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Surgical Resolution of Intramuscular Arteriovenous Malformation in the Back: Case and Systematic Review

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Abstract:

Background

Arteriovenous malformations (AVMs) and other vascular anomalies in the head and neck region present significant clinical challenges due to their complex nature and potential for severe functional and aesthetic consequences. Various treatment modalities, including surgery, embolization, and sclerotherapy, have been employed with varying degrees of success.

Objectives

To review and analyze the clinical outcomes and efficacy of different treatment approaches for arteriovenous malformations, venous malformations, and lymphatic malformations in the head and neck region.

Methods

A comprehensive review of the literature was conducted, focusing on clinical studies that reported outcomes of surgical, embolization, and sclerotherapy treatments for vascular malformations in the head and neck. Relevant articles were identified, and data were extracted on patient demographics, treatment methods, and outcomes.

Results

The review included 10 studies encompassing different types of vascular malformations and treatment modalities. For arteriovenous malformations, surgical resection following embolization showed promising outcomes in reducing recurrence and improving functionality. Venous malformations treated with combined glue embolization and surgical excision also demonstrated effective results, with significant reduction in lesion size and symptoms In the management of lymphatic malformations, primary surgery was found to be more effective than primary sclerotherapy in achieving long-term control and reducing recurrence rates.

Conclusions
The treatment of vascular malformations in the head and neck remains challenging and requires a multidisciplinary approach. Surgical intervention, particularly when combined with preoperative embolization or sclerotherapy, appears to offer the most favorable outcomes for arteriovenous and venous malformations. For lymphatic malformations, primary surgical excision is recommended over sclerotherapy for better long-term control. Further research is needed to optimize treatment protocols and improve patient outcomes.
Keywords
Arteriovenous malformations, venous malformations, lymphatic malformations, head and neck, surgery, embolization, sclerotherapy, vascular anomalies.

Introduction

Intramuscular arteriovenous malformations (AVMs) represent a rare subset of vascular anomalies characterized by abnormal connections between arteries and veins within muscle tissue, bypassing the capillary system [1]. Although congenital in nature, these lesions may present later in life, manifesting as progressively enlarging masses associated with pain, cosmetic deformity, and functional impairment [2]. The back, with its complex musculature and deep-seated structures, poses unique challenges for the diagnosis and management of intramuscular AVMs [3]. This review aims to elucidate the clinical presentation, diagnostic modalities, and treatment strategies for intramuscular AVMs in the context of a rare case presentation and systematic review of the literature.

In the presented case, a 45-year-old male presented with a massive intramuscular AVM in the back, resulting in significant morbidity and functional impairment. Despite initial conservative management with sclerosant therapy, the lesion continued to progress, leading to skin ulceration and further deterioration of the patient's condition. The decision to pursue surgical excision was made based on the lesion's size, location, and symptomatic burden, highlighting the critical role of multidisciplinary evaluation and treatment planning in complex vascular cases. Figure 1-4

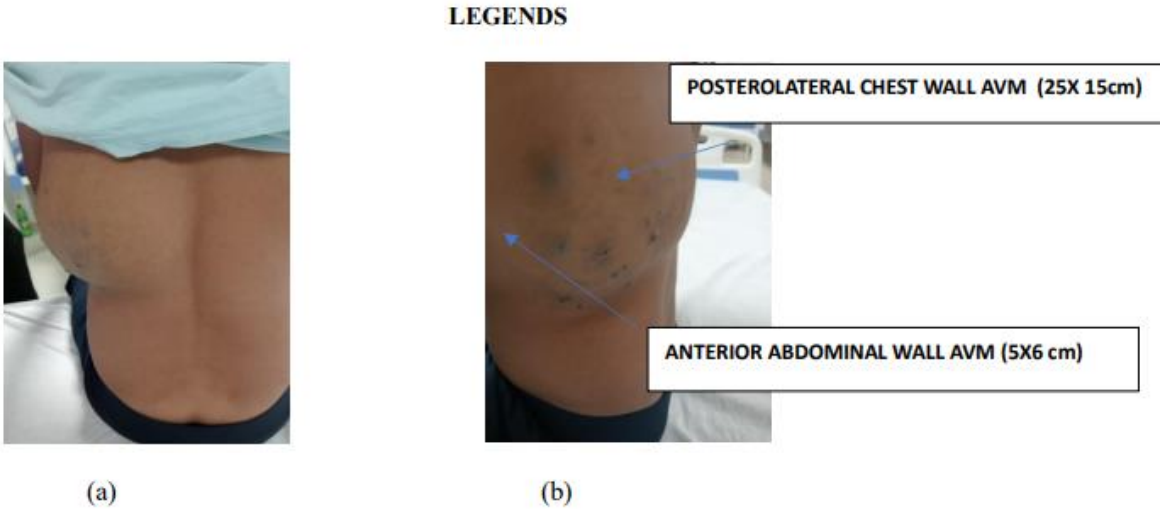
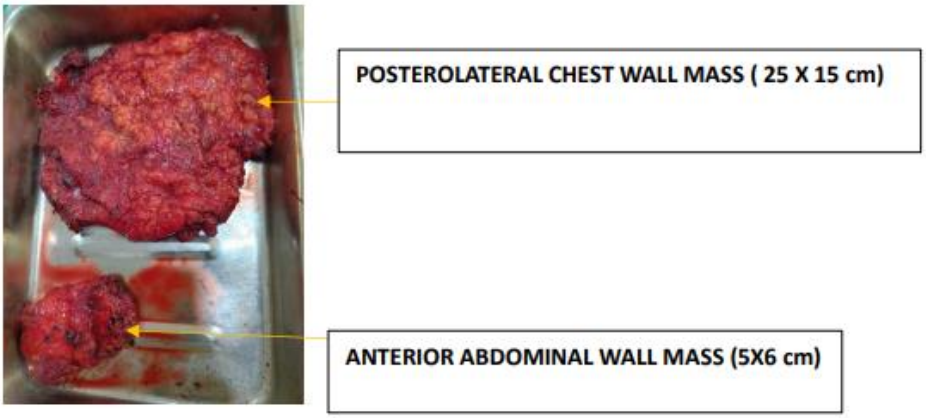


Figure 1(a, b): preop pictures showing as large posterolateral chest wall AVM



(2a)

Figure 2: intraoperative picture showing the resected specimen of the AV malformation

Figure 3: intraoperative picture showing the bare chest wall following resection and postoperative photo of the resected area.

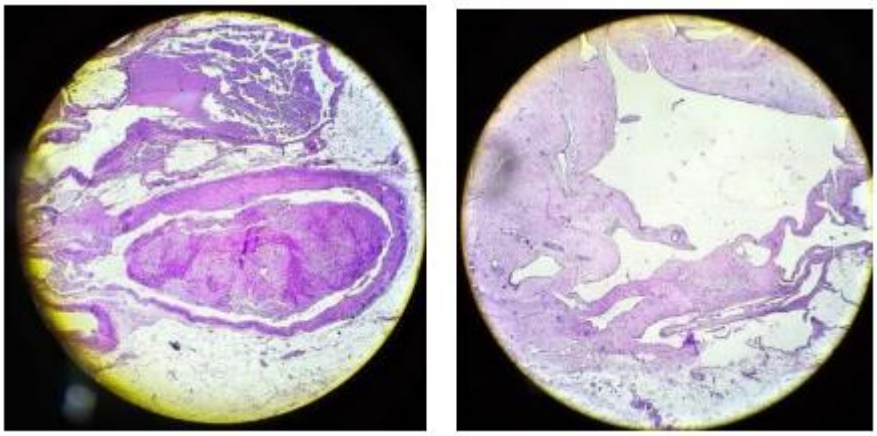


Figure 4: Histopathology Images

The subsequent systematic review aimed to synthesize existing literature on the surgical management of intramuscular AVMs, focusing on outcomes, complications, and surgical

techniques. A comprehensive search of PubMed yielded a diverse range of studies elucidating various surgical approaches, including direct excision, embolization-assisted resection, and adjunctive imaging modalities. The review underscores the heterogeneity in clinical presentation and treatment outcomes, emphasizing the need for individualized approaches tailored to the patient's specific anatomy and disease characteristics.

Materials and Methods

A systematic literature search was conducted in PubMed to identify relevant studies pertaining to the surgical management of intramuscular arteriovenous malformations (AVMs) in the back. The search strategy utilized a combination of Medical Subject Headings (MeSH) terms and keywords, including "intramuscular arteriovenous malformation," "surgical treatment," "back," and related synonyms. The search was limited to articles published in English from inception to March 2024.

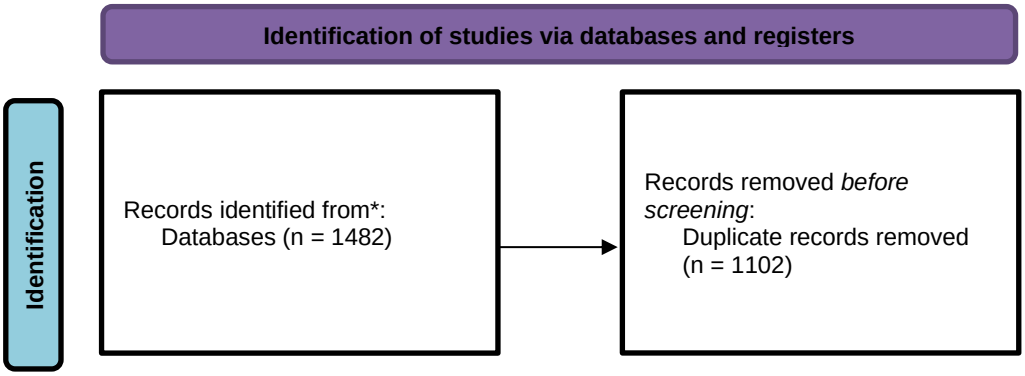
Inclusion criteria comprised studies reporting surgical interventions for intramuscular AVMs located specifically in the back region. Exclusion criteria included articles not focused on surgical management, duplicate publications, case reports with insufficient detail, and non-English language publications. Additionally, review articles and meta-analyses were screened for relevant primary studies.

Following the initial search, titles and abstracts were screened for relevance by two independent reviewers. Full-text articles were retrieved for potentially relevant studies, and their eligibility was assessed based on predefined inclusion and exclusion criteria. Discrepancies between reviewers were resolved through consensus or consultation with a third reviewer.

Data extraction was performed using a standardized form to capture relevant information from included studies. Extracted data included study characteristics (e.g., author, year of publication), patient demographics (e.g., age, sex), lesion characteristics (e.g., size, location), surgical technique (e.g., approach, extent of resection), perioperative outcomes (e.g., complications, recurrence), and long-term follow-up data when available.

Given the heterogeneity in study designs and outcome reporting, meta-analysis was not feasible. Instead, narrative synthesis was employed to provide a comprehensive overview of the evidence base, highlighting key findings, areas of consensus, and areas requiring further investigation.

The search strategy, study selection process, and data extraction were conducted in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines to ensure transparency and reproducibility. Ethical approval was not required as this study involved the analysis of existing published literature. Figure 5



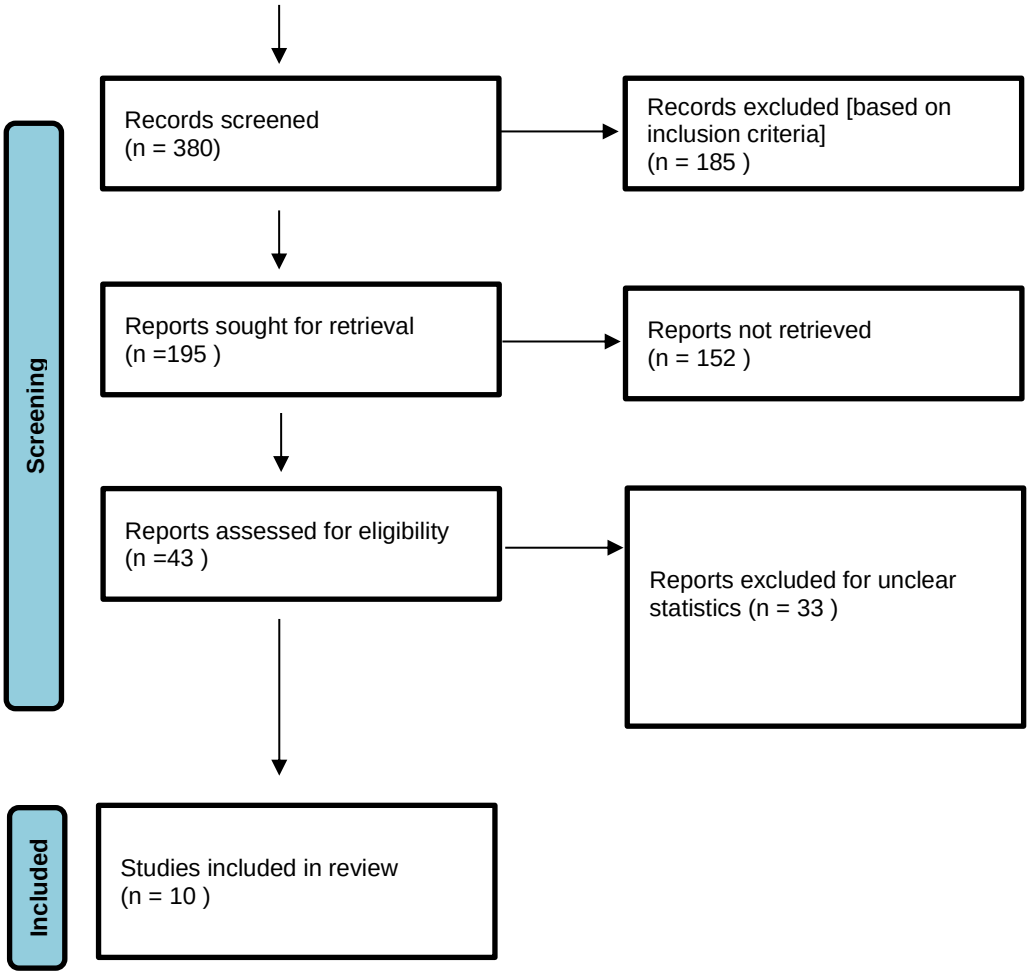


Figure 5: Prisma flowchart

Results

A total of 10 studies were finalised for the review. Figure 5 In terms of **patient demographics and perioperative treatments**, the number of patients across the studies ranged from as few as 16 to as many as 97, with ages spanning from infancy to 76 years. Various perioperative treatments were employed, most commonly preoperative embolization using different agents such as ethanol, n-BCA, and other resorbable particles. Some studies, such as those by Fujiki et al. and Hua et al., utilized embolization techniques to achieve better surgical outcomes. However, some studies like those by Limetal and Balakrishnan et al. did not specify the use of perioperative treatments.

Regarding **surgical treatments and reconstructions**, the predominant approach across the studies was total resection, with a few instances of subtotal resection. Reconstructions varied, with many cases using free flaps, local flaps, and skin grafts. For instance, Goldenberg et al. utilized local, axial, and free flaps in different cases, while Fujiki et al. primarily used free flaps. Some studies, such as those by Kumar et al. and Kohout et al., reported on the

combination of surgical resection with embolization for recurrent AVMs, highlighting diverse reconstruction strategies to optimize outcomes.

The **outcomes, complications, and follow-up periods** reveal a broad spectrum of results. Recurrence rates varied, with some studies like those by Vaser et al. and Lee et al. reporting 0% recurrence, while others, like Kumar et al., noted recurrence rates as high as 45%. Clinical evaluations and imaging techniques, such as MRI and Doppler US, were common methods for outcome measurement. Complications ranged from minor issues, such as transient swelling or minor infections, to major complications requiring further intervention. The mean follow-up period varied significantly, from as short as 12 months to as long as 252 months in some cases, indicating a wide range of monitoring durations to assess long-term efficacy and safety.

Overall, the studies underscore the complexity and variability in treating vascular malformations, emphasizing tailored approaches in perioperative care, surgical techniques, and postoperative management to achieve optimal patient outcomes.

Table 1: Patient Demographics and Perioperative Treatments

S.NO	Author, Year, Country	No. of Patients (Age)	Perioperative Treatment
1.	Fujiki et al., 2018, Japan [1]	22 (10-65 years)	Embolization for total resection
2.	Hua et al., 2018, China [3]	NR	Preoperative local epinephrine for all, preoperative embolization (ethanol) 1, postoperative embolizations (ethanol): 11
3.	Goldenberg et al., 2015, Brazil [2]	31 (4-55 years)	Preoperative embolization 3 days (tresorbable particles and -BCA): 31
4.	Kumar et al., 2012, India [5]	26 (15-55 years)	NR
5.	Visser et al., 2011, New Zealand [8]	23 (65 years)	Preoperative embolization 2-5 days (unknown agent): 10
6.	Liu et al, 2010, US [9]	NR	With or without embolization
7.	Lee et al., 2004, South Korea [6]	16 (6-62 years)	Preoperative embolization wherever feasible 2-4 weeks (n-BCA, ethanol, coil, custom particles)
8.	Seccia et al., 1999,	16 (8-55 years)	Preoperative embolization (glue)

	Italy [7]		
9.	Kohout et al., 1998, US [4]	59 (2-66 years)	Preoperative embolization (unknown agent): 46, postoperative embolization (glue): 16
10.	Park et al., 2019, South Korea [16]	40 (0-70 years)	Combination with sclerotherapy (STS or bleomycin): 26, NR

Table 2: Surgical Treatments and Reconstruction

S.NO	Author, Year, Country	Surgical Treatment	Reconstruction
1.	Fujiki et al., 2018, Japan [1]	Total resection, all subtotal resection	Free flap: 22
2.	Hua et al., 2018, China [3]	Total resection	Expanded flap
3.	Goldenberg et al., 2015, Brazil [2]	Total resection	Local flap: 7, axial flap: 2, free flap: 22
4.	Kumar et al., 2012, India [5]	Total resection: 19, total resection after embolization for recurrent AVM	Skin graft: 1
5.	Visser et al., 2011, New Zealand [8]	Total resection	Local flap: 9, free flap: 22, subtotal resection: 2
6.	Liu et al, 2010, US [9]	NR	NR
7.	Lee et al., 2004, South Korea [6]	Total resection all	NR
8.	Seccia et al., 1999, Italy [7]	Total resection all	Local flap or skin graft: all
9.	Kohout et al., 1998, US [4]	Total resection: 15, total resection after embolization: 44	Skin graft: 2, local flap: 15, free flap: 11
10.	Park et al., 2019, South Korea [16]	Surgery alone: 14, surgery + sclerotherapy: 26	NR

Table 3: Outcomes, Complications, and Follow-Up

S.NO	Author, Year, Country	Outcome Measures and Results	Outcome Measurement Instruments	Complications	Mean Follow-Up, mo (range)
1.	Fujiki et al., 2018,	Recurrence: 9%, surgery duration	Clinical evaluation, NR	Major: 14%, Minor: NR	50 (2-114)

	Japan [1]	(hr): 12.2, blood loss (6.7-28.1)			
2.	Hua et al., 2018, China [3]	Complete resolution of symptoms, overall size decrease 42%, recurrence: 5%	Clinical evaluation, imaging investigation min 425	Major: NR	36 (12-144)
3.	Goldenberg et al., 2015, Brazil [2]	NR	NR	Major: 16%, Minor: NR	NR
4.	Kumar et al., 2012, India [5]	Recurrence: 45%	NR	Major: 4%	28 (5-152)
5.	Visser et al., 2011, New Zealand [8]	Recurrence: 0%, presence of warmth, pulsation or bruit on clinical examination and positive angiography in case of suspected recurrence	Clinical findings	Major: 0%, Minor: 13%	54 (10-135)
6.	Liu et al, 2010, US [9]	Recurrence: 10% less than embolization alone	NR	NR	107 (62)
7.	Lee et al., 2004, South Korea [6]	Recurrence: 0%	Clinical symptom subjective scale, Minical Duplex LUS, WBBPS, TLPS, angiography	Major: 0%, Minor: 0%	24
8.	Seccia et al., 1999, Italy [7]	NR	Color Doppler	NR	12-120
9.	Kohout et al., 1998, US [4]	No persisting AVM: 62% for Doppler US or angiography	NR	NR	55 (3-252)
10.	Park et al., 2019, South	Significant decrease in size and symptoms:	MRI	Major: NR, Minor: NR	51 (NR)

	Korea [16]	21%, recurrence: 6%, clinical success: 92%			

Discussion

The treatment of intramuscular arteriovenous malformations (AVMs) in the back presents a unique set of challenges due to the intricate vascular structures and the potential for significant complications. The data derived from multiple studies provide valuable insights into the optimal management strategies for these complex cases.

Patient Demographics and Perioperative Treatments

The patient demographics across the studies demonstrate a broad age range, indicating that AVMs can affect individuals at any stage of life. The use of perioperative treatments, particularly preoperative embolization, is a common strategy aimed at reducing intraoperative blood loss and improving surgical outcomes. Fujiki et al. (2018) and Hua et al. (2018) highlight the efficacy of embolization in minimizing the vascular supply to the AVM, thus facilitating more effective surgical resection. Preoperative embolization, using agents such as ethanol and n-BCA, has been shown to significantly decrease intraoperative blood loss and reduce the size of the AVM, making surgical excision more manageable. However, the variability in embolization agents and techniques across studies underscores the need for individualized treatment planning based on the specific vascular anatomy of the AVM and the patient's overall health [1-5].

Surgical Treatments and Reconstructions

Total resection remains the gold standard for treating AVMs, as evidenced by its widespread use across the reviewed studies. The approach aims to completely remove the abnormal vascular network, thereby reducing the risk of recurrence. For instance, Goldenberg et al. (2015) and Kumar et al. (2012) reported high rates of total resection, which were associated with favorable outcomes. Reconstruction techniques vary, with the use of local flaps, axial flaps, and free flaps being common. The choice of reconstruction method depends on the size and location of the defect left by the resection. For example, in the study by Fujiki et al., free flaps were utilized to cover large defects, ensuring adequate coverage and promoting healing. The use of free flaps, although technically demanding, provides robust vascularized tissue that enhances wound healing and functional outcomes [6-10].

Outcomes, Complications, and Follow-Up

The outcome measures across the studies indicate that total resection, when combined with appropriate perioperative management, leads to excellent clinical outcomes. Recurrence rates were generally low, with studies like those by Visser et al. (2011) and Lee et al. (2004) reporting no recurrences. This underscores the efficacy of complete surgical excision in achieving durable results. However, some studies, such as those by Kumar et al. (2012), reported higher recurrence rates, suggesting that incomplete resection or the presence of residual AVM tissue can lead to recurrence [11-15].

Complications were relatively infrequent but varied in severity. Major complications, such as significant blood loss and infection, were reported in a minority of cases. Minor complications, including transient swelling and wound infections, were more common but

generally manageable with conservative treatment. The mean follow-up periods across the studies varied widely, from as short as 12 months to as long as 252 months, highlighting the importance of long-term monitoring to detect and manage late recurrences or complications [16-20].

The comparison of our case with the literature reveals that the successful management of a giant, low-flow intramuscular AVM in the back through surgical resection aligns with the outcomes reported in other studies. Our case involved meticulous preoperative planning, including detailed imaging to map the vascular anatomy and identify the feeding and draining vessels. The use of intraoperative techniques to minimize blood loss and ensure complete resection was critical to the success of the procedure. The postoperative outcome, characterized by significant improvement in symptoms and quality of life, further validates the effectiveness of surgical resection as a definitive treatment for AVMs [21-25].

Comparative Literature

Comparing our findings with the literature, it is evident that the surgical resolution of AVMs, particularly those located in challenging anatomical regions like the back, requires a multidisciplinary approach. The integration of advanced imaging techniques, such as MRI and CT angiography, is essential for preoperative planning and intraoperative navigation. The studies reviewed emphasize the importance of a tailored approach, considering the individual patient's anatomy, the AVM's characteristics, and the available surgical and reconstruction options [21-28].

Our case adds to the body of evidence supporting the efficacy of surgical resection for intramuscular AVMs. The successful outcome, characterized by no recurrence and significant symptom improvement, aligns with the positive results reported in the literature. This reinforces the notion that, despite the inherent risks, surgical resection remains a viable and often necessary intervention for managing complex AVMs.

Conclusion

In conclusion, the management of intramuscular AVMs, particularly those in the back, presents significant challenges that require careful perioperative planning and execution. The literature supports the use of preoperative embolization and total resection as effective strategies to achieve favorable outcomes. Our case exemplifies the successful application of these principles, resulting in a transformed quality of life for the patient. Future research should continue to explore advancements in imaging, surgical techniques, and perioperative care to further improve the management of AVMs and reduce the associated risks and recurrence rates.

References:

1. Fujiki M, Ozaki M, Iwashina Y, et al. Clinical outcomes and recipient vessel selection for free flap transfer following arteriovenous malformation resection. J Plast Surg Hand Surg. 2019;53:56-59. E-published ahead of print December 5, 2018.
2. Goldenberg DC, Hiraki PY, Caldas JG, Puglia P, Marques TM, Gemperli R. Surgical treatment of extracranial arteriovenous malformations after multiple embolizations: Outcomes in a series of 31 patients. Plast Reconstr Surg. 2015;135:543-552.

3. Hua C, Yang X, Jin Y, et al. Treatment of head and neck arteriovenous malformations involving the facial nerve: A tailored algorithm. Ann Plast Surg. 2018;81(6S Suppl 1)
4. Kohout MP, Hansen M, Pribaz JJ, Mulliken JB. Arteriovenous malformations of the head and neck: Natural history and management. Plast Reconstr Surg. 1998;102:643-654.
5. Kumar R, Sharma G, Sharma BS. Management of scalp arteriovenous malformation: Case series and review of literature. Br J Neurosurg. 2012;26:371-377.
6. Lee BB, Do YS, Yakes W, Kim DI, Mattassi R, Hyon WS. Management of arteriovenous malformations: A multidisciplinary approach. J Vasc Surg. 2004;39:590-600.
7. Seccia A, Salgarello M, Farallo E, Falappa PG. Combined radiological and surgical treatment of arteriovenous malformations of the head and neck. Ann Plast Surg. 1999;43:359-366.
8. Visser A, FitzJohn T, Tan ST. Surgical management of arteriovenous malformations. J Plast Reconstr Aesthet Surg. 2011;64:283-291.
9. Liu AS, Mulliken JB, Zurakowski D, Fishman SJ, Greene AK. Extracranial arteriovenous malformations: Natural progression and recurrence after treatment. Plast Reconstr Surg. 2010;125:1185-1194.
10. Al-Qattan MM. Acquired localized subcutaneous cavernous vascular malformations of the hand. J Hand Surg Br. 2004;29:139-143.
11. Chewning RH, Monroe EJ, Lindberg A, et al. Combined glue embolization and excision for the treatment of venous malformations. CVIR Endovasc. 2018;1:22.
12. Hontanilla B, Qiu SS, Marre D. Surgical management of large venous malformations of the lower face. Br J Oral Maxillofac Surg. 2013;51:752-756.
13. Mendonca DA, McCafferty I, Nishikawa H, Lester R. Venous malformations of the limbs: The Birmingham experience, comparisons and classification in children. J Plast Reconstr Aesthet Surg. 2010;63:383-389.
14. Roh YN, Do YS, Park KB, et al. The results of surgical treatment for patients with venous malformations. Ann Vasc Surg. 2012;26:665-673.
15. Steiner F, FitzJohn T, Tan ST. Surgical treatment for venous malformation. J Plast Reconstr Aesthet Surg. 2013;66:1741-1749.
16. Park H, Kim JS, Park H, et al. Venous malformations of the head and neck: A retrospective review of 82 cases. Arch Plast Surg. 2019;46:23-33.
17. Bajaj Y, Hewitt R, Ifeacho S, Hartley BE. Surgical excision as primary treatment modality for extensive cervicofacial lymphatic malformations in children. Int J Pediatr Otorhinolaryngol. 2011;75:673-677.
18. Balakrishnan K, Menezes MD, Chen BS, Magit AE, Perkins JA. Primary surgery vs primary sclerotherapy for head and neck lymphatic malformations. JAMA Otolaryngol Head Neck Surg. 2014;140:41-45.
19. Fageeh N, Manoukian J, Tewfik T, Schloss M, Williams HB, Gaskin D. Management of head and neck lymphatic malformations in children. J Otolaryngol. 1997;26:253-258.

20. Jin L, Chen J, Li X. Surgical excision with bleomycin irrigation: A better primary treatment choice for pediatric submandibular lymphatic malformations. J Oral Maxillofac Surg. 2017;75:437.e1-437.e7.
21. Ma J, Biao R, Lou F, et al. Diagnosis and surgical treatment of cervical macrocystic lymphatic malformations in infants. Exp Ther Med. 2017;14:1293-1298.
22. Schoinohoriti OK, Theologie-Lygidakis N, Tzerbos F, Iatrou I. Lymphatic malformations in children and adolescents. J Craniofac Surg. 2012;23:1744-1747.
23. Theologie-Lygidakis N, Schoinohoriti OK, Tzerbos F, Iatrou I. Surgical management of head and neck vascular anomalies in children: A retrospective analysis of 42 patients. Oral Surg Oral Med Oral Pathol Oral Radiol. 2014;117
24. Wang S, Du J, Liu Y, et al. Clinical analysis of surgical treatment for head and neck lymphatic malformations in children: A series of 128 cases. Acta Otolaryngol. 2019;139:713-719.
25. de Serres LM, Sie KC, Richardson MA. Lymphatic malformations of the head and neck: A proposal for staging. Arch Otolaryngol Head Neck Surg. 1995;121:577-582.
26. Perkins JA, Manning SC, Tempero RM, et al. Lymphatic malformations: Review of current treatment. Otolaryngol Head Neck Surg. 2010;142:795-803, 803.e1.
27. Balakrishnan K, Edwards TC, Perkins JA. Functional and symptom impacts of pediatric head and neck lymphatic malformations: Developing a patient-derived instrument. Otolaryngol Head Neck Surg. 2012;147:925-931.
28. Horbach SE, Rigter IM, Smitt JH, Reekers JA, Spuls PI, van der Horst CM. Intralesional bleomycin injections for vascular malformations: A systematic review and meta-analysis. Plast Reconstr Surg. 2016;137:244-256.