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Surgical management of extensive facial vascular malformation with skin graft: A multidisciplinary approach—Case report

Shailendra Katwal[1], Pratiksha Paudel[2], Bidur Khatiwada[3], Srikant Agrawal[4] and Sahadev Bidari[3]

Abstract

Vascular malformations are rare congenital abnormalities of blood vessels that persist throughout life. Large vascular malformations affecting the facial region can be distressing and require meticulous management to achieve satisfactory outcomes and prevent recurrence. Here, we present a case of a 40-year-old man with a progressively growing tumor-like mass on the right side of his face, extending from the periorbital region to the chin. He was diagnosed with vascular malformations, which was further confirmed with contrast-enhanced magnetic resonance imaging. Eventually, he underwent surgical excision of the lesion with a split-thickness skin graft. The surgical management of this case was challenging due to the large size and complexity of the lesion, particularly because it was located in the facial region. This unique case report highlights the importance of diagnostic imaging techniques and effective surgical management in addressing large vascular malformations, especially those affecting the facial region while prioritizing desirable cosmetic outcomes.

Keywords

Dermatology, surgery, radiology, vascular malformations, lymphovenous, skin graft, blood vessel

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Introduction

Vascular malformations (VMs) are abnormalities in the structure of blood vessels that are present at birth and continue throughout life. VMs can be categorized based on the type of blood vessel involved.[1] VMs can occur in various parts of the body and vary in size, appearance, and symptoms. Head and neck areas are the most common sites for vascular and lymphatic malformations.[2] VMs are often asymptomatic but may present with rash or tumor-like growth, with cosmetic issues commonly observed in the facial region. Ultrasound and magnetic resonance imaging (MRI) are the preferred imaging modalities for the diagnosis of VMs.[3] Sclerotherapy, surgical excision and repair, radiofrequency ablation, and laser are among the treatment options available. Here, we present a case of a 40-year-old male with a rare lymphovenous malformation on the right side of his face, who underwent surgical excision and repair with a split-thickness skin graft.

Case presentation

A 40-year-old man presented to our dermatology clinic with a purplish, tumor-like growth on the right side of his face, which had been present since birth and had been growing gradually over several years. The lesion was non-blanching and was not accompanied by pain, fever, or itchiness. He had no history of seizures, breathing problems, or headaches. He had no developmental issues or growth problems and he has always been a non-smoker. There are no similar lesions in his family. Initially not bothersome, the growth began to affect his eyes, nose, and upper lip as it expanded, causing him distress. Over time, it spread extensively across the upper face to the chin while respecting the boundaries of the midline and ear, resulting in significant cosmetic concerns and discomfort in the affected area as assessed on subjective examination.

[1]Dadeldhura Subregional Hospital, Amargadhi, Dadeldhura, Nepal
[2]All Nepal Hospital, Kathmandu, Nepal
[3]Nepalese Army Institute of Health Sciences, Kathmandu, Nepal
[4]National Academy of Medical Sciences, Bir Hospital, Kathmandu, Nepal

Corresponding Author:
Bidur Khatiwada, Nepalese Army Institute of Health Sciences, Bhandarkhal, Kathmandu 44600, Nepal.
Email: drbidurkhatiwada@gmail.com

Upon examination, his blood pressure was 130/80 mmHg, and his pulse rate was 88 beats per minute. There were no pallor, icterus, cyanosis, lymphadenopathy, clubbing, or edema. On examination of his chest, bilateral air entry was normal, and equal and bilateral normovesicular sounds were heard. The abdomen was soft and non-tender, with the presence of bowel sounds. Upon neurological examination, his higher mental functions, 12 cranial nerves, and sensory and motor examinations were normal. On local examination, a large, rough tumor with red lobes of varying sizes was observed on the right side of his face (Figure 1). These lobes obstructed eye-opening and distorted the nose. They appeared purplish, shiny, lobular, pedunculated, and densely clustered together, with no signs of overlying inflammation. The ultrasound indicated a low-grade VM, revealing an ill-defined hetero-echoic area on the right side of the face. On contrast-enhanced computed tomography (CT), the mass displayed nodular and tubular enhancing structures, fat attenuation, and multiple scattered calcifications (Figure 2). Contrast-enhanced MRI revealed a lobulated soft tissue intensity lesion within the cutaneous and subcutaneous tissues on the right side of the face, extending superiorly to the preorbital region and caudally to the submental region (Figure 3). Few signal voids were observed within it. His complete blood count revealed a white blood cell count of 6000/mL and hemoglobin of 14.8 g/dL. Additional investigations revealed a C-reactive protein level of 5 mg/dL.

Subsequently, a multidisciplinary team of dermatologists, plastic surgeons, and ear, nose, and throat surgeons specialized in dealing with VMs performed the surgical excision of the lesion. Damage to the facial nerve was avoided with precise surgical techniques and intraoperative nerve monitoring. His intraoperative period was unremarkable with no instance of heavy bleeding. After the surgical excision, the wound was closed using a split-thickness skin graft (STSG). The histopathology examination of the excised mass confirmed the presence of venous and lymphatic components. His postoperative period was uneventful, and he was discharged on the fifth postoperative day. After 2 months, during a follow-up appointment at the Outpatient Department, it was noted that the skin graft was healing normally. He was satisfied with the treatment outcome. The repeat ultrasound of the right side of his face showed no findings suggestive of recurrence or vascular pathology. The patient refused the follow-up MRI due to financial constraints. The patient missed the scheduled follow-up appointment 3 months later.

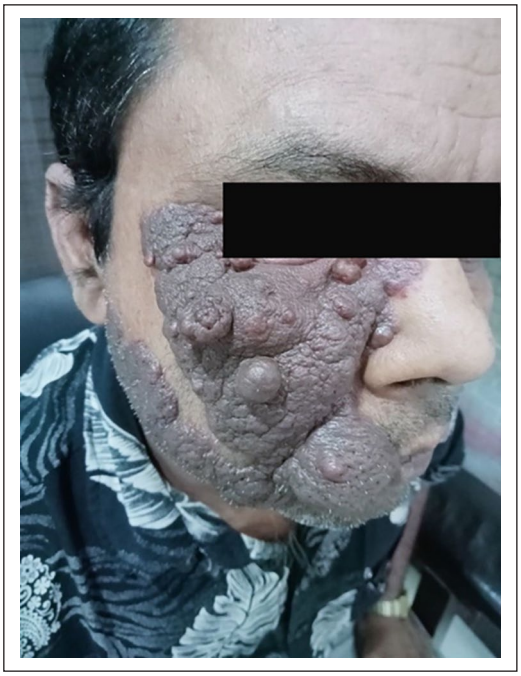

Figure 1. Purplish-colored nodular growth densely clustered on the right side of the face extending from the lower eyelid to the chin.

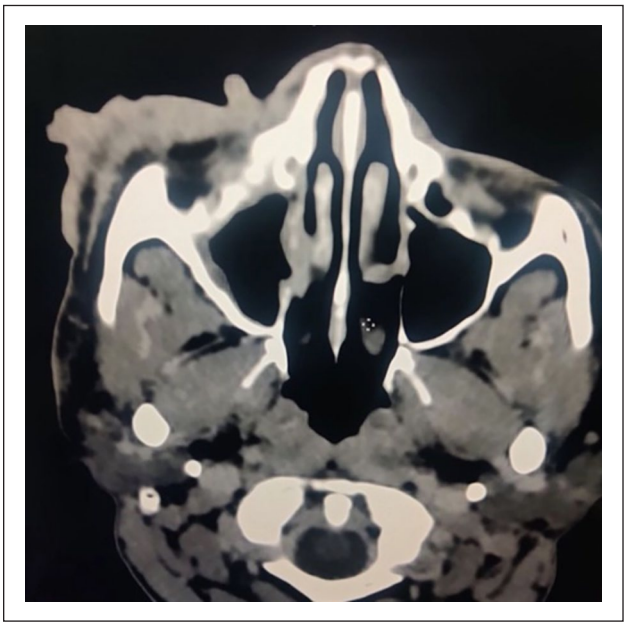

Figure 2. Post-contrast axial computed tomography images show enhancing nodular sheet-like mass on the right side of the face over the premaxillary and dorsum of the nose.

Discussion

VMs are structural abnormalities of the vascular system that are present from birth and persist throughout life.[2] Mulliken and Glowacki's classification system divided various vascular anomalies based on their histopathological makeup into vascular tumors and malformations, which can further be categorized based on the type of vessel involved (capillary, venous, lymphatic, and arteriovenous).[4] Venous and lymphatic malformations are among the most prevalent VMs found in the head and neck region.[2] Venous malformations in the head

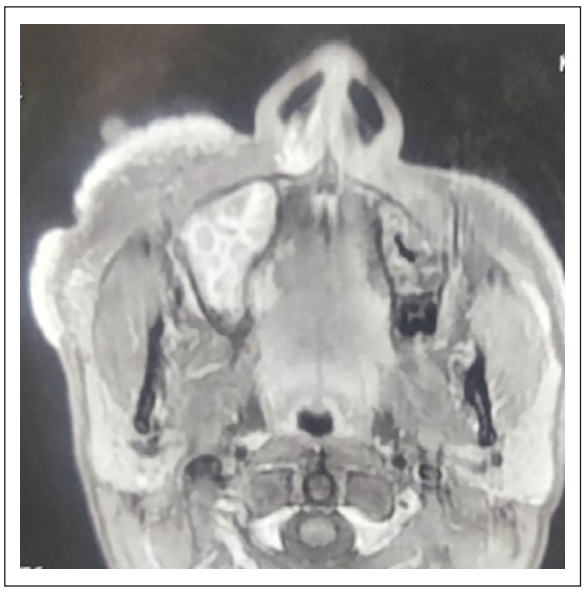

Figure 3. Axial T2-weighted magnetic resonance imaging image shows high signal intensity lobulated soft tissue lesion involving the cutaneous and subcutaneous plane of the right side of the face.

and neck regions constitute more than 40% of cases of VMs.[2] They occur equally in males and females.[5] VMs are unique in their lifelong growth without signs of natural regression or reduction, setting them apart from other conditions.[5] The main differential diagnosis includes other low-flow vascular anomalies such as capillary malformations and high-flow vascular anomalies (arterial or mixed arterial-venous).[2]

VMs often remain asymptomatic and may come to attention during childhood, adolescence, or early adulthood, depending on their size and location. They can present as a rash, tumor-like growth, or even discomfort such as dull aches aggravated by physical activity or temperature shifts, depending on their location. Lesions commonly appear as soft, compressible masses without pulsation, often displaying a bluish discoloration. Rarely, VMs can present with hemorrhage that requires urgent medical attention. A detailed medical history and thorough clinical examination are usually sufficient for diagnosis.[2] In our case, the patient presented with a purplish tumor-like growth on the right side of the face, which caused significant distress and cosmetic issues.

Most VMs are diagnosed based on clinical history and examination. However, in larger and deeper lesions, imaging aids in confirming the diagnosis, assessing morphology, and strategizing treatment. Ultrasound is the preferred initial investigation in outpatient settings due to its cost-effectiveness and easy availability. While it has limitations in visualizing deeper lesion components, it is crucial for determining the composition of lesions in terms of vascular space versus cellular matrix ratio.[3] Contrast-enhanced MRI is the imaging modality of choice due to its superior soft tissue resolution and ability to delineate the morphology of lesions, often revealing flow voids.[2,3,6] Biopsy is essential for a definitive diagnosis, but it carries a risk of bleeding due to abundant vascular flow. In our case, ultrasound revealed a low-grade VM on the right side of the face, while contrast-enhanced CT and MRI confirmed nodular structures with scattered calcifications and soft tissue lesions extending from the preorbital to submental regions.

Effective management of VMs is essential to minimize the risk of severe bleeding and enhance the quality of life for affected individuals. As there is no single preferred method, multidisciplinary approaches are advocated, and treatment decisions should be individualized based on each case. Before initiating active treatment, factors such as size, location, depth, and proximity to adjacent structures should be carefully evaluated. Medical treatments aim to mitigate symptoms such as venous stasis, vascular expansion, and thrombus formation, using options like low-molecular-weight heparin and compression garments, although their efficacy may be limited.[2] Sclerotherapy is often used as an initial nonsurgical treatment option for symptomatic patients, but its efficacy lacks conclusive evidence.[7]

Surgical excision is recommended for symptomatic and localized large lesions, failure of nonoperative methods, or persistent aesthetic concerns. The primary surgical options for VMs include radical excision, debulking surgery, and marginal vein resection.[8] In cases of cervicofacial lesions, surgery may prioritize debulking procedures over complete eradication, especially for diffuse, symptomatic lesions that cannot be entirely removed.[9] The surgical excision of the VMs should involve a skin-sparing incision to minimize disfigurement, followed by primary closure whenever feasible. Full-thickness skin grafts are preferred for the face, while split-thickness grafts (STSGs) are effective for superficial tissue losses due to their ability to cover large areas, promote quicker healing with minimal donor site morbidity, and achieve satisfactory cosmetic results.[10]

Surgical excision of facial VMs can pose the risk of major blood loss, iatrogenic injury, deformity, facial nerve injury, and incomplete resection.[11] Following the initial excision, there is a 50% recurrence rate, primarily attributed to residual anomalous tissue.[12] Other treatment options encompass laser treatment and high-frequency radio-wave ablation of the lesions. Nevertheless, there is a lack of conclusive data supporting their efficacy.[2] In our case, surgical excision of the lesion was performed based on the patient's preference, the large size of the lesion, and aesthetic concerns. STSG was selected for its ability to cover large areas, expedite healing, minimize donor site morbidity, and provide favorable cosmetic outcomes. The patient was followed up after 2 months postoperatively and subsequent examination revealed a normally healing skin graft. On subjective assessment, the patient expressed satisfaction with the cosmetic outcome. The repeat ultrasound of the right side of the face showed no findings suggestive of recurrence or vascular pathology. The patient was

scheduled for a subsequent follow-up appointment after 3 months but he was lost to follow-up.

Conclusion

This case report highlights the importance of thorough knowledge and a comprehensive understanding of lesions, as well as diagnostic and treatment methods, in the management of VMs. Accurate diagnosis and tailored, meticulous intervention form the cornerstone in the treatment of extensive lesions involving the facial region. Our case further underscores the significance of adopting a multidisciplinary approach in achieving satisfactory cosmetic outcomes, especially in similar clinical circumstances.

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None.

Author contributions

S.K. and P.P. designed the study and collected and analyzed the data. P.P. and B.K. wrote the manuscript. All authors participated in the manuscript revision. They all approved the final version for publication and agreed to take accountability for all aspects of the work.

Data availability

In this case, the data supporting the findings are available upon request to the corresponding author.

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Ethical approval

Our institution does not require ethical approval for reporting anonymized case reports.

Informed consent

Written informed consent was obtained from the patient(s) for their anonymized information to be published in this article.

ORCID iDs

Shailendra Katwal https://orcid.org/0000-0001-7903-8789
Bidur Khatiwada https://orcid.org/0000-0003-4650-889X
Srikant Agrawal https://orcid.org/0000-0002-0371-4821

References

1. Almeida de Arruda JA, Andrade ES, Andrade RA, et al. Vascular malformation of the face: a rare case with follow-up of 18 years. *Acta Stomatol Croat* 2017; 51(4): 332–335.
2. Fowell C, Verea Linares C, Jones R, et al. Venous malformations of the head and neck: current concepts in management. *Br J Oral Maxillofac Surg* 2016; 55: 3–9.
3. McCafferty IJ and Jones RG. Imaging and management of vascular malformations. *Clin Radiol* 2011; 66(12): 1208–1218.
4. Mulliken JB and Glowacki J. Hemangiomas and vascular malformations in infants and children: a classification based on endothelial characteristics. *Plast Reconstr Surg* 1982; 69(3): 412–422.
5. Muqtadir F, Hiremath S, Agarwal S, et al. Vascular malformation of cheek: a case report. *J Evol Med Dent Sci* 2015; 4(15): 2617–2620.
6. Munechika M, Tobino K, Okahisa M, et al. A case of huge lymphatic and venous malformations of the mediastinum. *Respir Med Case Rep* 2018; 26: 53–55.
7. Chen AW, Liu YR, Li K, et al. Efficacy of sclerotherapy with radio-opaque foam guided by digital subtraction angiography for the treatment of complex venous malformations of the head and neck. *Br J Oral Maxillofac Surg* 2015; 53(9): 809–813.
8. Roh YN, Do YS, Park KB, et al. The results of surgical treatment for patients with venous malformations. *Ann Vasc Surg* 2012; 26(5): 665–673.
9. Ramakrishnan K, Palanivel I, Narayanan V, et al. Management of vascular malformations in the oral and maxillofacial region: a systematic review. *J Stomatol Oral Maxillofac Surg* 2021; 122(6): 588–599.
10. Bhandari PS, Sadhotra LP, Bhargava P, et al. Management strategy for facial arteriovenous malformations. *Indian J Plast Surg* 2008; 41(2): 183–189.
11. Li J, Chen J, Zheng G, et al. Digital subtraction angiography-guided percutaneous sclerotherapy of venous malformations with pingyangmycin and/or absolute ethanol in the maxillofacial region. *J Oral Maxillofac Surg* 2010; 68(9): 2258–2266.
12. Bai Y, Jia J, Huang XX, et al. Sclerotherapy of microcystic lymphatic malformations in oral and facial regions. *J Oral Maxillofac Surg* 2009; 67(2): 251–256.