

Knowledge towards Thalassemia and Willingness to Screen among Students in Public Senior High School 3 Bandung

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Abstract

Background: Thalassemia carrier screening is a major preventive measure potentially influenced by the level of knowledge, particularly in adolescents. Therefore, this study aimed to analyze the effect of health education on knowledge of thalassemia in adolescents and its association with their willingness to do thalassemia screening.

Methods: A cross-sectional study was conducted using data regarding knowledge of thalassemia before and after health education sessions from 229 students at Public Senior High-School 3 Bandung. All participants attended a one-day health education in July 2019. A questionnaire was filled in to measure their knowledge regarding thalassemia before and after the session, including knowledge on etiology and definition, risk of disease, clinical manifestations, treatment, complication, prognosis, and disease prevention. Only data with complete questionnaire responses were included. These responses were scored quantitatively and analyzed for their association with participants' willingness to screen.

Results: Participants were knowledgeable concerning thalassemia before the health education session (median, range: 60.0, 25.0-90.0), and knowledge was increased significantly after the education session (median, range: 80.0, 35.0-100.0) with an increased median difference=19.99 (p-value <0.001). Although there was no significant association between the overall post-test score on participants' willingness to screen (p-value >0.05), the willingness was slightly associated with improved knowledge regarding the risk of disease (OR: 1.02; 95%CI: 1.00-1.03; p-value <0.005).

Conclusion: Health education regarding thalassemia significantly increases general knowledge of thalassemia. However, improving knowledge is not significant in influencing adolescents' motivation to take the screening tests.

Keywords: Health education, knowledge, screening, thalassemia

Introduction

Thalassemia is an inherited abnormality of the globin chain influencing the structure and function of hemoglobin. Since it is an autosomal recessive disease, individual carrying only one thalassemia mutation, named thalassemia carrier does not show any symptoms. This condition lead to difficult clinical detection. Nevertheless, when a thalassemia carrier is married to a thalassemia carrier partner, there is a 25% possibility of having an affected child. Ineffective bone marrow erythropoiesis and excessive hemolysis in thalassemia patients cause an increased need for blood

transfusions.¹ A previous systematic review identified 64–89% of major beta-thalassemia patients requiring regular blood transfusions every 2 and 4 weeks.² Also, infants and children affected by thalassemia will have physical development disturbance, thus increasing the burden of disease. Therefore, in countries with a high prevalence of thalassemia carriers, preventive strategy is essential to reduce the risk of thalassemia in the next generation. Among population worldwide, 1.5% are indicated as thalassemia carriers. Mostly, they live in the “thalassemia belt”, extending along the shores of the Mediterranean and throughout the Arabian Peninsula, Turkey,

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Iran, India, and Southeast Asia, including Indonesia.^{3,4}

Several preventive measures have been proposed in Indonesia, such as genetic counseling and carrier screening, as the best options for facing thalassemia.^{5,6} However, these approaches have been relatively unsuccessful, particularly in adolescents, due to a lack of willingness to screen in the target population.⁷ A previous study in West Java showed that only about 50% of adolescents were willing to screen for thalassemia voluntarily, while 9.3% were suspected of having minor beta-thalassemia.⁷ Possible reasons for this barrier are a lack of knowledge, that adolescents have lower knowledge on thalassemia than adults and a lack of understanding of the concept of thalassemia carriers.^{8,9} Another study in West Java showed that many high-school students were unaware of the disease inheritance and the importance of genetic screening tests before providing educational modules.¹⁰ Furthermore, it is well known that health education can improve the knowledge and perspectives of individuals regarding a situation or disease, particularly asymptomatic condition such as thalassemia carrier.¹¹ A previous study in Malaysia has indicated an increase in the rate of genetic screening tests immediately after implementation of health education.¹² However, another study in Turkey showed that the implementation of hemoglobinopathies screening test significantly decreased the prevalence of newborns with thalassemia.¹³ This finding indicates the urgency to undertake screening tests early in life before they decide to get married and plan a pregnancy. Currently, there is still limited study that can explain in-depth how improving knowledge through health education can increase willingness to participate in genetic screening tests for adolescents in Indonesia.

Therefore, this study aimed to analyze the effects of health education on knowledge on thalassemia in adolescents and its association with their willingness to screen. It is expected that with the improvement of knowledge through health education, adolescents would be more open to thalassemia screening tests.

Methods

This study was cross-sectional using secondary data from the Academic Leadership Grant (ALG) project at the Medical Genetic Working Group, Faculty of Medicine, Universitas Padjadjaran. This project involved students

from Public Senior High School (*Sekolah Menengah Atas Negeri, SMAN*) 3 Bandung, Indonesia. This high school was selected because the high school was recognized as a high rank public school with good information exposure. Final grade students, who were mostly adolescents, were invited to participate in a one-day health education session on thalassemia which was held in July 2019. Health education session was held in parallel in each class. It was started with a presentation related to thalassemia by a qualified counselor, using the same presentation slides for each class, and followed by an interactive discussion. This health education was part of the community service provided by the Faculty of Medicine, Universitas Padjadjaran. Before and after the session, participants were asked to fill in a questionnaire, developed by the Medical Genetic Working Group Universitas Padjadjaran, including experts in thalassemia, genetics, and public health. Questionnaire development has been described in the previous article.¹⁴ Data from all participants attending the health education session were included in this study, but data of participants who failed to complete both questionnaires were excluded from the analysis. With 80% power of the study, we calculated a minimum of 178 participants required to find statistical significance for 20% knowledge difference. Ethical clearance has been provided by the Ethical Committee of the Universitas Padjadjaran Bandung, with number 887/UN6.KEP/EC/2021.

The questionnaire was developed to measure participants' knowledge of thalassemia, particularly regarding the etiology and definition, risk of disease, clinical manifestation, treatment, complication, prognosis, and disease prevention. Each aspect of knowledge was measured by 2 to 5 questions and thus contained 20 True-or-False questions (Table 1). This questionnaire has been developed by experts in this field and was considered relevant to the presentation of the counselor. Each answer to this questionnaire was assessed. The correct answer was scored 1 and zero otherwise, and subsequently summarized based on the related aspects of knowledge. Finally, the scores were transformed to a scale of 0–100, according to the percentage of correct answers. The levels of knowledge were then quantified as an overall general knowledge and separately according to the above-mentioned aspects of knowledge. In addition, participants were also asked about their willingness to get

Table 1 Characteristics of Students from Public Senior High School 3 Bandung

Characteristics	(Median, Range)	n (%)
Age	17(14-18)	
Gender		
Male		87 (38.0)
Female		142 (62.0)
Experience in thalassemia education		
Never		212 (92.6)
Ever		2 (0.9)
Unclear		15 (6.6)
Family history of thalassemia		
Yes		1 (0.4)
No		228 (99.6)
Relationship with thalassemia family members		
No relatives with thalassemia		228 (99.6)
First-degree relatives		1 (0.4)

thalassemia screening after participating in a health education session, with “yes” or “no” answers.

The quantitative level of knowledge pre and post education session, which was then called the pre-test and post-test scores, was checked for the normality of the data distribution. To measure the effect of health education on participants’ knowledge, a comparative analysis using the Wilcoxon Signed Rank test between pre-test and post-test was performed. The difference in scores between pre-test and post-test was considered as knowledge improvement. Furthermore, the association of knowledge improvement and post-test score on participants’ willingness to screen was measured by performing the independent t-test and the Mann-Whitney U test, respectively. Five percent type I of error was adjusted in this study, and thus the p-value <0.05 was considered statistically significant.

Statistical analysis and data visualization were performed in R Version 4.0.

Results

Of 298 individuals who participated in the thalassemia education session, 229 had completed the pre-test and post-test. Most respondents were late adolescents with a median age of 17 years and had never gotten in thalassemia education or had family/relative with thalassemia. Only one respondent had a first-degree relative with a history of thalassemia. Details of these characteristics are summarized in Table 1.

Overall, participants were already knowing about thalassemia before the education session (median, range: 60.0, 25.0–90.0). After the education session, participants showed better knowledge about thalassemia (median difference of pre-test and post-test=19.99,

Table 2 The Score of Knowledge Related to Thalassemia Before and After Health Education sessions among students from Public Senior High-School Bandung

Aspects of Knowledge	Pre-test Score		Post-test Score		P-value
	Median	Min-Max	Median	Min-Max	
Etiology and definition	66.7	0-100	100.0	0-100	<0.001
Risk of disease	50.0	0-100	50.0	0-100	<0.001
Disease manifestation	66.7	0-100	100.0	0-100	<0.001
Treatment	50.0	0-100	50.0	0-100	0.18
Complication	100.0	0-100	100.0	0-100	<0.001
Prevention	80.0	20-100	100.0	0-100	<0.001
Overall knowledge	60.0	25-90	80.0	35-100	<0.001

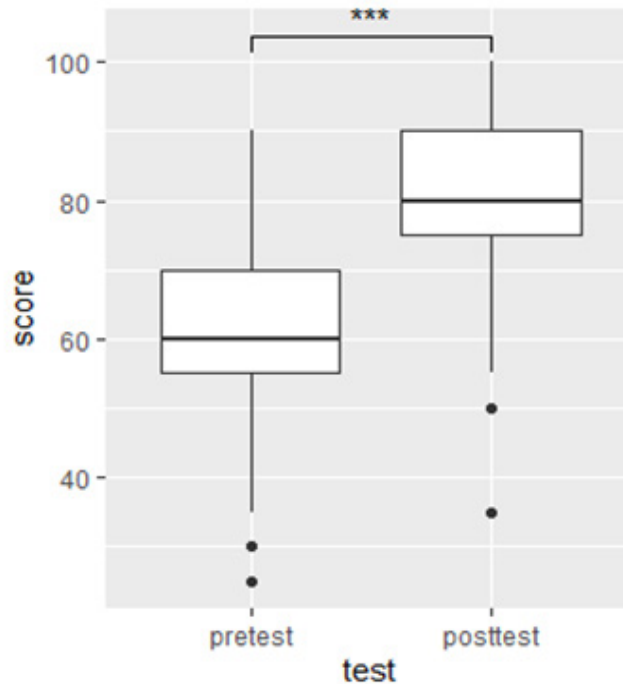


Figure 1 Different Test Scores Regarding Knowledge of Thalassemia Before and After Thalassemia Education Sessions

Note: In the box plots, the boundary of the box closest to zero indicates the 25th percentile, a black line within the box marks the median, and the boundary farthest from zero indicates the 75th percentile. Whiskers above and below the box indicate the 10th and 90th percentiles. Points above and below the whiskers indicate outliers outside the 10th and 90th percentiles. (*)/(**)/(***): statistically significant, NS= Not significant.

p-value <0.001) (Figure 1 and Table 2). The slightest median differences were seen in treatment and disease risk (Table 2), but the knowledge distribution of disease risk has shown the most prominent shift after attending the education session.

Although most respondents were willing to get a genetic screening test for thalassemia after attending thalassemia education (87.3%), the willingness to take the screening test did not depend on the participants' knowledge of thalassemia. The overall post-test score and the overall score improvement did not significantly influence respondents' willingness to screen (Figure 2). However, this study showed that respondents willing to get the screening test had a higher level of knowledge (median: 80 vs. 75, p-value= 0.107). Also, there was a slight association between the willingness to screen and improved knowledge about the risk of thalassemia (OR: 1.02; 95%CI: 1.00, 1.03; p-value < 0.005).

Discussion

In this study, the effects of health education on knowledge on thalassemia in adolescents and its association with their willingness to screen was measured. This study found that health education at school has significantly improved adolescents' knowledge of thalassemia, and thus, this activity could have a good general understanding of thalassemia. A remarkable improvement in knowledge was found in the aspect of the risk of disease. Although it was found that the respondents had a high willingness to screen, good knowledge about thalassemia and a significant improvement in knowledge were not associated with the willingness to test screening.

Several studies have shown significant effect of health education in improving knowledge and attitude towards inherited disease prevention, such as thalassemia.^{10,12} Similarly, the findings of the current study

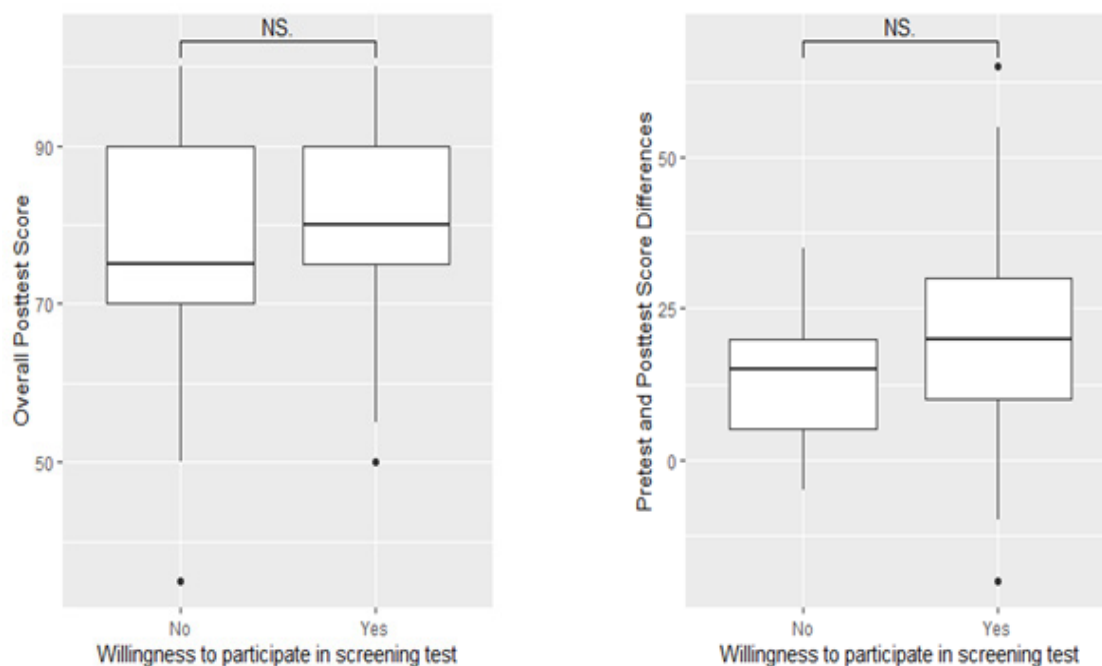


Figure 2 Association of Overall Post-test Scores on Respondents’ Willingness to Screen (A) and Association of Pre- and Post-test Difference Scores on Respondents’ Willingness to Screen (B)

Note: In the box plots, the boundary closest to zero indicates the 25th percentile, a black line within the box marks the mean, and the boundary farthest from zero indicates the 75th percentile. Whiskers above and below the box indicate the 10th and 90th percentiles. Points above and below the whiskers indicate outliers outside the 10th and 90th percentiles. (*)/(**)/(***)= statistically significant, NS= Not significant.

show an improvement in knowledge of thalassemia after providing health education to the respondents, especially about the risk of the disease. These findings agree with the previous studies showing the effectiveness of educational modules in improving the level of knowledge about thalassemia prevention.^{10,12} However, these previous studies showed that the level of knowledge was not retained in a long-term, indicating the need for a long term or sustainable availability of accessible resources to make the knowledge lasts longer.

This study identified that improved knowledge of thalassemia was accompanied by a high rate of willingness to perform screening tests. However, this study failed to identify a statistical association between knowledge improvement and motivation on screening tests. Although this study found a significant association between willingness to screen and risk of disease, the effect size was considered too small to influence willingness to screen. These results are inconsistent with previous studies, which indicate a lack of

knowledge and understanding of the personal concept of disease as a barrier to screening tests.^{9,11,15} This discrepancy is possibly because knowledge is not the only factor driving the motivation to perform genetic tests, but rather personal perspectives on the test itself.^{16,17} A previous study has identified that anxiety of invasive procedures, which also apply to the thalassemia screening tests, can influence the willingness to take screening tests.¹⁸ Despite their knowledge about the benefits of genetic testing in thalassemia prevention, there are still some contradictory effects of the test. These potential side effects might influence the motivation to get the test, particularly related to reproductive attitude. For example, positive findings in a genetic test for thalassemia, especially in reproductive age, are considered to negatively influence marriage opportunities.¹⁵ Also, evidence of a negative perspective on living with thalassemia and worries of being stigmatized potentially increase the fear of finding the disease, which is also one of the barriers to a screening test.¹⁹

Families provide a strong motivation for the screening test in which a better understanding of parents could encourage their children to get the test.²⁰ Therefore, family support and motivation are considered essential to complete a good understanding of thalassemia and thus to improve the willingness of adolescents to participate in thalassemia screening tests.

Some limitations needed to be addressed. This current study used only a few question items to identify robust associations from each aspect of knowledge about the motivation for a genetic screening test. It can be seen in the low density of data distribution in each aspect of knowledge. In addition, this study could not deeply elucidate why respondents were unwilling to take the screening test due to the limited information available. However, this study also had some strengths. It has identified a comprehensive understanding that health promotion and education can improve the awareness of thalassemia among adolescents and the response rate to genetic screening tests compared to previous studies. In addition, this study used a reproducible method to evaluate the effectiveness of health education practically, especially related to the risk and prevention of thalassemia. Although health education was performed separately in several groups, the use of standardized materials has maintained the validity and reliability of the measurements. Also, current study included more than the required sample size to conduct a powerful analysis

Further analysis is needed to understand in-depth how health education about thalassemia in adolescents can improve their perspective towards thalassemia prevention, particularly genetic screening test. Factors influencing the willingness to screen need to be studied in-depth to improve strategies to increase public's willingness to take genetic testing. Also, further validation of the improved knowledge of thalassemia risk on willingness to test is required.

In conclusion, health education regarding thalassemia can significantly improve adolescents' knowledge of thalassemia, but it fails to associate with a high willingness to screen. Further study on factors associated with knowledge and willingness to screen is required to validate our findings and identify more considerable factors influencing such preventive measures.

Acknowledgment

We would like to acknowledge Ms. Putri

Halleyana Adrikni Rahman for her assistance in data collection and data entry, and late Prof. Dadang Sjarif Hidajat Effendi for funding this research.

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