

Article title: Economic Evaluation of Screening Strategies for Thalassaemia Prevention: A Case Study From Thailand

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Authors' information: Phyllis Tenkorang¹, Phrutsamon Wongnak², Benjaporn Kiatpakdee³, Kittiphong Paiboonsukwong³, Na'ima Nasir¹, Caesar Atuire¹, Chris Painter^{1,2,4}, Wirichada Pan-ngum^{2,5*}

¹Centre for Tropical Medicine and Global Health, Nuffield Department of Medicine, University of Oxford, Oxford, UK.

²Mahidol-Oxford Tropical Medicine Research Unit (MORU), Faculty of Tropical Medicine, Mahidol University, Bangkok, Thailand.

³Thalassemia Research Center, Institute of Molecular Biosciences, Mahidol University, Nakhon Pathom, Thailand.

⁴Lao-Oxford-Mahosot Hospital Wellcome Trust Research Unit (LOMWRU), Microbiology Laboratory, Mahosot Hospital, Vientiane, Lao People's Democratic Republic.

⁵Department of Tropical Hygiene, Faculty of Tropical Medicine, Mahidol University, Bangkok, Thailand.

***Correspondence to:** Wirichada Pan-ngum; Email: pan@tropmedres.ac

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Supplementary file 1

Table S1: Evaluated comparators with their full description

Comparator	Description
Post-conception screening — Strategy 1	This is the current screening strategy implemented in Thailand. It is targeted at pregnant couples* who visit health facilities for antenatal care (ANC). CBC & DCIP testing is routinely offered to all pregnant couples in Thailand as part of a bundle of ANC tests. Couples may be subjected to further testing based on the results of these initial tests. If couples are both found to be potential thalassaemia carriers, the woman may be tested further using CBC & Hb typing. If her results are normal, couples can continue with routine ANC. If she has abnormal results, her partner is also tested using CBC & Hb typing. Abnormal results for the male partner warrants conducting DNA analysis for both partners after which PND is offered if the couple is found to be at high-risk of having a severe thalassaemia baby and they presented for ANC before 20 weeks' gestation. If severe thalassaemia is confirmed in the foetus after PND, an abortion is recommended to the couple. All steps in this screening process are voluntary, and couples may accept or decline further testing at any point. If the couple presented for their initial ANC visit late (after 20 weeks' gestation) and they both have abnormal results from CBC & Hb typing, the results of the DNA analysis conducted is documented for the benefit of future pregnancies, but they are not offered PND or abortion per the clinical guidelines in Thailand.
Pre-conception screening with targeted DNA analysis — Strategy 2	An alternative strategy may be targeted at Thai couples who have decided to conceive. Couples will both be tested initially using CBC & Hb typing, which have been identified as the primary tests for thalassaemia screening based on their increased sensitivity and specificity when used in combination. If both partners have abnormal results, further testing is done using DNA analysis and couples at high-risk of having severe thalassaemia babies are counselled to reconsider their decision to conceive.
Pre-conception screening with universal DNA analysis — Strategy 3	An alternative pre-conception screening strategy where couples who have decided to conceive are screened for thalassaemia using DNA analysis as the first-line test. For a genetic disease like thalassaemia, DNA analysis is the most definitive diagnostic test, although expensive and requires technical expertise. We explored this policy option for the Thai government in their consideration of implementing pre-conception screening.
Combination of pre- and post-conception screening — Strategy 4	This alternative presents a policy option for the use of a combination of strategies for thalassaemia prevention in Thailand as no single screening strategy has been identified as the best option for preventing thalassaemia globally or locally. Strategy 4 combines pre-conception screening using targeted DNA analysis with post-conception screening, where couples who have been identified to be at high-risk for severe thalassaemic babies but fail to reconsider their decision to conceive are offered PND during pregnancy. The subsequent steps are the same as in the post-conception screening strategy.
No screening	We evaluated all four strategies against a “no screening” arm where no thalassaemia screening in Thailand was assumed. This was done to ensure all relevant alternative strategies were included in the economic evaluation. Also, comparing the current screening strategy in Thailand to a “no screening” strategy was a first step in assessing its cost-effectiveness before considering other alternatives.
* The term “couple” as used in this study refers to male-female sexual relationships, both stable and unstable, which can result in reproduction	

Table S2: Model assumptions and justification

Model assumption	Explanation/justification
In the baseline analysis for post-conception screening, we assumed that 80% of pregnant Thai couples present early for ANC services	The exact value of this parameter has not been accurately captured and is explored further in the sensitivity analyses. This assumption also implies an ideal situation in which Thai couples initially present for ANC together. We acknowledge that this may not always be the case in reality and the absence of male partners during initial ANC visits may delay post-conception screening
The possible phenotypes of babies born to carrier partners are severe thalassaemia (T), carrier (C) or healthy, non-carrier babies (H)	These were simplified for ease of computation and interpretation of this work whilst noting that more than 60 genotypes for thalassaemic syndromes are possible, with varied phenotypes and clinical presentations
We assumed that the tests for thalassaemia screening were readily available across all regions in Thailand and their acceptability by couples was the only limitation	In reality, screening tests for thalassaemia may be unevenly distributed across Thailand and this has implications for the inferences made from the models. This assumption is discussed in the main text
We assumed that extensive genetic counselling was done at all points during the screening process by well-trained personnel and ignored the nuances of behavioural effects on the choices of couples	Inadequate genetic counselling may heavily influence whether couples accept or decline various procedures or tests. Personal, socio-cultural and religious factors also affect choices relating to conception. Modelling the effect of these behavioural factors on screening uptake was beyond the scope of this study and could be considered in future models for thalassaemia prevention in Thailand
In the baseline analyses for pre-conception screening, we assumed that 50% of couples at high-risk of having babies with severe thalassaemia will reconsider their decision to conceive	This data is currently unavailable as pre-conception screening is yet to be implemented in Thailand. It was explored in the sensitivity analyses to assess its effect on the model results. This is a major assumption which affects the potential cost-effectiveness of pre-conception screening in Thailand
We assumed all screening tests used in these strategies have high sensitivity and specificity. We excluded the possibilities of false positives and false negatives	Barring CBC, the various tests used in the screening strategies have been identified to have relatively high sensitivities and specificities. Their combined use in a multi-step approach, in our described strategies, also increases their diagnostic accuracy
We assumed the life-expectancy of individuals with homozygous β -thalassaemia and β /Hb E	Life-expectancy of thalassaemic patients depends on the type, severity of disease and resources available for treatment. A conservative life-expectancy of 30 years was chosen for this CEA. This implies that the CET used (which was calculated using this life-expectancy)

thalassaemia in Thailand to be 30 years	may be an underestimation. Hence, the screening strategies evaluated may be more cost-saving than suggested
We assumed the prevalence rates of β /Hb E thalassaemia in pregnant and non-pregnant women in Thailand to be the same — 16%, based on the prevalence studies we reviewed	The choice of screening strategies for haemoglobinopathies is highly dependent on disease prevalence. The prevalence rates we assumed, based on the literature we reviewed, appear biased towards Northern Thailand. This may have implications on the inferences drawn from our models. A formal systematic review may have addressed this by presenting more cohesive prevalence rates. We explored the uncertainty around these prevalence rates in scenario analyses

Table S3: Distributions, lower and upper bound values of parameters used in the sensitivity analyses

Number	Parameters	Included in deterministic sensitivity analysis (DSA)?	Base value (lower bound, upper bound)	Included in probabilistic sensitivity analysis (PSA)?	Distribution
1	CBC & Hb typing cost per person (THB)	Yes	390 (252.39, 557.08)	Yes	Gamma
2	CBC & Hb typing cost per couple (THB)	Yes	780 (504.77, 1114.16)	Yes	Gamma
3	DNA analysis cost (THB)	Yes	6000 (3882.88, 8570.42)	Yes	Gamma
4	PND cost (THB)	Yes	5500 (3559.31, 7856.22)	Yes	Gamma
5	Abortion cost (THB)	Yes	3000 (1941.44, 4285.21)	Yes	Gamma
6	Probability of one partner having trait	No [*]	-	Yes	Dirichlet
7	Probability of both partners having trait	No [*]	-	Yes	Dirichlet
8	Probability of both partners not having trait	No [*]	-	Yes	Dirichlet
9	Probability of couple agreeing to PND	Yes	0.58 (0.48, 0.67)	Yes	Beta
10	Probability of couple agreeing to abortion	Yes	0.67 (0.57, 0.76)	Yes	Beta
11	Probability of early presentation	Yes	0.80 (0.72, 0.87)	Yes	Beta
12	Probability of reconsidering decision to conceive	Yes	0.50 (0.40, 0.60)	Yes	Beta
13	Probability of man having trait	No [*]	-	Yes	Beta
14	Probability of woman having trait	No [*]	-	Yes	Beta

* These parameters were explored separately in a scenario analysis to evaluate how thalassaemia prevalence rates in Thailand affect the ICERs

Table S4: Assumptions for budget impact analysis and justification

Assumption	Explanation/ justification
The budget impact of the upfront costs of implementing the screening strategies was estimated over a one-year time horizon	We assumed one pregnancy per couple per year
We used the average number of births in Thailand over 3 years (2022, 2023, 2024) as an estimate of the eligible number of pregnant couples to be screened in the post-conception screening strategy	Data on the estimated number of pregnant couples who are screened annually for thalassaemia in Thailand is not widely available. The number of births was thus used as a surrogate, assuming all these births were from Thai couples who have access to the universal coverage scheme. We assumed 50% of these couples will require further thalassaemia testing based on the initial CBC & DCIP tests done as part of routine ANC. This was further explored in scenario analyses, by evaluating the annual budget impact of upfront costs, with 75%, 50% and 25% uptake of post-conception screening
Considering pre-conception screening is yet to be implemented in Thailand, we assumed it will be accessible to all Thai population in the reproductive age group. We assumed this age group to be 18 – 45 years for this evaluation	We divided the Thai population in the reproductive age group by 2 to estimate the number of couple units. Each male — female pair was assumed to be a couple unit, ignoring the possibilities of multiple partners and same sex partners. We then explored the impact of the upfront costs of implementing pre-conception screening among this reproductive population, on the annual budget, at 75%, 50% and 25% uptake rates
Only direct costs, as used in the CEA, were captured in the BIA	The BIA was done from the perspective of the budget holder i.e. the payer of the universal coverage scheme in Thailand (NHSO)

Table S1: Threshold analysis results

Strategy	Parameter	Threshold value for becoming (or ceasing to be) cost-saving
Strategy 1: post-conception	CBC & Hb typing cost per person (THB)	977.67
	Probability of couple agreeing to PND	0.27
	Probability of couple agreeing to abortion	0.33
	Probability of early presentation	0.38
Strategy 2: pre-conception, targeted DNA analysis	CBC & Hb typing cost per couple (THB)	2,032.13
	DNA analysis cost (THB)	54,911.40
	Probability of couple reconsidering decision to conceive	0.22
Strategy 3: post-conception, universal DNA analysis	DNA analysis cost (THB)	2,185.73
	Probability of couple reconsidering decision to conceive	Not defined
Strategy 4: combined strategy	DNA analysis cost (THB)	86,349.38
	Probability of couple reconsidering decision to conceive	Not defined
	CBC & Hb typing cost per couple	2,836.95

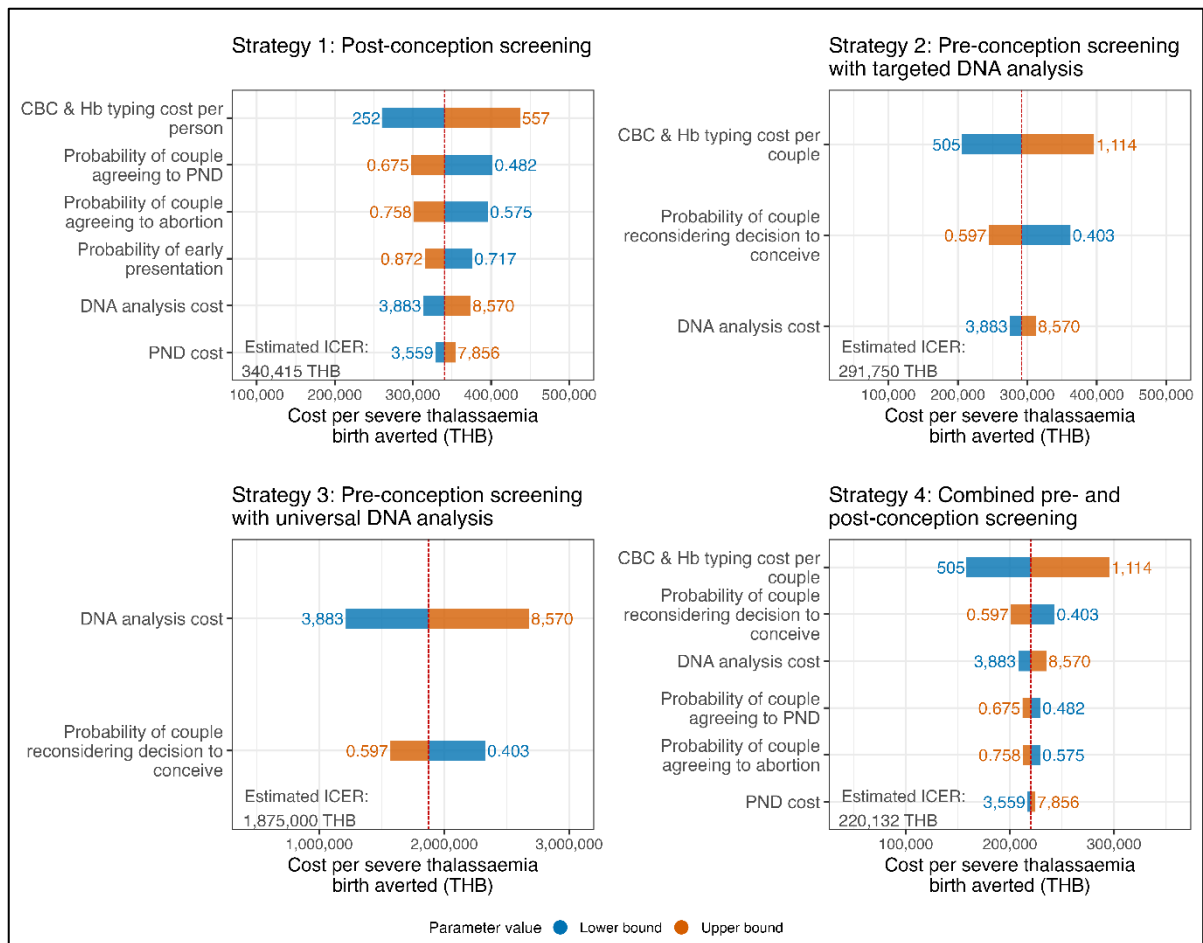


Figure S1: Univariate Deterministic Sensitivity Analyses (DSA) for the Cost-effectiveness Analyses of Thalassaemia Screening Strategies in Thailand. The tornado diagrams show how a change in parameter values between their lower and upper bounds affect the estimated ICERs. The vertical, red dashed line represents the estimated baseline ICER for each strategy. Horizontal bars represent the absolute change in ICER values as each parameter value varies between its lower (blue) and upper bounds (orange), ranked by the magnitude of the absolute change. Abbreviations: THB, Thai Baht; ICER, incremental cost-effectiveness ratio; PND, prenatal diagnosis; CBC, complete blood count; Hb, haemoglobin.

Table S6: Uncertainty intervals for the benefit-cost ratios (BCRs)

Strategy	BCR (using lower scenario for the lifetime management cost of severe thalassaemia)	BCR (using the average lifetime management cost of severe thalassaemia)	BCR (using higher scenario for the lifetime management cost of severe thalassaemia)
Strategy 1	0.80 [0.53 – 1.18]	2.01 [1.34 – 2.95]	2.79 [1.86 – 4.10]
Strategy 2	0.94 [0.60 – 1.48]	2.34 [1.51 – 3.69]	3.26 [2.09 – 5.14]
Strategy 3	0.15 [0.09 – 0.25]	0.36 [0.22 – 0.63]	0.51 [0.30 – 0.88]
Strategy 4	1.24 [0.83 – 1.86]	3.10 [2.09 – 4.65]	4.32 [2.90 – 6.46]