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HOUSEHOLD COST OF SICKLE CELL DISEASE AMONG PATIENTS OF SICKLE CELL CLINIC, TEMA GENERAL HOSPITAL, GREATER ACCRA REGION

BY

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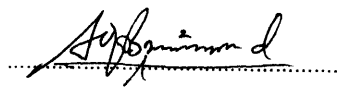
DECLARATION

I hereby declare that apart from precise references which have been accordingly acknowledged, this submission is my own work towards my MPH dissertation and that, to the best of my knowledge, it contains no material previously published by another person nor material which has been accepted for the award of any other degree of the University or elsewhere.

Lawrence Ofori-Boadu



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12/10/17

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DEDICATION

I dedicate this thesis to the staff at Sickle Cell Clinics in Ghana and all sickle cell disease clients at these clinics. I sincerely hope that this work would help open accesses for policy development on improved and comprehensive coverage of care for all people living with sickle cell disease. To God be the glory.

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ABSTRACT

Introduction: Sickle Cell Disease (SCD) is progressively gaining the recognition that was once ignored despite its increasingly higher prevalence of 2-3 % in the World and Ghana. The high household cost of care associated with this chronic disorder leads to stress not only on people living with the disease but their households-parents, caretakers and families. The purpose of this study was to determine the household cost of sickle cell disease among patients of Sickle Cell Clinic (SCC) at Tema General Hospital in the Greater Accra Region of Ghana.

Method: The study was a cross-sectional cost of illness study designed to quantitatively determine the direct, indirect and the intangible cost of 78 SCD patients who attended sickle cell clinic at Tema General Hospital over a period of 12 months. The household cost was the summations of direct and indirect cost incurred when SCD patients assesses healthcare. Sensitivity analysis was used to test the robustness of the results. Likert's scale was used to assess intangible cost of SCD. Composite intangible cost of SCD was estimated.

Results: The mean age of the SCD population was 20 years. The total cost of SCD at SCC estimated was GHS 97,615.20 with direct cost being 88% and indirect cost 12%. The mean monthly household cost of SCD was estimated at GHS 1,251.48 (SD 833.20). The total composite intangible cost indicates that low intangible cost accounted for 52% (41), moderate intangible cost 45% (35) and high intangible cost 3% (2). Functional loss in terms of intangible cost of SCD patients with episodes of pain was the highest (7.36) and stigmatization (3.19) the lowest.

Conclusion: The household cost of SCD at the SCC is very high. There is the need to intensify education on SCD to bring the majority of SCD patients in the community to the clinic. This will help draw policy maker's attention for a complete and all-inclusive cost care plan (NHIS)

for SCD patient to further reduce cost. As indirect cost minimizes, more SCD patients will attend clinic regularly and emotional sufferings associated with SCD will be under control.

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LIST OF ABBREVIATION

BD:	Bipolar Disorder
COI:	Cost of illness
CVM:	Contingent Valuation Method
ERC:	Ethical Review Committee
GHS:	Ghana Health Service
HCM:	Human Capital Method
HbSC:	Heterozygous hemoglobin SC
HbSS:	Homozygous hemoglobin SS Sickle cell
NHIS:	National Health Insurance Scheme
OPD:	Out of Patient
PI:	Principal Investigator
SCC:	Sickle Cell Clinic
SCA:	Sickle Cell Anemia
SCD:	Sickle Cell Disease
TGH:	Tema General Hospital
TMA:	Tema Metropolitan Assembly
WHO:	World Health Organization
WTP:	Willingness to Pay

CHAPTER ONE

1.0 INTRODUCTION

1.1 BACKGROUND

Sickle cell disease (SCD) is a long-lasting hereditary disease affecting many people worldwide and this term is used to embracing all disease states where hemoglobin S (Hb S) or beta-S gene is present (Dennis-Antwi, Dyson, & Ohene-Frempong, 2008). The formation of hemoglobin S is due to mutation that results in exchange of the amino acid valine for glutamic acid at the 6th place of the beta globin chain, producing sickle cell development (Ingram VM, 1956). It is identified that about 150,000 to 300,000 offspring are born every year with the disorder in Africa (Sadarangani, Makani, Komba, 2009). The sequence of the disease differs with some kids showing extreme signs needful of regular hospital attendance (Olatunya, Ogundare, Fadare, Oluwayemi1, 2015). Sickle cell disease gives rise to a range of other body weakness that can cause death. Over 2 million United State residents are estimated to be carrying either a single of both defective hemoglobin gene. Many of those affected are of African origin (Sebil, 2011).It is projected that about 100,000 Americans have SCD (Grosse, Odame, Atrash, Amendah, Piel, Williams, 2011). Although SCD is associated with major morbidity, currently more than 90 percent of children with SCD in the United States of America and the United Kingdom survive into adulthood (Woods et al., 2016) However, their lifespan remains shortened by two or three decades compared to the general population (Powars, 2005).The most common SCD genotypes comprise hemoglobin SS (HbSS) and the compound heterozygous conditions hemoglobin S β 0-thalassemia (HbS β 0-thalassemia), hemoglobin S β+-thalassemia (HbS β +-thalassemia), and hemoglobin SC disease (HbSC). HbSS and HbS β 0-thalassemia are clinically very similar and usually referred to as Sickle Cell Anemia (SCA). These genotypes are associated with the most severe clinical manifestations (Ohene-Frempong

et al., 2008). The progression of the illness differs extensively with some young ones showing plain displays necessitating recurrent hospital visits (Sadaragani et al., 2009). It is documented that SCD has the propensity to drain the funds of families in emerging countries where the high level of poverty and unfair distribution of wealth and incomes exist (Olatunya et al, 2015). Even though some studies on household health costs exist in Ghana, only a limited number were on SCD in spite of its long-lasting nature and high occurrence in the country (Kyerewaa Edwin, Edwin, & Etwire, 2011). Sebil, (2011) conducted a study on social cost of sickle cell disease within three institutions –Korle Bu Teaching Hospital, Sunyani Regional Hospital and Komfo Anokye Teaching Hospital- and found the total cost per annum to be GHS 2,724.00 (US$ 1,816.00). Scrutinizing the current household cost of SCD will help direct opinion leaders and law makers outline appropriate policies to lessen the household cost and guide all key stakeholders in their best of cost-effective actions in taking care of people with the disease condition. It will also help the families to strategize for the health care requirements of their relatives with SCD.

1.2 Problem Statement

Sickle cell disease remains an increasing problem among clients and households worldwide (Woods et al., 2016). Despite reductions in mortality associated with early screening, sickle cell disease is still associated with high consumption of medical resources (Grosse et al., 2011). Many cost of care research done among patients with SCD have concentrated on one or two types of care, such as inpatient hospitalizations and physician visit (Kauf et al., 2009). Factors contributing to this burden include socioeconomic status of clients and their caretakers; and the disease complications such as recurrent anemia with blood transfusions, painful joints, acute chest syndrome, osteoarthritis and stroke. Cost of accessing and seeking health care has always been a burden to patients of SCD due to the chronicity of the disease condition (Ohene-

Frempong et al., 2008). Majority of families or clients may indirectly lose money from income due to loss of hours or days spent at hospitals for consultations or admission. Frequent visits of SCD clients incur loss of valuable time and money which may psychologically affect households. Quality of life is affected since patients tend to spend all productive time at the hospital without attending to social, family and spiritual engagements. Probing the economic problem of SCD on families will help to guide opinion leaders outline suitable plans to do away with the challenge and guide health workers in their choice of measures cost effective way of taking care of children with the disease condition (Kauf et al., 2009). It will also help the families to plan for the health needs of their relatives with SCD (Olatunya et al., 2015). Studies on household cost of sickle cell disease (SCD) are small in spite of its increasing numbers among newborns in emerging typical African countries (Ngolet et al., 2016).

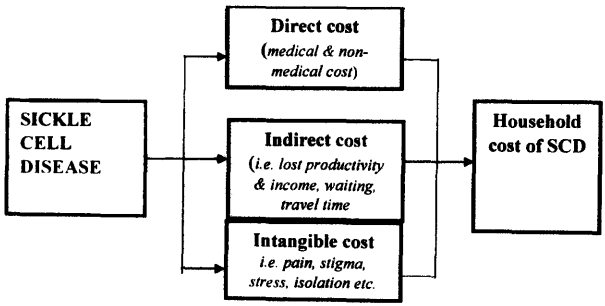
The main focus of this study is to clearly document total healthcare cost and the intangible cost of SCD households at the Sickle Cell Clinics. This may help SCD households to plan appropriately to minimize losses and be able to support the health needs of their relatives with this long-lasting condition.

1.3 Conceptual Framework

A sickle cell disease patient habitually needs recurrent hospital appointments for the management of this long-lasting illnesses requiring families to pay for the resources (Kauf et al., 2009). The long lasting nature of condition with its associated crises has an associated household cost. From Figure 1 the household cost of SCD arise from various cost components grouped into three main cost. A direct cost is either medical or non-medical cost. A direct medical cost include cost of medical folder, consultation, medication, laboratory investigations, X-rays, blood transfusion processing cost. Direct non-medical cost of SCD include

transportation and feeding cost whilst seeking health care. Many clients miss clinical review due to inability to afford transportation fees (Kauf et al., 2009). Indirect cost of SCD are absenteeism from work and loss of wages –all contributing to the total loss of productivity when SCD patient seeks health care. Intangible cost of SCD is mostly ignored by many but contribute to substantial loss on the part of the SCD patient. For the purposes of this study, pain, stigmatization, social isolation, self-esteem, and stress constitute intangible cost of SCD. Descriptive analysis was employed on intangible cost evaluation, and patients assessment on level of pains, stigma, social isolation, stress and low self-esteem were used to estimate the intangible cost.

Figure 1 A conceptual framework of Household cost of SCD



1.4 Justification

Despite the high consumption and usage of medical assets, sickle cell disease (SCD) patients pay the full cost of healthcare, including other indirectly associated and intangible cost that are not broadly documented (Kauf et al, 2009). Sickle cell disease clients require peculiar attention and this necessitate a wide-ranging of care plan categorized into medical and non-medical needs. To prioritize well-being for persons with SCD, a total approach that involves qualified medical and non-medical staff that meets the total needs of persons with SCD is desirable (Dennis-Antwi et al., 2008). Documentation on household cost of SCD is scanty in Ghana (Sebil, 2011). The focus of this research is to determine household cost of SCD patients at Sickle cell clinic in the Tema General Hospital. Findings from this study will help identify, quantify and update household cost of SCD at sickle cell clinics in Ghana.

1.5 OBJECTIVES

1.5.1 General Objective

The main objective was to determine household cost of SCD among patients of sickle cell clinic at Tema General Hospital, Greater Accra Region.

1.5.2 Specific Objectives

The specific objectives were:

(1) To estimate direct cost of SCD on households.

(2) To estimate indirect cost of SCD on households.

(3) To assess the intangible cost associated with SCD to households.

CHAPTER TWO

2.0 LITERATURE REVIEW

2.1 Introduction

Sickle Cell Disease (SCD) is a long-lasting medical conditions associated with the formation of hemoglobin S (HbS) and another irregular hemoglobin. SCD affects millions of people worldwide predominantly black people in Africa, Europe, America, Asia, Arabs and those of Asian ancestry (Ohaeri & Shokunbi, 2002). Sickle-cell disease is the commonest hemoglobin disorder, mostly in sub-Saharan Africa where 75% of 300, 000 babies are born year with hemoglobin disorders. Homozygous form of Sickle Cell Disease (SCD) is related with very high child death. (Olatunya et al., 2015). WHO estimates that 70% of deaths are avoidable by putting in place preventive measures which encompass early diagnosis, information, education, and prophylaxis of infections. The non -accessibility to care and medicines is linked to financial challenges (Grosse et al., 2011). The genes associated with HbSS and HbSC forms the largest population in people living with SCD and form the population base of this research (Kyerewaa Edwin et al., 2011). Studies in the sub region indicate a population of 30% for people carrying one abnormal gene and 2% of children born in Ghana have SCD (Ohene-Frimpong et al., 2008). It is therefore very important to assess and quantify how much it cost households or clients with SCD in the event of accessing health care in hospitals within specified period over their lifetime.

2.2 Disease Burden of sickle cell disease

Sickle cell disease is a chronic illness and more often than not, sufferers go through problems such as recurrent pain, infections, and sometimes the affected persons may suffer stroke (de Montalembert, 2008). A detailed medical care package is well planned and managed by group of medical expects with knowledge in blood related diseases. It emphases on delivering care to

children and adults and other specialized care as and when it is required. (Grosse et al., 2011). Pain management is based on staying away from things that can provoke painful crises, a major antecedent of discomfort. Commonly used ones analgesics include paracetamol and ibuprofen. Adequate water intake is key to successful and early alleviation of pain. Haemotransfusion and blood products are crucial as SCD patients normally have higher requirement for blood due to persistent destruction with superimposed hyper-hemolytic episodes. Nutrition is important and supports immune function and proper development. Folic acid and vitamin supplements are useful adjuncts (Grosse et al., 2011). Very key factor manipulating the quality of life in SCD patients is self-care. Self-care involvements encourage effective management approaches to ease signs and attain a healthier quality of life. Many patients with SCD successfully manage their disease in the outpatient setting without the need for frequent health care utilization and do not require frequent hospitalizations and/or emergency department visits. Potentially, much could be learned from this group in terms of living productively with SCD. Sickle cell clinics have been very useful to clients and families of SCD as it creates a call point to addressing their special needs. Sickle cell clinic provides both physical and psychological needs of households and clients regularly reducing morbidity significantly (Osei-akoto, Akoto, Brobby, & Baffoe-bonnie, n.d.). One key important strategy recognized includes psychological, vocation choice and achievement issues, diet, sponsorship, information, suitable and adequate physical exercise and then good medicine. Personal care, the most vital aspect of self-management, is a sense of being aware of certain behaviors that promote or negatively affect health. Putting all these approaches together, SCD patients are proficient of attaining a near-normal life period with a realistic quality of life.

2.3 Economic burden of SCD

Sickle cell disease patients when in crises condition or in pain are always sent to the health care centers where they are either treated and discharged or admitted to ward (Sebil, 2011). A child with SCD often requires frequent hospital visits for the treatment of chronic illnesses necessitating households to bear financial costs (Olatunya et al., 2015). The continual and routine schedule of hospital review with its associated cost has become a burden to many households and clients. In many instances financial challenges lead to absenteeism from clinics, neglect of regular attendant or hospital visits, resulting to complications of SCD (Sebil, 2011). In spite of decreases in illness and death related with screening, SCD continue to be associated with high utilization of medical resources. Many cost of care studies conducted among patients with SCD have focused on one or two types of care, such as inpatient hospitalizations and physician visits (Kauf et al, 2009). Due to the reduced life-expectancy linked with SCD and varying follow up schedules, most available cost material is concentrated on children. Also, few reports describe the cost of care for people with SCD directly associated to SCD. Thus, although there is enough evidence to justify investments in screening, prophylaxis, and treatment for African children with SCD, better data are needed to estimate their cost effectiveness (Grosse et al., 2011).

The United Nation resolved in 2008, the recognition of SCD as a public health issue, and urged all member states to raise awareness of SCD on June 19 of each year (Grosse et al., 2011). In spite of all efforts to call for worldwide efforts to bring the disease to light, comparatively only few inputs and recognition to evaluating the problem of SCD and how to reduce it in Africa, where about 85% of children with SCD are born (Grosse et al., 2011). The WHO Regional Office for Africa suggested a sickle cell disease policy in acknowledgment that the condition is significant cause of child death in many developing countries. Parents are unable to go to work

putting pressure and affecting their earnings which on the other hand are continuously being drained from direct cost of purchasing medicines and periodic laboratory tests (Kauf et al., 2009) Information gathered on persons with sickle cell disease in Florida medical aid program to estimate all the cost components related to SCD on age whilst accessing care. Out of 4,294 patient recruited, total costs of health increased with age, from $892 to $2,562 per patient-month in the respective children and the aged groups. Average cost per patient-month was $1,389. Overall, 51.8% of care was directly related to SCD, the majority of which (80.5%) was associated with inpatient hospitalizations. Particularly, indirect costs were considerably higher compared to that of the whole United States populace. The fallouts suggest a lifetime cost of care be an average of $460,151 per patient with SCD (Kauf, Coates, Huazhi, Mody-Patel, & Hartzema, 2009). Interventions such as wide coverage and effective insurance policy supports regular clinic attendance of patients and prevent SCD complications and admissions, reducing the substantial economic drain of the disease.

2.4 Social Burden of Sickle cell disease

Social issues such as stigma has been noted to be commonly associated with chronic disease (Biljana, Serette, Tim , 2007). Sickle cell disease patients have to endure some psychological pain and rejection from society such as isolation, outcast, condemnation, disgrace and name calling, so deserves our special consideration in all matters of human endeavor (Aisiku, Barakat, Lutz, Smith-Whity, 2007). Socially, both clients and parents are unable to attend family and societal gatherings due to frequent illness and hospital visits. Neighbors see you as unfriendly as you persistently deny invitations to social gatherings (Lubkin, Larsen, 2006). Psychologically, clients output are affected when complications with associated loss of function sets in. Stigmatization is a prominent parameters that affect a client's social life. The

perception of patients with SCD are that they are treated unfairly/differently from other inpatient clients-discrimination from staff, lack of attention and no respect (Lubkin et al, 2006). Pain, fear, functional limitation and stigmatization contribute to loss of output on the part of the person and hence a real loss.

2.5 Determination of Household cost

2.5.1 Cost of Illness studies

Cost of Illness COI) is defined as the value of the supply that are used or inevitable as a result of a health problem (Tarricone, 2006). These encompass the aggregation of all expenditures on regular schedule visits, the outpatient and inpatient treatment including investigations, and admissions. Two methods of direct costing have been described, micro-costing and gross-costing, which are quite identical in course to bottom-up and top-down approaches respectively. The more preferred and precise micro-costing involves summing up each single cost component that has contributed to the provision of the service, while gross-cost is obtained by dividing total costs of the service unit by the total number of services produced in a period of time (Tarricone, 2006). While direct cost estimation involves direct recording of actual monetary resources, indirect and intangible cost estimation may on the other hand be associated with more difficultly and uncertainty.

Human Capital Method (HCM): This is the method adopted in most COI studies (Jo, 2014), having been used at least since the 17th century (Tarricone, 2006). This method tries to put monetary value to an individual on the premise that society would have benefitted from the productivity of the individual should he/she have continued to live in full health. It thus assumes or calculates the human capital per their current socio-economic status. (Jo, 2014). Using the human capital method to estimate the cost of accidents in Thailand and Vietnam, it

came out that, loss of output and human cost was a heavy burden on the economies of both countries (Kudebong, Wurapa, Nonvignon, Norman, Awoonor-Williams, Aikins 2011). The human capital approach seems well suited for developing countries and as such, will be employed in this study to estimate indirect cost even though it can also be used to estimate intangible cost.

Willingness to pay method (WTP): Willingness-to-pay method (WTP) estimates the amount of money people affected would pay to avoid having that disease condition .Thus, willingness to pay (WTP) method estimates productivity loss by attempting to measure the amount that an individual is eager to pay in order to reduce or eliminate the morbidity associated with the illness. Several methods have been suggested to achieve these, however the commonest is the contingency valuation method (CVM). This method has generally been used in the estimation of indirect cost (Jo, 2014). The household cost will be the sum total of the direct, indirect and intangible cost of SCD. Willingness-to-pay method is commonly used in developed countries but difficult to use in developing countries because extensive surveys to obtain perceived risk and acceptable payment to avoid a given hypothetical level of risk is difficult to conduct (Kudebong et al., 2011). COI is over time become a standardized gauge for policy makers. Cost-of-care estimates are important inputs to health care planning, research prioritization, and the economic evaluation of new SCD treatment strategies

2.5.2 Direct cost of Sickle Cell Disease:

Direct cost refers to the resource consumption in the provision of help intervention (Browner et al., 2013). Direct costs of a disease are those generated by the resources used in treating or coping with the disease, including expenditures for medical care and the treatment of the illness. Direct cost usually represent the cost associated with medical resource utilization which include the consumption of in-patient, out-patient and pharmaceutical services within the

health care delivery system (Boccuzzi, 2003). Direct costs include all resources associated with the provision of an intervention or treatment for an illness. As direct costs are easily and accurately identified and estimated this cost component has been included in many cost studies (Feng et al, 2008). Direct costs can be understood as the additional use of resources of a certain value that is directly associated with providing the treatment. This will be estimated by summing all the cost incurred by patients due to the SC disease and its related complications on medical goods and services e.g. consultation, diagnostic test and medication. Direct non-medical cost is estimated by summing non-medical goods and services e.g. travelling cost, diet and other subsistent cost due to the SCD and its related complications and comorbidities (Amon, 2016).

2.5.3 Indirect cost of Sickle Cell Disease

Indirect cost addresses the potential resources that are lost as a results a SCD. They include societal cost morbidity, disability, and premature mortality. Indirect cost represents the impact-present and future, of opportunity losses by the individual as a results of consequence of SCD (Sebil, 2011). Indirect costs refer to productivity loss incurred by an illness and are very important in cost of illness studies as they can be substantial (Maclean et al., 1998). Indirect costs are defined as an economy's lost potential on productivity resulting from illness-related absences or impaired performance at the workplace (Anders et al., 2013). The calculation assumes that the wages correspond to the marginal product of labor. In this work, indirect costs not only consisted of the gross income and non-wage labor costs but also include sickness benefits (Amon, 2016). The latter often are not conceived to be indirect costs in related studies and are itemized separately instead. Indirect costs therefore comprise payments in the form of sick pay or sickness benefits that employees who are incapable of working receive during illness, either from the employer or from the health insurance or employers' liability insurance

association. Estimation of indirect cost was based on human capital approach (or income approach). This was estimated output or productivity losses based on total work hours lost and total travel and waiting times SCD household spent at SCC.

2.5.4 Intangible cost of Sickle Cell Disease

An intangible cost of a disease is an unquantifiable cost relating to a recognizable source. Intangible costs are defined as pain and sufferings of patients because of a disease, which are usually measured by using the reduction in quality of life (Belotti, 2003). Stigma and discrimination are so high that SCD may sometimes lose their livelihood and even accommodation (Kyerewaa Edwin et al., 2011). Thus the psychosocial burden of SCD is not insignificant.

2.5.4.1 The Likert's Scale

Rensis Likert developed a scale for measuring assertiveness or character traits. The typical Likert's scale is a 5- or 7-point ordinal scale used by respondents to rank the degree to which one agree or disagree with a statement (Scales & Sullivan, 2013). As a result of difficulty of measuring attitudes, character, and personality traits and the procedure for transferring these qualities into a quantitative measure for data analysis, the scale was developed (Boone *et al.*, 2012). Quantifying stigma, pain, emotional stress suffered by SCD patients and households makes it appropriate to describe them using five dimensions for patients.

Also ordinal scale responses can be rated, but the distance between responses cannot be measured. Thus, the differences between "not at all," a little "moderately," quite a bit "and "extremely" on a frequency response of Likert scale should not necessarily be equal. One cannot assume that the difference between responses is equidistant even though the numbers assigned to those responses remain same. Amoakoh & Aikins, (2013) estimated household

economic costs of Buruli ulcer in Ghana and described the intangible cost suffered by Buruli ulcer patients and their households using the Liket's Scale without quantifying in monetary terms. The study therefore intends using five dimension Liket's scale for SCD patients.

2.5.5 Cost of Illness studies for other chronic diseases

Cost-of-illness (COI) studies provide useful information on the economic burden that diseases imposes on a society. COI studies are key sources of evidence used in economic evaluations. The study aimed to compare general overview of COI studies for other chronic diseases such as Bipolar Disorder (BD) and Buruli Ulcers (BU) and to discuss their cost of care and methodological issues that might potentially influence results. The primary outcome of BD review was the annual cost per BD patient. The review showed that numerous COI studies have been conducted for BD since 1995. However, these studies employed varying methods, which limited the comparability of findings (Jin, Huajie, McCrone, 2015). The commendations provided by the review of BD can be used by those conducting COI studies and those evaluating them, to increase the credibility and reporting of study results. In 2013, annual societal costs per patient with Bipolar Disorder varied from US$1,904 to US$33,090. Production losses made up to 20–94 % of the total societal cost of bipolar disorder, but have only been reported in 30 % of included studies (Woods et al., 2016).

In a retrospective one year cost study on household cost of out-patient treatment of Buruli ulcer in Ghana: a case study of Obom in Ga South by Hannah Brown Amoakoh and Moses Aikins (2013), the total household cost was US$ 35,915.98, of which about 65% was cost suffered by children with a mean cost of US$ 521.04. The mean annual household cost was also US$570.09. The direct cost was 96% of the total cost. Non-medical cost accounted for about 97% of the direct cost with a mean cost of US$529.27. The main cost drivers of the household costs were transportation (78%) and food (12%). Caregivers and adult patients lost a total of

535 productive days seeking care, which gives an indirect cost valued at US$ 1,378.67 with a mean of US$21.88. A total of 365 school days were lost by 19 BU patients (mean, 19.2 days). Functional loss and pain were low, and stigma rated moderate. Most children suffering from BU (84%) were socially isolated.

Cost of illness studies do not control the most appropriate course of action with respect to the disease studied as such studies are not evaluative. Yet provision is made on the extent of the influence of an illness on society or a part of people. COI studies provide a framework and important evidence for cost estimation in economic evaluations. The key procedural points to consider when revising a COI study include analysis, cost component, costing method and sensitivity analysis (Jin et al., 2015). Understanding the costs of an illness helps policy makers to determine which disease condition requires to be addressed urgently by health care and prevention policy. Cost-of-illness studies are capable of pinpointing the financial impact a disease has on public health programs. For employers, such studies can indicate which diseases have an especially large effect on their costs. Cost-of-illness studies gives important information for cost-effectiveness and cost-benefit analyses as well as provide the framework for estimating costs. Lastly, COI is useful in educating, informing and enlightening policy makers in planning, financing as well as relevant in chronic diseases that weigh heavily on health expenditures (Costa *et al.*, 2012). Sensitivity analyses would be conducted to test all important parameters and key assumptions used in COI studies, and a range of possible cost estimates reported.

2.6 Cost of SCD in Ghana

Persons born with SCD have peculiar requirements and necessitate a comprehensive care profile organized into medical and non-medical services, and self- management strategies

(Kyerewa Edwin et al., 2011). To improve health care for people with SCD, a comprehensive technique which includes a well trained professionals with diverse specialty, and a distinct societal provision organization that meets total needs of persons with SCD is desirable. (Dennis-Antwi et al., 2008). Amongst the few studies done in Ghana regarding cost of SCD, Sebil (2011) estimated household cost of SCD as GHS 2,724.00 (US$1,816) per annum. Out of this cost, direct cost constituted GHS 2,280.00 (US$1520) per annum whilst productive loss (indirect cost) per annum was GHS 440.00 (US$293). There were several assumptions in this study since most patients interviewed could not quantify their losses as others were also not formal employees with regular income. Many patients attending sickle cell clinic rely on the National Insurance Health Scheme (NHIS) and out of pocket means to pay for the direct cost of healthcare. Any factor that affects the income of the client will directly affect attendance in seeking medical and non-medical care.

2.7 CONCLUSION

Over the years, SCD has scarcely been regarded as a disease of any public health significance. The health status of children with serious genetic disorders such as SCD has been submerged in statistics on death from the major childhood diseases in Ghana, such as malaria, acute respiratory infections and malnutrition-related conditions (Ohene-Frempong, 2007). Despite the high carrier rate of 30% with 2% of all new borne babies having SCD in Ghana (Dennis-Antwi, JA et al (2008); very little study has been done on household cost of the disease. Cost of illness study studies done on Bipolar disorders (Huajie, 2016) indicates that notwithstanding the many studies on cost of illness, its helpfulness is limited and does not regulate suitable progression of action with respect to disease to study though it provides some outline and indication for cost estimate.

This study will use the HCA to estimate the indirect cost in view of the fact that it seeks to quantify the productivity loss of SCD patients seeking healthcare services. Consequently, the intangible cost will not be quantified but described using Likert's scale. It is therefore foreseen that this cost-of-illness study in respect of SCD patients care will provide a basis for cost profile that will inform caregivers and policy makers in the clear understanding of the disease process and requisite health resources to SCD clinics making them more accessible and affordable.

CHAPTER THREE

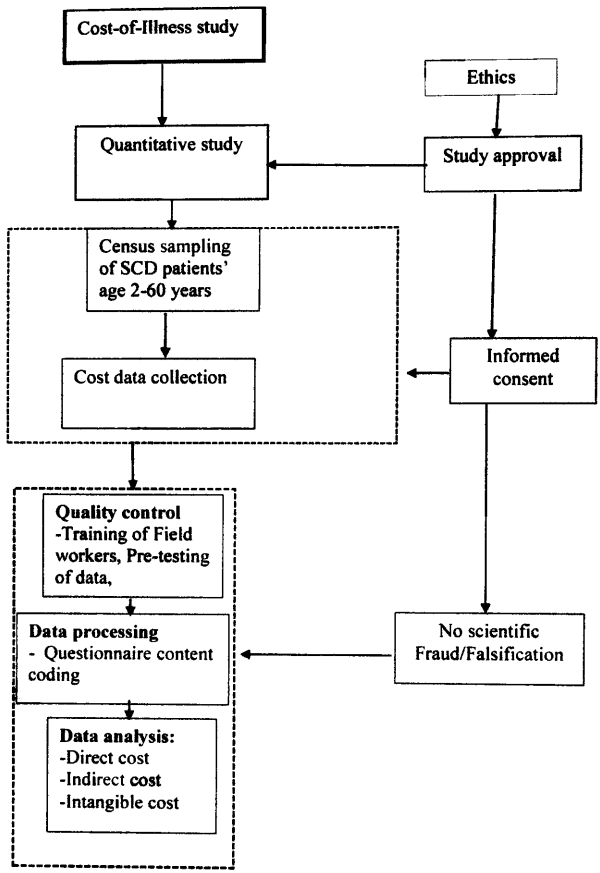
3.0 METHODS

This chapter describes the research methods and design used in the study, the setting and context under which the study was conducted as well as the target population of the study. It further explains various steps for collecting data as well as ethical considerations observed.

3.1 Study design

A retrospective cross sectional cost of illness (COI) study of SCD patients who attended the sickle cell clinic at Tema General Hospital. As described in Figure 2, it was a quantitative method with a cost analysis designed to determine direct, indirect and intangible costs of SCD patients. The study was conducted in June, 2017.

Figure 2. Schematic diagram of the study design



3.2 Study Area

The study was conducted at the sickle cell clinic (SCC), Tema General Hospital (TGH) in the Tema Metropolis of the Greater Accra Region of Ghana. Tema General Hospital is a semi-regional facility and the main referral hospital in Tema Metropolis. It is one of the biggest hospitals in the Greater Accra Region of Ghana and its strategic location makes it accessible to the major towns in the southern part of Volta region as well as Ashaiman, Adentan and Kpone-Katamanso Municipalities which were all carved out from Tema Metropolis in 2011. The estimated 2016 population of Tema Metropolis is 351,616 (as projected from the 2010 Census), and a Tema Central sub metro population of 101939 making it the second largest-population of the ten districts in the Greater Accra Region, after Accra Metropolis. Estimated outpatient department (OPD) attendant of Tema General Hospital is 69,720 per annum with an average attendance of 194 per day. Annually 1053 patients (both old and new) of SCD attend sickle cell clinic out of which 153 were admitted in 2015. Average attendance of four (4) regular visits per client was recorded in 2015 (TGH Annual Report, 2015). Also averagely, 250 new and old patients (both children and adults) regularly attended the sickle cell clinic every quarter (TGH Annual report, 2015). The sickle cell clinic at Tema General Hospital was started in 2003 with a weekly clinic consultation and counselling sessions. The clinic now runs a bi-weekly specialized consultation where SCD patients attend clinical consultations and counseling's on Wednesdays and Fridays respectively. Since 2015 the hospital now operates a Pediatric and Adult clinics instead of the former single clinic for all clients. The adult clinic is headed by a principal Medical Officer and the children clinic by a Pediatrician. There are two nurses, and two support staffs together with the two doctors forming the team that runs the clinic with a common register (TGH Annual report, 2015). Though two Physicians run the clinics for adults and children, records and activities are managed together by one team of SCD caregivers. All patients have to pay their medical bills through self-financing, except those

who are enrolled in the National Health Insurance Scheme (NHIS) in which a portion of their expenditure is catered for. The unit has four (4) reserved beds at all times for the Adults patients at the general wards and 2 beds for the pediatric patients at the Children ward.

3.3 Study Variable

The dependent variable of this study was household cost. The independent variable were direct, indirect and intangible cost of SCD.

In reference to Table 1, **direct cost** was made up of medical and non-medical costs. Medical cost included cost of registration, consultation, medicines, laboratory tests, imaging and blood transfusion. Non-medical cost included cost of transportation, food and water, communication and other few specified items.

Indirect cost: This included working hours loss (productivity loss) by SCD patients, the number of days caretakers had absented themselves from work, waiting and travel times of both patients and caretakers.

Intangible cost: Perception of patient's pain, social isolation, stress, self-esteem and stigma was assessed. This was done with the use of a Likert scale. The individual choices were graded and an average score obtained.

The socio-demographic characteristics for each patient was obtained, including age, sex, marital status, monthly income, NHIS beneficiary status, and educational status.

Table 1 Independent Variables of household cost of SCD

Household cost	Category of Care	Cost Description
Direct Cost	Medical	1.Consultation 2.Medicines 3. Laboratory investigations 4. Diagnostic test 5.Registration
	Non-medical	1.Travel 2.Food 3.Water/snacks 4.Communication
Indirect Cost		1.Working hours lost (productivity losses) 2.Waiting time 3. Travelling time
Intangible Cost		1.Stigmatization 2.Social isolation 3.Pain 4.Stree 5.Self esteem

3.4 Study Population

The study population was all patients with SCD enrolled at the sickle cell clinic at Tema General Hospital within the ages of 2 -60 years.

Inclusion Criteria

All SCD patients who attended the Tema General Hospital Sickle Cell Clinic (Out Patient) with hemoglobin genotype Hb SS and Hb SC within the ages of 2-60 years, were included in the study. All those who were above 18 years and gave consent and between 2-17 years who assented were included.

Exclusion Criteria

The study excluded all SCD with genotype HbSS and HbSC within age's 2-60years who refused to consent or assent. Patients within the selected group of SCD on admission at the hospital during the period of study were excluded.

3.5 Sample size

Minimum sample size was obtained using the number of persons on the Sickle Cell Register who attended clinic over a period of 12 months from June 2016 to May, 2017. The total number of persons on the Sickle Cell Register as at December, 2016 were 1053, averaging 87 persons per month (TGH Annual report, 2016). With this finite population of 87, the sample size of SCD patients was determined by adopting the following statistical formula for the minimum sample size calculation (Yamane, 1967)

$$n = N/ 1+N (e)^2$$

Where:

n = sample size

N= Population size

e = the acceptable sampling error (level of precision).

p = degree of variability

A 95% confidence level and p = 0.5 were assumed, N=87; e =0.05 (5%)

$$n= 87/ 1+87(0.05)^2$$

$$n = 87/1+0.22$$

$$n= 87/1.22 = 71.3$$

Assuming a non-response rate of 10%.

Non-responders = 71.3 × 10% =7.13

Actual sample size taken was 71.3 +7.13 = 78.43; Approximated to 78 SCD patients.

3.6 Sampling Procedure

Records of attendance of patient who visited SCC in June, 2017 and between the ages of 2-60 years were used for selection to constitute the sample size of 78 SCD patients. At the clinic all patients reporting for review or counselling are made to write their names. The first person to arrive at the clinic with his or her folder is selected first followed by the next patient according to the daily attendance list. All patients within the required age group were picked according to the order on the list of attendance per time of arrival and after consent or assent given. Averagely 15 SCD patients within the inclusion criteria were obtained during Wednesday clinic and Friday counselling sessions and within 5 weeks a total of 78 patients were interviewed.

3.7 Data Collection Technique and Tool

Four data collectors with one field supervisor were used for the data collection. Structured questionnaires were used to collect data from SCD patients attending TGH sickle cell clinic. The questionnaire (Appendix C) was divided in five (5) sections: The first part being the socio-demographic information. The second section dealt with health status information. The third section dealt with direct medical and non-medical cost. The indirect cost borne by households of SCD patients on accounts of absenteeism from work, travel and waiting time over the selected period were captured in the fourth section and the last section captured variables for estimating intangible cost which included information on stigmatization, pain, social isolation, stress, and self-esteem. Experienced research assistants were used.

3.8 Data processing

The investigator and assistants visited the clinic and cross checked completed questionnaires for completeness, consistency and accuracy. During this process any other error in the data was rectified. All the close ended questions in the questionnaire were pre-coded and the open-ended questions coded after reviewing the responses and developing a coding manual. Data entry – coding, compilation and editing were done using Epi Info version7.2. Data was imported unto Microsoft Excel 2013 version for cleaning

3.9 Data Analysis

This constitute the background characteristics of participants, estimation of direct, indirect and intangible cost as well as the sensitivity analysis of cost of SCD.

3.9.1: Background characteristics of participants

The background characteristics of the research participants were obtained- sex, age in completed years, marital status, education level in ordinal form, socio-economic, and Stata /IC 14.1 version was used for most of the analysis.

3.9.2 Estimation of direct costs

This was made up of medical and non-medical cost. From the questionnaire in appendix C, the medical cost included registration fees, consultation fees, and cost of medicines, laboratory investigation fees, imaging fees, and blood processing fees for transfused patients. Consultation and registration fees were obtained from a payments bill in the patient's folders. Cost of medicines purchased over the counter and or purchased outside hospital were obtained from receipts and also verbally. Cost of blood processing and routine laboratory investigations such as Full blood count, Malaria parasite screening were also obtained as part of bills in the patient's folders or receipts paid out of pockets. These were computed together for both patients with and without access to the National Health Insurance Scheme (NHIS). The cost

adjustment policy being practiced in the TGH ensured almost equal fees for both NHIS holders and non NHIS holders for all goods and services bought. Average cost per visit was calculated and documented.

The non-medical related costs included the following: **Transport cost**: This covered the cost of caretakers and patients conveying to and fro the hospital for regular review and anytime SCD crises –pain, anemia, chest pains-occur. Fares paid per visit were enquired from SCD patients or care takers, recorded and total calculated. **Food and Drinks cost**: The estimated cost of food and drinks as well as call phone calls (miscellaneous) SCD patient and caregiver incurred during the period of waiting in the hospital were computed. Cost of each item were ascertained, documented and total sum calculated. **Other miscellaneous costs:** The costs of other miscellaneous such as phone call made during care were computed by adding up the miscellaneous costs incurred by households in seeking care.

The total direct cost was ascertained by summation of all components of medical-related and non-medical expenses.

3.9.3 Estimation of indirect costs

Indirect cost was calculated with human capital method which is intended to ascertain the cost of human capital as the present value of the patient's upcoming wages. This is obtained by multiplying the daily minimum wage rate by the number of days of workdays lost for the patient as well as his/care giver(s). Productivity loss was therefore valued using the 2017 national minimum wage in the country. The valuation of productive time lost to patient relied on the national minimum wage per day of GHS8.80 as at June, 2017 (Ministry of Finance and Economic Planning, June 2017).

Table 2. Estimation of Household indirect cost

No	Category	Cost estimation approach
1.	Valued Days lost to all patients	This is the summation of days lost to patients who are due for SCD clinic per month except students
2.	Valued Days lost to those who accompany the patient to the hospital.	This is the sum total of days lost to household members as a result of clinic visit with patient per year
3.	Productive /Valued traveling time loss	This is the sum total of the number of hours spent by the patient as well as household members as travelling time to seek treatment at clinic per year
4	Productive /Valued waiting time loss	This is the sum total number of hours spent by the patient as well as household members as waiting time to seek treatment at clinic per year
4.	Total Indirect cost	This is the overall aggregation of the total productivity loss in travel, waiting and days absent from work multiplied by the minimum daily wage of Ghana(GHS 8.8)

3.9.4 Total household cost estimation

The overall household cost was estimated by the summation of the total direct cost and total indirect cost. The average cost per patient was determined by dividing the overall cost by the SCD patients sampled.

3.9.5 Determination of intangible costs

The intangible cost to household was not quantified in monetary terms. Likert scale was used to score the domains of intangible cost. A five dimension Likert scale was used, in which

patients were asked to rate statements under each dimension as (1)'Not at all' (2) ' A little' (3) 'Moderately' (4) 'Quite a bit' (5) 'Extremely' in respect of the pain, social isolation, stress, stigma and self-esteem. The scores of each domain of the intangible cost was used descriptively for patients and household caretakers. The mean of the responses for each dimension and their individual items under them were then displayed graphically in Microsoft Excel 2013. Table 3 indicates the scale and scores of the intangible cost.

Table 3. Composite Intangible Cost

No	Domain	Scale	Score range
1	Stigmatization	1. Not at all 2. A little 3. Moderately 4. Quite a bit 5. Extremely	2-10
2	Social isolation	1. Not at all 2. A little 3. Moderately 4. Quite a bit 5. Extremely	2-10
3	Pain	1. Not at all 2. A little 3. Moderately 4. Quite a bit 5. Extremely	2-10
4	Stress	1. Not at all 2. A little 3. Moderately 4. Quite a bit 5. Extremely	3-15
5	Self Esteem	1. Not at all 2. A little 3. Moderately 4. Quite a bit 5. Extremely	3-15

NB: Numbers 1-3 variable scores estimated from 5 questions by 2 responses and numbers 4-5 variables scores estimated from 5 questions by 3 responses.

The composite intangible score was obtained by adding the dimensions in each domain and multiplying by the number of questions giving a score of 12-60. This score was reclassified into low, moderate and high intangible cost with their corresponding ranges using tertile statistic as shown in Table 4.

Table 4. Composite Dimension of Intangible cost

Score	Dimension	Upper range limit
Composite Scores	Low	12-28
	Moderate	29-45
	High	46-60

3.9.6 Sensitivity analysis of cost of SCD

Sensitivity analysis (SA) is the process of testing the robustness of the study's conclusions to variations in the various assumptions made in the course of the analysis, changing the value of some parameters, variables and/or model structure in a meaningful way to examine the influence of these changes on the study results. The components on which the sensitivity tests were conducted were medication and wage. The presence of uncertainty associated with these items led to their selection. The test was conducted by increasing the two cost components by 3%, 5% and 7% respectively.

3.10 Quality Control

The following measures were adopted to ensure data quality.

Training of Interviewers

Research assistants who could speak English and Akan were recruited and a- day training session organized for them on how to conduct interviews before embarking on the field work. The training content included an overview of the project, objectives of the study, and interviewing skills.

Pre-testing and Review of Instruments

The questionnaire used in the study was reviewed and pre-tested at Ridge Hospital, which had similar working environment and sickle cell clinic as Tema General Hospital. Research assistants were supervised to ensure standardization in the way the questions were asked. The essence of the pre-testing was to improve validity of the data collection tools before the actual field work. This process allowed for any ambiguous questions to be clarified and also gave the investigator an idea of the length of time it might take for a questionnaire to be administered. The participants in the pretested area gave their opinion on whether or not the length of time was reasonable or too long, and also the user friendliness of the questionnaire. The questionnaires from the pretested study was entered into Epi Info version7, screened with Microsoft Excel 2013, and cross tabulation done using Stata/IC 14.1 version. Results from the pretest were used to make adjustment and fill gaps identified in the questionnaire.

3.11 Ethical Clearance

To ensure that the research meets ethical standards, clearance were sought from the Ghana Health Service Ethics Review Committee.

3.12.1 Access and approval of study area.

The principal investigator (PI) visited the study area and informed the Tema General Hospital about the intension to undertake a research in their facility. An introductory letter was taken from the School of Public Health, University of Ghana for the Management of Tema General Hospital to conduct the study. Approval was given by the Management of Tema General Hospital and study successfully conducted.

3.12.2 Study Subjects

The study included all SCD patients who attend sickle cell clinic at the Tema General Hospital and within the ages of 2-60 years.

3.12.3 Privacy and Confidentiality

In order to ensure privacy and confidentiality, the questionnaire were coded and names of respondent were not be required in filling the questionnaire. The interview were conducted in an isolated places with the individual patients to ensure privacy. Information obtained from patients were kept confidential between the PI and the study participants.

3.12.4 Risks and Benefits

Apart from the time that were lost by study subjects in answering questionnaires, there were no risk or cost associated in participating in the study. Participants did not gain any direct benefits. However it was expected that the results of the study will contribute towards policy decision on cost of caring for chronic diseases like SCD in Ghana.

3.12.5 Voluntary Withdrawal

Participation in the study was voluntary. Participants were free to pull out from the study at any point in time. Participants were however encouraged to fully participate to ensure that findings from the study were a true reflection of the household cost of SCD amongst clients attending SCC at Tema General Hospital. There were no incidence of withdrawal

3.12.6 Informed consent and consenting process

Participants were given the opportunity to give consent or assent after explaining the objectives, benefits, risk, confidentiality, and the process of data collection with a questionnaire, to all patients before the study commenced. Participants were involved individually with their caretakers to endorse a consent or assent form that contained a well spelt out instructions on the process of data collection. The decision to take part in the study was absolutely voluntary and refusal to take part was not by any means affected the care and treatment of the patients.

3.12.7 Compensation

Study participants were not given any compensation for participating in the study. This was made known to participants before they were recruited. However participants were given snacks and water after questionnaires were administered.

3.12.8 Data storage, security and usage

Data files were password protected. Hard copy and electronic data stored in locked file cabinets, and access limited to the PI and the Supervisors of the study. Data stored or kept safe for at least one year after research published and then destroyed by burning.

3.12.9 Conflict of interest

The PI had no conflict of interest in the study.

3.12.10 Proposal Funding

This study was self-sponsored in partial fulfillment for a Masters of Public Health Degree from the School of Public Health, University of Ghana.

CHAPTER FOUR

RESULTS

The study result is presented in this chapter. The various results section are: background characteristics of study patients, direct treatment cost of SCD patients; indirect treatment cost of SCD patients and the intangible cost associated with SCD.

4.1 Background Characteristics of SCD patients

The socio-demographic background characteristics of SCD patients with genotypes HbSS and HbSC involved in this study are shown in Table 4.1 More females patients 55.1% (43) compared to males 44.9% (34) were involved. Majority of the patients 51.3% (40) were below 18 years, 21.8% (26) were between the ages of 18 and 30 years whilst only 15.4% (12) were above 30 years old. The mean age of the SCD population was 20 years. Also among the patients who are not married, majority 51% were minors who are below the age of marriage in Ghana (18 years), whilst the remaining 48.7% are 18 years and above. The married SCD patients were only11.5% (9). Majority of the SCD patients 91% (71) had at least received some form of formal education whilst very few 9% (7) has had no formal education. Among those with formal education, those with primary education constituted the majority 29.5% (23) followed by 25.6% (20) who had secondary, 24.4% (19) attained the level of junior high and the least of 11.5% (9) had tertiary level education. About 82% (64) of the patients were unemployed whilst 18% (14) were employed. Amongst the employed patient are 6.4 % (5) who were in the formal sector and 11.6% (9) informal. Within the unemployed group 29.5% (23) were students, 25.7% (20) unemployed adults, 24.4% (19) were apprentices in diverse vocational trades, one housewife (1.3%) and one retired worker (1.3%). About 15.4% (12) patients stopped working due to SCD whilst no reasons were assigned to the rest 10.2% (8).

The average monthly income of less than GHS1000.00 was received by 57.7% (45). Registered National Health Insurance Scheme (NHIS) users were 86.5% (69). Homozygous hemoglobin-S genotype (Hb SS) were 65.4% (51) and constituted the majority .The average attendance of SCD patient at SCC was 58.3% (7) out of expected 12 regular visits within a year.

Table 5 Socio-demographic characteristics of SCD

Characteristic	Number (%)
Sex	
Male	35 (44.9)
Female	43 (55.1)
Age	
< 18	40 (51.3)
18-30	26 (33.3)
>30	12 (15.41)
Marital Status	
Married	9 (11.5)
Not married	69 (88.5)
Educational Level	
No education	7 (9.0)
Primary	23 (29.5)
Middle/JSS/JHS	19 (24.4)
Secondary	20 (25.6)
Tertiary	9 (11.5)
Employment Status	
Employed	14 (18.0)
Unemployed	20 (25.6)
Students	23 (29.5)
Apprenticeship	19 (24.4)
Housewife	1 (1.3)
Retired	1 (1.3)
Monthly income	
< GHS 1000	45 (57.7)
> GHS1000	33 (42.3)
Mean(SD)	GHS 1,168.97
Insurance status	
Insured	69 (88.5)
Not insured	9 (11.5)
Sickling genotype	
HbSS	51 (65.4)
HbSC	27 (34.6)
Total	**78 (100.0%)**

Table 6.Total Household cost of SCD

Cost component	Number	Cost (GHS)	Average cost (SD)	Median	Cost profile (%)
Direct cost					
Direct medical cost					
Registration/Folders	78	4,695.00	60.19 (40.25)	50.00	4.8
Consultation	78	7,580.00	97.18 (57.38)	90.00	7.8
Laboratory investigations	78	16,055.00	205.83 (140.48)	166.00	16.5
Medicines/drugs	78	30,578.00	392.03 (623.13)	222.50	31.3
Diagnostic tests	58	2,820.00	48.62 (36.44)	45.00	2.8
Other	29	1,345.00	46.38 (77.29)	0.00	1.4
Sub total		63,073.00	808.63 (696.07)	625.00	64.6
Direct non-medical					
Transportation	78	11,181.00	143.35 (156.93)	100.00	11.5
Food	78	6,175.00	79.17 (106.95)	50.00	6.3
Drink/water cost	77	3,235.50	42.02 (57.26)	20.00	3.3
Miscellaneous	67	2,198.00	32.81 (57.33)	15.00	2.3
Others	6	74.00	12.33 (9.09)	15.50	0.1
Sub total		22,863.50	293.12 (304.04)	210.00	23.4
Total direct cost		85,936.50	111.75 (782.15)	888.00	88.0
Indirect Cost					
Patient					
Valued productive days lost	54	6,099.50	112.95 (98.03)	83.60	6.3
Valued travelling time	78	329.23	4.22 (3.51)	3.30	0.3
Valued waiting time	78	523.27	6.71 (3.33)	6.60	0.5
Caretaker					
Valued productive days lost	52	4,087.93	78.61 (70.45)	70.40	4.2
Valued travelling time	56	273.68	4.89 (5.94)	3.30	0.3
Valued waiting time	56	365.09	6.52 (3.21)	6.60	0.4
Total indirect cost		11,678.70	149.73 (142.03)	122.87	12.0
Total cost		97,615.20	1,251.48(833.2)	988.45	100.0

Table 6 shows that the total estimated non-medical cost was GHS 22,863.00 with a mean of GHS 293.12 and a median of GHS 210.00. Food cost formed 6.3% of the total cost profile, soft drinks/water cost was 3.3% of total cost profile whilst miscellaneous cost (mainly call time) contributed 2.3% with a mean of GHS 32.81.

4.2 Direct Treatment Cost of SCD at SCC

The direct cost was made up of two main components i.e. direct medical and direct non-medical cost incurred by SCD patients and household.

4.2.1 Direct medical cost

The components of direct medical costs were registration, consultation, laboratory investigations, medicines, other diagnostic test and other treatment such as blood transfusion. Table 6 shows distribution of direct medical cost by study patients. Cost of medicines constituted a bulk of total cost profile (31.3%). The mean medicine costs was GHS 392.00 with a median of GHS 222.50. Laboratory investigation cost recorded the second highest share of the cost profile with GHS 16,055.00 with a mean of GHS 205.83 and a median of GHS166.00. Consultation and registration/folder cost were GHS 7,580.00 and GHS 4,695.00 respectively and their respective means were GHS 97.00 and GHS60.00. The total direct medical cost estimated was GHS 63,073.00 with a mean cost of GHS 808.63 and a median of GHS 625.00

4.2.2 Direct non-medical cost

The components of direct non-medical costs were travel cost, food cost, drink/water cost, miscellaneous and other cost. Approximately half of the total non-medical cost profile composed of travel cost. That is, for SCD patients travel cost constituted 11.5% of the total cost with a mean of GHS 143.35 and a median of GHS 100.00. This formed the major expenditure of the direct non-medical cost.

4.3 Indirect Cost of SCD

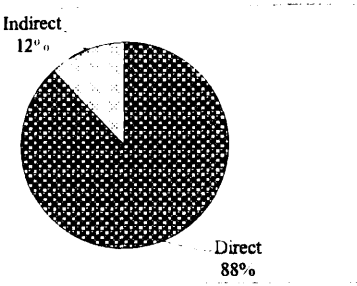
The indirect cost estimated the productive work hours lost as a result of SCD by using the human capital approach. Table 6 shows the percentage distribution of total time spent travelling to and from SCC by both employed and unemployed SCD patients, productive work time lost by employed and unemployed with the exception of students and total waiting time spent at the SCC and hospital whilst seeking health care or treatment. The percentage work time lost in the month preceding patients' clinic visit in relation to overall time spent by patients on SCD management was 52.2% , followed by 35% which constituted valued productive days lost by caregivers in taking care of the SCD patients , then 4.6% which is valued waiting time at the hospital for treatment by patient, then 3.1% waiting time of caregivers whilst SCD patient was receiving care, with 2.8% and 2.3% constituting the travel time to-and-from the hospital by patients and caregivers respectively. The overall valued productive time lost due to SCD patients receiving care was 10.5% (90 days) of total cost. Again the total productive lost hours due to waiting time at the hospital to receive medical care for both patients and caregivers was 0.9% of the total cost profile followed by 0.6% constituting total travel time.

The indirect cost estimation was done for only productively engaged SCD. The total productive days lost by SCD patients was 60 days whilst that lost by caregivers was 30 days. The valued productive time lost by SCD patients was GHS 6,099.50 with a mean of GHS112.95 whereas the valued productive time lost by caregivers was GHS 4,087.00 with a mean of GHS 78.61. The total indirect cost estimated was GHS 11,678.70 with a mean of GHS 149.73.

The total treatment cost of SCD was estimated by summation of direct cost and indirect cost. As shown in Table 6 the total treatment cost for SCD at the SCC was estimated as GHS 97,615.20 with mean of GHS 1,251.48 and median of GHS 988.45.

Figure 3 presents percentage distribution of treatment cost. Direct cost constituted the greater portion of the total treatment cost profile which was 88%.

Figure 3. Distribution of Total treatment cost of SCD

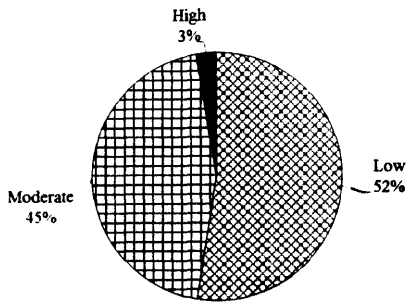


The share of direct cost to total treatment which was very high (88%) and can largely be attributed to the influence of three main products-medicine cost, Laboratory investigation cost and transportation over the total treatment cost. Medicines cost constituted 31.3% of the total treatment cost profile, Laboratory investigation cost 16.5% while transportation cost was 11.5% together making up 59.3% of the total treatment cost profile. The mean cost of medicines, laboratory investigation and transportation were GHS 392.03, GHS 392.03 and GHS 143.45 respectively per annum. Mean valued productive days lost by both patients and caregivers were GHS112.95 and GHS 78.61 respectively per annum.

4.4 Intangible Cost associated with SCD

The total composite intangible cost indicates that highest composite SCD score was low dimension accounting for 52% (41) of the total score. The least score was the high dimension constituting 3% (2) as shown in Figure 4.

Figure 4. Total Composite intangible cost



Also the means of the five intangible cost dimensions for SCD patients was computed as in Table 7. The dimension with the highest mean was patients feeling pain in social situations (7.36) whereas the one with the least mean was patients bothered by stigma (3.19). The estimated means for low self -esteemed in public, being stress up with disease condition, or feeling socially isolated in the event of sporting activity or employment were 7.19, 7.12 and 3.77 respectively.

Table 7. Intangible cost components with Means

Variable	Observation	Mean	Std. Deviation	Minimum	Maximum
Stigma	78	3.19	1.81	2	10
Stress	78	7.12	2.57	3	15
Pain	78	7.36	2.22	2	10
Social isolation	78	3.77	2.29	2	10
Self-esteem	78	7.19	1.91	3	15

Figure 5. Distribution of Intangible cost by Sickling genotype HbSS and HbSC

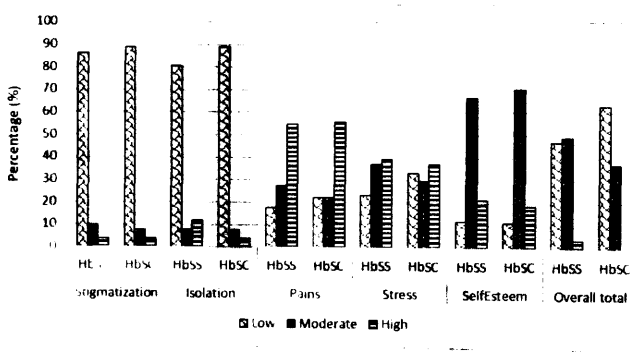
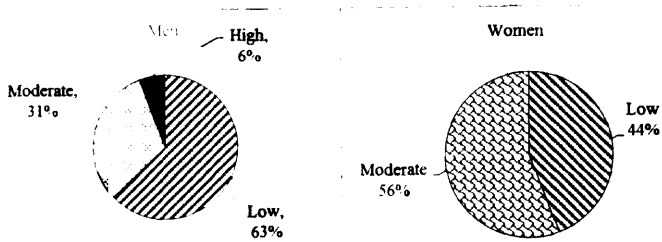


Figure 5 demonstrates intangible cost of the five dimensions used in respect to hemoglobin genotypes HbSS and HbSC. Overall intangible total cost indicated a least score of high dimension component, followed by moderate dimension and the highest intangible score being low dimension. Genotype HbSS was identified as having higher intangible score compared to

HbSC. For individual dimensions, pain is very prominent, followed by stress, then self-esteem, social isolation and the least being stigmatization.

Also composite intangible cost by sex as indicated in Figure 6 shows that majority of women (56%) constituted moderate composite dimension followed by 44% constituting low dimension with no woman in a high dimension composite score. Comparatively 63% of men constituted the highest dimension composite score which is low, followed by 31% moderate and 6% high composite scores.

Figure 6. Intangible cost distribution by Sex



4.6 Sensitivity analysis of cost of SCD

The components on which the sensitivity tests were directed as in Table 8 were medication and wage. The test was conducted by increasing the two cost components by 3%, 5% and 7% respectively. One way sensitivity analysis was conducted by varying the cost of medication by 3%, 5% and 7% resulted with 0.9%, 1.6% and 2.2% respective increases in total household cost of SCD. Similar analysis conducted on wage rate generated percentage increases of 0.4%, 0.6% and 0.8% respectively in total treatment cost. Again the 3%, 5% and 7% variations in medication respectively resulted in 0.1%, 0.2% and 0.3% increases in direct cost. At this same level of variations in wage rate resulted in 0.3%, 0.5% and 0.7% increases in indirect cost.

Alterations in both medication and wage rate by 3%, 5% and 7% resulted in a percentage reduction in direct cost in proportions to total household cost and thus a percentage rise in indirect cost in proportions to total treatment cost. There were 1.3%, 2.2 %and 3.0% increases in total treatment cost when both medication and wage rates were varied by 3%, 5% and 7%. The results of the sensitivity analysis showed that the cost estimates were sensitive to changes in wage and medicine cost variables and the estimated treatment costs were robust because varying assumptions and uncertainty caused an insignificant change in total SCD household costs.

Table 8. Sensitivity Analysis chart

Scenario	Cost component	*Percentage change in parameter	Total cost		Percentage change in total cost	Proportion of total cost		Percentage change in proportions of cost	
			GHS	USD**		Direct	Indirect	Direct	Indirect
Base scenario		0	97,615.20	22,388.81	0.0	88.0	12.0	0	0
Variation (One-way Sensitivity Analysis)	Medication	3	98,532.54	22,599.21	0.9	88.1	11.9	0.1	-0.1
		5	99,144.10	22,739.47	1.6	88.2	11.8	0.2	-0.2
		7	99,755.66	22,879.74	2.2	88.3	11.7	0.3	-0.3
Variation (One-way Sensitivity Analysis)	Wage rate	3	97,965.56	22,469.17	0.4	87.7	12.3	-0.3	0.3
		5	98,199.14	22,522.74	0.6	87.5	12.5	-0.5	0.5
		7	98,432.71	22,576.31	0.8	87.3	12.7	-0.7	0.7
Multi-variation (Multi-way Sensitivity Analysis)	Medication and Wage rate	3	98,882.90	22,679.56	1.3	87.8	12.2	-0.2	0.2
		5	99,728.04	22,873.40	2.2	87.7	12.3	-0.3	0.3
		7	100,573.17	23,067.24	3.0	87.6	12.4	-0.5	0.5

*The cost of medication and wage rate was independently and concurrently varied by 3%, 5%, 7% increments.

** USD 1.00 is equivalent to GHS 4.38 (Bank of Ghana average interbank exchange rate, June 2017)

CHAPTER FIVE

DISCUSSION

This chapter presents the discussions of the study. The outline is based on the objectives of the research. It includes summary and discussion of the key findings of the study related to published literature on cost of SCD treatment as well as pain and stigmatization associated with the disease.

Summary of findings include the fact that 65.4% of SCD patients are genotype HbSS and 34.6% HbSC, indicating HbSS genotype is more prevalently. This is in line with Anim et al (2016) whose work on Prevalence of Psychological Symptoms among Adult with SCD in KBTH, Ghana had 65% of the population being HbSS and 35% being HbSC. Given that HbSS is a more severe form of the disease than HbSC genotype and HbSS persons experience more pain and psychological distress than HbSC (Konotey-Ahulu, 1991). This findings agree with the previous research.

A mean age of 20 years indicated majority of SCD patients are young. This observation supports WHO statement that individuals with homozygous gene S do not have defends against Malaria in Africa and consequently die before attaining the age of procreation (WHO, Regional office for Africa, 2015). The disease appeared more among females (55%). However it may also confirms that more women report to the health centers when they are ill than men (TGH Annual report, 2015).

Many SCD patients 82% (64) are unemployed as a result of disease complications that prevent them from attending work every day, leading to loss of jobs or loss of interest formal employment. Average monthly income of the employed SCD patients was GHS 1,523.33 with the unemployed patient's monthly income being GHS 1,123.90. The mean (monthly) cost for SCD were GHS 1,251.48. This implies that averagely 83% of all income received by

employed SCD patients are spent on health care and all monies received and even more from relatives, friends and loans by the unemployed, were spent on SCD health care. It therefore indicates that total annual income of SCD patient and their care givers (GHS 91,180.00) were all spent on healthcare.

The direct estimated cost (88%) constituted greater proportion of the total treatment cost profile with 64.6% being direct medical and 23.4% direct non-medical. Even though the total indirect cost in percentage is (12%) out of total treatment cost, the monthly indirect cost of GHS 149.73 was higher compared to the monthly direct medical cost which is GHS111.75. This means that averagely money spent by households on indirect cost every month is higher. The high cost of SCD has been recognized in Ghana and Nigeria with in-patients whose caregivers pay for lodging, or regularly visited them (Sebil, 2011 & Ngolet, 2015). A low indirect cost in this study resulted because these patients stayed in their homes and only attended clinics for regular review. Many patients were students (23%) whose productivity loss was also not valued.

The mean of intangible cost estimated for SCD patients was highest for pain (7.36) and least for stigmatization (3.19). Attendance of 1053 annually (TGH Annual Report, 2015) with average monthly attendance of 87 patients at SCC is woefully inadequate. Despite the carrier rate of 30% with 2% of all new borne babies having SCD in Ghana (Dennis-Antwi et al., 2008), Tema Metro with a population size of 315,616 will produce 6,312 SCD patients. Thus only 16% of estimated SCD population in the study area attends clinic regularly.

5.1 Direct Cost of SCD

This study recorded a high direct cost of the total household cost of SCD patient estimated as 88%. This high cost recorded compared to the indirect cost was attributed to fact that the

indirect cost of many participant who were students in this study were not computed.Sebil (2011) estimated a similar high direct cost of 84% for social cost of SCD in three SCC centers in Ghana. Also Kauf (2009), estimated a high SCD related costs (80.5%) that was consistent with previous cost studies and suggested that the total cost of care for SCD is large and equivalent to other chronic diseases and conditions such as bipolar depression. Average annual societal costs per patients with bipolar disorder varied from US$1,904 to US$33,090. Production losses made up to 20–94 % of the total societal cost of bipolar mania (Jin et al., 2015). This total cost falls within the total cost range of this study.

Das Gupta and Guest (2002) also estimated the non-healthcare direct medical cost of mental healthcare (chronic disease) was 79% of the total direct cost and this further supports the findings of the study. Their direct medical cost accounted for the direct mental healthcare and treatment (in-patient day's costs and out-patient cost).

The results of this study is again in line a study by Amoakoh and Aikins (2013) on household cost of Buruli ulcer (a chronic disease) in Ghana where it established that 97% of the total was from direct cost.

The findings of this study is consistent with Casado et al., (2006) study on Multiple sclerosis. Casado (2006) found direct cost accounting for a large proportion (60%) of the total cost of multiple sclerosis.

5.2 Indirect Cost of SCD

The total indirect cost estimated for SCD at SCC was GHS 11,678.70 constituting a low proportion (12%) of the total cost of SCD. The indirect cost estimation was done for only productively engaged SCD. The total productive days lost by SCD patients was 60 days whilst that lost by caregivers was 30 days. The valued productive time lost by SCD patients was GHS 6,099.50 with a mean of GHS112.95 whereas the valued productive time lost by

caregivers was GHS 4,087.00 with a mean of GHS 78.61. The total indirect cost estimated was GHS 11,678.70 with a mean of GHS 149.73. This result is in line with Sebil (2011) whose research on social cost of SCD in three different clinics in Ghana yielded 17% of total household cost being indirect cost.

The cost due to care giving by household members made up the second highest proportion of the total indirect cost incurred by SCD patients. This findings of low indirect cost of chronic disease is in line with a study of Buruli ulcer in Ghana by Amoakoh and Aikins (2013) that estimated indirect cost of out-patient treatment as low as 4% of the total cost because respondents did not include admitted patients.

Productivity losses could have been over or understated because of recall and respondents not knowing the exact and actual hours lost due to SCD. This can also be addressed if a prospective study instead of retrospective study such as this study is conducted.

5.3 Total Health Cost of SCD

The total cost of SCD estimated to be GHS 97,615.20 with monthly average cost of GHS 1,251.48 was high. It came to bare that 83% of all income received by employed SCD patients are spent on health care whilst all monies received and more from relatives, friends and loans by the unemployed, are spent on healthcare.

The total cost of this study is however lower in comparison to that estimated by Sebil (2011) (GHS 2,724.00 per month). The difference between these costs can be attributed to the cost components that were estimated including admissions.

Olatunya (2015) in a study on the Financial Burden of sickle cell disease on households in Ekiti, Southwest Nigeria, estimated total health expenditure of all the households as US$ 26,607.59 with a range between US$73 and US$1,303, and a mean of US$ 240 ± 215 (GHS

1,051.00) per household. The mean total cost is almost equivalent to the mean total cost of this study (GHS 1,251.48).

5.4 Household Intangible Cost of SCD

The intangible cost for SCD patients were described using the composite scores to determine the variations of patients' response to the combined intangible cost items. The highest composite score was low dimension making up 52.2% (45) of the total score and the least score was the high dimension constituting 3% (2). The highest composite score in the dimension of low implies that the response of more than one-thirds of SCD patients to the composite intangible cost items was minimal as compared with their reactions to the individual intangible cost items. The scores further uncovered that less than two-third of SCD patients responded above average or were expressively affected by the composite intangible cost variables. Two SCD patients were minimally affected by the composite variables. However, notwithstanding the level of dimension, SCD patients to a large extent express pain, suffer some level of stress and low self-esteemed and therefore need psychological care. Whilst patients who scored moderate and high dimensions are likely to constantly seek care and counseling, those in the score of low dimension may not need much regular treatment since they are not much affected. In Amoakoh and Aikins (2013) study on estimated household economic costs of Buruli ulcer in Ghana, intangible cost suffered by Buruli ulcer patients and their households were measured with the Likert's scale without quantifying the value in monetary terms.

The findings of this study revealed that SCD patients suffer painful crises in social situations as a result of their illness (mean score of 7.35). As such they were unable to attend school regularly. This observation was worsened with low self-esteemed with mean score of 7.19 of SCD patients who persistently express their inability to fully participate in any competitive activity. SCD patients also suffer some emotional stress resulting from their crises. Stress

from emotions and painful crises contributed to a mean of 7.12. A greater proportion of patients indicated some bodily discomfort when walking. The score in the domain of social isolation was low with a mean of 3.77 with stigmatization being the least (3.19). This means that SCD patients received least stigmatization. Contrary to this study SCD patients were perceived to be suffering from high level stigmatization. Social issues such as stigma has been noted to be commonly associated with chronic disease (Biljana et al., 2007). Stigmatization is a noticeable parameters that affect a client's social life (Kyerewaa Edwin et al., 2011). Any defect of stigmatization being least may be corrected in further studies when questionnaires are administered at the household level and intangible cost of each member of the household estimated.

Limitations of the study

Documentation on household cost of SCD and individual health bills or records keeping for chronic disease condition like SCD was very challenging in many households. Even though bills in patient's folders were verified in addition to the routine review cost of medication, laboratory investigations and blood transfusions from the hospital service charges book, most bills paid were obtained by recall on the part of patients or caretakers hence the challenge of recall bias as a limitation.

The study was silent on cost of admissions and complications that affect quality of life.

Sensitivity Analysis on medication cost and wages were done to assess the validity and robustness of the research of the study despite the limitations.

CHAPTER SIX

CONCLUSION AND RECOMMENDATIONS

6.1 Conclusion

The household cost of SCD patients can be characterized into direct, indirect and intangible costs. Household cost at SCC constituted a registered population at outpatient care of people with SCD.

Household cost of SCD is high. Direct cost was the highest of the total cost. Medicines and laboratory investigation cost together form the bulk of the total direct medical cost .The mean indirect cost is higher compared to mean direct cost of SCD. Low intangible composite cost was the highest score and the high intangible composite cost, the lowest. The study further recognized that SCD patients suffer emotionally in pain with less stigmatization in society.

Despite a high participation of NHIS by SCD patients, high cost of care still affects regular attendance at SCC, with women and genotype HbSS being predominant attendants. The need for households' education on how to reduce valued days lost without affecting SCD patients' regular attendance to clinic is key.

6.2 Recommendations

This study targeted at estimating the cost incurred by SCD patients in seeking care at a sickle cell clinic. Recommendations made include:

1. The total household cost of SCD patient at SCC in this study should serve as a reference point that will inform SCD patients and caregivers to plan for their health care..

2. There is the need to intensify public education on how to reduce direct and indirect cost of care as well as minimizing the intangible cost associated with SCD.

This can be done if household members and society are educated on how to manage the psychological aspects associated with chronic diseases like SCD.

3. Finally, further studies should be conducted on impact of wider coverage of medical cost by NHIS on attendance of SCD patients to SCC in Ghana.

REFERENCES

Amoakoh, H. B., & Aikins, M. (2013). Household cost of out-patient treatment of Buruli ulcer in Ghana: a case study of Obom in Ga South Municipality. *BMC Health Services Research, 13*, 507.:10.1186/1472-6963-13-507

Anim, M. T., Osafo, J., & Yirdong, F. (2016). Prevalence of psychological symptoms among adults with sickle cell disease in Korle-Bu Teaching Hospital, Ghana. *BMC Psychology, 4*, 53. http://doi.org/10.1186/s40359-016-0162-z

Aisiku, M., Barakat, P., Lutz M., Smith-Whity K., (2007). Is treatment adherence associated with better quality of life in children with sickle cell disease? *Quality of life Research. 14.407-411 doi: 10.1007/s11136-004-5328-0*

Anders, B., Ommen, O., Pfaff, H., Lüngen, M.,Lefering R., Thüm, S.,(2013). Direct, indirect, and intangible costs after severe trauma up to occupational reintegration – an empirical analysis of 113 seriously injured patients

Amon, K.S., (2016). Economic burden of Type 2 Diabetes Mellitus- A case study of patients attending Eastern Regional Hospital Outpatients clinic. (MPH Dissertation, University of Ghana, School of Public Health, Ghana)

Barron, A. C., Lee, T. L., Taylor, J., Moore, T., Paso, M. H., Graham, T. B. & Brunner, H. I. (2004). Feasibility and construct validity of the parent willingness-to-pay technique for children with juvenile idiopathic arthritis. *Arthritis Care & Research, 51*(6), 899-908.

Biljana M., Serette K.,Tim M.,(2007). Sickle cell disease:-Experience of care givers in managing the disease in children in Western Region of Jamaica.

Boccuzzi S.J., (2003) .Contemporary Cardiology-Cardiovascular Healthcare Economics-Indirect health care cost.

Boone L, Bart S, Vansteenkiste M, Braet C, 2012. Is there a perfectionist in each of us? An experimental study on perfectionism and eating disorder symptoms

Casado, V., Martínez-yélamos, S., Martínez-yélamos, A., Carmona, O., Alonso, L., Romero, L., Arbizu, T. (2006). Direct and indirect costs of Multiple Sclerosis in Baix Llobregat (Catalonia, Spain), according to disability. *BMC Health Services Research, 8*, 1–8. :10.1186/1472-6963-6-143

Das Gupta, R., & Guest, J. F. (2002). Annual cost of bipolar disorder to UK society. *British Journal of Psychiatry, 180*, 227–233. doi:10.1192/bjp.180.3.227

Dennis-Antwi, J. A., Dyson, S., & Frempong, K. O. (2008). Healthcare provision for sickle cell disease in Ghana: challenges for the African context. *Diversity & Equality in Health and Care.*

Farrell, K., Dent, L., Nguyen, M. L., Buchowski, M., Bhatt, A., & Pilar Aguinaga, M. (2010). The relationship of oxygen transport and cardiac index for the prevention of sickle cell crises. *Journal of the National Medical Association, 102*(11), 1000.

Felts, W., & Yelin, E. D. W. A. R. D. (1989). The economic impact of the rheumatic diseases in the United States. *The Journal of rheumatology*, *16*(7), 867-884.

Gabriel, S. E., Crowson, C. S., Campion, M. E., & O'Fallon, W. M. (1997). Indirect and nonmedical costs among people with rheumatoid arthritis and osteoarthritis compared with nonarthritic controls. *The Journal of rheumatology*, *24*(1), 43-48.

Gail M. Sullivan and Anthony R. Artino Jr (*2013*) Analyzing and Interpreting Data From Likert-Type Scales. Journal of Graduate Medical Education: December 2013, Vol. 5, No. 4, pp. 541-542

Grosse, S. D., Odame, I., Atrash, H. K., Amendah, D. D., Piel, F. B., & Williams, T. N. (2011). Sickle cell disease in Africa: a neglected cause of early childhood mortality. *American journal of preventive medicine*, *41*(6), S398-S405.

Gupta, S., Hawker, G. A., Laporte, A., Croxford, R., & Coyte, P. C. (2005). The economic burden of disabling hip and knee osteoarthritis (OA) from the perspective of individuals living with this condition. *Rheumatology*, *44*(12), 1531-1537.

Ingram, D., & New, A. (1956). A Specific Chemical Difference between the Globins of Normal Human and Sickle-Cell An/Emia H/Emoglobin. *Nature*, *178*.

Jin, H., & McCrone, P. (2015). Cost-of-illness studies for bipolar disorder: systematic review of international studies. *PharmacoEconomics*, *33*(4), 341-353.

Jo, C. (2014). Cost-of-illness studies: concepts, scopes, and methods. *Clinical and Molecular Hepatology*, *20*(4), 327–37. http://doi.org/10.3350/cmh.2014.20.4.327

Kauf, T. L., Coates, T. D., Huazhi, L., Mody-Patel, N., & Hartzema, A. G. (2009). The cost of health care for children and adults with sickle cell disease. *American journal of hematology*, *84*(6), 323-327.

Konotey-Aholu FID. The Sickle cell disease patient, London; Macmillan Press Ltd, 1991. Google scholar

Kudebong, M., Wurapa, F., Nonvignon, J., Norman, I., Awoonor-Williams, J. K., & Aikins, M. (2011). Economic burden of motorcycle accidents in Northern Ghana. *Ghana Medical Journal*, *45*(4), 135–42.

Kyerewaa, A.E, Edwin, F., & Etwire, V. (2011). Controlling sickle cell disease in Ghana ethics and options. *Pan African Medical Journal*, *10*.

Lanes, S. F., Lanza, L. L., Radensky, P. W., Yood, R. A., Meenan, R. F., Walker, A. M., & Dreyer, N. A. (1997). Resource utilization and cost of care for rheumatoid arthritis and osteoarthritis in a managed care setting. The importance of drug and surgery costs. *Arthritis & Rheumatism*, *40*(8), 1475-1481.

Leardini, G., Salaffi, F., Montanelli, R., Gerzeli, S., & Canesi, B. (2002). A multicenter cost-of-illness study on rheumatoid arthritis in Italy. *Clinical and experimental rheumatology*, *20*(4), 505-516.

Liang, M. H., Cullen, K. E., Larson, M. G., Thompson, M. S., Schwartz, J. A., Fossel, A. H., ... & Sledge, C. B. (1986). Cost-effectiveness of total joint arthroplasty in osteoarthritis. *Arthritis & Rheumatism*, *29*(8), 937-943.

Lubkin, I., and Larsen, P. (2006). Chronic Illness: Impact and Interventions (6th ed.).

Sudbury: Jones & Bartlett Publishers.

Luce, B. R., Manning, W. G., Siegel, J. E., & Lipscomb, J. (1996). Estimating costs in cost-effectiveness analysis. *Cost-effectiveness in health and medicine*, *3*.

MacLean, C. H., Knight, K., Paulus, H., Brook, R. H., & Shekelle, P. G. (1998). Costs attributable to osteoarthritis. *The Journal of rheumatology*, *25*(11), 2213-2218.

Maetzel, A., Li, L. C., Pencharz, J., Tomlinson, G., & Bombardier, C. (2004). The economic burden associated with osteoarthritis, rheumatoid arthritis, and hypertension: a comparative study. *Annals of the rheumatic diseases*, *63*(4), 395-401.

Majumdar, S., Robertson, Z., Robinson, A., Starnes, S., Iyer, R., & Megason, G. (2010). Outcome of hematopoietic cell transplantation in children with sickle cell disease, a single center's experience. *Bone marrow transplantation*, *45*(5), 895-900.

Masserini, A. B., Zeni, S., Cossutta, R., Soldi, A., & Fantini, F. (2003). Cost-of-illness in systemic sclerosis: a retrospective study of an Italian cohort of 106 patients. *Reumatismo*, *55*(4), 245-255.

McGann, P. T. (2014). Sickle cell anemia: an underappreciated and unaddressed contributor to global childhood mortality. *The Journal of pediatrics*, *165*(1), 18-22.

Modell, B., & Darlison, M. (2008). Global epidemiology of haemoglobin disorders and derived service indicators. *Bulletin of the World Health Organization*, *86*(6), 480-487.

National Heart, Lung, and Blood Institute. (2014). Evidence-based management of sickle cell disease: expert panel report 2014. *Bethesda (MD): National Heart, Lung, and Blood Institute, National Institutes of Health.*

Ngolet L.O., Engoba M.M., Kocko. I.,Dokekias A.E.,Mombouli J.V.,Moyen G.M., (2016). Sickle-Cell Disease Healthcare Cost in Africa: Experience of the Congo

Ohene-Frempong, K., Oduro, J., Tetteh, H., & Nkrumah, F. (2008). Screening newborns for sickle cell disease in Ghana. *Pediatrics*, *121*(Supplement 2), S120-S121.

Ohaeri, J. U., & Shokunbi, W. A. (2002). Psychosocial burden of sickle cell disease on caregivers in a Nigerian setting. *Journal of the National Medical Association*, *94*(12), 1058–70. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/12510705

Olatunya, O. S., Ogundare, E. O., Fadare, J. O., Oluwayemi, I. O., Agaja, O. T., Adeyefa, B. S., & Aderiye, O. (2015). The financial burden of sickle cell disease on households in Ekiti, Southwest Nigeria. *ClinicoEconomics and Outcomes Research : CEOR*, *7*, 545–53. https://doi.org/10.2147/CEOR.S86599

Opoku A., (2015). Tema General Hospital Annual report

Osei-akoto, A., Akoto, A., Brobby, N. A. W., & Baffoe-bonnie, B. (n.d.). Alex Osei-Akoto Dept of Child Health SMS-CHS KNUST.

P"latt, O. S., Brambilla, D. J., Rosse, W. F., Milner, P. F., Castro, O., Steinberg, M. H., & Klug, P. P. (1994). Mortality in sickle cell disease--life expectancy and risk factors for early death. *New England Journal of Medicine*, *330*(23), 1639-1644.

Quinn, C. T., Rogers, Z. R., McCavit, T. L., & Buchanan, G. R. (2010). Improved survival of children and adolescents with sickle cell disease. *Blood*, *115*(17), 3447-3452.

Rizzo, L. B., Costa, L. G., Mansur, R. B., Swardfager, W., Belangero, S. I., Grassi-Oliveira, R., ... & Brietzke, E. (2014). The theory of bipolar disorder as an illness of accelerated aging: implications for clinical care and research. *Neuroscience & Biobehavioral Reviews*, *42*, 157-169

Sadarangani M, Makani J, Komba AN, (2009). An observational study of children with sickle cell disease in Kilifi, Kenya. ; 146:675–682.

Sebil W. (2011). Social cost of Sickle Cell Anemia to households: A case study of three sickle cell clinics in Ghana.

Tarricone, R. (2006). Cost-of-illness analysis. What room in health economics? *Health Policy (Amsterdam, Netherlands)*, *77*(1), 51–63. doi:10.1016/j.healthpol.2005.07.016

Woods, K., Karrison, T., Koshy, M., Patel, A., Friedmann, P., & Cassel, C. (2016). Association of Schools of Public Health Hospital Utilization Patterns and Costs for Adult Sickle Cell Patients in Illinois Hospital Utilization Patterns Costs for Adult Sickle MD Patients in Illinois, *112*(1), 44–51.

World Health Organisation [Accessed March 10, 2015] ;Fifty ninth World Health Assembly Sickle Cell Anaemia-report by the secretariat provisional agenda. 2006 11:4. items. Available from: http://www.who.int/pdffiles/WHA59/A59_9-en.pdf

World Health Organization. (2008). The Global Burden of Disease: 2004 update. *2004 Update*, 146. http://doi.org/10.1038/npp.2011.85

WHO Regionaloffice for Africa. Sickle cell disease prevention and control, 2015 http:/www.afro.who.int/en/nigeria/nigeria-publication/1775 . google scholar.

Yamane, Taro. 1967. *Statistics, An Introductory Analysis*, 2nd Ed., New York: Harper and Row.

Yawn B. P. et al. (2014). Management of sickle cell disease: summary of the 2014 evidence-based report by expert panel members. *Journal of American Medical Association.* 10;312 (10):1033-48. doi: 10.1001/jama.2014.10517.

APPENDICES

APPENDIX A: CONSENT FORM

Research Topic: Household costs of sickle cell disease among patients of sickle cell clinic, Tema General Hospital, the Greater Accra Region.

Introduction

My name is Dr. Lawrence Ofori-Boadu, a post-graduate student from the School of Public Health, University of Ghana. I am carrying out a study in this unit to find out the household cost of Sickle Cell Disease clients who report to this hospital/clinic and I would be very glad if you could participate in it. Your participation would involve a 30 minutes interview with me to answer some questions about yourself and how much you or your caretaker have spent on you.

You may not have any immediate or direct benefits from my interview but your responses would be helpful in policy planning and formulation of recommendations to appropriate authorities to help come out with better way to manage healthcare cost among SCD to reducing cost burden on households.

The only discomfort, if any that you would face by accepting to take part in this study perhaps is your time. If you indeed decide to take part, you are allow to withdraw whenever you wish to, and are also allowed to skip answering any of the questions that you are not very comfortable with.

The information you would provide is going to be treated with strict confidentiality. Apart from my research team and members of the ethics committee of GHS, nobody shall have access to the information since it shall be under lock and key. We also assure you that your name shall not appear or be mentioned in any report that will come out from this study.

Before taking Consent

Do you have any questions you wish to ask about the study? Yes [____] No [____]

If yes, please, indicate the questions below:

In case you have any questions later please, do not hesitate to contact Lawrence Ofori-Boadu, Department of Health Policy, Planning and Management (Tel 0244013372), School of Public Health, University of Ghana. Email: eo74ofori@yahoo.com.

For any further enquiry about research, kindly contact

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PARTICIPANT'S CONSENT

I have read or I have let somebody read or translated all the necessary information that I need to know concerning this study and have fully understood it. I have decided on my own accord without any coercion to take part in this study. However by deciding to participate in this study, I am not waiving any of my personal rights by signing or thumb printing this consent form.

Signature: OR

....

L/R Thumb Print

Interviewer's statement

I, the undersigned, have explained this consent to the subject in English language/Ga/Twi, and that she/he understands the purpose of the study, procedures to be followed, as well as the risks and benefits of the study. The participant has fully agreed to participate in the study.

Name and Signature of Interviewer.....

Date.....

APPENDIX B. ASSENT FORM

TITLE OF RESEARCH: HOUSEHOLD COST OF SICKLE CELL DISEASE AMONG PATIENTS OF SICKLE CELL CLINIC, TEMA GENERAL HOSPITAL, GREATER ACCRA REGION.

INVESTIGATOR: LAWRENCE OFORI-BOADU

The investigator named above is doing a research study.

These are some things we want you to know about research studies:

We are asking you to be in a research study. Research is a way to test new ideas. Research helps us learn new things.

Whether or not to be in this research is your choice. You can say Yes or No. Whatever you decide is OK. We will still take good care of you.

Why am I being asked to be in this research study?

You are being asked to be in the study because you have SCD.

What is the study about?

I need to learn more about the best way to manage cost of healthcare among young people with SCD and come up with the safest and best way of managing your household cost

What will happen during this study?

If you agree to be in this study, you will have no monetary reward.

What else should I know about the study?

If you feel sick or afraid that something is wrong with you, tell an adult at once. You do not have to answer any questions that are asked of you.

What are the good things that might happen?

People may have good things happen to them because they are in a research study. The doctors hope to learn about the best to manage household cost in children with SCD. The doctors might find out something that will help other children like you.

What if I don't want to be in this study?

You do not have to be in the study if you do not want to. The doctors will still take care of your condition. If you don't want to be in this study, you can continue to get your medical care at the sickle cell clinic.

Who should I ask if I have any questions?

If you have any questions about this study, you or your parents can call Lawrence OFORI-BOADU on 0244013372 or
Hannah Frimpong

GHS-ERC Administrator
Office: +233 302 681109
Mobile: **233 (0) 243235225 or 0507041223**
Email: Hannah.Frimpong@ghsmail.org

Signatures

Before deciding if you want to be in the study, ask any questions you have. You can also ask questions during the time you are in the study.

If you sign your name below, it means that you agree to take part in this research study.

_____	_____
Your Name	Age
_____	_____
Your Signature	Date
_____	_____
Signature of Person Obtaining Consent	Date
_____	_____
Signature of Witness	Date

APPENDIX C: QUESTIONNAIRE

Topic: Household Cost of Sickle Cell Disease among patients of Sickle Cell Clinic, Tema General Hospital, Greater Accra Region.

This is a research carried out on sickle cell disease clients who attend sickle cell clinic at Tema General Hospital. I will therefore like to take few minutes of your precious time to answer these questions. You are assured that answers you give will be strictly confidential and your name will not be mentioned in my response. Thank you

Qn. No.	Question	Response
		Respondent ID: |__|__|__|
Section 1	**SOCIO-DEMOGRAPHIC INFORMATION**	
1	What is your sex 1. Male 2. Female	☐
2	What is your age in years (i.e. age at last birthday)?	|__|__| years
3	What is the highest level of school you attended? 1.No education 2.Primary 3.Middle/JSS/JHS 4. SSS/SHS/Secondary/Vocational/Technical 5. Tertiary	☐
4	What is your current marital status? 1. Married 2. Not married	☐
5	What is your employment status? 1. Employed 2. Unemployed (*If **Unemployed**, answer Qs. 6 & 7)* 3. Student/Apprentice 4. Housewife 5. Retiree	☐
6	If Unemployed, why are you not working now? 1. Unable to work due to illness 2. Other (please specify).....	☐

7	If Unemployed, have you been looking for a job in the last 12 months? 1. Yes 2. No	☐
8	If employed, in which sector are you employed? 1. Formal sector 2. Informal sector	☐
9	If Employed, what is your average monthly income? (i.e., salary plus other monies from other sources)	GHS .....
10	If unemployed/minor what is the average income of your caregiver	
11	What level of social income status do you/ or your care takers belong to? 1. High 2. Medium. 3 Low	
12	Are you an NHIS beneficiary? 1. Yes 2. No	☐
Section 2	**HEALTH STATUS INFORMATION**	
	The recent Hemoglobin level checked is	☐ g/dL
13	What is your Sickling genotype 1. SS 2. SC 3..Others	☐
14	Have you any of the following complication over the past 12 months? (a) Hemolysis (cola-like urine) (b) Priapism (c) Painful crises (d) Osteomyelitis (e) Stroke (f) Bone fracture (g) Acute Chest Syndrome (Pneumonia) (h) Retinal disease (i) Others, please specify	☐ ☐ ☐ ☐ ☐ ☐ ☐ ☐
15	Have you been on admission over the past 12 months? 1. Yes 2. No	☐
16	If Q15 is YES, how many times have you been admitted during the last 12 months?	☐

17a	How many times have you attended SCD clinic in the past 12 months?	☐
17b	How many times have you attended clinic OUTSIDE TGH in the past 12 months?	☐
Section 3	**DIRECT COST INFORMATION**	
18	***Medical cost: how much have you spent during your hospital visit or stay in the management?***	GHS
	(a) Registration/Folders	☐
	(b) Consultation	☐
	(c) Laboratory investigations	☐
	(d) Medicines/drugs	☐
	(e) Other diagnostic tests (such as scan etc.)	☐
	(f) Other, specify: _____	
19	***Direct Non-medical cost: how much did you spend/pay for (you and accompanying relative)?***	GHS
	(a) Estimate of total travel cost (to and from the facility)	☐
	(b) Food cost	☐
	(c) Drinks/water cost	☐
	(d) Miscellaneous costs (i.e., phone calls/phone credits, other consumables used due to this illness)	☐
	(e) Others, kindly specify:	
20	Did you rely on financial help from other source(s) for treatment, apart from normal income? 1. Yes 2. No	☐
21	What are the sources (multiple responses possible)? 1. Relative 2. Friend 3. Savings 4. Loan/Grant 5. Other (Specify)	☐
22	How much money did you receive from the identified source(s)?	GHS.....

Section 4	INDIRECT COST INFORMATION	
23	How many **days** in total have you absented from work/school (if applicable) as a result of admission/attendance to clinic? (excluding travel and waiting time)	|__|__|__| days
24	How many minutes in total do you estimate to have spent travelling to and from the health facility?	|__|__|__| Mins.
25	How many minutes did you spend waiting before you were called to see the doctor or health officer for treatment?	|__|__|__| Mins.
26	Did anyone from your household accompany you from home to the health facility? 1. Yes 2. No	☐
27	If anyone did accompany you to the health facility, what is his or her employment status? 1. Employed 2. Unemployed 3. Student 4. Housewife	☐
28	How many days did the person who accompanied you absented from work/ school?(**excluding travel and waiting time)**	|__|__|__| Days
29	Did the person who accompany you, come with you from the house and stay with you for treatment and take you back home? 1. Yes 2. No	☐
30	How many hours/minutes in total did he/she travel to and fro to be with you in the health facility?	|__|__|__| Mins.
31	How many hours/minutes/days in total did he/she spend with you when you were receiving treatment in the health facility?	|__|__|__| Mins/hours/Days
32	Total time spent	|__|__|__| Mins/hours/Days

Section 5	INTANGIBLE COST INFORMATION	
	Please, rate the following statements concerning STIGMATIZATION	
33	Because of SCD people look down on me 1. Not at all 2. A little 3. Moderately 4. Quite a bit 5. Extremely	
34	I am discrimination in terms of marriage partner I can choose 1. Not at all 2. A little 3. Moderately 4. Quite a bit 5. Extremely	
	Please, rate the following statements concerning SOCIAL-ISOLATION	
35	Because of the SCD I am deprive access to certain sporting activities at school 1. Not at all 2. A little 3. Moderately 4. Quite a bit 5. Extremely	
36	I think I am sidelined by others because of my illness 1. Not at all 2. A little 3. Moderately 4. Quite a bit 5. Extremely	
	Now kindly rate again, the following statements concerning PAIN.	
37.	How much physical pain do you suffer 1. Not at all 2. A little 3. Moderately 4. Quite a bit 5. Extremely	

38	Does painful crises incapacitate you from embarking on physical activity? 1. Not at all 2. A little 3. Moderately 4. Quite a bit 5. Extremely	
	Please rate the following statements concerning STRESS	
39	I suffer emotionally because I constantly become depressed 1. Not at all 2. A little 3. Moderately 4. Quite a bit 5. Extremely	
40	Frequent painful crises stress me up. 1. Not at all 2. A little 3. Moderately 4. Quite a bit 5. Extremely	
41	My episodic anemia state with blood transfusion stress me up. 1. Not at all 2. A little 3. Moderately 4. Quite a bit 5. Extremely	
	Now kindly rate, the following statements concerning SELF-ESTEEM	
42	I feel I do not have much to be proud of. 1. Not at all 2. A little 3. Moderately 4. Quite a bit 5. Extremely	
43	I think that overall, people find me boring to talk to. 1. Not at all 2. A little 3. Moderately 4. Quite a bit 5. Extremely	

44	I feel that I'm a person of worth, at least on an equal plane with others. 1. Not at all 2. A little 3. Moderately 4. Quite a bit 5. Extremely	

APPENDIX D: ETHICAL APPROVAL LETTER

GHANA HEALTH SERVICE ETHICS REVIEW COMMITTEE

In case of reply the number and date of this Letter should be quoted

Research & Development Division
Ghana Health Service
P. O. Box MB 190
Accra
Tel: +233-302-681 109
Fax: + 233-302-683 624
Email: ghsers@gmail.com

My Ref: GHS/RD/ERC/Admin/Conf/vol.v/53
Your Ref: No.

Lawrence Ofori-Boadu
University of Ghana
School of Public Health
Legon, Accra

The Ghana Health Service Ethics Review Committee has reviewed and given approval for the implementation of your Study Protocol.

GHS-ERC Number	GHS-ERC: 109/04/17
Project Title	Household Cost of Sickle Cell Disease among Patients at Sickle Cell Clinic, Tema General Hospital, Greater Accra Region
Approval Date	24th April, 2017
Expiry Date	23rd April, 2018
GHS-ERC Decision	Approved

This approval requires the following from the Principal Investigator

- Submission of yearly progress report of the study to the Ethics Review Committee (ERC)
- Renewal of ethical approval if the study lasts for more than 12 months,
- Reporting of all serious adverse events related to this study to the ERC within three days verbally and seven days in writing.
- Submission of a final report after completion of the study
- Informing ERC if study cannot be implemented or is discontinued and reasons why
- Informing the ERC and your sponsor (where applicable) before any publication of the research findings.

Please note that any modification of the study without ERC approval of the amendment is invalid.

The ERC may observe or cause to be observed procedures and records of the study during and after implementation.

Kindly quote the protocol identification number in all future correspondence in relation to this approved protocol

SIGNED .....
PROFESSOR MORRIS AIKINS
(GHS-ERC VICE-CHAIRPERSON)

Cc: The Director, Research & Development Division, Ghana Health Service, Accra