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SICKLE CELL DISEASE SCREENING: PERCEPTIONS OF PARENTS WITH POSITIVE NEWBORNS AT KORLE BU TEACHING HOSPITAL

BY

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DECLARATION

I, Irene Kanyoke hereby declare that this research is my work except for references derived from other people's work which have been duly acknowledged. I assert that this work was done by me and supervised by Dr Faustina Hayford Blankson and has not been submitted in whole or part to any institution for the award of any degree.

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DATE: 7th July 2022

DR. FAUSTINA HAYFORD BLANKSON

(SUPERVISOR)

DATE: 7th July 2022

DEDICATION

I dedicate this work to my family and friends for their immense support and encouragement throughout this period.

I also dedicate this work to all the parents and children at the Paediatric Sickle Cell Clinic of Korle Bu Teaching Hospital, Accra.

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My appreciation to God for His blessings throughout my time doing this research.

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Abstract

Background: Sickle Cell Disease (SCD) is a genetic blood disorder caused by the presence of abnormal haemoglobin (Hb S) leading to acute pain episodes, Anaemia, acute chest syndrome, priapism and organ damage. One of the main interventions for managing the disease is by screening babies at birth for early detection and comprehensive clinical management leading to prolonged life. Early enrolment and comprehensive care are some of the underlying principles of universal newborn screening. There is however a lack of understanding and knowledge of the practice from the perspectives of parents.

Aim: This study aimed to explore the experiences of parents of newborns regarding the receipt of positive sickle cell results at the Paediatric Sickle Cell Clinic of Korle Bu Teaching Hospital.

Method: An explorative approach to qualitative research was used, and data was collected by using semi-structured individual interview guides to interview mothers and a father of newborns attending the Paediatric Sickle Cell Clinic of Korle Bu Teaching Hospital. A purposive sampling method was used and data collected was analysed using NVivo software to identify relevant codes which were then organized into themes. The Socio-Ecological Model was used to underpin this study.

Data analysis was done using thematic analysis. Open coding was used to identify initial codes after which they were categorised into 5 themes and sub-themes. The 5 themes identified were: emotional response after disclosure, decision to share results, care and management of newborns at home, knowledge of genetics of SCD, and adherence to routine follow-up.

Result: Parents of positive sickle cell disease newborns at the Paediatric sickle cell clinic of Korle Bu Teaching reported varied emotional responses to receiving the results due to prior knowledge of the disease or personal experiences with the disease. Parents also reported improving their knowledge of SCD. Prior knowledge of SCD and personal experience impact how parents react to positive NBS results of their newborns. Parents of newborns usually

adhere to follow-up procedures due to a desire to see their children healthy. However, some default on follow-up due to difficulties such as COVID 9 pandemic and financial difficulties.

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LIST OF ABBREVIATIONS

SCD	Sickle Cell Disease
SCA	Sickle Cell Anaemia
NBS	Newborn Screening
P-SCD	Possible sickle Cell Disease
KBTH	Korle Bu Teaching Hospital
PSCC	Paediatric Sickle Cell Clinic
KATH	Komfo Anokye Teaching Hospital
Hb	Haemoglobin
HU	Hydroxyurea
GVHD	Graft Versus Host Disease
HPLC	High-Performance Liquid Chromatography
IEF	Iso-Electro Focusing
NCDs	Non-Communicable Diseases
ASH	American Society of Haematology
GoG	Government of Ghana
NICU	Neonatal Intensive Care Unit
PICU	Paediatric Intensive Care Unit

DEFINITION OF TERMS

Universal Newborn Screening: testing all newborns for disorders that usually otherwise are not found at birth.

Confirmatory test: final, verified testing after initial diagnostic screening

Possible Sickle Cell Disease: someone diagnosed with sickle cell disease prior to confirmatory tests being done

Comprehensive follow-up care: regular medical checkups by a multi-disciplinary team for prevention and management of the health problems of a patient.

CHAPTER ONE

INTRODUCTION

1.1 Background of the Study

Sickle Cell Disease (SCD) is a hereditary disorder characterized by the presence of abnormal haemoglobin the most common of which is the haemoglobin S (Hb S) (Asare et al., 2018).

SCD clinically presents as acute haemolytic and vaso-occlusive crises which can lead to complications including stroke, organ damage, priapism and acute chest syndrome and it is estimated that between 300,000 to 500, 000 babies are born every year with SCD worldwide (Russo et al., 2019). Studies show that 7 in 10 SCD-related deaths are preventable and the high prevalence of morbidity and mortality in Africa is made worse by inadequate systemic screening for the disease, follow-up care, shortage of facilities for SCD management, poor research and intervention protocols (Hsu et al., 2018).

Sickle Cell Disease was declared a major public health issue that needed to be addressed by the African Union in 2005, the World Health Organisation in 2007 and the United Nations General Assembly in 2009 (Hsu et al., 2018).

Asare et al (2018) estimate that about 2% (15,000) of all babies in Ghana are born with sickle cell disease annually. Studies have shown that children below 5 years of age are at a higher risk of SCD-related complications (Runkel et al., 2020). According to Makani et al (2017), children between 6 months and 3 years have the highest levels of SCD- related deaths with acute chest syndrome and acute splenic sequestration being the most common causes.

SCD has a huge socio-economic impact not only on parents but also on the community and even the country at large (Hsu et al., 2018) and as such, over the years, there have been several disease management interventions for the disease. One of these interventions is Newborn Screening (NBS).

Newborn screening for SCD is the process by which blood from a baby is collected in the first few days after birth and tested for SCD (Hsu et al., 2018). The main objective of newborn screening is to allow for the early identification of infants with the disease and the provision of follow-up care to help reduce morbidity and mortality (Miller et al., 2010). Newborn screening for SCD is a laudable idea (Segbefia et al., 2019) and comprehensive follow-up care which involves a multi-disciplinary approach must include the families and caregivers of newborns with SCD.

Due to the aforementioned deficiencies, this study aims at exploring the experiences of parents of newborns who test positive for sickle cell disease at Korle Bu Teaching Hospital so that appropriate programmes can be developed and implemented.

1.2 Problem Statement

Newborn screening for SCD followed by prophylactic penicillin and effective vaccinations have been attributed to a remarkable reduction in morbidity and mortality in children under 5 years (Quinn et al., 2010; Streetly et al., 2018).

In Korle Bu Teaching Hospital (KBTH), NBS for SCD has been ongoing since June 2017, and all newborns at the hospital are screened for SCD and those identified as being positive for possible Sickle Cell Disease (P-SCD) are tracked and enrolled in the KBTH Paediatric Sickle Cell Clinic (PSCC) for follow-up specialist care (Segbefia et al., 2019). An infant is identified as having possible sickle cell disease (P-SCD) when confirmatory tests are yet to be conducted after the initial NBS diagnosis.

At the KBTH PSCC, parents of positive newborns diagnosed via NBS, are called and scheduled for a clinic appointment where initial positive results are disclosed and general and genetic counselling on SCD is done for parents after which confirmatory tests are done and babies started on prophylactic penicillin. Those who do not show up for the initial clinic appointment

are then informed of the positive results of their newborns via telephone calls and given new appointment dates for clinic (Segbefia et al., 2021).

According to unpublished records from the Newborn Screening programme in KBTH, a total of 211 newborns with P-SCD were identified through screening out of a total of 11,617 newborns screened at KBTH from June 2017 to December 2019 representing 1.8% of the total screened (Segbefia et al., 2021). A total of 195 of these P-SCD babies were tracked and parents given the results while the remaining 16 could not be successfully tracked due to inaccurate or insufficient information. However, raw data from the clinic show that only 123 of these parents had accepted the initial diagnosis and enrolled their newborns in the KBTH PSCC as of December 2019 with the average age of enrolment being 6 months, usually, after babies start showing symptoms (Segbefia et al., 2021). There is also quite a high attrition rate in terms of parents attending subsequent clinic visits after the initial visit (Segbefia et al., 2021).

This poses a huge challenge in the timely enrolment and commencement of penicillin prophylaxis, as well as the provision of comprehensive follow-up care which are underlying principles of NBS for SCD.

Records from the KBTH PSCC also indicate that some of those enrolled did not follow up with subsequent review appointments till their babies developed severe symptoms (Segbefia et al., 2021). To date, some of them have not shown up despite home visits by public health nurses.

It can therefore be summarized that there is a problem of reluctance of parents to accept initial SCD diagnosis and to comply with follow-up clinic procedures. This is a huge concern for the NBS programme in Ghana and needs to be researched especially given that there are plans for more regional NBS programmes to commence and even for the national scale-up of the programme in the country.

Nonetheless, previous studies conducted on the various newborn screening programmes concentrate on documenting the clinical and implementation information. As such, there is a paucity of information about the experiences of parents of newborns who have tested positive for SCD within the Ghanaian context. There is also a paucity of information on the attitudes of parents towards follow-up procedures for SCD as well as the responses of parents to counselling and education about SCD.

Even though a few studies have been done exploring the experiences of parents of children with SCD in Ghana, there is no exploration into the reaction of parents to initial positive results, especially newborns identified through the NBS programme as well as how parents respond to in-clinic counselling. There is little information on how this could influence parents' attitudes towards acceptance of the initial positive diagnosis and subsequent clinical attendance.

Therefore, there is the need to explore the experiences of parents with sickle cell positive babies identified through the NBS process at the PSCC of KBTH to be able to better understand the current care practices in terms of, in-clinic genetic counselling, disclosure of positive SCD results, as well as the general attitudes of parents toward follow up care. This exploration would provide a better understating of the attitudes of parents towards clinic enrolment and follow-up visitations to the clinic and serve as the basis for further research into the implementation of interventions to meet the goal of early identification, early enrolment and comprehensive follow-up care of SCD positive newborns.

1.3 Research Questions

The research questions that guided the study were:

1**. Research Question 1 (RQ1):** What are the reactions of parents towards the communication of positive results?

2**. Research Question 2 (RQ2)**: How does in-clinic counselling and support form part of comprehensive follow-up care?

3**. Research Question 3 (RQ3):** How do parents of SCD newborns adhere to follow-up clinic procedures?

1.4 General Objective

The main objective of this study was to explore the experiences of parents of positive sickle cell disease newborns at the Paediatric Sickle Cell Clinic of Korle Bu Teaching Hospital.

1.5 Specific Objectives

1. To explore the reactions of parents of PSCD newborns towards disclosure of initial positive results at the Paediatric Sickle Cell Clinic of KBTH.
2. To explore how in-clinic counselling and support form part of comprehensive follow-up care at the Paediatric Sickle Cell Clinic of KBTH.
3. To inquire into how parents of SCD-positive newborns adhere to follow-up clinic care at the Paediatric Sickle Cell Clinic of KBTH.

1.6 Theoretical Framework

This study used an adaptation of Urie Bronfenbrenner‘s Social-Ecological Model (SEM) developed in 1979. The SEM has been adapted by public health experts over the years for research into understanding issues affecting well-being. Thus, this framework was adapted for this study to better explore the multifaceted factors that affect parents' experiences.

1.6.1 Social-Ecological Model of Health

This framework was first developed as a conceptual model in the 1970s by Bronfenbrenner to study human development and was later modified into a theoretical framework. This theory looks at how various factors such as individual factors, interpersonal connections/networks, community influence, institutions and policy influence health (Kilanowski, 2017).

The SEM framework has been used by various health experts to gain an understanding of how the various factors interplay to affect health and thus is useful in health prevention, control and promotion. The SEM has 5 levels of influence on health behaviour:

- Intrapersonal or individual factors include individual characteristics such as age, education, sex, attitudes, knowledge, feelings, personality, health history, etc.
- Interpersonal factors include networks, interactions or relationships with other people in society.
- Institutional or Organizational level includes policies, rules, and regulations within an organisation or institution such as churches, schools, hospitals, etc. that may influence health.
- Community includes societal norms, cultural or religious values, etc. that exist among groups, individuals or organisations.
- Public Policy factors include local or national policies and laws that govern or support health care delivery.

This study, however, used an adapted version that examines the interplay and interconnectedness of individual (intrapersonal) and interpersonal factors that affect the experiences of parents of positive SCD newborns at the Paediatric Sickle Cell Clinic of Korle Bu Teaching Hospital. This model was used to study how the factors at each level interact and may influence parents' experiences. This study, therefore, modified the SEM to examine the interplay of factors only at the individual (intrapersonal) and interpersonal levels only. The

SEM was suitable as a theoretical framework for this study because it provided a robust platform for understanding how individual and interpersonal factors interact.

1.6.1.1 Intrapersonal Influence

The interpersonal stage of the model looks at the individual's characteristics such as knowledge, skills, emotions, age, income, etc. This stage of the model posits the individual at the centre of the theory and examines how certain characteristics of individual influences certain health behaviours (Kilanowski, 2017). The researcher, therefore, examined how individual influences played a role in parents' reactions to positive results, how they played a role in their responses to counselling at clinic and how these factors played a role in the attitudes of parents to follow-up clinic procedures. This study adapted this model to better explore the role of individual influences in the experiences of parents of newborns who tested positive for SCD at the PSCC of Korle Bu Teaching Hospital.

1.6.1.2 Interpersonal Influences

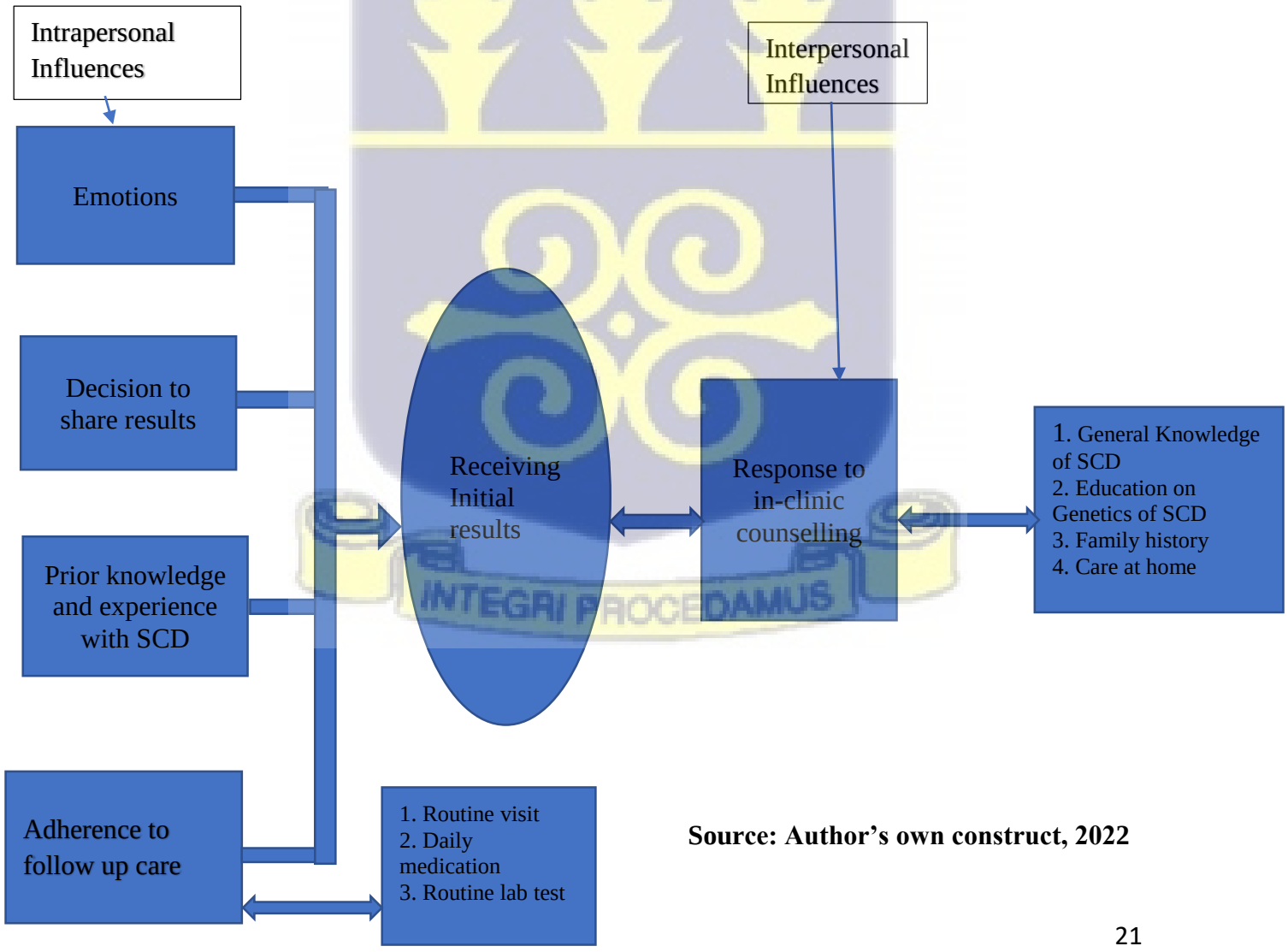
This stage examines the relationship, interactions and interconnectedness that an individual has with other people such as family, friends, etc. influence health behaviour. It looks at the network of interactions between an individual and other people and how this interaction influences health. This model helped examine the relationship and interactions between clinic staff and parents. The interpersonal stage helped explore how the interactions in clinic helped explain how to in-clinic counselling and education about sickle cell disease form part of comprehensive follow-up care. It helped explain the relationship between parents and clinic staff and how this led to parents being educated on knowledge of SCD, genetics of SCD, management of the disease, as well as general management of the disease by parents.

1.7 Conceptual Framework

This study used an adaption of the Socio-Ecological Model to explain the various concepts of the study: parents' reactions to receiving the initial positive results, counselling and care parents have received from clinic staff and parents' attitude to follow-up care.

The framework illustrated how individual factors such as emotions, prior knowledge of SCD, influences parents' reaction to receiving initial positive results, their decision to share results and their attitudes toward follow-up care. The framework also looked at how parents' networking or relationship with clinic staff helped them gain knowledge about SCD genetics and care of SCD newborns at home. It also helped the researcher explore how the interaction between these factors could influence early enrolment and encourage adherence to follow-up care procedures at clinic.

Figure 1.1 Illustration of Social-Ecological Model Adapted from Bronfenbrenner (1979)



Source: Author's own construct, 2022

1.8 Rationale for using this theoretical framework

The rationale for using the Socio-Ecological Model of Health was to enable the researcher to develop a robust platform to explore the experiences of parents on the individual and also how a connection of networks may influence their experiences. The SEM was therefore ideal for this study as a theoretical framework because it provided the bedrock on which the individual and interpersonal factors such as knowledge of SCD, care provision by clinic staff, education and counselling by clinic staff could all influence parents' reactions to disclosure of positive results, their response to counselling and education by clinic staff, the role of parents in comprehensive follow-up care and their attitudes towards follow up clinic procedures.

1.9 Justification of Study

This study will provide information about the factors impacting parents' acceptance of the initial SCD diagnosis of their newborns and how in-clinic counselling could influence a change in attitude. This can be very useful for the NBS programme in Ghana when it is finally implemented and scaled up to the national level because measures can be put in place to curb the non-acceptance of positive results as well as the dropout rate of parents at clinic.

This study will lay the foundation for further targeted research into better processes and guidelines for communicating positive results of newborns to parents that will increase the acceptance of these results. It will also lay the foundation for further research into the specific counselling and educational needs of parents of newborns at clinic to solve the problem of attrition by parents at clinic as well as non-acceptance of positive SCD results.

In general, findings from this study could be used to develop guidelines and SOPs for clinical health care providers with a focus on improving the current methods.

Knowledge gained from this study can also be used to develop SOPs and manuals for caregivers and parents of newborns with sickle cell disease at the PSCC.

CHAPTER TWO

LITERATURE REVIEW

2.0 Introduction

The literature review was conducted through a web search of relevant phrases, topics, and words from Google and various research journals such as Research Gate, and PubMed, among others via the University of Ghana Library system. The Boolean operators "AND" and "OR" were used to widen or restrict the search. The literature review will cover an overview of sickle cell disease, disease burden and management of SCD, newborn screening policy and implementation in Ghana, knowledge and perception of sickle cell disease, responses to counselling and attitudes to follow-up care.

2.1 Overview of Sickle Cell Disease

Sickle cell disease (SCD) is a recessive genetic disorder characterized by the formation of abnormal haemoglobin (Hb) called Haemoglobin S (HbS). The abnormal Hb is formed due to nucleotide substitution of valine for glutamine acid within the ß-globin gene (Makani et al., 2017). SCD occurs due to the inheritance of two identical alleles of the HbS (HbSS) which is sometimes also called Sickle Cell Anemia (SCA) and sometimes due to the inheritance of two or more heterogeneous recessive alleles of the ß-thalassemia mutations(HbS/ß°, HbS/ ß+) (Russo et al., 2019). The other clinically significant variant of SCD is the HbSC (Makani et al., 2017). Other variants such as D Punjab, Hb E, and Hb B(Bart's) are less commonly known in Sub-Saharan Africa (Modell & Darlison, 2008).

De-oxygenation and polymerization of HbS lead to the production of irregular red blood cells known as sickled cells (Makani et al., 2017) and as these sickled cells are dense and brittle,

they easily get trapped in certain organs and this usually leads to vaso-occlusive crises (Russo et al., 2019).

SCD in infants generally presents later in infancy and newborns usually appear healthy and asymptomatic. Once they become symptomatic, children with SCD present with dactylitis, anaemia, acute chest syndrome, jaundice, splenic sequestration, and pneumococcal septicemia (Avard et al., 2016).

2.2 Disease Burden and Distribution of SCD

SCD was originally thought to be discovered in 1910 by an American Doctor (Chakravorty & Williams, 2015), yet the disease has been known in certain parts of Africa well before the 20th century albeit by different local names (Makani et al., 2017). Even though SCD has been known to originate from malaria-prone areas, global migration over the years means that a lot of children with SCD are being born all over the world (Chakravorty & Williams, 2015).

It is estimated that over 400, 000 babies are born with SCD globally, (Hsu et al., 2018) and nearly 67% of all these SCD cases are found in Africa (Pule & Wonkam, 2014). SCD is a life-long disease of the blood and people with SCD usually live with incidences of painful crises, stroke, and other complications. According to Ohene-Frempong *et al.*, (2005) and Asare *et al.*, (2018) close to 15,000 (2%) of all neonates are born with SCD in Ghana every year. As such, the disease has a huge socio-economic impact not only on patients but on their families and the country as a whole (Hsu et al., 2018).

According to a study by Asare *et al.*, (2018), there were a total of 20,788 clinic visits made by 5451 adolescent and adult SCD patients to the Ghana Institute of Clinical Genetics (the adult sickle cell clinic) in Korle Bu Teaching hospital between 2013 and 2014. Over 900 of these patients were referred to other departments such as Obstetrics, Orthopaedics, Ophthalmology, general surgery, urology, and nephrology for specialist care due to complications of SCD. The

life expectancy for people with SCD in developed countries is 44 – 55 years (Wastnedge et al., 2018) due to long Oheneterm disease intervention and management programmes such as NBS, Hydroxyurea (HU) treatment and blood transfusion (Makani et al., 2017). There is, therefore, hope that Africa can achieve such progress as well.

2.3 Management of SCD

Specialists normally focus on clinical management of SCD, and four main approaches are used: supportive, symptomatic, preventive and abortive. Supportive management includes quality nutrition and regular intake of vitamin supplements (Niekerk, 2015). Symptomatic management is usually aimed at alleviating symptoms and it includes blood transfusions and intake of iron supplements to increase the amount of normal Hb in the blood (Niekerk, 2015). Preventive management of the disease aims to prevent disease complications such as stroke by using Hydroxyurea (HU) and pneumococcal vaccines. HU is a medication initially intended for cancer treatment but after several clinical trials proved to have great efficacy in preventing vaso-occlusive crises and end-organ dysfunction (Agrawal et al., 2014) it has been used for SCD management. The abortive option includes using Nitric Oxide (NO) to stop prolonged pain episodes (Pule & Wonkam, 2014).

Stem cell transplantation from bone marrow has been known to be the only treatment option for SCD. There are however known complications such as infertility and Graft versus host disease (GVHD) associated with it (Pule & Wonkam, 2014). This procedure is also very expensive and more than 90% of all global stem cell transplantation procedures were done in high-income countries (Pule & Wonkam, 2014). It is important to note there are only 6 countries in Africa where Hematopoietic stem cell transplantation is available namely Morocco, Algeria, Tunisia, Egypt, Nigeria and South Africa (Harif et al., 2020).

Clinical management of SCD mainly aims to prevent or minimize disease complications to reduce morbidity, increase the quality of life and improve the survival rates of people with SCD (Pule & Wonkam, 2014).

One other way that haematologists manage SCD is by screening for SCD. Screening involves testing to identify individuals who are at risk of a particular disease to take preventative or symptomatic management or action (Niekerk, 2015). According to (Modell & Darlison, 2008), screening for SCD is a disease management and prevention strategy that usually is undertaken in 3 ways. The first is pre-marital screening where potential parents are given genetic counselling and are advised to check their phenotype before marriage. For couples with an affected child, the probability of having another child with the disease is 25% and parents are usually made aware of this. The second is prenatal screening and diagnosis where the fetus is screened for SCD before birth so that parents can have the knowledge and opportunity to terminate the pregnancy or take precautions to reduce complications. The third is newborn screening where neonates are screened for the disease in the first few days of life and once identified, clinically managed to reduce complications and death (Modell & Darlison, 2008). These three screening processes are usually done in tandem to optimize the impact of SCD management.

Longitudinal studies conducted over the years, especially in Jamaica and the USA have provided proof that interventions such as screening, clinical management of SCD with blood transfusion therapies and HU treatment have had meaningful and valuable impacts on the quality of life of patients and have also improved the chances of survival for patients (Makani et al., 2017).

2.4 Newborn Screening for SCD

Newborn Screening for SCD is an important public health initiative practised in most developed countries to help identify neonates who are born with the disease (Ulph et al., 2017). The main aim of screening newborns for SCD is for early detection and management of the disease (Runkel et al., 2020). Early management of the disease usually includes the use of penicillin prophylaxis and pneumococcal vaccinations (Ohene-Frempong et al., 2005) to avoid disease complications and symptoms such as vaso-occlusive crises (Runkel et al., 2020). Newborn Screening programmes in most high-income countries usually screen for a variety of disorders such as SCD, cystic fibrosis, congenital hypothyroidism, phenylketonuria, and maple syrup urine disease, among others (Ulph et al., 2017).

The NBS process varies from country to country and some places use the opt-in option where parents thought to be at high risk of having children with SCD are offered NBS services where they can choose to have their babies screened (Avard et al., 2016). This differs from the practice in the USA and Canada, where there is universal screening with parents having the option to opt-out (Chudleigh et al., 2016). The first option is usually practised where SCD is not very common.

In Ghana and other African countries where SCD is prevalent, universal screening for SCD is recommended. NBS for SCD usually starts with genetic education about SCD given to parents before screening (Ulph et al., 2017). Blood from newborns is then taken from the cord or the heel of the neonate after birth and tested for SCD (Chudleigh et al., 2016). Blood from the heel is usually recommended over cord blood and this is because, better samples and results are obtained with heel stick blood (DeTolve, 2016).

In high-income countries, the High-Performance Liquid Chromatography (HPLC) method is used to detect SCD in the blood of neonates. However, in lower-income settings such as Ghana, the HPLC method can be expensive and as such the Iso-Electro Focusing (IEF) and

Electrophoresis methods are usually recommended for use to detect SCD in Neonates. These methods, however, are not 100% full proof due to the high levels of Hb F in neonates (Makani et al., 2017) and as such, these tests are repeated for newborns at 6 months of age when the Hb F are at lower levels (Hsu et al., 2018). These repeat tests are what are known as confirmatory tests (Makani et al., 2013).

NBS for SCD has been known to have made remarkable improvements in the life expectancy of children born with SCD. According to a study in Dallas by Quinn *et al.*, (2010) on the survival of children with SCD, NBS was known to have increased the survival rate of these children to about 98%. Another significant cohort study detailing the remarkable improvement in survival of children under 5 with SCD is the Jamaican Experience study by King *et al.*, (2007), where children with SCD were screened for the disease at birth and followed for years.

2.5 Newborn Screening for SCD – Policy and Implementation

NBS for SCD programmes are very effective in developed countries where there are nationwide screening programmes. These programmes are known to reduce under 5 mortality in children with SCD by over 90% (Streetly et al., 2018). In Africa, the only nation running a nationwide NBS programme that includes SCD is Egypt. Morocco has an NBS programme but does not include SCD due to the low number of children born with it in the country. Eleven other African countries including Ghana, Benin, Cameroon, Nigeria, Angola, DRC, Malawi, and others have conducted some form of pilot or regional NBS programmes (Therrell et al., 2020). However, only Ghana and Benin have comprehensive NBS for SCD programmes i.e., where all babies are screened for SCD and even with that they are being run at regional levels with delays in scaling up to the national levels (Therrell et al., 2020). Nigeria is known to have the greatest burden of SCD in the world with over 40 million people being carriers of the sickle cell trait and over 150,000 babies born with the disease every year (Muoghalu, 2016). Yet,

Nigeria does not have a national NBS for SCD programme and as of 2017, had only a few pilots and region-based programmes being run in certain parts of the country known to have a high prevalence of SCD (Hsu et al., 2018).

NBS for SCD was started in Ghana in 1995 at the Komfo Anokye Teaching Hospital (KATH) as a disease management intervention. All newborns at KATH and Tikrom, a small community near Kumasi were screened for SCD and babies who tested positive for Possible Sickle Cell Disease (P-SCD) were picked up, enrolled in the KATH Sickle Cell Clinic and provided with comprehensive follow-up specialist care (Ohene-Frempong et al., 2005). By 2006, the programme had expanded to other community hospitals and maternity homes in Kumasi and had screened over 220000 newborns, making it the largest cohort in Africa. Two per cent (2%) of the total newborns screened were diagnosed with SCD (Ohene-Frempong et al., 2008). This study set the platform for other pilot and regional NBS for SCD programmes in Africa. Unfortunately, as of 2016, NBS was still only being undertaken in Kumasi and a few select health facilities in the Ashanti Region (Segbefia et al., 2021).

In 2012, the Ministry of Health after several stakeholder meetings and consultations came up with a policy for non-communicable diseases in Ghana.

Even though NBS for SCD was adopted as part of the National Policy on Non-Communicable Diseases (NCDs) in Ghana in 2012 (Ministry of Health, 2012), it has never been fully implemented and scaled up to the national level. As such, the few NBS programmes currently ongoing in the country are funded through various partnerships with donor organisations.

Presently, the only NBS programmes in the country are the Ashanti Region NBS programme being run in a few select health facilities including KATH, the Pfizer-SickKids - funded KBTH NBS programme which includes NBS screening in KBTH and Ussher polyclinic as well as the American Society of Hematology (ASH) - funded NBS pilots in Ridge and 37 military

hospitals. All these programmes collaborate with the Sickle Cell Foundation of Ghana, the national body mandated to coordinate all NBS for SCD programmes in the country. Even though the Ghana Health Service, the Ministry of Health and the GoG have made some effort to claim these NBS programmes that are currently ongoing, full ownership of these programmes hasn't been fully asserted by these institutions and as such, they have not been able to scale them up to national level. There is therefore an issue with sustainability especially when these foreign donations and partnerships end. For instance, the Ashanti Region NBS programme has faced serious issues with sustainability since funding from foreign donors ended. There have been calls for the NHIS to cover the cost of testing for NBS and the cost of HU in Ghana but as it stands, this hasn't happened yet.

According to Public Health experts, there needs to be comprehensive policy and implementation for SCD prevention and management including genetic counselling, neonatal screening, clinical management and care as well as help for families and people with SCD (Makani et al., 2013).

2.6 Knowledge and perceptions about SCD and NBS

SCD is not a new phenomenon in Africa and as such, there is a bit of knowledge about the disease among the general populace. One key management strategy for SCD is parental education and counselling. Parents' knowledge of SCD and NBS is key for a successful intervention. Assessing parents' knowledge serves as a baseline to determine their attitudes towards acceptance of positive SCD results as well as their attitudes towards further clinical management of the disease. A study by Chudleigh *et al.*, (2016) in the UK to explore the experiences of parents receiving positive SCD and CF results for their newborns noted that all 7 families who participated in the study for SCD had prior knowledge about the disease. Five had prior knowledge due to family history whiles 2 knew, due to experience with close friends

with the disease. According to the study, most parents were able to give adequate information about SCD, the genetic connection as well as ways of managing the disease. These parents also made known that they had received adequate information about NBS and their chances of having children with the disease and as such the parents were subsequently able to accept the results of the screening despite initial anxiety. This can be attributed to the adequate knowledge about the possible outcomes and implications that they had prior to screening. This study shows a positive connection between pre-NBS counselling and the acceptance of initial results by parents. However, much is not known about this connection in the Ghanaian setting.

There have also been several studies conducted to understand the perceptions of people about SCD. These studies sought to understand people's knowledge of the disease in order to know the impact of certain interventions in certain communities as well as how people's perception of the disease could influence their acceptance of a positive diagnosis for a family member as well as their attitudes towards preventive interventions.

A study was conducted by Dennis-Antwi *et al.*, (2019) in Kumasi to ascertain the religious beliefs and attitudes of people towards SCD and how these attitudes would affect public health interventions. The study found that religion in Ghana was part of the Ghanaian socio-cultural landscape and as such religion had a positive impact on coping with SCD as well as embarking on behavioural change. There was however a negative link between religion and certain SCD management practices such as genetic counselling and prenatal testing. The study noted that reliance on religion could lead to unacceptance of certain SCD interventions as well as people making ill-informed decisions about SCD interventions.

2.7 Responses of parents towards receiving initial positive SCD results

Initial responses of parents to positive results are key in understanding their acceptance of results. Research shows that inadequate education and preparation can increase levels of anxiety for parents receiving positive carrier results for SCD (Ulph et al., 2015). According to a study by Ulph *et al.*, (2015) on the effects of disclosing positive newborn carrier results of SCD to parents, it was noted that parents experienced anxiety and distress initially but after more education and counselling, they appeared to understand the results and were more accepting of it.

Another study by Kai *et al.*, (2009) exploring the communication of newborn carrier results for SCD, noted that untoward anxiety and distress of parents to initial disclosure of results was mostly influenced by how the results were communicated and their prior knowledge of the disease rather than the result itself. The study notes that parents' anxiety subsided with more understanding of the results. This study shows that prior knowledge of SCD and how positive results are communicated have an impact on the acceptance of results. This study, however, did not indicate whether these factors also affected parents' attitudes towards subsequent clinic follow-up care.

2.8 Attitudes of parents After Receipt of Results

Research shows that parents face some distress and anxiety upon the receipt of initial positive NBS results. However not much is known about the attitudes of parents towards follow-up clinical care and how the receipt of positive results influences this attitude. Much is also not known about the impact of in-clinic counselling on parents' attitudes towards follow-up clinic care. However, the study by Chudleigh *et al.*, (2016) indicated that most parents did not share the positive results with friends and relatives due to the fear of stigma. The study also made it

known that the disclosure of the results resulted in some arguments and friction between the parents of the newborns.

Stigma is known to be one of the biggest barriers to sickle cell disease. According to a systemic review of previous studies on the stigma of SCD by Bulgin *et al.*, (2019) stigma plays a key role in altering the care-seeking behaviours of patients with SCD due to experiences of discrimination.

Parents of newborns who receive positive results of their newborns tend to share the results with family and friends (Ulph et al., 2015).

2.9 Response to In-Clinic Counselling

Counselling in the form of emotional and psychosocial support is known to be one of the needs of patients with sickle cell disease (Yahaya et al., 2013). Patients have been known to cope with embarrassment, and stigma as well as the clinical manifestations of the symptoms of the disease (Yahaya et al., 2013). It is also known that sickle cell disease patients suffer when they don't have an understanding of the daily medication needed (Yahaya et al., 2013).

Genetic counselling and education on the prevention of complications and infections are very essential measures which are sometimes not readily available to patients with sickle cell disease (Adebowale, 2014). Education on the genetics of sickle cell disease, complications and symptoms, newborn testing, pre-marital counselling, and knowledge of the disease is needed as well as awareness creation by the masses to help guide the management of the disease (Adebowale, 2014).

Counselling is known to be an effective tool for self-understanding and adjustment for people with chronic conditions and sickle cell is no exception (Adegboyega, 2020). Guidance and

counselling professionals are essential to provide personalized assistance to individuals (Adegboyega, 2020).

A study by Adegboyega (2020) to assess the counselling needs of adolescents found that sickle cell disease was a life-altering disease and as such, there was a need for collaboration between the family of patients and counsellors to develop a comprehensive approach to address psychosocial effects.

There is also the need for parental education for SCD for neonates who test positive for SCD to have resources for clinical support (Johnso et al., 2015). It is recommended that once a diagnosis is made, parents of newborns should be taken through education and counselled. They should be given practical information on care for their child, expected illnesses, medications and genetics of the disease (Makani et al., 2013).

2.10 Attitudes towards Follow up Care

Comprehensive management of Sickle cell disease in young children involves periodic, continuous, monitoring and follow-up for physical, laboratory and medical evaluations (Johnso et al., 2015). Management of neonates with SCD usually involves the services of a team including specialist doctors, nurses, ophthalmologists, pharmacists, lab technicians, radiologists and social workers and caregivers (Johnso et al., 2015).

It is therefore necessary that newborn screening be followed by the enrolment of positive newborns into clinic for monitoring by this multidisciplinary team (Makani et al., 2013).

Follow-up care includes the provision of adequate and appropriate counselling and advice as well as routine and periodic clinic visitation, daily folic acid supplements and other medications, regular physical examinations and routine lab testing (Makani et al., 2013). Long-term care provided by a multidisciplinary team is an objective of newborn screening for sickle cell disease (Makani et al., 2013).

2.11 Gaps in knowledge

A review of the literature shows the dearth of information about SCD and NBS. However, it also reveals the gaps in available information on the following: a link between knowledge of SCD and acceptance of initial results as well as effects on attitudes towards subsequent clinic care of parents of positive SCD newborns.

There is also a paucity of information on the current clinical practices and protocols for disclosure of results to parents of newborns in Ghana. Most literature about SCD in Ghana usually focuses on adults and the clinical, psychosocial and socio-economic impact of the disease on patients and their families. Little is known about NBS in Ghana and the impact of positive results on families. Most of what is known is from the 2 studies ongoing in Ashanti Region and Korle Bu Teaching Hospital. Information from these studies focuses on the screening data as well as the clinic enrolment and other data on clinical manifestations of the disease. There is a paucity of information on NBS from the perspective of parents of newborns in the Ghanaian context and this study will provide information on that to identify gaps in service and recommend ways for intervention.

CHAPTER THREE

METHOD

3.0 Introduction

This chapter presents a detailed description of the methods that the researcher used in conducting the research. It explains the research setting, design, target population, sampling method and size, data collection, handling, processing and analysis. It also includes information on validity and how threats to validity were managed as well as how ethical considerations and requirements were met during this research process.

3.1 Study Site

The study was conducted at the Paediatric Sickle Cell Clinic of the Korle Bu Teaching Hospital (KBTH). KBTH is a tertiary health facility in Ghana, established by Sir Gordon Guggisberg on 9th October 1923 as a general hospital. KBTH offers specialist and referral care, and it has 3 national centres of care as well as 14 sub-BMCs and Departments one of which is the Child Health Department.

The Child Health Department which is opposite the Accident and Emergency Centre has a 197-bed capacity with the following units: The Outpatient department including the Sub-specialty clinic venue where various clinics are held on different days, the Emergency Room, the NICU, the PICU, the Babies Unit, PS1, P2A, P2C, and P3B.

The Hematology clinic of which the Paediatric Sickle cell clinic is a part is held every Tuesday and Thursday at the sub-speciality clinic venue. Sickle Cell Disease and Thalassemia patients are seen on Thursdays and newborns diagnosed with SCD through the NBS programme as well as patients with other haematological conditions such as haemophilia, etc. are seen on Tuesdays. The Clinic attends to an average of 80 patients per week.

3.2 Study Design

This study used a qualitative research design with an explorative approach. Data was collected with the use of a semi-structured interview questionnaire. This study was explorative because there is a paucity of information in Ghana about the responses of parents on disclosure of positive NBS results as well as how or if this influences follow-up clinic attendance. Individual in-depth interviews were used because of the sensitive nature of the topic, which would require privacy.

This study was conducted within a 1-year frame and field data collection was done within 3 months.

3.3 Study Participants

The study population were mothers or fathers of newborns enrolled in the Pediatric Sickle Cell Clinic of KBTH.

3.4 Inclusion Criteria

All mothers or fathers of newborns diagnosed via the KBTH NBS programme and enrolled in the clinic.

3.5 Exclusion Criteria

Parents of newborns between 6 weeks to 4 years old, diagnosed via the KBTH NBS programme and enrolled in clinic but attending clinic for the first time. Parents of newborns, diagnosed via the KBTH programme, enrolled in clinic but less than 6 weeks or more than 4 years.

3.6 Sampling Method and Size

The researcher used the purposive sampling method to recruit participants. This method was used because the researcher needed parents of newborns who met a stipulated criterion and could provide some information about their experiences.

According to Francis et al., (2010), data saturation for theory-based qualitative studies can be decided by using 2 principles: 1. establishing an initial minimum sample size for analysis. And 2, specifying the stopping criterion i.e. how many more interviews will be conducted without new ideas. Prior to starting data collection, a total of 36 interviews were expected to be conducted, continuing until saturation. However, due to time constraints and other challenges, the sample size was modified to 18 with the stopping criterion being 3. Saturation was achieved after 24 parents were interviewed.

3.7 Recruitment of participants

Participants were recruited from the Paediatric Sickle Cell Clinic of KBTH. They were mothers and a father who met the inclusion criteria. In one case, both parents were available in the clinic on a particular day, as such, they were both interviewed together. Participants were recruited at the clinic using the purposive sampling method. The researcher used 2 interview instruments: a questionnaire to collect socio-demographic information and a semi-structured individual interview guide created by the researcher.

3.8 Pilot Study

The interview guide was piloted at the Paediatric Sickle Cell Clinic of KBTH once approval was received from the Institutional Review Board of KBTH (KBTH-IRB). Five participants were interviewed for the pilot study using semi-structured individual interview guides to conduct individual in-depth interviews. This provided the interviewer with a better

understanding of the interview guide as well as a chance to hear some likely questions some of the interviewees had. Piloting was done in October 2021, once approval was received from the KBTH IRB. The interviews were transcribed and modifications to the interview guide were made based on feedback from the pilot before the commencement of the study. The modifications to the interview guide included the addition of 2 more questions asking about the content of the disclosure of the results and the person who disclosed the results to participants during their initial visit or call. Transcripts from the piloting were not added to the main interviews for analysis.

3.9 Data Collection Approach

A Semi-structured interview guide was used to solicit information. Recruitment and commencement of interviews began from the first week of November 2021, once approval had been given from the KBTH IRB. A semi-structured individual interview guide was developed by the researcher using the conceptual framework and objectives of the study and included open-ended questions and socio-demographic questions. These interviews were conducted at the PSCC venue during clinic days. Each face-to-face interview per participant was conducted in a private space at the clinic venue whiles observing all COVID'19 protocols. Patients were required to wash their hands before entry into the clinic venue, have their temperature taken and wear face masks whiles inside, per the KBTH COVID 19 guidelines. Face masks were provided for those who did not have one with them, and hand sanitisers were provided at the entrance. The interviewer and interviewee were seated at least 6 feet apart from each other, each wearing a face mask. All members of the research team were also provided with face masks and hand sanitisers and trained on how to observe proper COVID 19 protocols. Items such as pens, ink pads, etc. were sanitized before being passed from interviewer to interviewee and vice versa during consent administration. Since patients attending the clinic are usually scheduled and given appointment dates before the visitation date, staff nurses usually have an

idea of who would be attending the clinic on a particular day. Each clinic day, an announcement was made by a staff nurse at the clinic about the study and the inclusion criteria were explained to participants. As such, individuals who fit the inclusion criteria and were willing to participate were interviewed during clinic hours.

Interviews were conducted after the participants had received services at the clinic and were exiting. Each Interview took between 40 minutes to an hour and was conducted in Twi or English and tape-recorded for transcription later. A field diary was used to take field notes which included notes taken during each interview and other observations made. Thirteen interviews were conducted in English whiles eleven were conducted in Twi.

NVivo software was used to assist in the analysis and coding of data. This software is used for qualitative and mixed methods research to analyse unstructured texts, audio etc. obtained from interviews, focus group discussions, surveys, social media, etc. into themes and patterns.

3.10 Data Handling

To ensure the privacy and safety of data collected, all audiotape recordings, field notes and memos were kept under lock and key and only accessible to authorized persons.

Sufficient controls were put in place to preserve the safety of the data as well as to guarantee its accuracy and devoid of any bias. As such, the researcher used various validation strategies which are further discussed under validity and threat to validity.

A research assistant, well versed in English and Twi languages was recruited and trained thoroughly on how to use reliable and accurate techniques to conduct interviews using an interview guide through demonstration and role-playing. Thus, interviews were conducted by the researcher with the assistance of the research assistant who made notes in the field diary.

3.11 Validity

Qualitative research uses various validation strategies to ensure trustworthiness, credibility, and transferability in a study.

Trustworthiness in a research study is how much confidence there is in a researcher's data, interpretation and methods used (Connelly, 2016). This usually reflects the quality of the study and as such, there is the need for researchers to clearly outline their protocols and procedures which are considered worthy of consideration.

Credibility refers to the confidence in the truthfulness of the findings of a research study. This is equivalent to internal validity in a quantitative research study. Credibility is examined by questioning if standard procedures of the qualitative approach chosen by the researcher were indeed used in the study (Connelly, 2016).

The transferability of a research study usually examines the degree to which the results of a study can be generalized or transferred to other settings or contexts (Connelly, 2016).

3.12 Validity and Threat to Validity

The three main threats to validity are researcher bias, reactivity and respondent bias. These threats to validity were addressed in this study by using various validation strategies such as reflexivity, iteration, constant comparative methods, saturation, triangulation, thick, rich descriptions, codebook and peer debriefing.

Credibility was achieved in this study by using a codebook, which served as a trail of evidence as well as the use of a field diary and a memo to record all observations and notes during interviews and analysis of data. Data was triangulated using the memo notes, field diaries and transcriptions from interviews to ensure trustworthiness and credibility.

Saturation was achieved in this study by utilizing the 2 principles of achieving saturation (Francis et al., 2010) where the initial minimum sample size was established before data collection and the stopping criterion was also established.

Another way of ensuring trustworthiness and credibility in this study was the use of 2 peer-debriefers in the person of my academic supervisor and a Haematologist nurse specialist and a researcher at the sickle cell clinic of Korle Bu Teaching Hospital. Transferability, another quality of validity was achieved by using rich, thick descriptions and quoting participants' voices under each theme.

The researcher used an iterative process of checking interpretations and analysis of data with the peer-debriefer as well as constantly comparing responses of participants to help influence the validity of the study.

3.13 Data Processing and Analysis

Data Analysis using thematic analysis was done after data collection had been concluded. The transcripts from the pilot were not added to those from the main study. Audiotaped interviews from the main study were transcribed verbatim after each interview and those conducted in the local languages were then translated into English. Thematic Analysis following the 5 phases of thematic analysis as defined by Braun & Clarke, (2006) was used.

Open coding (codebook) using an inductive approach was used as a first step in identifying initial codes. Similar codes were then clustered to form categories after which the relationships between the emerged codes were analysed and categorized into themes.

3.14 Ethical Considerations

Ethical approval for this study was sought and acquired from the Institutional Review Board of Korle Bu Teaching Hospital (KBTH). The approval reference number for the study was KBTH-IRB /00094/2021.

Consent was sought from participants before interviews began. Consent forms were given to parents to read and sign, whiles those who could not read had the consent translated verbally into the local language for them, after which they thumb printed their consent. Participants were assured of the confidentiality and anonymity of data by assigning pseudonyms to them and assuring them their real names would not be used. Also, to ensure the privacy and safety of data collected, all audiotape recordings, field notes and memos were kept in private under the investigator's care and accessible to authorized persons only.

No compensation was paid to participants of the study and participants were not exposed to any potential risk. However, participants were informed of this and made aware of how their contribution could help in formulating interventions and changes in current clinical care provision.

Participants were assigned unique codes and pseudonyms as identification. All information gathered was treated with the utmost confidentiality. The recordings and other research materials will be appropriately discarded after 5 years.

3.15 Reflexivity

Reflexivity is mainly used in qualitative research to establish a criterion of rigour and to monitor the researcher's subjectivity in generating credible findings. The researcher employed introspection and reflection during the interview stage and the analysis of data. Another way of using reflexivity to control subjectivity was the use of memos and field notes both at the interview stage at the data analysis stage. The memos and field notes provided details regarding context after each interview whiles memos were written at the analysis stage to help explore and act as a reminder of how initial codes would be defined and how patterns would be organized into themes.

This study provided an understanding of the perceptions of parents whose newborns tested positive for sickle cell disease. It provided insight into the reactions of parents to the disclosure of positive results as well as their attitudes towards follow-up clinic care. It provided information on the current practices as well as identified gaps so that appropriate interventions could be implemented. It provided an understanding of what influences parents' acceptance of initial positive results of sickle cell disease.

The principal investigator is a staff of the Korle Bu Teaching Hospital and is a member of the team that provides care to patients at the Paediatric Sickle Cell Clinic. The principal investigator, however, declares no conflict of interest concerning this study as this study is being self-sponsored in partial fulfilment towards the award of a Master of Public Health degree from the University of Ghana.

CHAPTER FOUR

RESULTS

4.0 Introduction

This chapter presents the experiences of parents of positive newborns with sickle cell disease attending the Paediatric Sickle Cell Clinic of Korle Bu Teaching Hospital. The results of data analysis are presented in this chapter as themes and sub-themes.

4.1 Socio-demographic Characteristics

A total of twenty-four (24) mothers and a father of newborns who had tested positive for SCD at birth and enrolled in the clinic were interviewed. Four of the participants had babies who were less than one year old. Four more participants had children aged one year to two years and sixteen had children aged two years one month to three years, and eleven months.

The participants consisted of seventeen (17) married and three unmarried parents. Two other participants were not married but in a relationship with the father of their babies whiles two were separated from their husbands. All participants lived within the Greater Accra Region. Nine of the participants had completed tertiary education, three had completed primary six, five had completed Junior High School (JHS), six had completed vocational or Senior High School (SHS), and one had not had any formal education at all. Five of the participants had more than one child enrolled in the clinic. Ten (10) of the participants also had the disease or had partners who had the disease whiles fourteen (14) participants had the SCD trait. More than half of the participants reported earning less than 500 cedis for the household per month. This reflects the socio-economic costs of the disease and provides insight into how socio-economic challenges influence parents' experience at clinic. The results are found in the table below.

Table 4.1 Summary of socio-demographic information of participants

Archival number	Age of child (years and or months)	Marital status of participant (s)	Place of residence	Educational level of participant (completed)	Number of children	Number of children with known SCD	One parent with known SCD
KBTHPSCCII001	1.7 years	married	Tabora	Tertiary	2	1	mother
KBTHPSCCII002	0.8	married	Accra	JHS	2	1	Mother
KBTHPSCCII003	0.7	Not married/in relationship with child's father	Dansoman	JHS	1	1	None
KBTHPSCCII004	1.8	married	Korle Gonno	SHS	1	1	none
KBTHPSCCII005	2.11	married	Ashaladza	Primary 6	2	1	none
KBTHPSCCII006	3.11	Married	Dansoman	Tertiary	3	1	father
KBTHPSCCII007	3.10	married	Kaneshie	SHS	2	1	mother
KBTHPSCCII008	0.9	married	Bortianor	none	5	1	none
KBTHPSCCII009	3.11	married	Amasaman	Primary 6	3	1	None
KBTHPSCCII010	2.11	married	Odorkor	Tertiary	2	1	Father
KBTHPSCCII011	3.10	Not married	Teshie	SHS	1	1	Mother
KBTHPSCCII012	3.1	married	Larterbiokorshie	JHS	2	1	None
KBTHPSCCII013	3.2	Separated	Ablekuma	JHS	5	2	Mother
KBTHPSCCII014	2.6	Not married	McCarthy Hill	SHS	1	1	Mother
KBTHPSCCII015	3.9	married	Sowutuom	SHS	4	3	None
KBTHPSCCII016	2.6	married	Ashongman Estates	Tertiary	2	1	None
KBTHPSCCII017	2.11	married	Afienhya	Tertiary	1	1	Mother
KBTHPSCCII018	2.0	married	Ablekuma	Tertiary	4	3	None

KBTHPSCCII019	0.7	Not married	Sowutuom	Primary 6	3	2	Mother
KBTHPSCCII020	3.8	separated	Fadama	Tertiary	2	1	None
KBTHPSCCII021	2.3	married	Banana Inn	Vocational	2	2	None
KBTHPSCCII022	2.0	Not married. In relationship with baby father	Korle Gonno	JHS	1	1	None
KBTHPSCCII023	2.1	married	North Legon	Tertiary	2	1	None
KBTHPSCCII024	2.4	married	Ablekuma	Tertiary	1	1	None

Data Analysis was done after data collection had concluded. Audiotaped interviews were transcribed verbatim after each interview and those conducted in the local languages were then translated into English.

4.3 Findings

The emerged codes were categorised using the socio-ecological model indicating the experiences of parents of positive newborns. The codes were categorised into two themes based on the socio-ecological framework, namely: intrapersonal influences and interpersonal influences. A summary of the codes and themes can be found in the codebook in appendix 5. The results are presented as follows: Intrapersonal influences (Emotional Response after Disclosure, Decision to Share Results, and Adherence to Routine Clinic) and Intrapersonal influences (Care and Management of the Disease at Home, Knowledge of Genetics of SCD).

4.3.1 Intrapersonal Influences

Participants reported a range of emotional reactions, feelings, as well as decisions made after disclosure of positive results as well as certain reasons for their attitudes towards follow-up care.

> *"So, when they called me, I got worried. So, when they called me told me, I said it's not true. And my child was a big baby. It was when he was hospitalized in Korle Bu for one week that I truly believed. Yes.*
>
> *(Female participant)*

4.3.1.1 Emotional Response after Disclosure

Participants were called on the phone and scheduled to visit the clinic after which they were informed about the results. One of the main themes identified was the emotional response of participants towards the results after disclosure.

Scared

Participants reported that they felt scared when they were initially told of the positive results. For participants, this reaction was due to the little knowledge or previous experience or preconceived ideas they had had about the disease.

> *"You know, life is hard now, and I was scared that if it happens that she is a sickler, what would I do?"*
>
> *(Female Participant, 41 years, JHS,)*

> *"I got scared. You know, it is money issues. So when they told me he had the disease, I got very scared".*
>
> *(Female participant, 41 years, Primary 6)*

> *"It's not easy oo. It's not easy ((with tears in voice)). Me knowing how it is. Especially, me, if I didn't even know how it is. But me knowing how the pain, hmmm, the, the, hmm, it's not easy oo. I'm facing a lot inside me… And I've been keeping this*

in me. And I don't know what to do. I don't know what to do. Hmm, it's really, difficult thing, ever since I got to know my son, has this disease.

(Female participant, 27 years, SHS)

Worried

Some participants also reported being worried after receiving the results. They reported they got worried because of the knowledge that sickle cell disease had a lot of socio-economic ramifications. As such, a positive diagnosis was very worrying for them.

Because, I can sometimes look at the child, and shake my head and think to myself "this disease too where is it coming from"? That I gave birth and need to be at the hospital every 3 months. It's a lot of worry"

(Female participant, 34 years, J.H.S)

Not surprised

Some of the participants on the other hand had not been surprised by the positive results of their newborns as they already had prior knowledge of the family history of the disease and as such, knew of the possibility of their baby testing positive for the disease. Thus, they were not surprised by the result.

"I wasn't surprised when they told me. Knowing that my other girl already had the disease. I understood".

(Female participant, 34 years, SHS)

So him ((pointing to child with SCD at clinic)), when his results came, I wasn't surprised because I knew the dad is SC. so I wasn't surprised at all.

(Female Participant, 34 years, Tertiary)

"I wasn't really surprised. Because I had been told that if I give birth to more children, all my children could have it"

(Female participant, 40 years, Primary 6).

Shocked/surprised

There were however several participants who reported being shocked or surprised by the results even though they had prior knowledge of the disease and knew that they had the trait and could therefore give birth to a child with the disease.

> *It did help. Because, even before we got married, I was contemplating. And then when the news came, even though it came as a shock, I knew, this was what was before us. And then. So, it was quite okay.*
>
> *(Female participant, 33 years, Tertiary)*

Disbelief

Some participants also, reported that they did not believe the results when they were initially told about the results. This was because they were not expecting the results. Some participants, however, then came to believe and accept the positive results when their children started showing symptoms of the disease.

> *"And then later she called and informed us that he had the disease but I didn't believe it. And then when we were 10 months, he started getting sick".*
>
> *(Female participant, 34 years, Primary 6)*

> *"That first day they gave me the results, I didn't believe it".*
>
> *(Female participant, 39 years, no education)*

However, some of the participants reported that even after receiving the positive results and having attended the clinic for a while they and or their partners still did not believe the results.

> *"no. he still doesn't believe. He said he donates blood to people in Korle Bu all the time. So if he had the disease, they wouldn't allow him to".*
>
> *(Female Participant, 39 years, no education)*

> *"So it's only my husband and I that know. And we don't believe in that thing, so, we don't see our child as SC".*
>
> *(Female participant, 28 years, Tertiary)*

4.3.1.2 Decision to Share Results

For most participants, the decision to share the positive results they had received at the clinic was based on different reasons. Mothers who came alone for that first clinic visit reported that they had shared the results with their partners or the child's father when they got home.

> *"I called my sister. And I told her, my sister has 4 children, so I told her that they said my baby has this disease and when she also gave birth, did they tell her something similar? And she said no. So, I was surprised. So when I got home, I told the father and I asked him whether his side of the family has any disease like this. And he said no".*
>
> *(Female participant, 27 years, JHS)*

Shared results with a partner only

Some however reported that apart from their partner or the child's father, they had not shared the results with any other person. They stated they had kept the results just between the 2 of them and not shared it with any other person.

> *"Well, I didn't want anyone to know. I just wanted it to be between me and my husband so I didn't tell anyone"*
>
> *(Female participant, 41 years, JHS)*

Fear of social exclusion and stigma

Some of the reasons parents gave for not sharing the results with extended family and friends included fear of judgment, social exclusion and stigma.

> *"Hmm, for my family, I don't really mind telling them, but it's because, I don't want people to say the child is this or the child is that. Yes, I don't want someone to stand somewhere and tell another person about it and say something like oh this child is beautiful but ahh, she has this disease. And then it will reduce her marks or something. I don't want that. Yes, you see people will talk. Maybe they don't mean anything bad but"*
>
> *(Female participant, 41 years, JHS)*

> *"Nobody oo. I've not told anybody. Because of the stigma, I don't want to, to tell anybody… Because of what I went through, I don't want to tell people about it. My mom didn't keep quiet and people would tease me and all that. So. I don't want that".*
>
> *(Female Participant, 32 years, SHS)*

Shared results with family and friends

Some parents, however, did not mind sharing the results with some family and friends as they felt those people were closer to them and were part of providing care for the children. As such, they felt they needed to know to help them care for the child.

> *"Yes. I told the caretaker. Because if I have to leave, and you don't give the medication, I will blame her. And then recently, I told my mother-in-law… and then my parents, my siblings are also aware. So that in case there is something to give, I tell you, chilled things are not for her. I'll tell you please, hold on".*
>
> *(Female participant, 33 years, Tertiary)*

4.3.1.3 Adherence to Routine clinic

Parents reported that they attended the clinic regularly, sometimes, every two months. They reported that they routinely and regularly brought their child to the clinic for checkups and follow-ups. Others also reported that they attended the clinic as and when they were scheduled and given an appointment to visit.

Adherence due to the expectation for a healthy outcome for child

Parents reported attending the clinic regularly because they wanted their children to remain healthy.

> *"I've been coming to clinic for the past 7 to 8 months. Every month. I'm here. Yes, sometimes even 2 weeks. In a month, I can come like 2 times … for me, I always say that because of his health. I want him to be healthy. And for his breathing to be fine. Because sometimes, when we come, they check everything, his breathing, they do a physical exam of him to make sure everything is fine. So, I bring him to make sure everything is fine. Because when we are home, everything is fine. But, when I'm given a date to come, I have to come".*
>
> *(Female Participant, 30 years, SHS)*

Adherence due to parents' personal experience with the disease

Others also reported adhering to regular monthly or bi-monthly clinic visitation due to previous experience of parents living with the disease and as such found it necessary to keep the child healthy.

> *"Because I have that experience. So I don't want her to pass through the stress I have gone through, so I come to clinic".*
>
> *(Female participant, 36 years, SHS)*

Non-adherence due to financial difficulties

There were however some who reported that they sometimes missed appointments due to circumstances such as financial difficulty

> *"Madam please, you see, for my son, I love him. And it's the doctor who knows, so whatever they tell me I have to listen and do it. But sometimes, when I'm unable to come on the date, it's usually because of financial difficulties. I may be lacking money for the labs .... But financially, it's been difficult. Because of money issues. I was supposed to bring him last month but because I didn't have money, I couldn't come".*
>
> *(Female participant, 41 years, Primary 6)*

Non-adherence due to COVID-19 pandemic

Some of them also reported that they did not attend the clinic for a while due to the COVID 19 pandemic. They reported that even though they were scheduled for regular clinic visitations, some of these circumstances prevented them from visiting the clinic regularly.

> *"We come. Every 3 months, we come. But when he was 2 years, we were asked to come and do some labs. But we couldn't do it. Because of the COVID. I was scared so I didn't come. And then I was called in November to bring her. And we will come again in February".*
>
> *(Female participant, 34 years, JHS)*

Adherence to daily medication

For the basic medication, all parents reported that they strictly followed the guidelines of daily medication for their children. Parents reported always refilling the medication and stated that they always made sure to give the child medication every day, as prescribed by specialists.

> *"Every day, I give her the medication. I follow all the instructions I'm given. I give her every day. So she is fine and she is very fast too".*
>
> *(Female participant, 31 years, Tertiary)*

> *"Every day. I give her the medication. Every day. When it finishes, I go back and buy. Yes, the other time I came, the doctor wrote another prescription with the same medication for me so, now I know it is the same medication I have to keep giving her. So once it finishes, I buy more. Once it runs out, I call my husband and if he has money, he sends it".*
>
> *(Female participant, 41 years, JHS)*

All parents also reported making sure to refill the prescription every time it finished.

> *"And so because of that, I don't play with the medication. Once it finishes, I make sure I refill it. And he is fine. He has not been sick since. Once in a while he gets a cold, and when that happens, I take him to Amasaman, then they give us medication and we go back home. That's it".*
>
> *(Female participant, 34 years, Primary 6)*

Routine laboratory tests

Parents reported that they were routinely asked to do lab tests such as full blood count, reticulocyte count, and sometimes liver function tests, among others. This they stated, was done routinely, most often once a month. Participants reported that they usually adhered to whatever lab requests they were given to do.

> *"And if I have to do any labs, if I have money, I hurry and do it, so the doctors can check his blood levels. So, I believe what they say is good for me".*
>
> *(Female participant, 41 years, JHS)*

> *"Oh it's usually the lab we frequently do"*
>
> *(Female participant, 41 years, Primary 6)*

4.3.2 Interpersonal Influences

Parents reported a range of intrapersonal influences due to their network with staff at the clinic. They were educated on the care and management of their newborns at home as well as the genetics of SCD during counselling and educational sessions as part of comprehensive follow-up care provided at the clinic.

4.3.2.1 Care and Management of Newborns with SCD

Parents reported that they received education on SCD during regular counselling and educational sessions with trained staff at the clinic. As such, they were able to acquire lots of knowledge on SCD.

Preventive measures

Parents reported they were counselled on how to care for their newborns at home and how to take preventive measures to reduce illnesses and painful episodes at home.

Nutrition and diet for newborns

They reported that they were counselled on how the nutrition or diet of the child is important and could improve the health of the child.

> *"They said to pay attention and take care of him well, his eating habits. The food he eats... They said I should pay attention to that. So that he doesn't become sick. Yes".*
> *(Female participant, 39 years, no education)*

Hydration

Some parents also stated that they were advised to make sure the child drank lots of water and was always hydrated. Thus, they were advised on the importance of hydration for their children with SCD.

"Yes. I give him lots of fruits. He drinks water too. A lot. I make sure he wears pullovers and stuff".

(Female participant, 31 years, Tertiary)

Warm clothes

Parents reported that they were also educated on the need to keep their babies warm.

"Hmm, they said many things. How to how to, ehh, is it, prevent it, ehehn, in terms of not to take something cold. And all those kinds of things. You will know the way when it's cold, you should put something, like cardigan, all those kinds of things. And you have to take your medication always, no crises, for the 0 months to 5 years. You won't have any problem".

(Female participant, 36 years, SHS)

Awareness of treatment options

Medication literacy was one of the things imparted to parents as well and as such, parents reported that they were now aware of the medication as well as other treatment options necessary for managing the disease. Parents reported being advised to give their children penicillin, zinc supplements, folate supplements and Hydroxyurea

He's taking, umm, brufen, zincovit, folic acid, penicillin V. but recently he was given the … I've forgotten the name… yes. Hydroxyurea

(Male and female participants, 28 and 28 years, JHS and Tertiary)

"And the doctor said not to think too much of it. So, when he keeps taking the medicine, small small it will become fine. Maybe the SS too can change. Because it's good to treat them when they are little. Before they grow up".

(Female participant, 30 years, SHS)

Awareness of danger signs

Parents informed us they were counselled on what danger signs to watch out for and actions to take to mitigate any symptoms at home. Parents were counselled and gained knowledge on watching out for danger signs such as yellow eyes, fever, swollen feet and hands, constant crying, etc.

"They said for sickle cell, if I see he is in pain or he gets a fever, if his urine become coke colored, I should bring him".

(Female participant, 28 years, Vocational)

"They said he may get complications from the disease. When they are babies, maybe you will see one of their hands swelling or their feet swollen. Or their eyes. And when they get temperature, you have to bring them to the hospital and not stay in the house. It might be infection".

(Female participant, 32 years, Tertiary)

Awareness of possible complications from the disease

Parents stated that they were counselled on the possible complications that could emerge from the disease. They reported that they were taught about how the disease could affect other parts of the body such as the eyes, the kidneys, etc.

"They told me the disease can affect other parts of the body. That's why i was so sad when i couldn't get the eye test for my son when his eye got swollen".

(Female participant, 41 years, primary 6)

4.3.2.2 Knowledge of Genetics of SCD

Participants reported that they were educated on the nature of the disease as well as the genetics of the disease and the possible causes of the disease.

Inheritance of SCD

They reported that they were taught that the disease was inherited from both parents. Parents were made aware of the fact that they both had to have the trait to be able to have a child with the disease and that the disease was found in their genes and was passed down to their child. They also reported being informed of the possibility of having other children with the disease.

"Yes. The day I brought the results, the doctor asked that both my husband and I come. So, we came and he explained everything to us. They explained everything. That

because my husband is AS and I'm AC, that's how come. He took my husband's S and took my C. the older one took my husband's S and my A".

(Female Participant, 31 years, Tertiary)

"They told us. Ehh. They told us that it's genetic. If one of the parent has, then probably 1 or 2 of your children will have it"

(Female participant, 32 years, Tertiary)

Blood disorder

The parents also reported that they were also educated on the fact that sickle cell disease is a blood disorder and that it is sickle cell shaped instead of round and breaks easily thus causing painful episodes.

"They took me through the sickling. How it forms. How their blood cells look like. Is it the white one or the, the red one? Yea, that looks like a bofloat, and theirs is like an arch, the moon, half-moon, which is very hard not like the others that are soft that can pass through all the blood. Theirs because is hard, sometimes, it gets broken and chokes the veins that you have to give them the (massage) and all that. So, I was taught all that".

(Female participant, 34 years, Tertiary)

"They told me that there is S in the child's blood. And she showed me how it is, with a diagram, how the S looks in the blood, she drew it and showed me. And how that for us, the blood is not round but is curved. And that is how come it happens that way. And so, they will try and treat it. She said it is something that's happening in Africa. Sickle cell disease. And she said so nowadays when they see that a child has the disease, they advise parents to bring the child to hospital for care so that it can prevent certain complications".

(Female participant, 41 years, JHS)

Family history of the disease

Participants also stated that they were asked if they were aware of any known family member with the disease. They reported that they were informed that sickle cell was a familial disease passed down from parents to the child and could affect other family members.

"Because my father, for instance, he also has it … we are 3. My sister also has the disease. Hers is worse".

(Female participant, 37 years, Primary 6)

"It's me and our last born (meaning youngest sibling) who have the disease in our family".

(Female participant, 41 years, primary 6)

4.3. 3 Emerged Themes and Sub-themes

The codes were organized and six (5) themes emerged after analysis. These were: Emotional Response after Disclosure, Decision to Share Results, Care and Management of Newborns with SCD, Knowledge of Genetics of Sickle cell disease, and Adherence to Routine clinic. Out of these 5 themes, a total of fourteen (19) sub-themes were identified. A list of the themes and the sub-themes are presented in Appendix 5.

Summary of Findings

In summary, findings from this study revealed the various experiences of parents of newborns diagnosed with SCD at birth. All participants lived in the Greater Accra Region. Seventeen of them are married, three are unmarried, two are unmarried but in a relationship with their partners, and two of them are separated from their partners.

Parents with newborns who tested positive for SCD revealed a wide range of experiences concerning the themes identified. Their experiences were categorised according to the socio-ecological model's two constructs namely: intrapersonal influences and interpersonal influences. They expressed that they had experienced certain emotional responses after the disclosure of the results such as being scared, not being surprised, and disbelieving or not expecting the results.

The participants also stated that they had received counselling and education on SCD and as such now had knowledge of genetics and causes of SCD, knowledge on SCD, as well as family history of the disease.

Parents again stated that they followed routine clinic and medication guidelines.

CHAPTER FIVE

DISCUSSION

This chapter discusses the findings of the study. The main themes and the sub-themes have been presented as they relate to the objectives of the study.

5.1 Reactions of parents to disclosure of initial positive results

The first objective of this study was to explore how parents reacted when the initial positive results were disclosed to them. There were therefore two themes that emerged from the analysis of the results that provide an answer to this question: Emotional response to the disclosure of results and the decision to share results.

5.1.1 Emotional Response to the disclosure of results

This study concentrated on the emotional responses parents felt or expressed regarding the results as well as their decision to inform or not inform close family and friends about the diagnosis.

The findings of the study uncovered that fear and worry are some of the emotions parents feel when given initial positive results of their newborns. This doesn't differ much from a study by Chudleigh et al. (2016) where they explored the experiences of parents of newborns receiving positive SCD carrier results. In that study, parents reported a range of emotions such as relief, devastation, guilt, denial, surprise and shock. As much as these emotions may not have been the same, they were still negative emotions that were felt by parents receiving positive results. Some known effects of newborn sickle cell carrier status disclosure to parents are anxiety or distress (Ulph et al., 2015).

From this study, disbelieving initial positive results is another reaction of parents to the disclosure of positive results. Some parents expressed that they didn't believe the results

initially but later accepted while others stated that they and or their partners still did not believe the results. This is somewhat worrying as it has implications on whether or not parents would then accept the results and enrol their child in the clinic for care. Parents' reasons for disbelief were due to misconceptions and misinformation about the disease and as such, they believed that their children could not have the disease.

Another group of participants stated that they were not surprised by the results. This they said was because of their prior knowledge of the disease in their family.

One important thing this study revealed was that prior knowledge of the disease and or personal experience with the disease has a huge impact on how parents react to positive results of SCD. Parents who had prior knowledge due to family history of the disease stated that they were not surprised by the results whiles those who may have heard about the disease but did not know their carrier status were scared, worried and surprised by the results of their newborns. Thus, prior knowledge of the disease impacts parents' emotional reactions to the disclosure of positive SCD results of their newborns. This is consistent with a study by (Niekerk, 2015), where she found out that parents of children with the disease had misconceptions about the disease due to incorrect or inaccurate information and this, therefore, affected their expectations of treatment and disease progression.

5.1.2 Decision to Share Results

This study's findings revealed the decision to share the initial positive results was something that many parents did not take lightly due to issues such as stigma. In this study, most parents of newborns decided to share the positive results/ or diagnosis with someone, at least a partner with some preferring to keep the diagnosis just between them and their partners. Some parents' decision not to share the results was based on the desire to not subject their child to stigma and also not make their child less socially accepted among friends and even family. A few of the

parents however made decisions to share the results with their partners and a very few close family members.

This is in contrast with a study by Ulph et al., (2015) documenting parents' responses to receiving sickle cell carrier results for their newborns, where parents felt a responsibility towards sharing the results with their extended family and parents had no struggle with informing extended families about the results. For others, the decision to share with at least just their partners was because of the expected support they could receive from them.

A study by Chudleigh et al., (2016) also contrasts the findings in this study. In Chudleigh et al., (2016) study, most of the parents of newborns who had received positive SCD carrier results of their newborns shared results with family and friends. In this same study, however, some of the parents who had lived in Africa or had family back in Africa hesitated to inform that side of their family in Africa because they felt their families had a bleak outlook on the disease. It can therefore be said that geographical context and culture influence peoples' experience of stigma and this in turn influences parents' decision to inform other people about the results of the newborns.

Social acceptance and stigma were thus a very major part of the decision of parents to either share or not share the positive SCD results of their newborns.

This may an implication in the acceptance of initial positive results as well as adherence to follow-up clinic procedures such as routine clinic visits. This is because parents who are afraid to share results with close family and friends may also be afraid or worried to send their child to a clinic for regular visits for fear of people discovering that the child has the disease.

5.2 In-clinic Counselling and comprehensive follow-up care

The second research question aimed to look at how parents responded to in-clinic counselling and education and how this formed an essential part of comprehensive follow-up care. It aimed

to provide insight into parents' relationships and interactions with staff at the clinic in terms of the counselling and education provided by trained staff at the clinic. Analysis of data yielded two themes that answer this question: knowledge of care and management of newborns with SCD and knowledge of genetics of SCD.

5.2.1 Care and Management of Newborns with SCD

Counselling patients with sickle cell disease is an essential tool in ensuring effective adjustment (Adegboyega, 2020).

Educating parents on how to care for their children at home is one of the ways of reducing bacterial infections in children (Makani et al., 2013). Parents should be advised on giving their children enough fluids to reduce dehydration. They should also be advised on wearing warm clothes in cold weather, and the need for ensuring proper diet and nutrition (Johnso et al., 2015; Makani et al., 2013).

This study's findings uncovered that parents of newborns were counselled and received a lot of knowledge on the nature of sickle cell disease such as preventive measures and care at home for their child. Parents stated that they were advised during counselling sessions with staff to make sure to take care of their newborns at home by ensuring they stayed hydrated, warm and had adequate nutrition.

The findings of the study further revealed that parents of newborns were advised on the treatment options available for their children. As such, parents knew what medication they were prescribed and how that medication played a role in managing the disease.

This is consistent with a study by Colah et al., (2018) documenting the Indian experience of newborn screening for SCD where parents were given daily folic acid and penicillin prophylaxis for their newborns. It is also consistent with the Jamaican experience study by King et al., (2007) where parents of newborns diagnosed via newborn screening were

counselled on the need for routine penicillin prophylaxis. Medications such as folic acid, vitamin c, and penicillin have been known to reduce infections among infants with SCD.

Parental education on possible complications of the disease on their children was also divulged from the findings. Clinic staff educated parents on the possible complication outcomes of the disease such as end-organ damage, and splenic sequestration, among others.

Findings also showed that parents of newborns were educated on watching out for danger signs such as yellow eyes, coke-coloured urine, bloated stomach, swollen joints, and excessive crying. These, they revealed were the first indications or symptoms of impending infections or other complications of the disease. Parents were advised to watch out for these and report to the nearest health centres.

This is consistent with the study by King et al., (2007) of the Jamaican experience of newborn screening where parents were educated on how to watch out for symptoms and also check out for splenic sequestration by performing splenic palpation.

In general, the parents of newborns who have tested positive for SCD receive advice and counselling on the nature of sickle cell disease. The evidence from this study reinforces the need for involving parents in the comprehensive follow-up care of newborns with SCD.

5.2.2 Knowledge of Genetics of Sickle Cell Disease

Once a sickle cell diagnosis has been confirmed after the newborn screening, parents need to be educated on the type of SCD their child has, the genetics of the disease and how to manage the disease at home (Johnso et al., 2015). As part of comprehensive care provided for managing sickle cell disease in newly diagnosed infants, parents play a huge role in managing the disease. As such, parental education and counselling are essential tools.

Educating parents on the genetics of sickle cell disease is essential as uncovered by this study. Parents were educated on what the disease was, its causes, how the disease is inherited and

how family history plays a role in the inheritance of the disease. Findings from this study showed that as part of comprehensive follow-up care for newborns diagnosed with SCD at birth, parents need to be taken through several counselling and educational sessions or programmes with trained SCD nurses or staff trained in genetic counselling. This is to provide information about the genetic make-up of the disease, the way the disease is inherited through the genes of biological parents and how the history of the disease in the family may play a role in families having a better understanding of the disease make up.

This is similar to the Jamaican Experience study by King et al., (2007) where after newborn screening and diagnosis, parents were counselled on the genetics of the disease and how to perform spleen palpitations at the first clinic visit and then given more education during subsequent clinic visits. This study revealed that mortality and morbidity were reduced due to preventive measures adopted by clinic staff and parents as part of comprehensive follow-up care.

These findings from this study, therefore, revealed that, as part of comprehensive follow-up care for newborns diagnosed with sickle cell disease, parents play a huge role in this and should receive education and counselling on the genetics of sickle cell disease to get a better understanding of how to prevent painful episodes and other complications.

Prior knowledge about SCD could lead to people forming misconceptions about the disease thus affecting treatment plans and this was uncovered by findings from the study. As such, what counselling and education do, is that they clear these misconceptions and wrong ideas about the disease and the need for treatment. This thus can be important where there is the need to improve adherence to follow-up care by parents in the clinic.

5.3 Parents' adherence to follow-up care procedures

The third research question sought to explore parents' attitudes towards follow-up care procedures, i.e. Adherence or non-adherence. One major theme emerged from this section, namely routine clinic care.

5.3.1 Adherence to Routine clinic

Routine clinic visitation, regular physical examinations and recording of clinical and laboratory information of patients with SCD is important (Johnso et al., 2015). Routine clinical procedures for parents at the Korle Bu Teaching Hospital included routine lab tests, daily medication and routine clinic visitation for physical examinations.

It was discovered from the findings of the study that parents adhered to these routine clinic requirements due to their desire to see their children healthy as well as from their own experiences with the disease. This is in contrast with a study by Ingerski et al., (2017) into the clinic attendance of youth with SCD on Hydroxyurea where it was found that family size, financial capacity, incidence and non-incidence of painful episodes were reasons why the youth were more likely to attend clinic regularly.

There were however some parents who could not adhere to these routine procedures due to socio-economic challenges as well as the COVID 19 pandemic as shown by findings from the study. The findings of COVID 19 are similar to a study by Kenney et al., (2021) into how COVID-91 affected ambulatory services and comprehensive care for people with SCD which revealed that COVID 91 induced stress and anxiety contributed significantly to an increase in patients' no-show and patients rescheduling. It is therefore important for hospital administration to plan better to be able to adjust to continue to provide care for newborns whose parents may be too anxious or apprehensive to attend a clinic during a pandemic.

Sickle cell disease is known to cause financial difficulties for families of people with the disease (Da Silva De Jesus et al., 2018) and financial difficulties were one of the reasons why parents could not adhere to the routine clinic visitation and lab tests as they could not afford it.

5.4 Limitations of the Study

One limitation of the study is that this study did not explore the role of psychosocial factors in influencing parents' acceptance of positive results and their attitudes toward routine follow-up care.

Another limitation of this study is that the opinions of the clinic staff and other people involved in the provision of comprehensive care were not included. This may have given a more thorough insight into the experiences of parents of newborns at the clinic, especially with regards to the counselling and education offered to parents.

Finally, this study was a cross-sectional study and it was conducted at just one point in time at one health centre and as such, findings cannot be generalized.

Parents were taught about the nature of the disease as well as management of the disease thus cementing the importance of involving caregivers in the comprehensive follow-up care of newborns with SCD.

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CHAPTER SIX

SUMMARY, CONCLUSIONS, AND RECOMMENDATIONS

This chapter summarizes the findings of the study and recommendations.

6.1 Summary of study

The study utilized an adaptation of the Socio-Economic Model of Health to explore the experiences of parents of newborns who had been diagnosed with sickle cell disease at the Paediatric Sickle Cell Clinic of KBTH. Ethical approval was obtained from the Institutional Review Board of Korle-Bu Teaching Hospital (KBTH-IRB). Piloting was done at the same clinic with 6 participants and modifications were made to the semi-structured individual interview guide after which data collection began. A total of 24 interviews were conducted and consent was obtained from parents before the interviews. Recruitment of participants, as well as data collection, transcription and analysis, was done between November 2021 and March 2022. The data was analysed by open coding to generate a list of codes which were then categorised into themes and sub-themes using NVivo.

6.2 Conclusions

It was discovered from findings from this study that parents felt different emotions when the positive results were disclosed to them. Fear, Worry, Surprise, Shock, and Disbelief are some of the emotions parents feel when the initial positive SCD results of their newborns are disclosed. The reasons for these emotional reactions stem from prior knowledge of the disease, misconceptions about the disease as well as parental experience with the disease. Some parents decide to share the results with their close family and friends whiles others decide not to share the result. Reasons for this decision include the fear of social exclusion and stigma as well as cultural and geographical context.

This study also discloses that counselling parents on preventive measures and other home care measures help to reduce infections and painful episodes. Education on genetics of SCD is also an important part of the comprehensive follow-up care process. As such, it was discovered that parents play an important role in providing comprehensive follow-up care for newborns who have tested positive for SCD.

Parents' adherence to routine clinic procedures such as routine visitations, regular laboratory testing and daily medication was also discovered by findings. Parents adhere to these follow-up procedures due to the desire to keep their newborns healthy as well as the parents' own experience with the disease which makes them understand the need to ensure their babies are followed up. There are however some parents who do not adhere to these procedures due to certain challenges such as a pandemic and other financial difficulties.

6.3 Recommendations

Considering the conclusions of the study, the following recommendations are being made:

KBTH Paediatric Sickle Cell Clinic

The Clinic Administration should:

- Make efforts to increase genetic education about sickle cell disease to pregnant mothers at the Ante-Natal clinic by liaising with the Public Health Unit. SCD education can be included as part of the Ante-natal talks offered every morning.
- Offer continuous counselling sessions for mothers at the clinic i.e. keep counselling and educating parents about SCD and the care and management of their children in multiple, continuous counselling sessions. This is to make sure they do not forget all the things they have been taught.

- Develop SOPs about how clinic staff should monitor parents' follow-up activities i.e. routine visitations, lab tests, and medication.
- Conduct more research to assess the counselling offered to parents by the pre-screening nurses. This would help find gaps in the genetic education given to parents prior to screening their newborns and provide the opportunity to improve upon the process. Thus, more parents especially those with the SCD trait, would have an idea of the possibility of their child having the disease. This would help reduce the non-acceptance of results by parents.

KBTH Administration

The KBTH Administration should:

- Lobby the NHIS, MoH, Ministry of Finance, GHS and other governmental agencies and stakeholders to get all the basic/routine SCD medication and all the rest of the routine labs not yet under NHIS to be covered by the NHIS. This will reduce the financial burden on parents greatly.
- Offer an Amenity fund/ patient fund to parents of children with SCD. This can be done by using IGF or by appealing to NGOs and other agencies to donate to the fund. As such, parents who cannot afford medication and other routine services can apply for and access this fund to help pay for these services.

Parents of Newborns diagnosed with sickle cell disease

- Parents should advocate for various health sector stakeholders such as the Ministry of Health, NHIS, and Ghana Health Service to put all lab tests and all medications for sickle cell disease under the NHIS. This will reduce the financial strain on parents and help increase attendance.

- Parents (both mother and father) should endeavour to know their genotype especially whiles the mothers are pregnant. This would prepare them for positive results for their newborns. This study revealed that prior knowledge of the disease and parents' previous experience with the disease played a major role in their acceptance of the initial positive results of their babies.
- Parents should form support groups where they can provide information, advice, and general support to each other. These support groups can then organise and advocate for more education on SCD and newborn screening.

The Ministry of Health (MoH) and Ghana Health Service (GHS)

- Liaise with stakeholders to get all SCD medication and other lab tests not covered by the NHIS covered by the NHIS.
- Work together to implement newborn screening for SCD and other genetic conditions nationally.
- MoH should liaise with GHS to offer SCD care at the primary health centres with referrals to a specialist in case of complications. This can be done by training and getting paediatricians stationed at all District hospitals. This will make it easier for parents to adhere to routine follow-up visitations since they would be able to attend a clinic at a health centre closest to them.

Chapter Summary

In this chapter, the summary, the limitations, conclusions and recommendations of the study were presented. This study used the Socio-Ecological model of health to explore the experiences of parents of newborns diagnosed with sickle cell disease at the Paediatric Sickle Cell Clinic (PSCC) of Korle Bu Teaching Hospital (KBTH). There were five themes presented

in the findings. The limitations of this study included the omission of psychosocial factors in determining parents' response to positive results among others. Recommendations were made to the PSCC administration, KBTH, Ministry of Health and Ghana Health Service and parents of newborns enrolled in the clinic.

References

Adebowale, T. (2014). The Role of Social Welfare Counselling in the Eradication of Sickle-Cell Disease in Nigeria Beyond 2025. *African Research Review*, *8*(2), 1. https://doi.org/10.4314/afrrev.v8i2.1

Adegboyega, L. O. (2020). Counselling Needs of Sickle-Cell Anaemia Adolescents in Ekiti State, Nigeria. *Ethiopian Journal of Health Sciences*, *30*(6), 1005–1010. https://doi.org/10.4314/ejhs.v30i6.19

Agrawal, R. K., Patel, R. K., Shah, V., Nainiwal, L., & Trivedi, B. (2014). Hydroxyurea in sickle cell disease: Drug review. *Indian Journal of Hematology and Blood Transfusion*, *30*(2), 91–96. https://doi.org/10.1007/s12288-013-0261-4

Asare, E. V., Wilson, I., Benneh-Akwasi Kuma, A. A., Dei-Adomakoh, Y., Sey, F., & Olayemi, E. (2018). Burden of Sickle Cell Disease in Ghana: The Korle-Bu Experience. *Advances in Hematology*, *2018*. https://doi.org/10.1155/2018/6161270

Avard, D., Kharaboyan, L., & Knoppers, B. (2016). Newborn screening for sickle cell disease: Socio-ethical implications. *First Do No Harm: Law, Ethics and Healthcare*, *January*, 495–509. https://doi.org/10.4324/9781315582450-39

Braun, V., & Clarke, V. (2006). *Using thematic analysis in psychology*.

Bulgin, D., Tanabe, P., Carolina, N., Carolina, N., Hill, C., & Carolina, N. (2019). Stigma of Sickle Cell Disease: A Systematic Review. *Issues in Mental Health Nursing. Taylor and Francis Ltd.*, *39*(8), 675–686. https://doi.org/10.1080/01612840.2018.1443530

Chakravorty, S., & Williams, T. N. (2015). Sickle cell disease: A neglected chronic disease of increasing global health importance. *Archives of Disease in Childhood*, *100*(1), 48–53. https://doi.org/10.1136/archdischild-2013-303773

Chudleigh, J., Buckingham, S., Dignan, J., O'Driscoll, S., Johnson, K., Rees, D., Wyatt, H., & Metcalfe, A. (2016). Parents' Experiences of Receiving the Initial Positive Newborn

Screening (NBS) Result for Cystic Fibrosis and Sickle Cell Disease. *Journal of Genetic Counseling*, *25*(6), 1215–1226. https://doi.org/10.1007/s10897-016-9959-4

Colah, R. B., Mehta, P., & Mukherjee, M. B. (2018). Newborn screening for sickle cell disease: Indian experience. *International Journal of Neonatal Screening*, *4*(4), 1–8. https://doi.org/10.3390/ijns4040031

Connelly, L. (2016). Trustworthiness in qualitative research. *MedSurg Nursing*, *25*(6), 435+. https://go.gale.com/ps/anonymous?id=GALE%7CA476729520&sid=googleScholar&v=2.1&it=r&linkaccess=abs&issn=10920811&p=AONE&sw=w#:~:text=Trustworthiness or rigor of a,Pilot %26 Beck%2C 2014).&text=Credibility of the study%2C or,Polit %26 Beck%2C 2014).

Da Silva De Jesus, A. C., Konstantyner, T., Lôbo, I. K. V., & Braga, J. A. P. (2018). Socioeconomic and nutritional characteristics of children and adolescents with sickle cell anemia: A systematic review. *Revista Paulista de Pediatria*, *36*(4), 491–499. https://doi.org/10.1590/1984-0462/;2018;36;4;00010

Dennis-Antwi, J. A., Ohene-Frempong, K., Anie, K. A., Dzikunu, H., Agyare, V. A., Okyere Boadu, R., Sarfo Antwi, J., Asafo, M. K., Anim-Boamah, O., Asubonteng, A. K., Agyei, S., Wonkam, A., & Treadwell, M. J. (2019). Relation Between Religious Perspectives and Views on Sickle Cell Disease Research and Associated Public Health Interventions in Ghana. *Journal of Genetic Counseling*, *28*(1), 102–118. https://doi.org/10.1007/s10897-018-0296-7

DeTolve, G. R. (2016). Newborn screening. In *Health Care for People with Intellectual and Developmental Disabilities Across the Lifespan* (Vol. 31, Issue May). Springer International Publishing. https://doi.org/10.1007/978-3-319-18096-0_55

Francis, J. J., Johnston, M., Robertson, C., Glidewell, L., Entwistle, V., Eccles, M. P., & Grimshaw, J. M. (2010). What is an adequate sample size? Operationalising data

saturation for theory-based interview studies. *Psychology & Health*, *25*(10), 1229–1245. https://doi.org/10.1080/08870440903194015

Harif, M., Weisdorf, D., Novitzky, N., Szer, J., Mahmal, L., Benakli, M., Ben Othman, T., Bazuaye, N., McGrath, E., Eldridge, P. W., Torjemane, L., Madani, A., Ahmed Nacer, R., Belkhedim, R., Rasheed, W., Ahmed, S. O., Kodera, Y., Aljurf, M., Niederwieser, D. W., & Quessar, A. (2020). Special report: Summary of the first meeting of African Blood and Marrow Transplantation (AfBMT) group, Casablanca, Morocco, April 19–21, 2018 held under the auspices of the Worldwide Network for Blood and Marrow Transplantation (WBMT). In *Hematology/ Oncology and Stem Cell Therapy* (Vol. 13, Issue 4). King Faisal Specialist Hospital & Research Centre. https://doi.org/10.1016/j.hemonc.2019.05.003

Hsu, L., Nnodu, O. E., Brown, B. J., Tluway, F., King, S., Dogara, L. G., Patil, C., Shevkoplyas, S. S., Lettre, G., Cooper, R. S., Gordeuk, V. R., & Tayo, B. O. (2018). White Paper: Pathways to Progress in Newborn Screening for Sickle Cell Disease in Sub-Saharan Africa. *Journal of Tropical Diseases*, *06*(02). https://doi.org/10.4172/2329-891x.1000260

Ingerski, L. M., Arnold, T. L., Banks, G., Porter, J. S., & Wang, W. C. (2017). Clinic Attendance of Youth With Sickle Cell Disease on Hydroxyurea Treatment. *Journal of Pediatric Hematology/Oncology*, *39*(5), 345–349. https://doi.org/10.1097/MPH.0000000000000859

Johnso, P. L., Lombardo, F. A., & Moore, C. D. (2015). Options in the management of sickle cell disease. *U.S. Pharmacist*, *40*(7), 4–7.

Kai, J., Ulph, F., Cullinan, T., & Qureshi, N. (2009). Communication of carrier status information following universal newborn screening for sickle cell disorders and cystic fibrosis: Qualitative study of experience and practice. *Health Technology Assessment*,

13(57), 1–82. https://doi.org/10.3310/hta13570

Kenney, M. O., Becerra, B., Beatty, S. A., & Smith, W. (2021). Physicians' opinions of covid-19 ambulatory care constraints: A survey of sickle cell clinicians. *Journal of Ambulatory Care Management*, *44*(4), 325–330. https://doi.org/10.1097/JAC.0000000000000386

Kilanowski, J. F. (2017). Breadth of the Socio-Ecological Model. *Journal of Agromedicine*, *22*(4), 295–297. https://doi.org/10.1080/1059924X.2017.1358971

King, L., Fraser, R., Forbes, M., Grindley, M., Ali, S., & Reid, M. (2007). Newborn sickle cell disease screening: The Jamaican experience (1995-2006). *Journal of Medical Screening*, *14*(3), 117–122. https://doi.org/10.1258/096914107782066185

Makani, J., Ofori-Acquah, S. F., Nnodu, O., Wonkam, A., & Ohene-Frempong, K. (2013). Sickle cell disease: New opportunities and challenges in Africa. *The Scientific World Journal*, *2013*. https://doi.org/10.1155/2013/193252

Makani, J., Williams, T. N., & Marsh, K. (2017). Sickle cell disease in Africa : burden and research priorities Sickle Haemoglobin and Sickle Cell Disease. *Ann Trop Med Parasitol*, *101*(1), 3–14. https://doi.org/10.1179/136485907X154638.Sickle

Miller, F. A., Paynter, M., Hayeems, R. Z., Little, J., Carroll, J. C., Wilson, B. J., Allanson, J., Bytautas, J. P., & Chakraborty, P. (2010). Understanding sickle cell carrier status identified through newborn screening: A qualitative study. *European Journal of Human Genetics*, *18*(3), 303–308. https://doi.org/10.1038/ejhg.2009.173

Ministry of Health, G. (2012). *National Policy for the Prevention and Control of Chronic Non-Communicable Diseases in Ghana* (Issue August).

Modell, B., & Darlison, M. (2008). Global epidemiology of haemoglobin disorders and derived service indicators. *Bulletin of the World Health Organization*, *86*(6), 480–487. https://doi.org/10.2471/BLT.06.036673

Muoghalu, C. O. (2016). The Burden of Sickle Cell Disease to Parents of Sufferers in Nigeria. *International Journal of Genetic Science (2016) 3(1) 1-5*. https://doi.org/10.15226/2377-4274/3/1/00113

Niekerk, V. K. (2015). Knowledge and experiences of parents with children affected by Sickle Cell Disease in Cape Town [University of Cape Town]. In *Journal of Chemical Information and Modeling* (Vol. 53, Issue July). http://library.wur.nl/WebQuery/wurpubs/fulltext/353506

Ohene-Frempong, Bonney, A., Tetteh, H., & Nkrumah, F. K. (2005). Newborn screening for sickle cell disease in ghana: 270. *Pediatric Research*, *58*(2), 401. https://doi.org/10.1203/00006450-200508000-00299

Ohene-Frempong, K., Oduro, J., Tetteh, H., & Nkrumah, F. (2008). SCREENING NEWBORNS FOR SICKLE CELL DISEASE IN GHANA: TABLE 1. *Pediatrics*, *121*(Supplement 2). https://doi.org/10.1542/peds.2007-2022uuu

Pule, G., & Wonkam, A. (2014). Treatment for sickle cell disease in Africa: Should we invest in haematopoietic stem cell transplantation? *Pan African Medical Journal*, *18*, 1–5. https://doi.org/10.11604/pamj.2014.18.46.3923

Quinn, C. T., Rogers, Z. R., McCavit, T. L., & Buchanan, G. R. (2010). Improved survival of children and adolescents with sickle cell disease. *Blood*, *115*(17), 3447–3452. https://doi.org/10.1182/blood-2009-07-233700

Runkel, B., Klüppelholz, B., Rummer, A., Sieben, W., Lampert, U., Bollig, C., Markes, M., Paschen, U., & Angelescu, K. (2020). Screening for sickle cell disease in newborns: a systematic review. *Systematic Reviews*, *9*(1), 1–9. https://doi.org/10.1186/s13643-020-01504-5

Russo, G., De Franceschi, L., Colombatti, R., Rigano, P., Perrotta, S., Voi, V., Palazzi, G., Fidone, C., Quota, A., Graziadei, G., Pietrangelo, A., Pinto, V., Ruffo, G. B., Sorrentino,

F., Venturelli, D., Casale, M., Ferrara, F., Sainati, L., Cappellini, M. D., … Forni, G. L. (2019). Current challenges in the management of patients with sickle cell disease - A report of the Italian experience. *Orphanet Journal of Rare Diseases*, *14*(1), 0–9. https://doi.org/10.1186/s13023-019-1099-0

Segbefia, C., Goka, B., Welbeck, J., Amegan-Aho, K., Dwuma-Badu, D., Rao, S., Salifu, N., Oppong, S. A., Odei, E., Ohene-Frempong, K., & Odame, I. (2021). Implementing newborn screening for sickle cell disease in Korle Bu Teaching Hospital, Accra: Results and lessons learned. *Pediatric Blood and Cancer*, *68*(7), 1–7. https://doi.org/10.1002/pbc.29068

Segbefia, C., Goka, B., Welbeck, J., & Oppong, S. (2019). Implementing newborn screening for sickle cell disease in Accra, Ghana: Results and challenges. *Pediatric Blood and Cancer*, *Supplement 2*, S266. http://ovidsp.ovid.com/ovidweb.cgi?T=JS&PAGE=reference&D=emexa&NEWS=N&AN=627305253

Streetly, A., Sisodia, R., Dick, M., Latinovic, R., Hounsell, K., & Dormandy, E. (2018). Evaluation of newborn sickle cell screening programme in England: 2010-2016. *Archives of Disease in Childhood*, *103*(7), 648–653. https://doi.org/10.1136/archdischild-2017-313213

Therrell, B. L., Lloyd-Puryear, M. A., Ohene-Frempong, K., Ware, R. E., Padilla, C. D., Ambrose, E. E., Barkat, A., Ghazal, H., Kiyaga, C., Mvalo, T., Nnodu, O., Ouldim, K., Rahimy, M. C., Santos, B., Tshilolo, L., Yusuf, C., Zarbalian, G., & Watson, M. S. (2020). Empowering newborn screening programs in African countries through establishment of an international collaborative effort. *Journal of Community Genetics*, *11*(3), 253–268. https://doi.org/10.1007/s12687-020-00463-7

Ulph, F., Cullinan, T., Qureshi, N., & Kai, J. (2015). Parents' responses to receiving sickle

cell or cystic fibrosis carrier results for their child following newborn screening. *European Journal of Human Genetics*, *23*(4), 459–465. https://doi.org/10.1038/ejhg.2014.126

Ulph, F., Lavender, D. T., & Bennett, R. (2017). Consent for newborn screening and storage of blood samples. *British Journal of Midwifery*, *25*(11), 730–732. https://doi.org/10.12968/bjom.2017.25.11.730

Wastnedge, E., Patel, S., Goh, M. Y., & Rudan, I. (2018). The global burden of sickle cell disease in children under five years of age : a systematic review and meta-analysis. *Journal of Global Health*, *8*(2), 1–9. https://doi.org/10.7189/jogh.08.021103

Yahaya, L., Durosaro, I., & Nuhu, M. (2013). Challenges and Counselling Needs of Sickle Cell Patients Attending University of Ilorin Teaching Hospital. *Journal of Science and Sustainable Development*, *5*(1), 57–63. https://doi.org/10.4314/jssd.v5i1.5

APPENDICES

Appendix 1: Background/Socio-Demographic Data Questionnaire

Code …….

1. Gender ……
2. Age ……..
3. Marital status ……
4. Nationality …….
5. Religion ……
6. Place of residence ……
7. Educational level of mother…….
8. Educational level of father ……
9. Number of children (incl. child enrolled in clinic) ……
10. Phenotype of child (the child enrolled in clinic) ……
11. Phenotype of Mother……..
12. Phenotype of Father …….
13. Phenotype of other children (if any) ……..

    …….

    …….
14. Occupation of Mother …….
15. Occupation of Father ……..

Appendix 2: Interview Guide

a. Reaction to Disclosure of Initial Results

1. How old was the baby when you received the initial results?
2. How were the results communicated to you?
3. Were you alone when the results were disclosed?
4. Can you please share with me exactly how you felt when you were told about the results?

   Probe

   - Sad?
   - Anxious
   - Disbelief
   - Suspicious?
   - Surprised?
   - Not surprised? i.e expecting the results?
   - Other emotions not mentioned? Please share with me.
5. Can you share with me how you felt when you received the final confirmatory tests?
6. Before this, did you know about sickle cell disease? If yes, what did you know about it?

b. Responses to in-clinic counselling and support

7. Can you let me know all information you have received about the disease?
8. Do you know about your family history of the disease?
9. What information have you received regarding danger signs?
10. What information have you received about managing the disease?

11. What information have you received about how to avoid complications of the disease?
12. What other support have you received from the hospital or clinic?

c. Attitude towards follow-up clinic

13. Tell me about your clinic attendance since the diagnosis
14. Factors that determine whether you miss an appointment or not
15. What medication is your child on?
16. Have any of your other children been diagnosed with SCD?
17. Has your child been hospitalized since the initial diagnosis?
18. Sharing of diagnosis with spouse and other family members of friends
19. What has the relationship between you and your spouse /baby' father been since the diagnosis?
20. What support have you received from friends and family?
21. Is there anything else you would like to share?

Thank you.

Appendix 3: Informed Consent Form

Study Title

Sickle Cell Disease Screening: Perceptions of Parents with Positive Newborns at Korle Bu Teaching Hospital

Researcher

Irene Kanyoke, School of Public Health, University of Ghana
P.O. Box LG 43, Legon, Accra

General Information about the Research

My name is Irene Kanyoke, a postgraduate student at the University of Ghana and I am conducting a study titled ***Sickle Cell Disease Screening: Perceptions of parents of Positive Newborns at Korle Bu Teaching Hospital.***

I would like to have a conversation with you in either English, Twi or Ga regarding your experiences as parents of a newborn who tested positive for sickle cell disease. The researcher or assistant will interview you and he/she may be accompanied by another member. This interview will take between 45 minutes to an hour. This conversation will be recorded with an audio device and we will also be taking notes. It will be individual one-on-one interviews and as such, we would like to get your consent to participate in this study. This is part of the informed consent process.

Purpose of the study

This study aims to explore the experiences of parents of positive newborns at the clinic. This study aims to explore your reaction to receiving the initial positive results, how you responded to the counselling you have received so far at the clinic as well as what informs your decision to come for follow clinic. This study will help give insight into the current clinic practices and an understanding of the experiences of parents. The findings from this will be used to develop interventions to help improve current care practices at the clinic. As such, you will be asked questions about your experiences both at the clinic and in other areas of your life.

Risks/ Discomforts

Although there are no known risks or discomfort associated with this study, there will however be some sensitive questions and as such, you may feel some discomfort answering these

questions. The study team has taken all precautions to reduce discomfort and minimize any unknown risks.

Possible Benefits

There are no direct benefits from participating in this study. However, information from this study will be useful in developing interventions to help improve current care practices and provision at the clinic. There will be no financial cost to you because of you participating in this study apart from the time spent.

Confidentiality

All information obtained from you in this study will remain confidential and will be protected by us to the best of our ability. All information will be kept private and only the research team will have access to your information. Neither your name nor the name of your child (ren) will be used in documents or publications associated with this study. All other information such as demographic information will be anonymized by using codes instead of real names and quotes by you will NOT be cited with the code given and not by your real name. All recordings, memos and diaries will be kept under lock and key and only accessible by only the research team. All interviews will be kept for 5 years and then discarded.

Compensation

No compensation is provided for participation in this study. However, your time is appreciated as it will help provide an understanding of the experiences of parents of positive newborns in the clinic.

Voluntary Participation and Right to Leave the Research

Your participation in this study is completely voluntary. You may decide whether to participate in this interview or not. In case you decide to participate, you can always change your mind and withdraw from this study at any point in time during the interview. If you decide not to participate or withdraw at any point during the interview, your child will still be able to get the health care services he/she usually gets at the facility.

Conditions for Additional Information

In case you have any questions, concerns or comments, you can contact the Principal Investigator at: Irene Kanyoke, Korle Bu Teaching Hospital, Department of Child Health,

Mother's Hostel, Room 119. Telephone: +233 20 198 25 63 / + 233 54 822 13 35. Email: irenekanyoke@yahoo.com

Your Rights as a Participant

This research has been reviewed and approved by the Institutional Review Board of Korle Bu Teaching Hospital for Medical Research (KBTH- IRB). If you have any questions about your rights as a research participant, you can contact the IRB office between 8 am – 5 pm through the landline 0302666766 or email address rdo@kbth.gov.gh.

VOLUNTEER AGREEMENT

The above document describing the benefits, risks and procedures for the research title "*Sickle Cell Disease: Experiences of parents of Positive Newborns at Korle Bu Teaching Hospital"* has been read and explained to me. I have been allowed to have any questions about the research answers to my satisfaction. I agree to participate as a volunteer.

_____ _____

Name and signature/thumbprint of volunteer **Date**

If volunteers cannot read the form themselves, a witness must sign here:

I was present while the benefits, risks and procedures were read to volunteer. All questions were answered and the volunteer has agreed to take part in the research.

_____ _____

Name and signature/thumbprint of witness **Date**

I certify that the nature and purpose, the potential benefits and the possible risks associated with participating in this research have been explained to the above individual.

_____ _____

Name and signature/thumbprint of person obtaining consent **Date**

Appendix 4: Codebook

Research Question/objective **(open coding)**	**Themes** **(quotes from interviews)**	**List of Codes**	**Emerged Themes**	**Attitude** **(from Theoretical framework)**	**Construct** **(from SEM)**
Reactions to the disclosure of positive results	*"You know, life is hard now, and I was scared that if it happens that she is a sickler, what would I do"?* *"Because that first time she told me, I got scared. That's why I didn't believe it. I was worried".* *"So him ((pointing to child with SCD at clinic)), when his results came, I wasn't surprised because I knew the dad is SC. so I wasn't surprised at all".* *'he still doesn't believe. And I tell him that, if he didn't have it, the nurses wouldn't have told me he has it".* *"It did help. Because, even before we got married, I was contemplating. And then when the news came, even though it came as a shock"*	1.Scared 2.Worried 3. Not Surprised 4.Disbelief 5.Surprised/shocked	1. emotional response after disclosure	Influences	Intrapersonal (individual)
Reactions to the disclosure of positive results	*"well, I didn't want anyone to know. I just wanted it to be between me and my husband so I didn't tell anyone"* *"nobody oo. I've not told anybody. Because of the stigma, I don't want to, to tell anybody"* *"Yes. I told the caretaker. Because if I have to leave, and*	1. Shared results with partner only 2. fear of social exclusion stigma 2. shared results with family and friends	2. Decision to share results	Influences	Intrapersonal

	you don't give the medication, I will blame her. And then recently, I told my mother-in-law... and then my parents, my siblings are also aware".				
In-clinic counselling and comprehensive follow-up care	*"How to how to, ehh, is it, prevent it, ehehn, in terms of not to take something cold. And all those kinds of things. You will know the way when it's cold, you should put something, like cardigan, all those kinds of things. And you have to take your medication always, no crises, for the 0 months to 5 years. You won't have any problem".* *"When they are babies, maybe you will see one of their hands swelling or their feet swollen. Or their eyes. and when they get temperature, you have to bring them to the hospital and not stay in the house. It might be infection".*	1. Preventive measures/care at home (nutrition, hydration, warm clothes, 2. awareness of treatment options 3. awareness of possible complications 4. danger signs	3. Care and Management of Newborns with SCD	Network (with clinic staff)	Interpersonal
In-clinic education and comprehensive follow-up care	*"Yes. She said like, it was from both my husband and I. and that I have S in my blood and my husband also has S in his blood. And that's how come the child has the S".* *"They took me through the sickling. How it forms. How their blood cells look like. Is it the white one or the, the red one? Yea, that looks like a bofloat, and theirs is like an arch, the moon, half moon, which is very hard not like the others that are soft that can pass through all the blood. Theirs because is hard, sometimes, it gets broken and chokes the veins that you have to give them the (massage) and all that. so, I was taught all that"*	1. Inheritance of SCD 2. blood disorder 3. family history of disease	4. Genetics of sickle cell disease	Network with clinic staff	Interpersonal

Adherence to Follow-up procedures	*"I've been coming to clinic for the past 7 to 8 months. Every month. I'm here. Yes, sometimes even 2 weeks. In a month, I can come like 2 times".* *"So, when they tell me to come even every 2 weeks, I am ready to come more than 2 weeks. I'm ready to come check if the blood level is normal before I can be happy".* *"Madam please, you see, for my son, I love him. And it's the doctor who knows, so whatever they tell me I have to listen and do it. But sometimes, when I'm unable to come on the date, it's usually because of financial difficulties. I may be lacking money for the labs .... But financially, it's been difficult. Because of money issues. I was supposed to bring him last month but because I didn't have money, I couldn't come".* *"And so because of that, I don't play with the medication. Once it finishes, I make sure I refill it"* *"oh it's usually the lab we frequently do"*	1. Adherence to routine clinic visits (due to expectation of healthy outcome for child, and due to parents' personal experience with disease) 2. Non-adherence to clinic visits (due to COVID financial difficulty) 3. Adherence to daily medication 4. routine lab tests	5. Adherence to routine clinic	Influences	Intrapersonal

Appendix 5

And the date of this
Letter should be quoted
My Ref. No. KBTH/MS/ES/21
Your Ref. No......

KORLE BU, ACCRA.

Tel: +233 302 667759/673034-(
Fax: +233 302 667759
Email: Info@kbth.gov.gh
pr@kbth.gov.gh
Website: www.kbth.gov.gh

7th July, 2021

IRENE KANYOKE
SCHOOL OF PUBLIC HEALTH
UNIVERSITY OF GHANA
LEGON

SCIENTIFIC AND TECHNICAL COMMITTEE APPROVAL
PROTOCOL IDENTIFICATION NUMBER: KBTH-STC 00094/2021

The Korle Bu Teaching Hospital Scientific and Technical Committee (KBTH-STC), on 7th July, 2021 approved your submitted study protocol.

TITLE OF PROTOCOL: "Sickle Cell Disease Screening: Experiences of Parents with Positive Newborns at Korle-Bu Teaching Hospital, Ghana"

PRINCIPAL INVESTIGATOR: Irene Kanyoke

This approval requires that you **forward your approved document to Korle Bu Teaching Hospital –Institutional Review Board (KBTH-IRB) for the ethical aspect of the proposal to be assessed before the project can be initiated.**

This STC approval is valid till 30th December, 2021

You may, however, request extension of the approval period, or renewal as the case may be, should the study extend beyond the stated period.

Upon completion, you are required to submit a final report on the study to the STC. This is to enable the STC ensure among others that, the project has been implemented as per the approved protocol. You are also required to inform the KBTH-STC and Research Directorate of any publications that may emanate from the research findings.

Kindly note that, should the need arise, the KBTH-STC or IRB may institute appropriate measures to satisfy itself that study is being conducted according to the highest scientific and ethical standards.

Please note that any modification to the study protocol without Scientific Technical Committee (STC) approval renders this approval invalid.

Sincere regards,

Prof. G. Obeng Adjei
Chairman, KBTH-STC

Cc: The Chairman, KBTH-IRB

Appendix 6

In case of reply the number
And the date of this
Letter should be quoted
My Ref. No. KBTH/MD/23/21
Your Ref. No.....

KORLE BU TEACHING HOSPITAL
P. O. BOX KB 77,
KORLE BU, ACCRA.

Tel: +233 302 667759/673034-6
Fax: +233 302 667759
Email: Info@kbth.gov.gh
pr@kbth.gov.gh
Website: www.kbth.gov.gh

16th September, 2021

IRENE KANYOKE
SCHOOL OF PUBLIC HEALTH
COLLEGES OF HEALTH SCIENCES
UNIVERSITY OF GHANA, LEGON

"SICKLE CELL DISEASE SCREENING: EXPERIENCES OF PARENTS WITH POSITIVE NEWBORNS AT KORLE BU TEACHING HOSPITAL"

KBTH-IRB /00094/2021

INVESTIGATOR: Irene Kanyoke

The Korle Bu Teaching Hospital Institutional Review Board (KBTH IRB) reviewed and granted approval to the study entitled "Sickle Cell Disease Screening: Experiences of Parents with positive Newborns at Korle Bu Teaching Hospital"

Please note that the Board requires you to submit a final review report on completion of this study to the KBTH-IRB.

Kindly, note that, any modification/amendment to the approved study protocol without approval from KBTH-IRB renders this certificate invalid.

Please report all serious adverse events related to this study to KBTH-IRB within seven days verbally and fourteen days in writing.

This IRB approval is valid till 30th August, 2022. You are to submit annual report for continuing review.

Sincere regards,

DR. DANIEL ANKRAH
VICE CHAIR (KBTH-IRB)
FOR: CHAIR (KBTH-IRB

Cc: The Chief Executive Officer, KBTH
The Director of Medical Affairs, KBTH

Appendix 7

In case of reply the number
And the date of this
Letter should be quoted
My Ref. No. KBTH/MA/C/312
Your Ref. No......

KORLE BU TEACHING HOSPITAL
P. O. BOX KB 77,
KORLE BU, ACCRA.

Tel: +233 302 667759/673034-6
Fax: +233 302 667759
Email: Info@kbth.gov.gh
pr@kbth.gov.gh
Website: www.kbth.gov.gh

23rd September, 2021

IRENE KANYOKE
SCHOOL OF PUBLIC HEALTH
COLLEGES OF HEALTH SCIENCES
UNIVERSITY OF GHANA
LEGON

INSTITUTIONAL APPROVAL: KORLE BU TEACHING HOSPITAL-SCIENTIFIC AND TECHNICAL COMMITTEE/INSTITUTIONAL REVIEW BOARD (KBTH-STC/IRB/00094/2021

Following approval of your study entitled "Sickle Cell Disease screening: Experiences of parents with positive Newborns at Korle Bu Teaching Hospital" by the Korle Bu Teaching Hospital-Scientific and Technical Committee/Institutional Review Board.

I am pleased to inform you that institutional approval has been granted for the conduct of your study in Korle Bu Teaching Hospital.

Please contact the Head of Department to discuss the commencement date of the study.

Please note that, this institutional approval is rendered invalid if the terms of the Institutional Reviewed Board/Scientific and Technical Committee approval are violated.

Sincere regards,

Dr. Harry Akoto
Ag. Director of Medical Affairs
For: Chief Executive

Appendix 8

MEDICAL DIRECTORATE
KORLE BU TEACHING HOSPITAL

23rd September, 2021

THE HEAD
SICKLE CELL UNIT
KORLE BU

LETTER OF INTRODUCTION – IRENE KANYOKE
"SICKLE CELL DISEASE SCREENING: EXPERIENCES OF PARENTS WITH POSITIVE NEWBORNS AT KORLE BU TEACHING HOSPITAL"

I have the pleasure to introduce to you the above named Investigator from School of Public Health, University of Ghana, Legon. Irene Kanyoke sought and has been granted approval to conduct a study entitled: "Sickle Cell disease screening: Experiences of parents with positive newborns at Korle Bu Teaching Hospital" in your Department.

She is to contact you to discuss the commencement date of the study.

Please verify her identity with a Government issued National ID card and accord her the needed assistance.

Attached is the Scientific and Technical Committee and Institutional Review Board approval, which specifies the terms.

Sincere regards,

Dr. Harry Akoto
Ag. Director of Medical Affairs
For: Chief Executive

Cc: The Head
Dept. of Child Health
Paediatric Sickle Cell Clinic
Korle Bu