Stigma and illness uncertainty: adding to the burden of sickle cell disease
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Sickle cell disease (SCD) is one of the world’s foremost genetic diseases and has been declared a public health priority by the World Health Organization [1]. It can have multiple physical manifestations, such as recurrent pains, strokes, leg ulcers, among other ailments [2] and even though increasing, life span is still significantly shortened for persons living with the disease. The disease also has potential for multiple psychosocial effects, such as loss of independence, feelings of isolation and loneliness, poor interpersonal relationships and stigma.

Based on Goffman’s social theory of stigma, stigma is defined as ‘a phenomenon where an individual is rejected due to an attribute or behavior that is deeply discredited by society’ [3]. Health-related stigma results on the basis of a ‘socially discredited’ medical condition [4]. Persons with SCD frequently describe being stigmatized due to society’s misunderstandings about their disease, as well as due to their race as SCD is more prevalent in minority populations [5,6]. Even in Jamaica where it might be expected that stigmatization that may be due to racial disparities will not be experienced, patients frequently describe experiences of stigma [7] and delay in receiving care. They describe being ostracized due to fear of contagion and being thought of as ‘drug seekers’. Many will cope by hiding and withdrawing from social contact, disclosing only to few persons their SCD status, and not revealing when they might be in need of medical help [7].

Recurrent acute and chronic pain experiences are the hallmark of SCD [8] and often patients describe these and the quest for pain relief to be responsible for their experiences of stigma and isolation [9]. Many times patients are described as being ‘drug seekers’ as opioid analgesics are frequently required for pain relief in SCD. Attitudes of healthcare providers have been frequently found to be negative with patients experiencing undue delay in getting medicated for severe pain [10–12]. This sets in place an unrelenting cycle where some patients present recurringly due to...
under-treated pain and healthcare providers get more frustrated with providing for their needs. On the other hand, anticipating stigma can delay seeking healthcare when in need [4]. Empowering individuals with an illness that causes them to experience stigma can be a useful strategy to reduce the associated stigma. Listening to patients’ accounts, they describe part of their coping mechanisms include discussing the illness with their families and communities as well as educating them about SCD improves the support they receive from these persons [7]. It is also implicit that better understanding of their disease by the patient can lead to improved self-care as well as higher confidence in negotiating healthcare access [13]. When patients feel a loss of control over their lives due to their illness, especially when an illness may be unpredictable and life long, then it becomes a cognitive stressor and impacts adjustment and quality of life.

The concept of ‘illness uncertainty’ has been explored both in acute and chronic illnesses [14,15] and Mishel has defined it as the inability to determine the meaning of illness related events [16]. The influence of illness uncertainty has been examined in life-threatening conditions such as cancer [17–20], cardiopulmonary conditions [21,22] and HIV [23,24] and is found to be an additional psychosocial stressor on patients [25] and families [26]. Individuals living with SCD and their families may also experience uncertainty in everyday life due to the unpredictability of onset of a painful crisis and unforeseen complications, such as infections or the fear of early death.

The issue of illness uncertainty can also play an important role in both the patient’s and family’s experience and may influence the quest for a cure, decision-making about treatment options and discussing prognosis [27]. Reducing uncertainty can impact on reducing adverse psychological outcomes and improving coping [28,29]. Whether reducing uncertainty surrounding their illness will reduce stigma experiences is unknown.

The present study’s aim was to understand the stigma and illness uncertainty experiences in adults living with SCD; whether there is any relationship between these two outcomes; and to further identify factors which may worsen or improve these negative experiences. A secondary objective was to present initial psychometrics on the ‘Stigma in SCD’ and ‘Mishel’s Illness Uncertainty’ scales which have not been used in Jamaicans with SCD previous to this study.

Methods

The study population

All adult patients with SCD, ≥18 years of age, attending the Sickle Cell Unit (SCU), an outpatient clinic, at University of the West Indies (UWI), Mona Campus during the study period (November 2015–April 2016) were eligible for the study. Only well patients were recruited (i.e., those attending for routine, health maintenance visit or very minor illnesses, and not experiencing any acute event, such as painful crises, febrile episode or acute chest syndrome) during their visit. Patients who were ill at presentation to the clinic were excluded. Patients with significant cognitive problems which could hinder their ability to understand the questions were also excluded.

Procedures

Ethical approval was granted by the UWI Ethics Committee. A study coordinator not involved directly with clinical care of persons with SCD, approached adult patients at the SCU while they waited to see the doctor. Informed consent was given.

The questionnaire included information on socio-demographics, general health, Stigma in Sickle Cell Disease Scale (Adult) questionnaire [30] and Mishel Uncertainty in Illness Scale (Adults) questionnaire [16,31,32]. The socio-demographic section consisted of 13 questions and consisted of items, such as educational attainment, employment status, household possessions (used as an indicator of economic status) and crowding (calculated by dividing the number of persons living in the household by the number of rooms).

The Stigma in Sickle Cell Disease Scale (Adult) was adapted by the U.S.A. co-authors on this study [30], from the Berger’s HIV stigma scale [33,34] and the epilepsy stigma scale [35]. It consists of 40 items which are answered on a 4-point Likert scale ranging from 0 (not at all) to 3 (often) and total scores of up to 120. Higher scores imply greater level of stigma. It has been piloted in adults with SCD in the U.S.A. and Nigeria and shows internal consistency reliabilities of 0.86. The instrument’s four (4) components/subscales with factor analysis ranging from 0.79 to 0.89 are societal impact regarding the disease and isolation, personal feelings of shame, rejection, guilt, etc., treatment when in pain and concerns for the future, and sense of burden and needing assistance (0.79).

The Mishel Uncertainty in Illness Scale (Adults) [31] consists of 33 questions on a 5-point Likert scale with responses ranging from 1 (strongly disagree) to 5 (strongly agree) and total scores ranging between 33 and 165. Higher scores imply higher levels of illness uncertainty. The scale was developed not only for acutely ill adults but has also been widely used in patients with chronic illnesses, such as breast cancer, hepatitis C, prostate cancer and chronic obstructive pulmonary disease [20,22,32,36–39]. The instrument has four subscales: complexity, unpredictability, ambiguity and consistency with coefficient alphas between 0.65 and 0.86 for the subscales and 0.87 for
the overall scale. One of the investigators (RL) sought and received permission from Dr. Mishel to use the tool in this study.

The questionnaires were interviewer administered in English by the study coordinator in a quiet and private room within the clinic. Each participant took between 25 and 40 min to complete the study.

**Statistical data analysis**

All data were entered in Microsoft Excel 2013 and analyzed using the STATA SE 14.0 for Windows. Gender-specific estimates of means and proportions along with estimates of variability were determined for all socio-demographic and outcome variables. These variables were further compared using two-sample t test, the Wilcoxon rank-sum test, chi-squared test and analysis of variance as indicated. Pairwise Pearson’s correlation coefficient was calculated between the two outcomes’ mean scores.

Factor analysis with varimax rotation was done for both outcome scales. Items that had loadings greater than 0.4 were retained. Several models ranging from 2 to 5 factor solutions were examined and the most parsimonious model was determined based on: (1) eigenvalues >1; (2) total variance explained by the model and (3) intuitive meaningfulness of the factors described [40]. Mean subscale scores were calculated for each outcome. Cronbach’s alpha internal reliability coefficients were calculated for all stigma and illness uncertainty scales and subscales.

Age and sex controlled multiple linear regression analyses were conducted for each outcome to determine predictive variables.

**Results**

**Demographic**

Table 1 reports on socio-demographics of the study population. One hundred and one adult patients participated (45.5% males; mean age: 31.6 ± 10.4 years) with 72.2% having homozygous (SS) disease and 22.8% had heterozygous (SC) disease. Almost one-fifth (19%) had attained tertiary education, 33% had post-secondary or vocational attainment, 42% had secondary education as highest level of schooling and 6% had primary education only. Forty-one percent were employed in full-time positions, 40% were unemployed and the remaining had part-time employment. There were no sex differences in any socio-demographic variables, such as age, marital status, education level, household possessions or crowding.

**Stigma and illness uncertainty**

The overall mean stigma score was 33.6 ± 21.6 (range: 2–91), and there was no sex difference in the scores (males: 32.3 ± 21.3, females: 34.7 ± 21.9; p-value: 0.58). Internal consistency reliability score was 0.94. Factor analysis of the stigma scale (Table 2) showed that a 4-factor solution was the most parsimonious model and accounted for 59.1% of the total variance. The first factor (variance explained: 36.5%) was labeled ‘Society’s beliefs’ and included items, such as ‘I feel blamed’, ‘I fear loss of job’ and ‘I fear my significant other will reject me’. The second factor (variance explained: 9.4%) was labeled ‘Disclosure concerns’ and included items, such as ‘I regret having told persons’, ‘I am careful who I tell’ and ‘It’s easier to avoid friendships than telling them status’. The third factor was ‘Self-esteem/social conduct’ (variance explained: 7.2%) and included items, such as ‘I have lost friends’ and ‘People’s attitude make me feel worse about myself’. The fourth factor (variance explained: 6.1%) was labeled ‘Personal feelings of guilt/shame’ and included ‘I feel ashamed of my illness’ and ‘I feel my illness affects how my family is treated’ Table 4 shows that all subscales had very good reliabilities (Cronbach’s α: 0.6–0.9) and that the first two factors had higher means than the third and the fourth factors.

The overall illness uncertainty score was 85.9 ± 16.5 (range: 47–129), with females having higher scores, though not significant, than males (females: 88.7 ± 13.5, males: 82.6 ± 19.2; p-value: 0.07). Factor analysis

| Table 1. | Socio-demographic characteristics and main outcomes in study population by sex. |
| --- | --- |
| | Total (n = 101) | Males (n = 46) | Females (n = 55) | p-value |
| Age, years (mean (SD)) | 31.6 (10.4) | 30.9 (10.1) | 32.2 (10.7) | 0.53 |
| Genotype, N (%) |  |  |  |  |
| SS | 73 (72.2) | 32 (69.6) | 41 (74.6) | 0.76 |
| SC | 23 (22.8) | 11 (23.9) | 12 (21.8) |  |
| Others | 5 (5) | 3 (6.5) | 2 (3.6) |  |
| Employment status, N (%) |  |  |  | 0.21 |
| Full time | 41 (41.4) | 19 (41.3) | 22 (41.5) |  |
| Part-time | 19 (19.2) | 12 (26.1) | 7 (13.2) |  |
| Unemployed/student | 39 (39.4) | 15 (32.6) | 24 (45.3) |  |
| Marital status, N (%) |  |  |  | 0.38 |
| Never married/ separated/divorced | 64 (64.6) | 27 (60) | 37 (68.5) |  |
| Married | 9 (9.1) | 6 (13.3) | 3 (5.6) |  |
| Common law/visiting | 26 (26.3) | 12 (26.7) | 14 (25.9) | 0.97 |
| Highest level of schooling completed, N (%) |  |  |  |  |
| Primary and below | 6 (6) | 3 (6.5) | 3 (5.6) |  |
| Secondary | 42 (42) | 19 (41.3) | 23 (42.6) |  |
| Tertiary | 33 (33) | 16 (34.8) | 17 (31.5) |  |
| No. of persons in home, N (%) |  |  |  | 0.13 |
| 1–5 | 77 (76.2) | 38 (82.6) | 39 (70.9) |  |
| 6–9 | 20 (19.8) | 8 (17.4) | 12 (21.8) |  |
| ≥10 | 4 (4) | 0 (0) | 4 (7.3) |  |
| Crowding index, mean (SD) | 0.89 (0.69) | 0.79 (0.89) | 0.98 (0.69) | 0.22 |
| Household items, mean (SD) | 11.1 (3.5) | 10.6 (3.3) | 11.4 (3.6) | 0.25 |
| Stigma score (mean (SD)) | 33.6 (21.6) | 32.3 (21.3) | 34.7 (21.9) | 0.58 |
| Illness uncertainty score (mean (SD)) | 85.9 (16.5) | 82.6 (19.2) | 88.7 (13.5) | 0.07 |
once again revealed a 4-factor solution (Table 3) accounting for 46.1% of the total variance. Factors could be labeled as 'Ambiguity' (variance: 23%), 'Inconsistency' (variance: 11%), 'Unsurety' (variance: 6.9%) and 'Unpredictability' (variance: 5.2%). Internal consistency reliabilities were very good for all subscales ranging from 0.69 to 0.89 (Table 4). 'Unpredictability' and 'Inconsistency' had higher mean scores than the other two factors.

The two outcomes were significantly correlated though with a small correlation coefficient of 0.31.

**Predictors of main outcomes**

Table 5 reports on age and sex adjusted multiple regression analyses examining effects of key social factors on the main outcomes. Stigma was not associated with age or sex, but was significantly higher in those who lived in more crowded households (Coef: 7.89, \( p \)-value: 0.002) but lower in those who had higher household possessions (Coef: −2.26, \( p \)-value: 0.001).

Illness uncertainty was higher in females (Coef: 6.94, \( p \)-value: 0.02) and was significantly lower in those with a tertiary education as compared with those with a primary education (Coef: −16.68, \( p \)-value: 0.03).

**Discussion**

Although there is some research done in the past to study experiences of stigma in SCD, there has been no formal study in our Jamaican setting, where SCD is the most common genetic disorder and patients and families often report facing stigma and discriminatory behaviors at various levels. As noted by Weiss and Ramakrishna [41], cultural factors can affect stigma experiences and so it is important to study its presence and effects in diverse populations.

The study reports on the stigma experiences of our study population, encompassing the various domains of stigma as determined by the factor analysis results. Patients have described enacted stigma faced, as they report on society’s beliefs regarding their illness, and disclosure concerns they have, possibly arising.
from these experiences. In a genetic disorder, such as SCD, where disclosure is important for both seeking timely healthcare and in reducing the further transmission of disease, this would be an area that will require interventions at multiple levels: patient, families and other structural levels. Patients also describe self-perceived or ‘felt’ stigma through their experiences of lower self-esteem and personal feelings of guilt or shame. The felt stigma is thought to be even more disruptive in the lives of patients than enacted stigma [42] and can lead to withdrawal and inability to seek support [43]. Stigma research in the HIV/AIDS epidemic identifies similar stigma experiences that limit access to prevention, care, and treatment services [44]; and tackling stigma and discrimination have been identified as one of five key imperatives for success while developing a sustained response to the epidemic [45]. Stigma hence requires interventions at the patient level, including possibly psychotherapy and counseling.

The multidimensionality of stigma described in this study mirrors the results from another study that reports on patient experiences in the U.S.A. [46]. They also similarly report the ‘anticipated stigma and discrimination from society’ to be a bigger burden than personal feelings of guilt or shame. Other studies within the U.S.A. have posited that it is the high rates of disability and the stigma attached to this illness that may limit care-seeking for the higher levels of depression that are associated with it [47]. Experts also report that many psychosocial adjustment issues in SCD children may be wholly triggered or exacerbated by stigmatization of these individuals [48]. Even young adults, in whom unpredictable painful crises may be the norm, may feel inadequate to navigate the healthcare environment and so avoid the healthcare system whenever possible [49]. Nurses in emergency departments, intensive care units and surgical wards are shown to have equally negative attitudes towards persons with SCD [50]. Canadian mothers of babies with SCD describe facing coping challenges as well as stigma experiences which they ascribe to racism and which contribute to their feelings of social isolation [51]. It is important to note these similarities despite differences in location of the studies. Whereas stigma is thought to be partly a product of SCD being a ‘minority’ disease in countries,
Table 5. Multiple regression model examining stigma and illness uncertainty outcomes in adults with SCD.

| Outcome          | Adjusted R² | Predictors                  | Coefficient | SE     | p-Value |
|------------------|-------------|-----------------------------|-------------|--------|---------|
| Stigma score     | 0.29        | Age                        | −0.27       | 0.20   | <0.001  |
|                  |             | Female sex                 | 3.43        | 3.89   | 0.38    |
|                  |             | Genotypea                  | −3.50       | 4.53   | 0.44    |
|                  |             | SC                         | 18.44       | 8.48   | 0.03    |
|                  |             | Married statusb             | 7.12        | 6.94   | 0.31    |
|                  |             | Common law/visiting         | −8.06       | 4.34   | 0.06    |
|                  |             | Household items             | −2.26       | 0.68   | 0.001   |
|                  |             | Crowding                   | 7.89        | 2.53   | 0.002   |
|                  |             | Highest level of schooling completedc | −1.41 | −1.41 | 0.83 |
|                  |             | Post-secondary/vocational   | −8.92       | −8.92  | 0.20    |
|                  |             | Tertiary                   | −16.68      | −16.68 | 0.03    |
| Uncertainty score| 0.23        | Age                        | −0.24       | 0.15   | 0.12    |
|                  |             | Female sex                 | 6.94        | 3.01   | 0.02    |
|                  |             | Household items             | −1.10       | −1.10  | 0.06    |
|                  |             | Crowding                   | 2.32        | 2.32   | 0.25    |

*aReference category: SS genotype; bReference category: single; cReference category: primary.*

such as the U.S.A. or Canada, the results are similar in our population where those considerations are not relevant.

The study is one of the few that examines the experiences of illness uncertainty in adult patients with SCD. Illness uncertainty in a person living with a chronic illness affects their coping and generates stress and has in fact been viewed as one of the biggest challenges to successful adaptation [52]. Our patient population frequently describes experiences of ambiguity and unpredictability. The disease is variable in its course among and within individuals, and despite continuing efforts at understanding markers of disease severity; it still remains difficult to predict which patient will have more or less severe disease. Furthermore, a well-functioning individual can suffer acute and potentially significant complications at any time. It makes it difficult for patients to plan their lives and future direction, as from childhood itself they can have lower school attendance, cognitive dysfunction and thereafter lower functioning as adults. Persons with SCD face numerous psychological and social problems, such as depression, loneliness and poor quality of life [53–56]. Illness uncertainty itself is also associated with poorer psychological functioning in various chronic diseases [29,39,52], and further studies can help to better define this relationship as well as seek to determine if illness uncertainty may be one of the mediators to psychosocial dysfunction in SCD.

The positive relationship between stigma and illness uncertainty, though small, requires further understanding; there is only minimal literature which has assessed this relationship [57]. The concept of stigma is multifactorial [58] and involves many processes, including structural, personal, etc. [59] and even this level of correlation with illness uncertainty in the study is meaningful. This relationship could be exploited in developing interventions for reducing stigma experiences. Better understanding by patients of their disease process, which could aim to reduce ambiguity and inconsistencies especially, could lessen the stigma that individuals face and which no doubt adds to the daily psychological burden with which they cope. Jenerette et al. [60] have described that decreasing ‘vulnerability factors’, such as complications of SCD and recognition and response to them, can improve health outcomes, such as stigma.

Few social factors are seen to be associated with stigma and illness uncertainty in this study. Poorer socioeconomic status appears to worsen stigma and higher education seems to be associated with lower illness uncertainty. Other works also report that stigma is in fact more prevalent in those who are socially disadvantaged especially, poorer and less educated [58,61]. As poorer persons will usually have lower health-seeking behaviors [62,63], the co-existence of stigma can increase their experiences of unmet health needs and lower their perceptions of quality of care they receive [64]. Higher educational achievement was associated with lower levels of illness uncertainty, but whether this is related to greater knowledge about disease is uncertain as latter was not examined in this paper. However, previous work in our population has reported that females have higher knowledge about their SCD [65], and with the current study reporting that women have higher illness uncertainty; it suggests that disease knowledge may not be correlated with illness uncertainty. The results suggest that healthcare professionals will need to employ more appropriate strategies to impart health information which moves beyond simply imparting knowledge. Focus of interventions will include reducing uncertainty their patients face, especially related to disease complications, treatment plans and the unpredictability associated with their illness. Studies are also needed to understand why women face higher levels of illness uncertainty and gender-specific strategies may need to be developed to reduce the same.
Higher levels of social support can mitigate against both stigma [66] and illness uncertainty [22] Family and other personal support networks can assist a person to make better sense of their illness; peer-support can assist in coping and undermining their stigma experiences; and structural factors, such as more accessible quality healthcare services can improve patients’ healthcare experiences and reduce adverse outcomes.

One of the main limitations of the current study is that it was conducted in a clinic setting and experiences of those not routinely attending for health maintenance care could be different. Secondly, qualitative assessments could provide more depth to our understanding of stigma experiences as the scores on the stigma measures appear to be actually smaller than what is anecdotally reported by the patients. Also, no controls were employed in this study and so it is difficult to determine whether the effect of lower socioeconomic status on stigma is direct or worsened by the presence of SC, though our experience points to an interaction of both being most likely. Additionally, many other factors such as self-efficacy, self-esteem and the impact of complications that have been experienced as result of their SCD that may be implicated in stigma experiences of individuals were not assessed in this study. This may be one of the reasons why the models explored here explain limited variance in our outcomes. However, within psychosocial research, meaningful conclusions can be drawn about the effect of these significant predictors on the outcomes (Refs).

The study also provides confidence in using the current stigma scale used as well as the Mishel’s Illness Uncertainty scale for use in adult patients with SC. The stigma scale displayed a robust factor structure, mirroring the factors that are important theoretically of stigma experiences in SCD, and assesses both felt and enacted stigma. Studies in various other populations with SCD are needed to provide further evidence for continued use of both measurement scales. Ongoing and prospective studies will be needed to further assess their robustness and response to changes in patients’ lives.

Disclosure statement
No potential conflict of interest was reported by the authors.

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