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Utilization of Healthcare Resources by Vascular Anomaly Patients: An Assessment of Healthcare Burden by Lesion Complexity

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Background: Vascular anomalies (VAs) are heterogeneous lesions. Symptoms vary widely by lesion type and complexity. VA patients often require life-long interdisciplinary care; however, there is a paucity of data on the healthcare utilization of VA patients, and their burden on the healthcare system remains largely unquantified. We hypothesize that healthcare utilization by complex lymphatic malformation (LM) and venous malformation (VM) patients will be significantly higher compared with simple LM and VM patients.

Methods: A retrospective, longitudinal study was performed of LM/VM patients seen through multidisciplinary VA clinics between January 1, 2019 and December 31, 2020. Data were collected from each patient's first presentation through December 31, 2021 and included number of office visits, imaging studies, specialists involved, procedures, hospitalization data, and approximate costs, normalized to per year utilization. Patients were divided into "simple" and "complex" LMs/VMs. Involvement of the airway, more than one anatomic area, and/or complex lymphatic anomalies were defined as "complex."

Results: In total, 28 simple and 29 complex LM patients and 51 simple and 18 complex VM patients were identified. Complex LM and VM patients had significantly higher numbers of imaging studies, specialists involved, procedures and hospitalizations, and costs incurred. Complex LM patients also had significantly higher per year office visits.

Conclusions: VA care is chronic and costly, especially for complex LM/VM patients. LM/VM complexity was a predictor for increased inpatient and outpatient healthcare utilization and higher costs. Better awareness of the healthcare utilization trends of LM/VM patients will allow for improved counseling for these patients regarding prognosis and expectations. *(Plast Reconstr Surg Glob Open 2023; 11:e5348; doi: 10.1097/GOX.0000000000005348; Published online 16 October 2023.)*

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INTRODUCTION

Vascular anomalies (VAs) are congenital lesions of the vascular system that may affect the capillaries, arteries, veins, lymphatics, or a combination of different vessel types. VAs include vascular malformations, which arise due to abnormalities in vasculogenesis, and vascular tumors, which are lesions with hyperplastic endothelium. Vascular malformations are classified according to their flow characteristics and involved vessel subtypes. Lymphatic malformations (LMs) and venous malformations (VMs) are among the most prevalent slow-flow vascular malformations, affecting 0.025%–1.0% of the population.[1,2] LMs and VMs have diverse clinical presentations, and symptoms may vary depending on the lesion subtype, involved tissues, and affected anatomic areas. LMs/VMs can be severe burdens with high morbidity and mortality, and affected

Disclosure statements are at the end of this article, following the correspondence information.

individuals often require life-long interdisciplinary care. Morbidities commonly seen in affected patients include pain, functional impairment, infection, and hematologic abnormalities such as hemorrhage and painful coagulopathy.[2–7] Patients with complex LMs/VMs may have additional severe complications such as airway obstruction, pleural effusions, bony destruction, protein-losing enteropathy, and malnutrition.[6,8–10] LM/VM patients are frequently managed by multidisciplinary teams that include dermatologists, hematologists, interventional radiologists, plastic surgeons, orthopedic surgeons, otolaryngologists, pediatric general surgeons, geneticists, and social workers. Treatment traditionally consists of surgical debulking, sclerotherapy, and, more recently, pharmacotherapies. These interventions are typically not curative, recurrence is common, and patients often require multiple procedures and hospitalizations.[11–15] Additionally, long-term pharmacotherapeutics have potential side effects that require regular monitoring, posing an additional burden to patients.[16]

Despite a high need for chronic specialized care, there are little data on the healthcare utilization or burden of LM/VM patients. A 2017 study analyzing resource utilization for pediatric LM patients found that complications from LMs were responsible for an average of 1619 hospitalizations per year in the United States, and that the average cost of each admission in 2009 was $46,184 ($70 million per year).[17] These estimates do not account for productivity losses for patients and their caretakers and likely do not reflect the current costs of hospitalization. Kim et al found on retrospective review of 7485 pediatric inpatients with VAs [hemangiomas, arteriovenous malformations (AVMs), and LMs] that AVM patients had higher average costs, number of computed tomography scans, and average hospital length of stay (LOS).[18] This study was limited by the authors' inability to specifically identify the factors driving higher utilization among AVM patients, though the authors hypothesized that a greater need for high-acuity care and more complex procedures may be contributing factors.

To our knowledge, an expanded analysis of the inpatient and outpatient healthcare utilization of patients with LM/VMs has not been conducted. Furthermore, utilization patterns according to lesion level of complexity have not been studied and may help to predict the heterogeneity of healthcare needs of VA patients. We hypothesize that patients with complex LMs and VMs will utilize significantly higher healthcare resources compared with those with simple LMs and VMs.

Takeaways

Question: What are the annual costs incurred by slow-flow vascular anomaly (VA) patients, and how do utilization and costs differ between patients with complex lymphatic (LMs) and/or venous malformations (VMs) and simple LMs/VMs?

Findings: Complex LM and VM patients had significantly higher numbers of imaging studies, specialists involved, procedures and hospitalizations, and costs incurred relative to patients with simple LMs/VMs.

Meaning: VA patients have significant healthcare needs, and VA complexity predicts increased inpatient and outpatient healthcare utilization; it will inform third-party payors of expected utilization and costs.

PATIENTS AND METHODS

An institutional review board–approved (IRB AAAT7002) retrospective, longitudinal study was performed of congenital LM and VM patients seen through the university faculty practice of one surgeon (J.K.W.) and the clinics of the multidisciplinary VA program within a single institution. Patients who were seen at least once between January 1, 2019 and December 31, 2020 were included. The electronic medical records of all study patients were reviewed, beginning with the patient's initial visit to our institution through December 31, 2021. Data were collected on healthcare utilization, including number of office visits, imaging studies, specialists involved, procedures, hospitalizations, hospital days, and average hospital LOS, normalized to per year utilization for each patient. Emergency department visits were classified as office visits if patients were not admitted and as hospitalizations if patients were admitted. Using publicly available national average costs for office visits, imaging studies, and hospitalizations, approximate costs incurred for each patient in one healthcare system were calculated and normalized to per year utilization. Normalizing to per-year utilization accounted for differences in length of follow-up across patients in our study.

LM and VM patients were categorized as either complex or simple. Involvement of the airway and/or more than one anatomic area was defined as complex. Anatomic areas were defined as head, neck, extremity (more than one extremity is considered complex), chest wall, abdominal wall, back and flank, thoracic/pleural, intraperitoneal, and pelvis/perineum/genitalia. Patients with a diagnosis of Klippel Trenaunay syndrome; congenital lipomatous overgrowth, vascular malformations, epidermal nevi, and scoliosis/skeletal/spinal abnormalities (CLOVES); generalized lymphatic anomaly (GLA); and kaposiform lymphangiomatosis (KLA) were classified as complex LMs. LM and VM patients without these features were categorized as simple (Fig. 1).

STATISTICAL ANALYSIS

Descriptive statistics including means and SDs were calculated to analyze healthcare utilization characteristics and to compare costs incurred by simple and complex LM/VM patients, and Welch *t* test assuming unequal variance was used to compare continuous variables. Fisher exact test was used to determine significant differences in categorical variables. A value of *P* less than 0.05 was considered statistically significant for all comparisons. Analyses were performed in GraphPad (Prism 9.0.0; San Diego, Calif).

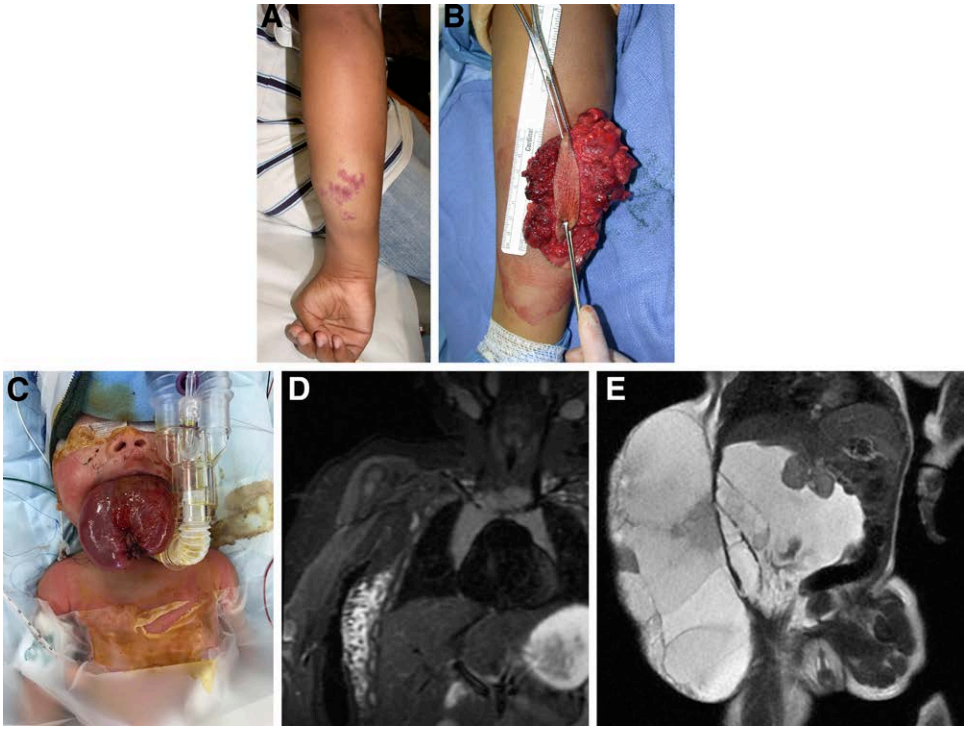

Fig. 1. Clinical spectrum of simple and complex VMs and LMs. A-B, Simple VM of the arm on clinical exam (A) and intraoperatively (B). C, Complex VM with airway involvement. D, Microcystic simple LM of the chest wall, T2 weighted postcontrast STIR sequence MRI. E, Complex LM of the abdominal wall extending into the intraperitoneal space, T2 weighted postcontrast MRI.

RESULTS

Demographics

A total of 126 patients met inclusion criteria for the study. Fifty-seven LM patients, including 28 simple and 29 complex patients (including nine Klippel Trenaunay syndrome, CLOVES, GLA, and KLA patients), and 69 VM patients, including 51 simple and 18 complex patients, were identified for analysis. LM patients ranged in age from 2 months to 35 years at the time of initial office visit within the study period and 58% of LM patients were women. VM patients ranged in age from 5 months to 67 years and 51% of patients were women.

Outpatient Utilization

To assess differences in outpatient healthcare utilization between simple and complex LM/VM patients, we analyzed the number of office visits, imaging studies performed, and specialists seen for each patient and normalized these values to per year utilization. Relative to simple LM patients, complex LM patients had a significantly higher number of office visits per year (3.9 versus 1.8, $P = 0.011$). Complex VM patients, however, did not incur a significantly higher number of office visits compared with simple VM patients, although there was a trend toward more visits (4.3 versus 2.8, $P = 0.061$, Fig. 2A).

The number of imaging studies performed for each patient was also documented as a metric of outpatient healthcare utilization. The most common imaging studies were magnetic resonance imaging and ultrasound. Both complex LM and VM patients required significantly more imaging studies than simple LM and VM patients, respectively (LMs: 5.5 versus 1.1, $P = 0.0004$; VMs: 5.1 versus 1.6, $P = 0.0087$, Fig. 2B).

Finally, we quantified the number of specialists seen per year by each patient. Complex LM patients were seen by significantly more specialists per year relative to simple LM patients (5.0 versus 3.0, $P < 0.0001$). A similar pattern of utilization was observed in VM patients, with complex VM patients being seen by an average of 4.6 specialists per year compared with 3.1 specialists for simple VM patients ($P = 0.0075$, Fig. 2C).

Inpatient Utilization

Yearly numbers of procedures and hospitalizations were quantified for simple and complex LM and VM patients to compare inpatient healthcare utilization across these groups. Procedures included direct treatments for VAs, such as sclerotherapy and debulking procedures, and supportive treatments such as tracheostomies and gastrostomy tube placements. Complex patients, regardless of VA subtype, were significantly more likely to require one or more procedures; 72.4% of complex LM patients versus 18.6% of simple LM patients required one or more procedures ($P < 0.0001$), whereas 79.3% of complex VM patients versus 51% of simple VM patients required one or more procedures ($P = 0.024$, Fig. 3A).

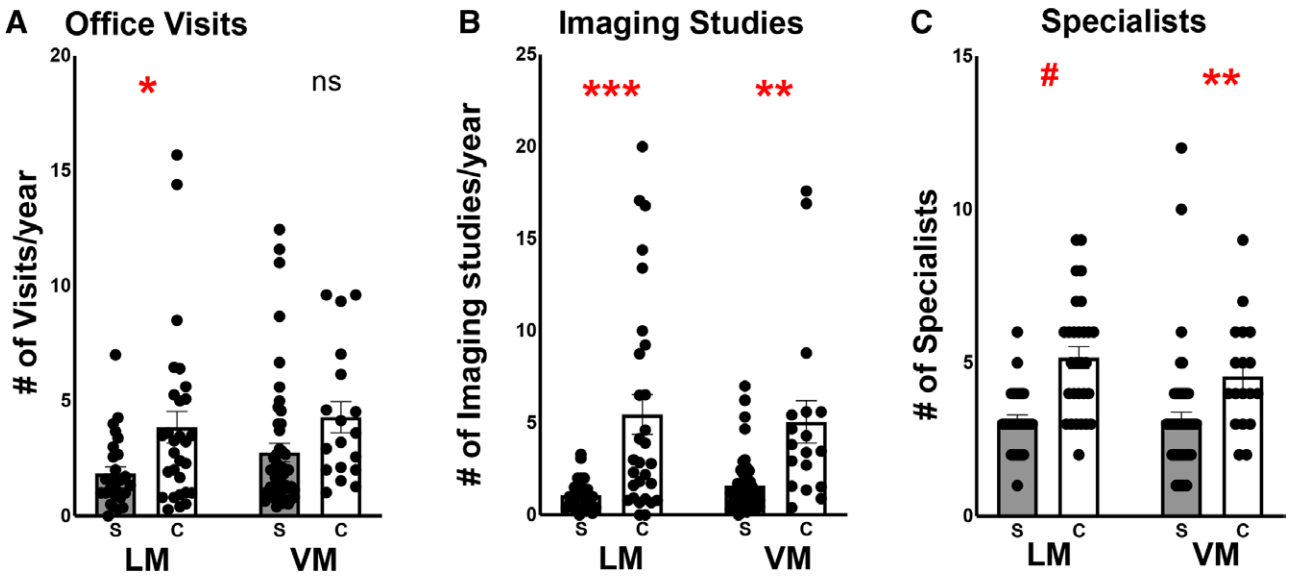

Fig. 2. Outpatient healthcare utilization by LM/VM patients. A, Average number of office visits per year for simple (S) and complex (C) LM (left) and VM (right) patients (LM: $P = 0.011$, VM: $P = 0.061$). B, Average number of imaging studies performed per year for simple and complex LM (left) and VM (right) patients (LM: $P = 0.0004$, VM: $P = 0.0087$). C, Average number of specialists seen per year for simple and complex LM (left) and VM (right) patients (LM: $P < 0.0001$, VM: $P = 0.0075$). Error bars represent mean ± standard error of the mean. *$P < 0.05$, **$P < 0.01$, ***$P < 0.001$, #$P < 0.0001$.

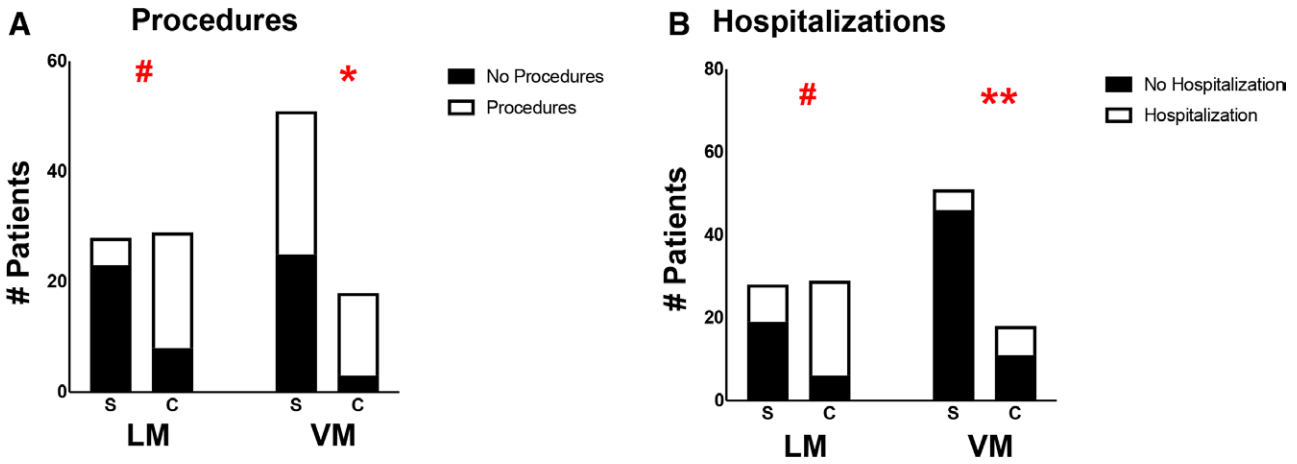

Fig. 3. Procedures and hospitalizations of LM/VM patients. A, Number of simple (S) and complex (C) LM (left) and VM (right) patients who had previously undergone one or more procedures vs no procedures (LM: $P < 0.0001$, VM: $P = 0.024$). B, Number of simple and complex LM (left) and VM (right) patients who had previously undergone one or more hospitalizations vs no hospitalizations (LM: $P = 0.0005$, VM: $P = 0.0098$). *$P < 0.05$, **$P < 0.01$, #$P \leq 0.0005$.

Complex LM/VM patients were also more likely to be hospitalized. Indications for hospitalization included planned admission after procedures as well as unplanned admission for management of complications such as sepsis and bleeding. For LM patients, 80% of complex patients relative to 32.1% of simple patients required one or more hospitalizations ($P = 0.0005$). For VM patients, 39% of complex patients versus 10% of simple patients required one or more hospitalizations ($P = 0.0098$, Fig. 3B).

We next performed subgroup analyses of simple and complex LM/VM patients who had electronic medical record documentation of procedures or hospitalizations. Among LM patients requiring procedures (n = 26), complex and simple patients underwent similar numbers of procedures (1.1 versus 0.8, $P = 0.25$). Likewise, for VM patients requiring procedures (n = 41), no significant difference was found between the number of procedures required by complex and simple patients (0.63 versus 0.47, $P = 0.29$, Fig. 4A).

The subgroup of simple and complex LM/VM patients who required one or more hospitalizations was also analyzed. Complex and simple LM patients who had one or more hospitalizations (n = 31) had similar average numbers of hospitalizations per year (0.87 versus 0.54, $P = 0.062$). However, complex LM patients had a significantly

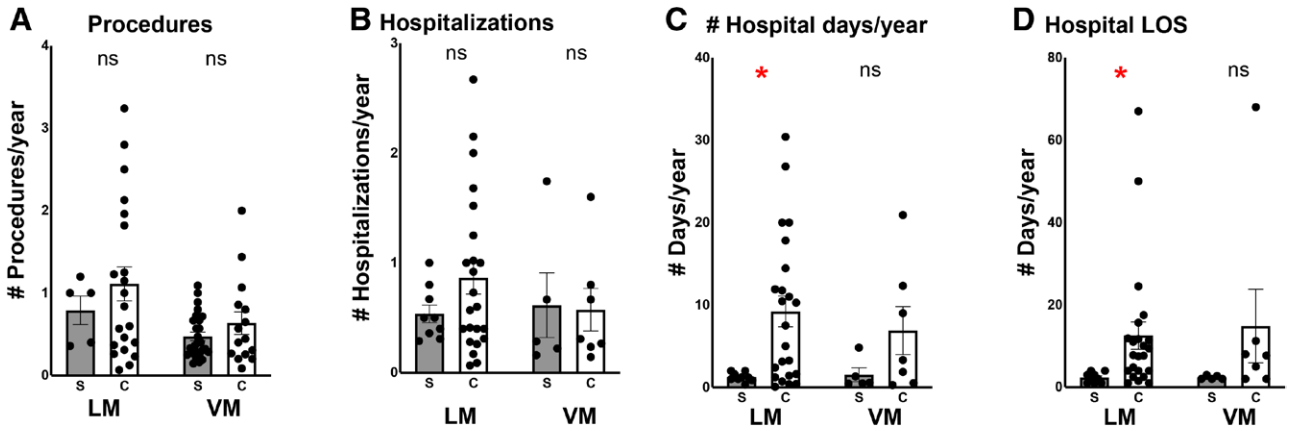

Fig. 4. Subgroup analysis of all patients requiring procedures and/or hospitalizations. A, For patients who underwent one or more procedures, average number of procedures per year for simple (S) and complex (C) LM (left) and VM (right) patients (LM: $P = 0.25$, VM: $P = 0.29$). B, For patients who underwent one or more hospitalizations, average number of hospitalizations per year (LM: $P = 0.062$, VM: $P = 0.91$), (C) hospital days per year (LM: $P = 0.0003$, VM: $P = 0.12$), and (D) hospital LOS for simple and complex LM (left) and VM (right) patients (LM: $P = 0.0055$, VM: $P = 0.21$). Error bars represent mean ± standard error of the mean. *$P < 0.006$.

higher number of hospital days per year (9.2 versus 1.2, $P = 0.0003$) and a longer average LOS when hospitalized (12.2 versus 2.1 days, $P = 0.0055$). Of the VM patients who required one or more hospitalizations (n = 12), there was also no significant difference in the number of hospitalizations per year between complex and simple patients (0.57 versus 0.61, $P = 0.91$). In contrast to LM patients, there were also no significant differences in the number of hospital days per year (6.9 versus 1.5, $P = 0.12$) or average LOS (14.0 versus 2.3 days, $P = 0.21$) between complex and simple VM patients (Fig. 4B). The lack of statistical significance in these comparisons despite a trend toward an increased number of hospital days and hospital LOS in complex VM patients may be attributable to the small number of simple (n = 5) and complex (n = 7) VM patients who were hospitalized in this study cohort.

Costs

VA care can be costly and includes fees for office visits, diagnostic tests, imaging studies, procedures, and inpatient admissions. Care for complex LM/VM patients in our study was significantly more costly than care for simple LM/VM patients. We used publicly available national averages to determine cost estimates for office visits, imaging studies, and hospitalizations for each patient. Costs of procedures, including provider fees and hospital costs, were not included in our analysis as these data were not available inpatient records. Excluding procedure-related costs, we found that complex LM patients incurred an average annual cost of $22,174, whereas simple LM patients incurred an average cost of $2442 ($P = 0.0001$; Table 1). A similar trend was observed for VM patients, with complex VM patients incurring an average of $19,814 annually relative to $3814 incurred annually by simple VM patients ($P = 0.026$, Table 1, Fig. 5A).

To further characterize costs incurred by simple and complex LM and VM patients, we analyzed the costs of office visits, imaging studies, and hospitalizations separately. Costs of office visits were significantly higher for complex LM patients compared with simple LM patients ($1002 versus $480, $P = 0.011$); however, complex VM patients did not incur significantly higher costs annually for office visits relative to simple VM patients ($1116 versus $717, $P = 0.061$, Fig. 5B). Imaging costs were significantly higher for complex LM and VM patients, with

Table 1. Annual Costs Incurred by LM and VM Patients

Average Annual Costs	Simple LM	Complex LM	*P*
LM patients			
Overall cost	$2379	$21,572	**0.0001**
Office visits cost	$493	$973	**0.017**
Imaging studies cost	$1151	$4019	**0.0001**
Hospitalizations cost	$2480	$20,725	**0.0004**
Average Annual Costs	**Simple VM**	**Complex VM**	***P***
VM patients			
Overall cost	$3814	$19,814	**0.026**
Office visits cost	$717	$1116	0.061
Imaging studies cost	$2793	$11,800	**0.0087**
Hospitalizations cost	$3101	$17,738	0.13

LM, lymphatic malformation; VM, venous malformation.
P values in boldface are statistically significant.

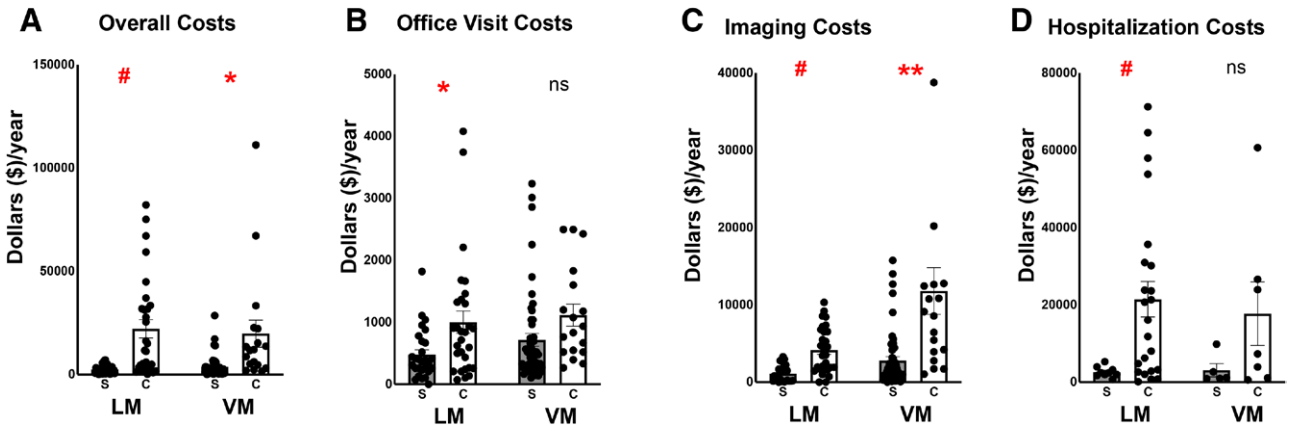

Fig. 5. Annual costs incurred by LM/VM patients. A, Average annual costs incurred (dollars) by simple (S) and complex (C), LM (left) and VM (right) patients (LM: $P = 0.0001$, VM: $P = 0.026$). B, Average annual cost of office visits for simple (S) and complex (C), LM (left) and VM (right) patients (LM: $P = 0.011$, VM: $P = 0.061$). C, Average annual cost of imaging studies for simple and complex LM (left) and VM (right) patients (LM: $P = 0.0005$, VM: $P = 0.0087$). D, Average annual cost of hospitalizations for simple and complex LM (left) and VM (right) patients (LM: $P = 0.0005$, VM: $P = 0.13$). Error bars represent mean ± standard error of the mean. *$P < 0.05$, **$P < 0.01$, #$P \leq 0.0005$.

complex LM and VM patients incurring an average annual cost for imaging studies of $4158 and $11,800, respectively, and simple LM and VM patients incurring average annual costs of $1110 and $2793, respectively (LM: $P = 0.0005$, VM: $P = 0.0087$, Fig. 5C). Costs of hospitalizations were significantly higher in complex LM patients compared with simple LM patients ($21,452 versus $2648, $P = 0.0005$), but no significant differences were observed between average annual costs of hospitalizations for complex and simple VM patients ($17,738 versus $3101, $P = 0.13$, Fig. 5D). Of note, the lack of significance for cost of hospitalization between simple and complex VM patients may be attributable to the low number of hospitalized simple (n = 5) and complex (n = 7) VM patients.

DISCUSSION

LMs and VMs include a heterogeneous group of conditions with symptoms ranging from pain and functional impairment to life-threatening emergencies such as sepsis, coagulopathy, and airway obstruction. Thus, healthcare needs vary widely between patients with different levels of lesion complexity. Minimal research has been conducted to estimate the public health burden of VAs or to analyze factors that predict increased healthcare utilization. Our retrospective analysis confirmed that lesion complexity positively correlates with increased healthcare utilization and cost. compared with simple LM and VM patients, complex patients demonstrated increased needs in the outpatient setting (higher number of office visits, imaging studies, and specialists involved) and in the inpatient setting (higher number of hospitalizations, procedures, and hospital days per year).

Our study is the first to demonstrate a direct association between LM/VM complexity and increased healthcare utilization in both the inpatient and outpatient settings and shows that complexity is a driving factor contributing to increasing costs for LM/VM patients. Although this may be intuitive, this study provides clear documentation of these differences. This data may help patients and providers better anticipate potential future healthcare needs and costs, and improve counseling and expectations for patients and their families. Moreover, these data will provide a framework for third-party payors of typical utilization patterns of VA patients and demonstrates that they have significant healthcare needs. Few studies have investigated the healthcare burden in VAs, and studies that analyzed healthcare utilization and costs in LMs and other VAs such as hemangiomas and AVMs did not include outpatient utilization or factors that increased costs.[17,18] Although our cost estimation was incomplete due to inaccessibility of procedural and diagnosis-related group costs, we were able to estimate the healthcare utilization of LM/VM patients in both the inpatient and outpatient settings. Because complex patients were significantly more likely to require procedures, the true cost differential would likely increase if procedural costs were included.

When we analyzed the subgroup of LM/VM patients who required procedures or hospitalizations, simple and complex patients did not differ significantly in their average number of procedures or hospitalizations per year. Thus, although it is less likely for a simple LM/VM patient to require a procedure or be hospitalized, if they required either, the number of procedures or frequency of hospitalizations were comparable to those of complex patients. This information may be helpful for patients and providers in predicting prognosis and need for healthcare services.

Limitations to our study include its retrospective design and inclusion of patients from a single surgeon's practice and the clinics of the multidisciplinary VA program within one academic center, which may not be reflective of utilization patterns in other settings. Furthermore, our study analyzed patients affected by two prevalent VA subtypes, LMs and VMs, but a more comprehensive analysis could include patients with additional VAs. Additionally, our study estimated costs using publicly available data for average costs of office visits, imaging studies, and hospitalizations and did not include costs of procedures. We did not have information for DRGs or CPT codes by proceduralists and surgeons, and

thus could not factor surgeon and facility fees into our cost analysis. Thus, our study likely grossly underestimated the true costs incurred by LM/VM patients. These estimates also do not account for regional differences in costs of inpatient and outpatient services. Finally, we did not include emergency department visits as a separate variable; some of our patients used emergency department visits as part of their primary care for their VAs, which would have confounded true emergency visits. Future studies are needed to investigate VA patient factors, beyond lesion complexity, that may impact healthcare utilization, such as patient age, which has been found to affect healthcare costs and LOS in the inpatient setting for pediatric and adult AVM patients.[18–20]

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DISCLOSURE

The authors have no financial interest to declare in relation to the content of this article.

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