Stigma and African Genomics Research: An Exploration of Stigma Associated with the Genetic Attribution of Rheumatic Heart Disease

Marlyn Collin Faure

Thesis Presented for the Degree of

MASTER OF SCIENCE

In the Department of Medicine

UNIVERSITY OF CAPE TOWN

Supervisors:
Associate Professors Jantina de Vries and Mark Engel

June 2018

DECLARATION

I, Marlyn Collin Faure, hereby declare that the work on which this dissertation/thesis is based is my original work (except where acknowledgements indicate otherwise) and that neither the whole work nor any part of it has been, is being, or is to be submitted for another degree in this or any other university.

I empower the university to reproduce for the purpose of research either the whole or any portion of the contents in any manner whatsoever.

Signature: Signed by candidate

Date: 7 June 2018

ACKNOWLEDGMENTS

There are many people and moments that have brought me to this place. While these many of these people and places cannot be named, I would like to acknowledge the following persons:

- Creator who has gifted me with life in these historical moments where as a brown person I have access to elite spaces of higher education.
- My parents, grandparents and all those who came before them who have sacrificed so much under centuries of oppression – it is their life-blood that continues to push me, and it's on their shoulders that I stand.
- My supervisors Jantina de Vries and Mark Engel who both supported me, pushed me and saw what I often could not. To you I will remain indebted for your wisdom, guidance and honesty.
- A community of people who believed in me and supported me in so many ways, from offering up their homes for me to work in, feeding me, giving me process space, and constant forgiveness for my absence over extended periods.
- Finally to my partner, Parusha who stood by me (even across continents), who cheered and cared for me, and believed in me even when I did not believe in myself. Your love held me.

ABBREVIATIONS

ARF	Acute Rheumatic Fever
ISMIS	Internalized Stigma of Mental Illness Scale
FGD	Focus group discussion
RHD	Rheumatic Heart Disease
RHDGen	Genetics of Rheumatic Heart Disease
SAX	Genomics of Schizophrenia in Xhosa speaking South Africans
WHO	World Health Organisation

LIST OF FIGURES

LIST OF TABLES

ABSTRACT:

Introduction:

There is mixed evidence about how genetic attribution of disease may impact on stigma. One theory, based on essentialism, argues that knowledge of genetic attribution may increase stigma, while attribution theory argues that bio-genetic explanations may result in individuals feeling a decreased sense of personal responsibility about the disease. Most empirical studies shows mixed evidence. These studies however are mostly conducted in Western contexts. This study then is one of the few studies investigating the impact of genetic attribution on stigma in an African context. Specifically, this paper explores the question of genetic attribution with RHD patients in the Western Cape.

An important part of this exploration was the use of video content to stimulate discussion in focus groups. Many studies include visual methods and justify the use of visual method based on the assumption that visual methods are more effective at stimulating discussion and generating richer data in qualitative research. In addition to explore the impact of genetic attribution on stigma, this thesis also evaluates the efficacy of visual in qualitative research.

Methods:

Given that this paper has two components, one investigating the impact of genetic attribution on stigma, and a methodological component, this thesis presents findings of three sub-studies. The primary study (Study 3 in this thesis) related to stigma and genetic attribution, 11 focus group discussions were conducted using vignettes to explore the impact of genetic attribution on stigma with RHD patients. These vignettes were developed into films and used to stimulate discussion in FGDs. Thematic coding analysis was used to analyse data.

For the methodological component, one study, presents a systematic review of evidence related to the efficacy of visual methods in qualitative research (Study 1 in this thesis). The final study, is an empirical evaluation of the efficacy of visual methods (Study 2 in this thesis).

A before/after study designed was conducted to evaluate the efficacy of visual methods. Six of the FGDs watched the video clips produced from the FGDs, while the other five had the vignettes read to them. Another source of evidence for the evaluation was using the coding density calculated by NVivo 11 software.

Results:

For the primary study investigating the impact of stigma, the finding show that stigma has a negligible impact of stigma amongst RHD patients in the Western Cape.

For the methodological component, the systematic review finds evidence that visual methods are more effective at generating richer data. The evaluation study however finds no difference in results before and after each stimuli, when compared between groups who watched the video or heard the vignettes read.

Conclusion:

For the primary study investigating the impact of stigma, one of the reasons no evidence was found was because of the low level stigma reported. In instances where stigma is reported, I argue that it is in the context of RHD in this population, the impact of genetic attribution on stigma is displaced given that individuals having multiple explanations models of genetics is just one. Additionally, this population is forced navigate more immediate challenges such as cultural norms, and structural inequality related to the enduring impact of South Africa's racialised apartheid history.

In relation to the methodological component, I argue that results from the systematic review is difficult to generalise given the small number of included studies, and the lack of detail described in the studies, used to evaluate claims that visual methods are more effective. The result of the evaluation finds no difference between the groups which may be there are no differences between these methods, or the questionnaire may have been inappropriate. This study nonetheless is still the first to empirical evaluate such claims.

Contents

INTRODUCTION

This thesis explores the impact of genetic attribution on stigma amongst Rheumatic Heart Disease (RHD) patients in the Western Cape, or, in other words, whether framing an illness in genetic terms changes the perception of stigma associated with the illness. It also explores the effect of using visual methods a prompts in qualitative research. It forms part of a larger study, namely the "Stigma in African genomics research on Schizophrenia and Rheumatic Heart Disease" study or "Stigma in African Genomics" for short funded by the US National Institutes of Health (NIH) (Fund number 5U01HG008226). The Stigma in African Genomics study is a mixed methods study investigating the impact of genetic attribution on internalised and associative stigma with schizophrenia and rheumatic heart disease (RHD) patients in the Xhosa population in the Western Cape, South Africa. The parent study was comprised of two components. First, the study used the Internalized Stigma of Mental Illness Scale (ISMIS), a quantitative scale, to investigate self-reported levels of stigma [1]. The second component of the parent study conducted 12 focus group discussions (6 focussing on schizophrenia and 6 on RHD) investigating the impact of genetic attribution of schizophrenia and rheumatic heart disease. Three vignettes were developed to stimulate discussion about the relationship between stigma and genetic attribution of disease. The parent study drew on patients who were enrolled in two genomic studies, both based at the University of Cape Town, namely the "Genomics of Schizophrenia in Xhosa speaking South Africans" or SAX study and the "Genetics of Rheumatic Heart Disease" or RHDGen study.

OBJECTIVES OF THESIS

This thesis has two components. The first relates to the use of visual methods in qualitative research. The Stigma in Genomics Study used vignettes to stimulate discussion in FGDs. These vignettes were developed into short videos by a professional filmmaker. The use of film

in this parent study was premised on the assumption that visual methods are effective at stimulating discussion and generating richer data when compared to traditional methods such as text based methods or traditional interviews using only questions. I questioned this assumption, and designed my research project in such a way as to also explore whether the use of visual methods in qualitative social science research indeed leads to 'richer data'. The first component of this thesis empirically evaluates this claim. The second component of this thesis investigates the impact of genetic attribution of RHD on stigma with the mixed ancestry or coloured (a term which will be explained later) population in the Western Cape, using the same methodology as the parent study but in a different population group.

I conducted three sub-studies to investigate these two questions and are presented separately in the chapters of this thesis. Study 1 reports on a systemic review synthesising evidence relating to the efficacy of visual methods in qualitative research. Study 2 reports on an empirical investigation comparing film and text-only vignettes in the focus group discussions (FGDs) conducted for this study. Study 3 reports on the relationship between stigma and genetic attribution of RHD. Study 2 and 3 draw on 11 FGDs conducted with RHD patients for this study.

a) Study 1 & 2: comparing the efficacy of film and text-based vignettes in generating rich data in qualitative research

Part of the Stigma in Genomics study used FGDs to investigate the effect of genetic attribution on stigma. Vignettes were developed about a fictitious character who develops either schizophrenia or RHD. These vignettes were then developed into video clips by a professional filmmaker. These clips were used to stimulate discussion. When these vignettes were developed it was assumed the use of visual methods, such as video clips, were more effective at stimulating richer discussion than text-based or verbal-based only methods. When I started

this project focussing on RHD patients in the coloured population, I began to question this assumption and decided to investigate this claim.

The use of visual methods is now well established across a variety of disciplines. A literature review by Pain explored why researchers choose to use visual methods and identified two main reasons: first, that visual methods are more effective at generating richer data; second, that visual methods allow participants to have more control in the research process by mitigating the power differences between researcher and participants [2]. Yet there is little empirical evidence to support such claims [2].

In Study 1 (Chapter 3) of this thesis, I used a systemic review method to summarise evidence related to the efficacy of visual methods to stimulate discussion and generate richer data. Using a comprehensive search strategy I searched 11 databases across various disciplines including education, marketing, business studies, and sociology, anthropology and health sciences. This systematic review is the first of its kind. The results of the systematic review found that there is evidence to support claims that visual methods are more effective at generating richer data in qualitative research. However, there are also about concerns about the robustness of this data. In addition, the review shows that more evidence is needed to substantiate claims that visual methods are efficient at stimulating discussion and generating richer data in qualitative research. Additionally, the findings also reflect that no clear evaluation methods currently exist to evaluate such claims.

Study 2 (Chapter 4) seeks to fill these gaps by contributing empirical evidence about the impact of film vs text-only vignettes in eliciting rich data. It presents the results of an empirical study comparing the efficacy of visual methods with text-only methods in the context of focus groups discussions. As I conducted 11 FGDs to examine the effect of genetic attribution, I designed the study so as to enable me to compare the data generated in FGDs using a filmed vignette versus that generated using a text-only vignette. Specifically, 6 of the FGDs used video clips to stimulate discussion and 5 FGDs used a text-only vignette. To evaluate these methods, I used a before/after study design to compare how participants responded to a set

of questions before and after the stimuli. I also compared the coding density of the transcripts and supported my results with observations recorded in field notes. Contrary to the results of the systematic review, I find that there is no significant difference between the use of visual methods and text-based methods in qualitative research.

b) Stigma and Genetic Attribution: A qualitative study

The impact of stigma has a long history but it was only when the sociologist Erving Goffman published his influential work, *Stigma: Notes on the Management of Spoiled Identity* [3] in 1963 that stigma began to be taken seriously as an area for academic investigation. There has since been a proliferation of empirical and conceptual work investigating a wide range of aspects of stigma and its impact on human wellbeing. What is critical for this discussion, is how stigma is defined and measured. Stigma has been conceptualised in two broad categories. The first follows Goffman's approach which conceptualises stigma as a static concept which marks or sullies individuals or groups [4, 5] . This definition has been contested by others, arguing that stigma does not only exist in static ways but as a social process. Link and Phelan have been most influential in conceptualising stigma as a social process relying on unequal power relationships [6].

The academic interest in stigma studies has been documented particularly in relation to health [7]. Importantly, recognising and trying to alleviate the impact of stigma on health has been critical for developing approaches to deal with complex diseases such as mental illnesses. The impact of stigma on a person's wellbeing can range from feeling a sense of personal responsibility, negatively affect self-esteem, lead to social exclusion and discrimination and influence treatment-seeking behaviour and adherence [8, 9].

The genomic revolution has drastically changed how we understand health, disease and treatment. These advancements have also resulted in number of other ethical [10] and social challenges including how genetic knowledge impacts on stigma relating to illness [11]. There

are currently two views on how genetic attribution may impact stigma. The first argues that genetic attribution could increase stigma given that individuals may believe that they are less likely to recover, and experience further social exclusion and discrimination as people are less likely to believe that the condition will improve [12]. The second view argues that bio-genetic explanations of disease may decrease stigma as individuals may feel less responsible for their disease, and the public may have greater sympathy for individuals if they are less at fault [12]. Empirical studies have found evidence in support of both of these theories [13, 14]. Most of these studies however, have been conducted in Western contexts and have focussed on public attitudes, with only few studies including participants living with disease. Currently there are only three published studies in the African context exploring the impact of genetic attribution on stigma [15-17]. These studies also provide mixed evidence. Yet genomics research is increasing, for instance through activities like the H3Africa Consortium [18] , and questions about how an increase in genomic knowledge may impact on stigma associated with conditions is key to ensuring that genomics research is conducted in a responsible and beneficial manner.

This study then is one of the first studies to investigate the effect of genetic attribution conducted in the African context, and the first to do so in South Africa. The results of this study are presented in Study 3 (Chapter 5) of this thesis. It reports on the evidence generated in 11 FGDs conducted with RHD patients in the Western Cape. In this study, I argue that in the context of RHD in the coloured population, the impact of genetic attribution on stigma is displaced. The first reason is that bio-genetic explanations is one of many explanations individuals draw on to understand their disease causation. The second reason, is that individuals are forced to navigate more immediate challenges such as cultural norms and structural inequality related to the enduring impact of South Africa's racialised apartheid history. Whilst the chapter presents some insights generated about RHD in this population, this description is not reflective of the depth of the empirical data on that topic that I collected, which will be presented in a separate publication.

1. CHAPTER 1: LITERATURE REVIEW

1.1 INTRODUCTION

This thesis is primarily concerned with how genetic attribution influences experiences of stigma amongst RHD patients in the Western Cape. A key concept to investigating this relationship, and one that will be first explored in this chapter is stigma and the complexities related to how stigma has been conceptualised. I then discuss stigma and its relationship to health. In the next section, I explore theoretical and empirical studies exploring the impact of genetic attribution of disease on various kinds of stigma. I subsequently focus on empirical studies which have investigated the effect of genetic attribution of disease on stigma on the African continent. Noting the paucity of work in this area, I lastly focus on Rheumatic Heart Disease as focussed on in this thesis.

1.2 STIGMA: CONCEPTS AND COMPLEXITIES

How stigma has been defined and constructed has often been contested. The word 'stigma' initially referred to a tattoo which marked someone's devotion to service in temples. Later the meaning of these markings changed to label individuals as prisoners or criminals [19]. While stigma as a social phenomenon has a long history, it was only until sociologist, Erving Goffman's seminal work, *Stigma: Notes on the Management of Spoiled Identity* [3] that studying stigma gained significant traction in academia.

Goffman was interested in the experiences of people who lived with mental illnesses, had physical deformities or who were generally associated with deviance [20]. From his studies, Goffman formulated an understanding of stigma as an "attribute which is significantly discrediting". Goffman argued that deviance from normative social arrangements marked an individual as having a "spoiled identity" [3]. Generally Goffman's work has been the primary source of understanding and conceptualising stigma.

Research on stigma has proliferated since Goffman published his pioneering work on stigma in 1963 [6]. Stigma has subsequently been studied with respect to a variety of fields including, exotic dancing [21, 22] leprosy [23], obesity [24], cancer [25], disability [26], HIV/AIDS [27], sexuality [28], unemployment [29, 30] and deviance and criminality [31, 32].

How stigma has been conceptualised subsequently can generally be categorised by two broad approaches. The first draws on Goffman's approach. Here, stigma studies tend to focus on understanding stigma as an individual trait which marks an individual or group. Goffman's work has been influential on many studies – these studies have generally understood stigma as a static concept which an individual possesses. For example Stafford and Scott conceptualise stigma as a "characteristic of persons that is contrary to a norm of a social unit" [4]. And Byrne in his study on mental illness defined stigma as a "sign of disgrace or discredit, which sets a person apart from others" [5]. Contesting these definitions, Parker and Aggleton assert that one of the reasons stigma has been so narrowly defined, is that Goffman's idea of stigma has often been somewhat misinterpreted and employed as a "kind of thing" (italics theirs), an attribute which serves as a marker of difference, and is "mapped onto people" [20]. In this understanding of stigma, it is the body which serves as the site for the production of difference [20]. Studies which have used such static conceptions originate from social psychology [20] and have often been criticised for being highly individualistic [6]. In more recent scholarship, these conceptualisations have been criticised, with scholars asserting that stigma should rather be defined as process.

In response to this understanding, other scholars have problematized these conceptions as being too static, and not fully capturing how stigma operates in real-world contexts. The second approach conceptualises stigma as a process and has been pioneered by Link and Phelan. This approach moves away from the understanding of stigma as an attribute sullying a body, and conceptualises stigma as a process rooted in asymmetrical power relationships [6]. They outline five contingent components necessary for stigma to exist. The first is labelling, which occurs when people are assigned labels which attribute social value and results in the

production of difference. In the second component, the label is attached to negative stereotypes which further seeks to entrench normative boundaries of belonging. The third component relies on assigning people into distinct and often hierarchal groups. The fourth occurs once people have been labelled, stereotyped, categorized as being different, they lose social status as full members of society and are discriminated against. The combination of these interacting forces often results in loss of life chances as groups who are discriminated against are often excluded from critical basic rights or services such as income, education or healthcare. The final component is rooted in existing power relationships – Link and Phelan argue that for an individual or group to be stigmatised, those who stigmatize must be in a position of power to ensure that negative labels and categories are embedded within cultures and structures in order to negatively impact the lives of individuals and groups [6].

Within this broad conceptualisation of stigma as a process, Bos et al. drawing on the work of Pryor and Reeder, note that stigma can also be manifested in four distinct yet interrelated ways [33]. The first is public stigma which refers to the "cognitive, affective, and behavioural" response to an individual who is perceived to be stigmatised [33]. The second type of stigma is self or internalised stigma. This refers to the negative impact the stigmatised individual or group may experience, and to the individual accepting or internalising society's beliefs and attitudes [33, 34]. The third type is associative stigma which refers to the "social and psychological reactions to people associated with a stigmatized person (e.g., family and friends)". This also includes people's responses to being affiliated with a stigmatised individual [33]. The fourth relates to structural stigma which exists at the macro-level, refers to how "rules and policies" limit life chances of stigmatized groups [34].

1.3 STIGMA AND HEALTH

There is a long history of the relationship between stigma and health. The relationship between stigma and conditions like epilepsy dates back 4000 years, while other forms of disease-

stigma only have relatively recent histories about which we have much to learn [35]. Over the last few decades, the number of studies related to stigma and health has grown considerably. Major and O'Brien found that the volume of articles just using the word 'stigma' in the PsychInfo database had significantly increased between 1990-2004 (n=2321), when compared with 1965-1989 (n=603) [7]. Weiss noted a similar pattern when searching health sciences databases [36].

One of the reasons stigma is increasingly receiving attention is due to its often devastating impact on the already compromised health of individuals and groups. Disease stigma significantly impacts the lives of individuals and groups, and includes low-self-esteem and isolation, decreased adherence levels and help-seeking behaviour, and decreased disclosure of infectious diseases. For example, the founding Director for the World Health Organization's former Global Programme on AIDS, noted that the fight against stigma faced by those living with HIV and AIDS is just a critical as combating the disease itself [9]. Studies investigating stigma relating to HIV have also found that stigma negatively impacts adherence [8], and fear of disclosure which may have impact on support systems [37]. High levels of stigma are often associated with lower incomes, poorer mental and physical health [38]. A systematic review investigating evidence about the impact of stigma on patients with lung disease found a positive relationship between stigma, poor quality of life and high levels of social stress. Furthermore, working on stigma related to mental health, Clement and colleagues, through a systematic review on the impact of stigma on mental-health patient's help-seeking ability found that internalised stigma was a primary barrier for help-seeking, and other forms of stigma were another major barrier to seeking help [39].

One of the reasons disease-stigma is particularly intractable is that illness or disease aetiology is seldom exclusively the source of stigma. Rather disease-stigma relies on existing social arrangements and has the potential to further entrench specific forms of stigma contingent on social, economic and political contexts. Weiss, who also defines stigma as a process, argues that health studies that investigate stigma must be:

> shaped by consideration of needs to formulate disease- and culture-specific interventions that consider psychological processes of individuals, social dynamics of institutions, and various social and economic processes… [36].

If disease stigma exists at the intersection of bio-medical, cultural and economic factors, in localised contexts, then changes in these contexts would impact disease stigma. Genetic research has irrevocably changed disease aetiology and care. There are important questions which must be asked in relation to such developments. What impact do bio-genetic explanations have on stigma? How does knowledge of genetic attribution influence the public's perception of disease? How does knowing that a disease is genetic change how individuals think about their disease, their responsibility in developing the condition, and the treatment they need to seek out? How do these bio-genetic explanations influence the kind of support provided to those living specific conditions? These and other questions are critical to think about, especially when considering how stigma is influenced by the genomic revolution.

1.4 STIGMA AND GENOMIC RESEARCH

The development and use of genomic methods have revolutionised how we think about health, disease and treatment. The bedrock of the impact of this advancement lies in its ability to be "predictive, preventive, personalised and participatory" [40]. It is this ability to predict that shifts how medicine is practised from 'reactive to proactive" [40]. Genetic medicine uses genomic-wide associations to measure variations in DNA sequences in an attempt to identify a common genetic marker of diseases [41]. The ultimate object of GWAS is to "make predictions about who is at risk and to identify the biological underpinnings of disease susceptibility for developing new prevention and treatment strategies" [41].

While these technologies hold the potential to significantly aid prevention and care, there are legitimate concerns over increasing geneticisation of diseases as a step towards eugenics [42]. For example, routine screening of foetuses for disabilities with the option to abort has led

to such concerns being raised in disability-rights literature [43] . Additionally, there are also other factors to consider when considering genomic research. Another important consideration is the influence of bio-genetic explanations on disease stigma, given its potential to reduce stigma, yet such claims remains highly contested [44]. Kong and others, in the context of mental health, are suspicious of claims that knowledge of genetic attribution will decrease stigma. Part of their argument rests on how laypeople do not think about genetics as being the sole cause of a disease, but rather understand genetics in a "probabilistic, nonabsolute rather deterministic manner" [44]. While theoretical frameworks to understanding directionality have been in existence for a while, empirical studies on the impact of genetics on disease stigma date back only about a decade ago [12, 45-47].

Currently the literature reflects that there are two potential directional pathways for how genetic knowledge can impact stigma. The first theoretical model suggests that genetic knowledge could increase both public and internalised stigma. This model is based on genetic essentialism, which argues that individual health outcomes are pre-determined by genetic make-up, and in this sense this theory holds that "we are our genes" [12]. Phelan notes three implications of this theory on disease stigma. First, that the individual living with a disease is considered to be essentially different, second that the condition is severe and unrelenting, and third, that there is a strong belief that family members will also develop the condition [12]. The second model argues that genetic knowledge may result in a decrease in stigma, because knowing that the cause of illness is a result of genetics could reduce self-blame, where individuals may feel less responsible for their condition. In addition, other people may be more sympathetic given that the individual cannot be held responsible for the condition [45]. This model is rooted in attribution theory which was proposed by Weiner, based on the idea that humans are interested in creating causal links between events. As such he asserts that when people are believed to be in control of negative outcomes (in this a particular case illness and/or associated behaviour) they are more likely to be held responsible. Whereas if they are perceived to have little or no control over the outcome they are more likely to be shown

sympathy [48, 49]. As Link and Phelan observe, it also entirely possible for both models to exist concurrently [6].

The results of many empirical studies, conducted to investigate the relationship between disease stigma and genetic attribution, are complex and reflect the complicated relationship between genetics, stigma and health. There are a number of reasons why results are heterogeneous and no conclusive evidence exists about the impact of knowledge of genetic attribution on stigma. First, most studies have focussed on the impact of bio-genetic explanations on public perception, most frequently through quantitative methods such as large-scale telephone surveys. Second, there is a paucity of studies which have included participants who live with specific conditions to provide personal insight on experiences of disease stigma, and the potential impact of genetic attribution [44]. This is important because stigma studies have long been criticised for their bias in foregrounding perspectives of those who are outside disease communities [6]. Third, if stigma is conceptualised as a process contingent on contextual factors, then we also have to consider where much of the empirical studies are located. The majority of studies originate in Western contexts, reflecting a serious limitation to understanding how genetic knowledge may influence disease stigma in non-western contexts.

The results of studies which investigate the impact that genetic knowledge could have on public perception reflect a complex relationship between genetic knowledge and stigma. Importantly, much of the empirical studies have focussed on mental illness. Phelan and others have, through telephonic surveys of US public perceptions tested various reactions to genetic causation vignettes. In one study, they found that genetic causation had no significant relation to attribution theory (i.e. it had no effect on the public's perception of blame and punishment). They did find that knowledge of genetic attribution supported a genetic essentialist theory in that people were more likely to believe that mental illness was more serious and that children were also likely to develop the conditions if the disease was genetic [12]. In another study Phelan and colleagues tested public perceptions of treatment recommendations and

strategies if they knew that depression and schizophrenia were genetic. They found in cases where genetic attribution was made explicit, respondents were more likely to recommend specific treatments such as seeing a psychiatrist or hospitalisation, however, treatment was thought to be unlikely to effectively treat depression or schizophrenia [46]. Some studies of public perception also conclude that genetic knowledge had a negligible effect on public perception. A systematic review on public perception of blame and responsibility towards people with mental illness, found that biogenetic explanations either had no significant impact on alleviating perceptions of blame, or in studies focussing on schizophrenia, perceptions of blame were increased [13]. Mixed results were found by Kvaale et al., who through a meta-analysis found that while biogenetic explanations of mental illness were more likely to reduce blame, it also increased perceptions that people living with mental illnesses are dangerous, and strengthened a desire to increase social distance [14].

The results from the few studies which have focussed on investigating the impact of genetic attribution with people in a specific disease community are also inconclusive. Exploring these questions with stigmatised groups is important, given many of the empirical studies have often focussed on perceptions of stigma by those who do actually experience disease stigma [6, 44, 47]. A US-based study, this time through in-depth interviews with individuals suffering from breast cancer, sickle cell disease, cystic fibrosis and people living with deafness, investigated the meaning that individuals would attach to genetic causation in relation to stigma. The results reflect how association with genetic attribution is contingent on disease-contexts. For those who were deaf, a genetic cause created a point of connection and belonging to other family members who are also deaf. While those living with sickle cell disease and cystic fibrosis felt that a bio-genetic explanation of their disease relieved a sense of personal responsibility or lessened feelings of inferiority. However, there was also evidence to suggest that participants blamed their parents for not preventing the disease. In the case of breast cancer, genetic attribution decreased a sense of personal responsibility, but respondents also reported on feeling more vulnerable given the increased likelihood of other family members developing

breast cancer. Importantly, the researchers concluded that, while the results may be mixed, where patients did report on experiences of stigma, it was not exclusively related to genetic attribution but rather these experiences, must be understood "within the lived experience of a particular condition" [47]. Easter, who interviewed US women who suffered from eating disorders, also explored the impact of genetic aetiology on subjective experiences of stigma. This was also one of the first studies to explore this question with patients who have a mental illness. Her findings also illustrate the complexity of the relationship between genetic attribution and stigma. Her results show that bio-genetic explanations were more likely to decrease a sense of personal blame and responsibility. However some respondents also reported that it may lead to a sense of helpless and pessimism about the impact of the disorder [50]. A different set of results were found by Meiser at al, which largely supported attribution theory in families with high-incidence of bio-polar disorder. They found that their respondents believed that bio-genetic explanations were more likely to reduce stigma given that "it shifted the locus of control and responsibility away from the individual towards the role of heredity" [51]. Furthermore they found that respondents often insisted on interpreting genetics in non-deterministic ways (i.e. must be seen as interacting with environmental factors) [51]. Also finding some evidence to support attribution theory, a Dutch study involving people living with depression, found that respondents expressed that knowledge of genetic attribution could reduce stigma by alleviating self-blame. Their results show that biogenetic explanations helped participants have more open discussions with their family about the disease and its causes. However they reported that respondents did not think that biogenetic explanations would not change social stigma [52].

These studies show that the impact of knowledge of genetic attribution is inconclusive. In studies whose participants are part of a disease community, there is some evidence supporting both essentialist and attribution theories. Furthermore, what these studies reflect is that stigma, when conceptualised as a process, is contingent on socio-cultural

arrangements. Most studies have focussed on Western context and very little is known about how biogenetic explanations may influence stigma in other contexts.

1.4. STIGMA IN AFRICAN GENOMICS

When it became clear that genomic medicine had the potential to revolutionise the field, there were concerns about the need for developing countries, especially on the African continent, to be part of this revolution, given global health disparities [53, 54]. Some serious concerns were raised about the need for capacity building, public engagement and support, and funding from local and global communities [53]. Subsequently, there have been several genomic initiatives that have played an important role in ensuring that African scientists and populations are included in the genomic revolution. The Human Heredity and Health in Africa or H3Africa, funded by the National Institute for Health and The Welcome Trust, and African Center of Excellence for Genomics of Infectious Diseases or ACEGID, funded by the World Bank, are examples of such initiatives [55]. While these initiatives are critical in ensuring that African populations benefit from biomedical technological innovation, this work has also created important ethical challenges in relation to local contexts and necessary global collaborations [10]. One important challenge relates to understanding how an increase in genomic research could influence disease-related stigma in African research contexts.

As noted previously, there is a lack of studies which seeks to understand the impact of biogenetic explanations of disease stigma on African populations. There are only three studies that have investigated this relationship in the African research context. De Vries and colleagues working a project focussing on the genetics of malaria, in Kenya and the Gambia, investigated how genomic research could cause stigma for members of a particular ethnic group if that group were found to have a genetic predisposition to developing stigmatized diseases [10]. This question is especially relevant when there is existing tension between ethnic groups and when research focusses on diseases with pre-existing stigma. They interviewed key stakeholders, fieldworkers, members of ethics committees and members of

funding bodies. Their results show that genomic research would be unlikely to cause stigma, though it could increase stigma that already exists [10]. Furthermore, participants generally understood stigma to be a kind of mark which would label the group as different or deviant. And while malaria was not stigmatised, some respondents expressed concerns that data could be used for research on conditions that are stigmatised in secondary analysis. There is precedent for such concerns given the well documented case of the Havasupai where secondary use of data and samples were found to be potentiality stigmatising even though primary use was not [56]. Linked to this is the concern that if attention is paid to already stigmatised diseases, then stigma may increase for those patients [57]. Tekola and colleagues, in a qualitative study working on consent with people in Ethiopia who live with podoconiosis, a condition which causes serve swelling of the lower limbs, found that pre-existing stigma levels attached to podoconiosis, influenced people's decision to be part of the study. Given the high levels of stigma attached to the condition often as a result of it being genetic (which was well known in communities), participants suggested that consent needed to be sought from the entire family and not just the individuals living with the condition, as stigma could increase for the entire family [57, 58]. Pointedly they found while most participants knew that podoconiosis is familial, participants instead chose to deny the genetic aetiology as it could further increase stigma for family members [57]. In another study, Marsh and colleagues reported on a study on sickle cell disorder and genetics in Kenya in which they found that gender often mediated experiences of stigma. They found that mothers of children who had sickle cell disorder were blamed for their child's condition. This was the case even when information was offered that the genetics of both parents pay a contributing role, or when there was clear evidence that the disease was inherited from the father's side. Their findings show how stigma experiences in this context are never exclusively mediated by biomedical explanations but rather occur at the complex intersection of genetics, culture, and structural economic and gendered inequalities [59]. These findings indicate strong support for understanding stigma as a process contingent on a variety of factors.

The results from these studies reflect how disease stigma is embedded in cultural, gender, socio-economic beliefs and structures. Moreover, the dearth of studies focussing on African populations also suggests that there is a clear need to understand disease stigma and how genetic knowledge may influence stigma. Furthermore, only 2 of these studies included participants from disease communities. This study would be the first to investigate the influence of genetic knowledge with RHD patients in South Africa.

1.5 RHEUMATIC HEART DISEASE IN SOUTH AFRICA

Rheumatic Heart Disease (RHD) is a chronic heart condition that is caused by untreated infection with Streptococcus pharyngitis [60]. In some instances, untreated Strep A can cause rheumatic fever, which can result in an autoimmune response which attacks the tissues of heart valves. Over time and with continuous infections with Strep A, permanent damage can be caused to heart valve tissue. This can potentially result in unalterable and fatal heart damage [60] which can only be effectively treated through surgery.

RHD is a disease which largely only affects the global poor with the condition virtually having being eradicated in high-income countries. The residual burden of this disease is carried by middle and low income or indigenous populations in high-income countries [60, 61]. Furthermore, evidence exists which shows that high levels of poverty are correlated to high incidence rates [61]. At a global level, RHD is still one of the most prevalent cardiovascular diseases in young adults and children [62].The World Health Organisation (WHO) estimates that approximately 15-16 million people are living with RHD with around 233 000 annual deaths occurring as a direct result of acute rheumatic fever or RHD. The WHO also notes that these figures are conservative with incidence rates possibly being much higher [60]. In sub-Saharan Africa, 30 out of 1000 children are affected, “leading to the designation of this region as the ‘hotspot’ of RF and RHD in the world” [61]. This is especially troublesome given that preventative measures are both effective and inexpensive [63]. The use of penicillin, in

primary care, to treat sore throats will prevent RHD from developing and prophylactic use of penicillin in RHD patients effectively prevents continuous infections and further damage to cardiac valves. Like other countries, South African incidences of RHD are correlated to socio-economic inequity, such as poverty, over-crowded households, poor nutrition and low levels of education [64]. Furthermore, living with Acute Rheumatic Fever (ARF) and RHD, have high and enduring social and economic implications for both patients and families of patients. Some of these costs are increased school drop rates, and loss of income due parent absenteeism [61].

The RHDGen project aimed to contribute to how we understand RHD by investigating genetic factors that contribute to disease development. This was described as one of the four pillars of the global action to eradicate the disease [65]. RHDGen aimed to determine genetic factors which may affect susceptibility of patients to develop ARF and rheumatic heart disease, post Strep A pharyngitis infections. A systematic review of published twin studies, by Engel et al. found heritability of developing rheumatic fever to be 60% [66]. RHDGen brought together research from a number of African countries to conduct a genome-wide association study of patients and unaffected groups for control. The majority of samples were collected in Cape Town, though the cardiac unit at Groote Schuur Hospital.

Currently no work exists on the impact of genetic knowledge on stigma relating to RHD in South Africa (and the rest of the continent). Most studies investigating stigma and genetics have focussed on HIV/AIDS and mental health. Additionally, very little work has been on the stigma of cardiovascular diseases. This study thus is the first to explore how knowledge of genetic attribution will impact potential stigma related to RHD patients in South Africa.

2. CHAPTER 2: RATIONALE AND DESIGN OF THE STUDIES

2.1 OVERVIEW

This study's objective is to explore the effect of genetic attribution on stigma, amongst RHD patients in the Western Cape. The study uses FGDs to explore this question with patients based at Groote Schuur Hospital in the Western Cape. As part of the FGD design, short videos were produced to stimulate discussion during the FGDs. This design was premised on the assumption that the use visual method is more effective at stimulating discussion and generating richer data than other traditional methods such as text-based methods. Part of this this study presented in this thesis, also included an embedded study which evaluated the efficacy of visual methods in the context of focus group discussions.

This study forms part of a larger NIH-funded study entitled "Stigma in African Genomics Research on Schizophrenia and Rheumatic Heart Disease" – the "Stigma in Genomics study" for short. This study investigates the impact of genetic attribution on stigma associated with schizophrenia and Rheumatic Heart Disease (RHD) within the Xhosa population in the Western Cape. The study reported in this thesis expands the methodology for the broader Stigma in Genomics study to the mixed ancestry population in the Western Cape of South Africa.

The Stigma in Genomics study drew on both quantitative and qualitative methods to investigate the role of genetic attribution on stigma. In that project, the investigators conducted 12 FGDs, 6 with Xhosa patients who have schizophrenia and 6 with Xhosa RHD patients. An internalised stigma scale was administered before each group discussion to quantitatively measure levels of internalised stigma in each patient group. After administering this scale, patients watched a video about a fictional character who develops either RHD or schizophrenia patients. The population for the larger study

however only focussed on the Xhosa population and primarily investigated the effect of internal and associative stigma and the impact of genetic knowledge on levels of stigma.

Instead, the study reported in this thesis investigates the impact of genetic knowledge on patients who have RHD within the mixed ancestry or coloured population only. Given that stigma experiences are also influenced by social factors and the cultural environment it was important to investigate this question within this particular population also. In addition, it was important to include this group given that it comprises a large number of patients who attend Groote Schuur. For reasons of efficiency and because we did not have access to mixed-ancestry schizophrenia patients, the study reported in this thesis also distinguishes itself from the Stigma in Genomics study in that it only focused on RHD and not schizophrenia.

Beyond the inclusion of the coloured or mixed ancestry population, the study reported on in this thesis also had a strong methodological component. As previously mentioned, in the Stigma in Genomics project the investigators used videos on the basis of an assumption that the use of visual material in qualitative research stimulates discussion and richer data collection. I questioned this assumption, and thus designed my research project in such a way as to also explore whether the use of visual methods in qualitative social science research indeed leads to 'richer data'. To this end, the study reported on in this thesis had two important methodological components: first a systematic review which established evidence for the efficacy of visual materials in qualitative research and explored the tools used to evaluate such methods which is reported on Chapter 3, Study 1. The second methodological component is an original empirical study which evaluates the efficacy of visual methods in qualitative research, which is reported on Chapter 4, Study 2 of this thesis.

2.1.1. The study population

Before describing specific demographics of the population in this study, it is important to briefly explain the term ‘coloured’ or mixed ancestry. The term coloured was used as a racial classification for people of mixed race under colonialism and apartheid in South Africa. This group was made up of slaves brought from the East, including countries such as Indonesia, Java and Batavia, and also included groups from Madagascar and indigenous groups like the Khoi and the San [67]. Through intermarriage with local Nguni people, white settlers and other groups who settled as slaves or free people, a group of ethnic diversity began to emerge and became known as ‘coloured’ [67, 68]. Under apartheid this was term was used as an official race classification and was used to differentiate between African blacks, whites, Indians and Chinese. This classification under the apartheid government was designated sub-human status in relation to whites, and preferential treatment in relation to African blacks [69]. Under the Group Areas Act of 1950 this population, like other non-white groups, had to legally live in areas designated for coloured people only, which in Cape Town in the Western Cape was an area that came to be known as the Cape Flats. As a result of dispossession and forced removals, this part of Cape Town is well known for many social ills such as high levels of crime, gangsterism, alcohol and drug abuse, and poverty.

While the term ‘coloured’ is contested and sometimes substituted with the less connoted term ‘mixed ancestry’, as a member of this population I will use the term ‘coloured’ in relation to the study population because the term captures the ethnic diversity and cultural association, and signals this group’s political and social history.

The coloured people makes up about 8.8% of South Africa’s total population and 49% of Western Cape’s population [70]. Participants are generally of lower socio economic status with 50 % of the group earning no income and earning a maximum of R3200

(250 USD) per month [70]. In terms of language, a large proportion of this group speaks Afrikaans as first language with the remaining speaking English as a first language.

The sample included a total of 52 participants (n=52). In terms of gender, 90% of the sample were women, while the remaining 10% were men. The average age of participants was 59 years, with the youngest participant being 35 and the oldest 78. In terms of level of education, only 4% of participants completed a tertiary-level qualification, and the majority of participants did not complete high school. In terms of socio-economics, most participants were are on a government pension or disability grant which is a monthly income of R1500 (121 USD).

Many of the participants recruited for this study were enrolled for the RHDGen study which was a study focussing on the genetics of RHD [71]. We therefore worked on the assumption that many of the participants had previously been exposed to genomic research.

2.2. STUDY 1: SYSTEMATIC REVIEW OF VISUAL METHODS

2.2.1. General Description of the Study

Systematic reviews use a rigorous method to summarise data from a collection of studies, specifically giving increased power to detect an association between risk factors and the outcome of interest. Using the systematic review method, we investigated whether the use of visual materials was an effective stimulant to generate richer data in qualitative research.

2.2.2. Specific Objectives of the Systematic Review

There were two primary objectives for the systematic review. First was to review whether the use of visual materials was more effective than other stimuli at generating

richer data in qualitative discussions. The second objective was to summarise how various studies evaluated the efficacy of visual materials in qualitative research contexts.

2.2.3. Methods of the Systematic Review

The methods for this systematic review are described in detail in Chapter 3: Study 1.

2.3. STUDY 2: EFFICACY OF VISUAL METHODS

2.3.1. Introduction

This study is primarily interested in empirically testing whether the use of visual methods are more effective at stimulating discussion and generating richer data in focus group discussions. I used three different methods to test claims that visual methods are superior to text-based methods in qualitative research. Results from the systematic review in Study 1 showed that there is no clear reproducible evaluation method which exists in the literature. While the methods described below have important limitations (discussed in Study 2, Chapter 4), they are first attempt to conduct such an evaluation.

I used three approaches to evaluating the efficacy of visual material, namely questionnaire data, coding of transcripts and researcher field notes. What follows first is a description of the methods related to the questionnaires used in the FGDs. Section 2.3.9 discussed methods related to the use of the transcripts and field notes.

2.3.2. Study Design and Setting

We conducted this study in Groote Schuur Hospital, the largest tertiary hospital in the Western Cape. The Western Cape has the largest population of coloured people in the South Africa. Patients who took part in the study had either previously been enrolled in other studies related to RHD or are currently being recruited for new studies relating to RHD. Many of the patients regularly attend Groote Schuur hospital for treatment and to receive medication.

In the Stigma in Genomics study, researchers had already developed vignettes and FGD topic guides by the time I joined the team to conduct this study, and I used these materials as the basis for my own project. The Stigma in Genomics study had developed three vignettes relating to the causation of RHD, namely genetic, mixed or environmental. The vignette tells the story about a fictional character who develops RHD. Details about the content of the video can be found in section 2.4.

There are a number of reasons why researchers choose to include visual methods. Some of these reasons include that they are more effective at rapport building, generating rich data with specific reference to topics that are difficult to probe using other means, and aided in creating a more reflective space [2]. This study is specifically interested in empirically testing the efficacy of visual methods in stimulating discussion and generating richer data in qualitative research.

In order to test the efficacy of visual methods versus other methods, I designed a comparative study where I conducted 6 FGDs using the video vignette (2 for each scenario), and 5 FGDs using the text-based vignette that I read out to the participants during the FGDs, as is more common in this kind of research. While it was my intention to have one additional FGD which used a text-based vignette, there were some challenges with recruiting additional patients and I had to abandon that attempt in the interests of time. These challenges are explained in section 2.4.4. Both the read and video groups either watched or listened to same vignette about the factitious character,

Enrico, who develops RHD. This was important in the process so that if there was a difference between the read and video groups, it would be likely attributed to the stimulus.

This study used a before/after study design to trace changes in individual responses before and after the stimulus. Before participants were exposed to either watching the video or hearing the read vignette, they were asked to complete a questionnaire which asked them to respond personally to statements relating to RHD, using a 1-4 Likert scale (Appendix 1). After completing the questionnaire, participants then watched the video or listened to the vignette being read to them. Details about the development of the questionnaire can be found in Section 2.3.6. After this, participants were told that questions that were posed would not be answered immediately and they were not told about post-exposure questionnaire. The same questionnaire was immediately administered after the focus group ended. Figure 1.1 is a graphic representation of the structure of the evaluation component of the study with each scenario.

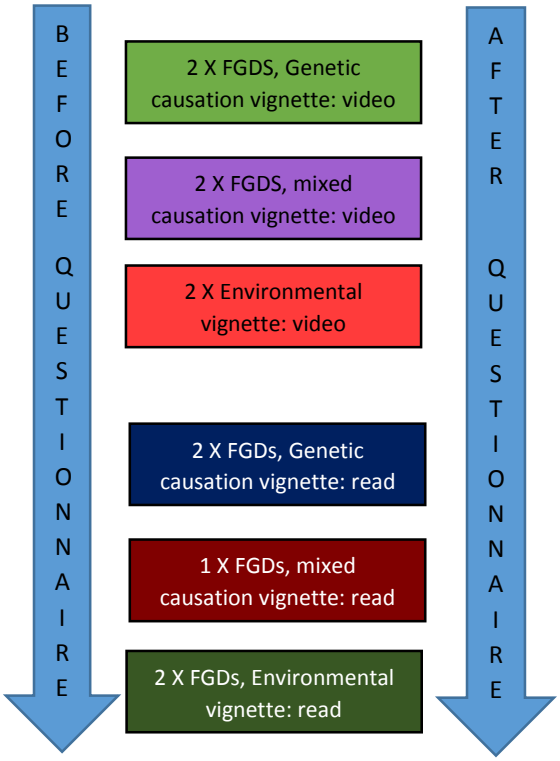

Figure 1.1: Schematic representation of evaluation process

2.3.3. Specific Objectives of the Study

- To evaluate if the use of visual material in FGDs is more effective than textual methods as a stimulant for stimulating discussion and eliciting richer data.
- To explore evaluation methods which aid in assess such novel methods.

2.3.4. Sampling Procedures

The study population consisted of coloured RHD patients who had previously enrolled in studies relating to RHD. A list of patients was obtained from clinical trial registry managed by the clinical trial manager of these studies listed above. Patients were phoned by myself as the researcher or the research assistant who worked on the project. Patients invited to the FGD discussion at Groote Schuur Hospital. The only criteria for being enrolled was that patients had to be coloured and have RHD.

2.3.5. Data Collection and Handling

2.3.5.1. Design of Questionnaire

The results of the systematic review provides evidence that the use of visual methods may yield richer data than other forms of stimulants in qualitative research. The review however found no consistent or standardised evaluation tool or methods which could be used to test assumptions made by researchers about the efficacy of visual materials in generating rich data. An important component of this second study was to design an evaluation plan which could test the response of participants to various stimuli. Given that qualitative data is subjective, it was important to isolate subjective responses to visual stimuli. In order to effectively evaluate the effect of the responses, a questionnaire was designed to test participants' responses in relation to six domains, namely, general interest, personal engagement, empathy, willingness to share in a group, and emotional response. Three

statements were provided under each domain. The questionnaire used a 1-4 Likert scale to measure participants' responses (see Appendix 1).

From the systematic review, it became clear that such an evaluation tool did not exist in existing literature. Additional searches were conducted to check if such a tool exists and none were found.

In consultation with a colleague, a research psychologist and PhD student running the broader Stigma in Genomics study with Xhosa RHD and Schizophrenia patients, I developed a tool that tested responses to living with RHD, talking about RHD, and the importance of sharing about RHD in group settings. Given that the evaluative study had a pre- and post- component, the questionnaire had to test general attitudes, opinions and responses rather than focus on specific details about the vignette. In other words it could not include any questions which referenced the story participants were about to be exposed to given that I wanted measure the change in responses before and after the stimulus.

2.3.5.2. Testing and Revising Questionnaire

When the first draft of the questionnaire was completed, I conducted one focus group with 14 participants using cognitive interview methods. This process helped to ensure that questions were clear and comprehensible to the study population. Participants involved in the cognitive interviews were of a similar background in terms of language, race and socio-economic demographics but they did not have RHD. To recruit participants I drew on people from my own social network who I knew shared similar demographic background of the patients enrolled in the RHDGen study.

At the outset participants received an explanation about the study, the structure of the study, and the purpose of the cognitive interviews. Participants then watched one of the videos produced for the FGDs. The questionnaires were then handed out and participants were given a chance to read through each item in the questionnaire. I read through each item individual

asking participants if they question was clear, if they could explain to me what they thought each item meant when they heard it being read and if there was a phrase or word that they did not understand. At first, participants thought they needed to provide opinions about the actual statement. I then re-explained that they needed to simply feedback what they had understood by each statement.

The session was recorded on a mobile device. After the session, I listen to the recording and made the changes participants suggested. Most of the suggestions were about changing words which were unclear. Most of the items were understood by participants as they could feedback what was meant by the item.

2.3.5.3. Administering the Questionnaire

Following finalisation of the questionnaire, I started the FGDs. Prospective participants were invited to participate in FGDs relating to stigma and genetic attribution of RHD. After signing a consent form, the before-questionnaire was administered to all participants present. Participants were not told that there would be a subsequent after-questionnaire and were just asked to complete the questionnaire given to them. The questionnaire also asked participants to include basic demographic information and two questions about RHD. The forms were anonymous so participants could feel more comfortable answering questions more honestly. Each participant was given a number from 1-12 before the initial questionnaire was administered. Participants were asked to write their numbers on both before- and after-questioners which would allow participants to remain anonymous but also allowed the researcher to match the before- and after- questionnaires.

Each item was read out in English and Afrikaans. Participants were also asked if they understood each item that was being read. The 1-4 Likert scale was explained, with each option from ‘Strongly Disagree’ which represent 1 to ‘Strongly Agree’ which represented 4. After completing the before-questionnaire, it was collected. Depending on the intervention,

participants were told they would either watch a video or listen to vignette with questions. Participants were told that questions would not be discussed immediately but at a later time during the session. After the stimulus was provided, participants were given the post-questionnaire. Participants were given the same instructions for completing the after-questionnaire. Each item was read aloud in English and Afrikaans and participants were given the opportunity ask questions.

It is important to note that given the age of the patients in the group, there were a few who had eyesight problems and could not read. In these cases, either I or my research assistant would sit with the person and ask them which score they would give to each item.

2.3.6. Data Handling and Record Keeping

The before- and after- questionnaires were stored together with consent and demographic forms in a file that I kept at the University of Cape Town. A purpose-designed database was created using Epi Info and used to capture the data by the researcher. The software was also used to analyse the data. The files were stored on the researcher's password-protected computer and was backed up on an external hard drive.

2.3.7. Analysis

The questionnaire data

The primary purpose of using the questionnaire data was to measure if there was a difference in response rates between groups who received a video or read vignette. In order to map any difference between the two groups we had to be able to compare the difference in scores between the two groups. In order to do this, the data for all questionnaires were divided between video and read groups. Within each group all of the individual scores for each were added and the average score for each item, for before and after scores, was calculated. It was important to compare each item and calculate averages for each individual questionnaire

given that each item measured a specific domain. The average scores for each before and after item were used to calculate the difference between the scores. The average difference for each item were used to compare the groups.

FGD transcripts

Another approach used to evaluate whether the use of visual media was superior to text-only vignettes, was to compare the differences in the percentage coverage for each group, when the same coding scheme was applied. This method was by Collier in his studying evaluating the use of photographs in in-depth interviews. While he describes this process in vague details he notes that created a coding scheme using the content to create 10 codes which "supplied [them] with an approximate statistical picture of our results" [15]. I tried to develop a similar test. The details of how the coding was developed can be found in section 2.4.8 of this chapter. NVivo 11 was used to store and code each transcript. One of the functionalities of this software is that it calculates the percentage coverage for each code [72]. Figure 1.2 below is a screen shot from NVivo displaying the percentage coverage for the child code 'General understanding':

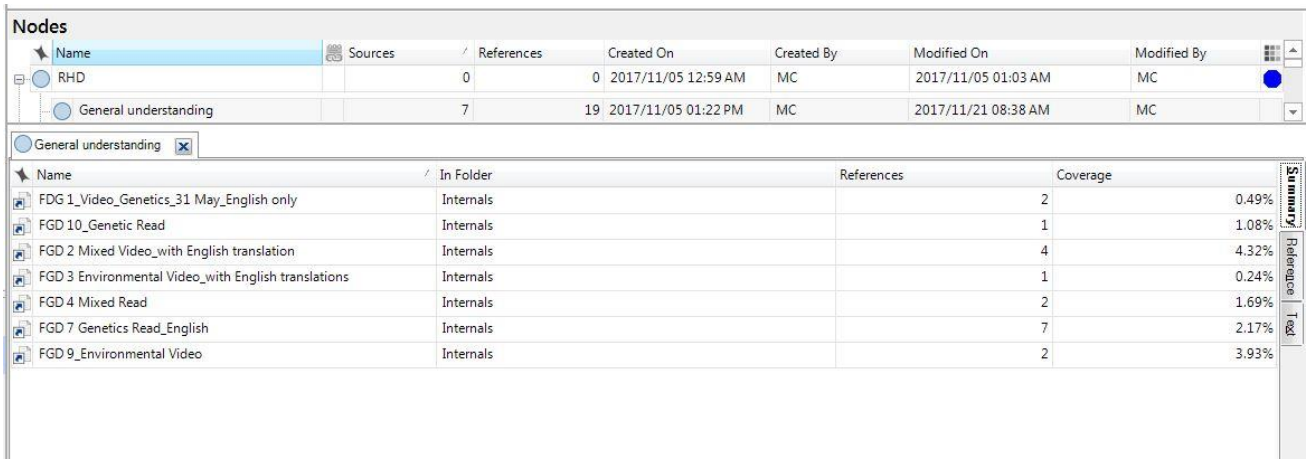

Figure 1.2: 'General understanding' code percentage coverage per FGD transcript

After the qualitative analysis for Study 3, I chose codes which were most prominent in my analysis. I added the percentage of coverage for each parent code, across all 11 transcripts stratifying by video or group. I then calculated the average percentage coverage for each parent code, for both video and read groups. Lastly I compared the average percentage of coverage for each code between video and read groups.

Field notes and researcher observations

I also used field notes to track differences between the video and read groups. All of three included papers in the systematic review (Study 1) used field notes as a tool of evaluation. A research assistant was present in each FGD and primarily played an observational role. We both made notes of the FGDs in relation to the content as well as any observations relevant to the comparison between stimuli. These notes were not used as a primary source of data but rather to support interpretation of results from the questionnaire and transcript data.

2.4. STUDY 3: STIGMA AMONG RHD PATIENTS: EFFECTS OF GENETIC KNOWLEDGE ON STIGMA

2.4.1. Introduction

This study focussed on exploring the relationship between stigma and the genetic attribution of disease with RHD patients in the Western Cape. Focus group discussions, using vignettes about a factious character who develops RHD, were used to explore this question. A theoretical thematic analysis was used to analyse data generated from FGDs.

2.4.2. Study Design and Setting

Focus group discussions can be defined as “interactive discussion between six to eight pre-selected participants, led by a moderator and focussing on a specific set of issues” [73]. By using FGDs, I was able to specifically explore how various people experienced living with RHD within a particular social and cultural context to which this group belonged.

Reasons for using qualitative methods were (1) methods reported on in this thesis were set by the larger project and, (2) there is very little evidence published about the relationships stigma and genetic attribution in African contexts. Thus, qualitative methods help exploratory methods to understand this relationships before creating clear hypotheses which can then be tests using other approaches such quantitative methods

After piloting, the vignettes were turned into short videos with the help of a professional film maker. Each story is told using a voiceover, and questions related to each scenario are displayed at the end of each scenario. These questions were used as a guide to structure the discussion. I played or read the first part of the scenario and then paused at each questions for discussion. Once all the questions had been discussed, we moved to the next scenario. Importantly, participants were first asked to complete the before and after questionnaire discussed in section 2.3 before actually having the group discussion. Figure 1.3 below depicts the structure of the process:

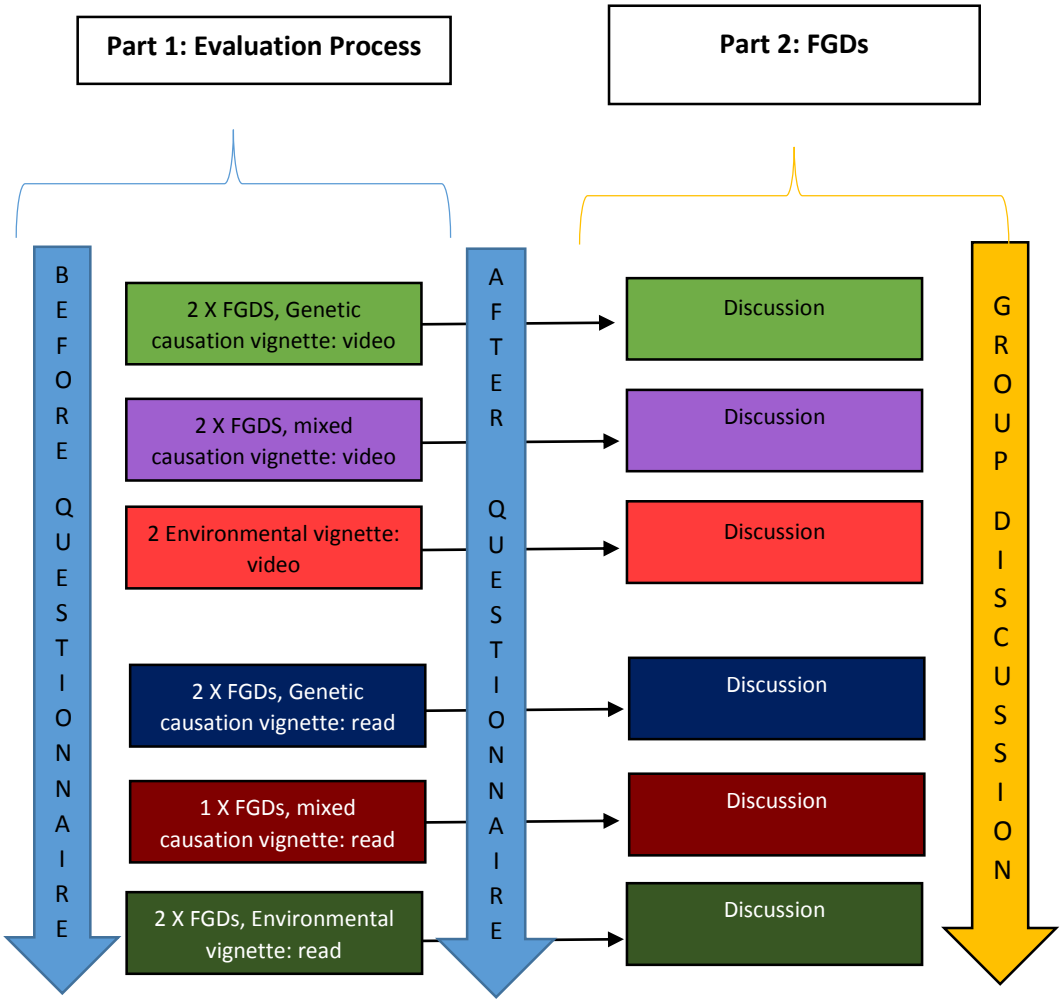

Figure 1.3: Schematic representation of evaluation process and focus group discussion

Because this project was interested in explored the impact of stigma associated with the genetic attribution of RHD, three casual models were explored. Each of the FGDs focussed on one of these three causal-model. The first casual-model explored the role of genetic attribution of RHD. The second causal-model drew on both a biogenetic explanation and environmental explanation. Here the scenario explores how Enrico may have developed RHD as a result of a history of RHD in his family and his impoverished living environment. The third casual model focusses the environmental causation only. Figure 1.4 illustrates the structure of each FGD and causal model explored:

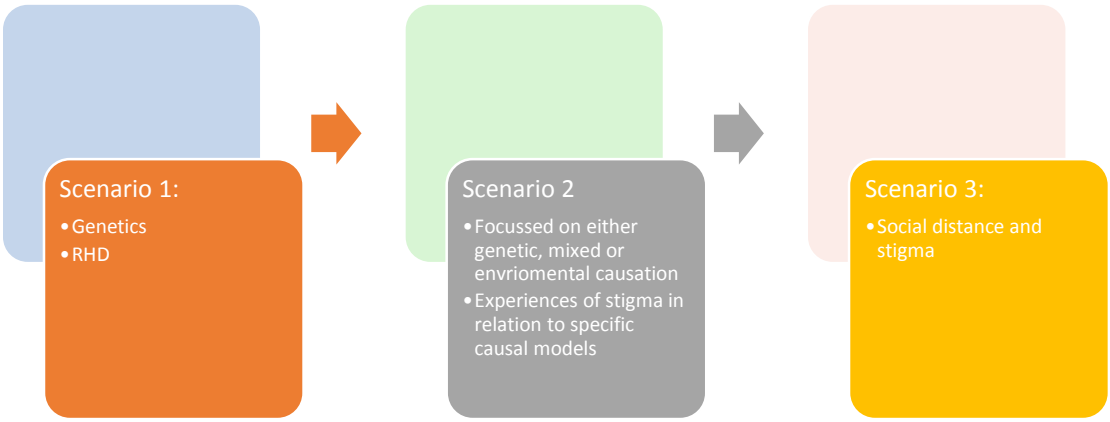

Figure 1.4: Structure of FGDs

Each FGD started with a discussion of the disease of RHD which investigated topics such as disease causation, consequences and severity of the condition, effect of the disease on the person and their social lives, and stigma. The FGD also explored culturally specific understandings of illness, and where possible tensions in the groups between traditional and modern views. After introduction of the vignette, the FGD topic guide first explored participants' associations with or understandings of genes, genetics and disease causation by exploring the associations that come to mind upon hearing 'genes' [74]. It then explored participants' feelings about the likelihood that the patient's condition (as represented in the video) can be remedied through treatment and so forth. Lastly, I explored the consequences of genetic attribution for a desire to maintain social distance [12], which has been used as one means of measuring the effect of genetic attribution on stigma. This component of the FGD topic guide explored questions like "Would you be happy for Patient X to marry your child/ be the parent of your grandchildren", "Would you be likely to be friends with Patient X" and so forth (see Appendix 2 for screen shots of videos).

2.4.3. Specific Objectives of the Population-based Study

The primary objective of this study is:

a) To investigate the role of genetic attribution of RHD on levels of stigma

2.4.4. Sampling Procedures

While the same participants who formed part of the empirical study described in section 2.3 were also enrolled in this study, it is important to note the following information pertinent to the sampling process for the FGDs.

Initial recruitment drew on the RHDGen database, meaning that all those participants had previously consented to participation in a genomic study. On that basis, it was likely that participants had some basic understanding of genetics and/or genomic research. Similarly, many of these initial participants would also have been involved in patient awareness days where talks and information sessions were offered on RHD and genetics. Moreover, some of the participants would have been part of the patient advisory group that was established in 2016 and would also have exposed them to some of this research.

However, I had to include other databases of RHD patients given that many of the patients in the RHDGen database, had either changed their contact details and not informed us, could not make any of the scheduled FGDs due to poor health or work, or were deceased. In total, for this project I recruited about half (n=30) participants who had previously enrolled in RHDGen. It was not clear before the FGDs what participants recruited from these databases understood about genetics. Nonetheless, the FGD guide explored general understanding

of genetics at the outset of the discussion. When analysing the data, we did not observe a major difference between the FGDs conducted with people who had previously enrolled in genomics and those who had not. I progressively distributed participants across of each of the 3 FGD scenarios and the video/read methods. This allowed for participants to be spread amongst those participants who had previously been enrolled in genomic studies.

2.4.5. Data Collection and Handling

2.4.5.1. FGDs

Over the period of March to November 2017, I conducted 11 FGDs. While we aimed to for each groups to have a minimum of 6 and maximum of 12 participants, the average number of participants in one FGD was 5 with 4 being minimum and 9 being the maximum. While it was our intention to have at least 6, participants would often cancel on the day owing to poor health or transportation challenges given that most participants had to use public transport to get to the hospital. Despite this though I found that FGDs with a smaller number of participants worked well in terms of fostering a lively discussion about the study. The FGDs lasted on average for about 90 minutes. Each participant was given a number to use when they spoke. This was done so participants could remain anonymous but their voices could still be distinguished when the discussion was later transcribed. There was also an observer present in the FGDs who took notes during the discussion. After the discussion the observer and I reflect on the FGD and shared insights, observations and reflections. The notes from this were typed up by the observer, and these field notes were a data source in their own right.

2.4.5.2. The FGD Guides

Discussion guides in FGDs are important for the research as they act to provide structure to explore a particular topic [73]. When I joined this study, the guide was already developed as part of larger project (see Appendix 3). The guide explored three areas, namely general understanding of genetics and RHD, social stigma and associative stigma. After the guide was developed, a video depicting the vignette was created for the larger study and voiceover were originally done in IsiXhosa with subtitles appearing in English (see section, 2.4.1. for full description of guide).

In the study reported on in this thesis, the same topic guide and videos were used. However given the coloured population speak Afrikaans and English, I translated the three vignettes to Afrikaans and then had these changes made to the videos (through an Afrikaans voiceover and English subtitles). I also changed some minor details in the initial vignettes to be more culturally relevant to this population. These changes primarily related to the character's name and where he lived. In the groups where the vignettes were read, participants were given the option of hearing the vignette in English or Afrikaans or both.

2.4.5.3. FGD: Transcription and Translation

The FGDs were transcribed by an independent transcriber who was not part of the FGDs. The person first transcribed the FGDs verbatim, in both English and Afrikaans depending on the language of the FGD. The transcriber, who is fluent in English and Afrikaans then translated Afrikaans text to English. Once I received the transcripts, I randomly checked the quality of the initial transcription against the recorded FDGs and the quality of the translation.

2.4.6. Data Handling and Record Keeping

The FGDs were recorded on two separate audio recording devices and were then downloaded onto a laptop and password protected cloud storage drive as a backed-up of the recording. Notes were taken by the observer present in the FGDs, which later typed up and sent me via email.

The completed transcripts were saved on a password protected laptop and saved on an external hard drive. The transcripts were imported into NVivo11 software [72]. NVivo11 was used to code all of the transcriptions [72].

2.4.7 Analysis

A thematic analysis was used to analyse the data, with all codes being derived from the data. Thematic analysis is described Braun and Clarke as a “method for identifying, analysing and reporting patterns (themes) within data” [75]. They describe a 6-stage process for thematic analysis [75]. The first step involves becoming familiar with the data, the second step requires generating first-level coding, the objective in the third step is to look for relationships between codes and generating themes (including second-level coding), the fourth step involves reviewing and refining these themes, in the fifth step the themes should be named and defined, in the final step requires the research to be written up [75].

I have generally followed these steps in my analysis. To immerse myself in the data, I listened to the audio recording of the each FGD twice before starting the initial coding process. I then used three transcripts, one for each causal-mode, doing line-by-line coding to develop first level of coding. My supervisor also used the same set of transcripts to develop a coding scheme. Once we met and compared coding schemes, I went to the data to develop a hierarchical scheme based on our discussion. Once I

drafted this scheme, my supervisor and I met again to discuss the hierarchical coding scheme. I then applied this hierarchical coding to a fourth transcript which was not used to develop this coding scheme. After this, I reviewed the coding scheme. I then developed themes based on the coding scheme. We met again to review the themes in relation to the data and codes, after which I went back to refine the themes once more before then applying it to the entire dataset.

In relation to deciding on codes and themes, I specifically drew on what Braun and Clarke term “theoretical thematic analysis” which they differentiate from more inductive thematic approaches [75]. They argue this approach is less interested in describing the scope of the data but rather in focussing on specific aspects driven the researcher’s questions and interest. One of the reasons this approach was important for this project was because questions related to stigma and the genetic attribution for disease was at centre of the NIH-funded study. Another reason this specific question was foregrounded in the analysis was because of the lack of research related to this question set in African contexts. Given that there is growing body of literature related to stigma and the genetic attribution of disease, much of this theory has been formulated in relation to psychiatric research in high-income and Western contexts.

3 ETHICS

Ethics approval for the study was given by UCT’s Faculty of Health Sciences Human Research Ethics Committee prior to commencement of the study. Research will take place in accordance with UCT’s Research Ethics Policy, the Declaration of Helsinki (Update 2013) and Good Clinical Practice guidelines. All participants gave written informed consent to be part of the study (see Appendix 4).

3. CHAPTER 3

STUDY 1: ARE VISUAL METHODS MORE EFFECTIVE IN COLLECTING QUALITY DATA FOR QUALITATIVE RESEARCH? A QUALITATIVE SYSTEMATIC REVIEW

3.1 BACKGROUND

The use of visual material in qualitative and mixed method studies is increasingly common, partly due to an oft-repeated assumption that such material generates more discussion and richer data in comparison to text-only based methods; yet, there is limited evidence to support these claims [2]. Reproducible evaluation methods allows for the testing of visual material in qualitative research as an effective stimulus for discussion and richer data generation. This study aimed to fill this gap by, first, presenting the results of a systematic review examining evidence relating to the efficacy of visual methods in fostering the collection of good quality data for qualitative research. Secondly, the chapter will summarise evidence about how the efficacy of visual methods have been evaluated.

Visual materials have been incorporated into a variety of qualitative studies across multiple disciplines. Visual material can generally refer to drawings, pictures or photographs, and audio-visual materials such as videos. Historically, the use of visual methods in research is rooted in anthropology and subsequently in sociology from the mid-1900s [76]. Researchers involved in early studies argued that visual methods could capture experiences and phenomena in ways that text-only methods could not [76, 77]. Since then, the use of visual methods in research has become more prominent across various disciplines such as psychology, education, geography and health sciences [2] One of the more common ways in which visual methods have been utilized is as data to aid analysis as part of the research process. For example, as part of research on experiences of illness, Guillemin asked women with various heart conditions to draw how they experienced their illness [76] .

Another popular visual method which is increasingly gaining popularity is photovoice and refers to a participatory method where participants are given cameras or video cameras to capture their lived realities and direct the research process. In many instances participants would bring the images or video recordings to an interview or focus group to discuss what was captured [78, 79]. This method is now well established and has been used extensively across various geographical contexts [80-82]. Furthermore, these methods are often used to help empower marginalised groups. A study by Foster-Fishman et al. found that the use of photovoice has a significant positive impact on research participants [83]. In their study, interested in experiences of community life, they asked residents to take photographs capturing meaningful experiences of living in particular neighbourhoods. Participants were also asked to reflect on specific questions related to their photographs and were subsequently discussed in focus groups. Through interviewing participants about their experience of taking photographs as part of the research process, they found that participants had great levels of self-confidence, increased awareness of their environment, and developed resources for social and political action.

Other ways visual methods have been used, include the use of visual material as a stimulus to prompt participants to share their experiences. Epstein et al., in a study assessing the therapeutic efficacy of campsites on children with cancer, used pictures of campsites during in-depth interviews to stimulate conversations between the child and the researcher. They found that showing children pictures of campsites helped children engage more deeply with their perceptions of campsites as being therapeutic [84].

There are multiple reasons why visual materials are used in qualitative research, but most often authors justify their use of visual materials describing their conviction that traditional methods cannot yield the same richness that visual methods [85]. For example Keller justifies the use of visual material by presenting the argument: "…to both foster and move qualitative inquiry beyond the verbal…" [86]. Additionally, there has been some attempt to evaluate the

efficacy of specific visual methods. Meo, through her study on identities among Argentinian high school leaners, states that her interviews where photographs were used, were much longer than the interviews without the use of images. She interpreted this to mean that the use of photographs have greater potential to stimulate richer discussion [16]. Additionally, Samuels interviewed monks using photographs her participants generated. She noted the interviews using photographs were much longer and generated quantitatively more data than interviews which did not use visual material [87]. However, there is very little evidence to support such claims and no empirically reproducible methods have been proposed to evaluate the effectiveness of the use of visual material to stimulate discussion and generate richer data in qualitative research [2].

3.1.1. Why is this review needed?

Evidence based methods refers to the meticulous, clear, and prudent use of evidence in making decisions about research, incorporating both individual knowledge and proficiency, and the most reliable evidence. A critical tool of evidence based methods is the systematic review, which is a method that summaries evidence with the goal of minimising bias through systematically identifying, evaluating and integrating all relevant studies related to a particular topic according to a method made explicit at the outset of the review. A key objective of the systematic review is to highlight areas where there is limited evidence, allowing researchers and funders to plan relevant primary studies [88]. Additionally, systematic reviews are intrinsically designed to reduce bias in evaluating existing research, allowing a pathway for key stakeholders and decision-makers to have evidence related to specific questions [88]. Critically, systematic reviews require considerable less resources in comparison to primary studies [88].

Here, I present the results of a systematic review aiming to evaluate the efficacy of using visual material to stimulate discussion and generate richer data in qualitative research. In addition, I

summarise how these studies have evaluated the use of visual material. To date no systematic review has evaluated the efficacy of visual materials in qualitative research.

3.1.2. Objectives

There are two outcomes for this systematic review. The first is to evaluate the efficacy of using visual material to stimulate discussion and generate richer data in qualitative research. The second is to summarise how the efficacy of visual materials in qualitative research has been evaluated.

3.2. METHODS

3.2.1. SEARCH STRATEGY AND STUDY SELECTION

Types of studies

Studies were included if they aimed to evaluate the effectiveness of visual methods in enriching data generated through discussion. Thus, we considered qualitative studies as eligible for inclusion.

Types of participants

Any participants participating in a related research study.

Types of interventions

Studies were included if they used one or more of the following:

For studies to be included, it needed to have used any visual (photographs, drawings, other image media) or audio-visual material (recordings or videos) that was either provided by the researcher or generated by the participant as a stimulus for discussion within a qualitative

research context, such as an in-depth interview or focus group, to stimulate richer discussion and data collection.

Studies needed to have evaluated the use of these visual methods to generate data. We were not strict about the method evaluation but rather that some kind of evaluation component was documented in the study.

Comparison

Some studies may have included components where no visual material was used to stimulate discussion or research may have read vignettes to participants to stimulate discussion without the aid of visual materials.

Types of outcome measures

Primary outcomes

Improved / enriched discussion (defined as contributing to longer length of discussion OR greater variation or increase in the volume of coded data generated).

Secondary outcomes

Evaluation methods employed in assessing the effectiveness of visual methods

3.3. SEARCH METHODS FOR IDENTIFICATION OF STUDIES

3.3.1. Electronic searches

We used a comprehensive search strategy. There was a high probability of retrieving irrelevant papers given that we were interested in methods rather than specific research content. One reviewer tested the strategy to check if it retrieved relevant papers. We searched in title and

abstract only for visual methods search strings which used words such as “photo-elicitation” or “photo-interview”, and qualitative methods search strings to include words like “focus group” or “in-depth interview” and evaluation search string to include words such as “evaluate” or assessment”. (See Appendix 5 for the complete search strategy). When we included the evaluation search string, we searched in the full text since we did not want to miss any studies which have evaluated their methods but failed to mention this in the abstract or title. This was also important given that we searched in a variety of disciplines where there are not necessarily standardised norms to write abstracts with different kinds of information being included across various disciplines and journals.

Since this review is a methodological review, multiple databases from a number of disciplines were included in the search. In total, we searched in 10 databases:

- Academic Search Premier
- Business Source Premier
- Africa-Wide Information
- CINAHL
- Communication & Mass Media Complete
- EconLit
- ERIC
- MEDLINE
- Psych Info
- PubMed

We did not limit the search to any particular date or language, although we only searched using English keywords and search strings.

3.4. Results

3.4.1. Search Strategy

Two reviewers independently selected relevant papers based on title and abstract. The initial search retrieved 992 papers which, after removing duplicates (n=871), resulted in a total of 425 hits. After applying the inclusion criteria, a total of 201 papers were retrieved for full text analysis. A search of the reference lists in these manuscripts yielded one additional paper for inclusion. Another paper was included which was found referenced in one of the included papers. See Figure 3.1 for summary of searches:

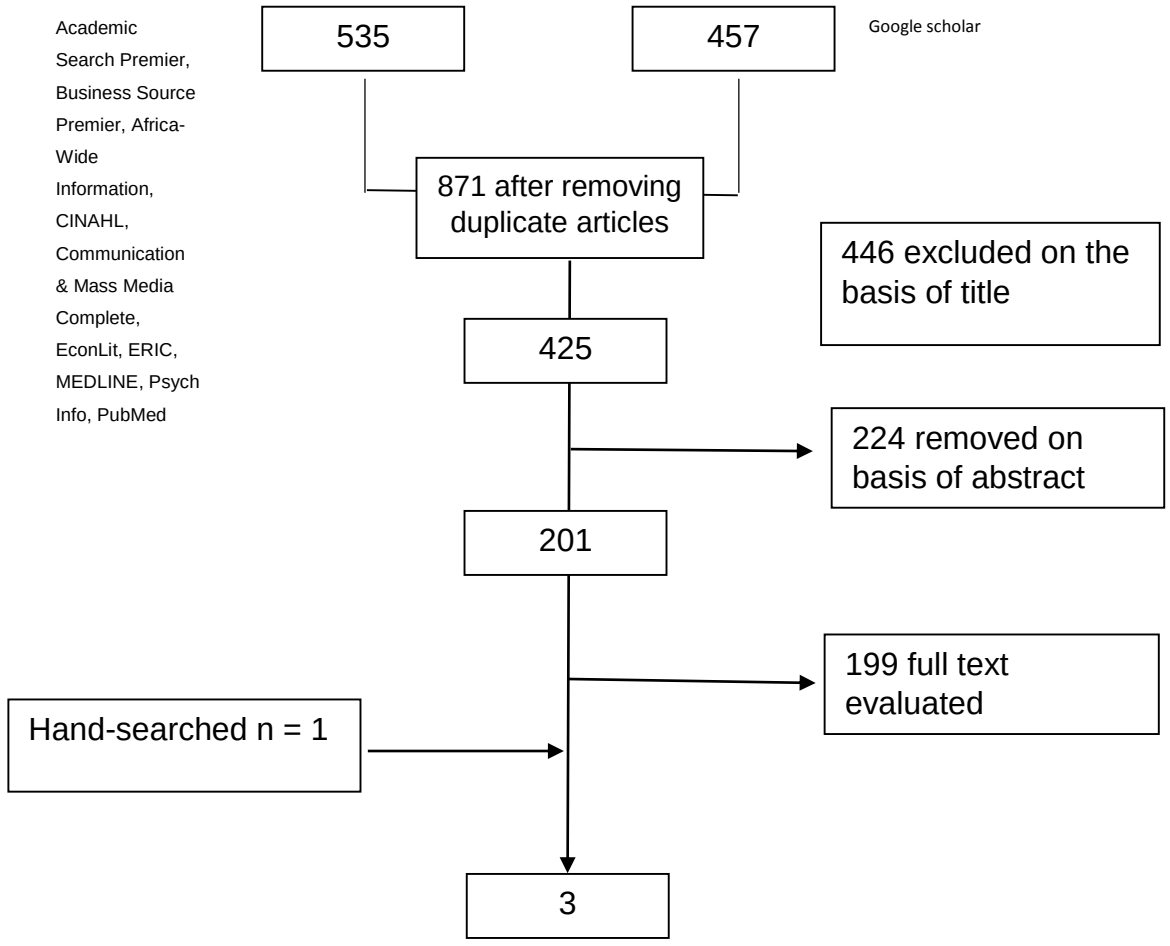

Figure 3.1 Flow diagram of search results

3.4.2. Included studies

Description of studies

After applying the inclusion criteria, three papers were included in the systematic review. Two of these were published in 2010 [16, 17] whilst the third, was published in 1957 [15]. Table 3.1 summarises the characteristics of the three included studies:

Table 3.1: Summary of included studies

	Collier, 1957	**Meo, 2010**	**Cooper and Yarbrough, 2010**
Population	French speaking Acadian families living in Bristol, Canada	15 to 17year old high school students in Buenos Aires, Argentina	Older women who help other women during childbirth in Rural Guatemala
Intervention	Structured interviews questions plus photos (research generated)	Structured interviews questions plus photos (participant generated)	Structured questions used in FGDs plus photos (participant generated)
Comparison	Structured questions alone	Structured questions alone	Structured questions alone
Outcome	Data richness measured by interview length and coding density	Data richness measured by interview length	Data richness measured by interview length

The Collier (1957) study describes and evaluates the effectiveness of the use of photographs in interviews [15]. Collier, an anthropologist, conducted an experiment to evaluate whether photographs are effective prompts in in-depth interviews. He conducted three thematic interviews with each of the participants, interested in exploring how French-speaking Acadians would assimilate into their English speaking environment, where many of them worked – this was done in relation to their physical environment. The participants were divided into two groups answering structured questions, with the difference that one group utilized photographs (taken by the principal researcher) in addition in the interview process. At the end

of both sets of interviews, the participants who were interviewed without the use of photographs were also shown the photographs to explore if they still felt the same way.

The second study by Meo was published in 2010 [16]. As part of a larger study aiming to evaluate how children are socialised in relation to class, education and identity in two schools in Buenos Aires, Argentina, the author included an evaluative component exploring whether the use of photographs in interviews elicits richer data compared to interviews which did not use photographs. Unlike Collier's study where the researcher provided the images to use in interviews, Meo asked participants to generate their own photos which were used to stimulate discussion in the subsequent interviews. She enrolled twenty students who were initially interviewed without any images. She then gave them each a camera with which to take photographs at school and at home. She then did a follow-up interview, using the photos they had brought to the interview [16].

The final study by Cooper and Yarbrough was also published in 2010 [17]. This was part of a larger study investigating how social and economic factors determine health outcomes in Guatemala. Unlike the previous two studies which employed in-depth interviews, this study used focus groups to collect data. There were two phases, the first used a traditional focus group approach with a text-only topic guide. Specific participants were then selected and asked to take photos of their surroundings. These specific participants were the invited to another focus group where their photos were used to further explore questions relating to how their environment determines their health.

Findings

Given the variability across the studies in terms of objectives, and in different academic disciplines, we were unable to conduct quantitative analysis, and thus, present a qualitative systematic review having utilized detailed, rigorous and explicit methods.

This systematic review found evidence for the use of visual material stimulating discussion and yielding richer data in qualitative studies. The evidence reflects that interviews using photographs allowed people to speak more specifically and clearly about what they were being asked, whereas in verbal-only interviews participants often provided non-relevant information. Second, authors report that interviews using photos were longer in duration than interviews which used text or verbal only methods. This implied that visual material stimulates richer discussion as compared with traditional methods. Finally the results show the use of visual methods allowed unexpected (and relevant) information, responses and emotions to emerge.

In addition, this systematic review also found evidence for the efficacy of visual methods beyond the two defined outcomes. First, the use of visual methods created empathic distance between the participants and the content of the research. That is, the images allow the participants to reflect at a distance about their personal experience [15].Second, in the interviews that used photos, photos allowed participants to have more control over what was discussed in the research process [16]. Lastly, the use of photographs allowed participants to explore abstract concepts and make connections between abstract concepts and their experiences [17].

The following table summarises how each study evaluated their findings:

Table 3.2: Summary of evaluation methods

Collier, 1957	**Meo, 2010**	**Cooper and Yarbrough, 2010**
Researcher observations	Researcher observations	Research observations
For interviews that did not use photos, an additional interview was done using photos – this interview was longer and more substantial than the previous interviews.		
10 codes were used to code all transcripts – no significant difference was found in terms of number of responses but there was significant difference in the quality of responses, with the photo-interviewing yielding much richer data.		

Evaluation Assessment of the reliability of the evidence

The included papers in this review have provided evidence to support claims that the use visual of methods are effective at stimulating discussion and generating richer data. However, the methods used to evaluate these claims need are not sufficiently rigorous. Two of three included studies provided insufficient details about the evaluation methods used to substantiate their findings and therefore makes it difficult to generalise such claims [16, 17]. Moreover, 2 of 3 studies relied solely on researcher observations as evidence of the efficacy of visual methods in eliciting richer data [16, 17]. Evaluations based on observations may be biased and call into question the validity of findings. Additionally, studies used different outcomes measures to test for efficacy which makes results difficult to compare. These factors calls into question the overall validity of the evidence presented in this review and it is unlikely such results can be generalised.

3.4.3. Excluded studies

The primary reason why most studies did not meet the inclusion criteria (n = 165) is as a result of these studies not evaluating whether the use of visual material were effective. Other papers were excluded either because they were not empirical (n = 18), or they were not conducted in a qualitative research context (n = 11), or they were evaluating the efficacy of photovoice as a research empowerment tool (n = 3) or finally because they were not be relevant (n = 2).

3.5. DISCUSSION

This study has found evidence to support claims that visual material is more effective than text-only materials at stimulating discussion and richer data in qualitative research. All three studies, which used both participant- and researcher-generated interviews, found that visual media allowed participants to make connections they did not make where visual methods were not used, and that participants were able to provide more relevant information about the specific questions being asked. Collier found, during his in-depth interviews which used only text-based methods, that participants would often provide a great deal of irrelevant information [15]. Both Collier and Meo suggest that the reason for this is because the visual media allow participants to focus on something specific when being asked questions [13, 15].

What remains unclear however is how these studies have defined what they mean by ‘rich data’. Indeed they all make the claim and show evidence to support their claim that using visual media allows for richer data collection but they all fail to define what is meant by richer data collection. This question becomes important when findings rely on comparing one method versus another. While we agree that determining what is meant by richer data in qualitative methods is necessarily subjective, researchers should nevertheless be able to define what rich data in their study means in order to allow for a comparison of two datasets. Failure to define what is meant by ‘richer data’ does not mean that researchers do not have a preconceived idea of what it is. For example, in their discussion on the efficacy of photo-

elicitation to help participants make abstract connections and articulate complex ideas, Copper and Yarbrough note that “Photographs show the action and show or suggest the context… This might be one of the reasons we found the data from phase two to be richer, more reflective, and more contemplative” [17], suggesting that ‘richer data’ for them meant data that is more reflective and contemplative. Similarly Collier based his claims on richness on data being more relevant to questions asked by the researcher and did not consider information provided by participants which were not related to the question as ‘rich data’ [15]. However, in both papers these definitions of what was meant by richer data was not mentioned previously when comparative claims between the two groups were being made, and were apparently not explicitly considered in the design of the evaluation strategy.

There were two objectives of this study. The first was to summarise evidence to either support or reject claims that the visual of media in qualitative research is more effective at stimulating discussions and yielding richer data. This is especially pertinent given that it is an oft-repeated yet rarely substantiated claim in qualitative research [2]. Most of the papers that were excluded from this study did not include any evaluation component. This was found for papers across disciplines using a variety of visual methods and related to both participant- and researcher-generated images and other forms of visual media. Evaluating qualitative research methods is now an established practice [89]. The same rigour must be applied to novel methods of research, especially as technology is increasingly facilitating these novel methods.

The second objective was to summarise how researchers evaluated the efficacy of visual methods. While all three studies were explicitly interested in testing assumptions about the use of visual material and included an evaluation in their research design, only Collier used and reported on various mechanisms he used to evaluate these assumptions [15]. Both Meo and Cooper and Yarbrough seem to rely on their observations although they do not report directly on what they base their claims on [16, 17]. Meo, for example refers to interviews using photos being longer [16], while Cooper and Yarbrough refer to participants making abstract connections when photos are included [17].

Another reason why it is critical that evaluation methods are made explicit is that there may be differences when participants generate their own images as opposed the images or visual material being provided by researcher. In both papers which did clearly report on they evaluated which method was more effective, the participants provided their own images which could also imply that participants may have been more inclined to speak about their lived experience through the images they captured. While this observation does not relate evaluation methods directly, it is important as it contextualises the evidence.

This systematic review has been limited primarily by the scope searching. While a comprehensive search strategy was developed, many of the databases searched did not have the functionality to use multiple search words or search strings to simultaneously search though catalogued articles. It is therefore possible that some papers were missed during searching. Additionally it was also beyond the scope of this review to search grey literature. The primary reason for this is that a methodological review of this nature which spans across a variety of disciplines would require a great deal of time and resources to find and locate relevant studies which are not yet published or searchable.

Implications for practice and research

While the evidence seems to suggest that the use of visual materials aids in generating richer data in the qualitative research environment, we suggest caution given the lack of rigour applied in the evaluation in the studies included in this review. There certainly is scope for further studies to address this topic. We recommend incorporating well-defined outcomes evaluation in assessing the evidence.

CHAPTER 4:

STUDY 2: GENERATING RICHER DATA IN FOCUS GROUP DISCUSSIONS? AN OBSERVATIONAL STUDY COMPARING VIDEO- AND TEXT-BASED APPROACHES

4.1. INTRODUCTION

This chapter builds on Chapter 3 (Systematic Review). Briefly, visual methods in qualitative research are increasingly popular across various disciplines [90]. Rooted in anthropology and sociology, visual methods were first used in the mid-20th Century. In broad terms, this approach can be defined as using visual material (photographs or audio-visual material) to generate discussion and can include for instance photo-elicitation or the use of photographs or drawings to stimulate discussion. Visual material can be provided by the researcher or be created by research participants [91].

One illustration of the enormous popularity of visual methods in qualitative research is the establishment of journals specifically dedicated to the use of visuals methods in social science and humanities, including titles such as Visual Studies, Visual Anthropology, Visual Anthropology Review, Visual Communication and the Journal of Visual Culture [92]. There has also been an increase in membership of organisation specifically focussing on using visuals method. These include the International Visual Sociology Association (IVSA), the recent British Visual Sociology Group, the ISA Visual Sociology Thematic Group, the Visual Communication Studies Division of the International Communication Association (ICA), and the International Visual Literacy Association (IVLA) [92]. Yet despite growing traction of the variety of visual methods, there still remains clear methodological gaps for doing research using visual methods. For instance, Pauwels notes that:

> Unfortunately there is little integration with respect to the findings and practices of visual methods, especially between the social sciences and the humanities and behavioural sciences. Visual methods, therefore, seem to be reinvented over and over

> again without gaining much methodological depth and often without consideration of long existing classics in the field. [92]

While Pauwels' discussion is specifically focussed on the use of visual methods as data in social sciences research, this kind of observation is equally relevant for how visual methods are used in traditional methods.

4.2. BACKGROUND TO THIS STUDY

Harper argues that methods which incorporate photo-elicitation versus interviews alone, are more superior because it stimulates parts of our brains which are older and more adept to responding to visual cues as opposed to words only [77]. There are many other reasons why visual methods are used by researchers and why such methods are increasingly gaining popularity [90]. Moreover, a literature review by Pain shows how researchers use such claims to justify their use of visual methods [2]. However, our systematic review reported in Study 1 (Chapter 3) of this thesis, revealed the scarcity of evidence to support claims that visual methods are more effective at generating richer data, thus supporting Pain's assertion that such claims remains largely unsubstantiated and further empirical evidence is required to test these assumptions.

In addition, our systematic review showed a lack of consistency in methods for evaluating the efficacy of visual methods. Of the three included studies, only one made explicit the methods used in its evaluation [15], while the other two studies relied on observations made by researchers during the study [16, 17]. As the use of visual methods becomes increasingly popular, it is important to evaluate such novel methods given the amount of time and resources the incorporation of visual methods may require. While some studies ask participants to draw, other studies may require the making of films or the purchasing and distribution of cameras which comes at great research costs. Given both time and financial commitment, it becomes important to evaluate the efficacy of such methods in relation to claims about the superiority of visual methods in eliciting richer data in qualitative research.

The study Stigma in African genomics research on Schizophrenia and Rheumatic Heart Disease, provided the opportunity to explore the efficacy of using film as a prompt in FGDs. The study used vignettes depicting a fictional character who develops RHD to explore the impact of genetic attribution on disease stigma. These films were to be used in focus groups to help stimulate discussions (details of the film can be found in the Methods, Chapter 2).

4.3. METHODS

Participants for this study comprised 11 focus groups in total. Five of the FGDs had the vignette read, while the remaining six watched the short film. I used three methods to evaluate how effective the use of the film was at stimulating discussion and gathering richer data in qualitative methods. Given that I found no validated evaluation tool to test the efficacy of visual material, I designed a comparative study to test these assumptions. I defined richer data as coding density or the frequency of codes that occurred in each transcript (as one way to define richer data). Given how challenging these methods are to evaluate, I used three different methods to triangulate approaches to ensure greater internal validity.

Specifically, I did the following:

1. Questionnaire

A before/after study design was used to measure the change in participants' responses and willingness to talk about their experiences of RHD before and after they either watched the video or listened to the vignette being read. Building on existing instruments in psychology and with the help of research psychologists in our team, I designed a short questionnaire that covered 6 domains, namely: general interest (of RHD); personal engagement; empathy; willingness to share in a group; and emotional responses. Each domain generally had three items (or: questions). Each item involved a statement about RHD relating to one of the above mentioned domains. The questionnaire used 1-4 Likert scale with 1 representing 'strongly agree', 2, 'disagree', 3, 'agree' and 4, 'strongly agree' (See Appendix 1 for the questionnaire).

The questionnaire was piloted with a group of people to the same racial population as the participants, and who came from a similar class background. The questionnaire was adjusted after piloting phase. It was administered before participants either watched the video or listened to the vignette being read. After the stimulus, I immediately administered the same questionnaire.

The data was captured on data managing software Epi Info and was then analysed in Stata and Microsoft Excel. The questionnaires were used in an attempt to measure differences in responses rates before and after a particular stimulus. All the answers for each of the 16 items in the questionnaire were aggregated for the 'read' and 'video' groups respectively. The average scores, for each 'before' and 'after' item were calculated, the difference or change in scores was calculate between each 'before' and 'after' individual score, for both video and read groups. I compared the differences in mean scores for each item, between the read and video groups. While the questionnaire used an ordinal 1-4 Likert scale, it was still helpful to compare before and after scores. While I could not statically calculate if these is a significant difference, given the ordinal scale, the value of this approach has valuable in seeing the distribution of change in scares when comparing these two stimuli.

2. Coding comparison

I also used the coded transcripts of each FGD to compare which group (read or video) generated richer data in relation to the discussion. As the results of the systematic review shows, one other study used the transcripts of the conducted interviews to compare between interviews [15]. In that case, the researcher counted the number of codes used in each of the transcripts and subjectively evaluated the quality of responses of each participant [15]. In my second approach to evaluating the quality of data generated, I adopted a similar approach. Using the statistics generated by NVivo11 [72], I compared the percentage coverage of selected themes per transcript. In order to measure 'richness', I conducted this part of the evaluation *after* I had written up my analysis of the data as relating to the research question (presented in Chapter 5). Looking at the analysis, I examined which themes in the data were

most informative in elucidating the study topic. I identified major themes in the data which covered important aspects of the analysis (comprising 6 parent nodes and 22 child nodes). I added all of the percentage coverages for each theme across video and read transcripts and compared the average percentage coverage for selected themes with respect to both video and read groups.

3. Field notes

I also incorporated the field notes I made immediately following the FGDs. During the FGDs, an observer was also present who assisted in managing the group and who also took detailed notes during the FGDs. These notes included observations about participants' reactions to either being read a vignette or watching a video. Specifically, I used my field notes in the discussion section to explain some of the results found.

4.4. RESULTS

Questionnaire Data

Out of the 94 questionnaires (47 participants each completing two questionnaires), there was a combined total of 736 data points (464 data points for video, 272 data points for read group) representing the change in individual score for each item on the questionnaire, total of 9 missing data points. Table 4.1 and 4.2 shows the change in scores for each questionnaire item, for video and read groups respectively.

Table 4.1: Video group: average change in scores before and after video stimulus

Mode	Q1D	Q2D	Q3D	Q4D	Q5D	Q6D	Q7D	Q8D	Q9D	Q10D	Q11D	Q12D	Q13D	Q14D	Q15D	Q16D
V	0	0	0	0	0	0	0	0	0	0	0	0	0	0	1	0
V	0	-1	0	0	0	0	0	0	0	0	0	0	0	0	0	-1
V	0	0	0	0	0	0	0	0	0	0	0	0	0	0	-1	0
V	1	0		0	1	0	0	-1	-1	0	0	0	0	0	0	0
V	0	1	1	0	0	0	0	0	0	0	0	0	0	0	0	0
V	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0
V	0	0	1	0	-1	0	0	1	0	0	0	0	0	0	0	-2
V	0	0	0	0	0	0	2	1	1	0	0	0	0	2	0	0
V	0	0	0	0	0	0	1	1	1	1	0	0	0	1	1	0
V	-1	0	0	0	0	0	0	0	0	0	0	0	0	0	1	-2
V	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	2
V	0	0	0	0	0	0	-1	-1	1	-1	0	0	0	-1	-1	2
V	0	0	0	1	0	1	1	1	0	0	0	1	1	1	2	0
V	0	0	0	-1	0	-1	0	0	1	0	0	0	0	0	0	0
V	0	-1	1	-1	-1	-1	-1	-1	-1	0	0	0	0	1	2	-2
V	0	0	-1	0	0	0	0	0	0	1	0	0	0	0	0	0
V	0	1	0	0	1	0	1	0	0	-1	-1	1	0	0	0	0
V	0	0	0	0	-1	1	0	0	0		-1	-1	0	0	0	0
V	0	0	0	0	0	0	3	1	0	0	0	0	0	0	-1	0
V	0	1	0	1	0	0	0	0	0	-1	-1	-1	-1	-1	-1	0
V	0	1	1	0	-1	0	-1	-1	1	0	0	1	0	0	0	1
V	0	0	0	0	0	0	2	1	0	2	1	0	0	1	1	3
V	1	1	0	0	-1	0	0	1	1	0	0	-1	0	-1	-2	0
V	0	1	0	0	0	0	1	0	0	-1	-1	-1	0	-1	-1	1
V	0	0	0	0	0	1	0	-1	0	1	0	0	0	-1	0	0
V	0	0	0	0	-1	1	0	-1	-1	0	-1	-1	1	0	0	0
V	0	0	0	0	-1	-1	-1	-1	-1	-1	-1	-1	-1	-1	-1	-1
V	0	1	1	0	0	0	1	1	0	1	1	1	1	1	1	-2
V	0	0	0	0	0	0	0	0	0	1	1	0	0	1	1	0
Average	**0.03**	**0.17**	**0.14**	**0.00**	**-0.17**	**0.03**	**0.28**	**0.03**	**0.07**	**0.07**	**-0.10**	**-0.07**	**0.03**	**0.07**	**0.07**	**0.00**

Table 4.2: Read group: average change in scores before and after read stimulus

Mode	Q1D	Q2D	Q3D	Q4D	Q5D	Q6D	Q7D	Q8D	Q9D	Q10D	Q11D	Q12D	Q13D	Q14D	Q15D	Q16D
R	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0
R	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	2
R	0	0	-1	-1	-1	-1	0	-1	-1	1	0	0	1	-1	-1	0
R	1	1	0	0	0	-1	0	1	1	1	0	0		0	1	0
R	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	
R	0	0		-1	1		0	1	0	-1	0	0	-1	0	0	0
R	0	0	0	0	0	0	0	0	0	0	0	0	0		0	0
R	1	0	0	0	0	0	-1	0	0	0	0	0	0	0	0	-1
R	0	-1	0	0	0	-1	1	1	0	1	0	-1	1	0	0	0
R	0	0	0	0		0	0		1	1	0	-1	1	0	0	0
R	0	0	0	0	0	0	0	0	0	1	1	1	1	1	1	-1
R	0	0	0	1	1	1	0	0	0	0	0	0	0	0	1	-3
R	0	0	0	0	1	-1	0	0	-1	0	0	-1	0	0	0	1
R	0	-1	0	-1	0	0	0	0	0	0	0	0	0	0		1
R	0	0	1	0	0	0	0	-1	0	1	0	0	0	0	0	0
R	0	0	0	0	1	0	0	0	0	0	0	0	0	0	1	0
R	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	2
Average	**0.12**	**-0.06**	**0.00**	**-0.12**	**0.19**	**-0.19**	**0.00**	**0.06**	**0.00**	**0.29**	**0.06**	**-0.12**	**0.19**	**0.00**	**0.19**	**0.06**

△ *Scores represent the individual differences between before and after responses to the intervention was administered to each individual participant. The final row represents the average change for each item. A positive score indicates a favourable outcome. That is, people were likely to think RHD is important to discuss, more willing to share in a group, and greater empathy towards others living with RHD. A zero represents no change in score. Green indicates a positive shift. Yellow indicates a negative shift.*

From both tables, the majority of scores show no change before and after the video or read stimulus. In the video group, 67.88% of the scores remained the same, and in the read group 74.26% of the scores remained unchanged. While 31.68% of the scores changed in the video group and in the read group 30.96% of the scores changed.

The graphs below, Figures 4.1 and 4.2 reflect the change in scores for video and read groups respectively:

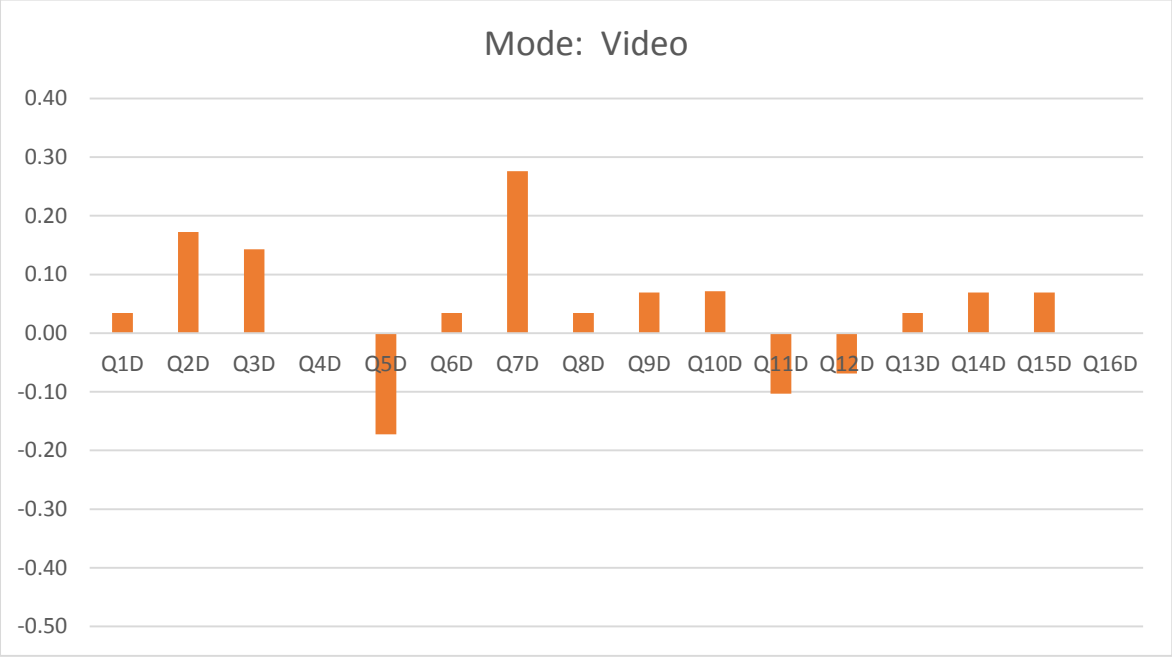

Figure 4.1: Change in scores: video group

Participants in the video group showed a shift in a positive direction (change in scores in 10 of 16 items in the positive direction). That is, they seemed to more open to discussing RHD after having watched the video. Item 7, measuring empathy showed greatest change in the positive direction.

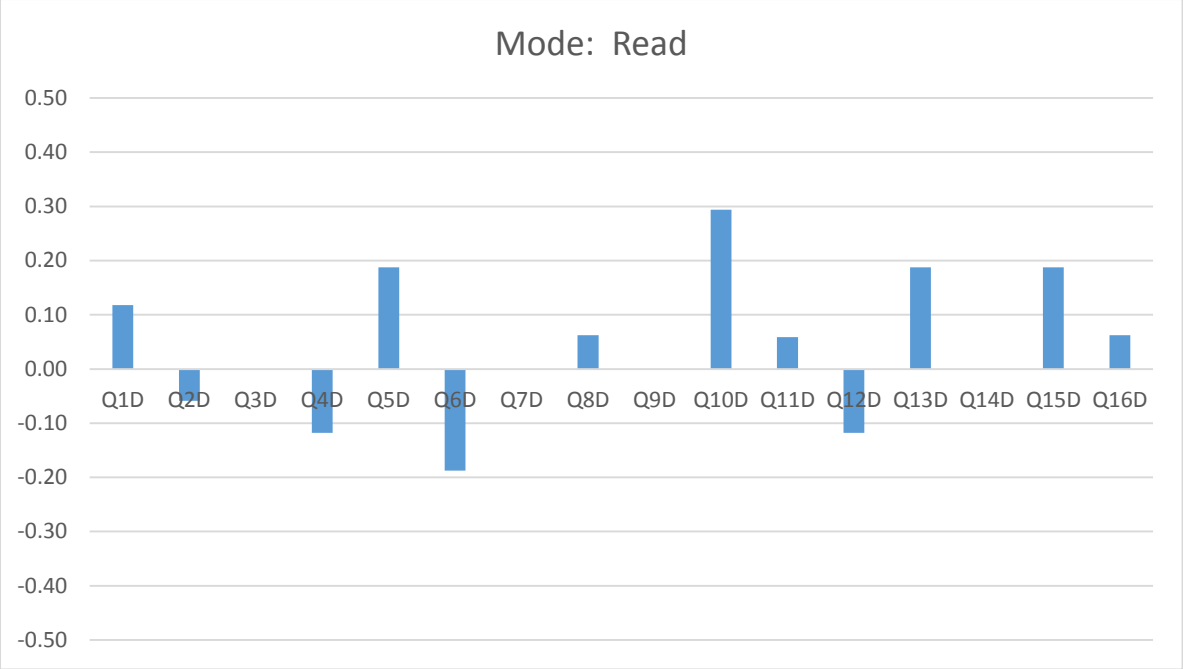

Figure 4.2: Change in scores: read group

For participants in the read category, changes in the positive direction occurred in 8 items shifting the positive direction), with 4 items shifting in the negative direction.

Data Richness

Figure 4.3 reflects the average percentage of coverage of selected codes, comparing video to read groups:

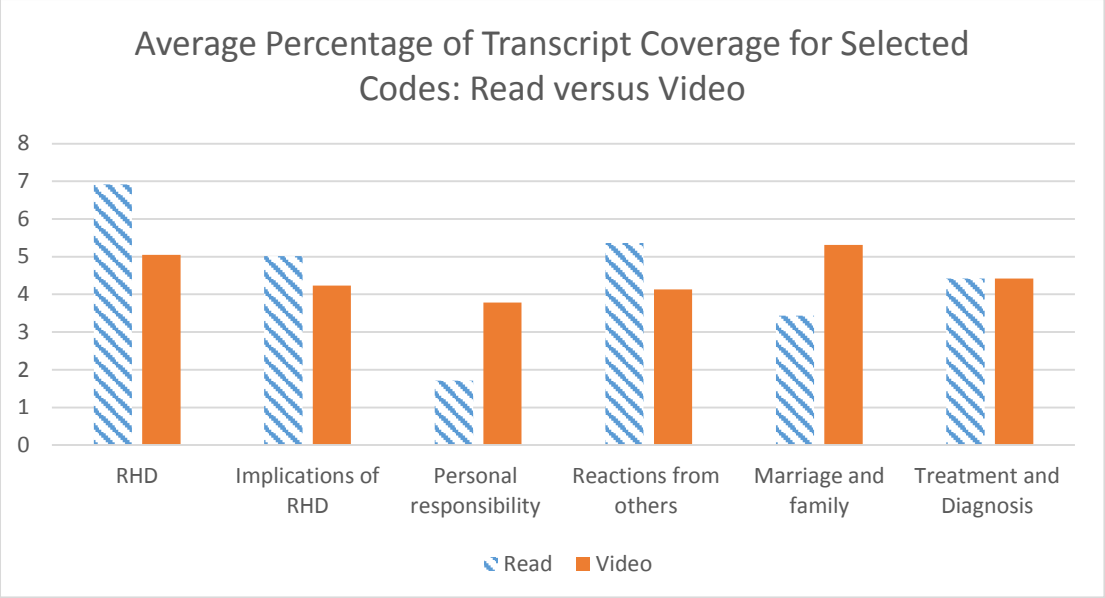

Figure 4.3: Average percentage of transcript coverage for selected codes: read versus video

There is no discernible trend or pattern when comparing reading to video groups. Differences between code coverage between the two groups seem to appear random. The 'treatment and diagnosis' code cover the exactly the same percentage coverage between the two groups. It appears that the use of video resulted in greater (transcript) coverage for issues relating to personal responsibility, and marriage and family. At the same time results in the table suggest that there is less coverage for RHD, Implications of RHD and Reactions from others, for the video groups when compared to the group groups. .

Similarly, Table 4.3 below reflects that there is not a significant difference in the change in scores between the video and read groups. The greatest differences between the two groups are domains labelled 'Empathy' and 'Emotional response'. With regards to the 'Empathy' domain, in the video group nearly half of the scores changed, whereas in the read only about a quarter of the scores change. This domain measured how participants relate to other people with RHD.

Table 4.3: Percentages of scores unchanged and changed by domain for both video and read groups

	Video		Read	
	Unchanged	Changed	Unchanged	Changed
General Interest	78.16%	19.54%	84.31%	13.73%
Personal engagement	70.00%	22.99%	70.59%	25.49%
Empathy	58.62%	40.52%	72.06%	26.47%
Willingness to share in a group	73.56%	27.59%	78.43%	19.61%
Emotional response	55.17%	44.82%	66.67%	27.45%

4.5. DISCUSSION

This study found no considerable difference between the use of visuals methods or textual methods in generating discussion and richer data in the context of qualitative research. The primary concern of this chapter was to empirically investigate the effectiveness of the use of visual methods in qualitative research to stimulate discussion and generating richer data when compared to text-based methods through FGDs between groups employing text-only (read) and video-based vignettes. Using a before/after study design to test the change in response in to how important participants this it is discuss RHD, willingness to share in groups about RHD, and emotional responses and empathy towards people living with RHD, we found that the majority of individual scores, 67.88% in the video group and 74.26% in the video group, remained unchanged after the respective stimulus.

While a hypothesis may predict that a greater proportion of scores should change after the intervention, such high percentages of unchanged scores may be as a result of the fact that people already felt quite strongly about the importance of RHD, their interest in sharing their stories and a belief that RHD must be spoken about more publically. Nevertheless, the results also show that there is no considerable difference in the change scores between the two groups. What is clear is that for most items, there is in both groups, a shift in response. We acknowledge however, that this is most likely due to a few scores for a particular item skewing the data, given that most scores remained unchanged and were counted as 0.

The greater shift in the video group scores result may be explained by the fact that seeing the life story of Enrico, one of the characters in the film may have caused a greater sense of empathy with people who have RHD compared to simply hearing about him in the read vignette. Additionally, what needs to be highlighted is that, in the video group there seems to be a greater change in the positive direction after the stimulus, when compared to the read group.

There is a marginal difference in the items which shifted in the negative direction between the two groups; the read group also had more unchanged items versus the video group. There are at least two possible reasons for this. First it may indicate that watching a video may result in people becoming more open to talking about RHD and feeling a greater sense of empathy. Second, it may also be that more people in the read groups had already responded positively and did not change their responses in the after-questionnaire. What is important to note is this only refers to changes in scores which made up about one third of the scores in each group.

Results measuring richness of data in the transcripts also show no considerable difference between the two groups when considering percentage coverage of transcripts for particular themes. As the researcher facilitating the FGDs, I took notes afterwards. I also had a research assistant present in each of the FGDs who took notes during the FGDs. We also debriefed about each session, and in our debriefing sessions we particularly considered the comparative aspect of our study. In both of our experiences there was no notable difference between the groups which watched the video or had the vignette read to them. In fact, as the facilitator reading the vignette, I often felt more engaged with the participants because it forced me to be actively involved in the process. Whereas when the video was played, it felt easier to disengage while the video was being played given my familiarity with the video. We also did not notice a difference in responses from participants who were in either group. There was neither a significant difference in the lengths of the FGDs nor the length of the transcripts between the two groups.

This paper has provided evidence, drawing on three different evaluation components, to show that there is no significant difference between the efficacy of using film versus text-only vignettes as prompts in the focus group discussions we conducted. This is important given that a recent literature found that most researchers incorporate visual methods into qualitative arguing that such methods are more superior to other forms of stimulating discussion [2]. The results of this study contrasts with the findings of our systematic review (Chapter 3), thus raising concerns about the robustness of the evidence extracted from the included papers. This may be explained by the fact that two of the three studies only drew on field notes and reported no clear or systematic methods used to evaluate the use of visual methods. The remaining study by Collier described a clear research design to evaluate the use of photographs in in-depth interviews; however, the evaluation process was vague and focussed only on the coding density [15]. Thus, drawing on a variety of evaluation methods, provides confidence in our findings that film is not more effective at stimulating discussion or generating richer data in focus group discussions than text-only vignettes.

Limitations

One major limitation of this study has been in the analysis due the kinds of data collected. The questionnaire employed a 1-4 Likert scale and used a before/after study design. It was difficult to find an appropriate non-parametric statistical test to evaluate the differences between two dependent data points. This constricted my ability to make more precise claims about the effect of visual and read stimuli. Future studies of this nature would benefit from using a tool which collected continuous data so that more sophisticated tests could be used. Or if ordinal data is used, the study must be designed in such a way that it allows for the use of non-parametric tests.

The second limitation relates to the use of questionnaire data. While I conducted cognitive interviews to ensure that questions were understandable to this population, it was beyond the scope of this study to validate the questionnaire. Therefore this poses as an important

limitation to the generalisability of the results of the questionnaire. Future studies should validate such questionnaires to ensure that concepts are being operationalised appropriately.

Third, another reason for visual methods not having significant impact is possibly because the FGDs were facilitated well and therefore the impact of visuals methods was rendered redundant. While this is possible, it is unlikely that given that I do not have vast experience in facilitating FGDs of this nature.

The final limitation of this study relates to the nature of both evaluation methods. Both methods employ quantitative evaluation methods to measure the effect of visual versus read stimuli. While such quantitative methods have provided important insights, especially when making comparison, it is difficult to quantify concepts like richness of data or effective discussions. What studies should consider is how to include qualitative evaluation methods to broaden findings. As an attempt to include a qualitative component, I used field notes that were created by myself and an observer (and we had debriefing sessions after each FGDs). These insights however proved difficult to use as a source of evidence given we had to largely rely on observations.

CHAPTER 5: STUDY 3:

EXPLORING THE EFFECTS OF GENETIC KNOWLEDGE ON STIGMA ASSOCIATED WITH RHD

5.1. INTRODUCTION

This study focuses on understanding the impact of genetic attribution on stigma relating to Rheumatic Heart Disease (RHD) in the Western Cape. The project worked with patients who were enrolled in the RHDGen genomics research project, based at UCT and Groote Schuur Hospital. In this project I explored two types of stigma – 'internalized stigma', defined as the stigma experienced by patients, and 'associative stigma', or the stigma that is experienced by others as a result of their biological or other association with patients living with a stigmatised condition. While this study generated a wealth of data on how RHD patients view their condition, the current chapter will specifically reported on data relating to the question of how genetic attribution influences stigma experiences of people living with RHD.

The relationship between stigma and genetics may have two effects. Namely, on the one hand genetic information could help reduce stigma by decreasing personal blame or responsibility for developing the condition [11]. On the other, genetic information could increase stigma by making conditions absolute and unchangeable, with few options for recovery or treatment [93].

Most studies focussing on the effect of genetic attribution of disease have been conducted in Western contexts. This study is one of the first to explicitly investigate the relationship between genetic attribution and disease stigma in an African population, and one of the first undertaken in a non-Western research context. This may be important because different cultural belief systems about disease may have very different effects on stigma [94].

5.2. METHOD

As described in Chapter 2, Section 2.4, to understand the relationship between genetic knowledge and its impact on stigma among RHD patients, I conducted 11 FGDs with 52 mixed-ancestry research participants who had previously participated in a genomic study. I used vignettes to explore their beliefs and experiences relating to living with RHD. Each vignette explored one of three causal pathways for developing RHD, namely, genetic, partially genetic and non-genetic (environmental). Each focus group was organised around one of these three vignettes. Topic guides were created for each of three vignettes (see Appendix 3). After piloting, the vignettes were turned into short videos with the help of a professional film maker. The vignettes or videos depicted a fictitious character, Enrico, who develops RHD and learns the cause of his condition, based on one of the three casual pathways explored.

Each vignette was split up in three stages. After each stage was played or read, group discussions were moderated around a number of topics relevant to explorations of stigma and genetic attribution. The first stage of each vignette explored participants' associations with or understandings of genes, genetics and disease causation by exploring the associations that come to mind upon hearing 'genes' [74]. The second stage in the vignette consisted of an exploration of how having RHD will impact on the patient's life, such as how he is treated by others, the probability of finding work, getting married or having children. In the third and final stage, the vignettes explored the consequences of a desire to maintain social distance [12] (in relation to someone who has RHD), which has been used as one means of measuring the effect of genetic attribution on stigma. This component of the FGD topic guide explored questions like "Would you be happy for Enrico to marry your sister?", and, "Would you be likely to be friends with Enrico" (for full topic guides see Appendix 3).

The FGDs were recorded and where necessary, simultaneously transcribed and translated from Afrikaans to English by a professional transcriber. The transcripts were imported and analysed in NVivo 11 [72] . I listened to the recording of the FGDs in the original language, and read the transcripts twice. I developed open codes using transcripts from one of each of

the three casual-models for the first level of analysis. Two other members of the research team independently also developed a set of open codes from the same sample of transcripts. Based on multiple rounds of open coding and in discussion with the research team, I then developed a hierarchal coding scheme which contained 11 main themes and 29 subthemes. The hierarchical coding scheme was applied to one transcript and refined, after which it was applied to the entire dataset. In this chapter, I draw on the text coded under themes related to general understanding of genetics and RHD and stigma experiences of RHD patients to explore how genetic attribution may impact on stigma associated with rheumatic heart disease.

See Chapter 2, section 2.4 for a more comprehensive account of the methods used in this study.

5.3. RESULTS

5.3.1. Understanding disease causation

Relating to genetics

Before exploring ideas around experiences of stigma, it is important to develop a general sense about how participants understand genetics. In some instances participants reported that they did not know what genetics is. One participant stated clearly, “I truly don’t understand what genetics is”. However, many others expressed what they thought genetics meant. The most common way participants explained genetics, was in relation to hereditary characteristics in a particular family:

Participant: Something that's passed on.

Facilitator: Something that's passed on, where, how?

Participant: Like, it automatically comes to mind like someone in the family had it or something in your, I don't know, something like that.

Other participants spoke about genetics as something embedded within a particular family, as if part of the family's genealogy. One participant in a FGD which explored mixed causation of RHD expressed her understanding as:

> Genetics is something that comes down in the family, it grows…like a cell, it develops forward, it's been planted in the family as it goes, the family tree, it carries on from the one to the other one, sometimes it skips the first… and then the second one gets it, so it runs in the family…"

And while the idea that a disease is passed on from parents or grandparents was a consistent theme, participants frequently noted how it did not necessarily have to be transferred in every generation. One expression of this sentiment was:

> Say now for instance your grandfather has it, now it's to his daughter, either his daughter or sons, one of their children will have it again, it's not always the same hey, it's not always that it happens.

What was perhaps most unclear about genetics for many participants was how diseases were actually passed down from one generation to another. While people had some notion that genes are hereditary and came from their grandparents or their parents, the actual biological mechanisms of transfer were more difficult for participants to describe perhaps owing to how thinking about genetics is an abstract concept to many. When I asked people to explain how they thought genes were transferred from one generation to another, one participant expressed this confusion often shared by participants:

Participant: It actually depends, it is actually in your blood, if they say you have inherited it from your family, a heart problem.

Facilitator: And it's something in your blood?

Participant: I won't say it's something in your blood, you inherit it from your family, and it's not necessarily in your blood.

Another participant related their understanding about how genes were passed through the process of birth:

> would think it's at birth, or, how can I now put it, it's not something you give physically to a person or a child, so I think it's in the make-up of the body and a baby, when developing, I think it takes it from there.

Despite expressing some difficulty in understanding how genes are passed on, I found that overall participants had a clear understanding of genetics as being something that was passed on through family lineage. Even though people were relating genetics to their personal experiences of their own family's genetic history, only one participant equated genetics with stigma, namely, "genetics, people will think it's a disorder, some disorder that you have, in your genetic system, your combination of human being, so there's something wrong". In most other responses about genetics, participants attached no social value or judgment to their understanding of genetics.

Causes of RHD

In general participants were quite conversant about their condition. I gauged whether participants understood what caused their RHD. This was important because it provided a sense of how participants not only interpreted the cause of their illness, but also the degree to which participants attributed genetics as a casual factor. Participants identified a range of causes for their illness, including genetic, bacterial and environmental. While each causation-vignette emphasised different causation pathways, the causation attributed to RHD by participants was often inconsistent with what the vignette emphasised. This is important to

note as it shows that what people described as the cause for their illness was not simply a reflection of what was suggested in the vignette.

Some participants reported that having RHD was a result of their family's genetics. This was generally reported by participants who could directly trace the history of RHD in their family. For instance, one participant in response to what causes RHD said:

> …my mother has chest problems, this asthma, my father's side, are heart people, my father's brothers, many of them are died because of heart attacks, and one of the doctors said it's in our genes.

Of those participants who attributed the cause of RHD to genetics, one participant understood genetics to play a deterministic role:

> I think it's because, if I think of myself, my mother had a heart disease…. so because of genetics, you can get it from both sides, your mother or your father, and I got it from both, so there's no way you're going to back out, because it's genetic, it's there in your gene.

But other participants who also believed that RHD was caused by genetic factors, seemed to believe genetics to only be a contributing factor. For instance, one participant shared that of her 12 siblings, she was the only child who had developed RHD. Another expressed a more flexible understanding of genetic causation in relation to their own experience, responding to the likelihood of Enrico's children also developing RHD:

> I think no, because not one, ok my one was born with a heart failure but she's never had any problems, so I don't think so, sometimes not your kids will have it, their kids will have it, but not your kids , like in our family, my mother, like my mother's sisters and brothers, none of them have a heart disease, but most of the nephews and nieces, like their kids, like one or two in each family have a heart disease…maybe it skipped one generation…

Other participants reported that it was an infection, sore throat or germs which caused them to develop RHD. For instance, one participant related her own story of RHD:

> I think sore throat as a child, it's got a big part in this. I mean, I suffered a lot with tonsillitis, even up till today I still have my tonsils and like the doctor always explained to me, the germs that's here, closest organ is your heart so that's where your germs go settle..."

Another participant claimed, "It's a germ, it's not a germ that I can pass onto you, it chooses somebody…and there it's by you".

For many other participants, they attributed the cause of RHD to a materially impoverished environment. One participant noted his condition was caused by environmental factors:

> Rheumatic fever, I mean, it's something that you pick up, cos for me it wasn't genetic, no one else got heart problems, for me it was the environment, it must have been an infection, maybe not doing something properly, it just got worse.

Others spoke more directly about their own experiences of living in poverty, and in crowded homes, which resulted in increased infections:

> I think it can be, your way of life, your conditions, that you live under…I think it could be something like that because I mean, why would you get a sore throat all of a sudden, or a germ if it's not in the air somewhere, you know, I mean, no one ask for these things to happen to you… I remember myself as a child, there were always a lot of people in the house… I lived in a time where a three bedroom house and a lounge, you had four families, a family in each room and the lounge…people…had to cook, they had to wash, they had to do everything in their room, you see, so... it could've brought on the germ thingy…

Another participant from a mixed-causation vignette FGD reflected on that fact that it was after her family was forced to move to the Cape Flats, where houses did not have proper insulation, that she started to get throat infections, "…we used to move to Lavender Hill, we were the first people that moved… the walls and stuff, it was very cold…from there onwards, I was sick…" Lavender Hill was one the areas people of colour were forced to move during Apartheid. These areas were usually associated with small houses, built very close together, and often had to accommodate large, and sometimes multiple families.

Overall, most participants had a clear sense of what they think caused RHD. While some responses were unexpected such as smoking or drinking alcohol, most participants reported on causes of RHD as having multiple pathways of causation, most notably, genetics, bacteria or their environment.

5.3.2. Experiences of Stigma

In order to understand whether people with RHD experienced stigma, we explored a number of questions in relation to how Enrico's life may be affected by RHD, how people may react to him having RHD, their chances of being employed and so forth. These questions were specifically explored in relation to the three causation pathways described previously.

It became clear that RHD impacts the lives of people in multiple ways. When we asked people how Enrico's life would be affected by having RHD, participants across all three causation-vignettes said that he would live a "normal life". One participant summed up this sentiment by saying:

> From my point of view, go for your check-ups, the doctor finds out you got rheumatic fever, you take your tablets, I don't think it will affect your life, I've got a normal life.

However some participants reflected on ways RHD did negatively impact on their lives. These largely related to the physical limitations of living with a chronic heart disease. For instance one participant reported on not being able to sleep after having an operation and others, mostly women, reported on not being able to do as many household chores. This compromised ability to participate in (sometimes communal) physical activities was one of the most prominent themes reported on. One participant noted:

> It was a problem because I loved athletics, I loved ballet… and I couldn't be part of it anymore because of my getting tired, and so I just had to take a step back and realize, ok, I can't…

Social Responses to RHD

One of the ways to measure stigma is to gauge how people living with a particular condition are treated. We explored with participants, how they thought Enrico might be treated by family and friends. We found that people consistently reported that family played a supportive role in their lives. While participants generally reported that friends would also be supportive, some participants also reported on experiences of social exclusion and labelling.

We asked how participants thought Enrico's family may treat him after receiving his diagnosis. Across the three causation vignettes participants generally reflected that they felt "supported" and said family was "encouraging. One participant in the genetic-causation FGD reflected on her personal experience and the history of heart of disease in their family:

> My family is very caring towards me, my father died of a heart attack, my brother died of a heart attack, I'm the only one now in the family that has it, so I have a problem, they all cluck around me and I appreciate their love.

Most participants felt that the cause of RHD would not matter to family members. One participant reflected:

> It won't make a difference. When I had rheumatic fever, my back used to pain, when it was still fine, I was in hospital for almost nine months. When I was fine, when I was healthy, what stayed behind was the heart valve, but I lived a normal life, they treated me like a normal child.

Another participant reported on how proud her family was of her for living a long life. She reflected, "My family is proud of me, since the age of 30 I had a heart complaint and I'm still going strong…they're even looking forward to my 80th birthday."

Only one participant reported on feelings of guilt but this was as a result of environmental factors rather than genetics. In a mixed-causation FGD, a participant felt that Enrico's family may feel guilty for living in crowded home:

> I think the family would feel guilty, because maybe having all the other family living with them… caused no ventilation in the house… and I think the parents will feel more to, if they can, try to improve their living conditions.

We also explored whether participants thought Enrico may be treated differently by his friends. Many participants felt that Enrico would not be treated any differently by his friends, or that "true friends" would not treat him differently. This was a consistent finding across the three causation-vignettes. However, some participants also reported experiences of social exclusion or being labelled as "sick" (either based on their own experiences or as a possibility for Enrico). Some participants did express the potential for social exclusion as a result of not being able to take part in particular activities with friends. Another participant in a FGD which focussed on genetics as a causal pathway, said:

> Most of the time you can't be with your friends, they might be taking part in sports, you're tired all the time, you can't take part in sports, so you will feel left out.

One participant reflected on her son's experience as being labelled as sick and therefore being perceived as unable take part in social activities, resulting in social exclusion:

> Aunty's son [referring to herself], he didn't have friends, he stayed indoors because he couldn't play the way he wanted to… They [his friends] didn't want to play with him, they teased him, they were scared, they were scared to play with him because he was sick…

Another participant in the environmental-causation FGD shared his experience of a friend who had RHD:

> They will treat him differently, if he must play soccer, he can't play soccer… I saw it on the soccer field, the one [guy] had even had chest pains and the flu, and when they had to play, they said no, he must play, and he said he can't play, he's sick.

When I asked participants if he would be looked down on by others, my findings reflect a similar pattern. Many participants across all three causation-vignettes felt Enrico would not be looked down on by other people. One participant said:

Participant 1: I don't think so actually

Facilitator: Why not?

Participant 1: Because its life threatening, but it is not contagious, I can't give it to you

Participant 2: They treat us like normal people

Participant 1: Yes, like normal people

Other participants did feel that Enrico may be treated differently but this was often as a result of people being ignorant. One participant reflected on how ignorance could lead to someone like Enrico being teased:

> …when they don't understand, then they say ignorant things, but when they understand that it's something bigger and this is the problem and this, so it's the information that's out there. Someone will say something silly like you're slow or you're this or you're that and it will make you feel bad… but I think it only comes from when people don't understand.

Other participants related this kind of ignorance to how other people with TB or AIDS are treated. The participant noted:

> It can be either like, if you say somebody's got TB, although it's curable now, people will still stay away… the misinformation about HIV for example, people will think, oh they sit on the same seat, they're going to get AIDS… like those kind of things so it depends on society.

From these results it is clear that where participants have had experiences where they labelled or excluded, it was mediated by cultural and other social factors.

Impact on marriage and having children

Another way to investigate disease stigma is to understand how disease (including disease causation) may influence people's decisions to get married and have children. Overall, participants reported that having RHD would have a marginal influence on decisions to get married and have children.

When I asked participants if having RHD would impact Enrico's chances of getting married, many participants felt that it would not. A consistent response often related to romantic notions of marriage, "where love conquers all" as one participant said. In fact some respondents suggested that RHD would be irrelevant when it came to marriage. In one of the FGDs which explored genetics as a causal pathway, one participant shared:

> When I was diagnosed with a heart problem, when I met my boyfriend, I forgot about a heart disease, you don't think about that, you just think about the love, and all the birds and the bees flying…

Another participant commented, "there's no sore throat, there's no tonsillitis… there's only stars". While agreeing with these sentiments, another participant insisted that honesty and disclosure about Enrico's condition to his partner was really important. The participant said, "if he gets married he must tell her what his condition is… she will understand if she loves him… this is what I have… they can have a normal life.

Another participant from a different FGD shared this sentiment reflecting on Enrico's decision saying, "…he's got to think how he is going to support the family, except for the love-making… he's got to think mentally how can he manage it, it's going to put more pressure on him…it will also affect him". For this community, traditional values were seen as being important. Getting married and having children, is seen an indicator of "having a normal life" (a phrase often repeated by participants). An important part of traditional values in this context is the man's ability to provide for his family. Moreover, consistent responses of "love is all that matters" (i.e. being in a relationship with the prospect of having children) and being able to financially provide for your family primarily influences decisions to get married.

Finally in relation to decisions about getting married, some participants did report on internalised stigma which may influence decisions to get married. One participant reflected on being sick and needing care, "[he] probably [does] not want to get married… he is already a burden to his family members… because he needs frequent care… he's going to feel it's unfair to involve somebody else." Another participant felt that maybe Enrico's potential partner may

struggle to “[be] with somebody who’s got a germ”. In a different FGD another participant reflected on her own experiences of feeling undesirable, “For me it was like, who would want me now, and who would want to get married to me and all that, I’m sick.”

When I explored whether participants thought Enrico should have children, participants generally agreed that having RHD would not affect his decision to have children. Similarly, having children was almost an obvious choice in this population. This question was often met with quick responses such as “yes he will have children” or “of course”. One participant who was aware that she may not have children reflected on the fact that it did not matter to her partner, “…and I told him, listen here, there’s a 50/50 chance for babies, for a family. And then he said, just remember, babies wasn’t there before me and you”. This response was however an anomaly, in that most participants indicated that having RHD did not influence their decision to have children. It is important to note, that the majority of the participants were women and many of them reported that they did know they had RHD when they fell pregnant for the first time.

For women living with RHD, pregnancy is an increased risk and for many participants, it was during pregnancy that RHD first manifested and was initially diagnosed. However, RHD was generally not reported as being an obstacle to have children. Additionally, gender emerged as important theme where participants emphasised how men and women living with RHD, are differently impacted by the disease, in relation to having children. Women often reported that men, “don’t think about this” and “a man can make a baby, a woman must get a baby”. Only one participant insisted that her husband actually thought about the risk of children and was concerned about the impact of pregnancy on her health. Many participants (including men sometimes) however felt that women were forced to think about having children more seriously given the direct implications of pregnancy on the body of woman with chronic heart disease body. For instance, one participant spoke about how RHD patients who are female are told to not have children, “if it’s a lady they will discourage her to have children, they will say you mustn’t have children.” Female participants often shared their experiences of doctors issuing

serious warnings to not have children or stop having children given the increased pressure on the heart during pregnancy. At the same time, female participants also reflected on the pressures placed on them from their partners to have children. In one example a participant reflected on her experience of being told to stop having children, but also her husband's desire for more children:

> because, they told me that… when I had one child, and then they said, if the second one is a boy, I have to stop, but I was still very young [they] wanted sterilise me, so I said no, I'm still too young to go for a sterilisation… my husband wanted another child and they became four… they don't have a heart problem, not one of them.

In this population, getting married and having children has an important social function. Likewise, values such as love and the ability to take care of your family significantly influence people's decisions to get married.

Impact on finding work

As another way of investigating experiences of stigma, we asked participants how living with RHD (in relation the specific causal pathways) may impact the chances of Enrico finding employment. There were mixed responses to this question across the various vignettes. In some instances participants were clearly discriminated against, while other participants shared experiences of RHD impacting their ability to both find and retain work.

Some participants felt it would not impact on his ability to find work. When asked if Enrico would struggle to find work, one participant felt, "Not at all…if he's willing to work, and he's able to work, and he's healthy to work." However many others felt while he will find work, it will limit the kind of work he is able to do. Participants expressed that he would not be able to do "heavy duty work" but "light duty" or "clerical work". One participant who worked in construction said:

> …I've been in construction up till today, so I'm not sitting in an office all day, but you're physically involved, so it gets to you sometimes, it gets to you, when you must sit down and you must take a breather, but it won't put you down, and say I must quit this job, if it's your job, that is what you've done all the years, you're going to do your utmost to carry on with that… most of the people here had heart operations, I never had one, I can still manage… but it gets to you sometimes, you can just bend over to pick up something, when you get up you can't breathe, take a break then you can carry on again, but certain people won't be able to do it, it might be worse than what I got…

Participants often shared the importance of perseverance. While this can be framed as resilience, in this context, much of this need to "keep going" is also related to a context where finding work is difficult, especially given low levels of formal education and high levels of unemployment. Related to this may also be perceptions that people with RHD require additional accommodation and support, in the workplace.

Some participants felt that Enrico would be less likely to find a job than keeping an existing job. When participants spoke about finding employment, two important obstacles were often reported. The first was that RHD patients regularly need to attend hospital appointments – as often as once a month, with the implication of missing an entire day's work. In some instances this was highlighted as a serious problem in finding and retaining work. One participant said:

> You get that good bosses that will accept you, then you get that stingy type… that will look down upon you… a lot of bosses do that, it's like a stigma… then they just tell you, look sorry, the job isn't good enough for you.

Another participant shared how she was told not come back to work, "take it from me when I was working, my boss said, if I stay out of work every time to go to hospital then I might as well stay at home." The second barrier was being sick which also seen as resulting in missing work. One participant described how she lost her job after being sick, and presenting her medical certificate. Her boss's response was "[you have a] weak problem [poor excuse], you need to stay away".

Other participants reported on being able to work and attend hospitals without it being a problem when they were employed. One participant summed up this sentiment, "I worked for

32 years for this firm, and I went to hospital every month, to Groote Schuur and you just bring a certificate…" Alongside people reporting on their work not being impacted however, was the need to "prove yourself". One participant said:

> I think you have to work double as hard to see that you are capable of doing the job, I said to myself, I will show you I will do what I want to, I came here to do, and if you excel they will just go on, they won't discriminate against you.

Another participant felt similarly:

> If you prove yourself and you do what you have to do and tell your employer, listen I need this one, not a day off, I will even just go and then come back, but don't take advantage. If you finish off at 9 o' clock, you don't want to go in to work, so I think you, the person also need to put in that extra. They're allowing you this, so you also don't misuse it.

While people often spoke about how it's possible to have a full working life, the theme of needing to "prove yourself" could also be understood as a strategy to combat possible perceptions that people with RHD (or other chronic illnesses) are lazy and constantly need to take time off work.

5.3.3. Stigma through Social Distance

Another way to measure stigma is through social distance. That is, how people feel about being seen to be in a close social relation to someone with RHD. Here I explored how participants would feel about being friends with Enrico, and Enrico meeting their family.

I first asked participants how they would feel being friends with Enrico. Most participants said that they would be comfortable being friends with Enrico. A phrase that people often used was, "there's nothing wrong with him". In addition, participants often felt there was no reason that having RHD should matter or determine whether they should be friends. One participant

noted however the difficulty in being friends with someone who has a heart disease. Reflecting on her own experience, she said:

> I have mixed feelings about that… I had a friend who had a heart condition and so when she passed away, it was very emotional for me… it was extremely hard for me.

A similar trend emerged when we explored whether participants would be comfortable to introduce Enrico to their families. Again, most participants said that would not be an important factor. In fact, many participants expressed:

> Participant 1: Exactly and you don't introduce somebody like that, here's Enrico
> Participant 2: And he has rheumatic fever, you don't do that (laughs)
> Participant 3: Not the first time, not the first time

When I asked if they would feel comfortable if Enrico wanted to get married to their sister, many participants reported what is of primary importance is that they love each other. It was also important that Enrico discloses the details of his condition.

5.3.4. Structural Stigma

While we did not set out to explore structural stigma, as we intentionally were investigating internalised and associative stigma, a theme related to structural stigma emerged. This theme arose specifically in relation to how participants spoke about living with and managing RHD. While participants did consistently feel that you can "live a normal life", they often also spoke about the need to have a positive attitude and change of lifestyle in order to be healthy. While these are important strategies to manage RHD, participants also recognised constraints imposed on their ability to change their lifestyle. This recognition is important because it highlights how structural discrimination renders this group more vulnerable to developing diseases like RHD, and increases the barriers to managing the condition well.

Participants across all three causation-vignettes spoke about the need to have a "positive attitude" to live a healthy life. This was often in response to living with a chronic illness, being young and having a heart disease, the need to often go to hospital and have operations. One participant reflected on the first time she had an operation:

> I think having a positive outlook towards your disease also does help… I discovered that being in hospital after I had an operation, I was very positive and when other people came in for the operations… because if you're not going to be positive about it, you are definitely going to take longer to recover than what you would if you have a positive outlook.

Another participant reflected on being diagnosed with RHD and being uncertain about the future:

> Participant: Because that time I was sick and I now don't know the future, because he's the doctor and I came to him because he diagnoses me with rheumatic fever…. and by that time I was still young, I was only 20 before they did the operation but I got the disease from 7 years old already.
>
> Facilitator: So what made you say to the doctor when you heard, you can't do this and you can't do that, what made you say I'm still going to do it.
>
> Participant: Because I'm positive, I was positive by that time

While these discussions can be framed as strategies for resilience, some participants also reflected on the enduring impact of structural inequality on the lives on people living with RHD. Participants noted the limitations that someone like Enrico may face. People from both the mixed- and environmental-causation vignettes reflected on factors outside of Enrico's control, like his impoverished environment. One participant noted:

> …it depends how he look at things, it mustn't become, because you have rheumatic heart disease…, I can't do this… you need to push yourself. Okay, in his circumstances, it's so bleak, his mother's support is there for him but there's so many other conditions that can affect his wellbeing

Another participant observed how difficult it may in fact to live a healthy life with RHD under difficult circumstances:

> …depending on the treatment that he received, if it was positive for him or negative for him, and if he, in this time got himself out of this environment that he was in, maybe improved his life… But I mean that is speaking in a wishful world where everything is fine. It's not like that, you know, many people don't have the opportunity, many people have the operations, they come home, they don't have the amenities, they don't have the comfort, so there's that element of them not healing properly, so there's those kind of things that come to plan.

Similarly, people spoke about the importance of changing their lifestyles such as an improved diet or living a more peaceful life. Another person from a FGD which focussed on mixed-causation pathway, reflected on the importance improving his living conditions:

> I think they might make a change in his living standards… because they've been living with him all the time and they weren't aware… they may say, maybe if we changed our life style, this wouldn't have happened…

Another participant from a FDG which explored environmental factors, noted the importance of changing your diet but also noted how many people may not be able to afford healthy food:

> A person has to eat healthy. I think as a person has heart problems, or had heart diseases, and grew it, then you must eat healthy… Lots of our people cannot afford healthy food… I was 23 or 22 years old, then I would go to the doctor, then they would give me thick white medicine. They said it's for the heartburn. I would use it and then I would go away for a few hours, and then when I eat again, and I breathed, there would be a pain on heart… And so he [the doctor] told [me], it's under the coloured communities, the Muslims, the Indians, because they from younger ages the wrong food… you must eat properly. You eat too much curry, samosa, pies, and pastries… But some of the food is very cheap to make, it's unhealthy, our people can't afford healthy food, we can't. We will stay here, disadvantaged, we will stay behind. That's just part of life, that's how it is.

Since RHD largely impacts the lives of poor people, participants had personal experience of constraints created by unjust structures, partly as a result of South Africa's history of racial inequality. Structural impacts are enduring, and are compounded in relation to those living with RHD. What is interesting is that responses such as "having a positive attitude" or the solution is simply change your lifestyle or insistence on a living a "normal life" can also be seen as a response to being rendered powerless to the circumstances which allowed for RHD to

develop, and for the potential difficulty in managing the condition such as importance of eating healthily.

5.3.5. Knowledge of genetic attribution and stigma

From what we have reported on, it is clear that participants had a general understanding of what is genetics is, namely that is something passed down from generation to another. There is also evidence to suggest that participants think of genetics as only one casual pathway for developing RHD. In fact, multiple casual pathways were attributed to developing RHD, most notably genetics, germs or throat infections and environmental factors. In terms of stigma, patients overall did not describe RHD as a stigmatised condition. However, there is some evidence of experiences of stigma, these include, internalised stigma, labelling, social exclusion and some forms of discrimination. Where patients have described experiences of stigma, these were often mediated by social, cultural and economic factors.

Only one participant reflected on the impact knowledge of genetic attribution may have on public perception. The participant, in response to reflecting how Enrico may be treated by other people, spoke about perceptions of having a heart disease at a young age because of lifestyle choices:

> …when you have a heart disease, people look at you and they think you did something wrong, they either think you partied a lot or you consumed some or the other substance a lot, like drinking or drugs, so if they realise that it's hereditary, something he inherited due to his family's bloodline, they might actually be more supportive and try and understand and learn more about the disease.

When participants were asked to reflect on the likelihood of Enrico's children developing RHD participants did not seem to think his offspring would inherit the condition. In this context, because genetics is understood as non-deterministic in that it may not necessarily be passed on to every generation or may be not an influencing factor at all, it seems to mitigate its impact

on the decision making process related to reproduction. One participant from a FGD which explored the genetics as a causal pathway shared:

> For me, it's not like your children will inherit it, because they told me I have rheumatic fever, my son didn't inherit it, and I have two sons, one is 54 and the other 34, and they both didn't inherit it, so their children's children might inherit it, but they didn't get it, perhaps it skip two generations or so, then it will come up again, but it might come out somewhere in the generations

Given that RHD is generally not described as a stigmatised condition, knowledge of genetic attribution may not simply as significant for this population. In addition, when experiences of stigma are reported on, these experiences are often mediated by others factors, other than the having the disease alone.

5.4. DISCUSSION

The results of his study show that while RHD is not typically known as a stigmatised condition, the RHD patients we interviewed do describe some stigma. These findings show that stigma does not merely exist as 'mark' as previous studies have conceptualised it. Rather, following on from Link and Phelan [6], these findings show how stigma operates as a process rooted in unequal power relationships. Participants described experiences of social exclusion, where they could for instance not fully participate in sporting activities. Other experiences of social exclusion also related to being labelled as 'sick'. In addition, participants reflected on the difficulty of finding or retaining work – this was specifically linked to needing to go hospital or clinic in order to collect treatment, which requires taking time off work. Finally, there was some evidence of internalised stigma where participants believed that because they were sick, they could not be able to find a partner or that there is something wrong with them.

While there is evidence of stigma in our study, these experiences did not interact with genetics. This is especially notable given that there was a general understanding among participants that genetics related to the hereditability of disease in the family. In many instances

respondents could connect the development of RHD to a family history of heart disease. Why does genetic attribution then not have a more immediate effect? Much of the studies cited in Western contexts found some evidence to support that genetic attribution may decrease stigma by alleviating personal responsibility and blame, or increase stigma through beliefs that conditions are severe and that treatment would be ineffective. Why then has genetic attribution had no impact on experiences of stigma in this population? There at least two possible explanations for this finding.

First, when we explored with respondents what they think causes RHD, they presented a number of casual explanations. These included genetics explanations where participants shared how other members of their family also suffered with heart disease. Others asserted that it was related to germs or bacteria which caused them to get sick. Others spoke about constantly having a sore throat as a child which eventually led to developing RHD. And in other instances, participants spoke about their impoverished living conditions which caused them to developed RHD. Such findings corroborates arguments by Kong [44]. In relation to refuting the efficacy of bio-genetic explanations on reducing stigma in the context of mental health, she notes how people do not interpret genetics as the only causal factor in disease aetiology. Studies by Condit shows the complexity and variation of complex aetiological frameworks used by laypeople, with genetics being only one factor in how laypeople understand disease-causation [95]. In addition to this, genetics is not necessarily understood be absolute or deterministic. Participants often insisted that a genetic disease does not necessarily mean that everyone in the family will develop the condition. The fact that participants use multiple causal-models to explain RHD is an important mitigating factor in limiting the influence of bio-genetic explanations on stigma.

The second reason may relate to the fact that genetics may not be as immediate given the local context. Here there two important examples. The first relates to the cultural context and the second to the fact that individuals in this population are juggling the impact of structural inequality associated to South Africa's history of racialized oppression. The importance of

cultural contexts was highlighted when I explored how RHD may impact an individual's life. For instance, I asked participants if Enrico would still get married or have children knowing he has RHD. The most common reported response was that having RHD (and knowing that it was genetic) would not influence his decision to get married or have children. In some instances female participants reflected on how they defied their doctors' pleas to not having children given the real risk of maternal death in the context of RHD. For this population though having children or getting married has great social and cultural importance which may displace considerations related to RHD and genetic attribution. Such contextual findings also related to the material realities faced by many in this population.

Related to how genetics may not be considered as significant in this context, it may also be that the impact of poverty, inequality and impoverished living conditions may be more immediate to managing RHD. This study only focussed on the coloured community in the Western Cape, a community whose social and economic position has been marginalised through apartheid, and its impact is still crippling on coloured communities across the country. Many of the participants spoke of the importance of a change in lifestyle, such as eating healthy food, or the significance of having a positive attitude or improved living conditions. And while these are legitimate expressions of resilience and living well, some participants also noted how difficult it is to eat healthy or change your living conditions when you are trapped in poverty. In such circumstances changing one's attitude may be the only thing one can change. For example, a recent study shows that the majority of South Africans are unable to afford a moderate diet with sufficient nutrition to live a healthy life, as a result of unregulated food markets, monopolised by four large retailers [96]. In order to manage a cardiovascular disease like RHD, maintaining a healthy diet is critical, but what does this mean if those who carry the burden of the disease are unable to afford to make this imperative change? While responses related to having a positive attitude or lifestyle changes may be framed as resilience strategies, in contexts of harsh inequalities, these responses can also framed as having to take personal responsibility for life outcomes [97], in response to enduring unequal structures which

participants are powerless to change. Genetics in this contexts unlike perhaps in Western contexts, may not feature as a priority in terms of difficult daily realities people have to live through. Moreover, for this population and for other groups who have a similar history of racialised inequality, the conditions under which people are stigmatising. For example living in poverty, not being to provide for your family or communities ridden by constant gang violence conditions are already stigmatised. As an example, when I recruited patients, many expressed a desire to be part of the research but were unable to come because leaving their homes was too dangerous given high levels of crime and gang violence. Such materially stigmatised conditions play a critical role in disempowering individuals and groups, and certainly from our results, mitigate the influence of genetic attribution.

Implications for Research

One of the reasons stigma becomes important in genetic research is that there is a concern that genomic research may create or exacerbate stigma in communities. While this is an important consideration, what do such findings mean for genomic research in contexts where people are struggling with many other structural and environmental difficulties such as poverty? In their study on the impact of genomic research on stigma conducted in Kenya and the Gambia, de Vries et al. asserted that local contexts must be taken into account [98]. This is especially important in relation to global research collaborations, where the global North and South may have very different research priorities. What we need are more studies conducted in local African contexts (and generally non-Western contexts). Moreover, these studies must include local researchers (or research familiar with the local contexts) who understand context factors, especially when relating to Western scientific paradigms.

Most research related to investigating the impact of genetic attribution on disease stigma focusses on either internalised stigma or public stigma. Yet, these findings highlight the importance of structural stigma which has received little attention in stigma studies. Furthermore, what we do not understand well enough is the relationship between structural

forms of stigma and genetic attribution. In other words, in the context of unequal social arrangements, which already often stigmatised, how do these structural conditions relate to bio-genetic explanations? For example, with multi-factorial diseases like RHD, how do structural factors relate to genetic susceptibility? Kong notes that one of weaknesses of relying on exclusively on bio-genetic explanations to reduce stigma, in the context of mental health, is that such a focus does not take into account how stigma related to mental illness plays out in the relation to structural factors such as economic or gender inequality [44]. Another way to frame this, would be in relation to power, or how the ability control a situation impacts an individual's ability to take care of themselves. Richman notes how individuals with low power are less likely to achieve goals such as treatment adherence [99]. In the context of structural inequality that often renders individuals powerless, how do perceptions of genetic attribution influence stigma? Such questions are important because they not only inform research priorities, but can also be informative for anti-stigma work. Furthermore, our findings also illuminate how these various aspect of stigma are interrelated.

Such findings also have implications for how stigma studies are designed. In their survey on stigma concepts and studies, Bos et al. emphasise the lack of knowledge of various aspects of stigma relating to each other:

> More empirical research on the interrelatedness of the different stigma manifestations is necessary. For example, we need to know if efforts to decrease structural stigma (e.g., changing discriminatory laws) can yield lower public stigma [33].

As an example of this, Angermeyer et al. in a study conducted in Germany, tested if there was a change in public attitudes towards people living with various disease including depression and HIV, related to public stigma (measured through social distance) and structural stigma (measured through levels of opposing to public funds used for specific diseases) [13]. They found while there was no significant change in people wanting to maintain social distance, there was an increase in public opposition towards cutting funds related to treatment of depression and decrease in resistance towards cutting funds related to HIV. Their findings, as

well findings present here underscore the need to understand both structural stigma, and how various manifestations of stigma relate to each. In addition, how would genetic attribution impact these kinds of interacting stigma? This is especially important given that these forms of stigma do not necessarily occur independently but co-exist together and interpedently in real-world contexts [33].

Strengths and Limitations

This study provided insight into the stigma experiences of RHD patients in the Western Cape. Since very little other work has been on stigma related to cardiovascular disease, or in relation to stigma and genetic attribution in non-western contexts, these findings are new. It was also focussed on the relationship between genetic attribution and stigma and found that genetic attribution does not influence experiences of stigma in this population. While these are novel findings, this study focussed on the coloured population in the Western Cape which, which was socio-economically homogenous group but religiously and linguistically diverse. What is needed is research with a more diverse (in terms race, ethnic, geographic and socio-economic) sample which could potentially provide greater insight into mechanism of stigma related to RHD (or other cardiovascular diseases). Another limitation of this study, was that my findings offer limited insight into a relationship between genetic attribution and stigma in this context because RHD is not severely stigmatised disease. More research focussing on this relationship, in non-Western contexts is needed on diseases which are more explicitly stigmatised.

CONCLUSION

This thesis has explored the impact of genetic attribution on stigma amongst Rheumatic Heart Disease patients in the Western Cape. Through the use of focus group discussions, I explored how understanding RHD as having a genetic causation could affect the stigma associated with RHD. This thesis is part of a larger study, The Stigma in African genomics research on Schizophrenia and Rheumatic Heart Disease study based. This study enrolled Xhosa patients, with either RHD or schizophrenia patients attending Groote Schuur Hospital in Cape Town.

This thesis only focussed on RHD patients in the coloured population attending Groote Schuur hospital. As part of the larger study, short video clips were produced to stimulate discussion in the FGDs. When I started working on this project, I began to question the assumption that the use of visual material is more effective at stimulating discussion and generating richer data in qualitative research. As part of the study exploring the impact of genetic attribution on stigma, I also designed an embedded study exploring the efficacy of visual material in qualitative research.

I have reported on the three sub-studies conducted. Study 1 and 2 related to the efficacy of visual methods in qualitative research. Study 3 explored the impact of genetic attribution on stigma amongst RHD patients in the Western Cape.

In Study 1, I led a systematic review which summarised evidence for the effectiveness of visual methods in qualitative research. This study is the first of its kind to synthesise this evidence. Searching 10 databases across various disciplines, three studies met inclusion criteria. All three studies provided evidence that the use of visual method is more effective at generating richer data when compared to textual methods. However, it was difficult to trust this evidence given that only study clearly described how the use of visual methods was evaluated, while the other studies apparently only relied on perceptions of the researcher.

Based on the results of the systematic review, in Study 2, I designed an evaluation study comparing the use of film and textual methods as prompts in focus group discussions. Of the

11 focus groups exploring the impact of genetic attribution on stigma, video clips were used as prompts in six, and text-based vignettes as prompts in five. I used three approaches for this evaluation study. First I used a before/after study design where participants completed a questionnaire before and after the prompts and mediated discussion. Second, I used transcript coding density calculated by NVivo 11 [72] as a proxy for richness of data. Third, I used the field notes that I and an observer took during the FGDs. Contradicting the evidence of the systematic review, across all three approaches, no differences were found between the use of film and textual methods in stimulating discussion and generating 'richer' data in FGDs.

Study 3 focussed on exploring how framing RHD as having a genetic cause could impact on stigma associated with RHD. Theoretically, genetic attribution could possibly increase stigma as conditions are perceived to be untreatable and individuals may feel personally responsible, or may decrease stigma as individuals or groups may be perceived as not being responsible for the disease. Whilst empirical studies provide evidence to support both of these hypotheses, almost no studies on this topic have been conducted in Western contexts. This study, focussing on internalised and associative stigma found that for coloured RHD patients in South Africa, genetic attribution had a negligible impact on stigma. I explained this finding by describing that whilst RHD is not a typically stigmatised condition, when experiences of stigma are reported they are not connected to disease aetiology but are rather displaced by other factors. One reason is that genetics explanations are seen as one of many possible explanations for developing RHD. The second reason relates to the socially and economically unequal context of South Africa, where there are more immediate factors which are pertinent to impact patients' lives such as poverty, inequality, community violence and cultural factors. Against that background, genetics is almost 'too small to matter', at least in the context of a complex illness such as RHD.

This study has been one of the first studies focussing on exploring the impact of genetic attribution on stigma associated with illness in an African context, and the only study to focus on stigma related to cardiovascular disease. As such, it is important to understand how stigma

interacts in local contexts, given its contingency on social, political and economic factors, and not just bio-medical ones. Furthermore, as ‘stigma’ is often considered as a risk to genomics or genetic research in Africa, this study suggests that it may not always be considered such.

REFERENCES

1. Boyd Ritsher, J., P.G. Otilingam, and M. Grajales, *Internalized stigma of mental illness: psychometric properties of a new measure.* Psychiatry Research, 2003. **121**(1): p. 31-49.
2. Pain, H., *A Literature Review to Evaluate the Choice and Use of Visual Methods.* International Journal of Qualitative Methods, 2012. **11**(4): p. 303-319.
3. Goffman, E., *Stigma; notes on the management of spoiled identity*. 1963, Englewood Cliffs, N.J.: Prentice-Hall.
4. Stafford, M.C., & Scott, R. R. , *Stigma deviance and*

social control: Some conceptual issues, in *The Dilemma of Difference*, L.M.C. M. Becker, Editor. 1989, Plenum: New York
5. Byrne, P., *Stigma of mental illness and ways of diminishing it.* Advances in Psychiatric Treatment, 2000. **6**(1): p. 65-72.
6. Link, B.G. and J.C. Phelan, *Conceptualizing stigma.* Annual review of Sociology, 2001: p. 363-385.
7. Major, B. and L.T. O'Brien, *The Social Psychology of Stigma.* Annual Review of Psychology, 2005. **56**(1): p. 393-421.
8. Katz, I.T., et al., *Impact of HIV-related stigma on treatment adherence: systematic review and meta-synthesis.* Journal of the International AIDS Society, 2013. **16**: p. 18640-n/a.
9. Chambers, S.K., et al., *A systematic review of the impact of stigma and nihilism on lung cancer outcomes.* BMC Cancer, 2012. **12**(1): p. 184.
10. de Vries, J., et al., *Ethical issues in human genomics research in developing countries.* BMC Medical Ethics, 2011. **12**(1): p. 5.
11. Phelan, J.C., *Genetic bases of mental illness–a cure for stigma?* Trends in neurosciences, 2002. **25**(8): p. 430-431.
12. Phelan, J.C., *Geneticization of deviant behavior and consequences for stigma: The case of mental illness.* Journal of Health and Social Behavior, 2005. **46**(4): p. 307-322.
13. Angermeyer, M.C., et al., *Biogenetic explanations and public acceptance of mental illness: systematic review of population studies.* Br J Psychiatry, 2011. **199**(5): p. 367-72.
14. Kvaale, E.P., N. Haslam, and W.H. Gottdiener, *The 'side effects' of medicalization: A meta-analytic review of how biogenetic explanations affect stigma.* Clinical psychology review, 2013. **33**(6): p. 782-794.
15. Collier, J., *Photography in anthropology: a report on two experiments.* American Anthropologist, 1957. **59**(5): p. 843-859.
16. Analí a Inés, M., *Picturing Students' Habitus: The Advantages and Limitations of Photo-Elicitation Interviewing in a Qualitative Study in the City of Buenos Aires.* International Journal of Qualitative Methods, 2010. **9**(2): p. 149-171.
17. Cooper, C.M. and S.P. Yarbrough, *Tell Me—Show Me: Using Combined Focus Group and Photovoice Methods to Gain Understanding of Health Issues in Rural Guatemala.* Qualitative Health Research, 2010. **20**(5): p. 644-653.
18. The, H.A.C., *Enabling the genomic revolution in Africa: H3Africa is developing capacity for health-related genomics research in Africa.* Science (New York, N.Y.), 2014. **344**(6190): p. 1346-1348.
19. Mason, T., *Stigma and social exclusion in healthcare.* 2005.
20. Parker, R. and P. Aggleton, *HIV and AIDS-related stigma and discrimination: a conceptual framework and implications for action.* Social science & medicine, 2003. **57**(1): p. 13-24.
21. Thompson, W.E. and J.L. Harred, *Topless dancers: Managing stigma in a deviant occupation.* Deviant Behavior, 1992. **13**(3): p. 291-311.
22. Thompson, W.E., J.L. Harred, and B.E. Burks, *managing the stigma of topless dancing: a decade later.* Deviant Behavior, 2003. **24**(6): p. 551-570.

23. Opala, J. and F. Boillot, *Leprosy among the limba: Illness and healing in the context of world view.* Social Science & Medicine, 1996. **42**(1): p. 3-19.
24. DeJong, W., *The Stigma of Obesity: The Consequences of Naive Assumptions Concerning the Causes of Physical Deviance.* Journal of Health and Social Behavior, 1980. **21**(1): p. 75-87.
25. Bloom, J.R. and L. Kessler, *Emotional Support Following Cancer: A Test of the Stigma and Social Activity Hypotheses.* Journal of Health and Social Behavior, 1994. **35**(2): p. 118-133.
26. Cahill, S.E. and R. Eggleston, *RECONSIDERING THE STIGMA OF PHYSICAL DISABILITY.* Sociological Quarterly, 1995. **36**(4): p. 681-698.
27. Malcolm, A., et al., *HIV-related stigmatization and discrimination: Its forms and contexts.* Critical Public Health, 1998. **8**(4): p. 347-370.
28. Herek, G.M., *Beyond "Homophobia": Thinking about sexual prejudice and stigma in the twenty-first century.* Sexuality Research & Social Policy, 2004. **1**(2): p. 6-24.
29. Vishwanath, T., *Job Search, Stigma Effect, and Escape Rate from Unemployment.* Journal of Labor Economics, 1989. **7**(4): p. 487-502.
30. Biewen, M. and S. Steffes, *Unemployment persistence: Is there evidence for stigma effects?* Economics Letters, 2010. **106**(3): p. 188-190.
31. Rasmusen, E., *Stigma and Self-Fulfilling Expectations of Criminality.* The Journal of Law and Economics, 1996. **39**(2): p. 519-543.
32. Elliott, G.C., et al., *Understanding stigma: Dimensions of deviance and coping.* Deviant Behavior, 1982. **3**(3): p. 275-300.
33. Bos, A.E.R., et al., *Stigma: Advances in Theory and Research.* Basic and Applied Social Psychology, 2013. **35**(1): p. 1-9.
34. Livingston, J.D. and J.E. Boyd, *Correlates and consequences of internalized stigma for people living with mental illness: A systematic review and meta-analysis.* Social Science & Medicine, 2010. **71**(12): p. 2150-2161.
35. Keusch, G.T., J. Wilentz, and A. Kleinman, *Stigma and global health: developing a research agenda.* The Lancet. **367**(9509): p. 525-527.
36. Weiss, M.G., J. Ramakrishna, and D. Somma, *Health-related stigma: Rethinking concepts and interventions.* Psychology, Health & Medicine, 2006. **11**(3): p. 277-287.
37. Skinner, D. and S. Mfecane, *Stigma, discrimination and the implications for people living with HIV / AIDS in South Africa : original article.* SAHARA : Journal of Social Aspects of HIV / AIDS Research Alliance, 2004. **1**(3): p. 157-164.
38. Logie, C. and T.M. Gadalla, *Meta-analysis of health and demographic correlates of stigma towards people living with HIV.* AIDS Care, 2009. **21**(6): p. 742-753.
39. Clement, S., et al., *What is the impact of mental health-related stigma on help-seeking? A systematic review of quantitative and qualitative studies.* Psychological Medicine, 2015. **45**(1): p. 11-27.
40. Hood, L. and S.H. Friend, *Predictive, personalized, preventive, participatory (P4) cancer medicine.* Nature Reviews Clinical Oncology, 2011. **8**: p. 184.
41. Bush, W.S. and J.H. Moore, *Chapter 11: Genome-Wide Association Studies.* PLOS Computational Biology, 2012. **8**(12): p. e1002822.
42. Savulescu, J. and J. Kerin, *The "geneticisation" of disease stigma.* The Lancet, 1999. **354**: p. SIV16.
43. Shakespeare, T.O.M., *Choices and Rights: Eugenics, genetics and disability equality.* Disability & Society, 1998. **13**(5): p. 665-681.
44. Kong, C., M. Dunn, and M. Parker, *Psychiatric Genomics and Mental Health Treatment: Setting the Ethical Agenda.* The American Journal of Bioethics, 2017. **17**(4): p. 3-12.
45. Phelan, J.C., R. Cruz-Rojas, and M. Reiff, *Genes and stigma: The connection between perceived genetic etiology and attitudes and beliefs about mental illness.* Psychiatric Rehabilitation Skills, 2002. **6**(2): p. 159-185.

46. Phelan, J.C., L.H. Yang, and R. Cruz-Rojas, *Effects of attributing serious mental illnesses to genetic causes on orientations to treatment.* Psychiatric Services, 2006.
47. Sankar, P., et al., *What is in a cause? Exploring the relationship between genetic cause and felt stigma.* Genetics in Medicine, 2006. **8**(1): p. 33-42.
48. Weiner, B., *An Attributional Theory of Achievement Motivation and Emotion*. Vol. 92. 1985. 548-73.
49. Corrigan, P.W., *Mental Health Stigma as Social Attribution: Implications for Research Methods and Attitude Change.* Clinical Psychology: Science and Practice, 2000. **7**(1): p. 48-67.
50. Easter, M.M., *"Not all my fault": Genetics, stigma, and personal responsibility for women with eating disorders.* Social Science & Medicine, 2012. **75**(8): p. 1408-1416.
51. Meiser, B., et al., *Implications of genetic risk information in families with a high density of bipolar disorder: an exploratory study.* Social science & medicine, 2005. **60**(1): p. 109-118.
52. Laegsgaard, M., et al., *The perceived and predicted implications of psychiatric genetic knowledge among persons with multiple cases of depression in the family.* Acta Psychiatrica Scandinavica, 2010. **122**(6): p. 470-480.
53. Singer, P.A. and A.S. Daar, *Harnessing Genomics and Biotechnology to Improve Global Health Equity.* Science, 2001. **294**(5540): p. 87-89.
54. Bustamante, C.D., F.M. De La Vega, and E.G. Burchard, *Genomics for the world.* Nature, 2011. **475**: p. 163.
55. Folarin, O.A., A.N. Happi, and C.T. Happi, *Empowering African genomics for infectious disease control.* Genome Biology, 2014. **15**(11): p. 515.
56. McGregor, J., *Racial, ethnic, and tribal classifications in biomedical research with biological and group harm.* The American Journal of Bioethics, 2010. **10**(9): p. 23-24.
57. Tekola, F., et al., *Impact of social stigma on the process of obtaining informed consent for genetic research on podoconiosis: a qualitative study.* BMC Medical Ethics, 2009. **10**(1): p. 13.
58. Tekola, F., et al., *Tailoring consent to context: designing an appropriate consent process for a biomedical study in a low income setting.* PLoS Negl Trop Dis, 2009. **3**(7): p. e482.
59. Marsh, V.M., D.M. Kamuya, and S.S. Molyneux, *'All her children are born that way': gendered experiences of stigma in families affected by sickle cell disorder in rural Kenya.* Ethnicity & health, 2011. **16**(4-5): p. 343-359.
60. Carapetis, J.R., M. McDonald, and N.J. Wilson, *Acute rheumatic fever.* The Lancet, 2005. **366**(9480): p. 155-168.
61. Robertson, K.A. and B.M. Mayosi, *Rheumatic heart disease: Social and economic dimensions.* SAMJ: South African Medical Journal, 2008. **98**: p. 780-781.
62. Robertson, K.A., J.A. Volmink, and B.M. Mayosi, *Towards a uniform plan for the control of rheumatic fever and rheumatic heart disease in Africa-the Awareness Surveillance Advocacy Prevention (ASAP) Programme.* South African Medical Journal, 2006. **96**(3): p. 241-245.
63. Irlam, J., et al., *Primary Prevention of Acute Rheumatic Fever and Rheumatic Heart Disease With Penicillin in South African Children With Pharyngitis A Cost-Effectiveness Analysis.* Circulation: Cardiovascular Quality and Outcomes, 2013. **6**(3): p. 343-351.
64. Barth, D.D., et al., *Effect of distance to health facility on the maintenance of INR therapeutic ranges in rheumatic heart disease patients from Cape Town: no evidence for an association.* BMC health services research, 2015. **15**(1): p. 1.
65. Carapetis, J.R. and L.J. Zühlke, *Global research priorities in rheumatic fever and rheumatic heart disease.* Annals of pediatric cardiology, 2011. **4**(1): p. 4.
66. Engel, M.E., et al., *Genetic susceptibility to acute rheumatic fever: a systematic review and meta-analysis of twin studies.* PLoS One, 2011. **6**(9): p. e25326.

67. Erasmus, Z., & Pieterse, *Conceptualising Coloured identities in the Western Cape Province of South Africa*, in *National identity and democracy in Africa*, M. Palmberg, Editor. 1999, Human Sciences Research Council Cape Twon. p. 167-187.
68. Saul, J.S. and P. Bond, *South Africa - The Present as History From Mrs Ples to Mandela and Marikana.* 2014.
69. Posel, D., *Race as Common Sense: Racial Classification in Twentieth-Century South Africa.* African Studies Review, 2014. **44**(2): p. 87-114.
70. Africa, S.S., *Census 2011: Community Profile Databases*. 2012, Statistics South Africa: Pretoria.
71. Zühlke, L.J., et al., *Group A Streptococcus, Acute Rheumatic Fever and Rheumatic Heart Disease: Epidemiology and Clinical Considerations.* Current Treatment Options in Cardiovascular Medicine, 2017. **19**(2): p. 15.
72. *NVivo qualitative data analysis Software*. 2015 QSR International Pty Ltd.
73. Hennink, M., I. Hutter, and A. Bailey, *Qualitative research methods*. 2015, London: SAGE Publications Ltd.
74. Solomon, G., et al., *Understanding of genetic inheritance among Xhosa-speaking caretakers of children with hemophilia.* Journal of genetic counseling, 2012. **21**(5): p. 726-740.
75. Braun, V. and V. Clarke, *Using thematic analysis in psychology.* Qualitative Research in Psychology, 2006. **3**(2): p. 77-101.
76. Guillemin, M., *Understanding Illness: Using Drawings as a Research Method.* Qualitative Health Research, 2004. **14**(2): p. 272-289.
77. Harper, D., *Talking about pictures: A case for photo elicitation.* Visual studies, 2002. **17**(1): p. 13-26.
78. Webster, M., *Similarities and differences in the meanings children and their parents attach to epilepsy medications.* Social Science & Medicine, 2017. **177**: p. 190-197.
79. Rosenthal, A.I., et al., *Community effect of alcoholism: A photovoice study in saint vincent and the grenadines.* Psychiatric Quarterly, 2016.
80. Wang, C. and M.A. Burris, *Photovoice: concept, methodology, and use for participatory needs assessment.* Health Education & Behavior: The Official Publication Of The Society For Public Health Education, 1997. **24**(3): p. 369-387.
81. Killion, C.M. and C.C. Wang, *Linking African American mothers across life stage and station through photovoice.* Journal of Health Care for the Poor and Underserved, 2000. **11**(3): p. 310-325.
82. Gallo, M.L., *Picture this: Immigrant workers use photography for communication and change.* Journal of Workplace Learning, 2002. **14**(2): p. 49-57.
83. Foster-Fishman, P., et al., *Using Methods That Matter: The Impact of Reflection, Dialogue, and Voice.* American Journal of Community Psychology, 2005. **36**(3/4): p. 275-291.
84. Epstein, I., et al., *Photo Elicitation Interview (PEI): Using Photos to Elicit Children's Perspectives.* International Journal of Qualitative Methods, 2006. **5**(3): p. 1-9.
85. Harrison, B., *Seeing health and illness worlds – using visual methodologies in a sociology of health and illness: a methodological review.* Sociology of Health & Illness, 2002. **24**(6): p. 856-872.
86. Keller, C., J. Fleury, and A. Rivera, *Visual Methods in the Assessment of Diet Intake in Mexican American Women.* Western Journal of Nursing Research, 2007. **29**(6): p. 758-773.
87. Samuels, J., *Breaking the Ethnographer's Frames: Reflections on the Use of Photo Elicitation in Understanding Sri Lankan Monastic Culture.* American Behavioral Scientist, 2004. **47**(12): p. 1528-1550.
88. Greenhalgh, T., *How to read a paper: Papers that summarise other papers (systematic reviews and meta-analyses).* BMJ, 1997. **315**(7109): p. 672-675.
89. Patton, M., *Qualitative Research & Evaluation Methods (Thousands Oaks, Sage).* 2002.

90. Pink, S., *Interdisciplinary agendas in visual research: re-situating visual anthropology.* Visual Studies, 2003. **18**(2): p. 179-192.
91. Glaw, X., et al., *Visual Methodologies in Qualitative Research: Autophotography and Photo Elicitation Applied to Mental Health Research.* International Journal of Qualitative Methods, 2017. **16**(1): p. 1609406917748215.
92. Pauwels, L., *Visual Sociology Reframed: An Analytical Synthesis and Discussion of Visual Methods in Social and Cultural Research.* Sociological Methods & Research, 2010. **38**(4): p. 545-581.
93. Mehta, S. and A. Farina, *Is being "sick" really better? Effect of the disease view of mental disorder on stigma.* Journal of Social and Clinical psychology, 1997. **16**(4): p. 405-419.
94. Yang, L.H., et al., *Culture and stigma: adding moral experience to stigma theory.* Social science & medicine, 2007. **64**(7): p. 1524-1535.
95. Condit, C.M., *How the public understands genetics: non-deterministic and non-discriminatory interpretations of the "blueprint" metaphor.* Public Understanding of Science, 1999. **8**(3): p. 169-180.
96. Ledger, T., *An Empty Plate: Why We are Losing the Battle for Our Food System, why it Matters, and how We Can Win it Back*. 2017: Jacana Media.
97. Crandall, C.S. and R. Martinez, *Culture, Ideology, and Antifat Attitudes.* Personality and Social Psychology Bulletin, 1996. **22**(11): p. 1165-1176.
98. de Vries, J., et al., *Investigating the potential for ethnic group harm in collaborative genomics research in Africa: Is ethnic stigmatisation likely?* Social science & medicine, 2012. **75**(8): p. 1400-1407.
99. Richman, L.S. and M.R. Lattanner, *Self-regulatory processes underlying structural stigma and health.* Social Science & Medicine, 2014. **103**: p. 94-100.

APPENDICES

APPENDIX 1: BEFORE/AFTER QUESTIONNAIRE: EVALUATION STUDY

	Questionnaire: Mode *(Genetic, Mixed, Environmental)*, **Medium:** *Video or Read*, **Focus Group Number**					
	Sex:	**M / F**				
	Age:					
	Highest level of education:	**Number:**				
	Where do you live?	**URBAN / PERI-URBAN / RURAL**				
	Have you had an operation?	**How long ago?**				
		1	**2**	**3**	**4**	
		Strongly disagree/	**Disagree**	**Agree**	**Strongly Agree**	
General interest						
1	Rheumatic Heart Disease (RHD) is an important disease to talk about with everyone.					
2	RHD should be something that everyone learns about.					
3	I think it's important to have conversations about RHD.					
Personal engagement						
4	I am really interested in sharing my experiences of having RHD.					
5	I find it meaningful to have other people with RHD tell their story about living with RHD.					
6	I feel I have a duty to talk about living with RHD.					
Empathy						
7	I feel sad when I meet other people with RHD.					

8	I easily relate to other people with RHD.					
9	When I see other people with RHD, I think of my own experiences of living with RHD.					
		1	**2**	**3**	**4**	
		Strongly disagree/	**Disagree**	**Agree**	**Strongly Agree**	
10	Hearing about people living with RHD, helps me think and talk about my own experiences.					
Willingness to share in a group						
11	It is easy to talk about my experiences of living with RHD.					
12	I am willing to talk to other people about having RHD.					
13	I feel it is important for me to share my experiences of living with RHD with other people.					
Emotional response						
14	I feel sad when I see other people living with RHD.					
15	It upsets knowing that many other people are living with RHD.					
16	I do not like to talk to other people about having RHD.					

APPENDIX 2: SCREEN SHOTS OF VIDEOS USED IN FGD

Figure A: Enrico is introduced

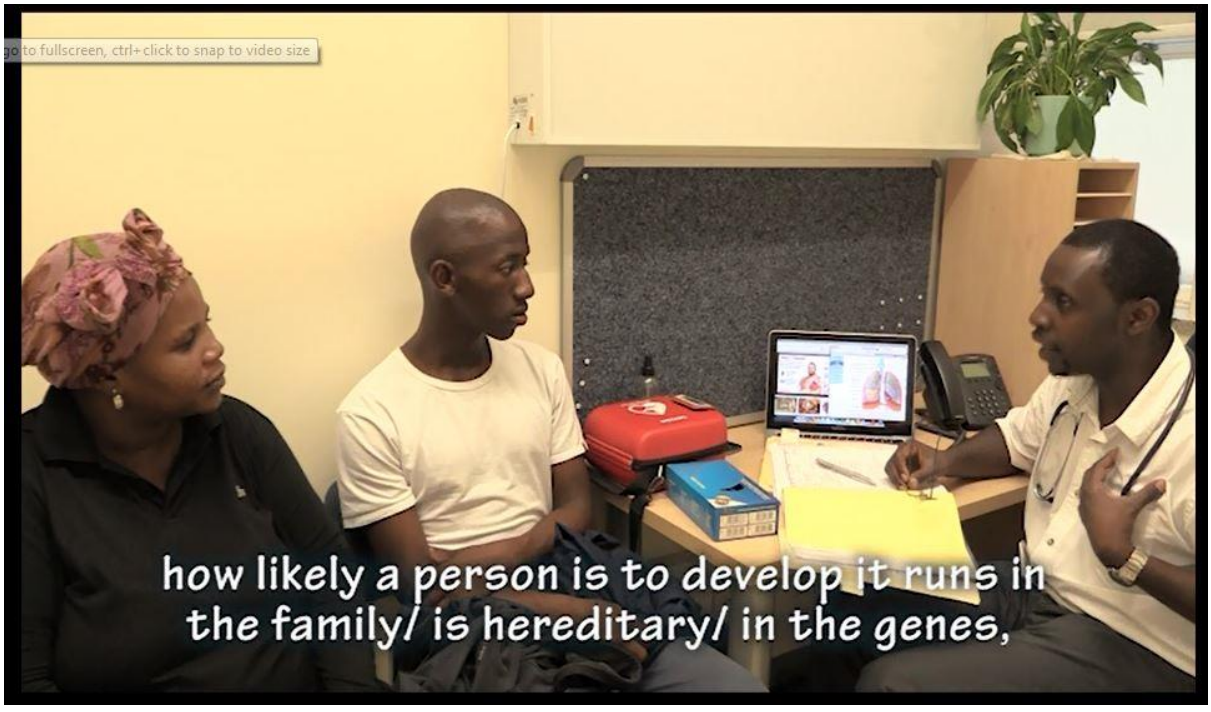

Figure B1: Genetic causation of RHD explained to Enrico and his mom

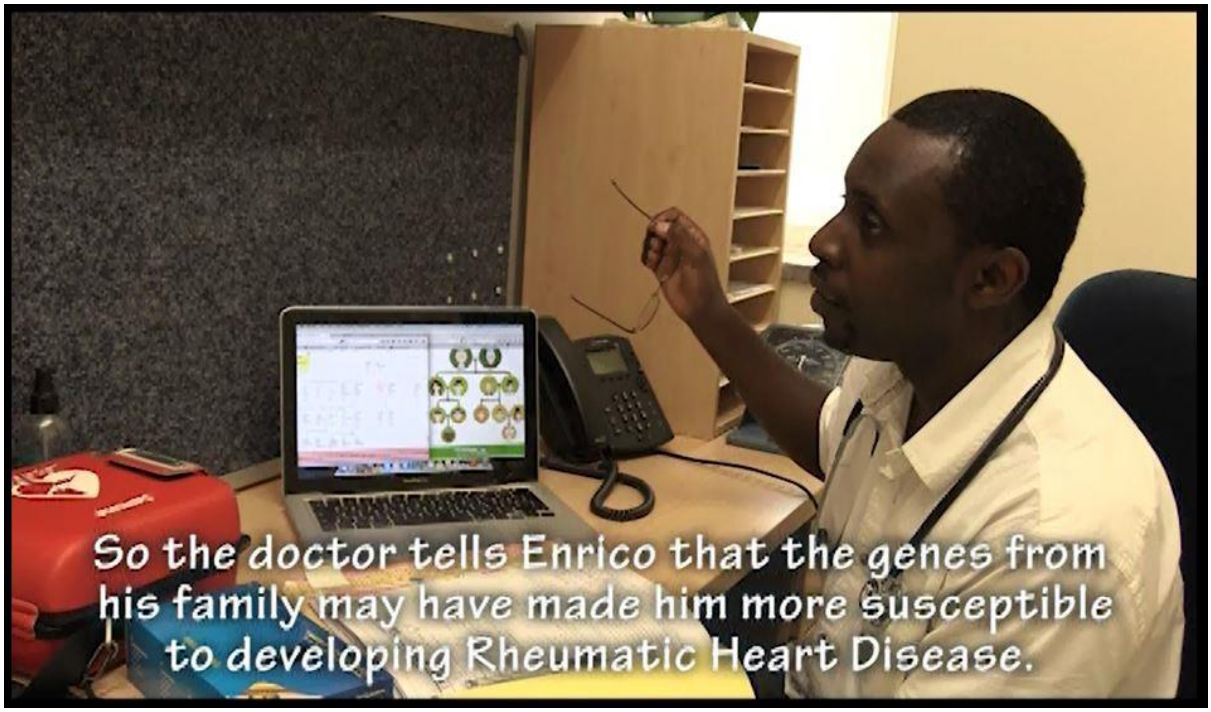

Figure B2: Genetic causation of RHD explained to Enrico and his mom

Figure C1: Mixed causation of RHD explained

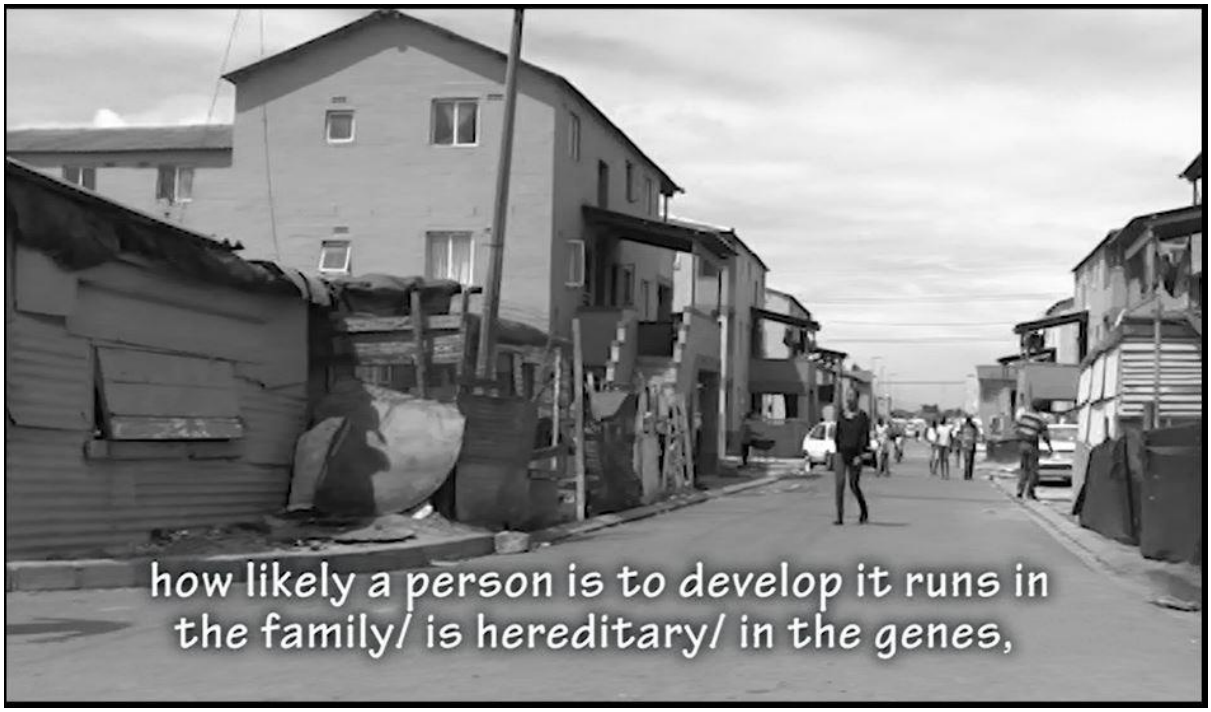

Figure C2: Mixed causation of RHD explained

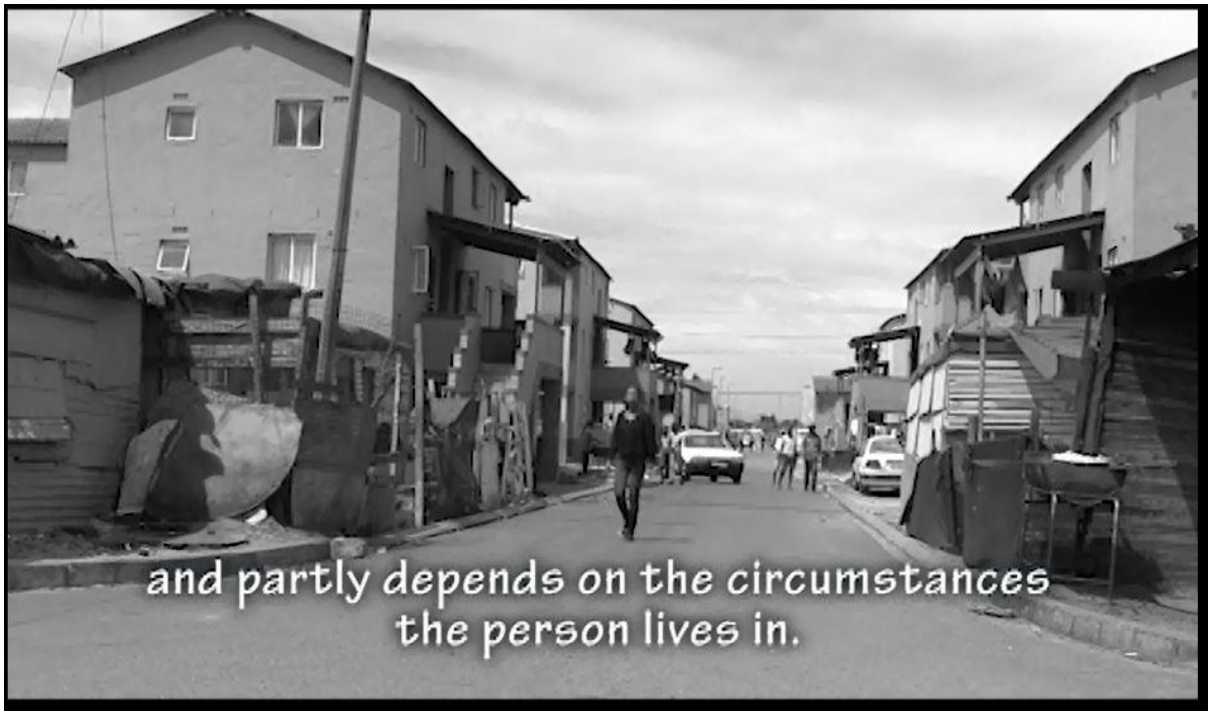

Figure C3: Mixed causation of RHD explained

Figure D: Exploring stigma and social distance

APPENDIX 3: FOCUS GROUP DISCUSSION TOPIC GUIDE

Vignette: Stage 1

Enrico is 26 years old and lives with his mother in an RDP house in the Bonteheuwel area. He is unemployed and gets a government disability grant each month to support them. Enrico has rheumatic heart disease or what is also called valve disease, and first became ill four years ago. He then became very tired and was often short of breath. He also went to the hospital many times for simple things that other people would not get very ill of, like flu. His mother found this very strange and worrying. She took him to the hospital where the doctor told them that Enrico had rheumatic heart disease and began treating him.

- What do you know about rheumatic heart disease?
- What do you think causes it?
- How will having rheumatic heart disease affect Enrico's life?
- How do you think other people will respond to Enrico when they know he has rheumatic heart disease?
    - Is it likely that Enrico will be looked down on by others?

Vignette: Story stage 2

(i) *Genetic*: Enrico wanted to know what had caused his rheumatic heart disease. The doctor explained that although this disease is triggered by an infection, how likely a person is to develop it runs in the family/ is hereditary/in the genes, which means that it was passed down from his parents and previous generations in the family. He asked if Enrico remembered anyone else in the family who had similar problems. Enrico remembered having an uncle who also had heart problems.

- So the doctor tells Enrico that his rheumatic heart disease would not have happened but for something passed down in his family. If other people find out about it, how will this explanation affect Enrico's life? [After open-ended responses, follow up with specific probes below, if not already mentioned; to explore further ask "How?"]
    a. Will it change how his family relates to him?
    b. Will it change how his friends relate to him?
    c. Will it affect the chances that a boss would give him a job?
    d. Will it affect the chances that he'll get married?
    e. Will it affect his decisions about having children?
    f. Will it change the chances that he will want treatment?
    g. How else might it change his life?

(ii) *Mixed environmental/genetic*: Enrico wanted to know what had caused his rheumatic heart disease. The doctor explained that although this disease is triggered by an infection,

how likely a person is to develop it partly runs in the family/ is hereditary/in the genes, and partly depends on the circumstances the person lives in. The doctor asked where Enrico grew up. Enrico tells him that for most of his life, she lived in a shack they shared with his aunt. The doctor asked if Enrico could remember whether he had ever had a really sore throat, followed by pain in his body. He remembered that he was very ill when he was about six, and then again in his early twenties. The doctor tells Enrico that his disease is caused by a combination of things in his blood (his genes), the sore throat and where he grew up.

- So the doctor tells Enrico that his rheumatic heart disease would not have happened but for something passed down in his family and the circumstances in which he grew up. If other people find out about it, how will this explanation affect Enrico's life? [After open-ended responses, follow up with specific probes below, if not already mentioned; to explore further ask "How?"]
    a. Will it change how his family relates to him?
    b. Will it change how his friends relate to him?
    c. Will it affect the chances that a boss would give him a job?
    d. Will it affect the chances that he'll get married?
    e. Will it affect his decisions about having children?
    f. Will it change the chances that he will want treatment?
    g. How else might it change his life?

(iii) *Environmental*: Enrico wanted to know what had caused the rheumatic heart disease. The doctor explained that although this disease is triggered by an infection, how likely a person is to develop it partly depends on the circumstances the person lives in. For example, people who live in crowded, poorly ventilated houses are more likely to get RHD. He asked if Enrico could remember whether he had ever had a really sore throat, followed by pain in his body. He remembered that he was very ill when he was about six, and then again in his early twenties.

- So the doctor tells Enrico that his rheumatic heart disease would not have happened but for the circumstances in which he grew up. If other people find out about it, how will this explanation affect Enrico's life? [After open-ended responses, follow up with specific probes below, if not already mentioned; to explore further ask "How?"]
    a. Will it change how his family relates to him?
    b. Will it change how his friends relate to him?
    c. Will it affect the chances that a boss would give him a job?
    d. Will it affect the chances that he'll get married?
    e. Will it affect his decisions about having children?
    f. Will it change the chances that he will want treatment?
    g. How else might it change his life?

Vignette: Stage 3

Enrico's mother is friendly with your parents and you all happen to meet up one day. Enrico's mother wants to introduce you to her son and hopes that the two of you would become friends.

- How would you feel about becoming friends with Enrico?
- How would you feel about introducing Enrico to other people in your family?
- How would you feel if Enrico wanted to date or marry your sister?
- Do you think Enrico's children would also develop this disease? All of them, or just some of them?

APPENDIX 4: CONSENT FORMS TO BE USED FOR THIS STUDY

Title of the Study: Stigma in African genomics research on Rheumatic Heart Disease

Introduction and summary
We would like to speak with you about rheumatic hearth disease ('valve disease') and how it affects your life. We are mostly interested in knowing whether having this disease causes you to feel like there is a stigma on you – that is, whether it makes other people look at you in a negative way. We would also like to talk with you about genetic research: what you understand about it, and if it could change stigma relating to your disease. We are asking you to take part in a group discussion on these topics. The other people in the group are patients with the same disease as you have. We will also ask you to do answer some questions on paper. All the discussions will take place in English and Afrikaans, depending on your preference. We will take about two hours for the discussion.

Objective
We hope to use the findings of this study to understand better if genetic research could influence the stigma relating to rheumatic heart disease.

The Researchers
This study is conducted by Marlyn Faure of the University of Cape Town. The project is led by Drs. Jantina de Vries and Megan Campbell at the University of Cape Town (UCT). They work with other researchers at UCT and in the United States.

Participants
We will conduct this study with patients who have rheumatic heart disease (RHD). We hope to speak with no more than 120 patients with RHD. We contacted you because you also participated in the RHDGen project.

Methods
If you agree to participate, you will be part of a group discussion with other patients that have the same disease as you do. The group will be small, about 6-10 people. The group discussion will consist of four parts:

1. We will first ask you to answer some questions on paper. This will include questions about your disease experiences;
2. We will then show you a video or tell you a story about a patient with your disease, and what the doctor tells him about how he got it;
3. We will ask you to complete the same questions again;
4. We will then conclude with a last group discussion where we talk about that story, and whether the story would change any of the stigma you may experience with your disease.

Risks and benefits
There are some risks relating to taking part in this study. Most of all, you could find it upsetting to talk about your disease and any stigma you will feel. We will talk with you about how you feel after the group discussion, to make sure you are not upset. If you would like to talk to us after you've left, then you can call us on 021 650 5716 and we will make sure to put you in touch with people who can help.
The benefit of this study is that it will help us understand whether genetic research can decrease stigma for rheumatic heart disease. With the findings, we will be able to do better

genetic research in the future. It may also be a benefit for you to be in a group of people who have the same disease as you do, to talk about how the disease makes you feel.

Privacy
We will record and write down the group discussion. At the start of the interview, we would like to get some information about you – including your name – but we will not share your name with anybody. When we write the discussion down it will not have your name on it. We will share the writings between all the researchers on the project. When we report on our research, we may use some of the sentences that you said. If we use one of your sentences, it will appear together with a brief description of you (for instance, 'Rheumatic Heart Disease patient'), and a code for the discussion (for instance, Focus Group 06).
However, it is important to remember that the discussion will take place in a group. We will ask everybody in the group to keep the discussion confidential, but it is possible that somebody could talk to others about it. If you want, you can use another name for yourself during the discussion, so that nobody in the group will know your real name. It is up to you if you want to do that.

Withdrawal
You do not have to answer all the questions during the group discussion. For instance, if we ask a question and it makes you feel uncomfortable, you can just tell us. Also, you can decide to leave the discussion at any time.

Compensation
We would like to thank you for the time that you took to be part of our study. We will compensate you R100 for your time and for transport.

Contact Information
If you have any questions or comments about this project, or if you want to talk more about the project, you can call us on 021 650 5716 (telephone Jantina de Vries). You can also call Peggy Mgwayi at 021 650 6361, who can put you in touch with the RHD Patient Advisory Group. If you prefer, you can also send us an email: jantina.devries@uct.ac.za.

APPENDIX 5: FULL SEARCH STRATEGY

Search string 1: “Photo-elicitation” OR “photo elicitation” OR “Photo-interviews” OR “Elicitation technique” OR “visual method” OR “visual methods” OR “visual methodology” OR “visual material” OR “visual stimulus” OR “visual illness narratives” OR Video Intervention/Prevention Assessment OR “Video intervention” OR “Visual narratives” OR “Visual ethnography” OR “Applied visual anthropology” OR Photovoice OR “Visual sociology” (in both title or abstract)

AND

Search string 2: “qualitative research” OR “focus group” OR “group discussion” OR interview

AND

Search string 3: Evaluat* OR effect* OR assessment OR review