# **Economic Burden of Hairy Cell Leukemia: A Systematic Review**

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# **Background and Objectives**

- Hairy Cell Leukemia (HCL) is a rare, slow-growing B-cell malignancy affecting primarily adults aged 50-60 years.<sup>1</sup> It accounts for approximately 2% of adult leukemias, with an incidence of 0.3-0.4 cases per 100,000 persons annually.<sup>2</sup>
- The disease often leads to significant disability, requiring extensive caregiver support and impacting the patient's quality of life. This is especially true given the need for prolonged treatment and frequent hospitalizations.
- First-line therapies, including purine analogs (cladribine), and newer therapeutic options like BRAF inhibitors, have been developed to manage the disease.<sup>3,4</sup>
- However, these treatments, along with the need for extended hospital stays and long-term followup care to monitor for relapse, create substantial challenges for the healthcare system.<sup>5,6</sup>
- In lower- and middle-income countries, where access to advanced treatments may be limited, the burden of HCL can be even higher.<sup>2</sup>
- This systematic literature review (SLR) aims to evaluate the economic impact of HCL, synthesizing available data on direct healthcare costs, treatment-related expenses, and the cost-effectiveness of various therapeutic approaches.

# Methods

- sources.
- and minimize bias.
- trends.

#### Fig 1: PRISMA Flowchart



Abbreviations CER: cost-effectiveness ratio; HCL: hairy cell leukemia; MA: meta-analysis; NMA: network meta-analysis; PICOS: population, intervention, comparator, outcome, study design/setting; PRISMA: Preferred Reporting Items for Systematic reviews and Meta-Analyses; RUR: Russian Ruble; SLR: systematic literature review; vs: versus; USA: United States of America; USD: United States dollar

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References

• A comprehensive literature search was conducted in the EMBASE, MEDLINE, and Cochrane CENTRAL were searched from inception to November 2024 using OVID database and supplemented with hand-searching of grey literature

• Articles were selected based on prespecified PICOS criteria and reviews aligned with PRISMA statement<sup>7</sup> as well as the Cochrane guidelines.<sup>8</sup>

 Title, abstract, and full-text screening were conducted by 2 independent investigators to ensure consistency

 Data on study characteristics, economic evaluation methods, and outcomes were extracted.

• A qualitative synthesis summarized findings on the economic burden of HCL treatments, comparing cost data and study outcomes to identify

d	Records removed before screening (n=13)
n=434)	
ed	Records excluded (n=389)
for	
ed for	Reports excluded: 26 Population (n=6) Study Design (n=7) On topic SLR/MA/NMA (n=3) Outcome (n=10)
N	

## **PICOS Criteria**

- The searches captured studies in all languages; however, non-English citations were excluded during the screening process.
- Inclusion Criteria: Studies involving adult patients ( $\geq$ 18 years) with HCL, any HCL treatment (single-agent, combination, or supportive care), economic evaluations, and outcomes related to economic burden (costs, direct and indirect).

## Results

### **Study Characteristics**

- Out of 434 records identified, 6 studies were included after screening (Fig 1). Among these, 4 studies were retrospective analyses, while 2 were economic evaluations.
- The studies were conducted in the USA (n=2), Russia (n=1), India (n=1), both the USA and France (n=1), and 1 study had an unknown location.

# Discussion

Conclusion



Cost (\$, USD) • The findings highlight the substantial economic burden of HCL treatment, particularly for patients requiring inpatient care or experiencing complications like myelosuppression and opportunistic infections. Myelosuppression significantly increases costs, emphasizing the need for treatment strategies that minimize hematologic toxicity. Given the high inpatient costs, strategies that reduce hospitalizations—such as early intervention and optimized outpatient care—could help alleviate the financial burden. • The cost-effectiveness of cladribine combined with interferon alfa suggests that combination therapies may provide better financial and clinical outcomes. IFN-alpha-2b demonstrated notable cost savings over chlorambucil, reinforcing its role as a more sustainable treatment option. However, affordability remains a challenge, particularly in low-income settings where out-of-pocket expenses make splenectomy a more accessible but clinically inferior option. These insights underscore the need for cost-conscious treatment approaches that balance efficacy with affordability while minimizing complications that drive up overall healthcare expenditures.

#### The findings underscore the significant economic burden of HCL, with inpatient care and chemotherapy being the primary cost drivers. The need for more affordable and cost-effective therapeutic alternatives is critical to alleviating the financial burden on both healthcare systems and patients.



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