ORIGINAL ARTICLE

# Multidisciplinary vascular malformations clinic in Hong Kong

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**Objective** To review clinical characteristics, imaging modalities, and treatment outcomes of patients referred to a multidisciplinary clinic for management of vascular malformations.

**Design** Retrospective case series.

**Setting** Multidisciplinary vascular malformation out-patient referral clinic in a teaching hospital in Hong Kong.

**Patients** The 141 attendees of the clinic from August 2005 to November 2011.

**Main outcome measures** Management and treatments offered, and responses to treatment.

**Results** Of the 141 patients, 46% were diagnosed to have low-flow vascular malformations, 16% were diagnosed to have high-flow vascular malformations, and 15% were diagnosed to have a haemangioma. Prior to attending the clinic, approximately one third (32%) of the patients had a clinical diagnosis that was consistent with the final diagnosis. Overall, the radiological and clinical diagnoses were consistent in 43% of the patients. Magnetic resonance imaging and ultrasonography were the most commonly used imaging modalities. Of the 73 patients who received active treatment, 70% had a good response, 12% had minimal improvement, 8% had no change, and 7% had a recurrence or a major complication; in 3% of the patients the outcome was unknown.

**Conclusion** From this retrospective case series, it is evident that confusion still exists over vascular malformations and haemangiomas. Multidisciplinary clinics have a role in providing an accurate diagnosis and facilitating appropriate management and treatment plans. Magnetic resonance imaging and ultrasonography had demonstrable utility in determining the extent of the lesions and flow type.

**New knowledge added by this study**

- Vascular lesions can be broadly divided into vascular malformations and haemangiomas.
- The preferred management of these two categories of vascular lesion is very different.
- A significant number of medical practitioners are unaware of or unable to make an appropriate clinical diagnosis of such vascular lesions.

**Implications for clinical practice or policy**

- Multidisciplinary teams (MDTs) should be established to manage patients with such complex vascular lesions.
- Patient management by MDTs is likely to involve more experienced caregivers and provide more evidence-based treatment options whilst reducing the number of hospital visits.

**Key words**
Hemangioma; Patient care team; Port-wine stain

*Hong Kong Med J 2013;19:116-23*

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## Introduction

Vascular malformations are congenital developmental abnormalities in vascular anatomy that cause significant morbidity.[1] They are often confused with haemangiomas. Mulliken and Glowacki[2] described distinct characteristics that differentiate these two anomalies, including their natural history and histology. This classification system was later updated by the International Society for the Study of Vascular Anomalies.[3] Haemangiomas are vascular tumours involving hyperplasia of the endothelial cells. They are usually absent at birth, but

become evident shortly after, and are characterised by a rapid proliferative phase followed by slow involution. Conversely, vascular malformations are present at birth, grow proportionately with the child and do not regress spontaneously. Vascular malformations can be further subdivided according to flow type.[4] High-flow types include arterial malformations, as well as arteriovenous fistulas and malformations. Low-flow types include venous, lymphatic, capillary and mixed malformations.

Confusion over the classification of vascular malformations can lead to inappropriate management, such as waiting for 'haemangiomas' to regress spontaneously.[5] Various centres therefore run multidisciplinary clinics dedicated to the diagnosis and management of such vascular lesions.[1,4] Expertise from a multidisciplinary team facilitates accurate diagnoses, and helps guide imaging and treatment decisions. In addition, joint clinics also eliminate the need for multiple visits to see different specialists, thus avoiding conflicting diagnoses and treatment plans.

At the Prince of Wales Hospital (PWH) in Hong Kong, a multidisciplinary Vascular Malformation Clinic (VMC) was established in 2005, with the


## 香港的跨學科血管畸形診所

**目的** 探討被轉介到跨學科血管畸形診所的病人的特徵、其放射影像模式及治療效果。

**設計** 回顧研究。

**安排** 香港一所大學教學醫院的跨學科血管畸形轉介門診部。

**患者** 於2005年8月至2011年11月期間141位被轉介至以上診所的病人 。

**主要結果測量** 所給予的治療，以及病人的治療反應。

**結果** 141名病人中，46%被診斷患有低流量型血管畸形，16%有高流量型血管畸形，15%患有血管瘤。32%的病人在跨學科血管畸形診所的診斷結果與轉介前的診斷吻合。放射診斷與臨床診斷只有43%吻合。磁力共振成像與超聲是最常用的放射影像模式。73位病人接受了積極治療，其中70%有理想效果，12%有輕微進步，8%無變，7%有復發或併發症，3%的成效未知。

**結論** 本報告顯示血管瘤與血管畸形的分類仍然比較混亂。跨學科門診有助提供正確的診斷以及協調適當的治療模式。磁力共振與超聲波可以用來分析病灶範圍及流量型，從而有助分辨血管畸形與血管瘤。


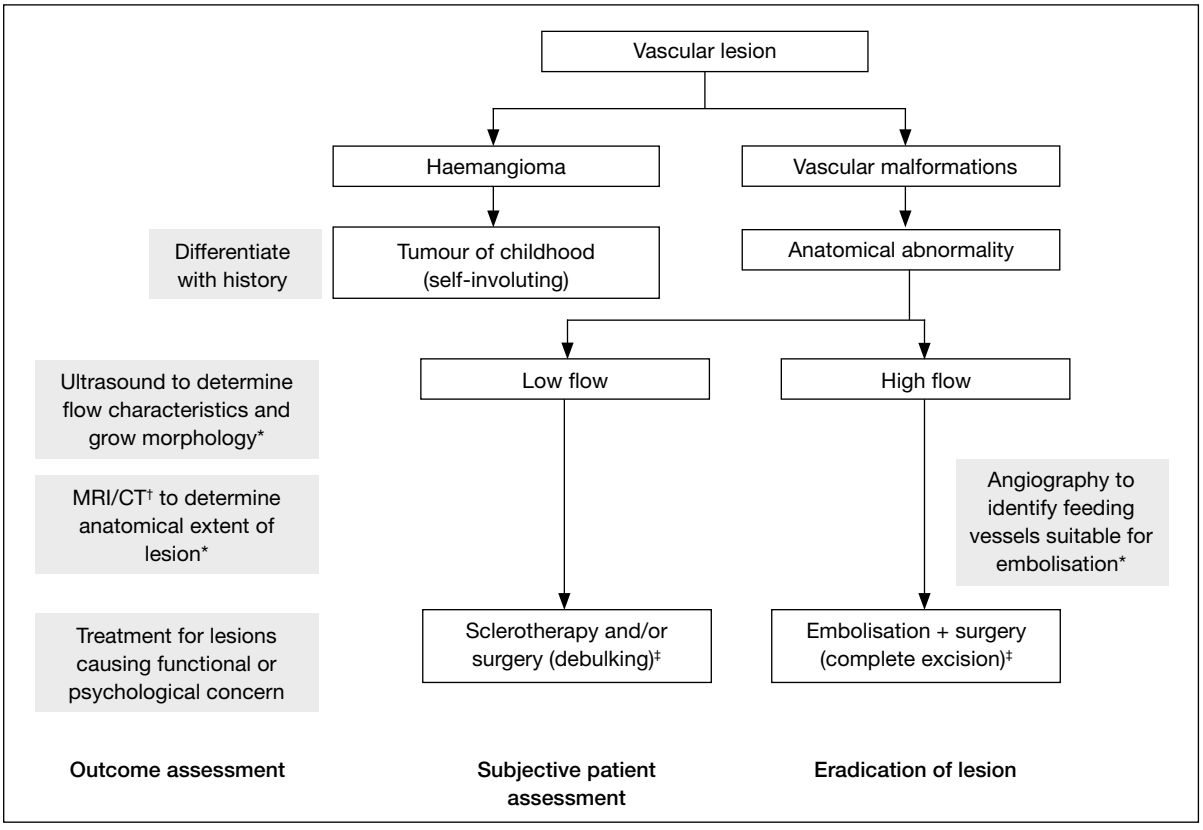


FIG 1. Simplified algorithm depicting the management flow to deal with vascular lesions

* Imaging diagnosis noted

† MRI denotes magnetic resonance imaging, and CT computed tomography

‡ Tissue diagnosis noted

intention of managing vascular malformations as distinct from haemangiomas, which are most often dealt with by paediatricians. Referred patients are seen jointly by plastic, vascular, and orthopaedic surgeons, together with an interventional radiologist. The collective clinical diagnosis is made based on the history and physical examination. Further radiological investigations may be ordered to substantiate the diagnosis and guide management. The imaging modalities used include ultrasound, magnetic resonance imaging (MRI), computed tomography (CT), and angiography. Conservative or active treatment options are then offered, based on patient preferences and suitability (Fig 1). Notably, capillary vascular malformation (also known as port-wine stains) are routinely seen in laser clinics and not referred to the VMC.

## Methods

Electronic records of patients seen at the PWH VMC from August 2005 to November 2011 were reviewed retrospectively. A database was created using Microsoft Access, with fields including patient age, gender, first VMC attendance, anatomical location of lesion, type of malformation (diagnosis at clinic), previous diagnosis, treatments offered, outcomes, as well as radiological and pathological findings. Clinic notes, and radiology and pathology reports were reviewed to input relevant data into the various fields.

Data analysis was performed using Microsoft Excel to outline characteristics of patients seen at the clinic, including the diagnosis, age at onset, and anatomical location of lesion. The management and treatment options undertaken and offered were evaluated. The diagnosis noted in the radiology and pathology reports were cross-checked for consistency with the clinical diagnosis.

Outcomes of patients who underwent active treatment after attendance at the VMC were divided into three categories: 'good response', 'minimal improvement', or 'no change'. Outcomes were based on the subjective evaluation of the patients as noted in the clinical record. Good response referred to complete lesion reduction with optimal functional and cosmetic outcomes. Minimal improvement referred to a suboptimal cosmetic outcome. No change referred to static lesions and those that recurred. Patients having a recurrence or any complication were recorded.

Outcomes were determined based on subjective comments from both the clinician and patient perspective, and had been recorded in the notes at the latest follow-up visit to the multidisciplinary clinic. There were no objective measures included in this study.

## Results

### Patient characteristics

A total of 141 patients had attended the VMC over the 7-year study period; 62% were female and 37% were children (Table 1). The head and neck was the most common anatomical location for vascular lesions. Venous vascular malformations (VVMs) were the most common, followed by arteriovenous malformations (AVMs), lymphatic, capillary, and mixed types (Table 2). There was one case with a lymphatic malformation and a VVM at a different anatomical location, in addition to 19 cases of non-specific vascular malformations. Cases were classified as 'non-specific' when there was no assigned type in the notes or evidence of flow type. There were four patients diagnosed with Sturge-Weber syndrome (SWS) and four with Klippel-Trenaunay syndrome. For lesions not classified under the term vascular malformation, haemangioma was the most common diagnosis, of which 62% were in females.

Only 32% had a previous clinical diagnosis that was consistent with the diagnosis at the VMC. Notably, 54% of the vascular malformations diagnosed at the VMC were previously diagnosed as haemangiomas. In contrast, all patients previously labelled as having a haemangioma had been correctly diagnosed. Overall, the radiological and clinical diagnoses were consistent in 43% of the patients.

Swelling and pain were the most common complaints associated with these lesions; 35% and 20% of patients presented with these symptoms, respectively. Whilst 8% presented with enlarging lesions, 4% presented with recurrences after

TABLE 1. Demographic data and predominant locations of lesions

| Demographics/lesion location | No. (%) of patients (n=141) |
|---|---|
| Children (<18 years old) | 52 (37) |
| Gender | |
| Female | 87 (62) |
| Male | 54 (38) |
| Locations | |
| Head and neck | 61 (43) |
| Oropharynx | 4 (3) |
| Upper extremities | 19 (13) |
| Lower extremities | 30 (21) |
| Trunk | 8 (6) |
| Groin, pelvis, and perineum | 3 (2) |
| Oral, head, neck | 6 (4) |
| Upper extremity and trunk | 3 (2) |
| Head, neck, and upper extremity | 2 (1) |
| Other combinations of multiple locations | 5 (4) |

previous surgery or other treatment. Notably 8.5% patients indicated concerns with disfigurement, 7.1% presented with bleeding lesions, and 4% reported discomfort or itch; 6% had functional impairment in relation to the lesion. The latter included sleep disturbance, obstruction of vision, speech impairment, decreased range of movement, and obliteration of ears (due to increasing lesion size).

### Management and treatment outcomes

Further imaging investigations were performed on 74% of all patients referred to the VMC, of which MRI was the most commonly used (for 79%), followed by ultrasound (for 59%), angiography (for 26%), and CT (for 16%).

Active treatment was undertaken for 52% of all patients, whilst 48% received conservative treatment, including observation and pressure garments (Table 3). Among active treatments, surgery was used in 22% of the patients, followed by sclerotherapy in 12%. Other modalities include embolisation, laser therapy, intense pulsed light (IPL), and propranolol.

Regarding high-flow AVMs, 73% of the patients undergoing active treatment achieved a good response, but 20% had minimal improvement. Surgery was the most common treatment, with a good response in seven out of eight treated patients (Table 3). Embolisation was used in three cases, two of whom had a good response and minimal improvement ensued in one. Two patients received sclerotherapy, with a good response in one and minimal improvement in the other.

For low-flow vascular malformations, 75% of patients undergoing active treatment had a good response, 8% showed minimal improvement, 10% had no change, and 8% had a recurrence. For VVM, sclerotherapy was successful for all patients treated. In the majority, the sclerosant used was sodium tetradecyl sulfate (STS); ethanol was used in one patient. Surgery was also a popular treatment option for VVM patients; 82% achieved a good response and 18% resulted in recurrence or residual disease. Pressure garments were used with good effect in two patients.

For capillary malformations, there was no change in the patient treated with IPL. Sclerotherapy appeared successful in one patient, but another had a suboptimal cosmetic outcome. One patient received a combination of sclerotherapy and surgery. Of the three patients diagnosed with SWS, successful treatment was achieved with dermal laser therapy in one, surgery in another, and pulsed dye laser (PDL, Vbeam) in one who had had previous IPL and surgery. All five lymphatic malformations were treated surgically and had successful outcomes.

Haemangiomas were mainly treated conservatively; seven out of 11 displayed involution over the course of follow-up at the clinic, and four exhibited no change. Three were treated surgically and had a good response. Two were treated with sclerotherapy, one received propranolol and four received multiple modalities of treatment.

TABLE 2. Diagnoses at vascular malformation clinic

| Diagnosis | No. (%) of patients (n=141) |
|---|---|
| High flow | |
| Arteriovenous malformation | 22 (16) |
| Low flow | |
| Venous malformation | 46 (33) |
| Lymphatic malformation | 10 (7) |
| Capillary malformation | 6 (4) |
| Mixed malformation* | 3 (2) |
| Multiple-type malformations at different sites | 1 (1) |
| Complex | |
| Sturge-Weber syndrome | 4 (3) |
| Klippel-Trenaunay syndrome | 4 (3) |
| Haemangioma | 21 (15) |
| Other | |
| Vascular malformation (uncategorised)† | 19 (13) |
| Varicose veins | 2 (1) |
| Lipoma | 1 (1) |
| Lipoblastoma | 1 (1) |
| Enlarged lymph nodes | 1 (1) |

* All mixed vascular malformations contained lymphatic and venous components

† Uncategorised vascular malformations that were not classified at initial assessment

TABLE 3. Management/treatment decisions*

| Management/treatment | No. (%) of patients | | | |
|---|---|---|---|---|
| | High-flow VM (n=22) | Low-flow VM (n=65) | Haemangioma (n=21) | All patients (n=141) |
| Further diagnostic workup | 20 (91) | 61 (94) | 16 (76) | 104 (74) |
| Conservative treatment | | | | |
| Observation | 3 (14) | 21 (32) | 11 (52) | 61 (43) |
| Pressure garment | 0 | 5 (8) | 1 (5) | 7 (5) |
| Active treatment | | | | |
| Surgery | 8 (36) | 17 (26) | 3 (14) | 31 (22) |
| Sclerotherapy | 2 (9) | 13 (20) | 2 (10) | 17 (12) |
| Embolisation | 3 (14) | 2 (3) | 0 | 5 (4) |
| Laser | 0 | 1 (2) | 0 | 4 (3) |
| Intense pulsed light | 0 | 1 (2) | 0 | 1 (1) |
| Propranolol | 0 | 0 | 1 (5) | 2 (1) |
| Other | 0 | 0 | 1 (5) | 1 (1) |
| Multiple | 1 (5) | 5 (8) | 4 (19) | 12 (9) |

* VM denotes vascular malformation

In all, five patients had recurrences of their vascular malformations. Only one patient (with a lower-limb AVM) suffered significant complications culminating in amputation, following multiple recurrences treated by embolisation and surgery. This patient also had heart failure. Recurrence with progressive enlargement occurred in three patients with VVMs. In one of these, there was clinically noticeable enlargement 3 years post-excision, for which sclerotherapy was offered. Another entailed progressive enlargement evident on radiological scans 4 years postoperatively, but the patient declined further treatment. The remaining patient with a VVM on the left thigh was managed conservatively with pressure garments and massage. Though radiology suggested progressive enlargement, the patient did not consent to any other form of active treatment. One patient experienced subjective recurrence of a haemangioma in his finger, however no further treatment was pursued as there was no lesion causing functional or cosmetic disturbance. In summary, of the 73 patients who received active treatment, 70% had a good response, 12% had minimal improvement, 8% had no change, and 7% had a recurrence or a major complication; in 3% of the patients the outcome was unknown.

## Illustrative case

A healthy female infant developed a swelling of her left face at the age of 30 days. This rapidly increased in size and the mother sought help. The baby was admitted to the Plastic Surgery Service at the PWH with a 6-cm diameter bluish mass in the left parotid region, with normal overlying skin. The patient was diagnosed to have a subcutaneous haemangioma and referred to paediatricians for treatment with propranolol. Regrettably the lesion did not regress,[6] for which reason intralesional steroid therapy was given but again there was no response. The mother sought the advice of various doctors in the Mainland, all of whom recommended surgery and she returned to the PWH VMC. Figure 2a shows the 7-month-old child with a lesion over the left cheek. An MRI indicated large feeding and draining vessels medially and a lesion that appeared to arise from and displacing the parotid gland. Medially the lesion extended to the left parapharyngeal fat. The patient was admitted for digital subtraction angiography, which revealed a highly vascularised lesion with a tortuous feeding vessel arising from the left external carotid artery with multiple draining veins (Fig 2b). No embolisation was performed. A radical excision was performed 2 days later, whereby a large vascular


FIG 2. (a) Preoperative view with an expanding lesion not responsive to propranolol or intralesional steroids. (b) Large medial feeding vessels. (c) Skin reflected to reveal a subcutaneous lesion with a surface element resembling a typical 'strawberry naevus'. (d) The lesion isolated on the vascular pedicle

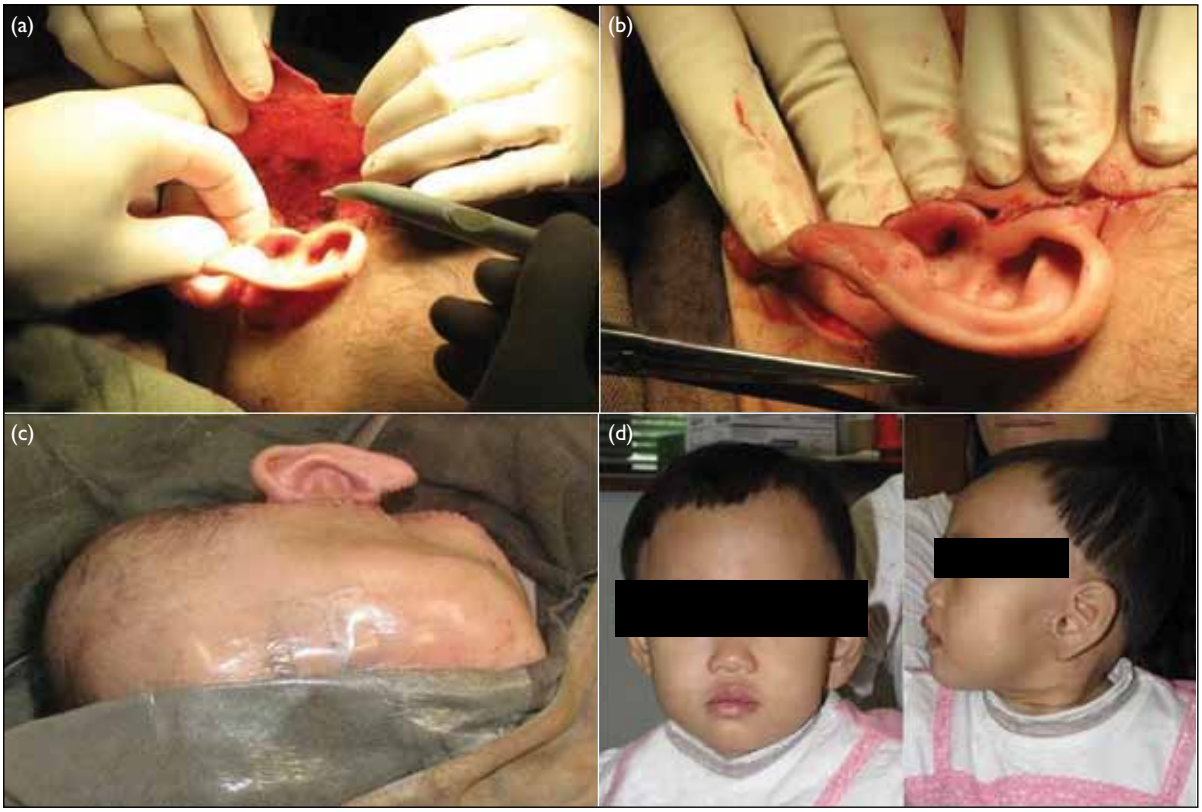


FIG 3. (a) Allogenic fibrin sealant prepared from the mother's blood using the Vivostat (Vivostat, Denmark) system was sprayed into the concave defect. (b) The skin was moulded into shape. (c) The wound was closed without a drain. (d) Seven months postoperation, the concave deformity remains. Autologous fat augmentation combined with revision of the ear lobe will be performed later

lesion 10 x 7 x 5.5 cm was removed (Figs 2c and 2d). The skin was redraped in the concave excision bed and fixed using fibrinogen tissue glue prepared from the mother's plasma (Figs 3a-3c). The child made an uneventful recovery and was last reviewed 7 months postoperatively (Fig 3d) when the mother confirmed that the child's speech was developing well. Moreover, oral competence was intact and eye closure could be achieved, but there was still residual weakness of buccal muscles (innervated by a branch of the facial nerve). Pathology of the lesion was reported as a capillary haemangioma.

## Discussion

This is the first case review of patients presenting to a VMC in Hong Kong. Despite existing classification systems for such anomalies, in this case series the high number of inconsistent diagnoses prior to attending the clinic indicates that confusion still exists. Confusion with haemangiomas can lead to delayed management of lesions such as port-wine stains, which are low-flow vascular malformations. If not treated early, they can progress to become nodular and refractory to treatment.[7] In addition, port-wine stains are associated with SWS, whereby patients can develop seizures, developmental delay, and glaucoma; if left undetected the patients can become blind. This reinforces the need for more education regarding early accurate diagnoses of vascular lesions. It is also distressing to encounter adults waiting for their 'haemangiomas' to spontaneously involute, which will not happen if the lesion is a vascular malformation. Part of the role of the clinic is to increase undergraduate and postgraduate awareness of the importance of appropriate diagnoses of vascular lesions.

Radiological investigations are essential to determine the nature and extent of vascular malformations accurately prior to surgery, interventional radiology, or other treatment.[8] The most commonly used imaging modality in this case series was MRI, which is consistent with current literature outlining its benefits in planning resection or other therapy, as physical examination alone often underestimates the anatomical extent of vascular malformations.[9] Recourse to multiplanar MRI views also provides better soft tissue contrast and is considered the best modality for identifying large vascular lesions, and the addition of MR angiography can help to differentiate low- and high-flow malformations.[10]

Ultrasound was the other main imaging

modality used at the VMC. Various studies support the use of ultrasound as the initial investigation of choice, due to its easy access, low cost, ability to assess lesion haemodynamics, and facilitate distinguishment between most vascular malformations from haemangiomas.[8,11] Ultrasound is also particularly useful for low-flow lesions, as it allows determination of the vascular space to cellular matrix ratio. Lesions predominantly consisting of vascular spaces are thought to respond better to sclerotherapy.[12] In our series, angiography was mainly used for therapeutic embolisation of AVMs. Its advantages include visualisation of vessel anatomy, arteriovenous shunting and fistulas, and as an aid to plan embolisation therapy. However, it is a more invasive procedure than the alternatives.[13] The least utilised imaging modality in our clinic was CT, which was mainly used to assess suspected bone involvement. Recent reviews suggest a limited role for CT in the workup for vascular malformations, due to the unnecessary ionising radiation exposure it entails and the inferior soft tissue detail it provides in comparison to MRI.[12,13] Nevertheless, it is a sensitive means of detecting intralesional phleboliths (characteristic of VVMs) and for the assessment of bony involvement.[12]

It is acknowledged that this is a retrospective review and that outcomes were based on the subjective assessments detailed in the medical record. Ideally, a prospective assessment could include more objective evaluation.

In this case series, 70% of all patients reported good responses to active treatment. For high-flow vascular malformations, surgery was the main treatment, and yielded good outcomes in 88% of those treated. Complete surgical excision with immediate reconstruction is accepted as the gold-standard treatment for AVMs and is reported to be the most successful for small, localised lesions.[14] Consistent with our experience, embolisation has also been reported to be a useful alternative treatment. For more extensive lesions, it can be performed independently or prior to surgical interventions, but reported complications range from skin necrosis to limb loss.[15] In our series, one patient was referred following an episode of right heart failure. His extensive AVM involving the left leg was subjected to embolisation and debulking surgery several times, but eventually he underwent amputation at the level of the hip.

Sclerotherapy is the preferred treatment for VVMs,[4,13] and following endothelial destruction, fibrosis, and shrinkage of the lesion it gives rise to luminal obliteration. In our series, all the VVMs treated using sclerotherapy alone had good outcomes, the commonly used sclerosants being STS, ethanol, and hypertonic saline.[13] Conservative treatment options included compression garments that reduce swelling. Prophylactic aspirin use should be considered to prevent thrombotic events and phlebolith formation, as VVMs are often associated with coagulation abnormalities.[16]

Pulsed dye laser is a recognised treatment of choice for capillary malformations.[17] Several treatments are usually required to achieve a lighter colour. Of the two patients treated with 595-nm PDL, one had a good response, whilst in the other the lesion did not get lighter. Interestingly, a long-term study found re-darkening of capillary malformations after PDL treatment (after 10 years).[18] Patients must be prepared for multiple treatment sessions and be counselled about possible re-darkening after treatment.

Historically, surgical resection has been the preferred treatment for lymphatic malformations, but recent studies indicate the effectiveness of sclerotherapy, particularly with the sclerosing agent OK-423 (group A *Streptococcus pyogenes* treated with benzylpenicillin potassium) that achieved an 86% success rate after four treatments.[18] Sclerotherapy is thought to be more effective in macrocystic lymphatic malformations.[13,19] The majority of haemangiomas behaved as expected, and resolved with observation. Propranolol administered for haemangiomas during the proliferative phase was effective.

## Conclusion

This retrospective case series highlights confusion over the nomenclature of vascular lesions. Imaging, including MRI and ultrasound, demonstrated utility for the initial evaluation of vascular malformations. The interventional radiologist plays a significant role in planning sclerotherapy and interpretation of radiological findings. The vascular surgeon has a major role in achieving vascular control of major vessels associated with aggressive AVMs. The orthopaedic surgeon has a major role in amputation and dealing with deep-seated vascular malformations involving muscle and bone. The plastic surgeon has a major role in the surgical excision of most soft tissue lesions. From the patient's perspective, bringing multiple specialists together in a single clinic decreases hospital visits, enhances consensus decisions, improves diagnostic accuracy, and facilitates appropriate management.

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