

Hemophilia in Indonesia: A Cost of Illness Analysis Using National Health Insurance Data (2022–2023)

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ABSTRACT

Hemophilia is one of the highest contributors to catastrophic costs in the National Health Insurance (NHI) program. This study aims to estimate the Cost of Illness (COI) of NHI participants with hemophilia from a payer and partial productivity loss perspective, using secondary data from BPJS Kesehatan to examine direct medical costs from the perspective of a third-party payer, and average hourly wages by province in Indonesia to assess patient productivity loss. The research method is a cross-sectional study using a quantitative design. The findings indicate that the average COI per participant was IDR 728,832,232. After applying sampling weights to estimate the national population, the total COI for hemophilia during 2022–2023 was IDR 3.02 trillion, with 99% attributable to direct medical costs. These findings indicate that although hemophilia affects a relatively small number of participants, it imposes a substantial financial burden on the NHI system. Significant differences in COI medians were observed across age, gender, length of stay, special drug usage, membership segmentation, ward class, hospital region, hospital type, treatment type, and severity. Hemophilia, as a high-cost, low-frequency condition, requires careful management to avoid further strain on NHI funding.

Keywords: BPJS Kesehatan; Cost of Illness; Hemophilia; National Health Insurance

INTRODUCTION

Hemophilia is a rare hereditary disease transmitted through the X chromosome that impairs blood clotting. Apart from affecting physical health, hemophilia also impacts the social and psychological quality of life of patients, who mostly rely on lifelong medical care (S. L. Chen, 2016; Coffin et al., 2023). In Indonesia, although it was estimated that there are 25,000 individuals with hemophilia, data from 2022 shows that only 3,155 cases were detected (WFH, 2023), highlighting a significant gap in case detection. Despite detection rates and the use of factor VIII therapy being far below international standards, the cost of hemophilia treatment remained high, particularly within the NHI system. In 2022, hemophilia medical costs amounted to IDR 650 million and increased substantially in the following year, reaching IDR 1.2 trillion in 2023 (DJSN, 2023). The rising claims for hemophilia services are attributed to changes in reimbursement policy, particularly the introduction of updated top-up tariffs for anti-hemophilic factor therapies under Minister of Health Regulation (PMK) No. 3 of 2023, replacing the previous tariff structure in PMK No. 52 of 2016. (Kemenkes, 2016, 2023; Pharadiba et al., 2024).

This pattern is consistent with the broader rare disease literature, which characterizes such conditions as high-cost and low-frequency, with high per-patient treatment costs despite

relatively small patient populations, posing challenges for cost predictability and budget management (Banks et al., 2021). To date, evidence on the economic burden of hemophilia in Indonesia remains limited, particularly from the perspective of the NHI payer using administrative claims data. Furthermore, previous studies primarily focused on clinical management or international cost estimates, with limited national-level analysis of cost components and associated patient characteristics within Indonesia. In this context, this study aims to estimate the economic burden of hemophilia through a cost-of-illness (COI) analysis from a payer and partial productivity-loss perspectives using administrative claims data.

METHODS

Research Design

This study employs a descriptive observational design with a cross-sectional approach. The research analyzes the economic burden of hemophilia from a payer and partial productivity loss perspectives using secondary data from BPJS Kesehatan to examine direct medical costs from the perspective of a third-party payer. Moreover, the productivity loss was estimated using average provincial wages multiplied by inpatient days, assuming the patient is employed or, in the case of children who are not yet employed, viewed from the perspective of the caretaker.

Population and Sample

This study utilized the official BPJS Kesehatan administrative claims sample dataset, which represents approximately 1% of the national population for the 2022–2023 period. The inclusion criteria were NHI participants with active membership status during the study period and at least one recorded claim with ICD-10 codes D66, D67, D68.0, or D68.1. As a result, total of 47 participants (1,804 visitations) met these criteria in the sample dataset. Records with incomplete cost information were excluded. All eligible cases within the administrative dataset were included (total sampling).

Data Collection Technique

The data were based on BPJS Kesehatan classification and coding systems. To estimate population-level costs, sampling weights provided in the BPJS Kesehatan dataset were applied using the weighting procedure in SPSS. Weighted frequencies were used to approximate the total number of hemophilia participants nationally ($n = 4,143$). Total cost estimates were then extrapolated by multiplying weighted participant counts by mean cost values. Disease severity was defined according to the Indonesian Case-Based Groups (INA-CBGs), with outpatient cases classified as severity level 0 and inpatient cases categorized as mild (severity I), moderate (severity II), or severe (severity III). Membership segmentation followed NHI participant categories, including Contribution Assistance Recipients (PBI), Non-Wage Recipient Workers (PBPU), and Wage Recipient Workers (PPU). Special drug usage referred to high-cost medicines reimbursed under BPJS Kesehatan special drug codes DD-06-II and DD-10-II, primarily coagulation factor concentrates used in hemophilia management.

Data Analysis

Data analysis was performed using SPSS for univariate and bivariate tests. Non-parametric tests were applied due to non-normal distribution and unequal variances. The Mann-Whitney U test was used for variables with two categories (e.g., age, gender, and special

drug usage). In comparison, the Kruskal-Wallis test was used for variables with more than two categories (e.g., membership segmentation, hospital region, hospital type, treatment type, and severity). Due to the limited sample size, multivariate regression analysis was not performed.

RESULTS AND DISCUSSION

There were 47 participants (sample) who met the inclusion criteria and extrapolated into 4,143 participants (population). All participants were active members of BPJS Kesehatan, with no deaths recorded during the study period.

Table 1. Characteristics of NHI Participants with Hemophilia 2022-2023

Variable	Unweighted		Weighted	
	n	%	n	%
Gender				
Male	41	87.2	3166	76.4
Female	6	12.8	977	23.6
Age				
< 18 years	26	55.3	2216	53.5
≥ 18 years	21	44.7	1927	46.5
Hospital Region				
Regional I	27	57.4	2599	62.7
Regional II	6	12.8	115	2.8
Regional III	1	2.1	12	0.3
Regional IV	7	14.9	598	14.4
Regional V	6	12.8	818	19.7
Membership Segmentation				
Non-Worker	1	2.1	3	0.1
PBI	14	29.8	1748	42.2
PBPU	13	27.7	681	16.4
PPU	19	40.4	1710	41.3
Hospital Type				
Class A Type/Equivalent	21	44.7	2003	48.3
Class B Type/Equivalent	16	34.0	1509	36.4
Class C Type/Equivalent	8	17.0	617	14.9
Class D Type/Equivalent	2	4.3	14	0.3
Treatment Type				
Only Outpatient	32	68.1	3104	74.9
Only Inpatient	3	6.4	27	0.6
Both Outpatient and Inpatient	12	25.5	1011	24.4
Severity				
Outpatient	43	2.4	3995	96.4
Mild (Inpatient Severity I)	3	0.2	135	3.3
Moderate (Inpatient Severity II)	1	0.1	12	0.3
Special Drugs Usage				
Non-Special Drugs	18	38.3	1779	43.0
Special Drugs	29	61.7	2363	57.0

Based on Table 1, the study population was predominantly male (76.4%), with slightly more than half of the patients aged under 18 years (53.5%). Most patients were treated in Regional I hospitals (62.7%), followed by Regional V (19.7%) and Regional IV (14.4%). In

terms of membership segmentation, PBI (42.2%) and PPU (41.3%) were the most common categories. Nearly half of the patients received care in Class A hospitals (48.3%), while the majority were managed as outpatients only (74.9%). Most cases were classified as outpatient severity (96.4%), and more than half of the patients received special drugs (57.0%).

Table 2. Treatment Cost per Episode Based on Treatment Types

Treatment	Mean (IDR)	Median (IDR)	SD	Min-Max (IDR)	Total Cost (IDR)
Outpatient	18,602,803	20,988,000	8,519,480	138,100– 27,191,300	31,085,283,000
Inpatient	22,771,332	29,675,400	9,478,882	2,369,600–36,874,600	2,891,959,200

Table 3. COI of NHI Participants sample data with Hemophilia 2022-2023

Cost Type	Mean (IDR)	Median (IDR)	SD	Min-Max (IDR)	Total Cost (IDR)
Treatment Cost (Direct cost)	722,920,047	486,217,700	850,078,037	176,200 – 3,422,847,700	33,977,242,200
Productivity Loss (Indirect cost)	5,912,186	4,502,340	6,329,173	103,940 – 25,571,400	277,872,724
COI	728,832,232	489,024,080	855,895,321	290,740 – 3,448,419,100	34,255,114,924

Table 3 presents the components of the COI based on the study sample (n = 47). The results indicate that total costs were largely driven by direct medical expenditures of IDR 33,977,242,200, while productivity loss accounted for only a small share (IDR 277,872,724).

Table 4. COI Based on the Number of Samples and Population

Cost Types	Total Cost (IDR)	
	Sample (n = 47)	Population (n = 4143)
Treatment Cost	33,977,242,200	2,995,057,754,721
Productivity-Loss	277,872,724	24,494,186,598
COI	34,255,114,924	3,019,551,937,176

After extrapolation, the total COI for the hemophilia population was estimated at IDR 3.02 trillion, of which direct medical costs accounted for approximately 99%, while productivity losses contributed around 1%.

Table 5. Relationship between characteristics and COI (n=4,143)

Variable	n	%	Median (IDR)	p-value
Gender				
Male	3166	76.4	812,122,400	<0.001
Female	977	23.6	537,128	
Age				
< 18 years	2217	53.5	296,629,176	<0.001

Variable	n	%	Median (IDR)	p-value
≥ 18 years	1928	46.5	946,418,040	
Hospital Region				
Regional I	2599	62.7	591,064,612	<0.001
Regional II	116	2.8	296,629,176	
Regional III	12	0.3	5,510,764	
Regional IV	599	14.4	7,363,688	
Regional V	819	19.7	537,128	
Membership Segmentation				
Non-Worker	3	0.1	1,056,156	<0.001
PBI APBN or APBD	1747	42.2	489,024,080	
PBPU	682	16.4	1,062,713,516	
4PU	1713	41.3	4,956,440	
Hospital Type				
Class A Type/Equivalent	2005	48.3	875,862,528	<0.001
Class B Type/Equivalent	1509	36.4	489,024,080	
Class C Type/Equivalent	618	14.9	4,956,440	
Class D Type/Equivalent	13	0.3	5,510,764	
Treatment Type				
Only Outpatient	3107	74.9	296,629,176	<0.001
Only Inpatient	27	0.6	5,510,764	
Both Outpatient and Inpatient	1011	24.4	812,122,400	
Severity				
Outpatient	3995	96.4	296,629,176	<0.001
Mild (Inpatient Severity I)	135	3.3	1,181,842,808	
Moderate (Inpatient Severity II)	12	0.3	5,510,764	
Special Drugs Usage				
Non-Special Drugs	1780	43.0	537,128	<0.001
Special Drugs	2365	57.0	1,009,893,640	

Gender

This study finds that most NHI participants with hemophilia are male (76.4%), aligning with previous research showing that Hemophilia A occurs in approximately 1 in every 5,000 males, with males predominating across all severity levels of hemophilia (Coffin et al., 2023; Salen & Babiker, 2023). In the United States, females contributed only 18-20% to cases of mild hemophilia and less than 3% to more severe cases (van Galen et al., 2021). This pattern may also reflect underdiagnosis and undertreatment of hemophilia among females, which has been increasingly recognized in recent literature (d'Oiron et al., 2021). There is a significant difference in the COI between males and females, in accordance with a finding that showed that the cost of treating Hemophilia A in males is much higher than in females, both in commercial schemes and Medicaid. Despite female patients experiencing emergency room visits and inpatient care for bleeding, only a few received FVIII therapy, which was one of the reasons why male patients incur significantly higher medical costs for hemophilia (Batt et al., 2023).

Age

The majority of hemophilia patients are children below 18 years old (53.5%), which aligns with data from the World Federation of Hemophilia, showing that 56% of Hemophilia A cases and 65% of Hemophilia B cases in Indonesia occur in the 0-18 age group (WFH, 2023). Despite the higher proportion of child patients, the COI for adult patients is significantly higher. The findings are consistent with more extensive research demonstrating that the cost of hemophilia care typically rises with age, with adults paying higher total direct and indirect costs than paediatric patients as a result of more intensive treatment and long-term healthcare use. From the payer perspective, annual healthcare expenditures are also consistently higher among adults, reflecting more frequent treatment and long-term disease management needs. Primarily due to intensive prophylactic therapy, adolescents and young adults (15–30 years old) are frequently the most expensive group within this pattern. Evidence from Portugal similarly shows higher annual costs in paediatric hemophilia A compared to adults and peak costs in the 21–30 age group for hemophilia B. In older adults (31–50 years), although treatment utilization may decline, costs can increase again up to fivefold due to inhibitor-related complications. (Calvo et al., 2023; S. L. Chen, 2016; Y. Chen et al., 2023; da Silva Etges et al., 2024). As BPJS claims data do not capture complication status, the relationship between age and inhibitor-related costs could not be directly assessed in this study. Although the average cost per episode was higher among children, the substantially higher overall COI observed in adults suggests that total direct and indirect costs remain greater in this group. This is primarily driven by higher treatment frequency among adult patients, resulting in greater cumulative per-patient costs and, consequently, a higher overall COI.

Hospital Regional

In terms of regional distribution, the majority of hemophilia patients are from Regional I (62.7%), which mainly includes urban areas. A study found that hemophilia patients primarily reside in metropolitan areas (John et al., 2018; Liedl, 2022). Additionally, the highest median COI is found in Regional I, while the lowest is found in Regional V, despite Regional V having the second-largest proportion of participants. This finding aligns with a study comparing treatment costs for Thalassemia and Hemophilia in Jakarta and NTT, which noted that Jakarta (representing Regional I) had substantially higher claim costs compared to NTT (representing Regional V) (Heniwati & Thabrany, 2016). This pattern may reflect regional tariff structures under the INA-CBGs system, where higher-class hospitals are more concentrated in urban regions. Consequently, claim costs in Jakarta remain high due to the abundance of Class A and B hospitals, while in NTT, costs are lower due to the predominance of Class C and D hospitals, which have lower tariffs (Kemenkes, 2024). This will be further discussed in the following section.

Hospital Type

This study finds that most participants mainly seek treatment at Class A hospitals (48.3%), which aligns with the study in China reporting that 66.3% of hemophilia patients are treated in advanced hospitals (tertiary hospitals) (Huang et al., 2022). Median COI was highest in Class A hospitals, aligning with previous findings that higher-level hospitals are associated with higher treatment costs (Huang et al., 2022). Previous studies have described how disparities in hospital classification across regions may lead to unequal reimbursement levels under the INA-CBGs system, contributing to fiscal imbalances between developed and

underdeveloped regions (Heniwati & Thabrany, 2016). The pattern observed in this study is consistent with this earlier evidence.

Membership Segmentation

In the current research, most participants belonged to the PBI (Premium Assistance Recipients) segment (42.2%). Although the PBPU (non-wage recipient worker) segment accounted for the smallest proportion of participants, it had the highest median COI, followed by PBI participants. This pattern is further contextualized by the low active participation rate among PBPU members (51.91%), who account for approximately 99% of inactive BPJS participants (DJSN, 2023). Premium payment deferral is commonly driven by income instability, limited flexibility in the payment system, unaffordable contribution levels, and the prioritization of other essential household expenditures over health insurance (Dartanto et al., 2020). Similar challenges related to contribution compliance among informal workers have been reported in the universal health coverage systems in Thailand, raising concerns about long-term financial sustainability (Lim et al., 2023). Moreover, given that PBI participants are fully financed by government subsidies and represent the largest patient volume, their substantial cost share reflects a significant expenditure component within the NHI financing structure.

Treatment Type

According to this study, most participants utilize only outpatient care (74.9%), in line with the World Federation of Hemophilia, which reported that only 10% of hemophilia patients and only 13% patients with severe hemophilia require inpatient care (World Federation of Hemophilia, 2021). The episodic treatment cost for inpatient care is higher than that for outpatient care, despite being significantly lower in total (see Table 2), which is consistent with the findings of previous studies (S. L. Chen, 2016; da Silva Etges et al., 2024). Moreover, the COI pattern shows that the frequency of visits and treatment duration are associated with higher total COI, with participants utilizing both outpatient and inpatient care recording the highest costs due to their longer cumulative treatment durations.

Severity

This study finds hemophilia visits primarily fall under severity level 0 or outpatient (96.4%). While the composition of outpatient visits is the highest, mild severity has the highest median COI. This shows a contrast with a finding in Spain and US that treatment costs were higher for patients with higher severity than in moderate patients, as well as higher in Hemophilia B than in Hemophilia A, especially those with inhibitor complications (Buckner et al., 2021; Peral et al., 2024; Rocha et al., 2015). This difference is due to the higher number of inpatient visits among patients with mild severity, accumulating higher total costs despite not being as severe as those with complications. However, severity in this study was based on INA-CBGs administrative classification, which may not fully reflect clinical severity.

Special Drugs Usage

The majority of participants in this study receive treatment with special drugs (57%), which highlights the importance of coagulation factor concentrates in hemophilia management. In this study, participants who received special drugs had a significantly higher median COI compared with those who did not ($p < 0.001$). Despite factor VIII consumption in Indonesia

remaining low (0.28 IU/capita) compared with the international standard of 1 IU/capita, treatment still relies predominantly on plasma-derived products (87%), indicating limited access to recombinant therapies (WFH, 2023). Consistent with previous studies, coagulation factors accounted for the largest share of direct medical costs, contributing more than 70% of total treatment costs (Huang et al., 2022).

Although the cost-effectiveness ratio of preventive care is relatively higher than that of episodic treatment, which may be unaffordable in many low and middle-income countries, the costs associated with restoring musculoskeletal health are also high when considering postoperative complications and the economic burden (Serban et al., 2021; Rodriguez-Merchan et al., 2022). However, when the costs of Coagulation Factor Replacement Therapy (CFRT) are excluded, prophylaxis is associated with lower direct medical and non-medical costs, lower indirect costs, and lower total costs, as well as higher health-related quality of life (HRQoL) scores compared with episodic treatment (Rodriguez-Santana et al., 2022b, 2022a). Since 2021, national guidelines have recommended a gradual shift from on-demand to prophylactic therapy, but implementation has been uneven across regions due to funding and availability constraints (Angchaisuksiri et al., 2024). While earlier studies suggest that prophylactic treatment may reduce long-term complications and costs in resource-limited settings (Ibrahim et al., 2023; Kemenkes, 2021; Maimon et al., 2024; Seth et al., 2023), this evidence is presented for contextual comparison. The present study did not assess treatment regimens or clinical outcomes, the observed cost patterns thus reflect current treatment practices under the NHI scheme.

This study has several limitations that should be considered when interpreting the findings. First, the cross-sectional design and use of secondary data restrict the ability to establish causal relationships and assess changes in hemophilia treatment costs over time. Furthermore, the analysis was based on a relatively small sample ($n = 47$), and the extrapolation of average costs to the national hemophilia population may introduce uncertainty. In addition, the use of administrative claims data from BPJS Kesehatan may not fully capture clinical severity and treatment complexity, which could affect the accuracy of cost estimates. Moreover, the estimation of indirect costs was limited to productivity loss, calculated using average provincial wages and inpatient days, which does not account for individual employment status, age, informal sector work, or economic adjustments such as inflation and therefore may underestimate the true productivity loss associated with hemophilia. Other relevant cost components, including transportation, caregiver time, and out-of-pocket expenditures, were not included due to data limitations.

CONCLUSION

This study shows that the COI of hemophilia under the NHI scheme varies significantly across patient, clinical, and service-related characteristics, including age, gender, special drug usage, membership segmentation, hospital region and type, treatment type, and disease severity. The total estimated COI for hemophilia under the NHI scheme during 2022–2023 reached approximately IDR 3.02 trillion, with direct medical costs accounting for nearly all expenditures. Cost variation across age, hospital type, treatment pattern, and special drug usage indicates that resource utilization is highly concentrated among specific patient groups. Given its high per-patient cost despite relatively low prevalence, hemophilia represents a significant financial burden for the NHI system. Furthermore, these findings emphasize the importance of more targeted resource allocation, such as severity-based treatment and the

evaluation of prophylactic treatment strategies, in managing long-term costs. Finally, future studies should incorporate longitudinal designs and include broader indirect and non-medical costs to provide a more comprehensive economic evaluation.

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