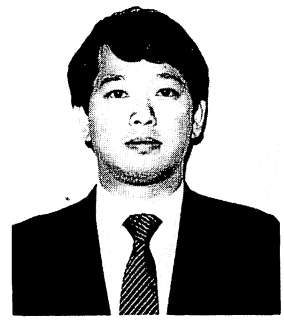

Embolization in High-flow Arteriovenous Malformations of the Face

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Five patients with arteriovenous malformations of the face (4 males and 1 woman; age range, 11–38 years) were treated by selective embolization through the feeding arteries with polyvinyl alcohol particles. Immediate gross angiographical obliteration was obtained in 4 patients, with pronounced reduction of the arteriovenous shunt in the fifth. Clinical symptoms including bleeding, swelling, pulsations, bruit, and disfigurement improved in all the patients followed up for a period of 6 to 21 months. Polyvinyl alcohol particle embolization, without surgical resection, though palliative could be useful in select patients. The classification and diagnosis of congenital vascular malformations is briefly reviewed and treatment discussed.

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The classification and comprehensive understanding of congenital high-flow vascular malformations of the face is still not clear. Nevertheless, their management remains a challenge to the clinician, more so to an interventional neuroradiologist. Advances in selective angiography and development of newer embolic materials offer fresh avenues for treating such lesions. We present our experience with 5 patients with arteriovenous malformation (AVM) and discuss the diagnostic and therapeutic problems, including transarterial particulate embolization.

Material and Methods

The Table gives the clinical details of our patients. There were 4 males and 1 woman, aged 11 to 38 years (mean, 24.6 years). Diagnoses from referral clinics included "high-flow hemangiomas" in 3 and "hemangioma/AVM complexes" in 2 patients. Multiple partial resections, electrocoagulation, or both were performed in Patients 2 and 5, possibly due to lack of proper angiographic interpretation.

Embolization therapy was decided on jointly by the interventional neuroradiologists (neurosurgeons in the team) and the plastic surgeons. Detailed discussions were held with patients and families before embolization was decided on.

The embolic material used was polyvinyl alcohol (PVA) particles (250–600 μm) injected using 6.0 to 7.0 French-size catheters with a tapering tip, catheterized selectively in the feeding pedicle of the AVM. Because bilateral external carotid arteries (ECAs) had been surgically ligated in Patient 5, multiple feeders to the AVM were used for this purpose. These procedures were performed under local anesthesia except in Patient 3.

Illustrative Patients

Patient 1

A 21-year-old man noticed a swelling of his left cheek (Fig 1), which did not bother him, at age 12 years. Since the age of 19 years, oral bleeding occurred almost every week. On admission, a pulsatile swelling of the left cheek and upper lip was obvious. He pointed to a bleeding site on the hard palate posterior to the left incisor, which nevertheless appeared normal.

Angiography revealed an AVM involving the left cheek and the upper lip, fed mainly by the left facial and transverse facial arteries and partially by the right facial artery. It drained into the superficial facial veins. Through these feeders, embolization was performed,

Summary of 5 Patients with Arteriovenous Malformations of the Face

Patient No.	Age (yr)	Sex	Location	Symptoms	Feeders	No. of embolization	Results	Follow-up (mo)
1	21	M	L cheek, upper lip, maxilla	Swelling, bleeding, pulsation	Bil facial artery, L transverse facial artery	1	↓ Swelling, no bleeding, no pulsation	9
2	38	M	L cheek, upper lip	Swelling, bleeding	L facial artery, L transverse facial artery	1	↓ Swelling, no bleeding	8
3	11	M	L ear	Disfigurement, bruit, pulsation	L posterior auricular artery	1	No bruit, no pulsation	6
4	32	F	L ear	Bleeding, disfigurement, swelling, pulsation	L posterior and anterior auricular artery, occipital artery	3	↓ Swelling, no bleeding, no pulsation	21
5	21	M	R cheek, ear, maxilla temporal	Bleeding, disfigurement, swelling	Bil ascending and deep cervical artery, Bil vertebral artery	7	↓ Swelling, no bleeding	18

Bil = bilateral; L = left; R = right; ↓ = decreased.

resulting in gross disappearance of the malformation. The patient refused subsequent surgery and a follow-up at 9 months after embolization showed no recurrence of the swelling or bleeding.

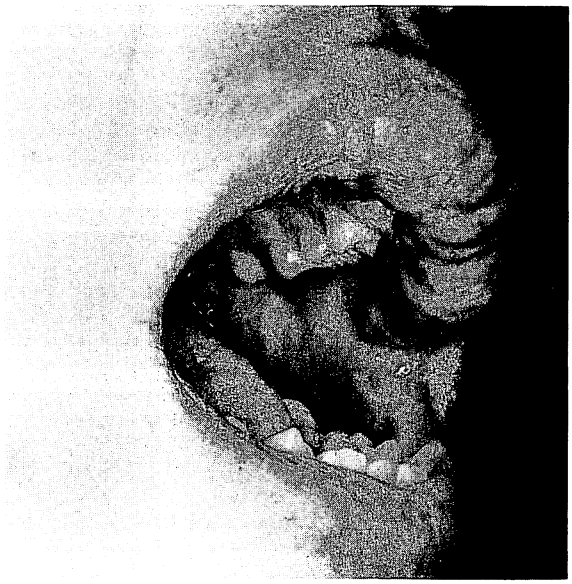
Patient 4

A 32-year-old woman was referred for an obvious ear disfigurement (Fig 2). She had a birthmark on her left ear, which had gradually increased in size and become pulsatile, but was not treated until her pregnancy. At 31 years, during her first pregnancy, she bled from the ear lobe, which was treated by only suturing. A second bleed occurred 4 months after delivery. On admission, her left ear was pulsatile and prominently disfigured. Angiography revealed an AVM of the ear, which was mainly fed by the left posterior auricular artery. The left anterior auricular and the left occipital arteries also contributed; drainage was into the left superficial temporal vein and the left jugular vein. PVA embolization through these feeders showed gross disappearance of the AV shunt and her ear became smaller and nonpulsatile. After this embolization, she was free of bleeding for approximately 1 year.

A follow-up angiography at the end of 1 year revealed some degree of recanalization of the AVM and therefore PVA embolization was repeated during the same session. She refused any surgery on the ear and preferred annual or biannual PVA embolization instead. She has been free of bleeding for 21 months.

Patient 5

This 21-year-old man had a congenital swelling of the right cheek, which gradually increased in size (Fig 3). Since the age of 17 years, he had undergone seven surgical procedures (within 5 years), including a partial resection, electrocoagulation, ligation of both ECAs, suturing of the bleeding sites, and skin grafting. Due to the increasing swelling of the right side of his face, he was referred to us. The first angiogram, done outside, at the age of 17 years showed an AVM mainly fed by the right ECA. Being a conventional angiogram it did not disclose the precise extent of the lesion. Angiography was hence repeated. As a result of the ECA ligation, there was a complexity of feeders supplying the AVM, from the deep and ascending cervical arteries on both sides, from both vertebral arteries, and small branches from both internal carotid arteries. The stumps of both ECAs were seen on angiography. Embolization through a microcatheter was abandoned because it could not deliver large amounts of PVA particles. PVA embolization was therefore performed by direct punctures of the left superficial temporal artery and the right occipital artery, and by placing the catheters at the origins of both deep and ascending



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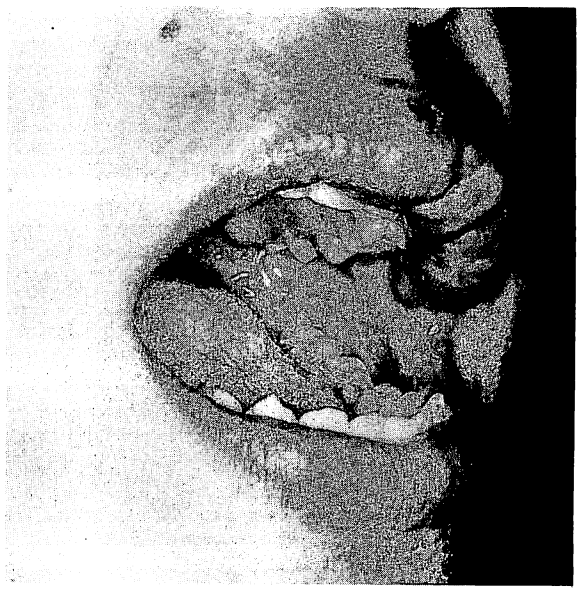

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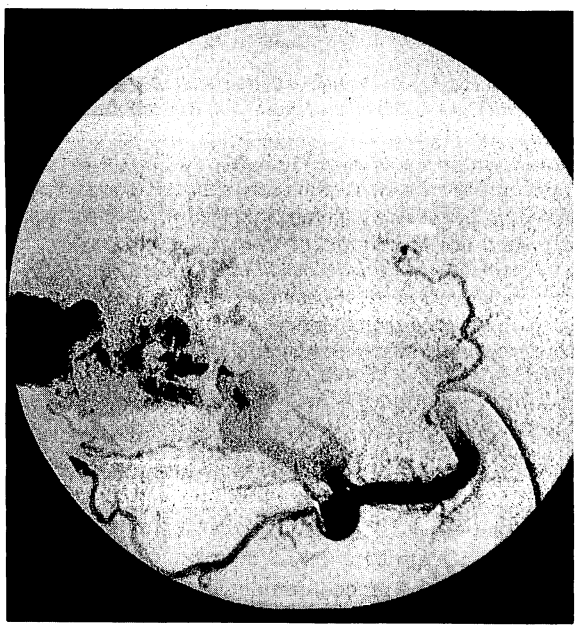

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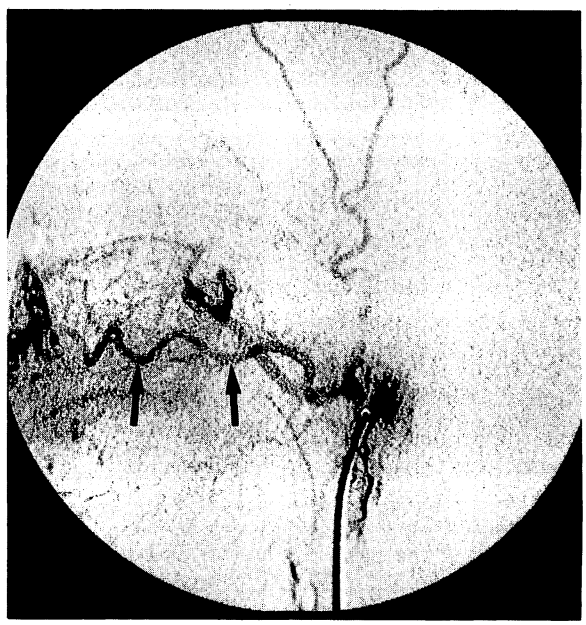

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Fig 1. Patient 1; 21-year-old man with an arteriovenous malformation (AVM) involving the left cheek, upper lip, and maxilla. (A) Preembolization photograph, showing the pulsatile, swelling of the left upper lip. (B) Follow-up photograph taken 1 month after embolization. Reduction of the swelling and disappearance of pulsation were noted. (C) The left facial artery injection reveals the AVM and the aneurysmal dilatation at the upper lip. Draining veins were the superficial facial veins (not shown). (D) The left transverse facial artery (arrows) *is moderately contributing to the AVM.*

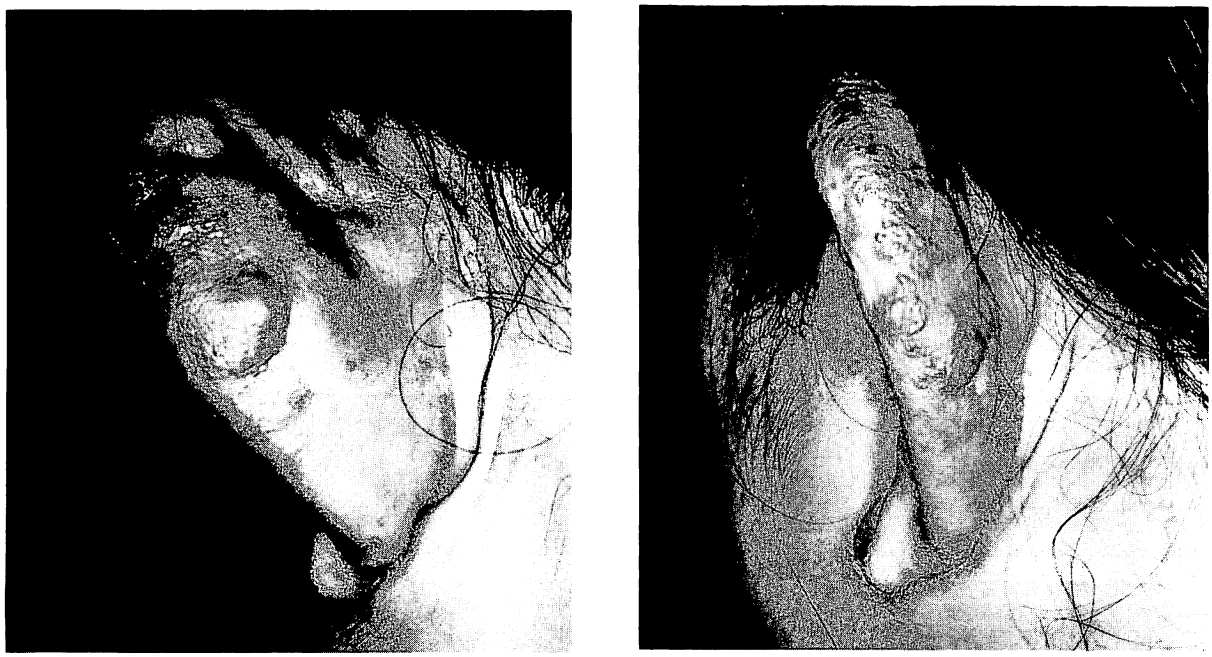

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Fig 2. Patient 4; 32-year-old woman showing arteriovenous malformation (AVM) of the left ear. (A) Preembolization photograph. (B) Postembolization photograph. Reduction of the size and protrusion of the ear are observed. (C) The left posterior auricular artery injection shows the AVM occupying the entire ear lobe and draining into the left superficial temporal vein, and then to the left external jugular vein. (D) The left external carotid injection performed at the 1-year follow-up, which shows partial recanalization of the AVM. Subsequent to the control angiography, embolization was again performed in the same session.

cervical arteries bilaterally, with care being used regarding their anastomoses with intracranial vessels. After seven sessions of embolization, the swelling appreciably reduced in size and no bleeding has been noted during the past 18 months. Embolization for him is regarded as a palliative procedure and may be repeated when necessary.

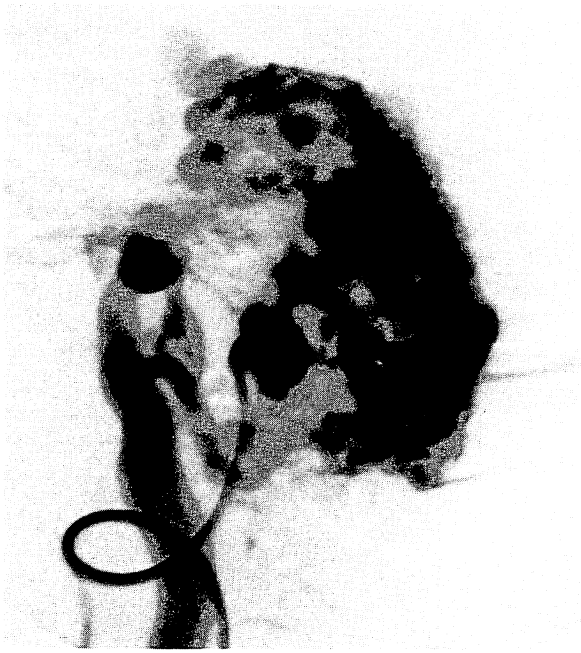
Results

Three patients were embolized in a single session. Patient 4 needed three and Patient 5, seven sessions. Immediate angiographical results showed almost total disappearance of the AVMs in all patients except Patient 5, in whom only reduction of the arteriovenous shunts could be obtained. Swelling (4 of 4 patients), pulsations (3 of 3 patients), and bruit (1 patient) were either markedly reduced or disappeared after embolization. Acute arterial bleeding was controllable (4 of 4 patients). Follow-up periods varied from 6 to 21 months with a mean of 12.4 months.

There were no procedure-related complications. Local pain, swelling, and low-grade fever usually lasted from 3 to 7 days after the embolization.

Discussion

At present, vascular malformations of the face pose a multifaceted problem to the clinician by way of their classification, diagnosis, and treatment. We shall briefly review each of them.



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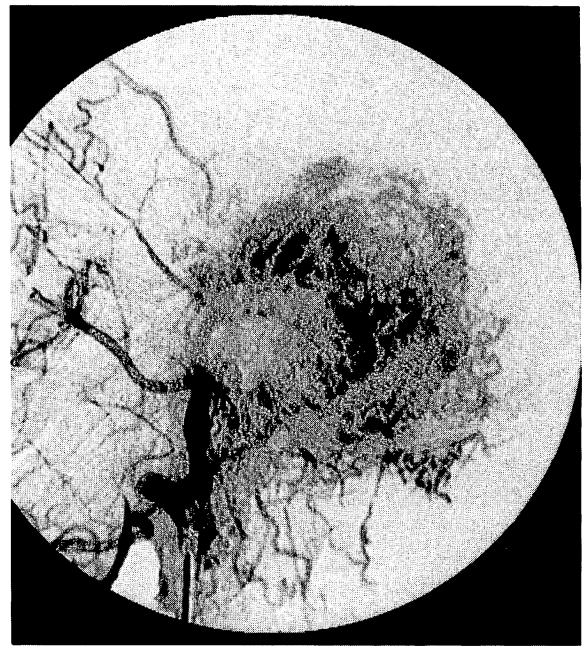

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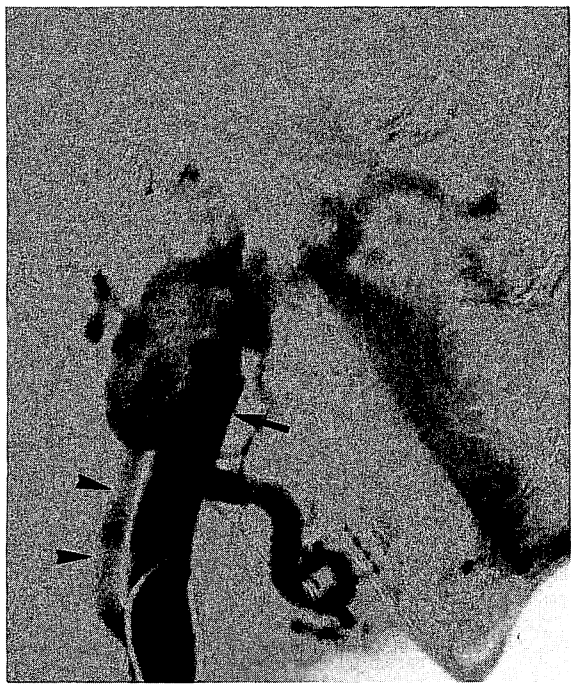
Classification

Various classifications based on embryology, histology, and clinical and hemodynamic characteristics have been offered, but none is universally accepted. Multiplicity of terminologies has finally led to various confusing nomenclatures.

Embryologically these lesions are thought due to failure of the common embryological anlage to differentiate into an artery and a vein. According to Woollard [1], there are the following three identifiable stages of vascular system differentiation: (1) undifferentiated capillary network, (2) retiform, and (3) vascular maturation. Developmental failure in the undifferentiated capillary stage results in capillary or cavernous hemangiomas; the retiform stage, in vascular malformations; and that in the mature end stage results in port-wine stains and lymphangiomas [2]. Because the developmental failure can occur at different stages of development, it is not surprising that a given congenital lesion could contain more than one form of the anomaly.

According to the classification of Edgerton [3], congenital arteriovenous malformations are included under type 4 hemangiomas, that is, arteriovenous fistulae. Mulliken and Glowacki [4] classified pediatric vascular lesions, based on clinical behavior and endothelial cell characteristics, into the following two categories: hemangiomas with a life cycle showing proliferation and involution, and vascular malformations that can be subdivided depending on the vessels involved, that is, any combination of arteries, capillaries, veins, or lymphatics with or without fistulas. According to their classification, hemangiomas exhibit increased cellular activity whereas vascular malformations are cellularly stable. Hemangiomas are benign endothelial neoplasm with a natural history of proliferation and involution, whereas vascular malformations grow commensurately with the patient and never involute. We believe the classification of Mulliken and Glowacki [4] is useful in assessing the prognosis and helps in the treatment planning. Clinical deterioration is due to proliferation of the endothelial cells in hemangiomas and hemodynamic changes in vascular malformations.

Differentiating hemangiomas from vascular malformations histopathologically without clinical and angiographical (hemodynamic) data, at times, can be difficult. Hemangiomas may be present at birth, but usually appear in early infancy, and exhibit rapid growth with subsequent slow regression and do not affect bone [4, 5], whereas vascular malformations usually are present at birth and become clinically



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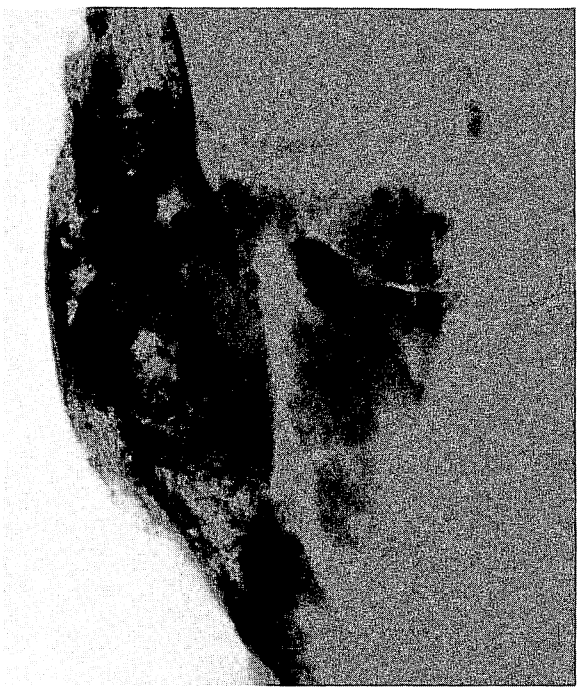

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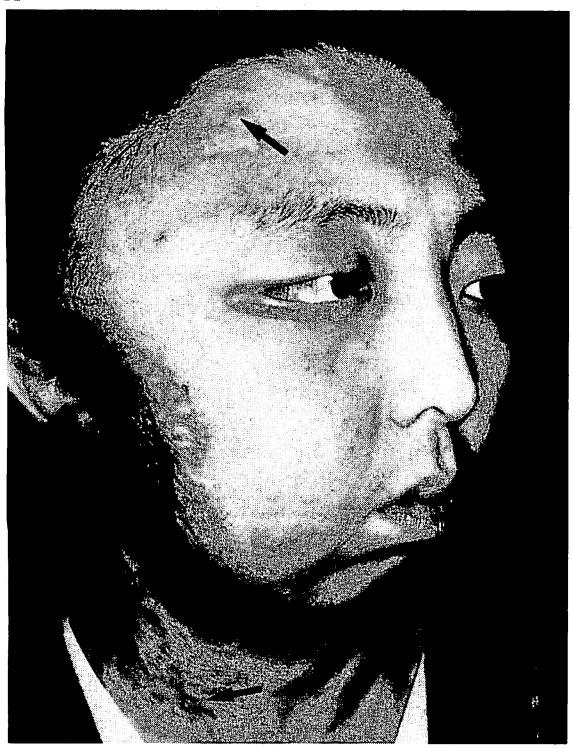

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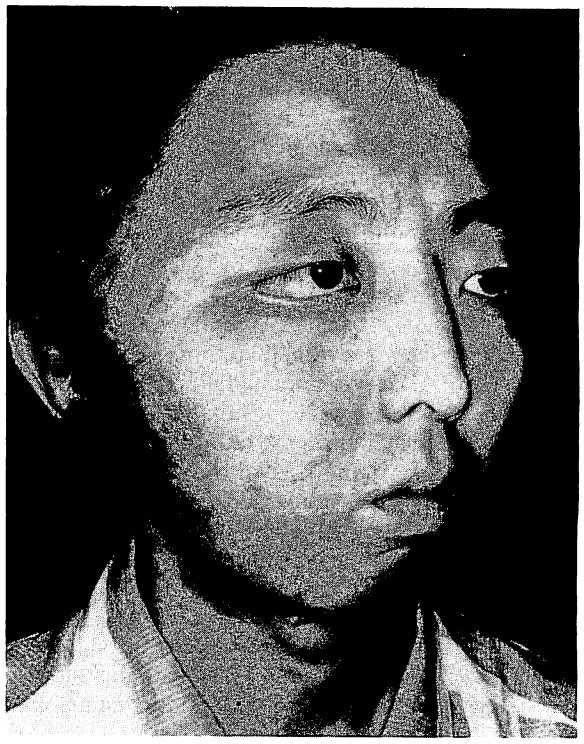

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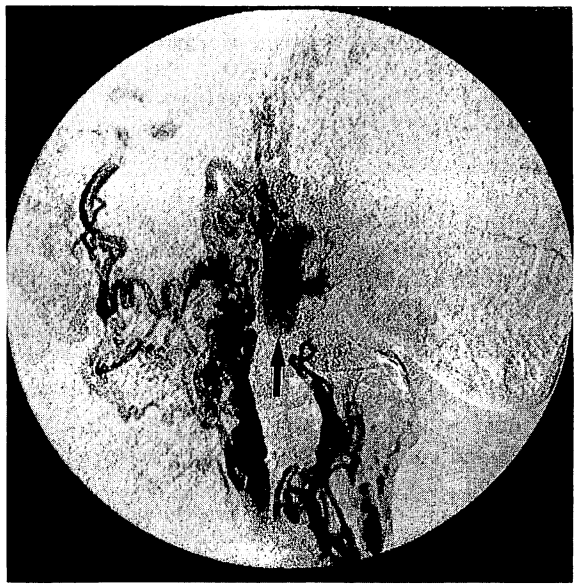

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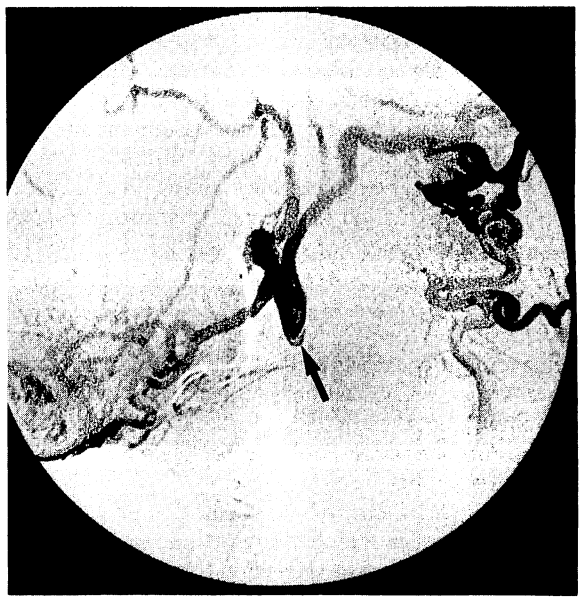

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Fig 3. Patient 5; 21-year-old man with an arteriovenous malformation at the right cheek, temporal region, and maxilla. (A, B) Right common carotid angiography at the age of 17 years. Lateral view showing markedly enlarged external carotid artery (arrow) *in comparison with the internal carotid artery* (arrowheads). *Anteroposterior view showing the tumor-stain involving the right face with ill-defined margin. (C) Preembolization photograph, which shows the pronounced swelling of the right face with scars and skin graft. Note the dilated, collateral superficial arteries at the neck and the forehead* (arrows). *(D) Postembolization photograph, which shows the reduction of the swelling and disappearance of the superficial dilated arteries. (E) The right deep cervical artery injection discloses the retrograde anastomosis from the right occipital artery to the stump of the ligated right external carotid artery* (arrow). *(F) The right ascending cervical artery injection also discloses the anastomosis to the ligated external carotid artery* (arrow). *It is difficult to delineate the lesion due to rich collateral pathways. (G) The left deep cervical artery injection shows the stump of the ligated left external carotid artery* (arrow), *which gives blood supply to the lesion crossing over the midline.*

apparent at a later time after local trauma, infection, and hormone changes associated with puberty or pregnancy. They are often associated with bony abnormalities [4, 5].

Diagnosis

In the absence of a widely accepted classification, differential diagnosis is problematic. Precise clinical and histopathological examination does not always give a clear diagnosis. Clinical course is an important clue in certain situations.

Angiography is essential both for diagnosis and therapy [6, 7]. Superselective catheterization enables precise delineation of the morphology of the lesion and aids in understanding its hemodynamics [6]. Szilagyi and colleagues [8] suggested the presence of angiographically nondemonstrable AVMs (microfistulas) in addition to the demonstrable ones (macrofistulas). We do not think, however, that microfistulas really exist. Visualization of newer collaterals after proximal ligation could be easily mistaken for "conversion of micro to macrofistulas." Computed tomographic scanning and magnetic resonance imaging may add the anatomical and histological information.

Angiographically, hemangiomas are well circumscribed lesions exhibiting intense tissue staining, usually organized in a lobular pattern. Vascular malformations are diffuse lesions consisting entirely of vessels without any intervening tissue staining [9]. Arteriovenous shunting, which is commonly seen in vascular malformations, does not distinguish between the two [9].

Treatment

Management of AVMs of the head and neck includes observation, embolization, surgical resection, or a combination of these [10]. Radiotherapy does not seem helpful. Embolization could be used as a preoperative procedure aiding surgical excision or as a primary method of treatment, whether curative or palliative. The advantages of embolization include the following: (1) a selective access to the lesion through the feeding arteries, sparing the normal tissues, (2) easy repeatability, (3) performance under local anesthesia, permitting continuous monitoring of the patient's neurological status, (4) a provocative Xylocaine test can be performed to assess the possibility of damage to the cranial nerves, especially to the facial nerves, and (5) reduction in size and vascularity helps during surgical resection. The disadvantages include, as follows: (1) various procedure-related complications [11], (2) difficult embolization in patients with prior vascular ligation, and (3) difficulty with complete occlusion of the AVMs, and recanalization and collateralization can occur.

Embolic materials can produce temporary or permanent embolization and can be liquid or solid. The commonly used embolic materials for AVMs of the head and neck are isobutyl-2-cyanoacrylate or *N*-butyl-2-cyanoacrylate as liquids and PVA, silk sutures, and microcoils as solids. Embolic materials used in this series were solely PVA particles (250–600 μm), which were large enough to spare the vessels to the cranial nerves and less likely than the liquid embolus to cause necrosis of the normal tissue. Occlusion with PVA particles, however, can recanalize subsequently.

The ideal goal of a cure is not easy to achieve in patients with head and neck AVMs. Embolization carries the risk of a stroke, cranial nerve ischemia, skin necrosis, bleeding, blindness, and adverse hemodynamic changes, and so on [11], and should be performed by an adequately trained neuroradiologist. Proximal ligation may yield transient benefits but creates problems for subsequent management. Total surgical resection of AVMs of the head and neck may not always be possible. Extensive abrasion involving the face, for this basically nonmalignant lesion, can result in disfigurement unacceptable to the patients in spite of good quality facial reconstruction. PVA particulate embolization can be curative in select patients, such as those with small AVMs fed by a single pedicle, but this is generally regarded as palliative and is useful when the AVM is inoperable, when clinical manifestations are mild, and when the patients refuse surgical excision. PVA embolization, however, "stabilizes" the lesion, which then should be carefully followed up.

Unless our overall understanding of AVMs of the head and neck improves, diagnostic and therapeutic dilemma shall remain unchanged. A close interaction between the plastic surgeons and interventional neuroradiologists is mandatory to "chalk out" various protocols, and long-term follow-up studies are necessary to assess the efficacy of the various modes of therapy.

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