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ORIGINAL RESEARCH

Cost-Benefit Analysis of Genetic Testing as a Prenatal Diagnostic Tool for Thalassemia: A Single-Center Study From Central Thailand

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Purpose: This study aimed to evaluate the costs and benefits of genetic testing, specifically mutation analysis and prenatal diagnostic testing, for the confirmation of thalassemia in at-risk pregnancies in Thailand, providing crucial insights to inform public health policy decision-making.

Patients and Methods: We analyzed the costs and benefits of following standard screening guidelines, which included a sequence of tests such as mean corpuscular volume (MCV)/mean corpuscular hemoglobin (MCH) with dichlorophenol indophenol precipitation (DCIP), hemoglobin (Hb) typing, genetic testing, and amniocentesis. A decision-tree model was employed for this analysis. The study compared the scenarios with and without genetic testing, adopting a societal perspective that accounted for costs during pregnancy and the lifetime of a child born with thalassemia. Both one-way and probabilistic sensitivity analyses were conducted to account for uncertainties in the parameters used.

Results: The results revealed that adhering to the standard screening program with genetic testing resulted in a cost-savings of approximately 490 USD per prevented thalassemia case. Among the diagnostic methods, the specificity of the MCV/MCH with DCIP showed a higher degree of sensitivity relative to other testing methods, significantly influencing the outcomes. From a governmental perspective, with a full uptake of genetic testing, the incremental budget required was estimated to be 3.7 million USD (131 million THB) for one year.

Conclusion: These findings are particularly valuable for policymakers, as they provide robust evidence supporting potential revisions to the reimbursement structure within Thailand's Universal Health Coverage benefit package, facilitating better management of thalassemia and improving prenatal care.

Keywords: cost-benefit analysis, genetic testing, prenatal screening, thalassemia, pregnancy

Introduction

Thalassemia represents a diverse spectrum of genetic disorders caused by the reduced synthesis of alpha or beta chains of hemoglobin (Hb), and it is inherited, meaning at least one parent must be a carrier of the disease.[1] The prevalence of α-thalassemia is notably high in Asia, especially Southeast Asia, where it is estimated to affect 22.6% of the population,[2] while β-thalassemia is more commonly observed in Mediterranean regions.[1] In Thailand, the prevalence of α-thalassemia is approximately 20.1%,[2] with β-thalassemia affecting between 3% to 9% of the population.[3] Projections based on gene frequencies and annual births suggest that approximately 1.2% of newborns in Thailand will have severe thalassemia.[4]

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Consequently, the Thai government has implemented a strategic plan for the prevention and control of thalassemia in pregnancy, aiming to reduce the incidence of this condition through early detection and intervention.[5]

Genetic testing serves as the definitive step in prenatal screening, facilitating the identification of couples at risk for severe thalassemia and providing them with crucial information to make informed reproductive decisions and pursue prenatal diagnosis if necessary.[6] Both the National Health Service (NHS) and the American College of Obstetricians and Gynecologists recommend antenatal thalassemia screening protocols similar to those adopted in Thailand.[6–8] The screening process in Thailand begins with a pregnant woman undergoing a combination of the single tube osmotic fragility (OF) test or red cell indices such as mean corpuscular volume (MCV) and mean corpuscular hemoglobin (MCH) along with dichlorophenol indophenol precipitation (DCIP) tests.[8] If these initial screening results are positive, the pregnant woman's partner is also invited to participate in the same screening tests.[8] Couples who both test positive for any of the screening tests (OF, MCV, MCH, or DCIP) then proceed to hemoglobin typing (Hb typing) and/or DNA analysis. Couples identified as high-risk for having a child with severe thalassemia are advised to undergo prenatal diagnosis (PND), commonly performed via amniocentesis, to confirm the diagnosis in the fetus.[8,9]

The Thalassemia Prevention and Control Program, initiated by the World Health Organization (WHO), and the Ministry of Public Health of Thailand in 1992, was designed to reduce the incidence of severe thalassemia in newborns and enhance the quality of life for individuals affected by the disorder.[4] In 2014, the initiative gained further support from the National Health Security Office (NHSO) through the Thalassemia Prevention and Control in Pregnancy Program.[4] While thalassemia screening is included in the Universal Health Coverage (UHC) benefit package for pregnant women, comprehensive coverage for genetic testing especially for β-thalassemia remains limited across Thailand's health insurance schemes.[10] Despite a formal request from the NHSO, no economic evaluation of genetic testing for prenatal thalassemia screening has been conducted to date. This gap in knowledge limits the ability to make evidence-based decision about the potential adoption of a universal prenatal screening policy. In response, this study seeks to evaluate the cost-benefit of incorporating genetic testing into prenatal thalassemia screening in Thailand. The analysis compares scenarios that include genetic testing against those without it and estimates the budget impact assuming a 100% uptake rate for each scenario. The findings of this study aim to fill the current knowledge gap and provide critical insights for decision-makers, helping to inform potential revisions in national policies and health coverage for prenatal thalassemia screening.

Materials and Methods

Study Design

We conducted a cost-benefit analysis using a decision-analytic model to compare the costs and benefits of standard thalassemia screening program with genetic testing versus screening without genetic testing for pregnant women in Thailand. The study population included cohorts of pregnant women of all age groups, along with their at-risk spouses. Spouses underwent screening if the pregnant woman's initial test either MCV, MCH, or DCIP yielded positive results, following the Royal Thai College of Obstetricians and Gynecologists Clinical Practice Guidelines.[8] We modeled the costs and benefits during pregnancy and throughout the lifetime of individuals with thalassemia from a societal perspective. An annual discounting rate of 3% was applied to both costs and outcomes according to the Thai Health Technology Assessment Guidelines.[11] Cost-benefit calculation was measured as net benefit and benefit-to-cost ratio. Net benefit (Δ benefit-Δ cost) reflects the incremental difference between the benefits and costs of the standard screening program with genetic testing versus screening without genetic testing. Benefit-to-cost ratio (Δ benefit/Δ cost) represents the efficiency of the investment in the screening program with genetic testing. Here, Δ benefit denotes the difference in benefits between the standard screening and screening without genetic testing, while Δ cost represents the difference in costs between the two approaches. Additionally, we estimated the financial implications of adopting each screening strategy by multiplying the per-person total cost from the governmental perspective by the number of 700,000 single pregnancies occurring annually in Thailand.[12]

Model Structure

The decision-analytic model employed in this study was derived from the Royal Thai College of Obstetricians and Gynecologists Clinical Practice Guidelines.[8] To ensure the accuracy and reliability of the model, face validity was performed

by three obstetricians. The model simulates two scenarios ie, standard thalassemia screening program involving genetic testing and screening without genetic testing. Figure 1 depicts decision tree model used in this study. The process starts with all pregnant women either accepting or declining MCV/MCH with DCIP screening test (Figure 1a). If they decline the screening, the pregnancy outcomes include normal newborns, miscarriage, and severe or non-severe thalassemia in newborns. If they accept the screening, results can be either positive or negative, determined by the test's sensitivity and specificity. In the case of a negative test result, the pregnancy outcomes (normal newborns, miscarriage, or thalassemia) are monitored. For a positive test result, the at-risk husband is then offered MCV/MCH with DCIP screening to determine whether both partners are carriers of thalassemia (Figure 1b). If both are positive, the couple is then offered a Hb typing test to confirm their carrier status (Figure 1c). If the Hb typing test is positive, genetic resting for either α-thalassemia, β-thalassemia or both is then offered to the pregnant woman (Figure 1d). In contrast, pregnant women in the scenario of screening without genetic testing would proceed directly to a final diagnosis to assess whether they have babies with thalassemia or not (Figure 1e). If a final diagnosis is needed, the pregnant woman is offered amniocentesis, which has the following outcomes: procedure-related loss, such as miscarriage due to the test, true or false positives, and true of false negatives, depending on the test's sensitivity and specificity. If the pregnant woman declines the amniocentesis, the outcomes are spontaneous abortion or live birth with either severe thalassemia, non-severe thalassemia, or a normal newborn.

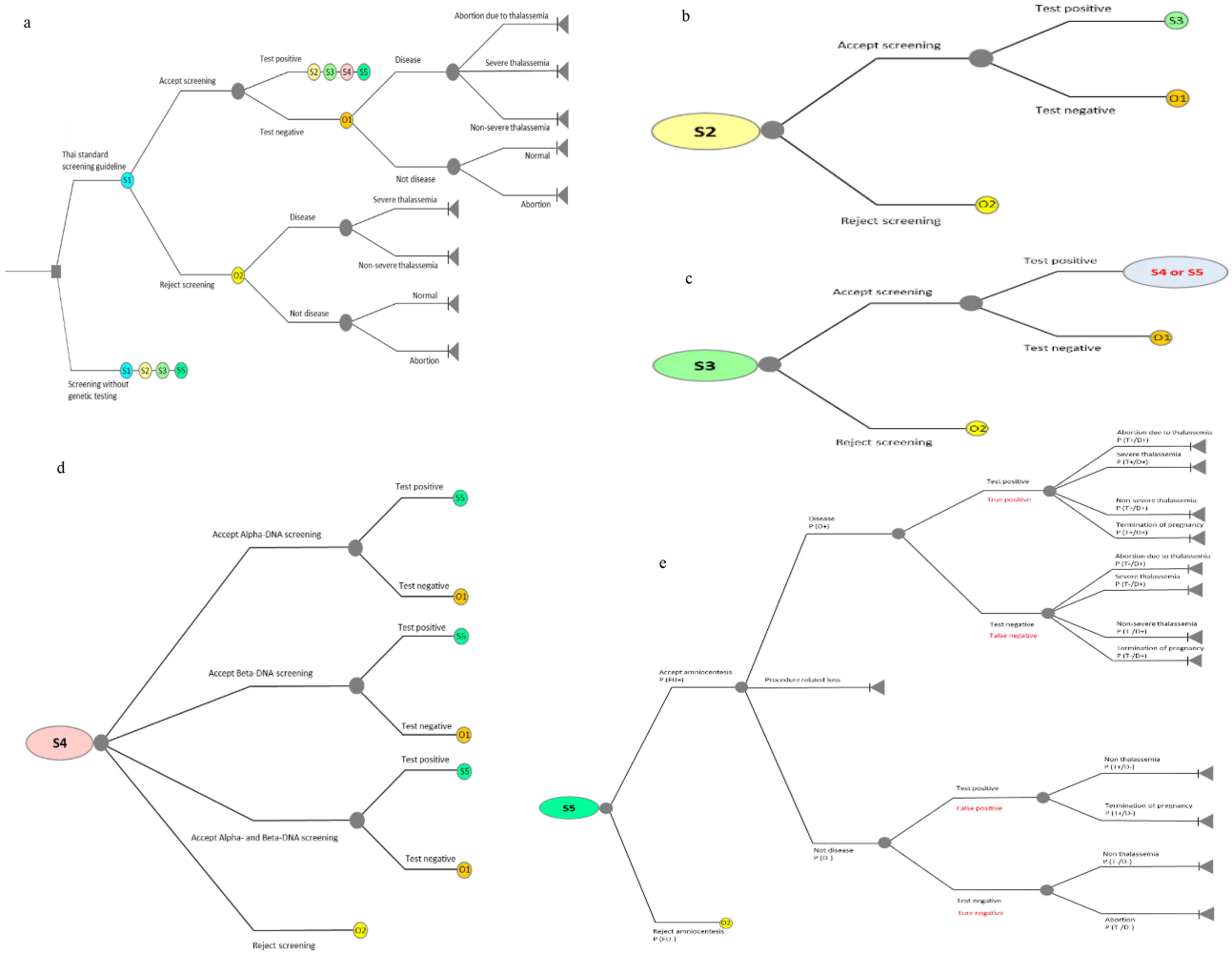

Figure 1 (**a**) Overall decision-tree model, (**b**) Decision-tree model for at-risk husbands screened by MCV/MCH with DCIP (S2), (**c**) Decision-tree model for couple screened by Hb typing (S3), (**d**) Decision-tree model for pregnant women screened by genetic testing (S4) (**e**) Decision-tree model for pregnant women diagnosed by amniocentesis (S5).

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Model Parameters

The input parameters manipulated in the model were categorized into four groups, as follows: epidemiological data and probabilities, screening performance, cost, and benefit. The values of these parameters are detailed in Table 1.

Epidemiological Data and Probabilities

We obtained epidemiological data on the disease and probabilities for screening and uptake rates from a previous study which collected data from a retrospective chart review of 4,062 pregnancies in Siriraj hospital database from January 2019 to December 2019.[13] The ethics of this study was approved by Siriraj Institutional Review Board (SIRB) (MU-MOU COA Si 1053/2020) and the ethics committee waived the requirement for informed consent. All methods were carried out in

Table 1 Input Parameters Used in the Model

Parameter	Type of Distribution	Mean	Standard Error	Reference
Probability of occurrence of events as for various scenarios				
Probability of a positive test result of MCV/MCH+DCIP in pregnant women	Beta	0.303	0.030	[9,13]
Probability of a positive test result of MCV/MCH+DCIP in risk husbands	Beta	0.330	0.033	[9,13]
Probability of a positive test result of Hb typing in couple	Beta	0.325	0.032	[9,13]
Probability of a positive test result of genetic testing for α-thalassemia	Beta	0.738	0.074	[9,13]
Probability of a positive test result of genetic testing for β-thalassemia	Beta	0.019	0.002	[9,13]
Probability of a positive test result of genetic testing for α- and β-thalassemia	Beta	0.014	0.001	[9,13]
Prevalence of thalassemia	Beta	0.010	0.001	[9]
Prevalence of severe thalassemia	Beta	0.005	0.001	[13]
Prevalence of abortion in all pregnant women	Beta	0.061	0.006	[13]
Prevalence of abortion in thalassemia cases	Beta	0.042	0.004	[13]
Prevalence of abortion in normal case after amniocentesis	Beta	0.007	0.001	[13]
MCV/MCH+DCIP acceptance rate in pregnant women	LOG normal	0.989	0.099	[13]
MCV/MCH+DCIP acceptance rate in risk husbands	Beta	0.279	0.028	[13]
Hb typing acceptance rate in risk couple	Beta	0.826	0.083	[13]
Genetic testing acceptance rate for α-thalassemia	Beta	0.180	0.018	[13]
Genetic testing acceptance rate for β-thalassemia	Beta	0.064	0.006	[13]
Genetic testing acceptance rate for α- and β-thalassemia	Beta	0.034	0.003	[13]
Amniocentesis acceptance rate after genetic testing for α-thalassemia	Beta	0.130	0.013	[13]
Amniocentesis acceptance rate after genetic testing for β-thalassemia	Beta	0.476	0.048	[13]
Amniocentesis acceptance rate after genetic testing for α- and β-thalassemia	Beta	0.303	0.030	[13]
Amniocentesis acceptance rate after Hb typing	Beta	0.017	0.002	[13]
Rate of pregnancy termination in case of alpha-thalassemia	Beta	0.875	0.088	[13]
Rate of pregnancy termination in case of beta-thalassemia	Beta	0.923	0.092	[13]
Rate of procedure-related abortion in all pregnant woman	Beta	0.002	0.000	[13]

(*Continued*)

Table 1 (Continued).

Parameter	Type of Distribution	Mean	Standard Error	Reference
Direct medical costs per test or visit (USD)				
Cost of MCV/MCH+DCIP	Gamma	6.50	1.30	[17]
Cost of Hb typing	Gamma	7.63	1.53	[17]
Cost of genetic testing for α-thalassemia	Gamma	22.62	4.52	[17]
Cost of genetic testing for β -thalassemia	Gamma	59.37	11.87	[13]
Cost of ultrasound	Gamma	25.45	5.09	[17]
Cost of normal labor	Gamma	235.51	47.10	[13]
Cost of cesarean section	Gamma	332.49	66.50	[13]
Cost of medical services procedure-related loss	Gamma	83.12	16.62	[13]
Cost of elective termination	Gamma	166.24	33.25	[13]
Cost of termination of pregnancy/miscarriage	Gamma	277.07	55.41	[13]
Direct non-medical costs per visit (USD)				
Cost of travel	Gamma	4.20	0.84	[18]
Cost of food per visit	Gamma	1.55	0.31	[18]
The opportunity cost of pregnant women	Gamma	2.37	0.47	[18]
The opportunity cost of a caregiver	Gamma	2.82	0.56	[18]
Indirect costs (USD)				
Loss from abortion due to amniocentesis or intentional abortion in normal case	Gamma	490,848.49	98,169.70	Calculated from GDP 20
Productivity loss due to a decrease of working capacity in beta-thalassemia	Gamma	278.82	55.76	[22]
Direct benefits (USD)				
Cost avoidance of severe α-thalassemia	Gamma	665.58	133.12	[23]
Cost avoidance of severe β-thalassemia	Gamma	148,824.45	29,764.89	[13]
Cost avoidance of non-severe thalassemia	Gamma	46,706.64	9,341.33	[13]
Indirect benefit (USD)				
Productivity gain of caregiver severe α-thalassemia	Gamma	7,438.14	1,487.63	[20]
Productivity gain of caregiver severe β-thalassemia	Gamma	257,600.68	51,520.14	[20]
MCV/MCH+DCIP	LOG normal	0.838	0.943	[16]
Hb typing	LOG normal	0.990	0.990	[13]
Genetic testing for α-thalassemia	LOG normal	0.990	0.990	[13]
Genetic testing for β-thalassemia	LOG normal	0.990	0.990	[13]
Amniocentesis	LOG normal	0.990	0.990	[13]

Abbreviations: MCV, mean corpuscular volume; MCH, mean corpuscular hemoglobin; DCIP, dichlorophenol indophenol precipitation; Hb, hemoglobin; USD, US dollar.

accordance with International Guidelines for Human Research Protection such as the Declaration of Helsinki, Belmont Report, CIOMS Guidelines and International Conference on Harmonization in Good Clinical Practice (ICH-GCP). Additionally, this study utilized data from published studies focusing on the outcomes of prenatal thalassemia screening in Thailand.[9,20] The calculations were performed using a formula derived from an economic evaluation of disease screening,[21] where the probability of a positive test is defined as (sensitivity × prevalence) + [(1-specificity) × (1-prevalence)]. Here, prevalence is the number of cases that tested positive on the screening divided by the total number of cases screened, while sensitivity and specificity represent the performance of the screening test.

Screening Performance

We used sensitivity and specificity data from a previous study which retrieved data from Siriraj hospital database[13] and published studies.[19]

Cost

The resource utilization costs in this study were evaluated from a societal perspective, encompassing direct medical costs, direct non-medical costs, and indirect costs adjusted to 2023 values using the consumer price index (CPI). Subsequently, costs were converted from Thai baht (THB) to USD at an exchange rate of 35.37 THB per 1 USD (2023 prices). All future costs and health outcomes were discounted to their present values at a rate of 3% per annum. Direct medical costs, including those associated with prenatal screening and diagnostic tests, ultrasound examinations, medical services related to procedure-related loss, delivery procedures (elective termination, normal labor, cesarean section), and service care for termination of pregnancy or miscarriage, were derived from price lists of governmental and hospital databases.[13,14] The ratio of total cesarean sections to the number of normal deliveries was 54:46.[13]

Direct non-medical costs comprised travel, food, and opportunity costs of pregnant women and caregivers during the screening test, sourced from general hospital data on the Standard Cost List for Health Technology Assessment (HTA), a recognized reference cost list in Thailand.[15] Indirect costs reflected productivity losses due to miscarriage resulting from definitive diagnosis or termination of a non-thalassemia case, calculated using a human capital approach.[19] Productivity loss or income loss was estimated by multiplying the working age range by the Thai Gross Domestic Product (GDP) per capita per year (6,908.8 USD).[22] The working age range was assumed to be 45 years (15–60 years). Following the recommendation from Thai HTA guidelines, considering different cost values over time, future values of total expected productivity loss (FV) were adjusted to present values (PV) using an annual discount rate of 3% based on the formula: $PV = FV \times [1/(1+r)^n]$, where PV = present value, FV = future value, r = discount rate, and n = each year in the future.[11] Additionally, we assumed an annual income growth rate of 4%, derived from the income growth rate during 1990–2023 in Thailand.[23] Furthermore, productivity loss due to a decrease in working capacity in β-thalassemia was obtained from a published study by Riewpaiboon et al.[17]

Benefit

We utilized direct benefit data as a measure of cost avoidance achieved by averting the occurrence of severe thalassemia types α and β, including non-severe thalassemia in children resulting from each screening. This benefit data was derived from a previous study,[13] which involved determining the average total healthcare costs of patients with severe thalassemia type β and non-severe thalassemia. Concurrently, the costs of treating patients with severe thalassemia type α were obtained from a previous study utilizing governmental databases.[18] Moreover, we estimated indirect benefits in terms of productivity gains for caregivers who were not required to care for children with severe thalassemia due to the prenatal screening test. The human capital approach was employed by multiplying the average expected survival of severe thalassemia patients with the expected income of caregivers, referencing the annual Thai GDP per capita (6,908.8 USD).[16] We assumed a one-year survival for fetuses with severe thalassemia type α and a 30-year survival for those with severe thalassemia type β, based on expert opinions and a published study by Dhanya et al.[24] For caregivers of children with severe thalassemia type β, the future values of total expected productivity gain were adjusted to their present values using a discount rate of 3%[11] and an income growth rate of 4% per year.[23]

Table 2 Cost-Benefit Analysis Results (USD)

Thalassemia Screening Modalities	Costs	Benefits	Δ benefit-Δ Cost	Δ benefit/Δ Cost
Screening without genetic testing	427.76	1,505.54		
Standard screening program with genetic testing	427.73	1,995.48	490	Cost saving

Uncertainty Analysis

We conducted both one-way and probabilistic sensitivity analyses (PSA) to assess the model's sensitivity to each input parameter. One-way sensitivity analysis involved varying each input parameter within its 95% confidence interval (CI), and the resulting range of net benefit values was presented using a Tornado diagram. Furthermore, we simultaneously evaluated the uncertainty of all parameters through a 1,000 Monte Carlo simulation. Probabilities were assigned a beta distribution, performances followed a log-normal distribution, and cost and benefit parameters adhered to a gamma distribution. The PSA results were depicted as a cost-benefit plane.

Results

Cost-Benefit Analysis

The decision-analytical model employed in this study simulated a scenario wherein pregnant women and at-risk husbands underwent tests for screening without genetic testing, as well as a standard screening program with genetic testing throughout the pregnancy and over the lifetime of a thalassemia-affected newborn. Table 2 presents the estimated total costs, benefits, net benefit, and the benefit-to-cost (B/C) ratio. From a societal perspective, the analysis revealed that the implementation of a standard thalassemia screening program with genetic testing resulted in a net benefit of 490 USD compared to screening without genetic testing. Consequently, a cost saving was observed with the adoption of the standard thalassemia screening program with genetic testing.

Uncertainty Analysis

The results of the one-way sensitivity analysis for the standard thalassemia screening program with genetic testing are illustrated in Figure 2. The net benefit was the most sensitivity to the specificity of the MCV/MCH with DCIP test, with

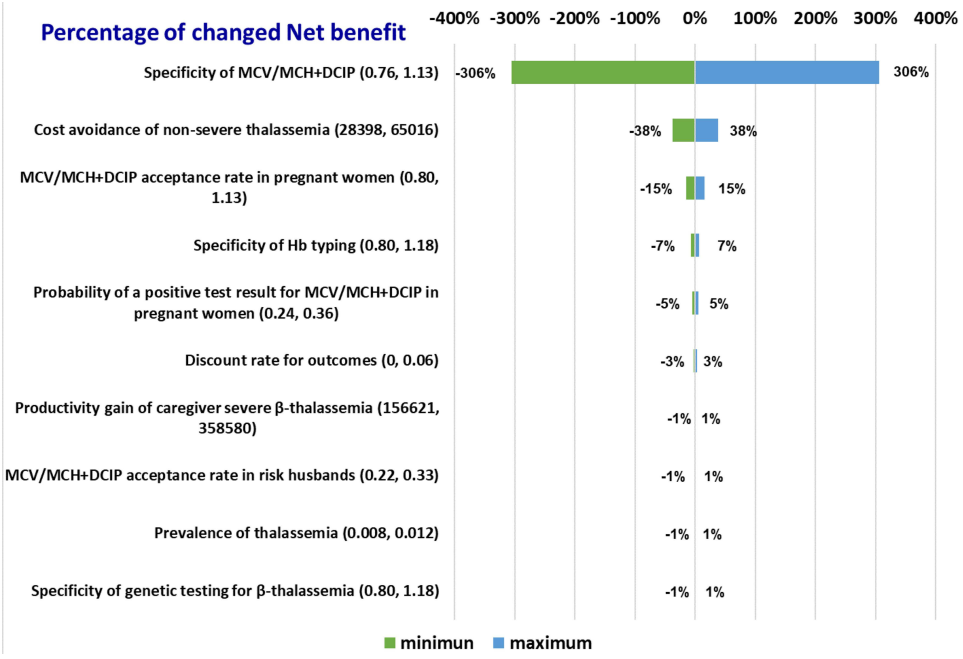

Figure 2 Tornado diagram for the standard thalassemia screening program with genetic testing.
Abbreviations: MCV, mean corpuscular volume; MCH, mean corpuscular hemoglobin; DCIP, dichlorophenol indophenol precipitation; Hb, hemoglobin.

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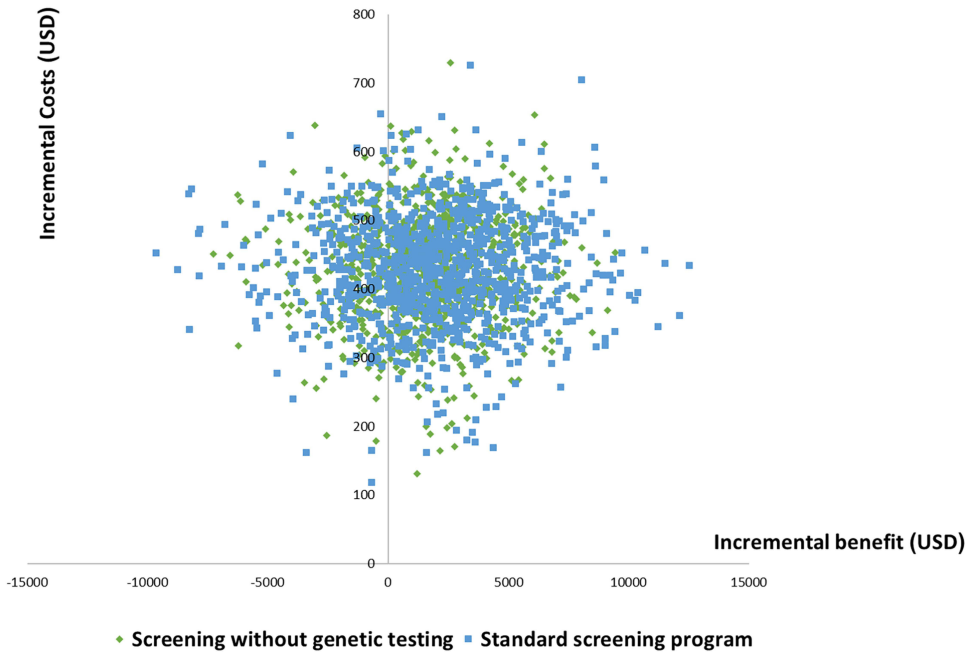

Figure 3 Cost-benefit plane. Green diamonds represent the cost and benefit outcomes of screening without genetic testing, while blue squares represent those of standard screening with genetic testing.

subsequent sensitivity observed in the cost avoidance of non-severe thalassemia, the acceptance rate of MCV/MCH +DCIP in pregnant women, the probability of a positive test result for MCV/MCH+DCIP in pregnant women, and the discount rate for outcomes. Figure 3 portrays the outcomes derived from the PSA. The cost-benefit plane revealed that the majority of simulations were situated in the northeast quadrant.

Budget Impact Analysis

Table 3 provides insight into the budgetary impact of each screening test from the governmental perspective over the course of one year. The incremental budget, amounting to approximately 3,703,457 USD (130,991,266 THB) was incurred for the standard thalassemia screening program with genetic testing, assuming a 100% uptake rate.

Table 3 Budget Impact Analysis of Each Screening Test With a 100% Uptake Rate

Screening Test	Genetic Testing Not Included in UHC		Genetic testing Included in UHC		Incremental Budget (USD)
	Cost Per Test	Total Budget	Cost Per Test	Total Budget	
MCV/MCH+DCIP in pregnant woman	6.50	4,502,583	6.50	4,551,880	49,297
MCV/MCH+DCIP in at-risk husbands	6.50	380,359	6.50	1,378,221	997,862
Hb typing	7.63	243,559	7.63	1,068,955	825,396
Genetic testing for α-thalassemia	22.62	21,093	22.62	514,017	492,924
Genetic testing for β-thalassemia	33.93	11,315	59.37	1,349,294	1,337,978
Total incremental budget (USD)					3,703,457

Abbreviations: MCV, mean corpuscular volume; MCH, mean corpuscular hemoglobin; DCIP, dichlorophenol indophenol precipitation; Hb, hemoglobin; UHC, universal health coverage; USD, US dollar.

 https://doi.org/10.2147/CEOR.S500802

Discussion

This study represents the first comprehensive evaluation of the cost-benefit of genetic testing for prenatal thalassemia screening in comparison to screening without genetic testing among Thai pregnant women and at-risk husbands suspected of carrying thalassemia in Thailand. The findings indicated that the standard thalassemia screening program with genetic testing was more cost-saving than screening without genetic testing. Moreover, results from the PSA suggested that while the costs were comparable, genetic testing proved more beneficial than screening without genetic testing. Our finding was consistent with several previous cost-benefit and cost-effectiveness analysis studies, which suggested that thalassemia screening program in pregnancy was more cost-saving than no screening.[25–28] Consequently, genetic testing for β-thalassemia is currently fully reimbursed in Thailand's health benefit packages.[29]

While all tests in the prenatal thalassemia screening strategy are provided free of charge, the uptake rate for each screening test was notably low, particularly in the case of test acceptance among husbands during the initial screening, registering at only 27.9%.[13] This low acceptance rate has implications for the potential loss of identifying suspected husbands who may be carriers of thalassemia, increasing the risk of having offspring with thalassemia. Additionally, accessibility to screen for Hb typing and genetic testing was limited across general hospitals, tertiary hospitals, and university hospitals. The survey conducted across 95 hospitals in Thailand revealed that Hb typing was available in 67.4% of the facilities, while genetic testing was accessible in only 24.2%.[23]

Although the Royal Thai College of Obstetricians and Gynecologists has established clinical guidelines for prenatal screening and diagnosis in pregnant women, variations in strategies exist among different hospitals.[23,30] The approach adopted depends on the prevalence of thalassemia in each region of Thailand, including the Northeast. In this context, pregnant women and their husbands decide to undergo simultaneous screening of MCV and MCH with DCIP initially, given the high prevalence of Hb E in the Northeast of Thailand.[4,23] Furthermore, certain university hospitals opt for screening Hb typing instead of DCIP to streamline the process, reduce waiting times, and enhance test acceptance among at-risk husbands.[30] This screening approach bears similarities to the thalassemia screening in Australia and the carrier screening for thalassemia and hemoglobinopathies in Canada.[31,32]

Despite the significant findings presented herein, it is imperative to acknowledge several limitations. Firstly, the uptake rate for screening and epidemiological data was derived solely from one hospital located in the central region of Thailand, characterized by a low to medium prevalence of thalassemia. As a result, our analysis may underestimate the potential benefits associated with identifying new thalassemia cases. Secondly, the intangible benefit of pregnant women's willingness to pay for screening was not taken into consideration, this omission may underestimate the overall benefit of screening, leaving room for future studies to explore this aspect further. Lastly, the current study revealed that the percentage of screening genetic testing for both α- and β-thalassemia was 100%, higher than the prevalence of either genetic testing for α- or β-thalassemia in Thailand in 2019, which stood at 28–87%.[9] Therefore, this would result in an overestimation of the budget impact.

Conclusion

This study provided supportive evidence that from a societal perspective, genetic testing, when incorporated into the prenatal thalassemia screening strategy, was more cost-beneficial than screening without genetic testing for pregnant women and at-risk husbands with suspected thalassemia carriers in Thailand, offering long-term benefits that outweighed the costs to society as a whole. The estimated annual budget impact of screening with a 100% uptake rate during pregnancy amounted to 3.7 million USD (131 million THB). Our research contributes valuable insights that may guide policymakers advocating for the incorporation of genetic testing for β-thalassemia into Thailand's UHC benefit package, aligning with the WHO and Thailand's operational strategy to prevent and control new thalassemia cases. Further investigations should be undertaken to assess the inclusion of various prevalence rates in determining the most value-centric strategy in future studies.

Ethics Approval

Our study used data from previous published studies. The ethics of this study was approved by Siriraj Institutional Review Board (SIRB) (MU-MOU COA Si 1053/2020). The need for informed consent for this study was waived by the ethics committees, as all data were obtained from published studies. All methods were carried out in accordance with

International Guidelines for Human Research Protection such as the Declaration of Helsinki, Belmont Report, CIOMS Guidelines and International Conference on Harmonization in Good Clinical Practice (ICH-GCP). All procedures performed in the study were in compliance with international guidelines for human research protection such as Declaration of Helsinki, the Belmont Report.

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Disclosure

The authors report no conflicts of interest in this work.

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