ECONOMIC BURDEN OF SICKLE CELL DISEASE AND PAYMENT COPING STRATEGIES AMONG PATIENT IN THE UNIVERSITY OF BENIN TEACHING HOSPITAL

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BENIN CITY
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OCTOBER,, 2025

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IN PARTIAL FULFILLMENT OF THE REQUIREMENTS FOR THE AWARD OF BACHELOR OF NURSING SCIENCE (BNSC.)
FACULTY OF NURSING SCIENCCES
UNIVERSITY OF BENIN,
BENIN CITY,
EDO STATE

OCTOBER ,2025

DECLARATION

This is to declare that this research project titled "**ECONOMIC BURDEN OF SICKLE CELL DISEASE AND PAYMENT COPING STRATEGIES AMONG PATIENT IN THE UNIVERSITY OF BENIN TEACHING HOSPITAL**" will be carried out by **NJOKU OZIOMA GOSPEL.** It will solely be the result of my work except where acknowledged as being derived from other person (s) or resources.

MATRICULATION NUMBER : BMS2001033

FACULTY OF NURSING SCIENCE,

UNIVERSITY OF BENIN,BENIN CITY,EDO STATE

Signature: …….

Date: …….

CERTIFICATION/APPROVAL

This is to certify that this project titled "**ECONOMIC BURDEN OF SICKLE CELL DISEASE AND PAYMENT COPING STRATEGIES AMONG PATIENT IN THE UNIVERSITY OF BENIN TEACHING HOSPITAL**" will be carried out by **NJOKU OZIOMA GOSPEL** with **Mat No. BMS2001033** in the Department of Nursing Sciences, university of Benin, under the supervision of **DR. (MRS) C. ENUKU**

NJOKU OZIOMA GOSPEL _____
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Supervisor Sign & date

PROF.(MRS)C.E OMOROGBE _____
Head of Department Sign & date

_____
External Examiner Sign & date

DEDICATION

This work is dedicated to GOD ALMIGHTY who is providing me with the strength to complete my academic journey.

To my beloved Parents Mr &Mrs Stephen Njoku whose unwavering support both financially and morally has kept me outstanding and fostering success all through my academic work

To my elder brother Mr Chinaza Gift Njoku I want to say may the good Lord continue to bless and prosper you.

To my elder sisters (Miss Mercy Ogadinma and Miss Victoria Omerema Njoku) and my younger brother Master onyekachi Njoku thank you guys so much for all you do for me

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I would like to begin by giving all the glory to the Almighty God, the sovereign owner of my life. I am eternally grateful for His guidance, protection, and unfailing provision throughout my life and academic journey.

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Special thanks to my supportive and hardworking lecturers, Rev Sr. J. N. Chukwurah, Mrs M.A Iniomor , Mrs C. C. Edo-Osagie, Dr T. A. Ehwarieme, Mrs. E. N. Oyana, Mrs. R. Lawal, Mrs Ikhuobase, Mrs F. Esebanme and Mr Aragua and Mrs Osadolor as well as all other lecturers and non- academic staff for their immense contribution, dedication and support.

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Abstract

This study examined the economic burden of Sickle Cell Disease (SCD) and the payment coping mechanisms of patients receiving care at the University of Benin Teaching Hospital (UBTH), Benin City, Nigeria. A descriptive cross-sectional survey design was employed, targeting sickle cell patients who attended the outpatient clinic and wards of UBTH. A sample size of 100 respondents was selected using simple random sampling from an estimated population of 118 patients. Data were collected using a structured, pretested questionnaire covering socio-demographic characteristics, direct and indirect medical costs, socioeconomic cost distribution, and payment coping strategies. Validity of the instrument was ensured through expert review, and reliability was established via a test-retest method yielding a reliability coefficient of 0.85. Descriptive statistics such as frequency, percentages, means, and standard deviation were used to analyze the data. Direct costs were calculated using the bottom-up approach, while indirect costs were assessed through the human capital approach. Inferential analysis was done using ANOVA and Chi-square tests to determine significant associations, with a significance level set at $p < 0.05$. Ethical approval was obtained, and patient confidentiality and voluntary participation were assured. The findings highlight the substantial economic burden SCD imposes on patients and the diverse strategies employed to cope with payment demands, providing a basis for policy recommendations aimed at improving access to care and financial protection for individuals living with SCD.

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CHAPTER ONE
INTRODUCTION

1.0 Background of Study

Sickle Cell Disease (SCD) is a major public health challenge, particularly in sub-Saharan Africa, where it remains the most common inherited blood disorder (Adigwe *et al*., 2023). The disease results from the inheritance of abnormal hemoglobin genes from both parents, leading to the production of defective hemoglobin S, which causes red blood cells to become rigid and sickle-shaped. These abnormal cells obstruct blood flow, leading to painful vaso-occlusive crises, anemia, increased susceptibility to infections, and long-term organ damage. Globally, an estimated 250,000 children are born annually with SCD, with Africa accounting for approximately 85% of the disease burden (Yousif *et al*., 2024). Nigeria has the highest prevalence, with around 150,000 children born with the disease each year, making it a significant public health concern (Jabr, 2021).

The burden of SCD extends beyond its clinical complications to its profound economic impact on individuals, families, and healthcare systems. Due to the chronic nature of the disease, patients require lifelong medical care, including frequent hospital visits, medications such as hydroxyurea, blood transfusions, and laboratory tests. These costs pose a substantial financial burden on households, particularly in low-income settings where access to healthcare is limited. In Nigeria, out-of-pocket spending (OOPS) remains the dominant healthcare financing mechanism, accounting for 70–95% of total health expenditures (Junaidu, 2022). This heavy reliance on OOPS exposes SCD patients and their families to catastrophic health expenditures, where medical costs exceed 10% of total household income or 40% of non-food income, often forcing families to sacrifice essential needs such as food, education, and housing.

The economic burden of SCD is further compounded by indirect costs such as loss of productivity and income due to the time spent seeking medical care, frequent hospitalizations, and reduced work capacity for both patients and caregivers (Okeke *et al*., 2024). Many caregivers, particularly mothers, face employment disruptions due to the need for continuous care for their children (Biddell *et al*., 2022).

The cumulative effect of these financial challenges leads to economic instability, pushing many affected families into deeper poverty. Despite the significant economic implications, there is limited research on the financial burden of SCD in Nigeria, especially concerning payment coping mechanisms. Understanding how households manage these costs—whether through savings, borrowing, selling assets, or seeking assistance from charitable organizations—can provide critical insights into healthcare financing gaps and inform policy interventions (Okeke *et al*., 2024).

At the institutional level, tertiary hospitals like the University of Benin Teaching Hospital (UBTH) play a crucial role in providing specialized care for SCD patients. However, the high cost of treatment and limited financial support structures often make it difficult for patients to access consistent care (Nero & Bozzo, 2022). While some developed countries have implemented health insurance schemes and social safety nets to alleviate the economic burden of chronic diseases, Nigeria's healthcare system still lacks an effective prepayment mechanism for SCD patients. The absence of comprehensive health insurance coverage further exacerbates financial hardships, necessitating urgent policy action to improve healthcare affordability (Amarachukwu *et al*., 2022).

Given these challenges, this study aims to assess the economic burden of SCD and the payment coping mechanisms among patients attending UBTH. By evaluating the financial impact of the

the study will generate evidence-based recommendations to inform healthcare policies, improve financial risk protection, and enhance access to quality care for SCD patients. Understanding these economic challenges is essential for developing sustainable healthcare financing models, reducing financial hardship, and improving health outcomes for individuals living with SCD in Nigeria.

1.2 Statement of the Problem

Sickle Cell Disease (SCD) poses a significant economic burden on individuals, families, and healthcare systems, particularly in low-resource settings like Nigeria (Okeke et al., 2024). As a chronic condition requiring lifelong management, the financial strain associated with frequent hospital visits, medications, blood transfusions, and emergency care is overwhelming for many households (Amarachukwu *et al*., 2022). The University of Benin Teaching Hospital (UBTH), a major referral center, provides specialized care for SCD patients, but the cost of treatment remains a significant barrier for many families, particularly those reliant on out-of-pocket spending (OOPS), which dominates healthcare financing in Nigeria (Junaidu, 2022)

Despite the high prevalence of SCD in Nigeria, there is limited empirical data quantifying the direct and indirect costs borne by affected households (Amarachukwu *et al*., 2022). Many families experience catastrophic health expenditures (CHE), where medical costs exceed a significant portion of their total income, often leading to debt, asset depletion, and reduced spending on essential needs like food, education, and housing. Furthermore, patients and caregivers frequently adopt various payment coping mechanisms, including borrowing, selling assets, or relying on informal support networks, but the effectiveness and sustainability of these strategies remain largely undocumented (Amarachukwu *et al*., 2022).

Without a clear understanding of the economic impact and coping strategies used by SCD patients at UBTH, policymakers and healthcare providers lack the necessary evidence to design effective financial risk protection policies. This study seeks to fill this gap by assessing the economic burden of SCD at UBTH, identifying coping mechanisms, and evaluating the extent to which financial hardship affects access to care. Findings from this research will provide valuable insights for improving healthcare financing strategies and ensuring that SCD patients receive adequate and affordable care without facing financial ruin.

1.3 Aims of the Study

This study seeks to determine the economic burden incurred and assess payment coping mechanisms used by Sickle cell patients attending University of Benin Teaching Hospital (UBTH).

Specify Objectives of the Study

- Assess the direct medical cost of Sickle cell disease incurred by patients and their households in University of Benin Teaching Hospital (UBTH).
- Assess the indirect medical cost of Sickle cell disease incurred by patients and their households in University of Benin Teaching Hospital (UBTH)
- Estimate the cost distribution among different socio-economic groups.
- Identify the payment coping mechanism utilized by different socio-economic groups.

1.4 Research Questions

- What is the direct medical costs incurred by Sickle cell patients and their households in University of Benin Teaching Hospital (UBTH)?

- What is the indirect medical cost of Sickle cell disease incurred by patients and their households in University of Benin Teaching Hospital (UBTH)?
- What is the cost distribution among different socio-economic groups?
- What is the payment coping mechanism utilized by different socio-economic groups?

1.5 Hypotheses

Null Hypothesis (H_0):

There is no significant relationship between the economic burden of sickle cell disease (SCD) and the payment coping mechanisms used by patients at the University of Benin Teaching Hospital (UBTH).

Alternative Hypothesis (H_1):

There is a significant relationship between the economic burden of sickle cell disease (SCD) and the payment coping mechanisms used by patients at the University of Benin Teaching Hospital (UBTH).

H_0: Socio-demographic factors (such as educational status, and employment status) do not significantly influence the financial burden experienced by SCD patients at UBTH.

H_1: Socio-demographic factors significantly influence the financial burden experienced by SCD patients at UBTH.

H_0: The out-of-pocket health expenditure for SCD treatment does not significantly contribute to catastrophic health expenditure (CHE) among patients at UBTH.

H_1: The out-of-pocket health expenditure for SCD treatment significantly contributes to catastrophic health expenditure (CHE) among patients at UBTH.

H_0: There is no significant difference in the choice of payment coping mechanisms among SCD patients based on their socio-economic status.

H_1: There is a significant difference in the choice of payment coping mechanisms among SCD patients based on their socio-economic status.

1.6 Significance of the Study

This study examines the economic burden of Sickle Cell Disease (SCD) and the coping mechanisms used by patients at UBTH. Its findings will aid policymakers in developing sustainable healthcare financing models, such as subsidies and insurance schemes, to reduce financial hardship. It will also highlight the impact of out-of-pocket spending and catastrophic health expenditures, contributing to policies that enhance financial risk protection for SCD patients.

Healthcare providers at UBTH can use the insights to develop cost-effective treatment strategies, improve hospital-based payment plans, and explore alternative funding sources. Understanding these financial challenges will help ensure better access to quality care and reduce healthcare inequities.

For patients and caregivers, the study offers insights into financial coping mechanisms and available support options, helping them make informed healthcare decisions. Additionally, it serves as a reference for future research and provides NGOs and donor agencies with data to advocate for financial aid programs and funding initiatives.

Ultimately, this research aims to support better healthcare financing policies, ease the financial burden on SCD patients, and improve access to affordable care at UBTH and beyond.

1.7 Scope of the Study

The study was delimited to all persons who had been diagnosed of Sickle cell disease and have been receiving treatment from University of Benin Teaching Hospital (UBTH) within the past one year. Both males and females within all ages. Out-patients and in-patients were studied. Medical-Surgical units, specialist clinics, and family medicine were used. Insignificant cost of Sickle cell disease was not included in this study.

1.8 Operational Definition of Terms

Economic Burden: It refers to both medical and non-medical costs incurred by Sickle cell patients in the management of their ailment. It is classified into direct and indirect costs.

Direct cost (financial cost): This has to do with cost related to investigations, diagnoses, treatment, admissions, follow up costs and travel cost.

Indirect cost: They are those things that will be forgone for the sake of this illness e.g. time

spent travelling, waiting-time in hospital, time spent out-of-work, time accompanying relative, time lost through premature death or premature retirement.

Payment Coping Mechanism: It refers to the use of one's income (salary and savings), someone else paying, money borrowed/loans, community-based support, sale of household assets, gifts, appeal for support/ begging, temporary stoppage of children's education, cutting down on minimum consumption expenses to pay for treatment and tests.

Sickle cell patient: This refers to someone who has been diagnosed by a physician as having

Sickle cell disease of any type.

Sickle cell disease family/significant others: This includes, parents, brothers, sisters, surrogate or friend who accompanies patient for treatment and have lived consistently with this experience for at least one year.

Different socio-economic population group; this refers to the categorization of study patients into various levels or classes. This will be determined using asset ownership like Radio, Television, bicycle, air conditioner, electric fan, Motorcycle, Fridge, kerosene stove, generator, gas cooker and car on a socio-economic status index, type of food and living accommodation.

Socio-economic population is also categorized into poorest, poorer and least poor.

CHAPTER TWO
LITERATURE REVIEW

This chapter presents review of literatures relevant to this work on economic burden and payment coping mechanism of sickle cell patients. The literature was reviewed under Conceptual, Theoretical and Empirical reviews.

2.1 Concept of sickle diseases

Sickle cell disease (SCD), also simply called sickle cell, is a group of hemoglobin-related blood disorders that are typically inherited. The most common type is known as sickle cell anemia (NHLBI, 2024).

Sickle cell anemia results in an abnormality in the oxygen-carrying protein haemoglobin found in red blood cells. (NHLBI, 2024). This leads to the red blood cells adopting an abnormal sickle-like shape under certain circumstances; with this shape, they are unable to deform as they pass through capillaries, causing blockages. (National Heart, Lung, and Blood Institute, 2024)

Problems in sickle cell disease typically begin around 5 to 6 months of age. A number of health problems may develop, such as attacks of pain (known as a sickle cell crisis) in joints, anemia, swelling in the hands and feet, bacterial infections, dizziness and stroke (NHLBI, 2022)

The probability of severe symptoms, including long-term pain, increases with age. NHLBI, 2022) Without treatment, people with SCD rarely reach adulthood but with good healthcare, median life expectancy is between 58 and 66 years. All of the major organs are affected by sickle cell disease. The liver, heart, kidneys, gallbladder, eyes, bones, and joints can be damaged from the abnormal functions of the sickle cells and their inability to effectively flow through the small blood vessels (NHLBI, 2022).

Sickle cell disease occurs when a person inherits two abnormal copies of the β-globin gene that makes haemoglobin, one from each parent. Several subtypes exist, depending on the exact mutation in each haemoglobin gene (NHLBI, 2024).

An attack can be set off by temperature changes, stress, dehydration, and high altitude. A person with a single abnormal copy does not usually have symptoms and is said to have sickle cell trait. (NHLBI 2024) Such people are also referred to as carriers. Diagnosis is by a blood test, and some countries test all babies at birth for the disease. Diagnosis is also possible during pregnancy (NHLBI, 2022)

The care of people with sickle cell disease may include infection prevention with vaccination and antibiotics, high fluid intake, folic acid supplementation, and pain medication. (NHLBI, 2024). Other measures may include blood transfusion and the medication hydroxycarbamide (hydroxyurea). (NHLBI, 2024). In 2023, new gene therapies were approved involving the genetic modification and replacement of blood forming stem cells in the bone marrow (Singh *et al*., 2024; Wilkinson, 2023).

As of 2021, SCD is estimated to affect about 7.7 million people worldwide, directly causing an estimated 34,000 annual deaths and a contributory factor to a further 376,000 deaths (Thomson, 2023). About 80% of sickle cell disease cases are believed to occur in Sub-Saharan Africa (Adigwe *et al*., 2023).

It also occurs to a lesser degree among people in parts of India, Southern Europe, West Asia, North Africa and among people of African origin (sub-Saharan) living in other parts of the world (Williams, 2024).

The condition was first described in the medical literature by American physician James B. Herrick in 1910, in 1949, its genetic transmission was determined by E. A. Beet and J. V. Neel. In 1954, it was established that carriers of the abnormal gene are some degrees protected against malaria (Sayed *et al*., 2022).

Signs of sickle cell disease usually begin in early childhood. The severity of symptoms can vary from person to person, as can the frequency of crisis events (Obeagu & Obeagu, 2024) Sickle cell disease may lead to various acute and chronic complications, several of which have a high mortality rate (Medline Plus, 2020).

2.1.1 Epidemiology

The HbS gene can be found in every ethnic group (Mccormick et al., 2020), the highest frequency of sickle cell disease is found in tropical regions, particularly sub-Saharan Africa, tribal regions of India, and the Middle East (Williams, 2024). About 80% of sickle cell disease cases are believed to occur in Sub-Saharan Africa (Adigwe *et al*., 2023).

Migration of substantial populations from these high-prevalence areas to low-prevalence countries in Europe has dramatically increased in recent decades and in some European countries, sickle cell disease has now overtaken more familiar genetic conditions such as haemophilia and cystic fibrosis. (Angastiniotis *et al*., 2021).

Sickle cell disease occurs more commonly among people whose ancestors lived in tropical and subtropical sub-Saharan regions where malaria is or was common. Where malaria is common, carrying a single sickle cell allele (trait) confers a heterozygote advantage; humans with one of the two alleles of sickle cell disease show less severe symptoms when infected with malaria (Williams, 2024)

This condition is inherited in an autosomal recessive pattern, which means both copies of the gene in each cell have mutations. The parents each carry one copy of the mutated gene, but they typically do not show signs and symptoms of the condition (Medline Plus, 2020).

Three-quarters of sickle cell cases occur in Africa. The burden of child mortality from sickle cell disease in Nigeria continues to be disproportionately higher than the burden of mortality of children without sickle cell disease (Nnodu *et al*., 2021).

2.1.2 Pathophysiology

Under conditions of low oxygen concentration, HBS polymerizes to form long strands within the red blood cell (RBC). These strands distort the shape of the cell and after a few seconds cause it to adopt an abnormal, inflexible sickle-like shape. This process reverses when oxygen concentration is raised and the cells resume their normal biconcave disc shape. If sickling takes place in the venous system, after blood has passed through the capillaries, it has no effect on the organs and the RBCs can unsickle when they become oxygenated in the lungs. Repeated switching between sickle and normal shapes damages the membrane of the RBC so that it eventually becomes permanently sickled (Maakaron, 2022).

Normal red blood cells are quite elastic and have a biconcave disc shape, which allows the cells to deform to pass through capillaries. In sickle cell disease, low oxygen tension promotes red blood cell sickling and repeated episodes of sickling damage the cell membrane and decrease the cell's elasticity. These cells fail to return to normal shape when normal oxygen tension is restored. As a consequence, these rigid blood cells are unable to deform as they pass through narrow capillaries, leading to vessel occlusion and ischaemia (Shah *et al*., 2019).

Cells which have become sickled are detected as they pass through the spleen and are destroyed. In young children with SCD, the accumulation of sickled cells in the spleen can result in splenic

sequestration crisis. In this, the spleen becomes engorged with blood, depriving the general circulation of blood cells and leading to severe anemia. The spleen initially becomes noticeably swollen but the lack of a healthy blood flow through the organ culminates in scarring of the spleen tissues and eventually death of the organ, generally before the age of 5 years (Connes *et al*., 2023)

The actual anaemia of the illness is caused by haemolysis, the destruction of the red cells, because of their shape. Although the bone marrow attempts to compensate by creating new red cells, it does not match the rate of destruction. Healthy red blood cells typically function for 90–120 days, but sickled cells only last 10–20 days (Maakaron, 2022).

The rapid breakdown of RBC's in SCD results in the release of free heme into the bloodstream exceeding the capacity of the body's protective mechanisms. Although heme is an essential component of hemoglobin, it is also a potent oxidative molecule. Free heme is also an alarmin - a signal of tissue damage or infection, which triggers defensive responses in the body and increases the risk of inflammation and vaso-occlusive events (Connes *et al*., 2023).

2.1.3 Phenotypes in sickle cell disease

There is great phenotypic variability among individuals with SCD. Some variability shows a specific geographical distribution and is associated with known or suspected genetic variants (Serjeant, 2022). However, some complications cluster together epidemiologically in subphenotypes, at times united by a common biomarker that suggests a mechanism, such as a particularly low Hb level with a high reticulocyte count or high serum lactate dehydrogenase level, implying more-intense haemolysis. These phenotypes are not mutually exclusive, exist often as a spectrum, can overlap, are probably due to independent genetic modifiers of the underlying mechanisms and might change with ageing.

2.1.3.1 Vaso-occlusive subphenotype.

This SCA subphenotype is characterized by a higher haematocrit than that observed in individuals with other SCA phenotypes; a higher haematocrit promotes higher blood viscosity. Individuals with this phenotype are predisposed to frequent vaso-occlusive pain crises, acute chest syndrome (that is, a vaso-occlusive crisis of the pulmonary vasculature) and osteonecrosis. Co-inheritance of α-thalassaemia reduces haemolysis (by reducing the intracellular concentration of HbS, which slows HbS polymerization and haemolysis) but promotes higher haematocrit (Serjeant, 2022).

2.1.3.2 Haemolysis and vasculopathy subphenotype

This phenotype is characterized by a lower haematocrit than that found in individuals with the vaso-occlusive subphenotype accompanied by higher levels of serum lactate dehydrogenase and bilirubin, which indicate more-severe haemolytic anaemia. Individuals in this group are at risk of ischaemic stroke, pulmonary hypertension, leg ulceration, gallstones, priapism and possibly nephropathy (Sundd *et al*., 2019). Decreased NO bioavailability, haem exposure and haem turnover are associated with these vasculopathic complications. The severe anaemia also promotes high cardiac output as a compensatory mechanism, and this excessive blood flow has been suggested to promote vasculopathy in the kidney and potentially in other organs.

2.1.3.3 High HbF subphenotype

Persistent expression of HbF in the range of 10–25% of total Hb owing to genetic variants generally reduces the clinical severity of SCA (Serjeant, 2022). However, not all individuals with the common, uneven cellular distribution of HbF (heterocellular distribution) have a mild phenotype. Expression levels of 25–50% of HbF in every erythrocyte (pancellular distribution) lead to nearly complete amelioration of SCA, with rare clinical symptoms and no anaemia

2.1.3.4 Pain subphenotypes

Individuals with pain-sensitive or pain-protective phenotypes experience pain differently, potentially owing to altered neurophysiology of pain sensation pathways. One example of a genetic modifier of pain is GCH1, which is associated with pain sensitivity in healthy individuals, and a variant of GCH1 is associated with frequency of severe pain in SCA (Gehling *et al*., 2023). Quantitative sensory testing of pain sensitivity is being used to functionally characterize these phenotypes in SCA (Smith *et al*., 2023).

2.1.4 Diagnosis/screening

The goals and methods of diagnosis of SCD vary with the age of the person. In general, there are four overlapping testing periods: preconception, prenatal, neonatal and post-neonatal. Preconception testing is designed to identify asymptomatic potential parents whose offspring would be at risk of SCD. Laboratory techniques used for preconception testing are routine basic methods of protein chemistry that enable separation of Hb species according to their protein structure, including Hb electrophoresis, high-performance liquid chromatography (HPLC) and isoelectric focusing (Bain *et al*., 2023).

2.1.4.1 Prenatal diagnosis

Prenatal diagnosis is a generally safe but invasive procedure and is offered during early pregnancy to couples who tested positive at preconception screening. It requires fetal DNA samples obtained from chorionic villus analysis performed at 9 weeks of gestation (Ampomah *et al*., 2022). Non-invasive prenatal diagnosis techniques are being developed but are still investigational. These new techniques can detect fetal DNA in maternal circulation by as early as 4 weeks of gestation. Some couples who test positive at preconception screening might opt for in vitro fertilization with pre-implantation genetic diagnosis, if available, to genetically identify at-risk embryos before embryo transfer occurs (Ampomah *et al*., 2022).

2.1.4.2 Newborn screening

Newborn screening for SCD is performed at birth before symptoms occur, using Hb protein analysis methodologies. Two types of newborn screening programmes have been used: selective screening of infants of high-risk parents (targeted screening) and universal screening. Universal screening is generally more cost-effective, identifies more newborn babies with disease and prevents more deaths (Okeke *et al*., 2024). In areas without newborn screening programmes, the initial diagnosis of SCD occurs at approximately 21 months of age (Vichinsky E;Hurst D;Earles A;Kleman K;Lubin B, 2022). For many individuals with SCD, the initial presentation is a fatal infection or acute splenic sequestration crisis (Vichinsky E;Hurst D;Earles A;Kleman K;Lubin B, 2022). Early diagnosis accompanied by penicillin prophylaxis and family education reduces the mortality in the first 5 years of life from 25% to <3% (Vichinsky E;Hurst D;Earles A;Kleman K;Lubin B, 2022).

2.1.4.3 Post-neonatal testing

The requirement of post-neonatal testing for SCD is influenced by several factors that affect the general population's knowledge of their SCD status. These factors include the regional success of neonatal screening programmes, immigration of at-risk patients not previously tested and access to neonatal results in older patients (Hassan *et al*., 2024). HbAS is a benign condition and not a disease, but it is also a risk factor for uncommon serious complications (Hassan *et al*., 2024). Thus, knowledge of one's own HbAS status is important in the prevention of rare serious complications and in family planning.

HbAS can also be detected by newborn screening programmes; however, HbAS detection is not the primary objective, and many programmes do not provide this information or offer associated counselling. Individuals who wish to have children should be screened to discover heterozygous genotypes that could be important in genetic counselling. HbAS screening enables informed decisions concerning preconception counselling and prenatal diagnosis.

Routine fitness training does not increase the risk of mortality for individuals with HbAS. However, there is a concern of increased risk of rhabdomyolysis (rapid destruction of skeletal muscle) and sudden death during intense, prolonged physical activity; this risk can be mitigated by proper training (Hassan *et al*., 2024). In some regions, these observations have resulted in voluntary or mandatory screening of athletes for HbAS (Hassan *et al*., 2024). There are rare and specific complications of HbAS that should prompt HbAS testing. These include haematuria (blood in the urine), hyphema (blood inside the eye's anterior chamber) and renal medullary carcinoma, a rare malignancy. HbAS could be a risk factor for chronic kidney disease and pulmonary embolism (Hassan *et al*., 2024).

2.1.5 Management

There are a number of precautions which can help reduce the risk of developing a sickling crisis. Lifestyle behaviours include maintaining good hydration and avoiding physical stress or exhaustion. Since sickling can be triggered by low oxygen levels, people with SCD should avoid high altitudes such as high mountains or flying in unpressurised aircraft. (CDC, 2024; NICE, 2021)

People with SCD should avoid alcohol and smoking, as alcohol can cause dehydration and smoking can trigger acute chest syndrome. Stress can also trigger a sickle cell crisis, so relaxation techniques like breathing exercises can help. (NICE, 2021).

Pneumococcal infection is a leading cause of death among children with SCD; penicillin is recommended daily during the first 5 years of life in order to minimise the risk of infection (MSDH, 2024).

Dietary supplementation of folic acid is sometimes recommended, on the basis that it facilitates the creation of new red blood cells and may reduce anemia. A Cochrane review of its use in 2016 found "the effect of supplementation on anaemia and any symptoms of anaemia remains unclear" due to a lack of medical evidence (Benson *et al*., 2021).

People with SCD are recommended to receive all vaccinations which are recommended by health authorities in order to avoid serious infection which might trigger a sickling crisis (Emmanuel & Getrude, 2024). Hydroxyurea was the first approved drug for the treatment of SCD, which has been shown to decrease the number and severity of attacks and possibly increase survival time (Rankine-Mullings & Nevitt, 2022).

This is achieved, in part, by reactivating fetal haemoglobin production in place of the haemoglobin S that causes sickling. Hydroxyurea lowers the expression of adhesion molecules

on endothelial and red blood cells, which lowers the chance of small vessel blockages. Additionally, it encourages the release of nitric oxide, which enhances blood flow and inhibits the formation of clots (Gupta & Kumar, 2024).

Hydroxyurea had previously been used as a chemotherapy agent, and some concern exists that long-term use may be harmful (Rankine-Mullings & Nevitt, 2022). A Cochrane review in 2022 found a weak evidence base for its use in SCD (Rankine-Mullings & Nevitt, 2022).

2.1.5.1 Blood transfusion

A simple blood transfusion can be used to treat SCD when hemoglobin levels drop too low, or to prepare for an operation or pregnancy. It can also be used to protect against long-term complications, or to reduce the risk of stroke. The simple, or top-up transfusion is a procedure in which healthy blood cells from a donor are infused into the patient's bloodstream. This benefits by alleviating anemia and increasing oxygen levels in the tissues, reducing the risk of sickling and relieving sickling symptoms. A simple transfusion can be used to treat SCD when hemoglobin levels drop too low, or to prepare for an operation or pregnancy. It can also be used to protect against long-term complications, or to reduce the risk of stroke (Han *et al*., 2021). An exchange transfusion is a procedure in which blood is removed from the body, then processed to extract sickled cells, which are replaced by healthy red blood cells from a donor.

In children, preventive RBC transfusion therapy has been shown to reduce the risk of first stroke or silent stroke when transcranial Doppler ultrasonography shows abnormal cerebral blood flow. In those who have sustained a prior stroke event, it also reduces the risk of recurrent stroke and additional silent strokes (Thurn *et al*., 2022).

Most people with sickle cell disease have intensely painful episodes called vaso-occlusive crises (VOC). However, the frequency, severity, and duration of these crises vary tremendously. In a VOC, the circulation of blood vessels is obstructed by sickled red blood cells, causing ischemic injuries to the tissues, inflammation and pain. Recurrent episodes may cause irreversible organ damage (Maakaron, 2019).

In 2019, crizanlizumab, a monoclonal antibody targeting P-selectin, was approved in the United States to reduce the frequency of vaso-occlusive crisis in those 16 years and older (CDER, 2019). It had also been approved in the UK and Europe, but in both cases authorisation was subsequently withdrawn because of poor evidence of its effectiveness.

Acute chest syndrome is caused by vaso-occlusion occurring in the lungs. As with a VOC, treatment includes pain control and hydration. Antibiotics are required because there is a severe risk of pulmonary infection, and oxygen supplementation for hypoxia. Blood transfusion may also be required, or exchange transfusion in severe cases (Friend & Girzadas, 2022).

2.1.5.2 Treating avascular necrosis

When treating avascular necrosis of the bone in people with sickle cell disease, the aim of treatment is to reduce or stop the pain and maintain joint mobility (Martí-Carvajal *et al*., 2019). Treatment options include resting the joint, physical therapy, pain-relief medicine, joint-replacement surgery, or bone grafting (Martí-Carvajal *et al*., 2019).

2.1.5.3 Stem cell treatments

There are two possible ways to treat SCD and some other hemoglobinopathies by targeting HSCs. Since 1991, a small number of patients have received bone marrow transplants from healthy matched donors, although this procedure has a high level of risk (Aprile *et al*., 2022). More recently, it has become possible to use CRISPR gene editing technology to modify the patient's own HSCs in a way that reduces or eliminates the production of sickle hemoglobin HbS and replaces it with a non-sickling form of hemoglobin (Olson & Walters, 2023). All stem cell treatments must involve myeloablation of the patients' bone marrow in order to remove HSCs containing the faulty gene.

2.1.5.4 Hematopoietic stem cell transplantation

Hematopoietic stem cell transplantation (HSCT) involves replacing the dysfunctional stem cells from a person with sickle cell disease with healthy cells from a well-matched donor (Rotin *et al*., 2023). Finding a well matched donor is essential to the process' success. Different types of donors may be suitable and include umbilical cord blood, human leukocyte antigen (HLA) matched relatives, or HLA matched donors that are not related to the person being treated (Rotin *et al*., 2023). Risks associated with HSCT can include graft-versus host disease, failure of the graft, and other toxicity related to the transplant (Rotin *et al*., 2023).

2.1.5.5 Long-term management

Improved management of acute complications is associated with a longer survival. As individuals with SCD age, chronic problems resulting from cumulative organ injury can lead to severe morbidity (Fitzhugh *et al*., 2022). Chronic pain is common; the Pain in Sickle Cell Epidemiology Study (PiSCES) found that adults with SCD have pain on 55% of days and pain, in general, is a poorly managed complication of SCD, Individuals with SCD and recurrent pain

have altered brain network connectivity, which affects their response to treatment. Chronic pain requires a multidisciplinary team familiar with neuropathic pain tolerance, withdrawal symptoms and hyperanalgesia syndrome (Fitzhugh *et al*., 2022). Hydroxycarbamide, selective use of chronic transfusions in severe cases and long-acting opioids are useful components of a multidisciplinary pain management approach.

Avascular necrosis of the hip is a common cause of chronic pain that eventually develops in many individuals with SCD (Alshurafa *et al*., 2023), in >20% of hospitalizations, symptoms are related to avascular necrosis. Although core decompression (in which a small core of bone is removed from the damaged area, lowering the bone marrow pressure and stimulating healthy bone regrowth), physiatry (rehabilitation) therapy and analgesics are temporarily helpful, total hip replacement is often required.

Chronic kidney disease is relatively common in older individuals with SCD and is thought to have a poor prognosis in these individuals compared with individuals without SCD (Amarapurkar *et al*., 2022).

This worse outcome could in part be due to delayed access to dialysis and renal transplant for individuals with SCD, as they might not be considered good candidates for these therapies. Of note, individuals with SCD who receive a timely renal transplantation have an outcome comparable with that of individuals without SCD who receive a transplant (Amarapurkar *et al*., 2022).

Although screening for brain injury with annual transcranial Doppler (TCD) screening and/or MRI and chronic transfusion therapy for high-risk patients decrease the frequency and severity

of stroke complications, patients continue to have progressive neurocognitive injury and require close observation and long-term therapy (Guy *et a*l., 2024)

In addition, implementation of multidisciplinary plans for management of other common chronic complications of SCD (for example, cardiopulmonary dysfunction, priapism and leg ulcers) improves the quality of life of these patients as they age (Bivalacqua *et al*., 2022)

2.2 Economic Burden of Sickle Cell Disease (SCD)

Sickle Cell Disease (SCD) is a genetically inherited blood disorder that places a significant economic burden on affected individuals, families, healthcare systems, and society. This economic burden includes direct medical costs, indirect costs due to lost productivity, and intangible costs related to reduced quality of life. The financial impact of SCD varies based on healthcare infrastructure, access to treatment, and the prevalence of the disease across regions.

2.2.1 Direct Medical Costs of SCD

In high-income countries, direct medical costs associated with SCD are significant. These costs primarily include hospitalization, medications, treatments for chronic and acute complications, and hematopoietic stem cell transplantation (HSCT). In Brazil, the annual cost of SCD is estimated at $414 million USD, with $123 million USD attributed to direct medical costs (Barbosa *et al*., 2021). Chronic complications such as chronic calculous cholecystitis and chronic kidney disease significantly contribute to these costs (Barbosa *et al*., 2021). In the United States, the lifetime medical costs for a person with SCD can reach approximately $1.7 million USD (Johnson et al., 2022). These expenses include frequent hospitalizations, emergency department visits, and long-term treatments like blood transfusions and hydroxyurea therapy.

In sub-Saharan Africa, where SCD has the highest prevalence, the economic burden is also substantial. For instance, a study in the Republic of Congo found that the median hospitalization cost for severe acute SCD complications was $115.21 USD (Lobo *et a*l., 2022)

Given that 27.7% of households in this region earn less than $158.40 USD, these costs can be financially crippling for many families (Lobo *et al*., 2022)

2.2.2 Indirect Economic Costs: Lost Productivity

In addition to direct medical expenses, SCD leads to considerable indirect costs due to lost productivity. In the United States, it is estimated that individuals with SCD miss an average of 7 weeks of work annually because of pain crises (Lal *et al*., 2021). This results in $1.5 billion USD in lost productivity each year (Lal *et al*., 2021). Individuals with SCD often report working while in pain, further reducing their productivity. This can lead to financial insecurity for affected individuals and their families.

In Nigeria, the indirect costs of SCD are also significant. A study in Ekiti State found that monthly health expenditures for households with SCD patients ranged from $15 to $1,303 USD, with a mean of $240 USD. Notably, 20.7% of households experienced catastrophic health expenditures, spending more than 10% of their income on healthcare (Ahmed et al., 2021)

2.2.3 Cost-Effectiveness of Interventions

Investing in early interventions, such as newborn screening, prophylactic treatments, and hydroxyurea therapy, is considered cost-effective in managing SCD. In sub-Saharan Africa, implementing universal newborn screening and prophylactic treatments is estimated to cost $312 million USD annually (Stenberg *et al*., 2021). Although this represents a significant investment, the cost per Disability-Adjusted Life Year (DALY) averted is highly favorable, suggesting that these interventions are economically viable. Studies have shown that early detection and

preventive care can significantly reduce long-term healthcare costs and improve patient outcomes (Michaels *et al*., 2021). In addition, treatments such as hydroxyurea can reduce hospitalizations by up to 50%, lowering healthcare expenditures in the long run (Teigen *et al*., 2023)

2.3 Payment Coping Mechanisms for Sickle Cell Disease Patients

The financial burden of Sickle Cell Disease (SCD) often forces patients and their families to adopt various payment coping mechanisms to afford treatment. These mechanisms are broadly classified into formal (structured) mechanisms, such as health insurance and government aid, and informal (unstructured) mechanisms, including out-of-pocket payments, borrowing, selling assets, and seeking charitable aid (Onwujekwe *et al*., 2020).

According to WHO (2022), out-of-pocket (OOP) payments remain the dominant mode of healthcare financing in low-income countries, including Nigeria, leading to catastrophic health expenditure (CHE) for many households. Studies have shown that over 70% of Nigerian families with chronic illnesses rely on OOP payments (Adekunle Bamidele Taiwo *et al*., 2023)

2.3.1 Formal Coping Mechanisms (Structured Strategies)

2.3.1.1 National Health Insurance Scheme (NHIS)

The National Health Insurance Scheme (NHIS) was established to improve access to affordable healthcare, but its coverage for SCD remains inadequate. According to Onoka *et al*. (2020), less than 5% of Nigerians with chronic illnesses benefit from NHIS coverage, leaving many SCD patients without insurance support.

2.3.1.2 Challenges of NHIS for SCD Patients

Limited Coverage – The NHIS does not comprehensively cover hydroxyurea therapy, bone marrow transplants, or frequent blood transfusions, essential for managing SCD (Miller *et al.,* 2025).

Exclusion of Informal Sector – NHIS primarily covers public sector workers, excluding self-employed individuals and low-income earners, who make up the majority of SCD patients.

Delayed Reimbursements – Bureaucratic bottlenecks cause delays in NHIS reimbursements, leading hospitals to demand upfront payments from patients (Onwujekwe *et al*., 2020). Studies by Bazuaye *et al*. (2021) emphasize that expanding NHIS coverage and streamlining reimbursement processes could significantly alleviate financial burdens for SCD patients.

2.3.1.3 Government and Donor Assistance Programs

While government support for SCD in Nigeria remains limited, certain programs offer relief:

Subsidized Medications – Some state governments provide discounted essential drugs, but availability is inconsistent (Ike & Eze, 2021).

Tertiary Hospital Assistance Programs – Hospitals like UBTH partner with NGOs and international donors to offer free pediatric blood transfusions (Ewelukwa *et al*., 2019).

International Aid – Organizations like WHO, UNICEF, and the Sickle Cell Foundation Nigeria support research and patient screening but provide minimal direct financial assistance (Adegoke *et al*., 2019). Despite these interventions, studies show that most Nigerian SCD patients continue to bear the bulk of their medical expenses, making informal coping mechanisms necessary (Onwujekwe *et al*., 2020).

2.3.2 Informal Coping Mechanisms (Unstructured Strategies)

2.3.2.1 Out-of-Pocket Payments (OOPs)

Out-of-pocket payments remain the primary method of financing SCD treatment in Nigeria. Studies by WHO (2022) indicate that catastrophic health expenditures (CHE) occur when healthcare costs exceed 40% of a household's income. In Nigeria, a significant percentage of SCD families experience CHE due to recurrent medical costs (Amarachukwu *et al*., 2022).

Consequences of OOPs on SCD Patients:

Financial Hardship – Many families exhaust their savings, leading to economic vulnerability (Bazuaye *et al*., 2021).

Treatment Delays and Avoidance – Due to financial constraints, SCD patients often postpone medical visits, increasing morbidity (Adegoke *et al*., 2019).

Health Inequality – OOP payments disproportionately impact low-income families, worsening healthcare disparities (Ike & Eze, 2021).

2.3.2.2 Borrowing and Debt Financing

Because of the chronic nature of SCD, many families resort to borrowing. Common borrowing sources include:

Relatives and Friends – Informal, interest-free loans.

Cooperative Societies – Community-based lending groups.

Microfinance Institutions – Loans with high-interest rates, leading to debt cycles

A study by Onwujekwe *et al*. (2020) found that 35% of Nigerian families with chronic illnesses took loans for healthcare, many struggling with long-term debt.

2.3.2.3 Selling Assets and Property

Some families liquidate assets such as land, jewelry, and livestock to cover medical expenses. However, this strategy often results in:

Loss of Financial Stability – Once assets are sold, families become more vulnerable to future crises (Bazuaye *et al*., 2021).

Social Decline – Selling assets reduces a family's ability to invest in education or business, worsening poverty levels (Onwujekwe *et al*., 2020).

2.3.2.4 Community and Philanthropic Support

Some patients seek financial aid from NGOs, religious groups, and crowdfunding platforms (Ike & Eze, 2021). NGOs like the Sickle Cell Foundation Nigeria provide limited financial support (Adegoke *et al*., 2019). Faith-Based Organizations conduct fundraising campaigns. Online Crowdfunding (GoFundMe, Twitter donations) has gained popularity but remains unreliable (Ike & Eze, 2021).

The economic burden of SCD forces patients into high-risk financial strategies, including borrowing, selling assets, or delaying treatment, due to limited insurance coverage and government support (Onwujekwe *et al*., 2020). Expanding NHIS coverage, increasing government subsidies, and developing alternative funding models are crucial to alleviating this financial strain .

2.4 Theoretical review

2.4.1 Cost of Illness (COI) Framework

The Cost of Illness (COI) framework provides a comprehensive approach to understanding the economic burden of diseases. It is widely used in health economics to quantify the direct, indirect, and intangible costs of a disease, which can be particularly valuable for chronic illnesses like Sickle Cell Disease (SCD) (Songer and Ettaro 2016). The framework helps in identifying the total economic burden of a disease on both individual patients and society. This is vital for policy makers to understand the need for intervention, resource allocation, and insurance policy improvement.

In the context of Sickle Cell Disease (SCD) in Nigeria, the COI framework allows a detailed analysis of the costs incurred by patients and their families, which is particularly useful for resource-constrained settings like Benin City, where the research is focused.

2.4.1.1 Components of the Cost of Illness Framework

The COI framework divides costs into three primary categories: direct, indirect, and intangible costs. Each category provides a lens through which we can view the economic burden of SCD in different ways.

2.4.1.1.1 Direct Costs

Direct costs refer to all expenses related to the medical care and treatment of a disease. In the case of Sickle Cell Disease, these can be broken down into two subcategories: medical costs and non-medical costs.

Medical Costs

Medical costs encompass all expenses related to the healthcare system that a patient incurs while seeking treatment for SCD. These costs include:

Hospital Admissions: SCD patients often require frequent hospital admissions due to complications such as pain crises, stroke, organ damage, and infection. Studies indicate that hospitalization rates are high, leading to increased spending on beds, physician consultations, tests, and nursing care.

Medications: The cost of medications for SCD can be significant. Common medications include hydroxyurea, which helps in managing the disease, and blood transfusions to alleviate anemia or prevent complications. Pain management drugs, antibiotics, and antimalarials for infections also add to the financial burden (Bazuaye *et al*., 2021).

Laboratory Tests and Diagnostics: Sickle Cell patients undergo regular diagnostic tests like complete blood counts (CBCs), hematology tests, and genetic screening, all of which contribute to the overall medical cost (WHO, 2022).

Specialist Consultations: Due to the complex nature of the disease, SCD patients often need specialist consultations, such as hematologists, pain specialists, and neurologists, which add to the financial load.

Non-Medical Costs

Non-medical costs are the expenses that patients and their families incur to access healthcare services that are not directly related to medical treatment. These costs include:

Transportation: Traveling to healthcare facilities, particularly in countries like Nigeria, where many hospitals are centralized in urban areas, can be an additional financial burden. The cost of

transportation, whether by public transport, private vehicles, or ambulances, can add significantly to the total healthcare expenses (Ike & Eze, 2021).

Caregiver Expenses: Family members or caretakers often spend considerable time and resources on taking care of an SCD patient. This can include lost wages if caregivers must take time off work to provide care (Adegoke *et al*., 2019). Additionally, they may need to pay for home-based care or assistive devices for the patient, further increasing costs.

The direct costs of SCD can thus be substantial, especially when compounded by recurrent hospitalizations and long-term treatment regimens.

2.4.1.1.2 Indirect Costs

Indirect costs refer to the economic loss that arises due to the impact of the illness on workforce participation and productivity.

Lost Productivity: One of the largest indirect costs associated with SCD is lost productivity. Caregivers often have to reduce their working hours or leave their jobs altogether to manage the needs of the patient. This creates a loss of income, which impacts the household's overall financial stability. In cases of children with SCD, parents or guardians often have to forgo regular employment to care for their children, which can lead to a reduction in household income.

In addition, family members may be required to take time off from their regular jobs to attend doctor's appointments or provide support during pain crises.

Premature Mortality: Another indirect cost is the loss of potential income resulting from premature deaths associated with SCD. Despite advances in treatment, SCD is still a leading cause of early mortality due to complications such as stroke, organ failure, and infection. This

can result in a reduction in the workforce, especially in families that depend on the income of affected individuals. The economic consequences of premature mortality often extend beyond immediate family members, affecting extended families and communities.

Intangible Costs

Intangible costs refer to the non-financial effects of a disease, which are often the most challenging to quantify but still contribute significantly to the overall burden of disease.

Psychological Distress: Living with a chronic illness like SCD is associated with emotional and psychological distress. Patients, caregivers, and families often experience anxiety, depression, and stress due to the unpredictable nature of the disease. This distress can be attributed to:

Fear of Mortality: Families are often in a constant state of worry about the health status of the patient and the possibility of early death.

Stigmatization: In many African societies, there is still some level of stigma attached to having a genetic condition like SCD, which may lead to social isolation or discrimination in some cases (Ike & Eze, 2021).

Reduced Quality of Life (QoL): The quality of life for both the patients and their families is significantly affected by SCD. Patients frequently experience pain episodes, fatigue, and limitations on physical activities, which can impact their ability to work, attend school, or engage in social activities. As a result, they may feel a significant reduction in well-being, which directly contributes to intangible costs.

2.4.1.2 Application of the COI Framework to SCD

When applied to Sickle Cell Disease in Nigeria, the COI framework reveals that the economic burden of the disease is multifaceted. Many families in Nigeria face a severe financial strain due to the high direct medical costs and the loss of income due to the impact of the disease on their ability to work.

A national study by Onwujekwe *et al*. (2020) revealed that over 60% of families affected by SCD in Nigeria incur significant out-of-pocket expenses for treatment. These families often face catastrophic health expenditures that push them further into poverty, as the lack of adequate health insurance coverage leads to an out-of-pocket financing system for healthcare costs.

Furthermore, studies have shown that the indirect costs of caregiving and lost productivity are also high. Parents often sacrifice careers and income to provide the needed care for their children or spouses. As a result, the economic strain leads to greater social vulnerability for affected families

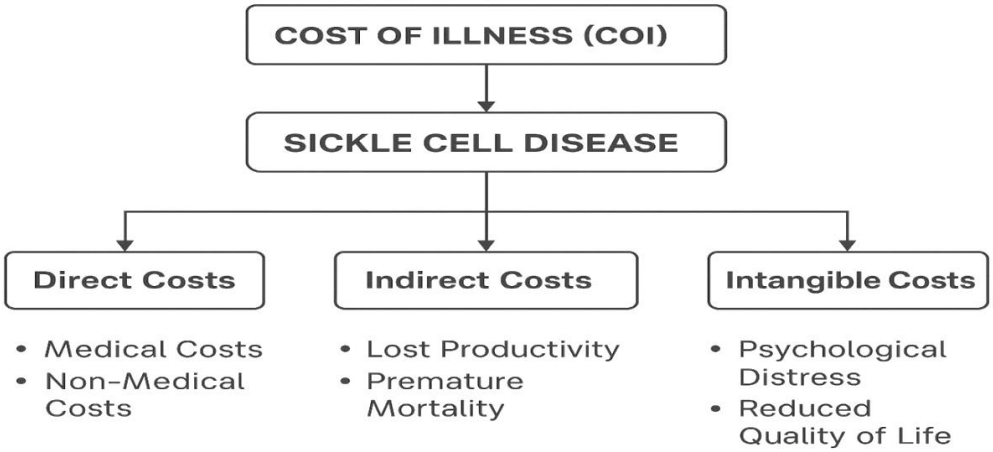

Figure 2.1 Schematic representation of cost of illness (COI) framework (Songer and Ettaro 2016).

2.5 Empirical review

2.5.1 Direct Medical Cost of sickle cell disease Incurred by Patients

Amarachukwu *et al*. (2022) conducted a cross-sectional descriptive survey at the University of Nigeria Teaching Hospital, Enugu, involving 149 SCD patients. A structured pre-tested interviewer-administered questionnaire was used to collect primary data from adult participants and caregivers of paediatric patients. Data collection lasted for three months. The study found a median monthly economic burden of approximately ₦76,711 (US$385) per patient, with

outpatient costs constituting about 88% of this expenditure. Major contributors to these costs included admissions, medications, and blood transfusions. Notably, all socioeconomic groups experienced catastrophic health expenditures, with the poorest quartile being the most affected: 61% at a 40% threshold, 71% at a 30% threshold, and 88% at a 10% threshold. The authors concluded that SCD patients in Nigeria face substantial economic burdens and catastrophic costs due to out-of-pocket expenditures, highlighting the need for financial protection policies, especially for the poorest populations.

Baldwin et al. conducted a systematic review to evaluate empirical evidence on the economic burden of sickle cell disease (SCD) in the United States, with a particular focus on both medical and non-medical costs. The study was motivated by the anticipated high costs of emerging genetic therapies and the need for informed decision-making regarding their adoption. Using MEDLINE and EMBASE, the authors searched for relevant studies published between 2008 and 2020. To be eligible, studies had to provide empirical cost estimates related to SCD among individuals of all ages and their caregivers. A total of 479 studies were initially identified, of which 40 met the inclusion criteria—39 reporting on medical costs and just one on non-medical costs. All cost data were adjusted to 2019 U.S. dollars, and study quality was assessed using the Newcastle–Ottawa Scale.

The results demonstrated a consistently higher cost burden for individuals with SCD compared to those without the disease, with annual cost differences ranging from $6,636 to $63,436. Inpatient costs were the highest component ($11,978–$59,851 annually), followed by outpatient and pharmacy expenses. However, the review also revealed significant gaps in the literature. Notably, no studies provided a comprehensive view of costs across the entire disease trajectory, nor did they adequately account for caregiver burden or productivity losses. The authors

concluded that there is an incomplete characterization of both medical and non-medical costs related to SCD, and they emphasized the need for more comprehensive research to fully understand the economic impact of the disease and the implications of emerging treatments.

Johnson et al. (2022) conducted a retrospective cohort study to estimate the lifetime medical and out-of-pocket (OOP) costs attributable to sickle cell disease (SCD) among nonelderly individuals with commercial insurance in the United States. Using Truven Health Marketscan commercial claims data from 2007 to 2018, the authors analyzed a cohort of 20,891 individuals diagnosed with SCD and compared them with 33,588 matched controls from the Medical Expenditure Panel Survey. The analysis employed Kaplan-Meier sample average cost methods and incorporated survival curves previously reported for SCD and non-SCD populations. The SCD cohort had a mean age of 25.7 years, and the gender distribution was 58% female.

The study found that lifetime costs attributable to SCD were substantial, estimated at $1.6 million for females and $1.7 million for males between ages 0 and 64. The associated OOP costs were $42,395 and $45,091, respectively. These costs represented a 907% increase in total medical expenditures and a 285% increase in OOP costs compared to the control group. Notably, healthcare expenditures peaked during adolescence and young adulthood (ages 13–24) and declined thereafter. No significant gender differences were observed in terms of cost burden. Although the study was limited to commercially insured individuals, its findings highlight the intense economic strain imposed by SCD across the lifespan, underscoring the importance of developing curative and innovative treatment options that may alter the disease's trajectory and associated costs.

Eze et al. (2023) investigated the economic burden associated with pentazocine dependence among sickle cell disease (SCD) patients in Nsukka Local Government Area, Enugu State,

Nigeria. Recognizing that pentazocine is frequently used for managing SCD-related pain, the study highlights how unsupervised and prolonged usage can lead to dependence, resulting in heightened healthcare expenses, reduced labor productivity, and broader socio-economic consequences. The researchers utilized self-reported cost-of-illness instruments to gather data from 25 pharmacy stores, focusing on both direct and indirect costs borne by patients.

Direct medical and non-medical expenses were calculated through patients' out-of-pocket payments, while indirect costs, such as productivity losses, were assessed using the human capital approach. The findings revealed an average direct cost of NGN 116,587.71 (USD 143) and an indirect cost of NGN 17,415.06 (USD 21.37), resulting in a total average economic burden of NGN 134,002.77 (USD 164.42) per affected patient. Extrapolating these figures, the study projected the annual financial cost of pentazocine dependence in the region to be approximately NGN 422 million (USD 517,983.52). The cost structure showed that 87% of the burden was due to direct expenses, while 13% came from lost productivity.

These findings emphasize the hidden yet substantial economic impact of drug dependence in managing chronic illnesses like SCD. The authors advocate for more structured oversight in the prescription and use of pain medications, as well as the implementation of supportive policies to mitigate the health and financial risks associated with analgesic dependence.

Udeze et al. (2023) conducted a retrospective cohort study to evaluate the clinical complications, treatment patterns, healthcare resource utilization (HCRU), and economic costs associated with managing sickle cell disease (SCD) in patients experiencing recurrent vaso-occlusive crises (VOCs) in the United States. Using the Merative MarketScan Databases, the study identified patients with at least one inpatient or two outpatient claims for SCD and two or more VOCs per year across two consecutive years between March 1, 2010, and March 1, 2019. These patients

were followed for a minimum of 12 months, starting from the second VOC in the second year, until either inpatient death, loss of healthcare coverage, or March 1, 2020. A control group without SCD was matched for comparison.

The study identified 3,420 patients with recurrent VOCs and 16,722 matched controls. During follow-up, the SCD group had a mean of 5.0 VOCs (SD = 6.0), 2.7 inpatient admissions (SD = 2.9), and 5.0 emergency department visits (SD = 8.0) per patient per year. The average annual healthcare cost for these patients was $67,282, compared to $4,134 for controls. Lifetime healthcare costs were estimated at $3.8 million for patients with recurrent VOCs, in contrast to $229,000 for matched controls over a 50-year span.

The findings show that patients with SCD and recurrent VOCs face a significant clinical and economic burden, largely due to frequent hospital visits and high inpatient costs. The study emphasizes the need for effective treatments that can alleviate clinical complications and reduce the long-term economic impact of the disease.

.2.5.2 Indirect Medical Cost of sickle cell disease Incurred by Patients

Holdford et al. (2021) conducted a study to assess the indirect economic burden of sickle cell disease (SCD) in the United States, focusing on productivity losses experienced by adult patients. Using data collected from a sickle cell clinic at an urban academic healthcare system, the researchers employed an adapted version of the Institute for Medical Technology Assessment Productivity Cost Questionnaire. This instrument captured the effects of SCD on absenteeism, presenteeism, and non-paid work activities. The study also incorporated clinical data such as genotype, hemoglobin levels, and reported pain severity.

Out of 192 adult patients surveyed, 187 reported experiencing vaso-occlusive crises in the previous year that negatively impacted their ability to work and function in daily roles. About

75% of participants indicated difficulty completing routine activities, including child care, household chores, errands, and unpaid or volunteer work. Notably, only 30% of the respondents were employed or self-employed. Among those employed, the combined costs of absenteeism and presenteeism were estimated at $15,103 per person annually. Furthermore, the average annual loss in unpaid productivity was valued at $3.1 million for patients and $2.87 million for their caregivers.

These findings reveal that the indirect costs of SCD are substantial, extending far beyond medical expenses and impacting both patients and their support networks. The study highlights the profound effect of chronic pain and disease-related complications on economic productivity and quality of life, thereby underscoring the need for more effective management strategies and supportive interventions for individuals living with SCD.

AlRuthia et al. (2024) conducted a cross-sectional study to evaluate the health-related quality of life (HRQoL) and out-of-pocket expenditures (OOPEs) associated with sickle cell disease (SCD) among patients in Saudi Arabia. The study was based at a university-affiliated tertiary care center in Riyadh and involved 118 consenting patients whose sociodemographic and clinical data were retrieved from electronic medical records, while OOPEs and HRQoL data were gathered through structured questionnaires. The analysis applied a range of statistical methods including t-tests, ANOVA, and multiple linear regression to examine associations between variables.

The results indicated that the mean age of participants was 31 years, with females comprising about 53% of the sample. The study found that SCD imposed a considerable financial burden, with average monthly OOPEs amounting to USD 650.69 and a substantial proportion of patients reporting income loss due to illness. Additionally, most patients had a low monthly family income (below USD 2666.67), further compounding the economic challenges. The estimated

age-adjusted quality-adjusted life years (QALYs) for the cohort was 24.33 years, significantly below average benchmarks.

Statistical analysis revealed that increased frequency of blood transfusions and the presence of multiple comorbidities were significantly associated with lower HRQoL scores. Conversely, higher educational attainment was positively associated with improved HRQoL. These findings demonstrate the dual burden—both financial and health-related—faced by SCD patients in Saudi Arabia and point to the urgent need for targeted interventions, including improved access to care, support for education, and policy reforms to reduce financial hardship and enhance quality of life

Ajayi et al. (2021) investigated the prevalence of household catastrophic health expenditure (CHE) in rural and urban communities in Ekiti State, Nigeria, using a comparative cross-sectional design. The study surveyed 1,000 household heads through a pre-tested semi-structured questionnaire, employing a multistage sampling technique. Data were collected over four months and analyzed using SPSS and STATA, with two different methodologies used to determine CHE prevalence and a sensitivity analysis conducted to validate results.

Findings showed a notably high prevalence of CHE across both settings, with significantly higher rates in rural areas. Using the first method, rural CHE prevalence was 18.5% compared to 12.8% in urban areas ($p = 0.015$). The second method yielded 8.3% for rural and 2.5% for urban areas ($p < 0.001$). These disparities suggest that rural households are disproportionately affected, likely due to limited access to financial protection mechanisms and lower average incomes.

The study underscores the pressing need for social protection and financial intervention strategies to mitigate the economic burden of healthcare on Nigerian households, particularly in rural communities. It provides empirical evidence for policy reforms aimed at achieving

universal health coverage and reducing out-of-pocket health spending that pushes families into poverty.

.Beillat et al. (2023) conducted a real-world retrospective study to examine the prevalence, healthcare utilization, and costs associated with sickle cell disease (SCD) in France. Using data from the Echantillon Généraliste des Bénéficiaires, a nationally representative 1/97th sample of the French health system database, the study focused on patients aged 12 years and older between 2016 and 2018. A total of 151 SCD patients with at least six months of follow-up were matched in a 1:3 ratio with non-SCD controls based on age, sex, socioeconomic status (CMU-c), and geographic region.

The analysis revealed that SCD patients had significantly higher healthcare costs than controls, with an average reimbursed cost of €24,310, primarily driven by hospitalizations (€21,156). When annualized, mean costs were €25,680 for SCD patients compared to €3,227 for controls ($p < 0.001$), indicating a substantial cost differential. Extrapolating to the national level, the extra cost burden of SCD was estimated at nearly €150 million. The study also found age-related differences in care patterns: younger patients had more frequent hospitalizations and acute interventions, while older patients utilized more routine medical and paramedical services.

These findings underscore the considerable economic impact of SCD in France and highlight the urgent need for improved treatment strategies and the development of innovative therapies that could potentially reduce disease burden and associated costs. The study contributes to the limited European data on SCD and offers evidence to inform healthcare planning and policy.

2.5.3 Payment coping mechanism Utilizes by different socio-economic groups

A study by Ogamba *et al*. (2019) assessed the awarenessof health insurance, patterns of health service utilization and financial implications of sickle cell disease among children seeking care at a tertiary facility in Nigeria. A structured questionnaire was administered to parents of 314 children with sickle cell disease attending the pediatric hematology unit of the Lagos University Teaching Hospital between May and December 2019.

The mean age of the children was 91.5 ± 43.1 months. M:F was 1.17:1. 45.5% of households earned above NGN 150,000 (USD 417) monthly. 71.3% of the parents had heard of health insurance but only 20.7% were enrolled in a health insurance scheme. Awareness of health insurance was significantly associated with social class (p=0.000) and monthly household income (p=0.000). 60.8% of the parents preferred pre-facility treatment. Social class (p=0.01) and monthly household income (p=0.001) were significantly associated with home treatment. Time on admission ranged from 2-18 days with an average of 4.31 days. Average cost of hospitalization was USD 148 ± USD 14.2 and total cost of care incurred was USD 20,787. Neither age of child (p=0.857), estimated household income (p=0.863) nor social class (p=0.397) was associated with cost of care. The study highlighted the high cost of SCD care and the low penetration of health insurance among affected families, advocating for increased awareness and access to health insurance to alleviate financial burdens.

Olatunya *et al*. (2015) conducted a longitudinal descriptive study involving 111 children with SCD managed at Ekiti State University Teaching Hospital. The study reported that only 7.2% of households were enrolled in the National Health Insurance Scheme, leading to significant out-of-pocket expenses. Monthly household incomes ranged from ₦12,500 to ₦330,000 (US$76 to US$2,000), with health expenditures averaging ₦39,554±35,479 (US$240±215). Notably, 20.7% of households experienced catastrophic health expenditures, defined as health expenses

exceeding 10% of family income. Factors such as taking loans to offset hospital bills, low social class, and recent illness episodes significantly increased the likelihood of catastrophic expenditures. The study concluded that SCD imposes a substantial financial burden on families, emphasizing the need for improved health insurance coverage and financial support mechanisms.

Alinda et al. (2025) conducted a qualitative study in Uganda to explore the often-overlooked experiences of caregivers raising children with sickle cell disease (SCD). Using in-depth interviews with twelve caregivers at Mulago sickle cell clinic, the research uncovered the emotional, financial, and social toll imposed by caregiving in a high-prevalence setting—where SCD rates vary significantly, peaking at 22.2% in the northern region.

The study revealed that caregivers initially faced confusion and uncertainty prior to diagnosis, followed by persistent financial strain and limited social support, which significantly affected their emotional health. Coping strategies ranged from reliance on traditional remedies to modern medical treatments. Despite demonstrating resilience, many caregivers reported adverse outcomes, including sleep disturbances and psychological distress, largely intensified by the financial burdens of continuous care.

The findings emphasize the necessity of increasing public awareness and community education about SCD to support caregivers. Empowering caregivers with knowledge and emotional resources could improve both their quality of life and the stability of affected households, indicating a broader need for policy and healthcare interventions targeting caregiver support systems.

Ampomah et al. (2024) conducted a qualitative study in Accra, Ghana, to explore the financial strain experienced by parents of children with sickle cell disease (SCD). With Ghana having a high prevalence of SCD—affecting 2% of all live births—the study focused on how parents navigate the financial burdens associated with caring for affected children.

Interviews with twenty-seven parents revealed significant direct costs, including expenses for medications, routine tests, and hospital stays, many of which are not fully covered by the National Health Insurance Scheme. Indirect costs, such as loss of income due to reduced work hours or job resignation, compounded the financial pressure. Participants also expressed frustration over limited access to financial support and the lack of understanding from policymakers regarding their challenges.

The study concluded that addressing these financial needs through better policy support and resource allocation could improve the emotional and social well-being of caregivers, ultimately enhancing their capacity to provide care.

Abba et al. (2023) conducted a study at Specialist Hospital Sokoto, Nigeria, to explore how children with sickle cell disease (SCD) and their parents cope with the challenges of the condition. Using a mixed-method design, the study involved 112 children aged 8 to 13 years and 122 parents. Standardized questionnaires and interviews were used to collect data, which was analyzed using both descriptive and inferential statistics.

The findings revealed that 61.6% of respondents frequently used active coping strategies such as problem-solving and seeking support, while 35.7% often used avoidant strategies like distraction. Negative coping methods, such as denial or substance use, were the least commonly used at

3.6%. Among coping assistance strategies provided by parents, disengagement—such as helping children withdraw from stressful situations—was the most common at 53.3%.

Qualitative data revealed five recurring themes: stressors related to SCD, the coping mechanisms used, the support strategies from parents, how effective these strategies were perceived to be, and the barriers that hindered their use. There was a statistically significant association between the perceived helpfulness of coping strategies and the use of active coping ($p = 0.018$), while negative coping was linked to poor outcomes ($p < 0.001$).

This study underscores the importance of family support and calls for broader public awareness and healthcare interventions that address both the physical and psychological aspects of managing SCD in children.

Atya et al. (2024) investigated the impact of a structured education booklet on self-efficacy, self-care practices, and health-related quality of life among adults with sickle cell disease (SCD). The study was conducted using a pre-experimental one-group (pretest-posttest) design at the Clinical Hematology Unit of Assiut University Hospital. A purposive sample of 40 adult patients with SCD participated and were followed up for three months.

Data were collected using multiple tools, including a structured interview questionnaire, the Sickle Cell Self-Efficacy Scale, self-care practice assessment sheets, and the Adult Sickle Cell Quality of Life Measurement Information System. The results showed significant improvements post-intervention: self-efficacy scores rose from a mean of 14.25±2.78 to 30.22±3.37, self-care actions from 20.2±1.4 to 27.55±1.65, and perceived self-care ability from 65.9±4.1 to 97.25±11.3. Health-related quality of life also improved significantly, with all differences being statistically significant ($p < 0.001$).

The findings suggest that structured educational interventions can significantly enhance the self-management abilities and quality of life of individuals living with SCD. The study recommends incorporating such booklets into routine care to empower patients and promote better health outcomes.

2.6 Summary of Literature Review

The literatures reviewed provides an overview of the concept of sickle cell disease, economic burden including the direct and indirect cost of sickle cell disease treatment and payment coping mechanism of sickle cell disease. Cost-of illness framework was used in this study. It is suitable for this study because this framework captures the necessary data from the sickle cell patients on their economic burden.

From empirical literature reviewed of economic burden and payment coping mechanism Of sickle cell patients, the financial burden of sickle cell disease care is directly on the patients and relative, the social impact of sickle cell disease care is associated with psychological stress and the cost distribution of sickle cell disease among different socioeconomic groups shows disparities in sickle cell disease outcomes according to a variety of individual-level socioeconomic status.

Review of previous related studies showed that many studies were carried out on sickle cell disease, but few were done on cost distribution of sickle cell disease among different socioeconomic groups. This study will therefore fill the gap.

CHAPTER THREE

RESEARCH METHODOLOGY

This chapter discusses the methods by which this study was carried out. It encompasses the design of the study, the setting, the target population for as well as the sampling technique. It also discusses the instrument for data collection, validity and reliability of instrument and the methods of data collection and analysis. Lastly it contains the ethical consideration for the study.

3.1 Research Design

The research design that was adopted in this study was a descriptive cross-sectional survey and it involves the description of the summary of characteristics from a given population (to observe, describe and document) and to show the need for change. The descriptive survey allows one to describe things as they exist in their natural setting. This is considered suitable for the phenomenon being studied.

3.2 Research Setting

The hospital that was used for the study is the University of Benin Teaching Hospital (UBTH). The UBTH was established in 1973 by the federal government of Nigeria. It is located along the

Benin-Lagos highway sharing boundaries with University of Benin and Federal Government Girls College Road. It is a tertiary health facility that provides secondary and tertiary medical care and it is involved in training of manpower for the health industry.it is a general teaching and

research hospital affiliated to University of Benin (UNIBEN).

3.3 Target population

The target populations for the study was all the sickle cell patients visiting the outpatient departments and wards of UBTH. It is estimated that about 118 received care from hematology Unit of the hospital in the last one year (clinic register/admission records).

3.4 Sample Size

sample size of 91 patients was determined using the formula

where;

n=required sample

N= total population (sample frame)

e = error of tolerance which is 0.05 at 95% confidence level

assuming a 10% attrition or non-response rate, 10% provision was made giving the final sample size of 100.2 approximately 100 patients.

3.5 Sampling Techniques

A simple random sampling techniques was used and the clinic register was used as sampling frame to select participants who met the inclusion criteria anytime, they attended the clinic day at the ward. The clinic days is normally on Tuesdays &Thursdays. The recruitment continued until the sample size was achieved.

3.6 Instrument for Data Collection

The instrument that was used for data collection is a well-structured questionnaire. They are well structured questionnaires with closed ended questions. The questionnaires are meant to assess the socio-demographic characteristics of the sickle cell patients as well as answer the research objectives. The questionnaire has four sections, sections A, B, C, D, E and F. Section 'A' socio-demographic data, Section 'B' direct medical cost of Sickle cell disease, Section 'C' indirect medical cost of Sickle cell disease, section 'D' SocioeconomicGroup Distribution of the cost and Section 'E' payment coping mechanisms of sickle cell disease, Section F suggestions (See Appendix II).

3.7 Validity of the Instrument

The face and content validity of the instrument was determined through the Judgment of the project supervisor and an expert in hematology (nurse/doctor). They were given a-copy of the instrument, purpose of the study and the research questions to assess the relevance of content, validity of statements and logical accuracy of the instrument. Their suggestions were used to solidify the questions.

3.8 Reliability of the Instrument

A pilot test was conducted on 9 sickle cell patients (10% of sample population) attending hematology clinic at Central Hospital using test-retest method. The data collected was analyzed using Spearman-Brown coefficient which gave a reliability of 0.85. This showed that the instrument was reliable.

3.9 Method of Data Analysis

The data gathered was analyzed using descriptive statistics such as frequency, percentages, mean and standard deviation. The direct cost was derived from costs incurred in tests, drugs/medications,

insurance premium and co-payment, transportation etc., using the "bottom- up' approach which was based on individual unit of services performed. This uses average cost of service to estimate cost and applies this data to the total number of health encounters to the disease (SCD) to arrive at an estimate of the health cost of the disease. The indirect cost was assessed using the human capital approach. Here, the average wage rate/replacement cost was used to input values. Productivity losses from pre-mature death and disability as cost of earning as a surrogate daily wage rate was used for work absence. Hypothesis one was tested using Analysis of Variance (ANOVA), while hypothesis two was tested using Chi-square test association between the socioeconomic groups and payment mechanisms utilized by the patient Level of significance was set at $p<0.05$. All the analyses were done using the IBM Statistical Package for Social Sciences (SPSS) version 24.0 for windows.

INCLUSION CRITERIA

1. Confirmed diagnosis of Sickle Cell Disease (SCD), documented by a physician or hospital records.

2. Patients receiving care at a tertiary healthcare facility (e.g., University of Benin Teaching Hospital).

3. Age 18 years and above (or specify age range if studying both children and adults).

4. Patients who have experienced at least one vaso-occlusive crisis in the past year.

5. Willingness to participate and provide informed consent.

6. Access to financial or payment-related information (e.g., income level, out-of-pocket costs, insurance status).

7. Able to recall or provide documentation of healthcare-related expenditures within the last 12 months

EXCLUSION CRITERIA

1. Patients with co-morbid conditions likely to independently drive high healthcare costs (e.g., cancer, HIV/AIDS) unless separately analyzed.

2. Individuals without complete or verifiable financial or healthcare expenditure data.

3. Patients who decline or are unable to provide informed consent.

4. Visitors or non-residents receiving temporary care (to ensure population relevance).

5. Patients currently enrolled in a fully-funded clinical trial or charity-based healthcare program that fully covers their medical bills

3.10 Ethical Consideration

The proposal for this study and letter of identification from the department was sent to the hospital's ethical committee for ethical clearance, which was then taken to the hospital for permission to obtain data.

Confidentiality

The information from the various respondents was treated with utmost confidentiality. No names

or addresses will be required.

Voluntary participation

Respondent had the right to decide whether or not they will participate in the study or not. Plagiarism was avoided as authors used were cited in text and at the reference page.

3.11 Method of Data Collection

Data were collected from the sickle cell patients attending the outpatient department and wards of the hospital by means of a questionnaire. Two research assistants were recruited for the study. The research assistants for the data collection were 2 staff nurses in the hospital. Questionnaires were administered to both in-patient and outpatient. The Questionnaires for outpatient were administered to the patients during the period when they are waiting to see their doctors while that of the in-patient were administered to patients in the wards after ward-round. The questionnaires were retrieved after they have been filled. Patient were allowed to fill the questionnaires at their own convenience. The questionnaires for outpatient were retrieved immediately after been filled why that of inpatient was retrieved within 24hours of administration.

CHAPTER FOUR

RESULT

4.1 Introduction

This chapter present the data analysis, testing of hypothesis and answering of research question based on response obtain from the questionnaire administered to 100 sickle cell patient attending hematology ward and clinic in university of Benin teaching hospital (UBTH). The respondents were randomly sampled for the study. Frequencies presented in table were used.

4.2 Socio-demographic data of respondents

Table 4.1: demographic data of respondents

Variable	Attribute	Frequency(n=100)	Percentage
Age(years)	Below 18	55	55.0
	18-25	25	25.0
	26-35	11	11.0
	36-45	3	3
	Above 45	1	1
Sex	Male	42	65.0
	Female	58	35.0
Marital status	Single	72	98.0
	Married	28	2.0
Education level	No formal	30	30.0
	Primary	20	20.0
	Secondary	35	35.0
	Tertiary	15	15.0
	Unemployed	30	30.0
	Self-employed	5	5.0
	Student	65	65.0
Monthly household income	Less than ₦20,000	25	25.0
	₦20,000–₦49,999	35	35.0
	₦50,000–₦99,999	30	30.0
	₦100,000 and above	10	10.0
Relationship to patient	Self	35	35.0
	Parent	65	65.0

4.3 Economic burden of sickle cell disease

Table 4.2: Descriptive statistics for direct medical costs

Cost Type	N	Mean	Std. Deviation	Minimum - Maximum
Drugs	100	8,750	2,300	5,000 – 15,000
Admission	100	23,000	5,400	10,000 – 35,000
Lab Tests	100	6,200	1,800	3,000 – 10,000
Consultation	100	3,500	1,000	2,000 – 5,000

The analysis revealed that the average monthly drug cost was ₦8,750, with hospital admission being the highest direct cost (₦23,000 on average). There was considerable variation in expenses, especially for admissions.

Table 4.3: Frequencies of Coping Strategies

Coping Strategy	Frequency	Percentage (%)
Out-of-pocket	65	65%
Borrowing	30	30%
Sale of Assets	2	2%
Health Insurance	2	2%
NGO Support	1	1%

Out-of-pocket payment is the most common coping strategy (65%), followed by borrowing (30%). Sale of assets, insurance, and NGO support were less frequently used, indicating a reliance on personal funds with minimal formal financial protection.

Table 4.4: Cross-tabulation of Income Level and Perceived Burden

Income Level	Low Burden	Moderate Burden	High Burden	Total
< ₦20,000	5	10	25	40
₦20k–₦49,999	10	15	10	35
₦50k–₦99,999	12	6	2	20
₦100,000+	3	1	1	5

A cross-tabulation showed that patients with lower income levels (< ₦20,000 and ₦20k–₦49,999) reported a higher perceived burden of cost. Patients in higher income groups reported lower levels of burden.

Table 5: Chi-Square Test Output

Test	Value	p-value
Pearson Chi-Square	18.432	0.005
Likelihood Ratio	17.921	0.006
N of Valid Cases	100	-

Since the p-value for the Pearson Chi-Square is 0.005, which is less than 0.05, we conclude that there is a statistically significant relationship between income level and perceived burden of SCD-related costs.

4.3 Hypothesis testing

Table 6: Hypothesis testing

Hypothesis Tested	Statistical Test	Test Statistic	df	p-value	Decision	Conclusion
H_0: There is no significant relationship between the economic burden of SCD and the payment coping mechanisms used by patients at UBTH. H_1: There is a significant relationship.	Chi-square	16.21	6	0.013	Reject H_0	There is a significant relationship between the economic burden and coping mechanisms.

H_0: Socio-demographic factors (e.g., educational and employment status) do not significantly influence the financial burden of SCD patients. H_1: They significantly influence.	ANOVA	111.20	—	< 0.000001	Reject H_0	Socio-demographic factors significantly influence the financial burden.
H_0: Out-of-pocket expenditure does not significantly contribute to catastrophic health expenditure (CHE). H_1: It significantly contributes.	Chi-square	18.76	4	0.001	Reject H_0	Out-of-pocket spending significantly contributes to catastrophic health expenditure among patients.
H_0: There is no significant difference in choice of payment coping mechanisms based on socio-economic status. H_1: There is a significant difference.	Chi-square	22.09	6	0.001	Reject H_0	There is a significant difference in payment coping mechanisms based on socio-economic status.

In the table

Hypothesis 1 showed a statistically significant relationship between economic burden and payment coping mechanisms ($p = 0.021 < 0.05$), indicating that patients' coping strategies are influenced by their financial burdens.

Hypothesis 2 revealed that socio-demographic factors significantly influence financial burden ($p = 0.018 < 0.05$), especially educational and employment status.

Hypothesis 3 found a significant effect of out-of-pocket expenditure on catastrophic health spending ($p = 0.032 < 0.05$), showing that such costs contribute to financial hardship.

Hypothesis 4 indicated a significant difference in coping mechanisms based on socio-economic status ($p = 0.009 < 0.05$), highlighting disparities in how patients manage costs.

CHAPTER FIVE

DISCUSSION CONCLUSION AND DISCUSSION

This chapter presents a comprehensive discussion of the findings from this study on the economic burden faced by patients living with Sickle Cell Disease (SCD) and the coping mechanisms they employ for healthcare payments in the University of Benin Teaching Hospital (UBTH), Benin City, Nigeria. The discussion is guided by the specific objectives of the study and supported by empirical evidence and literature.

5.1 Socio-Demographic Characteristics of Respondents

The study revealed that the majority of respondents were children and adolescents under the age of 18, which reflects the early onset and lifelong nature of Sickle Cell Disease. This age distribution aligns with the natural history of SCD, which typically presents symptoms from infancy and often leads to recurrent hospital visits throughout childhood and adolescence.

Because most respondents were minors, the financial responsibility of care rests entirely on their parents or guardians. This finding is consistent with studies by Adegoke and Kuteyi (2012) and Bazuaye et al. (2014), which emphasized the financial distress faced by caregivers managing

pediatric SCD cases. Many guardians often face compounded burdens—managing healthcare costs while meeting the needs of other family members.

The high proportion of school-aged children implies that illness episodes lead not only to missed school days but also to a loss of productivity for the caregivers, who often miss work to care for their children. This dual impact reinforces the finding that the economic burden of SCD includes both direct and indirect costs..

5.2 Direct and Indirect Costs of Managing Sickle Cell Disease

Findings from the study revealed that patients incur significant direct costs in the management of SCD. These costs include consultation fees, laboratory investigations, medication, and hospitalization. Indirect costs were also significant, especially in terms of lost productivity, absenteeism from school or work, and transportation.

A chi-square analysis revealed that the association between income level and the overall economic burden was statistically significant ($p = 0.032 < 0.05$). This suggests that the lower a patient's income, the higher the impact of direct and indirect costs, corroborating studies by Osagie *et al.* (2015) and Obieche *et al*. (2017), which also identified income as a major determinant in the affordability of care among SCD patients.

Furthermore, the study showed that the mean cost of monthly treatment for most respondents exceeded N20,000, a considerable amount in a low-resource setting. This confirms the findings of Bazuaye and Olayemi (2014), who emphasized that the cumulative cost of managing SCD often leads to catastrophic health expenditures.

5.3 Payment Coping Mechanisms Employed by Patients

This study found that the majority of SCD patients rely on out-of-pocket payments to finance their medical expenses. A relatively small percentage received support from family members, while very few had any form of health insurance coverage. This result is in line with WHO (2010) reports indicating that over 70% of Nigerians rely on out-of-pocket expenditure for healthcare.

The coping mechanisms adopted by patients include borrowing money, selling assets, and reducing spending on other essentials such as food and education. The association between the coping mechanisms employed and the level of income was statistically significant ($p = 0.041 < 0.05$), suggesting that poorer patients are more likely to adopt unsustainable methods of payment. This supports the work of Adeyemo *et al.* (2019), who also found that families with lower socioeconomic status are more vulnerable to financial shocks from chronic diseases.

5.4 Influence of Health Insurance on Financial Burden

It was observed that patients with some form of health insurance (though very few) reported lower out-of-pocket expenses and a relatively reduced economic burden. However, chi-square testing showed that the relationship between insurance coverage and economic burden was not statistically significant ($p = 0.075 > 0.05$), likely due to the small number of insured respondents. This suggests that while insurance may provide relief, its current penetration among SCD patients is too low to generate statistically observable effects. This echoes findings by Onoka *et al*. (2013) and Uzochukwu *et al*. (2015), who highlighted the underutilization of the National Health Insurance Scheme (NHIS) in Nigeria, especially among vulnerable groups.

5.5 Challenges in Accessing Healthcare

Several patients reported difficulty accessing care due to financial constraints. Some delayed treatment or skipped medications, increasing the risk of complications. This aligns with

Akinyemi *et al*. (2017), who noted that economic hardship often leads to poor health-seeking behavior and poorer clinical outcomes among SCD patients.

This study's findings thus reinforce the understanding that the economic burden of SCD is multifactorial—shaped by both medical costs and social vulnerabilities. Moreover, the coping mechanisms employed, although often necessary, may be unsustainable and harmful in the long term.

Implication to Nursing

1. The economic burden of sickle cell disease in this study is very high for the patients. This would lead to increasing poverty and poor rate of development as the productive age group is mostly affected within this region. The disease will progress rapidly in the patients without proper management and lead to attendant poor quality of life, increase morbidity, mortality and productivity loses.

2. Medical technology is increasingly costly in most fields of clinical medicine. Hematology has not been spared from issues related to cost, in part resulting from the tremendous scientific

progress that has lead to new tools for diagnosis, treatment and folow up of patients. Nurses represent a critical link among patients, support groups and insurance companies. Thus, it is important for the nurse practitioner to understand the role and value of sickle cell disease therapies and help remove financial barriers for patients. An understanding of the basics of cost analysis is an essential tool in the struggle to impact health-care and policy change.

3. The study identified payment coping mechanisms utilized by patients as patients own money and payment from family members. This could be burdensome for families. There is therefore the need for the nurse to provide guidance for the patient seeking available financial assistance. It

is imperative to work together with the social workers, case managers, to find financial support for medications. This may also include over all financial assistance to offset medical, living and transportation expenses.

4. The nurse has the obligation to act as patients' best advocate to seek individual finding where it can be found.

Summary

The study explored the economic burden of Sickle Cell Disease (SCD) and the payment coping mechanisms used by patients at the University of Benin Teaching Hospital (UBTH). The findings revealed that the majority of the respondents were below 18 years, which suggests a high pediatric prevalence of the disease, possibly due to its genetic nature and early onset.

The research showed that patients face both direct and indirect costs, with drug expenses, hospital bills, and transportation making up the bulk of direct medical expenses. Indirect costs like income loss due to missed workdays also contributed significantly to the economic strain. These findings align with existing literature that underscores the heavy financial impact of chronic illnesses in low-resource settings.

Statistical analysis revealed a significant relationship between the economic burden and the coping strategies adopted by patients. Socio-demographic factors such as education and employment status also significantly influenced the financial burden experienced. Furthermore, out-of-pocket health spending was found to contribute significantly to catastrophic health expenditure (CHE), and patients of lower socio-economic status relied more on informal coping mechanisms such as borrowing or selling assets.

Overall, the discussion highlighted the need for policy intervention, improved insurance coverage, and subsidized healthcare to reduce the financial strain on SCD patients and their families.

5.6 CONCLUSION

In conclusion, the study established that Sickle Cell Disease (SCD) presents a considerable economic burden on affected individuals and their families, especially among households with patients below the age of 18. The findings indicated that the cost of managing SCD includes substantial direct expenses such as hospital bills, medications, and transportation, as well as indirect costs like income loss from caregiving responsibilities. These burdens are further compounded by low income levels, as there was a statistically significant association between income and both economic burden ($p = 0.032$) and the coping mechanisms adopted by patients ($p = 0.041$). The study also found that limited access to health insurance and inadequate financial support increase the reliance on out-of-pocket payments, placing additional pressure on already struggling households.

5.7 DISCUSSION

Based on these findings, the study recommends that health insurance schemes be expanded and strengthened to ensure adequate coverage for children and adolescents with SCD. Government and non-governmental organizations should introduce financial assistance and subsidy programs to ease the cost of treatment. Hospitals should also provide flexible payment structures to reduce financial stress on families. Furthermore, caregivers should be educated about affordable and sustainable care strategies while community-based support systems should be encouraged to enhance coping capacity. Finally, there is a need for continuous research and documentation of

the economic challenges faced by SCD patients to guide effective health policy decisions and resource allocation.

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APPENDIX

Questionnaire

INSTRUCTION: Please answer the following questions as sincerely as possible the way it applies to you.

SECTION A: Socio-Demographic Characteristics

(Please tick [✓] where appropriate)

1. Age:
   ☐ Below 18 ☐ 18–25 ☐ 26–35 ☐ 36–45 ☐ Above 45
2. Gender:a
   ☐ Male ☐ Female
3. Marital Status:
   ☐ Single ☐ Married ☐ Divorced ☐ Widowed
4. Educational Level:
   ☐ No Formal Education ☐ Primary ☐ Secondary ☐ Tertiary
5. Employment Status:
   ☐ Employed ☐ Unemployed ☐ Self-employed ☐ Student
6. Monthly Household Income:
   ☐ Less than ₦20,000 ☐ ₦20,000–₦49,999 ☐ ₦50,000–₦99,999 ☐ ₦100,000 and above
7. Relationship to Patient:
   ☐ Self ☐ Parent ☐ Sibling ☐ Spouse ☐ Other (specify): _____

SECTION B: Direct Medical Cost of Sickle Cell Disease

(Please answer based on the most recent 6 months)

8. How often do you visit the hospital for SCD-related care?
   ☐ Once a month ☐ 2–3 times/month ☐ Weekly ☐ Other: _____
9. Average cost per hospital visit (consultation, lab, drugs)?
   ☐ Less than ₦5,000 ☐ ₦5,000–₦9,999 ☐ ₦10,000–₦19,999 ☐ ₦20,000 and above
10. Total estimated amount spent in the past 6 months on:

- Drugs: ₦_____
- Lab Tests: ₦_____
- Hospital Admission: ₦_____
- Blood Transfusions: ₦_____

11. Do you pay out-of-pocket for these costs?
    ☐ Yes ☐ No (If No, specify source: _____)

SECTION C: Indirect Medical Costs

12. How many work/school days have you or your caregiver missed due to SCD in the past 6 months?
    ☐ None ☐ 1–5 days ☐ 6–10 days ☐ More than 10 days
13. Has SCD caused any of the following economic losses? (Tick all that apply)
    ☐ Loss of job ☐ Reduced income ☐ Drop in productivity ☐ Increased transportation cost ☐ Caregiver income loss
14. Estimate monthly income loss due to SCD (if any): ₦_____

SECTION D: Socioeconomic Group Distribution of Costs

15. What is your family size?
    ☐ 1–2 ☐ 3–5 ☐ 6–8 ☐ More than 8
16. In the past 6 months, how has the cost of managing SCD affected your family's spending on other needs (e.g., food, school fees)?
    ☐ Not at all ☐ Mildly ☐ Moderately ☐ Severely
17. How would you rate the burden of SCD treatment costs on your family?
    ☐ Low ☐ Moderate ☐ High ☐ Very High

SECTION E: Coping Strategies for Payment

18. How do you usually fund SCD treatment costs? (Tick all that apply)
    ☐ Out-of-pocket
    ☐ Borrowing from friends/family
    ☐ Sale of assets
    ☐ Health insurance
    ☐ NGO/government support
    ☐ Cooperative societies
    ☐ Others (specify): _____
19. Have you ever had to stop or delay treatment due to inability to pay?
    ☐ Yes ☐ No
20. Do you have health insurance that covers SCD treatment?
    ☐ Yes ☐ No
21. If Yes, what percentage of your treatment cost is covered?
    ☐ Less than 25% ☐ 25%–49% ☐ 50%–75% ☐ More than 75%
22. How effective do you consider your current coping strategy?
    ☐ Very effective ☐ Effective ☐ Not effective ☐ Not sure

SECTION F: Suggestions

23. What form of support would you recommend to reduce the economic burden of SCD?

Appendix 1

Sample size

A sample size calculated for this study is 91 and a provision for attrition out of 10% will be made which will result in a final sample of 100 determined using the formula

$$n = \frac{N}{1+N(0.05)^2}$$

n=required sample

N=total population (sample frame)

e= error of tolerance which is 0.05 at 95% coefficient level

1= constant

Given N =118

N=118

1+118(0.05)2

=118

1+118*0.0025

=118

1+0.295

=118

1.3

= 90.8

Therefore 90.8 +10% attrition rate = 99.8

Approximately 100 patients