









Benefit Package of Universal Coverage Scheme for Hereditary Angioedema (HAE) Caused By C1 Esterase Inhibitor Deficiency (C1-INH)

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OBJECTIVES

Hereditary angioedema (HAE) is a rare and life-threatening genetic disease. This disease can lead to recurrent attacks of severe swelling occurring in areas such as the face, gastrointestinal tract, and larynx. These attacks can result in hospitalization or death. Thus, effective medications are necessary for treating the acute attacks. However, Thailand did not specify medication for treatment and prophylaxis. Therefore, effective strategies for universal health coverage for the management of HAE patients remain a challenge in Thailand. This review aims to determine the extent of research in the published literature that examine benefit package and strategies for universal health coverage for the management of HAE patients in Thailand.

METHODS

On 5th January 2022, Embase, Scopus, OpenGrey, PubMed, Science direct, Clinicaltrials.gov, and Cochrane were searched for literature published from inception. Studies evaluating the efficacy and safety of medication for treating acute attacks in HAE patients were included. Additionally, this study performed a survey of data from the drug company, experts in the field, and stakeholders. Our survey asked about the situation, cost, and health service systems for screening, diagnosis, treatment, and referring HAE patients in Thailand.

RESULTS

Of 4,834 articles identified, 28 studies were included. Twenty-four studies are original articles, one study was a systematic review and network meta-analysis, and three studies were cost-effectiveness analyses. From current trials, first-line therapy such as Plasma-derived C1 inhibitor, Recombinant C1 inhibitor Conestat alfa, Icatibant, and Ecallantide showed benefits over placebo. However, the study related to the full economic evaluation of first-line therapy in Thailand has not been provided.

Figure 1 **PRISMA Flow** 4,834 records identified through database searching in January 5, 2022 412 PubMed **1,207 EMBASE** 1,042 Science direct 1,890 SCOPUS 255 Cochrane 0 OpenGrey 28 Clinicaltrials.gov 1,511 duplicate records removed 3,323 records screened on the basis of title and abstract 3,166 excluded on title and abstract screening. 157 full-text articles assessed for eligibility. 129 excluded on full-text articles screening. 37 non-target population 21 not measure interesting outcome. 26 in vitro study 39 other study design 6 same participants 28 trials included in qualitative synthesis. 24 records are original article 1 record is systematic review and network meta-analysis 3 records are cost-effectiveness

Table 1 Estimated number of HAE cases in Thailand (2023-2027)

	2023	2024	2025	2026	2027
Thai population	67,903,079	68,785,819	69,680,035	70,585,876	71,503,492
Predicting the number of cumulative patients with HAE (1:50,000 of the population)	1,358	1,376	1,394	1,412	1,430
Patients who have been screened					
New suspected HAE patients screened with a C4 level (0.0001% of the population)	68	69	70	71	72
New cases with low C4 levels (35%)	24	24	25	25	25
New patients with confirmed HAE disease (88%)	21	21	22	22	22
Patients with confirmed HAE accumulated from the previous year	63	84	105	127	149
Total confirmed cases of HAE	84	105	127	149	171

Table 2 Budget Implications of HAE Diagnosis

Unit: Baht	Step with C1	Step with gene mutation	All at one				
Cost of diagnosis and confirmation							
1 st year (2023)	130,720	152,720	330,720				
2 nd year (2024)	130,720	152,720	330,720				
3 rd year (2025)	134,500	156,500	344,500				
4 th year (2026)	134,500	156,500	344,500				
5 th year (2027)	134,500	156,500	344,500				
The cost of transporting specimens							
1 st year (2023)	280,000	280,000	240,000				
2 nd year (2024)	280,000	280,000	240,000				
3 rd year (2025)	290,000	290,000	250,000				
4 th year (2026)	290,000	290,000	250,000				
5 th year (2027)	290,000	290,000	250,000				
Including expenses for both diagnosis and delivery of specimens							
1 st year (2023)	410,720	434,720	570,720				
2 nd year (2024)	410,720	434,720	570,720				
3 rd year (2025)	424,500	448,500	594,500				
4 th year (2026)	424,500	448,500	594,500				
5 th year (2027)	424,500	448,500	594,500				
Total 5 years	2,094,940	2,214,940	2,924,940				
Lowest average budget per person	13,780	13,780	23,780				
Maximum average budget per person	33,780	39,780	23,780				

Table 3 Budget Implications of HAE Treatment

Unit: Baht	Supportive treatment with Fresh Frozen Plasma	Treatment with Icatibant	Difference				
Budget burden on payer perspective							
1 st year (2023)	16,557,798	23,363,948	6,806,150				
2 nd year (2024)	20,745,696	29,191,645	8,445,949				
3 rd year (2025)	25,065,344	35,316,259	10,250,915				
4 th year (2026)	29,449,590	41,423,153	11,973,563				
5 th year (2027)	33,769,237	47,547,767	13,778,530				
Budget burden on patient perspective							
1 st year (2023)	807,900	452,100	-355,800				
2 nd year (2024)	1,011,000	564,900	-446,100				
3 rd year (2025)	1,222,200	683,400	-538,800				
4 th year (2026)	1,434,900	801,600	-633,300				
5 th year (2027)	1,646,100	920,100	-726,000				
Total budget							
1 st year (2023)	17,365,698	23,816,048	6,450,350				
2 nd year (2024)	21,756,696	29,756,545	7,999,849				
3 rd year (2025)	26,287,544	35,999,659	9,712,115				
4 th year (2026)	30,884,490	42,224,753	11,340,263				
5 th year (2027)	35,415,337	48,467,867	13,052,530				
Total 5 years	131,709,765	180,264,872	48,555,107				

CONCLUSIONS

HAE may affect the budget of Thailand if patients do not receive appropriate treatment. Thus, screening, diagnosis, and effective medication for treating acute attacks are essential measures to decrease the overall HAE-related healthcare cost. However, full economic evaluation studies of first-line therapy in Thailand are needed.