HEAD & NECK SURGERY

OCTOBER 1984
VOLUME 7/NUMBER 1

4.6.17

 ISSN No. 0148–6403

We report on the histologic and anatomopathologic aspects of cutaneous hemangiomas, stressing the involutive and noninvolutive aspects of these malformations. We analyze its clinical evolution, approaching the propedeutic and therapeutic methods, and present a detailed commentary on the conservative methodology of a series of 473 consecutive patients, encompassing a follow-up of 1 to 18 years.

HEAD & NECK SURGERY 7:47–59 1984

CLINICAL EVALUATION AND SURGICAL TREATMENT OF HEMANGIOMATA

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Hemangiomas are benign malformations of the blood vessels that develop independently of the normal adjacent circulatory system.[1,2] Its classification becomes somewhat difficult due to its great variation in size, shape, site, and histological picture.[3–7]

The etiologic factors and natural history of these lesions still remain obscure, resulting in great controversy regarding their treatment.[8,9] Various authors have observed that the majority of hemangiomas tend to involute spontaneously.[8,10,11] Lister,[12] in 1938, was one of the first to report the expontaneous involution of hemangiomas, establishing its evolutive picture, which is characterized by a rapid growth during the first 6 months of age, an arrest in growth from 6 to 12 months, with a tendency to involute slowly and progressively until 5 years of age. Thompson[13] reported 411 cases treated conservatively and followed for 7 years without any treatment. It was impossible to localize the site of the original lesion in 33% of this group since a discreet cutaneous atrophy was observed in 57%. Only 3% of the cases studied did not show involutive characteristics. On revising 326 cases, of which 135 underwent surgery and 62 were treated conservatively, Van der Werf stated that the percentage of favorable results was greater when a conservative approach was followed.

More recently, several articles indicate that almost 95% of hemangiomas show signs of involution.[10,14–16] In this article, we present a methodology and analysis of a conservative treatment approach in a series of 473 consecutive cases with hemangiomas (Table 1), encompassing a follow-up of 1 to 18 years. Two hundred twenty-one patients did not undergo any surgical treatment before the pre-school age, with remarkable involution in 149 of the cases and growth in 46. Twenty-six cases remained unchanged.

In accordance with the current medical literature, we believe that the best cosmetic results are obtained through a conservative approach.[17,18]

CLASSIFICATION

Nowadays, the classification of hemangiomas follows different points of view. These include the histopathologic, morphologic, hemodynamic, and clinical points of view. The histopathologic classification is a result of alterations identified in histopathologic findings. Its correlation to the clinical picture and to the behavior of the lesions is very labile.[19] This is the case when we consider the strawberry nevus, which can vary from a capillary hemangioma[10,20] to a cavernous one.[21,22]

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Accepted for publication May 11, 1984.

0148-6403/0701/0047 $04.00/0

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Table 1. Treatment of hemangiomas in 473 patients.

Treatment	No. of patients
Early surgical treatment	101
Resection following obliterating treatment	151
Conservative treatment	
Involution	149
Noninvolution	46
Unchanged	26

Table 2. The morphologic classification of hemangiomas.

Smooth	Tumoral
Nape erythema	Strawberry nevus
Nevus flammeus	True cavernous
Nevus araneus	Mixed
	Diffuse hemangiomatosis
	Hemangiolipoma
	Hemangiofibroma
	Cirsoid aneurysm

In fact, it is accepted that both types of vessels are frequently present and that the histopathologic picture depends entirely on the lesion being found in a proliferative or regressive static phase. Capillary hemangiomas are so designated because they are composed of blood vessels that, for the most part, conform to the caliber of normal capillaries. Although any organ or tissue may be involved, they usually occur on the face, scalp, and thorax and can vary from a few millimeters up to several centimeters in diameter. They may take the form of large, flat, map-like red-blue discolorations referred to in this pattern as "port-wine stains." Characteristically, they are bright red to blue and on a level with the surface of the skin or slightly elevated and circumscribed. The intense red hue has led to its designation as the strawberry hemangioma, although this term has been used rather loosely with application to capillary cavernous and mixed lesions. Not infrequently, a capillary hemangioma may be overlying a cavernous element.

Arteriovenous fistulas occur somewhat later during fetal development and are classified in macro- and microfistulas on the basis of gross and angiographic studies. The most common symptoms of patients afflicted are deformity, swelling, pain, hyperhidrosis, and hyperthermia of the affected region. Limb hypertrophy as well as varicose vein are often associated. The histopathologic examination will help differentiate a simple vascular anomaly from another and/or associated to other tissues, such as lymphatics (lymphohemangioma), fat cells (hemangiolipoma), and fibroblasts (hemangiofibroma). According to the histologic characteristics, hemangiomas are divided into three groups:[9] capillary, cavernous, and mixed. The morphologic classification (Table 2) is of poor application, being limited to the contours of the region involved. It seems to us that the growth of hemangiomas involves alterations in their architecture and in the level of intensity of their blood flow. Thus, having established the hemodynamic classification of hemangiomas (Table 3), we must keep in mind that the alteration from a low blood flow to a high blood flow indicates a worse prognosis, for it basically changes an inactive lesion into an active one. Cases have been reported in which traumatic factors led to a subsequent excessive growth of the hemangioma on the affected area. Simultaneously, several authors remain unsatisfied with the selective links between the great afferent vessels. This treatment occasionally results in an alteration of the hemangioma picture, with formation of a new system of arteriovenous connections. The monitoring of these architectural alterations and of the degree of blood flow between arteries and veins or through arteriovenous fistulas can help predict the course of congenital hemangiomas (Fig. 1). Thus, in addition to photographic routine and serial measures, Merland et al.[23] be-

Table 3. Hemodynamic classification of hemangiomas.

Low blood flow	High blood flow
Capillary	Arteriovenous fistulas
Cavernous	Arteriovenous malformations
Mixed	

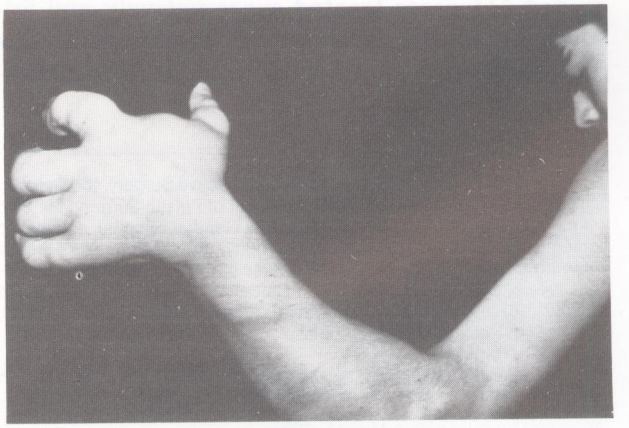

FIGURE 1. Hemangioma of the limb, which frequently leads to gigantism.

Table 4. Clinical classification of hemangiomas.

Involutive	Noninvolutive
Superficial (strawberry nevus)	Port-wine stain
Associated (superficial/deep)	True cavernous
Deep	Racemose angioma
	Arteriovenous malformation
	Diffuse hemangiomatosis
	Vascular spiders
	Angiofibroma
	Telangiectasis
	Malign (very rare)

Table 5. Associated syndromes.

Hemangioma	Syndrome
Capillary	Sturge-Weber
	von Hippel-Lindau disease
Cavernous	Maffucci (dyschondroplasia)
	Blue rubber nevus
Mixed	Klippel-Trenaunay-Weber (hypertrophic hemangiectasia)
	Hepatic disease (Laennec cirrhosis)
Vascular spiders	Pregnancy
	Vitamin B deficiency

lieve that a superselective arteriography helps in studying these malformations. Bingham[14] employed the Doppler machine to identify the number of arteriovenous fistulas. Repeated examinations to prove if the number of these fistulas remain stable or if there is any increase in number will furnish its involutive or noninvolutive characteristics. Being a noninvasive method, the Doppler method can be used in newborns or children, in contradistinction to arteriography, which can only show the momentous architecture of the lesions. These relatively new concepts for monitoring the blood flow in hemangiomas play an important role in the choice of the appropriate treatment, with great prospects of remaining fundamental for the treatment of these lesions.[5,24]

The nomenclature for clinical classification may seem confusing due to the variety and mixture of morphologic, histopathologic, and clinical characteristics. Despite this, the essence of the clinical classification lies in the possibility of a spontaneous or nonspontaneous regression of the lesion.[24] In this manner, it is more relevant to think in terms of involutive and noninvolutive lesions (Table 4).

Eighty to 90% of cutaneous hemangiomas involute spontaneously; some authors indicate that there is an age limit in which this normally occurs. Nonetheless, there are exceptions in spontaneous involutions and these are particularly related to involutive hemangiomas of the mucous membrane that persist in adult life.[7,24]

Several factors alter the clinical picture of the disease, such as ulceration, necrosis, secondary infection, hemorrhage, trauma, and wound healing, thus hindering an accurate definition of the natural history of the lesion. Adding to this, hemangiomas are frequently associated to other syndromes, which alter the general clinical picture[24] (Table 5, Figs. 2 and 3).

TREATMENT

The treatment of hemangiomas is mainly conservative since the majority involute during the first years of life.[15,16,25] Patients must undergo periodic revisions in their treatment, since the lesion will be monitored through photographic measures, xeroradiography, ultrasonography, and the Doppler method. Superselective arteriography is a propedeutic method that we use to evaluate the extent of the hemangioma lesion. We applied this

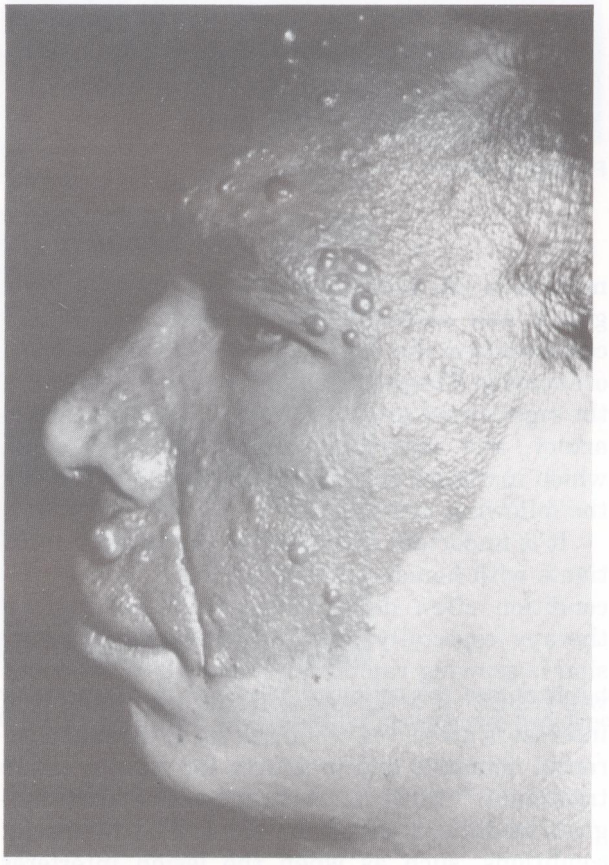

FIGURE 2. Sturge-Weber syndrome.

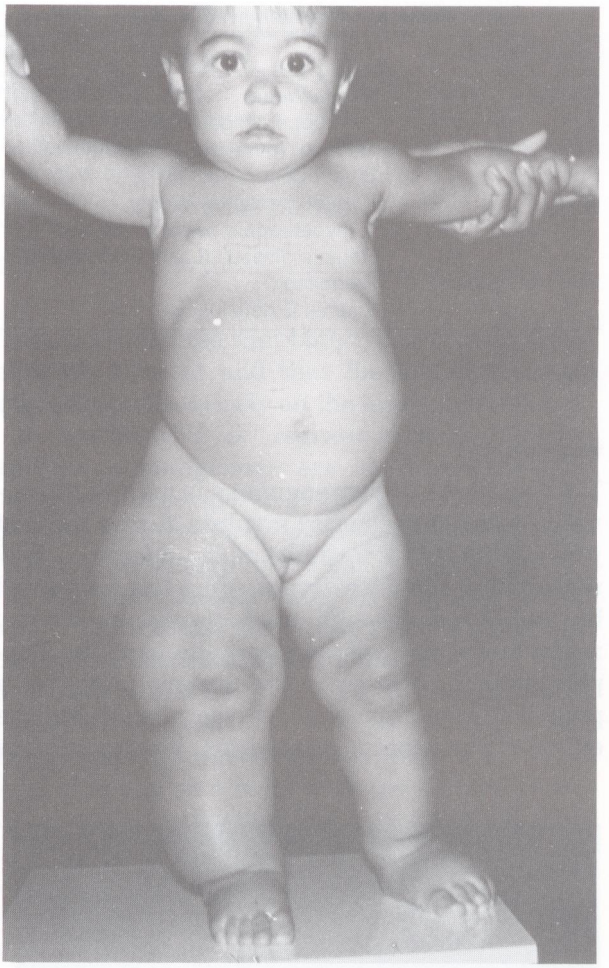

FIGURE 3. Klippel-Trenaunay syndrome.

method to one patient with a cavernous hemangioma and various arteriovenous fistulas that compromised the inferior third of the face and oral cavity. The patient underwent obliteration of the right facial artery, the right external carotid artery, and some branches of the left artery, which increased the collateral circulation during the following months.

It is important to explain to the parents of patients with hemangiomas that the nature of this condition often demands an immediate active therapy, especially during the phase when the lesion is growing rapidly and anatomic distortions will result.[1] It is our opinion that the child's best interest is preserved when the surgeon resists parental demands and pressures for an aggressive treatment.[26] However, it is obvious that early surgical treatment should be considered in special conditions, such as when the lesion interferes with sight,[11] breathing, and suction, when it is in the presence of atypical lesions, when the diagnosis is doubtful, when severe bleeding or ulceration do not respond to conservative therapy, or when the lesion is growing rapidly.[27] Some lesions that can be immediately resected are considered for surgical treatment, particularly in cases of cavernous hemangiomas that can lead to regional tissue destruction due to multiplication and proliferation of cells[5,6] (Figs. 4 and 5).

If the hemangioma presents rapid development, treatment should be indicated in the first months of life, thus avoiding a late and more complicated surgical procedure[4–7,24,28] (Fig. 6). Large hemangiomas, where an augmentation in the incidence of thrombocytopenia and/or intravascular disseminated coagulopathy is present, are another indication for surgery in children. The Kasabach-Merritt syndrome seems to be a defined entity, but its etiopathology is unknown.

When thrombocytopenia occurs, it is almost always fatal, and the indication for an early surgical approach can be life-saving.[29]

Involutive Hemangiomas. Thompson[13] and Moore[30] reported the use of pressure to control hemangiomas and showed two satisfactory results. This procedure has been recommended for localized lesions on the limbs, where elastic pressure is applicable.[31–33] The senior author has been using various facial splints made of easy molding material (Orthoplast^{R}). The exact pressure mechanism is unknown but seems to be the end result of a narrowing of the vessel lumen, blood flow stasis, and subsequent thrombosis (Fig. 7). In 1976, Edgerton[34] and Yamagishi et al.[35] reported using steroids injected systemically or locally for limiting the rapid growth of hemangiomas and inducing a premature involution. Nevertheless, they observed that corticosteroids must be administered during the growth period.[36–39]

Noninvolutive Hemangiomas. This group includes those patients who show no evidence of resolution until the pre-school age.[40] Surgical treatment consists of excising both capillary and cavernous hemangiomas whenever possible, followed by direct repairing or advancement and/or flap rotation, or skin grafting (Fig. 8).

When the extension and/or localization of the lesion renders the excision difficult, the senior author prefers the obliterating method. This treatment can be curative or can constitute an important step in preparing for surgery, diminishing

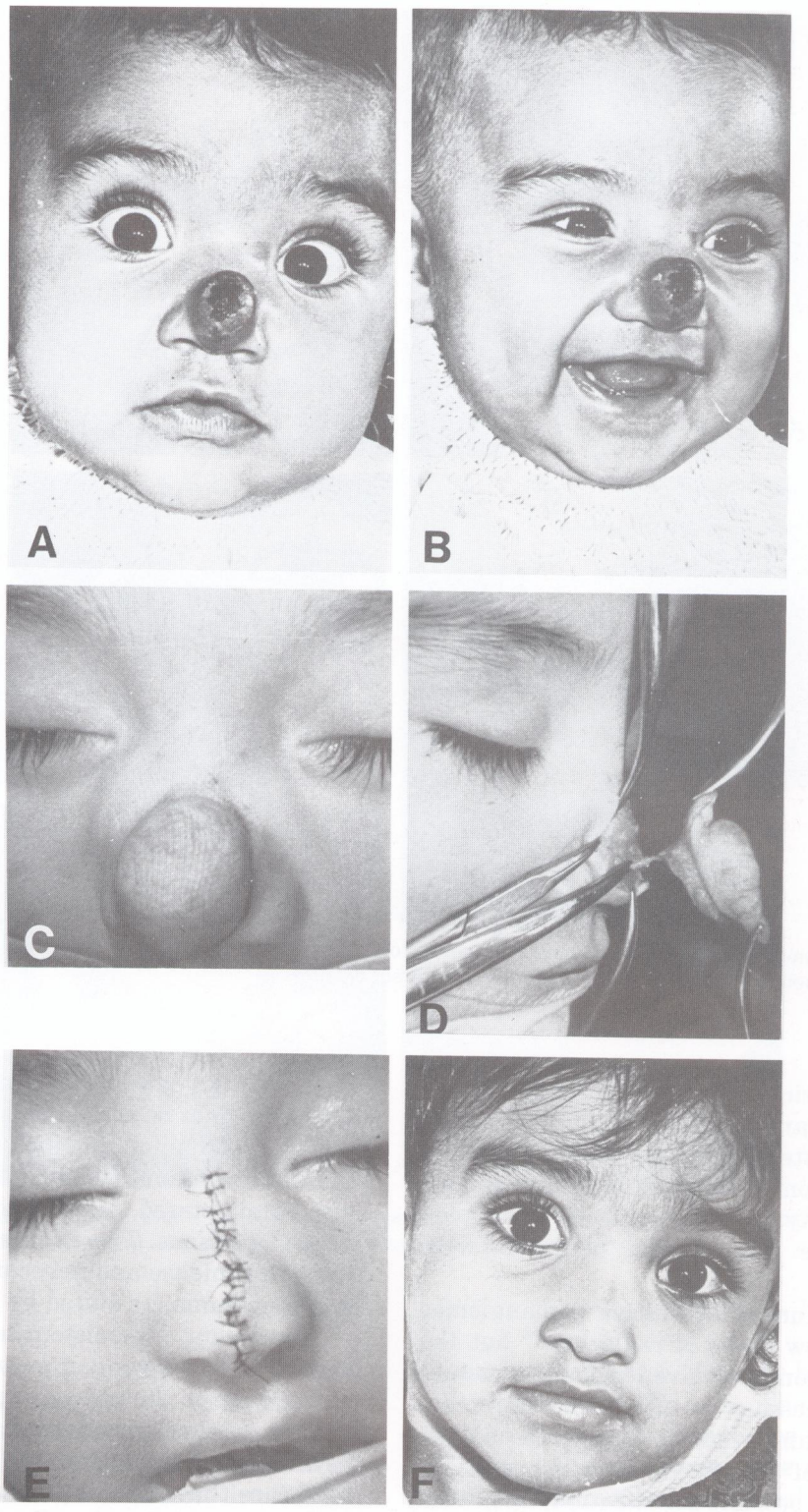

FIGURE 4. (A–C) Four-month-old female patient with a mixed hemangioma who underwent sclerosing and carbon dioxide therapies elsewhere. She presented with progressive ulceration. **(D–E)** She underwent resection of the hemangioma and direct closure. **(F)** Two-year follow-up.

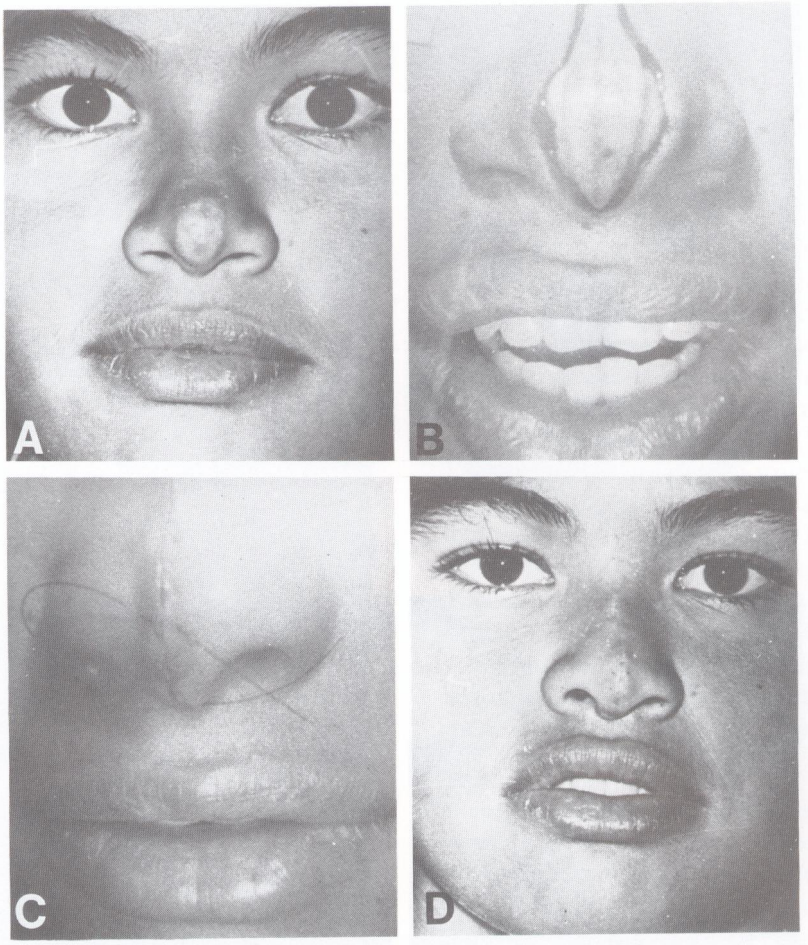

FIGURE 5. (A) Thirteen-year-old patient with nasal tip hemangioma, which increased progressively, with bleeding episodes. **(B–D)** A longitudinal cuneiform resection was performed plus direct closure.

the size of the lesion which will be resected subsequently.[41,42] Various methods of obliteration have been suggested, such as electrocoagulation, diathermy, carbon dioxide, and radiotherapy; the great majority of these methods have been abandoned since the results obtained were poor.

The endothelium of involutive hemangiomas is sensitive to low doses of radiation,[46] but the endothelium of noninvolutive lesions is resistant (the dosages necessary to affect the tumor are more noxious than curative). Due to the deleterious potential of its effects, such as radiodermitis, neoplasia, possible sterilization, and facial atrophy, we contraindicate radiotherapy for treatment of hemangiomas.[3,21,33,47–55] The senior author prefers obliteration with sclerosing agents, particularly in cases of cavernous hemangioma, or in cases of capillary hemangioma associated to deep cavernous hemangioma, and when surgery cannot be considered.[20,56,57] He gives preference to Ethamolin[R], since it attempts to reach the main afferent vessels.[3,53] Routine injections are administered, with 1-month intervals between injections. The sclerosing agent is diluted in hyaluronidase and novocaine to avoid necrosis and hyperemia, as stated by Lewis[58] (Fig. 9).

Treatment of persistent hemangiomas not resulting in loss of contour or anatomy may present a challenge to the plastic surgeon.[46]

The nevus flammeus can provoke significant psychological problems and the surgeon must take care that the treatment does not result in a more severe deformity. Besides considering the cosmetic characteristics of such lesions, it is also important to define the presence or absence of a subjacent and deeper element, since a badly conducted operation may result in the transforma-

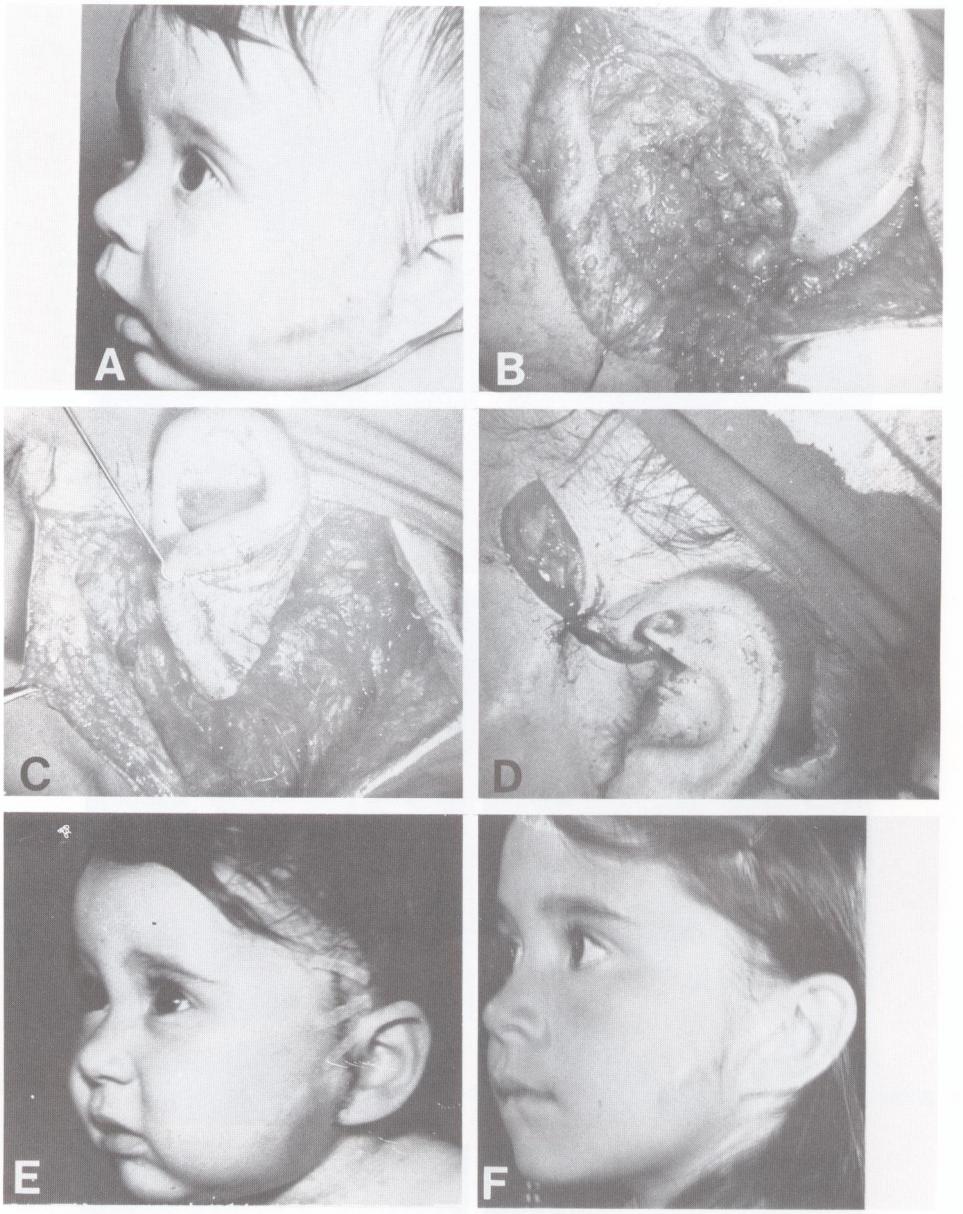

FIGURE 6. (A) Six-month-old patient with a left hemifacial mixed hemangioma. **(B–E)** The patient underwent resection of the hemangioma followed by undermining and advancement of flaps. **(F)** Five-year follow-up.

tion of a low blood flow state into a high blood flow state with disastrous results.

Port-wine stains can be treated by excision and direct closure. More extensive areas may benefit from serial excisions or from local flaps. The latter is preferable to skin grafting since they produce better results. Sometimes this becomes impossible due to the inconvenience of various operations and, occasionally, to technical difficulties related to the localization of the lesion.[5,33,54] In these cases, the use of grafts becomes necessary after excision[24] (Fig. 10). The extension of the grafted area can eventually be reduced by means of consecutive resections. The cutaneous dermal over-grafting, as recommended by Rees and Cassan,[54] improves the aspect of the grafted area.[59,60] Dermabrasion is another method for treating port-wine stains, and is indicated when the depth of the lesion does not surpass the superficial layer of the dermis. A biopsy can help determine the

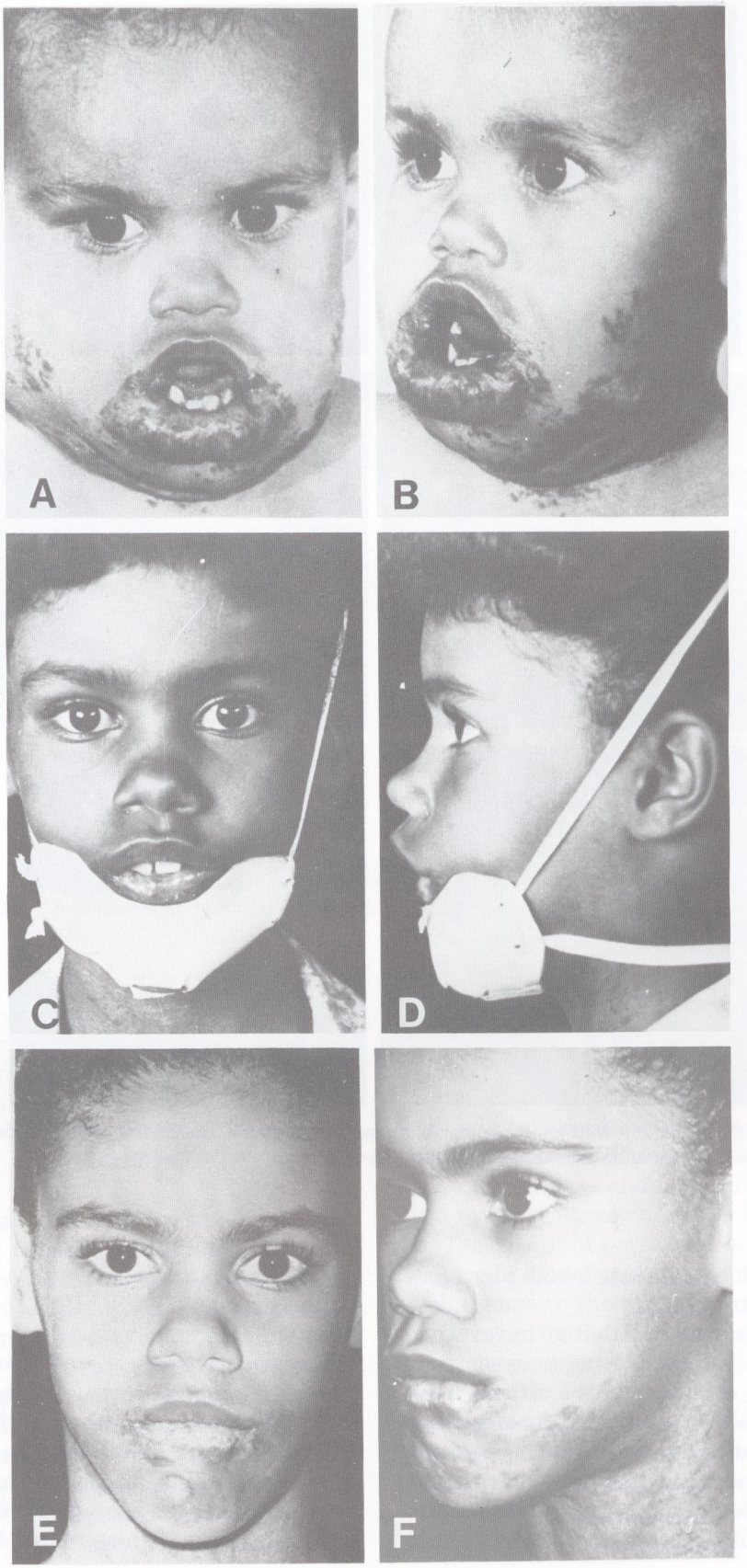

FIGURE 7. (A,B) Eighteen-month-old patient with a mixed hemangioma involving the lower third of the face and neck. **(C,D)** The patient underwent several partial resections and compression treatment. **(E,F)** Nine-year follow-up.

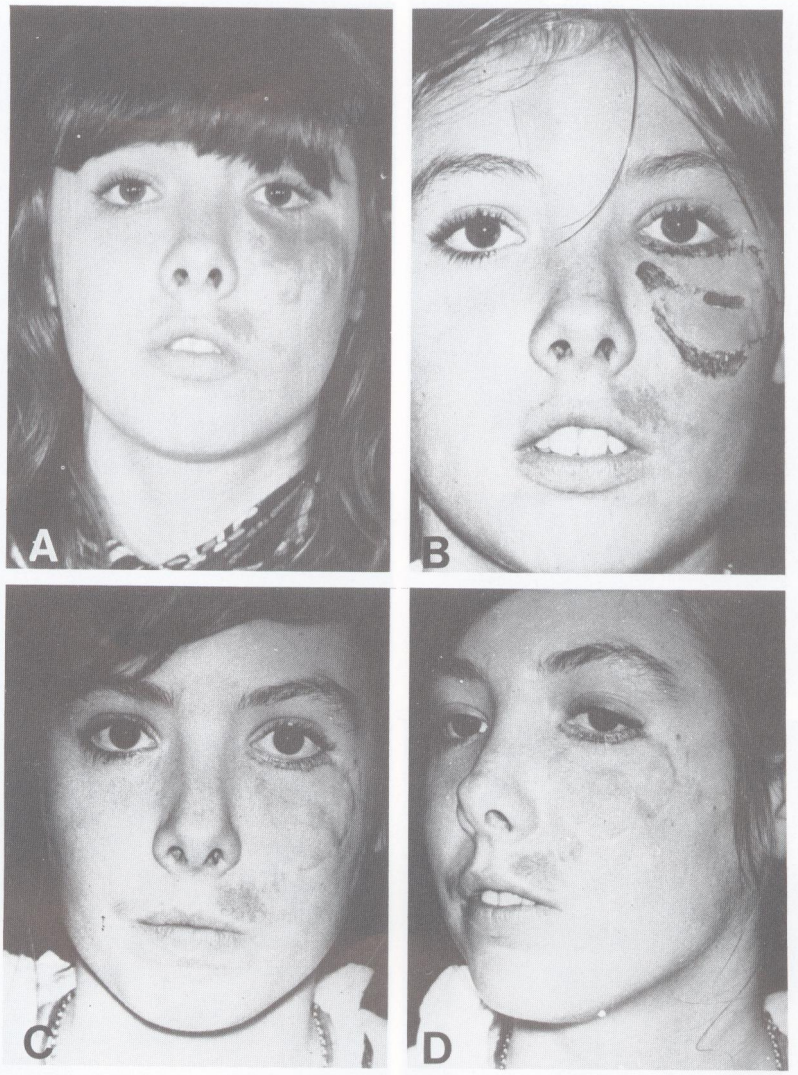

FIGURE 8. (A) Patient with nevus flammeus. **(B–D)** She underwent resection and full-thickness grafting with subsequent resections and advancement of the lower flap.

level of the lesions and the indication for this procedure. Some authors advocate serial dermabrasions since this can lead to thrombosis of the lesion. Conway and Hugo[61] advised tattooing, which yields acceptable results in certain cases. Cryotherapy has been indicated for treating port-wine stains. But one of the most interesting elements is the use of the Argon laser, since a growing number of articles are reporting successful treatment with it.[59,62–66] This type of treatment seems to produce superior results; it is a safe and effective method in comparison to other treatments used formerly. Hemangiomas with arteriovenous malformations and fistulas are more difficult to treat, since a hemodynamic evaluation is essential. A superselective arteriography can determine the extension and nature of the lesion.

Embolization employing muscle, silicone, hooks, and a large variety of other agents has been employed by various authors. Such methods can be curative or can be employed before surgical excision, allowing for a more secure and efficacious operative technique.

Care must be taken when an incomplete excision of the lesion is considered, since it can change the lesion to a high-risk type.

Techniques, such as the Doppler method and selective arteriography, seem to be the key to a difficult case therapy, allowing for a more rational planning of the treatment.[24,54]

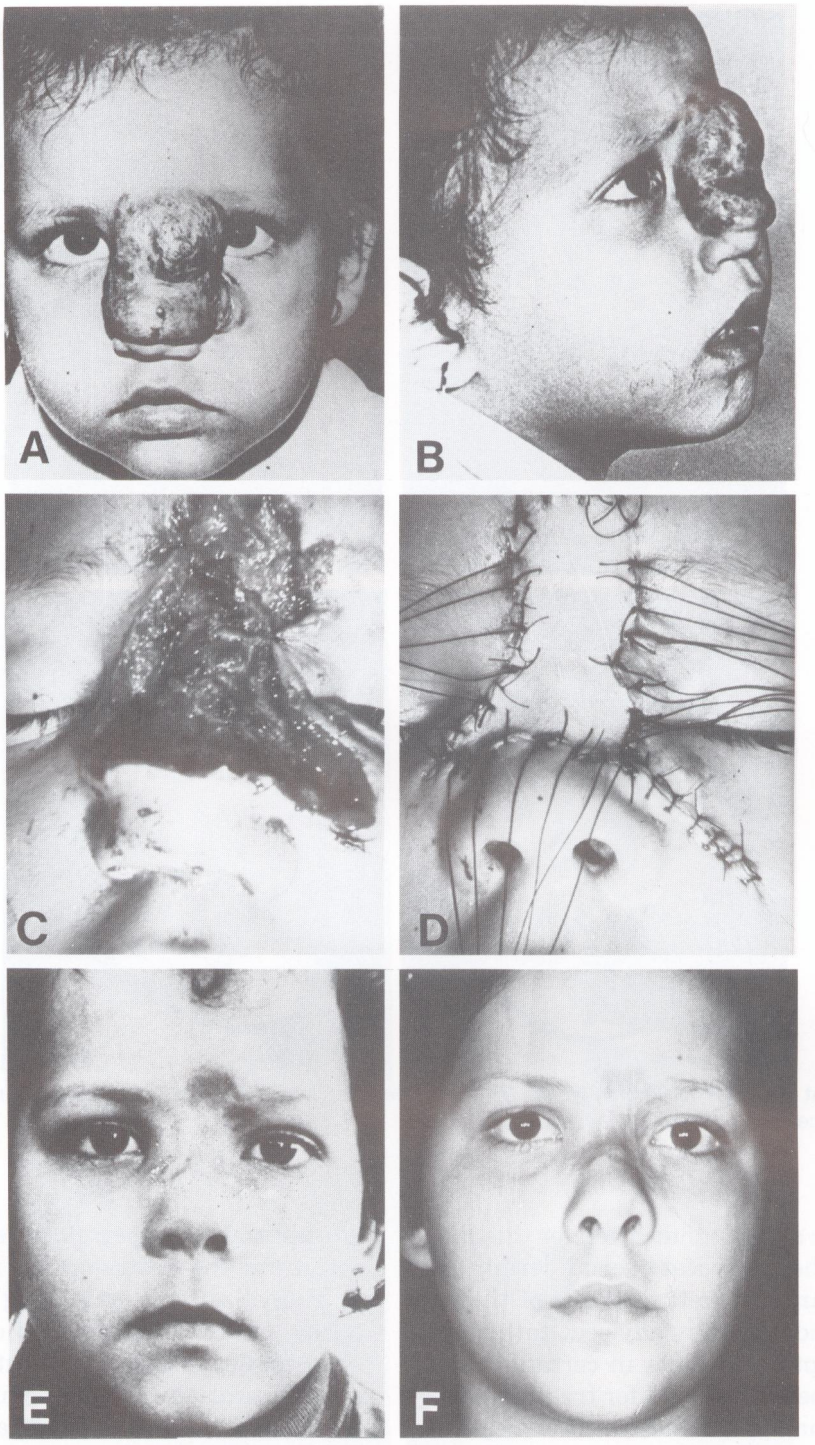

FIGURE 9. (A,B) A patient with a port-wine stain on the glabellar region, which increased up to 6 months of age. **(C–E)** The patient submitted to exeresis and a retroauricular skin graft. **(F)** Fifteen-year follow-up.

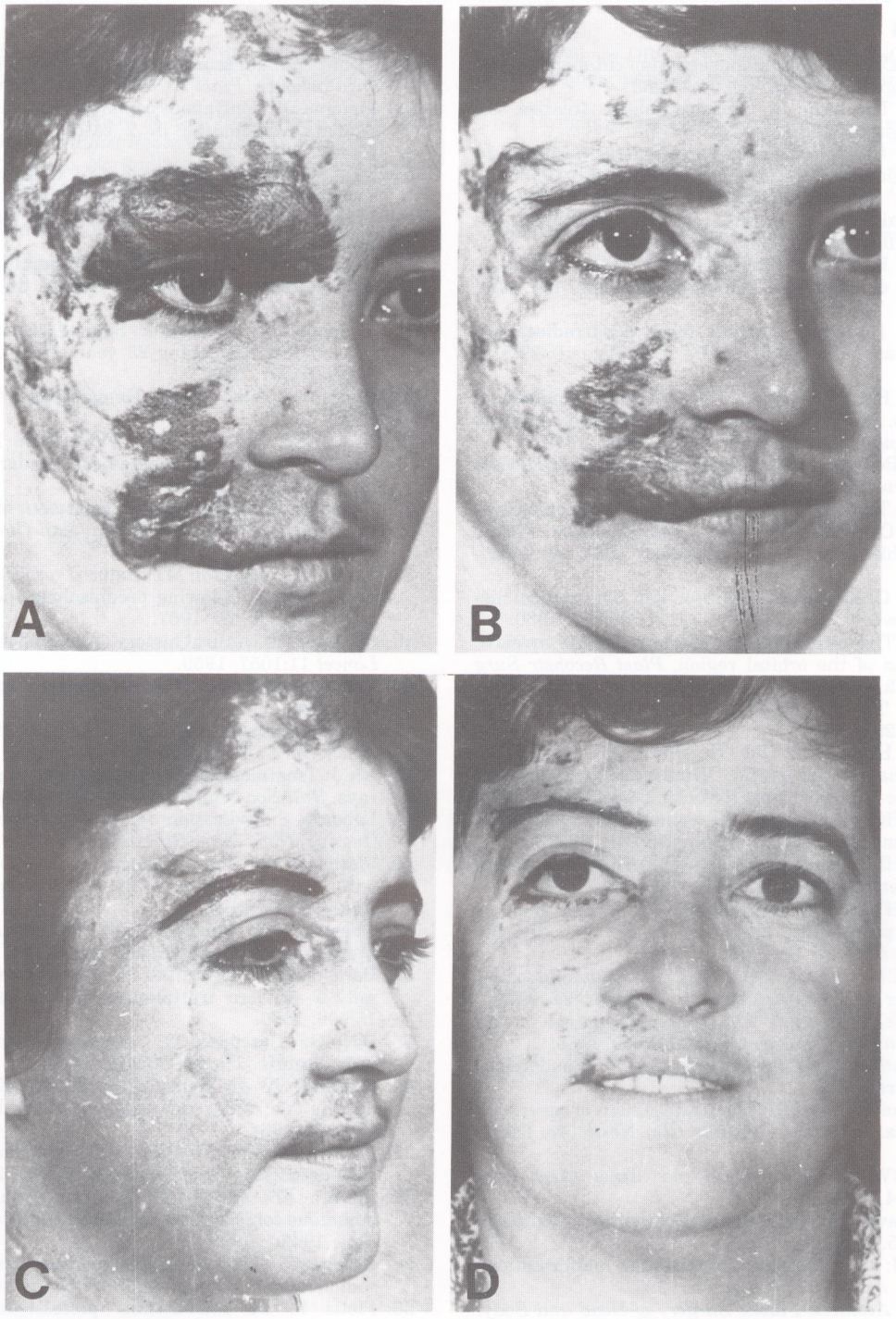

FIGURE 10. (A) Twenty-three-year-old patient with right hemifacial mixed hemangioma. The patient underwent sclerosing therapy followed by multiple resections and reconstruction of the facial lesions, including total reconstruction of the upper eyelid and right eyebrow. **(B)** Six-month follow-up, **(C)** 12-month follow-up, **(D)** 18-year follow-up.

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REFERENCE: (2)OT 155
Pitanguy, Ivo; Caldeira, Alberto M. L.; Calixto, Carlos Alberto; Alexandrino, Adelson
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Head Neck Surg. 7:47-59, October 1984
Table I

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