptualisation of hope

When an infant is diagnosed with CF, without hope for their future there seems little point of vigilance in treatment and health awareness, and little point of sometimes struggling and sometimes embracing life with CF. However, from devastation of a CF diagnosis rises hope for a child's life; hope is primarily for a child's future in terms of longevity, quality, normality and health.

According to the Ortony, Clore and Collins Classification System of Emotions (1990), hope is a primary emotion that refers to feeling pleased about the prospect of a desirable event; it is a positive attitude based on an expectation of positive outcomes related to events and circumstances in one's life or the world at large. Hope has been linked to the perceived availability of successful pathways related to goals, combined with a determined plan for reaching goals along with a perception of successful agency related to the goals (Synder, et al., 1995).

AREST CF provides a goal-oriented positive orientation towards the future (Cutcliffe & Herth, 2002; Samson, et al., 2009); it offers parents a pathway to achieve their new end goal after a child's diagnosis irrevocably changes initial parent and family goals. AREST CF offers a window of opportunity, of hope, to treat a child as early as possible. The notion of *earliest surveillance leading to earliest possible treatment available in the world* is perceived by parents as the best chance their child has at avoiding morbidity and achieving increased life expectancy, which creates hope for a child's future. Therefore, early surveillance for CF lung disease is seen by parents as a successful pathway for their new goals that includes a perception of successful agency related to such goals. That is, the sense of control created by early surveillance acts in both existential and practical terms in reaching parents' alternative goals for their child created as part of the *new normal* after assimilation of diagnosis into the family. Moreover, agency has been reported as a significant factor in predicting subjective well-being in parents of children with special needs (Shenaar-Golan, 2015). As AREST CF provides a pathway to achieving new goals and a sense of agency towards achieving them, it may well contribute to their subjective well-being.

Parents' construction of hope for a child's future – their lifeworld, occurs through their beliefs in treatment as the best option because of perceived direct clinical benefit to the child health outcomes generating assurance in child health. Recall that hope for a child's future could be constructed early on when uncertainty in morbidity and mortality are understood (in sub-theme *Understanding CF*). Hope can then become crystallised over the first few years in early surveillance through belief that early surveillance and subsequent treatment will provide direct clinical benefit.

Uncertainty is a precondition for hope, as hope is about possibilities (Verhaeghe, van Zuuren, Defloor, Duijnste & Grypdonck, 2007). As early surveillance functions to reduce

perceptions of uncertainty about disease progression, it may moderate the relationship between hope and uncertainty. That is, early surveillance may decrease uncertainty, which could increase hope. Additionally, hope in the face of uncertainty contributes to adaptation (Samson, et al., 2009; Truitt, Biesecker, Capone, Bailey & Erby, 2012), suggesting that positive parental outcomes from early surveillance (i.e., increased hope, reduced uncertainty, agency and perceived control) may function as moderators of adaptation and effective coping, which could ultimately influence resilience.

Whilst it is a complex concept, there is a consensus that hope has a role in helping individuals to cope with existential challenges, including the ability of a parent to cope with their child's life limiting disease (Samson, et al., 2009). Hope has been reported as a protective factor against psychological distress in parents of chronically ill children (Barlow & Ellard, 2006; Ogston, et al., 2011), functioning as a moderator between stress and maladjustment, suggesting a buffering effect against maladjustment (Horton & Wallander, 2001). Based on outcomes such as these, hope is understood to provide the basis for resilience (Petersen & Wilkinson, 2015).

Whilst parents in my research fluctuate between positive and negative outlooks on life, regardless of parents' transitory state of being, their hope never fades. All adverse parental experiences and outcomes that result from early surveillance are transcended by the hope they sense from early surveillance for their child's long-term health. That is, adverse parental experiences associated with early surveillance are a by-product of hope for long-term child health, which can reduce parental anxiety and emotional impact of CF (Wong & Heriot, 2008). In context of my research, it appears that hope afforded by participating in early surveillance functions to reduce psychological distress associated with parenting a child with CF. The interpretation of hope as a lived experience for parents whose children undergo early surveillance supports the notion of short-term parental adversities caused by early surveillance (e.g., fear and helplessness at relinquishment, guilt from negative annual review results, anxiety and ambivalence from cognitive dissonance) as a parental compromise for long-term child, parent, family, and CF community benefit.

Knowledge is the outcome of early surveillance for CF lung disease; about a child's current health status and also about CF disease progression in early childhood. Being part of a program that is the first in the world to examine the earliest signs of lung disease in young children with CF, parents not only carry hope for their child but they also carry hope for future CF generations. In terms of hope for future CF generations, parents are contributing to science through research, which they perceive as hope for children in the future through their contribution to research now. Ultimately, there is hope for a cure for CF, however this more global element of parents' hope is secondary to the direct hope for their child's future.

Knowledge is power. Power affords hope. Therefore, hope is the essential meaning of early surveillance for parents. Hope for the future, child, CF community - a cure. The diagram below depicts processes and factors for parents' construction of hope. The diagram illustrates how belief in early surveillance as the best treatment option because of perceived direct clinical benefit generates perceptions of improved child health, which creates a sense of assurance in child health. Assurance in child health from improved health outcomes facilitates hope for their child's future.

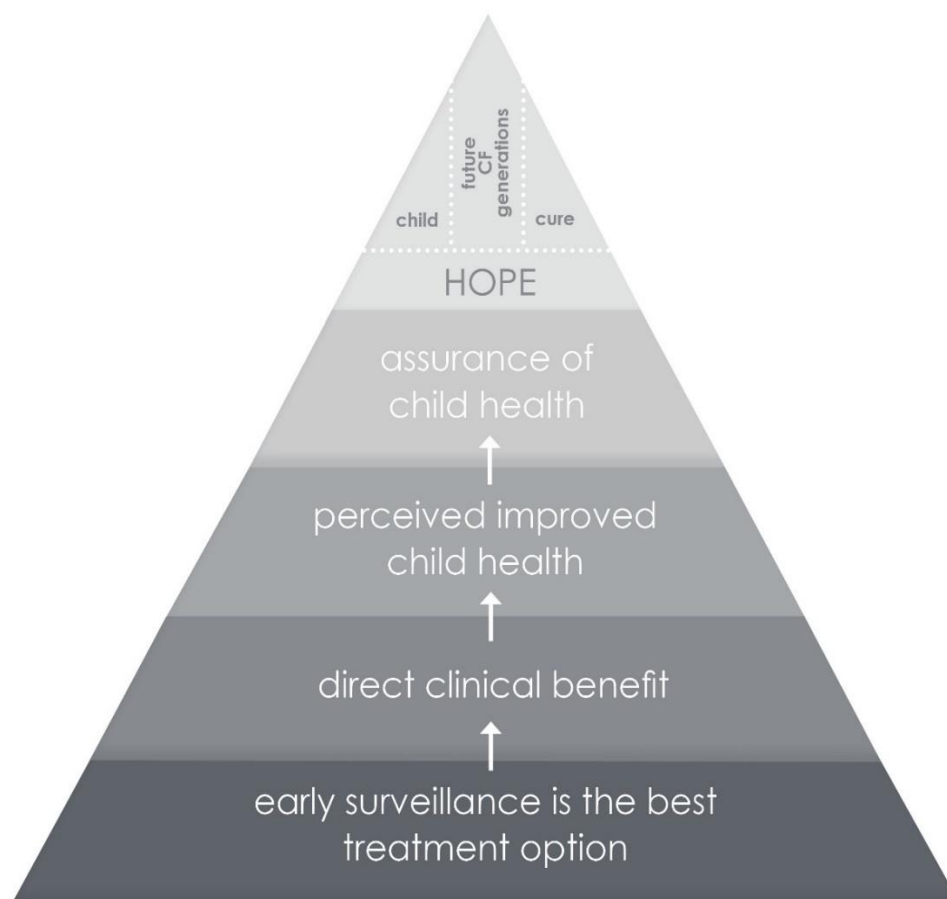


Figure 10. How hope is constructed by parents

Hope for the future is both a product of, and transcends, CF and early surveillance. In a medical context, hope is "strongly linked to the consumption of medical technologies, particularly therapies that promise to offer cures," (pp. 115) (Petersen & Wilkinson, 2015). Whilst early surveillance for CF lung disease does not claim to offer a cure, dramatic technological advances in medicine, such as those offered by AREST CF, offer more potential opportunities for hope for recovery than were available in the past (Gengler, 2015). In context of my research, hope is for improved health outcomes and hope is for future CF generations. Ultimately, hope is for a cure. As a dominant parental outcome of early surveillance, hope is interpreted as a lived experience for parents of children undergoing early surveillance of CF lung disease. Hope appears to overcome fear and reduce stress, which likely contributes to resilience; and it appears to develop from perceived

control over disease progression afforded to parents by AREST CF providing a sense of existential and practical agency to achieve new end goals.

5.9 Summary and conclusion

The underlying aims of this chapter were to describe how parents in Western Australia and Victoria whose children have CF constructed meaning and understanding about the experience of parenting within the context of early surveillance for CF lung disease in the first 7 years of life, and how they cope. Five major themes emerged and were explored, as were their respective sub-themes, these were, 1) Redefined Expectations and Reimagined Identity; Redefined Reality and the sub-themes of Collapsed and Redefined Expectations, and *New Normal*, 2) Understanding the Unknown and Understanding Uncertainty and the sub-themes of Understanding CF, and Understanding Early Surveillance for CF Lung Disease, 3) Good Days and Bad Days; Fluctuation Between Positive and Negative Outlook on Life and the sub-themes of Bad days, Good Days, and Fluctuation Between Good and Bad days, 4) Annual Surveillance is a Significant Event and the sub-themes of Indicator of Treatment Efforts, Parental Outcomes Directly From Early Surveillance, and Fear From Diagnosis Exacerbated Each Annual Review can Result in Anxiety, and 5) Early Surveillance is in the Best Interest of My Child with sub-themes of Perceived Clinical Benefit to Child Health and Cognitive Dissonance Results in Anxiety or Ambivalence. Evidence was provided for each theme through the provision of examples drawn from the data and linked with the literature. Lastly, hope for the future was interpreted as an over-arching and pervading parental outcome from their child's early surveillance.

There is much empirical and anecdotal evidence regarding adjustment and adaptation, or normalisation, for parents of newly diagnosed infants (Glasscoe & Smith, 2011; Hodgkinson & Lester, 2002; Hopkins & Gallo, 2012; Knafel, et al., 2010; Ray, 2002). Diagnosis of CF via NBS and its treatment become assimilated into family life, and parents' experiences from my research provide further support for the importance of adaptation for long-term parent mental health and family functioning, and child physical health. However, the most salient finding from parents' experiences of their child's CF diagnosis related to the collapse of child, parent and family expectations and rebuilding expectations and parental identity, which were commonly discussed outcomes for parents in my research. These experiences also affected the way parents viewed their child's and family's future by influencing their expectations of family life both in both positive and negative ways. This was particularly evidenced by parents recalling negotiation from devastation of diagnosis to acceptance of their new way of life.

In addition, both the parental role and parental identity were reconstructed to include the carer who administered intensive and time-consuming treatment. In particular, parents appeared vigilant about administering treatment, which resulted in high self-efficacy for treatment

management. Although some of the above factors led to early adverse responses and reactions, parents described how support from medical staff and from family and friends, along with time to develop understanding and experience of treatment management, contributed greatly to their negotiation of parenting a child with CF. Support was identified as important to parents' experiences of diagnosis and treatment management, and highlights the importance of drawing attention to available support mechanisms and resources. It also raises awareness of those parents without family support or who are geographically isolated may struggle to cope effectively and might encounter barriers to successful management, adjustment and adaptation.

Despite the emphasis of treatment management of paediatric CF primarily focussing on child health outcomes, findings from my research support that a more holistic approach to caring for children with CF is needed that takes into account the well-being of the family. For example, parents frequently referred to intra-psychic processes that resulted from becoming primary carer for their child. Undervaluing these psychological aspects important for treatment management restricts possibilities for addressing the more holistic needs in the promotion and maintenance of health and well-being, and potentially limits opportunities for the families' adaptation and resilience. Moreover, findings from my research offer insight based on parents' lived experiences that can be used to improve delivery and implementation of future policy and practice for diagnosis of CF via NBS and its management in the early years of life.

Another salient finding from my research indicated that constructed thoughts about early surveillance for covert CF lung disease at both sites were primed by biomedical discourse used to describe their early surveillance motivations. Findings particularly highlighted that constructs such as *disease progression* and *early treatment* were well-recognised by parents, as well as influencing their health beliefs and behaviours towards their children. However, though parents' initial comments about their experiences of their child's early surveillance frequently related to adverse experiences and outcomes, their experiences also reflected that the positive psychological factors influencing their experience of early surveillance were of equal, if not greater significance. This was evidenced by the volume of parents' discussions focussing on beneficial long-term outcomes associated with early surveillance. Significantly, these factors affected perceptions of themselves as parents and the future of their child.

A particularly noteworthy finding from my research was that it was more common than not that parents expressed feelings of hope and control when reflecting on their early surveillance experience. Feeling hopeful and in control because of, and grateful for, their experience was a common way for parents to express how they attributed meaning to early surveillance and ultimately influencing their appraisal of their child's condition and their early surveillance experience itself. Despite the challenges faced by parents, findings from my research indicate that parents had

good emotion regulation. This was primarily evidenced by their maintenance of a positive attitude expressed through acceptance of change resulting from their child's diagnosis and the regulation of the dynamics between adverse and beneficial outcomes of early surveillance.

Moreover, how parents constructed meaning of their early surveillance experience appeared to be primarily mediated through temporal factors mostly reflected in context of gaining knowledge and experience over time, which facilitated understanding. Making comparisons with others (i.e., children, families) also allowed parents to construct meaning about their early surveillance experience, as well as provided a way of thinking about, and planning for their child's, and family's futures. In addition, the notion of gratitude was found to mediate participants' constructions of meaning around their early surveillance experience with parents using it to create a perception of appreciation, enabling them to maintain a sense of indebtedness for their early surveillance experience. Lastly, support from medical and research staff, family and friends was a primary coping resource, permitting successful management of practical and psychological aspects of parenting a child with CF in context of early surveillance.

Therefore, findings from my research support the notion that the early surveillance experience is not an isolated event; that it permeates the lives of parents and their families who experience it. These findings suggest that constructions of early surveillance and meanings attributed to it influence constructions of CF and its management. However, insight from my research also highlights the psychological and social content of parents' experiences that were of equal, if not greater significance to their experience of early surveillance. Importantly, findings from my research provide a particularly strong foundation for the centrality of hope, existential belief systems and personal meaning orientations to the early surveillance experience. Isolating parenting and family processes described in this chapter, and identifying how they might function together to explain, and predict, family outcomes of managing CF is fulfilled in the next chapter by applying the research findings to the The Resiliency Model of Family Stress, Adjustment and Adaptation (McCubbin & McCubbin, 1993).

CHAPTER 6**Parenting a child with CF in the context of early disease surveillance: Adapting to paediatric chronic disease from a theoretical perspective**

This chapter presents further discussion of the research findings, with particular reference to applicable theoretical frameworks through which to view the findings. Psychological factors identified in the preceding chapter that may contribute to adaptive coping within families are applied to the Resiliency Model of Family Stress, Adjustment and Adaptation. How the model is applicable to families whose children with CF undergo early surveillance is speculated, along with how some psychological factors that may contribute to coping can be negatively influenced by the early surveillance experience. By applying the research findings to the Resiliency model, considerations are made for how constructions of experience and attributions of meaning may contribute to coping with lived experiences. Such considerations demonstrate how parents might adapt to parenting children with a chronic condition who undergo early surveillance for covert disease progression.

Resilience is accepting your new reality, even if it's less good than the one you had before.

You can fight it, you can do nothing but scream about what you've lost, or you can accept it and try to put together something that's good...Elizabeth Edwards

6.1 Introduction

As theoretical models are developed from previously tested knowledge and identify a plan for interpretation of findings (Bogdan & Bilken, 2006), they provide a background that supports explanation of phenomena that includes concepts and relationships intended to be interpreted. In this way, theoretical frameworks guide research findings, provide a foundation for their application, and can drive further exploration and analysis. Therefore, theoretical frameworks advance understanding of research findings by being able to apply findings beyond their current context as well as providing an opportunity to support or refute a model's components within the findings' current context.

The Resiliency Model of Family Stress, Adjustment and Adaptation (McCubbin & McCubbin, 1993) provides a structural guide for exploring and identifying relevant variables of interest and their potential associated relationships and outcomes as analysed and interpreted in the previous chapter. As such, concepts within the Resiliency Model may begin to indicate how parental processes can influence, and be influenced by, systemic, family and personal factors that in turn influence parental coping of, and ultimately adaptation to, CF specifically, and paediatric chronic illness more generally.

The current chapter widens the scope of the research findings and their interpretation as they might apply within the family context. In this way, the chapter provides a stepping stone to broader applicability of the research findings, not only to the family context but to the theoretical advancement of the Resiliency Model. Lastly, the chapter argues for the use of the Resiliency Model with families whose children undergo early surveillance for CF lung disease for therapeutic intervention as well as further research.

6.2 Application of Resiliency Model of Family Stress, Adjustment and Adaptation

The Resiliency Model of Family Stress, Adjustment and Adaptation (McCubbin & McCubbin, 1993) attempts to explain why some families recover and are deemed resilient whilst others remain vulnerable and some deteriorate under the same circumstances. Family resiliency can be defined as "positive behavioural patterns and functional competence that individuals and the family unit demonstrate under stressful or adverse circumstances, which determine the family's capacity to recover by maintaining its integrity as a unit whilst ensuring, and where necessary restoring, the well-being of family members and the family unit as a whole (McCubbin, Thompson, & McCubbin, 1996, pp. 5)". Given the observation that families in my research generally appeared to cope

successfully with a psychologically and logistically challenging situation, a strength-based approach to understanding how parents manage their child's CF is appropriate.

As adaptation to a paediatric chronic condition is the most desired outcome for families, the adjustment phase of the model will not be a focus of the current chapter (refer to Figure 1 in Chapter 3 for a diagram of the adjustment phase). As such, the adaptation phase of the Resiliency Model serves as a framework to view how parenting processes and outcomes may contribute to family resiliency in the context of my research. Moreover, the model lends itself to recognising how early surveillance might contribute to those processes identified in the model as important factors for family adaptation and resiliency. By applying my research findings to the Resiliency Model, this chapter presents how processes and outcomes of parenting a child with CF in the context of early surveillance might contribute to adaptation to paediatric chronic disease. See the following page for a diagram of the adaptation phase of The Resiliency Model of Family Stress, Adjustment, which the remainder of the chapter addresses.

The Adaptation phase of the model begins by recognising that an abundance of stressors, strains and transitions impact upon the initial family crisis situation. However, due to the nature of my research, such factors were not identified. It is therefore acknowledged here that parents and their families are likely to have a multitude of historical, economical, developmental and personal difficulties that can influence the way a family initially experiences a crisis situation. Once the family crisis situation has occurred (i.e, for families in my research, it is the diagnosis of CF), parents start to institute new patterns of family functioning. My thesis will apply the research findings to the Resiliency Model from this point on, highlighting factors and relationships the model deems relevant for adaptation to paediatric chronic disease.

Adaption Phase

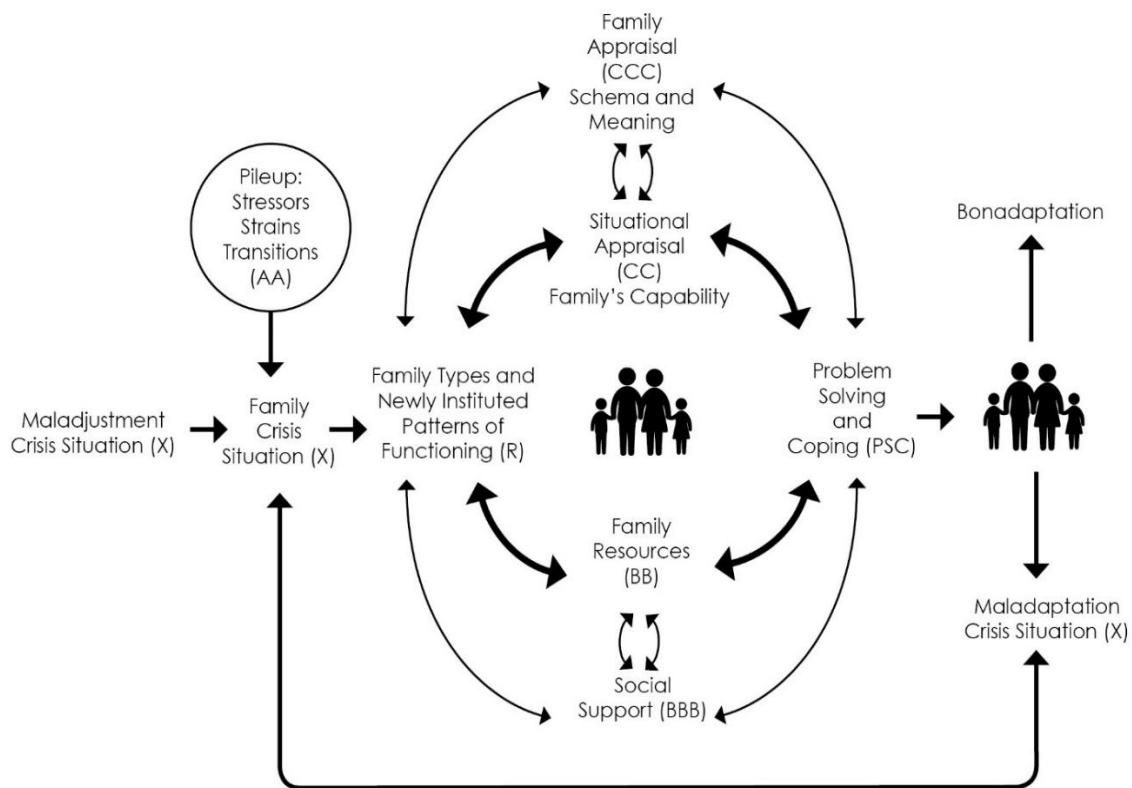
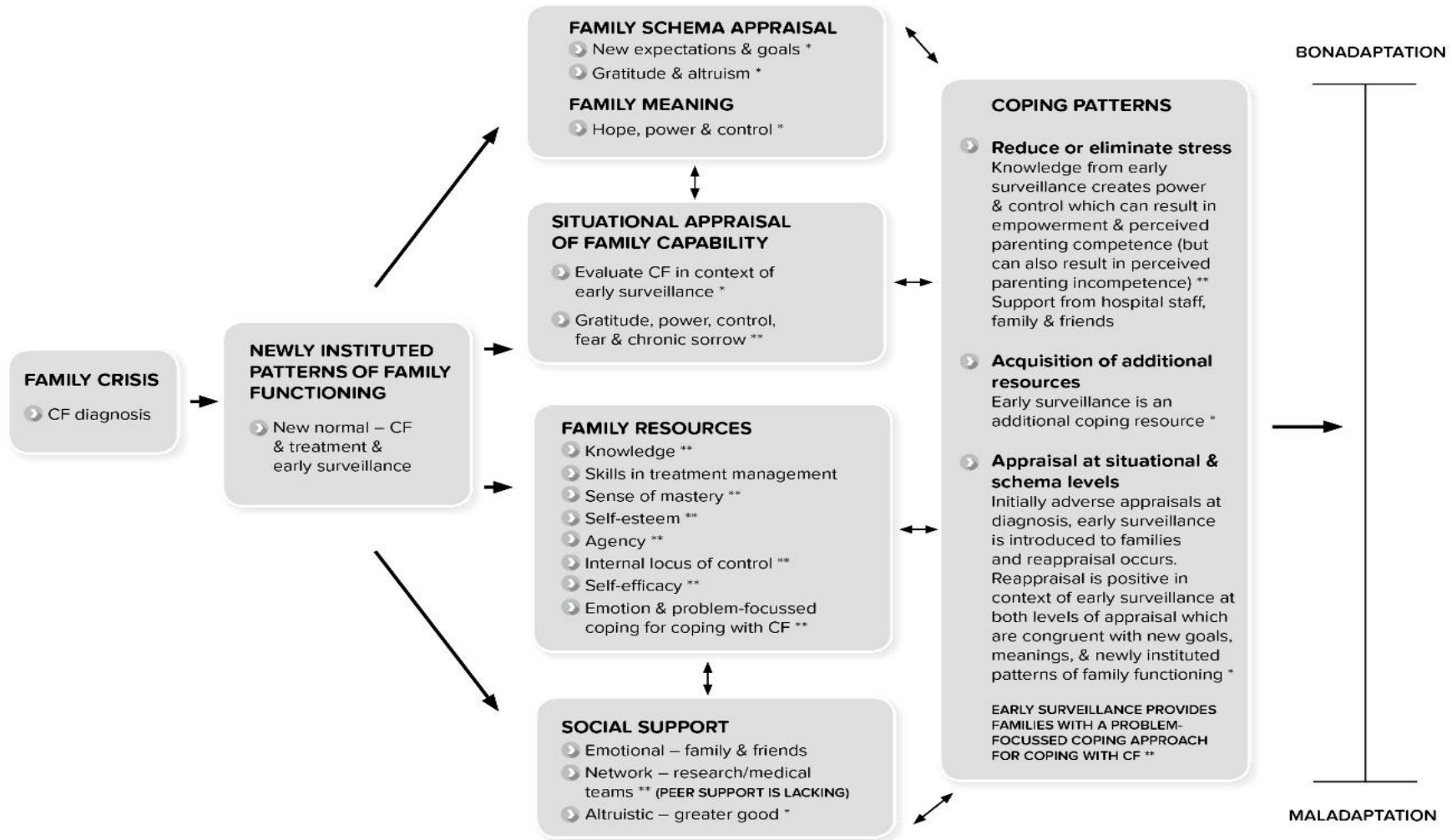


Figure 11. Adaptation phase of The Resiliency Model of Family Stress, Adjustment and Adaptation. Adapted from “Families, Health and Illness: Perspective on Coping and Intervention,” by H. I. McCubbin and M. A. McCubbin, 1993, p. 23. Copyright 1993 by Mosby.

By applying the research findings to the Resiliency Model, considerations can be made for how constructions of experience and attributions of meaning identified in the previous chapter may contribute to coping with lived experiences of parenting children with CF who undergo early surveillance for lung disease. Applying the research findings to the Resiliency Model enables identification of essential interacting components of family life and can explain, as well as predict, family processes and outcomes of managing a paediatric chronic condition. The following page outlines how constructs and processes identified in the previous chapter might function together to explain, and predict, parent and family outcomes of managing a paediatric chronic condition, specifically in the context of early surveillance for CF lung disease. The remainder of the chapter will detail the diagram’s constructs and relationships for adapting to CF in the context of early surveillance from the point of establishing family patterns following diagnosis (i.e., the crisis situation).



* Early surveillance participation positively influences construct

** Early surveillance participation can either positively or negatively influence construct, depending on whether the child's surveillance results are un/favourable

Figure 12. Research findings applied to The Resiliency Model of Family Stress, Adjustment and Adaptation

6.2.1 Newly Instituted Patterns of Functioning.

Families have a wide range of established patterns of functioning that offer stability to family life which, according to the model (McCubbin & McCubbin, 1993), may not be adequate to manage illness within the family system. Therefore, newly instituted patterns are needed to keep the family functioning with stability while managing the illness and its related hardships. The major changes in the family patterns of functioning found in my research were; establishing a home regimen for the child's treatment management, and redefining present and future expectations about parenting and family life. Essentially, the process of normalising the condition and its treatment represents a newly established pattern of family functioning that re-establishes family stability and resources, thereby promoting adaptation in families.

Re-establishing family patterns after introduction of CF is not achieved overnight. Trial and error practices and strategies must be negotiated and compromised to fit within the family's already-developed schema (i.e., blueprint) for their family functioning that has evolved over time (McCubbin & McCubbin, 1993). By establishing both practical and existential patterns of functioning through normalisation and redefinition, parents are able to reaffirm their family that constitutes a redefined sense of reality including managing a child's chronic condition. How effective these newly established patterns of family functioning become is determined by the degree to which they are congruent with the family's schema (McCubbin & McCubbin, 1993), discussed shortly.

By assessing changing family patterns, HCPs can determine strategies for intervention by distinguishing adaptive and destructive patterns of functioning that are tailored to address illness-specific and related hardships. Assessing both new and modified patterns of functioning can identify what these families deem important to them, and the strengths and resources they have. Moreover, assessing family functioning when faced with their child's chronic condition can begin to shed light on what meaning families attach to themselves, the illness and their future (McCubbin & McCubbin, 1993).

Normalising CF management in practical terms and normalising both CF and its management in existential terms, influences all other components identified in the Resiliency Model as important factors for adaptation. Therefore, these patterns of family functioning contribute to what resources a family has and chooses to draw upon. For example, maintaining a home treatment regimen enables parents to develop a high sense of self-efficacy for keeping their children healthy. Self-efficacy is a family resource that is influenced by newly instituted patterns of family functioning. At the same time, self-efficacy influences a family's coping patterns. Similarly, established patterns of family functioning contribute to the level of social support families draw upon and accept as part of their adaptation. For example, successful normalisation and acceptance of CF into family life is likely

to enable parents to draw upon social support that further facilitates, as well as validates, their adaptation strategies and outcomes.

A family's situational and family schema appraisals are also influenced by what patterns of family functioning parents establish as part of their aim to achieve adaptation to their child's chronic condition. Moreover, early surveillance becomes part of the newly established patterns of functioning. As such, early surveillance becomes part of the fabric for how a family will adapt to their child's CF. Accordingly, early surveillance contributes to development, establishment and maintenance of the remaining components identified within the Resiliency Model as important factors for adaptation.

6.2.2 Situational and family appraisal.

The Resiliency Model (McCubbin & McCubbin, 1993) emphasises two levels of family appraisal that play a role in a family's ability to manage a crisis situation, which shapes the course of family adaptation. Firstly, situational appraisal of the family's capability is the critical relationship between the illness-related demands on the family and their capabilities and strengths to manage these demands. The family's situational appraisal would reveal their in/adequacy to manage the situation and serve as the basis for additional coping or changes in family patterns of functioning. Why the family's situational appraisal becomes crucial in shaping the family's response to illness is that through the process of situational appraisal (including evaluation of the illness, assessment of family capabilities and strengths, and evaluation of alternative courses of action and coping strategies), the family ultimately comes together as a unit to cope with the illness.

6.2.2.1 Situational appraisal.

By applying my research findings to the Resiliency Model, parents appraise their family's situation by evaluating CF in the context of early surveillance. The inherent link between the two brings a range of emotions for parents including fear and chronic sorrow. However, parents were also able to experience altruism and gratitude and perceived control and power that came from early surveillance participation. Other parents of children with CF have also reported both positive and negative appraisals of their child's condition (Baldaccino, et al., 2012), however their appraisals were not in the context of early surveillance. It appears that for parents in my research, early surveillance outcomes seemed to off-set both the early surveillance experience and negative responses to having a child with CF.

Cystic fibrosis and early surveillance certainly place significant burden on parents, their children and their families. They both demand psychological and practical attention from parents that can impact upon families. However, CF within the context of early surveillance provides parents with strength that seemed to transcend CF and early surveillance. For example, early surveillance

was appraised as a family resource in terms of the knowledge acquired on an annual basis about a child's health. Parents were able to use both their child's early surveillance results and the early surveillance principles as sources of family strength. That is, knowing of their child's health and knowing they are contributing to something greater were sources of strength for parents. Therefore, whilst parents are placed under additional burden of early surveillance, the transcendent strength provides a perception of adequacy in managing and coping, both with CF and early surveillance. Social comparison appraisals at this level that function as sources of family strength for coping with adversity are discussed in the section: family resources – strengths and coping strategies as family capability.

6.2.2.2 Family appraisal.

According to the Resiliency Model, the other level of appraisal that contributes to a family's ability to manage a crisis situation is family appraisal, consisting of schema and meaning. When faced with an illness-induced family crisis that demands changes in organisation and patterns of family functioning, the family is required to appraise its past and its future to give meaning to the illness and to the resulting changes within the family system. This meaning can facilitate adaptation (McCubbin & McCubbin, 1993).

A family's schema consists of value attached to shared or accepted goals, priorities and expectations that have been developed and shaped over time. The family schema, unlike other components in the Resiliency Model, is relatively stable and it is used as a major reference point against which situational appraisals are contrasted and shaped. The family schema is more stable and enduring than situational appraisals because it captures the family's values and internal sense of identity and goals.

As part of establishing and implementing new patterns of functioning, the existential (as opposed to practical) normalisation of CF into the family created changes in family schema. Appraisal at this level included assessing and redefining goals, priorities and expectations that were congruent with introduction of CF into the family system to give meaning to the illness and to the changes made. Family schema appraisal is posited as the most stable component in the model (McCubbin & McCubbin, 1993). Changes at this level contributed to disintegration of previously-established family goals, priorities and expectations, which initially created instability and uncertainty about the family's future goals and expectations.

The situational appraisal of a child's condition within the context of its unique management gave parents a new reference point to re-establish goals, priorities and expectations that matched their newly-established patterns of family functioning. That is, early surveillance as a family source of strength functioned as a framework to redefine goals, priorities and expectations associated with

their child's CF. Being part of a unique treatment program, and gaining information about a child's health, enabled parents to give meaning to their required family changes because of their involvement in the early surveillance program. Consequently, family schema was redefined within the context of early surveillance to give meaning to CF and to the changes made. Whilst fostering family coping and legitimising changes in family functioning, the family's schema has as one of its central functions the development of family meanings (McCubbin, Thompson, & McCubbin, 1996).

6.2.3 Meaning-making.

Due to required changes to family goals, priorities and expectations, as well as changes to the family's pattern of functioning due to a child's chronic condition, the family's schema appraisal plays a central role in how families give meaning to the illness and the family's changes. This level of appraisal functions to develop congruency between changes in the family's schema and their instituted patterns of family functioning. Developing a sense of family meaning to changes created by a family crisis is a difficult and demanding process, achieved only through perseverance, patience, negotiation and shared commitment. Therefore, meaning-making takes on critical, and some would argue central, importance in how families cope with their child's chronic condition, ultimately contributing to family adaptation.

Parents take on new roles as primary carers of the sick child and develop new patterns on family functioning around CF management. At the time of diagnosis, parent struggle to make sense of, and give meaning to, their child's condition because the family's original family schema is threatened. Therefore, they battle to give meaning, and thus legitimacy, to the new parental roles and family patterns. Over time, and with input from early surveillance, parents are able to introduce new meaning to the changes they initially struggled with.

Parents were able to experience perceptions of power and control over their child's condition because of their participation in early surveillance through knowledge and ability to act on findings. Perceived power and control facilitated a sense of gratitude for the opportunity to participate in early surveillance. Perceived control and power over disease progression, along with a sense of gratitude for early surveillance, combined to give parents the meaning of hope; hope for their child, hope for their family, hope for the future. These new meanings, which do more than explain the illness, appeared to bring legitimacy and value to the family changes and seemed to give parents a sense of purpose and meaning beyond the illness itself. With this, the overall changes in the family unit gain both meaning and acceptance, and seemed to be the basis of the family's emerging sense of calm and commitment.

As family meaning fosters congruency between newly instituted patterns of functioning and family schema appraisal, it appeared that hope from early surveillance facilitated congruency

between normalisation of CF and its treatment (inclusive of early surveillance practices) and parents' redefined goals, priorities and expectations for their family. Hope is a mediator between a family's actions and beliefs: families act in a certain way because of the belief it will benefit their child – and that belief is facilitated by what they are told the early surveillance program will do for their child. That is, hope mediates the relationship between normalisation of CF and its management in the context of early surveillance and a family's redefined future expectations.

Newly instituted patterns of family functioning are the most predictive component within the Resiliency Model of un/successful coping because they are foundational to all other processes that follow (McCubbin & McCubbin, 1993). Family meaning must match new patterns of family functioning for adaptation to be likely. Parents in my research were able to create congruency between changes in family patterns to accommodate CF in their daily lives, and the meaning attributed to those changes. Participating in early surveillance for CF lung disease (i.e., changes in family patterns) empowered parents through information acquisition and the ability to act on the findings, which seemed to function as a mechanism for congruency between changes in family behaviour arising from CF diagnosis and the meaning created from those changes. Parents attributed hope to their child's' future from empowerment afforded by early surveillance. Whilst the Resiliency Model posits the reciprocal nature of newly instituted patterns of functioning, family schema and situational appraisal, and family meaning-making in adapting to chronic illness within the family, my research findings have shown how these elements of the Resiliency Model might function to facilitate coping with, and adaptation to, CF in the context of early surveillance.

6.2.4 Family resources – Strengths and coping strategies as family capability.

McCubbin and McCubbin (1993) define capability as the potential a family has for “meeting all of the demands it faces” (pp. 45). They emphasise two types of capabilities; resources that a family has, and coping behaviours and strategies, which is what a family does as individual members and as a unit. Whilst the model identifies individual, family and community resources, my thesis will focus on individual resources from the perspective of the parent. Adaptive resources are characteristics that facilitate adaptation (McCubbin & McCubbin, 1993). A number of resources highlighted in the family stress literature as critical for meeting demands during illness within a family (Clark, Gong & Kaciroti, 2014; Fink, 1995; Knafel, et al., 2013; Tak & McCubbin, 2002; Wallander, Varni, Babani, Banis & Wilcox, 1989) have been found in my research. Firstly, acquiring knowledge and skills through education, training and experience means that family members can perform tasks with greater efficiency and ease. Knowledge also helps parents to understand why tasks associated with illness management are necessary. Acquiring knowledge itself about their

child's condition is another component of this family resource because knowledge in this instance is seen by parents as a form of power.

From a philosophical perspective, knowledge is an epistemological matter. In the current context, knowledge creates a sense of control over CF – it appears that knowledge is power. Foucault's (1980) popular theorising of the complex relations between knowledge and power provide a structural approach to understanding how the mechanisms of knowledge and power can create a sense of control. He asserted that; power is not a thing but a relation with knowledge, power increases capacity to control, and control from power is not simply repressive but it is productive. Theory and research both attest to a positive association between knowledge thorough education and a sense of control (Ross & Sastry, 1999; Pallas; 2000; Schieman & Plickert, 2008). Notwithstanding the psychological adversity of gaining knowledge about a child's disease progression, parents are able to subjugate their child's condition that has been portrayed to them as an uncontrollable entity. Moreover, a sense of control shares conceptual ground with self-efficacy and internal locus of control (Rotter, 1966), which are salient attributes for parents in my research. Therefore, belief in the ability to conduct home treatment (i.e., self-efficacy); accountability for child health (i.e., internal locus of control) and a sense of control over disease progression combine to create this sense of power over their child's condition, which, according to the Resiliency Model, acts as a resource that positively contributes to a family's coping patterns.

Secondly, a sense of mastery is an important adaptive resource for coping with paediatric illness; it is the belief that the actions one performs will contribute to control over the circumstances in one's life (Reber & Reber, 2001). Based on definitions in the paediatric medical literature, my thesis uses this term to refer to parents' perceived competency and subsequent confidence in their ability to achieve the required home treatment regimen. Achieving the home treatment regimen gives parents a sense of mastery, which supports parental self-esteem. However, in context of my research, sense of mastery can be supported or disrupted by early surveillance results (i.e., un/favorable review results), which in turn can affect parental self-esteem.

Lastly, self-esteem is an adaptive resource (McCubbin & McCubbin, 1993). It is a positive judgement about one's self-worth (Reber & Reber, 2001). For parents in my research, self-esteem emanated from their competency and confidence in being able to conduct home treatment adequately. Therefore, sense of mastery is influenced by how well parents can establish and maintain CF management. Moreover, early surveillance results are constructed by parents as an indication of how well they have been conducting home treatment, which therefore, means that early surveillance results can either support or disrupt development and preservation of self-esteem.

Sense of mastery and self-esteem have been highlighted by many researchers of family stress as critical factors in the family stress process because they are essential for effectively managing demands (Folkman, Lazarus, Gruen & DeLongis, 1986; Hobfoll & Spielberger, 1992; Raina, et al., 2005). Therefore, an important mechanism that may link stress to negative family outcomes is the weakening of self-esteem due to failure of mastery (Cantwell, Muldoon & Gallagher, 2014; Pearlin, Menaghan, Morton & Mullen, 1981; Silver, Bauman & Ireys, 1995). Perceived mastery of CF management is affected by early surveillance because unfavourable review results can undermine parents' perceptions of their ability to conduct home treatment, resulting in adverse effects on their parenting self-esteem.

Accordingly, the family coping resources (as well as constructed relationships between them) of acquiring knowledge and skills, sense of mastery, and self-esteem can be influenced both by a family's capability for CF management and by early surveillance. Therefore, acquiring knowledge from early surveillance about a child's condition is an adaptive mechanism for coping with CF. However, depending on what the early surveillance results yield for parents, sense of mastery and self-esteem can either function as coping mechanisms for coping with CF or they can hinder parents' ability to cope effectively.

Coping behaviours and strategies are identified by the Resiliency Model as what a family does as individual family members and as a system to meet the demands it faces (McCubbin & McCubbin, 1993). This component of the Resiliency Model relies on Lazarus' and Folkman's (1984) definition of coping to explain that coping is viewed as a process that involves continuous adjustments based on appraisals between the person and the situation. As my research focussed on parental coping strategies as conceptualised within Lazarus' and Folkman's framework, this section speculates their contribution to family coping patterns and categories for adaptation to managing CF, and how they are potentially influenced by early surveillance.

Supported by my research findings, parents of children with CF (Tack, et al., 2014; Priddis, et al., 2010; Zubrycka, 2015), and other chronic conditions (Coffey, 2006; Greening & Stoppelbein, 2007; Tong, et al., 2010), primarily use emotion-focussed coping strategies for coping with knowledge of their child's condition. That is, emotion-regulation became a source of coping when it was understood (i.e., appraised) that CF (i.e., the stressor) could not be removed (Lazarus & Folkman, 1984). Applied to parents in my research, being able to down-regulate psychological burden through venting, social comparison, and staying in the present functioned to minimise personal adversity that helped parents to manage knowing their child has CF.

Downward social comparisons were a prevalent coping strategy parent employed to cope with their child having CF, but also to cope with their child undergoing early surveillance for lung

disease. Parents not only compared their child (and their family) to those who were worse off, but they also compared their own families to families who were not given the opportunity to track their child's health as sensitively as what AREST CF offers. Such appraisals enabled parents to feel a sense of gratitude for their given situation within the context of wider society, and to cope with their sense of fear about uncertainty in their child's morbidity and mortality. Situational appraisals that involved downward social comparisons functioned as sources of family strength in their capacity to cope with the demands they faced.

As resources within a family system, individual coping strategies can influence how the family functions, both in relation to the child's illness, but also more broadly as a synergistic unit that works together to harmonise and cultivate itself. The Resiliency Model emphasises how coping strategies and behaviours of individual members have the capacity to influence family resources of cohesion, organisation, communication, hardiness, integration and stability. These factors represent family functioning, essential for the family system's ability to thrive and for their well-being (McCubbin & McCubbin, 1993). Moreover, balanced family coping (consisting of integration and personal growth) has been reported as a predictor of positive health trends in children with CF (Patterson, et al., 1993). From the point of view of the Resiliency Model, findings from my research and Patterson et al.'s findings indicate how a family might adapt to their child's CF, which can influence a child's long-term health outcomes.

6.2.5 Social support.

Support from individuals, such as family members and friends, and institutions, such as medical and community services, can help the way a family copes with an illness stressor (McCubbin & McCubbin, 1993). The study of social support has received the most attention in literature on stress associated with family adaptation (Crnic, Greenberg, Ragozin, Robinson & Basham, 1983; Kazak & Marvin, 1984; Paynter, et al., 2013; Tak & McCubbin, 2002). It has most often been viewed as a primary buffer or mediator between stress and health breakdown (Horton & Wallander, 2001; Kazak, 1997). The authors of the Resiliency Model use Cobb's (1976) early definition of social support who defined it as information exchange at the interpersonal level that provides; emotional support (showing individual family members they are loved and cared for); and network support (showing family members they belong to a network of communication involving mutual support and understanding). McCubbin and McCubbin (1993) added another form of support to their model that they felt contribute to a family's capacity to adapt to their child's chronic condition; altruistic support, which is information given in the form of goodwill from others for having given something of oneself.

6.2.5.1 Emotional support.

The primary form of support for parents in my research was emotional support, which appeared to buffer or mediate fear parents experience around the time of their child's annual review and for fear associated with uncertainty of disease progression in their children. As shown in the previous chapter, emotional support from family and friends helped to regulate personal burdens, which in turn helped parents to cope with knowledge of their child's chronic condition. A question to ask is – are parents whose children undergo early surveillance of CF lung disease in need of emotional support specifically for their early surveillance experiences? I believe the answer to this question is *yes* because of fear associated both with medical procedures and with knowledge of covert disease progression. It is beneficial to give parents such information about their children but as HCPs, we need to offer relevant support to manage what it is presented to them. Moreover, the avoidant coping strategies parents use around the time of annual review may induce depression, anxiety and/or stress (Sheehan, et al., 2014).

My research findings lay a foundation for understanding specific support that parents need, and can help to identify appropriate interventions and support strategies. By applying my research findings to the Resiliency Model, emotional support from family and friends is an important resource that parents could draw upon to help institute new patterns of family functioning (showing the bidirectional nature of processes within the Resiliency Model). Additionally, emotional support may buffer against adverse outcomes from a child's early surveillance procedures by positively influencing coping patterns around the time of a child's annual review.

6.2.5.2 Network support.

Since the discovery that infections associated with CF can be transmitted between patients, network support has been difficult to offer parents. Changes in clinical practice and community support have meant that parents and their families are segregated from each other to avoid cross-infection of children with CF (Bell & Robinson, 2008). There are community-based support programs offered by Cystic Fibrosis Western Australia that take segregation into consideration (e.g., parent weekends, dinners and coffee mornings in suburbs and regional areas where only one adult or child with CF is allowed to attend), and with the introduction of the Internet, there are online support networks that give parents and their families network-support (e.g., CF Smart and CFMatters Facebook pages which are both supported by Cystic Fibrosis Western Australia). Additionally, network support is available to parents from medical and research staff, giving them a source of coping beyond their immediate networks. However, for parents in my research network-support from peers, specifically around and regarding early surveillance, is currently lacking. As an important component for adaptation to chronic illness emphasised by the Resiliency Model, such network-

support could enhance parents' capacity to adapt to their child's CF in the context of early surveillance. Moreover, parents in my research highlighted how establishing and maintaining relationships and networks were important for them to manage their early surveillance experiences. To facilitate such network support, a formal online support network for AREST CF families where they could share their stories, ask questions and give advice to each other could form network-support aimed at developing mutual support and understanding. Mentor training could be offered to a number of consumer advocates who could moderate the online network so they can be formally supported throughout the process.

6.2.5.3 Altruistic support.

Altruistic support has unique beneficial elements in the context of my research. Parents have a sense of altruistic support from knowing they are giving something of themselves and their child to advancing scientific knowledge about CF to hopefully help future generations of children born with CF. Whilst the experience of early surveillance is undoubtedly unpleasant, parents are uniquely supported by the knowledge of their altruistic principles. According to the Resiliency Model, giving to something that is deemed bigger than oneself can help families to cope with their child's chronic condition, and can be a source of family strength that also helps parents to cope. Therefore, it altruistic support may exist for families coping with CF in the context of early surveillance.

6.2.6 Coping patterns.

The Resiliency Model indicates relationships exist between family resources and coping strategies, social support, situational and family-schema appraisal and meaning-making with generalised coping patterns. The Resiliency Model (McCubbin & McCubbin, 1993) characterises the family system as a resource-exchange network, whereby coping occurs within the system and as part of the system. In this way, coping behaviours are the actions for this exchange of resources, resulting in coping patterns that either facilitate or impede family adaptation to paediatric chronic illness.

Whereas strategies are defined in the Resiliency Model as intentional efforts to reduce or manage specific illness-related demands, they are grouped into patterns that represent more generalised ways of responding that transcend different kinds of stressful situations. The authors of the Resiliency Model identified four broad patterns of coping that facilitate adaptation to illness, which are inherently either emotion or problem-focussed coping approaches; efforts to reduce or eliminate stresses and associated hardships, acquisition of additional resources to manage illness and associated hardships, management of family system tension, and shaping appraisal at both the situational and schema levels. How these coping patterns are influenced by family resources, social support, situational and schema appraisal and family meaning-making (as indicated by the Resiliency

Model) will be outlined. Further, how coping patterns are influenced by early surveillance through its contribution to family resources, situational and schema appraisal and family meaning-making will be outlined.

6.2.6.1 Family resources contribute to family coping patterns.

As a family resource for coping with paediatric chronic disease, gaining the most detailed information available in the world about a child's condition gives parents a level of knowledge about their child's health that few parents in the world receive. Parents perceive they are gaining as much information as is conceivably possible about their child's disease progression. Whilst information-seeking about the disease itself is a coping strategy, the sensitivity and uniqueness of early surveillance results appears to give parents an additional element to this particular coping strategy. Parents perceive a sense of control and empowerment in managing their child's disease, which, according to the Resiliency Model, can promote coping with CF more generally.

Additionally, gaining a sense of parenting competence and parental self-esteem by mastering a child's home treatment regimen can facilitate coping. Early surveillance results can confirm these parental constructs when favourable annual review results are received, which serves to validate parents' treatment efforts throughout the year. The sense of empowerment, control, competence and self-esteem are over-arching factors for parents in my research. However, unfavourable annual review results can undermine these positive experiences.

Whilst parents believe that early surveillance is in the best interests of the child, it can have short-term, adverse psychological effects on parents. Whilst parents are willing to tolerate these for the sake of long-term benefits on child health, there is the possibility that these adverse effects may be experienced on a longer-term basis not be discernable through my research. A longitudinal study of how individual resources identified in my research are influenced by early surveillance results annually could be of benefit to understanding any long-term effects.

Lastly, social support is posited in the Resiliency Model to have both a direct effect on coping patterns, as well as an indirect effect by acting as a family resource. Emotional support and altruistic support are well-provided for parents in my research, thereby positively contributing to family resources and coping. However, lack of network support could be detrimental to a family's capacity to cope with CF and/or early surveillance, which in turn contribute to adaptation to CF.

Social support has been identified in the literature as a family factor that fosters resiliency (Benzies & Mychasiuk, 2009). Whether it is instrumental, emotional or practical, support is vital to families during times of stress and helps maintain good physical and mental health (Black & Ford-Gilboe, 2004; Walsh, 2003). Therefore, the levels of support identified in the Resiliency Model, and

how well they are fulfilled, can influence family coping patterns by providing parents with, or preventing them from accessing, extraneous sources for individual and family coping.

6.2.6.2 Situational and family schema appraisals and meaning-making contribute to family coping patterns.

According to the Resiliency Model (McCubbin & McCubbin, 1993), how a family shapes both their situational and family schema appraisals will contribute to how a family copes with their child's condition. Cognitive appraisals play a central role in how one will respond to a stressor, and therefore what coping mechanisms are activated (Lazarus & Folkman, 1984). This process is embedded in the Resiliency Model as an outcome for how families adapt to their child's chronic condition. In their primary appraisal, parents evaluated their child's illness as threatening and harmful. When the condition was considered in this way, negative outcomes were found. However, social comparisons and early surveillance functioned to change parents' perceptions of their child's CF from a hopeless and uncontrollable situation to one of hope and control, which indicates the relationship between individual persons and their specific environment (Lazarus & Folkman, 1984). Consequently, this change appeared to lead to a positive re-evaluation of the child's condition, which may lead to subsequent changes in ways of coping. For example, appraisals of a situation as hopeful and controllable may activate the coping mechanism of sense of mastery for parents, which is a critical factor in a family's ability to cope with a crisis situation (McCubbin & McCubbin, 1993).

In this way, the situational and family appraisals of CF in the context of early surveillance, the resultant creation of hope for their child's and family's future, and activated coping strategies based on situational and family appraisals could operate synergistically to reduce the burden of CF on families. For example, social comparison is an appraisal that functions as a coping strategy contributing to a family's capability to meet the demands it faces. Not only does the appraisal appear to reduce the psychological burden associated with parenting a child with CF, but the appraisal appears to activate adaptive coping strategies, further enabling the family to cope with burden associated with their child's CF. Moreover, the early surveillance results themselves (and subsequent treatment) are a mechanism for eliminating, or at least reducing, the perceived severity of CF. Therefore, these outcomes represent a problem-focussed coping approach for families managing CF in the context of early surveillance.

Problem-focussed coping aims to eliminate or reduce the source of stress in practical ways, therefore directly reducing stress (Lazarus & Folkman, 1984). Literature shows that problem-focussed coping in an array of circumstances is an effective method of decreasing burden associated with a stressor (Billings & Moos, 1981; Chao, 2012; Moos, Brennan, Fondacaro & Moos, 1990; Nes & Segerstrom, 2006). This approach has both behavioural and cognitive elements for dealing directly

with the stressor, therefore changing the perception of, and relationship with, the stressor. The aim of AREST CF is to delay or prevent disease progression. By engaging in the AREST CF early surveillance program, parents are adopting a more problem based coping approach. Parents primarily utilise emotion-focussed coping strategies for coping with the stress of early surveillance procedures and processes. Early surveillance provides parents with a practical, problem-focussed approach to manage stress caused by CF, directly reducing the stress by changing the perception of, and relationship with, the stressor.

Entering a child into the early surveillance program grants parents a sense of control over CF. Early surveillance participation changes parents' perceptions of CF through the early, proactive management of the disease afforded by early detection. Parents felt as though they had taken as much control as they possibly could over CF by entering their child into early surveillance; which parents attribute to the uniqueness of this type of CF management. Access to the information from early surveillance allows parents to cope more generally with CF. Therefore, a sense of mastery and acquiring knowledge and skills makes parents feel as though they are taking control of the situation that has been presented to them – affording them a problem-focussed coping mechanism to coping with CF. By feeling a sense of control and a sense of mastery, parents' self-esteem is supported. Therefore, the empowerment and perceived control (i.e., taking control) parents experience from acquiring knowledge (i.e., information-seeking) about their child's condition, as well as from their understanding of sensitivity and uniqueness of early surveillance, promotes coping with CF more generally. It is conceivable that parents whose children undergo early surveillance may be less daunted by, more optimistic about, and perceived to be more in control of their child's CF than parents whose children do not undergo early surveillance, which may have implications for parents' attitudes and values around treatment adherence, as well parental identity and parental mental health and quality of life more generally.

Lastly, the constructs of situational and family schema appraisals and meaning-making emphasised in the Resiliency Model (McCubbin & McCubbin, 1993) interact directly with what is labelled as situation and family schema appraisal coping. That is, what a family values and prioritises (i.e., family schema appraisal), and what they think of their situation (i.e., situational appraisal) and the meaning they attach to it (i.e., meaning-making), can be a generalised coping pattern for adapting to a child's chronic condition. Gratitude, perceived control and power from situational appraisal, along with the meaning of hope created from both levels of appraisal, emerged as coping mechanisms through which parents were able to feel comfort and content with their child's CF in the context of early surveillance; maybe *because of* the context of early surveillance.

With appraisals identified at the individual level, and their influence on coping discerned, how these appraisals might function synergistically or antagonistically to produce family adaptation could be explored in the future. As the Resiliency Model emphasises family-level appraisal as a determining factor of whether other illness dimensions become problematic, identifying individual-level factors is an important part of creating a comprehensive understanding of how families adapt to paediatric chronic illness. With little research conducted on the family-level appraisal component of the Resiliency Model, the framework could be used to distinguish family appraisals that represent the unique experience of early surveillance and how those factors might facilitate or impede family adaptation to CF. How the unique experience of early surveillance might compare to other families experiencing paediatric chronic illness can also be explored to determine commonalities and points of difference that can be viewed in the broader context of paediatric chronic conditions.

6.2.7 The family adaptation process: Bonadaptation and maladaptation.

The final outcome within the Resiliency Model is family adaptation, where families engage in direct response to the excessive demands of an illness and depleted resources, and realise that systemic changes are required to restore functional stability and improve family satisfaction within their given situation (McCubbin & McCubbin, 1993). The Resiliency Model highlights how coping strategies play a critical role in adaptation, which is the point where my thesis ends and future research must take over. My thesis has highlighted what parents must cope with in relation to their child's CF in the context of early surveillance. My thesis has further highlighted numerous ways that parents strive to cope with their child's CF, and particularly in the context of early surveillance, and how early surveillance can facilitate or impede coping, and how engagement in surveillance itself, can be used as a coping mechanism.

6.3 Summary and conclusion

Using the Resiliency Model as a guiding framework, this chapter has shown how families might adapt to paediatric chronic illness through a series of inter-related processes and resources. The first change required for family adaptation is that newly instituted patterns of family functioning must be engaged. Parents in my research described how they normalised CF and its treatment and redefined future expectations, which helped them to form new patterns of practical and existential family functioning.

Appraisals at both levels and meaning-making were positively influenced by participating in early surveillance, whereas early surveillance participation could have either positive or negative effects on family resources such as sense of mastery and self-esteem, depending on what annual review surveillance results yielded. Therefore, the Resiliency Model aptly highlights how parents'

adaptation to their child's CF can be influenced by early surveillance, and can therefore be used as a guiding framework to develop and implement tailored interventions.

For example, application of the Resiliency Model to my research findings highlights that parents generally normalise CF into the family and establish patterns of family functioning that incorporate the treatment regimen. Additionally, parents develop expectations that are congruent with their newly instituted patterns of family functioning because of the hope they attribute to their family meaning. Applied to the Resiliency Model, my findings highlight that hope for a child's future establishes a link between newly instituted patterns of family functioning and their expectations of those new patterns. The combination of compatible patterns (or actions) and expectations (or thoughts) contribute to effective family coping with paediatric chronic illness. The process from establishing patterns of family functioning to effective family coping demonstrates how the Resiliency Model can be used in clinical practice as a framework to assess how families might adapt to paediatric chronic disease in the context of early surveillance for disease progression. Due to the uniqueness of early surveillance for CF lung disease, how a program such as that run by AREST CF functions to facilitate coping has not previously been reported. Indeed, it would seem that early surveillance *is* a primary mechanism for coping with CF.

Subsequently, it is possible that parents whose children undergo early surveillance for CF lung disease have more coping mechanisms than parents whose children do not undergo early surveillance for CF lung disease. Therefore, parents in my research may be more resilient for future burden of CF because of the additional coping mechanisms parents construct from their early surveillance involvement. However, the possibility that these parents are more vulnerable to future burden of CF must be considered because of the additional stressors of early surveillance, and their effects on parents, identified as part of my research. Moreover, problem-focussed coping is generally more effective than emotion-focussed coping, which may mean that parents cope effectively with their child's CF but not so well with their child's early surveillance.

In conclusion, the Resiliency Model attempts to explain how families adapt to paediatric chronic illness by isolating individual, family and community properties and processes that interact and shape the course of family behaviour over time. My thesis has been able to isolate individual factors from parents' perspective that are posited within the Resiliency Model to contribute to family adaptation to paediatric chronic illness. Additionally, considerations for how constructions of experience and attributions of meaning may contribute to coping with lived experiences were made. My thesis has demonstrated use of the Resiliency Model as a framework that would assist in determining how families might respond to CF within the context of early surveillance, which may lead to family resiliency and adaptation. The model presented within my thesis characterises how

families might respond to CF within the context of early surveillance so that targeted interventions can be tailored to family needs that build individual and family strength. The model can therefore be aptly used as a framework to assess individual and family-level factors that may function as predictors of family adaptation in the context of early surveillance for CF lung disease. The factors identified in my research that are outlined in the model presented in the thesis can be tested longitudinally through mixed-effects logistical regression analyses to determine indicators of health vulnerability and resilience. Subsequently, intervention and support can target specific individual and family-level factors that may reduce risk of maladaptation, as well as increase adaptation and resilience.

This chapter has provided a stepping stone from my research questions about parental perspectives to the more holistic biopsychosocial model of family existence, health and resilience. The Resiliency Model of Stress, Adjustment and Adaptation distinguishes psychological processes, and coping strategies and styles that likely contribute to coping with a paediatric chronic condition. Using this particular theoretical framework to interpret and extrapolate findings has allowed application of my research findings beyond their current context that will hopefully drive further exploration and analyses.

Lastly, the relationships identified in the Resiliency Model are bi-directional, meaning that each component within the model has a reciprocal nature with its associated components. Therefore, adaptation to CF is not static, nor are the elements of family life that contribute to it. As families move through their developmental trajectory and as illness-related crises demand attention, parents need to reconsider and adjust to changes for their families to continue adaptation to a child's progressive condition. The implications of my research findings and their theoretical and practical application to best support parents during diagnosis and early surveillance for CF lung disease are reviewed in further detail in the next chapter.

CHAPTER 7**Summary and conclusions: Key research findings, implications, strengths, limitations and future directions**

In this chapter, key findings are summarised to give an account of, and the reasons for, parents' experiences of parenting children with CF in the context of early surveillance. Findings and interpretations from the data are presented and positioned within the context of the main research aims and questions. The significant aspects relating to parents' experiences of early surveillance are also summarised along with how parents cope, how they construct their experience and what meaning they attribute to early surveillance. Implications of this research for service delivery pertinent to clinical care as well as implications for family theory, policy and practice are discussed. To conclude the chapter, strengths, contributions and limitations of this research are reviewed, avenues for future research outlined, and lastly, post-reflections of conducting the research are described.

*Hope is the thing with feathers, that perches in the soul, and sings the tune without the word,
and never stops at all...Emily Dickinson*

7.1 Key findings that apply to research aims and questions

The primary aim of this research was to explore the lived experience of parenting children with CF in the context of early surveillance. An additional aim was to explore how parents cope with the lived experience of parenting. Moreover, my research aimed to explore how parents construct their experience and attribute meaning to early surveillance for CF lung disease. The aims of the research were achieved using in-depth semi-structured interview data to explore experiences of CF diagnosis, treatment management and early surveillance. Examining parents' experiences, constructions and attributions of meaning in the context of current conceptualisations of adaptation to paediatric chronic conditions also facilitated achievement of the research aims. The exploratory nature of the research was essential given the current focus on early intervention for chronic conditions; as a response to recent national and international clinical and political agendas on early intervention (Jenkins, 2005; Wise, da Silva, Webster & Sanson, 2006), and due to the dearth of research about the lived experience of parenting a child with CF in the context of early surveillance from the perspective of parents themselves. The key findings from this research are summarised in the next section and reviewed in context of the research questions.

7.2 The lived experience of parenting a child with CF in the context of early surveillance:

Implications for policy and practice

Research questions one and two:

1. *What is the lived experience of parenting children with CF in the context of early surveillance?*
2. *What are parents' experiences of early surveillance?*

Essentially, the aim of all research is to contribute to the knowledge base about an area of interest (Mays & Pope, 2000). In my research, parents' lived experiences and personal accounts both informed and extended existing knowledge about the experience of parenting a child with CF. A salient finding was that common experiences and thoughts were shared by parents, regardless of their gender or the age of their child. As such, findings and interpretations relating to research questions one and two (as outlined above) were presented collectively. Where relevant, findings unique to mothers or fathers, or parents of children younger or older than 3 years of age were presented separately. A summary of the key findings relevant to research questions one and two are reviewed in the next section and their contribution to contemporary understandings of parenting a

child with CF and in particular, are discussed in relation to implications they have for future policy and practice.

7.2.1 Policy and practice implications.

The current national and international political and clinical agendas for early intervention (Jenkins, 2005; Wise, da Silva, Webster & Sanson, 2006) prompted my research to explore the lived experience of parenting a child within the context of early surveillance for CF lung disease, and to investigate parents' experiences of the early surveillance process itself. The unique context of early surveillance for CF lung disease gave me the opportunity of exploring the lived experience of parenting children who undergo early and aggressive treatment for CF lung disease.

As previously established, the biomedical view of chronic disease has largely constructed early diagnosis and intervention as a modern conception for prevention of disease progression rather than from a therapeutic perspective. My research found that while personal belief orientations and clinically contextual factors function to advocate early diagnosis and surveillance to parents, it was also evident that psychological and social aspects of the experience were of equal or greater significance to the meaningful understandings parents constructed about their experience of parenting a child within the context of early surveillance. The findings of my research highlight a need to evaluate the way in which diagnosis and early surveillance are constructed and normalised, both in terms of the practical and experiential, so that a holistic view of CF care can be used as a basis to support parents through such a unique process.

By its very nature, normalisation of paediatric chronic illness and its treatment into the family context may buffer and protect its members against its existence as well as against future burdens of the condition. Moreover, though life is disrupted at diagnosis, early diagnosis resulting from NBS allows normalisation to occur very early in the child's life (Cederborg, et al., 2011). Therefore, normalisation of the condition and its treatment, both as a process and an outcome should be an endeavour of the family, clinicians and other HCPs working with these families. The more established and routine a family can become, the more accessible normalisation, and its benefits, will be. The inextricable link identified in my research between CF and early surveillance meant that parents' normalisation of CF was referenced in relation to early surveillance, giving parents an additional reference point that functioned to create hope and control in a situation that was initially constructed as hopeless and uncontrollable. Therefore, understanding how parents appraise circumstances presented to them, and what factors contribute, becomes a critical component of normalisation when evaluating how parents construct their experiences and attribute meanings.

The design of the AREST CF early surveillance program focusses on physical aspects of a child's health and as such, parents' main motivation for becoming involved in the program included to address their child's current health condition, and as a preventative measure to preserve their child's health. Among the features identified as important to parents' experiences included knowledge of a child's current health status as well as any suggested treatment as a result of the surveillance. Therefore, parents' accounts of their child's early surveillance provide support for the perceived importance of early intervention and preventive action against disease progression from both a parental and clinical perspective.

Though loss of connection between parents of children with CF due to recent understandings of cross-infection is substantiated (Bell & Robinson, 2008), parents desire connection with other parents under the same unique circumstance (Duffy, 2011; Eatough, et al., 2013; Kerr, et al., 2004; Shields, et al., 2008). Importance of this aspect of early surveillance does not appear fully appreciated, and therefore alternative considerations have not been fully explored. This particular finding emphasises that connection with other parents and families could be an essential component of parental adaptation to their child's CF within the unique context of early surveillance (Barcroft, 2015). Findings also indicated that parents respond positively to a well-structured and comprehensively yet simplistically explained program with experienced and trustworthy staff that relate and communicate well.

Development of rapport with parents early in the therapeutic relationship is of paramount importance to clinicians because of the ongoing medical care required for CF patients. Emphasising importance of the therapeutic relationship, Swallow & Jacoby (2001), Salgado, et al. (2011) and Gjengedal et al. (2003) reported how this type of relationship provided a consistent source of support for parents. Spiers, et al. (2011) suggested that such relationships are integral for parents' confidence to administer home treatment, and Bull & Grogan (2010) asserted that it was vital that parents felt confident in the health professional they were relinquishing their child to. Given the importance of the therapeutic relationship for CF patients, clinicians and researchers introducing early surveillance need to be considerate of parents' experience during this time. For example, parents were more likely to feel confident about their child's involvement when they felt a sense of competence and trust in the hospital staff.

Notwithstanding these factors, the most salient finding to emerge from parents' experiences of the early surveillance program was that knowledge acquisition about a child's health status was critically important to parents' sustained involvement. In particular, a sense of control over disease progression and sense of hope for the child's future contributed to parents' positive experiences of the early surveillance program. Whilst there are undoubtedly adverse experiences, reactions and

outcomes that require consideration for intervention and support strategies, parents' perceptions of themselves were generally positively influenced by their involvement. This was particularly evidenced by their perceived parenting competence being influenced by the level of self-efficacy for achieving their child's home treatment regimen and orientation towards an internal locus of control for their child's health, which contributed to increased parenting self-esteem. Moreover, feelings of altruism and gratitude for the opportunity to participate in early surveillance positively influenced parents' lives.

However, receiving knowledge about a child's covert disease progression as part of involvement in early surveillance for CF lung disease appears double-edged. That is, parents perceive both control over disease progression and also hope for their child's future, yet they can become acutely attuned to how their child's condition is likely to progress, which can result in fear (Glasscoe & Smith, 2011). Dimensions of fear in the context of early surveillance for CF include fear of losing a child to CF, fear of a child getting sick from disease severity, and fear of knowing a child is getting sick through knowledge of early surveillance results. These dimensions of fear are inextricably linked to, and affected by, each other. For example, fear that a child will die young is fear of disease progression, and so because the child may die of disease progression, the disease, too, becomes an object of fear. Then, because early surveillance provides knowledge of disease progression, which for parents can be viewed as an indication of the child's life expectancy early surveillance, too, becomes an object of fear.

Parents want knowledge of their child's often covert disease progression so, if present, it can be treated as soon as possible; but they are also fearful of it because of what it means for their child and their family. Therefore, it appears that parents simultaneously fear and desire knowledge of their child's covert disease progression. The early surveillance program provides parents with detailed information about their child's disease progression, but parents have not been provided with tools for how to manage such information. Furthermore, is the issue of perceived accountability for child health by parents as primary carers and receiving detailed information about their child's disease progression. Given the findings of my research, there is an opportunity to support parents through such experiences by connecting parents with other families in the AREST CF program so personal experiences can be shared in such a way that mutual understandings are created. Such support could enable parents to develop realistic expectations about the early surveillance experience, as well as about their parental role in the care for their child.

The adverse experiences, reactions and outcomes that occurred of parents need to be considered as likely influencing parental mental health. Among the challenges experienced by parents were; feelings of self-judgement and blame if their child's annual review results showed

some disease progression; relinquishment of their child for medical procedures, and cognitive dissonance around the decision to enrol their child in medical research. Becoming familiar with the processes and procedures, and with the research and medical staff appeared to mitigate adverse responses. These findings bring attention to the issues that parents encounter and that hospital staff must attend to so that parents can be better supported in their quest to keep their children healthy. A reciprocal opportunity exists for future research to consult with medical and research staff about their experience of running such a program, as well as their experiences of parents and children attending. This type of research may identify where staff perceptions are the same and different to parents, as well as any assumptions or anxieties for staff.

Moreover, my research has identified a need to assess whether parental anxiety is prevalent among parents of children undergoing early surveillance of this nature in CF, and determine what resiliency techniques might mitigate it, if it indeed needs addressing. Based on my findings and reports in the paediatric medical literature (Bearden, et al., 2012; Caes, et al., 2014; Harper, et al., 2013; Karlsson, Englund, Enskär & Rydström 2014; Kwan, Chiu, Gan & Chan 2016; Scholten, 2013), intervention for fear and anxiety, lack of control and helplessness, cognitive dissonance, or damage to parent identity may be necessary to support parents throughout their child's annual early surveillance procedures. For example, narrative psychotherapy is a forum where parents could express their fears and anxieties in a safe and supportive environment, where concerns can be validated and, at the same time, demystified and moderated. As an in-the-moment strategy, parents could use mindfulness techniques to help regulate either anxiety associated with relinquishment or cognitive dissonance during their child's medical procedures. Mindfulness strategies have demonstrated ability to reduce stress (Brown, Warren & Richard, 2003) and anxiety (Evans, et al., 2008; Hofmann, Sawyer, Witt & Oh, 2010), and could replace the distraction technique of coping with relinquishing a child for medical procedures. Mindfulness-based education could form part of the narrative psychotherapy. The social work or psychological annual assessment could include provision for mindfulness education and an assessment for anxiety using a brief screening tool such as The Generalised Anxiety Disorder Questionnaire (GAD-7) (Spitzer, et al., 2006). The GAD-7 has recently been recommended for use with parents of children with CF by The Cystic Fibrosis Foundation and the European Cystic Fibrosis Society Guidelines Committee on Mental Health (Quittner, et al., 2015). Additionally, the Reaction to Diagnosis interview (Pianta & Marvin, 1993) is a brief measure of grief associated with diagnosis that the social worker or psychologist could administer to parents to identify early, potential psychopathology.

Recently, substantial attention has been given to the role of translational research in public health systems and the provision of health care services (Oborn, et al., 2010; Ogilvie, et al., 2009).

This view emerged from recognition that health systems across the world often did not reach their full potential in the provision of health services (World Health Organization, 2004), and that health knowledge did not always translate into practice (Woolf, 2008). Findings from my research demonstrate a gap exists between the translation of the current lived experience and understandings of early diagnosis and early surveillance held by parents into clinical practices among the treating team. For example, improving or enhancing family resiliency appears an appropriate and sustainable approach to caring for children undergoing early surveillance within the wider family context. Benefits of such an approach should be systemic and reciprocal; benefits for child and the family system, and benefits for the child because of the care for the family system. A benefit might be that experiential effects of early surveillance from a psychological perspective are more clearly defined, which could result in targeted support for parents and their families as they embark on a unique experience that has little specific psychological support currently built into it.

In review, my research highlights the importance of continual conversation that needs to be had between the treating team and the parents about their experiences of parenting within the context of early surveillance for CF lung disease to ensure knowledge is better translated into policy and practice as real-world application. To sustain such a process, parents first need to feel appreciated, and that what they have to say is important and valued. Although recent clinical guidelines advocate for early intervention, parents in my research often expressed that their experiences with clinical practices and interactions with clinical staff were not always fully understood. As such, my research indicates a disconnect between clinical practice and the actual lived experience of early surveillance. My research highlights the substantial role of qualitative research in exploring the lived experience of parenting a child with CF undergoing early surveillance though moreover, underpinning factors have been identified for development of a framework that would serve not only to inform clinical practice and policy from a holistic perspective, but to critically assess how knowledge of the lived experience of parenting a child undergoing early surveillance for CF lung disease is translated into practice.

7.3 Parents' constructions and attributions of meaning to early surveillance

Research question three:

3. *How do parents construct their experience and attribute meanings to early surveillance for CF lung disease?*

A salient finding of my research was evidence that constructions of experience and attributions of meaning were foremost primed by the wider societal discourses used to promote early intervention in paediatric chronic disease. Parents' constructions of early surveillance were

particularly influenced by biomedical discourses prevalent in wider society that emphasised an early intervention understanding of paediatric chronic disease. Findings also revealed that the notion of early intervention was highly familiar to parents and was found to influence both how they thought about adherence to their child's treatment regimen as well as how they constructed meaningful understandings about the experience of early surveillance. In particular, proactive and preventive treatment was of paramount importance to parents given the chronic nature of the condition. For example, parents were keen to know their children were being treated as early as possible. This finding reinforced how discourses within social contexts can influence how parents related to their own personal experiences of CF in the context of early surveillance.

Moreover, parents revealed that making social comparisons with other families encountered within the hospital environment as well as with families whose child has CF, but who does not undergo early surveillance for CF lung disease, was a salient element of constructing meaning about their own experiences of CF. Making comparisons with others enabled parents to retain both a positive self-perception and perception of their family's present and future (Moola, 2012; Tanner, 2007). This tendency allowed parents to reflect on their fears, hopes and plans for the future (Giles, McIlrath, Mulac & McCann, 2010). Furthermore, making downward comparisons appeared to sustain parents' motivation to fulfil personally meaningful goals such as mastery of the treatment regimen and normalisation of the condition into family life, a finding that was consistent with previous research (Goldberg, et al., 1990; Heaton, 2014; Hughes & Beer, 2012). Additionally, the data suggest that the effects of making downward comparisons may not only be relevant for parenting a child with CF but also for the early surveillance experience, which resulted in hope for the future and gratitude for the present, a finding that would benefit from further research. Therefore, the current findings both support and extend existing social comparison literature (Festinger, 1964; Kwan, et al., 2003; Moola, 2012; Tanner, 2007).

My research supports the notion that meaningful understandings of early surveillance are more than the clinical outcome. Rather, with the influence of knowledge on constructing meaning about early surveillance, an opportunity presents itself to understand what becomes of importance for parents who enrol their children in early surveillance for disease progression. With parents' lived experiences reflecting personal outcomes of hope, gratitude, control and power, this finding suggests that early intervention initiatives need to embrace a holistic awareness of all aspects that influence parents' experience of the child's condition. It became evident that future policy decision making and models of practice should be encouraged to focus on the advocacy parental experience in a way that more closely aligns with their lived experiences. An area identified in my research where policy makers might translate knowledge into practice includes growing acknowledgement of

a link between self-efficacy, locus of control and self-esteem (Judge, Erez, Bono & Thoresen, 2002; Pruessner, et al., 2005; Wang, Zhang & Jackson, 2013; Wilson, et al., 2013), and how that link can be applied to parents' well-being in the context of early surveillance. For example, a resiliency approach to holistic care could be used to educate parents (and clinicians) about how taking ownership over their child's care will contribute to confidence in their redefined parental roles. Broader strategies may include reviewing the way early surveillance is introduced to parents, including timing, content and modality of information, or development and implementation of a standardised protocol for its introduction.

Moreover, my findings suggest that future policy and practice needs to better acknowledge and critically evaluate relationships between experiential, practical and psychological factors associated with early surveillance in shaping parents' thoughts and emotions around their child's and their family's future, their perceptions of themselves as parents, and their hopes, dreams and fears. A critical examination of the relationship between such factors would generate further understanding of both beneficial and adverse influences of early surveillance on parents' constructions of their experiences and attributions of meaning to them. Much like the sense of moral and social obligation held by the CIHR to ensure that outcomes of research proceed back to individuals who could benefit (Canadian Institutes for Health research, 2004), an essential component of the investigation would not just involve critically thinking about parental experience of clinical structures and practices. Rather, it would extend to recognise that those in positions of power who are in support of early surveillance need to consider parental consequences of such decisions and actions, which are now recognised as outcomes of my research. As demonstrated by my research, lack of both standard protocol and therapeutic support denies substantial adverse influence of early surveillance on parents, as well as negates potential misconceptions about research and clinical care that could affect autonomous decision-making. Moreover, lack of education about beneficial influence of early surveillance to parents also denies them full potential psychological and experiential benefits.

7.4 Coping with parenting children with CF in the context of early surveillance: Implications for theory, policy and practice

Research question four:

4. *How do parents cope with the lived experience of parenting in the context of early surveillance?*

Further to exploring the lived experience of parenting a child within the context of early surveillance for CF lung disease was to explore how parents cope with their experiences. Again, the

unique context gave me the opportunity of exploring how parents cope with the lived experience of parenting within the context of early and aggressive treatment for CF lung disease. The findings of my research have a number of important implications for future theory, policy and practice.

7.4.1 Theoretical implications.

My research has described and demonstrated that there are strong indications that parents generally cope well and adapt functionally to their child's CF diagnosis and treatment within the context of an early surveillance regimen. Parents' perceptions of hope, perceived control and power means that a resiliency perspective can further foster existing hope by inviting the family unit to recognise and call on their inherent strengths and coping abilities. Moreover, my research supports contemporary conceptualisations of resilience models for coping with paediatric chronic conditions, which if utilised in clinical practice, can encourage HCPs working with families to recognise resiliency and the inherent strengths within the family unit. If understood by HCPs, resiliency characteristics and inherent family strengths could become targets for strengths-based intervention. Lastly, a resiliency perspective would provide information needed to develop preventive interventions for families. For example, social comparison has been identified in my research as an inherent strength that reduces the intensity of burden and demands that families face, and activates adaptive coping strategies. Such information could be used in the development and implementation of preventative interventions.

Family resiliency theory highlights the complex but meaningful role that family properties, behaviours and capabilities play in buffering the impact of stressful life events, as well as facilitating the family's recovery in the face of family crisis, such as diagnosis of a paediatric chronic condition. From a salutogenic point of view, the family resiliency framework can be used to discover and build upon parents' and family's' existing strengths and resources, which makes it particularly well-suited to family-focussed, resilience-oriented work with families. A crucial initial step to such work is to identify existing and potential skills, attitudes and other resources that may enhance the family's overall growth and response to adverse circumstances. Such an approach runs counter to the traditional deficit-based models of assessment and intervention that have dominated training and practice in the helping professions. Traditional family assessment is based on the medical model assumptions and strategies aimed at identifying and treating family pathology and dysfunction (Goldenberg & Goldenberg, 2004). Using a resilience framework can help practitioners identify aspects of family resilience that may go underutilised by the deficit-based model traditionally used in clinical care.

My research has shown that parents use emotion-focussed and avoidant coping for managing early surveillance, yet they use early surveillance as an instrumental and problem-

focussed coping strategy for managing CF. Literature supports negative outcomes for emotion-focussed and avoidant coping styles including low emotional and social quality of life (McHugh & Otto, 2012), as well as depression, anxiety and stress (Sheehan, et al., 2014), suggesting that coping with early surveillance may be detrimental to parents' well-being. Additionally, is the concern of chronic sorrow around the time of the annual early surveillance review. However, instrumental and problem-focussed coping has been associated with high emotional and social quality of life (McHugh & Otto, 2012). Therefore, early surveillance may function to decrease social and emotional quality of life, as well as increase depression, anxiety and stress, and potentially trigger chronic sorrow around the time of annual review. However, early surveillance may function to increase emotional and social quality of life on a more long-term basis.

The potential for decreased social and emotional quality of life, along with depression, anxiety and chronic sorrow around the time of annual review as a function of increased social and emotional quality of life on a longer-term basis provides evidence that early surveillance is in the best interest of the child and their parents, but that it is a psychologically distressing experience for the family. Parents who behaviourally or psychologically disengage prior to annual review and/or cognitively distract themselves around the time of annual review may be at increased risk of low social quality of life during this time. This could be critical for parents' coping capacity because my research has shown that social support is integral to managing the early surveillance experience. Furthermore, that short-term negative experiences (i.e., cognitive dissonance, fear for their child's safety) appear a compromise for long-term positive outcomes (perceived control power, hope, gratitude, and altruism) may counteract the negativity experienced at annual review. Essentially, any psychological adversity experienced by parents as a result of their child's early surveillance exposure appears to be short-term adverse individual effects that are accounted for by long-term positive child health outcomes and beneficial psychological parental outcomes. However, there is still the concern of triggering chronic sorrow that by definition, is continuous and recurring.

My research reinforced prior literature that has emphasised the importance of social support for parents coping with paediatric chronic disease (Greening & Stoppelbein, 2007; Hoekstra-Weebers, et al., 2001; Horton & Wallander, 2001; Wiedebusch, et al., 2010). Unique to my research was the finding that social support contributed to multiple elements of the CF experience within the context of early surveillance. Support from family, friends and both the medical and research teams contributed to the way parents; managed their child's diagnosis and treatment regimen, normalised CF and the treatment regimen into family life, coped with their fear associated both with uncertainty of CF prognosis and with their child's early surveillance procedures. The data also support previous research that has described the importance of social support for coping with medical procedures

(Marsac, Donlon, Winston & Kassam-Adams, 2013; Peterson, 2013). Therefore, social support as a primary coping resource for fear associated both with early surveillance procedures and with fear of disease progression highlights the importance and value parents place on establishing and maintaining relationships and networks. These findings compliment theoretical advocacy in the literature arguing for the importance of social support in adaptation to paediatric chronic disease (McCubbin & McCubbin, 1993; Walsh, 2003). Lastly, findings support previous research regarding the importance of recognising and addressing such existential needs in holistic approaches to clinical care for children with chronic conditions (Bekenkamp, Groothof, Bloemers & Tomic, 2014; Shilling, et al., 2013).

The findings of my research also build upon prominent theories about the importance of developing and maintaining a perceived competent parental role as individuals transition, and adjust, to parenthood (Cast, 2004; McBride & Toller, 2011; O'Connor & Barrera, 2014). In my research, parents reported that perceiving themselves as a competent parent within the context of their child's condition was essential in exercising meaningful practices that contribute to perceptions of themselves as parents. Moreover, parenting practices and perceptions were found to be important markers for parents in understanding their own parenting experience. Despite challenges, parents were generally able to regulate their psychological states successfully as indicated by research into other paediatric chronic conditions (Clark, et al., 2014; Dardas & Ahmad, 2015; Knafl, et al., 2013; Ruiz-Robledillo, De Andrés-García, Pérez-Blasco, González-Bono & Moya-Albiol, 2014). Parents achieved psychological regulation (including emotion) through maintenance of a positive attitude, primarily by way of accepting and assimilating CF and its treatment into family life, as well as redefining parent identity. Parents were able to maintain hope (Barlow & Ellard, 2006; Faso, et al., 2013; Horton & Wallander, 2001; Kashdan, et al., 2002; Lloyd & Hastings, 2009; Ogston, Mackintosh & Myers, 2011; Petersen & Wilkinson, 2015; Samson, et al., 2009; Wong & Heriot, 2008), and negotiate the dynamics of gains versus adversities to achieve those gains. These findings provide support for the assumption that families with positive meaning and appraisal are likely to adapt to paediatric chronic disease, as well as be resilient to future burden of the condition (Black & Lobo, 2008; Lazarus & Folkman, 1984; McCubbin & McCubbin, 1996).

Lastly, normalisation of CF into the family setting and positive meaning-making about the condition were of primary importance to the CF experience within the context of early surveillance, which supports the Resiliency Model's emphasis on them as important processes for adaptation. Moreover, my research has highlighted how early surveillance can positively and negatively influence these processes. Therefore, these findings suggest that families who are able to draw meaningful inferences about CF and early surveillance, and establish newly instituted patterns of

family functioning are likely to adapt to paediatric chronic disease as well as be resilient to future burden of the condition (Black & Lobo, 2008; McCubbin & McCubbin, 1996). Furthermore, family processes and outcomes of adapting to CF should be viewed within the context of early surveillance because they are part of the same experience.

7.4.2 Policy and practice implications.

Policy and practice implications of my findings requires thinking about, and implementing, support strategies that promote and/or enhance adaptation to paediatric chronic disease within the unique context of early surveillance for covert disease progression. For example, resiliency-building strategies could be implemented as part of the annual review process when the social worker or psychologist within the multidisciplinary team offers his/her support to the family. Such strategies could inform a support program designed to build capacity and develop resources for parents' coping with adversity. The offer of support could include elements of education, as well as referral to resiliency-building community programs. Implementing support strategies through policy and practice, as well as thinking beyond clinical practice, is of primary importance so that parents can nurture their families as competent and supported parents.

Firstly, family resiliency substantially influences coping with, and adjustment to, life stressors and events such as paediatric chronic illness for all family members (Giallo & Gavidia-Payne, 2006; Knafl, et al., 2013; Van Schoors, et al, 2015). Early surveillance of covert disease has been shown to positively and negatively influence processes that make up family resiliency, that is, positive behavioural patterns and functional competence. Therefore, building and maintaining family resiliency should be a focus for paediatric clinical teams for families in their care because reinforcing family resilience may enhance families' coping and adaptation abilities/capabilities. Additionally, despite hardships and stresses of early surveillance specifically, and CF more generally, given the findings of my research, a strengths-based approach to managing families appears an appropriate model of support.

Implementing family resiliency frameworks that build program-specific experiences into existing support programs for parents of chronically ill children have shown promising outcomes (eg. Kieckhefer, et al., 2013; Mullins, et al., 2015). Accordingly, developing a resiliency program specific for early surveillance of covert disease could utilise elements of the early surveillance experience identified in my research that are known to support development of resiliency, for example, self-efficacy, locus of control, and adaptive coping (Kilic, Dorstyn & Guiver, 2013; Skinner, Pitzer & Steele, 2013; Wang, et al., 2013). Moreover, based on theoretical advocacy and support from my research, as well as others (Mitmansgruber, et al., 2015; Horton & Wallander, 2001), a number of strategies could be considered within a resiliency framework as being influenced by the early surveillance

process that are known to contribute to adaptation and resiliency when faced with stressful events. Firstly, strategies towards building family resiliency could: cultivate strong support networks both within and outside the family to help find understanding, practical advice and meaning (i.e., resources for social support); set goals that include hope for the future and motivation for the family to stick together (i.e., sense of coherence); learn as much about the condition, treatment and child health status as possible and educate family members about therapies and ongoing research (i.e., individual resource that contributes to family adaptation); and hold a belief in a greater entity than thyself (i.e., altruism). The adapted Resiliency Model presented in the previous chapter is a framework that could be used to guide development of a resiliency program. The program could focus on developing those family characteristics of resilience that I demonstrated as contributing to adaptation to paediatric chronic disease in the context of an early surveillance program.

Secondly, emotion and problem-focussed coping strategies I have identified assists our understanding of the role family resources play in parents' management of their child's CF, both practically and experientially. As family resources, these coping strategies contribute to a family's coping patterns. Problem-focussed coping strategies are not as commonly reported in the literature as emotion-focussed coping strategies are for parents of children with CF, as they have not traditionally been afforded such coping mechanisms. Due to the uniqueness of early surveillance, how a program such as AREST CF functions to facilitate coping has not previously been reported. Through access to, and execution of, both emotion and problem-focussed coping strategies, parents have a combination of coping patterns identified by the Resiliency Model for managing their child's CF in the context of early surveillance. In fact, despite immediate distress and discomfort, it would seem that early surveillance is a primary mechanism for coping with CF over the long-term.

Problem-focussed coping has been highlighted as an essential element that deserves more attention for the purposes of implementing family resiliency practices for those families whose children undergo early surveillance for CF lung disease. As families change and further coping mechanisms are needed, problem-focussed coping is a promising avenue for assisting families to develop coping strategies that currently do not receive much attention. Using the findings from my research as a basis, promoting problem-focussed coping strategies could positively influence services these families receive. Informing clinical practice through offering new ways to support families, family resiliency practices can be developed and refined.

Thirdly, given the importance of appraisals in response to stress (Lazarus & Folkman, 1984) and the importance of appraisals in adapting to paediatric chronic illness (McCubbin & McCubbin, 1993), developing a useful family schema is an essential process for families to engage in; because the way in which a family defines a crisis has a critical impact on how it copes (Hawley, 2000). Family

schema describes the shared values, goals, priorities and expectations of family members, and is a key element of highly resilient families (McCubbin & McCubbin, 1993). With knowledge that family schema is beneficially influenced by early surveillance, clinicians and other HCPs working with these families can help parents to construct or enhance their shared view of the world.

Family sense of coherence is a closely related construct to resiliency, which refers to a measure of the extent to which families feel confident that the outcomes of a situation will be favourable for them (Hawley, 2000). Sense of coherence originated from Antonovsky's (1987) salutogenic notion to health promotion. That parents whose children undergo early surveillance for CF lung disease have hope for favourable outcomes is likely to contribute to their sense of coherence, further validating the importance of family schema in the context of early surveillance. Therefore, therapy from a resilience perspective can focus on how a common view as a competent unit can be accessed, developed, enhanced and maintained as families seek to constantly adapt to their child's progressive condition. Moreover, encouraging a family to develop a collective view as survivors of distress is not only clinically useful in the present but can equip families for facing adversity in the future.

Lastly, my research has gone some way towards enhancing our understanding of how coping with similar events becomes more effective over time. Parental coping appeared to become more effective as knowledge about CF (e.g., prognosis), and experience with its treatment regimen (i.e., normalisation within the family setting, early surveillance procedures and home treatment) were obtained. Accumulation of both knowledge and experience facilitated creation of expectations, therefore changing the appraisal of the event in subsequent encounters. That is, more effective coping was facilitated by being able to manage subsequent encounters with the new knowledge and previous experience, which changes the appraisal of those subsequent encounters by building expectations into the appraisal. Therefore, parents of older children are likely to cope better than parents of younger children because they will likely have more, or different, coping skills that they have developed over time due to their experiences and accumulation of knowledge.

By understanding both the practical and experiential encounters of an event, parents can develop expectations and subsequent mechanisms to cope with future related events. Expectations contribute to parents' succeeding encounters by providing a psychological framework to orient themselves for what is likely to occur in the future, along with how they will react, and how they will cope. This practical and experiential orientation contributes to their ability to cope, or manage more effectively, with subsequent experiences. This information can be used to develop targeted activities aimed at supporting parental coping with their child's condition across the 7 years a child undergoes early surveillance. For example, talking to other parents with older children in the AREST CF program

may help parents to access that sense of experience and expectation through listening to other parents. In other words – talking about what it is like for parents who have been through it can help their understanding and their ability to cope. Additionally, a walk through the paediatric unit at the treating hospital, exposing parents to the procedures and environment might help parents to create expectations, and to begin to understand what their experience may be like, which may further contribute to their understanding and their ability to cope.

In review, my research substantially adds to a growing body of literature on the importance of facilitating adaptation to paediatric disease, as well as how parental appraisals and coping resources and abilities are important factors in the process to adaptation. Moreover, my research has highlighted how early surveillance contributes to parents' appraisals and coping resources and abilities. Accordingly, my research indicates a need to further characterise adverse outcomes that might hinder adaptation. However more importantly, my research highlights that families cope well, and appear resilient to burden of CF, because of early surveillance. Therefore, utilising a family resiliency approach to support families in achieving adaptation that is tailored to their unique needs and experiences will offer a modern and innovative approach to managing these families.

7.5 Further considerations: Future program development and implementation, and mental health care of parents and implications for their families

The Cystic Fibrosis Foundation and the European Cystic Fibrosis Society Guidelines Committee on Mental Health highlighted the inadequate and variable nature of CF mental health care across centres in Europe and the US (Abbott, et al., 2015). Additionally, a survey about experience of care administered in the US concurred with inadequate mental health care for patients with CF and their families (Homa, Sabadosa, Marrow & Marshall, 2015). The same disconnect seems to exist across Australian CF centres. A consensus statement released by the committee (Quittner, et al., 2015) recommended that CF teams provide sufficient education to help parents to develop coping skills and disease management.

I suggest developing guidelines for application of the Resiliency Model of Stress, Adjustment and Adaptation to address critical elements of parent and family functioning within the context of the AREST CF program. I believe that such an approach will ensure clinicians and other HCPs develop strategies for intervention based on a systematic diagnosis and evaluation of parent and family functioning under stressful circumstances. To implement additional services for parents and their families within the hospital setting, infrastructure and finance support is needed from the hospital. Evidence-based guidelines may support CF teams in leveraging funds for mental health professionals where this aspect of healthcare has traditionally been overlooked (Havermans & Staab, 2016). The social worker and/or psychologist require these additional resources if they are to support families

from a resiliency perspective and educate them on how to effectively cope with early surveillance. For the psychologist's role within the multidisciplinary CF team to be effective, Kerem and colleagues (2005) recommended that the post should be no less than 50% of their working time.

The Cystic Fibrosis Foundation and the European Cystic Fibrosis Society Guidelines Committee on Mental Health also recommended annual screening of anxiety and depression in parent caregivers, and if needed, referral for treatment to primary care or mental health services (Quittner, et al., 2015). Duff et al., (2016) evaluated the feasibility of these recommendations with 65 parents of young children with CF during outpatient appointments. They suggested that, depending on the size of the CF clinic, an annual 3-6 month screening period could be attainable. The following diagram is an algorithm set out by the committee for screening and treatment of depression and anxiety for parents and caregivers:

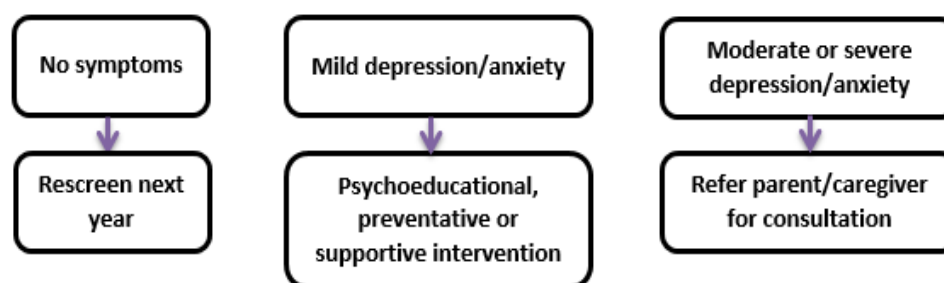


Figure 13. Screening and treatment of depression and anxiety: Algorithm for parents/caregivers

Preventative education and routine screening for anxiety and depression will create an opportunity for appropriate preventive and supportive interventions instead of reactive therapeutic intervention. Given the findings of my research, screening for depression should include awareness that chronic sorrow is a likely mental health issue for parents, especially those who experience multiple events associated with their child's condition. Going beyond the traditional deficit-model of psychological care is a modern approach to preventive and educational care for paediatric patients and their families. Therefore, assessment of both psychopathologic and resiliency factors would adequately address the multi-dimensional nature of how families cope with their child's chronic condition. Moreover, addressing mental health issues systematically is likely to improve health outcomes, quality of life and reduce healthcare utilisation (Burker, Sedway & Carone, 2004; Smith, et al., 2010).

Early surveillance techniques are promising, and clinical trials are needed to test which ones are the best markers of disease and indicative of prognosis. Such outcome measures can be used as clinical endpoints so that psychological distress experienced by parents can be substantiated, by showing that early surveillance does indeed prolong children's lives. Lastly, but just as importantly, parents' mental health and their ability to cope effectively are known significant predictors of family

functioning (Koutra, Triliva, Roumeliotaki, Lionis & Vgontzas, 2015; Pakenham & Cox, 2012; Robitschek & Kashubeck, 1999; Saunders, 1999), and both child physical and mental health (Sheidow, Henry, Tolan & Strachan, 2013; Szyndler, et al., 2005; Turner-Cobb & Steptoe, 1998). Moreover, family functioning is also a significant predictor of both child physical and mental health (Everhart, et al., 2014; Patterson, et al., 1990; Patterson, et al., 1993; Walsh, 2003). Therefore, considering parents' mental health as a primary predictor of how the psychosocial environment of the child influences their health outcomes will ensure the child and their family is supported in a systemic way, just as the family is a systemic unit.

In summary, my research offers important feedback about; the lived experience of parenting a child with CF in the context of early surveillance for lung disease, parents' involvement in the early surveillance program, and it invites consideration of the development and implementation of current and future practice and policy designed as a holistic approach to paediatric CF care. Specifically, despite short-term adverse parental reactions and responses to their child's unpleasant experience for which they feel ultimately responsible, the long-term benefits to the child and the family far outweigh immediate discomfort and distress. Importantly, my research has identified areas of concern and suggested areas for improvement to support parents as they embark on such a unique path of modern paediatric CF care. Undervaluing the non-clinical aspects of early surveillance participation for parents limits the possibilities for addressing the holistic needs of parents' psychological well-being. As early surveillance is inextricably tied to the CF experience, non-clinical aspects of CF care are of paramount importance to the maintenance of their child's health outside of the clinical setting.

7.6 Strengths and significance of the research

My research has been responsive to the need for obtaining contemporary parental perspectives, understandings and constructions of early surveillance for CF lung disease from both mothers and fathers. As unique as early surveillance for CF lung disease is, so too are parents' experiences that extend beyond current knowledge of parenting a child with CF. This information is timely given the likely uptake of the early surveillance framework for future therapeutic intervention trials in young children nationally and internationally.

A substantial strength of my research methodology was that it allowed for extension of current understandings of parenting a child with CF beyond local boundaries resulting in data collected from two national sites. Deriving themes from data collected with parents in Western Australia and Victoria made it possible to demonstrate the lack of site-specific influence of early surveillance on the construction of meaning that parents formed about their early surveillance experience. In particular, a salient finding was that both adverse and beneficial experiences and

outcomes asserted by parents in the Western Australian context were also found to have relevance for parents in Victoria, highlighting the significance of the findings as well as the unique contribution of my research.

A further strength of my research was the involvement of parents to provide information about their experiences of parenting within the context of early surveillance for CF lung disease. People are best positioned to report their understandings of, and meanings in, experience that they encounter (Denzin & Lincoln, 2005, 2008; Gelo et al., 2008; Patton, 2002), and it was therefore appropriate to utilise a qualitative methodology to provide parents the opportunity to express and explain their experiences from within the personal context with which they occur (Chwalisz et al., 2008; Corbin & Strauss, 2008; Creswell, 2007; Janesick, 2000). By consulting with parents about their experiences, I was able to gain insight into processes and principles of meaning-making about early surveillance. An additional benefit of my research methodology was that I was able to identify factors that both mediated and influenced how parents constructed understanding about their child's early surveillance for CF lung disease, their child's condition and their parental role. Moreover, this type of consultation allowed collection of in-depth information about the unique experiences of parents that perhaps would not have been identified through the quantification of personal experiences, or relying on secondary sources of information (Creswell, 2007). Consequently, any actions resulting from this shared information concerning parents' lived experiences of parenting a child with in the context of early surveillance are more likely to be responsive to their needs.

7.7 Limitations of my research

The primary aim of qualitative research is to generate findings that are beneficial and authentic within the context in which data was collected, which means that limitations exist as to the extrapolation of findings to other populations. Therefore, future research could assess the applicability of these findings amongst other population groups across different clinical contexts and settings. Additionally, given that participation in my research was voluntary, it is possible that some parents were representative of; a more motivated sample, parents who had a vested interest in the early surveillance program, or parents who were confident in discussing their experiences. Accordingly, future research with parents who are less confident in discussing their experiences would provide a more comprehensive account of the lived experience of parenting a child with CF in the context of early surveillance. Moreover, the amount of time between interview and annual early surveillance review varied because early surveillance reviews are conducted throughout the year at different times. Therefore, parents' recollection of events, and their associated emotions and cognitions could vary depending on time between review and interview.

Whilst not a limitation as such, as an explorative research design, it is impossible to state any sense of causality about how relationships and processes function to create outcomes. Therefore, undefined (that is, bi-directional or associative) relationships have been identified that warrant further investigation to determine direction and strength of relationships. Moreover, determining whether any significant relationships function as predictors of health resilience are important issues for future research so that mediators and moderators of health and family resiliency can be better understood. Accordingly, a longitudinal design investigating parenting a child within the context of early surveillance for CF lung disease to see if attitudes, outcomes and understandings change over time would add value to my research findings.

Lastly, the data collection method requires consideration in terms of how it may have affected the quality of my data. For optimal study recruitment, as well for convenience of data collection for parents, the limitation imposed by conducting one and two-parent interviews was deemed less important than obtaining a representative sample of both mothers and fathers. However, different methods for data collection may have influenced information discussed with me by potentially hindering couples' responses because of fear or concern about a partner's response to certain information that they may have expressed more openly and honestly if interviewed on their own. Therefore it must be acknowledged that methodological limitations of collecting data in this way may have compromised findings, such that data quality and content may have been different if parents were interviewed alone.

7.8 Directions for future research

Findings from my research emphasise the importance of shifting from a purely biomedical perspective when exploring the lived experience of parenting children with chronic conditions and relationships that mediate the experience. This involves continuing to challenge the unidimensional biomedical model in clinical care, as well as non-family systems assumptions, theories and perspectives in parenting children with chronic conditions. Findings from my research support a salutogenic family resiliency perspective, that is, a holistic perspective that acknowledges the power of the family in nurturance and development of its members. Therefore, I suggest that such an approach be considered when addressing issues associated with parenting children with chronic conditions. For example, acknowledging through policy and practice that pathways to family adaptation and resiliency are influenced by parents' appraisal of their ability to cope with their child's chronic condition within the family context with which it occurs.

Whilst descriptions and explorative research on parents' experiences encompasses a holistic approach to a child's clinical care, both prior research and my research suggest that understandings are still dominated by biomedical understandings of health (i.e., treatment regimen, nutrition)

despite the lived experience emphasising the importance of psychological and systemic factors in facilitating sense of personal control and parenting competence in conducting home treatment as parents adapt to managing their child's CF. Therefore, to effectively promote treatment adherence and a more holistic health message to the paediatric community, critical consideration needs to be given to how knowledge about the lived experience of a child's clinical care is better translated into clinical policy and practice, particularly the psychological and systemic aspects. Moreover, my research findings support recent theoretical and empirical literature that advocates for the use of salutogenic models of psychological care for people experiencing adverse events (e.g., Antonovsky, 1987; Seligman & Csikszentmihalyi, 2000) suggesting the continued necessity for future models of care to incorporate a strengths-based, rather than purely a deficit-based, perspective to supporting parents adapting to their child's chronic condition.

Findings from my research similarly indicated that the concepts of power and perceived control from knowledge about early disease progression would benefit from future research. I suggest these concepts and their relationships with perceived parenting competence and parenting practices such as treatment adherence be further delineated so that parents and children can be further supported in adapting to CF in the context of early surveillance. Moreover, with evidence emerging from the data that making social comparisons influenced how parents constructed meaning and that using social support influenced how parents managed their experiences, my research suggests that social connections provide a powerful medium to promote adaptive perspectives on paediatric chronic disease. Promoting parental and family adaptation to paediatric chronic disease would benefit from future research, investigating how social networks may be used to generate and sustain adaptive perceptions and how social connections can be effectively used to translate knowledge about paediatric chronic disease into the delivery and implementation of clinical agendas, policies and services.

To influence positive change in the experience of parenting a child undergoing early surveillance for CF lung disease, findings from my research revealed that adverse parental reactions and responses to both CF diagnosis and the early surveillance medical procedures are existing issues warranting additional attention. Coping with psychological adversity was a substantial finding of my research, pervasive across the condition and its clinical care. Fear, anxiety, guilt, chronic sorrow, cognitive dissonance and ambivalence recognised from my research as impacting upon parents' psychological well-being warrants further investigation. What role these outcomes play in the construction of meaning in; parenting, CF, early surveillance is of particular importance to address these issues. So too are the constructions of parenting a child within the context of early surveillance for CF lung disease important. Additionally, my findings suggest that there is benefit in examining

whether parents whose children undergo early surveillance for CF lung disease experience more adverse outcomes as a result of more and/or earlier hospitalisations than parents whose children who have less and/or later hospitalisations, or whether parents whose children undergo early surveillance are more resilient for future burden of CF for the same reasons.

Lastly, efficacy of the Resiliency Model is best demonstrated in studies designed to measure several critical variables with the intention of determining which variables, in what causal order, help to explain variability in family adaptation. Therefore, path analyses and structural equation modelling could be considered to assess variables identified in my study as important factors that might contribute to family adaptation within the context of early surveillance for CF lung disease. Moreover, how processes and pathways are common to other paediatric chronic conditions would shed light on commonalities and differences across those conditions that may be amenable to intervention and/or support within the wider context of families living with paediatric chronic illness.

To conclude, although beyond the scope of my research, an important consideration for future research based on my findings would be to further explore the role that clinicians, other HCPs and the hospital system more broadly plays on parental thinking, in promoting and influencing parental decision-making about their child's clinical care. This would assist in making visible those processes that maintain misconceptions about medical research and clinical components of early surveillance. Taking into account the overwhelming and hectic nature of CF diagnosis and multi-disciplinary nature of CF clinical care, identifying when and how such perceptions are developed and maintained could lead to better clinical understandings on both clinicians' and parents' behalves.

7.9 Final considerations

A primary goal of my research was to address a gap in knowledge about the lived experience of parenting a child undergoing a unique clinical care regimen for paediatric CF through consultation with parents about their experience. A related goal was to highlight how this knowledge can be used to develop policy and practice that more closely reflects parents' lived experience of CF in the context of early surveillance. In particular, this research aimed to understand influences on constructed experience and attributions of meaning in clinical care with a particular interest in the disjunction between policy and practice constructions and lived experiences. Although my research has identified the importance of familial and psychological factors to parents' experiences of their child's clinical care, findings suggest that parents' understandings of early surveillance were influenced by dominant social and biomedical discourses. It is important that these findings are used to inform existing and future policy decision making and models of best practice towards promoting adaptation and family resiliency for future burden of paediatric chronic disease. However, if this

knowledge is to be translated into practice so as to more closely align with parents' lived experiences, a critical review on how to more effectively translate policy into practice is required.

In conclusion, findings from my research encourage further thinking about how parents and their families strive to achieve normality when their child is diagnosed with a chronic condition, and more specifically, how fear, control and hope are implicated with knowledge of covert disease progression in both practical and experiential terms. Furthermore, findings from my research also provide evidence that parenting a child with a chronic condition is socially constructed, and therefore a modifiable experience. With evidence that clinicians, other HCPs and dominant discourses have an influence on parents' thinking, positive aspects of early surveillance through parents' experiences, particularly the psychological aspects need to be more effectively communicated and promoted in future clinical policy and research. Moreover, these aspects need to be reviewed as to how they can be more effectively translated into practice, specifically that constructions of CF and parenting are influenced by participation in an early surveillance program. Shifting the focus in this direction has the potential to significantly modify parents' constructions and understandings of parenting children within the context of early surveillance, which could therefore influence the broader parenting and family experience more positively, and ultimately influence children's long-term health outcomes.

Lastly, I would like to reiterate the centrality of hope expressed by parents in my research and the importance of hope for resilience and coping with adversities. The magnitude of hope, and the effects it has in the daily lives of individuals, can be seen in President Barack Obama's victory speech following his campaign for the parliamentary leadership in 2008 that included the iconic *hope* poster. His speech embraced and pulled at the heart-strings of the United States public. His speech was one of hope; hope *of* a nation, and hope *for* a nation. His speech eloquently articulated 10 references of hope for the future, one of which was:

I'm not talking about blind optimism, the kind of hope that just ignores the enormity of the tasks ahead or the road blocks that stand in our path. I'm not talking about the wishful idealism that allows us to just sit on the sidelines or shirk from a fight. I have always believed that hope is that stubborn thing inside us that insists, despite all the evidence to the contrary, that something better awaits us so long as we have the courage to keep reaching, to keep working, to keep fighting (Obama, 2008).

Obama's expression of hope in this particular excerpt epitomises the hope expressed by parents who took part in my research because; they do not underestimate the magnitude of their situation, they are not simply observers of their situation but are proactive and action-oriented, and despite the enormity of their situation, they are empowered to strive for a normal family life that requires drastic and challenging changes to their initial expectations, roles and functions.

My view is that the salutogenic approach of parental support is a well-suited approach to building and/or maintaining hope as a family characteristic that contributes to adaptation to paediatric chronic disease and promotes family resilience for future burden – both normative and disease-specific. The inherent link described at the beginning of my thesis between hope and empowerment emanates from belief in choice, and belief that chosen actions can generate a future different from the present (Groopman, 2005). Such beliefs cultivate motivation and capacity to move forward despite present challenges. Early surveillance for CF lung disease is a catalyst for empowerment through knowledge and a choice to act proactively, and belief that knowledge and actions will bring about a future for children and families that is different from what has been presented to them. Knowledge is constructed as control and the meaning attributed to control is hope; hope for their child, hope for CF generations, hope for the future, and hope for a cure. Hope is empowerment; motivation and capacity to strive for needs, desires and goals that might otherwise seem insurmountable. Hope is coping with challenges and burdens. Hope is resilience. As such, my thesis is a message of hope and resilience.

7.10 Postscript reflection

Notwithstanding ethical issues and challenges of investigating personal experiences of parents who have chronically ill children, the emotional challenges I might experience as a researcher was an aspect of humanistic research I failed to consider. As I read literature early in my doctoral research, I sensed parents' loss and sadness and I sensed their determination. My early readings primed me to think about ethical relationships and meaningful research methodologies for parents of chronically ill children. However, what it did not prepare me for was how to manage my own personal responses and affective states while conducting my research. In varying degrees and frequency, emotions and feelings such as sadness, empathy, enlightenment, passion, inspiration, gratitude and joy accompanied my journey. Such responses underpinned questions about potential biases I might hold as a researcher, as well as the place of qualitative psychological research in exploring understudied phenomena in understudied populations within understudied contexts.

However, key to confronting and constructively analysing these experiences and thoughts through the interactive researcher-participant dialogue (Ponterotto, 2005) was the reiterative practice of deep critical reflection (Charmaz & McMullen, 2011; Schwandt, 2000). Such endeavours were pivotal to challenging familiar assumptions of parenting practices, as well as opening my naivety to the complexity of parenting children with chronic conditions. Lastly, it is important to not overlook the ability of such parents to inspire and shape research. Throughout this journey I have never stopped marvelling at parents' capacity and commitment to their children and their families, and at their resilience in the face of adversity. The daily treatment regimen parents must follow

alone is profound. Such commitment deserves respect. It is this role modelling that has equally guided my own personal commitment as a researcher to the biopsychosocial model of clinical care from a salutogenic approach.

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Appendix A

Invitational Recruitment Letter – Princess Margaret Hospital



Government of **Western Australia**
 Department of **Health**
 Child and Adolescent Health Service



Date Name

Address

Dear Parent/s

I am writing to ask if you would be willing to be contacted by a researcher from The Telethon Kids Institute who is undertaking a project called 'The AREST-CF Early Surveillance Program for infants and children with cystic fibrosis (CF): What do parents experience and how do they cope?' and wishes to recruit suitable parents from this Hospital. Parents from this Hospital are needed for this study and from our records you would appear to be a potential participant.

Expected benefits to the participant and community

Potential benefits of taking part in this study to participants include:

- Participants may experience the satisfaction of participating in research that may benefit themselves, future patients and families
- Participants will be able to be involved as consumer advocates for people with CF, and their families
- Participants may benefit from the reflection that follows answering the types of questions and methods used in this study

Potential benefits of this study to the national CF community:

- May enhance school and socio-emotional experiences and development within the CF community (nationally and internationally)
 - May lead to a larger study that will generate support for all children living with cystic fibrosis, which may improve school and socio-emotional outcomes

The aim of this study is to explore parents' perspectives of what it is like to parent children who undergo early surveillance of CF. We want to help parents by supporting them and their families. This is very important as it may mean we can improve mental health and well-being for children with CF and families by supporting them in the early years of children's lives.

The people the researchers are looking for to help them with this study need to be:

- The parents or guardians (primary carers) of children aged up to years with a documented diagnosis of CF (positive sweat chloride >60 mEq/L) and/or a genotype with two identifiable mutations consistent with CF accompanied by one or more clinical features with the CF phenotype; ("parents" in this context means any person/s defined by the family itself as the primary caregivers for the child/ren)
 - Parents of children with CF undergoing early surveillance with AREST CF

If you do not wish to hear more about this study, or be contacted further, please complete the attached slip and return it in the envelope provided. If we do not hear from you within 3 weeks we will assume you are willing to be contacted and the researcher, Cindy Branch-Smith, will contact you shortly after to give you more information regarding the study. Should you wish to have further information about the study before making a decision as to whether or not you wish to be contacted please telephone the researcher (Cindy Branch-Smith on 08 94897601).

Whether or not you participate in this project, any future care you or your child receives at this Hospital will not be affected in any way.

This study has been approved by the Princess Margaret Hospitals Ethics Committee, Edith Cowan University Ethics Committee, Royal Children's Hospital Human Research Ethics Committee, and the confidentiality of all participants is assured.

Thank you for considering this

request. Yours sincerely

Doctor Tonia Douglas

A handwritten signature in cursive script that reads "Tonia Douglas".

CF Centre Director

Appendix B

Information Statement and Consent Form – Princess Margaret Hospital



Information statement

The AREST-CF Early Surveillance Program for infants and children with cystic fibrosis: What do parents experience and how do they cope?

Why are we doing the study?

Having a child with cystic fibrosis (CF) can be stressful for families and involve a lot of changes to lifestyle and family life. It can affect the health of family members too, especially emotional and mental health. Often the sicker the child is, the worse the impact of CF may be for families.

What we don't know is whether how a family works, the emotional health of parents, their parenting style and the social circumstances of families affects how parents experience the early surveillance program or how families cope with their child being involved with the early surveillance program. In other words, do certain factors in the social and family background lead to healthy or unhealthy coping styles and strategies and do certain factors in the social and family background have an effect on how parents experience the early surveillance program?

This is very important as it will mean we can promote mental health and well-being of parents and children by better supporting families and helping them make changes to the family setting that promote better health.

The aim of this study is explore and understand caregivers' coping and other experiences with being involved in the early surveillance program, and to understand if coping styles and strategies, and other experiences, are associated with specific domains within family and social systems.

Who is carrying out the study?

This study is being conducted by The Australian Respiratory Early Surveillance Team for Cystic Fibrosis (www.arestcf.org) at Princess Margaret Hospital for Children in Perth.

Do you have to take part?

No, your participation is purely voluntary, and if you choose not to take part that's just fine and you will not experience any consequences of your decision. Your decision to participate will not affect your child's care in any way. You may also choose to withdraw your consent at any time during the study.

What will you be asked to do if you decide to take part in this study?

Your child will need to do nothing extra if you participate in this program.

We will ask the parents to participate in an interview lasting between 1 and 1 ½ hours in length. During the interview, parents will be asked about their experiences with each of the clinical aspects of the program, including relinquishing your child for anaesthesia and obtaining annual results. Particular coping strategies that parents employ for each aspect of the program will also be established.

The interview will be carried out in hospital, at your home or another convenient place and we will make sure this happens at a convenient time for you if you choose to participate.

Is there likely to be a benefit to my child or family?

There may be no direct benefit to your child or your family by participating in this study, however, it is our experience that parents find thinking about their family, their children and how they parent quite a positive experience.

Is there likely to be a benefit to other people in the future?

This study looks at important questions for patients with CF and their families that we have yet to answer very well and will improve our understanding of the role of family and social factors in how families cope with being involved the program. This will help us to provide better all-round care for patients and families in the future.

What are the possible risks and/or side effects?

There are no anticipated side effects by taking part in this study.

What are the possible discomforts and/or inconveniences?

You may experience some inconvenience related to the time spent answering the interview questions as described above.

Where is your information kept?

The information we collect will be kept here at the hospital in a locked filing cabinet, held in a locked office. Information will be entered into a database that is password protected and accessed by the research team only.

What about my privacy?

The information we collect will be recorded in a separate file and kept strictly private. The study researchers will not know your name, as each parent will be given a codename. This further protects your family's privacy.

Who has approved the study?

This study has been approved by the Ethics Committees at Princess Margaret Hospital, Edith Cowan University and James Cook University (Queensland).

Who do I contact for more information about this study?

If you would like any more information about this study, please do not hesitate to contact one of the research team. They are very happy to answer your questions.

<i>Name</i>	<i>Title</i>	<i>Contact</i>
Cindy Branch-Smith	Principal investigator	9489 7818

Parenting Children with Cystic Fibrosis

Professor Julie Ann Pooley	Primary supervisor	6304 5591
Dr Tonia Douglas	Co-supervisor	9340 8830

Who to contact if you have any concerns about the organisation or running of the study.

If you have any concerns or complaints regarding this study, you can contact the Director of Medical Services at PMH (Telephone No: (08) 9340 8222). Your concerns will be drawn to the attention of the Ethics Committee who is monitoring the study.

What to do next if you would like to take part in this research:

If you would like to take part in this research study, please read and sign the consent form provided.

THANK YOU FOR YOUR TIME



Appendix C

Information Statement and Consent Form – Royal Children’s Hospital



Information statement and consent form

The AREST-CF Early Surveillance Program for infants and children with cystic fibrosis: What do parents experience and how do they cope?

Why are we doing the study?

Having a child with cystic fibrosis (CF) can be stressful for families and involve a lot of changes to lifestyle and family life. It can affect the health of family members too, especially emotional and mental health. Often the sicker the child is, the worse the impact of CF may be for families.

What we don't know is whether how a family works, the emotional health of parents, their parenting style and the social circumstances of families affects how parents experience the early surveillance program or how families cope with their child being involved with the early surveillance program. In other words, do certain factors in the social and family background lead to healthy or unhealthy coping styles and strategies and do certain factors in the social and family background have an effect on how parents experience the early surveillance program?

This is very important as it will mean we can promote mental health and well-being of parents and children by better supporting families and helping them make changes to the family setting that promote better health.

The aim of this study is explore and understand caregivers' coping and other experiences with being involved in the early surveillance program, and to understand if coping styles and strategies, and other experiences, are associated with specific domains within family and social systems.

Who is carrying out the study?

This study is being conducted by The Australian Respiratory Early Surveillance Team for Cystic Fibrosis (www.arestcf.org) at The Royal Children's Hospital and Princess Margaret Hospital for Children in Perth.

Do you have to take part?

No, your participation is purely voluntary, and if you choose not to take part that's just fine and you will not experience any consequences of your decision. Your decision to participate will not affect your child's care in any way. You may also choose to withdraw your consent at any time during the study.

What will you be asked to do if you decide to take part in this study?

Your child will need to do nothing extra if you participate in this study.

We will ask you to participate in an interview lasting approximately 1 hour in length. During the interview, you will be asked about your experiences with each of the clinical aspects of the program, including relinquishing your child for anaesthesia and obtaining annual results. Particular coping strategies that parents employ for each aspect of the program will also be established.

The interview can be carried out in hospital when you are visiting or staying, at your home or another convenient place, or by TeleHealth (telephone video conferencing using secure internet connection) or by telephone. We will make sure this happens at a convenient time for you if you choose to participate.

If you do not wish to participate, please complete the opt-out form included with this letter and return in the reply-paid envelope within 10 days. If you do not return the form, the principal investigator will contact you about your interest in the study.

Is there likely to be a benefit to my child or family?

There may be no direct benefit to your child or your family by participating in this study, however, it is our experience that parents find thinking about their family, their children and how they parent quite a positive experience.

Is there likely to be a benefit to other people in the future?

This study looks at important questions for patients with CF and their families that we have yet to answer very well and will improve our understanding of the role of family and social factors in how families cope with being involved the program. This will help us to provide better all-round care for patients and families in the future.

What are the possible discomforts and/or inconveniences?

You may experience some inconvenience related to the time spent answering the interview questions as described above. If you experience any level of distress as a result of discussing your experiences, you can be referred to the CF psychologist or CF clinic co-ordinator at the Royal Children's Hospital or an independent counselling service.

What are the possible risks and/or side effects?

There are no anticipated side effects by taking part in this study.

Where is your information kept?

The interviews will be audio-recorded and then destroyed once they have been typed into a Word document. The documents will be assigned a code so that you cannot be identified. A copy of the transcribed interview will be kept at RCH and a copy sent to the Telethon Institute for Child Health Research in Perth where the principal investigator will complete the analysis of the interviews. The documents at both sites will be kept in a locked filing cabinet, held in a locked office. Information will be entered into a Web application called SharePoint that is password protected and accessed by the research team only.

What about my privacy?

The information we collect will be recorded in a separate file and kept strictly private. The study researchers will not know your name, as each parent will be given a codename. This further protects your family's privacy.

Who has approved the study?

This study is a joint research project between the Royal Children's Hospital and Princess Margaret Hospital in Perth and has been approved by their respective Human Research Ethics Committees. This study is a PhD collaboration at Edith Cowan University (Perth) and James Cook University (Queensland) and has also been approved by their respective Human Research Ethics Committees.

Who do I contact for more information about this study?

If you would like any more information about this study, please do not hesitate to contact one of the research team. They are very happy to answer your questions.

<i>Name</i>	<i>Title</i>	<i>Contact</i>
Cindy Branch-Smith	Principal investigator	0416 580 255
Associate Professor John Massie	Associate Investigator	(03) 9345 5818
Judith Glazner	Associate Investigator	(03) 9345 5840

Who to contact if you have any concerns about the organisation or running of the study.

If you have any concerns and/or complaints about the project, the way it is being conducted or your rights as a research participant, and would like to speak to someone independent of the project, please contact:

Director, Research Development & Ethics, The Royal Children's Hospital Melbourne on telephone: (03) 9345 5044.

What to do next if you would like to take part in this research:

If you would like to take part in this research study, you don't need to do anything now. A member from the project team will contact you by phone to organise a convenient interview time and location, and to answer any questions you may have. The consent form on the next page will be collected at the meeting time prior to the start of the interview.

THANK YOU FOR YOUR TIME

CONSENT FORM

Research Project Title: The AREST-CF Early Surveillance Program for infants and children with cystic fibrosis: What do parents experience and how do they cope?

Version Number: 1 **Version Date:** 01/08/13

- I have read, or had read to me in my first language, the information statement version listed above and I understand its contents.
- I believe I understand the purpose, extent and possible risks of my involvement in this project.
- I voluntarily consent to take part in this research project.
- I have had an opportunity to ask questions and I am satisfied with the answers I have received.
- I understand that this project has been approved by The Royal Children's Hospital Melbourne Human Research Ethics Committee and will be carried out in line with the National Statement on Ethical Conduct in Human Research (2007).
- I understand I will receive a copy of this Information Statement and Consent Form

 Participant Name

 Participant Signature

 Date

Declaration by researcher: I have supplied an Information Letter and Consent Form to the participant who has signed above, and believe that they understand the purpose, extent and possible risks of their involvement in this project.

 Research Team Member Name

 Research Team Member Signature

 Date

Note: All parties signing the Consent Form must date their own signature.

Appendix D

Western Australian Counselling Services

Cystic Fibrosis Western Australia	(08) 9346 7333
Lifeline	13 11 14
Family Helpline	(08) 9223 1100
Parenting WA Helpline	1800 654 432

Appendix E

Victorian Counselling Services

Cystic Fibrosis Victoria	(03) 9686 1811
Lifeline	13 11 14
Lifeworks	1300 543 396
Parentline Victoria	13 22 89

Appendix F

Personal Reflections

Parents have not had a healthy child to begin with (he spent his first few months in hospital) but perceive him to be very healthy currently. Maybe their perception is based on the fact that they started with an unhealthy child. They do not perceive the program as positive at all, but as a necessity to the longevity of their child's life. As XXA is only 2, they're still learning about the program. These parents (not just the interviewees) must be so resilient, not only for their experiences, but to talk so openly about them.

Mum and Dad were willing to share their experiences openly. They were happy with their AREST CF involvement. Maybe, mainly because XXA has been so well. It did seem that that may be the case. I have expressed my thoughts previously about how I think parents' perceptions/experiences may be influenced by the health of the child.

Both parents were very open about their experiences and perceptions. Dad seemed like he felt that if he had a better 'measure' of XXA's disease severity, he would 'deal with it' better. Mum got quite emotional when discussing her views of the PMH staff, specifically the nurses. This was not a negative way, Mum was very grateful of the PMH staff. I wonder why you would choose to have a CF child when you find out of their condition in-utero. In this interview, I got the first sense of the evolution of coping. I identified some themes that have previously been discussed by other parents, that is, distraction and viewing the illness as not as bad as others. A new theme that came out was 'denial.'

Parent came across as the most confident and comfortable that I have interviewed in relation to her involvement in the program and in terms of her ability to cope with the procedures and processes. Her eldest son has CF and she agrees that has assisted her to deal with XXA's CF in a more effective way. She expressed how calm she is with the program and mentioned her belief in God as a major factor in her ability to stay calm.

Parent felt much more comfortable with her involvement in the program (i. e., her experience) over time, mainly due to knowledge and understanding of the procedures and processes that help to keep her child healthy. Healthy child and found out at 4 months old-cancer diagnosis analogy? Does this have any influence over tone/frame of mind of family life?

Appendix G

Interview Schedule

Before we start, I would like to thank you for your participation in the research project. Your comments and participation are greatly appreciated.

The purpose of this interview is to understand what you think, from your perspective, about your experiences with having a child with cystic fibrosis in the AREST-CF early surveillance program.

Can you tell me a story about caring for your child with CF?

Can you tell me a story about your child?

How did that make you feel?

Have there been similar experiences?

Have there been different experiences that made you feel the same way?

Can you tell me what it is like for you, and your family, to be involved with AREST CF?

What do you think it is like for your child to be involved with AREST CF?

Have you learnt anything about yourself from being involved with AREST CF?

If so, what have you learnt?

Is there anything you would like to add that you think I would like to know?

Examples of probes

What was that like for you?

Why do you think that was the case?

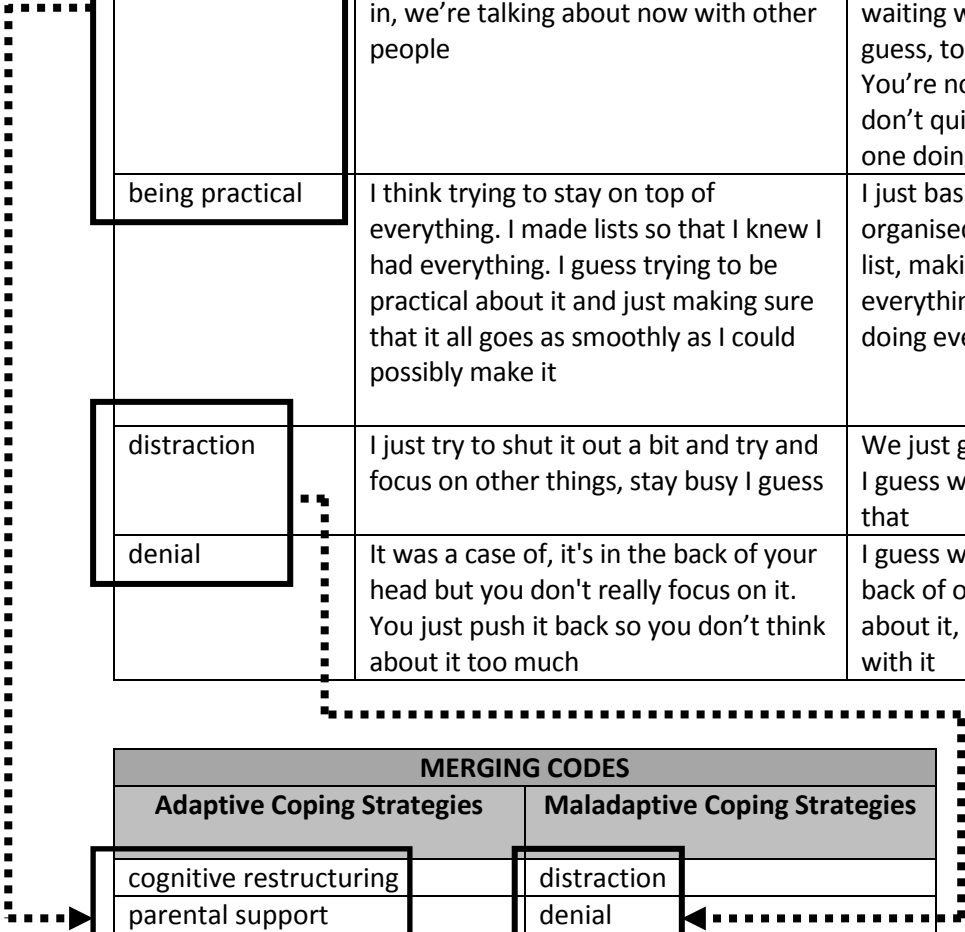
Can you give me an example/do you have further examples of this?

Appendix H

Initial Coding of Interview Transcripts

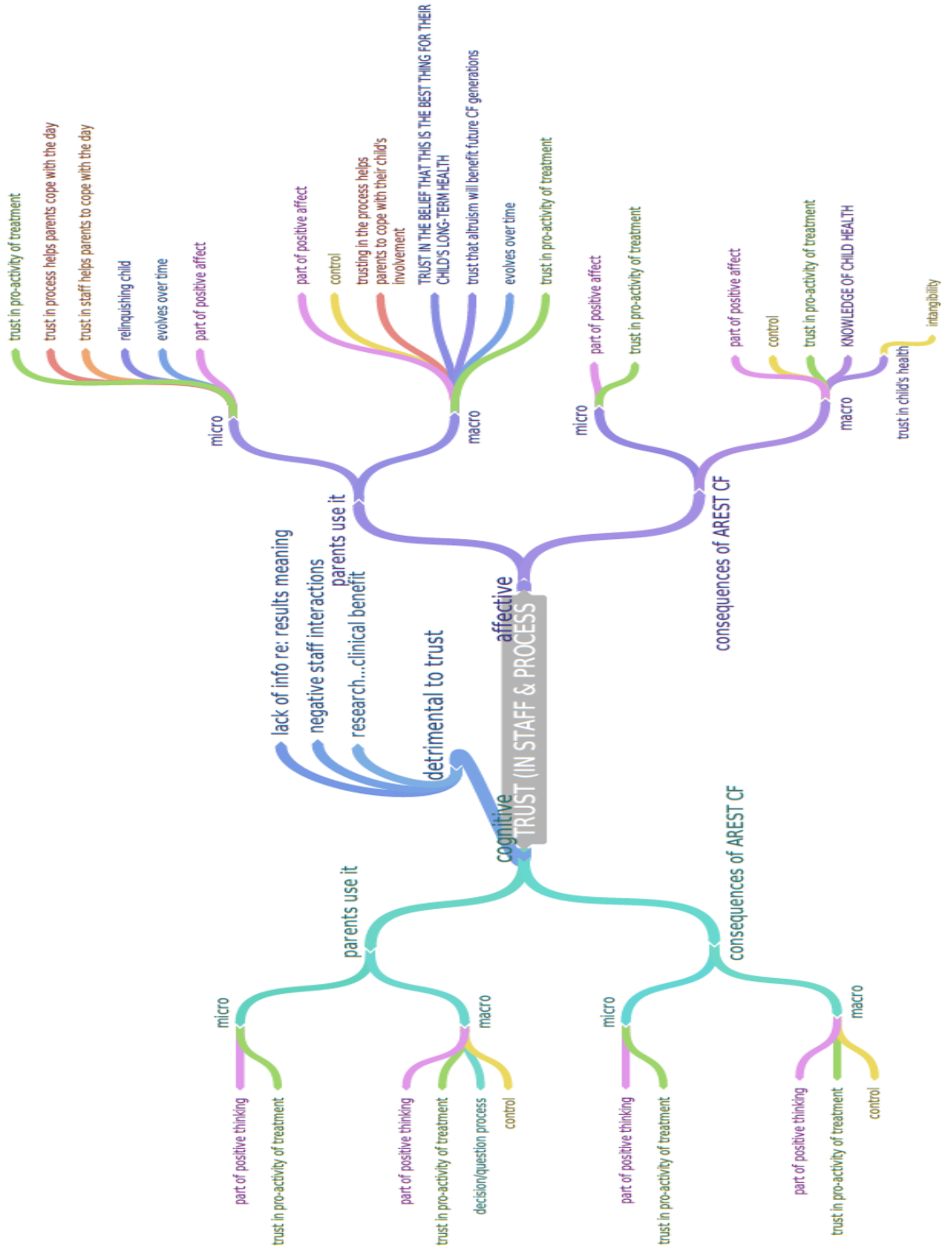
CODE	TRANSCRIPT	
cognitive restructuring	when you see some of those kids that are so physically or mentally disturbed, CF seems quite mundane	I kind-of normalise it and think it's the same, maybe not exactly the same for every family, but every family has issues and problems
parental support	I think we've started to let more people in, we're talking about now with other people	I talk to other parents that are there waiting with you, it helps you along I guess, to have someone to talk to. You're not totally on your own, you don't quite feel like you're the only one doing it, there is support around
being practical	I think trying to stay on top of everything. I made lists so that I knew I had everything. I guess trying to be practical about it and just making sure that it all goes as smoothly as I could possibly make it	I just basically tried to get as organised as I could so I just made a list, making sure that we had everything, making sure that I was doing everything
distraction	I just try to shut it out a bit and try and focus on other things, stay busy I guess	We just got on with our usual routine, I guess we preoccupy ourselves with that
denial	It was a case of, it's in the back of your head but you don't really focus on it. You just push it back so you don't think about it too much	I guess we kind-of almost put it to the back of our minds so we don't think about it, so we don't have to deal with it

MERGING CODES	
Adaptive Coping Strategies	Maladaptive Coping Strategies
cognitive restructuring	distraction
parental support	denial
being practical	



Appendix I

Thematic Mind-Map - Trust



Appendix J

Thematic mind-map – Cognitive dissonance

