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Direct percutaneous puncture digital-subtraction-angiography-based classification and treatment selection for soft-tissue arteriovenous malformations of maxillofacial region: a retrospective study

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Abstract. Treatment of arteriovenous malformations (AVMs) should be individualized based on the imaging findings. A total of 117 AVM cases were categorized into three types based on the angio-architectural characteristics: Type I ($n = 14$, no draining vein or diameter of the draining vein <2 mm); Type II ($n = 64$, draining vein diameter 2–6 mm); and Type III ($n = 39$, draining vein diameter >6 mm). Subjects were randomly allocated to one of two treatment groups: Group A ($n = 59$) received multipoint percutaneous ethanol injection (MPEI), while Group B ($n = 58$) received super-selective angiograms followed by embolization with gelfoam (EFAG) plus MPEI. Patients were followed up for 2–6 years. A significant between-group difference with respect to treatment outcomes was observed only for Type III cases ($P < 0.05$). Direct percutaneous puncture digital-subtraction-angiography-guided classification of AVMs provides easy-to-follow guidelines for its clinical management. EFAG plus MPEI with reduced procedure time and the amount of ethanol should be used for Type III AVMs.

Key words: arteriovenous malformations; maxillofacial region; digital subtraction angiography; draining vein; embolic.

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Arteriovenous malformations (AVMs) are congenital vascular malformations attributable to developmental defects of the vasculature. Classical AVM lesions comprise of snarled tangles of arteries and veins with no intervening capillary bed, thereby forming short-cuts for blood to pass from arteries directly to veins[1]. AVMs in the maxillofacial region are rare, but can be potentially life-threatening. Common symptoms of maxillofacial AVMs include bleeding, facial asymmetry, mobility of teeth, and headache. Nearly 60% of the cases are diagnosed at birth, while others are diagnosed during childhood or adolescence. The malformation typically increases in size over time[1]. Large AVMs can be disfiguring and painful, may cause erosion of the surrounding tissues, and may lead to fatal bleeding.

AVMs are considered the most challenging vascular anomalies to treat due to the high risk of haemorrhage and recurrence. Treatment should be individualized based on the angio-architecture. However, classification of soft-tissue AVMs based on the angio-architecture in the maxillofacial region is still ambiguous[1,2].

In this work, we propose a new angiographic classification for AVMs based on a study of angio-architectural characteristics in 117 patients with AVMs. The objective of this study was to demonstrate the utility of this classification system to guide treatment decision-making and to provide new insights related to the management of AVMs.

Materials and Methods

Patients

The institutional review board of our hospital approved the use of patient medical and imaging records for this study. From March 2008 to February 2015, a total of 117 consecutive patients with AVMs in the maxillofacial region were recruited at the Linyi Cancer Hospital (62 males and 55 females), median age of patients at the time of treatment was 23 years (range, 2 months to 57 years). Written informed consent for the procedure was obtained from all patients or their guardians after informing them about the advantages and the risks associated with the procedure.

All patients presented with progressively expanding AVM lesions in the maxillofacial region; the symptomatic history ranged from 2 months to 50 years. Eight patients had a history of blunt trauma, 26 had tissue ulceration and haemorrhage, 13 complained of recurrent episodes of epistaxis (including four patients with severe anaemia). Ninety-three patients had a pulsatile mass, and a vascular murmur was audible in 84 patients. The size of the lesions ranged between 9×20 cm and 2×3 cm, and the surface temperature of the involved area in all patients was significantly higher than that of the surrounding normal skin.

Imaging method

Digital subtraction angiography (DSA) was performed using the PHLIPS HD20 DSA system to confirm the diagnosis of AVMs in all patients. Selective angiograms of the ipsilateral internal carotid artery, external carotid artery, and vertebral artery were performed. Super-selective angiograms of the branches of the external carotid artery were performed on all subjects in order to clearly delineate the angio-architecture of the AVMs and the main feeding artery. Images of the nidus and the feeding arteries including frontal and lateral series were acquired with an exposure rate of 3 frames per second to examine the affected range, including the size, feeding artery, flow rate, draining vein, extracranial communicating branches and the arteriovenous anastomosis, if detectable. Draining vein of the lesion was identified via direct percutaneous puncture DSA with a 7-gauge butterfly needle inserting into the nidus. Two to four puncture sites were selected depending on the size of nidus. The direction and depth of needle were adjusted until blood outflow was observed in the connection tube to the butterfly needle. Then contrast medium was injected with high-pressure injector to delineate the vascular malformation. The affected region and draining vein with the maximum diameter were determined on frontal and lateral image series. Diameter of the draining vein was measured based on direct percutaneous puncture DSA, using a workstation (Allura Xper FD20, XINGYUN ISP) with 5F catheter as reference. The time to peak (TTP) for the corresponding draining vein was calculated as the time elapsed from the first frame that showed visible contrast medium to the frame that exhibited peak intensity. The diameters of the draining veins were the criteria for classification, while TTP provided an objective measure of the haemodynamic characteristics of the AVMs. AVMs were categorized into three types: Type I, no draining vein observed or draining vein with diameter <2 mm, for which the TTP is longer than that of the normal head and neck veins (indicative of low flow-volume/rate); Type II, draining vein with diameter ranging from 2 to 6 mm, for which the TTP is shorter than that of the normal head and neck veins (intermediate flow-volume/rate); and Type III, draining vein with diameter >6 mm, for which the TTP is shorter than that observed in Type II AVMs [indicative of high flow-volume/rate (with the exception of large aneurysm dilatation)].

Treatment procedure

One hundred and seventeen patients with AVMs in the maxillofacial region were randomly assigned to one of the two groups (Groups A and B). Lesion characteristics were comparable in the two groups. Patients in Group A (including six Type I, 32 Type II, and 21 Type III AVMs) were treated with multipoint percutaneous ethanol injection (MPEI) of the nidus, while patients in Group B (including eight Type I, 32 Type II and 18 Type III AVMs) were treated with super-selective angiograms of the external carotid artery branches followed by embolization of the feeding arteries with gelfoam (EFAG) plus MPEI of the nidus and the draining veins. Nevertheless, use of intravascular sclerosant can cause tissue necrosis and cardiopulmonary collapse during the procedure; in this study, hot compresses and vasodilators were administered in case of necrosis. In addition, a Swan–Ganz catheter was used to monitor pulmonary artery pressure in order to detect potential precapillary spasm and to promptly control the complication of cardiopulmonary collapse. The ethanol injection should be stopped immediately once an obvious rise in pulmonary artery pressure is noted.

Evaluation of clinical data and follow-up results

All patients were followed up for 2–6years after the initial procedure. The outcomes were categorized into three ranks: cured (asymptomatic with no recurrence or enlargement of lesion); effective (relief of symptoms and shrinkage of nidus by $>50\%$ with no further enlargement); and ineffective (no significant shrinkage or change in size).

Statistical Analysis

Chi-squared test was used to compare outcomes between the two groups. Data analysis was performed using IBM SPSS Statistics for Windows, version 15 (IBM Corp, Chicago, IL, USA). Statistical sig-

nificance was defined as a value of $P < 0.05$.

Results

DSA findings

The angiographic findings are summarized in Table 1. All cases presented classical imaging features of AVMs, including dilation and enlargement of the feeding artery with vascular malformation nidus and early venous drainage (Figs 1a, 2b, 3b) in routine DSA examination. Single or multiple anatomical areas were affected by the nidus of the AVMs. Of the 117 AVMs, 111 were fed by external carotid artery as the nutrient artery. Two AVMs located in the nose root and four AVMs located in the inter-eyebrow region were fed by the ophthalmic artery. Direct percutaneous puncture DSA images of the AVMs showed that 64 of 117 AVMs had draining veins ranging from 2 to 6mm, which were formed by miscellaneous draining veins surrounding the nidus and merged into the distal dilated veins; 39 cases had draining veins >6 mm in diameter, of which 18 cases had dilated draining veins surrounding the nidus and 21 had dilated draining veins measuring >6 mm at the mandibular angle; 14 AVMs showed only enhanced nodular or lump vascular niduses, but no draining veins of diameter <2 mm even after repeat punctures for multiple times.

Table 1. Digital-subtraction-angiography-based classification of 117 maxillofacial arteriovenous malformations.

Classification	Draining vein	Time-to-peak of draining vein	Diameter of draining vein
Type I	None	>7.38 ± 2.94 s	0 or <2 mm
Type II	Dilated	5.10 ± 1.30 s	≥2 mm and <6 mm
Type III	Aneurysmal dilatation	3.81 ± 0.35 s	≥6 mm

Time-to-peak of the normal veins in the head and neck was 7.38 ± 2.94 s.

Direct percutaneous puncture DSA-based classification

According to the diameter of the draining veins assessed by direct percutaneous puncture DSA examination, 117 AVMs were categorized into three types: Type I (14 cases); Type II (64 cases); and Type III (39 cases). For Type I lesions, no draining veins or only thin draining veins < 2 mm in diameter were observed after multi-point puncture (Fig. 1b). Routine DSA was performed despite the lack of evidence of the draining veins on direct percutaneous puncture DSA (Fig. 1a). Type II AVMs had significantly dilated draining veins with reticulate dispersion of communicating arteries converging into draining veins with diameter ranging from 2 to 6 mm (Fig. 2c). Type III AVMs were characterized by significantly enlarged veins (diameter >6 mm) that resembled venous aneurysm (Fig. 3c). TTPs of Type

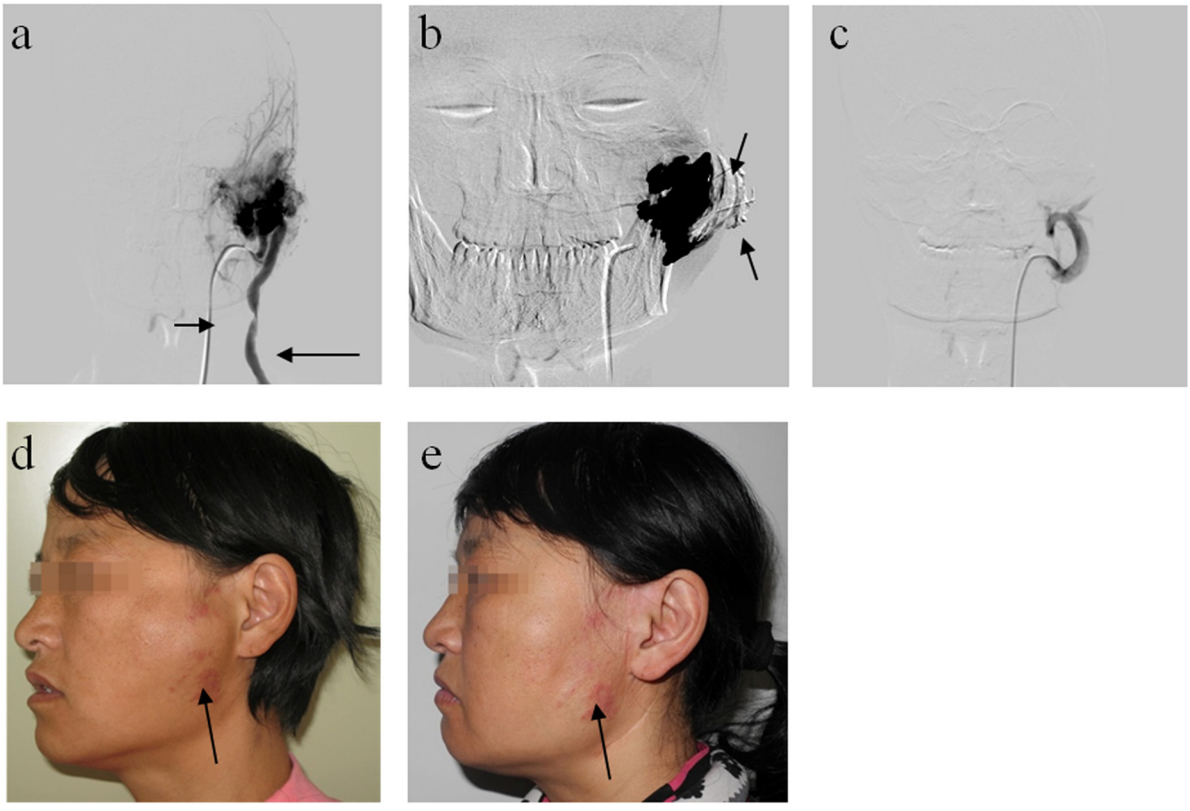

Fig. 1. Type I arteriovenous malformations with no evident draining vein. (a) Digital subtraction angiography (DSA) via femoral artery puncture shows the nidus in external carotid artery (short arrow) with a dilated draining vein (long arrow). (b) Direct percutaneous puncture DSA shows no obvious draining vein; lobulated nidus with acanthoid extentions (arrow indicates the puncture needle). (c) The blood supply of the feeding artery is blocked after embolization of the feeding arteries with gelfoam (EFAG). (d) After treatment with EFAG followed by multipoint (four points) percutaneous ethanol embolization (MPEI) of the nidus and the veins, no significant shrinkage of vascular nidus is observed (arrow indicates residual protrusion). (e) The mass was resected surgically (arrow indicates the preauricula incision).

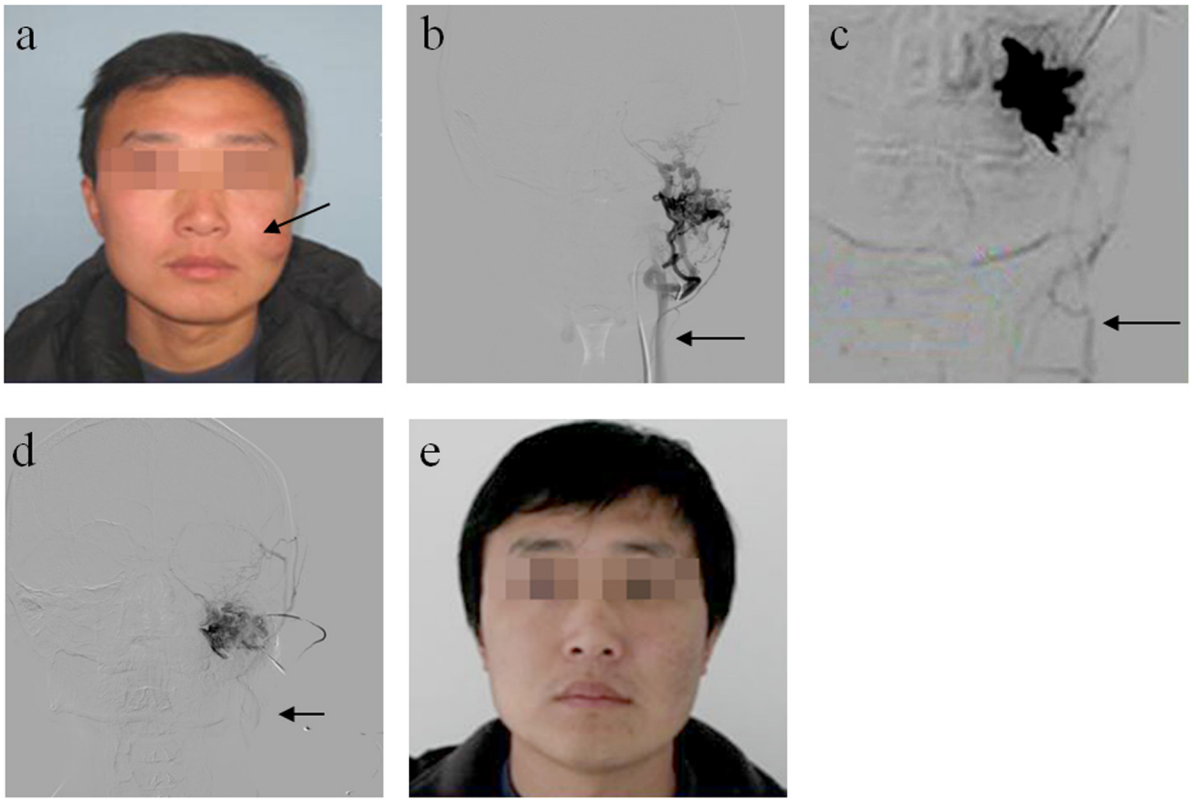

Fig. 2. Type II arteriovenous malformations (AVMs) with dilated draining vein. (a) Soft tissue lump in the left maxillofacial region before treatment (arrow). (b) Digital subtraction angiography (DSA) via femoral artery puncture shows arachnid enhanced lesion with dilated draining vein (arrow), confirmed as AVMs nidus. (c) Direct percutaneous puncture DSA image shows dilated draining vein surrounding the nidus (arrow). (d) After local multipoint percutaneous ethanol embolization (MPEI) the draining vein surrounding the nidus has almost disappeared in the DSA image (arrow). (e) The mass in the left maxillofacial area has disappeared after embolization.

I, Type II and Type III AVMs were 7.38 ± 2.94 s, 5.10 ± 1.30 s and 3.81 ± 0.35 s, respectively (Table 1). In addition, 14 Type I AVMs showed cluster or nodular lesion involving only one anatomical subregion. Of the 64 Type II cases, 29, 26 and 9 AVM lesions involved one, two and three or more anatomical subregions, respectively, and exhibited arachnid-like enhancement. 14, 17 and 8 AVMs of Type III cases involved one, two and three or more anatomical subregions, respectively, and exhibited cystoid and earthworm-like enhancement.

Clinical outcomes

The clinical outcomes are summarized in Table 2. All the 117 cases of AVMs (including 14 Type I, 64 Type II, and 39 Type III) were randomly allocated into two treatment groups: Group A (including six Type I, 32 Type II, and 21 Type III AVMs) were treated with MPEI of the nidus; and Group B (including eight Type I, 32 Type II, and 18 Type III AVMs) were treated with superselective angiograms of the external carotid artery branches followed by EFAG plus MPEI for the nidus and the draining veins. The general efficiency rates of Type I, II and III AVMs in Group A were 33.33%, 100% and 71.43%, while those in Group B were 37.50%, 100% and 94.44%, respectively. The corresponding cure rates in Group A were 0%, 75.00% and 14.29%, respectively; those in group B were 0%, 78.13% and 50.00%, respectively. There were no significant differences with respect to treatment outcomes of Type I and II AVMs between Groups A and B, while the treatment efficacy for Type III AVMs in Group B were significantly higher than those in Group A (cured: 50.00% vs. 14.29%; effective: 44.44% vs. 57.14%; ineffective: 5.56% vs. 28.57%, respectively).

Discussion

In the present study, routine DSA examination and direct percutaneous puncture DSA examination for maxillofacial AVMs were performed on all patients. Based on the diameter of the draining vein, AVMs were categorized into three categories. Subsequently, the patients were randomly assigned to one of the two treatment groups (MPEI only or EFAG plus MPEI). Treatment outcomes for the respective categories of lesions were compared between the two groups. A combination of draining vein diameter and the corresponding TTP was found to provide valuable guidance for selection of treatment approach and for prediction of overall prognosis. To our knowledge, this is the first report pertaining to classification of maxillofacial AVMs using direct percutaneous puncture DSA and its use in guiding treatment and predicting outcome. The easy procedure and quick evaluation process suggest its potential clinical value in practice.

AVMs are rarely seen in the maxillofacial region. Clinical symptoms usually vary with the anatomical location, including pain, ulceration, disfiguring mass, and haemorrhage. Episodes of acute bleeding and fatal complications were also reported in severe cases[3]. Schobinger staging system is widely used for evaluation of the development and severity of maxillofacial AVMs. This staging system categorize-

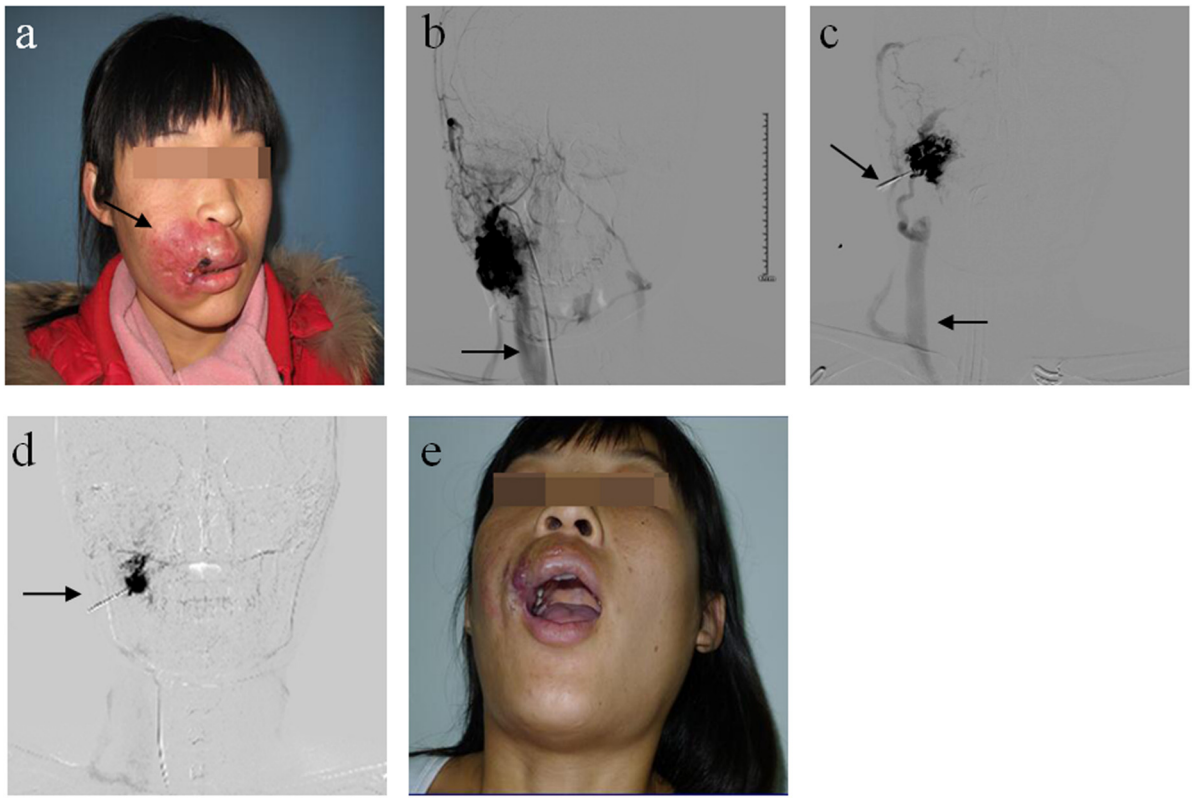

Fig. 3. Type III arteriovenous malformations (AVMs) with aneurysmal dilation. (a) Soft tissue lump in the maxillofacial region before treatment. (b) Digital subtraction angiography (DSA) via femoral artery puncture shows significant enhanced lesion with aneurysmal dilated draining vein (arrow) confirmed as AVMs. (c) Direct percutaneous puncture DSA image (arrow) shows aneurysmal draining vein (arrow). (d) After embolization of the feeding arteries with gelfoam (EFAG) followed by percutaneous ethanol embolization (MPEI) the mass has shrunk (arrow) and the large draining vein has disappeared. (e) The lesion has resolved after treatment.

sAVMs into four stages (quiescence, expansion, destruction and decompensation). Kohout et al.[1] further supplemented the staging system by grading the maxillofacial lesion based on the clinical presentation and the characteristics of endothelial cells. In addition, multiple imaging modalities have been used for classification of AVMs[1,2,4–8] and to differentiate the AVMs from soft tissue tumors[9,10]. A classification system based on DSA-assessed arterial–venous communications and niduses was proposed by Houdart et al.[4], which was later modified by Cho et al.[5]. Other classification systems based on the lesion angio-architecture have been proposed by Yakes[11], Liu[6] and Qin et al.[7] to facilitate management of AVMs. However, it is pertinent to mention here that the existing guidelines do not take the haemodynamic parameters into consideration. In our study, we proposed a classification model based on the diameter of the draining veins which reflects the flow characteristics of the AVMs and thereby facilitates the treatment decision-making.

Currently, standard treatment of AVMs is based on considerations such as location, flow characteristics, symptoms, functional disability, and cosmetic deformity, and optimal therapy can be varied with each case[12]. In particular, the flow characteristics are a key aspect of lesion characterization that helps identify the optimal treatment strategy[13]. More importantly, the nidus and the draining veins are considered as the main target for embolization. Ethanol (as in our study) denudes endothelial cells from the vascular wall; however, excessive dosage may increase the risk of pulmonary embolism and cause severe complications, such as skin necrosis and facial nerve damage. Therefore, the optimal treatment strategy should aim to minimize the dosage of ethanol to avoid these complications.

The feeding artery, draining vein, nidus connecting artery and vein of maxillofacial AVMs can be clearly imaged with routine DSA. Direct percutaneous puncture DSA of the vascular mass provides even more detailed information about the vascular malformation to guide direct sclerotherapy. In this article, we confirmed the hypothesis that diameter and TTP of the draining vein are key factors that determine the optimal treatment and predict the prognosis. Based on the draining vein diameter, we propose a new classifi-

Table 2. Outcomes of embolization therapy for 117 maxillofacial arteriovenous malformations.

Classification	Group	Cases	Cure (%)	Effective (%)	Ineffective (%)	$\chi 2$	*P*
Type I	A	6	0	2 (33.33)	4 (66.66)	0.024	0.877
	B	8	0	3 (37.50)	5 (62.50)		
Type II	A	32	24 (75.00)	8 (25.00)	0	0.087	0.770
	B	32	25 (78.13)	7 (21.88)	0		
Type III	A	21	3 (14.29)	12 (57.14)	6 (28.57)	6.984	0.008*
	B	18	9 (50.00)	8 (44.44)	1 (5.56)		

* Treatment outcomes for Type III cases in group B significantly outperformed those for Type III cases in group A.

cation system that classifies the AVMs into Type I [AVMs that lack effective draining veins and are largely composed of pathological solid tissues, with no dilated venous pool (no draining vein or draining veins measuring < 2 mm in diameter, longer TTP), which is indicative of their low flow-volume/rate], Type III [AVMs with draining veins >6 mm in diameter and shorter TTP, which indicates their high flow-volume/rate (except large aneurysm dilatation)], and Type II AVMs (the intermediate type).

Satisfactory results were achieved by taking measures guided by the classification system. The general efficiency rate and cure rate were up to 94.4% and 50.0%, respectively. Outcomes of Type I and II AVMs in Groups A and B were comparable ($P > 0.05$), while the Type III AVMs in Group B showed significantly improved outcomes than that of Group A. As for Type I AVMs, percutaneous ethanol injection failed to achieve significant mass shrinkage, while use of large amounts of ethanol entails a risk of complications. This is attributable to the nature of Type I AVMs (mainly composed of solid tissues with low flow-volume/rate), which readily allows the infiltration of the embolization agent into the interstitial space. Indeed, Type I AVMs failed to be effectively treated with ethanol embolization, but were successfully treated with surgical intervention (Fig. 1a–e). Therefore, surgical intervention should be considered as the first-choice treatment for Type I AVMs. A randomized controlled study has shown that Type III AVMs with abnormal dilated draining veins should be carefully evaluated and cautiously treated[14]. Ethanol embolization is not a desirable treatment for Type III AVMs as the augmented blood flow and increased volume rate is liable to dilute the ethanol immediately after its injection. Similarly, Type II (but not Type III) AVMs that have lower flow-volume/rate can be treated effectively with MPEI to nidus and the draining vein (Fig. 2a–e). Type III AVMs with high flow-volume/rate and dilated venous pool should be handled with EFAG first, followed by MPEI to nidus and draining veins so as to prolong the retention time of the sclerosing agents within the lesion; such a strategy helps reduce the dosage and improve the treatment efficacy (Fig. 3a–e).

There is a limitation of our study that needs to be considered while interpreting our results. Routine follow-up examination with DSA should be performed for all traceable patients to provide a more comprehensive assessment of the prognosis.

In conclusion, classification of AVMs in the maxillofacial region based on diameter of the draining vein may facilitate the selection of the optimal treatment approach and help predict the risk of complications as well as the overall prognosis.

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Competing interests

The authors declare that they have no conflicts of interest concerning this article.

Ethical approval

The institutional review board of our hospital approved the use of patient medical and imaging records for this study.

Patient consent

Written informed consent for the procedure was obtained from all patients and their guardians after informing them about the advantages and the risks associated with the procedure.

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