

A preliminary study of angiographic classification and its correlation to treatment of central arteriovenous malformation in the jaw

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Objective. To elaborate on the classification of central arteriovenous malformations (AVMs) in the jaw based on angioarchitecture and to evaluate the correlation of this classification with treatment options.

Study design. X-ray films and digital subtraction angiograms (DSA) of 25 cases with AVMs in the jaw were retrospectively reviewed to evaluate the appearance of bone resorption, feeding arteries, nidus of the malformations, and draining veins. Based on the findings a classification of angioarchitecture was recommended. Furthermore, the treatment results of these cases were reviewed to assess the correlation of this classification system with treatment options.

Results. The angiographic pictures of 25 cases with central AVMs could be divided into 5 types: Type I (n = 5) had diffused microarteriovenous fistulas (AVFs); type II (n = 8) had 1 large venous pouch with all the feeding arteries draining into it; type III (n = 7) had a large venous pouch as well as diffused microAVFs (I + II); type IV (n = 2) had multiple venous pouches; and type V (n = 3) had multiple venous pouches and diffused microAVFs (I + IV). The angioarchitecture corresponded well to the x-ray appearance in all cases except 1 (96%). With respect to the treatment outcomes, type II, III, and IV AVMs gained clinical cure in 100% of the cases, whereas type I and type V AVMs obtained clinical cure only in 60% and 33.3% of the cases, respectively.

Conclusions. Central AVMs in the jaw contained variant patterns of angioarchitecture and could be divided into 5 types.

This angiographic classification was helpful for decision making about appropriate therapy.

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Central arteriovenous malformations of jaws, although rare, are clinically important owing to the potential risk of life-threatening hemorrhage. Many therapeutic approaches have been proposed over the years with limited clinical success. Ligation of the external carotid artery (ECA) is completely proscribed, because even if it stops hemorrhage it never prevents recurrence from the rich collaterals and therefore it increases the difficulty of further treatment. Complete resection of the involved bone may be curative, but it involves severe blood loss and induces damage to the shape and function of the involved bone. Radiotherapy often fails owing to the level of cellular maturation along with the danger of radiation damage. Complete cure by arterial embolization is difficult, either with particles or glue, and may involve potential complications. Conservative resection after arterial embolization could be safe and curative, but it still involves functional deficits.^{1,2} Recently, some

authors have cured central AVMs by a venous approach with glue or microcoils, either via transosseous punctures or by a transfemoral approach, allowing preservation of the involved bone and teeth.³⁻⁸ In our experience, treatment options depend not only upon the patients' medical and economic situations, but also upon angioarchitecture of these high-flow malformations.² The aim of therapy should be complete obliteration of the lesion with preservation of the affected bone and teeth, and recurrence from insufficient therapy as well as associated impairment from excessive intervention should be prevented. In this study, imaging materials of 25 patients with central AVMs of the jaw in our hospital over the past 6 years were retrospectively reviewed to elaborate on a classification system of central AVMs on angioarchitecture. Furthermore, the treatment outcomes were reviewed to assess the correlation of this classification system with treatment options.

MATERIAL AND METHODS

Twenty-five patients with central AVMs of the jaw included 9 males and 16 females (male-female ratio 1:1.77), with a mean age of 17.6 years (ranging from 10 to 40 years). Eight AVMs were in the maxilla and 17 were in the mandible. Panoramic tomography was taken in all patients, as well as intraoral panoramic radiograph in 8 maxillary and 5 mandibular AVMs. Furthermore, 10 of the 25 patients had undergone a CT scan. All patients

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Table I. The correlation of x-ray and DSA appearance

	DSA type I	DSA type II	DSA type III	DSA type IV	DSA type V
X-ray type I	5				
X-ray type II					
X-ray type III					
X-ray type IV					
X-ray type V					

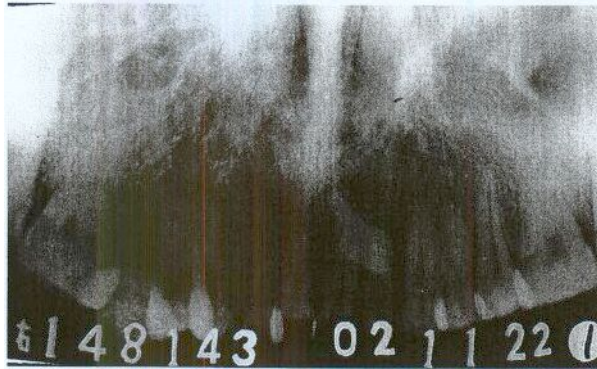


Fig 1. Right maxillary honeycombed radiolucency on the intraoral panoramic film.

experienced bilateral internal and external carotid angiography, followed by superselective angiography of ECA feeders with lateral and posterior-anterior projections. Moreover, vertebral angiograms were taken in 4 cases with ligated ECA. Generally, 35 angiography and embolization procedures were performed in these patients. Two experienced radiologists reviewed the patient's records for the following information: episodes of uncontrollable bleeding, radiographic appearance on plain and tomographic films, feeding arteries, nidus of the malformations, and draining veins. The correlation of radiographic and angiographic features was further analyzed. Additionally, the vascularization characteristics of the maxilla and mandibular AVMs were analyzed.

The courses of treatment was as follows: 3 cases of arterial embolization alone, 4 cases of arterial and venous embolization, 10 cases of arterial embolization and curettage, and 8 cases of arterial and venous embolization with curettage. All cases were followed up after the last treatment on a regular basis with a mean of 30 months (ranging from 6 to 60 months). The treatment outcomes were reviewed and categorized as anatomic cure, clinical cure, and persistent AVMs. An anatomic cure was defined as the disappearance of all symptoms with a radiographic demonstration of complete reossification of the affected bone. The clinical cure group included the anatomic cures, as well as patients where

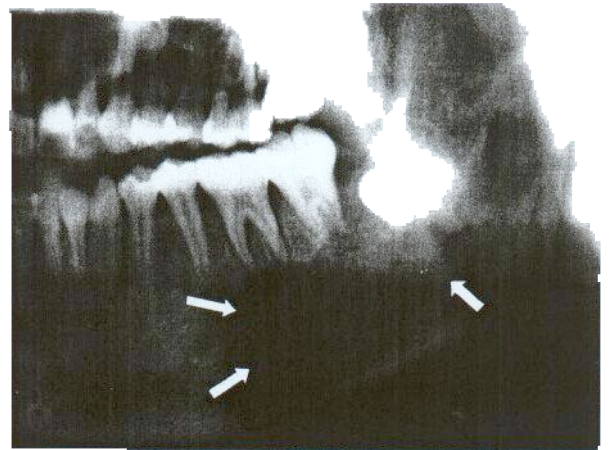


Fig 2. Left mandibular monocystic radiolucency (open arrow) on panoramic tomography.

there was a disappearance of clinical symptoms despite an incomplete reossification of the affected bone.

RESULTS

X-ray and angiographic findings

The x-ray findings were divided into 5 types (Table I): type I ($n = 5$) with honeycombed radiolucency (Fig 1), type II ($n = 9$) with well defined monocystic radiolucency (Fig 2), type III ($n = 7$) with monocystic and honeycombed radiolucency (I + II, Fig 3), type IV ($n = 1$) with multicystic radiolucency (Fig 4), and type V ($n = 3$) with multicystic and honeycombed radiolucency (I + IV, Fig 5).

The angiographic features could also be divided into 5 types (Table I). Type I ($n = 5$) were AVMs with diffused microarteriovenous fistulas (AVFs) that had multiple arterial feeders and drained via the anterior and posterior facial veins ($n = 2$) or merely by the posterior facial veins ($n = 3$, Fig 6). Type II ($n = 8$) had multiple arterial feeders that drained into a huge venous pouch and then into the anterior and posterior facial veins ($n = 6$) or only into the posterior facial veins ($n = 2$, Fig 7). Type III ($n = 7$) had 1 large venous pouch as well as diffused microAVFs (I + II, Fig 8). Type IV ($n = 2$) were with multiple large venous pouches (Fig 9). Type V ($n = 3$) had multiple large venous pouches together with diffused microAVFs (I + IV, Fig 10). According to the medical history, 1 AVM of type I, 5 of type II, 5 of type III, 1 of type IV, and 2 of type V had episodes of uncontrollable bleeding. In 24 cases (24/25), the radiographic appearance corresponded well to the angiographic feature, ie, the honeycombed radiolucency revealed diffused microAVFs on angiograms whereas a cystic radiolucency appeared as a large venous pouch on angiographic maps. The exception was in a bilateral

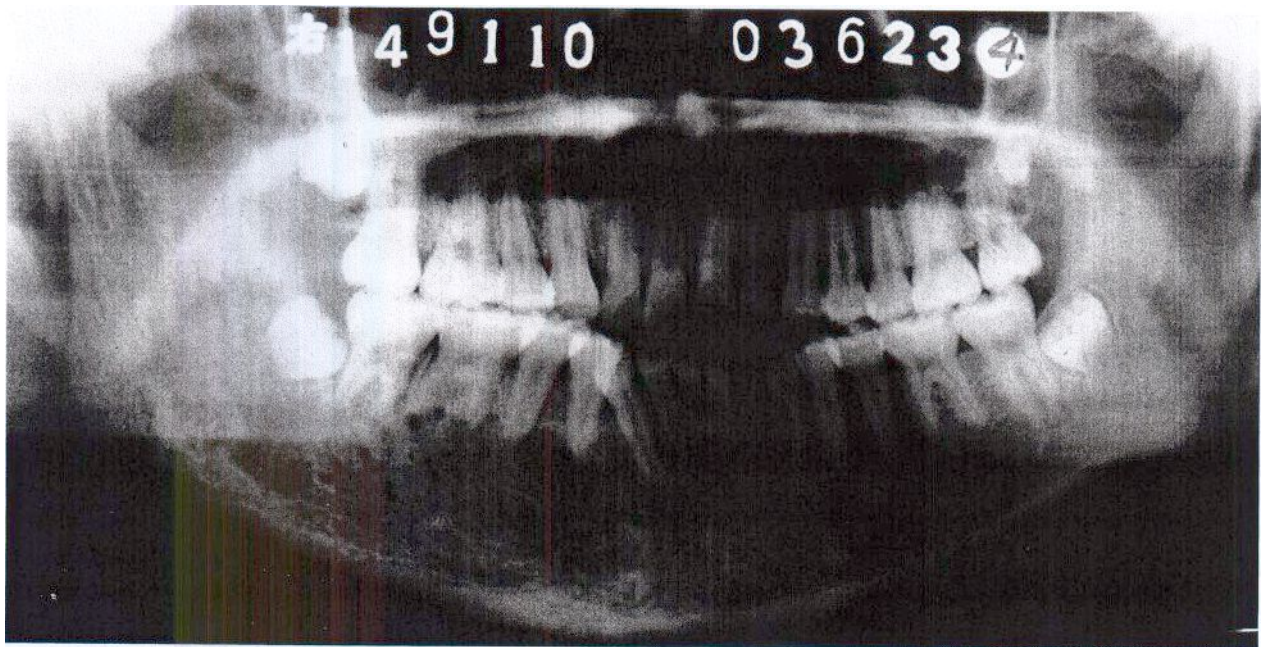


Fig 3. Monocystic and honeycombed radiolucency of the right mandible.

mandibular AVM that had a huge cystic radiolucency extending from one angle to the other of the mandible but exhibited multiple venous pouches on angiograms.

Angiographic features of maxillary and mandibular AVMs (Table II)

Of the 8 maxillary AVMs, 5 occupied the posterior portion of the maxilla, and 3 occupied the posterior and anterior maxilla. With the exception of a case with ligated ECA, 7 AVMs were fed mainly by the ipsilateral internal maxillary arterial branches as follows: 4 AVMs by the superior alveolar, infraorbital, descending palatal, and sphenopalatine arteries; 2 AVMs by the superior alveolar, infraorbital, and descending palatal arteries; and 1 AVM by the superior alveolar, infraorbital, and sphenopalatine arteries. According to the DSA classification described above, 3 AVMs were type I, 3 were type II, and 2 were type III.

Of the 17 mandibular AVMs, 11 involved 1 side of the mandible, and 6 were bilateral. With the exception of 2 AVMs, 1 having ligated ECA and 1 having undergone embolization 4 times before, 14 AVMs were mainly fed by ipsilateral ECA branches, as follows: 1 by the inferior alveolar artery, 7 by the inferior alveolar and facial arteries, and 6 by the inferior alveolar, facial, and lingual arteries. Of the 17 AVMs, 2 were type I, 5 were type II, 5 were type III, 2 were type IV, and 3 were type V. These cases demonstrate the complexity of vascularization of the mandibular AVMs.

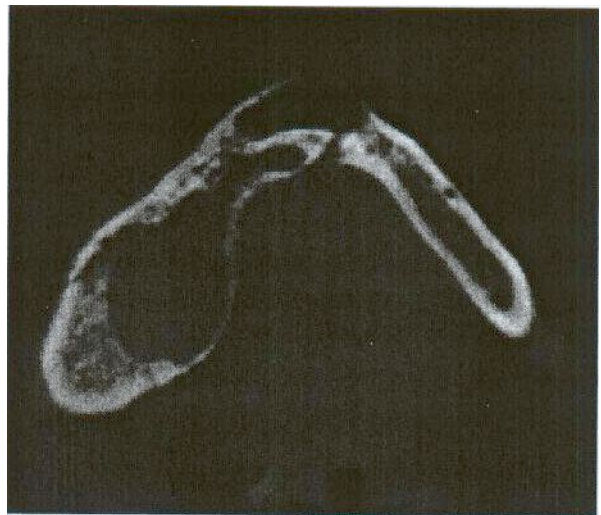


Fig 4. Axial CT showing multiple cystic radiolucency of the mandible.

Treatment outcomes of different types of AVMs (Table III)

Type I (n = 5 AVMs): Two AVMs obtained anatomic cure after embolization and curettage, 1 obtained clinical cure after embolization and curettage, 1 experienced persistent epistaxis even after 3 embolization and 2 curettage procedures, and 1 complained of recurrent gingival bleeding after 3 embolizations. The anatomic and clinical cures were obtained in 2/5 and 3/5 of cases, respectively.

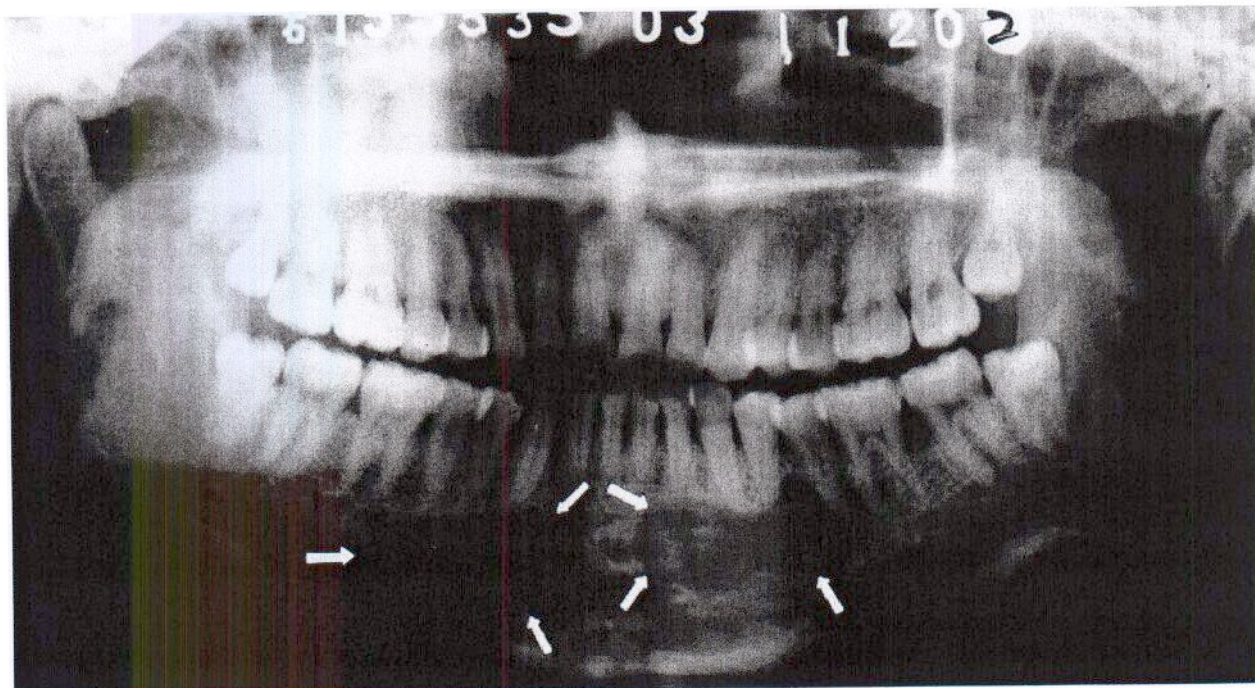


Fig 5. Panoramic film showing a multiple cystic (*open arrow*) and honeycombed lesion involving bilateral mandible.

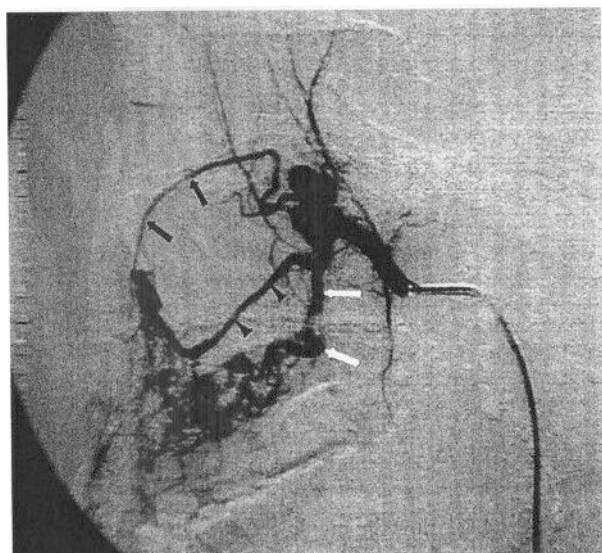


Fig 6. The IMA angiography showed a maxillary AVM with merely diffusely interwoven vessels (type I). The feeding vessels were from the descending palatine (*open arrow*), superior dental (*arrowhead*), and infraorbital arteries (*arrow*).

Type II-IV (n = 17 AVMs): Nine obtained anatomic cure after embolization and curettage, 4 obtained clinical cure after embolization and curettage, 2 obtained anatomic cure after arterial and venous embolization, and 2 obtained clinical cure after arterial and venous embolization. The overall anatomic and

clinical cure rates were 11/17 and 17/17 of cases, respectively.

Type V (n = 3 AVMs): One was anatomically cured after arterial embolization and resection, 1 experienced life-threatening hemorrhage 3 months after complete arterial embolization with glue and particles, and 1 had mild bleeding after 2 embolizations. The anatomic and clinical cures were both 1/3 of AVMs.

DISCUSSION

The treatment of high-flow vascular malformations of the head and neck region, either intracranial or extracranial, is a challenging and difficult field. The clinical presentations, prognosis, and treatment options of these lesions depend on the anatomic location, as well as the angioarchitecture, which is closely related to the hemodynamic changes. Cerebral AVMs were divided into I-V grades according to the size, location, and venous drainage patterns by Spetzler and Martin.⁹ With the advance of the cerebral AVM grading, the prognosis becomes worse and treatment becomes more difficult. Ling and Li divided cerebral AVMs into 5 types according to the angioarchitecture on superselective angiography,¹⁰ and this classification was helpful for decision making about appropriate treatment. Dural AVFs were classified as 4 types by Djindian in 1978¹¹ and as 5 types in 1995 by Cognard et al according to the venous patterns,¹² and these classifications were closely related to the prognosis and treatment approaches.

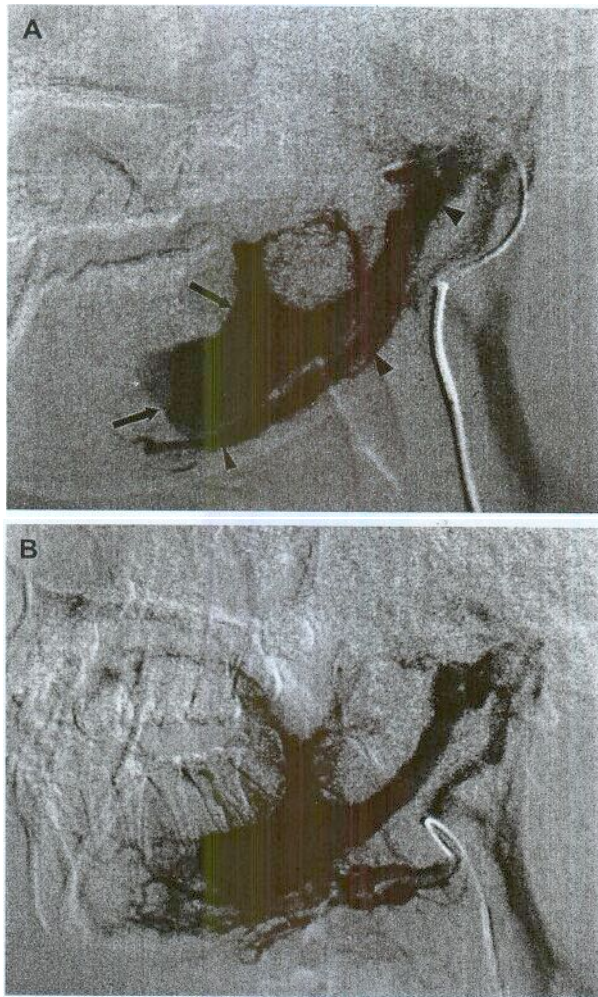


Fig 7. Angiography exhibited a huge venous pouch (A, arrow) within the mandible, fed by multiple feeders from the inferior dental (A, arrowhead) and facial arteries (B, type II).

Considering the high risks and variable treatment outcomes, central AVMs of the jaw are in need of a general and systematic analysis and classification system of angioarchitecture that incorporates and improves upon the individual descriptions in the literature.

In this study, the x-ray and angiographic features of central AVMs in jaws were divided into 5 types according to the analysis of 25 AVMs. The features of DSA corresponded well to that of radiography, therefore types I-V of the DSA classification system, based mainly on nidus patterns of the malformation, were elaborated on for the first time. Types I and II are the fundamental components of this angioarchitecture. Type III-V AVMs comprised multiple compartments of type I and/or type II fistulas and might represent different stages in the progressive advancement of these lesions, or they might result from incomplete endovascular or surgical interventions. Type I AVMs appeared as honeycombed

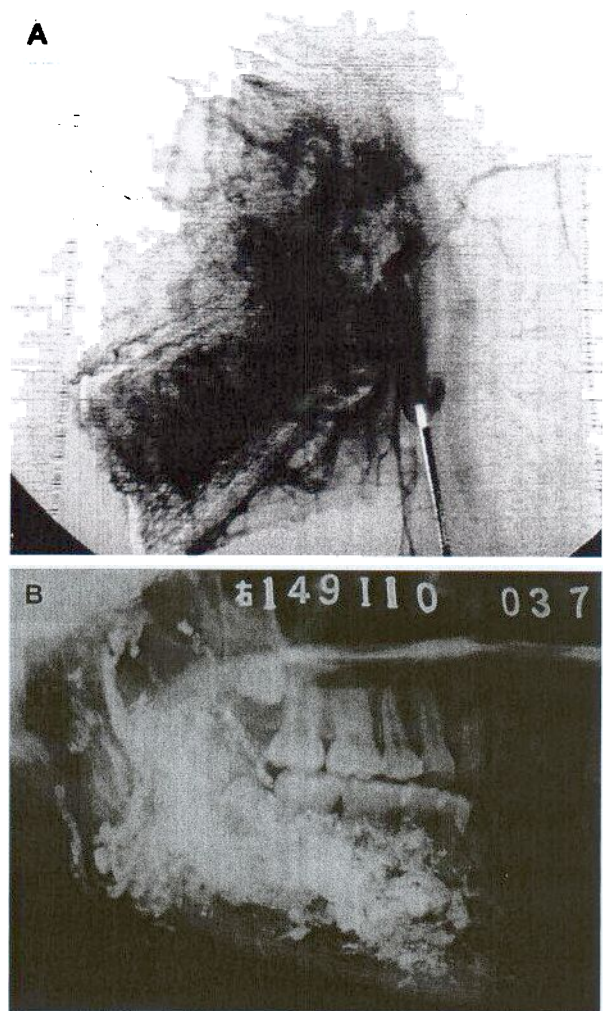


Fig 8. The ECA angiography opacified a mandibular AVM with a huge venous pouch, as well as diffused vessels (A) The post-embolic panoramic film showed the glue cast of the venous pouch and the surrounding diffused veins (B) (type III).

radiolucency on radiography and as diffusely interwoven vessels on angiography, which was similar to AVMs of other areas of the body. A type II AVM could be defined as an arteriovenous fistula that consisted of multiple arterial shunts in the wall of a single huge vein. This pattern of angioarchitecture might occasionally be found in dural AVFs but rarely in other AVMs. The pathogenesis of this pattern may be attributed to the high vascularization of the jaw, which involves an alveolar canal with accompanying arteries and veins, especially in the mandible. Once a congenital AVM develops, the afferent arteries could involve centrifugal feeders of the superior or inferior dental arteries, as well as centripetal feeders from the facial, lingual, descending palatal, and sphenopalatine arteries. In addition, the venous drainage is only via the alveolar veins, inducing progressive

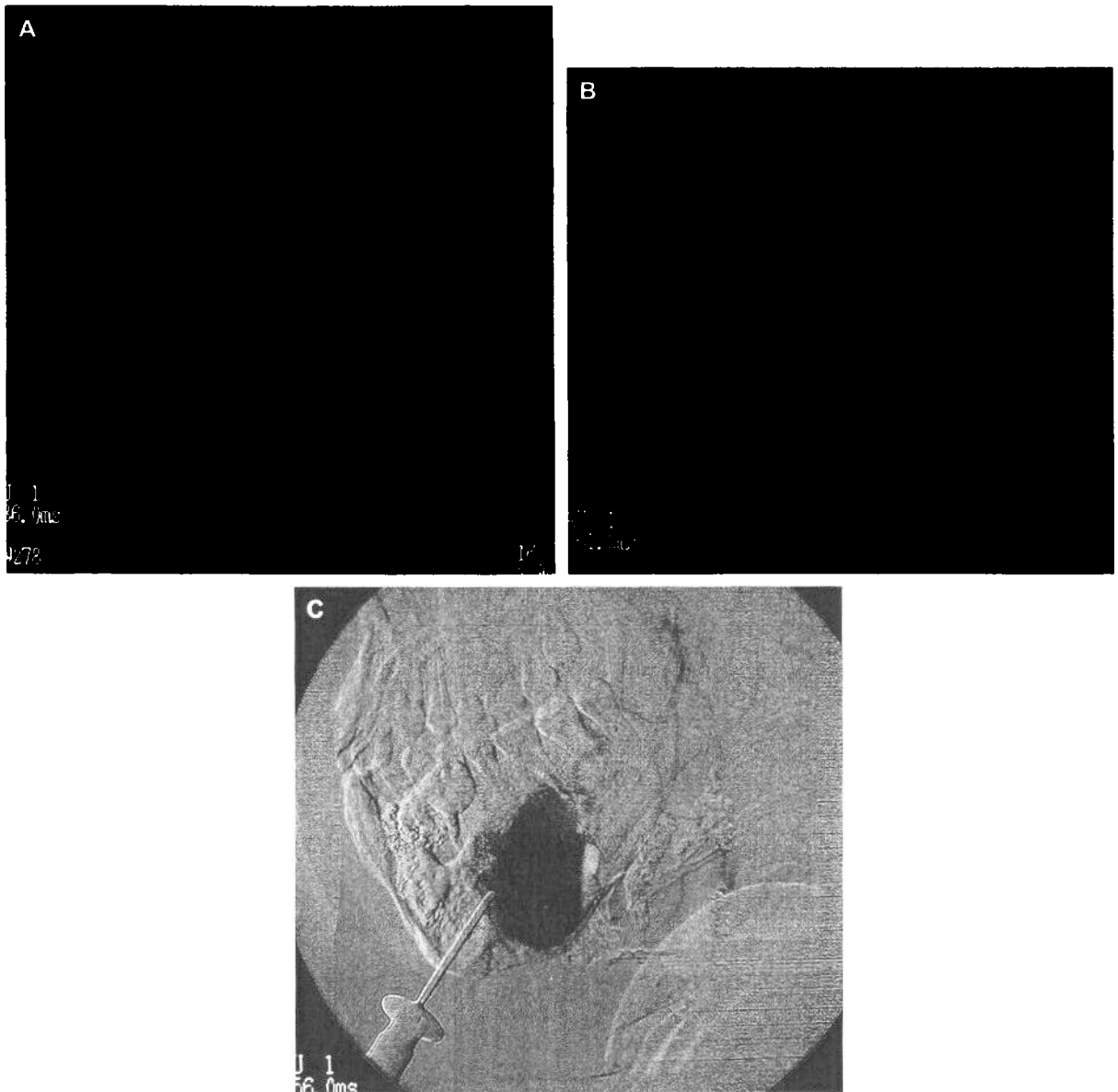


Fig 9. The right common carotid angiography showed a mandibular AVM with 2 venous pouches (A) (type IV). Venograms after percutaneous puncture via the incisor (B) and premolar area (C) opacified the pouches.

dilation of the veins. In our series, type II-V AVMs showed a higher prevalence rate of uncontrollable hemorrhage (13/20 AVMs) over type I AVMs (1/5 AVMs). This observation might also be attributed to the different patterns of angioarchitecture. The huge venous pouch of a type II-V AVM, which fills with high-pressure blood flow, serves as a direct pump for life-threatening bleeding once a rupture of the wall develops from the injury of the overlying teeth.¹³

Defining the patterns of angioarchitecture is beneficial for the decision making of an appropriate therapy. Complete occlusion of type I AVMs by arterial

embolization is rarely possible; superselective glue injection might increase the efficacy, but it produces a higher risk of complications, such as skin necrosis, cranial nerve ischemia, and even cerebral infarction. Conservative curettage is often incomplete, owing to the ill-defined margins of the AVM. In our cases, 3 of the type I AVMs were cured and 2 experienced persistent AVMs after multiple treatment procedures. Considering the unsatisfactory outcomes, we now recommend superselective arterial embolization, followed by complete curettage for this type of AVM, despite their comparatively benign behavior. For type II AVMs,

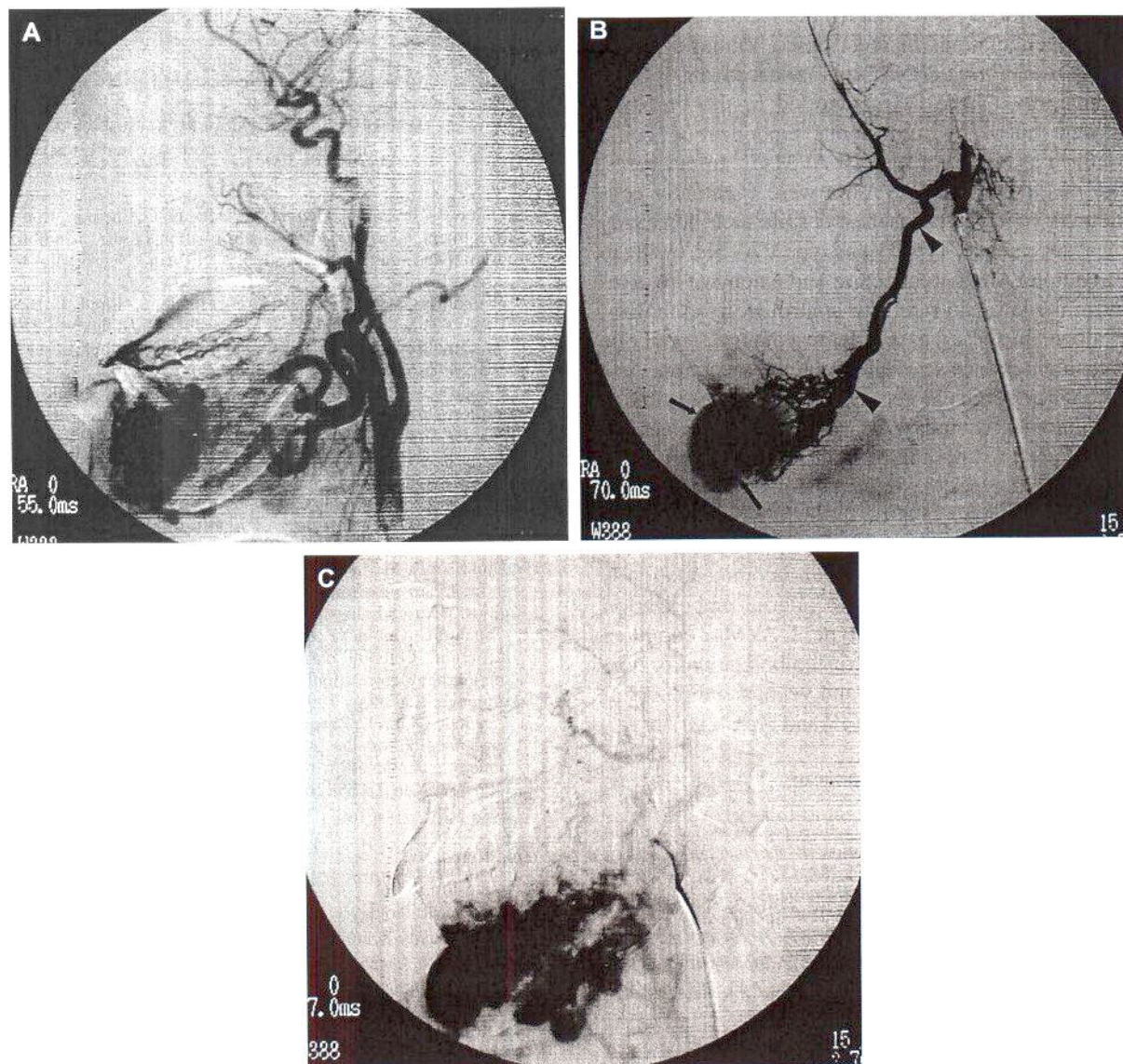


Fig 10. Right common carotid angiography showed the right mandibular AVM, which mainly occupied the anterior mandible and comprised 1 venous pouch and diffused vessels (A). B, The right internal maxillary arteriography showed the mental venous pouch (arrow) fed by multiple branches from the inferior dental artery (arrowhead). C, the left external carotid arteriography showed the left mandibular AVM, comprising 1 venous pouch in the mentum and diffused vessels in the body (type V).

Table II. Distribution of different DSA types of AVMs in the maxilla versus mandible

	Type I	Type II	Type III	Type IV	Type V
Maxilla	3	3	2	—	—
Mandible	2	5	5	2	3

a venous approach is easier than arterial embolization. Filling of the veins with occlusive agents will close all shunts on the wall. Type III AVMs comprise compartments of type I and type II fistulas, therefore complete obliteration of the complex nidus is mandatory,

Table III. Treatment records of different types of AVMs

	Type I	Type II	Type III	Type IV	Type V
Number of cases	5	8	7	2	3
Times of AE	10	10	10	2	4
Times of VE	2	5	4	1	1
Times of surgery	7	6	5	2	2
AC rate	2/5	7/8	3/7	1/2	1/3
CC rate	3/5	8/8	7/7	2/2	1/3

AE, arterial embolization; VE, venous embolization; AC, anatomic cure; CC, clinical cure.

otherwise it might serve as a trigger for recruitment of collateral circulation. In most cases, multiple procedures of intranidus embolization are necessary.

A majority of the cases reported in the literature that were cured by intravenous embolization could be defined as type II or type III AVMs.³⁻⁸ In our cases, 100% of type II and III AVMs were eventually controlled via variable approaches, despite their horrifying clinical appearance. Type IV and type V AVMs, owing to their complex angioarchitecture and extensive involvement of the jaw, can rarely be controlled by embolization alone. In these cases a large amount of embolic agents might induce severe foreign body reaction. As a consequence, surgical resection is commonly inevitable.

In summary, central AVMs of the jaw demonstrate variable patterns of angioarchitecture that could be divided into 5 types. Type I and II AVMs represent 2 fundamental components of these intractable lesions. Defining the angioarchitecture is helpful for the selection of an appropriate therapy. Our recommendation is a combination of superselective arterial embolization and complete curettage for type I AVMs, complete venous occlusion aided by arterial embolization for type II and III AVMs, and arterial and venous embolization followed by curettage for type IV and type V AVMs.

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