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ORIGINAL PAPER

Exploring how a genetic attribution to disease relates to stigma experiences of Xhosa patients with schizophrenia in South Africa

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Abstract

Background Over the past three decades, a range of international stakeholders have highlighted the possibility that genomic research may impact stigma associated with psychiatric disorders. Limited research has been conducted in Africa to investigate this relation.

Method In the present study, using focus group discussions, we explored the relation between genetic attribution and stigma among 36 Xhosa people with schizophrenia. We addressed three main questions: (1) What causal beliefs do Xhosa people with schizophrenia use to explain their illness and to what extent do genetic explanations play a role in these beliefs? (2) What are the internalised stigma experiences of Xhosa people with schizophrenia? (3) How do genetic explanations relate to stigma experiences, if at all?

Results Most participants were able to define genetics and some linked genetics to disease causation. Despite adequate knowledge of genetics and an emphasis on genetic explanations of schizophrenia in the study, most participants held a multitude of causal explanations including: psychosocial, environmental, and cultural. Moreover, participants rarely mentioned disease cause when describing their stigma experiences.

Discussion For this population group, there was no straight-forward relation between a genetic attribution and stigma. Therefore, we did not find evidence that genetic attribution may significantly increase stigma. Although North American and European literature provides conflicting evidence regarding this relation, there is increased consensus that biomedical explanations for psychiatric disorders may reduce blame. This study found evidence supporting that consensus. This study provides an empirical foundation to inform ongoing work on the psychosocial implications of psychiatric genomics research in non-Western contexts.

Keywords Genetic attribution · Schizophrenia · Stigma · Xhosa people

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Background

The possibility that genomics research may impact on stigma experienced by people living with a disease is a commonly held psychosocial concern often raised by ethics committees and investigated by ethical, legal and social implications (ELSI) researchers. Studies in North American and European contexts propose two theories in relation to genetic attribution and the possible stigma that may emerge for different population groups. The first, based on the attribution theory, proposes that genetic information could reduce stigma by reducing some degree of personal responsibility and self-blame for developing the condition [1–4]. The second suggests that genetic information could increase stigma by making conditions seem fundamental and unchangeable, with little possibility for recovery with treatment [1, 5]. The latter theory is arguably rooted in concerns over eugenics [6] and in early experiences with genetic screening for conditions such as sickle cell disease [4, 7].

Three decades ago, the World Psychiatric Association suggested that stigma could be reduced by disseminating information on neurobiological causes for psychiatric disorders [8]. The rationale was that stigma would be lessened if people believed mental illnesses were caused by biological phenomena and not behavioural actions. This assumption contrasted with the concern raised by ELSI scholars in the 1990s—which continues to be highlighted by ethics committees—that increased genetic information may heighten stigma. Three decades later, Kong et al. suggest that an increased focus on genetic causes for mental diseases may contribute to deterministic thinking and essentialist views, which may increase existing stigma of psychiatric illnesses [5]. In contrast, Appelbaum attests that while this is possible, genomic research is not likely to be the main cause for increased stigma experienced by patients with psychiatric disorders [9]. Consensus is, therefore, lacking on whether a biological explanation for psychiatric disorders may increase or decrease stigma [10, 11].

For instance, in a vignette-based US study, Phelan and colleagues found that genetic attribution had no effect on social isolation (used as a measure for stigma) [4]; similarly, Pescosolido et al. found no effect of biological attribution on stigma [10]. In New Zealand, Breheny also found that genetic attribution had no effect on participants' willingness to interact with an individual who had schizophrenia, depression or skin cancer [12]. In contrast, in Germany, Angermeyer and Matschinger found evidence that endorsing a biological cause for a mental illness is associated with a desire for social distance [13]. Similarly in England, Bennet, Thirlaway and Murray found that attributing schizophrenia to genetics did increase stigma [14]. Taken together, empirical studies on the implications of attributing genetic explanations to diseases present inconsistent evidence as to whether this relation may increase or decrease stigma for individuals living with a psychiatric disorder [2, 14]. Furthermore, this work has been undertaken in high-income countries [1–4], so we are particularly unsure of this issue in other contexts [15, 16].

Currently, there are only four published studies that have begun to investigate this concern in Africa, none of which have focussed on a psychiatric disease [16–19]. Two of these studies [16, 18] caution about the potential of increased ethnic stigmatisation as a result of genomics research, particularly for individuals from already stigmatised or marginalised population groups. In Ethiopia, Tekola et al.'s study focussed on podoconiosis—a highly stigmatised physical disease—found that participants were afraid to participate in genomics research due to fear of receiving genetic information about the origin of the disease, which could contribute to increased social stigma [18]. In Kenya, Marsh, Kamuya and Molyneux found evidence of associative stigma among mothers of children with sickle cell disease [19]. In South Africa, Faure et al. explored the association between genetic attribution and rheumatic heart disease and found no evidence of an effect [17]. Whilst Faure et al., Tekola et al., and Marsh et al. focused on understanding these phenomena in relation to physical diseases, there is currently no empirical research on the relationship between genetic attribution and stigma in psychiatric diseases in Africa [18, 19].

There is, however, a robust literature on the causal attributions of mental illness in African settings [20–25] In South Africa, Mbanga et al. found that caregivers of Xhosa people with schizophrenia tend to attribute mental disorders to a complex variety of psychosocial and cultural explanations, as opposed to a biomedical explanation [25]. Similarly, psychosocial causal attributions were echoed in a sample of members of the South African general public [26]. However, when symptoms are obvious and observable, as in psychotic symptoms of schizophrenia, this behaviour tends to more commonly be attributed to witchcraft or possession by evil spirits [25]. In fact, Naanyu reiterates that in South Africa and most African communities in general, magical aetiologies are regularly cited causal models for mental illness [27].

Schizophrenia is known to be a highly stigmatised disease [25, 28] and in this study, we focus on South African Xhosa people with schizophrenia. Research suggests that generally, the South African public tends to be more stigmatising towards people with schizophrenia in comparison to people with other mental disorders [25, 26]; however, the perceptions of the people living with schizophrenia themselves have largely been ignored in the South African literature to date.

Contemporary research differentiates among perceived stigma, endorsed stigma, anticipated stigma, felt/received stigma and enacted stigma. Perceived stigma is the belief that "most people" will devalue or discriminate against the stigmatised [29]. Endorsed stigma is understood as agreement with commonly held stereotypes about the condition or disease. Anticipated stigma is defined as an individual or group expecting that others will devalue or stigmatise them [29]. Felt/received stigma focusses on the stigmatised individuals, establishing their recollections of discriminatory or stigmatised behaviour towards them [29]. Enacted stigma refers to the experience of discrimination that a stigmatised individual is subjected to by others (i.e. members of the family or community) [30]. Internalised/self-stigma involves an individual experiencing shame and expecting discrimination when others know about their condition [31, 32]. Associative/courtesy stigma relates to the stigma experienced by caregivers or family members of an individual with a particular disease [33].

This study explores how genetic attribution for disease relates to the stigma experiences of Xhosa people with schizophrenia by interrogating: (1) What causal attribution models Xhosa people with schizophrenia employ to explain their illness and to what extent genetic explanations play a role in these causal models? (2) What are the internalised stigma experiences of Xhosa people with schizophrenia? and (3) How genetic causal explanations relate to their stigma experiences, if at all?

Method

Following a qualitative approach, we enrolled 36 Xhosa outpatients with schizophrenia from the Eastern and Western Cape Provinces in South Africa, in 6 focus group discussions (FGDs) conducted between February and April 2017. During the FGDs, participants watched one of the three video-vignettes of a 26-year-old Xhosa male character who has schizophrenia. The vignettes were identical other than the cause of schizophrenia being explained as either genetic, environmental or a combination of genetic and environmental causes. The vignettes had three segments embedded with questions exploring: (1) participants' general understanding of schizophrenia and genetics; (2) participants' perceptions of how one's life may change after being diagnosed with schizophrenia and knowing that the cause is genetic or non-genetic; and (3) how participants' knowing that the cause of the disease is related to genetic or non-genetic explanations may impact internalised and associative stigma experiences. The lead researcher guided the FG discussions around these broad questions.

Thirty-four Xhosa males (94%) and two Xhosa females (6%) participated in the study. Their mean age was 33.92 years (range 20–53 years). Thirty (83.33%) had secondary schooling or above, yet 32 (88.89%) reported being unemployed.

All participants previously consented to participating in a psychiatric genomic study (the Genomics of Schizophrenia in South African Xhosa people (SAX) project)[34] and were subsequently recruited to participate in this study. All participants provided written consent. Procedures involving patients were approved by the University of Cape Town (FHS204-2015). Permission to conduct this research in the selected sites was obtained from the local hospital head nurses in the relevant units and from the South African Department of Health. The authors assert that all procedures contributing to this work comply with the ethical standards of the relevant national and institutional committees on human experimentation and with the Helsinki Declaration of 1975, as revised in 2008.

Data analysis

FGDs were conducted in isiXhosa. All FGDs were tape-recorded, transcribed verbatim and translated from isiXhosa into English for thematic analysis [35]. English versions of transcripts were imported into NVIVO 11 software, which is a computer program for managing text data [36]. FGD data were analysed using inductive thematic analysis [35]. Notes taken during the FGDs, including the sequence of responses, were used to allocate quotes to participants. No differences in responses were observed in the data regarding which vignette was watched by participants—therefore, all data were analysed as one data set. First, data were analysed through multiple rounds of coding by two researchers, which led to the identification of general themes in the data. Thematic domains were further developed into specific codes for conceptual categories which were applied to all the transcripts.

Since the data covered multiple complex concepts such as genetics, stigma and their relationship, we decided to present the data according to the specific questions explored in the study. The reason for this was to more clearly and appropriately contextualise the data within the broader narrative of the FGDs.

Results

Causal explanations and genetics

Prior to exploring participants' causal explanations, it was important to understand what participants know about genetics and heredity, as well as how they relate genetics to schizophrenia. We asked: "What do you know about genetics?"

Many respondents referred to heredity, particularly the inheritance of biological features and genes:

> "I think that… I think that genetics has to do with genes. So let's say for example your father… Say you inherited a gene from your father, then you have that gene. Later you realise that that gene is related to the disease your father has. Then you end up having the same disease." (P.7: FGD2)
> "Genetics, what I know about genetics is like … if there is someone, like your father maybe, there are these things called chromosome networks, where if your father gets mentally disturbed you also end up falling in the same path. You would be aware that its genetics which caused that." (P.1: FGD2)

Notably, these two examples were shared by participants with tertiary level (post-secondary school) education. Not all participants demonstrated the same depth of understanding. Some highlighted a range of traits that could be inherited; while, others related genetics to the idea of passing down phenotypic traits or behavioural attributes or habits of a parent.

Following the question "Do you think [character name] may have children who have the disease?" many participants were able to consider the possibility that an individual could pass on their disease to their offspring:

> "If for example he has five children. Then two of the children may have the disease, because it can be understood that the one or two children who have it, got it through inheritance." (P.1: FGD6)
> "When it is genetic it means that he can still have a child or maybe a grandchild who may have the disease." (P.10: FGD4)

Exactly how these traits are passed down from parents to children was not clear to participants, but most were certain that they are "somehow" passed down in the bloodline. Participants also demonstrated knowledge that a disease can skip a generation and be passed down to grandchildren and not their parents.

Most participants seemed to consider that genetics possibly plays a role in the development of schizophrenia, with some explaining that other factors may trigger disease onset in the case of a genetic predisposition to the disease. For instance,

> "Uh, something that could also put his children at risk of getting the disease is their own decisions. For example, if a child uses drugs too much, then it will be more likely for the condition to emerge if it is genetic. Or for example if he abuses alcohol, it could also emerge. You see. Or if he is really hurt. For example, I have a niece whose mother has schizophrenia. I also am a schizophrenia. But for me, schizophrenia emerged because I was using weed [cannabis] for a long time, then I stopped, and then the schizophrenia began. So I always advise my niece that if she has anything that is hurting her feelings, she should speak to someone about that. I tell her that because she is likely to get ill, because this thing is genetic. You see for me, maybe I wouldn't have been like this had I not used drugs. So as for [character's] child, he/she could have the illness, even though the symptoms may not emerge. It depends on the decisions he/she makes and things he/she abuses." (P.10: FGD4)

This view was supported by many other participants who generally seemed to suggest that an individual's children would be unlikely to develop this condition if they were not exposed to circumstances that could act as a "trigger." In the quote above for instance, in addition to the possibility of substance abuse serving as a trigger, the participant alludes to the possibility that the niece's "emotional stress" may also serve as a trigger.

Despite their general understanding of genetics, most participants placed less emphasis on a genetic explanation for their disease and considered it in conjunction with a range of other non-biomedical causes, including: psychosocial, environmental and cultural explanations. Respondents specifically commented on the negative psychosocial factors (i.e. severe poverty, trauma, stress, and physical/emotional abuse) they experienced in the past, along with present experiences, as having an influence on the onset of their mental illness. Environmental factors (such as living in communities with high unemployment rates, severe poverty, incidents of crime and violence as well as drug abuse) were cited by many participants as playing a role in the onset of their disease. Furthermore, cultural beliefs (i.e. bewitchment—which they defined as being cursed or poisoned) were also commonly cited as possible triggers for developing their disease.

> "What I think is that it is caused by witchcraft. Because I have experience with those things when it comes to me and things that have happened to me. So I think that maybe it does happen that, there is Satanism, or witchcraft which is involved. No matter what may be the main cause of it, but I do think that those things do play a role in triggering it." (P.5: FGD2)

The emphasis on supernatural causes, particularly related to a bewitchment framework, articulated by many participants in our study has commonly been cited in other empirical work examining causal illness models of Southern African population groups [25, 27, 37, 38]. Overall, the participants in our study suggested that there could be a multitude of causal explanations for the onset of schizophrenia.

Stigma experiences

In answering the second question regarding the stigma experiences of Xhosa people living with schizophrenia, guided by the work of others [31, 39], this study found that participants conceptualised their internalised stigma experiences in relation to stereotypes (negative connotations), prejudice (ignorance or misinformation), and discrimination.

Many participants associated schizophrenia with negative connotations of being "dirty." For instance, participant 3 in FGD5 said: "When the illness starts, it makes him dirty, and he becomes darker." "Dirtiness" is a commonly cited stereotype about people with schizophrenia from the perspective of the general public [13], family members [25, 40], traditional healers [23, 24] and health care professionals [41]. In this study, we found that participants also related those stereotypes to themselves. Feelings of disgust and shame expressed by some about engaging with a person with schizophrenia suggest the internalisation of these stereotypes. This is exemplified by one participant asking, "Who would want to be around someone who has schizophrenia?" Prejudiced views regarding schizophrenia were reported to impact participants' self-esteem and confidence.

In describing discriminatory experiences, some participants associated schizophrenia with being treated as "disabled" and unworthy of having the same human experiences as others (e.g. marrying and having children) or being abused. One participant said:

> "He [character] cannot get married when he suffers from schizophrenia. His wife could be laughed at or she may speak badly, even to other women about him. He cannot fit into marriage when he has a mental disorder. Even me, I cannot fit into marriage because I have a mental disorder. I would be laughed at and even my wife would be laughed at. How could she marry a person with a mental disorder?" (P.4: FGD3)

The abovementioned view was not supported by all participants, but there were some who felt strongly about it. A few participants described experiences of violent behaviour perpetrated on them by members of society. Overall, it seems these participants have internalised the misinformed stereotypes and views often associated with schizophrenia.

Genetic explanation, schizophrenia and stigma

In addressing the third question regarding the impact of genetic attribution on stigma, when asked "How do you think [character's name] life will change after the doctor tells him that his disease is genetic?" participants reported anticipating stigma in relation to finding a spouse or having children. Importantly, given that participants did not mention cause in their responses, we cannot confirm that the anticipated stigma was based on the cause of the disease being associated with genetics, rather than the schizophrenia label or symptomatic behaviours. See for instance:

> "Others may say that they like him and act like they like him, however that may not be true… I think people may crucify him for his disease." (P.9: FGD1).

The fact that many participants failed to mention the cause being genetic in their responses (despite it being emphasised in the questions and vignettes) suggests that they did not consider that causal explanation as an important source for increased stigma.

When asked "How do you think his friends or family will relate to him when they know that the disease is genetic?", the general perception was that friends and family members would treat the individual as inferior. Furthermore, enacted stigma behaviours expected from community members included teasing, bullying or verbal/physical abuse. Many participants felt that someone who has schizophrenia is often treated as an outcast, as people tend to reject them and maintain a social distance. For instance, one participant said: "People may start abusing him, like his friends, family or elders" (P.7: FGD1). However, participants did not articulate these descriptions in conjunction with the cause of the disease—but rather attributed them to the disease label itself.

When asked "How would you feel about becoming friends with [character]?", most participants reported that they would not want to. With few exceptions, most participants demonstrated having endorsed stigma though reports of limited tolerance and an intention of social distance from the vignette character or any individual with schizophrenia. This was also found when we asked, "Would you allow your sister to date or get married to [character]?" as many participants either laughed or reported that they would disapprove of [the character] dating or marrying their sibling.

> "I would not agree for my sister to get married to someone who suffers from a mental disorder. That would even make other people to laugh at my sister. People like her friends for example." (P.4: FGD3)

Whilst most participants' reasoning for not wanting to be associated with the character was related to the fear of social rejection from others, one participant articulated the fear of having his sister bear children with ill health.

> "I also would not agree at all … since he is ill and my sister is well and you find out that even if he would have a family, he might have children that do not have good health, just like him." (P.2: FGD3)

Again, most of the responses were not related to the genetic origin of the disease. Interestingly, some suggested that a genetic explanation for their disease may reduce

personal responsibility and blame from their family or community.

> "No, maybe if they thought that he is a drug addict but they find out that it is caused by heredity, they will treat him differently, because they now understand that it is not drugs that caused his illness" (P.3: FGD5)

Overall, the responses of participants did not provide evidence to support the concern that genetic information may significantly increase stigma experienced by Xhosa people with schizophrenia. However, it is important to note that some respondents expressed the view that if the disease was related to genetics, then the individual might experience less self-blame, especially if their disease onset was related to previous drug abuse—as mentioned by the participant above. The reservations expressed by some participants in getting married and having children with someone who has schizophrenia suggest that participants do anticipate stigmatisation and rejection from society due to an association with a person who has the disease.

Discussion

We had three main findings. First, we found that despite two of the three vignettes and subsequent questions placing emphasis on genetic causes, Xhosa people with schizophrenia reported a multitude of causal explanations. These included genetic, environmental, psychosocial and cultural explanations. Our participants consistently considered these causal explanations in conjunction with one another, rather than exclusively. Most importantly, participants demonstrated an understanding that even if an individual has a genetic predisposition for schizophrenia, the development of the disease could be affected by other factors that they describe as a "trigger." Triggers included stress, severe poverty, substance abuse or bewitchment. Second, we found that participants do experience stigma, particularly in relation to marital relations and having children. Furthermore, internalised stigma experiences included the commonly held stereotype of "dirtiness," which is associated with mental illness in other international studies, [13, 25, 41, 42] as well as by the South African general public [26], traditional healers [23] and Xhosa-speaking schizophrenia patients' caregivers [25]. Additionally, participants' internalisation of prejudicial perceptions was evident in descriptions of shame for having the disease. As found in previous studies [39], discrimination experiences were described in the form of social distance and rejection. Importantly, in this study, we found that participants did not mention cause when describing social distancing and rejection, which suggests that the cause was not considered as important. Third, we found that an emphasis on a genetic attribution for disease is unlikely to increase stigma for these participants. However, as suggested by international research [3, 4], we found it possible that an emphasis on a genetic explanation may reduce self-blame for these participants, as posited by the attribution theory.

Our findings differ from those reported in a study from the UK, which found that a genetic explanation of schizophrenia could increase internalised stigma experiences [14]. Our findings are congruent with those of Condit et al.'s study in the US, which suggests that people generally perceive the causes of their disease to be multifactorial [43]. With participants' holding multifactorial models, it is difficult to conclude that a genetic explanation alone may increase stigma. This finding supports our previous work with rheumatic heart disease patients from the mixed ancestry community in South Africa, which found that genetic attribution is unlikely to increase stigma [17]. Indeed, the findings in this study are more supportive of the theory that biomedical explanations may reduce personal responsibility and self-blame, which is currently the most consistent perspective in the literature [11].

Possible reasons for our findings are: first, individuals from African cultural groups have consistently endorsed non-biogenetic causes for mental illness [25, 27, 37]. As in other cultures, the distinct cultural beliefs and practices held by Africans seem to play an important role in the ways in which many individuals conceptualise disease causation. Second, the complex nature of the science regarding genetics' role in causing schizophrenia may be too difficult to relay to people with the disease, especially those with a low level of education. We know that research suggests that there is a strong genetic component to schizophrenia [44]. However, the associations found only imply a partial role in genetic causality. How heritability translates into inherited genetic variants in the aetiology of schizophrenia is complex in itself [45].

Last, the most obvious causal explanation held by our participants is the experience of severe poverty. Many diseases prevalent in South African low-income communities are intimately related to the experiences of deprivation resulting from the country's history of apartheid and segregation [38]. Even though it is 26 years post-apartheid, its consequences are still strongly felt in low-income communities—which is especially evident in the high physical and mental disease burden in these communities [46].

While many of the participants reported stigma in multiple domains, these stigma experiences converged in a complex manner with the historical, cultural and psychosocial realities of their lives. Even though these participants shared internalised stigma experiences, these experiences were not primarily discussed in relation to their disease causal explanations. Instead, the heterogeneous and dynamic set of causal beliefs and stigma experiences described intersect with the realities of South Africa's inequities. In conclusion,

our findings present some reassurance that genetic explanations of illness seem exceedingly unlikely to substantively increase stigma experiences of Xhosa people with schizophrenia. Empirical evidence from our study suggests that the link between genetic attribution and stigma is complex and contextual as well as cultural realities of individuals ought to be taken into consideration in genomics studies. Given that there is very little African work on this topic, our findings extend understandings of the African context and could be important to consider for research and practice involving African or non-Western people with psychiatric conditions in genomics research.

Limitations

Several limitations need to be acknowledged when considering the results of this study. First, the sample enrolled is limited and strongly skewed towards young males (94.44%, with an average age of 33 years). Although the male predominance is representative of the sex difference in the larger SAX genomics study [34, 47] and in other genetics studies with Xhosa schizophrenia patients [48], the resultant data may not adequately represent the views of older Xhosa people, as well as Xhosa women with schizophrenia. In the larger study, however, we recruited sixty additional schizophrenia inpatients which included more females. In that dataset, we did not find notable differences in reports of stigma experiences between men and women. Second, we recognise that the qualitative nature of this study means that it cannot directly address a causal relationship between genetic attribution and stigma, but rather it offers an initial empirical exploration of this concern, which needs to be further assessed in future research.

Our evidence could contribute to the development of future quantitative studies with larger sample sizes, using tools that incorporate the elements identified in this study. Furthermore, our findings could also inform future qualitative studies investigating the relationship between genetics and stigma among different population groups in non-Western contexts. It is important for future studies to consider the historical, contextual and cultural uniqueness of the population groups being investigated.

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Author contributions All eight authors contributed to the conceptualisation of the work. OM conducted the research. All authors commented on the findings and data analysis strategy. OM wrote the first draft and all authors commented. Revisions were made by OM and all the other authors agreed to the suitability of the final manuscript. JdV and MC provided supervision of the work.

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Compliance with ethical standards

Conflict of interests On behalf of all authors, the corresponding author states that there is no conflict of interest. All authors are not aware of any affiliations, memberships, funding, or financial holdings that might be perceived as affecting the objectivity of the review.

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