Published in final edited form as:

Am J Med Genet A. 2019 May ; 179(5): 762–769. doi:10.1002/ajmg.a.61107.

Managing the need to tell: Triggers and strategic disclosure of thalassemia major in Singapore

Neha Kumar[1], **E Turbitt**[1,2], **Barbara B Biesecker**[1], **Ilana M Miller**[1], **Breana Cham**[3], **Katherine C Smith**[4], and **Rajiv N Rimal**[4]

[1]National Human Genome Research Institute, Bethesda, Maryland; [2]University of Technology Sydney, Graduate School of Health, Australia; [3]Department of Paediatric Medicine, KK Women's and Children's Hospital, Singapore, [4]Health, Behavior and Society, Johns Hopkins Bloomberg School of Public Health, Baltimore, Maryland

Abstract

This study explored patients' experiences and perceptions of living with thalassemia (an inherited hematologic disorder), perceptions of social stigma, and impact on disclosure decision-making. Semi-structured, in-person interviews were conducted in Singapore with 30 individuals: 16 thalassemia major patients and 14 parents of children with thalassemia. Findings were indicative of felt or enacted stigma that may have influenced disclosure decisions. Although affected individuals commonly disclosed their thalassemia diagnosis to family members, they either downplayed the condition with or avoided disclosure to unrelated individuals. Disclosure outside the family occurred only in response to triggers, such as questions about absences due to medical care. Health professionals should provide anticipatory guidance about disclosure strategies when managing individuals with thalassemia.

Keywords

Thalassemia major; stigma; genetic condition; Southeast Asia; disclosure decision making

INTRODUCTION

Thalassemia major is a chronic, hereditary, hematologic disorder. Untreated, thalassemia major can cause hemolytic anemia, and lead to cardiac disease, poor growth, bone changes, enlargement of the spleen, and other organ dysfunction (Cunningham, 2008). Treatment requires blood transfusions every 2–6 weeks along with iron chelation therapy via daily oral

Corresponding author at: Barbara B Biesecker, 701 13th St NW #750, Washington, DC 20005, United States, Phone: 202-370-2166 | bbiesecker@rti.org.

Conflict of interest: Author Kumar has no conflict of interest. Author Turbitt has no conflict of interest. Author Biesecker has no conflict of interest. Author Miller has no conflict of interest. Author Cham has no conflict of interest. Author Smith has no conflict of interest. Author Rimal has no conflict of interest.

Human studies and informed consent: All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2000. Informed consent was obtained from all patients for being included in the study.

medication (L1-deferiprone) and use of a subcutaneous infusion of Desferal by pump 5–7 nights a week for 8–10 hours to remove the resulting accumulation of iron (Cunningham, 2008; Olivieri & Brittenham, 2013a). While these treatments significantly impact individuals' daily lives, specifically at night, they also allow affected individuals to appear to lead relatively 'normal' lives where to others they seem ostensibly healthy.

Thalassemia major occurs worldwide but is more common in specific regions including Southeast Asia (Fucharoen & Winichagoon, 1992). Singapore is a developed multi-racial country in Southeast Asia with a population comprising mainly Chinese, Malay, and Indian ethnic groups. In Singapore, three percent of the population carries a pathogenic variant for thalassemia and face a higher chance for having an affected child (Fucharoen & Weatherall, 2016a). National preventative programs include public awareness campaigns, education and prospective screening services for couples planning a pregnancy (Fucharoen & Winichagoon, 1992; Weatherall, 2010).

In Singapore, the government has established an inclusive 'Singaporean' identity based on Asian values of emphasizing community over self. These community values are highlighted through the importance given to family, by taking part in the National Physical Fitness Award (NAPFA) test in school, carrying out compulsory military service for boys aged 18–21 years and putting an emphasis on achievement and success in general (National Library Board Singapore). This context is important for understanding the experience of stigma for individuals with thalassemia in Singapore.

Living with thalassemia may thus impact the family's honor or "face," community educational and employment opportunities, and abilities to participate in physical activities and military service. Further, the government ensures that the uniqueness of the three ethnic groups is maintained (Beng-Huat, 1998a; Thio, 2010). Identification with an ethnic group is likely to further influence the experience of stigma. While great importance is given to thalassemia major in Singapore, no research has explored the experiences and perceptions among those affected in this population.

Social stigmatization in the context of health has significant and varied effects on physical and mental health outcomes and contributes to inequities (Hatzenbuehler, Phelan, & Link, 2013). Individuals with stigmatized identities make an effort to decrease the personal and social effects of having an illness to maintain identities as 'good', 'normal' people, and to disavow identities as 'bad', diseased people (Goffman, 1963; Sandelowski, Lambe, & Barroso, 2004). Individuals can experience different types of stigma, for example: enacted or felt (Charmaz, 2000). Enacted stigma refers to its consequences, such as discrimination that arises from people being viewed as different. Felt stigma is stigma that is internalized as cognitions, affective responses or behaviors.

Stigma among individuals who suffer from illnesses that may not visibly manifest, such as HIV, epilepsy, or mental health disorders, have been described as "concealable stigmas" (Quinn & Chaudoir, 2009). The challenge of hiding a stigmatized identity depends on how the condition manifests and the effort to which patients are willing to go to keep it hidden. While people with concealable stigmatized identities may be able to hide their condition,

their self-identity as a stigmatized individual may remain and evidence suggests this identity can impact their psychological and physical wellbeing (Quinn & Earnshaw, 2013).

Despite the ability to appear healthy, a qualitative study in India indicated that individuals with thalassemia in disadvantaged settings may experience social isolation (Roy & Chatterjee, 2007). The literature suggests that thalassemia patients experience stigma and discrimination in social and professional settings (Atkin & Ahmad, 2001; Chattopadhyay, 2006; Shahraki-vahed, Firouzkouhi, Abdollahimohammad, & Ghalgaie, 2017; Wong, George, & Tan, 2011). Previous research with individuals affected by a milder form of thalassemia (thalassemia intermedia), found that some persons feel stigmatized (Ratip et al., 1995). The evidence of stigma, and the concealable nature of the condition, suggests that thalassemia may be described as a "concealable stigma" (Pouraboli, Abedi, Abbaszadeh, & Kazemi, 2014).

For individuals with concealable stigma, using discretionary disclosure strategies may be a powerful way to minimize the negative consequences of stigma. However, given the chronic and demanding nature of thalassemia, affected individuals may be confronted about their work and other daily life absences and face decisions about whether to disclose their condition. These decisions about whether, when, how, and to whom to disclose one's health status may be complex and challenging. Evidence suggests that the disclosure process involves weighing the perceived benefits against the perceived negative consequences (Johnson, Kass, & Natowicz, 2005; Sandelowski et al., 2004). To the best of our knowledge, disclosure patterns and decision making, including decisions about who to disclose to, when and how, have not been studied among individuals affected with thalassemia.

The objectives of this study are to: describe the impact of thalassemia on affected individuals' family, social and professional lives in Singapore, and better understand individuals' disclosure patterns; what they tell and to whom. We also aimed to contribute to literature on the disclosure decision-making process for individuals with chronic conditions and concealable stigmatized identities.

METHODS

Study design and theory overview

This study was carried out using an exploratory cross-sectional qualitative design. Semi-structured interviews were conducted in Singapore with individuals affected with thalassemia and parents of children with thalassemia. The discussion guide is included as supplemental material. Guided by social constructivism, a modified grounded theory approach was taken to create concepts and themes, emerging directly from the interviews (Charmaz, 1990).

Approval was granted from the National Human Genome Research Institute institutional review board (IRB) and the SingHealth Centralised IRB. Convenience sampling was used: participants were recruited from KK Women's and Children's Hospital Day Therapy Center and Genetics clinic, the Singapore Thalassemia Society. Snowball sampling was used whereby participants who previously participated in this study referred other individuals

with thalassemia or their parents. Recruitment and study participation lasted for three months from June 1 to August 30, 2010. Interviews were conducted until saturation was achieved; saturation was identified by the interviewer when no new concepts emerged from the interviews.

Informed consent and/or assent was obtained from all participants through verbal permission. Parents of participants under the age of 21 years (the age of majority in Singapore) also provided permission for their child to participate. Participants were also asked if their parent and/or child had participated in the study, or were scheduled or willing to do so. If they said yes, permission was obtained to establish parent-child dyads.

Inclusion criteria for individuals with thalassemia included: receipt of at least eight blood transfusions per year, 14 years of age and older (commensurate with IRB protocols, in Singapore, a child is a person below the age of 14, above this age, children may have the capacity to consent), and English speaking. Inclusion criteria for parents of children with thalassemia included; 21 years of age and older, having a child with thalassemia who required at least eight blood transfusions per year, having had caregiving responsibilities for this child, and English speaking. Parents themselves did not have thalassemia.

Procedures

Participation involved completing a one-page demographic information questionnaire and an interview. Most of the interviews were conducted in person during participants' blood transfusion appointments where individuals were in a private space for several hours. The lead investigator (NK) conducted all the interviews, which were audio recorded and transcribed verbatim. The interviewer completed a summary sheet following the interview to capture initial impressions of the interview. Along with interview data, these summary sheets were used to create emerging themes and shape data collection through modification of the interview guide as the study progressed. Data collection continued until no new concepts emerged.

The transcripts of the interviews were imported into NVivo (QSR International Pty Ltd.) and coded. Coding was performed the lead researcher (NK) and second coded by an independent coder (IM). Reliability between the coders was high (Cohen's kappa coefficient=0.84). Inductive thematic analysis was used to analyze the data (Braun & Clarke, 2006; Charmaz, 1990). The constant comparison method was used where similarities and differences in text coded under the same code were examined (Glaser, 1965). The role of demographic characteristics in leading to such similarities and differences was assessed. Additionally, data were obtained on each code from interviews with affected individuals and interviews with parents were compared for similarities and differences. Using this information, we identified themes and possible relationships among the themes. Illustrative quotes are included in the results. Some quotes were edited to aid readability; researcher modifications to the quotes are indicated by square brackets.

Reflexivity

The primary researcher (NK) was a genetic counselor in training at the time the study was conducted with specific interest in the experience of genetic disease in the cultural context of

Southeast Asia. NK had met with individuals with disabilities in Southeast Asia, and was concerned about the perceived limited support for individuals living with genetic disease in the region. These prior experiences and perceptions may have shaped the results of this study.

RESULTS

Participant Characteristics

A total of 30 participants completed the study. This included 16 individuals with thalassemia and 14 parents of children with thalassemia. There were five parent-child dyads. As parents were primarily interviewed to triangulate findings, only the demographic characteristics of the affected individuals are reported (Table 1). Given there were five parent-child dyads in this study, data were ultimately obtained from 25 individuals. Interviews lasted from 30–180 minutes; the length depended on how much time participants had available and how much they were willing to engage and share with the interviewer.

Major Themes

Living with thalassemia: medical management—Participants described their medical management requirements as painful and time-consuming activities that interfered with other activities, and made them feel different from ‘normal.’

> “Negatives, of course there’s the needles, the pain. The trauma.”
>
> (Patient #1, over 21)

Participants indicated that the biggest challenge was the infusion of Desferal due to its inconvenience and required frequency, and even more so because it offered no immediate benefit or relief. Many participants reported that they were not compliant with their Desferal treatment regimen. Most also found the monthly blood transfusion regimen challenging due to the time commitment required and its interference with other activities:

> “Like right now […] my iron is very high, partly it is due to the fact that I have not been chelating properly and it is due to the fact that I’m trying to lead the fullest of life possible.”
>
> (Patient #8, over 21)

However, blood transfusions did have an immediate impact on participants’ energy levels and participants were even able to extract some additional benefits. For example, the opportunity blood transfusions gave them to spend a day by themselves.

> Interviewer: *“…how about in terms of like right now you are sitting here [receiving a blood transfusion] rather than going shopping or whatever?*
>
> Interviewee: *“Oh, I’m okay…actually, I appreciate having one day every three weeks…I feel like taking a day off from work or I get to spend time with myself.”*
>
> (Patient #15, over 21)

Living with thalassemia: impact on social and professional life—The aspects of individuals’ lives affected as a consequence of living with thalassemia included: social life,

sporting activities, education, and employment. The challenges indicate patients and parents may feel different from ‘normal’ and socially devalued, experience discrimination and face psychosocial challenges. These responses suggest that perceptions of stigma are likely prevalent among individuals in Singapore living with thalassemia.

Study participants stated thalassemia had a negative impact on their social life in terms of their ability to make friends and participate in social activities. Participants described this was largely due to their time-consuming medical management demands, though some said this was also the result of feeling different from ‘normal’ and hence socially devalued:

> “…[you] don’t have that [many friends] if you [have] thalassemia…you wouldn’t be so like active or outgoing…[because] you feel like you are different from others so you don’t mix much.”
>
> (Patient #10, over 21)

Conversely, several participants expressed that as a result of thalassemia, they had the opportunity to interact with other individuals affected with thalassemia and thus become a part of a different social network.

Due to the physical impact of thalassemia, many participants talked about being to participate in normal physical education (PE) and sporting activities, including government-mandated programs such as the National Physical Fitness Assessment (NAPFA) and National Service (NS).

> “I would rather be normal and do PE rather than have [thalassemia] and don’t do PE.”
>
> (Patient #3, under 21)

Further, some participants felt thalassemia had an impact on their educational prospects. For most, having thalassemia did not impact their educational achievement. However, some participants described that they faced different parental expectations regarding their educational prospects and ability in comparison to their siblings and peers. Additionally, eight participants expressed that they were not allowed to go oversees for further education, a common practice in Singapore, because of their thalassemia.

> *“You can’t study overseas […] Because like if you go there, you’re debilitating their resources.”* (Patient #6, under 21)
>
> “… [My parents] mapped out her [sister’s] life, her career, her work but they didn’t want to put any expectations on me”
>
> (Patient #8, over 21)

For the majority, thalassemia had an impact on the type of jobs they pursued, as they had to consider the amount of physical activity and travel required, the amount of flexibility available, and whether the remuneration would be sufficient to cover thalassemia-related expenses:

> “Because of finances, it affects what kind of job I have to do. Honestly, I don’t like banking but because of money, I have to do it […] actually my passion is, I love

> animals […] I hope to do something that is animal-related […] I can't because it's not practical."
>
> (Patient #13, over 21)

Participants also reported that thalassemia had an impact on their ability to obtain a job, as employment applications in Singapore require medical conditions to be declared and the recruitment process often involves a medical exam by a physician.

A small minority of individuals did not feel concerned about discrimination when asked about specific circumstances, for example:

> Interviewer: *"Are you worried about employment discrimination at all?"*
> Interviewee: *"No, because they understand our condition. They know thalassemia, [they think] you may be more hard working, […] just to prove you can manage"*
>
> (Patient #10, over 21)

> "I told my principal […] I might take a couple of days off sometimes to see the doctor, but I will never let it affect my work. And he was very sympathetic. […] He knows about [thalassemia], and he says, sure, go ahead."
>
> (Patient #1, over 21)

Disclosure decision making: Influential factors—When participants made disclosure decisions they considered social, normative, strategic and practical factors. Participants described social factors predominantly pertaining to the nature and length of the relationship with the confidant. The closer the relationship between participants and their target of disclosure, the more likely they were to share full details of their thalassemia. For example, when one participant was asked if his friends knew about his thalassemia he replied:

> "No… it is very secret stuff… it is around the relationship in the family only."
>
> (Patient #8, under 21)

When asked about to whom s/he disclosed, another participant stated:

> "I only tell very, very, very close friends. Even those that I know for like a few years… I'm not very close to them, I will not tell them."
>
> (Patient #15, over 21)

One parent of a patient stated:

> "…his friends also, I think one or two only know, the rest he [didn't] want to tell them."
>
> (Parent #5 of patient under 21)

In terms of normative factors, participants considered how other people managed disclosure decision making and modelled their behavior on these other individuals. This included their friends with thalassemia and their parents.

Strategic factors described as influencing disclosure decisions included the affected individual's personal values, the perceived limited awareness of thalassemia in the

Singaporean community, and perceived benefits weighed against negative consequences of disclosure.

> *"With thal[assemia] [disclosure] becomes difficult because people have no idea what it is."* (Parent #14 of patient over 21)

> "So, a lot of people don't know what thal is, so there's no point explaining to them. I mean, maybe I tried, but they were still, they don't know what it means."
>
> (Patient #1, over 21)

Practical considerations influencing disclosure decisions included the impact thalassemia would have on various aspects of their life including physical activity, attendance at school or ability to secure and succeed at a job, particularly due to time commitment to medical interventions.

> "Uh, every time I went for an interview I only look for companies that does not do, does any medical examinations. And, so far none of the company as you know my condition."
>
> (Patient #13, over 21)

Different types of stigma further influenced disclosure decisions. Many participants described felt stigma and disclosure behaviors that were aimed at avoiding anticipated discrimination. Enacted stigma was experienced by several participants who reported discrimination or social devaluation as a result of their thalassemia. Participants also had negative self-perceptions as a result of their thalassemia and thus experienced internal stigma.

> "[at the time of the birth someone said why] don't you just leave her in a hospital and have another child […] you know, just dump her somewhere and have another child"
>
> (Parent #14 of patient over 21)

> Participant: *"I think most of the people, patients, that are working now, they don't tell [their employers] until maybe after they have [been hired]. So, there's still a stigma."*
>
> Interviewer: *"When you say stigma, what do you mean?"*
>
> Participant: *"If you say that you [have] thalassemia, they won't employ you."*
>
> (Parent #1 of under 21)

Disclosure decision making: Variation in the extent of disclosure—Some individuals fully disclosed their thalassemia. For many participants, full disclosure occurred in the context of active (or purposeful) rather than passive (accidental) disclosure and therefore was initiated by the individuals themselves. In this study, full disclosure often took place because of a participant's relationship with the confidant for example, fully disclosed their thalassemia to 'serious' dating partners.

> "I think the conversation [with my girlfriend] was something like, 'I've got something to tell you,' 'What is it, don't make me worried,' 'It is just that […] I

> have this anemic condition which requires once a month transfusions.'…Then you bring in the point about Desferal and L1 after that."
>
> (Patient #9, over 21)

Some individuals described downplaying their thalassemia when they disclosed their condition to others. This involved partial disclosure such as participants telling others about their blood transfusions but not about their iron chelation, in particular use of Desferal.

> "I downplay it a lot. So, in my opinion, how they see it is, not a big deal. Because really, my life goes on as normal. I go out with them and party. […] But that's because I downplay it a lot. I think a lot of them don't know that I inject myself at home."
>
> (Patient #1, over 21)

Further, participants indicated that when downplaying their condition, they tended not to tell others about the painful, demanding, and lifelong nature of medical management required and its impact on other aspects of their lives. They also avoided topics such as the psychological impact of living with thalassemia, the financial burden associated with it and their fears or concerns for the future. Many of the participants who made some form of disclosure to people beyond their family members described that they used downplaying terms such as *"anemic thing,"* or *"medical condition."*

Some individuals described making deliberate efforts to avoid disclosure of their thalassemia. They talked about using avoidance strategies when presented with a disclosure trigger and would not offer a sufficient response to the questions they were asked.

> "My colleagues say, 'why do you need to go for check-up?' because I say, 'I can't join you guys for lunch today, I have to go to the hospital.' […] Naturally they ask, 'are you okay?' At first, I say, 'Oh, it is just a regular checkup' But if they know I go monthly then I say, 'oh, the doctor needs to monitor my blood level" […] and then I stop there and I change the subject. So, I think they get the idea that I don't want to talk about it."
>
> (Patient #13, over 21)

Participants talked about using cover-up strategies, including 'white lies' in response to direct questions. One adolescent male reported that he was often asked about why he needed to take medication. In response, he would: *"tell a lie […] Maybe it is a vitamin or mineral tablet or something."*

> (Patient #2, under 21).

Disclosure decision making: Events triggering disclosure of thalassemia— Most participants did not wish to disclose their thalassemia to the extent possible. However, several circumstances or triggers prompted individuals to disclose thalassemia. For many participants, if not for these triggers, they may not ever have disclosed their condition. For others, these triggers served as an opportunity and convenient medium that enabled disclosure.

For most participants, the demands of medical treatment represent a major disclosure trigger, including taking time off from work or school for doctor appointments and explaining missing certain activities to accommodate blood transfusions.

> "So, when somebody asks me […] I say, 'oh, I need to go to hospital' and they go 'why do you need to go hospital?' Then I say, 'oh yeah, I forgot I have to tell you I have to go…' and I'll tell [them about thalassemia]."
>
> (Patient #11, over 21)

For many participants, thalassemia-related physical limitations represent another major trigger, and include inability to participate in physical education (PE), the National Physical Fitness Assessment (NAPFA) and National Service (NS). For other participants, questions about health status represent another trigger for disclosure, and typically arise during the process of applying for a job, undergoing a school- or employment-related medical examination, or recognition by others of visible signs associated with thalassemia such as appearing pale or tired.

> "[when] I go for [job] interviews, I will look at the forms […] I will just read carefully. Some of the medical questions they ask whether 'do you have any disease' or whether 'do you have any mental illness'. [Thalassemia] is not a disease. [Thalassemia] is not mental illness so I just put no […]. But if they mention pre-existing illness, I will just put [thalassemia]."
>
> (Patient #15, over 21)

Several study participants made a self-directed effort to disclose their thalassemia to certain individuals in their lives. For example, one participant disclosed her thalassemia to people close to her without any trigger because she anticipated that she would be asked questions and presented with disclosure triggers if she did not.

> "It's quite hard to hide it from them because like, I don't know, like why are you eating medicine in the middle of the day and that kind of thing, like where do you go on Saturdays. […] I just accept it and tell straightaway and that kind of thing."
>
> (Patient #6, under 21)

DISCUSSION

Disclosure patterns and decision making about disclosure emerged as components of primary importance to participants in our study. As such, disclosure decision making and the role of stigma within the decision-making process became a central focus of our findings.

Stigma has been associated with many chronic, invisible conditions because of the negative impact on an individual's identity and because living with the condition can make him or her different from 'normal'. Indeed, our results show that while a few participants did not feel stigmatized, many others recounted experiences that were indicative of felt or experienced stigma. Felt stigma was demonstrated among our participants who described worry about their ability to get a job or to get married. Participants in our study talked about enacted stigma in terms of experiencing discrimination in their social and professional lives. Individuals with stigmatized identities try to decrease the personal and social effects of

having a stigmatizing illness, to maintain identities as ‘normal’ people, and to disavow identities as diseased people (Sandelowski et al., 2004). One way to accomplish this is to utilize discretionary disclosure strategies, a major component of information control (Goffman, 1963; Kang, Rapkin, Remien, Mellins, & Oh, 2005).

Disclosure strategies employed by individuals with concealable stigmatized identities such as thalassemia include secrecy, spontaneous disclosure, protective disclosure, and preventive disclosure (Joachim & Acorn, 2000). In our study, some participants chose to downplay their thalassemia through being secretive about certain aspects of their condition; for example, by not telling about their iron chelation as this seemed to be a particularly invasive aspect of living with thalassemia. Participants did engage in spontaneous disclosure (full exposure of self with no or minimal control or planning over the disclosure) when presented with disclosure triggers and sometimes engaged in protective disclosure (planning deliberate and voluntary disclosure), particularly when disclosing to dating partners. Study participants tended not to utilize the preventive disclosure strategy (educating others to influence social judgements about the condition), though one participant did mention that proactive disclosure to employers may help them to be perceived as a ‘better’ worker.

The most important factor predicting the type of disclosure strategy utilized was participants’ social relationship with the confidant, with a distinction between people within and outside the family. Individuals tended to fully disclose their thalassemia to family members and dating partners. They utilized downplaying and nondisclosure strategies with people outside their family unit. Among our sample, this distinction between people within and outside the family may be related to the value placed on family within the Singaporean cultural context. However, studies of chronic illness in other countries have similarly found social relationships to be an important consideration for disclosure decision making. Among HIV positive individuals, disclosure to strangers and employers rarely occurs, whereas disclosure to healthcare providers and close friends and/or family was common (Bairan et al., 2007). Studies of adults with cystic fibrosis and individuals with Sickle Cell Disease (SCD; an inherited blood-borne disorder) have found similar patterns of disclosure based on closeness of social relationships (Kass et al., 2004; Modi, Quittner, & Boyle, 2010).

Our study showed that disclosure triggers generally prompted individuals to consider disclosing their thalassemia to people outside the family. Previous research around disclosure decision making and resulting models of the process have not included the role of disclosure triggers that participants talked about in our study (Chaudoir & Fisher, 2010). Participants described that they rarely considered revealing their condition without the presence of a trigger, and even in cases where a participant had decided to tell people outside their family, they almost always used triggers as an opportunity to make the disclosure. Triggers included medical management demands, apparent physical limitations due to thalassemia, and more general questions from others about health status. A recent study exploring the motivations of individuals with SCD describes “situation-related” reasons for disclosure (Derlega, Maduro, Janda, Chen, & Goodman, 2018). They give examples of situation-related reasons such as another person was present at the time of a pain episode, or another person knew about SCD-related hospitalization. However, they found this was a less frequently described motivation for disclosure compared to more frequently cited

motivations such as personal motivations: receiving emotional support, empathy and understanding.

Research and clinical implications

Our study has both clinical and research implications. Considering all participants were presented with disclosure triggers and had to find ways to manage and respond to triggers, health professionals, particularly genetic counselors, may consider raising the discussion of disclosure with patients and parents, early in the clinical relationship to plan ahead. This could involve general information about disclosure, resources for them to get additional support they may need, as well as opportunities to interact with other individuals with thalassemia. Such a discussion would provide patients with anticipatory guidance and thus enable them to manage disclosure triggers in a way that feels comfortable and appropriate to them. At the time of the study, genetic counselors were not involved in the management of patients with thalassemia. As genetic counseling grows as a profession in Singapore, there may be opportunities for them (for example) to support patients during treatment visits to help them mitigate the challenges they face with social stigma and managing disclosure decisions. Additionally, the Singapore Thalassemia Society could provide additional resources around how to navigate disclosure decision-making including via peer-to-peer support for individuals living with thalassemia in Singapore.

Based on our clinical understanding, the medical management of thalassemia is consistent across the world and thus would assume to impact individuals' lives in similar ways outside of Singapore. Further, prior accounts of the impact of thalassemia on individuals' social and professional lives in other parts of the world seem consistent with our findings (Atkin & Ahmad, 2001; Chattopadhyay, 2006; Shahraki-vahed et al., 2017; Wong et al., 2011), though our ability to make direct international comparisons is limited due to a paucity of research in this area. We can hypothesize that some aspects of living with thalassemia may be specific to Singapore due to some of the unique features of living in Singapore: the importance of family, participation in the NAPFA test in school, compulsory military training for boys aged 18–21 years, and emphasis on achievement and success in general. Future research endeavors should include conducting a larger, quantitative study assessing disclosure decision making in this population as well as others. This would provide insight into factors influencing disclosure decision making that are specific to the Singaporean context.

The findings of this study should be interpreted within the limitations of the study. As no comparative data was obtained from individuals living with thalassemia in other parts of the world, the findings cannot be assumed to be specific or unique to the Singaporean or Asian context. To our knowledge, no such evidence exists in other countries for comparison. Future research could, for example, explore the impact of living with thalassemia on family identity, given the importance placed on this concept in Singapore. Similar research in other parts of the world would help to determine what aspects of the study's findings are unique to the Singaporean context. As saturation was realized, the sample was of sufficient size to raise hypotheses to be tested. Yet, individuals who were willing to be interviewed may have been more likely to have had positive disclosure experiences and more open about their thalassemia. Additionally, demographic variables such as age, sex, race and educational

level may have influenced individuals' willingness to participate and their responses For example, a larger proportion of the sample was of Chinese ethnicity than of Malay or Indian. Lastly, as is common in qualitative research, the researchers' interests may have biased the study; however, the research team acknowledge this and were reflexive in designing and carrying out the study.

Conclusion

In describing the impact of living with thalassemia, many participants described perceptions of stigma in a variety of situations. Participants worried about their ability to get a job, or get married, and experienced discrimination in their social and professional lives. This stigma plays an important role in how participants approach decision-making about disclosure. We found that participants engaged in strategic discretionary disclosure, with many tending not to disclose their condition to people outside of their family and instead utilized nondisclosure and downplaying strategies. In general, the chronic and demanding nature of thalassemia served as a disclosure trigger and thus played an important role in shaping disclosure decisions. These findings should be considered as hypotheses to be tested in future large quantitative studies. Meanwhile, they are sufficient to suggest that providers provide anticipatory guidance on disclosure strategy planning to affected individuals.

Acknowledgements:

This research was supported by the Intramural Research Program of the National Human Genome Research Institute, National Institutes of Health.

This work was conducted to fulfill a degree requirement or as part of training.

Financial disclosures: This research was supported by the Intramural Research Program of the National Human Genome Research Institute, National Institutes of Health.

REFERENCES

Atkin K, & Ahmad WI (2001). Living a 'normal'life: young people coping with thalassaemia major or sickle cell disorder. Social science & medicine, 53(5), 615–626. doi:10.1016/S0277-9536(00)00364-6 [PubMed: 11478541]

Bairan A, Taylor GAJ, Blake BJ, Akers T, Sowell R, & Mendiola R (2007). A model of HIV disclosure: Disclosure and types of social relationships. Journal of the American Academy of Nurse Practitioners, 19(5), 242–250. doi:10.1111/j.1745-7599.2007.00221.x [PubMed: 17489957]

Beng-Huat C (1998a). Culture, multiracialism and national identity in Singapore (Chen K-H Ed.). New York, New York: Routledge.

Beng-Huat C (1998b). Culture, multiracialism and national identity in Singapore. Trajectories: inter-Asia cultural studies, 186–205.

Braun V, & Clarke V (2006). Using thematic analysis in psychology. Qualitative research in psychology, 3(2), 77–101. doi:10.1191/1478088706qp063oa

Charmaz K (1990). 'Discovering' chronic illness: Using grounded theory. Social science & medicine, 30(11), 1161–1172. doi:10.1016/0277-9536(90)90256-R [PubMed: 2360052]

Charmaz K (2000). Experiencing chronic illness (Albrecht GL, Fitzpatrisk R, & Scrimshaw SC Eds.). London, England: Sage Publications.

Chattopadhyay S (2006). 'Rakter dosh'—corrupting blood: The challenges of preventing thalassemia in Bengal, India. Social science & medicine, 63(10), 2661–2673. doi:10.1016/j.socscimed.2006.06.031 [PubMed: 16901596]

Chaudoir SR, & Fisher JD (2010). The disclosure processes model: understanding disclosure decision making and postdisclosure outcomes among people living with a concealable stigmatized identity. Psychological Bulletin, 136(2), 236–256. doi:10.1037/a0018193 [PubMed: 20192562]

Cunningham MJ (2008). Update on thalassemia: clinical care and complications. Pediatric Clinics of North America, 55(2), 447–460. [PubMed: 18381095]

Derlega VJ, Maduro RS, Janda LH, Chen IA, & Goodman BM (2018). What motivates individuals with sickle cell disease to talk with others about their illness? Reasons for and against sickle cell disease disclosure. Journal of Health Psychology, 23(1), 103–113. doi: 10.1177/1359105316649786 [PubMed: 27235142]

Fucharoen S, & Weatherall DJ (2016a). Progress toward the control and management of the thalassemias. Hematology/oncology Clinics, 30(2), 359–371. doi:10.1016/j.hoc.2015.12.001 [PubMed: 27040959]

Fucharoen S, & Weatherall DJ (2016b). Progress Toward the Control and Management of the Thalassemias. Hematology/oncology clinics of North America, 30(2), 359–371. [PubMed: 27040959]

Fucharoen S, & Winichagoon P (1992). Thalassemia in Southeast Asia: problems and strategy for prevention and control. The Southeast Asian journal of tropical medicine and public health, 23(4), 647–655. [PubMed: 1298071]

Glaser BG (1965). The constant comparative method of qualitative analysis. Social problems, 12(4), 436–445. doi:10.2307/798843

Goffman E (1963). Stigma: Notes on a spoiled identity. New York, New York: Simon & Schuster.

Hatzenbuehler ML, Phelan JC, & Link BG (2013). Stigma as a fundamental cause of population health inequalities. American journal of public health, 103(5), 813–821. doi:10.2105/AJPH.2012.301069 [PubMed: 23488505]

Joachim G, & Acorn S (2000). Stigma of visible and invisible chronic conditions. Journal of Advanced Nursing, 32(1), 243–248. doi:10.1046/j.1365-2648.2000.01466.x [PubMed: 10886457]

Johnson S, Kass NE, & Natowicz M (2005). Disclosure of personal medical information: Differences among parents and affected adults for genetic and nongenetic conditions. Genet Test, 9(3), 269–280. doi:10.1089/gte.2005.9.269 [PubMed: 16225407]

Kang E, Rapkin BD, Remien RH, Mellins CA, & Oh A (2005). Multiple dimensions of HIV stigma and psychological distress among Asians and Pacific Islanders living with HIV ollness. AIDS and Behavior, 9(2), 145–154. doi:10.1007/s10461-005-3896-9 [PubMed: 15933834]

Kass NE, Hull SC, Natowicz MR, Faden RR, Plantinga L, Gostin LO, & Slutsman J (2004). Medical privacy and the disclosure of personal medical information: the beliefs and experiences of those with genetic and other clinical conditions. American Journal of Medical Genetics Part A, 128(3), 261–270. doi:10.1002/ajmg.a.30057

Modi AC, Quittner AL, & Boyle MP (2010). Assessing disease disclosure in adults with cystic fibrosis: the Adult Data for Understanding Lifestyle and Transitions (ADULT) survey Disclosure of disease in adults with cystic fibrosis. BMC pulmonary medicine, 10(1), 46–53. doi: 10.1186/1471-2466-10-46 [PubMed: 20831811]

National Library Board Singapore. Singapore Infopedia: Shared Values. Retrieved from http://eresources.nlb.gov.sg/infopedia/articles/SIP_542_2004-12-18.html

Olivieri NF, & Brittenham GM (2013a). Management of the thalassemias. Cold Spring Harbor perspectives in medicine, 3(6), a011601. [PubMed: 23545573]

Olivieri NF, & Brittenham GM (2013b). Management of the Thalassemias. Cold Spring Harbor perspectives in medicine, 3(6), a011767. [PubMed: 23732853]

Pouraboli B, Abedi H, Abbaszadeh A, & Kazemi M (2014). Disease concealment: Experiences of thalassemia patients and their caregivers. Medical-Surgical Nursing Journal, 3(3), 121–128.

Quinn DM, & Chaudoir SR (2009). Living with a concealable stigmatized identity: The impact of anticipated stigma, centrality, salience, and cultural stigma on psychological distress and health. Journal of Personality and Social Psychology, 97(4), 634–651. doi:10.1037/a0015815 [PubMed: 19785483]

Quinn DM, & Earnshaw VA (2013). Concealable stigmatized identities and psychological well-being. Social and personality psychology compass, 7(1), 40–51. doi:10.1111/spc3.12005 [PubMed: 23730326]

Ratip S, Skuse D, Porter J, Wonke B, Yardumian A, & Modell B (1995). Psychosocial and clinical burden of thalassaemia intermedia and its implications for prenatal diagnosis. Archives of disease in childhood, 72(5), 408–412. doi:10.1136/adc.72.5.408 [PubMed: 7618906]

Roy T, & Chatterjee SC (2007). The experiences of adolescents with thalassemia in West Bengal, India. Qualitative Health Research, 17(1), 85–93. doi:10.1177/1049732306296400 [PubMed: 17170246]

Sandelowski M, Lambe C, & Barroso J (2004). Stigma in HIV-Positive women. Journal of Nursing Scholarship, 36(2), 122–128. doi:10.1111/j.1547-5069.2004.04024.x [PubMed: 15227758]

Shahraki-vahed A, Firouzkouhi M, Abdollahimohammad A, & Ghalgaie J (2017). Lived experiences of Iranian parents of beta-thalassemia children. Journal of multidisciplinary healthcare, 10, 243–251. doi:10.2147/JMDH.S132848 [PubMed: 28721064]

Thio L. a. (2010). Soft constitutional law in nonliberal Asian constitutional democracies. International journal of constitutional law, 8(4), 766–799. doi:10.1093/icon/mor001

Weatherall D (2010). Thalassemia as a global health problem: Recent progress toward its control in the developing countries. Annals of the New York Academy of Sciences, 1202(1), 17–23. doi:10.1111/j.1749-6632.2010.05546.x [PubMed: 20712767]

Wong LP, George E, & Tan J-AMA (2011). Public perceptions and attitudes toward thalassaemia: Influencing factors in a multi-racial population. BMC public health, 11(1), 193–201. doi:10.1186/1471-2458-11-193 [PubMed: 21447191]

Table 1:

Participant Characteristics: Individuals with Thalassemia Major

	Total (N=25*)
Age	
Under 21	14
Over 21	11
Gender	
Female	11
Male	14
Ethnic Group	
Chinese	12
Indian	3
Malay	6
Other	4
Education	
Primary or less	9
Secondary	5
College or more	11
Employment	
Not employed	14
Employed	11
Relationship Status	
Single	19
Dating	4
Married	2
Chelation therapy	
L1 only	4
Desferal only	8
L1 and Desferal	13

*As the focus of the study is on individuals with thalassemia, the table outlines only the characteristics of the affected individuals discussed in the interviews (not the parents).