Surgical Treatment of Facial Infantile Hemangiomas: An Analysis Based on Tumor Characteristics and Outcomes

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Background: Surgical treatment of infantile hemangiomas may interfere with patient appearance. The use of an algorithm is essential to select candidates. The objective of this study was to evaluate outcomes of surgical treatment based on tumor characteristics.

Methods: Seventy-four patients were treated surgically between 1997 and 2010. Demographics, tumor characteristics, surgical approach, and outcomes were evaluated.

Results: The female-to-male ratio was 5.7:1. Mean age and follow-up were 24 years and 33 months, respectively. Surgery was elective in 83.8 percent and emergent in 16.2 percent of patients. Most frequent locations were lips, nose, eyelids, and cheeks. Surgery was performed during the proliferative phase in 43 patients (58.1 percent), and growth-related deformity was the main indication. No significant association between sex and the presence of complications or treatment indication was observed. Patients who underwent emergency procedures were younger (p = 0.0031) and had a higher incidence of evolutional complications (p = 0.012). Also, they were more frequently operated on during the proliferative phase (p = 0.011). Favorable outcome of surgical treatment was observed in both simple and complex cases for facial contour, volume reduction, and need for reoperation. The best candidates for elective surgery were patients with localized eyelid, nasal, or lip hemangiomas, presenting growth-related deformities during the proliferative phase. For patients undergoing emergency procedures, the best candidates were nonresponders to pharmacologic therapy with segmental periorbital hemangiomas, treated by partial resection.

Conclusions: A profile of patients and their specific surgical approach was established. Satisfactory results could be achieved following the proposed algorithm. (*Plast. Reconstr. Surg.* 137: 1221, 2016.)

CLINICAL QUESTION/LEVEL OF EVIDENCE: Therapeutic, IV.

Infantile hemangiomas occurring in the face may represent a real problem to a child. Clinical significance is ultimately determined by the degree of tissue deformation.[1–4] Large dimensions; specific locations; and the presence of complications such as ulceration, bleeding, or infection indicate active treatment to minimize morbidity. Treatment modalities include pharmacologic therapy (systemic or intralesional) and interventional procedures (surgical resection or intralesional laser).[5–12]

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Surgical removal of infantile hemangiomas is a fast and definitive solution. Surgical approach during proliferative or involutive phases has been extensively questioned, mainly after the established use of oral beta blockers. In contrast, treatment of residual deformities during the involuted phase is easily justified because pharmacologic treatment is not effective anymore. Surgical treatment may definitively interfere in patient appearance, and this scenario strengthens the elaboration of precise indications for surgery and a comprehensive protocol for its approach.[5,9–11,13–17]

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The combination of clinical features and response to pharmacologic treatment are the main standpoints indicating surgery during the active phases of infantile hemangiomas. Planning must consider the indication for surgery (an emergency procedure or electively planned), approach (direct to the lesion or through a distant incision), and type of resection (partial or complete).

Over the past decade, an algorithm for surgical treatment was adopted. The goal of the present study was to evaluate outcomes of surgical treatment based on tumor-related clinical features.

PATIENTS AND METHODS

The present study was approved by the institutional ethical committee (protocol number 0931/09) and informed consent documents were obtained from parents or guardians. From February of 1997 to May of 2010, 208 pediatric patients with cervicofacial hemangiomas were treated at our unit. Conservative follow-up was adopted in 86 patients (41.3 percent). Active treatment by exclusive pharmacologic treatment (oral corticosteroid or propranolol) was applied in 48 patients (23.1 percent). A surgical approach was used in 74 patients, corresponding to 35.6 percent of cases. The indication for treatment modalities (Table 1) was based on dimensions, risk of growth-related deformities (resulting from mass effect caused by infantile hemangioma growth), and presence of complications (ulceration, bleeding, or infection). Indications for surgical treatment followed the algorithm illustrated in Figure 1.

In this series of 74 patients who underwent surgery, 63 were female (85.1 percent) and 11 were male (14.9 percent) (female-to-male ratio, 5.7:1).

Table 1. Indications for Treatment of Facial Infantile Hemangioma

Conservative (expectant)
IH <10 mm in diameter *and*
Absence of growth-related deformities *and*
Absence of systemic and local complications
Pharmacologic
IH >10 mm in diameter *and*
Presence of growth-related deformities except eyelids, nose, and lips *or*
Presence of systemic or local complications
Surgical
Presence of growth-related deformities in eyelids, nose, and lips *or*
Presence of growth-related deformities in other locations, not responding to pharmacologic treatment *or*
Presence of systemic or local complications not responding to pharmacologic treatment *or*
Involuted lesions with deformities

IH, infantile hemangiomas.

Fifty-one patients (68.9 percent) were white. The mean age at surgery was 30.9 months (median ± SD, 24 ± 27.2 months). Specific infantile hemangioma characteristics were evaluated in operative patients according to the following parameters:

Anatomical distribution: forehead, eyelids, nose, lips, cheeks, ear, cervicofacial, and hemifacial.
Involvement in extension: localized (respect anatomical unit) or segmental (compromise of more than two contiguous anatomical units or hemifacial).
Laterality: right side, left side, central, and bilateral. Unilateral cases in which the hemangioma slightly crossed the midline were not considered as bilateral.
Compromise in depth: superficial (only skin), deep (only subcutaneous tissue), and mixed.
Evolutional phase at surgery: proliferative, involutive, or involuted.
Presence of complications: local nonspecific (ulceration, bleeding, or infection), local specific (visual or airway obstruction), or systemic (respiratory failure, heart failure, or systemic infection).

Indications for surgery were classified as follows:

Emergency: functional involvement of organs and systems with obstruction of the visual axis or the airway.
Elective: presence of growth-related deformities, recurrent complications, and treatment of involuted hemangiomas.

Surgical tactic was evaluated considering:

Access to infantile hemangiomas: direct (through perilesional or intralesional incisions) or indirect (incisions distant to infantile hemangiomas with need for soft-tissue undermining).
Type of resection: partial (when no more than 50 percent of total volume was removed), subtotal (when more than 50 percent was removed), or total (complete excision).
Reconstruction method: primary closure or local flaps.

Follow-up was registered, as was the number of procedures performed per patient and the occurrence of postsurgical problems. To describe the profile of patients treated by surgery, all clinical characteristic and surgical information were crossed and statistical evaluation was performed.

Outcomes were analyzed by three independent plastic surgeons, not involved in patient

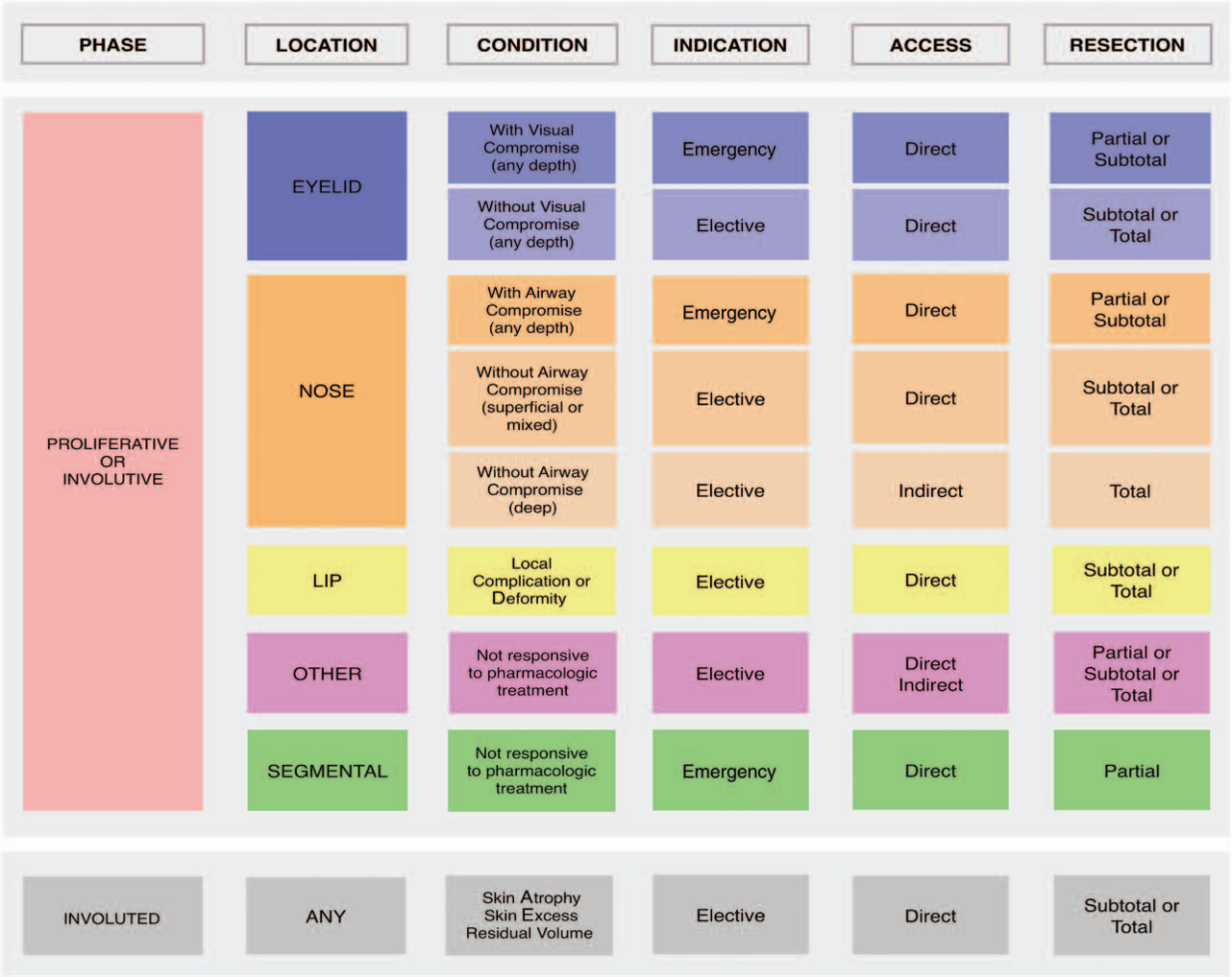

Fig. 1. Algorithm for indication of surgical treatment based on clinical features.

treatment. Preoperative and latest postoperative photographic documentation (at least 6 months after surgery) was used for analysis. In cases of patients with multiple lesions operated on, the area was identified with arrows on preoperative imaging. Questions were answered regarding surgical difficulty (easy, medium, or high), facial contour (worsening, slight improvement, or great improvement), volume reduction (worsening, slight improvement, or great improvement), and need for reoperation (not necessary, need for small additional procedures, or need for similar or larger procedures). Interobserver agreement and scores attributed to each parameter were evaluated statistically.

Statistical Analysis

Stata Statistical Software, Release 10 (StataCorp, College Station, Texas), was used to perform the statistical analysis in patients submitted to surgical treatment. Frequency distribution was used to describe categorical variables (sex, ethnicity, location, treatment categories, and evaluations), and measures of central tendency (mean and median) and variability (minimum, maximum, and standard deviation) were used to describe numerical variables (age and follow-up).

Fisher's exact test was applied to verify association between categorical variables. For analysis of the numerical variables, the Mann-Whitney nonparametric *U* test was applied for two category groups. The chi-square test was adopted to verify independence among scores of evaluators for quality of each item and the kappa index was used to check the degree of agreement among raters. A significance level of 5 percent was considered for all statistical tests.

RESULTS

Surgery was the unique approach for 57 patients (77 percent). In the remaining 17 cases, pharmacologic treatment was also used. Surgery was concurrent

in three cases (4.1 percent) or followed drug therapy because of partial response [14 cases (18.9 percent)] or absence of response [two cases (2.7 percent)].

A total of 90 surgical procedures were performed. The distribution of clinical variables at the moment of surgery (localization, extension, laterality, depth, evolutional phase, and evolutional complications) is summarized in Table 2.

Anatomical Distribution

The most frequent locations were lips, nose, eyelids, and cheeks, totalizing 54 cases (73 percent). The lips were involved in 20 patients, the upper lip in nine patients and the lower lip in 11 patients. Of the nine eyelid lesions, four involved the upper eyelid, three involved the lower eyelid, and in two patients both eyelids were compromised. Nasal hemangiomas occurred in 16 cases, and exclusive involvement of the nasal tip was found in 10 patients. The perinasal area was affected in five patients, and in only one case, it was restricted to the nasal dorsum.

Extension of the Affected Area

In 59 patients (79.7 percent), hemangiomas were restricted to a specific anatomical unit. In the remaining 15 cases (20.3 percent), either the hemangioma was larger than or affected more than one unit. Infantile hemangioma was classified as localized in 64 patients (86.5 percent) and segmental in 10 patients (13.5 percent).

Table 2. Distribution of Surgical Cases According to the Intraoperative Variables

Variable and Category	Frequency (%)
Involvement	
Localized	64 (86.5)
Segmental	10 (13.5)
Localization	
Forehead	5 (6.8)
Eyelids	9 (12.2)
Nose	16 (21.6)
Lips	20 (27.0)
Cheeks	9 (12.2)
Ear	2 (2.7)
Cervicofacial	5 (6.8)
Hemifacial	8 (10.8)
Superficial	18 (24.3)
Involvement in depth	
Deep	15 (20.3)
Mixed	41 (55.4)
Proliferative	43 (58.1)
Evolutional phase	
Involutive	22 (29.7)
Involuted	9 (12.2)
Laterality	
Right	20 (27.0)
Left	29 (39.2)
Central	22 (29.7)
Bilateral	3 (4.0)
Involvement of the midline	
Yes	33 (44.6)
No	41 (55.4)
Lesion restricted to the anatomical unit	
Yes	59 (79.7)
No	15 (20.3)

Laterality and Impairment of the Midline

Infantile hemangioma was unilateral in 49 patients (20 right and 29 left). In 22 patients (29.7 percent), lesions were centrally located, and in three patients (4.0 percent), the hemangioma compromised both sides of the face. However, considering unilateral cases with slight involvement of the contralateral side, the midline was compromised in 33 patients (44.6 percent).

Depth of the Affected Area

Mixed lesions were predominant and occurred in 41 patients (55.4 percent). Superficial infantile hemangiomas were seen in 18 cases (24.3 percent) and deep lesions were seen in 15 cases (20.3 percent). Nine of the deep hemangiomas (60 percent) were located in the nasal region.

Evolutional Phase

When surgery was performed, 43 of the hemangiomas (58.15 percent) were in the proliferative phase and 22 (29.7 percent) were in the involutive phase. Only nine patients (12.2 percent) had hemangiomas resected in the involuted phase. The female-to-male ratios were 6:1, 4:1, and 8:1 for patients operated in the proliferative, involutive, and involuted phases, respectively.

Presence of Complications

Before surgical treatment, 15 patients presented local nonspecific complications, accounting for 20.3 percent of the cases. Ulceration occurred in all 15 patients, concomitantly with infection in five cases and bleeding in one case.

Concerning specific complications, obstruction of the visual axis (total or partial) occurred in 10 patients (13.5 percent) and upper airway obstruction occurred in three (4.0 percent). No systemic complications occurred in patients submitted to surgical treatment.

Indications for Surgical Treatment

The majority of patients [62 (83.8 percent)] were treated under elective conditions (Figs. 2 and 3). Growth-related deformity was the indication in 44 patients. Lesions were located in the lips ($n = 14$), nose ($n = 11$), cheeks ($n = 6$), forehead ($n = 3$), eyelid ($n = 2$), ears ($n = 2$), and cervicofacial

Fig. 2. Patients submitted to elective direct total resection of proliferative infantile hemangioma. (*Above*) Direct closure and appearance after 1 year (*above*, *right*) in a patient nonresponsive to pharmacologic treatment at age 6 months (*above*, *left*). (*Below*) Use of purse-string sutures and appearance after 8 months (*below*, *right*) in a patient aged 8 months (*below*, *left*), nonresponsive to propranolol treatment.

region ($n = 3$), and three were hemifacial infantile hemangiomas. Treatment because of recurrent complications was performed in nine patients with infantile hemangiomas located in the forehead ($n = 2$), eyelids ($n = 2$), cheeks ($n = 2$), lips ($n = 2$), and cervicofacial region ($n = 1$). The remaining nine patients had involuted infantile hemangiomas, hemifacial in three cases or situated in the lips ($n = 3$), nose ($n = 1$), cheek ($n = 1$), and cervicofacial region ($n = 1$). Emergency removal was performed in 12 cases (16.2 percent). Obstruction of the visual axis was present in 10 patients (13.5 percent) (Figs. 4 and 5), and upper airway obstruction was the indication in two patients (2.7 percent) (Fig. 6).

Surgical Access and Type of Resection

Direct access was performed in 60 patients, through peritumoral incisions in 35 cases (47.3 percent) and transtumoral incisions in 25 cases

Fig. 3. (*Left*) Patient with involuting hemifacial infantile hemangioma after partial response to pharmacologic treatment. (*Center*) The first surgical treatment was performed at age 3 years by direct access with partial resection and primary closure. (*Right*) Final appearance after secondary procedure of tissue expansion.

Fig. 4. (*Left*) Patient with proliferative segmental orbital infantile hemangioma. (*Center*) Treatment was considered an emergency indication and was performed in conjunction with oral corticosteroid therapy by direct access with partial resection and primary closure at 6 months of age. (*Right*) Appearance after 6 years, showing involution of the nonresected portion of the hemangioma.

(33.8 percent). Indirect access through cervicotomy (neck infantile hemangiomas), open rhinoplasty approach (nasal infantile hemangiomas), or transpalpebral access (eyelid infantile hemangiomas) was performed in the remaining 14 cases. Total resection was performed in 49 cases (66.2 percent), subtotal resection was performed in nine cases (12.2 percent), and partial resection was performed in 16 cases (21.6 percent).

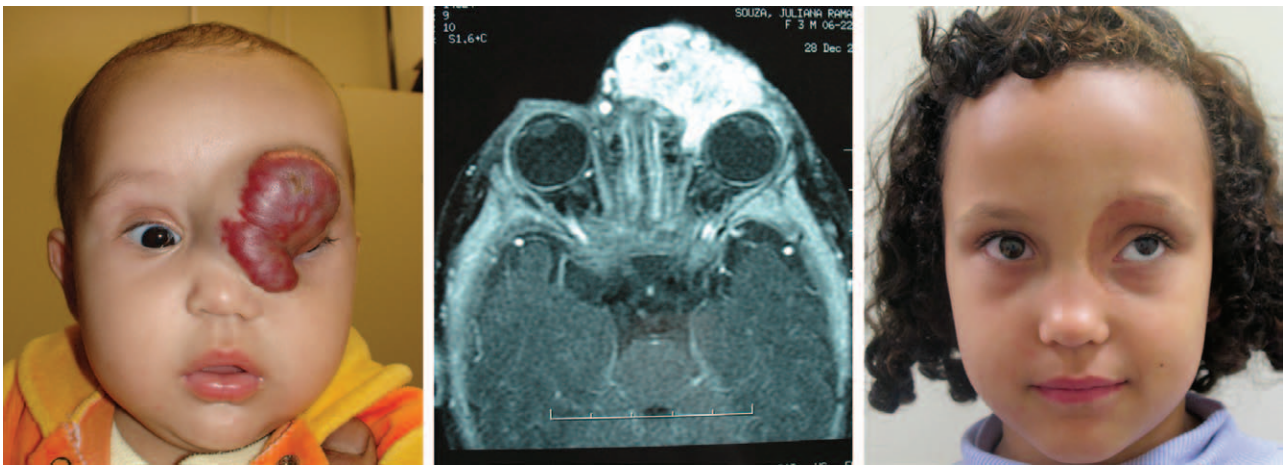

Fig. 5. (*Left*) Five-month-old patient with proliferative orbital infantile hemangioma. (*Center*) Magnetic resonance imaging scan showing periorbital involvement. (*Right*) Treatment was considered an emergency indication for surgery and was performed by direct access with total resection and primary closure. Appearance after 5 years.

Reconstruction Method

When partial and subtotal resections were performed, reconstruction was accomplished by primary closure (25 cases) (Figs. 2, 3, and 5). In total resections (Figs. 1 and 4), reconstruction was achieved by primary closure (14 cases), purse-string suture (eight cases), or advancement and rotation flaps (27 cases).

Patient Follow-Up

Mean follow-up was 33 months (median, 24 months; range, 6 to 60 months). Fifteen patients (20 percent) had postoperative follow-up longer than 5 years, and 13 patients (17.6 percent) had postoperative follow-up shorter than 12 months.

Postoperative Surgical Problems

Wound infection occurred in two cases (2.7 percent), tissue necrosis occurred in two cases (2.7 percent), and dehiscence occurred in one case (1.4 percent). Aesthetic complaints requiring additional treatment occurred in three cases (4.0 percent). In one case (1.3 percent), proliferation of the hemangioma was observed after partial resection, requiring additional pharmacologic treatment.

Surgical Patient Profile

Combined evaluation of demographic, clinical, and surgical data is summarized in Tables 3 and 4.

There was no significant association between sex and the presence of complications (ulceration, bleeding, or infection) or treatment indications. Patients who underwent emergency procedures were younger ($p = 0.0031$) and had a higher incidence of complications ($p = 0.012$). Also, emergency indications were more frequent in patients operated on during the proliferative phase ($p = 0.011$).

In patients submitted to partial resections, the incidence of complications was higher ($p = 0.001$) and emergency indication for surgery was more frequent ($p < 0.001$). Patients with segmental hemangiomas had more complications than those with localized hemangiomas ($p = 0.003$) (Tables 3 and 4).

Most of the operated facial hemangiomas were centrally located. Palpebral and nasal lesions were predominantly associated with emergency indications, and the number of additional surgical procedures was higher at these locations.

Patient Outcome

Concordance between evaluators was statistically confirmed. According to the kappa index, it was rated as satisfactory for facial contour (kappa = 0.3706, $p = 0.384$) and moderate for case difficulty (kappa = 0.4926, $p = 0.442$), volume reduction (kappa = 0.4122, $p = 0.432$), and need for reoperation (kappa = 0.5350, $p = 0.388$). Considering the average values of evaluations, there was a balance between easy, medium, and difficult cases. Changes in facial contour and volume reduction presented improvement in 99.5 percent. Regarding need for reoperation, it was considered not necessary in 40.8 percent of cases. The majority of suggested additional procedures (44.0 percent) were indicated for minor corrections (Table 5).

Fig. 6. (*Above*) Patient presenting a proliferative nasal hemangioma with airway obstruction. Surgical treatment was indicated at age 2 years because of absence of response to pharmacologic therapy. Treatment was performed in three sequential procedures, the first with subtotal resection and the two remaining for lining and minor correction. (*Below*) Late appearance after 5 years.

DISCUSSION

Continuous monitoring of patients until total resolution is crucial for assessing outcomes.[18–21] Newer concepts and knowledge gained in recent decades on biological behavior of infantile hemangiomas have directed therapeutic approach toward the pursuit of pharmacologic measures that may block proliferation or accelerate involution.[1,22–24]

However, there will still be a place for surgical treatment.[25,26] The most obvious examples are emergency indications and treatment of residual involuted infantile hemangiomas. There are advantages over pharmacologic treatment alone, especially in dramatic conditions, where any fast improvement will be considered beneficial.[14,25–32]

Most studies report overall treatment rates of approximately 20 percent, including surgical cases.[1,5,7,8,14,16,26] Otherwise, in the present study, the overall treatment rate was 58.6 percent (122 of 208 patients), with a high incidence of patients who were operated on [74 of 122 (60.6 percent)]. Patients referred to our clinic were usually potential candidates for active treatment and, more specifically, for the surgical approach, explaining in part this difference.

Emergency resection is often performed during proliferation and therefore in younger patients,

Table 3. Relationship of Demographic and Clinical Variables According to Complications (Ulceration, Bleeding, or Infection)

	Complications		
Variable	**No (%)**	**Yes (%)**	***p***
No.	59	15	
Age, mo			0.0327
Range	4–156	3–72	
Median	24	12	
Mean ± SD	33.5 ± 28.3	20.9 ± 19.9	
Indication			0.012
Emergency	6 (50)	6 (50)	
Elective	53 (85.4)	9 (14.6)	
Sex			0.999
Female	50 (79.4)	13 (20.6)	
Male	9 (81.8)	2 (18.2)	
Involvement			0.003
Localized	55 (85.9)	9 (14.1)	
Segmental	4 (40.0)	6 (60.0)	
Midline involvement			0.246
Yes	24 (72.7)	9 (27.3)	
No	35 (85.4)	6 (14.6)	
Restriction to anatomical unit			0.066
Yes	50 (84.8)	9 (15.2)	
No	9 (60.0)	6 (40.0)	
Type of resection			0.001
Partial and subtotal	14 (56.0)	11 (44.0)	
Total	45 (91.8)	4 (8.2)	

Table 4. Relationship of Demographic and Clinical Variables According to Treatment Indication

	Indication		
Category/Measure	**Emergency (%)**	**Elective (%)**	***p***
No.	12	62	
Age, mo			0.0031
Range	3–32	3–156	
Median	11	24	
Mean ± SD	14 ± 9.8	34.2 ± 28.2	
Extent of involvement			0.050
Localized	8 (12.5)	56 (87.5)	
Segmental	4 (40.0)	6 (60.0)	
Evolutional phase			0.011
Proliferative	11 (25.6)	32 (74.4)	
Involutional and involuted	1 (3.2)	30 (96.8)	
Midline involvement			0.352
Yes	7 (21.2)	26 (78.8)	
No	5 (12.2)	36 (87.8)	
Restriction to anatomical unit			0.059
Yes	7 (11.9)	52 (88.1)	
No	5 (33.3)	10 (66.7)	
Complications			0.012
None	6 (10.2)	53 (89.8)	
Bleeding, ulceration, or infection	6 (40.0)	9 (60.0)	
Type of resection			<0.001
Partial and subtotal	10 (40.0)	15 (60.0)	
Total	2 (4.1)	47 (95.9)	

as confirmed in this study. Essentially, in segmental infantile hemangiomas, partial resection is recommended, because the prime indication is functional. Confining surgery to the minimum necessary is an adequate and safe alternative for preventing sequelae arising from active surgical treatment.

Among elective cases, the decision must be considered under a protocol that includes all treatment alternatives. Although there has been a tendency toward pharmacologic treatment, it is important to emphasize that there are cases where the best treatment is still surgery. The adoption of an algorithm allows directing treatment in a more predictable fashion.

From all possible locations, surgery is a real option for cervicofacial hemangiomas.[1,21] The

Table 5. Treatment Outcomes Based on Experts' Survey

Question and Rating	Results (%)
Surgical difficulty	
Easy	35.0
Medium difficulty	33.0
High difficulty	32.0
Facial contour	
Great improvement	75.9
Slight improvement	23.6
Worsening	0.5
Volume reduction	
Great improvement	80.5
Slight improvement	19.0
Worsening	0.5
Need of reoperation	
Not necessary	40.8
Need for small additional procedures	44.0
Need for similar or larger additional procedures	15.2

indication is focused on cases with growth-related deformities and removal of the tumor potentially would allow adequate facial development. This condition is frequently seen in the nose, lips, and eyelids; even with complete involution, permanent deformities may develop. Surgical treatment, in the authors' opinion, prevents deformities caused by the interposition of infantile hemangiomas and is associated with satisfactory long-term results.

Supporting the surgical approach, there are studies suggesting that centrally located facial infantile hemangiomas may present lower regression rates.[8,18,28–33] Considering that central lesions are associated with increased psychosocial problems,[9,34] the predominance of surgical treatment in the eyelids, lips, and nose is justified.

A predominance of operations performed during the proliferative phase differed from more conventional treatment philosophies of delayed surgery.[11,16,32] Several overlapping aspects may explain and validate the early approach: consolidation of an algorithm for surgical indications, safety of anesthetic procedures, and a positive evaluation of outcomes.

The judicious selection of access, type of resection, and reconstruction method is fundamental for obtaining the best scar.[9,14,26] The concept of minimal possible scar is relevant, and the use of purse-string sutures, initially proposed by Mulliken et al., was applied, promoting a real reduction in the final scar dimensions.[35] When that is not appropriate, positioning of scar lines in strategic hidden locations is an alternative.

A clinical profile of candidates for surgery was defined: under elective circumstances, patients with proliferative localized eyelid, nasal, or lip hemangiomas with growth-related deformities are the best candidates for total resection under the direct (superficial or mixed infantile hemangiomas) or indirect (deep infantile hemangiomas) approach. For patients treated on an emergency basis, the best candidates are those nonresponders to pharmacologic therapy presenting complications, with segmental lesions, treated by partial resection under a direct approach (Fig. 1).

The objective survey performed by experts resulted in favorable outcomes in both simple and complex cases. Laypersons, parents, and health care professionals with or without expertise can perform outcomes evaluation. Laypersons generally tend to favorably overestimate results, particularly when clinical improvement is evident. It may downgrade a more critical evaluation. The surgeon who performs the procedure, by contrast, tends to be overly demanding in his or her results and may underestimate them.

When the objective of a study is to evaluate a technical result, specific knowledge is required, and experts not involved in treatment appear to be the best choice. The proportion of cases rated as "without need of reoperation" or "requiring small additional procedures" corroborates the impression that treatment goals have been achieved.

There are limitations in the study that should be considered. First, the retrospective design led to an evaluation of cases treated before the first publications on the effectiveness of beta blockers. In this condition, patients who underwent surgery had as alternative pharmacologic treatment with steroids. It could direct the option of surgery, considering side effects and lack of efficacy of using corticosteroids. Moreover, a comparative study of the various forms of treatment was not carried out; instead, the study focused on the analysis of surgical outcomes. Nowadays, with the possibility of pharmacologic treatments with fewer side effects, there is a need for new prospective comparative studies evaluating the outcome between the use of propranolol and surgery. However, there is still no response if the use of propranolol reduces the number of operations, decreases the morbidity of procedures, or only postpones the moment of definitive surgery. Considering the concern with growth-related deformities, the indication for surgery because it is grounded in clear and efficient criteria still seems a suitable alternative.

CONCLUSIONS

The results obtained in this study helped to establish the profile of patients treated by surgical resection and the specific surgical approach

for each situation. Satisfactory results could be achieved in both simple and complex cases when accurate indication criteria were followed.

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PATIENT CONSENT

Parents or guardians provided written consent for the use of patients' images.

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