Incidence of Post Surgery Peripheral Vascular Malformations of Limbs in SMS Hospital Jaipur

Chitresh Shekhawat¹*, Neetu Singh², Gaurav Sharma³

¹,²SMS Medical College and Hospitals, Jaipur, Rajasthan, India
³RVRS Medical College, MG Hospital Bhilwara, Rajasthan, India

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Corresponding author: Dr. Chitresh Shekhawat
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Abstract
Peripheral vascular malformations (PVMs) include a wide spectrum of diseases that can produce potentially life- or limb-threatening complications. The International Society for the Study of Vascular Anomalies (ISSVA) Classification System categorized vascular anomalies into two basic types, the vaso proliferative or vascular neoplasms and developmental vascular abnormalities.

Method: A retrospective, cross-sectional study was conducted for the period of 2 year. All patients under surgery OPD with a Peripheral limb vascular anomaly excluding upper limbs and lower limbs were taken into the study and investigated and followed up for at least 6 months.

Result: In this study, out of 38 patients 26 patients (12 (44%) males and 14 (56%) females) were diagnosed with vascular malformation of upper and lower limbs. It is observed that low flow lesions are more common in both upper and lower limbs. Male patients with these lesions tend to present much later in life than female patients.

Conclusion: AVMs show variance in site and type based on gender. Male patients were more likely present with high-flow AVMs in lower limbs whereas female patients typically presented with low-flow AVMs on the upper extremities.

Keywords: Peripheral limb vascular malformations, varicose veins, AVM.

Introduction
Peripheral vascular malformations (PVMs) include a wide spectrum of diseases that can produce potentially life- or limb-threatening complications. Many times, these lesions remain silent or asymptomatic for long time. PVMs are relatively common within the extremities and usually confined to the subcutaneous tissues and muscles. However, intra- articular and intraosseous extensions may be present and pose greater therapeutic challenges.

Incidence of vascular malformations is 1.5% [1]. 39.22% of these occur in the lower limbs [2].

Clinical history and physical examination remain essential in diagnosis but can underestimate their nature and size, with improvements in imaging playing an important role [3].

Vascular anomalies were classified originally by Mulliken and Glowacki in 1982 based on clinical features and biological behavior [4]. However, currently, the International Society for the Study of Vascular Anomalies (ISSVA) classification system is widely accepted. The International Society for the Study of Vascular Anomalies (ISSVA) Classification System places an
emphasized on the pathologic and hemodynamic features of vascular anomalies and was last updated in 2014 and utilized to categorize vascular anomalies into two basic types, the Vaso proliferative or vascular neoplasms such as hemangioma and developmental vascular abnormalities called congenital vascular malformations (CVMs)[5]. Hemangiomas are considered as most common tumors that start from few weeks of birth to 9-12 months by rapid growth [6].

The type of flow can be used to group vascular malformations into low-flow and high-flow malformations according to their vascular hemodynamics [5,7]. High-flow malformations are most commonly arteriovenous. Low-flow malformations include lymphatic malformations (LMs), capillary-venous malformations, and VMs, glomovenous malformations, and non-shunting mixed-lesions [8].

AVMs are rare, accounting for only 10-15% of all clinically significant Venous Malformations [9] with an estimated prevalence of 5-613 in 100,000 [10]. Thin skin covered by venous malformation can cause bleeding and formation of pool which is usually non dangerous and called as thrombophlebitis [11]. There are very few studies done on AV malformation in Indian subcontinent and there is paucity of medical literature. Therefore this study aims at determining the prevalence of arterio-venous malformations in central Rajasthan and studying the characteristics of patients presenting with AVMs.

**Material and Methods**

This retrospective, cross-sectional study was conducted at a tertiary care hospital. The study was done for the period of 2 year between Jan 2020 to Dec 2021. Approval was obtained from the Institutional Review Board (IRB) and Ethics committee. All patients diagnosed with a Peripheral limb vascular anomaly during the study period were considered potentially eligible for the study. Informed written consent was obtained from all patients. Patients diagnosed with vascular malformation other than limbs (upper limbs and lower limbs) were excluded. Pregnant patients and children under five years were not included. Lastly, patients lost to a minimum of six months follow-up were also excluded.

Monthly follow-ups were performed for at least six months. Ultrasound imaging was performed with Hitachi Arrieta 65. Clinical examination and ultrasound Doppler were performed at each follow-up to assess treatment efficacy. CT scan, MRI scan and MR angiogram was performed wherever required to access the extension of lesion and to plan the treatment modality. The recurrence rate and other complications were recorded.

Statistical analysis was performed using Statistical Packages for the Social Sciences program (SPSS) version 26 (IBM Inc., Armonk, USA). Quantitative variables like age were expressed as mean ± standard deviation. Qualitative variables like gender were expressed as frequencies and percentages. Patients were divided into two groups based on gender and were compared using Chi-square Test. A p-value ≤0.05 was considered as statistically significant.

**Results**

In the present study a total of 38 patients with vascular malformations visited our department during the study period. Out of 38 patients, 26 patients were diagnosed with vascular malformation of upper and lower limbs and were included in this retrospective analysis. In the study group 12 (44%) patients were males and 14 patients (56%) were females with male: female = 1: 1.3. Overall, the age ranges from 2 years to 54 years with mean age of whole study group was 24.2 years with males had mean age of 25.9±3 years and females 22.6±4 which shows that males presented late in life as compared to females (Image 1). The difference in age among males and females was statistically significant (p-vale=0.02). Out of total 26 cases, 16 patients had lesion in lower limbs and 10 patients had lesions in upper limbs. Patients were grouped according to type of flow of AVM in High flow and low flow malformations (Table 1). In our study 9 patients were diagnosed with high flow lesions whereas 17
with low flow lesions. The lesions in upper and lower limbs with flow velocity are shown in table 2 (Image 2). It is observed that low flow lesions are more common in both upper and lower limbs but percentage of low flow lesions in upper limbs is significantly higher than high flow as compared to lower limb and is statistically significant (p-value=0.003) (Image 3).

Various characteristic of patients between males and females are shown in table no 3. It was observed that in males symptoms are shown later in life as compared to females and the results are statistically significant (p = 0.02). In males the high flow lesions are more common whereas in females low flow lesions are more common and the difference is statistically significant (p=0.02). It was also observed that upper limb lesions are more common in females whereas lower limbs are more involved in males but the results had no significance statistically (Image 4).

The symptoms observed in the study group are shown in Table no. 4. The most common complaints were pain and swelling of the limbs. In our study, 2 patients did not have any complaints and the findings were incidental whereas one patient suffered from features of heart decompensation (Image 5,6).

Table 1: Type and flow of AVM

<table>
<thead>
<tr>
<th>Variables</th>
<th>N. (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Low Flow malformations</td>
<td>17 (65.38%)</td>
</tr>
<tr>
<td>High Flow Malformations</td>
<td>9 (34.61%)</td>
</tr>
<tr>
<td>Total</td>
<td>26</td>
</tr>
</tbody>
</table>

Table 2: Location of Vascular Malformations

<table>
<thead>
<tr>
<th>Parameters</th>
<th>Total</th>
<th>High Flow</th>
<th>Low Flow</th>
<th>P – value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Lower limbs</td>
<td>15 (57.93%)</td>
<td>7 (%)</td>
<td>8 (%)</td>
<td>0.013 (S)</td>
</tr>
<tr>
<td>Upper limbs</td>
<td>11 (42.06%)</td>
<td>2 (%)</td>
<td>9 (%)</td>
<td></td>
</tr>
<tr>
<td>Total</td>
<td>26</td>
<td>9</td>
<td>17</td>
<td></td>
</tr>
</tbody>
</table>

Table 3: Characteristics of male and female patients at time of presentation

<table>
<thead>
<tr>
<th>Age in years (Mean ± SD)</th>
<th>Male (n=12)</th>
<th>Females (n=14)</th>
<th>p-value</th>
</tr>
</thead>
<tbody>
<tr>
<td>25.9±3</td>
<td>22.6±4</td>
<td>0.004 (S)</td>
<td></td>
</tr>
<tr>
<td>Type of AVM</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>High-flow</td>
<td>7</td>
<td>2</td>
<td></td>
</tr>
<tr>
<td>Low-flow</td>
<td>5</td>
<td>12</td>
<td>0.02 (S)</td>
</tr>
<tr>
<td>Site of AVM</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Upper Limbs</td>
<td>3</td>
<td>8</td>
<td></td>
</tr>
<tr>
<td>Lower Limb</td>
<td>9</td>
<td>6</td>
<td>0.1 (NS)</td>
</tr>
</tbody>
</table>

Table 4: Signs and symptoms in peripheral AVM.

<table>
<thead>
<tr>
<th>Parameters</th>
<th>Low Flow</th>
<th>High Flow</th>
</tr>
</thead>
<tbody>
<tr>
<td>No clinical signs</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Skin nevus and/or skin heating</td>
<td>1</td>
<td>3</td>
</tr>
<tr>
<td>Mass, pulsating</td>
<td>3</td>
<td>9</td>
</tr>
<tr>
<td>Pain</td>
<td>10</td>
<td>12</td>
</tr>
<tr>
<td>Swelling</td>
<td>5</td>
<td>10</td>
</tr>
<tr>
<td>Ulcer</td>
<td>1</td>
<td>3</td>
</tr>
<tr>
<td>Bleeding</td>
<td>3</td>
<td>7</td>
</tr>
<tr>
<td>Heart decompensation</td>
<td>0</td>
<td>1</td>
</tr>
</tbody>
</table>
Image 1: Sex wise distribution of cases.

Image 2: Type and flow of AVM

Image 3: Location of Vascular Malformations
Image 4: Signs and symptoms in low and high flow peripheral AVM.

Image 5: Showing AV malformation in upper limb.

Image 6: CT angiogram showing multiple dilated arterial feeders from 3rd part of axillary artery with multiple arterial and venous channels. Findings suggestive of high flow AVM.
Discussion

AVMs are congenital vascular lesions that may not be apparent until later in life. AVMs may be simple or complex with some also show intra articular extension. These lesions require angiography to reach a diagnosis [1]. In many cases the AVMs can be life or limb-threatening, therefore early diagnosis and treatment is of imminent importance. A large AVM can also cause high-output cardiac failure due to the absence of an intervening capillary bed. Artery steal syndromes can also occur due to the presence of AVMs, resulting in under perfusion of tissues. Moreover, a large AVM closely related to a long bone can result in limb-length discrepancy either by limb undergrowth or overgrowth [5]. There may be a predictive risk of contractures after treatment with respect to the degree of muscular involvement of VMs [12,13].

The male to female incidence of AVMs in our study was 1: 1.3 which is in concordance with the study of Lee BB et al who also reported male: female ratio of 1: 1 to 1: 2 [9], whereas Usman, Rashid, et al reported a slightly higher male to female ratio of 1:3. Furthermore, the average age at presentation in female patients was younger compared to male patients [14]. Two factors might account for these findings. First, AVMs tend to grow under hormonal influences, enlarging after puberty and due to pregnancy [15]. Therefore, hormonal exposure could act as a confounding factor, leading to an earlier pickup in women.

Male patients tended to present with an AVM involving the lower limb whereas the upper limbs were more frequently involved in female patients. However there was no statistically significant correlation found. There are still very few studies where such association is observed. The effect of age and sex on location of intracranial AVM is done [16], however more studies are needed before similar conclusions can be drawn for peripheral limb AVMs.

Another difference we found was the nature of AVMs. Males more often presented with high-flow AVMs while females commonly presented with low-flow AVMs. The proliferating phase of infantile hemangiomas in its proliferation phase is also a high-flow lesion which is categorized as a vascular tumor rather than malformation which shows regression with time, unlike vascular malformations (Malik et al. 2021)[17]. Similar results was also published by Usman, Rashid, et al [14] and Nosher J [18]. The "Little AVMs", as defined by Stein et al. [19] are malformations on the spectrum between AVMs and venous malformations. These lesions present clinically as AVMs but behave radiologically as low-flow lesions. If we consider this little AVMs as low flow lesions this female predominance can be accounted, however the predominant opinion is that there are no gender differences in the prevalence of low-flow malformations and more studies in different sample population is needed to conclude.

There is more frequency to diagnose and classify vascular malformations incorrectly. Therefore the imaging modalities which are easily used and can interpret easily are very important. Thus, new techniques are required for better diagnosis, selection of treatment, and its monitoring due to improved understanding of pathogenesis, progression of angiogenesis in vivo [20].

Conclusion

Our study observed that there are significant gender differences in the clinical presentation of AVMs. Male patients with these lesions tend to present much later in life than female patients. Moreover, AVMs show variance in site and type based on gender. Male patients were more likely to present with high-flow AVMs in lower limbs whereas female patients typically presented with low-flow AVMs on the upper extremities. Due to lack of similar data on peripheral AVMs in the Indian population, Future longitudinal studies are needed before any decisive claims about these relations can be made. Larger studies are also needed to make recommendations for treatment regimens at the level of the general population.
Acknowledgement
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References
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