Effectiveness of Family Education in Thalassemia Screening Awareness: A Literature Review

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ABSTRACT

Thalassemia is a hereditary disease caused by the failure to form one of the four amino chains that form hemoglobin. The diagnosis of thalassemia carriage in extended family members serves as a significant prevention measure for those at increased genetic risk. Screening for thalassemia carriers by targeting Thaler-major large families can produce more cases of thalassemia carriers in an effort to reduce births with thalassemia. The study aimed to systematically examine family educational interventions in thalassemia screening. This systematic review was carried out using the PRISMA 2020 checklist. This research used databases like PubMed, Cochrane Library, Springer Link, and Base. The inclusion criteria were English-language articles with cross-sectional, case-control,and cohort designs published in the last 10 years (2013-2023). The research found that twenty studies met the inclusion criteria. Keywords used were awareness, thalassemia screening, extended family, and education. The results of the review showed that the majority of the research results described screening for thalassemia as a straightforward method in the preventative effort. Health education will increase awareness and consideration of screening for the prevention and control of severe thalassemia. It requires a comprehensive approach in which community education and promotional activities are tailored to local cultural and religious beliefs.

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1. INTRODUCTION

Indonesia is located along the "Thalassemia Belt" and is a hub for hemoglobinopathies [1]. Approximately 3.0–10.0% of the population has β-thalassemia (β-thal), while 2.6–11.0% have α-thalassemia (α-thal) [2]. Annually, approximately 2500 newborns are born with β-thal major (β-TM) each year. Currently, the primary treatment for β-TM in Indonesia is still supportive, such as blood transfusions and iron chelation therapy [3]. Some cities have poor hemovigilance systems, which increases the risk of transfusion-transmitted disease and reactions. The supply of iron chelators remains questionable; even in some remote locations, iron chelators do not

exist yet [4]. Low compliance with iron chelation therapy and maintenance of pre-transfusion hemoglobin levels above 9.0 g/dL are still the major problems in Indonesia [5]. National health insurance covers the costs of blood transfusions and iron chelation. As life expectancy has increased, so has the financial burden of thalassemia patients in Indonesia. As a result, enhancing prevention strategies may be the best solution for the current state of thalassemia in Indonesia [6]. Currently, around 9,000 thalassemia patients are receiving treatment at thalassemia centers across Indonesia. However, this may be underestimated for a number of reasons, including possible misdiagnosed cases due to a lack of access to health facilities [7]. Because thalassemia is an illness that causes financial difficulties, treatment has been covered in the benefit package for the poor (*JAMKSESMAS*) since 2010 and in the national health insurance program (*JKN*) since 2014. Cyprus is a pioneer in thalassemia prevention, having launched its program in 1980, resulting in a zero percentage birth rate for thalassemia major [8]. Iran, a conservative Islamic country, issued a fatwa permitting prenatal and medical abortion as a part of a thalassemia prevention campaign [9]. Thailand, which has a carrier prevalence rate of up to 40%, has implemented a preventative campaign since the 2000s. The clinical, psychological, and financial burdens are the greatest in Indonesia. Using Hardy Weinberg's rule for illness with recessive inheritance and the present population of 250 million, it is projected that 2,500-3,500 newborns are born with thalassemia major year, based on a prevalence of thalassemia carriers of 3-10%. For individuals at higher genetic risk, the identification of thalassemia carriers in extended family members is an important preventive measure [10]. As a result, our screening study of extended family members may offer an alternative to screening, the general public for the purpose of identifying present and prospective coworkers who run the risk of fathering thalassemia-affected children [11], [12].

2. METHOD

The systematic review was prepared using the guidelines of the PRISMA Checklist 2020. Four databases, including Pubmed, Cochrane Library, Springer Link, and Base, were employed in the review's literature search. The questions pertaining to this article were prepared using the PICO (Patient, Intervention, Comparison and Outcome) technique. This systematic review is one way of identifying, evaluating, and interpreting the availability of research that is relevant to the problem formulation related to the theme of education on thalassemia screening awareness in extended families. The type of data was secondary data obtained from articles in reputable national and international journals and then summarized, drawn conclusions, and found gaps in all the research results. Furthermore, the process of formulating problem questions using the PICOS framework was carried out as follows: 1) Population: families with thalassemia; 2) Intervention: education; 3) Comparison: educational intervention and no intervention; 4) Outcome: awareness of doing screening; 5) Study Design: quantitative, to obtain the research question "Does education about families with thalassemia cause someone's awareness of doing thalassemia screening?"

A number of internet databases, including Google Scholar, Springer Link, Science Direct, and Pubmed, were utilized to search for scientific literature. "Screening," "thalassemia," "extended," "family," "awareness," "education," and "extended family" were search keywords to find relevant sources. For Google Scholar, the keywords used were "thalassemia screening/*skrining thalasemia*" and "extended family thalassemia/*keluarga besar thalasemia*." The specified scientific articles were full-text articles in PDF format published in the 2013-2023 period and articles written in English and Indonesian. Exclusion criteria were identified for duplicate articles.

2.1 Articles Selection

There were four stages of the selection process, which are shown in the diagram [13], namely identification; all search database results were combined, and identical duplicates were identified and then excluded. The second stage was screening; human and full-text research

with 10 years of publication were screened for studies and then excluded. The third stage was eligibility; a feasibility test was carried out on appropriate articles, and the authors began to assess the quality by using an article quality assessment instrument [13]. The final stage was included; studies that have gone through those processes will be further reviewed systematically.

2.2 Quality Assessment

The Critical Appraisal Skills Program (CASP) checklist is a systematic review tool that serves as a roadmap for evaluating studies' viability in terms of validity and reliability before making evidence-based recommendations. The research design was taken into account when choosing the kind of CASP RCT to be employed [14].

2.3 Risk of Bias

Assessment of the risk of bias used the Cochrane Risk of Bias Assessment Tool [15]. This systematic review minimized the risk of bias by determining data extraction, including studies with the same research objectives, research design, which will be reviewed later.

2.4 Data Extraction

Data extraction was performed on the effectiveness of family education in thalassemia screening awareness, including author, year, country, research objective, research method, intervention, result, and conclusion. Data taken from each study were type of research, type of intervention, research result, population, time of publication, and language of publication [16].

2.5 Synthesis of Results

The results of a systematic review were the reporting of data selection in the form of a PRISMA flow diagram, the risk of bias, and explaining the implications and recommendations of the review as a conclusion from the synthesis of documents that has been carried out so far [16]. Data synthesis was conducted using a quantitative method for identifying research like educational interventions for families with thalassemia to raise awareness of thalassemia screening. The search and selection of literature can be seen in the following figure:

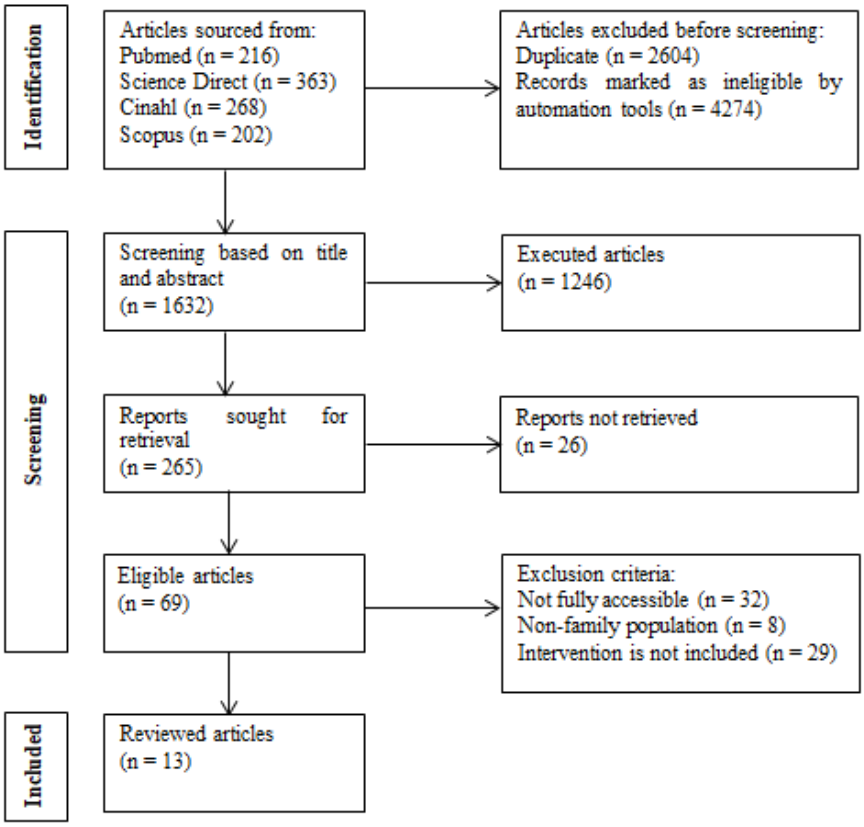

Figure 1. Article Search based on PRISMA

3. RESULTS AND DISCUSSION

3.1 Systematic Search Result

After employing AI to identify duplicates and remove systematic review articles that had been filtered by the search engine, a total of 4216 articles were found through the use of keywords and database sources on the internet. Of these, 1632 articles were returned after further filtering by title and abstract review. Based on the title and abstract review, 1246 articles are excluded because they do not match the research objective, and 69 articles are eligible. Of 69 articles, 32 cannot be fully accessed; 8 articles are not in the population of thalassemia families, and 29 articles show inappropriate interventions, leaving 13 articles remaining.

3.2 Characteristics of Research Articles

A total of 13 articles met the criteria; n = 4 (70%) articles come from India, Pakistan, Indonesia, Thailand, Malaysia, and Sri Lanka. The research designs were cross-sectional (n = 9) articles, cohort (n = 2) articles, and case control (n = 2) articles. The year of publication ranges from 2013 to 2023. There are two articles that do not mention the age range of the respondents involved in the research. The sample range in the research is 115-645 respondents.

Table 1. Data Extraction

No	Author, Year	Country	Title and Source Database	Method	Result
1.	Naresh Dattatraya Sonkawade, Aartu Avinash Kinikar, Rajesh	India	Screening of Extended Family Members of Thalassemia	Cross-sectional (N= 117)	Screening for carriers thalassemia disease in the Thaler extended family majors can earn

No	Author, Year	Country	Title and Source Database	Method	Result
	Kulkarni, Rahul Dawre, Chayat Valvi, Pragathi A Kamath Ethiop J Health Sci (2022)		Preventive Strategy		more thalassemia carrier cases in an effort to decline in the Thaler births major and as a national program [10].
2.	Baihaqi B.S., Hidayah A.N., Rujito L. Health Education and Health Promotion (2023)	Indonesia	Awareness of Non-Health Students about Premarital Genetic Screening	Cross-sectional descriptive study (N = 400)	A total of 355 respondents (88.75%) demonstrate positive behavior toward pre-marital genetic screening. There is no significant relationship between knowledge and attitude ($p > 0.05$), however, there is a significant relationship between attitude and behavior ($p = 0.021$). There is a relationship between attitude and behavior in pre-marital screening and genetics in the non-health field [17].
3.	Rishmitha , Sulochana Badagabettua, Archana M Vb , Vani Lakshmi R, Jomon C U Clinical Epidemiology and Global Health (2022)	India	Awareness on Thalassemia and Opinion of Carrier Screening among Young Women from Selected Undergraduate Colleges of Udupi District	Survey Descriptive Cross-sectional (N = 389)	There is a lack of awareness regarding thalassemia disease among young women, so it is urgent to revise the control thalassemia program [18].
4.	Susi Susanah, Nur Melani Sari, Delita Prihatni, Puspasari Sinaga, Jessica Oktavianus Trisaputra, Lulu Eva Rakhmilla, Yunia Sribudiani	Indonesia	Extended Family Thalassemia Screening as A Feasible alternative Method to be Implemented in Identifying	Control Case (N = 301)	In the control and case groups, 64 out of 150 (42.7%) and 16 out of 151 (10.6%) carriers are found ($p < 0.001$). ÿ thalassemia, ÿ/HbE thalassemia, suspected ÿ

No	Author, Year	Country	Title and Source Database	Method	Result
	J Community Genet (2021)		Carriers in West Java, Indonesia		thalassemia, and Hb variant ÿ-thalassemia are the types of thalassemia carriers in both groups [7].
5.	Rahat, M. A., Ullah, N., Saif, S., Rahman, H. ur, Rasool, A., Shah, M., Akbar, F., Ali, S., & Israr, M. (2023)	Pakistan	Knowledge, Attitudes and Practices regarding The Prevention of Thalassemia in Parents of Thalassemic Children	Survey Descriptive Cross-sectional (N = 200)	This study's knowledge, attitude, and practice are superior to those of earlier research. Programs for population screening thereby improve knowledge. However, resistance from society and religion needs to be overcome so that effective prevention practices can be further implemented [19].
6.	Mohammad Musa Meah, Zabeen Choudhury, Mohammad Bani Yeamin, Bimal Chandra Das, Jhulan Das Sharma American Journal of Pediatrics (2021)	Banglades h	Assessment of Awareness among Parents of Children with Thalassemia Major in Bangladesh: A Hospital based Study	Survey Descriptive Cross-sectional (N = 131)	Parents' knowledge about thalassemia major is the most important thing. The available treatment facilities are inadequate. To lessen the burden of thalassemia, it is the duty of society, government health workers, and educators to recognize the gravity of the issue, inform parents of children affected by the disease as well as the broader public, and raise public awareness of thalassemia prevention strategies and contemporary treatment options [20].

No	Author, Year	Country	Title and Source Database	Method	Result
7.	Nilanga Premawardhana Hematology &Transfusion International Journal (2017)	Sri Lanka	Parental Awareness and Cascade Screening of Thalassaemia	Cross-sectional (N = 200)	Wider awareness and screening programs targeting high-risk populations should be launched quickly because prevention is better for both patients and the government in the terms of cost [21].
8.	Fouzia Ishaq, Hasnain Abid, Farkhanda Kokab, Adil Akhtar, Shahi Mahmood (2013)	Pakistan	Awareness among Parents of β-Thalassemia Major Patients, regarding Prenatal Diagnosis and Premarital Screening	Cross-sectional (N = 115)	Parents' knowledge about thalassemia and its prevention is still lacking. Public health education targeted at the high-risk/target group is needed as a solution in this situation [22].
9.	Singh, Wade, Agrawal (2019)	India	Awareness about Thalassemia and Feasibility of Cascade Screening in Families of Thalassemia Major Patients	Cross-sectional (N = 230 parents with thalassemia)	68.5% of respondents have a positive attitude towards thalassemia [23].
10.	Pauzy et al. (2018)	Malaysia	Thalassemia Distribution based on Screening Programs in the Population	Cross-sectional (N = 645 volunteers)	There are 30% of thalassemia patients, 78% carriers of thalassemia, and 10% carriers of HbE. By optimizing thalassemia screening and counseling programs, thalassemia will become a disease that can be prevented in the future [24].
11.	Nailil Husna, Imanuel Sanka, Ahmad Al Arif, Chintya Putri, Elory	Indonesia	Prevalence and Distribution of Thalassemia Trait Screening	N = 241 individuals from the extended family of	44 volunteers are diagnosed as carriers of β-thalassemia. 30 volunteers are carriers of α-

No	Author, Year	Country	Title and Source Database	Method	Result
	(2017)			thalassemia	thalassemia and HbE disorder. One volunteer is diagnosed with αβ-thalassemia [25]
12.	Anupong Pansuwan et al (2021)	Thailand	Results from 8 Years of the Proficiency Testing Program for Diagnosis Hemoglobinopathies under the Prevention and Control Program of Thalassemia	Cohort	Most laboratories report acceptable MCV and MCH values, a decrease in test error and misinterpretation rates, good performance in the accuracy of thalassemia screening, competence in interpretation, and risk assessment [26].
13.	Ansari (2013)	Pakistan	Screening Immediate Family Members for Carrier Identification and Counseling: A Cost-Effective and Practical Approach	Cross-sectional (N = 188 siblings of 100 thalassemia patients)	Initial screening involves detecting Hb, MCV, and MCH levels, followed by electrophoresis examination in patients with low MCV and MCH [27].

The results of the review show that some studies describe thalassemia screening as a simple strategy in thalassemia prevention programs.

3.3 Discussion

In this study, thirteen articles were included about the theme of thalassemia screening and published from 2013 to December 2023. The research was conducted in Malaysia, Sri Lanka, Bangladesh, Thailand, India, Pakistan, and Indonesia.

3.3.1 Goals of Thalassemia Screening

It is possible to find additional carrier instances if the extended family members of children with thalassemia major are targeted. This could be a low-cost tactic that aids in the development of national thalassemia preventive initiatives [7], [28]. In West Java, Indonesia, the extended family approach that was utilized to obtain increased coverage appears to be a workable alternative for thalassemia screening. In practice, index patients for thalassemia major are their siblings and cousins who are before and during childbearing age [29]. It is advised that Indonesia's national preventive program use a combined strategy in terms of funding and resources, utilizing constrained potential options such as extended family screening [9].

3.3.2 Thalassemia Screening Awareness

This study suggests that the awareness and counseling services offered by population screening programs improve thalassemia patients' knowledge, attitude, and caregiver practice [30]–[32]. This study found a significant prevalence of thalassemia carriers (35%) among EFM children with thalassemia major. Because thalassemia is more common in developing nations, iron deficiency anemia should be eliminated when screening for carrier status [33]. The program must be implemented at the national level and result in effective policies that reduce religious and social conflict. It is an effective preventive measure that can be applied to lessen the illness load [19]. The most crucial thing to understand about thalassemia major is that parents are aware of it [34]. The complications and available therapeutic facilities are insufficient. Most of them are unaware of the pattern of inheritance or the need for premarital screening, which is a key method for preventing disease transmission. Furthermore, they have a limited understanding of transfusion-transmitted illnesses, the necessity of iron chelation therapy, and the many alternatives available to patients with thalassemia major. If parents of children with thalassemia had more knowledge, they could better comprehend the importance of the condition, safe blood transfusions, current treatment facilities, prenatal diagnostics, and other prevention strategies [17], [27], [35], [36].

3.3.3 Thalassemia Prevention Program

This is beneficial because: (1) it helps affected families prepare for having a sick child; and (2) it serves as an important secondary preventative measure [36]. The question is whether neonatal screening programs can successfully help achieve the intended results. In locations where effective pre-conception or pre-marital prevention initiatives are fully implemented, few events occur after birth. In this case, impacted newborns frequently come from families that have been told and have decided to give birth to the affected child. Despite the lack of preventive programs, newborns typically present clinically at an early age and require rapid treatment due to severe anemia. It appears unnecessary to design policies only focused on the early identification of thalassemia. However, if sickle cell disease detection programs existed, multiple thalassemia syndromes and their variants may be detected [37]–[39]. This can be effective for secondary prevention, but it can also be used to detect new cases early [21]. This set of mass educational events is critical in preparing and increasing community readiness for the next step in the thalassemia prevention program campaign, which is screening for thalassemia carriers among target populations. Carrier screening is the first critical step in thalassemia prevention [40] and Indonesia will face challenges because hundreds of ethnic groups will be tested [41]. Cultural diversity makes it difficult to apply new technologies or health interventions like job screening programs. This emphasizes the significance of systematic and widespread mass education activities tailored to local language and culture with the goal of enhancing community acceptability of screening programs. Implementation of a screening program can be classified into many levels of urgency depending on the target demographic [42]. The first target population is the extended families of thalassemia patients. This target population is thought to be the most important to screen. According to one study, around half of the extended families of thalassemia patients possess thalassemia gene mutations [43]. Screening the extended family of thalassemia patients varies their carrier status, which will serve as the foundation for counseling programs on reproductive planning [24].

To execute a nationwide thalassemia screening program in a systematic manner, a clear national health policy is required. A well-defined national health policy will serve as the legal basis for allocating the necessary resources for the program. This entails creating human resources and health-care infrastructure, spreading screening tools, and establishing roles and responsibilities for central and regional governments. The Ministry of Health is responsible for supplying suitable numbers of health personnel, developing competencies, and distributing them fairly around the country. Specific capacity-building programs should include hematology analysis, interpretation abilities, and competence for providing appropriate

counseling to the target group. There is also a need to establish a network of services based on academic centers to cover other tests, such as genetic analysis, to complete the detection procedure. The thalassemia screening program must be available to all community groups in Indonesia, regardless of socio-economic, cultural, and geographic background. A possible option to provide fair access to the thalassemia screening program is to include the program as part of the JKN benefits. Reducing the burden of thalassemia by implementing national prevention programs has proven beneficial in many endemic countries [44]. The extended family method, which can obtain higher coverage, seems feasible to be applied as a potential alternative thalassemia screening method in West Java, Indonesia. Practically, screening is focused on siblings and cousins before and during childbearing age of thalassemia major sufferers as index cases [45]. For the national prevention program in Indonesia, it is recommended to apply a combined approach regarding resources and budget, using limited prospective strategies and combining them with retrospective strategies such as extended family screening. Thalassemia education adopts disease carrier screening and the development of thalassemia prevention and control programs [32], [46]. It is the responsibility of the government, health workers, and the community to understand the density of the problem, educate parents of children with thalassemia, as well as the general public, and create public awareness about modern treatment facilities and aspects of thalassemia prevention. Inter-professional collaboration between stakeholders can strengthen the government's performance when considering policies for handling thalassemia in Indonesia [44], [47]. The program requires planning and investment to include public awareness, screening to identify disease carriers [20]. Strategies to control thalassemia need to include awareness programs that must be carried out to increase knowledge among parents of children with thalassemia. National and international guidelines provide recommendations regarding the most appropriate way to implement genetic carrier screening programs; however, this recommendation is not followed in many programs [9]. Education and socialization of thalassemia among minority groups at risk are still lacking. Although it is easy to argue that the lack of outreach has to do with language barriers and distrust of treatment, thalassemia prevention and control strategies include population screening for heterozygotes, genetic counselling, and fetal diagnosis with selective abortion of affected pregnancies [48], [49]. The concept of "community participation" is pushed forward, which is one of the most important principles of PHC, namely strengthening the implementation of thalassemia major awareness through educational programs, health counseling, premarital examinations, and campaigns [50]. Relevant health education techniques, which can be identified by the public, can effectively provide the understanding needed to raise public awareness. We propose the 'Bag and Ball' method, which includes role-playing for health education specifically regarding inherited genetic disorders [26]. Parents' knowledge about thalassemia and prevention efforts is still lacking [51]. It requires intervention in the form of public health education programs that concentrate on high-risk or target populations [23], [33], [52], and intervention in the form of a public health education program that concentrates on high-risk or target populations [22]. Wider awareness and screening programs that target high-risk populations must be launched immediately because prevention is better both for the patient and for the government in terms of cost. Prevention programs have reduced the birth prevalence of thalassemia in several countries, creating public awareness about modern treatment facilities and aspects of thalassemia prevention. The program requires planning and investment to include public awareness, screening to identify disease carriers, and cost-effective strategies that can help in the preparation of a national program for the prevention of thalassemia [53].

4 CONCLUSION

Health education will raise awareness of and encourage screening for severe thalassemia in order to prevent and control it. The efficacy of community-based thalassemia prevention and control was contrasted with health education. Local cultural and religious values must be taken

into consideration while developing a comprehensive approach to community education and promotional activities. Encouraging thalassemia screening also requires a thorough and multisectoral strategy. This entails raising awareness of the issue and promoting a perception of elevated risk, as well as enhancing patient education, thalassemia screening accessibility, and school-based promotion and screening initiatives. Targeting the Thaler main family for thalassemia carrier screening may result in more cases of thalassemia carriers and a decrease in the number of thalassemia generation births.

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