

A Systematic Review of Healthcare Resource Utilization and Direct Medical Costs in Hereditary Angioedema

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Background

- Hereditary angioedema (HAE) is a rare, genetic disease characterized by debilitating swelling episodes in various parts of the body¹
- HAE is associated with substantial and multifaceted burden for patients, caregivers, health systems²
- While previous literature has documented the economic impact of HAE on direct costs, productivity, and other indirect costs, no systematic literature reviews (SLRs) to date on this topic have been published
- As the HAE treatment landscape evolves, there is a need to better understand the economic value of HAE treatments, to inform decisions as well as to improve disease management, patient satisfaction, and quality of life

Methods

- We conducted an SLR on economic outcomes, including healthcare resource utilization (HRU) and costs, among patients with HAE, including those using prophylactic and/or on-demand HAE therapies, in accordance with PRISMA guidelines
- Searches were conducted in PubMed, Embase, and Google Scholar
- Article inclusion was limited to English peer-reviewed and grey literature published between January 1, 2007 and July 1, 2022
- Two independent reviewers assessed literature eligibility and abstracted data
- We adjusted all costs for inflation to 2022 USD

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Results

Table 1. SLR Study and Sample Characteristics

Characteristic	n (%)
Total Number of Publications	66 (100)
Sample Size, number of patients	
Minimum	1
Maximum	737
Publication Type	
Peer-Reviewed	39 (59.1)
Conference Proceeding	14 (21.2)
Economic Report	13 (19.7)
Study Design	
Observational	42 (63.6)
Economic Analysis/Modeling	23 (34.8)
Randomized Controlled Trial	1 (1.5)
Study Location	
United States	28 (42.4)
Europe	25 (37.9)
Other	13 (19.7)
HAE Treatment Type, Studies*	
On-demand treatment	20
Prophylactic Treatment	15

*32 studies assessed an HAE therapy; some assessed both on-demand and prophylactic treatments

- We identified 66 studies. The majority of studies were observational (64%) and peer-reviewed (59%), and the most common study location was the United States (42%) (**Table 1**)
- Among 53 peer-reviewed and congress proceedings, 79% assessed HRU and direct costs in HAE
- Findings in the HAE publications revealed that total annual direct medical costs reached up to \$950,361 per patient, with medication accounting for the majority total direct costs (up to 85%, or \$808,322)³ (**Figure 1**)
- Prophylaxis treatment was found to cost 2.42 times that of on-demand treatment only^{3,4}

Figure 1. Annual Medical Costs and Drivers Associated with HAE^{3,5}

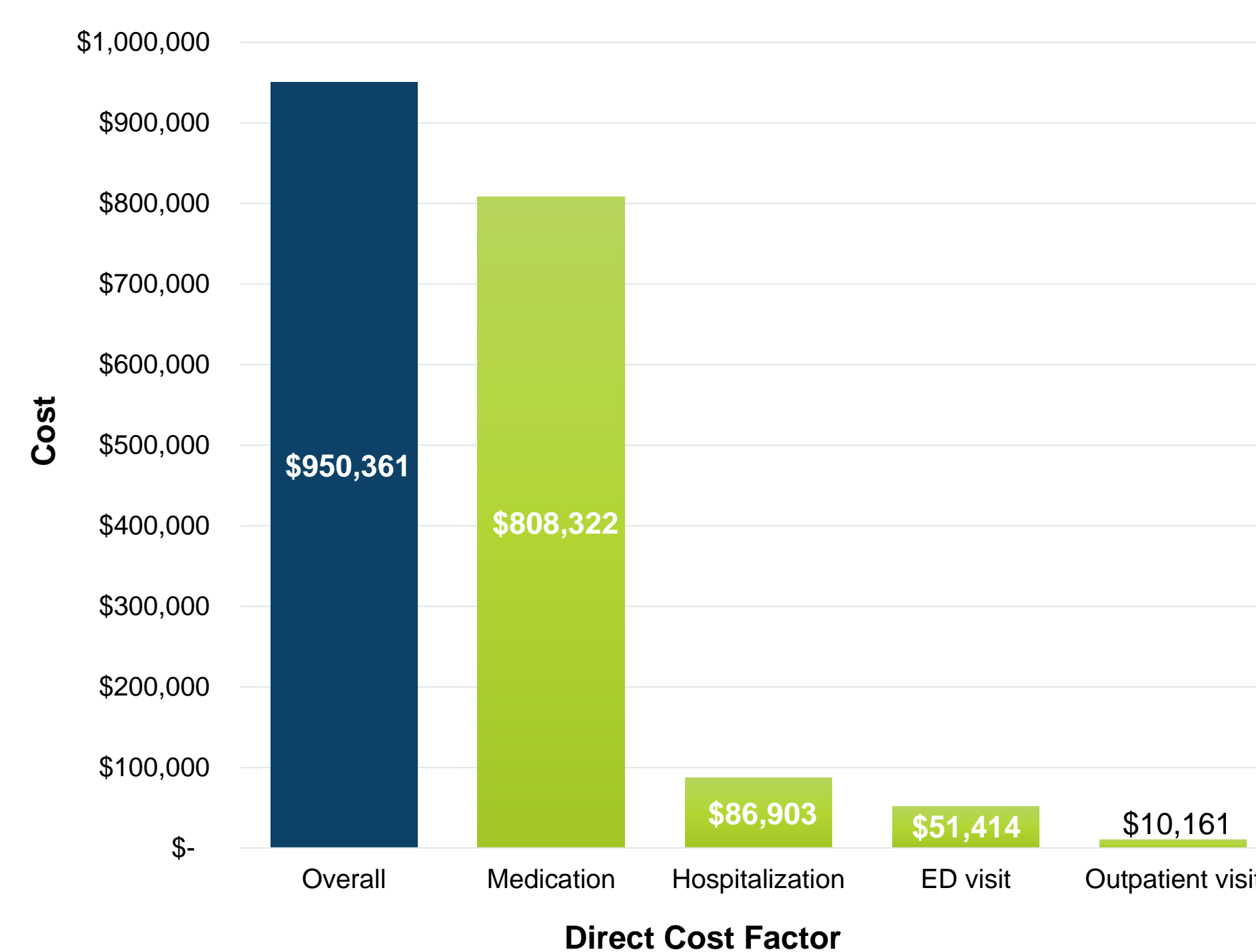
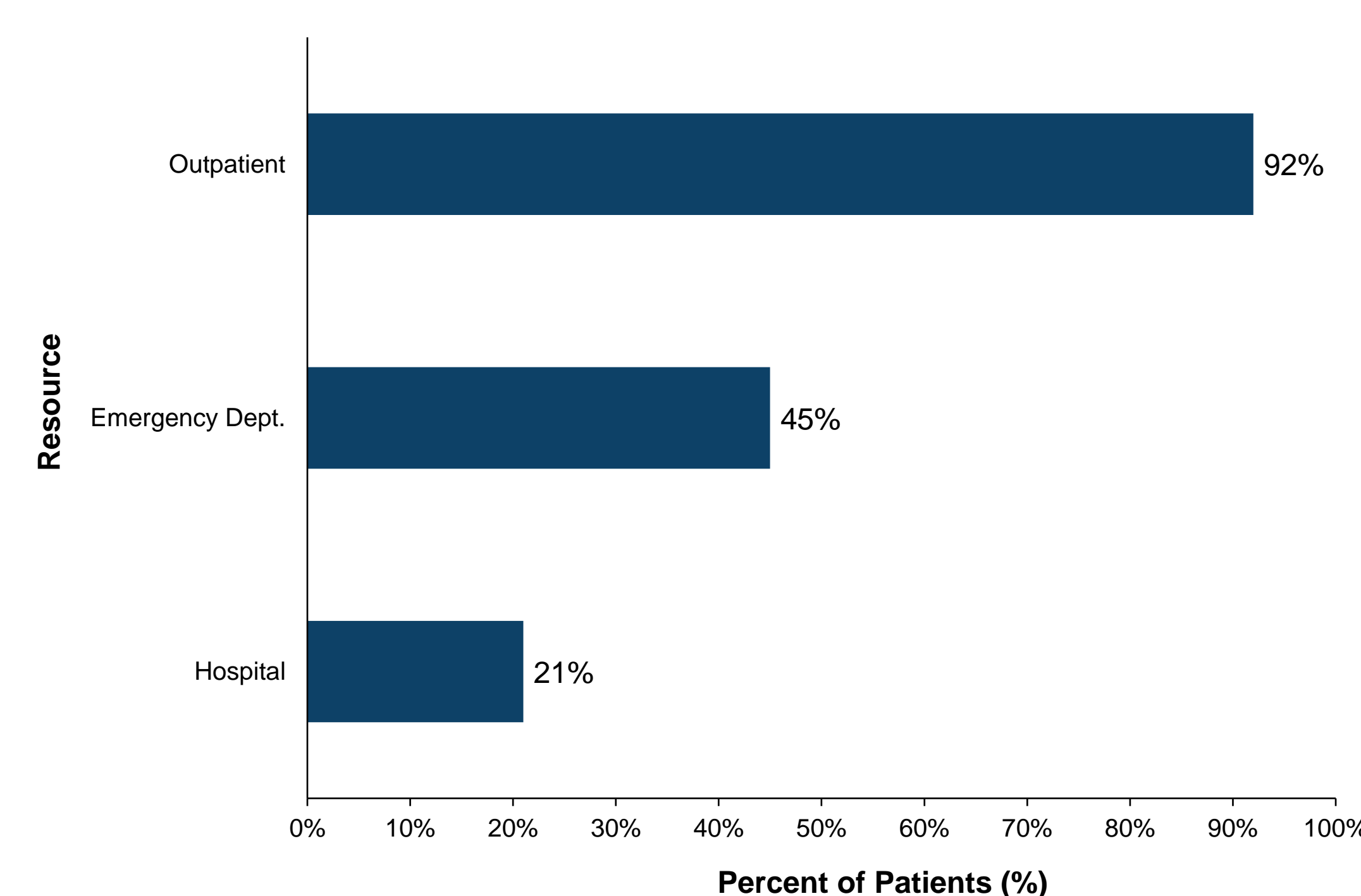
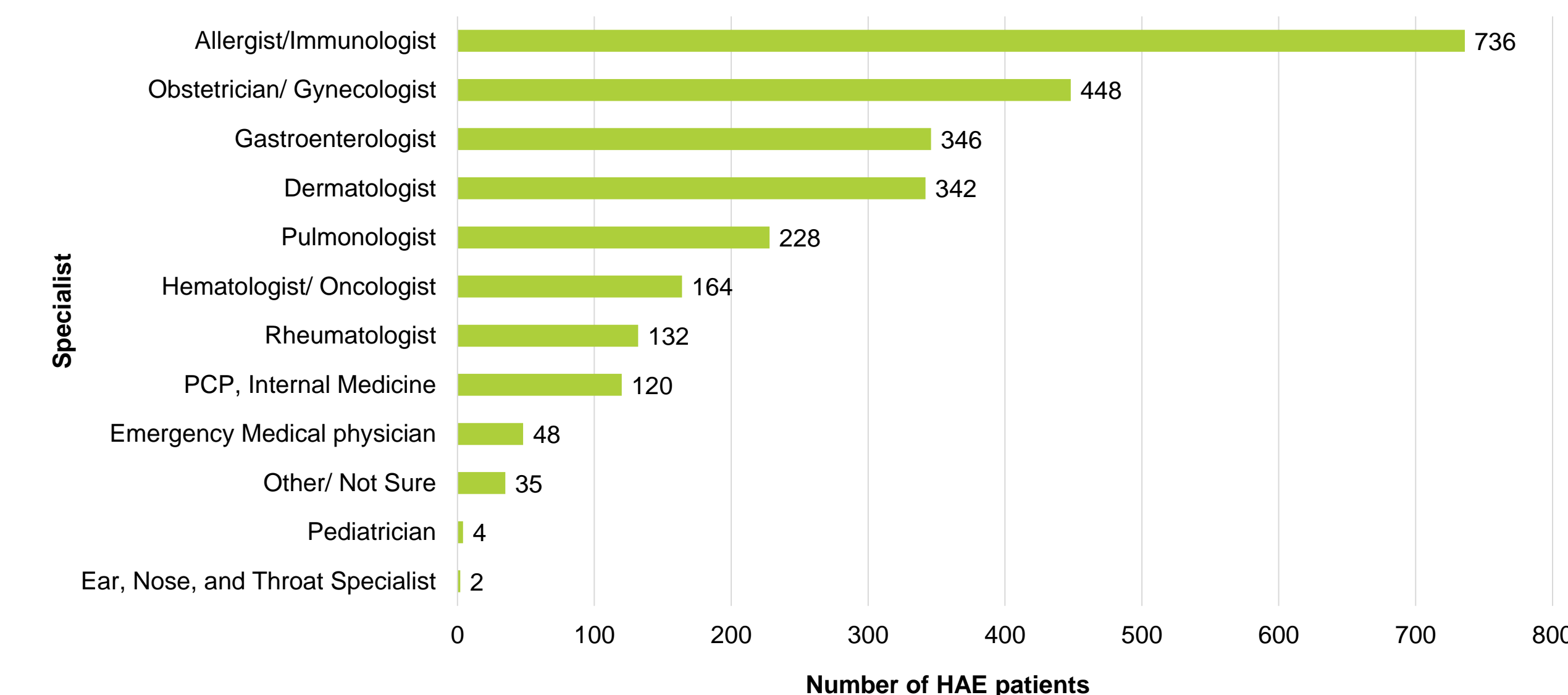


Figure 2. Percent of Patients Requiring at Least 1 Resource Visit Per Year^{6,7,8}



- For HRU outcomes, 92% of HAE patients using prophylactic and/or on-demand therapy required at least one outpatient visit,⁶ 45% required at least one emergency department (ED) visit,⁷ and 21% required at least one inpatient stay annually⁸ (**Figure 2**)
- Patients required between 2.7 – 3.7 outpatient visits,⁹ 1.94 ED visits, and 1.04 inpatient visits annually¹⁰ with a length of stay (LOS) per admission of up to 8 days¹¹
- Most HAE patients required specialist office visits, where the most common type of medical profession seen was an allergist/immunologist (28.3%)^{5,10} (**Figure 3**)

Figure 3. Medical Professionals Typically Seen by Patients with HAE^{4,9}



Conclusion

- This SLR found that, despite the emergence of new HAE therapies in the past decade, individuals with HAE still experience high HRU, regardless of using prophylaxis
- We found direct costs to be largely driven by medication costs, with prophylactic therapies costing more than twice that of on-demand therapies
- As the HAE treatment landscape evolves, there is opportunity to introduce patient-centered treatment options that improve disease control, thereby minimizing HRU and costs, as well as enhance patient satisfaction and quality of life

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