

BMJ Open Cross-sectional study on knowledge, attitude and practice towards thalassaemia among Indonesian youth

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ABSTRACT

Introduction Thalassaemia is an inherited blood disorder, for which definitive treatments remain largely inaccessible. The recommended approach to reduce the disease burden is by prevention through screening. Currently, the implementation of thalassaemia preventive measures is poorly regulated in Indonesia. Thalassaemia prevention and education are best targeted to the youth, but information on their awareness towards thalassaemia is limited. This study aims to investigate the knowledge, attitude and practice (KAP) towards thalassaemia among Indonesian youth.

Methods This observational study took place between January and May 2021. An online questionnaire was disseminated to Indonesian youth aged 15–24. Eligible respondents included carriers, unaffected individuals and individuals with unknown carrier status. The questionnaire comprised 28 questions to assess KAP. A cut-off of 75% was used to categorise participant's KAP into poor or negative and good or positive. Descriptive statistics, χ^2 test, logistic regression and Pearson correlation were performed for data analysis.

Results A total of 906 responses were gathered, and 878 were analysed. Most respondents had poor knowledge (62.1%), positive attitude (83.3%) and poor practice (54.4%) towards thalassaemia. The results implied that respondents had limited understanding regarding the types of thalassaemia and the difference between asymptomatic carriers and individuals without the thalassaemia trait. Many (82.6%) believed they were not carrying thalassaemia trait despite the fact that most (95.7%) never got tested. Age, education, gender, residence and family income were key factors that correlated with or predicted the youth's KAP towards thalassaemia. Older respondents and women were more likely to have good KAP.

Conclusion Thalassaemia screening targeted to the youth is urgently needed, and future interventions must consider sociodemographic factors that may affect how they perceive the disease. Social media appeals to the youth as an important source of information, but school, parents and health professionals should also be involved in delivering education about thalassaemia.

INTRODUCTION

Thalassaemia is an inherited blood disorder that affects the red blood cells, in which there is a genetic mutation causing the depletion

Strengths and limitations of this study

- First nationwide knowledge, attitude and practice study on Indonesian youth that provides baseline data for carriers, unaffected individuals and individuals with unknown carrier status.
- Incorporation of knowledge, attitude and practice questions that are specific to Indonesian cultural settings, which include perception of thalassaemia as a curse and impact of consanguinity on inheritance of thalassaemia.
- Online dissemination of the survey via social media allows us to reach a large number of Indonesian youth from all backgrounds who are located in all 34 provinces of Indonesia.
- The use of online self-administered questionnaire for data collection may discourage those with poor Internet connection, particularly in remote areas, from participating.
- Lack of background data on public perception towards thalassaemia in Indonesia means that some assumptions from expert opinions and educated guesses have to be made when developing the questionnaire.

or complete absence of alpha and/or beta-globin chains that serve as the backbone of haemoglobin.^{1 2} Indonesia, being one of the countries within the 'thalassaemia belt', was predicted to have 3.8% carriers of thalassaemia gene according to the Indonesian Ministry of Health in 2019.³ A twofold increase in the number of thalassaemia cases across the country was observed from 4896 cases in 2012 to 8761 cases in 2018.³ The latest data published in 2019 stated that there were around 10 500 patients with thalassaemia in Indonesia with 1500 new cases diagnosed every year.² Even worse, since there is currently no national thalassaemia registry in Indonesia, it is likely that there are a high number of undiagnosed cases in the country.²

Currently, definitive treatments for thalassaemia including bone marrow transplant and hematopoietic stem cell therapy are



still out of reach in Indonesia, especially among those within the low socioeconomic status. Therefore, thalassaemia prevention through population screening is the recommended approach in Indonesia.² In comparison to treatment for thalassaemia, which costs US\$ 28 500 per person annually, screening for thalassaemia only costs US\$29 per person.⁴ Cost–benefit analysis done in Northern Israel showed that each prevented case could financially support the prevention programme for up to 4.6 years.⁵ The success of thalassaemia preventive measures to reduce the number of new cases has been reported in many countries, for example, Cyprus, Greece and Italy.⁶ In Cyprus, a national prevention programme was first introduced in 1983 which involved mandatory premarital thalassaemia carrier screening as well as prenatal and antenatal diagnosis. Couples with known thalassaemia trait are also given genetic counselling along with the choice of abortion. As a result, the number of babies born with thalassaemia in Cyprus dropped from around 30 per year before 1983 to 0 in 2000.⁷ Thus, through thalassaemia screening, the number of incidence and economic burden of thalassaemia can be reduced.

However, there are several key issues regarding the implementation of thalassaemia preventive measures in Indonesia that need to be addressed. Currently, only Jakarta, the capital city of Indonesia, has mandated its citizens to undergo premarital thalassaemia screening which comprises mean corpuscular volume and mean corpuscular haemoglobin examination.² Thalassaemia screening is still not included as a national mandatory programme and has not been uniformly implemented in other regions in Indonesia.⁸ Furthermore, it is uncertain whether couples who have been screened would be willing to cancel their marriage over the test result. As discovered by a study in Iran, half of all couples still proceeded to marriage even though they were carriers of β -thalassaemia.⁹ Couples who are thalassaemia carriers still have the option of antenatal diagnosis to prevent their baby born with the disease in other countries, but this is not a feasible option in Indonesia as the current law does not allow abortion following antenatal diagnosis.²

A potential solution would be to perform a thalassaemia screening programme targeted to school-aged children and university students. Being aware of their carrier status from an early age can encourage the youth to be mindful in choosing marriage partners or seeking genetic counselling prior to having children. For thalassaemia education and prevention programmes for the youth to be successful, their level of knowledge, attitude and practice (KAP) towards thalassaemia must first be evaluated. To our knowledge, this is the first and largest nationwide study that investigates the KAP of Indonesian youth towards thalassaemia, and we hope the findings can help inform future policies and prevention programmes.

METHODS

Study design

The study design used was observational, cross-sectional using a newly developed online questionnaire. Eligibility criteria for the study's participants were as follows: (1) within the age range of 15–24 years; (2) willing to provide informed consent after reading and understanding the research background and aims; and (3) residents of Indonesia. Thalassaemia major patients were excluded from this study. The study was conducted from March 2021 until August 2021, which ended following completion of the manuscript. This study used the Strengthening the Reporting of Observational Studies in Epidemiology cross-sectional reporting guideline.¹⁰

Questionnaire design

The questionnaire for this study was self-developed by the research team following literature review and consultation with a professor of paediatric haematology involved in thalassaemia management for many years as well as a physician with expertise in public health research. The questionnaire was initially constructed in English then translated into Indonesian which is the spoken language of the target population. It comprised of 8 parts in the following order: consent and information letter, questions related to sociodemographic characteristics of the participants, source of information for thalassaemia, personal and family history of thalassaemia, questions to assess knowledge of thalassaemia (12 questions), questions to assess attitude towards thalassaemia (10 questions), questions to assess practice towards thalassaemia (6 questions) and future plans for thalassaemia screening. In total, there were 28 KAP questions for the participants to complete. A copy of the study questionnaire can be found in online supplemental material 1.

The questionnaire underwent validation and reliability test by a statistician to ensure representable interpretation of the result. Validity testing for questions in this study was done by calculating the Pearson and Point-Biserial correlation (for multiple-choice questions) between the score of each item and the total score of all items on certain variables or dimensions. If the *p* value is <0.05 (with a significance level of $=0.05$), then the item is said to be valid; if the *p* value is >0.05 , then the item is invalid. Cronbach's alpha was used to perform reliability testing. Cronbach's alpha value has a range between 0 and 1. A value of 0 indicates that the item is not reliable, and a higher Cronbach's alpha indicates a reliable question (do note that if the value is >0.95 , it can indicate redundancy). In this study, a question is deemed as reliable when the Cronbach alpha value was ≥ 0.6 . The methods and calculations above determined that all KAP questions used in this study were valid and reliable.

After finalisation of the question items, the questionnaire was then uploaded to Google Form and made online. Pilot testing of the questionnaire was conducted among 50 randomly chosen eligible participants to evaluate its readability and difficulty. Modifications of several

questions' wording to make them easier to understand were then made based on the feedback. The participants who were asked to complete the questionnaire for pilot testing were not included in the whole study sample.

Data collection

The minimum participants enrolled were calculated through the formula of $N = (Za^2 \times p \times q) / L^2$. Za represent the level of confidence, p represents the probability of an individual having good KAP towards thalassaemia, q represents the probability of an individual not having good KAP towards thalassaemia ($1-p$) and L represents the margin of error. The values of Za and L were 1.96 (95% CI) and 0.05, respectively. Meanwhile, the values of p and q were both 0.5. The value of p was set to 0.5 since previous data on the KAP of Indonesian youth towards thalassaemia were not available. Following the calculation, a minimum of 400 participants were required for the study.

This online questionnaire was then distributed through social media using convenience and snowball sampling methods. The social media platforms included Facebook, Twitter, Instagram, WhatsApp and LINE as they are among the most commonly used applications by Indonesian users. There was no incentive involved as participation was fully voluntary. Participants were asked to give their consent on the first page of the questionnaire, and they could contact a representative of the research team in case they had any questions. The use of Google Form as the survey platform enabled us to track the demographic distribution of the respondents in real time. Every week, a virtual meeting with the team was held to evaluate the progress and number of responses. If responses from a particular province or demographic group were lacking in comparison to responses from other groups, the research team would encourage participants from that specific group to distribute the questionnaire link with their network. For example, if there was a limited number of responses from middle school and high school students, the research team would enlist the help of more teachers and schools. All members of the research team were also responsible for distributing the questionnaire link to their social network. The responses from the online questionnaire were automatically compiled in Google Form and then downloaded as Microsoft Excel files. The file was encrypted with a password that could only be accessed by the research team. Data from all participants were safeguarded from any potential privacy breach and only used for this research purpose.

Outcome measures

For the KAP assessment, the widely adopted Bloom's cut-off points were as follow: 80%–100% (good KAP), 60%–79% (moderate KAP) and less than 60% (poor KAP).^{11–13} In this study, a modified Bloom's cut-off value of 75% was used to categorise participants' KAP into two levels: poor/negative and good/positive. This

cut-off value was also based on previously published KAP studies.^{14–17}

For the knowledge component, the questions were in the form of three-option multiple-choice questions. A correct answer was given 1 point while an incorrect or 'I do not know' answer was given 0 point. The number of correct answers by each participant was automatically scored by Google Form based on our answer key. A knowledge score from 0 to 8 indicated 'poor' knowledge, whereas a knowledge score from 9 to 12 indicated 'good' knowledge.

For the attitude component, a total score based on the responses to statements on a four-point Likert scale (1=strongly disagree; 2=disagree; 3=agree; 4=strongly agree) was computed. The number of points given corresponded to the chosen response from the four-point Likert scale, for example, a response of 'agree' was given 3 points. An attitude score from 10 to 29 indicated a 'negative' attitude, whereas an attitude score from 30 to 40 indicated a 'positive' attitude.

For the practice component, a total score based on the responses to statements on a four-point Likert scale (1=strongly disagree; 2=disagree; 3=agree; 4=strongly agree) was computed. The number of points given corresponded to the chosen response from the four-point Likert scale, for example, a response of agree was given 3 points. A practice score from 6 to 17 indicated poor practice, whereas a score from 18 to 24 indicated good practice.

Statistical analysis

Data were analysed using IBM SPSS V.24. Descriptive statistics were performed to summarise the sample sociodemographic characteristics and evaluate the overall distribution in KAP scores. Responses towards willingness to undergo thalassaemia screening and source of information for thalassaemia were also described. χ^2 test was used to assess the association between participants' sociodemographic characteristics and their KAP levels, followed by logistic regression to determine the relative importance of selected sociodemographic variables in predicting participants' KAP levels. Only variables that showed statistically significant results at least in two domains (knowledge, attitude or practice) in χ^2 test were chosen to analyse further using logistic regression. Statistical significance level used for inclusion in the logistic regression model was $p < 0.05$. Pearson correlation was used to assess whether KAP scores were associated with one another. The significance level was set at $p < 0.05$.

Patient and public involvement

As part of our commitment to patient and public involvement in our study, thalassaemia carriers as well as the general public were involved in the conception of the study, planning of the study protocol, finalisation of the questionnaire content and data collection. In particular, their feedback and comments helped

shape this study's research questions and future directions. During the data collection period, any eligible participant was encouraged to provide any criticism or suggestion by contacting one of the research team representatives through text or email. Most importantly, all participants were seen as partners rather than only subjects in this research as it was hoped that they would be empowered to help raise thalassaemia awareness among Indonesian youth with the help of our study findings.

RESULTS

A total of 906 respondents consented to and completed the online questionnaire that was distributed in January 2021. After the exclusion of 28 duplicate responses, 878 respondents were included in this study. The demographic characteristics of the respondents are displayed in [table 1](#), whereas their overall responses towards background questions related to thalassaemia are summarised in [table 2](#).

Most respondents had poor knowledge (62.1%), positive attitude (83.3%) and poor practice (54.4%) as seen in online supplemental material 2. In terms of knowledge, the majority (74%) knew the cause of thalassaemia, the difference between thalassaemia and leukaemia (78.6%), and the fact that thalassaemia is not contagious (83.9%). While the majority also knew that the trait can be passed from parents to offspring (85.1%), around a third of all respondents (36.3%) did not know that consanguinity increases the risk of passing thalassaemia trait. Regarding thalassaemia carriers, 46.7% knew that carriers do not always show the same symptoms as thalassaemia major patients, but only 33% knew that carriers do not always experience anaemia. Around half of all respondents (59.1%) recognised the appropriate treatments for patients with thalassaemia, and 38.3% knew that gene therapy and bone marrow transplantation can cure patients. More than half (52.4%) knew that thalassaemia can be prevented and can be detected from a blood test (80.8%), but 55.0% did not know that antenatal thalassaemia screening can be performed during pregnancy. Full details can be found in online supplemental material 3.

In terms of attitude, the majority either strongly disagreed (48.2%) or disagreed (34.4%) that they might be carrying the thalassaemia gene. Almost a third of all respondents (27.8%) either strongly disagreed or disagreed that couples who are carriers are still allowed to marry and reproduce as long as they have understood the risks involved. For other questions regarding attitude towards thalassaemia, most respondents adopted a positive attitude which can be either 'strongly agree' or agree. Full details can be found in online supplemental material 4.

In terms of practice, around a third of all respondents disagreed (30.8%) with the idea that they would undergo a thalassaemia screening test as soon as possible and that

Table 1 Distribution of respondents according to socio-demographic variables (N=878)

Variables	N (%)
Age group	
Adolescent (15–18)	256 (29.2)
Young adult (19–24)	622 (70.8)
Gender	
Male	261 (29.7)
Female	617 (70.3)
Education	
Never went to school	5 (0.6)
Elementary school or equivalent	0 (0.0)
Middle school or equivalent	21 (2.4)
High school or equivalent	226 (25.7)
Diploma or equivalent	74 (8.4)
Bachelor's degree	530 (60.4)
Postgraduate	22 (2.5)
Province	
Java Island	676 (77.0)
Outside Java Island	202 (23.0)
Father's occupation	
Healthcare provider	51 (5.8)
Not a healthcare provider	742 (84.5)
Unemployed	85 (9.7)
Mother's occupation	
Healthcare provider	87 (9.9)
Not a healthcare provider	538 (61.3)
Unemployed	253 (28.8)
Father's education	
Never went to school	2 (0.2)
Elementary school or equivalent	31 (3.5)
Middle school or equivalent	33 (3.8)
High school or equivalent	239 (27.2)
Diploma or equivalent	80 (9.1)
Bachelor's degree	343 (39.1)
Postgraduate	150 (17.1)
Mother's education	
Never went to school	4 (0.5)
Elementary school or equivalent	43 (4.9)
Middle school or equivalent	37 (4.2)
High school or equivalent	252 (28.7)
Diploma or equivalent	123 (14.0)
Bachelor's degree	319 (36.3)
Postgraduate	100 (11.4)
Health coverage	
Personal	249 (28.4)
National health insurance (BPJS)	430 (49.0)

Continued

Table 1 Continued

Variables	N (%)
Private insurance	118 (13.4)
Employer health insurance	81 (9.2)
Family monthly income	
IDR <2 million	107 (12.2)
IDR 2–5 million	225 (25.6)
IDR 5–10 million	221 (25.2)
IDR >10 million	325 (37.0)

BPJS, Badan Penyelenggara Jaminan Sosial; IDR, Indonesian's Rupiah.

they would do so when it might cost them money and time (30.0%). More than half had not previously encouraged their relatives to undergo thalassaemia screening, although the majority (86.1%) either strongly agreed or agreed that they would do so in the future prior to marriage. Regarding personal choice in picking a marriage partner and willingness in seeking more information about thalassaemia, most respondents adopted a good practice. Full details can be found in online supplemental material 5.

For knowledge, the six significant contributing factors were age group, gender, education, residence, mother's occupation and family monthly income. For attitude, the four significant contributing factors were age group, gender, education and family monthly income. For practice, the four significant contributing factors were gender, residence, father's education and mother's education. Father's occupation and healthcare coverage were not significantly associated with KAP (table 3).

Female gender, living outside Java Island and a family monthly income of Indonesian's Rupiah (IDR) >10 million were significant predictors of having good knowledge. Female gender, a family monthly income of IDR 5–10 million and a family monthly income of IDR >10 million were significant predictors of having positive attitude. Female gender and living outside Java Island were significant predictors of having good practice. There were other trends seen in the table, but none of the other variables were statistically significant predictors of good knowledge, positive attitude and good practice (table 4).

Spearman's rank correlation coefficient revealed that there was a statistically significant moderate positive correlation between attitude–practice ($r_s=0.453$, $p<0.001$) and a weak positive correlation between knowledge–attitude ($r_s=0.418$, $p<0.001$) and knowledge–practice ($r_s=0.340$, $p<0.001$).

DISCUSSION

With 878 responses gathered from all 34 provinces in Indonesia, this study's findings revealed the awareness level as well as behavioural patterns among the youth population towards thalassaemia. Overall, the majority

Table 2 Distribution of responses towards questions about the source of information for thalassaemia, personal and family history, and future plans for thalassaemia screening test

Background information	N (%)
Have you previously heard of thalassaemia?	
Yes	654 (74.5)
No	224 (25.5)
If yes, from what information source(s) did you hear about it?	
Friend	131 (8.2)
Family	119 (7.5)
School	335 (21.0)
Physician or other healthcare providers	217 (13.6)
Patients with thalassaemia	81 (5.1)
Social media	357 (22.4)
Television	72 (4.5)
Online news website or newspaper	80 (5.0)
Seminar	99 (6.2)
Others	102 (6.4)
Which one of the following describes your current condition?	
Carrier for thalassaemia gene	34 (3.9)
Healthy (not affected by thalassaemia)	506 (57.6)
I do not know	338 (38.5)
Is any member of your direct family a thalassaemia major patient, carrier, or receiving routine blood transfusion?	
Yes	53 (6.0)
No	448 (51.0)
I do not know	377 (42.9)
Have you previously undergone screening for thalassaemia trait?*	
Yes	37 (4.3)
No	833 (95.7)
If you have never undergone screening for thalassaemia trait, are you planning to get yourself tested in the future?	
Yes	407 (48.6)
No	53 (6.3)
I do not know	377 (45.1)
When are you planning to undergo screening for thalassaemia trait?	
As soon as possible	144 (16.4)
Before marriage	580 (66.1)
After marriage	5 (0.6)
After having offspring	3 (0.3)
I am not planning to	146 (16.6)

*Eight respondents were excluded from this question due to incomplete answers.

of our respondents (74.5%) claimed they had heard of thalassaemia. This was relatively satisfactory when compared with findings from other thalassaemia-prevalent countries including Bangladesh (33%), Saudi Arabia (48%), Turkey (58%), Bahrain (65%), Malaysia (76.4%), Italy (85%) and Greece (95%).^{18–24}

Table 3 Association of knowledge, attitude, and practice towards thalassaemia with sociodemographic variables of the respondents

Variables	Knowledge			Attitude			Practice		
	Good N (%)	Poor N (%)	P value	Positive N (%)	Negative N (%)	P value	Good N (%)	Poor N (%)	P value
Age group									
Adolescent (15–18)	75 (29.3)	181 (70.7)	0.001*	194 (75.8)	62 (24.2)	<0.001*	110 (43.0)	146 (57.0)	0.323
Young adult (19–24)	258 (41.5)	364 (58.5)		537 (86.3)	85 (13.7)		290 (46.6)	332 (53.4)	
Gender									
Male	68 (26.1)	193 (73.9)	<0.001*	204 (78.2)	57 (21.8)	0.009*	104 (39.8)	157 (60.2)	0.027*
Female	265 (42.9)	352 (57.1)		527 (85.4)	90 (14.6)		296 (48.0)	321 (52.0)	
Education									
Lower education	69 (27.4)	183 (72.6)	<0.001*	187 (74.2)	65 (25.8)	<0.001*	116 (46.0)	136 (54.0)	0.858
Higher education	264 (42.2)	362 (57.8)		544 (86.9)	82 (13.1)		284 (45.4)	342 (54.6)	
Residence									
Java Island	239 (35.4)	437 (64.6)	0.004*	555 (82.1)	121 (17.9)	0.093	286 (42.3)	390 (57.7)	<0.001*
Outside Java Island	94 (46.5)	108 (53.5)		176 (87.1)	26 (12.9)		114 (56.4)	88 (43.6)	
Healthcare coverage									
Personal	92 (36.9)	157 (63.1)	0.524	208 (83.5)	41 (16.5)	0.940	101 (40.6)	148 (59.4)	0.275
National health insurance (BPJS)	160 (37.2)	270 (62.8)		360 (83.7)	70 (16.3)		207 (48.1)	223 (51.9)	
Private insurance	52 (44.1)	66 (55.9)		96 (81.4)	22 (18.6)		56 (47.5)	62 (52.5)	
Parents' office/employer health insurance	29 (35.8)	52 (64.2)		67 (82.7)	14 (17.3)		36 (44.4)	45 (55.6)	
Father's occupation									
Healthcare provider	24 (47.1)	27 (52.9)	0.367	46 (90.2)	5 (9.8)	0.214	18 (35.3)	33 (64.7)	0.277
Not a healthcare provider	276 (37.2)	466 (62.8)		611 (82.3)	131 (17.7)		345 (46.5)	397 (53.5)	
Unemployed	33 (38.8)	52 (61.2)		74 (87.1)	11 (12.9)		37 (43.5)	48 (56.5)	
Mother's occupation									
Healthcare provider	46 (52.9)	41 (47.1)	0.007*	79 (90.8)	8 (9.2)	0.132	45 (51.7)	42 (48.3)	0.475
Not a healthcare provider	190 (35.3)	348 (64.7)		445 (82.7)	93 (17.3)		242 (45.0)	296 (55.0)	
Unemployed	97 (38.3)	156 (61.7)		207 (81.8)	46 (18.2)		113 (44.7)	140 (55.3)	
Father's education									
Lower education	115 (37.7)	190 (62.3)	0.921	246 (80.7)	59 (19.3)	0.132	160 (52.5)	145 (47.5)	0.003*
Higher education	218 (38.0)	355 (62.0)		485 (84.6)	88 (15.4)		240 (41.9)	333 (58.1)	
Mother's education									
Lower education	129 (38.4)	207 (61.6)	0.823	283 (84.2)	53 (15.8)	0.545	170 (50.6)	166 (49.4)	0.018*
Higher education	204 (37.6)	338 (62.4)		448 (82.7)	94 (17.3)		230 (42.4)	312 (57.6)	
Family monthly income									
IDR <2 million	33 (30.8)	74 (69.2)	0.003*	79 (73.8)	28 (26.2)	<0.001*	55 (51.4)	52 (48.6)	0.629
IDR 2–5 million	71 (31.6)	154 (68.4)		176 (78.2)	49 (21.8)		102 (45.3)	123 (54.7)	
IDR 5–10 million	81 (36.7)	140 (63.3)		197 (89.1)	24 (10.9)		99 (44.8)	122 (55.2)	
IDR >10 million	148 (45.5)	177 (54.5)		279 (85.8)	46 (14.2)		144 (44.3)	181 (55.7)	

Lower education level included those who never went to school until those who went to high school or equivalent. Higher education level included those who studied at Diploma, Bachelor's, and/or Postgraduate level.

* $P < 0.05$. χ^2 test was used.

BPJS, Badan Penyelenggara Jaminan Sosial; IDR, Indonesian's Rupiah.

Knowledge

Despite many claiming that they had heard about thalassaemia, most respondents (62.1%) still had poor knowledge about the disease. The number of KAP studies

on thalassaemia in Indonesia is still very limited, but this study's findings were consistent with those from a previous study among high school students in Pontianak City, West Kalimantan, Indonesia.²⁵ Even among medical

Table 4 Logistic regression analysis for selected sociodemographic variables associated with good knowledge, positive attitude and good practice towards thalassaemia

Variables	Knowledge			Attitude			Practice		
	OR (95% CI)	aOR (95% CI)	P value	OR (95% CI)	aOR (95% CI)	P value	OR (95% CI)	aOR (95% CI)	P value
Age group									
Adolescent (15–18)	1	1	1	1	1	1	1	1	1
Young adult (19–24)	1.711 (1.251 to 2.339)	1.298 (0.837 to 2.014)	0.243	2.019 (1.400 to 2.912)	1.445 (0.863 to 2.419)	0.162	1.159 (0.865 to 1.555)	1.414 (0.934 to 2.143)	0.102
Gender									
Male	1	1	1	1	1	1	1	1	1
Female	2.137 (1.553 to 2.940)	1.925 (1.388 to 2.669)	<0.001*	1.636 (1.131 to 2.366)	1.475 (1.006 to 2.164)	0.047*	1.392 (1.038 to 1.868)	1.373 (1.016 to 1.855)	0.039*
Education									
Lower education (never went to school until high school or equivalent)	1	1	1	1	1	1	1	1	1
Higher education (Diploma, Bachelor's, and Postgraduate)	1.934 (1.406 to 2.661)	1.345 (0.850 to 2.126)	0.205	2.306 (1.600 to 3.323)	1.454 (0.857 to 2.467)	0.165	0.974 (0.726 to 1.306)	0.724 (0.471 to 1.114)	0.142
Residence									
Java Island	1	1	1	1	1	1	1	1	1
Outside Java Island	1.591 (1.158 to 2.188)	1.600 (1.146 to 2.235)	0.006*	1.476 (0.935 to 2.329)	1.479 (0.920 to 2.378)	0.106	1.767 (1.286 to 2.426)	1.723 (1.245 to 2.384)	0.001*
Family monthly income									
IDR <2 million	1	1	1	1	1	1	1	0.816 (0.508 to 1.310)	0.400
IDR 2–5 million	1.034 (0.629 to 1.700)	0.935 (0.557 to 1.567)	0.797	1.273 (0.746 to 2.173)	1.122 (0.646 to 1.949)	0.683	0.784 (0.494 to 1.243)	0.844 (0.521 to 1.367)	0.490
IDR 5–10 million	1.297 (0.792 to 2.124)	1.186 (0.704 to 1.996)	0.521	2.909 (1.590 to 5.325)	2.576 (1.373 to 4.833)	0.003*	0.767 (0.483 to 1.218)	0.835 (0.526 to 1.325)	0.444
IDR >10 million	1.875 (1.178 to 2.984)	1.697 (1.034 to 2.785)	0.036*	2.150 (1.263 to 3.660)	1.817 (1.033 to 3.196)	0.038*	0.752 (0.486 to 1.165)	0.816 (0.508 to 1.310)	0.400

OR is unadjusted OR from univariate logistic regression analysis.

aOR is adjusted OR or OR that has been adjusted for all other variables from multivariate logistic regression analysis.

P value shown in the table is from the adjusted logistic regression model.

*P<0.05.

IDR, Indonesian's Rupiah.

students in Indonesia, who were expected to have significantly more knowledge of the disease, Dewanto *et al* and Tursinawati *et al* found that only 21.1% and 42.7% of their subjects, respectively, had good knowledge.^{26 27} From our survey, most respondents already knew the basic definition of thalassaemia, which is a blood disorder caused by abnormalities in red blood cells that has an autosomal recessive inheritance pattern. However, public awareness of the dangers of consanguineous marriages must be increased because consanguinity increases the risk of a child born with thalassaemia. Consanguineous marriages cause higher expression of recessive alleles, leading to clustering of mutations in a particular community. It was reported that the rate of consanguineous marriages was high in rural, impoverished and poorly educated communities.²⁸ There is a lack of information on the prevalence of consanguinity in Indonesia, but it was suggested to be relatively high in Asian countries with large populations.²⁹

A large proportion of the respondents still did not know much about thalassaemia carriers as well as the difference between carriers and thalassaemia major patients. This might be due to a poor understanding about the various types of thalassaemia, which is broadly divided into alpha and beta thalassaemia. Beta thalassaemia can be further classified based on its genotype, symptoms (major, intermediate, minor/trait) and the need for transfusion.³⁰ Since carriers of the thalassaemia trait usually do not exhibit any symptoms, they may never seek medical attention and live completely normal lives. If those who carry the trait are unaware of their condition, it is not surprising that other people are just as unaware. Even among Javanese mothers who had thalassaemic children, Widayanti *et al* found that they too did not know that they were carriers.³¹ The threat here is the assumption made by the public that being asymptomatic equals to not having the trait or disease. A Malaysian study found that the main reason behind parents' refusal of thalassaemia screening was the belief that the absence of thalassaemia in their family members meant their children were not at risk.³² Overall, these findings emphasise the importance of teaching youth about thalassaemia mode of inheritance, carriers, signs and symptoms, treatment, prevention and screening.

Attitude

Regarding attitude, most respondents (83.3%) showed positive attitude towards thalassaemia. The majority believed that everyone must undergo thalassaemia screening long before marriage and supported the incorporation of thalassaemia screening into a mandatory National Programme. Moreover, most of them agreed that receiving further information about thalassaemia would benefit them and their family. However, 82.6% of respondents either strongly rejected or rejected the idea that they might unknowingly be thalassaemia carriers. This might result from their poor knowledge regarding the difference between thalassaemia major patients, asymptomatic carriers and individuals without the thalassaemia

trait. Since attitude towards carrier testing was found to be a significant predictor of mothers' future reproductive plans, the youth must be made aware of the importance of carrier testing because they will soon become parents themselves.³¹ Failure to do so will lead to an increase in the rate of thalassaemic children being born every year. A study based in Malaysia found that despite the majority of its respondents showed positive attitude towards thalassaemia screening, only 13.6% of married participants actually got tested.²² Health education and genetic counselling are especially important in Indonesia as its current law does not support antenatal testing and subsequent abortion, which are offered as alternative family planning options in some other countries.³¹

Practice

Although the respondents showed a positive attitude towards thalassaemia, more than half (54.4%) had poor practice towards the disease. Almost all respondents had not undergone screening for thalassaemia trait, yet more than half responded that they were confident that they and their families did not carry the trait. The youth was equally divided in opinion; half of them planned to get tested while the other half was still unsure. Even among those who would be willing to get tested, the respondents preferred to do it before marriage rather than as soon as possible. As antenatal testing and abortion are not a feasible choice in Indonesia, finding out their carrier status just before marriage may complicate things for young couples due to strong social pressure to marry and fear of facing stigmatisation in Indonesia. Time and money were once again claimed to be important considerations for getting tested.³¹ This hopefully urges the national government to realise the importance of making thalassaemia screening a National Program alongside formal recommendation and subsidised coverage. At present, there is minimal regulation for thalassaemia screening in Indonesia as it is not mandatory. Some health centres offer thalassaemia screening tests, but these are individually organised. On the other hand, neighbouring countries such as Malaysia had already implemented thalassaemia screening programme at school since 2016.³²

Association between KAP and sociodemographic factors

Good KAP was more prevalent among young adults (age 19–24) than adolescents (age 15–18), as older youth with higher education generally had higher literacy to better understand thalassaemia from books and social media.³³ Women were more likely to have good KAP, which was consistent with findings from studies in Iran^{33 34} and Bahrain.³⁵ However, this was inconsistent with findings from a study in India, where men had better knowledge and a more positive attitude towards thalassaemia.³⁶ The reason behind our findings might be correlated with a previous study that revealed females were more likely than men to share health information on social media with their network.³⁷ Furthermore, since women tend to

be seen as the one carrying the major reproductive responsibility of conceiving and raising a child, they may feel an increased pressure to learn and receive information about hereditary diseases like thalassaemia. In contrast to women in Western societies, Javanese women in a study by Widayanti *et al* feared of stigmatisation due to their thalassaemia carrier status.³¹ Hence, having a thalassaemic offspring may affect women more than men, which can explain why women would be more likely to have better KAP. Good knowledge and good practice were also more likely to be found among those residing outside Java Island. This was a surprising result as most previous studies reported that the urban population had better KAP towards thalassaemia than the rural population. Therefore, we initially expected to find a similar trend as Java Island has been Indonesia's most developed island throughout decades. Nevertheless, this finding corroborated the claim made by Widayanti *et al*, which discovered that mothers living in rural areas were more open to receiving information about thalassaemia.³¹ This might be because those in rural areas were already more deprived of health information and thus more eager to learn about the disease.³¹

The χ^2 test also revealed that there were more respondents with higher education and high family monthly income who had good knowledge and positive attitude. However, practice was not significantly associated with either respondent's education or family monthly income. These findings indicate that having adequate knowledge and attitude do not automatically translate into having good practice. Thalassaemia education programmes targeted to the youth must therefore aim to improve not only knowledge but also attitude and practice. The programmes should also consider the diverse background of the Indonesian population, in which socioeconomic status is often not equally distributed across regions. The occupation and education level of parents also appeared to influence youth's KAP towards thalassaemia, but it was difficult to make any meaningful conclusion since the extent of this effect was not measured. Nevertheless, since family values are highly upheld in Indonesian culture, thalassaemia prevention and education programmes must incorporate parents' views and cooperation to be effective. Having parents as partners in delivering health information will allow it to be well-received by the youth, as the decisions of Indonesian youth are often influenced by values taught by their parents.

Social media as an important source of information and education tool

It is encouraging to learn that most respondents stated that they had heard of thalassaemia before. However, there is a subtle yet crucial difference between hearing about the disease and understanding it correctly. This is especially important since a significant portion of the respondents who had heard of thalassaemia claimed to receive information about it

from social media. The percentage of youth who use the Internet for seeking health information has vastly increased throughout the years, but it was suggested that only a small percentage believed that social media was a useful and reliable source.^{37 38} A major concern for youth seeking health information online is the difficulty to assess the reliability and validity of the information on each website they look at. Moreover, now with so many websites discussing similar health topics, they may also become overwhelmed and confused by information overload.^{38 39} Nevertheless, given that Indonesian youth is becoming increasingly reliant on the Internet, social media must be strongly considered as a powerful tool to deliver thalassaemia educational campaigns to this age group. The United Nations Population Fund Indonesia reported that 9 out of 10 Internet users in Indonesia are active on social media, compared with 7 out of 10 in the USA.⁴⁰ On the other hand, since health information from school is easier to be standardised and verified, education about thalassaemia and other genetic diseases must be further incorporated into the national curriculum. Parents and healthcare providers, especially those who interact closely with adolescents and young adults, must also be reminded of the importance of teaching the youth about thalassaemia prevention and screening from a young age.

Limitations

This study is not without limitations. Although this study was the largest of its kind, it might not be fully representative of the Indonesian youth population. The number of responses in some provinces was too small to be representative, so the results of this study cannot be generalised to all youth in Indonesia. Regarding gender distribution, there were significantly more women who participated in our study than men. This might lead to an overly optimistic prediction about Indonesian youth's KAP towards thalassaemia than what is actually happening in Indonesia right now, since women in our study were shown to be more likely to have good KAP. In addition, since the survey was disseminated online, some youth might be excluded due to the inability to access the Internet. Latest available figures from 2010 showed that 54.5% of male youth and 50.2% of female youth in Indonesian households had access to Internet or mobile phones. However, while 64.3% of youth living in urban areas had access to Internet or mobile phones, only 37.4% of youth living in rural areas had access.⁴¹ These numbers were expected to rise exponentially, but most recent data were not available online. The cross-sectional nature of this study also limited its ability to thoroughly interpret the association between sociodemographic factors and KAP towards thalassaemia. Future research is therefore needed to investigate further how certain sociodemographic factors influence public perception towards thalassaemia.

CONCLUSION

Considering the high economic and medical burden of thalassaemia in Indonesia, there is an urgent need to develop an effective prevention programme and incorporate it within the national scope. Education and prevention are best targeted to the youth population, with the hope that being aware of their carrier status can help guide their decisions when entering the stage of marriage and parenthood. Despite having a positive attitude towards thalassaemia, Indonesian youth generally had poor knowledge and poor practice towards it. The youth's KAP also appeared to be influenced by certain sociodemographic factors, so future intervention must take these into account. The four most important tools for delivering education about thalassaemia to the youth include social media, school, parents and health professionals.

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