

A qualitative exploration of adolescents with severe haemophilia and their caretakers regarding their future transition to adult services

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RCSI

ROYAL COLLEGE OF SURGEONS IN IRELAND

COLÁISTE RÍOGA NA MÁINLEÁ IN ÉIRINN

**A qualitative exploration of adolescents with severe
haemophilia and their caretakers regarding their future
transition to adult services.**

Volume 1 of 1

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LIST OF ABBREVIATIONS

CIT:	Critical Incident Technique
CNSp:	Clinical Nurse Specialist
CVC:	Central Venous Catheter
DoH:	Department of Health
HSE:	Health Service Executive
IHS:	Irish Haemophilia Society
MDT:	Multidisciplinary Team
PWH:	Person with Haemophilia
WFH:	World Federation of Haemophilia
WHO:	World Health Organization.

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Summary.

Title: A qualitative exploration of adolescents with severe haemophilia and their caretakers regarding their future transition to adult services.

Objective: To qualitatively explore the views of adolescents with severe haemophilia A or haemophilia B and their caretakers such as parents and multidisciplinary staff relating to transition from child to adult services.

Methods: Qualitative design using semi-structured interviews based upon the Critical Incident Technique (CIT).

Results: Thematic analysis using the CIT approach identified five themes all of which pertained to either the positive or negative aspects of the transition programme for adolescents with severe haemophilia and their parents and MDT. The main issues that emerged in the themes were the change in independence, meeting peers, being prepared for inevitable change, apprehension and communication levels.

Conclusions: The findings from this study are tentative and more research is required on this topic. The aim of this study is to offer initial evidence that can be used to positively affect change in current practice

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Dedication

Mr John C. Bradley, my hero.

Chapter 1: Introduction

1. Introduction

Before the introduction of advanced clotting factor replacement, the average life expectancy of a boy with severe haemophilia was just 16 years of age (Nordic Hemophilia Council 2015). However, Abali et al (2014) have suggested that due to successful prophylactic (also referred to as preventative) treatments over the last ten years; mortality in haemophilia has been decreasing.

In contemporary healthcare it may be possible to offer children with haemophilia a near normal life (Fischer et al 2017). Nowadays, children born with haemophilia and receiving proper treatment can look forward to a normal life expectancy (World Federation of Hemophilia 2018). These developments follow the introduction of much improved factor replacement products, resulting in haemophilia no longer being considered a disease of childhood (Canaro et al 2015).

Regardless of these findings, there have been limited studies exploring the transition of care from child to adult services for those with haemophilia (Chaplin 2016). Inadequate transition from child to adult services has been associated with a greater risk of non-compliance to treatment and therefore increased morbidity and mortality (Moynihan et al 2015). According to Chan et al (2011) adherence to the recommended treatment is an essential component to successful management of haemophilia. This provided much of the rationale for the student undertaking this study, exploring the views of adolescents with severe haemophilia in Ireland and their caretakers pertaining to their future transition to adult care. This research aims to be at the forefront of the future formation of a structured transitional care programme for those with severe haemophilia attending the children's hospital and the adult hospital.

This introductory chapter will explore the background to haemophilia, its prevalence in the population, treatment modalities, as well as how administration of contaminated blood products lead to the change of treatment both nationally and internationally. This chapter also introduces literature focusing on transitional care, and due to the paucity of available studies within the context of haemophilia, it also includes documents relating to transition in other chronic conditions. The long-term impact on the patient should their transition be deemed unsuccessful are further explored. In conclusion, this chapter highlights the need for the completion of this study considering the planned co- location of the national children's and adult haemophilia services to the one centre in the near future.

1.1 Background.

1.1.1 An overview of Haemophilia.

Haemophilia translates as love (philia) of blood (haemo) and is associated with prolonged and excessive bleeding (Fijnvandratt et al 2012). It is an incurable condition which will require treatment throughout the person's life. It is a mostly inherited blood disorder resulting in one of the clotting factors in the blood being absent or present in a reduced amount. It is an X –chromosome linked recessive disorder passed from a carrier mother to son in the majority of cases. However, about 30% of cases have no family history of haemophilia and occur following a genetic mutation (Kulkarni & Soucie 2011). Because of this mode of inheritance, haemophilia mostly affects the males with females being carriers. While females are usually asymptomatic carriers, they may also be symptomatic carriers with low factor FVIII or FIX levels (Di Michele et al 2014). There are no females with haemophilia in this study cohort

The general term haemophilia describes the two types of the condition. Haemophilia A is the deficiency of Factor VIII clotting

factor in the blood and haemophilia B is a deficiency of Factor IX clotting factor in the blood. Haemophilia A occurs more frequently than haemophilia B with 80-85% of people with haemophilia (PWH) having haemophilia A (World Federation of Hemophilia 2012). Both types share the same symptoms and inheritance patterns (The Haemophilia Society 2017). Haemophilia is an x-linked recessive disorder with the incidence of haemophilia A of 1:5,000 males and the incidence of haemophilia B of 1: 30,000 males (Lee et al 2006). Haemophilia occurs worldwide and is present in all racial groups (Kulkarni & Soucie 2011).

The lack of clotting factor causes people with haemophilia to bleed for longer, but not faster than those with normal clotting factor levels (Irish Haemophilia Society 2016). The severity of the person with haemophilia (PWH)'s condition depends on the amount of normal clotting factor present in their blood. In Table 1 Srivastava et al (2012) detail how the classification of severity of haemophilia is shown in blood plasma levels and its corresponding percentage range. They also show the bleeding episodes experienced for each severity.

Table 1: Relationship of bleeding severity with clotting factor level (from Srivastava et al 2012)

Severity	Clotting factor level	Bleeding episodes
Severe	$<1 \text{ IU dL}^{-1}$ ($<0.01 \text{ IU mL}^{-1}$) or $<1\%$ of normal	Spontaneous bleeding into joints or muscles, predominantly in the absence of identifiable haemostatic challenge.
Moderate	$1\text{--}5 \text{ IU dL}^{-1}$ ($0.01\text{--}0.05 \text{ IU mL}^{-1}$) or $1\text{--}5\%$ of normal	Occasional spontaneous bleeding; prolonged bleeding with minor trauma or surgery.
Mild	$5\text{--}40 \text{ IU dL}^{-1}$ ($0.05\text{--}0.40 \text{ IU mL}^{-1}$) or 5 to $<40\%$ of normal	Severe bleeding with major trauma or surgery. Spontaneous bleeding is rare.

The hallmark of a person with severe haemophilia is defined by Blanchette et al (2004) as someone with a normal factor activity level of less than one percent and has recurrent bleeds into joints and muscles from an early age. People with severe haemophilia will bleed after an injury but may also bleed spontaneously, most frequently into the muscles or joints. Spontaneous bleeding relates to a bleeding episode which has no relatable cause. People with moderate haemophilia bleed less frequently, for example only following an injury; however, they may also experience occasional spontaneous bleeding episodes. People with mild haemophilia tend to bleed infrequently and are sometimes not diagnosed until after surgery or injury occurs (Ljunc & Getenkort-Andersson 2015).

1.1.2 Historical References to Haemophilia.

Haemophilia was first mentioned as far back as in the second century AD in Jewish rabbinical writings. It was suggested that circumcisions should not take place if two of the infant's brothers died following excessive bleeding as the result of the procedure (Franchini & Mannacci 2012). These authors also state that the first use of the term Haemophilia was by Hopptt from the University of Zurich in 1828. Haemophilia was present through the European Royal families during the 19th and 20th centuries (Christopher & Chaithanya 2015). It was detailed by Castro et al (2012) that the subsequent marriages and children of Queen Elizabeth the first's affected daughters resulted in haemophilia B being present in the royal families of Spain, Germany and, as most frequently referenced; Russia.

1.1.2.1 Historical Treatment of Haemophilia 1930s- 1960s.

It has been stated by Ingran (1997) that the 1930's were the beginning of an extraordinary transformation in haemophilia

knowledge and treatment. They reported that this was when increased research and laboratory investigations began to demonstrate new evidence that enhanced the understanding of the processes of haemophilia. However, it was reported by Wong & Recht (2011) that prior to the 1940s, people with haemophilia were treated with supportive care and by the transfusion of whole blood or fresh plasma. Neither of these products contained enough clotting factor to be fully effective. As a result, PWH continued to experience major bleeds resulting in long term sequela. The first use of plasma as treatment for haemophilia occurred in the 1940s, followed by plasma concentrate in the 1950s, and this was followed by the use of cryoprecipitate in the 1960s (Kaufman & Powell 2013). Cryoprecipitate is prepared by thawing fresh frozen plasma, the precipitate of which is known to contain high levels of Factor VIII clotting factor (Sharma, Sharma & Tyler 2011). The introduction of commercial freeze-dried factor concentrate in 1968 greatly improved the quality of life for those with haemophilia by allowing home treatment to become possible (Kaufman & Powell 2013). This permitted PWH to continue their education, maintain employment and travel.

1.1.2.2 Contaminated blood products 1970s.

While the introduction of factor concentrates in the 1970s heralded a huge improvement in haemophilia care, it also resulted in the contraction of Hepatitis C Virus (HCV) and Human Immuno-deficiency Virus (HIV) by a high percentage of those who used it. It was explained by Lassila & Makris (2016) that pooled plasma concentrates used by PWH were made up from 20,000- 30,000 individual donations. The testing of donations for viruses at this time was limited to testing for Hepatitis B. However once testing for HCV and HIV became available it was evident that PWH were affected (Lindvall et al 2006). HCV and HIV acquired from factor concentrate was a grave concern in the 1970s and early 1980s (Zoulim & Bailly 2012). Almost all PWH became infected with HCV (Franchini &

Mannacci 2012). Approximately half of PWH in the USA eventually became infected with HIV and thousands died (National Hemophilia Foundation 2018).

1.1.2.3 The use of contaminated blood products in Ireland and how it changed the treatment of people with haemophilia (PWH) here.

In response to the international findings showing HCV and HIV infection occurring following the use of commercial factor concentrate, the “Tribunal of Inquiry into the Infection with HIV and Hepatitis C of persons with Haemophilia and Related Matters” (Lindsay 2002) took place in Ireland. The tribunal became known as The Lindsay Tribunal, named after the Judge who oversaw it from 2000-2002. It addressed the findings that 268 PWH in the Republic of Ireland had been infected with HIV and/or HCV following the use of contaminated blood products (Taylor & Power 2016). These products had been supplied to the PWH by the Blood Transfusion Service Board. The tribunal found that 69 people were infected with both HIV and Hepatitis C (Lindsay 2002). The Tribunal heard that HIV- infected donations were present in the Irish Blood supply in 1984. Before 1985 the commercial factor concentrate used for haemophilia treatment was generally not heat treated which would have killed the HIV virus (Lindsay 2002). The HIV/Hepatitis C crisis in Ireland during the 1980s and 1990s was the largest health scandal since the Thalidomide controversy in the 1960s (Taylor & Power 2016).

1.1.2.4 The Recommendations from the Lindsay Report (2002).

The recommendations in the report include:

- The formation of the National Haemophilia Council with the objective to make recommendations to the Minister of Health on all aspects of haemophilia care in Ireland. Its members include some of those who treat haemophilia as

well as members of The Irish Haemophilia Society. The Irish Haemophilia Society was formed in 1968 by members of the medical profession and PWH and their families to provide support and guidance to its members (Irish Haemophilia Society 2016).

- A national treatment protocol was introduced. This means that the standard care for all PWH is the same nationally. People with Haemophilia A are on one standard treatment and people with Haemophilia B are on another standard treatment.
- Medical teams working in the Haemophilia service throughout Ireland must be in regular contact.
- National and regional centres are subject to external audits biannually.
- The provision of services such as home nursing services for the terminally ill, hospice care, treatment abroad and home help services were made available to those affected.
- Anyone tested for a transfusion transmitted disease would be told beforehand and informed of the result in a timely manner. Accurate records should be taken and filed.
- In line with “The Hepatitis C Compensation Tribunal (Amendment) Act” (Department of Health 2002) financial compensation was provided.

All the recommendations from the Lindsay Tribunal affected how future generations of people with haemophilia were cared for in Ireland.

1.2. Current Treatment in Ireland

The World Federation of Haemophilia annual survey 2015 (2016) shows that there are now 839 people with haemophilia in Ireland. Six hundred and one (601) have Haemophilia A with nine percent (n: 54) between the ages of 14 & 18 years old. 238 have

Haemophilia B with eight percent (n: 19) between the ages of 14 & 18 years old. The primary aim of care for those with haemophilia is to prevent and treat bleeding by replacing the deficient clotting factor (World Federation of Hemophilia 2012). All the children with severe haemophilia in the student's clinical area obtain regular preventative treatments. This is the recommended treatment for all children with severe haemophilia (Schramm 2014). In line with a recommendation of the Lindsay Report (Department of Health 2002), each child should be reviewed by The National Paediatric Haemophilia team every six months.

1.2.1 Transitional Care

Transitional care describes a service which aims to ease a move from paediatric to adult services (Campbell et al 2016). It can be explained as encompassing the cognitive and social developments of the child with haemophilia (Young 2012). Transition from child to adult services has long been recognised as challenging Kelly (2014) and Brand et al (2015). It is especially difficult as it occurs during a time when there are also many life changes occurring (Vaks et al 2016). Adolescents with haemophilia have similar expectations as those without chronic disease, so all interventions should aim for providing a good quality of life (Garcia-Dasi et al 2016). By engaging adolescents with haemophilia and their caretakers early in their transition from child to adult services it prepares them for this inevitable process (Croteau et al 2016). Transition programmes for adolescents with haemophilia are currently in use in many countries including the United Kingdom (Khair et al 2013) and the United States (Young 2012).

1.3 Significance of the study

There are several reasons why this study is of significance to the adolescents with severe haemophilia and their caretakers in Ireland. To begin with, it was emphasised by Schwartz et al (2011),

Simmons et al (2013), Watson et al (2011) and Crowley et al (2011) that despite the benefits of structured transitional care being mentioned in many reports, there are limited published guidelines or assessment tools available for practitioners to use. By obtaining an understanding of all the underlying issues pertaining to transition in this group, these will be incorporated into the development of a new transition care programme. This tool will be available for practitioners to use.

The World Health Organisation (W.H.O) outlined the potential for clinical research to positively influence widespread population health (W.H.O 2012). It was also suggested by Coyne et al (2016) that the research conducted within the clinical area can promote patient care. This clinical research is being completed with the primary goal of improving patient care within this study group.

Adolescence is a time when plans for self- management of health are established (Sawyer et al (2012) & Srivastava et al (2012)). Studies completed by Crowley et al (2011), Moynihan et al (2015), Grant & Pan (2011), Brooks et al (2017), Andiman (2011) and Pai & Ostendorf (2011) found that transition between paediatric and adult centres is often poorly managed for those with chronic disability and can result in adverse health consequences. Focusing on haemophilia, Schrijvers et al (2016) found that prophylaxis adherence is frequently high except from adolescence to age 40. It is advised by Berntorf et al (2012) that if haemophilia is not correctly managed it can lead to lifelong disabilities. Just one bleed can lead to irreversible damage to soft tissue, a joint or even the central nervous system (Schrijvers et al 2016). It was detailed by Chaudry et al (2013) that patients who transfer to an adult centre with a poor or no structured transfer programme are at greater risk of not following their centre's treatment plan. It was also found that successful transitioning is increasingly identified as being essential as the numbers of children with chronic disease reach adulthood (O'Sullivan- Oliveria et al 2014). These studies identify the need for

a study relating to this topic to be completed in Ireland. The justification for this study design focusing specifically on adolescents with severe haemophilia is that this group are at greatest risk at time of transition to the adult hospital. Were they to decrease their level of compliance to their prescribed prophylactic factor treatment, it could lead to irreparable changes.

In recent years researchers have been encouraged to include the views of the patients and public in their studies, yet there is little evidence detailing its impact on care (Brett et al 2014). This is referred to as patient and public involvement (PPI). Successful transition for those with chronic conditions requires close collaboration between the patients, their families and the multidisciplinary team (Johnson et al 2016). The student counts this as another reason why this study is needed and the rationale for including the staff, patients and parents in the study.

1.4 Study Design.

This study took place in a hospital in the Republic of Ireland where children with severe haemophilia are transitioned from there to an adult hospital to continue their treatment. It was found by Fergran et al (2014) that most research on transition is based on quantitative research methods, and by using qualitative methods it allows us to gain a more concrete knowledge of the challenge's young adults face during this process. Qualitative research can offer insight into a specific experience and can raise awareness and further understanding of this experience (Ingham-Broomfield 2014). A qualitative approach, according to Thomas & Magilvy (2011), will provide a deeper understanding of specific events which may not be possible to explore in a quantitative research study. It is further explained by Gagliardi et al (2014) that due to the complexities in the modern healthcare environment there are many circumstances where qualitative research is required to aid in the planning, evaluation and assessment of quality improvements for patients. A

descriptive qualitative designed study conducted using the Critical Incident Technique (Flanagan 1954) was used in this study. This technique aims to identify behaviors and situations that have a significant effect on an activity and determine the positive and negative consequences of those behaviors on the activity (Peña & Rajas 2014). In this study the student asked the participants to relay the positive and negative views they had relating to their experience of transition from child to adult services.

Quantitative research methodology was further discounted due to the very small numbers of people with haemophilia in Ireland, and within that number an even smaller amount which the writer would be able to gain access to. As this study involved assessing the views of adolescents and adults with haemophilia and their caretakers, the author deemed a qualitative research approach to be most suitable for use in this study.

It was emphasized by Tong et al (2007) that studies which are deemed as poorly designed with inadequate reporting can lead to the inappropriate application of qualitative research in decision making in health care. For this reason, the author constructed a detailed study to ensure that it was well designed, and that findings were appropriately reported.

1.5 Research Question.

The research question under investigation in this study was:

“What are the views of adolescents with severe haemophilia and their caretakers when asked about their future transition to an adult service?”

The aim of this research study was to obtain the views of adolescents with severe haemophilia and their caretakers relating to their future transition to adult services.

The study objectives were:

1. Examine the views of adolescents with severe haemophilia towards transitioning to adult services.
2. Examine the views of the parents and multidisciplinary team members regarding the future transition of adolescent patients with severe haemophilia to adult services.
3. Examine the views of a sample group of recently transitioned adults with severe haemophilia.
4. Contribute to the development of a structured transition care programme for adolescent haemophilia patients transitioning to adult services.

1.6 Conclusion.

This chapter provided a background to haemophilia and the historical references to it, including historical treatment of the disease. It also detailed the worldwide contamination of blood products used by PWH which resulted in loss of life globally from the 1970's. The chapter discussed how the contaminated blood scandal affected Ireland and outlined some of the results of the Lindsay Tribunal in 2002 which changed the treatment of PWH here. The current treatment of PWH in Ireland was mentioned, as was the current transition process from child to adult services. The significance, aims, and objectives of this study were established. The study design was then outlined. The next chapter in this thesis will provide a literature review on the phenomenon under investigation.

Chapter 2: Literature Review

2.Introduction

The transition to adulthood for adolescents with medical complexities has become a topic of significant interest to healthcare professionals over the last decade (Joly 2016 & van Staa & Sattoe 2014) and has received worldwide attention (Ishizaki, Higashino & Kaneko, 2016). This literature review explored the process of transitional care in relation to adolescents and young adults with chronic health conditions as they move from paediatric to adult orientated health care services. While the studies explored here consist of the transitional care practices for people with a multitude of chronic conditions, the primary focus of this review is concerned with the transitional care practices relating to those with haemophilia. As this review will show, there has been a limited amount of published evidence in this specific area from 2007-2017. Chaplin (2016) concurs that there is very little published literature on effective transition for those with haemophilia. However, by reviewing published material pertaining to transition in other specialities, an extensive understanding of the transition from paediatric to adult care process can be explored.

Transition is defined as: "The process or a period of changing from one state or condition to another". (Oxford English Dictionary 2017). The World Health Organisation (W.H.O) defines an adolescent as any person between the ages of 10 and 19 years old (W.H.O 2014). However, Maturo et al (2011) state that traditionally the adolescent period lasts from ages 13 to 19 years.

2.1 Transition in Healthcare.

Transition from adolescence to adulthood is complicated for all youth, but especially for those with a chronic illness (Annunziato et al 2013, Croteau et al 2015, Quon et al 2015, El Dakhakhny et al 2014, Park, Adams & Irwin 2011, Bingham et al 2015, Andiman 2011).

Lyons, Becker & Helgeson (2014) suggest that the terms transition and transfer are often used interchangeably but they do not denote the same event. Transition is a dynamic and structured process, which involves planning and preparation, whereas transfer is often seen as a once off event involving the handover of care from one team to another (Rajendran & Iyer 2016).

There are four general models of transition (Mc Donagh (2005).

These are:

1. Direct transition where the adolescent moves from paediatric to the adult service without communication between the two centres.
2. Sequential transition where the adolescent is seen in a specially designed clinic.
3. Developmental transition where the adolescent is educated in relation to their condition.
4. Professional transition where there is a formal transfer of care and expertise between the paediatric and adult centres.

While sequential, developmental and professional transitions would seem likely to result in a successful transition, direct transition without communication between the two centres is the most likely to result in an unsuccessful transition. Patients who are transferred to an adult centre with either a poor or non-existent transition programme are the most likely to be lost to follow-up (Chaudhry et al 2013).

2.1.1 The importance of successful transition.

An unsuccessful transition from paediatric to adult services can risk delays and disconnection with healthcare practices for the adolescent, resulting in adverse outcomes in their care and poorer long-term health (Brooke et al 2017, Grant & Pan 2011). Similarly, inadequate transition for adolescents with chronic illness has been associated with an increased risk of non-adherence to medical treatment, and increased morbidity and mortality (Moynihan et al

2015). Transition difficulties are genuine for adolescents and can result in severe consequences when services fail them (Murcott 2014). Vaks et al (2017) also suggest that paediatric to adult healthcare transition can be especially difficult for these adolescents, as it occurs at a time in their lives when they are already experiencing many changes such as in education and personal development. However, transitioning is important both medically and socially as it allows the adolescents to become more independent with their care (Rauen et al 2013). The aim of all transitional care programmes is to increase the long-term health of those with chronic conditions. A coordinated transfer from the paediatric service to the adult service is a vital component of high-quality health care, especially for those with chronic health conditions (Wisk et al 2015). It is critical that a transition care programme meets the needs for not only the adolescent but also the parents and healthcare providers for it to be deemed successful (Brand, Dunn & Kulkarni 2015 & Johnson et al 2015). Importantly, despite agreement about the importance of successful transitional care, there remains a paucity of evidence relating to what is best practice in transitional care for adolescents with chronic illness (Watson et al 2011).

2.1.2 Transitional care for chronically ill youth in Ireland.

Begley (2013) found the absence of either policies or protocols in the vast majority of services relating to transition in Ireland to be concerning. While reviewing the Health Service Executive (H.S.E) website the only transition programme found was one specific to epilepsy (H.S.E 2017). The *Stepping Up* programme (2013) is the most comprehensive Irish online resource available. This a generic programme providing information for children and adolescents with chronic health conditions pertaining to transition.

By exploring what the barriers and facilitators are to transition for adolescents with chronic health conditions, it is envisaged that it will

provide information which will add to the knowledge base of this topic from an Irish perspective.

2.2 Search Strategy

The literature identified for inclusion in the study was located primarily through electronic searches. The electronic databases searched were: Cumulative Index to Nursing and Allied Health Literature (CINAHL), Web of Science, Scopus, Cochrane Library, Google Scholar and Medical Literature Analysis and Retrieval On-Line (MEDLINE).

The key search concepts used either individually or in combination during the searches were:

“Haemophilia Transition” “Adolescent transition” “Transition from child to adult services” “Child chronic illness and transition” and “Transition programmes”. “haemophilia A” “hemophilia A” “haemophilia B” “hemophillia B” “Christmas Disease” or “Factor VIII” “Factor 8” “Factor FIV” “Factor 9”. “Transition clinic”. “Transition to adult care” “paediatric to adult care” “transitioning issues” “transition process” or “transition plan” “continuity of patient care” “paediatric blood coagulation disorders inherited”

The literature included in this review met the following criteria:

1. Published between 2010 and 2017.
2. Available in the English language.
3. Published in peer reviewed journals, specialised organisation publications or government reports.
4. Included information on at least one of the key words.
5. Articles available through accessible library resources or inter-library loans were included.
6. Full article accessible.

A manual search was then completed after scanning of reference lists from relevant articles. The ultimate aim of this review was to

obtain a representative sample of accessible healthcare literature on recent research pertaining to transition.

The initial search identified 487 documents. These were then classed as being relevant or not relevant to the review. Those which were deemed not relevant did not contain advantageous information for the review, examples included: articles pertaining to transition in foster care, those which were not specific to transition health care despite containing some of the key words in their abstract, or studies which were not detailed enough to review thoroughly. The remaining articles include papers on transition in healthcare for adolescents with chronic illness, literature reviews as well as studies completed on the transition process. There was a limited amount of empirical studies relating to transition available. Only the articles containing information on studies were then appraised. Finally, forty-four studies were included in this review (see figure 1 and appendix 1). Each study was critically examined looking for the purpose of the study, methodology used, and study findings, as well as strengths and limitations.

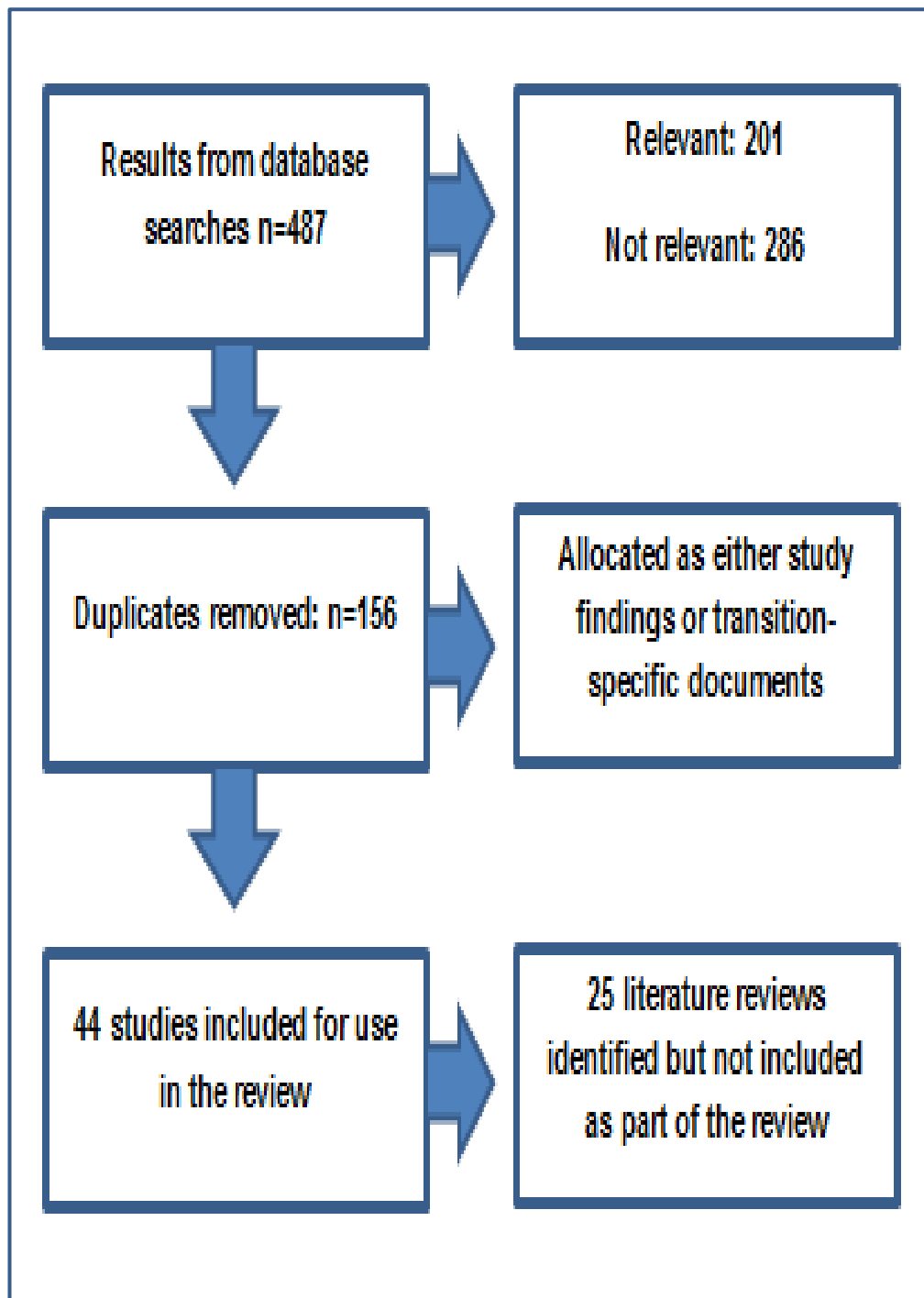


Figure 1: PRISMA Flow Chart

2.3. Themes emerging from the literature.

An integrative review focusing on transitioning adolescents and young adults with chronic diseases and or disabilities by Zhou, Roberts, Dhaliwal & Della (2016) identified many similar themes to the ones found by the student during her literature search. As a result, the studies being reviewed now will be grouped into several

categories similar to those identified by Zhou et al (2016). These are:

- Preparation for transition.
- Patient's outcomes post-transition.
- Barriers to the transition
- Facilitating factors to transition

2.3.1 Preparation for transition

Preparation, being the key to successful transitioning, was the main finding from a group of adolescents (n=9), stakeholders (n=21), health professionals (n=36) and parents (n=9) in a U.K. study pertaining to transitioning (Aldiss et al 2015). It is mentioned in this study that although the number of adolescents were small, it is imperative that their views were represented. Sawicki et al (2015) used a survey instrument and assessed the adolescents' experiences of health care transition preparation. The instrument was found to be reliable in measuring their preparation experiences in the three areas of transition: self-management, prescription medication and transfer planning. This study mentions as a limitation that the instrument does not include all the important aspects of health care transition in the quantitative study. Another limitation is that it does not contain an appropriate assessment tool for those with a cognitive or developmental delay. Another USA study, also by Sawicki, included youths (n=79) and parents (n=52) and used a questionnaire called the Transition Readiness Assessment Questionnaire (TRAQ) to obtain data (Sawicki et al 2014). The youth reported moderate readiness to transition, however one of the limitations of this study was that the outcomes were based on self-reporting and as such the transition readiness could not be validated by an external measure. Another limitation was the fact that the study took place in a single centre with a small sample of subjects.

A similar study assessing readiness for transition using the AM I ON TRAC adult questionnaire was completed by Moynihan et al (2015). The aim of the questionnaire was to assist nurses and other clinicians to identify what areas of care that the adolescent required assistance with prior to transition. This Canadian study recruited 201 youth through convenience sampling from a large paediatric hospital. The participants had not yet transitioned to adult services and were 12-19 years old. Once again, an exclusion criterion in this study were youth with developmental delay. The authors explain this is because it has been identified in the literature as a major obstacle to transition. However, when discussing the findings, they report that it is hospital policy for all patients to be transferred aged 18 to adult services, and that those who did not transfer at that stage might have some developmental delay or lack the knowledge or skills for adult care. This study had a very high participation rate of 93.5 %. The main finding was that 27% of 17-year olds, but 62% of 18-year olds were prepared for transition to adult services. This would suggest that there was a high percentage of people who did not transition despite being of adult age. There is no additional data in the study discussing what the outcome was for those who did not transition at 18 years of age. The authors also stated that they did not know if the completion of the AM I ON TRAC questionnaire would lead to improved transition outcomes. They recommended that further research should be completed on that score.

Simmons (2014) aimed to identify the information needs for children and teens with haemophilia in a USA study. Although not specific to transition, the study contains valuable information on this patient group who would have been transitioning in the near future. The purpose of this study was to determine what type of informational product was most suited to those who were due to transition to an adult centre. The authors compiled all the transition information available from documents, DVDs and webpages for example, and then asked adolescents and parents to attend a focus group to discuss these items. A total of 60 people took part in the online

focus groups, 40 adolescents and 20 parents. The two major themes that emerged were that adolescents were repeatedly being told they could not participate in sports although they had an interest in doing so, and that they were finding it difficult to explain haemophilia to different audiences such as peers or school personnel. This resulted in the development of two videos as educational tools for the adolescents relating to these issues. The limitations found in this study include the possibility of selection bias as participants were recruited through organisations that deal with people with haemophilia. Also, the fact that it was a qualitative study, results cannot be generalised to the larger population of youths with haemophilia. Another limitation that the student suggests is that although they were looking at items created for children aged 5 -12 years, this group was not involved in the study.

In a comprehensive study looking at one hundred and fourteen (114) parents of children with special needs in the USA aged between 12-18 years old in 2009-2010, the status of health care transition preparation was examined (Mc Manus et al 2013). In this study the authors explain that all children with special needs are scored on the transition score outcome scale. This scale has five questions to answer. The questions were:

1. Did the child receive anticipatory guidance on transition?
2. Have the doctors discussed transition with the adult provider?
3. Have doctors discussed further health care needs
4. Have doctors discussed insurance needs?
5. Does the caretaker report that the child has been encouraged to take responsibility for their own care?

Overall this study found that only 40% of the 4.5 million youths aged 12-18 years in the USA meet the national transitional outcome. While 78% increased their level of self-care, only 21% responded that their child discussed transition to an adult provider. The limitations mentioned with this study include that the results were reported by the parent and not the child, so this may under or

overestimate the true findings. It also does not allow for other factors such as health care provider characteristics, other transition support services or child-parent relationships which could influence the results.

Two studies assessing the perceptions of adolescents/young adults with chronic illness pre and post transition were reviewed (Rutishauser et al 2014 & While et al 2016). Using an anonymous questionnaire, Rutishauser et al (2014) compared 283 pre-transfer (participation rate 62%) and 89 post transfer (participation rate 54%) young people with chronic conditions in Switzerland. Their quantitative study found that there is some evidence that an older age at transfer results in a more successful transition and that anxiety and lack of information about transition were the most frequently reported barriers to successful transition. They found that barriers can be preventable or at the least, modified. The exclusion criteria in this study includes those with a cognitive disability but no specific rationale for this decision is included in the study. In the While et al study (2016) the sample group were 217 young people with cystic fibrosis, congenital heart defects or diabetes in Dublin, Ireland (participation rate 25%). Those with a cognitive condition were also excluded from this study as were those with current ill health, psychiatric conditions or family difficulties. No further information on this recruitment plan was given. The participants completed a questionnaire assessing their quality of life pre and post transition. One of their main findings was the importance of discouraging parental over protection as this negatively effects the quality of life of the participants. They also recommend that transition programmes should include strategies to promote self-advocacy skills. A limitation to this study was the low response.

Van Staa et al (2011 A) completed a cross-sectional study in a Dutch university hospital where adolescents aged 12-19 years old with chronic conditions participated in a web-based questionnaire (30% participation rate n=1087) examining their readiness for

transition to adult services, n=174 were excluded due to intellectual disabilities. In this hospital, the only transition program in use at the time was one for patients with haemophilia. The findings of this study show that 56% of the respondents perceived themselves as prepared for transition and that their general attitude towards transition was positive. A limitation to this study was the low response rate, which they suggest may be due to the participants being approached via an impersonal letter and were asked to access the questionnaire via the internet.

The views of health care providers regarding the transition process were examined in four studies explored in this section of the review: These studies were Begley (2013), Collins, Reiss & Saidi (2012), Ishizaki et al (2012) and Fair, Sullivan & Gatto (2015). Begley (2013) sent a postal questionnaire to physicians and nurse specialists (n=132) with n=71 responding (54%). The participants expressed dissatisfaction with the transition processes. Among the obstacles to successful transition were patient factors (56%) and developmental delay (18%). The limitations to this study relate to the unavailability of a database of health care professionals, so some relevant staff may not have been included. Therefore, some staff involved in the transition process may not have been contacted. Medical providers and social workers participated in the Fair, Sullivan & Gatto (2010) study examining the best practices when transitioning youth with HIV. A purposive snowball sampling method was used in the qualitative study, with 19 staff taking part. Many staff found that promoting independence and allowing the adolescents the chance to exert some control over their care was an important factor in transition. They mention as a limitation that while qualitative data analysis allows for a greater understanding of the issue at hand, it cannot be used to confirm a general relationship between the three factors. These factors were described as relating to patient, provider and the system.

Ishizaki et al (2012) conducted a survey in Japan between 41 paediatricians and 24 nurses relating to their perceptions on transition. Seventy eight percent (78%) of the paediatricians and 100% of the nurses reported that a transition programme was necessary. They found that the age of the patient, psychological development and social factors were important when determining their transition to adult services. They also found that healthcare professionals do not appropriately address any psychosocial problems and that a transition programme would assist with these issues. Collins, Reiss & Saidi (2012) explored the attitudes of paediatric hospitalists (staff) in a centre in the USA. The number of people involved in the quantitative study was 131 but no response rate was, so it is not known how many people were invited to participate. A 33-item questionnaire was employed. Eighty percent (80%) found that transitioning was a moderate or major problem for adolescents and only 1% were "quite prepared". The participants ranked lack of familiarity as being the biggest barrier to transition followed by lack of support from both paediatric and adult sectors. The main finding of this study was that transition programmes are vital, but that they are practically non-existent during the time this study took place. A limitation to this study is the small sample size and the authors suggest that a larger study should be completed to confirm these findings. The study states that it aimed to provide an overview of current attitudes to transition in paediatric hospitals, but that the perception from inpatients is that a better quality of care should be expected. They concluded that further studies and improved educational sessions should be developed.

A small study examining transition into adult healthcare for people with spina bifida in Scotland was examined by Levy (2014). The response rate was low at 12%, n=20. This study which used focus group methodology, found that people felt the paediatric hospital addressed the holistic needs of the child, but that the adult hospital is perceived as having less specialist knowledge and showed less respect towards the patient. They highlight the need for staff to be

trained in this area and outline the difficulties which may be encountered if the transition process is not successful. The authors mention though that it is possible that some of those who took part in the study were doing so as they were unsatisfied with the service provided. They also suggest that many people benefited from a successful transition from child to adult services.

McQuillan et al (2015) found in their Canadian study, that attending a transfer clinic was associated with improved adherence and renal function for a group of people who had received kidney transplants. The 32 patients were divided into those who had transitioned prior to the implementation of a transition clinic and those who had transitioned when the transition clinic existed. In the 1st year post transplant 18.8% of those who attended the transition clinic were not adherent in comparison with 62.5% in the 1st year for those who did not have a transition clinic. The limitations in this study include the fact that improvement in adherence may be due to the closer follow up post transfer rather than the transition clinic as well as that measures of non-adherence may be underestimated in both groups.

The student found while reviewing these articles, that the authors emphasised the need for further research to take place, and that the clinical guidelines relating to transition to be implemented. An overall dissatisfaction with the transition process was voiced by participants (Begley 2013).

2.3.2 Patient outcomes post transition.

The outcome of patients post transition from paediatric to adult services were mentioned in many of the studies included in this review and will be explored in more detail here. The nine studies focusing on the views of the young people will be examined first.

In a study in the Netherlands, van Staa & Sattoe (2015) followed up on 518 young people with chronic conditions who had transitioned from paediatric to adult services. Their web-based survey called *On Your Own Feet* which requested information on the experiences and satisfaction with the transition process had a response rate of 52%. Their study showed that 20% found their transition unsatisfactory, 50% felt prepared for transition and 24% had met the adult centre healthcare provider in advance. This research suggests that a successful transition programme should not be restricted to the paediatric centre and that all parties should be actively involved. The limitations of this study include the high non-response rate especially amongst males and those of non-Dutch ethnicity.

Hislop et al (2016) used a Q methodology approach in their UK based study to investigate all the possible views on the subject of transitioning from paediatric to adult centres for adolescents with chronic conditions. A purposive sample of 44 were approached with 41 completing the study. Participants had to be aged 14-22 years to be included. Those with an intellectual disability were excluded as it would be difficult for them to make hypothetical choices required as part of the methodology used. Four themes emerged in this study:

1. A laid back view of transition,
2. Anxiety about transition,
3. Wanting independence and autonomy during transition
4. Valuing social interaction to assist transition.

A key finding in this study was that young people do not have just one view on transition and that it is vital that transition programmes acknowledge this. Price et al (2011) also found similar results in their UK study relating to transition in diabetes care. The semi-structured interviews were completed with 11 people with 2 returning a year later for further interviews. The aims included an evaluation of the transition pathway in use. However, it resulted in generating many common themes in relation to transition with

adolescents including the need to recognise individuality in patients within the healthcare system. A similar finding was made by Gorter et al (2015), who designed a 4-year mixed method prospective cohort study assessing the use of transitional interventions to support youth with chronic illness. Fifty participants (47% response rate) with over 20 different conditions provided information on their transition experiences. The transition interventions were deliberately broad and holistic in their scope. They also found that goal setting is an important aspect of transition with 90% of participants setting transition goals. The limitations stated by the study include the fact that the mean age of the participants was 18 years old so that might account for their modest ratings in relation to transition supports as they may have yet to fully use them.

Chaudhry, Keaton & Nasr (2012) evaluated a transition programme for cystic fibrosis in the USA. In this quantitative study, 91 completed a 20-item questionnaire which included questions about patient satisfaction and feelings towards the different aspects of the paediatric and adult programmes. The findings showed that a well-planned transition programme might reduce the interruption of care for the patients. They found that some people did not recall discussing relevant items, such as meeting with their new doctor, or with their paediatric team prior to transition. The limitations to this study are that it was a self-reported retrospective study.

In a study by Oswald et al (2013) a survey was completed on 19-23-year olds who had transitioned. They found that only 21.5% of those included in the study had transitioned successfully to adult services. The study showed that those who received preventative healthcare were more likely to successfully transition to adult services than those who did not. It also showed a variation in health care approaches depending on the disability such as the specialised clinics available for cystic fibrosis or haemophilia but not for other disabilities. The main limitation to this study is that they

state a definition of a successful transition has not been independently validated so this may affect the findings.

In the second part of this section, the patient outcomes post transition as reported in studies exploring the views of staff, patients and parents will be examined.

Kime (2012) detailed the results of a 3-year multi centre UK study examining the views of young people with type 1 diabetes and their parents in relation to their transition to adult services. Over 300 people took part and Kime (2012) found that transition was a major area for concern with this group. They also found that none of the transition clinics included a member from the primary care team, this is despite the fact that the young person's G.P (General Practitioner) is often their first point of contact. Both the young people and the parents commented on how transition is a vital time in their lives and how greater emphasis should be placed on the primary care team assisting them through the process.

Strategies to encourage collaborations between primary and secondary care are required to provide optimal care for this patient group. A web-based survey by Sparud-Lundin et al (2017) on health care providers' attitudes towards transfer and transition in young persons with long term illness showed many areas requiring attention. Physicians and nurses were invited to participate. The authors had a thirty eight percent (38%) response rate (n=201).

One of the main findings was the importance of taking the professionals' perspective into consideration when developing a transition programme, as this will increase the possibility of it being implemented successfully. Seventy eight percent also found that the presence of a transition coordinator assists with transition. Most participants in this study did not consider primary care providers as active participants in the transition process. This study also disagreed that limited demand, as in few patients, was a barrier to organising a transition programme.

Another study examining the perspectives of patients and parents regarding transition (Chandra et al 2015) found that 23% had visited the emergency room between their last paediatric clinic visit and before their first scheduled adult clinic visit. This highlights the need for the transition programme to include an introduction to the adult centre before transition. There was an 88.5% response rate in this study (n=31). It also found that the repeated reminders sent out to the young adults were counterproductive in increasing their level of self-care and independence. Similarly, it was found in a qualitative study by Okumura, Saunders & Rehn (2015) that many young adults have been protected and continued to be treated as children, so are not prepared to function as independent adults once they have transitioned to adult services.

A qualitative approach using six focus groups of 10 young adults with chronic disease and 24 of their medical teams was used in a study by Huang et al (2011) exploring transition to adult care. The main themes identified by the patients which influenced transition were: parental involvement, communication difficulties between the paediatric and adult centres and difference in care models between the two. The themes identified by the medical teams were slightly different but did include reducing the involvement of the parents, the lack of standardised transition service protocols and the need for an overlapping period for the adolescents to attend both centres. The practitioners also found it vital that transition issues be addressed in a more direct fashion for it to be successful. The need for parents to slowly withdraw from disease management and allow their child to increase their independence was acknowledged by both parents and providers as being an important goal.

The parental perspectives of transition from paediatric to adult services for those with neurological disorders was examined in a study by Davies, Rennick & Majnemer (2011). A qualitative designed study with purposeful sampling was used. Seventeen parents of 11 young adults were recruited in the study. One of their

findings was the sense of abandonment felt by the parents during this process, which they also found very stressful. The parents reported that they could successfully navigate around the paediatric health care system, but only half the parents reported some level of confidence in navigating the adult service. Parents reported that they had received little preparation or guidance for the transition process and that the instability of the child's condition was not truly appreciated by staff. This study provides a greater understanding of parents' thoughts in relation to their child's transition and how it is necessary to include all relevant parties in transition programme design. The limitations in the study are that it took place in a single site and that further studies would be required to explore if parents' views change as the transition programmes develop.

Dogba et al (2014) evaluated a transition programme for those with osteogenesis imperfecta in Canada. They interviewed six patients, four parents and 15 staff in their study. In this centre, the patients can remain in the paediatric centre up to the age of 21 years to allow time for the patient to be assessed as ready for transfer. The strengths of this study were its strong approach based on partnership with parents and its model of increasing independence. The authors also declined to use direct quotes from the participants to maintain confidentiality. A limitation to the study was its small sample group and the fact that the interviewees knew each other, so this may have limited their willingness to identify any issues with the transition programme. The issues with the programme were identified using a SWOT analysis (strengths, weakness, opportunities and threats). The authors found that the staff in charge of the transition services had undergone numerous changes which resulted in a reduced focus on the transition service. They also stated that the transition needs of the patient varied according to their own motivation to attend the meetings.

A survey was developed to assess concepts identified as important for successful transition by Woodward et al (2011). This survey was

sent to 198 parents of youths and young adults with chronic diseases in the USA. A forty four percent response rate was obtained. Thirty-four percent of parents reported a need for counselling on nutrition, and 41% requested counselling on maintaining a healthy level of physical activity. Sixteen percent of participants included the need for further information relating to sexual health screening as a topic for inclusion during transition. The insights obtained from this study could allow for a more tailored transition approach specific to the needs of the adolescents. A limitation to this study was the description of functional limitations and health needs by parent respondents as of the low youth response (27%).

In a study evaluating a transition programme for paediatric rheumatology youth in the USA, Jensen et al (2015) designed a 10-item questionnaire. The response rate was 27% (n=57). A social worker was the transition coordinator and met with patients who were 16 years of age or older, supplied transition education material, assisted them developing a transition plan and assisted those making appointments with the adult services. They were then contacted again 6-8 months later. Despite this formal programme the success rate was 42%, which the author's state was unacceptably low. No further information is given as to how the success rate was so low, or if anything could have been done to increase that. It is stated that the satisfaction rate for the transition process was high, but this seems unlikely with the low success rate of the programme and the fact that 32% were lost to follow up. The limitation of this study as detailed by the authors that it took place in a single centre so may not be generalised to other populations. The student found that although these published studies pertained to the preparation of adolescents to transition to adult services were completed; there was a small participation rate in several of the studies. It is also not known if these programmes were continued following the completion of the studies. Once again, the lack of

structured protocols for adolescents transitioning from child to adult services is evident from these studies.

2.3.3 Barriers to successful transition.

Barriers to transition have been highlighted in many of the studies already reviewed here. One barrier that has been mentioned in a number of studies is the exclusion of those with intellectual disabilities from partaking in this research (van Staa et al 2011, Rutishauser et al 2014, Hislop et al 2016). While this may be related to the external validity of these studies, only one study specifically mentioned the inclusion of those with an intellectual disability in their study design (Gorter et al 2015). This may be seen as a gap in the already limited available research as the views of this study group is not being explored or included.

Garvey et al (2013) examined the barriers to successful transition in young adults with diabetes in the USA. They surveyed young adults aged 22-30 years old. The response rate was 53% (n=258). They noted that the development of evidence-based practice for transition programmes is critical for both paediatric and adult services dealing with those with chronic illnesses. Over 78% reported experiencing a barrier to transition such as not knowing how to contact their new adult provider or feeling upset about leaving their paediatric provider. The study found that lack of adult provider name and contact details were the barriers most strongly connected with a gap in care. The limitations to this study were that the information was gathered after the transition had occurred so might be subject to omission or bias. Huang et al (2011) mention the finding of healthcare staff in their study who found anxious parents and lack of information/poor communication between paediatric and adult centres as barriers to a successful transition for young people with chronic illness.

In a qualitative evaluation of a pre-consultation guide for young people with asthma, Milnes et al (2013) found that young people aged 14-19 years old have limited participation with professionals. This U.K study had 24 young adults and nine practice nurse participants and semi-structured interviews were conducted. The findings of this study showed that peer written information was most useful to the participants. They also found that some young people lacked the confidence to speak with the nurse themselves and would prefer a parent to be present. Some young people also found that their medical reviews were more centred on measurement of their vital signs, but they would also have wanted more from the consultation. The main finding in the study was that a similar pre-consultation guide could be useful for other young people with chronic conditions. A limitation to this study is the time lapse of three or more months between using the guide and partaking in the interview.

In a study by Szalda et al (2015), the practice supports and barriers to care for young people with chronic conditions were explored. The study was cross sectional and consisted of qualitative open-ended, semi-structured interviews with 22 providers who cared for adults with paediatric onset chronic conditions in the USA. Sixty-seven providers were invited to take part and 22 completed the interviews. Five major themes were identified as barriers. These were: a definition of the patient's medical team, lack of appropriate medical records, time constraints, lack of staff training and financial constraints. Although this study showed several barriers it also identified some facilitating factors for improving the care of young people with chronic conditions. These included: policies to formalise the process of new patient intake, the presence of patient portals and improving coordination in care. The limitations to this study include that it is exploratory in nature and is aimed to identify themes but not to measure outcomes. As it is qualitative in design it can uncover themes but not measure outcomes.

O'Sullivan –Oliveira et al (2014) analysed the perceptions of providers of transitional care from paediatric to adult services, and state that all healthcare providers regardless of discipline are increasingly aware of the importance of transition. A purposeful sample was recruited (n=28) and four focus groups were conducted. Six major themes emerged from the focus groups. These were:

1. Chronological age concerns as the adolescent may not be suitable for transition due to social support or education level as well as their age.
2. Provider barriers to transition such as negative views of the adult centre or poor structural support.
3. Resistance to transition. The participants stated that parents and patients exhibited a real fear of moving on.
4. Provider recognition of the importance of transition.
5. No institutional policies or standards for transition.
6. Resources for a successful transition.

The researchers state the importance of recognising how influential the staff are in preparing the patients and family for transition is vital for them to know. The study had some limitations which included that the findings from this one centre may not be open for generalisation in other areas. However, there was a large amount of information gained from this study which could benefit the transition service of this centre if acted upon clinically.

The student again found that the lack of standardised transition protocols is mentioned in articles discussing barriers to transition, and multiple studies have shown this as a concern, but it still remains an outstanding issue to be addressed.

2.4.4 Facilitators to a successful transition.

For the transition to be successful, all partners involved in the process; adolescents, parents and staff need to be prepared and educated about this process (van Staa et al 2011). Ciccarelli et al (2015) report on the work of a transitional support programme for medically and socially complex youth in the USA, aged 11-22 years of age. They find that while transition should be incorporated into health care for all youth, specific attention should be placed on those with more complex medical and social needs. In this study they assess the work completed in 2013 when they received over 200 new referrals with the average age of the patient being 16.3 years. Surveys were completed by eighty-two families (28.7% response rate) and eighty-one providers (30.7 % response rate) relating to the transition support provided. Families reported improvements to waiting times and total visit times, and receipts of reports. Physicians reported improved uploading of electronic records and requests for further information for patients on preventative care needs. Families who return to the transition clinic are an indication of its usefulness. The study confirmed that improvements in the transdisciplinary team are necessary in order to improve its functionality. The study also suggests that care coordinators are a valuable resource in a transitional care support service as they can link families to resources they may require. The limitations with this study include that it would be useful to show outcomes that can prove the usefulness of the programme such as improved population health and decreased health care costs but that is not shown in this study.

Coyne et al (2016) completed an e-health information intervention to support young people with chronic illnesses transitioning from paediatric to adult services. Data on information needs and preferences were obtained through a postal survey (n=20), and interviews (n=21) with adolescents or young adults. The participants preferred information that was trustworthy, easy to download and

did not use patronising language. They also requested information relating to the adult centre, as well as medication management. All these findings are facilitating factors to a successful transition for this patient group.

In a study by Babler & Strickland (2015) they explore the successful transition process of adolescents with diabetes to adult services.

This qualitative study, using grounded theory, has fifteen participants and each completed an in-depth interview. The purpose of the study was to gain an understanding of their experience living with type 1 diabetes. Study participants were asked open ended questions. The findings in this study included how the adolescents would say they were fine just to allay the concerns of their parents. The adolescents also detail situations when they feel they are able to transition from their parents, such as when they have the skills to take over. This study found that providing support for the adolescent is critical to allow them to obtain self-management. A limitation of this study is that as a qualitative study the results cannot be generalised.

Van Staa et al (2011) investigate the experiences and recommendations to improve transitional care of young adults, parents and providers in this Dutch study. They found that this topic had received little attention in the Netherlands and aimed to examine the experiences of this group there. Semi-structured interviews with twenty-four young adults aged 15-22 with chronic illness, twenty-four parents and seventeen healthcare providers were undertaken. Disease specific questions were added to gain more information on specific health care needs. The questions focused on expectations and experiences with transition for the patients and parents, and with attitudes towards transition and current practices with the healthcare staff. The results found that only those with haemophilia had a structured transition programme and that most patients had not been prepared for transition. Many young people and parents shared the same view that transition is

inevitable. Following transition to adult services however, it was found that those who had had frequent contact with their paediatric providers found it more difficult to establish a rapport with the adult service staff. The results also found that an improved level of organisation and communication between the paediatric and adult services was warranted. Preventing adolescents being lost in transfer is a major challenge in view of the challenges between the paediatric and adult centres which needs to be highlighted and addressed. A limitation to this study is that some of those with chronic conditions were non-responsive, and refusal rates were high, which could imply a selection bias. The possible benefits of a structured transition programme could not be established as only the haemophilia care team offered it at that time.

Mackie et al (2014) conducted a clinical trial on healthcare transition for youths with cardiac disease. The participants (n=66) were allocated to either usual care or a 1-hour nurse led one-to-one teaching session on transition to adult services. There were 58 participants. As seen in other studies, those with a developmental delay were excluded. It was stated that it believed the parent would decline participation. Following the transition intervention or usual care, all participants completed the Transition Readiness Assessment Questionnaire (TRAQ) one month and six months later. The findings of the trial showed that those who completed the nurse-led transition programme along with their usual care resulted in improved self- management skills. This intervention requires few resources other than nursing time and a quiet office and based on these findings it should be routinely provided in paediatric cardiac centres. A limitation to this study is that it took place in a single centre and was completed by a single nurse. However, study protocols were strictly adhered to throughout the process.

Exploring the experiences of youth with chronic health conditions, their parents and healthcare providers was the aim of the study by Nguyen et al (2016). This interpretive phenomenological study was

part of a larger longitudinal transition study. Purposeful sampling was used to select youths with a range of chronic conditions across 13 clinics in Ontario, Canada. A total of 11 youth, seven parents and seven healthcare providers were selected. Three themes emerged following analysis of the data. These were:

1. Youth and increasing independence:

Many young people showed their willingness to increase their self-management skills.

2. Parents as safety nets:

Parents reported that there were many instances when their child still requested assistance from them.

3. Healthcare providers as enablers and collaborators:

Many healthcare providers recognised the importance of assisting the young person with a gradual developmentally appropriate transition adult service.

The authors state that it is unrealistic for parents and adolescents to stop navigating the healthcare system together abruptly once the adolescent turns 18. The key findings of the study are that although the experiences of the youth, parent and healthcare provider are interconnected, the most significant relationship is the one between the youth and their parents. Once again, a study relating to transition, details that there is limited research-completed on this area and that further longitudinal studies would be beneficial. The study also details that its findings may not be representative of other groups of adolescents' transitions to adult services. However, the study does provide information on web pages that are currently available for adolescents transitioning. It also mentions that clinicians may not always have adequate time to spend with these patients due to time constraints and heavy patient loads. The student would believe that these issues would be a concern in many transition programmes although it is not necessarily documented.

2.5 Conclusion.

This literature review provides an overview and analysis of recent studies focusing on the transition process for adolescents with chronic conditions from paediatric to adult centres. It began with a definition of transition followed by explaining what transition relation to healthcare is, and why it is important. The search strategy was then included and following the identification of 44 studies, four emerging themes were found. These are: Preparation for transition, outcome post transition, barriers to transition and facilitators to transition. Regardless of the attention given to the importance of transition in medical organisations, few theoretical models or published guidelines exist to aid paediatric practitioners to accomplish successful transitions for their patients to adult services (Schwartz, Tuchman, Hobbie & Ginsberg 2011). Further qualitative and quantitative studies to examine transition programme effectiveness and outcomes are necessary to maximize the benefits for young adults with chronic conditions (Grant & Pan 2011).

The next chapter will detail the methods and methodology employed for the completion of the study

Chapter 3: Methodology and Methods

3 Introduction.

Research is defined as the rigorous and systematic process of enquiry, the aim of which is to describe phenomena and develop and test its concepts and theories (McCuskey & Guanyin 2015). This chapter contains a concise review of methodologies used in research. This is followed by an outline of the study design employed which is Flanagan's (1954) Critical Incident Technique. All qualitative researchers should account for the method they have chosen to use (Kramer-Kile 2012), this chapter details and rationalises the design of the study. The chapter will also elaborate on the population used, sampling method employed as well as data collection and analysis.

3.1 Research Paradigms

Research methodologies can be broadly categorised as either quantitative or qualitative in design (Meadows 2003). These two designs refer to the different kinds of data collection methods which can be associated with different views on how knowledge can be discovered and what knowledge is (Gambrill 1995). Mixed methods research, which combines the use of both quantitative and qualitative research methods, is also used and described as a third research paradigm (Ma 2012). Mixed method research is increasingly recognised as valuable as it may capitalise on the respective strengths of each research approach (Östund et al 2011).

Quantitative research can be described as involving the collection of standardised data from events, issues or practices using statistics to analyse them (Flick 2009). It can report statistically significant findings and produce statistical models (Meadows 2003). This methodology is interested in the frequency and distribution of events. It is described by Burns & Grove (2011) as a rigorous, formal, objective systematic process to propagate numerical data. Quantitative researchers generate predictions using deductive reasoning (Polit & Beck 2017). Deductive reasoning is also referred to as the "top down" approach where the researcher begins with general information and changes it into more specific information (Ingham-Broomfield

2014). There are four main types of quantitative research designs. These are Descriptive (where the subject is measured usually only once), Correlational (studies the relationship between two variables), Quasi-Experimental (estimates the impact of an intervention without random assignment) and Experimental (subject measured before and after the event). Quantitative research includes the use of data collected using questionnaires or surveys. It also includes the use of computer programmes for analysis of data, such as SPSS (Statistical Package for the Social Sciences). There are two types of sampling used most often in quantitative research according to Shields (2008). These are the representative sample and convenience sample. However, many other types of sampling occur within quantitative designed studies. Typically, sample sizes in quantitative research tend to be much larger than in qualitative research studies, this is to obtain a sample large enough to make a generalisation of the larger population studied (Holt 2009).

In contrast, the aim of qualitative research is to provide a rich, contextual understanding of the human experience through intensive examination of specific cases (Polit & Beck 2010). This methodology is characterised by its aims which are to understand some aspect of social life and its methods, which are to generate words rather than numbers for analysis (McCuskey & Guanyin 2015). Qualitative research can be described as a multifaceted method which analyses behaviour, society and culture through the examination of the person's own words and actions (Hogan, Dolan & Donnelly, 2009). Qualitative methodology uses inductive reasoning. This is a process in which generalizations are developed from the specific observations found within the research study data (LoBiondo-Wood & Haber 2014).

Some of the common qualitative research designs include: Ethnography, Phenomenology, and Grounded Theory. Ethnography is defined by Polit & Beck (2017) as a branch of human inquiry which aims to understand culture and customs of those involved in the study. Phenomenology is defined as aiming to describe the lived experience of those in the study (LoBiondo-Wood & Haber 2014). Grounded Theory is explained as an approach to

collect and analyse data that aims to develop theories which are grounded in real life observations (Polit & Beck 2018).

Qualitative researchers also build on and use the approaches, methods and techniques of hermeneutics, feminism, interviews, cultural studies, participant observation and survey research (Ryan- Nicolls & Will 2009). Qualitative research involves many different approaches and methods, but they all have a commitment to being interpretive and naturalistic in design (Cheek et al 2004). A key feature of qualitative research is how the observational data is collected. Observations are most often recorded by interview, field notes or recordings (Swafford 2014). The sample population in a qualitative study can be variable. It can vary from small groups, to institutions or just one individual (Ingham-Broomfield 2014). Overall, qualitative research has been used by health care researchers increasingly since the 1970s (Thorne 2011).

Quantitative and Qualitative Research:	
Quantitative research is used for:	Qualitative research is used for:
Recommend a final course of action.	Develop an initial understanding of an issue or problem.
Discover consensus on a particular issue.	Look for a range of ideas/feelings/views on something.
Project results to a larger population.	Understand different perspectives between social groups/ categories of people.
Identify evidence concerning a cause-and-effect relationship.	Uncover underlying motivations and attitudes influencing decision making.
Describe features of relevant groups of people.	Provide information needed to design a quantitative study.
Test hypotheses and examine specific relationships.	Explain findings from a quantitative study.
Identify and size market segments.	

Table 2. The differences between quantitative and qualitative research design (Farrell 2013).

As stated earlier, mixed methods methodology is another common approach. This methodology may offer many benefits over purely quantitative or qualitative approaches. According to Schifferdecker & Reed (2009), mixed methods can aid in the determination of the best tools possible for answering the hypothesis or question under examination. Mixed methods researchers collect and analyse data from both quantitative and qualitative methodologies

to address different but related questions (Polit & Beck 2017). This approach is being used more so in health science and is well suited to nursing and interdisciplinary research (Katz et al 2016). The main advantage to this methodology is its adoption of triangulation, where multiple methods and sources attempt to develop the research question from a variety of angles (Dodd 2008). However, mixed methods research should not be used under the assumption that more is better or that qualitative research is not complete without having a quantitative component (Dodd 2008).

All research methods have their strengths and weaknesses (Meadows 2003), and Jootun et al (2009) state that no single research method can be viewed as inherently superior to another. They find that the appropriateness of the method chosen should be appraised in relation to the research question asked. Similarly, Houghton et al (2013) state that qualitative research is being valued for its differences to quantitative research, rather than it being viewed as being methodologically inferior. Both quantitative and qualitative research continue to coexist in children's nursing to inform clinical practice and improve patient care (Clarke 2014).

In this study a qualitative methodology was used adopting the "Critical Incident Technique" (Flanagan 1954), this will now be explored in greater detail.

3.1.1 Design used: Critical Incident Technique.

The Critical Incident Technique (CIT) is a method of qualitative research developed by psychologist John Flanagan in the United States of America in 1954. This technique developed following studies completed on pilots in the American Armed forces in World War II. During this time there was a need to train flight crews in a very short time, and Flanagan devised an efficient and practical method to identify the successful training behaviors of crew members (Kenppainen 2000). CIT is an inductive, systematic method which allows for observations on human behavior in defined situations to be collected (Arvidsson & Fridlund 2005). A critical incident can be described as

a human activity which is complete enough to allow for predications and inferences to me made about the person accomplishing the deed (Bitner et al, 1990). The Critical Incident Technique has been used to study peoples' activities from a variety of professions since its inception (Schluter, Seaton & Chaboyer 2007). It has been used in many fields such as marketing, medicine, education, psychology and nursing (Mulholland, Barnett & Woodroffe 2015). Unlike other formats, CIT is highly focused on providing practical solutions to problems identified by participants during the interviews (Kenppainen 2000, Fitzgerald et al 2008).

CIT involves interviewing participants who are active in the areas under examination (Bradley 1992). It focuses on asking the participant to expand on positive and negative aspects of the incident. The sample size of the critical incident study should be based on the number of incidents rather than the number of participants, and a total of 100 incidents are sufficient for a qualitative analysis (Flanagan 1954). A critical incident is an event that results in a change to subsequent behavior and actions (Eriksson et al 2016). However, Pavlish et al (2011), Silén et al (2012) and Ivarsson et al (2014) chose to use different phrases other than a critical Incident for their nursing-based studies. These authors found that referring to an event as a critical incident caused some confusion as nurses would use this terminology about the physical condition of patients. Similarly, the student will be referring to "event" rather than "critical incident" for this study. Qualitative researchers do not usually label individuals involved in their studies as subjects but rather as informants or participants (Streubert & Carpenter 2011). Throughout this study, those involved will be referred to as participants.

The student chose to use this research method because, as stated by Kemppainen (2000), unlike other qualitative research methodologies which can focus on describing phenomenon, this methodology is highly focused on providing solutions to practical problems. Another advantage to using CIT is that it depends on the description of things as they are rather than as they should be (Narayanasamy & Owens 2001). It is also suitable for use for all the participant groups in this research study. Ensuring a great variation within

the sample group, while giving them the same information, is in line with the CIT tradition as stated by Arvidsson & Fridlund (2005). Adolescents, parents and MDT members were able to discuss the positive and negative experiences with reference to transitioning from child to adult hospital care during their interviews. Therefore, the one study design can be used for all participants and findings can be explored from all the viewpoints of the same area under examination. Another reason for this study design choice was that CIT interviewing comprises of a simple set of procedures which allows the participant to recall what they saw or felt without judging or expressing a personal view. This methodology has also been used globally by nurse researchers to explore numerous nursing issues (Bradbury-Jones & Tranter 2008).

In their study of children with chronic illness and their parents, Kelo et al (2012) found that the Critical Incident Technique (CIT) was suitable for use with children, although they found that it has seldom been used with this group. They stated the rationale for their method choice was that CIT helped the children and parents to be as specific as possible when describing events. This study shared many similarities with the authors study apart from study size. Their study was of 12 children age 5-12 years and 19 parents. Their study examined patient education during a hospital visit. The parent was also with the child during the interview- although the interviews took place in the patient's home. With the student's study a parent must be present during the interview process. It was very useful during the formation of the study design to see that data had been published in a similar area. Another study using CIT was Getrich et al (2016) which examined the use of focus groups with adolescents and parents. This study was also explored for similarities to the student's study. The main difference found was that their interviews were conducted separately, so that the parent and adolescent were not in the same room. They found that this enhanced participant comfort as well as their level of disclosure to the interviewer. In line with clinical guidelines, a parent must be present for any interaction with a child, so this option was not possible for the student's study. There was also a CIT study which involved examining how nurses communicate news to

hospitalized children (Peña & Rojas 2014). This study involved 30 children between the ages of 8 and 14. Unlike the student's study, these researchers used participant observation as well as semi-structured interviews. Interestingly, this study found that the majority of the nurses' who spoke to the children did not have their prior consent before imparting news to them. This issue was addressed in the student's study by providing age appropriate information leaflets and then obtaining assent and parental consent before any interviews took place.

Although the student chose CIT as the most methodology for this study, the limitations and weaknesses of this design should be acknowledged. The main weakness to this design as discussed by Byrne (2001), to which the student agrees, is that it elicits the best and worst aspects of the situation only. That suggests that routine incidents may not be reported, and therefore that data may not be discussed during the interview..Another weakness to this study design is that the critical incidents often rely on memory which could be distorted by the time the interview took place. This weakness is mentioned as a limitation to the study in chapter six.

3.1.2 The Process of Critical Incident Technique.

There are five steps identified in the CIT approach. These are:

Step one was obtaining the views of adolescents with severe haemophilia and their caretakers relating to their views on transition to the adult hospital.

Step two was to specify how the events were collected. The incidents were collected following the completion of semi -structured interviews.

Step three was the collection of data which is discussed in the next section of this chapter.

Steps four and **step five** are examined in detail in chapter four.

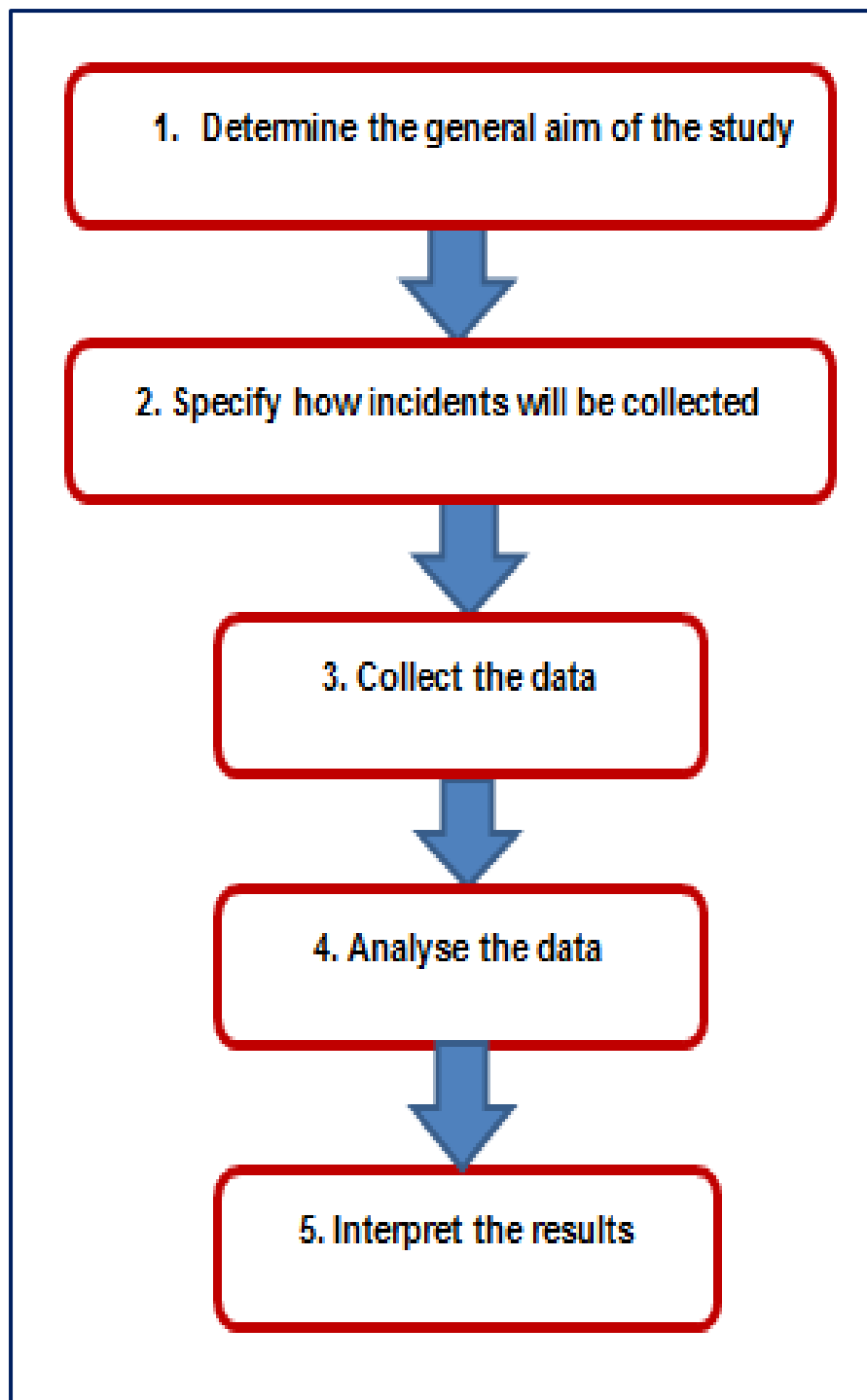


Figure 2. The five steps of Critical Incident Technique (Flanagan, 1954)

3.2. Sampling and Sample size.

As detailed in the most recent World Federation of Hemophilia global report (World Federation of Hemophilia 2016), of the 839 people with haemophilia in Ireland, 9% of people with haemophilia A (n: 54) and 8% of people with

haemophilia B (n: 19) in Ireland are aged between the ages of 14-18 years old. Similarly, this global report found that 41% of the population with Haemophilia A (n: 261) are within the age bracket of 19-44 years old. Thirty-eight percent of the population with Haemophilia B (n: 90) are within this 19-44-year-old age bracket. These are the two age groups which were relevant to this research study. The Consultant Haematologist from the paediatric Hospital provided a list of the adolescents with severe haemophilia due to transition over the next 5 years, and the Consultant Haematologist from the adult Hospital provided a list of people with severe haemophilia who have transitioned to adult services within the last 5 years. These lists have shown that there are 21 adolescents due to transition and there have been 23 adults who have transitioned.

The type of sampling employed in qualitative research is determined by the methodology selected and the topic under investigation, and not by the need to produce generalizable data (Higginbottom 2004). It has also been stated by Polit & Beck (2018) that the sample size in a qualitative study is frequently calculated on informational needs. For most qualitative studies, as with this study, the student was looking for a purposively selected sample of people who can illuminate the phenomenon they intend to study (LoBiondo-Wood & Haber 2014). This is a form of non-probability sampling where the researcher aims to sample only those relevant to the study (Bryman 2012). The aim of this research study was to interview people from each of the five groups as detailed below. For this reason, a purposeful sampling approach was employed as it was deemed the only suitable sampling technique to use in this study

Name of Study Group:	Name of Study Group:	Name of Study Group:	Name of Study Group:	Name of Study Group:
Adolescents with severe haemophilia.	Parents of an adolescent with severe haemophilia.	MDT Children's Hospital.	MDT Adult Hospital.	Adults who have completed the Transition from paediatric to adult hospital.
Number of Participants in this Study Group aim:	Number of Participants in this Study Group aim:	Number of Participants in this Study Group aim:	Number of Participants in this Study Group aim:	Number of Participants in this Study Group aim:
15	15	7	7	8

Table 3: Sample size for Inclusion in the study of “The views of adolescents with severe haemophilia and their caretakers on their transition to adult services”.

The adolescent group comprised of a number of those who will be transitioning from the paediatric to adult hospital within the next five years. Semi-structured interviews took place with the transitioning adolescents, once parental/caregivers' consent (appendix 2) and assent (appendices 3 & 4) had been obtained and ethical approval was granted (appendices 5-9). Interviews were completed using the appropriate interview guide (appendices 10 & 11). Information sheets (appendix 12 & 13) were also given to the adolescents prior to consent and assent being obtained. These interviews took place in a private room in the children's hospital: a parent was also present in line with hospital guidelines. The adolescents were asked their thoughts and expectations on transition. The clinical psychologist was aware of the interviews taking place and agreed to be involved should

any emotional support have been required after the interview. No psychological support was requested by any participant.

For the parents of adolescents with severe haemophilia and the MDT members consent forms were also available (appendices 16-19) Semi-structured interviews guides were also designed and used for the parents of adolescents with haemophilia (appendix 20) and MDT in the children's hospital (appendix 21). Study information sheets can be seen in appendices 20 & 21.

Semi-structured interviews were due to take place in the adult hospital for recently transitioned adults with severe haemophilia. The consent for this group can be read in appendix 22. An Information sheet (appendix 23) and interview guide (appendix 24) were also available. However, this did not occur so only data from the children's centre was collected. This was discussed further in section 3.3 page 70.

A total number of 26-52 participants were originally sought and 15 people took part in this study.

As part of this thesis a sample of one transcribed interview is detailed in appendix 25.

3.2. Recruitment

It is the policy of the Paediatric Department where the student works for the Consultant to inform all patients and parents/ caretakers of pending sponsored trials, academic trials or other relevant studies. Any trials/studies are always discussed at clinic in advance of them being offered to the patients for their consideration, and relevant information sheets are forwarded to them. This discussion took place between the Consultant and the adolescent patient and their parent/caretaker. The student was not present at these first meetings. Should they wish to take part the adolescent and parent/caretaker informed the staff at the time of their next visit.

With reference to data collection in the adult hospital: this did not take place as the Consultant who had agreed to oversee the study there ended his

employment and the student could not secure another similarly qualified Consultant within the time frame to complete data collection with the young adult group or MDT.

The people who were asked to participate met the inclusion criteria.

Inclusion criteria:

1. They must have been either an adolescent with severe haemophilia,
2. An adult with severe haemophilia who has transitioned from the Children's hospital,
3. A parent of an adolescent with severe haemophilia.
4. A member of the multi-disciplinary team (MDT) working in the area of haemophilia care in either the paediatric or adult hospital. This included the dentists, physiotherapists, clinical nurse specialists and psychologists.

Exclusion criteria:

1. Unable to or unwilling to participate in the study.
2. Individuals with mild or moderate haemophilia.

The recruitment process at the children's' hospital commenced once the Consultant Haematologist provided the number of patients with severe haemophilia aged between 12-18 years of age. An age appropriate information sheet was posted to these adolescents and their parents. The information letters provide a contact number/email address inviting potential participants and their parents/ guardians to contact the student to discuss the study. During the initial contact, it was offered to arrange for an outpatient attendance to discuss the study with the student to coincide with their regular appointment. This was rather than having to take additional time off from schools or work for the potential participants and their parents/caregivers. If the patient and parent / guardian wished to proceed with the study, they were provided with the consent form and patient assent during this visit. Forms were signed before any interviews were conducted. The patient and parents/guardians were informed that they could stop the process at any time, including during the interview.

MDT were sent an invitation letter in the internal mail and asked to contact the student should they wish to be involved. A time, date and venue in the children's hospital for the interview was then agreed. Consent Forms were signed before the interviews were conducted and the staff were reminded that they could stop the interview process at any stage.

For the adult groups in the adult hospital, a gatekeeper was identified and agreed to be involved with the recruitment process. An office was to be made available for use in the adult hospital where the interviews would have taken place.

All participants obtained a copy of their assent/consent forms. All participants were advised that they could withdraw from participating at any stage without prejudice. The aim was to complete the interviews within 6 months of their agreement to take part in the research study, and all data was collected within this timeframe.

3.3 Data Collection Step 3 Critical Incident Technique (CIT).

Although virtually anything may be considered data, the spoken word is the most common form of collecting data in health research (Grossoehme 2014). An interview is a method of collecting data in which closed quantitative style or open-ended qualitative style questions can be asked (Doody & Noonan 2013). In their CIT study Biagetti et al (2013) used interviews as their data collection method and stated that it gave them a structured and practical way to collect and analyse information, activities and their significance.

A semi-structured interview is described as including some predefined questions or topics leading to probing further as the participant responds and have been proven to produce excellent data providing insights into the experience, opinions and perceptions of participants (Halcomb 2015). The reason semi-structured interviews are such a popular data collection method is due to their versatility and flexibility (Kallio et al 2016). The student could also have used focus groups as a method of data collection. However, as the suitable participants were not attending the hospital at the same time this

was not practical. Therefore, in line with most CIT studies reviewed by the student, individual interviews were conducted instead as the data collection method.

Semi-structured interview guides were developed by the student in line with the CIT design. The objective was to obtain what the individual interviewee viewed as the positive and negative aspects of transition from the child to adult hospital from their own point of view. It was proposed that the findings from an adolescent with severe haemophilia would differ from those of a parent or MDT member. It was the student's aim to capture the findings of all the relevant groups of people involved with the care of the adolescent with severe haemophilia so that an encompassing view of the situation could be reviewed.

3.3.1 Data Collection.

Critical incidents in this study were reported during the completion of the fifteen semi-structured interviews. All the data was collected in private rooms within the research site. Consent forms were signed by the study participant and the student. The study participants were assured that no identifiable data would be reported, and that they would only be referred as a specific code. Only the student and the college had access to the codes which would identify the participants. All data was stored in a locked area within the hospital and all computer data was stocked securely in a password protected computer during and after data collection.

Some researchers suggest that the spontaneity of peoples' replies greatly enhance the content of the interview data (Bryman & Cassell 2006). However other researchers recommend advising the study participants in advance to think about critical incidences (Bott & Tourish 2016, Fitzgerald et al 2008, Schluter, Seaton & Chaboyer 2007). The quality of the interview data obtained depends on the ability of each participant to articulate their experiences for which they were recruited to explore (Sandelowski 1999). All participants in this study received written information leaflets specific to their

role in the study prior to consent. All the field notes taken by the student during the interviews were transcribed verbatim at the time of the interview and read back to the interviewee to insure the student had documented the data as intended.

3.3.2 Pilot Study.

Rigorous data collection is an integral part of qualitative research and therefore most of the time spent on study design should relate to this area (Ranney et al 2015). Pilot testing of an interview guide aims to confirm its relevance and identify any areas which may have required reformulation (Kallio et al 2016). To ensure optimal data collection was achieved, the student completed one pilot interview from each of the study groups. This allowed her to analyse the interview transcripts and identify any interview techniques and amend terminology used which required clarification. The data obtained from these pilot studies was included in the main study. Prompts used during the interview process of all participants included 'independence' and 'hospital environment. These prompts were used with the adolescent group but were not required with the adult groups.

3.4.3 Interview technique.

Following the completion of the pilot interviews, the student amended how she introduced herself to participants, as it became clear that all three pilot study participants were unsure of the specifics of her role.

She spent longer explaining the rationale for the study and offered to go through the information leaflets with participants again, as some pilot study participants were unclear as to the reasoning for the study.

Each study group had an age appropriate or group appropriate interview guide which the researcher used during the interview. Referring to the interview guide, the student then asked the participant what they thought as the positive outcomes towards transition to the adult hospital. This could have been one outcome or many. The student then asked the second question, which was what they are concerned about when they think about

transition to the adult centre. All interviews were performed by the student. The interview could also have been audio recorded, which is detailed in the information sheets provided to the participants. None of the participants wished to be recorded, so only the data transcribed verbatim for later analysis was available.

3.3.4 Terminology.

The student amended terminology used so that she could impart the same level of information more comprehensively. For example, she explained the term “transition” and explained the difference between transition and transfer as some pilot study participants were not completely sure as to the differences between the two.

The student included a short explanation of the transition process as it currently stands to the adolescences as they were in the 13-15-year-old category and she wished to allay any fears that they were going to be transferred to the adult hospital any time soon.

3.4 Rigour in Qualitative Research.

Rigor in qualitative terms, or reliability and validity in quantitative terms, are ways to establish confidence and trust in the research findings (Thomas & Magilvy 2011). The most frequently mentioned criteria for assessing rigour in qualitative studies in the literature reviewed by the author is that proposed by Lincoln and Guba (1985). The criteria of credibility, confirmability, dependability and transferability were mentioned in studies by Houghton et al (2013), Joosten et al (2009), Ryan-Nicolls & Will (2009), Tuckett (2005) and Thomas & Magilvy (2011).

How this research study-maintained rigour is now discussed. Rigour can also be deemed relating to the trustworthiness of the research findings,

Credibility: The credibility of a study is of the utmost importance in qualitative research (Swafford 2014). The results must represent the multiple realities in qualitative research for it to be deemed credible (Henderson & Rheault 2004). The credibility of this study was confirmed as the student had comprehensive knowledge of the topic being explored and was able to gain relevant information from the participants. The student also had a good understanding of the Critical Incident Technique methodology, so was able to extract the information gained and provide a credible study on this topic.

Confirmability: This refers to implied neutrality and states that a study is deemed confirmable when results are free from bias (Henderson & Rheault 2004). The student contacted all the people who met the inclusion and exclusion criteria once their permission to do so was obtained. Only the people who were willing to participate were interviewed. Therefore, selection bias was minimised as not all those suitable for inclusion took part. Researcher bias is minimised by confirming that Dependability and Transferability are intact. Confirmability was also established by the student providing a selection of quotations from the study participants.

Dependability: This is described as focusing in the stability of the data over time and in different conditions and contexts (Beck (2009). Dependability was assured in this study as the student completed all the interviews with the different study groups and used the same interview guides throughout the data collection.

Transferability: Nicholls (2009) explained that transferability is the evidence that is possible to relate developed theories from one group or context to another. It was also suggested by Amankwaa (2016) that by the investigator describing in sufficient detail the events surrounding the study that it can be transferred to other settings and situations. The student kept digital and handwritten records in line with ethical guidelines adherence. She also provided a description of the participants and the setting of the study. Therefore, it would be possible to transfer this study to a different setting or situation, for example for a similar subject group in a different hospital.

3.5. Ethical Considerations.

Prior to any contact with any possible participants in this research, ethical approval was granted by both the Ethical Committee in the Children's Hospital and that of the Adult Hospital following the submission of Application forms which addressed any ethical considerations. These committees adhere to the Declaration of Helsinki Ethical Principles for Medical Research involving Human Subjects (World Medical Association 2017) as well as the National Consent Policy (Health Services Executive 2017).

Only after written confirmation of ethical approval was obtained did the student begin to post out information leaflets to suitable participants. Student contact details were provided in these letters.

The student wishes to note that subsequent to her data collection in 2017, but prior to her thesis submission date in 2018, new legislation was introduced into Ireland relating to data collection. Statutory Instrument number 314 of 2018 (Irish Statute Book 2018) focuses on Health Research regulations contained within The Data Protection Act 2018. It relays how appropriate governance must be enforced regarding the completion of health research. It also includes details the necessity of Research Ethics Committee involvement, and the provision of training in data protection law for those involved in health research for example. The Introduction of General Data Protection Regulation (GDPR) came into force throughout the European Union on 25th May 2018 (Data Protection Commission 2018). It provides for higher standards of data protection for individuals and the increased obligations for organisations who process and hold personal data. The student continued to work within these frameworks when they were introduced as she had with the earlier legislation.

3.5.1 Assent and Consent.

It is of great importance that the voices of children be heard (Luchtenberg et al 2015). Having knowledge of children's experiences in relation to their life

situations is particularly important for professionals who care for them (Buckwald & Delmar 2011). In line with all international guidelines and the ethical approval granted for this study, assent forms were signed by each adolescent taking part in this study. A consent form for the adolescent's participation was also signed by their parent. Consent forms were also signed by all adult participants prior to interview. These were the parents of the adolescent with severe haemophilia and MDT.

3.5.2 Confidentiality.

Any information supplied to participants during the informed assent or consent process much include information as to how confidentiality and data storage is assured (Hiriscou et al 2014). The participant's confidentiality was protected by ensuring that all data was stored in a locked drawer within a locked room. All participants were given a code by the researcher so that their names would not be used. Only the student and her supervisors have access to the key to the coded data. Individual data/results were recorded on a password protected Microsoft Office document. Data were stored onto the V: drive of Royal College of Surgeons in Ireland (RCSI) server for 5 years, using a unique access code provided by RCSI to the researcher. Remote Access to RCSI is available on RCSI Connect (www.connect.rcsi.ie). The use and transfer of patient data associated with this study is governed by Irish Law 'Data Protection Act 1988 and Amendment' and the European 'Data Protection Directive 95/46/EC', which ensures the protection of individuals regarding the processing of personal data, including sensitive data, and the exchange of such data. The researcher has an obligation to protect the fundamental rights and freedoms of natural persons, and especially their right to privacy with respect to the processing of 'personal data' and 'sensitive data'. In accordance with the ethical approval obtained from the children's hospital, data is also stored on the password protected G: Drive in that hospital. All hard copies of forms and guides are stored in a locked drawer within the locked office of the student.

If a child were to reveal to the researcher, or it is observed by the researcher, that a child was at significant risk of harm, the researcher must divulge this information to the appropriate authorities. The child and parents/legal guardians were informed of this obligation during the consent/assent process and it is highlighted in the information leaflets.

3.6 Timescale.

The original ethics application was submitted in 14th March 2017 for the research site. This was then granted on 31st March 2017 (Appendix 5). Two further amendments to patient information sheets were made and ethical approval was also granted (Appendices 6- 9.) Interviews took place with the adolescents, parents and MDT in the research site between the 6th September 2017 and 11th December 2017.

Ethical approval to complete the research in the adult hospital was granted on 28th August 2017 (appendix 10) however, no data was collected as addressed in 3.3 on page 70

3.7 Conclusion.

In this chapter the methodology of research was discussed, and why the method of Critical Incident Technique was chosen as an appropriate research tool for use in this study. Information was provided on the population used, the sampling and recruitment methods. Data collection was also explored including the ethical issues when completing research.

Chapter 4: Findings

4. Introduction.

Analysis of data is one of the most challenging aspects to conducting qualitative research (Priest, Roberts & Woods, 2002). This chapter details the analysis utilized with this Critical Incident Technique (Flanagan 1954) study. The data was gathered in the three subcategories of:

1. Adolescents with haemophilia.
2. Parents of adolescents with haemophilia.
3. Multidisciplinary team members (MDT) working in haemophilia care.

Each subcategory is explored in this chapter, as are the categories of data extrapolated from the interview transcripts obtained from participants from these subcategories. The resulting categories are then analysed revealing the themes of:

- Independence
- Meeting peers
- Preparation for change
- Apprehension
- Communication

The findings are described in greater detail throughout this chapter and to illustrate some of these, excerpts from the participant's transcripts are provided.

4.1 Demographics

Resulting from the attempts to recruit study participants, a total of fifteen participants from the three study groups consented to take part in this study. These groups were:

1. Adolescents with severe haemophilia,
2. Parents of adolescents with severe haemophilia,
3. Multidisciplinary staff members from the paediatric haemophilia service.

All these participants met the inclusion criteria. The number of people who agreed to having information sheets provided to them is detailed in Figure 3

Four adolescents between the age of 13 and 15 years old took part. The adolescents were all male as is the case with all patients with severe haemophilia attending the acute site. Four parents of adolescents with severe haemophilia consented to be part of the study. The gender of the parents will not be provided as to maintain confidentiality. The majority of parents who attend the site with their child are their mothers, therefore if the father of the child attends and was interviewed then both he and his child would be more easily identifiable. The adolescents and parents were interviewed in a private room in the Out Patients Department (OPD) of the acute site. All adolescent interviews took place with a parent present along with the student. This is in line with hospital policy. Seven multidisciplinary team (MDT) members took part. Due to the small number of staff working in the various specialities within the haemophilia service, no further information will be provided in this article pertaining to their professional role. This is to maintain their confidentiality as was assured prior to their consenting to take part. These MDT members all have at least ten years' experience in their specialist role. The MDT were interviewed in either the student's private office or the MDT member's private office in the acute site.

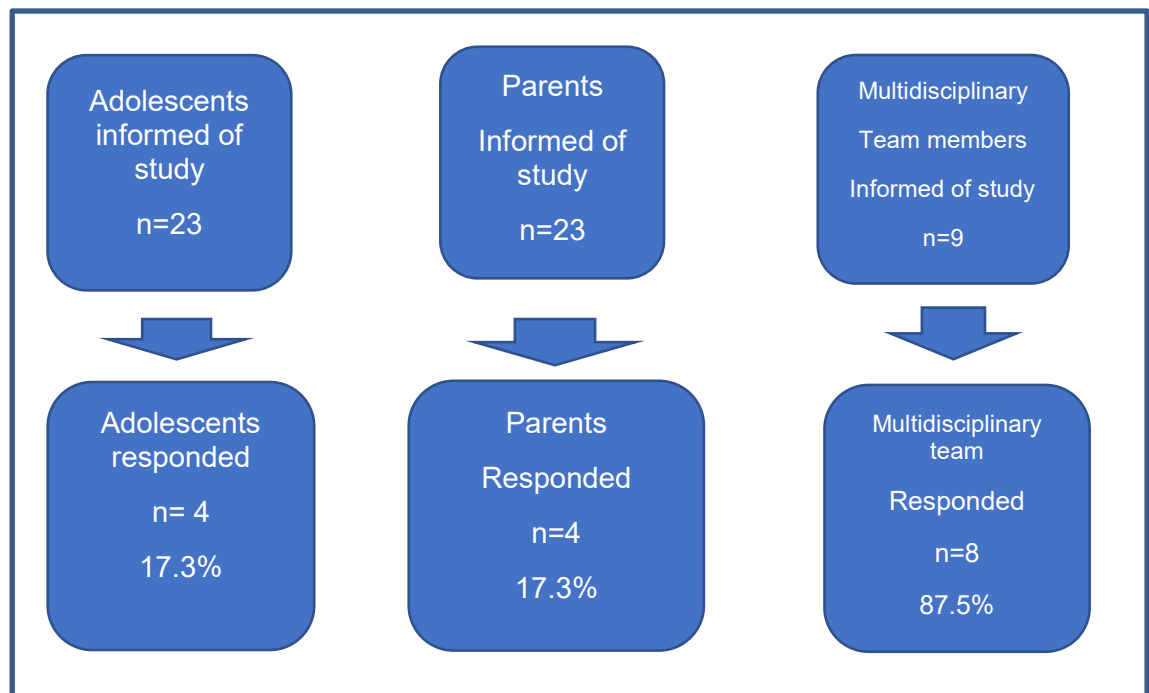


Figure 3: Demographics

4.1.1 Step 4 CIT approach: Data Analysis.

Thematic analysis was undertaken in accordance with the Critical Incident Technique. The student read through the field notes several times to obtain an overall view of the content and identify emerging themes. There are coding systems such as CAQDAS (Computer-assisted qualitative data analysis software) or Nvivo which could have been used to put the data collected into defined categories to analyse them. However, the student completed the analysis without the aid of software. Each transcribed interview was read repeatedly to analyse the data which was then coded and sorted into sub categories, categories and main areas in line with Flanagan (1954).

There are five steps involved in the analyses of data in the CIT approach.
These are

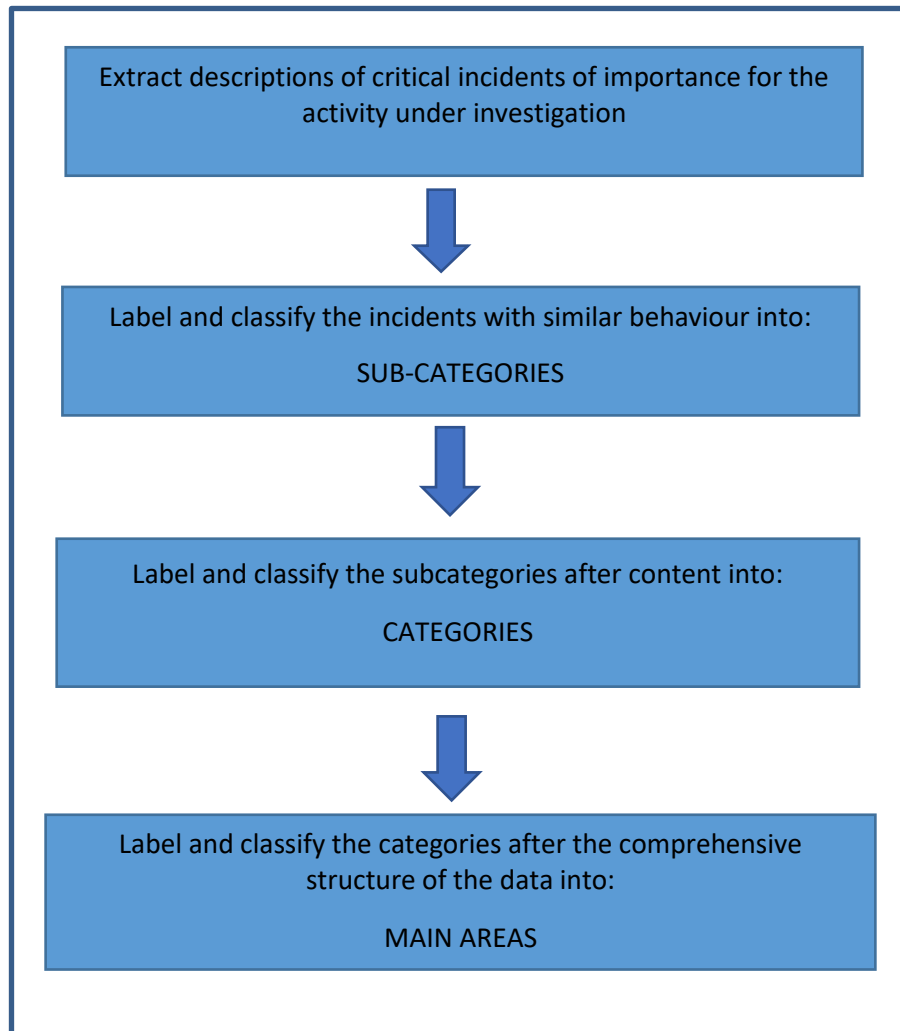


Table 4: Steps of data analysis in accordance with the Critical Incident Technique (Flanagan 1954)

4.2 Critical Incidents relating to the positive aspects of transitioning to the adult hospital.

Following the analysis of the interview transcripts there were 55 critical incidents identified as relating to the positive aspects to transitioning to the adult hospital. These incidents were labelled and classified into subcategorised groups before being categorised into similar themes.

These will now be explored in greater detail within each subcategory before the main themes of the study are presented.

Subcategory 1: Adolescents with severe haemophilia.

All the adolescents were aware that they would ultimately have to move on from the paediatric hospital to the adult hospital before the topic was broached with them by the student. The first question they were asked was:

“Can you tell me what you are looking forward to when you move to the adult hospital?”

There were eleven positive critical incidents identified in this group of four adolescents. Having reviewed all the interview data, the student deduced that the adolescents' positive views on transition to the adult hospital could be categorised as involving:

- Increased Independence
- New Beginnings

1. Increased independence

As adolescents this group will be distancing themselves from childhood activities and situations and are looking forward to the new opportunities offered to them as more mature and knowledgeable people in society.

One adolescent stated that being more independent and:

“...being able to organise my own appointments” (A1)

was what he was most looking forward to. Similarly, participant A3 stated that:

“Having the independence of going to the hospital on my own”

was the first thing that came to mind when he thought of transitioning to the adult hospital. During the transition process the adolescent will have increased their level of independence with their own care. They can now access their own veins or central venous catheter (CVC) to administer their factor replacement medication. They will be competent performing this skill following training with the Clinical Nurse Specialists (CNSp). All adolescents are required to be proficient in this task before they can transition to the adult hospital. They will also be able to contact the pharmacy to detail their requirement for medication prior to their transition. A task which would have been normally completed by their parents. They are now eager to increase the number of tasks that they can complete without requiring their parent to be present.

2. New Beginnings

The adolescents will be aware that the adult hospital has more opportunities to have a room of their own to be admitted to, rather than what the children's hospital has available. Most of the adolescents also mentioned that they are eager not to have young children, who they deem as noisy, close by. Two adolescent participants explained that they were most looking forward to the adult hospital because:

“It will be a different place so no babies and no screaming children” and “I’ll have my own room there and not sharing” (A2)

This view was reiterated by participant A4 who said that:

“There’ll be no small children, so it will be quieter”

The adolescents may have heard from other people who have transitioned to the adult hospital that it appears to have advantages as a hospital over the children's hospital. These could include such things as public transport links and the number of eateries.

Subcategory 2: Parents of adolescents with severe haemophilia.

The parents were also aware of the necessity for their child to transition to the adult hospital before the topic was brought up by the student. The parents interviewed were not expecting their adolescent to begin the transition process imminently, but all had views on this topic.

The first question the parents were asked was:

“What do you think are the positive aspects of your child transitioning to the adult hospital?”

From the parent group of four, there were sixteen critical incidents relating to the positive aspects of transition for their children to the adult hospital. To surmise, all the parent's data on the positive aspects to transition can be categorised as:

- Increasing responsibility
- Hospital environment
- Meeting older peers

1. Increased responsibility

Participant P1 said that they didn't know exactly what the positive aspects of transition would be but thought that:

“Maybe it will help him mature a bit?”

Participant P3 also suggested that:

“His responsibility is increased when transitioned- he can do more for himself”.

Another finding was that:

“In the adult hospital staff will talk to him not the parent and they would treat him more like an adult” (P2)

The parents were anticipating many positive changes for their adolescent once they had completed the transition process.

2. Hospital environment

Another area which was mentioned by the parent participants was that being in the same ward as younger children and babies can cause them to be disturbed frequently, and that this would be a positive aspect to their child transitioning to the adult hospital. Participant P2 found that:

“...When other children are noisy, and parents are making noise and not considerate of others”

They also highlighted the differences between the levels of maturity between the patients in the children's' hospital as being another positive aspect to their child leaving the paediatric hospital.

“The hygiene aspects ... nappies being changed in the bed beside your child's” (P2).

Also related to the hospital environment, participant P3 stated that parking would be better in the adult hospital. On a similar vein, participant P4 stated that:

“The adult centre is geographically closer to us and it's safer there”

As these related comments focus on the parent's views on the positive aspects of the hospital environment, the category of Hospital environment was identified.

3. Meeting older peers.

Several of the parents referred to their adolescents meeting older men with haemophilia as a positive aspect to their transition to the adult hospital.

People with haemophilia often have the chance to meet up at social and educational events, so the parents and adolescents may already know several adults with haemophilia who have already transitioned to the adult hospital. However, when they officially move to the adult hospital some of the parents felt that:

“... He gets to see how the adults get on” (P3)

“He’ll be meeting new people and it’s all integrated in the one centre” (P4).

Subcategory 3: Multidisciplinary Team members working in haemophilia care.

In the Multidisciplinary team (MDT) group of seven participants, there were 28 critical incidents identified as being related to the positive aspects of transition of the adolescent to the adult hospital.

The first question the MDT were asked was:

“In your role what do you see as the positive aspects of the current transition process of the adolescent with severe haemophilia to the adult hospital?”

After reviewing the transcripts, the student determined that the two categories found with the MDT group in relation to the positive aspects of transition to the adult hospital as being:

- Adolescent preparation.
- MDT preparation.

Adolescent preparation

The first positive finding with the current transition programme which all MDT participants mentioned was the increased level of responsibility in the adolescents during this time. Participant MDT6 stated that:

” They have to take more responsibility for their own health”

Participant MDT7 concurred when they said:

“We need them to grow up”

The MDT also mentioned how the transition programme is paramount for the continued care of the adolescents into adulthood. It should be:

“Emphasising what the adolescent can do” (MDT 1)

“Make them think and plan for the future.” (MDT2)

It was also suggested that:

“It is undignified to be around Disney cartoons at a time in their life when they want to strike out” (MDT 7)

It was noted by MDT2 that the transition programme will also clarify to the parents how little the adolescents really know about their condition.

MDT Preparation

While the MDT spoke of the preparation of the adolescent during the transition process, every MDT interviewed also spoke of the staff involvement in the transition programme. Most of the MDT staff mentioned they believed the adolescents were receiving a suitable transition to the adult hospital.

“What’s really good about our service is its individual approach “(MDT5)

“It’s a good system, a letter from each MDT is given from the paediatric hospital to the adult hospital (MDT 7)

“Paperwork to back up what we do” (MDT1)

“Very individualised programme” (MDT3)

“Adolescents are prepared over a number of years” (MDT4)

Following on from analysing all the data from the three groups regarding the positive aspects of transition from child to adult hospital, three main themes emerged:

Independence

All three groups mentioned the change in independence of the adolescent during the interviews specific to their study group.

Meeting Peers

Meeting with other people with haemophilia in the adult hospital was mentioned by adolescents, parents and MDT during interviews.

Preparing for change

Transition requires a change in circumstances, and each study group identified preparation for this change as a positive aspect to the transition.

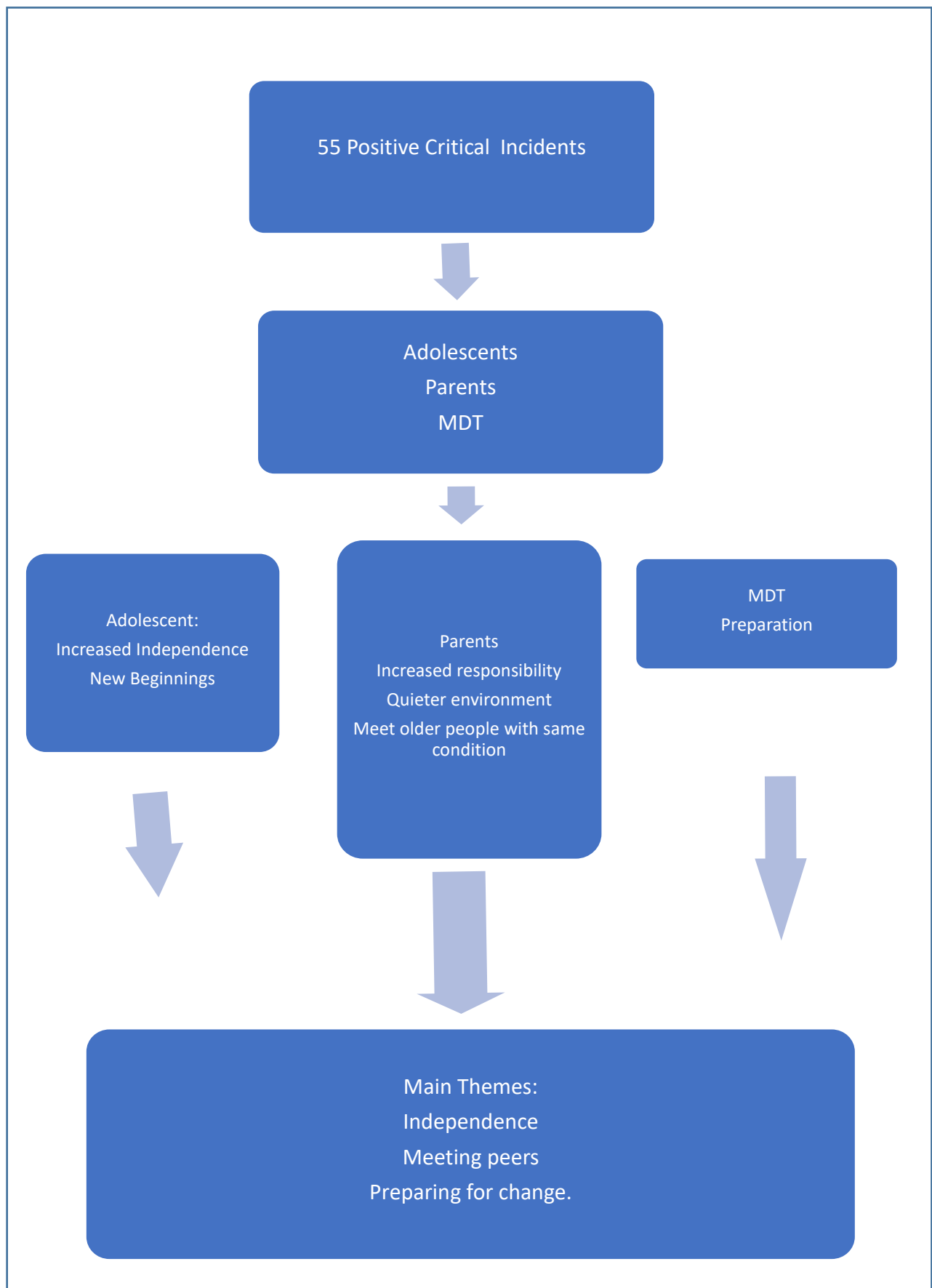


Figure 4 Steps of data analysis in accordance with the Critical Incident Technique (Flanagan 1954) Positive Aspects.

4.3 Critical incidents relating to the negative aspects of transitioning to the adult hospital.

Following the analysis of the data from the three subcategories there were 72 critical incidents identified relating to the negative aspects of transitioning to the adult hospital. As with the positive aspects explored earlier in this chapter, these incidents were labelled and classified into subcategorised groups before being categorised into similar themes.

These will now be explored in greater detail within each of the three subcategories.

Subcategory 1: Adolescents with severe haemophilia.

The second question the adolescents were asked was:

“Can you tell me if you have any worries about moving to the adult hospital?”

There were fifteen critical incidents identified by the adolescents as relating to the negative aspects of transitioning to the adult hospital. These were subcategorised as to pertaining to either:

- Lack of Familiarity
- Older Clientele

1. Lack of Familiarity

Most of the adolescent group stated that lack of familiarity with the adult hospital was a negative aspect to transitioning. These adolescents have attended this children’s hospital since their diagnosis as infants. As a result, they now know the hospital very well, and the routines associated within the various wards, canteen, outpatient’s department and car parking facility. They expressed their concern about leaving the familiar surrounding when they stated that:

“I won’t know anyone in the new hospital” (A1)

“...I wouldn’t know my way around” (A4)

“I could get lost (in adult hospital)” (A2)

They also mentioned the fact that they will not know any of the MDT team there as another negative aspect to transitioning to the adult hospital. The core haemophilia care MDT in the children's hospital have been in their posts for ten years or more. Consequently, the adolescents will not have witnessed a dramatic change in the MDT throughout most of their time in the children's hospital.

"I'm concerned about dealing with different staff" (A1)

"There'll be different doctors and nurses" (A2)

"I know the staff here, but I won't know any of the staff there" (A3)

The adolescents also know the hospital uniforms and can easily identify any staff member grade when they pass by. In the adult hospital it will take some time to recognise who is, for example, a staff nurse and who is a physiotherapist. In the children's hospital, the adolescents will have built-up long-term relationships with the staff there, often being able to name many of the MDT members on sight. Although it was not explicitly expressed during the interviews, the student believed that the role of the play specialist will be missed by the adolescents when they transition to the adult hospital. The play specialist plays a vital role in the MDT as they provide age appropriate assistance and distraction techniques during difficult procedures such as cannulations for example.

2. Older Clientele

Although participant A3 stated that mixing with older people was not disconcerting, two of the adolescent group mentioned that being around older clientele in the adult hospital was a concern to them.

"I'll be staying in a ward with loads of other people much older than me" (A1)

"There'll be older people there" (A2)

The adolescents are now used to being the senior members of the haemophilia patient cohort in the children's hospital. They understand their hospital care and of how the children's hospital works. It may be difficult for them to move to the adult hospital where they are the youngest, least experienced and are also unsure of their new surroundings.

In relation to the views of the adolescents towards their transition to the adult hospital, the younger adolescents interviewed expressed fewer concerns towards transition than the older adolescents. The student perceived this was because the younger adolescents are more reliant on their parents to organise and plan their care at this stage. The older adolescents are more proactive with their care and are more aware of the intricacies of organising hospital and home care. This is as their parents will be increasing their level of independence with their own care in line with their upcoming transition.

Subcategory 2: Parents of adolescents with severe haemophilia.

The second question the parents were asked was:

"Have you any concerns in relation to your child transitioning to the adult hospital?"

The parents identified 22 critical incidents as being the negative aspects of their child transitioning to the adult hospital. The negative aspect to transition to the adult hospital as expressed by the parents can be categorised in one word as: apprehension.

1. Apprehension for their adolescent.

The parents seemed particularly apprehensive about their adolescent moving to that specific adult hospital. The adult hospital may signify serious illness and death which has been faced by previous people with haemophilia who attended that hospital. Some sentiments which were expressed included:

"For myself and my family the adult hospital does not hold good memories"
(P2)

“They will face people at the end of their lives. It’s expected, but I really don’t want him to see that” (P2)

Another area of apprehension for their adolescents attending the adult hospital relates to the parent’s perceptions of that hospital as being very busy and overcrowded. The hospital having a cohort of patients with addiction issues was mentioned by some parents as being worrisome.

“Mixing with all sorts in the adult hospital...looking for something better than the children’s hospital will be hard.” (P3)

“You’re throwing them into a very adult environment in the likes of that hospital” (P1)

Another area which caused concern was the fact that, unlike in the paediatric hospital, in the adult hospital a parent will not be permitted to stay with them overnight.

P1 said that:

“He is going to have to stay on his own in an adult male ward with a mixture of people”.

P2 expressed similar concerns when they said

“I can’t stay with him and will have to leave him alone with grown men”.

2. Apprehension for parents.

Some of the parents mentioned that they will find it difficult adjusting to their new role in their adolescent’s care when they transition to the adult hospital. In the children’s hospital family centred care is paramount to the care of the adolescent .A parent can stay overnight, accompany the adolescent to theatre until they are anaesthetised, and must be present for any review with the MDT .It will be difficult for the parent to relinquish their control in their adolescent’s care when they have been involved in every decision made for the adolescent up until this point.

“Will be hard to give up my control as a parent” (P4)

“I’m more concerned than looking forward to it” (P1)

Some of the parents also mentioned the fact that they would have concerns about their adolescent’s compliance attending the adult hospital when the parent is no longer necessary in the organisation of appointments. One parent stated that it will be very difficult not to be able to call their adolescent’s team in the adult hospital to find out how he is progressing.

Subcategory 3: Multidisciplinary Team members working in haemophilia care.

The second question the MDT group was asked was:

“In your role have you any concerns in relation to the current transitioning process of an adolescent to the adult hospital?”

With the MDT group, there were 35 critical incidents relating to the negative aspects of the transition process as viewed by this group. These negative aspects to transition were found to be related to:

- Compliance
- Communication

1. Compliance

The issue of compliance to the transition process by the adolescents was mentioned frequently by the MDT during the interview process. Most of the MDT were concerned about how the adolescents would comply with their treatment once their parents are no longer there to focus them on their hospital care. Incidents discussed by the MDT relating to the compliance of adolescents to the transition process included:

“Adolescents may lose interest when they are half way through the programme” (MDT2)

Similarly, MDT1 found that:

“Some don’t follow up-, sometimes adolescents do not return phone calls”

While in the children’s hospital the parent will be the primary contact, but once the adolescent turns 16 years old they can also be involved with MDT scheduling of care. If the adolescent does not return the MDT’s phone call from the adult hospital it is believed that it is less likely that the MDT will continue to repeatedly try to contact them. Therefore, it is a concern for the MDT if the adolescent does not engage at this stage when they are attempting to contact them.

While the MDT quoted above suggest that some adolescents do not engage in the process of the transition, other MDT also suggest some possible reasons for their lack of compliance:

“Hard for them to have to take on a more independent role” (MDT3)

“The psychosocial background of the child is a critical predictor of how well it goes” (MDT7)

All the adolescents differ in their maturity level and willingness to engage in the transition process, they are experiencing many transitions such as preparing to finish school, not just transition to the adult hospital, during this time.

2. Communication

Pertaining to communication issues found during the review of the transcripts, several MDT mentioned that they were not always aware of the final meeting with a transitioning adolescent.

“Sometimes I only find out on the day that it’s their last visit” (MDT4)

“As an MDT, I am not always aware when someone is transitioning” (MDT5)

The opinion gathered by the student from these interviews was that the MDT would appreciate more communication between the haemophilia MDT

members to ensure that all relevant data is shared with all members more regularly. Another MDT stated that it would be good if parents knew in advance of the day the transition programme begins, as some only find out on the day it begins (MDTP1).

Communication between the MDT and adolescents is another area which was mentioned frequently as a concern by the MDT.

“...a lot of people think of transition as something that happens to them rather than something that happens for them!” (MDT7)

This suggests that some of the adolescents may view transitioning to the adult hospital negatively. The view that the adolescents who are actively engaged in the transition process are more likely to have a successful transition was mentioned by several MDT. It was also reiterated that those who were reluctant to engage in the transition process were the adolescents who would probably require more assistance with the process.

“The ones that turn up are the ones who know about their condition. The ones that don’t are the ones that you’d have issues with” (MDT2)

Similarly, MDT4 suggested that:

“If I could be critical about it, then the process could be more formal with checklists of competencies reached so we know when they’ve reached them”.

It was also suggested that current documentation could be improved.

“The current documentation is very broad- it should be more age appropriate, 12 to 15 years and 15 to 17 years old”.

By adopting a more structured programme each adolescent would have to complete a certain level of competency before they could move on to the adult hospital.

The MDT also acknowledged that, as they have treated the adolescent throughout their childhood, it can be difficult for them to change how they treat them.

“It’s hard to go from treating them as a child to treating them as an adult- can be a bit tricky” (MDTP1)

Following a comprehensive review of all the data from the three groups which the categories emerged, the main themes from the negative views of transition were classed as being:

1. Apprehension

The categories of lack of familiarity and older clientele showed that the adolescents expressed some level of apprehension when it came to transition to the adult hospital. The parents’ main concern relating to their adolescents’ transition was apprehension.

2. Communication.

Some of the adolescents showed a concern towards having to communicate with new staff, and one of the parents was concerned about not having the same level of communication with the medical team as they would have before. The MDT conveyed several concerns associated with communication.

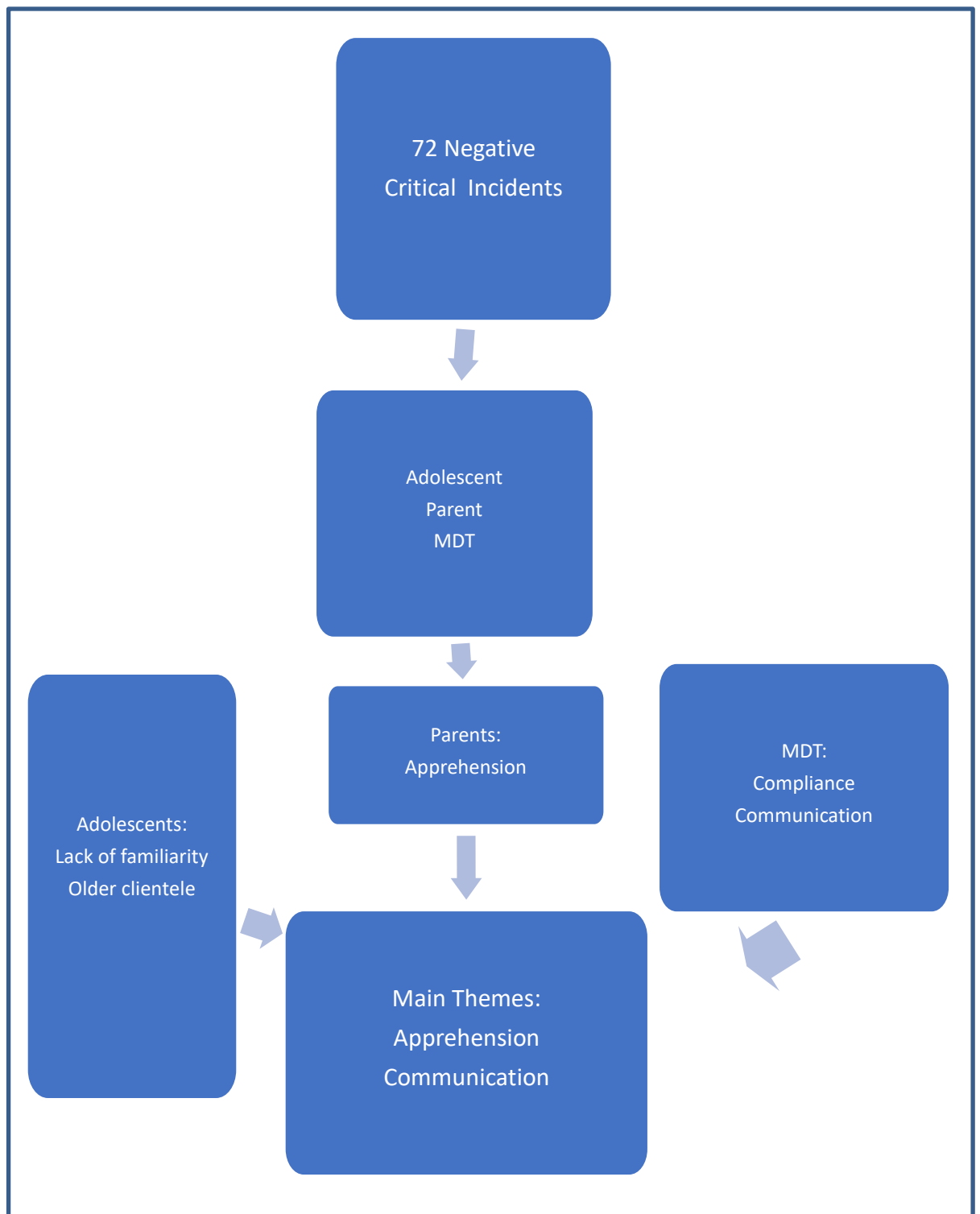


Figure 5 Steps of data analysis in accordance with the Critical Incident Technique (Flanagan 1954) Negative Aspects.

4.4 Interpreting the data.

This study aimed to examine the views of adolescents with severe haemophilia and their caretakers in relation to their transition from child to adult services. By including parents as well as multidisciplinary team members alongside the data collected from the adolescents with severe haemophilia, it provided a multidimensional view of haemophilia care for this patient group. It has been shown that there are many positive aspects to the transition process as it stands, but also several areas which could be improved. Both the positive and negative findings from this study are valid research data findings.

4.5 Conclusion

This chapter showed how the data was analysed using the stages of data analysis for CIT. The main themes from the positive views of transition were identified as were the negative. In the following chapter these findings will be further discussed with reference to how these findings can be interpreted.

Chapter 5 Discussion of Findings

5. Introduction

When data analysis is complete, there is a feeling that the study is finished, but there remains a need to interpret results (Gray 2017). This chapter discusses the key findings from this qualitative study. It reflects on the study results within the context of existing literature relating to treatment of adolescents with severe haemophilia and their caretakers.

To recap on the study results, the completion of 15 semi-structured interviews using CIT methodology revealed the main positive effects relating to transition from child to adult services as: a change in independence, meeting peers and being prepared for change. The negative effects to transition were identified as: apprehension and communication. This chapter has been divided into five sections, each section focusing specifically on one of the main themes identified during data analysis. While it has been noted earlier that there is a limited amount of data available on the views of the haemophilia community in relation to transition of care, the student will now discuss the findings from studies specific to other chronic conditions as well as haemophilia.

5.1. Independence

Haemophilia knowledge and education begins in early childhood, and self-infusion skills are taught by the MDT later in childhood (Khair et al 2013). This system also occurs in the hospital where this study took place. By ensuring that early planning of transition to adult services occurs, a gradual shift in responsibility and the ability for the adolescent to become independent with care will take place (Brand et al 2015).

This study showed that some of the adolescents were looking forward to the increased level of independence which would come with transition to the adult centre. Similarly, the results of studies by Nguyen et al (2016) and Babler & Strickland (2015) showed that adolescents in their studies welcomed their increased ability to self-manage. Nguyen et al (2016) found that the youth in their study reported that they had begun to manage their medical need independently in preparation of transitioning to adult services. Hislop et al (2016) found that some had a desire for withdrawal of parental

involvement in their care and increased independence. They felt that they were developmentally ready for a move from a paediatric environment with teddy bears and toys. Corbett et al (2011) also found, in their study relating to the transition pathway for diabetes that adolescents felt out of place in a children's unit as they got older. Some of the adolescents in this study also identified with the child-based environment as a reason why they were eager to transition to adult services. A MDT participant also stated that the childlike surroundings were much undignified for adolescents.

Babler & Stickler (2015) detailed that along with seeking to manage their chronic illness independently, the adolescents became aware that this can also be frustrating and challenging. The participants in this study did not vocalise any concerns about increased independence currently causing any difficulties.

5.2 Meeting Peers

Meeting peers was another theme identified during the analysis of the data. In the study by Hislop et al (2016), which focused on the views of young people with chronic health conditions transitioning from child to adult services, they found that their participants felt happier around people with the same condition and viewed that as productive to their care. That study does not mention specifically the age group of people with the same condition, but one parent in this study felt that it would be beneficial for the adolescents to meet older people with the same condition to see how they manage their care. It was a concern from one of the adolescents that they may be in a room with much older people, and another parent expressed concern about the fact that in the adult hospital the adolescent would be mixing with all types of patient.

5.3 Preparing for change

In this study, one parent stated that they were more concerned than looking forward to the transition to the adult centre. Meanwhile, in the study by van Staa (2011 B), they found that the adolescents did not seem overly worried about the prospect of transition and had developed a more relaxed attitude towards the event. Adolescents did however comment that transition should

be more focused on them while understanding how difficult it can be for a parent to let go (Aldiss et al 2015). The necessity for parents to be included in the transition process has been documented in numerous studies including van Staa et al (2011A) Huang et al (2011), and Aldiss et al (2015). Huang et al (2011) also emphasise the importance of the adolescent, parents and MDT being involved through the whole transition process to ensure a successful outcome. The transition programme as it currently stands in the student's clinical area includes the involvement of parents and the MDT during the transition process. While it was detailed by McManus et al (2013) that most adolescents were not receiving preparation for transition, this is not the case in the student's clinical area.

Preparation for transition during years preceding the actual process is deemed very important to the successful transition of the adolescent in studies by Chaudhry et al (2012), Nguyen et al (2016) and Croteau et al (2016). It was also found that those who received preventative healthcare, such as being involved in a transitional programme, were more likely to successfully transition from child to adult services than those whose preventative healthcare had been delayed or had not taken place (Oswald et al 2012).

5.4 Apprehension

All three study groups voiced some level of apprehension when it came to the transition process. Some of the adolescents explained how they would miss the staff and familiarity of the children's hospital, a finding which is also addressed in the study by Garvey et al (2013) examining transition for young adults with diabetes. Rutishauser et al (2013) also identified the long-term relationship that adolescents and parents have with the children's hospital MDT as a barrier to transition in their study exploring the readiness of adolescents with chronic conditions to transition. A parent in this study felt that the MDT were less strict and more accommodating than they would be in the adult hospital. Adult health care differs significantly from paediatric care in the type and level of support as well as family involvement (Davies et al 2011) and this finding was also reiterated by participants from the three groups as a cause for concern.

It was also suggested that it can be a challenge for both parents and MDT to relinquish responsibility for the adolescent's haemophilia management by Brand et al (2015) This was confirmed as a challenge by those groups in this study. The views of one MDT stated that the transition programme has a "one size fits all" approach when it doesn't really. This view is also reiterated by Dogba et al (2014) who stated, in their study examining transition for those with Osteogenesis Imperfecta, that a one size fits all approach to transition would be inappropriate. They suggest that each transition programme should be evaluated to create a theoretically sound individualised programme.

5.5 Communication

It is emphasized by van Staa et al (2014) that the health care provider's priority and main responsibility is to ensure a safe and smooth transition from paediatric to adult care. The results of this study showed that the MDT participants believe that the existent transition programme provides an individualised approach for each adolescent, and that this process occurs over a number of years. This is in line with studies by Kime (2012), examining transition for those with diabetes and Mc Quillan et al (2015), examining transition from renal care, who explained that transition from the child to adult service should not be a rushed process. It was also felt by Kime (2012) that more communication between the child and adult services was required for the adolescent during this period. While the student was able to attain the views of the MDT in the children's hospital, it was not possible to ascertain the views of the adult centre MDT. Garvery et al (2013) found that the lack of contact details for the adult hospital for those transitioning from the children's hospital may cause a gap in their care, but this was not addressed as a concern with the participants in this study. Studies on the transition process by Oswald et al (2012), Fegran et al (2014) and Szanda et al (2016) all emphasise the role of the nurse in this process. Oswald et al (2012) found that nurses play a very important role in transition both as facilitators and careful listeners. Similarly, Fegran et al (2012) state that nurses can facilitate individual transition plans, while Szalda et al (2015) explain that the paediatric nursing team can assist the adult team in relation

to the preparation of the adolescent prior to transition from the children's hospital. Parents noted that the paediatric staff addressed holistic needs of the adolescent more than the adult hospital in the study by Levy et al (2014) relating to transition. However, as it was not possible to include the views of the adult hospital MDT in this study as first expected, the student cannot include the views of this group. Adolescents shared the view that they say they have no concerns pertaining to transition in order to keep their parents calm in a study by Babler & Strickland (2015) but the adolescents in this study did not express this outlook.

5.6 Conclusion

In conclusion, this chapter discussed the findings of the study data under the themes identified during the data analysis. The views of the participants were included as were the results of other studies examining the transition process for those with chronic childhood illness. As this chapter has shown, these themes have been identified in other studies relating to transition from child to adult services from other childhood chronic illnesses. A thoughtful approach to the transition process is necessary for all the healthcare teams for successful transition to take place for these adolescents (Croteau et al 2015). The next chapter is the final chapter in this document and will discuss the strengths and limitations of the study as well as implications for practice, and how the study details will be disseminated.

Chapter 6: Conclusions and Recommendations

6. Introduction

This chapter will explore the strengths and weaknesses of this study and will then discuss the implication for practice from the viewpoint of the three study groups who participated. Recommendations for further research on this topic are then detailed followed by the aim of disseminating the findings. The student then details her own personal reflections on completing this study and concludes the chapter with her final views on this study.

6.1 Strengths and Limitations

Strengths

There are several strengths to this study.

- The first is that it obtained ethical approval from separate ethics committees prior to it being employed.
- Another strength is that, as shown in the literature review chapter, there is a distinct paucity of research completed with this study group in Ireland, and this study aimed to address this.
- The Critical Incident Technique (CIT) has seldom been used with school age children and their parents (Kelo et al 2012) so this study confirms that this qualitative research method is suitable for these research groups.
- The CIT recognises the potential for the excessive response burden on participants, so its interview technique consists of a simple set of procedures which are easy to follow (Yonas et al 2013).

Limitations

A limitation is described by Burns & Grove (2011) as a restriction in a study which may result in decreasing the credibility of its findings. The researcher is responsible not only for illustrating the value of the research findings but also its limitations (Flanagan 1954). There are number of potential limitations in this study.

- One limitation of the study would be that it was confined to a single site. This site is the main source for paediatric patients with severe

haemophilia, but there are other paediatric centres throughout the country which care for a small number of this patient group.

- Another possible limitation of this study is the homogeneous nature of the group, while this cannot be altered, it would be interesting to find if a more heterogeneous group shared the same concerns. Kontio et al (2011) mention the fact that their research participants came from the same cultural and demographic background as a limitation, and as a result, the student is including it here as a possibility.
- In line with hospital policy, adolescent interviews took place with the parent present. The parent in the group was interviewed on the same occasion. In a similar CIT study with parents and adolescents, Getrich et al (2016) suggested that conducting separate interviews for adolescents and parents would enhance disclosure and comfort levels. Therefore, by conducting the interviews with the adolescent and participating parent on the same occasion it could possibly be viewed as a limitation to this study.
- Although the response rate from MDT members was high (87.5%), the response rate from the adolescents and parents was low (17.5%). Therefore, it cannot be categorically stated that data saturation occurred in these groups as additional participants may have provided currently unknown data.
- It was originally planned to interview several MDT from the adult hospital, as well as PWH who had transitioned there within the last five years. Some internal changes prevented this from taking place, so this study was not as inclusive as originally designed.
- Since Critical Incidents rely on the ability to recollect events, incidents may be distorted if data is collected a long time after the event. While this study was examining the views on transition as the critical incident, and not recalling a specific occurrence, it is less likely that will be a limitation. However, it should still be included as a possible limitation.
- A limitation to the study involves the lack of recording of the interviews, as was requested by the participants. The possibility of

recall bias as well as the reliability of the student's field notes should be noted as possible limitations.

- The confidentiality of the participants was of paramount importance to the student. All study participants were made aware during the consent and assent process that names or identifying information would not be shared, but their words and views would be published. However, in a highly specialised area with a small population, their identity could possibly be deducted so the author has made every effort to remove any opportunities that could result in this occurring.
- Another limitation described by Chuan et al (2013) was that the participants may only divulge data which show examples of good clinical judgement. The student has no way to confirm or deny that this may have happened.
- Also, the student had not requested, as part of her ethical approval application, to ask those who didn't respond for their rationale as to why they did not to take part. This would have been useful data to collect if it had been possible to ascertain.

6.2 Implications for Practice.

The goal of completing this research was to ascertain the views of adolescents with haemophilia, parents of adolescents with haemophilia and multidisciplinary team members. As their views relating to transition to the adult hospital had not previously been examined here in the Republic of Ireland, it was critical to explore the impression of these groups towards this topic.

6.2.1 Implications for Adolescents.

Transitioning from a paediatric to adult hospital can be challenging for those with chronic diseases (Croteau et al 2015). An adolescent with haemophilia must gradually be introduced to taking responsibility for their own care and haemophilia management (Breakey et al 2014). Current practice allows for the completion of pre-transition training to take place in the children's

hospital with the adolescent, MDT and parent. This includes the adolescent becoming more aware of his treatment regime as well as ordering factor and other medical supplies by himself.

The results of this study found that the adolescents were both eager to embrace increased independence but nervous about their lack of familiarity of the adult hospital. All the participants were aware of the requirement for them to transition to the adult hospital when they reached adulthood. They were eager to move away from a children's hospital which they were outgrowing but were not fully aware of the protocols in place in the adult hospital. These results can be used to repeat the need for more education for the adolescent before they transition away from the children's hospital.

6.2.2 Implications for Parents.

It has been shown that not just the adolescent, but the parents and family members also need resources and information on transition to support their adolescent (Gorter et al 2011). During the transition process in the children's hospital a parent or nominated family member will always be present during any transition information or training. How the parents cope and manage with their change in role when their adolescent is transitioning to the adult hospital will impact on the adolescent as well as their own well-being (Heath, Farr & Shaw, 2016). This was one of the main reasons why the study was designed to include the parents of adolescents with haemophilia.

The views expressed by the parents of adolescents with haemophilia in this study included their enthusiasm to see the adolescents show a greater level of responsibility. They were also eager for their adolescent to have the chance to meet people of all ages with haemophilia in the Adult hospital. They were however apprehensive that their adolescent will be alone in the Adult hospital. In the Children's hospital a parent can stay overnight with them.

6.2.3 Implications for the Multidisciplinary Team.

Patients want to see a familiar face during transition, it was stated that this promotes a successful transition (Annunziato et al 2013) The professionals provide the practical aspects of psychological support for the patients throughout their treatment period in the children's hospital (Garcia -Dasi et al 2016).

In the children's hospital, the patients will have been treated by the multidisciplinary team (MDT) members for many years. A larger proportion of this MDT has remained unchanged for many years, so they will have built up a good rapport with each other. The results from this study show that the MDT main positive response to the transition of the adolescents, is their view that they are well prepared for the change. Each expert area such as Clinical Nurse Specialists (CNSp) has approved paperwork detailing the protocols involved with preparing the adolescent for transition. The participants were confident that by the time the adolescents were about to transition to adult services that they would be competent with self-care. It was voiced by several participants that the consultant would not allow them transition unless they were proficient with their own care. With relation to their negative findings of the transition process for adolescents, the MDT had two main concerns. The first one was their compliance to treatment once they were independent with their own care, and without a parent to ensure they were administering factor as required. Their other concern was with the level of communication the newly transitioned adolescent would have with the adult hospital. Unlike with the children's hospital, the adolescent will not have a parent to ensure they attend their out- patient's appointments, or order treatment. It is also less likely that the adult hospital will be contacting them prior to scheduled appointments or if they miss an appointment.

Recommendations for improving clinical practice would include increasing staff communication throughout the MDT so everybody is aware when the adolescent is leaving the children's hospital. Communication could also be improved between the adult and a children's hospital, so the adolescents and families are more aware of the adult hospital before they transition.

6.3 Suggestions for Further Research.

In this section of the chapter, suggestions for further research will be discussed. The completion of this research study was in line with the recommendations outlined in “The National Rare Diseases Plan for Ireland 2014-2018” (Department of Health 2014). This document highlighted the need for an increased number of qualitative designed studies to take place within the rare diseases’ population in the Republic of Ireland, of which haemophilia is one. While completing a literature review on Transition from child to adult treatment centres for those with haemophilia, the student was made aware of the paucity of published materials on this topic globally. There was also nothing published specifically on this topic in the Republic of Ireland. By providing new evidence on the thoughts and views of these study groups it is envisaged that it can inform new practice and policies. Other research questions which could suggested for further research could include:

- Vein training for children with haemophilia.
- Family support when there is no prior family history of haemophilia.
- Sibling groups with haemophilia, what are the pros and cons for each brother?
- Female siblings of a child with haemophilia. What are their factor levels and does it influence their life?

Rarely are the views of the adolescent or their families sought regarding the outcome of the transition process (Chaplin 2016). This is one reason why the student would like to see some collaborative research studies between the paediatric and adult hospitals take place. Some research questions relating to collaborative studies could include:

- The role of mentorship between adults with haemophilia and adolescents with haemophilia.
- Sports and exercise for PWH, does it increase or decrease with age?

This would allow for an all-encompassing view on topics to be explored. Research studies involving The Irish Haemophilia Society (IHS) could also

explore areas on haemophilia care such as peer support, and possibly other non-hospital based topics.

6.4 Dissemination of Findings.

Although research may be both intellectually satisfying and interesting to complete, there is little point in carrying it out if it is not disseminated to those who can make use of the newly generated knowledge (Gerrish & Lathlean 2015). The findings from this research piece will be shared in a poster format at the annual research days for the paediatric Hospitals as well as for the third level Institution. It will also be shared in the student's place of work. The student will also be able to share poster formatted information at national and international conferences such as ISTH (International Society on Thrombosis and Haemostasis) and EAHAD (European Association of Haemophilia and Allied Disorders). These findings will also be presented orally in the students' place of work. This will take place at department meetings as well as at the annual transition workshop for adolescents with haemophilia. The student will also present these findings at an Irish Haemophilia Society meeting. The main aim is to disseminate the data by publishing a piece in a high impact journal soon. Examples of journals which specialise in the area of haemophilia include "Haemophilia", "The British Journal of Haematology" and "Journal of Thrombosis & Haemostasis".

6.5 Personal Reflections of this Study.

Throughout the duration of this research study, I found maintaining a specific site for both hard and soft data storage to be invaluable. This allowed for prompt access to data throughout the two-year college period. The importance of maintaining up to date back up to soft data was something I was also very conscious of, having previously lost data completed for another third level course.

As I am employed in clinical research, this prepared me for the application to the various Ethics boards as required prior to the data collection. Having previously completed a master's degree, I had an idea of what to expect with this course, but the level of individualised support and guidance from my

academic supervisors here was paramount and greatly appreciated. The monthly group meetings with other post graduate students provided a marvellous opportunity to speak with others about research queries in general and was very beneficial. The opportunities provided for me to meet with visiting professors at the college and receive their feedback on my project was also a great privilege.

I was disappointed by the limited uptake of the parents and adolescents to partake in the study. I had posted out individual letters to all the adolescents and parents who met the inclusion and exclusion criteria, but only received confirmation of participation when they arrived at their scheduled Out Patient Department visit. Notwithstanding I managed to collect valid and useful data from these study groups. I was also discouraged by the inability to complete data collection from the adult hospital as detailed in the study design. There was nothing I could do to change the outcome of the adult hospital participation, but I found it disheartening as I had received approval from the Ethics board there to complete the study. Another change to the original study design was the data collection method. It was planned to use a Dictaphone to record the interviews and transcribe them once the interview had ended. However, I was informed by various people from within the sample groups that there was a reluctance to take part in a study which would be recorded. I returned to the Ethics board to amend the original consent data to include the phrase that interviews were not required to be recorded. Of the fifteen participants, no one favoured to have their interview recorded.

Overall, I have thoroughly enjoyed my time here completing this Master's Degree course. While there were some knock-backs they were not insurmountable with the support and guidance of college supervisors, fellow post graduate students as well as from my clinical supervisor. I will remain grateful for the opportunity to enhance my research skills which have been provided by the completion of this course.

6.6 Conclusion.

In conclusion it is important to reiterate the need for qualitative research studies to assist with the care of those with haemophilia. The development of evidence-based paradigms for health care transition for both paediatric and adult providers in chronic illness is critical (Garvey et al 2013) Similarly it has been shown that ensuring a timely and well managed transition from paediatric to adult focused care is a compelling component of high-quality health care for youths with chronic conditions (Wisk et al 2015).

While it has also been found that creating an organisational climate which values and supports the clinical nurses' participation in research is crucial for the further growth and development of this activity (Akerjordent et al 2012), improving the outcome for adolescents with haemophilia and their families will always be the main rationale for completing this study, as the good management of a chronic condition can ameliorate or eradicate the complications they may face (Nazzaro et al 2006).

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Appendices:

Appendix 1: Literature Search.

No.	First Author	Country	Health condition	Study design	Search Period	Groups	Data collection method	Prep.	Outcome	Barriers	Facilitating factors
1	Aldiss 2015	UK	Chronic disease	Qual	Oct 2011-Dec 2013	Adolescent Parent Professionals	Focus groups, workshops	*			
2	Sawicki 2015	USA	Chronic disease	Quant		Adolescents	Survey	*			
3	Fair 2010	USA	HIV	Qual		Transition staff	Interview	*		*	
4	Sawicki 2014	USA	Chronic Disease	Quant	2009-2011	Youth Parents	Surveys	*			
5	Simmons 2014	USA	Haemophilia	Qual		Adolescents with haemophilia parents	Focus Groups	*		*	
6	McManus 2013	USA	Chronic Disease	Quant	2009-2010	Parents	Survey	*		*	
7	Okumura 2015	USA	Chronic Disease	Qual		16-25 year olds Parent	Interview	*		*	*

						Professionals					
8	Moynihan 2014	Canada	Chronic Disease	Quan	April- June 2012	12-19 (200)	Questionnaire	*			
9	Mc Quillan 2015	Canada	Renal Transplant	Qual	Pre & post 2009	Adult (16) 17 (16)	Interview	*		*	*
10	Chaudhry 2013	USA	Cystic Fibrosis	Quan	Jan- Dec 2010	91 adults	Questionnaire		*	*	*
11	Garvey 2013	USA	Diabetes	Quant		Adults 22-30	Survey		*	*	
12	Levy 2014	UK	Spina Bifida	Qual		12-25 years N=12	Questionnaire	*	*	*	
13	Jensen 2015	USA	Rheumatology	Quant		16+ years	Questionnaire		*	*	
14	Szalda 2015	USA	Quan	Quant	Marc h-Aug 2011	Providers=22	Interview			*	*
15	Rutishauser 2011	Switzerla nd	Chronic Disease			298 adolescents 331 Parents	Questionnaire	*		*	
16	Woodward	USA	Chronic	Quant			Survey		*	*	*

	2012		Disease								
17	Kime 2012	UK	Diabetes	Quant	Over 3 years no date	N=300 no ages			*	*	*
18	Huang 2011	USA	Chronic Disease	Quant	2006- 2009	10 YA 24 Staff	Focus groups		*	*	*
19	Milnes 2013	UK	Asthma	Qual		14-18 Y Practice Nurses	Interview			*	*
20	Coyne 2016	R.O.I	Chronic Disease	Qual		AYA 14-25	Postal survey			*	*
21	Babler 2015	USA	Diabetes	Qual		11-15 yr				*	*
22	Mackie 2014	Canada	Heart Disease	Qual	Jan 2010- Nov 2012	15-17 yr	Questionnaire			*	*
23	Wells Collins 2012	USA				Paed hospitalists	Survey	*			

24	Byrant 2011	USA	Hemoglobinopathy	Qual		8 female 6 male 19-25	Interviews		*	*	*
25	Van Staa 2011	Netherlands	Chronic Illness	Qual		24 15-24 yrs 24 Parents 17 staff				*	*
26	Begley 2013	R.O I	Chronic illness	Qual		Consultants CNS ANP	Postal survey	*		*	
27	Nguyen 2016	Canada	Chronic illness	Qual	Marc h- April 2012	11 youths 18- 21 7 parents 7 healthcare providers	Telephone interviews			*	*
28	Ishizaki 2012	Japan	Chronic illness	Quant		Paediatricians = 41 Nurses =24	Questionnaire	*		*	
29	O'Sullivan	USA	All illnesses	Qual			4 focus groups			*	

	2014										
30	Van Staa 2014	Netherlan ds	Chronic illness	Qual		N=518 18-25 yr Pre & post	Survey		*	*	*
31	Van Staa 2011	Netherlan ds	Chronic illness	Quant	Oct- Dec 2006	12-19 yrs N=1087 Non response 64%	Questionnaire	*		*	*
32	Rutishauser 2014	Switzerla nd	Chronic illness	Quant		283 pre 89 post 14-25 r	Questionnaire	*	*	*	*
33	Ciccarelli 2015	USA	Chronic illness	Descriptive summary of transition support programme	2013	Primary care n=81 28.7% response Families n=82 30.7% response	surveys			*	*
34	Hislop 2016	UK	Chronic illness	Qual & Quant		14-22 Pre & post N=44			*	*	*
35	Fair 2011	USA	HIV	Qual		Medical providers & social workers	Interviews				*

						N=19					
36	Sterling 2013	Canada	Haemophilia	Qual	Jan- April 2010	12-19 yr N=11	Interviews	*		*	
37	Chandra 2015	USA	Liver Transplant			18-23 yr 39% Parents 61%	Survey Response 88.5%		*	*	*
38	Davies 2011	Canada	Neurological	Qual		17 parents 11 YA 18-21yr			*	*	*
39	Gorter 2015	Canada	Chronic illness	Mixed Methods		50 (47%) Post	Interviews		*	*	*
40	Oswald 2013	USA	Chronic illness			19-23 yr	Survey		*	*	*
41	Dogba 2014	Canada	Oetogenesis Imperfecta	Evaluation of programme	April –Oct 2013	Staff Paed & Adult services	Interview		*	*	*
42	Price 2011	UK	Diabetes			N=11 YP 16-25 yr	Interview		*	*	*
43	Sparud- Lundin 2017	Sweden	Chronic illness	Quant		Physicians Nurses Response 38%	Survey			*	*
44	White	R.O.I	Chronic illness	Quant	30	N=217 25%	Survey		*	*	*

	2017				Nov-30 Nov 2012						
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Appendix 2: Parental/Guardian Consent.

Appendix 2.

Consent Form: Parent

Reference Number:

Title of Study: Views on transition of adolescents with severe haemophilia to adult services.

Research Participant's Name:

1. I have had time to consider whether to take part in the study. My questions have been answered satisfactorily and I have received a copy of the Parent Information Leaflet.
2. I understand that my participation is voluntary and that I am free to withdraw at any time without my child's medical care or legal rights being affected.
3. I understand that all audio, written and digital data will be destroyed following the completion of the study in September 2018.

_____	_____	

Name of Parent/ Guardian	Date	Signature
_____	_____	

Researcher	Date	Signature

1 copy for research participant; 1 copy for researcher; 1 copy to be filed in the hospital notes.

Appendix 3: Assent Form 12-15 Years

Appendix 3.

ASSENT FORM 12-15 YEARS

Reference Number:

Title of Study: Views on transition of adolescents with severe haemophilia to adult services.

Research Participant's Name:

Please circle all you agree with:

Have you read the information form (or had it read to you)?	Yes / No
Has the researcher explained this study to you?	Yes / No
Do you understand what this study is about and have you asked all the questions you want?	Yes / No
Are you happy to take part in this research study?	Yes / No

If any answers are "no" or you don't want to take part, don't sign your name!

If you do want to take part in this study, please write your name and today's date below.

Your name: _____

Date: _____

The researcher who explained this study to you needs to sign too:

Print Name: _____

Sign: _____

Date: _____

When completed, I will get a copy of this signed and dated paper:

1 for the participant; 1 for the research site file; 1 original to be kept in medical notes

Appendix 4:

Assent Form 16–18 Years

Appendix 4:

ASSENT FORM 16–18 YEARS

Reference Number:
Title of Study: Views on transition of adolescents with severe haemophilia to adult services.
Research Participant's Name:

Young person (or, if unable, parent(s)/guardian(s) on their behalf) to circle all they agree with:

Has the study doctor or nurse explained this study to you?	Yes / No
Do you understand what this study is about?	Yes / No
Have you asked all the questions you want?	Yes / No
Have you had your questions answered in a way you understand?	Yes / No
Do you understand it's OK to stop taking part at any time?	Yes / No
Are you happy to take part in his study?	Yes / No

If any answers are “No” or you don’t want to take part, don’t sign your name!

If you do want to take part in this study, please write your name and today's date below:

Your name: _____

Date: _____

The researcher who explained this study to you needs to sign too:

Print Name: _____

Sign: _____

Date: _____

When completed, I will get a copy of this signed and dated paper:

1 for the participant; 1 for the research site file; 1 original to be kept in medical notes

Appendix: 5 Ethics Approval letter 1

Appendix: 5: Ethical Approval Letter 1.

Views on Transition to Adult Services

Principal Investigators: Dr. Beatrice Nolan, Ms. Róisín Bradley.

Dear Ms Bradley

At a meeting which took place on 14th March 2017, the Ethics (Medical Research) Committee reviewed the above study.

The Committee suggested that the title be amended from "*Views on Transition to Adult Services*" to e.g. "*The Views of Patients with Severe Haemophilia Transitioning to Adult Services*"

The Committee suggested that this study be extended to include other healthcare professionals from other disciplines e.g. Physiotherapy, Social Work, Psychology and Pharmacy.

In relation to Question 3.1 (a) of the Application Form, the Committee requested that the answer be amended as consent will be required.

The Committee requested that the data be analysed, coded and encrypted in this hospital before being transferred to RCSI. The Committee requested that the data also be stored in this hospital.

The Committee request that the amended documentation be forwarded to this Office for review by the Chairperson.

The study was approved subject to the above.

Appendix: 6

Ethical Approval letter number 2.

Appendix: 6

31st March 2017

REC Reference: GEN/548/17

**The views of patients with severe haemophilia and their caretakers
towards transitioning to adult services**

Principal Investigators: Dr. Beatrice Nolan, Ms. Róisín Bradley.

Dear Ms Bradley

Further to our previous correspondence dated, 24th March 2017, in relation to the above study.

[REDACTED] Chairperson, Ethics (Medical Research) Committee, has reviewed and approved the updated documentation submitted by you following the meeting which took place on, 14th March 2017, at which this study was approved.

This completes our records.

Yours sincerely

[REDACTED]

Ethics (Medical Research) Committee

Appendix 7:

Ethical Approval letter number 3

Appendix: 7.

16th August 2017

REC Reference: GEN/548/17

**The views of patients with severe haemophilia and their caretakers
towards transitioning to adult services**

Principal Investigators: Dr. Beatrice Nolan, Ms. Róisín Bradley.

Dear Ms Bradley

[REDACTED] Chairperson, Ethics (Medical Research) Committee, at a review which took place on 10th August 2017 reviewed your request for an amendment to the above.

[REDACTED] noted that the amendment to the Ethics (Medical Research) Committee's approval of this study will include the fact that recorded interviews would not be essential and that you will take notes during the interview and allow the interviewees to review these notes prior to completion of the interview.

The Amendment was approved.

Yours sincerely

[REDACTED]

Ethics (Medical Research) Committee

Appendix 8:

Ethical Approval letter number 4

Appendix: 8

20th November 2017

REC Reference: GEN/548/17

**The views of patients with severe haemophilia and their
caretakers towards transitioning to adult services**
Principal Investigators: Dr. Beatrice Nolan, Ms. Róisín Bradley.

Dear Ms Bradley

██████████ Chairperson, Ethics (Medical Research) Committee, at a meeting which took place on 14th November 2017 reviewed the following and his comments were disseminated to the Committee:

- Amendment Notification Form dated, 19th October 2017 and signed by Dr. Beatrice Nolan;
- R. Bradley – Transition Research - Assent Form 12 – 15 Years – Version 1.1 – 1st November 2017;
- R. Bradley – Transition Research - Assent Form 16 – 18 Years – Version 1.1 – 1st November 2017;
- R. Bradley – Transition Research – Interview Guide 12 – 15 Year olds – Version 1.1 – 12th October 2017;
- R. Bradley – Transition Research – Interview Guide 16 – 18 Year olds – Version 1.1 – 12th October 2017;

Cont'd/

- R. Bradley – Transition Research – Interview Guide Parents - Parent Q – Version 1.1 – 12th October 2017;
- R. Bradley – Transition Research – Interview Guide – Those who have already transitioned to Adult Services - Parent Q – Version 1.2 – 13th October 2017;
- R. Bradley – Transition Research – Interview Guide Staff - Staff Q – Version 1.1 – 12th October 2017;
- R. Bradley – Transition Research - Consent Parent for Child - Version 1 – 15th October 2017;
- R. Bradley – Transition Research - Consent Parent - Version 1.2 – 15th October 2017;
- R. Bradley – Transition Research - Consent Adults already transitioned - Version 1.2 – 12th October 2017;
- R. Bradley – Transition Research - Consent Staff - Version 1 – 15th October 2017.

The Amendment was approved.

Yours sincerely

A large, irregular black redaction mark covering the signature of the Ethics (Medical Research) Committee member.

Ethics (Medical Research) Committee

Appendix 9: Ethical Approval letter number 5

Appendix 9: Ethics letter 5


10th December 2017

REC Reference: GEN/548/17

**The views of patients with severe haemophilia and their
caretakers towards transitioning to adult services**
Principal Investigators: Dr. Beatrice Nolan, Ms. Róisín Bradley.

Dear Ms Bradley

Further to our previous correspondence dated 20th November 2017, in relation to the above.

 Chairperson, Ethics (Medical Research) Committee, at a review which took place on 10th December 2017, noted and approved the following additional Information Sheets:

Transition Research – Information Sheet – those who have already transitioned to the adult hospital – V 1.1 - December 08, 2017;

R. Bradley – Transition Haemophilia Staff Information Sheet – 08.12.17 – V 1.1;

R. Bradley – Transition Haemophilia Information Sheet – 12-15 age – Version 1.1 – December 8th 2017;

R. Bradley – Transition Haemophilia Research Pt age 16 – 18 Information Sheet – 8th December 2017;

R. Bradley – Transition Research – Parent Information version 1.2 - December 08th 2017;

Yours sincerely,



Ethics (Medical Research) Committee

Appendix 10:

Ethical Approval from Adult Hospital.

Appendix 10: Ethical Approval from Adult Hospital.

From: Research & Innovation
Sent: 28 August 2017 12:04
To: Roslin, Bradley
Subject: Research & Innovation Office Approval

Dear Roslin,

This email is for your records and to confirm that your research application, reference number 2017/2014, "The Viena of Teleria with Severe Hemiplegia and Their Caregivers towards Transitioning to Adult Services", has been logged with and approved by Research & Innovation Office (former R&D Hub).

Regards,
[Redacted Signature]

Research & Innovation Office

Appendix 11: REC Letter from RCSI

Appendix 11: REC Letter from RCSI.

<p>Royal College of Surgeons in Ireland The Research Ethics Committee 121 St. Stephens Green, Dublin 2, Ireland. Tel: +353 1 4022205 Email: recadmin@rcsi.ie</p> <p>Dr David Smith, Acting Chair Dr Niamh Clarke, Convenor</p> <p>1st October 2018</p> <p>Ms Roisin Bradley, [REDACTED]</p>	 <p>RCSI ROYAL COLLEGE OF SURGEONS IN IRELAND COLAISTE RÍOGA NA MÁINLEA IN ÉIRINN</p>
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Ethics Reference No:	REC1576
Project Title:	Assessing the views of adolescents and their caretakers regarding their future transition to adult services
Researchers Name (lead investigator):	Róisín Bradley [REDACTED]
Principal investigator on the project:	Dr Beatrice Nolan [REDACTED] Dr Barry White [REDACTED] [REDACTED] Dr Declan Patton (RCSI School of Nursing and Midwifery) and Prof Zena Moore (RCSI School of Nursing and Midwifery).

Dear Roisin,
Thank you for your Research Ethics Committee (REC) application.

The REC notes that the study is complete and therefore cannot provide retrospective ethical review/approval for the study. However since the study received ethical review and approval from both Crumlin hospital REC and St. James's hospital REC the RCSI REC can only accept this review and approval

Please note it is the responsibility of the PI/ Data Controller/Joint Data Controllers to ensure and monitor compliance with relevant data protection legislation and regulation.

We wish you all the best with your research.

Yours sincerely,

PP Dr Niamh Clarke (Convenor)
Dr David Smith (Acting Chair)

Appendix 12: Interview Guide 12-15-year olds.

Transition Research Interview Guide 12-15-year olds

Thank you for coming to talk with me today. My name is Róisín Bradley and I work here in [REDACTED] as a research nurse. This interview should take about 20 minutes and I will be recording it so I can review it later for my notes, is that ok with you?

< START RECORDING IF VERBAL CONSENT IS OBTAINED >

< STOP INTERVIEW IF CONSENT IS NOT RECEIVED >

If you would prefer it not to be recorded, I will take notes during our talk. I will then read back my notes to you at the end of the interview. This is to ensure I have documented the interview in accordance to your views. I will amend my notes should you request me to do so.

I want to let you know that what we talk about is only being used for our research and your name will not be used. If you like you can pick a nickname that we can use during the discussion.

Before we start, have you any questions?

1. Can you tell me what you're looking forward to when you move to the adult hospital?

Prompts: independence/care/environment/staff

2. Can you tell me if you have any worries about moving to the adult hospital?

Appendix 13: Interview Guide 16-18-year olds.

Appendix 13: Interview Guide for 16-18-year olds.

Transition Research Interview Guide 16-18-year olds

Thank you for coming to talk with me today. My name is Róisín Bradley and I work here in [REDACTED] as a research nurse. This interview should take about 20 minutes and I will be recording it so I can review it later for my notes, is that ok with you?

< START RECORDING IF VERBAL CONSENT IS OBTAINED>

< STOP INTERVIEW IF CONSENT IS NOT RECEIVED>

If you would prefer it not to be recorded, I will take notes during our talk .I will then read back my notes to you at the end of the interview .This is to ensure I have documented the interview in accordance to your views. I will amend my notes should you request me to do so.

I want to let you know that what we talk about is only being used for our research and your name will not be used. If you like you can pick a nickname that we can use during the discussion.

Before we start, have you any questions?

1. Can you tell me what you're looking forward to when you move to the adult hospital?

Prompts: independence/care/environment/staff)

2. Can you tell me if you have any concerns about moving to the adult hospital?

Appendix 14: Information Sheet 12-15 year olds.

PATIENTS 12-15 YEARS INFORMATION SHEET

Introduction

You are being asked to be involved in a research study because we would like to find out your views on transitioning to adult services.

Before you decide whether to take part it is important for you to understand what the study is and what you will need to do if you take part. Please read this sheet carefully and ask us if there is anything that you don't understand or if there is anything that you would like to know.

What is the purpose of this study?

We are interested to hear your views on moving from this hospital to the adult centre in the future. This research is part of a Masters by Research Degree course that Róisín Bradley, the research nurse, is currently completing. The research study would involve you taking part in an interview with the research nurse. There is only one interview required which should take approximately 20 minutes. The interview will be recorded on a Dictaphone or written down so that all your answers can be documented for further analysis. Your name or any identifying information will not be included. All data will be securely stored and saved directly onto the G: Drive in OLCHC where it will be analysed, coded and encrypted. It will also be stored on the V: drive of the Royal College of Surgeons in Ireland (RCSI) server. The study data will be securely stored and encrypted within the applicants (Research Nurse) unique project folder located within the RCSI V: drive. All study data (such as consent forms, participant leaflets, approval letters) will be scanned, encrypted, and saved onto the RCSI V: drive. Data

will be stored in this location for 5 years. Access will be limited to the researcher and her supervisors.

Are there any benefits to taking part?

This study should help us understand more about how you and others feel about moving on to another hospital.

Is there any disadvantage to taking part?

We hope to interview you on the same day as your routine clinic appointment. This would mean you would not have to schedule another visit to the hospital. Therefore there should be no disadvantages to taking part.

Do I have to take part?

It is your choice whether you would like to take part in the study or not. If you do not want to, you do not have to give a reason and you can change your mind at any time. If you decide that you don't want to be in the study now or later, you will continue to receive the same care.

Will anyone know I took part in the study?

All information that is collected about you will be kept within the hospital. Any information that leaves the hospital will have your name and address removed so that no-one will know that you took part.

Who can I contact for more information?

You can contact Róisín Bradley research nurse, [REDACTED] at:

(01) [REDACTED] and ask for bleep [REDACTED]

or

[REDACTED] – direct line

Appendix 15: Information sheet 16-18 year olds.

PATIENTS 16-18 YEARS INFORMATION SHEET

Introduction

You are being asked to be involved in a research study because we would like to find out your views on transitioning to adult services.

Before you decide whether to take part it is important for you to understand what the study is and what you will need to do if you take part. Please read this sheet carefully and ask us if there is anything that you don't understand or if there is anything that you would like to know.

What is the purpose of this study?

We are interested to hear your views on moving from this hospital to the adult centre in the future. This research is part of a Masters by Research Degree course that Róisín Bradley, the research nurse, is currently completing. The research study would involve you taking part in an interview with the research nurse. There is only one interview required which should take approximately 20 minutes. The interview will be recorded on a Dictaphone or written down so that all your answers can be documented for further analysis. Your name or any identifying information will not be included. All data will be securely stored and saved directly onto the G: Drive in OLCHC where it will be analysed, coded and encrypted. It will also be stored on the V: drive of the Royal College of Surgeons in Ireland (RCSI) server. The study data will be securely stored and encrypted within the applicants (Research Nurse) unique project folder located within the RCSI V: drive. All study data (such as consent forms, participant leaflets, approval

letters) will be scanned, encrypted, and saved onto the RCSI V: drive. Data will be stored in this location for 5 years. Access will be limited to the researcher and her supervisors.

Are there any benefits to taking part?

This study should help us understand more about how you and others feel about moving on to another hospital.

Is there any disadvantage to taking part?

We hope to interview you on the same day as your routine clinic appointment. This would mean you would not have schedule another visit to the hospital. Therefore there should be no disadvantages to taking part.

Do I have to take part?

It is your choice whether you would like to take part in the study or not. If you do not want to you do not have to give a reason and you can change your mind at any time. If you decide that you don't want to be in the study now or later, you will continue to receive the same care.

Will anyone know I took part in the study?

All information that is collected about you will be kept within the hospital. Any information that leaves the hospital will have your name and address removed so that no-one will know that you took part.

Who can I contact for more information?

You can contact Róisín Bradley research nurse, [REDACTED] at:

(01) [REDACTED] and ask for bleep [REDACTED]

or

■ – direct line

Appendix 16: Consent for parents

Appendix 16:

Consent Form: Parent

Reference Number:

Title of Study: Views on transition of adolescents with severe haemophilia to adult services.

Research Participant's Name:

1. I have had time to consider whether to take part in the study. My questions have been answered satisfactorily and I have received a copy of the Parent Information Leaflet.
2. I understand that my participation is voluntary and that I am free to withdraw at any time without my child's medical care or legal rights being affected.
3. I understand that any audio recordings will be destroyed following the completion of the study in September 2018.

Name of research participant

Name of Parent/ Guardian

Date

Signature

Researcher

Date

Signature

Appendix 17: Parent consent (telephone)

Consent Form: Parent (Telephone)

Reference Number:

Title of Study: Views on transition of adolescents with severe haemophilia to adult services.

Research Participant's Name:

1. I have had time to consider whether to take part in the study. My questions have been answered satisfactorily and I have received a copy of the Parent Information Leaflet.
2. I understand that my participation is voluntary and that I am free to withdraw at any time without my child's medical care or legal rights being affected.
3. I understand that all the study data will be securely stored and encrypted within the applicants unique project folder located within the RCSI V: drive. All study data (such as consent forms, participant leaflets, approval letters) will be scanned, encrypted, and saved onto the RCSI V: drive. Data will be stored in this location for 5 years. Access will be limited to the researcher and her supervisors.

Name of Parent/ Guardian

Date

Independent staff member to confirm telephone consent obtained from
named participant:

Independent staff member

Date

Signature

Researcher

Date

Signature

1 copy for research participant; 1 copy for researcher; 1 copy to be filed in
the hospital notes.

Appendix 18: Consent MDT.

Appendix 21:

Consent Form: Staff

Reference Number:

Title of Study: Views on transition of adolescents with severe haemophilia to adult services.

Research Participant's Name:

1. I have had time to consider whether to take part in the study. My questions have been answered satisfactorily and I have received a copy of the Staff Information Leaflet.
2. I understand that my participation is voluntary and that I am free to withdraw at any time.
3. I understand that all audio, written and digital data will be destroyed following the completion of the study in September 2018.

_____	_____	

Research Participant	Date	Signature

_____	_____	

Researcher	Date	Signature

1 copy for research participant; 1 copy for researcher; 1 copy to be filed in the hospital notes.

Appendix 19: Consent for MDT (Telephone)

Appendix 19:

Consent Form: Staff (Telephone)

Reference Number:

Title of Study: Views on transition of adolescents with severe haemophilia to adult services.

Research Participant's Name:

1. I have had time to consider whether to take part in the study. My questions have been answered satisfactorily and I have received a copy of the Staff Information Leaflet.
2. I understand that my participation is voluntary and that I am free to withdraw at any time.
3. I understand that all the study data will be securely stored and encrypted within the applicants unique project folder located within the RCSI V: drive. All study data (such as consent forms, participant leaflets, approval letters) will be scanned, encrypted, and saved onto the RCSI V: drive. Data will be stored in this location for 5 years. Access will be limited to the researcher and her supervisors.

Research Participant

Date

Signature

Independent staff member to confirm telephone consent obtained from
named participant:

Independent staff member

Date

Signature

Researcher

Date

Signature

1 copy for research participant; 1 copy for researcher; 1 copy to be filed in
the hospital notes.

Appendix 20: Interview Guide for Parents

Appendix 20:

Transition Research Interview Guide Parents

Thank you for coming to talk with me today. My name is Róisín Bradley and I work here in [REDACTED] as a research nurse. This interview should take about 20 minutes and I will be recording it so I can review it later for my notes, is that ok with you?

< START RECORDING IF VERBAL CONSENT IS OBTAINED >

< STOP INTERVIEW IF CONSENT IS NOT RECEIVED >

If you would prefer it not to be recorded, I will take notes during our talk. I will then read back my notes to you at the end of the interview. This is to ensure I have documented the interview in accordance to your views. I will amend my notes should you request me to do so.

I want to let you know that what we talk about is only being used for our research and your name will not be used. If you like you can pick a nickname that we can use during the discussion.

Before we start, have you any questions?

1. What do you think are the positive aspects of your child transitioning to the adult hospital?

Prompts: independence/care/environment/staff

2. Have you any concerns in relation to your child transitioning to the adult hospital?

Appendix 21: Interview Guide for MDT.

Appendix 21: Interview Guide for MDT.

Transition Research Interview Guide Staff

Thank you for coming to talk with me today. My name is Róisín Bradley and I work here in [REDACTED] as a research nurse. This interview should take about 20 minutes and I will be recording it so I can review it later for my notes, is that ok with you?

< START RECORDING IF VERBAL CONSENT IS OBTAINED>

< STOP INTERVIEW IF CONSENT IS NOT RECEIVED>

If you would prefer it not to be recorded, I will take notes during our talk. I will then read back my notes to you at the end of the interview. This is to ensure I have documented the interview in accordance to your views. I will amend my notes should you request me to do so.

I want to let you know that what we talk about is only being used for our research and your name will not be used. If you like you can pick a nickname that we can use during the discussion.

Before we start, have you any questions?

1. In your role, what are the positive aspects for an adolescent transitioning to the adult Hospital?

Prompts: independence/care/environment/staff

2. In your role have you any concerns in relation to the current transitioning process of an adolescent to the adult hospital?

Appendix 22: Information Sheet for MDT.

Appendix 22:

Views on Transition: STAFF INFORMATION SHEET

Introduction

You are being asked to be part of this study as your views on this topic would be very useful for the future development of a structured transition programme for adolescents with severe haemophilia.

Before you decide whether to take part, it is important that you understand the purpose of this study and what you would be required to do should you decide to take part.

What is the purpose of this study?

This research study is being carried out by Róisín Bradley Haemostasis & Thrombosis CNMII as part of a Masters by Research Degree course in The Royal College of Surgeons Ireland (RCSI). The title of the study is:

“What are the views of adolescents’ with severe haemophilia and their carers about their future transition to adult services?”

The research would involve you taking part in a once off interview with Róisín Bradley. There is only one interview required which should take approximately 20 minutes. The interview will be recorded on a Dictaphone so that all your answers can be documented for further analysis. Your name or any identifying information will not be included. All data will be kept on secure servers and on a password protected computer. Interview scripts and audio data will be kept in a locked cabinet within a locked room. Once the research study is finished all saved information will be destroyed. This will be completed by September 2018.

Are there any benefits to taking part?

The information gained from all participating staff will be used to assist with the development of a new structured transition programme for the benefit of all the stakeholders in this process. The stakeholders in this study are the adolescences, their parents and staff. The aim is to also obtain the views of recently transitioned people to ensure that all the relevant people are included.

Is there any disadvantage to taking part?

The interview would take place at a time suitable for each staff member so the aim is that it causes the least amount of disruption as possible. There are no obvious disadvantages to taking part and each staff member can withdraw their consent to take part at any stage without any reason being required.

Do I have to take part?

No, there is no obligation to take part in the study. As mentioned above, you can withdraw your consent to take part at any stage of the process.

Will anyone know I took part in the study?

Each participant will be given a code for identification of the data. No names will be used to assure confidentiality is maintained. The researcher Róisín Bradley will be the only key holder of the data. All the data will be kept in a password protected file and not accessible to anyone else.

Who can I contact for more information?

You can contact Róisín Bradley research nurse, [REDACTED] at:

(01) [REDACTED] and ask for bleep [REDACTED]

or

[REDACTED] – direct line

Appendix 23: Information Sheet for Parents

Appendix 23: Information sheets for parents.

Views on Transition to adult services

PARENT INFORMATION SHEET

Introduction

You are being asked to be involved in a research study because we would like to find out your views on the future transition of your child from child to adult services.

Before you decide whether to take part it is important for you to understand what the study is and what you will need to do if you take part. Please read this sheet carefully and ask us if there is anything that you don't understand or if there is anything that you would like to know.

What is the purpose of this study?

We are interested to hear your views about your experience of the future transitioning from the paediatric to adult centre of your child. This research is part of a Masters by Research Degree course that Róisín Bradley, a research nurse in Our Lady's Children's Hospital Crumlin, is currently completing. The research study would involve you taking part in an interview with this research nurse. There is only one interview required which should take approximately 20 minutes. The interview will be recorded on a Dictaphone so that all your answers can be documented for further analysis. Your child's name or any identifying information will not be included. All data will be kept on secure servers and on a password protected computer. Interview scripts and audio data will be kept in a locked cabinet within a locked room. Once the research study is finished the recorded information will be destroyed. This will be completed by September 2018.

Are there any benefits to taking part?

This study should help us understand more about how you and others feel about the transition process which your child will partake in in the future.

Is there any disadvantage to taking part?

We hope to interview you on the same day as your child's routine clinic appointment. This would mean you would not have to schedule another visit to the hospital. Therefore there should be no disadvantages to taking part.

Do I have to take part?

It is your choice whether you would like to take part in the study or not. If you do not want to you do not have to give a reason and you can change your mind at any time. If you decide that you don't want to be in the study now or later, your child will continue to receive the same care.

Will anyone know I took part in the study?

All information that is collected about you will be kept within the hospital. Any information that leaves the hospital will have your name and address removed so that no-one will know that you took part.

Who can I contact for more information?

You can contact Róisín Bradley research nurse, Our Lady's Children's Hospital Crumlin at:

(01) [REDACTED] and ask for bleep [REDACTED]

or

(01) [REDACTED] – direct line.

Appendix 24:

Consent Form: Those who have already transitioned to adult hospital.

Appendix 24:

Consent Form: Those who have already transitioned to adult hospital

Reference Number:

Title of Study: Views on transition of adolescents with severe haemophilia to adult services.

Research Participant's Name:

1. I have had time to consider whether to take part in the study. My questions have been answered satisfactorily and I have received a copy of the Transitioned People Information Leaflet.
2. I understand that my participation is voluntary and that I am free to withdraw at any time without my medical care or legal rights being affected.
3. I give permission for the researcher to look at medical records to get information on my history. I understand that the information will be kept confidential.
4. I understand that all audio, written and digital data will be destroyed following the completion of the study in September 2018.

Name of research participant

Date

Signature

Researcher

Date

Signature

1 copy for research participant; 1 copy for researcher; 1 copy to be filed in the hospital notes.

Appendix 25: Consent (Telephone) for those who have already transitioned to adult hospital.

Appendix 25:

**Consent Form: Those who have already transitioned to adult hospital
(Telephone)**

Reference Number:

Title of Study: Views on transition of adolescents with severe haemophilia to adult services.

Research Participant's Name:

1. I have had time to consider whether to take part in the study. My questions have been answered satisfactorily and I have received a copy of the Transitioned People Information Leaflet.
2. I understand that my participation is voluntary and that I am free to withdraw at any time without my medical care or legal rights being affected.
3. I give permission for the researcher to look at medical records to get information on my history. I understand that the information will be kept confidential.
4. I understand that all the study data will be securely stored and encrypted within the applicants unique project folder located within the RCSI V: drive. All study data (such as consent forms, participant leaflets, approval letters) will be scanned, encrypted, and saved onto the RCSI V: drive. Data will be stored in this location for 5 years. Access will be limited to the researcher and her supervisors.

Independent staff member to confirm telephone consent obtained from
named participant:

Independent staff member

Date

Signature

Researcher

Date

Signature

1 copy for research participant; 1 copy for researcher; 1 copy to be filed in
the hospital notes

Appendix 26: Information sheet for those who have already transitioned to the adult hospital.

Appendix 26. Information sheet for those who have already transitioned to the adult hospital.

Introduction

You are being asked to be involved in a research study because we would like to find out your views on your transition from child to adult services.

Before you decide whether to take part it is important for you to understand what the study is and what you will need to do if you take part. Please read this sheet carefully and ask us if there is anything that you don't understand or if there is anything that you would like to know.

What is the purpose of this study?

We are interested to hear your views about your experience of transitioning from the paediatric to adult hospital. This research is part of a Masters by Research Degree course that Róisín Bradley, a research nurse in *****, is currently completing. The research study would involve you taking part in an interview with the research nurse. There is only one interview required which should take approximately 20 minutes. The interview will be recorded on a Dictaphone or written down so that all your answers can be documented for further analysis. Your name or any identifying information will not be included. All data will be securely stored and saved directly onto the G: Drive in OLCHC where it will be analysed, coded and encrypted. It will also be stored on the V: drive of the Royal College of Surgeons in Ireland (RCSI) server. The study data will be securely stored and encrypted within the applicants (Research Nurse) unique project folder located within the RCSI V: drive. All study data (such as consent forms, participant leaflets, approval letters) will be scanned, encrypted, and saved onto the RCSI V: drive. Data will be stored in this location for 5 years. Access will be limited to the researcher and her supervisors.

Are there any benefits to taking part?

This study should help us understand more about how you and others feel about moving on to another hospital.

Is there any disadvantage to taking part?

We hope to interview you on the same day as your routine clinic appointment. This would mean you would not have to schedule another visit to the hospital. Therefore there should be no disadvantages to taking part.

Do I have to take part?

It is your choice whether you would like to take part in the study or not. If you do not want to, you do not have to give a reason and you can change your mind at any time. If you decide that you don't want to be in the study now or later, you will continue to receive the same care.

Will anyone know I took part in the study?

All information that is collected about you will be kept within the hospital. Any information that leaves the hospital will have your name and address removed so that no-one will know that you took part.

Who can I contact for more information?

You can contact Róisín Bradley research nurse, [REDACTED] at:

(01) [REDACTED] and ask for bleep [REDACTED]

or

[REDACTED] – direct

Appendix 27:

Interview Guide: Those who have already transitioned to adult services

Appendix 27: Interview guide for those who have already transitioned to adult services.

Interview Guide: Those who have already transitioned to adult services.

Thank you for coming to talk with me today. My name is Róisín Bradley and I work in **** as a research nurse. This interview should take about 20 minutes. I may be recording it so I can review it later for my notes, is that ok with you?

If this interview is recorded:

< START RECORDING IF VERBAL CONSENT IS OBTAINED>

< STOP INTERVIEW IF CONSENT IS NOT RECEIVED>

If you would prefer it not to be recorded I will take notes during our talk .I will then read back my notes to you at the end of the interview .This is to ensure I have documented the interview in accordance to your views.

I want to let you know that what we talk about is only being used for our research and your name will not be used. If you like you can pick a nickname that we can use during the discussion.

Before we start, have you any questions?

1. What do you think were the positive aspects of your transition to the adult hospital?

Prompts: independence/care/environment/staff

2. Were there any aspects of your transition that you would like to see improved?

(Prompts: independence/care/environment/staff)

Appendix 28:

Transition Research. Interview Transcript

Appendix 28:

Transition Research.

Interview Guide Staff.

Thank you for coming to talk with me today. My name is Róisín Bradley and I work here in **** as a research nurse. This interview should take about 20 minutes and I will be recording it so I can review it later for my notes, is that ok with you?

Date: 15th August 2017

Venue: MDT member's office, clinical site.

< START RECORDING IF VERBAL CONSENT IS OBTAINED>

< STOP INTERVIEW IF CONSENT IS NOT RECEIVED>

If you would prefer it not to be recorded, I will take notes during our talk .I will then read back my notes to you at the end of the interview .This is to ensure I have documented the interview in accordance to your views. I will amend my notes should you request me to do so.

I want to let you know that what we talk about is only being used for our research and your name will not be used. If you like you can pick a nickname that we can use during the discussion.

Before we start, have you any questions?

No

3. In your role, what are the positive aspects for an adolescent transitioning to the adult hospital?

No going back, left here one day here and next day in adult centre.

Children here were "mollycoddled" in the past.

Transition programme started when? What does haemophilia mean? They start to listen. How does it affect me? –These issues addressed.

Ideally starts at 11 and should be coming in on their own at the end.

Good to have a programme.

Nurse from ***** used to come here – adolescent clinic every 3-4 months, and for first clinic there a CNS from here would go there. This only happened a few times due to staff shortages- familiar faces when at the door in the adult site.

Preparing them for actual transfer itself: where you were going, who you would meet, the practical section of it.

The parents like to meet us-sometimes they might say to us what they wouldn't say to doctors.

When we “floated” (i.e. overall supervision of the patients rather than focusing on a select few) we actually saw more rather than seeing our own patients in clinics.

Prompts: independence/care/environment/staff .

4. In your role have you any concerns in relation to the current transitioning process of an adolescent to the adult hospital?

Current document very broad.

Should be age appropriate: 12-15 and 15-17.

Parents concerned about talking about sex.

Parents can be concerned as to what you'd be talking to them about.

I have an issue about all of a sudden talking to them about sex/adult issues.

Parents concerned about decrease in control, known them since they were babies and now taking to them about adult issues.

Mothers- control thing-as long as they are living under your roof they have control.

I don't feel comfortable about talking about sex- this information should be carried on to the adult centre. It should be a doctor who discusses these issues.

It has to be more age appropriate.

Army, Pilot, Gardaí are career choices which are not advised. Some might not discuss that they have haemophilia. The importance of teaching them from a young age so they can use this information when looking at future career choices and informing other people especially when out.

-Lost interest when half way through.

Frightens the parents that they are moving on. You have informed them- always advising them from day one.

Made parents aware how little the children knew.

One document for everyone.

One shoe fits all but it doesn't really.

Have to look at the maturity of the child.

The ones that turn up are the ones that know about their condition and the ones that don't are the ones that you have issues with.

(Prompts: independence/care/environment/staff