A MODEL FOR ENHANCEMENT OF SELF-CONCEPT OF PEOPLE WITH ALBINISM

by

MMUSO BARBARA JOAN POOE-MONYEMORE

submitted in fulfilment of the requirements

for the degree of

DOCTOR OF LITERATURE AND PHILOSOPHY

in the subject

HEALTH STUDIES

at the

UNIVERSITY OF SOUTH AFRICA

PROMOTER: PROF TR MAVUNDLA

JOINT PROMOTER: PROF AL CHRISTIANSON

June 2007

DECLARATION

I declare that **A MODEL FOR ENHANCEMENT OF SELF-CONCEPT OF PEOPLE WITH ALBINISM** is my own work and that all the sources that I have used or quoted have been indicated and acknowledged by means of complete references and that this work has not been submitted before for any other degree at any other institution.

Signature:-----------------------------------   **Date:**--------------------------------

MBJ Pooe-Monyemore

A MODEL FOR ENHANCEMENT OF SELF-CONCEPT OF PEOPLE WITH ALBINISM

STUDENT NUMBER: **683-487-6**

STUDENT: **MBJ POOE-MONYEMORE**

DEGREE: **DOCTOR OF LITERATURE AND PHILOSOPHY**

DEPARTMENT: **HEALTH STUDIES, UNIVERSITY OF SOUTH AFRICA**

PROMOTER: **PROF TR MAVUNDLA**

JOINT PROMOTER: **PROF AL CHRISTIANSON**

ABSTRACT

The purpose of this study was to develop and describe a model for the enhancement of self-concept of people with oculocutaneous albinism based on the experience of the participants. The need to conduct the study was triggered by the researcher's long-term interaction with people with oculocutaneous albinism, and observation that they are still stigmatised and marginalised by the community.

A purposive sample of fifteen participants, consisting of twelve females and three males with oculocutaneous albinism, aged between 18 and 48, participated in the study. The study was conducted in urban and semi-urban areas around the Gauteng Province in South Africa.

The study had two theory generative objectives, namely to identify the main concepts of the model from the findings, and to describe the model structure and process. A theory generative, qualitative, explorative, descriptive and contextual research design was used to achieve these objectives. Chinn and Kramer's (1995:92) four steps of theory-generation were used, namely concept analysis, construction of theoretical relationships, description of the model, and development of guidelines to operationalise the model.

Concept analysis involved identification, definition and classification of the concepts to develop the model. In-depth, phenomenological interviews were conducted to explore the participants' experience. Data analysis and interpretation revealed three main themes from the findings: the participants' perception of the self; experience with the external environment, and expression of need for development and growth. The main concepts deducted from the findings were **"self-concept"** and **"enhancement"** hence *"A model for enhancement of self-concept of people with albinism".*

Construction of theoretical relationships entailed using deductive reasoning to describe the interaction between the concepts of the model.

Description of the model entailed visually portraying and describing the structure and process of the model, as well as evaluating it.

Developing guidelines to operationalise the model entailed describing practical strategies to assist health professionals to enhance the self-concept of people with oculocutaneous albinism.

KEY CONCEPTS

Birth defects, congenital disorders, enhancement, oculocutaneous albinism, qualitative research, self-concept, and phenomenology.

ACKNOWLEDGEMENTS

There is an expression that no feast comes to the table on its own feet, and it can equally be said that no thesis comes to the shelf on its own wings. Accordingly, I am thankful to my God and Creator, from whom all blessings flow, for sending His earthly angels to carry me throughout this study. Special thanks, then, to the following people for their unwavering support:

- Prof TR Mavundla, my promoter, and Prof AL Christianson, my joint promoter, for their expert guidance
- The National Department of Health, for offering me a bursary for my doctoral studies.
- My mother, Sophie Baatseba Pooe, for being my source of inspiration
- My husband, Noah Andrew Monyemore, for his unconditional love and incredible patience, and my son, Thuto, for his love and understanding
- My sister, Mapilane, and brothers, Itumeleng and Nick, and their families as well as my in-laws for their constant encouragement and patience when I could not attend family events
- Mr Tony Ngwenya, Mrs Nomasonto Mazibuko and Ms Poppy Neke-Smith of the Albinism Society of South Africa, for their encouragement
- The research participants and their families, for their courage, acceptance and valuable inputs
- My spiritual mentor and dear friend, Dr Anniekie Ravhudzulo, for her special prayers and reassurance
- UNISA library staff members, especially Ms Talana Burger (Health Studies librarian), for their patience and help at all times
- Ms Lindi Madiseng and Mr Simon Jiane, for assisting me with procedures at UNISA
- Dr Sebolelo Amos, for her motherly love and encouragement, and Dr Nat Khaole, for his motivation and understanding
- Prof Jennifer Kromberg and Prof Jacquie Greenberg, for their encouragement
- Ms Iauma Cooper, for her patient and critical editing of the manuscript
- Ms Rina Coetzer, for her unbelievable patience to assist with the model and bibliography
- Drs T Modungwa, T Mabe, S Siphepho and SP Mkhabela, for evaluating the model
- My colleagues in the Cluster: Maternal, Child, Women's Health and Nutrition and Directorates: Health Promotion and National Human Resource Strategic Programmes.

KE A LEBOGA BAGAETSHO. MOTHO KE MOTHO KA BATHO BA BANGWE!
(Translation: Thank you fellow human beings. Support from other people is essential to one's success!)

DEDICATION

This thesis is dedicated to

- o *My mother, Sophie Baatseba Pooe*
- o *My late father, Ezekiel Mafatshe Pooe*
- o *My husband, Noah Andrew Monyemore*
- o *My son, Thuto*
- o *The research participants and the Albinism Society of South Africa*

TABLE OF CONTENTS   Page

TABLE OF CONTENTS   **Page**

TABLE OF CONTENTS   Page

TABLE OF CONTENTS Page

TABLE OF CONTENTS   **Page**

TABLE OF CONTENTS   Page

TABLE OF CONTENTS   **Page**

TABLE OF CONTENTS **Page**

TABLE OF CONTENTS **Page**

TABLE OF CONTENTS **Page**

TABLE OF CONTENTS   **Page**

TABLE OF CONTENTS   Page

LIST OF TABLES   **Page**

LIST OF FIGURES   Page

LIST OF ANNEXURES

Annexure A   ....Clearance certificate received from UNISA

Annexure B   ....Permission letter from the Albinism Society of South Africa

Annexure C   .... Request letter sent to the Albinism Society of South Africa

Annexure D   ....Consent letter

Annexure E   ....In-depth interview guide

CHAPTER 1

INTRODUCTION AND OUTLINE OF THESIS

"God gives us different passions so that everything He wants done in the world
will get done"   **Rick Warren**

1.1   INTRODUCTION

The diagnosis of a congenital disorder has lifelong medical and psychosocial implications for the person with the condition. Oculocutaneous albinism is a congenital disorder characterised by deficiency of a pigment called melanin in the skin, hair and eyes. Oculocutaneous albinism is therefore easily recognisable in dark-skinned people due to lack of the skin pigment (Christianson, Howson & Modell 2006:25).

Due to lack of this protective melanin, the person with oculocutaneous albinism has an increased risk of sun-induced skin disorders, including skin cancer. In addition, as a result of lack of the eye pigment, the affected person may also present with sensitivity to light referred to as photophobia, rapid eye movements referred to as nystagmus, and visual acuity (Christianson et al 2006:25; Winship 2003:174).

More often, the person has to cope with the with oculocutaneous albinism as a congenital disorder, and lack of knowledge and attitude of the community. A theory-generating phenomenological qualitative study was thus conducted to explore and describe the experience of people with oculocutaneous albinism. The main purpose of the study was to develop and describe a model for enhancement of self-concept of people with oculocutaneous albinism. The study was conducted around the urban and semi-urban areas of the Gauteng Province in South Africa.

This chapter describes the background and motivation for conducting the study, the problem statement, research purpose and objectives, the paradigmatic perspective, the research design and methodology, ethical requirements as well as measures for establishing trustworthiness of the study.

It should be noted that because of the qualitative nature of this study, the statement of the research problem, and purpose and objectives of the study are discussed before presenting the background. This was done to describe and understand the problem of oculocutaneous albinism upfront.

1.2   STATEMENT OF THE RESEARCH PROBLEM

Polit and Beck (2004:731) define a problem statement as" a situation involving an enigmatic or perplexing condition that can be investigated through a disciplinary inquiry". This section discusses the nature of the problem, the intention of the study as well as the research questions.

1.2.1   Nature of the problem

In her long-term interaction with people with oculocutaneous albinism, the researcher observed that they were stigmatised and marginalised. Discrimination is believed to be one of the key manifestations of stigmatisation. The *Save the Children Research Report* (2001:29) emphasises that stigma and discrimination have an insidious impact on the dignity and self-respect of the person being stigmatised. This, in turn, impacts on that person's self-image and self-confidence.

The researcher also observed with concern that despite the efforts of the Department of Health in South Africa to raise awareness and train health professionals, there is still a lack of knowledge on oculocutaneous albinism by both the health professionals and the community. Lack of knowledge, in turn, results in the community having negative attitude towards people with oculocutaneous albinism.

In most cases, the researcher observed that the negative attitude of the community was due to myths and superstitions about oculocutaneous albinism. Without doubt, this situation could result in low self-esteem, loneliness and isolation of the person. Isolation exacerbates the situation of affected persons, because it means that they may not able to access the essential services, such as health and educational facilities to meet their physical and developmental needs.

As marginalised individuals, people with oculocutaneous albinism may also not be able to attend social or religious gatherings to address their spiritual and psychological needs.

This is a vicious cycle that infringes on the Constitutional rights of people with oculocutaneous albinism. Section 27(1)(a) and (c) of the Bill of Rights of the Constitution of South Africa Act, 108 of 1996, emphasises that everyone has a right to access to health and welfare services (South Africa 1996:13).

In this regard, the researcher had a typical case in 1995 in a town in the North-West Province, where the rights of a learner with oculocutaneous albinism were infringed. A sixteen-year old learner with oculocutaneous albinism stayed away from school because of being ridiculed and called names. The mother then approached the genetics clinic to intervene. The researcher, who was a genetic nurse counsellor at the genetics clinic, and two social workers, visited the particular school to assess the situation so as to intervene accordingly. During discussions with the educators and learners, it emerged clearly that the most significant challenge was the myths and superstitions regarding oculocutaneous albinism. These myths and superstitions stigmatised the learner concerned. After further probing discussions with the educators and learners, an educational intervention was provided for them. Demystification of oculocutaneous albinism was successful in that the concerned learner returned to school.

In addition, as an experienced health professional working in the field of Human Genetics since 1989, with seventeen years' experience in this field, the researcher observed that one of the major challenges in the health system is the non-availability of reliable data on congenital disorders, including oculocutaneous albinism. Non-availability of reliable data negatively influences policy development and implementation programmes on congenital disorders, as well as budget allocation for programmes on congenital disorders. Most importantly, this affects the quality of care to be given to people with oculocutaneous albinism.

The above matters were therefore major concerns for the researcher, and this led to the development of the relevant research questions.

1.2.2 Research questions

The researcher wished to answer the following research questions:

- What is the life experience of people with oculocutaneous albinism and what does it mean to them to have the condition?
- What could be done in the clinical practice to improve the quality of life of people with oculocutaneous albinism?

Answering these questions meant exploring and describing the life-world of people with oculocutaneous albinism, for the purpose of developing and describing an appropriate model.

1.3 PURPOSE OF THE STUDY

In accordance with the research questions, the main purpose of this study was to develop and describe a model for enhancement of self-concept of people with oculocutaneous albinism based on their life-experience.

1.4 RESEARCH OBJECTIVES

To achieve the purpose, the study wished to meet the following objectives:

1.4.1 Identify the main concepts

In order to identify the main concepts of the model, the researcher explored and described the life-experience of people with oculocutaneous albinism.

1.4.2 Describe the model structure and process

This objective was achieved by

- Classifying the concepts of the model according to recipient, agent, context, dynamics, procedure and outcome/terminus.
- Describing the structure and process of the model.
- Formulating and describing guidelines for operationalisation of the model in practice.

1.5   BACKGROUND TO THE PROBLEM

This section discusses the stimulus to conduct the study based on the frequency of oculocutaneous albinism and its impact on health care services. Preliminary literature review is also discussed in this section.

1.5.1   Frequency of oculocutaneous albinism and impact on health care services

It is essential to first understand the genotype or genetic makeup of oculocutaneous albinism before discussing its frequency. Oculocutaneous albinism is inherited in an autosomal recessive manner. This implies that both parents appear unaffected but carry one defective gene of oculocutaneous albinism, and one normal gene each. The person with oculocutaneous albinism inherits two defective genes, one from each parent (Christianson et al 2006:25).

Mutations in seven different genes cause oculocutaneous albinism. Mutation is a change in the structure of deoxyribonucleic acid (Winship 2003:11). One form of oculocutaneous albinism, the tyrosinase positive oculocutaneous albinism (Type 2), is reported as the most common type among Southern African Blacks, and affects the skin, eyes and hair (Kerr, Stevens, Manga, Salm, John, Haw & Ramsay 2000:166). Type 2 oculocutaneous albinism is due to a mutation in the P-gene (small arm) on Chromosome 15 (Online Mendelian Inheritance in Man 2000).

Oculocutaneous albinism is found throughout the world, but the prevalence in different population groups varies widely (Kagore & Lund 1995:859). Delport, Christianson, van den Berg, Wolmarans and Gericke (1995:14) found a birth prevalence of 0.66 per 1,000 live births of oculocutaneous albinism in a rural area in South Africa. Venter, Christianson, Hutamo, Makhura, & Gericke (1995:15) recorded 0.23 per 1,000 live births of oculocutaneous albinism in an urban area in South Africa.

In the United State of America, the prevalence of oculocutaneous albinism among whites was estimated at 1 in 36 000, whereas, in certain isolated groups, such as the Hopi Indians in Arizona, oculocutaneous albinism is exceptionally high, and was estimated at 1 in 277 (Kagore & Lund 1995:859 citing Witkop, Quevedo, Fitzpatrick and King 1989:2905).

The estimates of oculocutaneous albinism among the Black population in Dar-es-Salaam, Tanzania was 1 in 1429, and indicates a high rate, compared to a prevalence rate in Soweto, South Africa estimated at 1 in 3900 (Kromberg & Jenkins 1982:384). Oculocutaneous albinism occurs in every ethnic or population group. However, certain ethnic groups show a higher prevalence as a result of traditional marriage patterns of consanguineous unions (Christianson et al 2006:21). Consanguinity refers to marriage between people who are blood relatives, such as cousins. First cousins share one-eighth of their genes, and are therefore at a greater risk of having the same defective gene (Winship 2003:14).

In this regard, in the early 1980s Kromberg and Jenkins (1982:385) found oculocutaneous albinism more common in the Southern Sotho and Tswana ethnic group compared to the Nguni ethnic group in Soweto, South Africa. The Southern Sotho and Tswana people appear to prefer a consanguineous marriage since among the Tswanas almost 42%, and among the Southern Sothos almost 27% have consanguineous relationships, compared to only 4% of Zulu people having consanguineous relationships (Kromberg & Jenkins 1982:385). However, the preference of consanguineous marriage among the Southern Sotho and Tswana people needs further investigation. It is also evident from this discussion that there is paucity of current epidemiological research on oculocutaneous albinism in South Africa, and most original research on this topic was undertaken by Kromberg and others in the early 1980s.

Nevertheless, congenital disorders are a global problem, and Christianson et al (2006:10) recently identify the previously hidden toll of congenital disorders. Christianson et al (2006:10) describe the global extent of the seriousness of congenital disorders, and how the impact of congenital disorders is still an unappreciated public health problem.

Christianson et al (2006:2) emphasise that the impact of congenital disorders is particularly severe in middle and low-income countries, where more than 94% of births with serious congenital disorders, and 95% of the deaths of these children occur. According to the World Bank (2005), middle and low -income countries are sometimes referred to as developing countries, and these countries have gross national income per capita of $826-10,065 and less than $826, respectively. An industrialised or high-

income country has a gross national income per capita of more than $10,065. South Africa is classified as a middle-income country.

Congenital disorders are more common in developing than in the industrialised countries, and their health burden has consistently been underestimated (World Health Organization (WHO) 1996:27; WHO 2000:17; WHO/March of Dimes 2006:12-15).

These reviews emphasise that relatively simple interventions can significantly reduce the burden of congenital disorders. The interventions include training of health professionals on congenital disorders, educating the public about congenital disorders, and establishing appropriate child health services to care for infants and children with congenital disorders (WHO 1996:27; WHO 2000:17 and WHO/March of Dimes 2006:12-15).

Furthermore, according to these reviews, there is evidence of enormous progress in medical genetics, and the care and prevention of congenital disorders in the industrialised nations. However, there is little or no impact of medical genetic knowledge and technology in the developing countries, where more than 80% of the world population is born and lives (Christianson & Modell 2004:6; WHO/World Alliance of Organisations for the Prevention of Birth Defects Report (WAOPBD) 1999:5; WHO/March of Dimes 2006:12-15).

The researcher therefore noted the technical knowledge on the genotype and cause of oculocutaneous albinism as a congenital disorder, its frequency and impact on the health care system as a challenge for South Africa as a developing country. Accordingly, the researcher identified a need to explore the life experience of people with oculocutaneous albinism and to develop a model that would serve as a framework for the health professional to enhance the self-concept of people with oculocutaneous albinism.

1.5.2 Preliminary literature review

Kromberg, Jenkins, Zwane, and Castle conducted several relevant original studies on oculocutaneous albinism in South Africa between 1984 and 1997. Other studies were conducted on congenital disorders and disabilities relevant to oculocutaneous albinism

(Ojofeitimi & Elegbe 1984; Bhagwanjee & Sterwart 1999*; Gaigher, Lund & Makuya 2002; Mweshi & Mpofu in 2001; Moodley 2003).

Despite the fact that some of the studies are not current, it is evident that international and national studies have been conducted on congenital disorders, oculocutaneous albinism and disabilities. It is also important to note that research to date on oculocutaneous albinism has looked mainly at its epidemiology, clinical features and to limited extent psychosocial implications.

Most importantly, little, if any research has been conducted to explore the life-world of persons with oculocutaneous albinism for the purpose of developing and describing a model for enhancement of their self-concept. The present study therefore took up Bhagwanjee and Stewart's (1999:15) challenge:

> The onus now rests primarily with researchers themselves to rise
> to the challenge of producing high quality research that will change
> the traditional conceptual models, influence future service provision
> programmes, address professional paradigm affecting the perception
> of disability issue and empower people with disabilities

In addition, the WHO/WAOPBD (1999:61-62) recommendations also emphasise the need to conduct this study:

- Research is an important component of medical genetics in developing countries, especially as insufficient data is available on the epidemiology of congenital disorders, the expectations of the population and the outcomes of genetic services; and
- Health services research with the purpose of finding new approaches to prevention and treatment of congenital disorders should be supported.

1.6 PARADIGMATIC PERSPECTIVE OF THE RESEARCH

Polit and Beck (2004:726) describe a paradigmatic perspective as "a way of looking at natural phenomena that encompasses a set of philosophical assumptions, and that guides the researcher's approach to inquiry". LoBiondo-Wood and Haber (2002:127) point out that "paradigm" comes from a Greek word meaning "pattern".

McKenna (1997:80) describe assumptions as "those basic givens, or accepted truths that are fundamental to theoretic reasoning, and are central components of a theory or model". The assumptions of this study were thus related to the process of enhancing the self-concept of people with oculocutaneous albinism, and were congruent with Neuman's system model, critical theory and Wiedenbach's prescriptive nursing theory as well as the rehabilitation self-empowerment assumptions (Burns & Grove 2001:77; Neuman 1995:18; Wiedenbach 1970:1057) (see chapter 5).

This study also used the assumptions of the naturalistic paradigm to address the basic philosophical questions (see table 1.1).

Table 1.1 Major assumptions of naturalistic paradigm

Assumption	Naturalistic paradigm
Ontologic What is the nature of reality in this study?	Reality of the life-world of people with oculocutaneous albinism was explored.
Epistemiological What is the relationship between the inquirer and that being researched?	The researcher conducted in-depth face-to-face interviews. Subjective interaction was thus facilitated, and it was assumed that knowledge is maximised in the findings.
Axiologic What is the role of values in the inquiry?	Subjective interaction and values were inevitable and essential in this study.
Methodology How should the inquirer obtain knowledge?	A phenomenological qualitative research method was used to conduct the study. The respondents shared their life experiences of oculocutaneous albinism.

Polit and Beck (2004:14) describe the naturalistic paradigm as an outgrowth of the pervasive cultural transformation referred to as postmodernism. According to Polit and Beck (2004:14), post-modern thinking emphasises the value of *deconstruction*, by taking apart old ideas and structures, and *reconstruction*, by putting ideas and structures together in new ways. Therefore, according to the post-modern thinking,

concepts were identified from the research findings. The concepts were then utilised as building blocks of the model for enhancement of self-concept of people with oculocutaneous albinism.

1.6.1 Meta-theoretical assumptions

According to Botes (1995:6), meta-theoretical assumptions are not testable, deal with the human being and society, and have their origin in philosophy. Botes (1995:6) emphasises that meta-theoretical or metaparadigm assumptions influence the research decisions throughout, and serve as a framework within which theoretical statements are made.

In this regard, the meta-theoretical assumptions reflected the researcher's belief regarding the experience of people with oculocutaneous albinism, and the need to enhance their self-concept. In the context of this study, the four concepts of nursing metaparadigm, namely, person, environment, health and nursing are described as follows:

1.6.1.1   Person

Person refers to an individual with oculocutaneous albinism who is in constant interaction with the environment, in which he or she may be stigmatised and prejudiced. Stigmatisation affects an individual's self-image, hence the need to develop a model that will enhance the self-concept of people with oculocutaneous albinism.

1.6.1.2   Environment

Environment refers to the internal and external factors or influences that surround the person with oculocutaneous albinism, and affect the person's life and development. Internal environment refers to an individual's inner strength, and external environment refers to external forces and interactive influences in the community. In this regard, the 'self-concept' of the person with ocolucutaneous albinism and the community's perception of oculocutaneous albinism have an important impact on the individual's self-image.

1.6.1.3   Health

The health of people with oculocutaneous albinism is viewed in response to the internal and external environments and the nursing care provided. People with oculocutaneous albinism are vulnerable to skin cancer due to the sensitivity of their skin to sunrays. Availability and utilisation of health policies and management guidelines will also have an impact in the health care system of people with oculocutaneous albinism. The knowledge of health care professionals on oculocutaneous albinism will enable them to provide quality health care to people with oculocutaneous albinism.

1.6.1.4   Nursing

Nursing care in this model is essential as the self-image of the person with oculocutaneous albinism will be assessed. Appropriate nursing intervention strategies will be used, so that the person with oculocutaneous albinism can maintain his or her integrity.

These meta-theoretical assumptions deal with human beings and society, and therefore provided an essential framework for the theoretical assumptions of this study.

1.6.2 Theoretical assumptions

Theoretical assumptions are testable, offer epistemic pronouncements about the research field, and give shape to the conceptual framework of the research and proposed model (Botes 1995:6). The theoretical assumptions of this study were also guided by Neuman's systems model; critical theory; and the definitions of terms.

It should be noted that although Neuman's systems model and critical theory were chosen as a point of departure for this study, the researcher entered the field with an open mind by applying bracketing and intuiting reasoning strategies (Burns & Grove 2001:606). Precautions were therefore taken not to force the research findings to fit within Neuman's systems model and critical theory. The research findings could only be contextualised within the relevant assumptions after data analysis.

1.6.2.1 Neuman's systems model

Neuman, a nursing theorist, indicates that the systems model depicts an open system in which people with oculocutaneous albinism and their environment are in dynamic interaction to maintain harmony. According to the Neuman's systems model, a person with oculocutaneous albinism as the client system is composed of five interacting dimensions (see table 1.2), namely physiological, psychological, socio-cultural, spiritual and developmental (Neuman 1995:10; Stanhope & Lancaster 2000:208).

The researcher considered these dimensions the basic needs of people with oculocutaneous albinism. According to Neuman (199510-15):

- Physiological dimension refers to the structure and functions of the body;
- Psychological dimension refers to the mental processes and relationships;
- Socio-cultural dimension refers to the system functions that relate to social and cultural expectations and activities;
- Spiritual dimension refers to the influence of spiritual beliefs; and
- Developmental dimension refers to those processes related to development over the lifespan.

Table 1.2 Definition of systems dimensions

Dimension	Definition	Examples
Physiological	Structures and function of the body/community	*Government programmes on oculocutaneous albinism and other congenital disorders *Transport system
Psychological	*Mental process and relationships *Cognitive and affective characteristics	*Happy/depressed *Intelligence level *Isolation of affected persons
Socio-cultural	*Refers to the systems functions *Social, economic, demographic, political, recreational, cultural, health characteristics and communication patterns/activities among subsets	*Communication patterns *Clinics/hospitals *Poor/middle class/affluent
Spiritual	*Moral, religious, and value systems of the individual *Influence of spiritual beliefs	*Moral-spiritual aspects on oculocutaneous albinism and other congenital disorders *Churches' role
Developmental	*Refers to processes related to development over lifespan *History, stage, and evolution of subsystems and aggregates in community.	*National registry of homes and education in the community.

Source: Neuman (1995)

To support Neuman's systems model, King maintains that each individual is a personal system, of which the relevant concepts are self, perception, growth and development, body image, space, learning and time. In this regard, perception is presented as the major concept of the personal system, as it influences all behaviours or to which all other concepts are related. King (1990:25) indicates further that perception is typically action oriented.

The researcher considered critical theory, with its characteristics of action feature, ideal to complement Neuman's systems model in this study.

1.6.2.2   *Critical theory*

According to Stanhope and Lancaster (2000:299), the aims of critical theory are to increase the depth of knowledge to shape reality and initiate action, and are concerned with a critique of society with envisioning new possibilities. This is in line with the post-modern thinking paradigm, which emphasises that knowledge can fit into new channels, and become operational, only if learning is translated into quantities of information. In this study the findings initiated action to develop and describe a model for enhancement of self-concept of people with oculocutaneous albinism.

Burns and Grove (2001:77) add that in critical social theory, the researcher attempts to uncover the distortions and constraints that impede free, equal, and uncoerced participation in society. Through this study, possible power imbalances for people with oculocutaneous albinism were exposed.

Burns and Grove (2001:77) cite Berman et al (1996) who describe the fundamental aims of critical social theory as follows:

- Research using critical social theory addresses an issue that is of concern to a group that is disadvantaged, oppressed or marginalised. People with oculocutaneous albinism have been generally marginalised (see section 1.2 and 1.5).
- The research process or results have the potential to benefit the group, immediately or in the longer term.
- People with oculocutaneous albinism will enjoy the long-term benefit of the study because the researcher also provided practical guidelines to operationalise the model for enhancement of self-concept of people with oculocutaneous albinism.

1.6.2.3   Definition of terms

For the purpose of this study, the following key terms are used as defined below.

- **Birth defects**

Birth defects are any abnormalities of the body structure or function that are present from birth. They can result from abnormalities of the genetic material, single gene defects or chromosomal abnormalities or when a combination of genes interacting with foetal environmental factors causing a multifactorial congenital malformation. These are genetic birth defects (Christianson et al 2006:14).

Post-conception, in which case the genes and chromosomes are normal, *birth defects* are caused by teratogens, physical or chemical agents that damage the embryo or fetus, and from intrauterine abnormalities that deform or disrupt the normal development of the fetus (Christianson et al 2006:14).

Some *birth defects* are visible at birth, for example oculocutaneous albinism, Down syndrome and neural tube defects, while others present clinically later in life, such as muscular dystrophy and Huntington's disease (Christianson et al 2006:14).

- **Congenital disorders**

The WHO (WHO/March of Dimes 2006:3) defines c*ongenital disorders* as "any potential pathological condition arising before birth. This includes all disorders caused by environmental, genetic and unknown factors, whether they are evident at birth or become manifest later in life."

According to the WHO (WHO/March of Dimes 2006:3), the definitions for the terms *birth defects* and *congenital disorders* were identical, and therefore they are interchangeable. In this study, the researcher therefore decided to use the term c*ongenital disorder.*

- **Enhancement**

Enhancement is a noun derived from the verb "enhance". *Collins English Dictionary* (2006:260) defines *enhance* as "to improve or increase in quality, value, or power".

In this study, *enhancement* is discussed in chapter 4, using various sources to identify its attributes to develop and describe the model for enhancement of the self-concept of people with oculocutaneous albinism.

- **Model**

LoBiondo-Wood and Haber (2002:110) define a *model* as "a symbolic representation of a set of concepts to represent an empiric experience".

- **Self-concept**

Blackwell's Nursing Dictionary (2005:545) defines *self-concept* as "the mental image one has, based on one's ideas and attitudes about oneself and one's personality". Chapter 4 provides comprehensive definitions of *self-concept* and the related attributes used to develop the proposed model.

1.6.3 Methodological assumptions

Botes (1995:7) states that methodological assumptions concern the researcher's view of the nature and structure of science and research in the relevant discipline, and direct the research design. The central methodological assumption in this study was that research should produce new knowledge on oculocutaneous albinism, and progress beyond the level of mastering the research procedures, but should be to the point of making a unique contribution to the nursing practice by developing a model.

It was therefore essential for the researcher to adopt a research design that was suitable to develop the proposed model.

1.7 RESEARCH DESIGN AND METHODOLOGY

Polit and Beck (2004:730) describe a research design as "an overall plan for addressing the research questions, including specifications for enhancing the integrity of the study". The researcher used a theory-generating research design to address the research questions. A theory-generating research design is qualitative, explorative, descriptive and contextual in nature (Chinn & Kramer 1999:133). In this regard, the researcher first explored and described the experience of people with oculocutaneous albinism so as to identify the main concepts of the model.

The researcher therefore used Chinn and Kramer's (1995:92) four steps of theory generation by to conduct the study, namely
- Concept analysis (concepts were identified, defined and classified);
- Construction of relationship statements;
- Description of the model; and
- Description of practical guidelines for operationalisation of the model.

Polit and Beck (2004:729) describe qualitative research as "an investigation of phenomena, typically in an in-depth and holistic fashion, through the collection of rich narrative materials, using a flexible research design". In-depth phenomenological interviews were used as data-collection techniques.

Fain (2004:220) emphasises that a qualitative phenomenological method aims at gaining a deeper understanding of the nature or meaning of the person's everyday experiences. On the other hand, quantitative research is an investigation of phenomena that lend themselves to precise measurement and quantification, and often involves a rigorous and controlled design (Polit & Beck 2004:729).

1.8 REASONING STRATEGIES

Bracketing, intuiting, analysis, inductive reasoning, synthesis and deductive reasoning were used as the reasoning strategies of this study (see table 1.3). These reasoning strategies were essential to facilitate logical arguments to assist with the exploration of life-experience of people with oculocutaneous albinism, and to develop and describe the model from the research findings (Polit & Hungler 1999:9).

Table 1.3 shows the relationship between the theory generation steps, the research design and the reasoning strategies.

Table 1.3 Overview of theory generation, research design and reasoning strategies

	Approach / method	Reasoning Strategies
STEP 1 Creating conceptual meaning	1.1 Concept identification* • Used purposive sampling • Conducted phenomenological in-depth interviews with people with oculocutaneous albinism • Data analysis: Collaizi phenomenological research method used (LoBiondo-Wood and Haber 2002:146) • Findings: used research findings to identify themes -Literature control done to compare the findings • Deducted main concepts of the model from findings	1. Bracketing 2. Intuiting 3. Inductive reasoning 4. Deductive reasoning 5. Analysis 6. Synthesis
	1.2 Concept analysis *concept analysis of key concepts done, using various methods (Chinn & Kramer 1999:81)	Analysis
	1.3 Concept definition (Theoretical) *put together defining attributes of concepts Results: well defined concepts	Synthesis
STEP 2 Interrelationship statements	Put concepts in relation with one another, and relationship statements from the model	Synthesis
STEP 3 Description of model	*Used Chinn and Kramer's (1995:107-119) theory description guidelines to describe the model *Evaluated the model using various strategies for theory evaluation (Chinn & Kramer 1999:119 and McKenna 1997:230)	Deductive reasoning
STEP 4 Developed practice guidelines	Developed and formulated guidelines to operationalise the model in practice. Used the model and literature	Deductive reasoning

1.9   ETHICAL REQUIREMENTS

Polit and Beck (2004:717) describe ethics as "a system of moral values that is concerned with the degree to which the research procedures adhere to professional, legal, and social obligations to protect the rights of the study participants". Ethical requirements pertinent to this study included obtaining approval to conduct the research, informed consent, privacy and confidentiality.

- The Research and Ethics Committee of the University of South Africa approved the research proposal (see Annexure A).

- The researcher got permission from the Research Committee of the Albinism Society of South Africa to collect data from the members of the Society (see Annexure B). This was in response to the request letter to the Albinism Society of South Africa (see Annexure C).

- Written informed consent was obtained from all the study participants (see Annexure D). Privacy and confidentiality were maintained throughout the research process. The legal implications of these ethical requirements are noted in Chapter 2.

Trustworthiness of the study was also established.

1.10   TRUSTWORTHINESS OF THE STUDY

Polit and Beck (2004:734) describe trustworthiness as "the degree of confidence that the qualitative researchers have in their data by using the criteria of credibility (truth-value); transferability (applicability); dependability (consistency); and confirmability (neutrality or verifiability)". These criteria are Guba's model for trustworthiness (Lincoln & Guba 1985:291), and were used to enhance the trustworthiness of this research. Polit and Beck (2004:35) emphasise that both qualitative and quantitative researchers want their findings to reflect the 'truth' (see chapter 2).

1.11   USE OF LITERATURE IN THIS STUDY

LoBiondo-Wood and Haber (2002:78) describe a literature review as "a systematic and critical review of the most important published and unpublished database, as well as conceptual literature". Chinn and Kramer (1999:129) emphasise that a literature review in theory-generating research is "comprehensive, and continues throughout the data-gathering and analysis phases". In this study, the literature review was therefore threefold as follows:

- The researcher conducted a preliminary literature review to verify the need for conducting the study and to focus the study (Polit & Beck 2004:56; Streubert & Carpenter 1999:20);

- After analysing the data, the researcher conducted another extensive literature review (literature control) to place the findings in the context of what is already known on oculocutaneous albinism; and

- An extensive literature review was done to define the key concepts and structure of the model (Streubert & Carpenter 1999:61) (see chapters 3, 4 and 5).

1.12   LIMITATIONS AND RECOMMENDATIONS

The limitations of this research were identified, and recommendations based on the findings made for clinical nursing practice, research, nursing education and policy making (see chapter 6).

1.13   OUTLINE OF THE STUDY

Chapter 1 introduces the study, and outlines the need for the study and background to the problem, the purpose and objectives of the study.

Chapter 2 describes the research design and methodology.

Chapter 3 discusses the research findings and literature control.

Chapter 4 describes the development of the model for enhancement of self-concept of people with oculocutaneous albinism.

Chapter 5 describes the model structure and guidelines to operationalise the model in practice.

Chapter 6 summarises and concludes the study, identifies its limitations, and makes recommendations.

1.14   CONCLUSION

This chapter outlined the study, including rationale for the study, problem statement, research questions, purpose and objectives of the study, as well as the paradigmatic perspective on which the study was based. The research design and methodology, ethical requirements and strategies to establish trustworthiness were also discussed.

Chapter 2 covers the research design and methodology.

CHAPTER 2

RESEARCH DESIGN AND METHODOLOGY

2.1   INTRODUCTION

Chapter 1 described the motivation and background for conducting the study. Chapter 2 discusses the systematic research design and methodology utilised to gather and analyse information. Since the main purpose of this study was to develop a model, the researcher conducted it in four steps of theory-generation: concept analysis; construction of theoretical relationships; description of the model, and guidelines to operationalise the model (Chinn & Kramer 1995:92). These four steps of theory-generation were undertaken to achieve the purpose and objectives of the study.

2.2   PURPOSE OF THE STUDY

The purpose of this study was to develop and describe a model for enhancement of self-concept of people with oculocutaneous albinism, based on the life-experience of these people. In line with this purpose, the relevant objectives were formulated.

2.3   OBJECTIVES OF THE STUDY

The study had the following objectives planned in two phases of theory-generation research approach:

2.3.1   Identification of the main concepts

The following objective for phase one was pursued:

- To explore the life-experience of people with oculocutaneous albinism so as to identify key concepts from the findings.

2.3.2   Description of the model structure and process

The following objectives for phase two were pursued:

- To classify the concepts of the model according to recipient, agent, context, dynamics, procedure and outcome/terminus;
- To describe the structure and process of the model; and
- To formulate and describe guidelines for operationalisation of the model in practice.

A theory-generating research design was relevant to achieve these objectives, and the research design is discussed in the next section.

2.4   RESEARCH DESIGN

In view of the main purpose of this study to develop a model, it is fundamental to understand the theory-generating research design, types of theory, and stages of theory generation before discussing the research methodology.

2.4.1   Theory-generative research design

The researcher used a theory-generative research design that was qualitative, explorative, descriptive and contextual. This was done to understand oculocutaneous albinism from the perspective of people who experience it, and to deduct key concepts from the research findings to develop the proposed model.

In this regard, Chinn and Kramer (1999:258) emphasise that in theory-generating research, the design must be consistent with the theory-generating orientation, so as to discover and describe relationships of empiric reality to construct the theory. Polit and Beck (2004:717) describe empiric as "evidence rooted in objective reality, and gathered using one's senses as the basis for generating knowledge".

2.4.1.1   Qualitative design

Polit and Beck (2004:729) describe qualitative research as "an investigation of phenomena, typically in an in-depth and holistic fashion, through the collection of rich narrative materials, using a flexible research design". Quantitative research is "an investigation of phenomena that leads to precise measurement and quantification, and often involves a rigorous and controlled design" (Polit & Beck 2004:729).

Qualitative data was collected from people with oculocutaneous albinism using in-depth phenomenological interviews. This was done to obtain an 'emic' perspective or insider's point of view of the study participants (Streubert & Carpenter 1999:17). Polit and Beck (2004:251) state that an 'emic' perspective is the local language, concepts, or means of expression that are used by the members of a group under study to name and characterise their experiences.

The theoretical framework was not predetermined so as to collect qualitative data from the study participants. Direct quotations of the research participants were captured during the research findings to reflect personal experiences. A holistic approach was therefore essential to obtain qualitative data.

2.4.1.2   Explorative design

Polit and Beck (2004:718) define an exploratory research as "a study that explores the dimensions of a phenomenon, or that develops relationship between phenomena".

Polit and Beck (2004:20) indicate that qualitative methods are useful to explore the full nature of a phenomenon that is not well understood. *Collins Thesaurus* (2006:252) lists the following synonyms for "exploratory": probe, scrutinise, search and analyse.

Oculocutaneous albinism is a phenomenon or a congenital disorder that is not well understood by the health professionals and the community. The following general question was thus helpful to probe the experience of the study participants: **Could you please share with me, as thoroughly and honestly as you can, your life-experience as a person with albinism.**

In this regard, the researcher explored the feelings and perceptions of people with oculocutaneous albinism to collect trustworthy qualitative data that could be described in the research findings. Furthermore, the key concepts deducted from the research findings were explored during the concept analysis to develop the proposed model.

2.4.1.3   *Descriptive design*

Polit and Beck (2004:716) define descriptive research as "a study with the main objective of describing an accurate portrayal of the characteristics of persons or situations". Polit and Beck (2004:192) explain further that the purpose of a descriptive design is to observe, describe, and document aspects of a situation as it naturally occurs, so as to serve as a starting point for theory generation. *Collins Thesaurus* (2006:179) lists the following synonyms for "descriptive": vivid, detailed and explanatory.

A naturalistic inquiry was thus undertaken in this study, and descriptive phenomenological in-depth interviews were used as the data-collection method. This was done to allow people with oculocutaneous albinism to describe their own life-experience, in their own words in as much detail as possible (Polit & Beck 2004:16).

The researcher also used a descriptive design to describe:

- the relationship of the concepts of the model;

- the structure and process of the model, and

- the guidelines for operationalisation of the model in practice.

For the benefit of prospective users of the proposed model, it is essential to understand the context in which the model was developed.

2.4.1.4   Contextual design

The context is the situation in which the research takes place, and comprises human, environment, professional, and organisational facilities (George 2002:206). Botes (1995:5) is of the opinion that the research context is only valid within a certain time-space, and is value context. In this regard, McKenna (1997:69) emphasises that concepts have different meanings depending on the context in which they are used.

The study was conducted around semi-urban and urban areas of the Gauteng province in South Africa. Data was collected from people with oculocutaneous albinism. Oculocutaneous albinism is a congenital disorder, characterised by hypopigmentation of the skin, hair and eyes.

The model was developed to serve as a framework for health professionals to enhance the self-concept of people with oculocutaneous albinism. Health professionals in the context of the proposed model are nurses, social workers, psychologists and doctors. At present, there is a lack of knowledge on oculocutaneous albinism by these health professionals. The proposed model is thus essential in the health profession.

2.4.2   Stages of theory generation

In developing a model that has value in the health practice, Chinn and Kramer's (1999:26-27) and Dickoff, James and Wiedenbach's (1968:431-434) stages of generating a theory were used as guidance.

Dickoff et al's (1968:431-434) four levels of theory development are

- **Level 1: Factor isolation** to name the theory;
- **Level 2: Factor relating** to see factors in relation to other factors;
- **Level 3: Situation relating** to see situations in relation to other situations; and
- **Level 4: Situation producing** to clarify the purpose of the theory.

Dickoff et al's (1968:431-434) levels of theory development can be compared to Chinn and Kramer's (1995:26-27) four stages of theory generation:

- **Stage 1: Creating conceptual meaning:** to clarify mental images of the concepts within the model;
- **Stage 2: Structuring and contextualising theory:** to organise relationships between and among concepts;
- **Stage 3: Generating and testing theoretic relationship:** to validate the relationships through empiric methods; and
- **Stage 4: Deliberately applying the theory:** to evaluate the effectiveness of a model in achieving the goals of health and nursing practice.

The next sub-section discusses four types of theory, and the relevance thereof to the present study.

2.4.3 Types of theory

Since theory-generation was pivotal to this study, it is important to understand the four related types of theory, namely meta-theory; grand theory; middle-range theory, and practice theory.

2.4.3.1   Meta-theory

Meta-theory is the first of the four levels of theory, and focuses on philosophical and methodological questions related to the development of a theory (Walker & Avant 1995:5).

At the level of meta-theory, the researcher considered:

➢ Proposing and critiquing the sources and methods of theory development.

➢ Analysing the purpose and kind of model needed.

➢ Proposing the criteria most suited for evaluating the model.

According to Walker and Avant (1995:13), meta-theory clarifies the methodology of the grand theory.

2.4.3.2   Grand theory

The grand theory is also referred to as macro theory, and describes large segments of the human experience (Polit & Beck 2004:115). Chinn and Kramer (1999:87) and Walker and Avant (1995:9) indicate that grand theories are abstract, and cover broad areas of concern within the nursing practice.

However, there is some disagreement on the value of grand theory in the nursing practice. In this regard, Fain (2004:67) indicates that grand theories are complex, and because they are not grounded in empirical data, they are usually not useful as guidelines for nursing practice.

Walker and Avant (1995:9) disagree with Fain ((2004:67), and argue that grand theories have made an important contribution in conceptually sorting out nursing from the practice of medicine by demonstrating the presence of distinct nursing perspectives. LoBiondo-Wood and Haber (2002:114) support Walker and Avant (1995:9), and list important grand theories in nursing practice, such as Roger's science of irreducible human beings, Neuman's systems model, Wiedenbach's prescriptive theory and Roy's adaptation theory.

In this study, the grand theory was based on Neuman's systems model and Wiedenbach's prescriptive theory, and these were used as assumptions of the proposed model (see chapter 5).

Walker and Avant (1995:13) explain that grand theories serve as guides for the phenomena of special concern at the middle-range level of theory.

2.4.3.3   Middle-range theory

Polit and Beck (2004:724) define middle-range theory as "a theory that focuses on only a piece of reality or human experience". According to Walker and Avant (1995:5), middle-range theory is less abstract, and is emerging to fill in the gaps between grand theories and nursing practice. Compared to grand theories, LoBiondo-Wood and Haber (2002:115 and Fain (2004:68) suggest that middle-range theory offers more direct application to research and practice. Fain (2004:68) adds further that the purpose of middle-range theory is to describe, explain, and predict phenomena.

The level of middle-range theory in this study was in view of describing the life-experience of people with oculocutaneous albinism from the perspective of people who have the experience. The middle-range theory in this study also predicts that the practical outcome of enhancing the self-concept of an individual is a person with a positive self-image.

2.4.3.4   Practice theory

According to Fain (2004:69), practice theories are sometimes referred to as prescriptive theories, and are important in developing a science of nursing practice. Fain (2004:69) compares practice theory to middle-range theory, and explains that practice theory are more specific, and produce specific guidelines for practice. Walker and Avant (1995:12) emphasise that the essence of practice theory is a desired goal and prescriptions for action to achieve the goal.

In this study, practice theory was fundamental because the goal of the study was to develop a model to serve as a framework for health professionals to enhance the self-concept of people with oculocutaneous albinism. The interventions to achieve the goal are described in detail by discussing the structure and steps of the model, and by formulating the guidelines to operationalise the model in practice.

The practice theory in this study justified the use of a theory generative research design that was qualitative, explorative, descriptive and contextual.

2.5 RESEARCH METHODOLOGY

Polit and Beck (2004:731) define a research methodology as a technique used to structure a study, and to gather and analyse information in a systematic fashion. As highlighted in the introduction of this chapter, the study was conducted in four steps of theory-generation, namely concept analysis; construction of theoretical relationships; description of the model; and guidelines to operationalise the model (Chinn & Kramer 1995:92; Dickoff et al 1968:415).

2.5.1 Step 1: Concept analysis

Concept analysis involved concept identification and definition as well as the classification of the concepts to develop the model. Concept analysis therefore incorporated the aspects of population, sampling, data collection, field notes, data analysis, literature control, ethical rigor and trustworthiness.

2.5.1.1 *Population and sampling*

- **Population**

Polit and Beck (2004:727) define population as the entire set of individuals having some common characteristics. A distinction is made between an accessible and a target population.

Polit and Beck (2004:711) define accessible population as the population of people available for a particular study, and that is often a non-random subset of the target population. LoBiondo-Wood and Haber (2002:242) define a target population as "the entire population the researcher is interested in, who meet the sampling criteria".

For this study, the accessible population would be people with congenital disorders to whom the researcher had reasonable access. The target population was both females and males with oculocutaneous albinism who met the sampling criteria.

- **Sampling criteria**

In this study, the participants had to meet the sampling criteria of having oculocultaneous albinism, thus having firsthand experience of the condition, and be able to articulate their own life-experience in a language the researcher understood. The sampling criteria facilitated the sampling process.

- **Sampling process**

The sampling process involved selecting a portion of the designated population to represent the entire population (LoBiondo-Wood & Haber 2002:24). The researcher used a non-probability purposive sampling technique to select the study participants.

Polit and Beck (2004:294) describe a purposeful sampling method as "non-probability sampling in which the researcher purposely selects study participants, based on personal judgement about which ones will be most representative". People with oculocutaneous albinism were therefore purposely selected as the key informants in this study to share their life-experience, and provide quality information on oculocutaneous albinism.

Interviews with the target population continued for a three-month period between January and March 2006. Interviews were conducted until data saturation was achieved to determine the sample size.

- **Sampling size**

According to Fain (2004:226), phenomenology tends to rely on small samples of participants, with guiding principles that all participants must have experienced the phenomenon under study, and are willing to describe it**.**

Therefore, after interviewing fifteen people with oculocutaneous albinism, the researcher felt that data saturation was achieved, when the ideas surfacing in the dialogue were previously heard from other participants (LoBiondo-Wood & Haber 2002:146). Saturation therefore refers to the participants' descriptions becoming repetitive, with no new or different ideas emerging during the data collection process (Fain 2004:226).

2.5.1.2   *Data collection*

Polit and Beck (2004:716) refer to data collection as "gathering of information to address a research problem". Chinn and Kramer (1999:130) state that in theory-generating research, data collection involves direct and indirect observations. Triangulation was therefore employed in this study, and data was collected using both indirect and direct methods.

Polit and Beck (2004:734) define triangulation as "the use of multiple methods to collect, and interpret data about phenomena, so as to converge on an accurate representation of reality". The purpose of using triangulation was to enrich the research findings, and use of the relevant tools was thus essential.

❑ *Data-collection tools*

The researcher was the main data-collection tool. Fain (2004:227) emphasises that the researcher is the primary study instrument in phenomenological research. The researcher asked all the study participants the following grand tour (broad) semi-structured question: Could you please share with me as thoroughly and honestly as you can your life experience as a person with albinism? In this regard, Polit and Beck (2004:77) point out that phenomenologists tend to ask meaning questions. The researcher used an interview protocol consisting of the grand tour question and biographical questions (see Anexure E).

Most importantly, the researcher managed to initiate a relaxed conversation with the participants during the interviews, and genuine reflection of the life-experience was encouraged. The researcher maintained good listening skills to determine the subsequent questions based on the participants' responses (Polit & Beck 2004:348).

❑ *Phenomenological in-depth interviews*

Phenomenological in-depth interviews were used as the data-collection method in this study. Polit and Beck (2004:727) define phenomenology as "a qualitative research tradition, with roots in philosophy and psychology, that focuses on the lived experience of humans". The researcher was physically present, and conducted the interviews with each of the fifteen study participants to elicit feelings and thoughts. The purpose of conducting the phenomenological in-depth interviews was to collect quality data, and identify concepts for the proposed model.

An advantage of using the phenomenological in-depth interviews in this study was that it was also a therapeutic encounter for some of the participants. It was evident during the conversation that some of the participants used the interviews to unburden their experiences to someone who was prepared to listen.

For example, at the start of the interview one of the participants remarked:

> *Hmm…but I'm glad you came to ask me this …for I bottled it up
> all the time …and I didn't know who to speak to about the staff…
> About why they criticise us..…*

Direct observation also includes keeping field notes and using an audiotape, and this also required the researcher to be physically present (Chinn & Kramer 1999:130). In this regard, the researcher kept field notes, and obtained permission to use a tape recorder during the interviews.

The researcher also noted that data collection in a phenomenological qualitative study might be more complex than in quantitative research, and a pilot study was undertaken to prepare for the main study (Polit & Beck 2004:196).

❑ *Pilot study*

Holloway and Walker (2000:121) point out that a pilot study helps to focus the research. Burns and Grove (2001:49) are of the opinion that a pilot study is essential to refine the data-collection process. However, Holloway and Walker (2000:121) maintain that there is no need to carry out a pilot study in a qualitative research project because the approach is developmental, and can encompass early attempts, depending on emerging and developing ideas. Nevertheless, Holloway and Warker (2000:121) emphasise that it is useful to undertake a "dry run", practising interview to increase the confidence and familiarity of the researcher with the research procedure.

The researcher therefore conducted a pilot study in the second week of January 2006, which was a week before finally conducting the main study. Phenomenological in-depth interviews were conducted with two females and one male with oculocutaneous albinism, and their ages ranged between 25 and 35 years.

During the pilot study, the researcher identified the need to re-phrase the grand tour question, and to re-sequence biographical questions. The researcher gained valuable experience in the fieldwork, and could determine the time required to prepare and complete the interviews. The pilot study was of benefit to the researcher to focus the study and this, in turn, enhanced the trustworthiness of the findings. The pilot study also helped the researcher to use appropriate facilitative techniques during the interviews.

□ *Facilitative communication techniques*

The researcher used facilitative communication techniques such as probing, clarifying, silence, minimal verbal response, non-verbal encouragement and summarising to facilitate conversation during the interviews.

• *Probing*

Polit and Beck **(**2004:728) describe probing as eliciting more useful or detailed information; for example, saying: "That is interesting, please elaborate."

• *Clarifying*

Collins English Dictionary (2006:141) defines clarify as "to make a point clearer or easy to understand". For example, the researcher said: "Let's see whether I understood you correctly, you said that …"

• **Silence**

Collins English Dictionary (2006:767) defines silence as "absence of sound". A brief silence was essential especially after a painful emotional response. This was done to give the participant a chance to recover and continue with the conversation.

• *Minimal verbal response*

In addition to silence, the researcher used minimal verbal prompts such as "Ee,……" (in some African languages "*ee*" means "yes") and "Hmm, .." to encourage the participant to continue with explanation of a point.

In this instance the researcher used the least possible words to facilitate the flow of the discussion (*Oxford Advanced Learners Dictionary of Current English* 2004:566).

- ***Non-verbal encouragement***

Collins English Dictionary (2006:551) defines non-verbal communication as "communication not involving words or speech". The researcher therefore used non-verbal communication techniques to set the tone for the interviews. For example, the researcher maintained a relaxed facial expression and posture, and used nodding of the head during the conversation to reassure the participant that she was listening.

- ***Summarising***

According to *Collins English Dictionary* (2006:831), summarising involves "a brief account of a process giving the main points of something". At the end of the interviews the researcher summarised the conversation, and used statements such as "To summarise what you said…."

The researcher also took field notes during the interviews to reflect and determine the progress in the field.

2.5.1.3   Field notes

Polit and Beck (2004:718) describe field notes as notes taken by researchers describing the unstructured observations they made in the field. Wilson (1989:420) describes the field as "the social-psychological area where the investigator gathers data to find answers in the central area of inquiry". Mouton (2004:104) supports the use of field notes, and emphasises that it is imperative that a researcher document the data-collection process as accurately, and in as much detail as possible to keep as historical record for other possible researchers.

The researcher kept the field notes to provide enough contextual information about time and place of the interviews, and to paint a picture of the life-experience of people with oculocutaneous albinism in a social situation (Polit & Beck 2004:383). Observational; analytic/theoretical; methodological, and personal field notes were thus kept (LoBiondo-Wood & Haber 2002:300, Polit & Beck 2004:718; Wilson 1989:434).

■ *Observational notes*

Polit and Beck (2004:726) define observational notes as "the observer's in-depth descriptions about events and conversations observed in naturalistic settings". According to Wilson (1989:434), observational notes are also referred to as descriptive notes.

After each interview, the researcher wrote descriptive notes, in terms of who was interviewed that day, where the interview was conducted, what transpired, and what the environmental situation was like. The researcher therefore recorded events experienced through watching, and listening to provide information about actions and context as completely and objectively as possible (Polit & Beck 2004:382; Wilson 1989:434).

Observational notes were essential to assist the researcher to remember important aspects of the interviews for data analysis. However, observational notes were taken with as little interpretation as possible (Wilson 1989:434).

■ *Analytic notes*

Polit and Beck (2004:734) define analytic notes as "notes detailing the researcher's interpretations of observed behaviour". The analytic notes were the researcher's efforts to attach meaning to the observational notes (Polit & Beck 2004:383).

Through the analytic notes, the researcher could interpret, infer and conjecture the observational notes to build an analytic scheme (Polit & Beck 2004:383; Wilson1989:435). The analytic notes were also helpful to formalise meanings into clusters of themes during analysis (Polit & Beck 2004:585).

■ *Methodological notes*

Polit and Beck (2004:723) define methodological notes as "the researcher's notes about the methods used during the data-collection process". The researcher took methodological notes during fieldwork to reflect the strength of using the phenomenological in-depth interviews to collect data, and the non-verbal observations during the conversation.

The methodological notes served as a reminder to the researcher about why probing of questions during the in-depth interviews was essential, and could be fruitful in the subsequent observations. Methodological notes were therefore essential for the researcher to critique her own tactics during data collection (Polit & Beck 2004:383; Wilson 1989:435).

■ *Personal notes*

Polit and Beck (2004:727) define personal notes as "written comments about the observer's own feelings during the research process". Due to the possible ethical and conflicting dilemmas in the field, it was essential for the researcher to make personal notes while in the field. The personal notes were therefore the researcher's written comments on her own feelings, reactions, reflections and experiences of oculocutaneous albinism (Polit & Beck 2004:383; Wilson 1989:435).

The researcher was alert that personal notes might inevitably give rise to personal emotions that challenge the researchers' assumptions on oculocutaneous albinism. The researcher therefore strove for personal notes not to have ethical, and conflicting implications during data collection and analysis (Polit & Beck 2004:382-383).

Data collection and analysis occurred simultaneously in this study. The analytic field notes taken during data collection, therefore, served as a starting point for the subsequent analysis (Polit & Beck 2004:383).

2.5.1.4   Data analysis

Chinn and Kramer (1999:133) point out that in theory-generating research, data analysis should be narrative, descriptive and qualitative analysis. Chinn and Kramer (1999:133) emphasise further that the data analysis process should be consistent with the qualitative research design and purpose of the study.

The purpose of this study was to develop a model, and the phenomenological qualitative and explorative research design and method adopted justified the use of qualitative content analysis. Furthermore, data analysis in qualitative research is an active and interactive process, and the researcher thus immersed in the data to search for meaning and develop an understanding on oculocutaneous albinism (Polit & Beck 2004:571).

Collaizzi's six basic principles of qualitative analysis method were therefore used to analyse the data in this study, namely transcribing; determining the meaning of each significant statement; aggregating formalised meanings into clusters of themes; exhaustive description; examining the evidence for interpretation; and presenting the findings (Polit & Beck 2004:571). Figure 2.1 represents these principles.

```
┌─────────────────────────────────────┐
│          Transcribing process        │
└─────────────────────────────────────┘
                    │
                    ▼
┌─────────────────────────────────────┐
│   Meaning of each significant statement │
└─────────────────────────────────────┘
                    │
                    ▼
┌─────────────────────────────────────┐
│    Aggregate formalised meanings into │
│           clusters of themes          │
└─────────────────────────────────────┘
                    │
                    ▼
┌─────────────────────────────────────┐
│          Exhaustive description       │
└─────────────────────────────────────┘
                    │
                    ▼
┌─────────────────────────────────────┐
│        Evidence of interpretation     │
└─────────────────────────────────────┘
                    │
                    ▼
┌─────────────────────────────────────┐
│         Presenting the findings       │
└─────────────────────────────────────┘
```
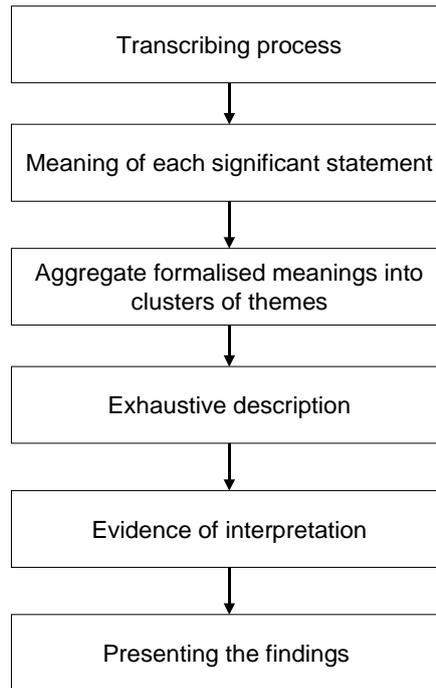
Figure 2.1

Schematic presentation of data analysis

▪ *Transcribing process*

The interviews were transcribed from the audiotapes, and prepared in an accessible visual written presentation using the Microsoft Word computer program. The researcher therefore transcribed the fifteen semi-structured interviews verbatim, listened to the taped interviews, and cross-checked with the field notes.

According to Chinn and Kramer (1999:130), tape-recorded interviews and field notes are the main data source for theory-linked research. The interviews were initially transcribed in the language spoken by the participants during the interviews, and then translated into English. The transcripts were given to a colleague who has knowledge of SeTswana to ensure that the translations of the responses were interpreted correctly.

Each transcription ranged from 16 to 20 pages in length, resulting in a total of 255 pages of the entire transcribed process. The researcher read, reread, organised, integrated and interpreted the transcribed document to make sense of the data. Important repetitive comments were highlighted during the initial transcription.

The researcher ensured that the transcripts were accurate and reflected the participants' experience. Bracketing and intuiting were applied as the reasoning strategies for transcribing. Non-verbal behaviours were also highlighted in the transcripts (Polit & Beck 2004:348).

The accuracy of the transcripts was essential to facilitate giving meaning to the participants' life-experience.

▪ *Meaning of each significant statement*

It was fundamental to analyse the data from the interviews and field notes without distorting the information. The researcher listened and re-read the transcripts carefully, with due sensitivity to the participants' life-experience. This was done until it was possible to make summaries with adequate description (Streubert & Carpenter 1999:60).

Sentences and paragraphs were assigned to labelled categories. The researcher did this process according to Collaizzi's analysis strategy, by reviewing each protocol and extracting significant statements (Polit & Beck 2004:585).

▪ *Aggregate formalised meanings into clusters of themes*

This process mostly involved identifying the themes, and the researcher focused on points of significance to formulate ideas on the participants' life-experience. In this instance, the researcher used inductive reasoning by moving from specific individual experience of oculocutaneous albinism, to a more general understanding of oculocutaneous albinism.

The themes were referred to the field notes and protocols again to validate the information, and also to take into consideration data that did not necessarily fit in the themes (Polit & Beck 2004:585). The main themes identified during analysis were: the participants' perception of the self; their experience with the environment; and their expression of need for development and growth. Categories and sub-categories of the themes were identified and described in detail.

▪ *Exhaustive description*

Exhaustive description formed an integral part of analysing and intuiting. The main themes were integrated into an exhaustive description of oculocutaneous albinism as experienced by the participants (Polit & Beck 2004: 585; Streubert & Carpenter 1999:51). The aim of exhaustive description was to communicate, and bring written and verbal description of distinct and critical elements of oculocutaneous albinism as experienced by the participants, and identify relationships of concepts for the proposed model (Streubert & Carpenter 1999:52).

The intent of the written document was to synthesise and capture the meaning of the participants' experience in a written form, without distorting or losing the richness of data (Streubert & Carpenter 1999:57) (see chapter 3).

Fain (2004:234) points out that the phenomenologist is not concerned with the frequency or prevalence of the theme, but rather, that all possible descriptions and understanding of the experience be uncovered. Verbatim excerpts were taken directly from the narrative data to enhance the findings.

Exhaustive description in this study also entailed detailed description of the structure and process of model. Furthermore, the researcher described guidelines for operationalisation of the model in practice as well as analysis and evaluation of the model (see chapters 4 and 5).

▪ *Evidence for interpretation*

As a final validating step, the participants were asked to review the research findings. This was done to test the reliability of interpretations to ensure that the findings reflected the thoughts and feelings of the participants, and not solely the researcher's ideas (Polit & Beck 2004:585; Streubert & Carpenter 1999:51). The researcher conducted follow-up interviews with six of the participants to clarify meanings of significant statements on oculocutaneous albinism.

In addition, experts in theory development and qualitative research were requested to analyse and evaluate the model. The inputs from these experts were fundamental as evidence for interpretation.

▪ *Presenting the findings*

Presenting the research findings is an important process, and required concentration, attention to detail, and time to describe the findings according to the themes, categories and sub-categories. This step also involved dissemination of the research findings, and writing a research report. Fain (2004:234) emphasises that in phenomenological research, dissemination of the findings means to let essential structures of the lived experience "be seen" through language. The research report described methods by which conclusions were arrived at, the significance and context of the findings, and the relationship of concepts to develop the proposed model.

2.5.1.5 Literature control

A literature control was fundamental to discuss the research findings and to develop the proposed model.

Chapter 3 discusses the literature review undertaken by the researcher to place the research findings in the context of what is already known on oculocutaneous albinism (Streubert & Carpenter 1999:61).

The themes were thus subjected to extensive literature control to make sense of the findings, and to identify the similarities and differences with previous research.

During the development of the model, the researcher used deductive reasoning to identify and define concepts of the model. This process also required extensive literature control. The researcher used Chinn and Kramer's (1999:84) descriptive components for theory definition which include clarifying the purpose of the theory; developing definitions that carry identifiable meaning to the theory; identifying relationships and structure of the model, and identifying assumptions that describe the theory (see chapter 5). The literature control enhanced the development of the proposed model.

2.5.2   Developing the model

The main purpose of this study was to develop a model for enhancement of self-concept of people with oculocutaneous albinism. In this regard, Walker and Avant (1995:3) state that theory development provides a way of identifying, and expressing key ideas about the essence of practice.

Walker and Avant (1995:131) emphasise that theory development is a sophisticated and complex level of theorising, since the theorist must deal with concepts, statements, linkages and definitions all at the same time. The researcher therefore used various sources to develop the proposed model (Chinn & Kramer 1999:58-60; Fain 2004:69; McKenna1997:57; Rodgers & Knafl 2000:81-83). Development of the proposed model entailed identifying the key concepts, defining the attributes of these concepts, and defining the core concept of the proposed model.

2.5.2.1   Identifying the key concepts

Fain (2004:83) refers to a concept as "a cluster of attributes". Chinn and Kramer (1999:54) describe a concept as "a complex mental formulation of experience that refers to perception of the world, and the totality of what is perceived".

After analysing data and developing the themes of the research findings, the researcher used deductive reasoning to identify the main concepts of the proposed model.

2.5.2.2   Defining attributes of the key concepts

Once the key concepts were identified, it was imperative for the researcher to define the attributes of these concepts, and to trace the origin of the words (Chinn & Kramer 1999:64). The researcher therefore undertook concept analysis, and used dictionary definitions, thesaurus, extended literature review, existing theories, and model and borderline cases to define the attributes of the identified concepts.

2.5.2.3   Defining the main concept

After identifying the attributes of the main concepts, the researcher engaged in formulating a concept definition. According to Chinn and Kramer (1999:74), identifying and defining the concepts specify the ideas on which the theoretic structure is built.

Chinn and Kramer (1999:76) differentiate between theoretical definitions and operational definitions. Theoretical definitions form the basis for and reflect empiric indicators for research, and convey the general meaning of the concept. Operational definitions indicate as clearly as possible how the concept is to be assessed in a specific study (Chinn & Kramer 1999:76).

Walker and Avant (1995:25) indicate that theoretical definitions are also referred to as conceptual definitions, and are the means by which the theorist introduces the reader to the critical attributes of the concepts.

Both the theoretical and operational definitions of "enhancement of self-concept" as the core concept of the model were provided, and contextualised to describe the structure of the proposed model (Chinn & Kramer 1999:74).

According to Walker and Avant (1995:137), concepts should be defined several times until a satisfactory outcome is achieved. The definitions adhered to Roussow's (1994:18) five rules to ensure that ambiguity is limited in the process of concept definition, namely

- The definitions must indicate the core characteristics of the concept.

- The definitions must not be circular.

- The definitions must not be too broad or too narrow.

- The definitions should not be stated in a figurative language.

- As far as possible, definitions should not be formulated negatively.

2.5.3   Step 2: Construction of the relationship statements

Chinn and Kramer (1999:77) attest that relationship statements describe, or predict the nature of the interactions between the concepts of the theory.

The researcher noted Chinn and Kramer's (1999:78) emphasis that the process of designing the relationship statements requires specific attention of the direction, strength, and quality of interactions between the concepts of the model. The researcher therefore used deductive reasoning to develop specific relationship statements using the concepts of the model. The researcher used Dickoff et al's (1968:431) survey list to classify the concepts of the model as follows:

- Agents: Who performs the activity?
- Recipients: Who is the recipient of the activity?
- Context: In what context is the activity performed?
- Procedures: What is the guiding procedure or technique of the activity?
- Dynamics: What is the energy source of the activity?
- Terminus: What is the outcome of the activity?

2.5.4 Step 3: Model description and evaluation

This sub-section discusses the processes to describe the model, and highlights the structure and process of the model as well as the critical reflection of the model.

2.5.4.1 Description of structure and process of the model

The researcher used Chinn and Kramer's (1999:84) process of theory to describe the structure and process of the model. This was done to create an objective description of the model as well as to form the basis for critical reflection of the model. Using Chinn and Kramer's (1999:84) guidelines, the following questions therefore facilitated description of the model:

- What is the **purpose** of the model?
- What are the **concepts that form** the model?
- How are the **concepts defined**?
- What is the nature of the **relationships**?
- What is the **structure** of this model?
- What **assumptions** underlie the model?

Once the model was described in detail by answering these questions, critical reflection of the model could be done.

2.5.4.2 Critical reflection of the model

Critical reflection of the model was essential to determine how well the model might be used in practice and research, and how it might be further developed. The researcher used Chinn and Kramer's (1999:100-109) critical reflection criteria to evaluate the model.

Experts in the field of theory development and qualitative research also evaluated the model using Chinn and Kramer's (1999:100) following critical reflection questions:

o **Is the model clear?**

This question addressed the clarity and consistency of the model.

o **Is the model simple?**

This question addressed the number of structural components and relationships within the model.

o **Is the model general?**

This question addressed the scope of experience covered by the model.

o **Is the model accessible?**

This question addressed the extent to which concepts in the model are grounded in empirically identifiable phenomena.

o **Is the model important?**

This question addressed the extent to which the model led to valued nursing goals in practice, research, and education.

Chapter 5 discusses in detail how these critical reflection questions were addressed to evaluate the model. In this regard, this was also helpful to develop guidelines to operationalise the model in practice.

2.5.5 Step 4: Guidelines to operationalise the model

Description of guidelines to operationalise the model in the nursing practice formed the final step of model development. According to Chinn and Kramer (1999:101-104), the final step in theory development involves application of theory. It was therefore essential for the researcher to ensure that the relationships of the concepts in the proposed model were systematically examined in the practice setting, and that the model achieved the desired outcomes and practice goals. The guidelines in Chapter 5 propose practical strategies to assist health professionals to enhance the self-concept of people with oculocutaneous albinism.

It was also fundamental for the researcher to use logical reasoning strategies throughout the research process.

2.6   REASONING STRATEGIES

Polit and Beck (2004:12) describe logical reasoning as "a method of knowing that combines experience, intellectual faculties, and formal systems of thought". Chinn and Kramer (1999:255) define logical reasoning as "a system of reasoning that deals with the form of relationships among propositions".

The researcher used the following reasoning strategies to generate knowledge on oculocutaneous albinism and to develop the proposed model: bracketing; intuiting; induction; deduction; analysis, and synthesis.

2.6.1   Bracketing

LoBiondo-Wood and Haber (2002:134) refer to bracketing as "a reasoning approach to identify and hold in abeyance, or 'put into brackets' preconceived beliefs, and opinions about a phenomenon under study". The researcher remained impartial, and applied bracketing, by putting in abeyance her own prior knowledge, thoughts, feelings and perception of oculocutaneous albinism during data collection and analysis. This was done to ensure pure description of data (Fain 2004:222; Streubert & Carpenter 1999:61).

However, Polit and Beck (2004:253) and Streubert and Carpenter (1999:47) warn that bracketing can never be totally achieved because of the close relationship individuals have with the world. Nevertheless, the researcher bracketed out the world and presuppositions on oculocutaneous albinism as much as possible so as to confront the data in pure form, and as described by people with oculocutaneous albinism (Streubert and Carpenter 1999:47). The researcher also noted that bracketing was essential to accomplish intuiting.

2.6.2   Intuiting

According to Fain (2004:222), intuiting refers to the researcher being immersed in the description of the lived experience to acquire a comprehensive and accurate interpretation. Streubert and Carpenter (1999:49) support Fain's (2004:222) definition, and describe intuiting as a process whereby the researcher begins to know about the phenomenon as described by the study participants.

In order to accomplish intuiting, the researcher avoided criticism, evaluation, or her own opinion, and became totally immersed in the participants' experience as they opened up, and shared their life-experience. Intuiting ensured that inductive reasoning was accomplished accordingly.

2.6.3   Inductive reasoning

Chinn and Kramer (1999:79) define inductive logic as "reasoning from the particular to the general". Fain (2004:65) supports Chinn and Kramer's (1999:79) definition, and indicates that inductive reasoning "is oriented towards discovery, and involves collecting observations that lead to conclusions".

The researcher used inductive reasoning in this study by obtaining and analysing data from the in-depth phenomenological interviews and field notes. Inductive reasoning was also used to identify suitable concepts for the proposed model.

Polit and Beck (2004:12) are of the opinion that both inductive and deductive reasoning are useful to organise phenomena, and both play an important role in nursing research.

2.6.4 Deductive reasoning

Chinn and Kramer (1999:79) define deductive logic as "reasoning from the general to the particular". Fain (2004:65) emphasises that deductive reasoning generates theory by beginning with known facts to test predictions, and validate existing relationships. Chinn and Kramer (1999:81) emphasise that in deductive logic, two or more premises are used to draw conclusions.

The researcher used deductive reasoning in this study in the following instances:

- During the literature control;
- To develop the structure of the model from the attributes of the concepts; and
- To formulate practical guidelines for the operationalisation of the model.

2.6.5 Analysis

According to Walker and Avant (1995:28), analysis is fundamental during theory development to clarify, refine, or sharpen concepts and statements. The theorist thus dissects a whole into parts, or concepts into parts so they can be better understood (Walker & Avant 1995:28).

In this study analysis as a reasoning strategy was used during data analysis for identification and classification of concepts, discussing concept analysis of the key concepts, identifying relationships between statements, and during literature control. During the development of the model, synthesis was done after concept analysis of the key concepts.

2.6.6 Synthesis

LoBiondo-Wood and Haber (2002:42) describe synthesis as "a combination or putting together, and combining of parts into a whole to make sense of it, and to explain relationships".

According to Walker and Avant (1995:28), the usefulness of synthesis in theory building is to construct a new concept, a new statement, or a new theory. Furthermore, Walker and Avant (1995:28) emphasise that synthesis works well where a theorist is collecting data without an explicit theoretical framework.

Polit and Beck (2004:572) maintain that at the end of the synthesis, the researcher can make some generalised statements about the phenomenon and about the study participants. Synthesis therefore has ethical implications. The Medical Research Council (2002:28) emphasises that researchers have a scientific responsibility to a particular profession, and to the study participants, to ensure that the conclusions and recommendations of a research project, yields knowledge worth knowing and that it is controlled and well verified.

In this study, synthesis was used as follows:
- By synthesising attributes of the concepts to form a definition of "enhancement of self-concept" as the core building block of the model.
- By synthesising the identified three steps to develop the model.
- By developing relationship statements of the model.
- By drawing conclusions and making recommendations for research in nursing practice.
- By undertaking the study without an explicit theoretical framework to synthesise, and yield fundamental knowledge for the nursing practice and for the study participants.

Achieving ethical rigor throughout the research process was essential.

2.7   ETHICAL RIGOR

The Medical Research Council (2002:4) define ethics as "a science of criteria, norms and values for human action and conduct". The Medical Research Council (2002:5) explains that the prime objective of ethics for health research is to promote health care, and its sole intent is to benefit the patient. The researcherobserved ethical rigor as a system of moral value, and adhered to professional, legal, and social obligations to promote health care, and protect the rights of the study participants (Polit & Beck 2004:717).

Most importantly, the researcher took heed of Fain's (2004:226) warning that because the researcher was the main data-collection instrument, several ethical considerations should be noted in research. In this regard, LoBiondo-Wood and Haber (2002:156) point out that "the personal nature of phenomenological qualitative research may result in considerable ethical implications compared to quantitative research".

Ethical rigor was therefore achieved in this study by subjecting the research proposal to ethical review. Informed written consent was obtained from the participants. The researcher also maintained the participants' privacy and assured them of confidentiality and anonymity. The legal justification of these ethical requirements is highlighted as well.

2.7.1 Research Ethics Committee review

The researcher sent the research proposal to the Health Studies Research and Ethics Committee of the University of South Africa. After careful consideration of the research proposal, the Health Studies Research and Ethics Committee of the University of South Africa granted the researcher permission to conduct the research (see Annexure A). This is in accordance with the Medical Research Council (2002:27) that all research involving healthy volunteers and patients must be subjected to the ethical review of a Research Ethics Committee.

The researcher also obtained approval from the Research Committee of the Albinism Society of South Africa to interview members of the Albinism Society of South Africa (see Annexure B).

2.7.2 Informed consent

The Medical Research Council (2002:9) emphasises the importance of written information, and consent forms for health research intervention.

2.7.2.1   Written consent letter

All the participants signed a written consent letter (see Annexure D). The participants were all competent people, capable of giving consent for the research (Medical Research Council 2002:8). Respect was given to the participants. The researcher explained to them in detail what would happen during the research process, in a language that they could understand, and ensured that they did understand.

Prior to signing the consent letter, full disclosure of the nature of the research was done. The researcher explained the content of the consent letter to the study participants. The consent letter clearly outlined the purpose of the study, the expected duration of the interview, request to use a tape recorder, the benefit of the study to the participant, the right to have access to the findings of the study, and the right to withdraw at any time (Medical Research Council 2002:26; Polit & Beck 2004:151).

The researcher explained to the participants that there was no risk of penalty or prejudice treatment if they did not participate in the study. The participants were also assured of confidentiality, and were given a chance to decide voluntarily to sign the consent letters. They were therefore not forced to participate in the research as this has legal implications.

2.7.2.2   Legal implication of informed consent

The researcher noted that informed consent has some legal obligations, as entrenched in the Bill of Rights of the South African Constitution. The Constitution of South Africa Act, 108 of 1996 is the supreme law in South Africa. Section 12(2)(c) of the Constitution of South Africa Act, 108 of 1996 (1996:8) states: "*Everyone has the right to bodily and psychological integrity, which includes the right… not to be subjected to medical or scientific experiments without their informed consent*" (South Africa 1996:8). The researcher addressed the literal interpretation of the Constitution by ensuring that the participants understood the content of the consent letter.

2.7.3 Privacy and confidentiality

Confidentiality is the researcher's management of private information to maintain the dignity of study participants (Medical Research Council 2002:26). The participants were therefore assured that data collected would be treated with confidentiality, used for the research purpose only, no one would have access to the interview data or the audio-tapes except another independent expert on qualitative research, and that the taped materials would be erased on completion of transcriptions of the tape. This reassurance facilitated the participants' confidence to discuss their life-experience openly and honestly (Bless & Higson-Smith 1995:103).

The researcher did not obtain identifying information, such as the participants' names and addresses, to safeguard their confidentiality and anonymity (Polit & Hungler 1999:139). Furthermore, the researcher did not link any information to any person, and did not publicise information in a manner that could identify the participant (Streubert & Carpenter 1999:38).

Role conflict emerged during the interviews as some participants requested further support and counselling. However, the researcher maintained the participants' confidentiality (Fain 2004:227). In this regard, the researcher referred those participants who expressed a need for support and counselling to the Albinism Society of South Africa.

The researcher maintained the privacy and confidentiality of the participants in view of the related legal implications. The Bill of Rights of the South African Constitution, Section 14(d) makes provision for the right to privacy: *"Everyone has the right to privacy, which includes the right not to have … the privacy of their communications infringed"* (South Africa 1996:8).

2.8   MEASURES FOR ENSURING TRUSTWORTHINESS

The researcher used Lincoln and Guba's (1985:290) model for trustworthiness to enhance the trustworthiness of this study. Lincoln and Guba (1985:290) describe trustworthiness as "a method of establishing, or ensuring scientific rigor in a qualitative research without sacrificing relevance". Polit and Beck (2004:35) affirm that qualitative researchers are as eager as quantitative researchers to have their findings reflect the true state of human experience.

According to Polit and Beck (2004:35), qualitative researchers enhance trustworthiness of their research to reflect the truth of the study, whilst quantitative researchers use the criteria of reliability and validity. Reliability refers to the accuracy and consistency of information. Validity concerns the soundness of the research evidence (Polit & Beck 2004:36). Streubert and Carpenter (1999:333) therefore emphasise that the research is believed to be trustworthy in a qualitative study, when it accurately represents the experience of the study participants.

The main purpose of the study was to develop a model to be used as a framework for health professionals to enhance the self-concept of people with oculocutaneous albinism. It was therefore essential to enhance the trustworthiness of this study for the following reasons (Polit & Beck 2004:35).

- The health professionals and consumers of research need to assess the quality of evidence offered in this study by evaluating the researcher's conceptual and methodological decisions.
- The proposed model and research cannot contribute evidence to guide clinical practice if the findings are inaccurate, biased, fail adequately to represent the experiences of people with oculocutaneous albinism, or are based on a misinterpretation of the data.

The researcher enhanced the trustworthiness of this study by using Guba's four criteria of trustworthiness (Lincoln & Guba 1985:290-330):

- Truth value, using strategies of credibility;
- Applicability, using strategies of transferability;
- Consistency, using strategies of dependability; and
- Neutrality, using strategies of confirmability.

2.8.1   Truth-value using strategies of credibility

Credibility refers to confidence in the truth of the data, and interpretations of the research findings (Polit & Beck 2004:715). Lincoln and Guba (1985:294) view credibility as an overriding goal of qualitative research. The strategies in achieving credibility in this study were prolonged engagement, persistent observation, triangulation, reflexive journal, member checking, peer debriefing, researcher credibility and structural coherence.

2.8.1.1   Prolonged engagement

According to Polit and Beck (2004:430 & 728), prolonged engagement is investment of sufficient time collecting data to have an in-depth understanding of the culture, language, or views of the group under study, and to test for misinformation and distortions. Polit and Beck (2004:430 & 728) are of the opinion that prolonged engagement is a first and very important step in qualitative research.

As a strategy to enhance prolonged engagement, a week before each interview, the researcher telephonically contacted the participants to establish rapport and request an appointment. Furthermore, phenomenological in-depth interviews were conducted, and the participants were given sufficient time to relate their life-experience. Each interview lasted for approximately 60 minutes.

During the interviews, the participants used a language they were comfortable with, and that the researcher could understand. This was done to facilitate the discussion, and to avoid misinformation and distortion of the data. Prolonged engagement was essential to built trust, and rapport between the researcher and the participants. This, in turn, facilitated the collection of accurate, useful and rich information on oculocutaneous albinism to establish the credibility of the research.

2.8.1.2 Persistent observation

To ensure persistent observation, the researcher was in constant collaboration with the Albinism Society of South Africa, therefore, the participants had access to the researcher if there was anything they wanted to add even after the main interviews. The interviews were also conducted until saturation of data, that is, to a point where a sense of closure was attained, and new data yielded redundant information (Polit & Beck 2004:731).

2.8.1.3 Triangulation

Polit and Beck (2004:734) refer to triangulation as "the use of multiple methods to collect and interpret data, so as to converge on an accurate representation of reality". In this instance, triangulation enhanced credibility of this study because two experienced promoters guided the researcher. The promoter is presently a research co-ordinator in the Department of Health Studies at the University of South Africa, and the co-promoter is presently an experienced clinical geneticist at the University of the Witwatersrand.

2.8.1.4 Reflexive journal

Confidence in the data was also established by maintaining a field journal or field notes throughout the study, and through the use of a tape recorder (Polit & Beck 2004:382-383). All the participants agreed to the use of the tape recorder during the interviews. Confidence in the quality of data from the field notes and tape recorder also determined transferability of data.

2.8.1.5   Member checking

Member checking was essential to provide feedback to the participants on the emerging data and interpretations, and to ensure that the researcher reflected the reality of the participants' life-experience (Polit & Beck 2004:432).

Member checking was done with a few of the participants after analysing the data. The main themes of the findings were discussed, and agreed upon by the participants. Furthermore, evaluation comments from experts in qualitative and theory development were integrated to develop the model. Literature control was done to discuss the research findings, and all sources and authors in the study are acknowledged with complete references.

2.8.1.6   Peer debriefing

Lincoln and Guba (1985:206) hold that peer review involves the researcher discussing the research process and findings with an impartial colleague who has experience with qualitative methods. Peer debriefing was done with a colleague who is also presently registered for a doctor's degree with the University of South Africa, developing a health promotion model for people with disabilities.

2.8.1.7   Researcher credibility

Researcher credibility refers to the faith that can be put in the researcher to enhance confidence in the research (Polit & Beck 2004:434). During data collection, the researcher was the main data-collection tool, and she also analysed the data (Burns & Grove 2001:425).

The researcher has good communication skills that she gained during the ten-year experience of counselling people with congenital disorders and their families (from 1989-1999). In addition, the researcher attended an Applied Management Technique Course on Effective Communication in 2000.

Between 2002 and 2003, the researcher attended courses on Scientific Writing and Qualitative Research Methods, which improved her academic and scientific writing skills. The researcher also gained valuable experience when she conducted in-depth interviews and focus group discussions to obtain her Master's degree in 2003. Therefore, the researcher's credibility ensured that she was aware of her role to guarantee that her own bias did not affect the outcome of the research (Polit & Beck 2004:335).

2.8.1.8 Structural coherence

According to Krefting (1991:220), structural coherence enhances credibility by ensuring that there are no unexplained inconsistencies between the data and interpretation of the data. The participants' life-experience was thus interpreted well, and the researcher ensured that loosely connected data was described in detail.

The research findings were also interpreted in a consistent manner, and discussed within existing theories and literature. The research process unfolded in a logical way for health professionals to have confidence in applying the proposed model in practice.

2.8.2 Applicability using strategies of transferability

The strategy used for achieving applicability is transferability. Transferability is analogous to generalisability, and is defined as "the extent to which the findings can be transferred to other settings or groups" (Lincoln & Guba 1985:203). Streubert and Carpenter refer to transferability as "fittingness", and describe it as "the probability that the study findings have 'meaning' to other similar situations". In this study, the techniques applied to ensure transferability were dense description and purposive sampling (Streubert & Carpenter 1999:59-61).

2.8.2.1   *Dense description*

Dense description refers to "a rich and thorough description of the research setting, and of observed transactions and processes provided in the research" (Polit & Beck 2004:41). Lincoln and Guba (1985:204) emphasise that an important mechanism for promoting transferability is the amount and quality of information qualitative researchers provide about the context of the study.

Sufficient descriptive data and analysis process is therefore provided in this study. The research findings are described in detail, and verbatim transcriptions of the collected data presented. The researcher described in detail the theory-generation steps to develop the proposed model, and practical guidelines to operationalise the model.

Furthermore, the researcher provided detailed descriptions of how the model was analysed and evaluated. In this regard, the researcher was guided by the following questions to determine the transferability or applicability of the model (Polit & Beck 2004:690-691):

- Will   the model "fit" in the present nursing practice?
- Is the philosophy of care underlying the model fundamentally different from the philosophy prevailing in the practice setting?
- Will the proposed model take too long to implement and evaluate?

Five broad criteria, namely clarity, simplicity, generality, empirical applicability and logical development were used to address these questions, and to describe in detail the analysis and evaluation of the model (Chinn & Kramer 1999:101; McKenna 1997:226) (see chapter 5).

Lincoln and Guba (1985:205) maintain, however, that expectations for determining whether the findings fit or are transferable rest with the potential users of the findings, and not with the researcher. Polit and Beck (2004:435) state further that transferability of the findings is, to some extent, a sampling and design issue, rather than an issue relating to the soundness of the data.

Purposive sampling was therefore used in this study to enhance transferability.

2.8.2.2 *Purposive sampling*

Fain (2004:225) defines purposive sampling as "a process of selecting persons who have lived a common experience, and are able and willing to describe it". Purposive sampling was used to select people with oculocutaneous albinism as study participants, based on their particular personal experience of oculocutaneous albinism (Streubert & Carpenter 1999:59).

According to Patton (1990:103), the "logic and power of purposive sampling lies in selecting information-rich cases, from which one can learn a great deal for the 'purpose' of the research". Information-rich data on oculocutaneous albinism was thus essential to guarantee dependability of data.

2.8.3 Consistency using strategies of dependability

Dependability refers to "the stability of data over time and over conditions" (Polit & Beck 2004:435). Streubert and Carpenter hold that dependability is a criterion met once the researcher has determined the credibility of the findings. The techniques used in this research to enhance dependability were triangulation, coding, and dependability audit.

2.8.3.1 *Data source triangulation*

The purpose of using triangulation to enhance dependability was to enrich the research findings. To establish triangulation in this instance, the researcher used both direct and indirect methods to collect data. In this regard, data collection methods included conducting in-depth phenomenological interviews, keeping field notes as well as the use of a tape recorder during the interviews.

Triangulation was also established during concept analysis by using several sources to define the concepts. These sources were dictionaries, thesaurus, existing theories, professional and classical literature, and model and borderline cases to define and clarify the key concepts of the proposed model.

2.8.3.2 Coding

Coding is a process used to identify recurring words or concepts within the data (Polit & Beck 2004:713). The researcher immersed in the data, and read and re-read the transcripts to identify common themes and concepts to grasp the experience of people with oculocutaneous albinism.

2.8.3.3 Dependability audit

An inquiry audit was established to enhance dependability. The inquiry audit involved scrutiny of the data and relevant supporting documents by an external reviewer (Polit & Beck 2004:435). An expert professional reviewed the tapes, interview transcripts and field notes. The researcher described in detail the research methodology, and the model and guidelines were analysed and discussed with colleagues in the Directorate: Women's Health and Genetics.   The inquiry audit also facilitated the confirmability of the data.

2.8.4 Neutrality using strategies of confirmability

Confirmability is the extent to which the research findings are free from bias, and refers to the objectivity or neutrality of the data (Polit & Beck 2004:435). Confirmability was established in this study by maintaining the researcher's reflexivity and confirmability audit.

2.8.4.1   Researcher's reflexivity

Researcher's reflexivity is discussed in detail as the researcher credibility under sub-section 2.8.1.7 of this chapter.

The researcher applied bracketing by putting in abeyance her own prior knowledge, thoughts and perception of oculocutaneous albinism, and became totally immersed in the participants' experience.

2.8.4.2 Confirmability audit

To establish a confirmability trail, a panel of experts experienced in qualitative research and theory development critically evaluated the entire process of the study. The objective of the confirmability audit was to illustrate as clearly as possible, the evidence and thought processes that led to the conclusions. For this purpose, the researcher recorded activities over time so that others can follow the research process undertaken (Streubert & Carpenter 1999:29).

2.9 CONCLUSION

This chapter discussed the research design and methodology in detail, including theory-generating research design and reasoning strategies, ethical considerations and criteria to enhance the trustworthiness of the research. The components of the proposed model, strategies to evaluate the model, as well as practical guidelines for operationalisation of the model were highlighted.

Chapter 3 discusses the research findings and literature control, and identifies the main concepts for the proposed model.

CHAPTER 3

RESEARCH FINDINGS AND LITERATURE CONTROL

3.1 INTRODUCTION

This chapter focuses on the research findings, and constantly compares the findings with those of previous studies. The findings are presented in a narrative form, and the participants' responses are quoted verbatim to support the findings.

Three main themes were identified during data analysis, namely perception of the self, experience with the external environment, and expression of need for development and growth of people with oculocutaneous albinism. These themes captured the emic views of the people with oculocutaneous albinism experiencing the phenomenon. In this regard, LoBiondo-Wood and Haber (2002:166) describe a theme as a label that describes data in a condensed format, to help the reader understand the emic view.

During data analysis the researcher identified the need to reflect the categories of the main themes within the dimension of Neuman's systems model, namely psychological, physiological, socio-cultural, spiritual, and developmental dimensions (Neuman 1995:15). Most importantly, the researcher entered the field without a preset theory of reference by using bracketing and intuiting reasoning strategies (see chapter 2).

In the next section, the researcher describes the sample to contextualise the findings within the biographical characteristics of the participants.

3.2 SAMPLE DESCRIPTION

The sample consisted of fifteen people with oculocutaneous albinism from urban and semi-urban areas in the Gauteng Province, South Africa. This section describes the demographic profile and the family history of the sample. Table 3.1 depicts the characteristics of the participants.

3.2.1 Demographic profile

The participants' demographic profile included gender, age, ethnic group, level of education, employment status, marital status, religious affiliation, sports/extramural activities, and residential area.

3.2.1.1 Gender distribution

Data was collected from fifteen participants, twelve females and three males, with oculocutaneous albinism. A random purposive sample was selected from a list of potential participants provided by the Albinism Society of South Africa. The list comprised more females (54%) than males (46%), hence there were more females in the sample. Nevertheless, the majority of females in this study was of significance according to studies on oculocutaneous albinism in the late 1980s. These studies raise concern that females with oculocutaneous albinism are in a particularly vulnerable position to be stigmatised and exploited in relationships, and thus seek help from support groups. Ezeilo (1989:1130) and Lonsdale (1990:24) maintain that, as a result of curiosity, women with oculocutaneous albinism may be sexually exploited rather than treated affectionately, and this in turn, may limit their quality of life and self-image.

To support Ezeilo (1989:1130) and Lonsdale (1990:24), one of the married female participants in the present study expressed her concern as follows:

> *You know sometimes I wonder whether the men who propose love to us*
> *really love us, or he's trying to get something from you. But I think sometimes*
> *its psychological, but there has been such remarks...*

In a study in Soweto, South Africa, in the early 1980s, however, Kromberg and Jenkins (1982:386) found more males (113) than females (93). Kromberg and Jenkins (1982:386) cite Froggart (1962) in Northern Ireland, who states that the sex difference of people with albinism was social, and males with albinism tend to come forward to be counted more readily than females.

At the same time, Kromberg and Jenkins (1982:386) also refer to Oettle's (1973) finding in Transkei, South Africa, of more females than males.

Kromberg and Jenkins (1982:386) then suggest that the sex ratio may be explained by the nature of the populations in which the studies were carried out.

3.2.1.2   Age distribution

The participants' ages ranged between 18 and 48 years, with a mean age of 33 years. The majority of the participants were between 25 and 35 years, while one was 18 and one was 48 years old.

Luand, Puri, Durham-Pierre, King and Brilliant (1997:734) indicate that as result of skin cancer, oculocutaneous albinism is associated with early death. According to Luand et al (1997:734), early death was especially noted in African countries closer to the equator like Tanzania and Nigeria, where only 10% of persons with oculocutaneous albinism survive beyond 30 years of age. Okoro (1975:475) found that among 517 people with albinism in the East Central State of Nigeria, 89% were aged 0-30 years while 10% were aged 31 to 60 years.

3.2.1.3   Ethnic group

Most of the participants were Tswana and Zulu speaking, each representing four participants respectively, followed by three who were Xhosa speaking. A minority of the participants were Southern Sotho, Ndebele, Venda and Coloured, each respectively represented by one participant. It is considered that the values and beliefs in a specific ethnic group could have influence on the experience of the people with oculocutaneous albinism, although no evidence of this was available in the literature.

Kromberg and Jenkins (1982:384) found the highest prevalence of oculocutaneous albinism in the Southern Sotho, followed by the Swazi and then the Tswana. The consanguinity rate of parents in the Tswana and Southern Sotho people accounted for the high prevalence rate in Kromberg and Jenkin's (1982:384) study. In this study, however, the prevalence of more Zulu people with oculocutaneous albinism may be attributed to the prevalent ethnic group in that geographical area.

Table 3.1 represents the participants' characteristics.

Table 3.1 Participants' characteristics (N=15)

Code	Gender	Age	Level of education	Present employment status	Marital status	Church attendance/ Religious affiliation	Sports/ Extramural activities	Residential area
P1	Female	23	Post school certificate	Unemployed	Single	Religiously affiliated and church attendance is often	None	Semi-urban
P2	Female	31	Post school certificate	Unemployed	Single	Religiously affiliated and church attendance is often	None	Semi-urban
P3	Male	31	Diploma	On learnership programme	Single	Religiously affiliated but seldom church attendance	**Snooker player**	Semi-urban
P4	Female	23	Diploma	On learnership programme	Single	Religiously affiliated and church attendance is often	None	Urban
P5	Female	23	Degree	Employed	Single	Religiously affiliated and church attendance is often	**Netball player and athlete**	Urban
P6	Female	33	Degree	Employed	Single	Religiously affiliated but seldom church attendance	None	Semi-urban
P7	Male	29	Diploma	Employed	Single	No religious affiliation	None	Semi-urban
P8	Female	31	Diploma	Employed	Single	Religiously affiliated and church attendance is often	**Participates in aerobics**	Semi-urban
P9	Female	25	Post school certificate	Part-time employment	Single	Religiously affiliated but seldom church attendance	**Used to be a tennis player**	Semi-urban
P10	Female	22	Diploma	On learnership programme	Single	Religiously affiliated and church attendance is often	None presently, but used to be a part-time model and under 14 netball coach	Semi-urban
P11	Female	24	Diploma	Employed	Single	Religiously affiliated and church attendance is often	None presently, but used to be an athlete	Semi-urban
P12	Female	24	Matric	Unemployed	Single	Religiously affiliated and church attendance is often	Church choir member	Semi-urban
P13	Male	35	Diploma	Employed	Married	Religiously affiliated and church attendance is often	None	Semi-urban
P14	Female	18	Attending school (Grade 11)	Still attending school	Single	Religiously affiliated and church attendance is often	None	Semi-urban
P15	Female	48	Diploma	Employed	Married	Religiously affiliated and church attendance is often	Participate in school music competitions	Semi-urban

(P= Participant)

3.2.1.4 Educational standard

The question on the level of education was asked to determine whether the level of education could influence the participants' life-experience of oculocutaneous albinism.

Table 3.1 shows that out of the fifteen participants, fourteen attended school to Grade 12 and matriculated. Only one, the youngest participant, was still attending school (in Grade 11 [Std 9]). The majority of the participants also had post-school qualifications including degrees, diplomas and certificates. Only one participant passed Grade 12 and had not been able to further her studies.

Three participants were presently enrolled for postgraduate studies and registered for a Bachelor of Arts degree, Diploma in Management and Diploma in Human Resources, respectively. It emerged that the level of education of the participants influenced their life-experience as presented in the contextual discussion of the main themes.

3.2.1.5 Employment status

It was essential to determine the participants' employment status to determine how this could influence their everyday life-experience of oculocutaneous albinism.

It is evident from Table 3.1 that almost half of the participants were employed. Of the fifteen participants, eight were employed, two of them on a contract basis; three were on management and business administration programmes, and three were not employed nor on any learnership programmes. Only the youngest participant was still attending school. During the interviews the participants indicated how their employment status influenced their life-experience, especially with regard to their self-esteem.

3.2.1.6   Marital status

The purpose of asking this question was to determine how marital status could influence the life-experience of people with oculocutaneous albinism.

Of the fifteen participants, thirteen were single and only two were married. The partners of both married participants did not have oculocutaneous albinism.

Of the thirteen single participants, three did not have partners (boyfriend or girlfriend). Of these, two had previously had relationships, and one had never been in a formal relationship. Ten of the thirteen single participants had partners, one of whom had a live-in partner. Of the ten single participants with partners, two of the partners also have oculocutaneous albinism, and eight do not.

Nevertheless, in line with the above description of the participants' marital status, the findings of Kromberg (1992:164) reveal that there was a positive attitude towards pigmented people marrying a person with oculocutaneous albinism, but a slightly less positive attitude towards allowing one's own child to marry a person with oculocutaneous albinism. Kromberg (1992:164) also noted that people with oculocutaneous albinism themselves were found to be slightly, but not significantly, less in favour of allowing their children to marry a person with oculocutaneous albinism.

3.2.1.7   Religious affiliation and church attendance

The question wished to identify the denominations to which the participants were affiliated, for the purpose of determining how the norms and belief in the churches could influence the life-experience of persons with oculocutaneous albinism (see table 3.1).

Of fifteen the participants, fourteen had a strong religious background and were affiliated to a particular religious group, whilst only one did not belong to any particular religious denomination. Those fourteen participants were affiliated to various churches: Anglican, Methodist, Lutheran, Apostolic and Islam, Dutch Reformed, Grace Bible and Zion Apostolic.

In South Africa individuals have a constitutional right to affiliate to any church of their choice (South Africa 1996:8). Raliphada-Mulaudzi (1997:46) is of the opinion that different churches have different norms and values, which may influence the attitude of the community on various issues.

During the interviews significance of the participants' strong religious background emerged clearly as they related their life-experience.

3.2.1.8 Sports/Extramural activities

This question was asked to determine the participants' social activity. The majority of the participants did not engage in any form of sports or extramural activities. Of the fifteen participants, only six engaged in some form of sports or extramural activity, including aerobics, snooker, music, and home exercises using videos. One of the active confident participants indicated that she used to be a part-time model, and was once a coach for under 16 netball players. Nine of the participants who were not engaged in any extramural or sports activities cited time constraints as a major barrier. The significance of this characteristic is described in detail in the contextual discussion of the main themes of the study.

3.2.1.9 Residential area

This section highlights the home area of the participants. Of the fifteen participants, thirteen were from semi-urban and two from urban areas (see table 3.1).

The residential area is of significance as the study participants shared their experience in view of beliefs, values and practices on oculocutaneous albinism from different areas. Jordaan and Jordaan (1998:799) maintain that beliefs in society affect the people's attitude to life, life styles and behaviour in an ethical and social context.

3.2.2 Family history

The participants' family history determined the family setting, number in the family, sibling rank, as well as other family members with oculocutaneous albinism. Table 3.2 indicates the participants' family history.

Table 3.2 Participants' family history

Code	Family setting	Number of children in the family	Sibling ranking	Immediate family members with oculocutaneous albinism	Extended family members with oculocutaneous albinism
P1	Nuclear	2	1st	None	Maternal aunt
P2	Nuclear	4	2nd	Sister	None
P3	Nuclear	2	1st	None	Paternal grandfather
P4	Nuclear	4	2nd	Sister	None
P5	Single parenting	5	3rd	None	Paternal nephew
P6	Nuclear	4	4th	2 brothers and a sister	Paternal aunt
P7	Nuclear	3	1st	None	None
P8	Nuclear	6	2nd	None	None
P9	Nuclear	3	3rd	None	Maternal great-grandmother
P10	Nuclear	4	3rd	None	Paternal great-aunt and nephew
P11	Single parenting	Only child	Only child	None	Paternal great-grandmother
P12	Extended family	3	1st	None	None
P13	Single parenting	5	5th	None	None
P14	Extended family	2	1st	None	None
P15	Nuclear	5	3rd	2 brothers	Uncertain

(P= Participant)

3.2.2.1 Family setting

Ten of the fifteen participants grew up in a nuclear family setting; three in a single-parent family setting, and two in an extended family setting. It emerged that the different family settings did have relevance in the participants' life-experience.

3.2.2.2   *Number of children in the family and sibling ranking*

Of the fifteen participants, only one was an only child, and the number of children in the other cases ranged between 2 and 6. Sibling ranking ranged from being the firstborn child to the fifth child. The relevance of the number of children in the families and the sibling ranking emerged as one aspect that facilitated the support that the parents provided to the participants.

3.2.2.3   *Family members with oculocutaneous albinism*

Table 3.2 shows that four of the fifteen participants had first-degree family members, brothers and sisters, with oculocutaneous albinism. None of the fifteen participants had parents with oculocutaneous albinism. Thus, all the parents are carriers of the recessive oculocutaneous albinism trait (Winship 2003:174).

The participants were also asked about the extended family members and cousins with oculocutaneous albinism. The results showed that seven of the fifteen participants did have extended family members with oculocutaneous albinism, for example great-grand aunts, grandparents, aunts and nephews. Seven of the participants indicated that they were not aware of extended family members with oculocutaneous albinism. One participant was uncertain of extended family members having oculocutaneous albinism, but could remember that some of them were very light in complexion, which could mean that they were carriers of the recessive oculocutaneous albinism trait.

3.3   CONTEXTUAL DISCUSSION OF THE MAIN THEMES

During analysis three main themes emerged from the findings of this study, namely
■   Perception of the self by persons with oculocutaneous albinism
■   Experience of persons with oculocutaneous albinism with the external environment
■   Expression of need for development and growth.

Figure 3.1 illustrates the main themes graphically.
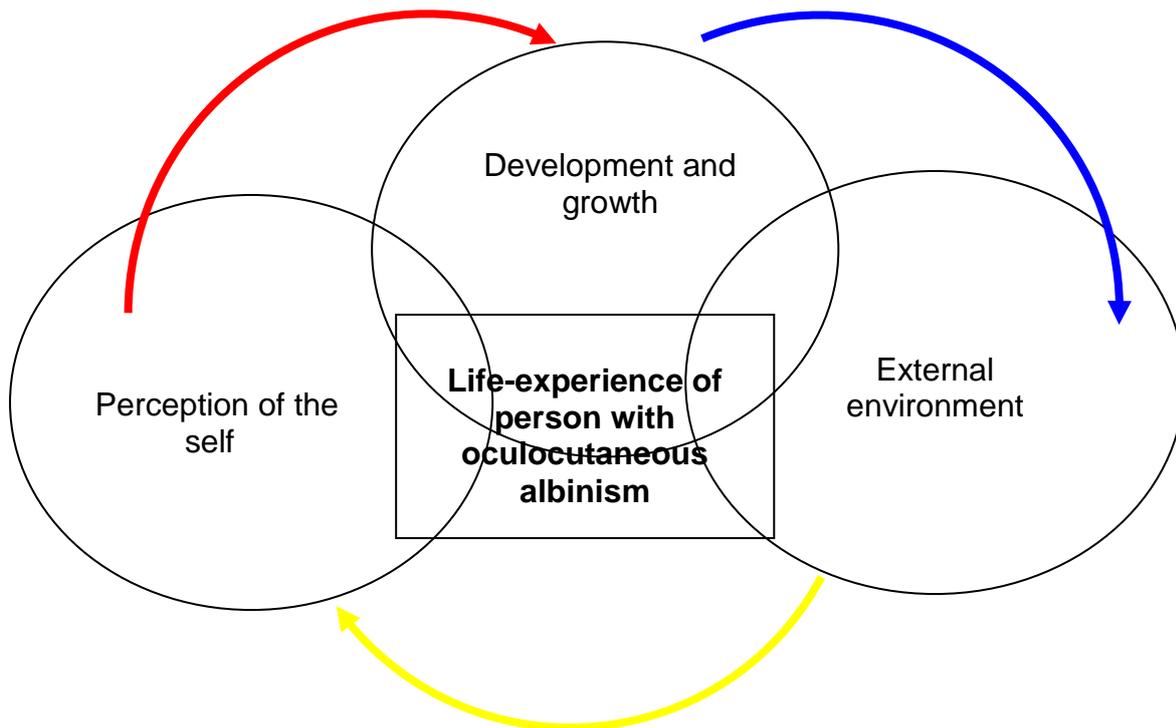
Figure 3.1

Main themes of the findings

Within the main themes, categories and sub-categories were identified as shown in Table 3.3. The researcher used bracketing and intuiting during data collection. Therefore, subjective description of the life-experience of people with oculocutaneous albinism was formed independently of any theoretical orientation. However, during data analysis, the researcher reflected the categories of the main themes within the dimensions of Neuman's systems model, namely psychological, physiological, socio-cultural, spiritual, and developmental dimensions.

The potential of Neuman's systems model in the findings lies in Neuman's description that the psychological, physiological, socio-cultural, spiritual and developmental dimensions occur simultaneously, comprehensively and are always present in a human being (Stanhope & Lancaster 2004:210; Neuman 1995:20). It emerged that these dimensions had fundamental comprehensive influences in the participants' life-experience.

Table 3.3 Overview of the themes, categories and sub-categories in participants' life-experience

THEME	CATEGORIES	SUB-CATEGORIES
Perception of the 'self' by persons with oculocutaneous albinism	Importance of the 'self' within the psychological dimension	Participants express need and importance of: *Self-concept *Self-actualisation *Self-esteem *Self-worthiness
	Expression of interdependence within the psychological dimension	*Relationship with the family *Relationship with the community
Experience of persons with oculocutaneous albinism with the external environment	Experience within socio-cultural dimension	*Participants express need for sense of belonging *Feeling of being stigmatised because of myths and superstition
	Experience within spiritual dimension	*Importance of strong religious belief *Role of the church in the participants' lives
Expression of need for development and growth	Experience within developmental dimension	Participants noting the important roles of: *Various departments *Private sectors *Support groups
	Experience within physiological dimension	*Care of the skin and eyes *Vulnerability to skin cancer

3.3.1 Participants' perception of the 'self'

The focus of the main theme of perception of the 'self' was the participants' recognising that they 'themselves' played an important role in their own lives, but most importantly also in relation to other people. The experience of the perception of the self-concept by persons with oculocutaneous albinism is in line with the description of perception and the self-concept in the literature review.

King describes perception as a process of human interaction with the environment, which influences behaviour, provides meaning to experience, and represents the individual's image of reality (King 1990:21).

Self-concept is a mental image an individual has of himself or herself, based on one's ideas and attitudes about one-self and one's personality (*Blackwell's Nursing Dictionary* 2005:545 and Del-Orto & Marinelli 1995:660).

In this regard, Jordaan and Jordaan (1998:616) point out that in everyday speech, the word "self" is linked to various nouns which express the notion that there are things that belong to the individual's essential nature, and that there is an inner self through which an individual experiences the self and the world.

The theme on the perception of the self by persons with oculocutaneous albinism is reflected within the psychological dimension of Neuman's systems model. According to Neuman, the psychological dimension refers to mental processes and relationship (Neuman 1995:58).   Two categories could therefore be identified within the theme on the perception of the self, namely the participants' noting the importance of the 'self' within the psychological dimension, and expression of interdependence within the psychological dimension.

3.3.1.1   Importance of the 'self' within the psychological dimension

The study found the role of the 'self' as experienced by the participants was important within the psychological dimension. The self-concept emerged from the majority of the participants. Of the fifteen participants, fourteen felt very strongly that the onus is on the 'self' effort of the person with oculocutaneous albinism to improve his/her self-worthiness:

> *So I started accepting albinism from such motivations, and I am the one*
> *facilitating the support group now…*
> *I grew up that way. That's why I even did part-time modeling. Because I*
> *knew…Yes I had this thing in my mind that I want to show the world…!*

The 'self' was found to be an important concept as it relates to other themes of this research. King also supports the importance of the 'self', and indicates that self is a composite of thoughts and feelings which constitute a person's awareness of his individual existence, his conception of who and what he is (King 1990:48).

In Manganyi, Kromberg and Jenkins' (1974:112) study on oculocutaneous albinism in South Africa, the persons with oculocutaneous albinism and the control group engaged in creating a self-portrait to determine their intellectual maturity, and body image differentiation.

Manganyi et al (1974:112) found that persons with oculocutaneous albinism found themselves confronted with a negative self-evaluation or identity, and the control group fared better in creating their self-portrait.

This study found that the participants' self-concept within the psychological dimension was one of the fundamental experiences in view of self-actualisation, self-esteem and self-worthiness (see figure 3.2).
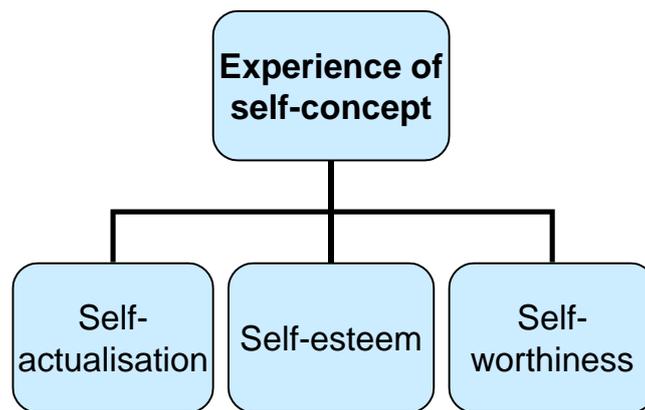
Figure 3.2
Participants' experience of 'self'

- **Participants' experience of self-actualisation**

The significance of the self-actualisation concept emerged in the findings, and the participants expressed the need 'themselves' to take ownership to raise awareness on oculocutaneous albinism. This emerged very strongly from the majority of the participants.

In this regard, Jordaan and Jordaan (1998:582) refer to Maslow, who points out the tendency of people to realise the self-actualisation process. Maslow emphasises that in the self-actualisation process, people strive to discover and realise their full and true potential.

The participants' tendency to realise their full and true potential was expressed as follows:

> *…that must be done by us people with albinism [raise awareness on oculocutaneous albinism]. We must make all Departments aware of our needs, to say what must be done.*

> *We must stand up as people with albinism...if we stand up …they [media] can see that these people, really, want to be known.*

> *I think we should go out to the people … Others they are still locked in the houses …there is no need to be afraid and hide away …ok, there is information on albinism...**but we must go out!***

Furthermore, the participants expressed a need to maintain self-esteem, and this is in accordance with Maslow's self-actualisation theory (George 2002:150).

- **Participants' experience of high self-esteem**

The study found a high self-esteem in most of the participants. Self-esteem is a complementary feature of the self-concept and is regarded as the most important and powerful value in the motivation system (*Oxford Textbook of Public Health* 2002:839).

Self-esteem is associated with having a positive attitude or acceptance of oneself, and is mostly associated with high performance. Maslow (1970:109-111) emphasises that the healthiest form of self-esteem is based on genuine respect shown to an individual by other people, because of the intrinsic human worth. The participants' self-esteem was noted as follows:

> *If I want to learn to drive. I can learn to drive, so I have just started, my eyes have not been tested, that is the other thing because I went to the testing station, and was tested, and could not succeed …and I told myself I will not go back there again…then they said I can drive ..but with binoculars…I bought them …they got it from America so…but I have started with lessons [driving lessons].*

According to Darling (1979:30), experimental studies show that people with high self-esteem have a greater willingness to perform as shown in this formula:

$$\text{Self-esteem} = \frac{\text{Success}}{\text{Pretensions (justifiable claim)}}$$

In supporting Darling (1979:30), Dell-Orto and Marinelli (1995:660) point out that the physical, emotional and intellectual resources enhance the social interaction of people, and build their self-esteem to achieve their goals in life.

It emerged from analysis of the participants' biographical characteristics that the self-esteem of the participants who socialised through extramural activities and sports was much higher than of those who did not.

It was also evident that the participants had the same experience as Salif Keita, an international singing star who has oculocutaneous albinism. Salif was rejected in his own community. However, Salif took ownership of his life (Danna 1998:100)*: "Salif Keita was rejected from birth because of his white skin. But he overcame the odds to become an International singing star, an ambassador for Africa, and fighter for people with oculocutaneous albinism."

People with oculocutaneous albinism are easy to recognise through their highly visible skin colour. It emerged that people with oculocutaneous albinism do not want to be discriminated on the basis of their visible light skin colour, or for any other reason. The participants were all from the Black population, making them highly visible in the community.

The study found that their skin colour also has a significant effect on their self-esteem. According to the participants:

> *...the only difference is the skin pigment…you know… that*
> *negativity towards albinism, it's always there, especially in blacks,*
> *it will always be there…JA!*

> ...*ke letlalo fela le shortang pigmentation, so ga go reye gore fa ke le*
> *yana*, *I am different. Colour e ke yona e different.* (It's only the skin
> that lacks pigmentation. So it does not mean I am in any way different.
> It's only the skin colour that is different.)

From these remarks, the participants did feel socially stigmatised on the basis of their skin. Christianson et al (2006:25) highlight the significance of the light skin colour of people with oculocutaneous albinism. Christianson et al (2006:25) describe oculocutaneous albinism as a highly visible birth defect in dark-skinned people. They state further that as a consequence of the visibility of the light skin colour, people with oculocutaneous albinism are often socially stigmatised.

Gaigher et al (2002:4) emphasise the relationship of the skin colour of people with oculocutaneous albinism (physical body) and the world. In a sociological study of children with oculocutaneous albinism at a special school in Limpopo Province, Gaigher et al (2002:4) found that the problems experienced by people with oculocutaneous albinism stem primarily not from their physical differences, but from the way others (persons in the world) respond to those differences, and from the social and physical environment that the persons with oculocutaneous albinism have to cope with.

Gaigher et al's (2002:4) findings and those of this study concur with Ndlovu's (2005:3) statement that people with oculocutaneous albinism "are tired of being abused, discriminated against, cursed and provoked wherever they go".

According to Dell-Orto and Marinelli (1995:601), stigma is usually associated with deviation from what is normally expected. Due to their highly visible skin colour, people with oculocutaneous albinism may be stigmatised, and viewed by the community as deviations from what is normally expected in everyday social interchanges. Stigma can therefore serve as a barrier to social integration, and alter or disrupt social relationships (Gaigher et al 2002:4; Kromberg & Jenkins 1984(b); Philpott 1995:144).

Jordaan and Jordaan (1998:616) emphasise that our very sense of whom we are, and our important social relations are intimately connected with our bodies. Jordaan and Jordaan add further that how people relate with their body has significance in their lives. In this regard, Urbani (1992:35) emphasises that

- Human existence in the world takes place through the body;
- The body is at the same time man's means of admission to the world (also to other people and things);
- Through our bodies we actively establish our own world; and
- When the body is [handicapped], the person involved cannot dissociate himself from it, because it is his or her body.

It is evident from the participants' experience that their highly visible skin colour influenced their interaction in the world as stated by Urbani (1992:35) above. The participants were stigmatised especially due to their skin colour. This is despite the fact that Urbani (1992:35) wrote about this 25 years ago!

Dell-Orto and Marinelli (1995:602) support Urbani (1992), and point out that body image and self-concept play an important part in an individual's ability to adjust to a disability. This study found that the participants' concept of body image encompassed more than a perception of physical appearance, it also included perception of feelings and thought.

A strong link thus emerged in the findings between self-concept, body image and self-esteem. The participants improved their self-esteem in various ways through motivation and joining support groups:

> *So I started accepting albinism as genetic problem*
> *from such motivations...and I am the one facilitating the*
> *support group now…*

At the same time, however, some participants raised concern that there was reluctance among other people with oculocutaneous albinism to take ownership on oculocutaneous albinism:

> *But it's just that we also are not strong … we tend to be reluctant*
> *On the other hand...Rona Ka borona (repeating)…hei…for*
> *instance, I don't know, I don't know! (emphasising a point)*
> *we are so reluctant, I don't know…maybe we are afraid of*
> *people or what…..!*

Self-esteem is an important concept also noted by the Albinism Society of South Africa (ASSA). The Chairperson of ASSA emphasises that the aim of ASSA is to enhance the self-esteem of people with oculocutaneous albinism (Ngakane 1998:13). It also emerged in the findings of this study that the self-esteem of the participants, who are members of ASSA, was high and they appreciated the support provided by ASSA:

> *Because what they do at Albinism Society is counselling …and*
> *we have like youth groups…awareness campaigns...*
> *you get a lot of information…and you also accept yourself…*
> *the main purpose of ASSA is to help you accept yourself…*

> *In 1999…I joined a support group…as per influence of a friend of*
> *my mother…and meeting with others helped me a lot, especially*
> *other older mature people with albinism who motivated me a lot…*

In this regard, Maslow (1970:110-115) points out that self-esteem and acceptance contributes significantly to people's experience of human dignity. Dignity is one of the rights recognised under chapter 2 of the Bill of Rights of the Constitution of South Africa Act, 108 of 1996. Section 10 of the Constitution provides that "Everyone has inherent dignity and the right to have their dignity respected and protected" (South Africa 1996:7).

However, this study found that not all the participants' self-esteem was high. Low self-esteem was apparent especially among the few participants who were presently not employed.

Their unemployment status clearly affected their self-worthiness:

> *And I still feel they [employers] don't want to give me chance.*
> *I went to so many interviews for work. They don't take me,*
> *I always get a letter to say it was unsuccessful, something*
> *like that. The other day I went to X [company] for an interview.*
> *I think I was so bottled up, and I told them "I know you're*
> *not going to take me because you guys see me and think,*
> *Oh no, she is not right" (Participant was emotional and sobbing.)*

With regard to the importance of work to the psychological and economic health of the individual, Dell-Orto and Marinelli (1995:767) cite Sigmund Freud who recognises love and work as the basic requirements of human existence. Various facets of the definition of work indicate the social impact of work in our lives. Dell-Orto and Marinelli (1995:767) define work as "goal-directed activity for social, economic or other desired accomplishments or outcomes".

Dell-Orto and Marinelli (1995:768) further cite Wright (1980:343), who indicates that the most important thing about work is not remuneration but its activity and product. This study found that the unemployment status of the few participants clearly affected their self-esteem.

Nevertheless, the high self-esteem of the majority of the participants contradicted Gaigher et al's (2002:6) findings of low self-esteem of all the learners with oculocutaneous albinism in a special school. Gaigher et al (2002:6) found that these learners seemed to be battling with a social desire to be with family and friends, as they were "forced" to choose friends from amongst the disabled children in the school. Gaigher et al (2002:6) describe this situation as "loss of self", which could be a form of suffering experienced by people with oculocutaneous albinism.

- **Participants' experience of self-worthiness**

In this study, self-worthiness emanated mainly from the high self-esteem of the majority of the participants. The participants experienced self-worthiness in different ways, especially with regard to employment status. It was evident from the

findings that there was a strong feeling that being employed improved one's self-esteem. For instance, during a debriefing session, one of the participants emphasised the following point:

> *My main concern is for me to get a job. I don't know. That's how I feel.*
> *Ja, maybe it will improve the way I feel now, maybe it will be different.*
> *Not working is making me feel less of myself…*

According to Maslow's self-actualisation theory, the esteem need is based on the fact that people need to feel self-respect and a sense of their own value, and also that they are respected and appreciated by other people (Maslow 1970:109-113). Self-worthiness in this study was influenced by needs for love, security, and stability as proposed in Maslow's self-actualisation theory. It therefore emerged that the study participants also needed to be loved and secure:

> *If you realise that they [family] have accepted you, then you accept*
> *yourself as well…and you say "Oh there is nothing wrong with me".*

On the other hand, it also emerged that a combination of lack of stability in terms of further education, employment and marital status brought about a lack of self-worthiness:

> *I have been unemployed since I left school [Std 10]. This year it's ten*
> *years now. And we have a ten years' school reunion, class of '96, and*
> *I don't think I will attend it, I told my friend, because I don't have a*
> *husband, I don't have child, I don't have a job, and all of the other*
> *friends have all those things.*

The sentiment expressed in the above life-experience of the need for self-worthiness was consistent with Erickson, Tomlin and Swain's (1983:56) modeling and role-modeling processes. These processes are holistic and describe individuals as being born with an inherent desire to fulfil their self-potential (Erickson et al 1983:59). Modeling is a process to develop an understanding of the client's world as the client perceives it, whilst role modeling is the facilitation of the individual in attaining, maintaining or promoting health through purposeful intervention (Erickson et al 1983:60-65).

On the other hand, Suinn (1979:111) questions the significance of marital status in enhancing an individual's self-worthiness, and maintains that marital status may be as much a cause as it is a result of emotional stability.

Suin (1979) is not current literature, but brings about an important discussion on marriage, which is one of the oldest institution in society. It emerged in the findings of this research that the majority of the participants were single and, like any other person, expressed a need for sense of belonging in a relationship. Suinn (1979:111) holds that the married state may bring with it certain stabilising features such as positive interpersonal relationships, sense of belonging and worth, and reassuring companionship.

Dell-Orto and Marinelli (1995:662) also emphasise the importance of a sense of belonging: "Most people with a disability are well-adjusted individuals with a positive self-image, and struggle for recognition as competent and productive people, just like individuals without disabilities".

3.3.1.2   Expression of interdependence within the psychological dimension

It was evident from the participants' experience that their self-concept and self-esteem were also influenced by their relationship with their families and the community. In this regard, the participants expressed a sense of interdependence to improve their self-esteem. Two sub-categories emerged from this category, namely relationship of people with oculocutaneous albinism with their family, and relationship of people with oculocutaneous albinism with the community.

- *Relationship with the family*

As the participants related their life-experience, the significant role of the family emerged as one of the important sub-categories. The family is regarded as the primary environment for rearing a child, and the family's role is regarded as a necessity, not as an opportunity   (Dell-Orto & Marinelli 1995:44). The findings of this study revealed that families of people with oculocutaneous albinism, and families without people with oculocutaneous albinism both have a social role to enhance the self-esteem of persons with oculocutaneous albinism.

According to Jordaan and Jordaan (1998:779), a social role refers to expected behavioural patterns, which people demonstrate in relation to other individuals, groups and communities in certain situations.

The findings of this study therefore reveal patterns of expected behaviours of families with regards to oculocutaneous albinism as a congenital disorder. One of the participants responded as follows when asked about her anchor in life:

> *…it's my family...it's my family ….nothing else…I mean it*
> *they are the only people that before you can convince people*
> *outside they [parents] should convince them [community]…*

The important role of the family is supported by Professor Anton van Niekerk from the University of Stellenbosch, in the Western Cape, South Africa, who pronounced an impact of genetics on families during a Human Genetics Conference in South Africa in 2003. Van Niekerk explained that the family has to cope with the genetic information and its implications, as the genes are passed down to the children (Genetic Science 2003:2), "Genetic science lives and functions at family level."

In line with van Niekerk's remarks, the Human Genetics Policy Guidelines emphasise that birth defects have enormous emotional, psycho-social, and financial implications for the family (Department of Health 2001:2). In this regard, this study found that the parents were playing their role to make sure that the participants had appropriate protective clothes, and also purchased sun protective factor (SPF) creams and other creams, which were apparently costly.

One of the participants shared the important role of the family as follows:

> *….and there was always Dermolin in the house to help with bites and burns*
> *…for my skin I use moisturiser with sun protective factor 30 and aquascream.*
> *The face one is R150,00 and the body one is R45,00 …*

They [parents] used to buy protective clothes, long sleeves, hats
and all those things ..and I used to wonder why we should dress up
that way...and I only understand it now as an adult in retrospect ...
to say okay it was to protect my skin...

The World Health Organization (2000:8) notes the significant role of the family in the lives of children with oculocutaneous albinism.

The WHO (2000:8) recommends that further work is essential to develop scientific principles and methods for measuring the financial and non-financial burden that birth defects/congenital disorders impose on the affected individual, families and communities. The WHO (2000:8) also recommends planning appropriate programmes on congenital disorders at the primary health care level. According to the Policy Guidelines for the Management and Prevention of Genetic Disorders, Birth Defects and Disabilities, oculocutaneous albinism is one of the priority congenital disorders (Department of Health 2001:6).

It emerged in the findings of this study that the positive attitude of both parents was essential to enhance the self-worthiness of the participants. Furthermore, the financial and non-financial burden of a child with oculocutaneous albinism is the responsibility of both parents. Most importantly, the findings also revealed that the role of the father in the participants' lives should not be underestimated:

Oh! to be honest my parents are very supportive. Very! They are
very proud of me, they don't hide me. My father talks about me, even
at work he used to talk of me "My child is an albino, the only girl". He
is very proud of me ...

...he (father) gave me support . Actually both my parents gave support
…and said I must not worry about those things (teasing and mockery in
the streets)…That's why I managed to attend mainstream school...up
until I matriculated …

One of the participants had two brothers with oculocutaneous albinism:

> *They [parents] accepted us and they were so loving…*

The significant role of mothers in the nuclear family emerged. The mothers recognised the rights of their children with oculocutaneous albinism to quality education, and quality health care. There were instances where the participants did experience problems at school. However, most of the time the mothers intervened at school on their behalf:

> *I think at primary [school]...there were a few teachers*
> *…I don't know whether they did not want to understand as*
> *such …I don't know…but let me say, they were ignorant.*
> *But my mother explained to them that I am a human being.*
> *Like after school I would relate to her that it was like this*
> *and this at school, and she attended to the situation as such…*

It also emerged that while acknowledging the role of both parents in the life of a child with oculocutaneous albinism, the participants felt that the most important person to be counselled was the mother. This was in view of the possibility of young mothers fearing being rejected in society after delivery of a child with oculocutaneous albinism:

> *Ahmmm … I can say at the clinics and hospitals, you can find*
> *out that especially these young mothers give birth to*
> *a child with albinism…so you find she does not understand*
> *and she worry how the community will treat her. So I think we*
> *should start there ... from the mothers. They must get counselling,*
> *and get enough education about albinism at the clinic...*

The role of parents in the education of their children is supported by Swick (1997:154), who found that parents have the potential to empower schools and communities in becoming caring places that embrace children in nurturing ways.

In a study to determine impact of the child's disability on an African family, Nkabinde and Ngwenya (1996:36) found the role of the parents to be critical.

Nkabinde and Ngwenya (1996:36) interviewed nine African parents of children with disabilities in various geographical locations, and found that the parents' major concern was to make sure that their children with disabilities received quality education.

One of the aspects that emerged in this study was that the participants' parents tried not to be overprotective of them.

It is assumed that not being overprotective enhanced the participants' self-esteem and confidence to take ownership of their live:

> *Let me say it starts in the family …they never took me like a different person…*

These findings concur with Gaigher et al's (2002:9) finding that nearly all the participants in their study indicated that their parents or guardians treated them similar to the rest of the family, despite their eyesight and skin problems.

According to Jordaan and Jordaan (1998:125), overprotectiveness is an excessive concern of parents over their child's behaviour. Furthermore, it stems from reality conditions like psychological conditions, social isolation, and underlying attitudes of rejection, of either the child or the parent (Jordaan & Jordaan 1998:125). Accordingly, the participants' parents appeared to have no underlying attitudes of rejection of their children, and were therefore not overprotecting them.

Gaigher et al (2002:10) emphasise the impact of lack of family support for a child with oculocutaneous albinism saying that if "rejection starts from birth, the chances are high that the person with oculocutaneous albinism will later succumb to society's negative attitudes and stereotyping".

This study also found that the extended family members played an important role in enhancing the participants' self-esteem. The participants related their experience of how the presence of an extended family member with albinism influenced being accepted in the family.

In this regard, the participants' biographical characteristics showed that within the first degree of the family tree, four of the fifteen participants had brothers and sisters with oculocutaneous albinism. None of the participants had parents with oculocutaneous albinism. In addition, seven of the participants did have extended family members with oculocutaneous albinism, for example great-grand aunts, grandparents, aunts and nephews. The participants noted the role of the extended family as follows:

> *They [paternal extended family members] play a big role cause they feel I am the only ...because as great grandmother is no more alive [paternal great grandmother had oculocutaneous albinism], they feel that I am the only one who is left in the family …so they are there in my life.*

- *Relationship with the community*

The participants emphasised that the parents of children without oculocutaneous albinism had an important role to educate their children to accept people with oculocutaneous albinism, and not to "mock" them. Nevertheless, the participants acknowledged the efforts of other parents who disciplined their children when they called people with oculocutaneous albinism names like "*lekgoa*" (white person).

> *And then ba bangwe ba, o kreye gore motho, e le motho a tsamaa le mama hae, Mm..and then a bare "mama bona umlungu' and then mama hae a ka se mo kgale, o kreiya ba bangwe ba tshega.* (crying).
> (Sometimes the young child may be in the company of his mother, and make derogatory remarks, and the mother does not reprimand her child, and the mother even goes to the extent of laughing).

> *And then ba bangwe o kreye mama hae a mo emisa/ warnisa (scold) a re: "nzo ko shaya o sa khulumi njalo."* (However, in other instances the mother does scold the young child when he or she makes those derogatory remarks, and the mother strongly reprimands her child: I'L BEAT YOU UP, DON'T SAY THAT.)

It therefore emerged that besides the family, the role of the community is essential to enhance the self-concept of persons with oculocutaneous albinism. This study found that there is a need to facilitate sense of community and belonging of people with oculocutaneous albinism. The participants emphasised that people with oculocutaneous albinism should be accepted in the community. At the same time, however, some of the participants expressed their appreciation that there was some improvement in terms of the community's attitude towards people with oculocutaneous albinism:

> *I think there is some improvement…most parents were afraid to expose*
> *their children (with albinism) to society ...but now I think they are free*
> *... they can play and do things done by other children …they don't have*
> *a problem of who will say what…so from that time to now ...I think*
> *things are all right...they are getting there...but not yet the*re!

It also emerged that the community's positive attitude in turn improved the confidence of the parents to let their children with oculocutaneous albinism socialise with their peers. The sense of community and belonging therefore enhanced the participants' self-concept. Beckman, Barnwell, Horn, Hanson, Gutierrez and Lieber (1998:131) emphasise the concept of sense of belonging. Beckman et al (1998:131) found that families of children both with, and without disabilities reported the importance of having a sense of connection with each other, through different contexts of extended families, neighbourhoods and in some more organised community activities.

It therefore clearly emerged in this study that the community played a fundamental role in the participants' socialising, and thus in enhancing their self-concept.

3.3.2 Participants' experience of the external environment

The study found that the external environment played an important role in the participants' life-experience to enhance their self-esteem. George (2002:290) points out that the external environment exists outside the person with oculocutaneous albinism, and is made up of all interactive influences that are outside the boundaries of the person.

In this study, two categories were identified from the participants' experience of the external environment. The categories are reflected in Neuman's systems model, namely experience in the socio-cultural dimension, and experience in the spiritual dimension.

3.3.2.1   Experience of the socio-cultural dimension

Neuman refers to the socio-cultural dimension as systems functions that relate to social and cultural expectations and activities (Neuman 1995:83). This study revealed that the socio-cultural dimension played a fundamental role in enhancing the participants' self-esteem. In this regard, the participants were striving to meet the social and cultural expectations so as to discover their full potential in the community. Two sub-categories were thus identified, namely expression of need for a sense of belonging, and a feeling of being stigmatised on the basis of myths and superstitions.

- *Participants' need for a sense of belonging*

Despite the high self-esteem of the majority of the participants, it emerged from the findings that there is a need for sense of belonging by people with oculocutaneous albinism. It is evident therefore that the study participants, one way or the other experienced social isolation in their lives. Jordaan and Jordaan (1998:672) point out that social isolation is a component of loneliness. Social loneliness is described as a lack of friends, and emotional loneliness is lack of a close and deep relationship with a special person (Jordaan & Jordaan 1998:672).

It was difficult for some of the participants to have friends at school, and social loneliness was thus felt mostly at the school level:

> *Hmm…I can start as when I was growing up…and realised that I am different*
> *from other children…and then it's where the problem starts …and when you*
> *go to school ...you don't have friends…because of albinism…and you*
> *think there is no one who loves you…then when it comes to home …*
> *you think they treat you differently…*

The participants therefore revealed the need for a sense of belonging in the family, at school, church and generally in the community.

⋏ *Sense of belonging in the family*

At the family level, some of the participants shared their experience of loneliness as they grew older. The concerned participants felt suspicious that their parents were discriminatory towards them because of oculocutaneous albinism. In this regard,one of the study participants remarked:

> *...because of albinism, and you think there is no one who loves you.*
> *Then when it comes to home, you think they [family] treat you differently,*
> *and when I was still young, I felt that cause I have albinism, they*
> *treat me differently. For example, I have a younger brother, who things*
> *were done for, and not necessarily done for me, and I felt somehow*
> *uncomfortable with it...*

The study also found that unemployment might exacerbate the tendency to social isolation in the family. Despite the evident family support provided, one unemployed participant experienced social isolation:

> *...and they [family] are also on the look out for me for job…but sometimes*
> *I feel they just hide it away. Maybe they feel ashamed of me….*

⋏ **Sense of belonging at school level**

It emerged from the findings that the school or educational institutions played a fundamental role in the participants' socio-cultural dimension. The type of friends met at school either facilitated a sense of belonging or worsened the social isolation:

> *Others [friends] asked 'Why are your eyes doing that [rolling]', and that thing*
> *hurt me. So at home they took me to special school. Because I did not*
> *have a problem there, because everybody is educated there. You see,*
> *at primary school, other teachers didn't like people with albinism at all.*
> *And hence my parents took me to a special school. At normal school*

> *I couldn't cope. Like in class I was very slow, and other children laugh at you and*
> *so on…*
> *At school you tend to be isolated ... and me and P [another learner with*
> *albinism at the same at school] we did not meet, because we thought*
> *both of us have albinism …and what would the people say to see us together…*

The findings of this study concur with Gaigher et al's (2002:6) finding that the majority of learners with oculocutaneous albinism (63%), were of the opinion that the problems they experienced at school were more of a social than an educational nature.

The significant role of teachers in the participants' lives emerged as a contributor to either acceptance or alienation. This was especially in view of the participants' poor eyesight, which in other instances influenced their progress at school. Nevertheless, the participants appreciated the support provided by some teachers:

> *...at school it was nice. I had nice friends. Teachers helped me very much.*
> *If there was stuff on the blackboard, they [teachers] helped me write it down,*
> *and if the teacher didn't write for me, then I would write from my friend's book …*

It also emerged from the findings that other teachers did not understand the participants' plight, especially their eye problems. The findings revealed that the negative attitude of some of the teachers affected the participants' self-esteem:

> *At primary [school], I think teachers didn't understand albinism. There was a*
> *time I stayed about a year not attending school because of eye problems.*
> *I was about 11-13 years old. Well, also my parents did not understand*
> *what's happening [on oculocutaneous albinism]. Because one doctor suggested*
> *to them why they don't take me to a special school. So my mother refused.*

However, it was evident that despite some teachers who did not understand the participants' plight, most teachers are sensitive to the needs of learners with oculocutaneous albinism. It emerged that most learners with oculocutaneous albinism could be classified as learners with special needs, due to their eye problems. Nevertheless, some of the participants did not agree that they should be classified as such because of the stigma attached to 'people with special needs'.

The positive and caring attitude of the majority of teachers towards the participants was consistent with Davies and Green's (1998:97) findings in a qualitative study of 113 teachers from six local schools in the Cape Town area in South Africa. Davies and Green (1998:97) found that most of the teachers (86%) indicated that it would be acceptable to them to teach learners with special educational needs, either without assistance, or with monthly consultation. Davies and Green (1998:100) also observed the effect of the presence of a learner with a special educational need in class, and the reaction such a child might evoke.

The present study also revealed that the presence of a learner with oculocutaneous albinism evoked various reactions from other learners:

> *Yes, ko mainstream it was a problem, ke gore o tla tsena fela a*
> *kwale dihlakanyana di le kana fela, so you are expected, gore o tomole mahlo*
> *o di bone, of which mahlo a o ga a a kgone (*participant reflecting saying it
> laughingly) *And ge o emmella o thoma o atamela mole ba re "EESH WA RE*
> *SIRA" dilo tse tshwanang le tseo* (At the mainstream school, it was
> a problem, because in class you will be expected to see the small
> writing on the board like all the other learners, and when you
> decide to come nearer the board, other learners scream at you
> that you are a nuisance!).

However, other participants seemed not to experience problems of having friends at school. Nevertheless, some of these friends were inquisitive and asked personal, sensitive questions:

> *At school, it was sharp, I managed to socialise with friends. I did not have*
> *problems with friends you see. But sometimes they will ask you silly*
> *questions, whether you bleed* [the participant saying it laughingly] *…and*
> *then I said "Oh yes, ke motho lenna* (meaning I am a human being too),
> *I am like you, its just my skin"...Oh hey! very silly questions like "Do*
> *you go to the toilet ?"…like that. Eei.. "Do you have a boyfriend?" As if you are not*
> *a human being, you see...*

It was evident from the discussions that friends played an important role in the participants' lives. In 1998 Salif Keita, one of the renowned people with albinism, expressed his appreciation of the role of friends in his life as follows: "I knew nothing about show business. Friends who looked after me helped me to have a visa, which I needed as a foreigner. I owe them a lot. Because of them I was able to get my own home and start a family."

In other instances friends of the participants seemed to be there for them to make them feel part of the group. The care and support from friends enhanced the participants' self-concept. Some of the participants' friends intervened on their behalf as follows:

> *And then ha ke tsamaa le dichumi tsaka, o kreye motho a le, a re "lekgowa",*
> *and* (raising voice a bit*) dichumi tsa ka o tla utlwa ba mo rasetsa ba re "Hee*
> *wena… Ha se thaka ya gago, and ha se chomi ya gago, o sa tlo re phaphela,*
> *ha nka go utlwa gape ke tlile go go betha.* (When I am with my friends, and
> somebody calls me a "white" person, my friends intervene and tell that
> person that next time they will hit that person if he or she can make that remark
> again)

⼊ *Sense of belonging at church*

It also emerged that the church played a significant role in the participants' socio-cultural experience (see section 3.3.2.2 on experience within the spiritual dimension). The important role of the church in the socio-cultural perspective, and the need for a sense of belonging was expressed as follows by one of the participants:

> *I didn't attend church for many years, think for 3 to 4 years …but now I*
> *attend church since I talked to the pastor's wife. Ja, people are now*
> *more nicer, especially those that I thought they did not like me…we*
> *cleaned the church one Saturday for a conference...and that day it was*
> *nice...and since then we see each other we greet each other nicely ...and*
> *smile and talk...*

⚐ *At community level*

The participants' social isolation at the community level also emerged in the findings**.** Some of the participants felt rejected by the community, and one of the participants raised this concern as follows:

> *I think people with albinism, they are sitting there, nobody is looking at them…*

The researcher noted that in the biological characteristics, most of the participants were not engaged in some form of extramural or sports activities. Nine of the participants indicated that they were not engaged in any extramural or sports activities. Only six were engaged in some form of extramural or sports activities. Most of the participants indicated that it is due to time constraints that they did not to participate in extramural or sports activities.

The researcher assumed that these study participants were telling the truth due to the trustworthy atmosphere that prevailed during the interviews. However, the findings of less participation in extramural or sports activities were contrary to Gaigher et al's (2002) study.

Gaigher et al (2002:4) found that all their participants took part in some form of sport. However, the researcher is of the opinion that this difference of engaging in sports or extramural activities could be attributed to the age ranges in the two studies. Gaigher et al's (2002:4) participants were aged between 5 and 18 years and, in addition, were all learners still attending school. In this study, the participants were between 18 and 48 years old, and only one was still attending school.

In this regard, the researcher is of the opinion that sports or extramural activities could be ways to enhance sense of belonging of people with oculocutaneous albinism in the community.

- ***Feeling of being stigmatised due to myths and superstitions***

It was evident from the findings that myths and superstitions on oculocutaneous albinism affected the participants' self-concept. The findings revealed that stigma on oculocutaneous albinism was also initiated by myths and superstitious beliefs in society. In this regard, Jordaan and Jordaan (1998:799) point out that beliefs in society affect people's attitudes to life, life style and behaviour in religious, ethical, political and social contexts.

All the participants shared their experience of various myths and superstitions on oculocutaneous albinism, and most importantly their reaction to them. It emerged clearly that these unscientific perceptions of oculocutaneous albinism affected the participants' everyday social life.

The most common myth and superstition expressed by all the participants was that people with oculocutaneous albinism do not die naturally, they simply disappear. Nevertheless, the participants differed in their reaction to this myth. Most of the participants thought that the myth that they disappear is a starting point to educate the community on oculocutaneous albinism, whilst other participants took it as an insult:

> *...those who don't understand that we are people, and so on, the*
> *issues of disappearing, we must be there all people with albinism.*
> *Ok, there is information on albinism, but we must go out!*

> *When I arrived [at high school] I got a friend, she was ok. But I*
> *realised she was sort of afraid of me, and I asked her, and she*
> *said "No Sipho [not real name], I used to hear all the stories like,*
> *I just think you will disappear". And it was like "Ao! my friend,*
> *what is the difference between me and you? Do you see this*
> *skin, how could you think I will disappear?"*

Other participants joked about the myth that they disappear:

> *…like the myths that we disappear we joke about it that "Oh! And*
> *then its less expense for the family if we disappear, then there is*
> *no funeral"* (laughing*). So everything that the people say it in a*
> *negative way, then we turn it into something positive.*

> *Hmmm, people like to say we don't die, we disappear, and we have*
> *turned it into a joke (*some laughter*). But at the moment, hey! Ja,*
> *they like to say that. And we respond that we die just like you, if ever*
> *we disappear we should just as well just have appeared from nowhere!*

The assumption was that the participants in this research decided to make realistic and rational choice to joke about the myths, in order to adjust in the community. In this regard, Krech, Crutchfield and Livson (1979:572) point out that a person may experience a considerable degree and persistence of frustration and conflict, but may continue to strive towards the goal and to look for new paths, and seek to make realistic and rational choices to adjust.

Dell-Orto and Marinelli (1995:260) emphasise that adjustment depends on various factors, which include the type and severity of the disability, the origin of the condition, support systems, self-perception stemming from social interaction with others, and the individual's ability to make autonomous decisions for the management of his or her life. From the findings it was evident that the adjustment factors identified by Dell-Orto and Marinelli (1995:260) were fundamental to enhance the participants' self-esteem during the adjustment process.

In other instances some of the participants 'ignored' these sensitive myths:

> *Nna ebile ke ntse ke sena taba le bone, ke ntse ke ba ignora.*
> *(I ended up not caring what they say, I ignore them.).*

It could be argued that the reaction of ignoring these myths, was just avoidance of further hurting by the perpetrators. Dell-Orto and Marinelli (1995:260) describe this reaction as an escape reaction, which is the tendency of people to escape from the frustrating situation.

However, Dell-Orto and Marinelli (1995:260) warn that the escape may only be temporary, and the original motive may still remain, and continue to induce tension.

The participants also related other myths that evoked emotional feelings. In some instances these myths provoked aggression on the part of the victimised participants. Krech et al (1979:572) are of the opinion that anger is more likely to occur, and to be expressed in an aggressive action against the thwarting object, and particularly if the thwarting seems to be deliberate or unreasonable. Therefore it was assumed that the aggressive reaction of some of the participants was that they felt the hurting myths was a deliberate provocation, and felt victimised**:**

> *I was about 15-16 years old when I hit them, for this tendency of spitting when*
> *they saw a person with albinism. They used to spit (inside the chest), they*
> *said they did that to prevent having a child with albinism. I hit them then*
> *to stop what they were doing!*

Only one female participant raised concern over the myth that if an HIV-positive person sleeps with a person with oculocutaneous albinism, that person will be cured of HIV and AIDS:

> *...others like if you sleep with a person with albinism you will not get HIV.*
> *It is bad because if the myth spread, we are in trouble, because people can*
> *take you and abduct you!*

When asked further whether the myth that an HIV-positive person could be cured by sleeping with a person with oculocutaneous albinism was well known, the participant replied:

> *It was well known, but here, I have not heard much of it. It was well known*
> *Especially at X area. The one [myth] that we disappear is well known all over.*

Dell-Orto and Marinelli (1995:259) also raise concern that the negative aspects of disability stem from social myths and assumptions. In this regard, Fine and Asch's (1988:19-20) assumption is that people with oculocutaneous albinism become "victims" of various myths and superstitions.

Kromberg (1992:165) concludes that the 'death myth' must be exposed as pure unsubstantiated fictitious superstition, so that people with oculocutaneous albinism can be accepted as normal mortals with the same pattern of birth, life and death, as others of the species.

On the other hand, it seems fairly clear from the findings of this study that the community consider other beliefs about oculocutaneous albinism in a positive way. Some of the participants related how in other instances, the community believed that having a child with oculocutaneous albinism may be regarded as good luck in the family:

> *People have this thing that I am a child who brings luck in the family,*
> *so at home they also had that about luck…*

In this regard, Kromberg (1992:159) indicates that in Central Africa, specifically in Ghana, people with albinism are believed to be special, and under divine protection. Furthermore, according to Kromberg (1992:159), in Congo, children with albinism are given to the king to be educated as sorcerers.

It emerged clearly that the participants' families were aware of these myths and superstitions. The families were supportive, and encouraged the participants to educate the community so as to eradicate these myths and superstitions. The participants' families also appeared to be stigmatised by these myths and superstition, hence their intervention:

> *...this thing that we [people with oculocutaneous albinism] disappear, is an*
> *old belief. And I asked my granny the origin of this belief. She explained*
> *that when a person with albinism passed away, he or she was buried*
> *in the evening, and when people asked where that person was, the*
> *answer was that that person was taken to the hospital and never*
> *came back, when the family knew what happened. I could not*
> *understand that perception. But after my grandmother explained to*
> *me. That's why people even today ... and my grandpa told me that*
> *we must spread the word … to eradicate this belief, we the people*
> *with albinism!*

The findings of this study contradicted Gaigher et al's (2002:10) on myths. Only two of the participants in Gaigher et al's (2002:10) study indicated that they were aware of only one common myth that people with oculocutaneous albinism "do not die naturally as they disappear". However, Gaigher et al (2002:10) attribute the possible reluctance of the study participants to talk about other myths as a way of denying the myths for fear of being seen as superstitious or ignorant.

Myths and superstitions also emerged in the findings of a study by Pooe-Monyemore (2003:107) as barriers to integrate children with disabilities into the community. Pooe-Monyemore (2003) found that

♥ child with a disability is a family affair, and the society should not know about it ("*segole sa ka*" concept in a protective way)

♥ the child should be hidden from the rest of the community for fear of:

  ➢ stigma on the family

  ➢ community member's reluctance to marry into a family where there is a child with a disability.

There are several definitions of stigma. Burris (1999:1231) describes stigma as "the imposition of a special, discrediting and unwanted mark on a person, or a specific category of persons in such a way that, in their interactions with others, they are looked at as fundamentally and 'shamefully different' by themselves and others". Save the Children Research Report (2001:29) indicates that discrimination is one of the key manifestations of stigma. Moreover, stigma and discrimination have an insidious impact on the dignity and self-respect of the person being stigmatised. This, in turn, impacts on that person's self-image and self-confidence.

Another significant finding that emerged was the participants' interpretation of oculocutaneous albinism as a disability. Various contradicting interpretations emerged, which also had an impact in terms of how society perceived a disability. It did seem that the interpretation was mostly influenced by the participants' personal experience, possible poor eyesight, the need for a disability grant, and fear of being stigmatised. The participants who supported the idea that oculocutaneous albinism was a disability shared their experience as follows:

...and they (community) don't want to accept that if you
are visually impaired, and you fall under people with disabilities
…for I think you will not get a disability grant if you don't have
*disability…so all of us (*people with oculocutaneous albinism*) we are*
visually impaired, but our vision (eye problems) differs…there are those
who are worse ...and there are those who are better…but then we
need to make it clear that we have this …because somewhere
along the line it's going to give us problems because now, I
managed to get this learnership with that understanding that
I do have a problem. So if you don't give people a clear
picture, they wouldn't understand what's going on…

Its okay, you see, but what I realised is sometimes you
lose a job because of your disability. So like in my case I
almost lost a job, cause people they did not know my condition,
especially my eyesight, so they wanted me to do something
…but after finding out that I will not be able to see well the
job I was expected to do

Other participants felt strongly that oculocutaneous albinism was not a disability. In this regard, responding to what information on oculocutaneous albinism is important for the community, one of the participants emphasised:

Hmmm… that we are also human beings. This condition of ours
[oculocutaneous albinism] is not a disability…you see that one.
That we are disabled …No I don't support so much!… they must
know that we are not disabled. We are human beings you see,
it's a condition… a genetic thing…that's why we are so….if
they can know that …people will be sharp ...

With regard to the role of cultural beliefs and unscientific perception of disability, most beliefs are negative. For instance, in an area of KwaZulu-Natal, South Africa, Philpott (1995:82) explored the needs and rights of people with disabilities. Philpott's (1995:82) significant views on the causes of disability are linked to curse or bewitchment, the technique of placing harmful medicines, black magic and not observing customs.

Superstitious beliefs are usually associated with visits to traditional healers. However, in this study none of the participants indicated that they ever visited a traditional healer. In their study, Gaigher et al (2002:7) found that only one child indicated that he had consulted a traditional healer once.

Salif Keita's father, however, consulted a sangoma a week after Salif's birth. On the advice of the sangoma, Salif and his mother had to immediately leave his birthplace of Djoliba near Mali in West Africa, to settle in France. The sangoma predicted a destiny outside the community of Djoliba for "the little boy with albinism". Salif Keita and his mother had to leave West Africa, because the sangoma indicated that the birth of a baby with a white skin was regarded as a curse in West Africa. However, Salif got support from his parents, and his self-worthiness was enhanced by the family and friends, and this led him to become an international singing star (Danna 1998:100).

Due to the sensitivity of some myths and superstitions shared by the participants, both the participant and the researcher were in a vulnerable position. In addition the researcher asked probing questions to elicit the necessary emotional information. The researcher nevertheless remained sensitive to and aware of the possibilities of ethical issues arising that might not have been anticipated, as in the case of the myths and superstitions. The researcher therefore employed the following measures during the interviews with the participants:

♦ The researcher noted that failure to provide an opportunity for the participants to talk about these myths and superstitions might be perceived as irresponsible, and thus she allowed them to express their emotions.

♦ After each interview the researcher asked the participants if there was need for a follow-up.

♦ The researcher also provided contact details for additional help.

♦ At the end of the interviews, a debriefing session was allowed

♦ In other instances the participants were referred to the Albinism Society of South Africa for further counselling.

♦ Most importantly, all the participants agreed to be contacted to verify the findings, and to ensure that the information was correctly stated (Medical Research Council 2002:27; Polit & Beck 2004:143; Streubert & Carpenter 1999:57).

It emerged that the spiritual dimension also played a fundamental role in the lives of the participants.

3.3.2.2 Experience within the spiritual dimension

Experience of people with oculocutaneous albinism within the spiritual dimension is reflected within the dimension of Neuman's systems model (Neuman 1995:100).

Two sub-categories could be identified in the participants' experience within the spiritual dimension, namely the participants' strong religious background, and the role of the church in their lives.

- ***Influence of the strong religious background***

The biographical characteristics revealed that the majority of the participants, that is, fourteen out of the fifteen, had a strong religious background. Of these, eleven indicated that they attended church service often, whilst three indicated that they attended church service seldom due to other pressing commitments. According to one participant:

> *Ah ...at church you know that…you would not see those things (stigma) in church because there is that certain way that people have to behave in a certain way...all of you are together there… so such things, it's unlikely that you will see …a person may behave in another way outside, but in church he/she will hide it...but I have never experienced anything. I used to be active in church, around 1989 and 1990s...I think I was around 12-13 years…and even there I didn't have a problem*

The participant who was not religiously affiliated indicated that there was still a need "to do some research" to decide which religious group to belong to. Therefore it appeared that the participant was not necessarily against religious beliefs:

> *I used to attend church when I was young…but not any longer…but my mother does attend church… I think for me not to attend church is personal, and I still must do research to decide.*

The participants' strong religious background concurred with a report in one of the African countries (Road map…1999:47). In an effort to involve the African churches on disability issues, personal testimony was given by people with disabilities during an International Church Forum in 1999, in Kinshasa, capital of Democratic Republic of Congo in Africa. At this international forum, the participants shared the sentiment that people with disabilities were more apt to lead a deeply spiritual life because they were often despised, or were always poor (Road map…1999:47).

In this study, the participants' strong religious beliefs could also be attributed to the strong family support that emerged from the findings. Mweshi and Mpofu (2001:30) stress the probable influence of the parents on the strong religious beliefs. Mweshi and Mpofu (2001:30) therefore found that there appeared to be a strong religious component to the belief system of many of the parents and caregivers of children with cerebral palsy who were interviewed.

- ***Role of the church***

The participants regarded the church as important in providing counselling. It is therefore essential that the clergy have appropriate counselling skills. It does seem in this study the church played a role in improving the majority of the participants' self-esteem:

> *I talked to my pastor in November last year (2005)…because I was not attending church anymore because then I had to "confess" …so he (pastor) wanted to know from my side why I stayed at home and stuff like that…and then we talked ...*

> *My reasons not to attend church…I don't have to …for me, I felt that the people don't like me, and why do I have to go to church…God is not going to give me a job…and stuff like that….I was very confused … but now I attend church since I talked to the pastor's wife.*

Dell-Orto and Marinelli (1995:628-631) identify the role of religion in the lives of people with oculocutaneous albinism.

Dell-Orto and Marinelli (1995:631) raise concern that religious professionals have been, and still are a widely untapped resource for assisting people with disabilities. In this regard, Dell-Orto and Marinelli (1995:631) emphasise that due to the philosophy of love and harmony in the church, a religious counsellor may be the only source of hope for a person with a disability, and the only professional trusted by both the individual and the family.

The participants also identified the need for development and growth, which was one of the main themes of the research findings.

3.3.3   Expression of need for development and growth

It emerged that the participants identified the need to adjust in the community. According to Dell-Orto and Marinelli (1995:601), the exact mechanism to which age affects adjustment of a disability either positively or negatively has not been established. Oculocutaneous albinism is a congenital disorder present at birth, and the participants' ages in this study ranged between 18 and 48 years.

Furthermore, Dell-Orto and Marinelli (1995:602) emphasise that the impact of disability on self-concept is related to environmental and social consequence, and can influence adaptation of the person in the environment. Therefore, the main theme of expression of need for development and growth was categorised into two categories according to Neuman's systems model, namely experience within the developmental dimension, and experience within the physiological dimension.

3.3.3.1   Experience within the developmental dimension

It emerged from the findings that the family, various government departments, private sectors, and support groups are of significance for the developmental process of the participants. Neuman refers to the developmental dimension as the processes related to development over the lifespan (Neuman 1995:110).

The developmental dimension was therefore closely linked to a growth and adaptation phenomenon. Levine defines adaptation as a life process by which, over time, people maintain their integrity as they respond to environmental challenges (Levine 1991:196). Unlike any other animal, the human being experiences a period of development and growth that is extraordinarily long (Jordaan & Jordaan 1998:89).

Suin (1979:117) maintains that the parents determine the type of environment in which the child will grow up: whether it will be one of harshness, rejection, warmth, acceptance, security or uncertainty.

The participants noted the important role of the family in their developmental stages. However, it also emerged from the findings that the role of various government departments in the participants' lives should not be underestimated.

- ***Role of various Departments***

The participants expressed their opinions on how various departments played a significant role in their development. The most important departments mentioned by the participants were the Departments of Health and Education.

♦ ***Department of Health***

The study found that the Department of Health played a major role, especially in terms of skin care treatments, providing counselling to people with oculocutaneous albinism and their families, and raising awareness of oculocutaneous albinism. Nevertheless, the participants differed in terms of the availability of information on oculocutaneous albinism in the Department. Some participants felt that the Department of Health was doing well to raise awareness of oculocutaneous albinism, whilst others felt that the information on oculocutaneous albinism was not enough:

> *I think…the main thing is…the hospitals and clinics like, well, at Z Clinic*
> *they have a day, Wednesday, is a day for people with albinism and give*

them counselling. And these are some aspects of what the Government is
doing, but when it comes to skin treatment … the most important thing is
skin treatment…

Gaigher et al (2002:8) also note the role of the Department of Health, and emphasise that specialised, accessible and people-friendly health care is necessary for the care of people with oculocutaneous albinism. Children with oculocutaneous albinism complained about the unfriendly and sometimes rude treatment they received from health officials (Gaigher et al 2002:8).

One of the participants felt prejudiced and shared her experience as follows when she was in labour (giving birth):

There was only one nurse …and she had a funny attitude: "Get in here",
Shouting. She did not treat me well… and I asked her why she was only
shouting at me … and she was later okay…

♦ ***Department of Education***

It emerged that teachers at school should be knowledgeable on basic issues on oculocutaneous albinism, and this information should be emphasised during their training:

When I was engaged with awareness, I wrote to a teachers college of
education to do an awareness… I think the college of educators
should be informed because not all of us go to special schools…
and I actually encourage people not to attend special schools. Rather
they must attend mainstream schools…teachers must be educated
how to treat such as person…and this must start during their training…

The important role of teachers and education is discussed in section 3.3.2.1 on the participants' experience within the socio-cultural dimension**,** and specifically the role of schools in enhancing a sense of belonging.

♦ *Department of Labour*

The Department of Labour's role was noted to help with employment for people with oculocutaneous albinism and learnership programmes. For example, three of the study participants were on practical learnership programmes.

Urbani (1992:35) maintains that through our bodies we actively establish our own world, which also applies to our occupational world. The findings of this study revealed that the participants who were unemployed, suspected that because of their highly visible skin colour, they were not given a fair chance to be employed:

> *...mostly they [people with oculocutaneous albinism] are*
> *unemployed… and so if …that's why I say people with*
> *albinism are not necessarily treated as normal people in all*
> *instances…it's hard for them to be employed …*

In this regard, Jordaan and Jordaan (1998:694) cite Retief and Cole (1995), who believe that throughout history, work or employment has boiled down to what people do to meet the demands of the environment and their circumstances.

♦ *Other Departments*

The participants also noted the role of other departments, including the Department of Social Development to grant disability grants to people with oculocutaneous albinism who qualify for a grant. The Department of Science and Technology also play a role to advance use of technology for people with oculocutaneous albinism.

♦ *Role of the media*

Almost all the participants highlighted the significant role of the media. However, most of the participants raised concern that the media did not do enough to raise awareness of oculocutaneous albinism:

> *Awareness on radio [*on oculocutaneous albinism*] is not raised enough*
> *in the media. That's why I say we must make them aware. Like*
> *there are commercials, which, if they can be made for people with*
> *albinism, regularly as others are made, so it will be important. So awareness*
> *is not raised as it should be. That's why most people don't understand*
> *what is albinism, and how to treat a child with albinism.*

However, only one participant had a slightly different opinion, and felt that the media was doing enough to raise awareness of oculocutaneous albinism, although it could do better. The participant therefore suggested that maybe there should be sensation on the topic on oculocutaneous albinism to make the story "attractive" to the media:

> *The media is doing enough…but you know what media does …you need*
> *a story that will sell, a story that will bring attention…I think media can*
> *do a little bit in raising awareness ...and then after that you need a more*
> *intense educational strategy or a more in-depth counselling strategy*
> *…media needs sensation…but someone needs to follow up...*

Other participants excitedly shared their experience of participating in the media programmes. It emerged clearly from the discussions with those participants who participated in the media, that their self-esteem was much higher, and they seemed to be more mature because of their exposure:

> *I appeared on Take 5, advising youth on learnership. Nna ke fihleletse [*meaning I have survived the ordeal*] but it was tough.*

> *Oh! I used to participate on TV (*television*). The first article of the Soul Buddys*
> *I did on TV … and there was an overseas TV documentary. They were*
> *doing a documentary on oculocutaneous albinism, and the support from ASSA*
> (Albinism Society of South Africa). *We were all involved. They wanted more*
> *information on oculocutaneous albinism. It's like research,*
> *that was in 2003 or 2004...for us it was awareness*

♦ *Role of the private sector*

Only one participant mentioned the role of the private sector, and felt that the government was doing enough to raise awareness on oculocutaneous albinism, but the private sector was not:

> *I think you are doing enough as government, they do have that knowledge, and have access to information. My concern is with the private sector. They don't have that information. I don't know whether they don't have access to information. For example, banks and private companies and so on and so on …I don't know whether it's because they don't want to know or they can't access information.*

♦ *Role of support groups*

It was evident from the findings that the participants valued the power and security of support groups for their own development and growth. Most of the participants belonged to the Albinism Society of South Africa. It emerged that support groups, especially the Albinism Society of South Africa (ASSA), played an important role in the participants' lives.

The participants appreciated how the Albinism Society of South Africa enhanced their self-esteem by providing counselling, raising awareness of oculocutaneous albinism, encouraging them to recognise their rights, and providing them with other support services. However, some participants also felt that Albinism Society of South Africa (ASSA) should be more active in other areas:

> *ASSA helped me to socialise with other people with albinism …but I think there is a need to have ASSA in other areas…because I used to be isolated …but now at ASSA I could relate, and say OK, we used to share experience*

> *I wonder... I think support groups they are ... if they run, they can come a long way...it's only that people become impatient …and I think most people who come to support groups its like "What am I going to benefit… What am I going to benefit?" in tangible things, like I will get out of that with R5 or when I go there I will get sunscreen…that will be nice as well*

> *maybe if we link with Department of Health and we get sunscreens supplied*
> *... but I think it is beyond sunscreen...*

Gaigher et al (2002:10) found that none of the participants in their study belonged to a support group, which might have accounted for their low self-esteem.

The role of the Albinism Society of South Africa (ASSA) confirms that a collection of people can only be considered a group if it displays certain attributes (Jordaan & Jordaan 1998:719).

According to the Chairperson of ASSA, the Society aims at enhancing the self-esteem of people with oculocutaneous albinism, and enables them to be respected members of the community. The Chairperson further explains that establishment of ASSA in 1992, was initiated by the community's rejection of people with oculocutaneous albinism (Ngakane 1998:13).

The findings discussed under the psychological dimension also confirm how ASSA played an important role in the participants' developmental stages.

3.3.3.2   Experience in the physiological dimension

The findings revealed that the physiological dimension was one of the important dimensions that should not be overlooked, as it was significant in the participants' self-esteem. Neuman refers to the physiological dimension as the structure and function of the body (Neuman 1995:185).

People with oculocutaneous albinism have highly visible light skin, which is susceptible to skin cancer, as well as visual problems (Winship 2003:175). The physiological dimension in the findings of this study, therefore, covered the participants' physical and health issues, especially with regard to skin and eye care.

All the participants were aware of the importance of their skin and eye care, and also of their vulnerability to skin cancer. It therefore emerged that the participants were aware of the importance of their appearance and body image:

...like when I was still young ...you must make sure that you are
not exposed to the sun…you must have creams for your skin
and then you must …I mean that care…you must understand
that it is more important than for the other children

At home they are aware that I have eye problems since I was
young…I have visited several doctors and they said the
spectacles don't make much of a difference in my eyes…even
when I read a book I bring it a bit closer…

However, the findings on maintaining good skin and eye care by the study participants contradicted Gaigher et al's (1992:7) findings. The majority of Gaigher et al's (1992:7) participants considered their skin and eye problems as "normal" conditions that did not necessitate special care. It could be assumed that the attitude on the care of the skin and eye in Gaigher et al's study (1992:7), might have been influenced by the age difference. In Gaigher et al's study the participants were 5 to 18 years old, whilst in this study, the participants were 18 to 48 years old.

Kromberg (1992:160) and Luande et al (1997:734) indicate that as result of skin cancer, oculocutaneous albinism is associated with early death. According to Kromberg (1992:160) and Luande et al (1997:734), early death was especially noted in African countries like Tanzania and Nigeria where only 10% of people with oculocutaneous albinism survived beyond 30 years of age.

In this study, it was evident that the participants were alert to the vulnerability of their skin to cancer, and their parents were also supportive in this regard. Accordingly, then, the survival age of people with oculocutaneous albinism in South Africa is higher than in other African countries, which might be attributed to various factors, including the spiritual dimension of the individual. However, this is an area that needs further research.

3.4 RESEACHER'S EXPERIENCE OF THE FIELDWORK

The researcher is a trained genetic nurse, and has extensive experience in congenital disorders and oculocutaneous albinism. She has been in the field of Human Genetics for seventeen years. This study therefore required that the researcher bracket out her prejudices, values and beliefs.

As a genetics co-ordinator, the researcher established rapport with the participants. The researcher is also a committee member of the Albinism Society of South Africa's Task Team that co-ordinates a project of essay writing on oculocutaneous albinism by the youth. The participants expressed delight to participate in the research, and most importantly, the fact that they could access the information on completion of the study.

The participants seemed relaxed and willing to share their life-experience with the researcher. Rich quality data was therefore obtained. Fifteen interviews were conducted over a period of three months between January and March, 2006. Thirteen of the fifteen interviews were conducted at the respective homes of the participants.

The researcher is not good at directions, and got lost in some instances. At times the researcher felt vulnerable, driving alone in an unfamiliar area, and sometimes late at night. Some of the working participants only agreed to be interviewed after hours at home. Nevertheless, the experience in the field was interesting to note the relationship of the participants with other family members, colleagues at work, and most importantly to determine the participants' adjustment in various settings.

The researcher kept field notes, and the personal notes were the researcher's comments on own feelings, reactions, reflections and experiences as a genetics co-ordinator. The researcher endeavoured not to have ethical and conflicting implications during data collection and analysis. The field notes were integrated into the themes and discussion of the research findings.

3.5 CONCLUSION

This chapter discussed the research findings and an extensive literature control was done. Most importantly, the findings reflected the participants' experience on oculocutaneous albinism, factual information, perceptions, ideas and the participants' feelings. Three main themes emerged from the findings, namely, perception of the self, experience with the external environment, and expression of need for development and growth.

It emerged that self-concept was an important aspect in the participants' life-world, and that the external environment played a fundamental role in enhancing their self-concept.

Chapter 4 describes the development of the model for enhancement of self-concept of people with oculocutaneous albinism, using the key concepts deducted from the research findings.

CHAPTER 4

DEVELOPMENT OF A MODEL FOR ENHANCEMENT OF SELF-CONCEPT OF PEOPLE WITH ALBINISM

4.1   INTRODUCTION

Chapter 3 discussed the research findings and literature control was done. Chapter 4 describes the development of a model to enhance the self-concept of people with oculocutaneous albinism. The key concepts deducted from the research findings were **"self-concept"** and **"enhancement"**. Accordingly, this chapter discusses the concept analysis of these key concepts.

Concept analysis of "self-concept" and "enhancement" led the researcher to develop a theoretical definition of **"enhancement of self-concept",** which became the core building block for the model**.** In this regard, Walker and Avant (1995:23) emphasise that clear definitions, careful observation and thinking are the best tools for a potential theory builder.

Various reference sources were used to develop the model (Chinn & Kramer 1999:58-60; Fain 2004:69; McKenna 1997:57; Rodgers & Knafl 2000:81-83; Walker & Avant 1995:37-35)**.** The researcher also used Dickoff et al's (1968:422) survey list to classify and put the concepts in relation to recipients, agents, dynamics, procedure and terminus to facilitate future use of the model.

4.2 CONCEPT ANALYSIS

Concept analysis is the core activity to develop a theory (Chinn & Kramer 1995:78; McKenna 1997:57). According to Walker and Avant (1995:37), concept analysis is "a strategy to examine the attributes of a concept". Concept analysis is therefore essential to create a conceptual meaning of a concept in practice, research or theory (McKenna 1997:57). In this regard, Chinn and Kramer (1995:212) describe creating a conceptual meaning as "a theory development process of identifying, examining, and clarifying the mental images that comprise the elements, variables or concepts within the theory".

The researcher specifically used Wilson's (1963) steps cited and modified by Walker and Avant (1995:39) to undertake concept analysis of "self-concept" and "enhancement":

- Select a concept (concept identification)
- Determine the aims or purposes of the concept analysis
- Identify uses of the concept
- Determine the defining attributes
- Construct model and borderline cases
- Identify antecedents and consequences
- Define empirical referents.

4.2.1 Concept identification

Selecting a concept is the first step of concept analysis, and entails identifying a concept that communicates the idea that the researcher wishes to convey in the model (Chinn & Kramer 1995:81). A concept is a key element of concept analysis, and this sub-section therefore first defines the term "concept", describes how "self-concept" and "enhancement" were deducted in the present study, and identifies the significance of these concepts.

4.2.1.1 Definition of "concept"

Concepts are fundamental for developing a relevant model or theory, and knowledge in a particular discipline. Chinn and Kramer (1995:58) and Fain (2004:69) describe concepts as building blocks of a theory.

To understand the definition of concepts further, Rodgers and Knafl (2000:9) cite Becker (1984), Jacox (1974), and Walker and Avant 1995) who define concepts as

- Attributes, dimensions or aspects of reality which interest the scientist;
- Words that describe objects, properties, events and relations among them;
- Labels, categories or selected properties of objects to be studied; and
- Social in nature and arise from personal interaction.

For the purpose of this study, the key concept, namely enhancement of self-concept was the main building block of the proposed model, and emerged from the participants' experience. Fain (2004:69) is of the opinion that concepts are defined and understood within a particular theory.

4.2.1.2 Selection of "self-concept" and "enhancement"

Self-concept and enhancement were the main concepts deducted from the research findings. It became evident that "self-concept" and "enhancement" represented the participants' life-experience. Avant and Walker (1995:40) suggest that the concept selected should be important to further the theoretical development.

Three main themes emerged from the findings, namely the participants' perception of the self; experience with the external environment, and expression of need for development and growth.

During the process of interacting with the findings, it emerged that the participants' viewed "self-concept" in their life-world. Moreover, the external environment, which comprises the family, community, and health professionals, especially the nurses, played a fundamental role to "enhance" the participants' self-concept.

4.2.1.3 Significance of "self-concept" and "enhancement"

Rodgers and Knafl (2000:81) describe the significance of a concept as "the ability of the concept to resolve problems, to characterise phenomenon adequately, and to improve knowledge on that phenomenon". Rodgers and Knafl (2000:31) add that when selecting a concept, the researcher should begin by identifying the nature of the problem to be addressed by the research.

The major research problem was with regard to people with oculocutaneous albinism being stigmatised and discriminated (see chapter 1). Discrimination is perpetuated by misconceptions, myths and superstitions. In turn, discrimination has an insidious impact on the dignity, self-image and self-confidence of person being stigmatised.

It emerged from the findings that there was a need to enhance the participants' self-concept or self-image, so that they "themselves" are empowered to address the misconception, myths and superstitions in the community. **"Enhancement of self-concept"** therefore became the key concept of the model to address the research problem.

4.2.2 Determine the aims or purposes of the concept analysis

Chinn and Kramer (1995:81) are of the opinion that clarifying the purpose of the concept analysis provides a sense of direction. Concept analysis was undertaken to develop the theoretical and operational definitions of the concepts. Rodgers and Knafl (2000:16) and Walker and Avant (1995:25 and 46) emphasise that theoretical and operational definitions of concepts are critical for theory building.

Theoretical definitions were the means by which the researcher introduced essential attributes of "self-concept" and "enhancement", and used other theoretical terms to define these concepts. Operational definitions reflected the theoretical attributes, and provided the means to classify "enhancement of self-concept" as the key concept of the model. Concept analysis was therefore essential to promote communication and clarify "enhancement of self-concept" (Walker & Avant 1995:25).

4.2.3 Identify uses of the concept

Rodgers and Knafl (2000:82) refer to "use" of the concept as the common manner of employing the concept, and the situations appropriate for its use. It was therefore essential to understand the use of "self-concept" and "enhancement' from valid knowledge and sources (Rogers & Knaff 2000:232).

The initial valid use of these concepts emerged from the interviews with the participants. The researcher then used dictionaries and thesaurus to define and provide synonyms for the concepts. Existing theories and professional and classical literature were also used to identify as many uses of the concepts as possible, and to extend the definitions beyond the limits of common linguistic usage of the concepts (Chinn & Kramer 1999:64; Walker & Avant 1995:40).

Identifying the use of "self-concept" and "enhancement" involved examining these concepts.

4.2.3.1 Examination of "self-concept"

In the process of examining "self-concept", the researcher first explored in detail the definition of the term "self" and then "self-concept".

- ***Definition of the term "self"***

In this study, the concept "self" initially originated from the interviews as an experience echoed by the participants. The participants felt that they 'themselves' played an important role in their own lives to enhance their self-concept so as to address misconceptions, and myths and superstitions on oculocutaneous albinism.

Collins English Dictionary (2006:745) defines "self" as "distinct individuality or identity of a person or thing; a person's typical bodily make-up or personal characteristics; or an individual's consciousness of his or her own identity or being".

Oxford Advanced Learners Dictionary of Current English (2004:1067) refers to "self" as a noun, and defines "self" as "type of person one is, especially the way one normally behaves, looks or feels; the self, which is one's personality or character that makes one different from other people; the inner self, which refers to the one's emotional and spiritual character".

Blackwell's Nursing Dictionary (2005:545) defines "self" similarly, and adds "total essence or being of a person or individuality; those affective, cognitive and spiritual qualities that distinguish one person from another; a person's awareness of his or her own being or identity; and a person's ego or consciousness".

Jordaan and Jordaan (1998:616) state that much of the psychological development of a person is bound up with the emerging sense of the self. Jordaan and Jordaan (1998:616) point out that in everyday speech, the word "self" is linked to various nouns that express the notion that there are things that belong to the individual's essential

nature; that there is an inner self through which an individual experiences the self and the world.

In spite of its importance, the phenomenon of the "self" is one of the most difficult to explain, because it is also formed by a generalised view of other people's perception (Dell-Orto & Marinelli 1995:660; Jordaan & Jordaan 1998:617). Jordaan and Jordaan (1998:617) argue further that the difficulty of explaining the "self-concept" might be partly because of the many applications of the "self" in an individual's personal and social life.

- ***Definition of "self-concept"***

Blackwell's Nursing Dictionary (2005:545) defines "self-concept" as "the mental image one has, based on one's ideas and attitudes about one-self and one's personality".

Collins English Dictionary (2006:746) defines self-image as "one's own idea of oneself or sense of one's worth". *Blackwell's Nursing Dictionary* (2005:545) defines "self-image" as "the total concept, idea, or mental image one has of oneself and of one's role in society".

Oxford Advanced Learners Dictionary of Current English (2004:1068) expands the definition of self-image to include appearance and abilities, and therefore defines self-image as "an opinion or idea one has of oneself, especially one's appearance or abilities".

Jordaan and Jordaan (1998:616) emphasise self-evaluation as an important premise of the self-concept. On the basis of the self-evaluation, the person should form a self-concept or self-image regarding physical, psychological and social attributes as portrayed in Figure 4.1.

```
┌──────────┐  ┌──────────┐  ┌──────────┐
│ Physical │  │Psychological│ │ Social   │
│attributes│  │ attributes │ │attributes│
│          │  │          │  │          │
│          │  │          │  │          │
│          │  │    │     │  │          │
│          │  │    ▼     │  │          │
│       ╲  │  │  ╭────╮  │  ╱          │
│        ╲ │  │  │Self-│ │ ╱           │
└─────────╲┘  └──│concept│─╱───────────┘
           ╲     ╲      ╱ ╲
            ▼     ╲____╱   ▼
```
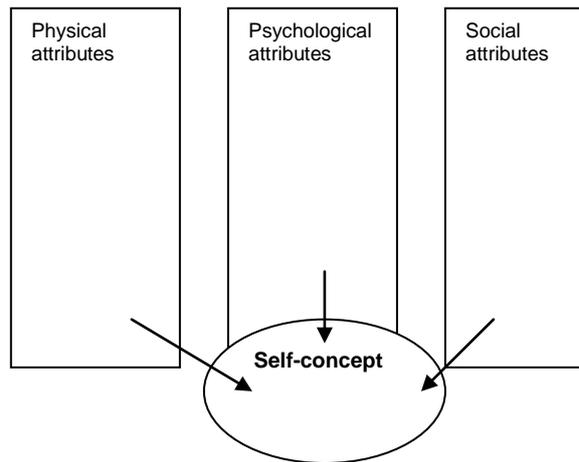
Figure 4.1

Attributes of self-concept

Source: Jordaan and Jordaan (1998:166)

The physical attributes relate to a person's appearance and body image. Psychological attributes are a person's talents, plans and needs. Social attributes are related to a person's tendency to be withdrawn or be submissive (Jordaan & Jordaan 1998:616).

Dell-Orto and Marinelli (1995:660) also refer to self-concept as "an image that an individual has of him or herself, and is formed by a conceptualisation process". However, Dell-Orto and Marinelli (1995:660) warn that in the process of forming the self-concept, inferiority and anxiety may create a devaluing effect, and there may be at least three adverse responses to this.

The first adverse response is "mourning", which refers to sadness, and results from the lack of functional skills; the second is "devaluation" when people see themselves as more handicapped than they really are; and the third is "spreading", which refers to those people who see their disability expanding from the original source to the whole body. All or any one of these responses may lead to an underestimation of oneself, and the self-concept formed is therefore not appropriate to the "real self" (Dell-Orto & Marinelli 1995:661).

In line with Jordaan and Jordaan (1998:616) (see figure 4.1), Dell-Orto and Marinelli (1995:660) explain that because the physical component is such an important factor in an individual's development, it could be argued that physical disability probably affects the formation of the self-concept.

Roy and others support the importance of the physical appearance to form the self-concept. In her adaptation model, Roy identifies the self-concept mode as one of the categories for assessment of behavior. According to Roy, the self-concept mode relates to the basic need for psychic integrity, and focuses on the psychological and spiritual aspects of the self. Furthermore, the self-concept mode is comprised of the *physical self* and the *personal self* (Roy 1991:196) (see figure 4.2). Figure 4.2 represents Roy's model diagrammatically.
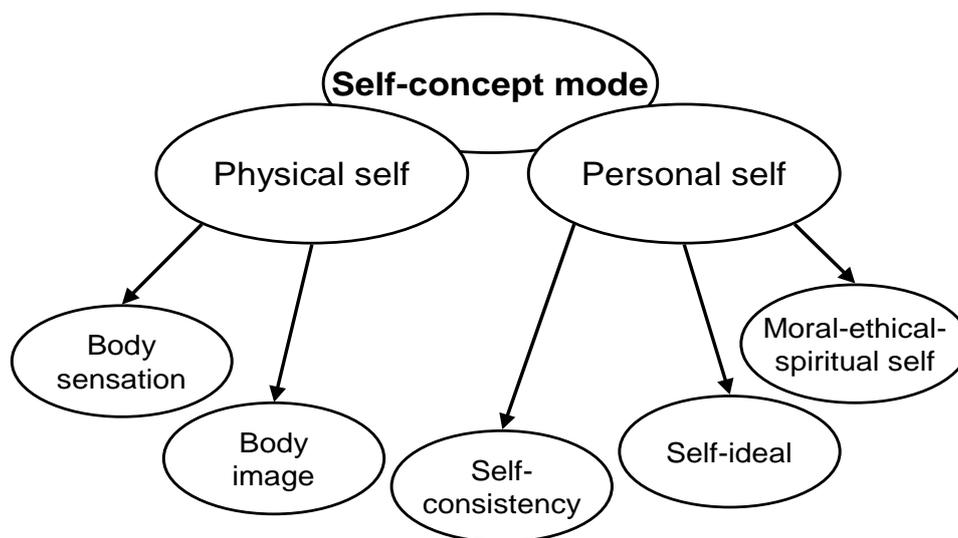
Figure 4.2
Roy's self-concept model
Source: Roy (1991)

According to Roy, the physical self has the components of body sensation and body image, and the personal self has the components of self-consistency, self-ideal, and moral-ethical-spiritual self. Body sensation is how the person *experiences* the physical self, and body image is how the person *views* the physical self. Self-consistency represents the person's efforts to maintain self-organisation and to avoid disequilibrium.

Self-ideal represents what the person expects to be and do, and the moral-ethical-spiritual self represents the person's belief system and self-evaluator **(Roy 1991:196)**.

Closely linked to the self-consistency described by Roy (maintain self-organisation and avoiding disequilibrium), is Levine's theory of integrity. In her theory, Levine discusses adaptation, conservation and integrity. Levine goes on to explain that adaptation is the life process by which conservation is achieved, and a process by which, over time, people maintain their wholeness or integrity. Levine refers to integrity as "being in control of one's life" (Levine 1991:151). Furthermore, Levine distinguishes between personal integrity, social integrity and structural integrity as a basic understanding of the self.

Levine explains that *personal integrity* focuses on a sense of self, and the "self" is defined, defended, and described only by the soul that owns it. *Social integrity* involves a definition of self that goes beyond the individual. In this regard, individuals use their relationships to define themselves. Levine emphasises that a person's identity is connected to the family, community, culture, ethnicity, religion, vocation, education, and socio-economic status. According to Levine, *structural integrity* focuses on the healing process, and supports the idea that a sense of structural integrity is more than an individual's physiological need (Levine1991:193).
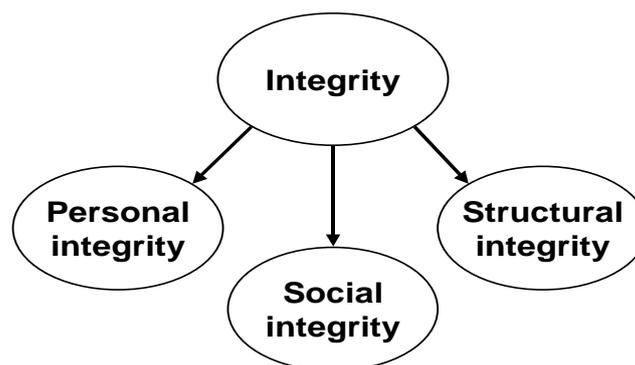
Figure: 4.3

Levine's description of integrity

Source: Levine (1991)

Collins English Dictionary (2006:415) defines integrity as "the quality of being whole or united, or the quality of being unharmed or sound".

Collins Thesaurus (2006:384) lists the following synonyms for "integrity": honesty, principle, honour, virtue, goodness, morality, purity, righteousness, probity, rectitude, truthfulness, trustworthiness, incorruptibility, uprightness, scrupulousness, reputability, cohesion, wholeness, soundness and completeness.

Stanhope and Lancaster (2004:139) cite Beauchamp and Childress (2001:36) and define integrity as "soundness, wholeness, and integration of moral character". From a research ethical point of view, Stanhope and Lancaster warn that people with compromised integrity are easy prey for potential scientific misconduct.

Clinebell (1992:178) cites Erik Erikson, a psychoanalyst, who describes ego (self) integrity as a growth process. According to Erikson (Clinebell 1992:178-179), ego integrity means

- Making peace with the passing years and with the inevitable frustration of some of one's dreams;
- Accepting one's finitude limitations; and
- Living and enjoying as much as possible the precious here-and-now moment.

Clinebell (1992:179) emphasises that the strength that results from ego integrity is wisdom. Furthermore, ego integrity is the cumulative result of growth throughout the life cycle, and an opposite of ego integrity is despair (Clinebell 1992:179).

A concept linked to integrity is self-determination. *Collins English Dictionary* (2006:746) defines self-determination as "the ability to make a decision for oneself without influence from outside". Philpott (1995:100) found that self-determination would not only benefit the person with a disability, but also benefit the community.

According to Dell-Orto and Marinelli (1995:660), social interaction, building of self-esteem and achievement of life goals are enhanced by physical ability, as well as emotional and intellectual resources.

The findings of the present study revealed the self-concept as it related to self-esteem, self-actualisation, and self-worthiness.

Self-esteem is a complementary feature of the self-concept associated with high performance, and is regarded as the most important and powerful values in the motivation system (*Oxford Textbook of Public Health* 2002:839).

Collins English Dictionary (2006:764) and *Oxford Dictionary of Nursing* (2004:435) define self-esteem as "respect for or favourable appreciation, and opinion of one-self". *Oxford Advanced Learners Dictionary of Current English* (2004:1068) refers to self-esteem as "feeling of being happy with one's character and abilities".

Collins Thesaurus (2006:638) and *Collins Paperback Thesaurus* (2004:650) list the following synonyms for "self-esteem": self-respect, confidence, courage, vanity, boldness, self-reliance, self-assurance, self-regard, self-possession, self-determination, faith in one self, pride in one self, and *armour-propre* (French).

Collins English Dictionary (2006:746) defines self-respect as "a feeling of confidence and pride in one's own abilities and worth". *Collins Thesaurus* (2006:638) and *Collins Paperback Thesaurus* (2004:650) list the following synonyms for "self-respect": pride, dignity, morale, one's own image and self-esteem.

DuBois (2003:938) discusses self-esteem from an educational point of view, and indicates that self-esteem is one of the most widely discussed markers of positive mental health for learners at school. DuBois (2003:945) points out further that in recent years, researchers have made significant progress in addressing questions about the basic nature of self-esteem, the factors that most influence its development during childhood, and the conditions under which high levels of self-esteem promote healthy overall adjustment at school. DuBois (2003:938) goes on to say that according to the Addictive Model of Self-Worth, the underpinnings of self-esteem are a sense of personal competence or efficacy, and a perception of being accepted and valued by others.

Oxford Advanced Learners Dictionary of Current English (2004:1068) defines self-worthiness as "a feeling of confidence in oneself that one is a good and useful person". *The World Book Dictionary* (1997:1891) defines self-worthiness as "a favourable estimate or opinion of oneself or self-esteem".

DuBois (2003:939) emphasises the importance of maintaining a sense of self-worthiness at an early stage of an individual's development. Supporting DuBois, Robinson and Noble (1995:64) suggest early socialisation practices of parents of gifted children to encourage early attainment of goals, high standards of excellence and self-direction of the learner.

According to Dell-Orto and Marinelli (1995:662), a sense of individuality and worth of people with disabilities is built by positive social awareness, legislation and technological advances.

Dell-Orto and Marinelli (1995:258) clarify that since social awareness influences self-acceptance, legislation is essential to ensure the rights of individuals with disabilities, and technological advances enhance the participation of people with disabilities in societal activities.

Oxford Advanced Learners Dictionary of Current English (2004:1067) defines self-awareness as "knowledge and understanding of one's own character". Self-knowledge therefore emerged as a related concept to self-awareness. In this regard, Stone-McCown and McCormick (1999:538-539) introduced the Self-Science Theory in the Education System for Grades 1 to 8 in London. The aims of the self-science theory were to legitimise self-knowledge as valuable subject matter, enhance self-esteem, and foster self-awareness and self-worthiness of learners.

Kubie (1978) (cited in Stone-McCown & McCormick 1999:539) emphasises the importance of self-knowledge: "What is the effective value of a knowledge of the external if we lack an equally deep personal insight? ... but self-knowledge in depth is a process, like education itself, and is never finished."

Self-awareness as it relates to self-concept is regarded as an important concept. Goleman (1997:43) refers to self-awareness as "the keystone to emotional intelligence". Goleman (1997:47) adds that self-awareness is a sense of ongoing attention to one's internal states, and it is a self-reflexive awareness the mind observes and investigates experience itself, including the emotions. Goleman (1997:48) is of the opinion that people who engage in self-awareness are sure of their own boundaries and, most importantly, they tend to have positive outlook on life.

Closely linked to self-awareness, is the self-actualisation concept. *The World Book Dictionary* (1997:1886) refers to self-actualisation as "self-realisation". *Collins English Dictionary* (2006:746) refers to self-realisation as "the fulfilment of one's own potential or abilities".

Oxford Dictionary of Nursing (2004:435) defines self-actualisation as "the tendency to realise and fulfil one's maximum potential". *Blackwell's Nursing Dictionary* (2005:545) defines self-actualisation as "the perceived competence of an autonomous system to satisfy the basic needs for homeostasis, safety, protection, feedback and exploration in due time".

Collins English Dictionary (2006:380) defines homeostasis as "the tendency of an organism to achieve a stable metabolic state by compensating automatically for violent changes in the environment and other disruptions".

Self-actualisation encompasses the social cognitive theory, self-efficacy and reciprocal determination (*Dictionary of Public Health* 1996:107). According to the *Dictionary of Public Health* (1996:107), social cognitive theory is "a general model of behaviour set forth by Bandura (1977,1982), emphasising effect of social environment and cognitive mediators such as beliefs on behaviour and the reciprocal effect of behaviour on environment". In the social cognitive theory, reciprocal determination is the continuous shared interaction, which emphasises that a person can shape the environment, and the environment can shape the world. However, the most prominent concept of social cognitive theory is self-efficacy (*Dictionary of Public Health* 1996:99).

Dictionary of Public Health (1996:105) refers to self-efficacy as "the internal state that a person experiences, such as competence or ability to perform a desired task".

Oxford Dictionary of Nursing (2004:435) defines self-efficacy as "an individual's estimate or personal judgement of his or her own ability to succeed in reaching a specific goal".

According to *Collins English Dictionary* (2006:581) "efficacy" is derived from the Latin "*efficere*" which means to achieve, and producing the intended result.

The Collins Thesaurus (2006:638) lists the following synonyms for "efficacy": Power, effectiveness, efficiency, value, success, strength, virtue, vigour, usefulness, potency, fruitfulness and productiveness.

According to Jordaan and Jordaan (1998:701), the need to achieve is associated with self-efficacy and internal locus of control. People with self-efficacy and internal locus of control believe that they can effect change where they feel change is needed, and that it is up to them to effect the change (Jordaan & Jordaan 1998:701).

Barrable (1995:11) describes the locus of control as "the point at which we place ourselves and cope with life". Barrable (1995:11-12) further explain the two poles of locus of control, namely, the inner perspective and outer perspective. At the pole of the inner perspective, an individual is the creator of circumstances, the person takes the responsibility of becoming part of an action, has freedom of choice, and the person also knows how to create, initiate, process and deal with circumstances. At the pole of the outer perspective, if a person believes things will happen to him or her over which he or she has no control, the person becomes a victim, and the person believes he or she is where he or she is because of blows, or blessings of life.

Driscoll (1994:23) refers to "self-efficacy" as the individual's evaluation of their performance capabilities related to a particular type of task. Furthermore, Driscoll (1994:23) suggests the following strategies to improve self-efficacy and motivation of learners at school:

- **Enhance relevance:** The educator should explain how instructions relates to learner's goals, and should build on learners previous experience;

- **Build confidence:** The educator should create positive expectations for success, provide opportunities for learners to successfully attain goals, and offer learners control over their learning; and

- **Generate satisfaction:** The educator should provide learners with opportunities to use newly acquired skills, and should provide them with positive feedback.

According to Abraham Maslow, well known for his hierarchy needs, self-actualisation is equivalent to optimal human functioning, and is actually people's striving to discover

and realise their full and true potential to reach their goals (George 2002:153-155). Maslow emphasises that the basic motive linking all the needs is the tendency towards self-actualisation.

Maslow goes on to emphasise the physiological needs, security needs, need for love (include need to belong in the sense of being accepted), need for esteem and appreciation, cognitive needs (need to know and understand) and aesthetic needs or need to appreciate beauty (Maslow 1970:109-111).

However, Levine, a nursing theorist, argues that Maslow discusses self-actualisation as reaching beyond, and not reaching into the person (Levine 1991:19). Levine therefore emphasises the 'inner self", and is of the opinion that self-actualisation should reach into the person. Levine goes on to explain that human beings have both a private and public self. The private self is unique and whole. A person can share fragments of the private life with others, and a separation between the self and other should be maintained (Levine 1991:19-25).

Self-acceptance as it relates to self-actualisation and self-worthiness emerged as another fundamental concept. Friedland (2005:64), in her classical book on "Self" discusses self-acceptance as an important prerequisite for taking excellent care of oneself. According to Friedland (2005:64), self-acceptance is a process that improves with effective psychotherapy and counselling. Friedland (2005:64) recommends three important statements to improve self-acceptance: "I am beautiful; I am valuable; and I am good enough"

In summary, the key elements of "self-concept" are influenced by the power of the "inner self" as one grows and develops in life, influenced by the environment to shape the "self", and most importantly to maintain the "self" as it relates to life and experienced by the person. It emerged from the various definitions and perspectives that "self" refers to the person's own identity, consciousness, appearance that makes the person different from other people, and own feelings of self-worthiness to boost the self-esteem and thus maintain self-efficacy and integrity. The "self-concept" is also influenced by the social attributes as it relates to the relationship of an individual with the family and the community.

4.2.3.2 Examination of the concept "enhancement"

The concept of "enhance" is invested in the notion of improving or advancing a particular phenomena. Enhancement is a noun derived from the verb "enhance".

Collins English Dictionary (2006:260) defines "enhance" as "to improve or increase in quality, value, or power".

Oxford Advanced Learners Dictionary of Current English (2004:385) defines "enhance" as "to increase or further improve the good quality, value or status of somebody or something".

Collins English Dictionary (2006:666) defines quality as "a degree or standard of excellence, or a distinguishing character or attribute, or formerly high social status".

Collins English Dictionary (2006:925) defines value as the desirability of something, often in terms of its usefulness exchangeability.

Collins English Dictionary (2006:810) defines status as "a person's position in society, or the esteem in which people hold a person, or the degree of importance".

Collins English Dictionary (2006:638) defines power as ability to do something, or a specific ability or faculty, or a prerogative or privilege.

Collins Thesaurus (2006:232) and *Collins Paperback Thesaurus* (2004:234) list the following synonyms for "enhancement": enrichment, strengthening, heightening, increment, embellishment, boost, betterment, augmentation, emelioration, elevation, intensify, magnify, reinforce, lift, exalt and complement.

Oxford Advanced Learners Dictionary of Current English (2004:384) defines "enrich" as to improve quality of something often by adding something to it. *Oxford Advanced Learners Dictionary of Current English* (2004:123) defines "boost" as "to make something increase, or become better or more successful". *Longman's Dictionary of Contemporary English* (1997:108) adds "to include encouragement, help a person to advance, to promote and praise".

Blackwell's Nursing Dictionary (2005:88) refers to a "booster dose" as "a later dose given to enhance the effect of an initial or previous dose, usually applied to antigens given for the purpose of producing specific antibodies".

Oxford Advanced Learners Dictionary of Current English (2004:63) refers to augment as to increase the amount, value or size of something. *Collins English Dictionary* (2006:44) defines augmentation as "a process to make or become greater in strength or number".

It is essential to understand enhancement in relation to the environment, and to consider Roger's principles of homeodynamics (Rogers 1992:28). Rogers grounds her theory of homeodynamics on three principles, namely integrality, resonancy and helicacy. According to Rogers, integrality is the continuous, mutual simultaneous interaction process between human and environmental fields. Resonancy then refers to the nature of the change occurring between human and environment. Helicacy deals with the nature and direction of change in the human-environment field. Rogers maintains that by using these three principles of homeodynamics, the nurse will help an individual in the environment to move to a higher, more diverse level of existence, which has direction to grow and develop (Rogers 1992:28-30).

Another relevant model to understand the concept "enhancement" is Johnson's behavioural system model. Johnson identifies the patients as the behavioural system, and three functional requirements of the system (Johnson 1980:207). The three functional requirements are: firstly, the system (or person with oculocutaneous albinism in this study) must be protected from noxious influences in the environment with which the system cannot cope; secondly, the system must be nurtured through the input of appropriate supplies from the environment, and finally, the system must be stimulated for use to enhance growth and prevent stagnation (Johnson 1974:373-374).

Rogers concurs with Johnson (1974:373) and emphasises that as long as the systems are meeting Johnson's functional requirements, the system is viewed as self-maintaining and self-perpetuating. However, Rogers warns that an imbalance in the behavioural system produces tension, which results in disequilibrium (Rogers 1992:29).

DuBois (2003:942) discusses enhancement from an educational point, and indicates that there is empirical evidence of effective intervention strategies for enhancing the self-esteem of learners. According to DuBois (2003:942), the intervention strategies to enhance self-esteem of learners are:

- Positive parent-child relationships;
- Stimulating home environment;
- Increased access of families to community-based resources; and
- Parent training and education.

DuBois (2003:942) emphasises that these intervention strategies not only benefit the young children, but also enhance the self-esteem of parents as caregivers to promote a positive sense of self in their young children.

Treffinger, Callahan and Vaughn (1995:37) from the University of Virginia in the United State of America (USA) reviewed several studies to describe models of enrichment in education. Treffinger et al (1995:37) discuss Renzulli's (1977) enrichment triad model, which had a major influence on the development and implementation of enrichment programmes in schools.

Renzulli's (1977) enrichment triad model involves three types of enrichment. *Type one enrichment* consists of general exploratory activities through which learners are exposed to a wide range of topics and issues that extend beyond the regular curriculum. *Type two enrichment* involves helping the learners to learn creative and critical thinking and problem-solving skills, as well as research skills. *Type three enrichment* consists of investigating real problems, in which the learners are involved in firsthand inquiry into problems and questions that are important and meaningful to them (Treffinger et al 1995:39).

Another relevant model in education is Feldhusen and Kollof's Purdue three stage enrichment model (Treffinger et al 1995:39). In line with the enrichment triad model described above, the Purdue three stage enrichment model is specifically designed to develop the learner's thinking skills, to stimulate application of those skills in problem solving, and to promote extension of those skills to independent projects (Treffinger et al 1995:39).

In the field of education in South Africa, Sonnekus (1996:16) suggests criteria to evaluate a particular enhancement programme. These criteria are meaning attribution, involvement, experience, self-actualisation and self-concept.

Sonnekus (1996:106) goes on to explain that *meaning attribution* influences the nature and quality of the person's relationship. *Involvement* is dependant on the meaning attribution and the quality of experience generated by the interaction between the participants in the relationship. *Experience* then reflects the unique reaction of an individual to specific situations (Sonnekus 1996:106-108). Sonnekus (1996:110) states that during the enhancement process, the dynamic quality of the self-concept makes it possible for the person to interact with and react to various environments, and in so doing the person can self-actualise himself or herself in different ways.

It is clear from the key elements of "enhancement" that the concept encourages improvement or advancement of something. During the enhancement or enrichment process, any imbalance in the "inner self" and the external environment may have an impact in the self-concept of the person, and thus cause disequilibrium. Therefore, it was fundamental to develop and describe the model for enhancement of the self-concept of people with oculocutaneous albinism, and use the main attributes of the main concepts for that process.

4.2.4   Determine the defining attributes

Attributes of concepts are regarded as fundamental to describe the characteristics of the concepts, and answer the question "What is this?" the researcher is discussing (Chinn & Kramer 1999:63). Rodgers and Knafl (2000:91) support Chinn and Kramer (1999:63), and explain that the attributes of concepts represent the primary accomplishment of concept analysis. McKenna (1997:63) adds that the attributes of a concept constitute a "real" definition, as opposed to nominal or dictionary definition that merely substitutes one synonymous expression for another.

The researcher identified clusters of the attributes most frequently associated with "self-concept".

4.2.4.1 Reduction process of attributes of "self-concept"

Clustering the attributes of "self-concept" was done to gain a broader insight into the concept, and to provide important information regarding contextual aspects of the concept (Chinn & Kramer 1995:98; Rodgers & Knafl 2000:95). Table 4.1 depicts the list of attributes the researcher clustered together for "self-concept".

Table 4.1 Attributes of "self-concept"

Ownership	Identity	Psychological
Possession	Inner self	Emotional character
Pride	Belong	Moral-ethical
Own opinion	Different from others	Spiritual
Worthy	Personal judgement	Behaviour
Personal attributes	Individuality	Abilities
Own mental image	Conception of who	Favourable appreciation
	Essence of being	
Awareness	Good enough	
Outlook on life	Faith in one self	
Knowledge	Integrity	**Physical being**
Understanding		Bodily make up
Self-evaluation		Appearance
Investigate		Different from others
Talents	**Ability**	
Plans	Actualise	
	Realisation	**Social being**
Adjustment	Maximum potential	Relationships
Competence	Motivation	Adjustment
Reach beyond	Self-determination	People's perception
Make decisions		Role in society
Accepted	**Self-worthiness**	Experience the world
Valued	Positive social awareness	
Reflexive	Legislation	
	Technological advances	
Balanced		
Fulfilment		
Absorption		
Control		
Consistency		
Avoid disequilibrium		
Relationships		

The researcher then clustered the essential attributes to identify related attributes of "self-concept. Table 4.2 represents the essential attributes and those perceived as related attributes of "self-concept".

Table 4.2 Essential and related attributes of "self-concept"

Essential attributes	Related attributes
Sense of ownership and identity	Possession Inner self Abilities and competence Appearance and character Faith in one-self
Adaptation	Feel worthy and valued Role in society Fulfilment Actualisation Appearance and character
Interdependence	Experience the world Relationships Feel valued and appreciated by others People's perception Belonging

4.2.4.2 Reduction process of attributes of "enhancement"

Clustering the attributes of "enhancement" was also done to provide important information regarding contextual aspects of the concept (Rodgers & Knafl 2000:95). Table 4.3 displays the attributes of the concept "enhancement".

Table 4.3 Attributes of the concept "enhancement"

Enrich	Positive environment	Having direction
Improve quality	Stimulating	Balanced
Advance	Awareness created	Avoid disequilibrium
Explore	Interaction	Prevent stagnation
Move higher	Role of family	Relationship
Make excellent	Role of community	Independence
Add something	Relationship independence	
Promote growth	Praise	**Power**
	Involve	Feel empowered
Strengthen		Maintain status
Encourage	**Skills**	Self-actualise
Boost	Improve critical thinking skills	Meaningful to person
Diverse	Improve problem solving skills	Sustain power
Quality of experience	Decision making	Positive sense of self
Increase		
	Increase value	**Improve status**
	Use capabilities	Position in society
	Feel self-worthy	Esteem elevation
	Cognitive abilities	Fulfil responsibilities
		Integrity

Table 4.4 represents the essential and related attributes of the concept "enhancement".

Table 4.4 Essential and related attributes of the concept "enhancement"

Essential attributes	Related attributes
To actualise potential	Positive sense of self Fulfil responsibilities Involved in decision making Promote growth
To enrich	Esteem elevation Boost what is already existing Stimulate Move higher
To improve quality	Improve skills: decision making, critical thinking and problem solving skills Add something to look better Balanced Create positive environment

4.2.4.3 Definition of related attributes of "enhancement of self-concept"

The essential and defining attributes central to enhancing the participants' self-concept are illustrated and summarised in Table 4.5 These attributes depict an advancing process of enhancing self-concept of a person with oculocutaneous albinism from a level of low self-image, to a high level of integrity of positive self-image. See section 4.3 for a theoretical definition of "enhancement of self-concept".

Table 4.5   Defining related attributes of enhancing self-concept of people with oculocutaneous albinism

---

Defining related attributes to enhance self-concept of people with oculocutaneous albinism

Step 1: Enhance self-awareness

- Enhance self-evaluation
  - physical attributes
  - psychological attributes
  - social attributes
- Enhance self-esteem
- Promote self-development and growth

Step 2: Support adaptation in the environment

- Involve in awareness creation
- Develop sense of belonging

Step 3: Maintain integrity

- **Self-efficacy**
- **Cognitive skills**
  - Decision making
  - Problem solving
  - Critical thinking

The next section constructs model and borderline cases.

4.2.5   Construction of model and borderline cases

The researcher constructed model and borderline cases to illuminate "enhancement of self-concept". McKenna (1997:64) is of the opinion that definitions from dictionaries and thesauri give the analyst an insight into the concept of interest. However, for abstract concepts such as those found in nursing, model and borderline cases are often valuable ways of gaining a deeper insight into the concept (McKenna 1997:64 and Walker & Avant 1995:42).

4.2.5.1 Model case

The constructed model case is a 'real-life' example of the key concept "enhancement of self-concept" of a person with oculocutaneous albinism, and includes all the critical attributes of the key concept (Walker & Avant 1995:42).

■ *Example of a model case*

A 23 year-old woman with oculocutaneous albinism visited the clinic for the first time one chilly Monday morning. She looked depressed, and complained that she could not make friends, and felt ridiculed and stigmatised at home and in the community. The young woman sobbed as she related that she had decided to seek counselling from the clinic nurse on this particular Monday morning. She elaborated that she thought the negative attitudes towards her were "because she has oculocutaneous albinism".

The young woman indicated further that she was the only one in the family with oculocutaneous albinism. She had two brothers and one sister who were all employed, and she was the only one unemployed. Both her parents did not have oculocutaneous albinism. However, she had learned from "neighbours" that her paternal great-grand mother, who had died five years ago, had also had oculocutaneous albinism. She thought her family never talked about albinism, as if they were "embarrassed to have her as part of the family".

When the nurse asked her whether she had a boyfriend, she replied sadly, "No. Even previously, I never really had a boyfriend. It's only a friend and it's only kissing around, stuff like that. I never had a boyfriend and I need someone to talk to. But it never happened!"

During the conversation with the young woman, the nurse realised that she had a low self-image. The nurse therefore identified that there was a need to enhance the young woman's self-concept. The advancing process of enhancing self-concept thus entailed firstly, the nurse as an agent enhancing self-awareness of the young girl. Secondly, the nurse supporting the woman to adapt in the external environment. Thirdly, as an outcome of the two previous processes, the young woman should have a positive self-image and thus maintain her integrity.

The nurse provided an enabling environment by encouraging the young woman to share and deal with her previous experience as an opportunity to grow. The nurse also encouraged her to indicate how she felt about herself (self-image), before she could conclude how her family and friends felt about her. In this regard, the nurse enhanced self-awareness by enhancing the young woman's self-evaluation.

The nurse used the three elements of physical, psychological and social attributes to enhance the young woman's self-evaluation. The first element was suggesting to her to identify her own feelings about having a highly visible light skin colour (physical attributes). The second element was encouraging her to recognise her talents and needs (psychological attributes). The third element determined her relationship with the rest of the other family members and the community (social attributes).

During the conversation, the young woman indicated that one of her talents was singing due to her good soprano voice. The young woman then indicated she was going to join the church choir. It was thus evident that her self-esteem was enhanced.

The nurse realised therefore that there was a need to promote the young woman's self-development and growth. The nurse thus provided her with essential knowledge on oculocutaneous albinism. The nurse explained the inheritance pattern of oculocutaneous albinism. Care of the skin and eyes was discussed, and the nurse explained that due to lack of melanin, the skin is sensitive to ultraviolet rays. The nurse then provided the young woman with pamphlets on oculocutaneous albinism, and referred her to the Albinism Society of South Africa for further counselling and support.

The nurse then and the young woman agreed that the counselling session continue a week later. In the meantime the nurse suggested that the girl should think of strategies to create awareness of oculocutaneous albinism, so that she develop a sense of belonging. The nurse also suggested that she to think of other development and growth initiatives that would promote her adaptation in the environment. The nurse and young woman agreed that they would discuss these strategies during the next counselling session.

A week later the young woman visited the clinic as agreed. This time she looked happy and requested more information on oculocutaneous albinism. The young woman related excitedly that she had joined the church choir to socialise with the other young members (adapt in external environment). The nurse then noted that she felt self-worthy, her self-esteem was boosted, and she thus recognised her potential in society. In this regard, it was evident that the young woman's self-development and growth was effectively promoted.

As one of the strategies to facilitate the adaptation process in the external environment, the nurse and the young woman agreed that she should give a talk during a "Youth imbizo" planned in that community. The young woman agreed to talk about "Albinism: being different in the community". It was essential therefore to provide her with cognitive skills, including critical thinking, problem-solving and decision making skills to prepare for the presentation.

The third counselling session, a week later, was arranged for the same day as the talk at the 'Youth imbizo'. It was essential at that time to determine whether the interventions applied by the nurse to enhance her self-awareness and to support her adaptation in the environment were effective. One of the strategies was therefore to evaluate whether she was strengthened to talk on oculocutaneous albinism to her peers. One of her brothers accompanied the young woman to the youth imbizo. On arrival the young woman was nervous, but indicated that she could not wait to tell her peers how important it is to give people with oculocutaneous albinism a chance to develop and grow in the community.

As she stood up, it was evident from her posture and the tone of her voice that she had faith in herself, and felt confident to talk about oculocutaneous albinism. The young woman shared her experience of albinism, and eloquently related her relationship in the community. She related how myths and superstitions as well as misconceptions on oculocutaneous albinism had nearly destroyed her life. She emphasised that these negative beliefs in society could result in people with oculocutaneous albinism having a low self-esteem. She expressed her gratitude and related how she survived the hardships by using the interventions given by the nurse at the clinic. She indicated that she felt she had the ability to succeed in reaching a specific goal in life.

The young woman then suggested that as a way forward that people who were interested in becoming members of a youth albinism support group should raise their hands. The majority of the youth raised their hands, and expressed their commitment to supporting her to <u>raise awareness of oculocutaneous albinism</u>. In this regard, the young woman maintained her <u>self-efficacy</u> by deciding to canvass for a youth albinism support group without the nurse's influence.

The young woman then concluded her presentation, thanked the audience, and indicated that for the first time in her life she felt <u>valued and appreciated by the family and community.</u> Most importantly, she thanked the nurse for supporting, and helping her to improve her <u>low self-image</u> to a <u>positive self-image</u>. It was thus evident that she had maintained her sense of integrity because she used her cognitive skills and also maintained her self-efficacy. She had announced publicly that she felt strengthened and was intending to register for a degree in Psychology with the University of South Africa the following year. At the end of the presentation, the audience applauded the young woman for her motivational eye-opening presentation on "Albinism: being different in my community". The young woman promised to keep in contact with the nurse to update her on her success in life.

■ *Discussion*

The above example of the model case shows the <u>advancing process</u> of enhancement of a person with oculocutaneous albinism's self-concept. All the attributes of "enhancement of self-concept" were noted. The young woman had a low self-image, and as a result of the nurse's interventions, she developed a positive self-image. The model case depicts the role of the nurse enhancing <u>self-awareness</u> by enhancing <u>self-evaluation,</u> which resulted in the young woman's <u>self-esteem</u> being boosted, which then facilitated her <u>self-development and growth</u>. As the process advanced, the nurse <u>supported her adaptation</u> in the external environment. The outcome in the model case reflected a person with oculocutaneous albinism whose <u>sense of integrity</u> was strengthened, who maintained self-efficacy and felt valued and appreciated by the family and community.

4.2.5.2   Borderline case

According to Walker and Avant (1995:43), borderline cases are examples or instances that contain some but not all of the critical attributes of the concepts being examined.

■ *Example of a borderline case*

A 21 year-old woman with oculocutaneous albinism visited the clinic for counselling. The nurse at the clinic was too busy to provide her with an <u>enabling environment</u> to provide counselling on skin and eye care. The young woman was depressed and felt that, because of <u>her skin and </u>eye problems the nurse was not giving her the necessary attention. She decided not to visit the clinic again, and blamed the nurse for not caring. The nurse realised that the young woman with oculocutaneous albinism did not honour subsequent appointments.

The nurse then visited her at home. During discussion, the young woman indicated that she felt stigmatised when the nurse did not give her the necessary attention at the clinic. She felt that as an <u>active member of the youth</u> (self-esteem) dance group, she did not deserve to be stigmatised. The nurse blamed the family and friends for the young woman's attitude, and did not encourage her to do self-evaluation. The young woman felt that she needed a <u>sense of belonging</u> and a healthy relationship with family and friends.

■ *Discussion*

The above example of a borderline case lacked the advancing elements to enhance the self-concept. The young woman with oculocutaneous albinism's self-evaluation was not enhanced. She felt self-worthy and was therefore a member of the youth dance group. The nurse failed to enhance the young woman's self-awareness. The nurse did not initially create a supportive and enabling environment, but the nurse rectified this by visiting her at home. Most of the attributes to enhance the self-concept were missing hence the young woman did not feel strengthened, and still needed a sense of belonging to boost her self-esteem and maintain her integrity.

4.2.6   Identify antecedents and consequences

Concept analysis of "enhancement of self-concept" also entailed identifying antecedents and consequence of this concept. According to Walker and Avant (1995:45), antecedents and consequences are useful theoretically to assist in further refining the critical attributes, and identifying the social context in which the selected concept is used. Antecedents and consequences also assist theorists in identifying the underlying assumptions of the concept (Walker & Avant 1995:45).

4.2.6.1   Antecedents of enhancement of self-concept

Walker and Avant (1995:45) refer to antecedents as "those events or incidents that must occur prior to the occurrence of the concept". In this regard, an antecedent of enhancement of self-concept would be a person with a low self-esteem, who felt that the family and the community did not value and accept him or her. Another related antecedent emanates from the assumption of Wiedenbach's prescriptive theory that people need stimulation in order to make the best use of their capabilities and realise their self-worthiness (Wiedenbach 1970:1059).

4.2.6.2   Consequences of enhancement of self-concept

Walker and Avant (1995:45) refer to consequences as "those events or incidents that occur as a result of the occurrence of the concept". Therefore, consequences of enhancement of self-concept would be a person's ability to maintain integrity and a positive self-image. In this regard, another assumption of Wiedanbach's prescriptive theory is that people are endowed with the unique potential to develop within themselves the resources that enable them to sustain and maintain themselves (Wiedenbach 1970:1057-1060).

4.2.7 Define empirical referents

Walker and Avant (1995:46) indicate that the final step of concept analysis is to determine the empirical referents for the critical attributes. Empirical referents refer to "classes or categories of actual phenomena that by their existence or presence demonstrate the occurrence of the concept itself" (Walker & Avant 1995:46). However, Walker and Avant (1995:46) are of the opinion that in some cases the empirical referents are the same as the critical attributes (see section 4.2.4).

4.3 THEORETICAL DEFINITION OF ENHANCEMENT OF SELF-CONCEPT

Synthesis of the main concepts enabled the researcher to arrive at the following definition of enhancement of self-concept in the context of this model:

Enhancement of self-concept is an advancing process whereby the health professional, as the agent, improves the low self-image of a person with oculocutaneous albinism, who is the recipient, to a positive self-image. In this process, the health professional enhances self-awareness by enhancing the person's self-evaluation and self-esteem. The health professional also supports the person's adaptation in the environment by promoting the person's self-development and growth thereby assisting the person to develop a sense of belonging, and involving the person in creating awareness on oculocutaneous albinism. The process leads to the person's self-efficacy, who has cognitive skills to maintain his or her integrity.

4.4 CLASSIFICATION OF THE MAIN CONCEPTS

Classification of the main concepts was done to facilitate future prescription to use the model. The researcher used Dickoff et al's (1968:431) survey list to classify the concepts of the model into agents, recipients, context, procedures, dynamics and terminus. Dickoff et al (1968:437) emphasise that the survey list calls attention to certain significant aspects of activity, and to certain dimensions, knowledge, or other resources relevant to the activity. The proposed model was therefore classified and described as agents, recipients, context, procedures, dynamics and terminus as follows:

4.4.1   Agent

According to Dickoff et al (1968:437) and Wiedenbach (1970:1059), an agent is the propelling force that moves the practice towards a goal, and is a person or thing that produces an effect and takes an active part, or the person who does the actual work. The agents in this model play a fundamental role to enhance self-concept of people with oculocutaneous albinism, and they are health professionals, including nurses, social workers, psychologists and doctors.

The health professional as an agent should therefore use the model to produce an effect of improving the low self-image of a person with oculocutaneous albinism as a recipient, to that of a person with oculocutaneous albinism who has a positive self-image. Wiedenbach emphasises that the health professional as the agent has the following four basic responsibilities (Wiedenbach 1970:1059-1060):

- To reconcile assumptions about realities on oculocutaneous albinism as a congenital disorder;
- To specify objectives of the practice in terms of behavioural outcomes that are realistically attainable for the person with oculocutaneous albinism; and
- To engage in related activities which contribute to self-realisation and to the improvement of health care practice.

Most importantly, Dickoff et al (1968:439) maintain that the internal and external resources of the agent should be determined. The internal resources include the health professional's skills, competence and knowledge of oculocutaneous albinism, as well as commitment, techniques, and policies on oculocutaneous albinism available to or through the health professional (Dickoff et al 1968:439; George 1995:183). The health professional's external resources include those resources other than the health professional or agent, that are available for maintaining, supporting, developing, protecting or extending the agent's capacity, power, or flexibility (Dickoff et al 1968:439).

4.4.2 Recipient

The recipient is the person who receives the activity. In this study, the person with oculocutaneous albinism is the recipient of the health professional's actions (Dickoff et al 1968:440). According to Wiedenbach, the person with oculocutaneous albinism as the recipient is vulnerable, depends on others for help, risks losing individuality, dignity, worth, and autonomy (Wiedenbach 1970:1059). The health professional as the agent should thus be supportive of the person with oculocutaneous albinism as the recipient.

4.4.3 Context

The context is the situation in which the activity takes place, and comprises human, environment, professional, and organisational facilities (George 1995:183). Dickoff et al (1968:441) and Wiedenbach (1970:1061) refer to the context as a framework, and indicate that to view an activity from the aspect of the framework is to view the activity from the aspect of the matrix of that activity or total context of that activity. The context of this model is people with oculocutaneous albinism in urban and semi-urban areas of the Gauteng province, South Africa. Oculocutaneous albinism is a congenital disorder characterised by hypopigmentation of the skin, hair and eyes. McKenna (1997:69) indicates that it is essential to note that concepts have different meanings, depending on the context in which they are used.

4.4.4 Procedure

The procedure comprises the protocol and devices through which the agent is enabled to attain a set goal (George 1995:183). According to Dickoff et al (1968:444), procedure emphasises the path, steps or pattern to perform an activity. Dickoff et al (1968:444) point out further that procedure does not stress the outcome or the particularising features of the activity.

The procedures are the three advancing steps that the health professionals as the agents engage to enhance self-concept of people with oculocutaneous albinism, who are the recipients of the procedures. In the first step, the health professional should enhance the self-awareness of a person with oculocutaneous albinism who has a low self-image. In the second step, the health professional should support the person to

adapt in the environment. The third step is maintaining the person's sense of integrity as an impact of the first two steps.

4.5.5 Dynamics

The dynamics are the energy or power sources for the activity. Dickoff et al (1968:446) explain that dynamics would explore power sources, which may be physical, biological, psychological or chemical for the agents and recipients.

The dynamics to enhance the self-concept of people with oculocutaneous albinism will be determined by the three steps in the procedure. The *psychological dynamics* are essential to enhance the person's self-awareness. In this regard, the self-awareness process needs the psychological dimension of motivation and the drive of both the health professional and the person with oculocutaneous albinism to advance and grow. Another dynamic relevant is the *physical dimension*, as the person with oculocutaneous albinism recognises and accepts that her or his skin colour makes her or him different from other people.

The *social dynamics* are essential to support the person's adaptation in the environment. The adaptation process will be enhanced by the person's relationship with the other family members as well as the community.

The *developmental dynamics* are essential at the point where the person maintains integrity as a result of the health professional's previous interventions to enhance self-awareness, and to support the person's adaptation in the environment

The dynamics also involves reflection and feedback on the use of the model. Reflection will allow the person with oculocutaneous albinism to deal with previous experience and use the opportunity to grow. Feedback will allow the health professional to determine the value of using the model in practice.

4.4.6 Terminus

The terminus is the desired outcome the agent wishes to attain through the procedures, or the end result or goal to be attained by the agent's action (Dickoff et al 1968:441;

George 1995:184). Terminus is therefore the person with oculocutaneous albinism with enhanced self-concept as evidenced by enhanced self-awareness, adaptation in the environment and integrity.

4.5 PROCESS OF ENHANCEMENT OF SELF-CONCEPT

The process of enhancement of self-concept of people with oculocutaneous albinism is schematically portrayed in figure 4.4. The process comprises the recipient, agent, context, procedure, dynamics, terminus, and is discussed in detail in chapter 5.
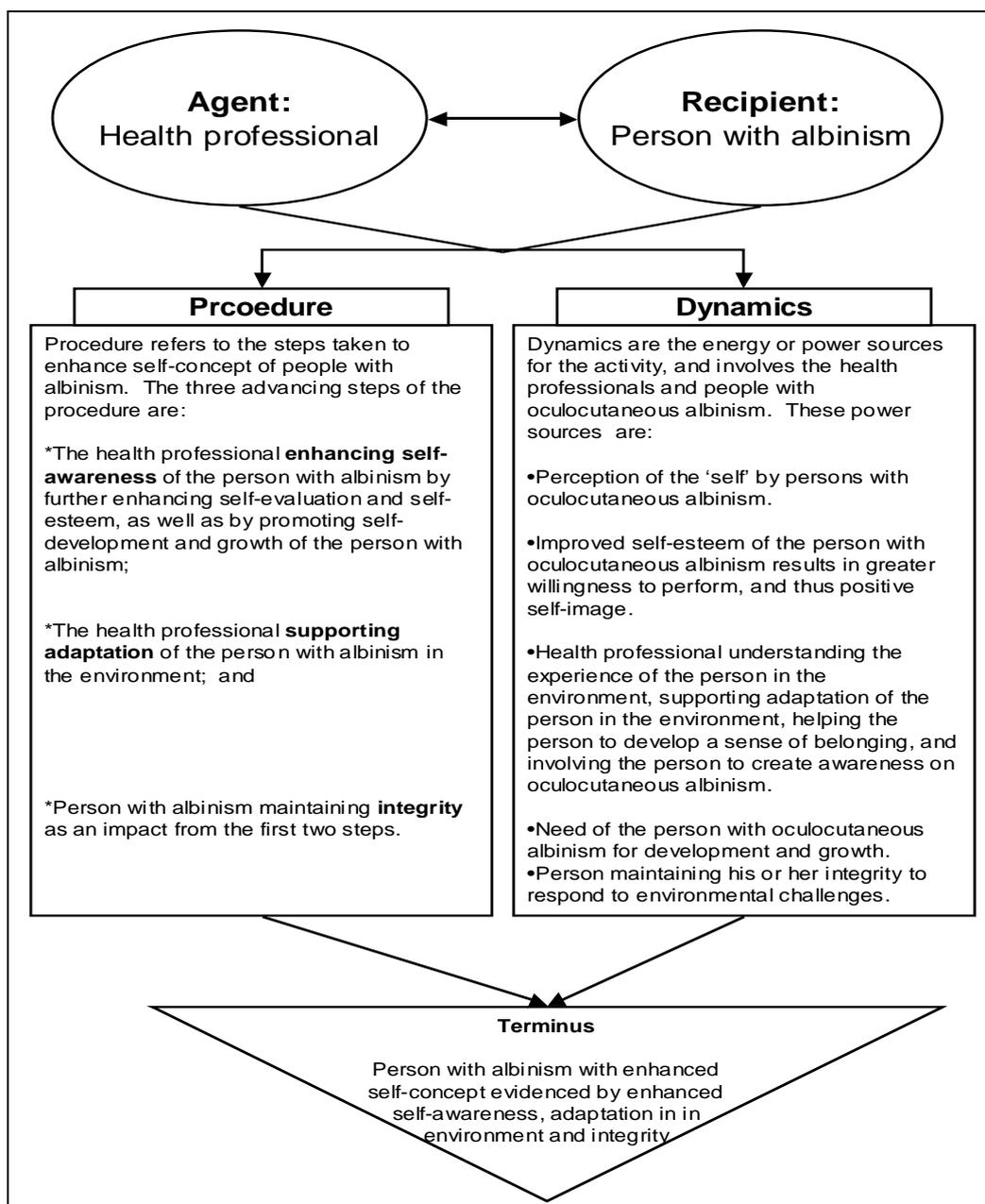
Figure 4.4
The process of enhancement of self-concept

4.6 CONCLUSION

This chapter defined the conceptual meaning of "enhancement of self-concept" to develop the model. This entailed concept analysis of "self-concept" and "enhancement" by identifying, examining and clarifying the attributes of these concepts.

Three steps of the model were identified. The first step entailed the health professional enhancing self-awareness of the person with oculocutaneous albinism. The second step entailed the health professional supporting the person's adaptation in the environment. The third step occurred as a result of the first two steps, and entailed the person maintaining his or her integrity.

Chapter 5 describes the structure and process of the model, and provides guidelines for operationalisation of the model in practice. It also uses various strategies to evaluate the model.

CHAPTER 5

DESCRIPTION OF A MODEL FOR ENHANCEMENT OF SELF-CONCEPT OF PEOPLE WITH ALBINISM AND GUIDELINES FOR OPERATIONALISATION OF THE MODEL

5.1 INTRODUCTION

Chapter 4 developed the proposed model by creating a conceptual meaning of "enhancement of self-concept" as the core building block. Through the concept analysis of the key concepts, the researcher identified three main steps to enhance the self-concept of people with oculocutaneous albinism. These steps entail the health professional *enhancing the self-awareness of the person with oculocutaneous albinism;* the health professional *supporting the person's adaptation process* in the environment, and finally, the person *maintaining his or her integrity* as an outcome of the first two steps.

Chinn and Kramer (1999:83) emphasise that once a theory has been developed, the questions for anyone interested in using or understanding it will ask "What is this?" and "How does it work?" This chapter therefore answers those questions, and focuses on describing the structure and process of the model. The guidelines for operationalisation of the model in practice are also formulated and described. These guidelines are proposed to meet the objectives of each of the three steps of the model.

Most importantly, the model was subjected to rigorous internal and external evaluation. This was done to determine its potential usefulness in nursing practice and research. The researcher used Chinn and Kramer's (1995:107-119; 1999:100-119), McKenna's (1997:223-230) and Pearson, Vaughan and Fitzgerald's (2005:226-228) strategies for theory evaluation.

5.2   OVERVIEW OF THE MODEL

An overview and detailed description of the model is provided in this section, using Chinn and Kramer's (1999:84-96) strategies for theory description. Figure 5.1 is a graphic representation of the model. The model is described under the headings:

■ **Orientation to the model**

- Purpose

- Assumptions

- Context

- Theoretical definitions

- Relationship statements

■ **Structure of the model**

153
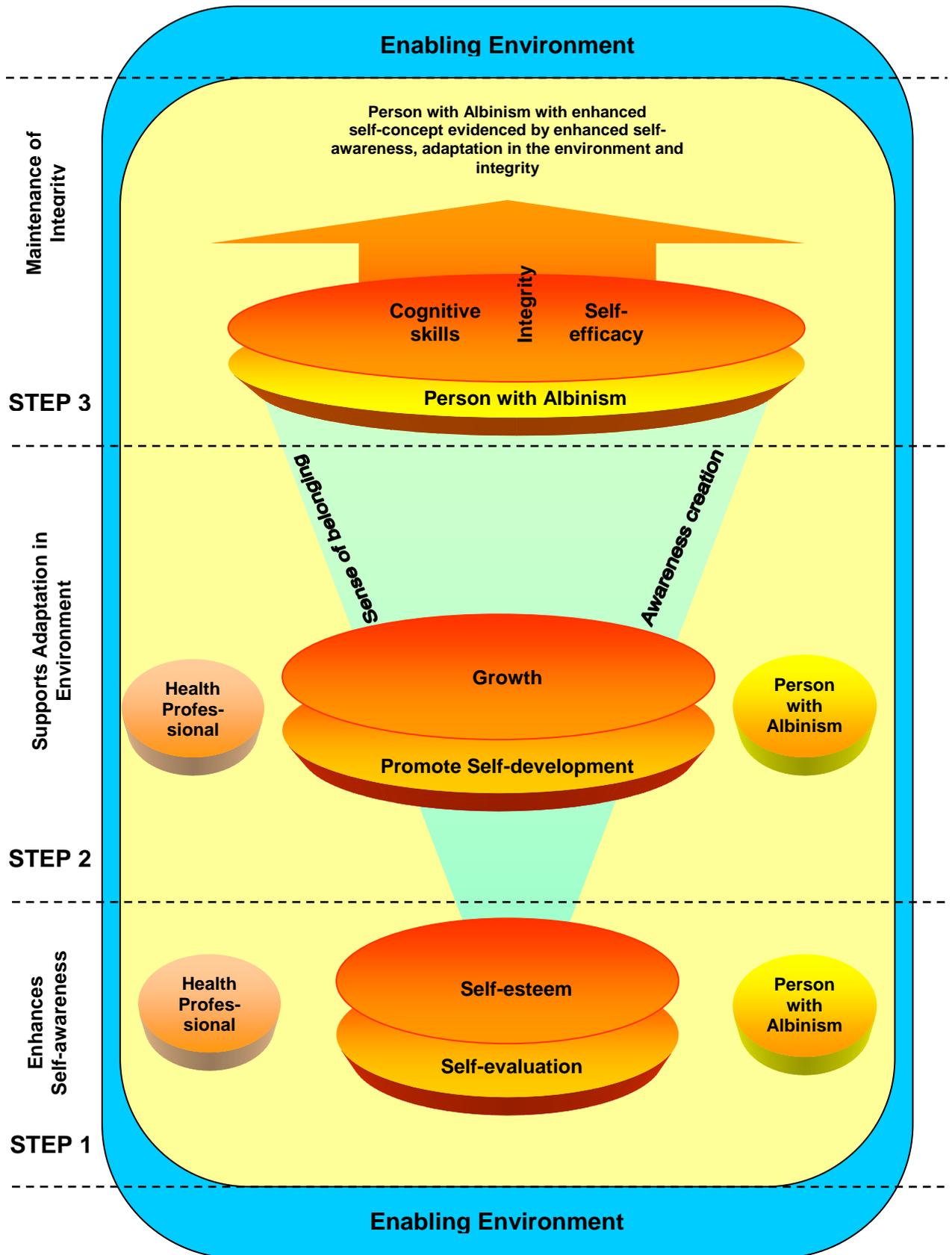
Figure 5.1

A model for enhancement of self-concept of people with

Albinism

5.2.1 Orientation to the model

Figure 5.1 portrays the model as an advancing step-by-step process to improve the self-image of the person with oculocutaneous albinism. The health professional should provide an enabling environment throughout the process, to enable the person to develop and grow towards a strengthening direction of maintaining integrity.

The model consists of the following three major steps:
- Step 1: Health professional enhancing self-awareness of the person with oculocutaneous albinism;
- Step 2: Health professional supporting adaptation of the person with oculocutaneous albinism in the environment; and
- Step 3: Person with oculocutaneous albinism maintaining his or her integrity.

The researcher used the three main themes of the findings to relate and formulate the three steps of the model (see chapter 3). Table 5.1 shows the relationship of the main themes of the findings with the steps of the model.

Table 5.1   Relationship of main themes of the findings with steps of the model

Main themes of the findings	Steps of the model
1st theme: Perception of the "self" by people with oculocutaneous albinism	Step 1: Health professional enhancing self-awareness of person with oculocutaneous albinism
2nd theme: Experience of people with oculocutaneous albinism with the external environment	Step 2: Health professional supporting adaptation of the person with oculocutaneous albinism in the environment
3rd theme: Expression of need for development and growth by people with oculocutaneous albinism	Step 3: Person with oculocutaneous albinism maintaining integrity

- ***Step 1: Health professional enhancing self-awareness of the person with albinism***

The first step of the model entails the health professional enhancing self-awareness of the person with oculocutaneous albinism. To ensure that the model objectively represents the life-experience of people with oculocutaneous albinism, after concept analysis (see chapter 4), the first theme of the findings was converted into the first step of the model, as shown in table 5.1.

The first theme of the findings was perception of the "self" by people with oculocutaneous albinism. The focus of this theme was an experience echoed by the participants, recognising that they 'themselves' play an important role in their own lives. Therefore, the role of the health professional to enhance the person's self-awareness is an important starting point of this model. In this regard, Goleman (1997:47) refers to self-awareness as "the keystone to emotional intelligence". The process of enhancing self-awareness thus entails the health professional enhancing the person's self-evaluation and self-esteem.

Most importantly, it emerged in the findings that the study participants shared the perception of the self in relation to the external environment (see chapter 3). This led to the second theme of the findings, and later to the second step of the model.

- ***Step 2: Health professional supports adaptation of the person with albinism in the environment***

The second step of the model entails the health professional supporting adaptation of the person with oculocutaneous albinism in the environment. The first step of the model is a prerequisite of the second step. After concept analysis, the second step of the model was also converted from the second theme of the findings as shown in table 5.1.

The second theme of the findings was identified from the experience of people with oculocutaneous albinism with the external environment. In this theme, it emerged that interaction of people with oculocutaneous albinism with the external environment influenced their self-image, sense of belonging, and impact of being stigmatised on the basis of myths and superstitions.

In step two of the model, the health professional should thus support adaptation in the external environment by promoting the person's self-development and growth. The development and growth initiatives entail involving the person in creating awareness of oculocutaneous albinism, and supporting the person to develop a sense of belonging in the family and community.

- *Step 3: Maintain integrity*

The third step of the model is for the person with oculocutaneous albinism to maintain his or her integrity. The third step was also converted from the third theme of the findings, as shown in table 5.1. The third theme of the findings was the participants' expression of need for development and growth. In this theme, it emerged that the development and growth process is influenced by various factors in the external environment. These factors include the role of the family and community, non-governmental organisations (NGOs), government departments, and the private sector.

The third step of the model is mainly the responsibility of the person with oculocutaneous albinism to maintain his or her integrity, and feel valued and accepted by the family and community. Successful interventions of the health professional during the first two steps are thus prerequisites of the third step of the model. The third step entails the person with oculocutaneous albinism maintaining self-efficacy, and having the cognitive skills to face the challenges in the environment (see section 5.2.2 for the steps).

5.2.1.1 *Purpose*

The purpose of this model is to provide a frame of reference for health professionals to enhance the self-concept of people with oculocutaneous albinism. The ultimate goal of the model is a person with enhanced self-concept evidenced by enhanced self-awareness, adaptation in the environment and integrity. Chinn and Kramer (1999:84) emphasise that the purpose of a theory is important because it specifies the context and situations in which the theory applies.

The next subsection discusses the assumptions of the model.

5.2.1.2 *Assumptions*

Chinn and Kramer (1999:95) and McKenna (1997:80) indicate that assumptions are central components of a theory or model, and are those basic givens or accepted truths that are fundamental to theoretic reasoning. The assumptions of the model were thus based on the findings of the study, and are congruent with Neuman's systems model, Wiedenbach's prescriptive nursing theory, and the rehabilitation self-empowerment assumptions (Dell-Orto & Marinelli 1995:258; Wiedenbach 1970:1058).

❏ *Neuman's systems model*

- Neuman's systems model depicts an open system in which people with oculocutaneous albinism and their environment are in dynamic interaction (Neuman 1995:12). According to Neuman, environment is the internal and external factors that surround a person. Both the internal (the self) and external environments thus affect the person's life and development (Stanhope & Lancaster 2000:207).
- Neuman depicts the person as a system composed of interactive dimensions, namely, physiological, psychological, socio-cultural, spiritual and developmental (Neuman 1995:12-15).

❑ *Wiedenbach's prescriptive theory*

Wiedenbach's prescriptive theory specifies the nature of the nurse's action, and the thinking process that determines the actions to implement the basic concepts of the model (Wiedenbach 1970:1060). The assumptions of Wiedenbach's prescriptive theory were adapted to people with oculocutaneous albinism as follows:

* Self-awareness and self-acceptance are essential for the integrity and self-worthiness of people with oculocutaneous albinism.

* People with oculocutaneous albinism need stimulation in order to make the best use of their capabilities and realise their self-worth.

* People with oculocutaneous albinism are endowed with unique potential to develop within themselves the resources that enable them to sustain and maintain themselves.

* People with oculocutaneous albinism basically strive towards self-direction and relationship independence, and the desire not only to make the best use of their capabilities and potentials, but also to fulfil their responsibilities.

* Whatever people with oculocutaneous albinism do represents their best judgement at the moment of doing it.

❑ *Rehabilitation self-empowerment assumptions*

Dell-Orto and Marinelli's (1995:258) rehabilitation self-empowerment assumptions were also adapted to people with oculocutaneous albinism as follows:

* Each person with oculocutaneous albinism is of great worth and dignity.

* Individuals with oculocutaneous albinism strive to grow and change in a positive direction.

* Each person with oculocutaneous albinism should have an equal opportunity to maximise his or her potential with societal help.

* People with oculocutaneous albinism should be free to make their own decisions about the management of their lives.

❑ *Assumptions of the model: enhancement of self-concept*

- The primary assumption of the model to enhance self-concept of people is that human beings support stimulation experts comments to make the best use of their capabilities so as to realise their self-worthiness.

- An enabling environment will enhance the self-concept of the person with oculocutaneous albinism.

- Enhancing self-awareness the person with oculocutaneous albinism will improve his or her self-esteem.

- Involving the person with oculocutaneous albinism in creating awareness of oculocutaneous will improve his or her self-esteem.

- Supporting the person with oculocutaneous albinism with adaptation in the environment will improve his or her self-esteem.

- The person with oculocutaneous albinism will develop a sense of belonging when the health professional supports him or her with adaptation in the environment.

- Self-evaluation will determine self-efficacy of the person with oculocutaneous albinism.

- The person with oculocutaneous albinism will use cognitive skills to maintain his or her integrity.

The next sub-section clarifies the context of the model.

5.2.1.3 Context

Chinn and Kramer (1999:74) emphasise that contextual placement describes the circumstances within which the theoretic relationships are expected to be empirically relevant. McKenna (1997:69) indicates that concepts have different meanings, depending on the context in which they are used.

The context of this model in terms of environment was urban and semi-urban areas around Gauteng province, South Africa. Gauteng is the smallest of the nine provinces in South Africa, by size, almost completely urbanised, and has the second largest provincial population. Its 17010 km^2 houses approximately 8 837 000 (19.7%) people, and the population density is 553.5 people/km.

The population is divided into 6 520 000 (74%) Blacks, 1 758 000 (20%) Caucasians, 338 000 (3.8%) Coloureds, and 218 000 (2.2%) Indians, and 97% of the population are designated as urban dwellers (South Africa 2001).

The Division of Human Genetics, National Health Laboratory Services and University of the Witwatersrand in South Africa run the medical genetic clinic at Chris Hani Baragwanath Hospital in Johannesburg. Furthermore, in an effort to improve access to health services of people with congenital disorders, the Medical Genetics Education Programme is underway in all the provinces of South Africa. The aim of the Medical Genetics Education Programme is to improve the knowledge and skills of health professionals on the management and care of congenital disorders, including albinism.

Oculocutaneous albinism is one of the priority congenital disorders characterised by hypopigmentation of the skin, hair and eyes. The researcher discovered that most of the health professionals around the Gauteng area, especially the nurses in the hospital and clinics still lack knowledge on oculocutaneous albinism and other congenital disorders. The model will therefore be a source of assistance for health professionals to enhance the self-concept of people with oculocutaneous albinism. In this regard, the health professional will also understand and address the person's physiological, psychological, socio-cultural, spiritual and developmental needs.

The next subsection discusses the theoretical definitions of the concepts to validate the model in the real world (Walker & Avant 1995:25).

5.2.1.4 Theoretical definitions

Walker and Avant (1995:25) point out that theoretical definitions are also referred to as conceptual definitions, and are the means by which the theorist introduces the reader to the critical attributes of the concepts. Enhancement of self-concept as the core concept of the model, and related concepts were therefore defined and contextualised to specify the ideas on which the model structure was built (Chinn & Kramer 1999:74).

5.2.1.4.1   Enhancement of self-concept

In the context of this model, enhancement of self-concept is an advancing process whereby the health professional, as the agent, improves the low self-image of a person with oculocutaneous albinism, who is the recipient, to a positive self-image. In this process, the health professional enhances self-awareness by enhancing the person's self-evaluation and self-esteem. The health professional also supports the person's adaptation in the environment by promoting the person's self-development and growth thereby assisting the person to develop a sense of belonging, and involving the person in creating awareness on oculocutaneous albinism. The process leads to the person's self-efficacy, who has cognitive skills to maintain his or her integrity.

5.2.1.4.2   Related concepts

The following related concepts were defined to give the model its particular character (Chinn & Kramer 1999:84).

- *Health professional*

The health professional is a person trained in the field of health care, who is proficient as a counsellor, teacher, resource and advocate, and is regarded as the first level of contact to help people with oculocutaneous albinism to solve their problems, propose solutions and implement the appropriate solutions (Stanhope & Lancaster 2004:996).

The health professional in the context of this model therefore refers to nurses, social workers, psychologists and doctors, who, as the agents, should be able to enhance the self-concept of the person with oculocutaneous albinism, and create an enabling environment.

- ***Person with oculocutaneous albinism***

In the context of this model, a person with oculocutaneous albinism is the recipient of the health professional's interventions. Oculocutaneous albinism is a congenital disorder of genetic etiology characterised by the absence of pigment in the skin, hair and eyes (see chapter 1 section 1.2.1 for the genotype of oculocutaneous albinism and mode of inheritance).

- ***Enhancement***

In the context of this model, enhancement refers to improving or strengthening the self-image of the person with oculocutaneous albinism. The role of the health professional as the enhancer will be facilitated by the use of his or her expert knowledge on albinism, and use of his or her cognitive skills to enhance the person's self-concept.

- ***Self-awareness***

Self-awareness entails the person's self-knowledge to foster self-worthiness, and understand his or her own identity, consciousness and feelings. It also entails the person becoming aware of his or her appearance that makes him or her different from others.

- ***Self-evaluation***

Self-evaluation is used to enhance self-awareness of the person with oculocutaneous albinism, and entails supporting the person to evaluate the physical, psychological and social attributes.

- ***Self-esteem***

Self-esteem will occur as an outcome of successful self-evaluation. The person's self-esteem should be improved, as it is regarded as the most important and powerful value in the motivation system.

Self-esteem thus entails having a favourable opinion about oneself, and feeling happy about one's character and abilities.

- *Development and growth*

Development and growth occurs as a result of adapting in the environment, and refers to a person developing and growing intellectually and emotionally so as to face other challenges in life. For development and growth to take place, the person with oculocutaneous albinism and the health professional need to take some initiatives.

- *Adaptation*

Adaptation of the person with oculocutaneous albinism entails adjusting to the environment so that the person develops a sense of belonging. Adaptation is thus influenced by the external environment as well as by the individual person (internal). During the adaptation process in this model, the health professional, as the agent, is regarded as the focal stimulus surrounding the person, and constitutes the greatest degree of change impacting on the person (Aggleton & Chalmers 2000:80).

- *Integrity*

Integrity refers to the person with oculocutaneous albinism being in control of his or her life. Integrity of the person will occur as a result of adaptation in the environment, and growth and development throughout the life cycle. Integrity also entails making decisions for oneself without influence from outside, and maintaining a positive self-image despite the challenges in the environment.

- *Cognitive skills*

Cognitive skills involve the use of intellectual and critical skills in problem solving and making decisions (Wilkinson 2001:13). The health professional should use cognitive skills to apply the model systematically and logically to enhance the self-concept of the person. The person with oculocutaneous albinism also needs

the cognitive skills to solve problems and use critical thinking skills to make decisions in life.

- *Self-efficacy*

Self-efficacy is associated with power and effectiveness, and means producing the intended result for the person with oculocutaneous albinism to maintain his or her integrity.

- *Enabling environment*

An enabling environment is a psychological process initiated by the health professional to encourage the person to use his or her experience of oculocutaneous albinism to develop and grow in a positive direction. An enabling environment should thus support the person to identify his or her self-worthiness. The environment should thus be calm, warm and relaxed. For successful enhancement of self-concept, there should be mutual trust and respect between the person and the health professional. An enabling environment should be one where the person is not discriminated against or stigmatised due to myths and superstitions. In this regard, the health professional should also educate the community on oculocutaneous albinism.

5.2.1.5 Relationship statements

Relationship statements describe, explain, or predict the nature of the interactions between the concepts of the model (Chinn & Kramer 1999:77). The researcher used deductive reasoning to develop the following specific relationship statements from the model:

(1) The central relationship statement of the model is that the process of enhancing self-concept takes place between the health professional as the agent, and the person with oculocutaneous albinism as the recipient, with an outcome of having a person with enhanced self-image.

(2) The health professional creates an enabling environment for the enhancement of self-concept of the person with oculocutaneous.

(3) The health professional improves the self-esteem of the person with oculocutaneous albinism by enhancing the person's self-awareness.

(4) The self-esteem of the person with oculocutaneous albinism improves with involvement in the awareness creation of oculocutaneous albinism.

(5) Improved self-esteem of a person with oculocutaneous albinism enables him or her to adapt in the environment with the support of the health professional.

(6) The health professional supports adaptation of the person with oculocutaneous albinism in the environment by involving him or her in the creation of awareness of oculocutaneous albinism.

(7) The health professional supports adaptation of the person with oculocutaneous albinism in the environment by assisting him or her to develop a sense of belonging.

(8) Self-evaluation determines self-efficacy of the person with oculocutaneous albinism.

(9) The person with oculocutaneous albinism uses cognitive skills to maintain his or her integrity.

The next subsection describes the structure of the model emerging from these relationship statements.

5.2.2 Structure and process of the model

The structure of the model refers to how the model is constructed, and gives an overall form to the conceptual relationships within the model (Chinn & Kramer 1999:92). The process and steps of the model as well as different shapes and colours used are described. Furthermore, this subsection discusses the health professional's attributes that enable him or her to enhance a person's self-concept.

5.2.2.1 Description of the model

The graphic representation of the model to enhance self-concept of people with oculocutaneous albinism in figure 5.1 is clear. Most importantly, the linkages of the concepts in the three steps of the model are understandable. Various shapes and colours are used which include blue, yellow, green, red and gold to evoke different moods during the process (Jordaan & Jordaan 1998:650).

Blue border of the model denotes a calm enabling environment created by the health professional throughout the process to enhance self-concept of the person with oculocutaneous albinism. The yellow background symbolises hope for the person.

The greenish funnel shaped linkages originate from the enhancement interaction of the health professional and person with oculocutaneous albinism. This funnel shape symbolise an advancing process, progressive development and growth, strength and direction for the person to ultimately maintain integrity. The 'greenish' funnel also denotes wealth of information on oculocutaneous albinism gained during the awareness creation activities, and sense of belonging initiatives.

The round yellowish and reddish shapes denote interaction of the health professional and person with oculocutaneous albinism during the processes. The oblong reddish and goldish structures denote the quest for warmth, and hope of the person during the process of enhancing self-evaluation. The oblong reddish shapes also symbolise quest of the person to attain cognitive skills and self-efficacy. The funnel ends with a reddish bold up-pointed arrow that symbolises energy and achievement of the person with oculocutaneous albinism to maintain integrity.

- **Steps of the model**

The reasoning of the model as an advancing process is easy to follow, and emerge from the relationships of the model. The process of the model therefore takes place in three overlapping steps as depicted in figure 5.1, namely

Step 1: Enhancement of self-awareness

Step 2: Support adaptation in the environment

Step 3: Maintenance of integrity.

❑ *Step 1: Enhancement of self-awareness*

In step 1 of the model, the role of the health professional as the agent is clearly depicted as an important intervention to enhance self-awareness of the person with oculocutaneous albinism. The process entails the health professional enhancing the person's self-evaluation and self-esteem as shown in figure 5.2.

Self-awareness is an essential concept of 'self-concept', and defined as "knowledge and understanding of one's own character" (*Oxford Advanced Learners Dictionary of Current English* 2004:1067). According to Goleman (1997:48), people with oculocutaneous albinism who engage in self-awareness are sure of their own boundaries and tend to have a positive outlook on life.
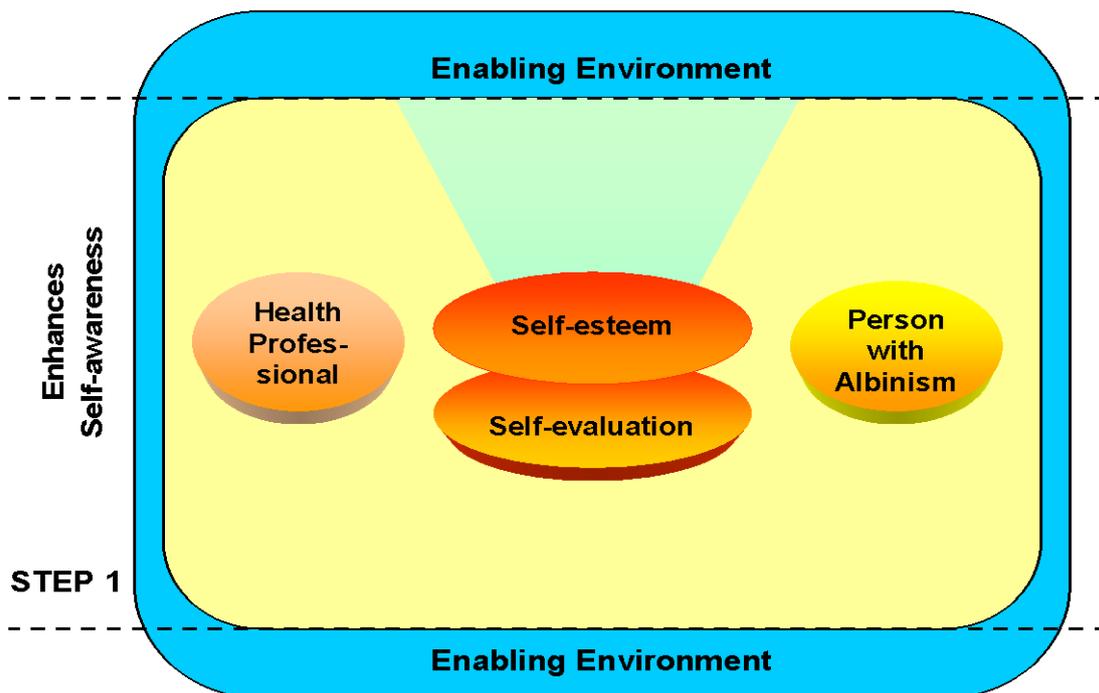

Figure 5.2

Step 1: Health professional enhancing the

self-awareness of the person with Albinism

Self-awareness is a keystone for the person to realise his or her maximum potential. The health professional should enhance self-awareness by enhancing self-evaluation, which entails physical, psychological and social dimensions. In this regard, the health professional should encourage and support the person to identify how he or she feels about his or her own appearance (physical), his or her abilities (psychological), and relationships with the family and community (social).

As a result of the successful self-evaluation interventions, the person's self-esteem should be improved. Self-esteem is the most important and powerful value in the motivation system, and is defined as "favourable appreciation and opinion of oneself" (*Oxford Textbook of Public Health* 2002:839). As a result of self-esteem, the person will have self-respect, which is defined as "a feeling of confidence and pride in one's own abilities and worth" (*Collins English Dictionary* 2006:746).

Self-acceptance is a prerequisite of self-esteem, and a process, which improves with effective psychotherapy and counselling (Friedland 2005:64). It is therefore fundamental that the health professional with expert knowledge should provide the necessary counselling and psychotherapy to enhance self-acceptance and self-esteem of the person with oculocutaneous albinism. The underpinnings of self-esteem are therefore a sense of personal efficacy and a perception of being accepted and valued by family and community.

A person with oculocutaneous albinism with an improved self-esteem will enable the health professional to promote the person's self-development and growth in the next step. The next step of the model therefore entails the health professional supporting the person in the environment.

❑ *Step 2: Support adaptation in the environment*
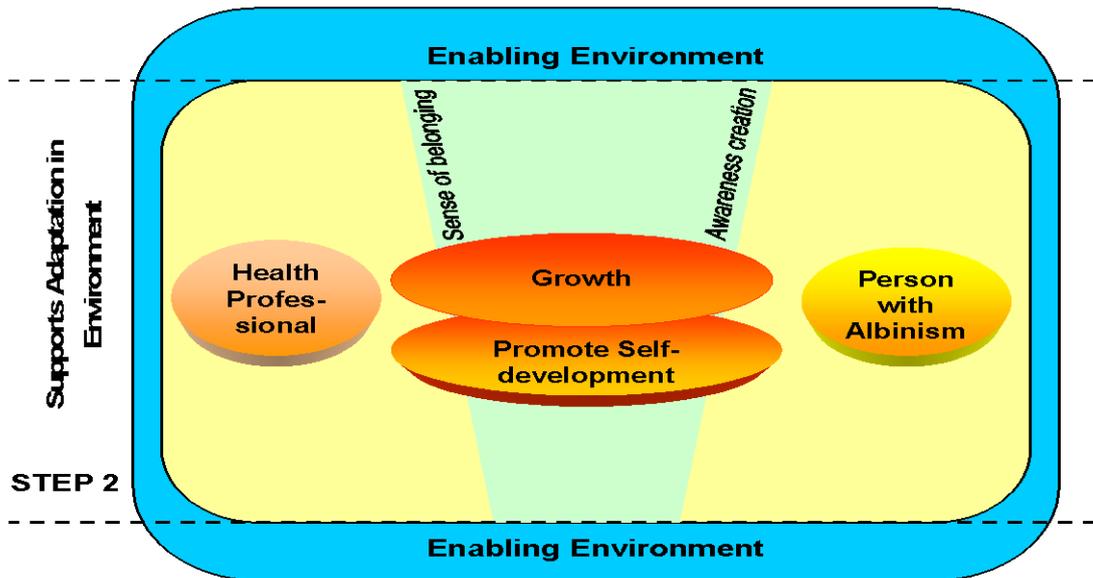
Figure 5.3

Step 2: Health professional supporting the person with Albinism in environment

In the second step of the model, the health professional supports the adaptation of the person with oculocutaneous albinism in the environment. This step entails the health professional promoting the person's self-development and growth, supporting the person to develop a sense of belonging, and involving the person in creating awareness of oculocutaneous albinism.

Environment is regarded as a fundamental concept in the nursing care. According to Pearson et al (2005:69), environment together with health, nursing and the person are domain concepts that are considered integral parts of any model. The external environment in this model may be outside stimuli that affect the development and growth of the person with oculocutaneous albinism.

Jordaan and Jordaan (1998:641) point out that the external environment may be one of harshness, rejection, uncertainty, warmth, acceptance or security. The development and growth initiatives should thus involve the family to help the person maintain a sense of belonging at an early stage of development, so as to feel accepted in the family and community.

The health professional should note the fundamental role of the family, which entails the early socialisation practices of parents, encouraging early attainment of goals, and supporting a high standard of excellence and self-direction of the person (DuBois 2003:939). However, the parents should not be overprotective of their children with oculocutaneous albinism because this may delay the individual's self-development and growth.

Other initiatives to promote a sense of belonging are encouraging the person to be involved in sports of interest, and having friends that the person can relate to. It emerged in the findings that psychosocial support of the person could be enhanced by involvement in sports and having supportive friends. Furthermore, belonging to a support group has been shown to have an significant impact on developing a sense of belonging, and improving self-esteem of a person with oculocutaneous albinism.

Once the person has developed a sense of belonging, he or she will feel empowered to create awareness on oculocutaneous albinism. Awareness creation involves both the health professional and the person marketing and mobilising oculocutaneous albinism, sharing experiences using the media, (for example, television, radio and newsletters), participating in relevant policy development and implementation, and being involved in developing relevant educational material. In the process of creating awareness, the person should be empowered to use cognitive skills to maintain his or her integrity to make decisions and solve problems, which leads to the third step of the model.

❑ *Step 3: Maintenance of integrity*
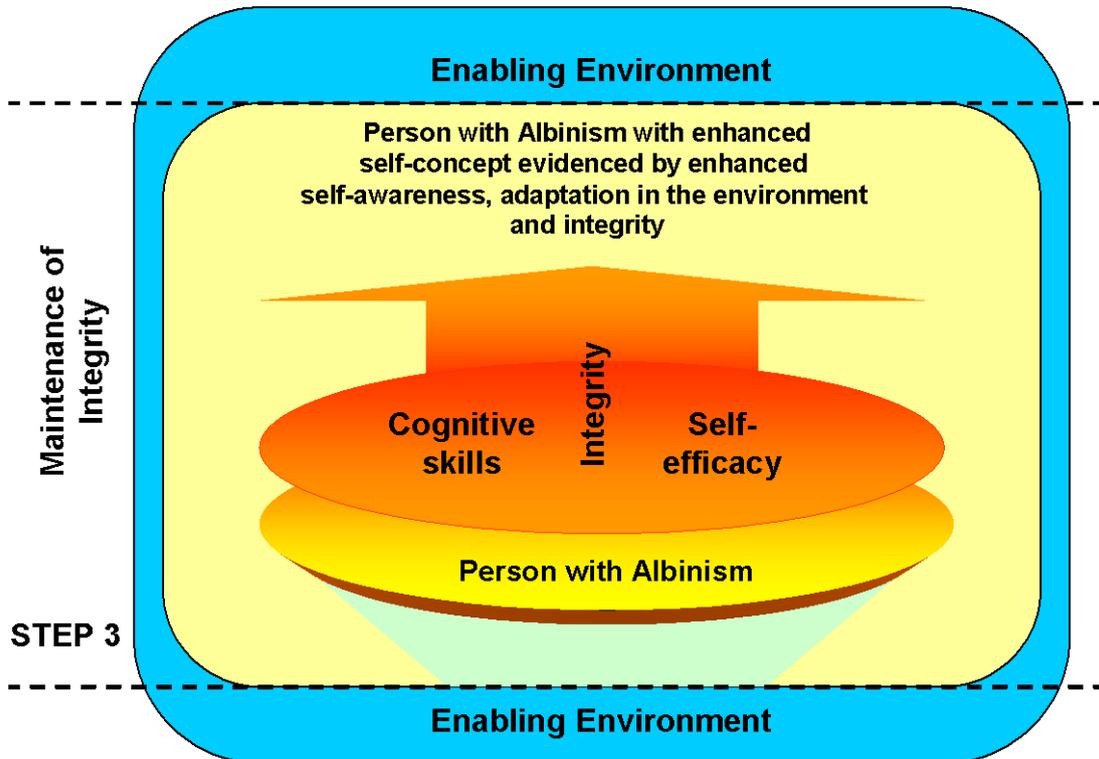
Figure 5.4

Step 3: Person with Albinism maintaining integrity

In the third step of the model, the person with oculocutaneous albinism should be empowered to maintain his or her integrity as an outcome of the first two steps. In this step, the person maintains self-efficacy and uses cognitive skills to adapt in the environment and maintain integrity as shown in figure 5.4. The third step is fundamental in that the first two steps were mostly the role of the health professional. However, the third step is mostly the responsibility of the person to sustain integrity in life.

Self-efficacy is described as the "internal state that a person experiences, such as competence or ability to perform or achieve a desired task" (*Dictionary of Public Health* 1996:105).

Integrity is associated with strength, self-determination and effectiveness. Clinebell (1992:179) emphasises that the strength that results from ego integrity or self-integrity is wisdom. The ego integrity of the person with oculocutaneous albinism is the cumulative result of development and growth throughout his or her life cycle. Clinebell (1992:179) warns that the opposite of ego integrity is despair.

Philpott (1995:100) supports self-determination associated to integrity, and reveals that self-determination would benefit not only the person with oculocutaneous albinism, but also the community. Self-determination is "the ability to make a decision for oneself without influence from outside" (*Collins English Dictionary* 2006:746).

In this regard, Levine distinguishes between personal, social and structural integrity. Personal integrity focuses on a sense of self of the person with oculocutaneous albinism, and the "self" is defined, defended, and described only by the soul that owns it. The advancing process of the model up to the third step depicts personal integrity as a development and growth process for the person. Social integrity, then, involves a definition of self that goes beyond the individual, and the person's identity is connected to the family, community, culture, ethnicity, religion, vocation, education, and socio-economic status. Structural integrity focuses on the healing process, and supports the idea that sense of structural integrity is more than a physiological need of an individual (George 2002:222).

The outcome of the interventions of all three steps of the model should be a person with oculocutaneous albinism with enhanced self-concept evidenced by enhanced self-awareness, adaptation in the environment and integrity. For the successful enhancement of self-concept, the health professional should possess certain attributes.

5.2.2.2 *Attributes that enhance self-concept*

Interpersonal relationship, communication, facilitation, leadership and advocacy are the essential attributes that the health professional requires to enhance self-concept of people with oculocutaneous albinism.

- ***Interpersonal relationship***

Interpersonal relationship pertains to the health professional's ability to maintain a healthy, polite and respectful attitude when interacting with the person. The health professional should be empathetic, so as to help the person deal with his or her emotions. The focus of empathy means sharing of feelings from the point of view of the person with oculocutaneous albinism.

- ***Communication***

According to Dennill, King and Swanepoel (1999:143), communication is the "exchange of factual information, perceptions and ideas between two people who strive to reach a mutual understanding". The health professional should be able to express facts on oculocutaneous albinism clearly and logically. The health professional should also have effective communication skills such as listening, motivation, reflection, self-disclosure and facilitation (Dennill et al 1999:152).

Self-disclosure is a process whereby the person will communicate personal information, feelings and thoughts both verbally and nonverbally to the health professional (Dennill et al 1999:146). The health professional should be sensitive to self-disclosure so as to maintain a healthy interpersonal relationship with the person with oculocutaneous albinism.

- ***Facilitation***

Collins English Dictionary (2006:281) defines facilitation as "making easier the progress of something". Synonyms of facilitation include help, promote, ease, pave the way for and speed up (*Collins Thesaurus* 2006:261).

Facilitation is one of the fundamental attributes that the health professional should have to influence the advancing process of enhancing the person's self-image. Facilitation entails helping the person to attain and maintain a positive self-image through the purposeful interventions of the model (George 1995:357).

In his or her facilitation role, the health professional should have the competence to help the person promote self-development and growth, and should build trust and confidence of the person with oculocutaneous albinism (Dennill et al 1999:152).

- *Leadership*

As a leader, the health professional should have initiative, which is the extent to which the health professional generates new ideas and improves where circumstances require (Stanhope & Lancaster 2000:629). Most importantly, as a leader, the health professional should empower the person by enhancing his or her self-image. Leadership requires advocacy skills so as to support the person with oculocutaneous albinism accordingly.

- *Advocacy*

Advocacy is the "instance of speaking or writing on behalf of someone else, and using persuasion in support of another person" (Stanhope & Lancaster 2004:132). The health professional should play the advocacy role, and support the rights of people with oculocutaneous albinism to be recognised as people with potential in the community.

Advocacy requires the skill of assertive communication channels within and among organisations. According to Stanhope and Lancaster (2004:144), health professionals' practical advocacy skills include recognising the ethical principles for effective advocacy, which recommend that the health professional should
- Act in the best interest of the person with oculocutaneous albinism.
- Keep the person informed.
- Carry out activities to enhance the person's self-image with diligence and competence.
- Act impartially and offer frank, independent advice to the person.
- Maintain the person's confidentiality.

Having developed and described the model, and described the attributes to enhance self-concept, it was essential to formulate and describe guidelines for operationalisation of the model in practice.

5.3   GUIDELINES FOR OPERATIONALISATION OF THE MODEL

The proposed guidelines describe strategies to assist health professionals to enhance self-concept of people with oculocutaneous albinism in a practical situation. LoBiondo-Wood and Haber (2002:294) emphasise that operationalisation of the model is essential to translate the concepts of the model into observable and measurable phenomena.

The guidelines are offered as suggestions and based on achieving the objectives of the three steps of the model: (1) enhancing self-awareness of the person with oculocutaneous albinism; (2) supporting adaptation of the person with oculocutaneous albinism in the environment; and (3) the person with oculocutaneous albinism maintaining his or her integrity

Table 5.2 depicts the relationship between steps of the model, set objectives and proposed key strategies for operationalisation of the model in practice.

Table 5.2 Relationship between steps of the model, objectives and key strategies for operationalisation of the model

Step of the model	Key Objectives	Key strategies
Step 1: Health professional *enhancing self-awareness* of the person with oculocutaneous albinism	*Assess self-image *Enhance self-esteem	*Enhance self-evaluation * Enhance self-knowledge
Step 2: Health professional *supporting adaptation* of the person with oculocutaneous albinism in the environment	*Development and growth *Awareness creation and develop sense of belonging	*Provide an enabling environment *Provide an enabling environment
Step 3: Person with oculocutaneous albinism maintaining *integrity*.	*Maintaining maximum potential *Self-efficacy and cognitive skills	*Develop a checklist *Build confidence to maintain self-efficacy and use cognitive skills

5.3.1 How to enhance self-awareness of the person with oculocutaneous albinism

Step one of the model is about enhancing self-awareness of the person with oculocutaneous albinism through self-evaluation, so as to improve the individual's self-esteem. Self-awareness is regarded as the keystone to emotional intelligence, and this step should thus be carried out with diligence. Two objectives were thus identified, namely to asses the person's self-image, and to enhance the person's self-esteem.

5.3.1.1 Assessment of self-image

The first step is to support the person with oculocutaneous albinism to share his or her life experience. This is essential to assess the person's self-image, and requires certain strategies and actions.

❑ ***Strategies and actions to asses self-image***

Assessment in the context of this model is a process of getting the facts about the person with oculocutaneous albinism to determine the person's self-image. Assessment should thus be done to identify the need to use the model, and is a vital process of operationalisation of the model in practice. During assessment, the needs and interests of the person with oculocutaneous albinism should be identified. The health professional should provide counselling to the person, and make a diagnosis of the person's self-image by observing his or her verbal and non-verbal responses.

According to Abdellah's nursing theory (Abdellah and Levine 1986:23), the health professional should be alert to the following signs of low self-image of the person with oculocutaneous albinism:

⚔ Lack of awareness of self as an individual with varying physical, emotional, and developmental needs.

⚔ Lack of accepting the optimum possible goals in the light of physical and emotional limitation.

⚲ Lack of identifying and accepting positive and negative expressions, feelings and reactions.

The purpose of assessment is to obtain a total picture of the person with oculocutaneous albinism, with an intention of identifying the need of the person. The health professional should therefore provide an enabling environment (Wilkins 2001:76). In this regard, the health professional needs critical thinking skill and knowledge of oculocutaneous albinism as a congenital disorder. The relevant skill and knowledge will help the health professional to decide which assessments to make, how much information is needed to make a diagnosis of the person's self-image, and recognise when information is missing (Wilkins 2001:78).

Furthermore, the assessment phase should be comprehensive, and the health professional should do detailed needs assessment of the person. The needs assessment, substantive needs and self-evaluation outcomes will influence the person's self-image.

- *Needs assessment*

Needs assessment of the person with oculocutaneous albinism should include learning about the culture, health beliefs, health behaviour, experience, skills and attitudes. These aspects influence the self-image of the person (Dennill et al 1999:154). George (2002:28) advocates a holistic approach during the assessment by considering the person's biological, psychological, social, and spiritual dimensions.

The biographical data, such as the person's age, will suggest his or her developmental stage. The person's health history, including family members, will be helpful to determine whether there are other family members with oculocutaneous albinism. The health professional should determine how the presence of another member with albinism influences the person's self-image.

- *Substantive needs*

It is essential during the assessment phase for the health professional to identify the substantive needs of the person with oculocutaneous albinism (Jordaan & Jordaan 1998:590). Substantive needs are the physical, socio-affective and self-esteem needs (Jordaan & Jordaan 1998:590 citing Davies 1991). Physical needs include everything associated with biological survival such as hunger, sleep, rest and activity.

Once the physical needs are met, the person becomes aware of the socio-affective need, which is a need to belong somewhere and to someone, and to give and receive love. Self-esteem need refers to the desire to cherish fairly consistent positive feelings towards self. Awareness of these needs therefore enhances the person's self-identity and sense of belonging, and consequently influences his or her self-image and the person should be able to do self-evaluation.

- **Strategies to enhance self-evaluation**

Strategies to enhance self-evaluation entail evaluating the person's physical, psychological and social attributes. The health professional should determine how the person feels about his or her appearance, which is the physical attribute. The abilities and talents of the person with albinism determine the psychological attribute. The social attributes pertain to the person's relationship with other family members and the community. The health professional should therefore enhance the person's self-evaluation to make a diagnosis of the individual's self-image.

To facilitate the process of self-evaluation, the health professional should ask the following questions:

* Does the person with oculocutaneous albinism regard self-acceptance as fundamental?
* Is the person with oculocutaneous albinism striving towards self-direction, and desire to use his or her capabilities to fulfil responsibilities?

*Is the person with oculocutaneous albinism given equal opportunity to maximise his or her potential with the necessary societal help?

* What are the internal and external factors that affect the person with oculocutaneous albinism's life?

5.3.1.2 Enhance self-esteem

To enable the person with oculocutaneous albinism to realise his or her self-worthiness requires certain strategies and actions.

❑ **Strategies and actions to enhance self-esteem of the person**

Once the person's self-evaluation is enhanced and a low self-image diagnosed, the following strategies are proposed to enhance self-esteem of the person with oculocutaneous:

⚔ The health professional should enhance self-esteem and self-knowledge by providing the necessary knowledge of oculocutaneous albinism to the person. This should include the pattern of inheritance, skin and eye care as well as the available resources on oculocutaneous albinism. Information should be provided on available support groups, and importance of using appropriate sun protective moisturisers. Self-knowledge is therefore essential to foster the person's self-esteem.

⚔ The person with oculocutaneous albinism should keep in mind that the concept of human "worth" is relative, and its meaning depends on who defines it. According to Prinsloo (1995:10), there are no absolute criteria according to which a particular human being can be defined as worthy or worthless. No matter what a person does, there is bound to be someone somewhere who will think that the person is not particularly worthy. In this regard, Prinsloo's (1995:10) emphasis on self-worthiness is as follows "You have to realise that the only person's opinion that really counts is your own."

- ⚹ Prinsloo (1995:10) maintains that the person should do things that he or she finds interesting and stimulating. The person should also confront daily problems and derive satisfaction from figuring out how to solve the problems.

- ⚹ According to Prinsloo (1995:10), the person with oculocutaneous albinism should fight back against invalid negative thoughts about him- or herself. The person should train him- or herself to recognise self-critical thoughts as they go through the mind, and should practise how to deal with these thoughts. Most importantly, the person should stop making self-torturing mental comparisons with other people.

- ⚹ Prinsloo (1995:11) emphasises that the person with ocolucutaneous albinism should not give power to other people by constantly seeking approval from them; for example "Do you think my boyfriend loves me or is just exploiting me?" If the person values other people's opinions more than his or her own, this automatically implies that those people are more competent in judging him or her. Other people will accept the power the person with oculocutaneous albinism gives them to be superior, and in this way the person becomes less able to build up his or her own self-esteem.

- ⚹ Prinsloo (1995:11) concludes that for the person with oculocutaneous albinism to meet the challenges of coping with problems, the person should think positively. Prinsloo (1995:11) emphasises that positive thinking can be a source of happiness and will promote personal development and growth.

5.3.2 How to support the person's adaptation in the environment

Two objectives were identified to support adaptation of the person in the environment in view of development and growth, and awareness creation.

5.3.2.1 Development and growth

To create an enabling environment that will promote self-development and growth of the person with oculocutaneous albinism.

❑ *Strategies to promote self-development and growth*

Once an individual's self-esteem is improved, the health professional should promote self-development and growth of the person with oculocutaneous albinism. According to Clinebell (1992:50), the health professional as the growth counsellor should actively encourage people with oculocutaneous albinism to set goals, develop workable plans for their own growth, and make contracts with themselves to implement those plans.

Clinebell (1992:50) is of the opinion that in the process of growth, there is a fundamental respect for the ways each individual wants to change and grow. Clinebell (1992:50) concludes that the choices about how one changes, the direction to take, and the speed, are the right and responsibility of the person. The ultimate goal of promoting the person's development and growth is to liberate his or her potential in all dimensions of his or her life, beginning with inner liberation. The environment should thus be non-threatening and non-judgemental so that the person develop a sense of belonging, which may be improved by creating awareness on oculocutaneous albinism can.

5.3.2.2 Awareness creation and sense of belonging

To improve a sense of belonging of the person by raising awareness on oculocutaneous albinism requires appropriate strategies.

❑ *Strategies to create awareness and improve a sense of belonging*

The health professional should involve the person and his or her family in creating awareness of oculocutaneous albinism. This, in turn, will enhance the person's self-image and sense of belonging. Accessible educational material should be developed that will educate the families and community on oculocutaneous albinism.

The person with oculocutaneous albinism should also be encouraged to participate in other community projects, such as sports, as this will enhance their sense of belonging. Parents should not be overprotective of their children with

oculocutaneous albinism, so that they do not feel different from other children, and thus have a sense of belonging in the family. Another strategy to improve a sense of belonging and increase awareness of oculocutaneous albinism include joining support groups to motivate and support each other.

5.3.3 How to determine the person's integrity

It is essential to achieve the following two objectives to support the integrity of the person with oculocutaneous albinism, which pertains to maintaining maximum potential and self-efficacy.

5.3.3.1 *Maintaining maximum potential*

To support the person with oculocutaneous albinism to use maximum potential so that the person feels valued and accepted by the family and community, certain strategies are needed.

❑ ***Strategies to support personal integrity***

Personal integrity is the cumulative result of the health professional's intervention to enhance the person with oculocutaneous albinism's self-concept.

A checklist should be developed to determine and support the person's personal integrity. The checklist should include the ten proposed questions to the person with regard to
- honest personal feelings on oculocutaneous albinism;
- whether the person has made peace with the passing years of experience;
- whether the person presently enjoys precious moments of his or her life;
- recognising cumulative result of development and growth throughout the life cycle;
- making decisions for him- or herself without influence from outside;
- being in control of his or her own life;
- connection of his or her own identity to the family and communication (social integrity);
- not becoming an easy prey to discrimination;

- accepting and dealing with his or her own strength and limitations; and

- inner motivation to perform a desired task in the community.

Integrity also entails maintaining self-efficacy and using cognitive skills to solve own problems.

5.3.3.2   Self-efficacy and cognitive skills

Relevant strategies are proposed to support the person with oculocutaneous albinism to maintain self-efficacy and use cognitive skills.

❑   **Strategies to improve self-efficacy and cognitive skill of the person**

The health professional should

- Build on the person's previous experience of oculocutaneous albinism to improve that person's self-efficacy.

- Create positive expectations for success, provide opportunities for the person with oculocutaneous albinism to successfully attain goals, and offer him or her control over his or her experience and growth.

- Provide the person with oculocutaneous albinism opportunities to use newly acquired cognitive skills to solve his or her own problems and make decisions (Driscoll 1994:34).

In addition to proposing the guidelines to operationalise the model in practice, it was fundamental to analyse and evaluate the model.

5.4   ANALYSIS AND EVALUATION OF THE MODEL

The purpose of subjecting the model to rigorous analysis and evaluation was to understand how well the model related to practice, research, and educational activities (Chinn & Kramer 1999:100). The researcher used various guidelines to analyse the internal structure and external validity of the model (Chinn & Kramer's 1999:100-119; McKenna 1997:223-230; Pearson et al 2005:226-228).

Most importantly, the researcher used Chinn and Kramer's (1999:100) questions for critical reflection of theory as a frame of reference to evaluate the model: (1) How clear is the model? (2) How simple is the model? (3) How general is the model? (4) How accessible is the model? and (5) How important and understandable is the model? The promoter also guided the researcher to develop a model that is easily understandable in practice and research. Furthermore, four experts in the field of theory development and qualitative research evaluated the model using Chinn and Kramer's (1999:100) critical reflection of theory. Three of them have doctorate degrees in nursing, and one in social welfare.

5.4.1 Evaluation of internal structure of the model

Evaluating the internal structure of the model entails description of the model itself. It also involves scrutiny of what the researcher has written to explain the perception of reality of the model in nursing (Pearson et al 2005:226). Five broad criteria, namely clarity, simplicity, generality, empirical applicability and logical development were used to analyse and evaluate the model internally (Chinn & Kramer 1999:101; McKenna 1997:226; Pearson et al 2005:226). These criteria therefore answer the question: **"How clear is the model?"**

5.4.1.1 Clarity

Clarity refers to whether the model is easy to understand, and ideas are consistently conceptualised (Chinn & Kramer 1999:101; Pearson et al 2005:226). The researcher determined the clarity of the model through semantic clarity, structural clarity, and semantic consistency (Chinn & Kramer 1999:101).

- **Semantic clarity** relates to the definitions of concepts in the model. In this model, the key concepts "self-concept" and "enhancement" were examined and defined in detail in such a way that relationships between the attributes of these concepts make sense, and are easy to understand. Chinn and Kramer (1999:101) also warn that if there are no definitions, or if only a few concepts are defined, clarity is limited. The researcher thus endeavoured to define all the related concepts of the model.

One of the experts who evaluated the model commented as follows on the semantic clarity of this model: "Self-concept and enhancement are clear enough and promote an understanding."

- **Structural clarity** refers to understanding the connections and reasoning within the model (Chinn & Kramer 1999:104). In this regard, Meleis (1991:158) is of the opinion that clarity of theories and models is enhanced by a visual representation of the theory or model. Figure 5.1 therefore graphically illustrated the model. The connections of the three steps of the model and structures are easy to understand, and described in detail. The graphic representation and linkages of concepts in Figure 5.1 are clear.

- **Semantic consistency** means that the concepts are used in ways that are consistent with the definitions. Chinn and Kramer (1999:103) warn that a theory that is inconsistently presented leads to confusion. In this model, definitions of the concepts were examined for consistency with each other, and in relation to the assumptions of the model. This in turn ensured the simplicity of the model.

5.4.1.2 Simplicity

This section answers the question: "**How simple is the model?**"

The model for the enhancement of people with oculocutaneous albinism is simplified in such a way that it can be used in a wide range of possible experiences in nursing practice. Comments from the experts of theory building confirmed that the model is simple and generalisable.

Torres (1990:13) emphasises that a theory should be relatively simple yet generalisable.

5.4.1.3 Generality

This section answers the question: **"How general is the model?"**

The generality of a theory refers to its breadth of scope and purpose. Scope addresses how broad the model is. The scope of the model addressed the question: "To what extent does the model accommodate all speciality areas of nursing practice?" (Chinn & Kramer 1999:106).

The model was developed and described so that it was adequate to achieve its main objective of enhancing the self-concept of people in general (Pearson et al 2005:226). Most importantly, the model was developed to be sufficiently broad to encompass other practical situations of congenital disorders or disabilities.

To confirm the generality of the model, one of the experts who evaluated the model commented as follows: "The model is broad enough to be used in a variety of situations in nursing. It can be useful in enhancing the self-concept of people in general." Generality meant that the model is easy to access and empirical applicability should be determined.

5.4.1.4   Empirical applicability

This section answers the question: "**How accessible is the model?"**

Empirical applicability addresses the accessibility of the theory, and determines the extent to which empiric indicators can be identified for concepts within the theory (Chinn & Kramer 1999:106). By identifying and defining the attributes of "self-concept" and "enhancement" as the key concepts that emerged from the findings, empirical accessibility of the model was achieved. In addition, a theoretical definition of "enhancement self-concept" as the key building block of the model was provided (see chapter 4).

Chinn and Kramer (1999:107) emphasise that if the concepts are defined, how they are to be empirically accessed is clearer. The concepts of the model are therefore empirically accessible in the clinical area to emphasise the importance of the model.

5.4.1.5 Logical development and importance of the model

This section answers the question: "**How important is the model?**"

The importance of this model pertains to its significance in nursing practice. In this regard, the model is valuable for creating a desired future in nursing practice to improve self-image of people with oculocutaneous albinism (Chinn & Kramer 1999:108).

The assumptions of this model, namely Neuman's systems model, Wiedenbach's prescriptive theory, and the self-empowerment assumptions, confirm that the model is important to the nursing practice (Chinn & Kramer 1999:108; Pearson et al 2003:226).

Internal evaluation of the model confirms that the model was developed in a logical way, was well thought of and constructed (see chapters 3 and 4). The model is therefore at an advanced stage of development and should thus be externally evaluated.

5.4.2 External validity of the model

The external validity of the model discusses and answers the question:
"**How important and understandable is the model?**"

The external validity of the model is concerned with the relationship of the model with the "real world", in view of the potential usefulness of the model in nursing practice and research (Meleis 1991:15; Pearson et al 2005:227).

5.4.2.1 Potential usefulness of the model in nursing practice

In terms of Pearson et al's (2005:227) criteria, the potential usefulness of the model in nursing practice took into consideration cost-effectiveness, relevance of the model to nursing practice today, and making the model understandable to health professionals.

- *Cost-effectiveness of the model*

The model is cost effective because there are no extra equipments to purchase when use the model. Nevertheless, quality time spent with the person with oculocutaneous albinism is essential to have an outcome of a person with a positive self-image, who has faith in him- or herself.

- *Relevance of the model for nursing practice today*

The model is relevant to nursing practice today as it is closely related to the enabling and self-empowerment concepts. The concept of enabling is based on the idea that in order to realise their freedom and assume greater responsibility for their health, individuals may require help in the form of know-how, resources and power to assume greater control (Dennill et al 1999:123). The model therefore enables the person with oculocutaneous albinism to be in greater control of his or her life and maintain integrity.

Stanhope and Lancaster (2004:612) point out that the outcomes of empowerment are positive self-esteem, the ability to set and achieve goals, a sense of control over life and changing processes, and a sense of hope for the future.

Furthermore, there is a link between the nursing process and the steps of the model. In this regard, the nursing process is described as a scientific approach by which the practice of nursing is applied in an orderly, creative and systematic manner to identify patient strengths, potential health problems and promote wellness (Wilkins 2001:9). The model is also constructed in an orderly and systematic manner to guide the health professional to assess the person's self-

image, promote the person's self-development and growth, and thus use intervention strategies to enhance that person's self-concept.

- ***Making the model understandable to the health professional***

The model was developed in such a way that it is easy for the health professional to understand the role of the health professional as the agent, the role of the person with oculocutaneous albinism as the recipient, procedures to enhance the person's self-concept, the dynamics involved, and finally the outcome of the interventions (see chapter 4 on enhancement of the self-concept).

5.4.2.2 Potential usefulness of the model in research

The potential usefulness of the model was determined by conducting a preliminary literature review to verify the need to undertake the study. In addition, the researcher determined how replicable the study is, whether the findings could be generalised, the appropriateness of the research design used, and whether the results were relevant in other fields (see chapters 2, 3 and 4). Pearson et al (2005:227) emphasise that if the theory remains untested, its usefulness is questioned.

In recognising the usefulness of this model, one of the experts who evaluated the model positively commented as follows: "The model comes at time when the nation of South Africa is fighting discrimination, and is trying to promote equity in all its programmes. The model will therefore become useful for health professionals and for nurses in particular. Only yesterday (Sunday, 29 April 2007), the phenomenon of albinism was highlighted by one newspaper. This means that people are becoming aware of the difficulties facing people with albinism, especially in the workplace. The model promises to be a useful tool that can be used by practitioners in the Employee Assistance Programmes. The model is cost effective, relevant and understandable."

5.5 CONCLUSION

This chapter described the structure and process of the model, and the three advancing steps of the model. Visual representation of the model enhanced structural clarity and connections of the concepts of the model. The researcher also described guidelines for the operationalisation of the model in practice. Most importantly, the model was subjected to rigorous internal and external evaluation to facilitate the potential use of the model in nursing practice and research.

Chapter 6 concludes the study, identifies its limitations, and makes recommendations for nursing practice, further research, nursing education and policymaking.

CHAPTER 6

CONCLUSIONS, LIMITATIONS AND RECOMMENDATIONS

6.1   INTRODUCTION

The previous chapters discussed in detail the theory-generating qualitative approach undertaken in this study, and a model to enhance the self-concept of people with oculocutaneous albinism was developed. Chapter 5 specifically described the structure and process of the model as well as practical guidelines for its operationalisation in practice.

Chapter 6 therefore draws conclusions on whether the purpose and objectives of the study were achieved. It also identifies the limitations of the study, and makes recommendations for the utilisation of the model in nursing practice, research, nursing education and policy making.

6.2 CONCLUSIONS

The main purpose of the study was to develop a model for the enhancement of self-concept of people with oculocutaneous albinism. This purpose was accomplished by using a theory-generative research design. The study was therefore conducted in four steps of theory-generation, namely concept analysis; construction of theoretical relationships; description of the model, and guidelines to operationalise the model.

The researcher used reasoning strategies of bracketing, intuiting, inductive and deductive analysis and synthesis to generate knowledge on oculocutaneous albinism, and to ultimately develop the model from the concepts deducted from the research findings. Ethical rigor was maintained throughout the research process, and trustworthiness of the findings established. These fundamental aspects are discussed in detail in chapter 2.

In order to develop the model, the researcher worked in two phases of theory-generation objectives: to identify the main concepts of the model from the participants' life-experience, and to describe the model structure and process.

6.2.1 Identification of the main concepts

The first objective was to identify the main concepts of the model from the participants' life-experience. This objective was achieved by using a qualitative, exploratory, descriptive and contextual research design.

6.2.1.1 Theory-generative research design and methodology

The theory-generative research design is qualitative, exploratory, descriptive and contextual in nature. Qualitative data was obtained by using in-depth phenomenological interviews to explore and describe the participants' life-experience. A purposive sample of fifteen participants, consisting of thirteen females and two males with oculocutaneous albinism, aged between 18 and 48, participated in the study. Interviews were conducted until data saturation.

Data from the interviews were subsequently analysed using Colaizzi's qualitative data analysis method (see chapter 2 for research design and methodology).

6.2.1.2 Findings and literature control

The findings were categorised into three main themes, namely participants' perception of the self; participants' experience of the external environment, and participants' expression of need for development and growth. A literature review validated the research findings (see chapter 3).

The focus of the first theme on perception of the self was an experience echoed by the participants, recognising that they 'themselves' play an important role in their own lives. The main concepts that emerged from the first theme were self-concept, self-actualisation, self-esteem and self-worthiness.

The literature review confirmed the importance of the self-concept by indicating that the "self" constitutes people's awareness of their individual existence. In the self-actualisation process, people strive to discover their true potential. The literature also confirmed that self-esteem is based on the fact that people need to feel a sense of their own value, and that they are respected and appreciated by other people. Most importantly, it emerged that the environment plays a fundamental role in the lives of the participants, and they shared their perception of the self in relation to the family and community.

The second theme of the findings was the participants' experience with the external environment. Here, it emerged that their interaction with the external environment influenced their self-image, sense of belonging, and feeling of being stigmatised on the basis of myths and superstitions. According to the literature reviewed, the external environment may be one of rejection, harshness, warmth, acceptance or uncertainty. It was evident from the findings that the external environment influences the development and growth of people with oculocutaneous albinism.

The third theme, then, was the participants' expression of a need for development and growth. It emerged that the external environment, consisting of the family, community, and various stakeholders, including NGOs, government departments, and the private sector, influences the development and growth process of the person with oculocutaneous albinism. The literature review confirmed the fundamental role of the external environment to improve the self-image of the person.

The key concepts of "**self-concept**" and "**enhancement**" were thus deducted from the findings. As the researcher interacted with the main themes of the findings, it became clear that there was a need to enhance the self-concept of people with oculocutaneous albinism, hence a *"A model for enhancement of self-concept of people with albinism".*

6.2.1.3 Concept analysis

Chapter 4 discusses the concept analysis of "self-concept" and "enhancement" as the main concepts deducted from the findings. Valid sources therefore created the conceptual meaning of **"enhancement of self-concept**" as the main building block of the model. The initial valid meaning of the key concept emerged from the interviews. The researcher then consulted dictionaries, thesaurus, existing theories, and professional and classical literature to identify the characteristic attributes of "enhancement of self-concept". Furthermore, model and borderline cases were used to gain insight into this concept.

6.2.1.4 Development of the model

After concept analysis of the main concepts, and in order to objectively develop the model from the participants' experience, the three main themes of the findings were converted into the three main steps of the model (see chapter 5). The main steps of the model are thus: (1) Health professional *enhancing self-awareness* of the person with oculocutaneous albinism; (2) Health professional *supporting adaptation process* of the person with oculocutaneous albinism in the environment; and (3) Person with oculocutaneous albinism maintaining *integrity*.

6.2.2   Description of the model structure and process

The second objective was to describe the structure and process of the model, and this was achieved (see chapters 4 and 5) by:

- Classifying the concepts of the model in relation to recipient, agent, context, dynamics, procedure and terminus according to Dickoff et al's (1968:422) survey list.
- Describing the structure and process of the model for the enhancement of self-concept in view of purpose, assumptions, context, theoretical definition, and relationship statements using Chinn and Kramer's (1995:107) guidelines.
- Formulating and describing guidelines for operationalisation of the model in practice.

6.2.2.1 Classifying the concepts of the model

Classification of the main concepts was done to facilitate future prescription to use the model. The researcher used Dickoff et al's (1968:431) survey list to classify the concepts of the model into agents, recipients, procedures, dynamics and terminus.

In this model, the health professional is regarded as the agent to enhance the self-concept of the person with oculcutaneous albinism. The person with oculcutaneous albinism is the recipient who benefits from the action of the health professional.

The procedure refers to the three advancing steps to enhance the self-concept, namely to enhance self-awareness, to support the person to adapt in the environment, and the person with oculocutaneous albinism maintaining his or her integrity. The dynamics are the energy source for the activity. Terminus is the outcome of the model, and should be a person with oculocutaneous albinism with enhanced self-concept as evidenced by enhanced self-awareness, adaptation in the environment and integrity.

6.2.2.2 Structure and process of the model

Chinn and Kramer's (1995:107-119) strategies for theory description were used to describe the structure and process of the model. These strategies described the purpose, assumptions, context, theoretical definition and relationship statements of the model. A graphic visual representation of the model depicting the structure and process of the model was created (see chapter 5).

The main purpose of the model is to provide a frame of reference for health professionals to enhance the self-concept people with oculocutaneous albinism. The assumptions of the model were thus related to the process of enhancing the self-concept of people. The assumptions were based on Neuman's system model, Wiedenbach's prescriptive nursing theory, and the rehabilitation self-empowerment assumptions.

Neuman's systems model depicts an open system in which people with oculocutaneous albinism and their environment are in constant interaction.

According to Wiedenbach's nursing theory, self-awareness and self-acceptance are essential for the sense of integrity and self-worthiness of people with oculocutaneous albinism. The self-empowerment assumption emphasises that individuals with oculocutaneous albinism strive to grow and change in a positive direction (Dell-Orto & Marinelli 1995:258; Neuman 1995:12; Wiedenbach 1970:1058).

The context is the situation in which the research takes place, and comprises human and environment (George 2002:185). The context of the model comprised people with oculocutaneous albinism in urban and semi-urban areas of the Gauteng province, South Africa.

Deductive reasoning was used to establish interrelationships between the concepts of the model. Relationship statements from the model were also established, which also guided the operationalisation of the model in practice.

6.2.2.3 Guidelines for operationalisation of the model in practice

The guidelines for the operationalisation of the model in practice were formulated and described by discussing the objectives of each step of the model. Strategies for achieving each stated objective were then suggested.

Most importantly, the model was subjected to rigorous internal and external evaluation using various strategies for theory evaluation. The internal evaluation criteria used were clarity, simplicity, generality, empirical applicability and logical development. The researcher specifically used Chinn and Kramer's (1999:100-109) critical reflection criteria to evaluate the model internally. The external evaluation of the model determined the potential usefulness of the model in nursing practice and research.

Sections 6.2.1 and 6.2.2 discussed above, conclude that the purpose and objectives of the study were achieved (see chapters 1 and 2).

6.3   LIMITATIONS OF THE STUDY

The researcher identified two methodological limitations of the study. Identifying the limitations of the study was essential for meaningful debate on the findings (Polit & Beck 2004:101).

The first limitation relates to the population of the study. The population from which the participants were selected was narrow. A purposive sample of fifteen participants, consisting of thirteen females and two males with oculocutaneous albinism, aged between 18 and 48, participated in the study. A possible question is what effect the inclusion of a wider population from people with other congenital disorders might make.

Secondly, the sample size and qualitative approach used could limit the generalisation of the study. Data was collected from the fifteen participants with oculocutaneous albinism using in-depth phenomenological interviews. However, the critical social theory emphasises that it is essential to raise concern of a group of people who have been disadvantaged, oppressed or marginalized (see chapter 1).

Therefore, interviewing a small group of people with oculocutaneous albinism was seen as a way of giving them a voice to share their everyday experience. The present findings and literature control confirmed that people with oculocutaneous albinism have been marginalised and discriminated. Most importantly, the number of participants in this study is in the scope of qualitative research, and the principle of data saturation was achieved as described in Chapter 2.

The researcher took cognisance of these limitations when interpreting the findings.

6.4   RECOMMENDATIONS

In the light of the limitations, the researcher makes the following recommendations for clinical nursing practice, further research, nursing education and policy making.

6.4.1 Nursing practice

In view of the purpose of this model to provide a frame of reference for health professionals to enhance the self-concept people with oculocutaneous albinism, the researcher recommends the use of the nursing process to utilise the model in the clinical nursing practice.

In this regard, George (2002:20) and Wilkins (2001:8-9) describe the nursing process as a scientific approach by which the practice of nursing is applied in an orderly, creative and systematic manner to identify the patient's strengths, potential health problems and promote wellness. George (2002:18) further supports the use of the nursing process to apply this model by emphasising that nurses should understand, and apply appropriate concepts and theories from nursing so as to provide the rationale for decision-making, judgements, interpersonal relationships, and actions.

The nursing process comprises five phases, namely assessment, diagnosis, planning, implementation and evaluation (Wilkins 2001:8). The nursing process will thus be helpful to enhance the self-concept of a person with oculocutaneous albinism in a creative and systematic manner. The person's strength will also be identified during this process. The model can thus be applied as follows, using the five phases of the nursing process:

- **Assessment:** The health professional should assess, collect and organise information on the person's self-concept. Re-assessment is also done during the planning and evaluation phases.

- **Diagnosis:** The health professional should make a diagnosis of the self-concept, and identify the person's problems and strengths.

- **Planning**

   *Planning outcomes:** The health professional should choose the desired outcomes based on the assessment and diagnosis with the person.

   *Planning interventions:** The health professional should choose appropriate interventions strategies proposed in the steps of the model to
   ⋏ Enhance the person's self-awareness.
   ⋏ Support the person's adaptation in the external environment.
   ⋏ Determine the person's integrity.

- **Implementation:** The health professional should carry out the plan of action identified during the planning phase, and use the model accordingly.

- **Evaluation:** In this phase, the health professional should determine whether the actions in the implementation phase achieved the intended aim of enhancing person's self-concept. This phase therefore requires the health professional to be able to follow-up on the person, and re-assess that person's self-image.

6.4.2 Further research

It is evident from this study that several priority issues of oculocutaneous albinism and related topics could be pursued in future research. Priority future research is categorised according to the three main themes of the findings, in view of self-concept, environment and development and growth.

6.4.2.1 Self-concept

Further research should be undertaken on the following topics:

- Explore the life-world of people with oculocutaneous albinism in a rural area to determine whether the findings of this study would be replicated, and determine whether a similar self-concept model would be developed.

- Explore the life-world of people with other congenital disorders, for example, cleft-lip and palate or neural tube defects, to determine whether the findings of this study would be replicated and a similar self-concept model developed.

6.4.2.2 Effect of external environment

The following topics are recommended for further research:

- Explore the cause-effect relationships in society to determine what causes the community tendency to stigmatise and prejudice persons with oculocutaneous albinism and/or other congenital disorders.
- Factors in the external environment that influence the quality of life and survival rate of people with oculocutaneous albinism.

6.4.2.3 Development and growth

The following topics should be researched:

- A phenomenological study to determine the physiological, psychological, social-cultural, spiritual, and developmental needs of people with oculocutaneous albinism and/or other congenital disorders.
- Creating an enabling environment for NGOs to promote self-development and growth of people with oculocutaneous albinism and/or congenital disorders.

6.4.3 Nursing education

The training modules and curriculum should improve the skills and knowledge of nurses on congenital disorders, including oculocutaneous albinism. Although nurses receive basic training on congenital disorders during their training, there is evidence that this basic training does not improve their knowledge on congenital disorders. Most importantly, the tutors should be trained in human genetics. The trained tutors should be able to share the necessary information on congenital disorders with colleagues and nurses in training.

The National Department of Health in South Africa is currently collaborating with academic institutions to support provinces with the Medical Genetics Education Programme (MGEP) training. This training programme aims at improving the knowledge and skills of health professionals on congenital disorders. Primary health care nurses, tutors, and midwives are especially encouraged to attend this genetics training programme. Furthermore, the contents of the Medical Genetics Education Programme should be included in nursing curricula (Department of Health 2001:30-31).

In addition, workshops and in-service training sessions on congenital disorders should be organised. Telemedicine demonstrations should be used to improve the diagnostic and management skills of health professionals on oculocutaneous albinism and other congenital disorders.

Student nurses should be guided to engage in community projects to form support groups on oculocutaneous albinism. Most importantly, the holistic approach of caring for persons with oculocutaneous albinism in view of physical, psychological, socio-cultural, developmental and spiritual growth should be emphasised in nursing education. Therefore, areas of core competencies on oculocutaneous albinism that should be incorporated in nursing education curricula as follows:

- **Education** on oculocutaneous albinism at all levels of care, namely primary prevention to reduce risk factors of skin cancer; secondary prevention to regain optimal system; and tertiary prevention to maintain wellness and protect the person with oculocutaneous albinism.
- **Marketing strategies** by encouraging the use of educational material, local radio and local press for wider publicity on oculocutaneous albinism.
- **Facilitating and networking** by encouraging people with oculocutaneous albinism to share their experience and skills with health professionals.

The inclusion of oculocutaneous albinism and/or congenital disorders in nursing education curricula should influence policy and practice.

6.4.4 Policy making

A policy is a course or principle of action, which provides guidelines to role-players to make decisions and implement a strategy (Dennill et al 1999:174).

The recommendations for policy therefore emanate mostly from the findings (see chapter 3).

It was evident from the findings that various government departments play an important role in the lives of persons with oculocutaneous albinism. Most importantly, the participants referred to the Departments of Health and Education. It is therefore essential that these Departments organise workshops to orientate health professionals on their policies pertaining to oculocutaneous albinism and/or other congenital disorders. People with oculocutaneous albinism and other congenital disorders should be involved in the development and implementation of these policies. This, in turn, would enhance their self-esteem.

The role of the private sector in the lives of employees with oculocutaneous albinism should not be underestimated. The advanced technology in the private sector is an advantage for health professionals to access information on oculocutaneous albinism and other congenital disorders from the Internet and other forms of technology. Policies on oculocutaneous albinism and other congenital disorders in the private sector should therefore be available and shared with employees.

The media has been identified as one of the main communication strategies to raise awareness of oculocutaneous albinism. Society uses the media for various reasons, and the media should therefore develop policies to deal with issues of concern in the community. It is evident that the media is an important tool to improve the knowledge of the community on oculocutaneous albinism and other congenital disorders, and most importantly to eradicate myths and superstitions.

6.5 THE RESEARCHER'S EXPERIENCE

The researcher experienced the study as a personal and professional self-development and growth journey. The study enriched the researcher's knowledge and understanding of "enhancement of self-concept". In addition, on 21 January 2007, a motivational speaker presented a topic entitled "self-concept layer" on the Motsweding Frequency Modulation (FM), a national radio station. The researcher was encouraged by the presentation, which confirmed the significance of this study.

Data was collected between January and February 2006. The researcher found transcribing the interviews, deducting the key concepts from the findings, developing attributes of the key concepts, developing the model, and drawing a graphic presentation of the model a strenuous exercise that demanded sufficient time and concentration. Furthermore, the researcher initially found the development of a model daunting, which at times caused her considerable anxiety. Nevertheless, the expert guidance of the promoters was very helpful to finally develop the model.

Coupled with her responsibilities as a programme manager of the Sub-directorate: Human Genetics at the National Department of Health, the researcher frequently felt overwhelmed. The post entails attending national and international conferences, and lot of travel around South Africa. The family responsibilities could also not be ignored. However, with the promoter and co-promoter's academic support, patience and guidance, the researcher completed the study.

6.6 CONCLUSION

The study explored the life-experience of people with oculocutaneous albinism, and developed a model for the enhancement of their self-concept. This chapter concluded the study, identified its limitations, made recommendations for clinical practice, research, nursing education, and policy making, and the researcher shared her experience in the present research.

In an essay in the 2005 National Schools Essay Competition on Albinism, the winner, a fifteen-year-old Grade 10 learner from Mpumalanga province, South Africa, succinctly captured the need to enhance the self-concept of people with oculocutaneous albinism:

> *Albinism can have a positive bearing if all people motivate persons with albinism. Help us to fight so that we can be understood and appreciated. We too, like any other children, teenagers and adults can go places, attain our dreams and aspirations and excel at that which we are good at!*

BIBLIOGRAPHY

Abdellah, FG & Levine, E. 1986. *Better patient care through nursing research.* 3rd edition. New York: Macmillan.

Aggleton, P & Chalmers, H. 2000. *Nursing models and nursing practice.* 2nd edition. London: Creative Print & Design.

Barrable, K. 1995. You Creator. *Woman's Forum.* Pretoria: Daan Retief.

Beckman, PJ, Barnwell, D, Horn, E, Hanson, MJ, Gutierrez, S & Lieber, J. 1998. Communities, families and inclusion. *Early Childhood Research Quarterly* 13(1): 123-150.

Bhagwanjee, AM & Sterwart R,1999. Disability research in South Africa, vision and imperatives for national co-ordinated approach. *South African Journal of Occupational Therapy* 29(1):15-17.

Blackwell's Nursing Dictionary. 2005. 2nd edition. London: Blackwell.

Bless, C & Higson-Smith, C. 1995. *Fundamentals of social research methods: an African perspective.* 2nd edition. Kenwyn: Juta.

Botes, A. 1995. The operationalisation of a research model in qualitative methodology. *Curationis* 1(1):4-9.

Burns, N & Grove, SK. 2001. *The practice of nursing research: conduct, critique, and utilization.* 4th edition. Philadelphia: Saunders.

Burris, S. 1999. Studying the legal management of HIV-related stigma. *American Behavioural Scientist* 42(7):1229-1231.

Chinn, PL & Kramer, MK. 1995. *Theory and nursing: a systematic approach.* 4th edition. St Louis: Mosby.

Chinn, PL & Kramer, MK. 1999. *Theory and nursing: integrated knowledge development.* 5th edition. St Louis: Mosby.

Christianson , AL & Modell, B. 2004. Medical genetics in developing countries. Annual *Reviews in Genomics and Human Genetics* (5):219-265.

Christianson , AL, Howson, P & Modell, B. 2006. *Global Report on Birth Defects: The hidden toll of dying and disabled children.* March of Dimes Birth Defects Foundation. New York: White Plains.

Clinebell, H. 1992. *Growth counselling:* h*ope-centered methods of actualising human wholeness.* Tennessee: Parthenon.

Collins English Dictionary. 2006. 2nd edition. Glasgow: HarperCollins.

Collins Paperback Thesaurus. 2004. 3rd edition. Glasgow: HarperCollins.

Collins Thesaurus. 2006. 2nd edition. Glasgow: HarperCollins.

Danna, C. 1998. Doomed by my white skin. *Drum Magazine*, 5 February: 100-101.

Darling, RS. 1979. *Families against society: a study of reactions to children with birth defects.* London: Sage.

Davies, J & Green, L. 1998. Mainstream teachers' attitudes to the mainstreaming of learners with special educational needs in primary classrooms: a Western Cape study. *South African Journal of Education* 18(2):97-100.

Dell-Orto, EA & Marinelli, RP (eds). 1995. *Encyclopedia for Disability and Rehabilitation.* London: Simon & Schuster Macmillan.

Delport, SD, Christianson, AL, van den Berg, HJS, Wolmarans, L & Gericke, GS. 1995. Congenital anomalies in black South African live-born neonates in an urban academic hospital. *South African Medical Journal* 85(1):11-15.

Dennill, K, King, L & Swanepoel, T. 1999. *Aspects of primary health care: community health care in Southern Africa.* 2nd edition. Halfway House: Thomson.

Department of Health. 2001. *Policy Guidelines for the Management and Prevention of Genetic Disorders, Birth Defects and Disabilities.* Pretoria: Government Printer.

Department of Health. 2005: *National Guidelines for the Care and Prevention of the Most Common Genetic Disorders, Birth defects and Disabilities.* Pretoria: Government Printer.

Dickoff, J, James, P & Wiedenbach, E. 1968. Theory in a practice discipline. Part 1: Practice oriented theory. *American Journal of Nursing* 7(5):415-435.

Dictionary of Public Health. 1996. *Health promotion and education: terms and concepts.* 4th edition. London: Sage.

Driscoll, MP. 1994. *Psychology of learning for instruction.* Boston: Allyn & Bacon.

DuBois, D. 2003. Self-esteem: early childhood. *Encyclopedia of Primary Prevention and Health Promotion.* New York: Plenum.

Erickson, HC, Tomlin, E & Swain, MA. 1983. *Modeling and role modeling: a theory and paradigm for nursing.* Lexington: Pine Press.

Ezeilo, B. 1989. Psychological aspects of albinism: an exploratory study with Nigerian albino subjects. *Social Science and Medicine* (9):1129-1131.

Fain, JA. 2004. *Reading, understanding and applying nursing research: a text and workbook.* 2nd edition. Philadelphia: Davis.

Fine, M & Asch, A. 1988. Disability beyond stigma: social interaction, discrimination and activism. *Journal of Social Issues 44 (1):3-21.*

Friedland, L. 2005. *Self: the essential health and life guide for today's woman.* Cape Town: Tafelberg.

Gaigher, RJ, Lund, PM & Makuya, E. 2002: A sociological study of children with albinism at a special school in the Limpopo province. *Curationis 1-11.*

Genetic science 'lives and functions' in families. 2003. *Genome News,* 6 June:2.

George, JB (ed). 1995. *Nursing theories: the base for professional nursing practice.* 4th edition. Englewood Cliffs, NJ: Prentice-Hall.

George, JB (ed). 2002. *Nursing theories: the base for professional nursing practice.* 5th edition. Englewood Cliffs, NJ: Prentice-Hall.

Goleman, D. 1997. *Emotional intelligence: why it can matter more than IQ.* London: Bloomsbury.

Holloway, I & Walker, J. 2000. *Getting a PhD in Health and Social Care.* London: Blackwell.

Jordaan, W & Jordaan, J. 1998. *People in context.* 3rd edition. First impression. Johannesburg: Heinemann.

Johnson, B & Christensen, L. 2000. *Educational research: quantitative and qualitative approaches.* Boston: Allyn & Bacon.

Johnson, DE. 1974. Development of a theory: A requisite for nursing as a profession. *Nursing Research* (23):372-377.

Johnson, DE. 1980. *The behavioural system model for nursing. Conceptual models for nursing practice.* New York: Appleton-Century-Crofts.

Kagore, F & Lund, MP. 1995. Oculoculataneous albinism among school children in Harare, Zimbabwe. *Journal of Medical Genetics* (32):859-861.

Kerr, R, Stevens, G, Manga, P, Salm, S, John, P, Haw, T & Ramsay, M. 2000. Identification of *P* gene mutation in individuals with oculocutaneous albinism in Sub-Saharan Africa. Research Article. *Human Mutation* (15):166-172.

King, IM. 1990. *A theory for nursing: systems, concepts and process*. Albany: Delmar.

Krech, D, Crutchfield, RS & Livson, N. *Elements of Psychology*. 1979. 3rd edition. New York: Random House.

Krefting, L. 1991. Rigor in qualitative research: the assessment of trustworthiness. *The American Journal of Occupational Therapy* 45(3):214-222.

Kromberg, JGR. 1992. Albinism in South Africa. IV. Attitudes and the death myth. *Journal of Birth Defects: Original Article Series* 28(1):159-166.

Kromberg, JGR, Castle, D, Zwane, EM & Jenkins, T. 1989. Albinism and skin cancer in Southern Africa. *Clinical Genetics* (36):43-52.

Kromberg, JGR & Jenkins, T. 1982. Prevalence of albinism in the South African population. *South African Medical Journal* (61):383-386.

Kromberg, JGR & Jenkins, T. 1984(a). Cultural influences on the perception of genetic disorders in the Black population of Southern Africa. *Journal of Biosoc Science (*6):147-157.

Kromberg, JGR & Jenkins, T. 1984(b). Albinism in the South African population. III Genetic counselling issues. *Journal of Biosoc Science* (6):99-108.

Kromberg, JGR & Jenkins, T. 1997. Specialist medicine. *Genetic services in South Africa and common ethical issues related to genetic information* X1X (5):54-56.

Kromberg, JGR, Zwane, M & Jenkins, T. 1987. The response of Black mothers to the birth of an albino infant. *American Journal of Disease of Children* (141):911-916.

Levine, ME. 1991. Conservation and integrity: Levine's conservation model*. In* n*ursing theories in practice* edited by ME Parker. New York: National League for Nursing.

Lincoln, YS & Guba, EG. 1985. *Naturalistic inquiry*. London: Sage.

LoBiondo-Wood, G & Haber, J. 2002. *Nursing Research: methods, critical appraisal, and utilization.* 5th edition. St Louis: Mosby.

Longman Dictionary of Contemporary English. 1997. New edition. London: Longman.

Lonsdale, S. 1990. *Women and disability*. Basingstoke: Macmillan.

Luand, PM, Puri, N, Durham-Pierre, D, King, RA & Brilliant, MH. 1997. Oculocutaneous albinism in an isolated Tonga community in Zimbabwe. *Journal of Medical Genetics* 34(9):733-735.

Manganyi, NC, Kromberg, JGR & Jenkins, T. 1974. Studies on albinism in the South African Population. I. Intellectual Maturity and Body Image Differentiation. *Journal of Biosocial Science* (6):107-112.

Mashele, M. 2003. *Critical thinking: a concept analysis.* Master's dissertation. MA (Cur). Pretoria: University of South Africa.

Maslow, AH. 1970. *Motivation and personality*. 2nd edition. New York: Harper & Row.

McKenna, H. 1997. *Nursing theories and models*. London: TJ Press.

Medical Research Council South Africa. 2002. Guidelines on Ethics for Medical Research: General Principles. 4th edition. Tygerberg: MRC.

Meleis, AL. 1991. *Theoretical nursing development and progress*. 2nd edition. Philadephia: Lippincott.

Moodley, L. 2003. *Facilitated problem definition: impact on families' perceptions of needs, resources and problems experienced in caring for young children with disabilities.* Doctoral thesis. Education. Pretoria: University of Pretoria.

Mouton, J. 2004. *How to succeed in your Master's and Doctoral studies: a South African guide and resource book.* 4th edition. Pretoria: Van Schaik.

Mweshi, MM & Mpofu, R. 2001. The perceptions of parents and caregivers on the causes of disabilities in children with cerebral palsy: a qualitative investigation. *South African Journal of Physiotherapy* 57(2):28-30.

National Schools Essay Competition on Albinism: 2005

Ndlovu, A. 2005. Stop the abuse of albinos. *Daily Sun*, 14 October:3.

Ngakane, L. 1998. The agonies of black souls with white skins: Mazibuko enhances self-esteem of people with albinism. *Woman 2 Woman City Press*, 4 January:13.

Nkabinde, ZP & Ngwenya TP.1996. Interviews with African parents of children with disabilities in South Africa. *Paidonomia* (19):1.

Neuman, B. 1995. The *Neuman Systems Model.* 3rd edition. Norwalk: Appleton & Lange.

Ojofeitimi, EO & Elegbe, I.1984. Mothers' belief as to causation of birth defects. *Journal of the Royal Society of Health* 104(3):116-118.

Okoro, A. 1975. Albinism in Nigeria. *British Journal of Dermatology* 92:485-492.

Online Mendelian Inheritance in Man (OMIM).2000. McKusick-Nathans Institute for Genetic Medicine, Johns Hopkins University (Baltimore, MD) & National Centre Biotechnology. Library of Medicine: http://www.ncbi.nih.gov.entrez/query.fcgi?db (last accessed 19 October 2005).

Oxford Textbook of Public Health. 2002. *The practice of public health.* 4th edition. New York: Oxford University Press.

Oxford Dictionary of Nursing. 2004. 4th edition. New York: Oxford University Press.

Oxford Advanced Learners Dictionary of Current English. 2004. 6th edition. New York: Oxford University Press.

Patton, MQ. 1990. *Qualitative evaluation and research methods.* 2nd edition. Newbury Park: Sage.

Pearson, A, Vaughan, B, & FitzGerald, M. 2005. *Nursing models for practice.* 3rd edition. New York: Butterworths-Heinemann.

Philpott, SC. 1995. *Amawoti: Responding to the needs and rights of people with disabilities.* Master's degree. Durban: University of Natal.

Polit, DF & Beck, CT. 2004. *Nursing research principles and methods.* 7th edition. Philadelphia: Lippincott Williams & Wilkins.

Polit, DF & Hungler, BP. 1999. *Nursing research: principles and methods.* Philadelphia: Lippincott.

Pooe-Monyemore, MBJ. 2003. *Integration of children with disabilities into the community: The role of the community nurse.* Master's dissertation. MA (Cur). Pretoria: University of South Africa.

Prinsloo, L. 1995. *Mind control for mood control.* Pretoria: Daan Retief.

Raliphada-Mulaudzi, F.M.1997. *Reproductive health rights of women in rural communities.* Master's dissertation. MA (Cur). Pretoria: University of South Africa.

Road map of concerns for disabled people in Zaire. 1999. *World Outlook* 47-48.

Robinson, NM & Noble, KD. 1995. *Social-emotional development and adjustment of gifted children.* University of Washington Center for Study of Capable Youth. Handbook of Special Education: Research and Practice. 1st edition. Volume 4. London: Wheatons.

Roussow, GJ. 1994. *Skilful thinking: an introduction to philosophical skills.* Pretoria: Human Sciences Research Council.

Rodgers, BL & Knafl, KA. 2000. *Concept development in nursing: foundations, techniques and applications.* 2nd edition. Philadelphia: Saunders.

Rogers, ME. 1992. Nursing science and the space age. *Nursing Science Quarterly* (5):27-34.

Roy, C. 1991. *Introduction to nursing: an adaptation model.* 2nd edition. Stamford, CT: Appleton & Lange.

Save the Children Research Report. 2001. South African Programme. *The role of stigma and discrimination in increasing the vulnerability of children and youth infected with and affected by HIV/AIDS.* Arcadia: Department of Nursing.

Sonnekus, IP. 1996. *Enhancing realistic academic self-actualisation: a psychoandragogical perspective.* Doctoral thesis. Pretoria: University of South Africa.

South Africa, Republic of. 1996. *Constitution of the Republic of South Africa Act, 108 of 1996.* Pretoria: Government Printers.

South Africa, Republic of. 2001. *Statistics census.* Pretoria: Government Printers. Statistics South Africa. South African National Census 2001. www.statssa.gov.za

Stanhope, M & Lancaster, J. 2004. *Community and public health nursing.* 5th edition. St Louis: Mosby.

Suinn, RM. 1979. *Fundamentals of Behavior Pathology.* 2nd edition. New York: Wiley.

Stone-McCown, K & McCormick, AH. 1999. *Self-science: emotional intelligence for children.* Instructional Design Theories and Models. Volume 11. A New Paradigm of Instructional Theory. London: Lawrence Erlbaum.

Streubert, HJ & Carpenter, DR. 1999. *Qualitative research in nursing.* 2nd edition. Philadelphia: Lippincott.

Swick, KJ. 1997. Working with families: a family-school approach for nurturing caring in young children. *Early Childhood Educational Journal* 25(2):151-153.

Treffinger, DJ, Callahan, CM, & Vaughn, VK. 1995. *Research on enrichment efforts in gifted education.* Handbook of special education. Research and practice. New York: Pergamom Press.

Torres, G. 1990. The place of concepts and theories within nursing. In nursing theories: the base for professional nursing practice. Edited by JB George*. Norwalk: Appleton & Lange.*

Urbani, U. 1992. *The problems surrounding children with genetic impairments.* Report No.0-019. Pretoria: Human Sciences Research Council.

Venter, PA, Christianson, AL, Hutamo, CM, Makhura, MP, & Gericke, GS. 1995. Congenital anomalies in rural black South African neonates: a silent epidemic? *South African Medical Journal* 85:15-20.

Walker, LO & Avant, KC. 1995. *Strategies for theory construction in nursing.* 3rd edition. Connecticut: Appleton & Lange.

Wilkinson, JM. 2001. *Nursing process and critical thinking.* 3rd edition. Englewood Cliffs, NJ: Prentice Hall.

Wilson, HS. 1989. *Research in nursing.* 2nd edition. Redwood City: Addison-Wesley.

Winship, WS. 2003. *Handbook of genetic and congenital syndromes.* 1st edition. Cape Town: Oxford University Press.

Wiedenbach. E. 1970. Nurse's wisdom in nursing theory. *American Journal of Nursing* (70):1057-1062.

World Bank. 2005. *Country Classification*.
http://www.worldbank.org/data/countryclass.html (last accessed 19 October 2005)

World Book Dictionary. 1997. New York: World Book Incorporation.

WHO – see World Health Organization.

World Health Organization (WHO). 1996. *Control of hereditary diseases*. Report of a WHO Scientific Group. Geneva: World Health Organization.

WHO. 2000. *Primary health care approaches for prevention and control of congenital and genetic disorders.* Geneva: WHO.

WHO/March of Dimes. 2006. Report of a joint WHO and March of Dimes meeting. *Management of birth defects and haemoglobin disorders.* Geneva: WHO.

WHO/WAOPBD
WHO/World Alliance of Organisations for the Prevention of Birth Defects Report (WAOPBD). 1999. *Services for the prevention and management of genetic disorders and birth defects in developing countries.* Geneva: WHO.