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Feasibility of preconception screening for thalassaemia in Indonesia: exploring the opinion of Javanese mothers

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Background. Thalassaemia has become a major public health issue in Indonesia. It has been estimated that up to 10% of the population carries a gene associated with beta-thalassaemia. Currently, there is no formal recommendation for thalassaemia screening. This study aimed to explore awareness of thalassaemia, and to explore attitudes regarding carrier testing among Javanese mothers.

Methods. A quantitative questionnaire, designed using constructs of the Theory of Planned Behaviour, was applied cross-sectionally.

Results. Out of 191 mothers who were invited, 180 agreed to participate (RR = 94%), of whom 74 had a child affected with thalassaemia. Both attitudes towards receiving information about thalassaemia, and attitudes towards carrier testing were very positive. Awareness of thalassaemia was poor. Mothers, both those with and without an affected child, had barely heard of thalassaemia, nor of carrier testing. However, all mothers, including those with an affected child expressed high levels of interest in carrier testing. Respondents did not perceive that they had any control over carrier testing, and feared stigmatization and being discriminated against if their carrier status was identified. Attitudes towards carrier testing explained 23% of future reproductive intentions, in addition to perceived stigmatization, education level and 'mother's age' ($R^2 = 0.44$; $p = 0.001$).

Conclusion. Responding mothers expressed high levels of interest in receiving information on both thalassaemia and carrier testing. The less educated and the more deprived they were, the keener they were to receive this information. Overall, awareness of thalassaemia was low. Even mothers with affected children seemed unaware of the inheritance pattern and the recurrent risk of having an affected child in a subsequent pregnancy, showing the need for genetic counselling in Indonesia. It is therefore recommended not only to raise awareness about thalassaemia, but to improve the education of healthcare professionals as well.

Keywords: thalassaemia; carrier screening; Javanese mothers; genetic awareness; Theory of Planned Behaviour (TpB); awareness; Indonesia

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Introduction

Haemoglobin disorders, such as sickle cell disease and thalassaemia, are severe autosomal recessively inherited blood disorders predominantly found in people originating from countries where malaria is or was prevalent (Lakeman *et al.* 2009). Thalassaemia is common in Indonesia, where there have been 13 mutations of beta-thalassaemia identified, of which the HbE, IVS-nt5, and Cd 35 mutations are the most prevalent (Setianingsih *et al.* 1998). The number of individuals suffering from this severe, life-shortening disorder is increasing. Indonesia has a population of around 200 million people. However, reported prevalence of thalassaemia varies greatly, due to poor registration, problems with diagnosis, lack of diagnostic laboratories and low levels of awareness (Timan *et al.* 2002). Carrier rates are therefore best estimates only and estimates vary from 5% (Wahidiyat and Wahidiyat 2006) to between 6 and 10% (Timan *et al.* 2002). From these figures, we can in turn estimate the number of haemoglobinopathy carriers in Indonesia at 12–20 million people, with some 4000–10,000 children born each year who are affected with a severe form of thalassaemia. From the point of view of the general population, affected individuals do not appear to have a family history of the disease, due to its recessive inheritance pattern, and the parents may be unaware of being a thalassaemia carrier. Carriers of beta-thalassaemia are usually unaffected themselves, but they may show mild symptoms of anaemia. Carrier couples face a risk of one in four in each pregnancy of giving birth to an affected child.

Both a carrier test, and a diagnostic test for haemoglobinopathies were developed decades ago (WHO 2006a). The World Health Organization (WHO) has recommended developing services which integrate treatment, carrier detection and genetic counselling for haemoglobinopathies (WHO 2006b, Modell and Darlison 2008). Prevention programmes have been applied in many countries, e.g. Cyprus, the Islamic Republic of Iran, and Bahrain, in which premarital screening as standard practice was effectively implemented in society, dramatically decreasing the number of children born affected with haemoglobinopathies (Dhamcharee *et al.* 2001, Samavat and Modell 2004, Wahidiyat and Wahidiyat 2006, WHO 2006a,b, Modell and Darlison 2008).

Though not expensive, and available, neither carrier nor diagnostic tests for haemoglobinopathies are commonly used in Indonesia (Pramoongjago *et al.* 1999). Furthermore, when children are diagnosed with thalassaemia, usually the parents do not receive information on the one in four risk they face of having additional affected children in each subsequent pregnancy. Until now, the focus of the government has been on infectious diseases and on malnutrition, and genetic and other screening programmes have not been highly prioritized (Wahidiyat and Wahidiyat 2006). Moreover, individuals affected majorly with beta-thalassaemia are not well integrated into society. Many affected children stay in their houses, and die before they reach the age of ten. Many parents are not able to afford the costs of treatment due to their low socio-economic status. Although blood transfusions are offered free at the hospital for those having health insurance, e.g., JAMKESMAS (insurance for the poor, but with only limited availability) or ASKES (insurance for those who work as civil servants), parents are still charged for the supportive drugs necessary. Moreover, patients undergoing a blood transfusion have to stay hospitalized for two or three days, during which one or both parents may not be able to work, meaning

loss of income. Blood transfusions are also offered by the Red Cross, but even in this case the parents have to pay a small amount of money, even when they are officially declared indigent. In such instances they are asked to pay IDR 80,000 (around US\$8), with the remaining costs covered by the Indonesian Red Cross. However, The Red Cross does not provide the additional iron-chelation drugs necessary as the second line of treatment, drugs that prevent patients from iron overload due to the large number of blood transfusions they receive.

In other Asian countries with high levels of thalassaemia, the population has demonstrated low levels of awareness and knowledge of thalassaemia (see, for example, Ahmed *et al.* 2002, on the situation in Pakistan). Therefore, even though thalassaemia is prevalent in Indonesia, at levels comparable to malaria, it was felt possible that the Indonesian population too may be relatively unaware of this disorder. As a starting point for planning any future public health programmes on haemoglobinopathies in Indonesia, it is important to explore the current state of awareness about thalassaemia within the Indonesian population. Further, it is important to explore possible screening possibilities (e.g., preconceptual screening). Therefore, this study aimed to explore awareness about thalassaemia, and both attitudes towards receiving information about thalassaemia, and attitudes towards carrier testing for thalassaemia, amongst Javanese mothers. The Javanese are one of the largest ethnic groups in Indonesia. Although the Javanese tend to attribute life-events somewhat more often to 'fate' and 'karma' compared with those from other Indonesian cultures, aspects of Javanese culture, beliefs and practice concerning family matters are not dissimilar to the Indonesian population overall. The study is aimed at mothers in particular, since mothers generally are regarded as the main caregivers for children, and this gendered approach to the research reflects the Javanese culture where women are responsible for the upbringing of children (Zevalkink and Riksen-Walraven 2001, Albert *et al.* 2005). In a European context, men were found unlikely to participate in a study on genetic testing in children in the Netherlands (Plass *et al.* 2010). Furthermore, in a North American context, women in the USA were seen as primarily responsible for decision-making regarding testing of children (Martens 2002). The sense that reproductive decision-making around genetics will be gendered seems likely to be even more applicable in a patriarchal society such as Indonesia.

Methods

Design

A cross-sectional study design was employed, using a quantitative structured questionnaire, making use of the constructs of the Theory of Planned Behaviour (Ajzen 1991). This theory has proven to be a good predictor of genetic testing behaviour in other contexts (Frost *et al.* 2001, Lakeman *et al.* 2008). It explains behaviour based on behavioural intentions, which are explained by attitude towards the behaviour, perceived control over the behaviour and social norms concerning the behaviour (in other words, your perception of the ideas of significant others concerning your behaviour).

Participants and procedure

From May to September 2009 adult Javanese mothers, i.e., aged over 20, both with and without children affected with thalassaemia, were invited to take part in the study. To be eligible for inclusion in the study, they had to be able to read 'Bahasa Indonesia', the official Indonesian language. Mothers with an affected child were recruited at the waiting rooms for thalassaemia patients at either the paediatric ward of Dr. Kariadi Hospital Semarang, or at the Red Cross branch in Semarang. Mothers without affected children were selected on the basis of stratification from various regions in Central Java Province, e.g., Semarang, Kendal, and Salatiga, since the mothers visiting the hospital originated from various regions as well. Both Semarang and Salatiga are urban areas, whereas Kendal is rural. The interviewer (CW) called at houses within various neighbourhoods (attempting to recruit from poor, rich, and middle class areas). In Indonesia it is not clear how complete or reliable existing registers, such as municipal archives, may be, nor is it possible to send postal invitations and/or questionnaires. If eligible for inclusion in the study, research purposes were explained to potential respondents, including the fact that the study would guarantee their confidentiality. The questionnaires were handed to the respondents personally immediately after they had agreed to participate. In order to enable the respondents to fill out the questionnaire, a leaflet was handed together with the questionnaire, explaining thalassaemia and thalassaemia carrier status. After having filled out the questionnaire, the respondents handed it directly back to the researcher. All materials were solely available in the Indonesian language. The research protocol was approved by the medical ethical review board of the Medical Faculty of the Diponegoro University, Semarang, Indonesia.

Measures

A quantitative structured questionnaire was developed, making use of the constructs of the Theory of Planned Behaviour (TpB). Accordingly, attitudes towards receiving information about thalassaemia, attitudes towards carrier testing for thalassaemia, social norms concerning carrier testing for thalassaemia and perceived behavioural control over carrier testing for thalassaemia were measured. In addition, attitudes towards receiving information about thalassaemia, awareness of thalassaemia and perceived stigmatization and discrimination due to carrier status were measured. At the end of the questionnaire demographic variables were asked: educational background (high: at least a completed bachelor level at university; average: completed senior high school; or low: completed elementary school at highest); occupation (blue collar: labourer; white collar: office worker; or housewife), age, marital status (married or widow) and residence (urban or rural).

Attitudes towards carrier testing for thalassaemia

Attitudes towards carrier testing for thalassaemia was measured through the statement: 'I feel that for me personally having myself tested for carrier status of hereditary blood disease (thalassaemia) would be...', by a semantic differential, using six word-pairs (good vs. bad, frightening vs. reassuring, important vs. unimportant, annoying vs. pleasant, worthless vs. valuable and wise vs. nonsense),

measuring each word-pair on a 5-point Likert scale. A score of 5 indicated a positive attitude, a score of 1 indicated a negative attitude. Items were re-coded if necessary. Cronbach's alpha was 0.8. All items were loaded on one factor using 'oblimin rotation' for factor analysis.

Social norms concerning carrier testing for thalassaemia

Social influence was measured by five items, e.g., 'I think my partner/family/neighbours/friends or colleagues/GP think(s) that I should have myself tested for carrier status of thalassaemia (disagree-agree)', on a 5-point Likert scale. A score of 5 indicated experiencing strong influence from significant others regarding carrier testing for thalassaemia, a score of 1 indicated experiencing weak influence from significant others to perform the test. Cronbach's alpha was 0.9. All items were loaded on one factor using 'oblimin rotation' for factor analysis.

Perceived behavioural control over carrier testing for thalassaemia

Perceived behavioural control was measured by four items: 'I could not afford carrier testing if I would like to' (disagree-agree); 'I am not capable of participating in carrier testing if this would cost much time' (disagree-agree). 'I could not afford carrier testing even if I would like to have myself tested' (disagree-agree); 'I am not able to participate in testing if this means that blood has to be taken from me' (disagree-agree). A score of 5 indicated a high feeling of control over carrier testing. A score of 1 indicated a low feeling control over carrier testing. Items were recoded if necessary. Cronbach's alpha was 0.5, which is rather modest, but comparable to what is found elsewhere in the literature (Beale and Manstead 1991). All items loaded on one factor, using 'oblimin rotation' for factor analysis.

Attitude towards receiving information about thalassaemia

Attitude towards receiving information about thalassaemia was measured through the statement: 'I feel that receiving information about the hereditary blood disease (thalassaemia) for me personally is...' by a semantic-differential, using six word-pairs (good vs. bad; frightening vs. reassuring; important vs. unimportant; valuable vs. useless; offensive vs. encouraging; confusing vs. understandable) on a 5-point Likert scale. A score of 5 indicated a positive attitude, a score of 1 indicated a negative attitude. Items were recoded if necessary. Cronbach's alpha was 0.7. All items loaded on two factors, using 'oblimin rotation' for factor analysis.

Awareness of thalassaemia

Awareness was measured by three items: 'Had you ever heard of hereditary blood disease (thalassaemia) before?' (Yes/No/Maybe/Don't know), 'Had you ever heard of carrier status of thalassaemia before?' (Yes/No/Maybe/Don't know), 'Do you know someone who suffers from a hereditary blood disease (thalassaemia)?' (No/Maybe/Yes). Scores were summed up and divided by three. 'Yes' for an answer received 2 points, 'Maybe' 1, 'No' and 'Don't know' received zero points. A score of 0 indicated

no awareness about thalassaemia, a score of 2 indicated high awareness of thalassaemia.

Perceived stigmatization and/or discrimination

Stigmatization and discrimination were both measured through one item: 'I think people would look differently at me if I turned out to be a carrier of thalassaemia' (agree–disagree), measuring discrimination; 'I would not marry (would not have married) someone (my husband) if I knew (had known) that he is a carrier of thalassaemia' (agree–disagree), measuring stigmatization. A score of 1 indicated strong feelings of discrimination/stigmatization. A score of 5 indicated weak feeling of discrimination/stigmatization. The items were analyzed separately.

Future reproductive planning

Intention of having more children in the future was measured through one item: 'Do you and your partner, plan to have more children in the future?' (No/Yes/I do not have partner).

Data analysis

Data were analyzed using SPSS 15 for Windows. For each determinant which was measured by multiple questions, internal consistency was determined by calculating Cronbach's alpha, including the scale 'if item deleted' if the individual item is removed from the scale. Exploratory factor analysis was carried out for determinants with three or more questions by unweighted least squares and oblimin rotation. Although data from Likert scales are ordinal in level of measurement, in order to compare groups data here are reported as means. Differences between groups (demographics) were assessed using independent sample *t*-Test and one-way ANOVA. Stepwise hierarchical linear regression was assessed to find the predictors of mothers' plans to have more children in the future. Significance level was determined at 0.05.

Results

Study population

Of the 191 mothers who were invited to participate in the study, 180 agreed (RR = 94%). Amongst the 11 mothers who were not willing to participate, there were four whose child had just died. Of the participating mothers, 74 had a child affected with thalassaemia. The majority were married, and only nine were widows. Reported level of education was low in 8% of the mothers, 42% had an average education level and 50% were highly educated (bachelor degree). Compared with the entire Indonesian population, the study participants were higher educated, since 48% of the Indonesian population is lower educated; 18% has an average education level and 34% are highly educated (National Economic Survey, BPS-Statistics of Jawa Tengah Province, 2006). The majority of participating mothers (57%) were working in the 'white collar sector', e.g., office worker, 38% were housewives and 5% were employed in the blue collar sector (labourer). Of the respondents 67% lived in urban

areas (see Table 1). Mothers without an affected child had a significantly higher education level, classified as 'high', compared with mothers who had an affected child, whose education level would be classified 'average' [$t(162) = 4.1$; $p < 0.001$].

Attitudes

The mean score for both attitude towards information and attitude towards carrier testing on a 5-point Likert scale was high (respectively $M = 4.1$; $SD = 0.65$, and $M = 4.1$; $SD = 0.67$) showing that all participating mothers were both highly positive regarding receiving information about thalassaemia and regarding carrier testing for thalassaemia.

Differences between groups

Mothers with and without a child affected with thalassaemia

Mothers who had a child affected with thalassaemia were no more likely to report feeling positive regarding receiving information about thalassaemia ($M = 4.0$;

Table 1. Sociodemographic characteristics of the participants.

	Mothers of one or more children affected with thalassaemia <i>N</i> (%)	Mothers who do not have a child affected with thalassaemia <i>N</i> (%)
Age (years)		
20–30	14 (20)	26 (25)
31–40	30 (40)	54 (51)
41–50	22 (30)	18 (17)
> 50	8 (10)	8 (7)
Marital status		
Widow	6 (8)	3 (3)
Married	68 (92)	103 (97)
Education level		
Low (elementary)	27 (36)	21 (19)
Average (senior high)	36 (49)	49 (46)
High (university)	11 (15)	37 (35)
Occupation		
Housewife	30 (41)	39 (37)
Blue collar (labourer)	6 (8)	3 (3)
White collar (office worker)	38 (51)	64 (60)
Living area		
Rural	22 (30)	46 (43)
Urban	52 (70)	60 (57)
Number of children		
1–3	56 (76)	103 (97)
4–6	15 (20)	3 (3)
> 6	3 (4)	–
Total number	74 (40)	106 (60)

SD = 0.74; $N = 76$) compared with mothers who did not have an affected child ($M = 4.1$; SD = 0.57; $N = 106$) [$t(178) = -1.2$; $p = 0.2$] (see Table 2).

Education level

Lower educated mothers had a significantly more positive attitude ($M = 4.5$; SD = 0.66; $N = 14$; $p = 0.03$) towards receiving information about thalassaemia [$F(2, 177) = 3.48$; $p = 0.03$] compared with average ($M = 4.0$; SD = 0.66; $N = 76$) and higher educated mothers ($M = 4.0$; SD = 0.63; $N = 90$). However, lower educated mothers were no more likely to report feeling positive towards carrier testing ($M = 4.4$; SD = 0.67; $N = 14$) [$F(2, 177) = 1.203$; $p = 0.3$] than were average ($M = 4.0$; SD = 0.67; $N = 76$) or higher educated mothers ($M = 4.0$; SD = 0.66; $N = 90$).

Social norms

Mothers did not report experiencing social influence from significant others (i.e., spouses, family members, neighbours, friends/colleagues and the GP) in their social

Table 2. Mean scores of the variables between mothers with and without a child affected with thalassaemia.

	Mothers of a child affected with thalassaemia ($N = 74$)	Mothers who do not have a child affected with thalassaemia ($N = 106$)	<i>t</i> -test values comparing the two groups
	Mean (SD)	Mean (SD)	p [$t(178)$]
Attitude towards receiving information about thalassaemia	4.0 (0.76)	4.1 (0.57)	0.2 (1.3)
Attitude towards carrier testing for thalassaemia	4.0 (0.71)	4.1 (0.63)	0.2 (1.5)
Social influences regarding carrier testing	2.6 (1.6)	2.4 (1.7)	0.4 (-0.8)
Perceived behavioural control over carrier testing	3.2 (1.1)	2.9 (1.0)	0.05 (-0.2)
Awareness of thalassaemia	0.9 (0.41)	1.1 (0.49)	0.002 (3.1)
Perceived stigmatization	1.8 (1.5)	2.7 (1.7)	<0.001 (3.6)
Perceived discrimination	2.3 (1.5)	2.7 (1.8)	0.1 (1.6)

Note: Mean scores were based on a 5-point Likert scale, (1) indicating a negative/unfavourable score and (5) a positive/favourable score. Except for Awareness which was measured on a scale ranging from 0 (negative) to 2 (positive). SD, standard deviation.

circle with regard to carrier testing for thalassaemia ($M = 2.5$; $SD = 1.63$). Mothers with a child affected with thalassaemia reported experiencing significantly more social influence from their general practitioner (GP) compared with those who did not have a child affected with thalassaemia [$t(178) = 4.2$; $p < 0.001$] (see Table 3).

Self perceived control

Mothers were uncertain whether or not they were in control over carrier testing for thalassaemia ($M = 3.0$; $SD = 1.05$). Lack of money ($M = 3.3$) and time ($M = 3.5$) were the most important reported barriers. The differences between groups in terms of self-perceived control are reported further below.

With and without affected child

Mothers who had a child affected with thalassaemia perceived significantly more control over carrier testing compared with those who did not have a child affected with thalassaemia [$t(178) = 2.01$; $p = 0.05$]. The mean score of mothers who have an affected child indicates that they tend towards perceiving themselves as having some control over carrier testing ($M = 3.2$; $SD = 0.95$; $N = 74$), whereas the mean score of the mothers who do not have a child affected with thalassaemia indicates that they perceive carrier testing for thalassaemia as somewhat beyond their control ($M = 2.9$; $SD = 0.96$; $N = 106$) (see Table 2).

Education level

Higher educated mothers perceived significantly less control over carrier testing ($M = 2.7$; $SD = 0.96$; $N = 90$) [$F(2, 177) = 5.6$; $p = 0.004$] compared with average ($M = 3.1$; $SD = 0.88$; $N = 76$) and lower educated mothers ($M = 3.1$; $SD = 0.93$; $N = 14$).

Table 3. Mean scores on the social norms scale of mothers with and mothers without child affected with thalassaemia.

	Mothers with a child affected with thalassaemia ($N = 74$)	Mothers without a child affected with thalassaemia ($N = 106$)	<i>t</i> -test values comparing the two groups
	Mean (SD)	Mean (SD)	p [$t(178)$]
Spouse ^a	3.0 (1.7)	2.9 (1.9)	0.8 (−0.2)
Family members	2.7 (1.9)	2.7 (1.8)	0.8 (−0.2)
Neighbours	2.1 (2.0)	1.9 (1.7)	0.6 (−0.5)
Friends/colleagues	2.2 (2.0)	2.2 (1.8)	0.8 (−0.2)
GP	3.8 (1.5)	2.6 (2.0)	<0.001 (−4.2)

Note: Mean scores were based on a 5-point Likert scale. A score of 5 indicated experiencing strong influence from significant others; a score of 1 indicated experiencing weak influence from significant others.

^a $N = 171$; $df = 169$.

Discrimination/stigmatization

Mothers expressed anticipated feelings of stigmatization towards thalassaemia carriers and fearing discrimination should they turn out to be a carrier of thalassaemia ($M = 2.5$; $SD = 1.76$). Overall responses tended towards the view that they would choose not to marry a carrier partner ($M = 2.3$; $SD = 1.66$).

Difference between mothers with and without affected child

Mothers who had a child affected with thalassaemia expressed significantly stronger feelings of anticipated stigmatization ($M = 1.8$; $SD = 1.5$) [$t(178) = 3.6$; $p < 0.001$], compared with mothers who did not have a child affected with thalassaemia ($M = 2.7$; $SD = 1.7$) (see Table 2).

Awareness

In general, awareness of thalassaemia was poor ($M = 0.9$ on the total awareness scale, ranging from 0 to 2; $SD = 0.9$; $N = 180$). Thirty-five percent of the mothers stated having heard of thalassaemia before (22% 'maybe'; 43% 'no'), whereas only 18% stated having heard of carrier testing for thalassaemia before (27% 'maybe'; 55% 'no'). The vast majority of mothers who did not have a child affected with thalassaemia responded that they may know a person with thalassaemia, but they were not sure (77% 'maybe'; 9% 'no'; 14% 'yes'). All mothers with an affected child answered in the affirmative to this question (see Table 4).

Difference between mothers with and without child affected with thalassaemia

A striking difference occurred when comparing mothers with and without an affected child. Mothers without an affected child were significantly more likely to state that they had previously heard of thalassaemia [$M = 1.1$ and $M = 0.9$; $t(178) = 7.3$;

Table 4. Mean scores on awareness of thalassaemia of mothers with and mothers without a child that is affected with thalassaemia.

	Mothers with a child affected with thalassaemia ($N = 74$)	Mothers without child affected with thalassaemia ($N = 106$)	<i>t</i> -test values comparing the two groups
	Mean (SD)	Mean (SD)	<i>p</i> [$t(178)$]
Ever heard of thalassaemia before	0.4 (0.8)	1.2 (0.8)	<0.001 (7.1)
Ever heard of carrier status of thalassaemia before	0.2 (0.6)	0.9 (0.7)	<0.001 (7.0)
Knowing someone that is affected with thalassaemia	2.0 (0.0)	1.1 (0.5)	<0.001 (-17.1)

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Note: Mean scores were based on a 3-point scale: (0) 'no'; (1) 'maybe'; (2) 'yes'.

$p < 0.001$], and of the carrier status for thalassaemia [$M = 0.9$ and $M = 0.4$; $t(178) = 7.0$; $p < 0.001$], compared with mothers who had an affected child. Mothers who had a child with thalassaemia were the least likely to state that they had previously been aware of thalassaemia or of the carrier status for thalassaemia (see Table 2).

Explaining future reproductive planning

Hierarchical linear regression was performed stepwise with attitude, social norm and perceived behavioural control in the first step, awareness, stigmatization and discrimination in the second step, and age, education level and living area in the third. Attitude towards carrier testing was found to be a strong predictor of future reproductive plans, explaining 23% of the variance in the first step (multivariate $\beta = -0.20$; $p = 0.005$) while stigmatization increased the explained variance by another 8% ($\beta = -0.18$, $p = 0.003$), in the second step, and education level ($\beta = -0.25$; $p = 0.001$), and 'mothers' age' ($\beta = -0.24$; $p = 0.001$) increased the explained variance with another 6 and 7% respectively in the third step. In looking at mothers with and without affected children separately, it appeared that 28% of the future reproductive planning of mothers with an affected child was explained by attitude towards carrier testing ($\beta = -0.12$; $p = 0.02$) in the first step, while another 23% was explained by 'mothers' age' ($\beta = 0.25$; $p < 0.001$) in the third step. On the other hand 21% of the future reproductive plans of mothers without affected children was explained by their perception of control over carrier testing ($\beta = -0.12$; $p = 0.02$) in the first step, in addition of 'mothers' age' ($\beta = 0.21$; $p = 0.001$) in the third step.

Differences between widowed and married mothers

Although the number of widowed mothers ($N = 9$) was far too small for statistical analyses, there were differences between this group and married mothers (see Figure 1). Widowed mothers perceived less influence of their social circle, were more in control over carrier testing and were found to be more aware of thalassaemia. However, they showed more feelings of fearing stigmatising, once carrier status had been confirmed.

Discussion

Awareness of thalassaemia amongst Indonesian (Javanese) mothers was found to be poor. Mothers whose child was affected with thalassaemia were found to be unaware of the fact that they themselves were carriers. Thus, they seemed to be unfamiliar with the inheritance pattern and recurrence risk of this autosomal recessive, severe, life-shortening disorder. However, all mothers showed high interest in receiving information about thalassaemia, and were positive about carrier testing for thalassaemia.

The majority of mothers, who were, for the greater part, well educated, had seldom heard of thalassaemia before. Surprisingly, it was the mothers who did not have a child affected with thalassaemia who said that they had heard of thalassaemia before, whereas mothers who had an affected child stated they had not. A possible explanation for this may be that mothers without affected children were more highly

Demographics			
<ul style="list-style-type: none"> • Widowed mothers were significantly older than married mothers $t(178)=-3.4$; $p=[.001]$ • Widowed mothers significantly more often had a 'white collar' job $t(178)=2.7$; $p=[.024]$ 			
Mean scores on variable outcomes			
	Widowed mothers (N= 9)	Married mothers (N= 171)	T-test values comparing the two groups
	Mean (SD)	Mean (SD)	p [t(178)]
• Attitude towards receiving information about thalassaemia	4.3 (0.64)	4.1 (0.66)	0.4 (0.8)
• Attitude towards carrier testing for thalassaemia	4.2 (0.8)	4.1 (0.66)	0.6 (0.6)
• Social influences regarding carrier testing for thalassaemia [†]	1.7 (2.6)	2.5 (1.6)	0.2 (-1.2)
• Perceived behavioural control over carrier testing for thalassaemia [†]	3.6 (1.2)	3.0 (1.0)	0.07 (1.8)
• Awareness of thalassaemia	1.0 (0.52)	1.0 (0.46)	1
• Perceived stigmatization [‡]	1.9 (1.4)	2.3 (1.6)	0.5 (-7)
• Perceived discrimination [‡]	2.9 (1.6)	2.5 (1.7)	0.5 (-7)

Figure 1. Differences between the small group of widowed mothers and the large group of married mothers.

Note: [†]Large differences though NS (not significant). Except for awareness, that was measured on a scale ranging from 0 (negative) to 2 (positive), all variables were measured on a 5-point Likert scale; a score of 1 indicating a negative/unfavorable score, a score of 5 indicating a positive/favorable score. SD, standard deviation.

educated compared with the other group, and therefore may be more aware of hereditary blood disorders and possible carrier status. However, another explanation may be that mothers with an affected child may have interpreted this question wrongly, answering in terms of whether they had heard of the word 'thalassaemia' before, since they already knew of the manifestations of the disease through their child with thalassaemia. The participating mothers were found to be uncertain about the extent to which they perceived they had control over carrier testing for thalassaemia. However, mothers who had an affected child tended to feel somewhat more in control. This may be due to the fact that they visited the hospital more often, and therefore had better access to healthcare. The main barriers for lacking control over carrier testing were limited financial resources, and the idea that testing would be time consuming. Most people in Indonesia do not have a health insurance, so they would have to pay for carrier testing. Less educated mothers, and mothers living in rural areas were even more in favour of receiving information about thalassaemia

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compared with more highly educated mothers, and mothers living in urban areas. A possible explanation may be that less educated mothers, and those living in rural areas may be more deprived in terms of receiving health information in general. Furthermore, less educated mothers may have less difficulty in accepting the probability that this may be 'something that could happen to them' compared with more highly educated mothers. Lower educated mothers would therefore appreciate receiving information more highly. They may also tend to be more receptive of information than more highly educated women, possibly because they perceive that they have less to lose in the way of social standing than the mothers who are graduates. Moreover, more highly educated mothers may be more aware of possible negative side-effects of receiving this information and would therefore ignore or deny this information, perhaps because they perceive the dangers of being discriminated against, or becoming stigmatized. However, no significant differences between more highly and less well educated women in terms of stigmatization and discrimination were found in this study. This is perhaps surprising, given that in other studies it has been found that more highly educated women were more likely to regard genetic testing (for example for cancer and schizophrenia) as relevant for both themselves and their families (Kreuter 1999, Bottorff *et al.* 2002, Prentice *et al.* 2005). Furthermore, more highly educated women may be more aware of the current limited availability of genetic testing within Indonesian society, and this may explain the finding in this study that they perceived less control over carrier testing for thalassaemia compared with less educated women.

Of particular concern for policy development in Indonesia was the finding that mothers who had a child affected with thalassaemia were usually unaware of their increased risk of having another affected child in the future. In Indonesia, the doctor only informs the parent(s) about the treatment of the child at the time of diagnosis. Anecdotally, it appears that some parents are being told by the doctor not to have more children in the future, without further explanation. This may explain why even mothers of affected children were found to be unaware of being carriers themselves and thus remained unaware of the inheritance pattern of their child's disorder. As beta-thalassaemia follows a recessive inheritance pattern, there often is no visible family history of thalassaemia upon which the parents can draw to assess their risk independently of testing. This finding emphasizes the need not only to educate the lay public, but to (better) educate Indonesian healthcare providers concerning the communication of genetic information and its implications.

The more positive their attitude about receiving information, the more mothers favoured participation in carrier screening for thalassaemia. In one sense this contrasts with the findings of Ahmed *et al.* (2002), who found that limited knowledge of thalassaemia influenced awareness, which in turn reduced willingness to participate in carrier testing for thalassaemia. In another sense it is consistent with Ahmed *et al.* (2002) as it confirms the relationship between knowledge, awareness and orientation to carrier testing, since in the Pakistan study carrier testing was regarded as unimportant, especially for those lacking a family history of thalassaemia. This arguably shows the importance of raising awareness in a society such as Indonesia where the whole population has an increased risk for being a carrier of a haemoglobin disorder.

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Furthermore, the current study shows that attitudes towards carrier testing were the strongest predictors of mothers' future reproductive intentions, in addition to

'mothers' age'. Thus it is at least possible that, once aware, mothers, especially the younger ones, may take information on this disorder and on the possibility of carrier status into account concerning their future reproductive plans. In contrast, a number of studies carried out in the Netherlands (Henneman *et al.* 2001, Poppelaars *et al.* 2004, Lakeman *et al.* 2009) found that not all people would take carrier status into account concerning their future family planning. Prenatal diagnosis would be opted for by 89%, abortion in case of an affected child by 68%, and refraining from (more) children by 27%. A lower proportion of minority ethnic groups in the Netherlands, compared with Dutch ethnic majority participants, would consider an abortion if the foetus was affected (Lakeman *et al.* 2008). In a study in Israel, Shiloh *et al.* (1995) found that carrier couples, once carefully counselled, adjusted their reproductive choices to their carrier status (Shiloh *et al.* 1995). The implementation of premarital programmes in some countries, for example, Cyprus, has led to a fall in the number of affected births. However, genetic information is sensitive to context and other studies have suggested that such contexts may mean that people may just want to know their carrier status, without linking consequences to this information (Ahmed *et al.* 2006).

Feelings of being stigmatized have been mentioned as an undesirable effect of genetic screening (Lakeman *et al.* 2008). Being a carrier might induce (feelings of) inequality, that is being treated 'differently', having fewer life chances compared with others, being considered a lesser being than others (having 'contagious' blood), and this is particularly the case for women (Burnes *et al.* 2008). In contrast to findings in studies carried out in Western societies, where members of the high risk population were found not to perceive feelings of discrimination or stigmatization should they prove to be a haemoglobinopathy carrier (Lakeman *et al.* 2008, Weinreich *et al.* 2009), the women in this study anticipated fearing both stigmatization and discrimination were they to turn out to be a haemoglobinopathy carrier. A possible explanation may be that in the Indonesian society a hereditary disorder, e.g., thalassaemia, is considered a 'curse' within the family. This is especially the case in the Javanese culture where having children is a cultural imperative as a way to guarantee the continuity of the family (Albert *et al.* 2005) with children having a responsibility to honour their family and to keep their family's name going in society. In such situations there is strong social pressure for parents to take care of and teach their children, and also provide their needs as they grow up. Meanwhile, having children is perceived as prestigious to the family. Besides adversely influencing the affected individual, a genetic disorder may therefore adversely influence the entire family dynamics (Meiser *et al.* 2001). Such wider family considerations are liable to influence the acceptance of hereditary disorders in society.

Although the group of widowed mothers was too small to be subject of statistical analysis, it was surprising to see that the widowed mothers felt more in control over testing for carrier status, perceiving less social pressure, and this may plausibly be due to the absence of a husband. They reported seeming much more in charge over their own lives, and decisions, compared with the married mothers. The possible relative freedom to act autonomously in relation to genetic information in the absence of a dominant male figure in a patriarchal society would merit further study.

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Limitations of the study

This study was carried out in Central Java, which is considered the heart of the Javanese culture, since the classical Javanese culture was established here. However, since the Javanese are the main ethnic group in Indonesia, and cultural differences between various ethnic groups in Indonesia are minor, the results of the current study may not only be applicable to the Javanese, but to the entire Indonesian population as well. Due to the Indonesian infrastructure it was not possible to randomly select mothers who did not have an affected child, and this limits the generalizability of the study. In order to make this group as comparable to the other group as possible, mothers were selected from various regions. Moreover, mothers with an affected child did not differ significantly on demographic variables from mothers without affected child other than on educational level. Furthermore, half of the respondents were highly educated. This is not representative for the entire Indonesian population where only about one third are highly educated. This may be due to the key inclusion criterion that they had to be able to read the official Indonesian language to such an extent that they were able to read the accompanying information leaflet and fill out the questionnaire. However, lower educated mothers very often are poor, and may not be able to supply the medical care needed to the child, due to the costs. These mothers may therefore be under-represented in the hospitals. Moreover, adult female literacy in Indonesia is 93% (UNICEF 2010). It could be argued that this relatively high level of literacy means that an inclusion criterion of being able to read 'Bahasa Indonesia' has not of itself greatly reduced the generalizability of the study. Only mothers participated in this study. Fathers and healthcare professionals did not. However, in exploring and raising awareness on thalassaemia in Indonesia it would be of the highest importance to take the opinion of fathers and healthcare providers concerning thalassaemia into account as well.

Key messages

Indonesian mothers were very interested in information on thalassaemia and carrier testing. Preconception screening for thalassaemia may therefore be feasible in the Indonesian society. Furthermore, information on their carrier status would reportedly influence future reproductive plans. This emphasises the need to educate healthcare professionals, improving their knowledge, and enabling them to give adequate counselling, including calculation of the recurrent risk of having an affected child. Furthermore, there is a need for patient-education programmes on thalassaemia into the Indonesian society, raising awareness, and decreasing possible discrimination or stigmatization. Further research is needed to ascertain how fathers' opinions should be taken into account, in order to avoid 'blaming the mothers'. Ideally, carrier screening for thalassaemia should be offered free of charge and be easily accessible, in order to overcome the perceived barriers to thalassaemia screening of lack of time and lack of money.

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