

Persistent Nasal Infantile Hemangioma: A Surgical Treatment Algorithm

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Abstract: Treatment of infantile hemangioma is usually medical. The nose is one of the most important aesthetic and functional units of the face; therefore, surgical management is preferred in persistent lesions (fibro-fatty tissue component) that do not respond to medical treatment. Herein, the authors analyze the results of surgical nasal hemangioma treatment in their center, a literature review and propose an algorithm for surgical management. A retrospective analysis of 23 persistent nasal hemangioma operated between 1996 and 2014 at our institution was made. The authors recollected the following demographic and clinical data: hemangioma subtype, phase of evolution, affected aesthetic nasal subunit, previous treatment, surgery type, complications and follow-up period. The Strasser scale was chosen for assessment of postoperative photographic results. Surgical treatment was performed. Age average was 6.8 years old (range 2–19). Mixed hemangioma was the most common subtype (83%). Surgery was mainly performed in the involution phase (87%). The most affected aesthetic subunit was nasal tip (44%). All lesions received previous medical treatment. The most common surgical technique was open rhinoplasty approach. Follow-up was an average of 26.6 months. Aesthetic results according to the Strasser method included: 3 excellent scores, 15 good, 5 regular, and none poor. Surgery was performed on patients in order to correct nasal persistent hemangiomas sequel. The nose has serious psychosocial impact and mixed infantile hemangiomas appear to have a higher rate of persistent hemangioma requiring surgery, but further studies are needed. Aesthetic outcome with surgical procedures chosen were acceptable and no complications were reported.

Key Words: Fibro-fatty tissue, hemangioma, nasal hemangioma, nasal reconstruction, persistent infantile hemangioma, surgery

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Infantile hemangioma is the consequence of postnatal vasculogenesis and angiogenesis. Hypoxia appears to play an important role as a contributory factor. Infantile hemangiomas (IH) have variable clinical presentations: superficial (only superficial dermis), deep (deep dermis and hypodermis), or mixed. They can be localized or segmental involving a large skin area. Initial treatment after diagnosis of persistent IH remains still controversial. Medical treatment of IH is well defined as well as its natural history: it tends to have a relatively short proliferative phase (approximately until 9 months old), followed by a stable phase (from 12 to 18 months), and then an involution phase (from 18 months), which in many patients produces almost complete remission of the original lesion. The majority of IH are mild and do not require surgery. Therefore, medical treatment, which consists in observation or administration of propranolol in order to accelerate the involution phase, is usually preferred. Main indications for pharmacological treatment of IH are vital risk (heart failure, respiratory distress), functional risk (amblyopia, swallowing disorders), painful ulceration and disfigurement (face involvement of nose and lips). Propranolol has been quickly adopted as the first-line medical treatment for complicated IH due to its high efficacy and security profile.¹

According to natural history studies of untreated IH it was proved that about 30% of lesions involute fully by 3 years of age, 50% by 5 years, 70% by 7 years, and over 90% by 9 years.² Thus surgical management should be expectant until this phase, unless vital risk is present. Moreover, factors such as recurrence, location, depth, the possibility of complications, and psychosocial impact on the child and family may change the therapeutic approach leading surgical intervention as the treatment of choice if the medical treatment has failure.³

The purpose of any treatment is based on 3 main principles: minimize the possibilities of complications that increase morbidity and mortality, reduce the psychosocial conflicts that may afflict the patient and family, avoid excessively aggressive procedures potentially at a high risk of undesired secondary effects or scarring in IH of otherwise good prognosis with a more conservative treatment.⁴

The nose represents an especial anatomic feature. Due to its particular location, IH limited to the nasal area involve special therapeutic considerations. The nose is one of the most important aesthetic and functional units of the face; therefore, surgical management is the treatment of choice in persistent lesions (fibro-fatty component and skin damage) that do not respond to medical treatment. The purpose of the surgeon should be to remove the IH and to restore normal facial features. Each of the facial zones has its own special features and challenges.⁵ We must remember that IH represent a proliferation of endothelial cells,¹ and therefore do not invade adjacent anatomic structures, but could displace them.

The aim of our study is to focus on the surgical management of persistent nasal IH through 23 clinical cases presentation, including an analysis of our results, a literature review, and to propose a surgical algorithm. Herein, we also present iconic clinical cases from our series with their particular surgical approaches.

METHODS

A retrospective analysis of 23 patients with persistent nasal IH after medical treatment was derived to plastic surgery unit from dermatology service was performed. They were incorporated between 1996 and 2014. The following data was analyzed: age, gender, IH subtype (superficial, mixed, deep), time of evolution at the time of surgery, affected aesthetic subunit, previous treatment (medical and surgical) and type of surgical procedure, complications, and follow-up period. Objective assessment for aesthetic results of nasal reconstruction was performed by 4 external and independent specialist individuals (plastic surgeons) with evaluation of a series of postoperative photographic results applying the Strasser score method.^{6,7}

RESULTS

A total of 23 patients underwent surgical intervention. Nine were females (39, 13%) and 14 were males (60, 87%). Average age was 6.8 years (range 2–19). The most frequent IH was mixed subtype (83%) followed by the deep subtype (13%) and finally superficial subtype (4%).

Of the total of patients, 87% underwent surgical intervention during the involution phase whereas the remaining 13% corresponded to a sequel phase.

The aesthetic nasal subunits affected with persistent IH in our surgical candidates case series were: 44% nasal tip, 17% sidewall subunit, 9% dorsal subunit, and 30% corresponded to a combination of several affected subunits.

Dermatological medical treatment was administered to all patients (corticosteroid until 2008, and propranolol thereafter and/or observation) prior to surgical excision. In 74% of the patients drug was indicated whereas 26% did not receive any previous pharmacological treatment. Two patients had received previous surgical treatment at different medical centers and therefore underwent surgical intervention in order to correct the nasal sequel. Surgery in our patients was indicated in the following settings: failure of local and/or systemic pharmacological treatment: evidenced in 17 patients (74%). To treat nasal sequel from previous surgery: in 2 patients (9%) and to correct incomplete involution with prior observational treatment, in 4 patients (17% patients).

The most frequently surgical techniques used were open rhinoplasty approach—9 patients, and forehead flaps—5 patients (for further results see Table 1).

There were no reported surgical complications in this case series (bleeding, obstruction, or infection).

An example of open rhinoplasty without damaged skin is shown as patient 1 (Fig. 1). An example of open rhinoplasty of a nasal hemangioma of the nasal tip with fibro-fatty tissue resection (skin overabundance) is shown as patient 2 (Fig. 2). An example of persistent nasal IH with damaged skin and its surgical resolution (in 3 times) with forehead flap is shown as patient 3 (Fig. 3). Other

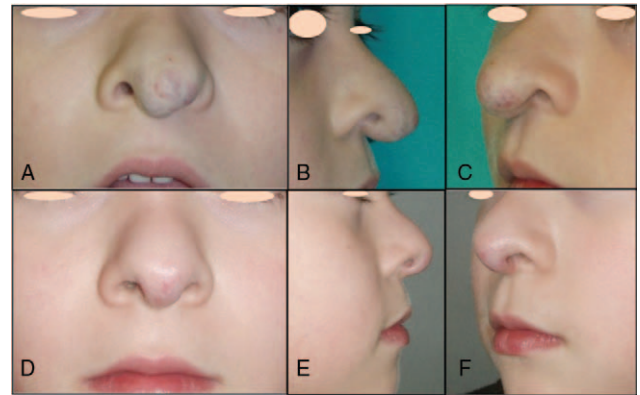


FIGURE 1. Patient 1. Before (A, B, C) and after (D, E, F) surgery image of patient with persistent IH of the nasal tip with good skin quality in whom open rhinoplasty was performed at age 6. As can be observed there are no growth alterations of the cartilage. Final Strasser score 2 (good).

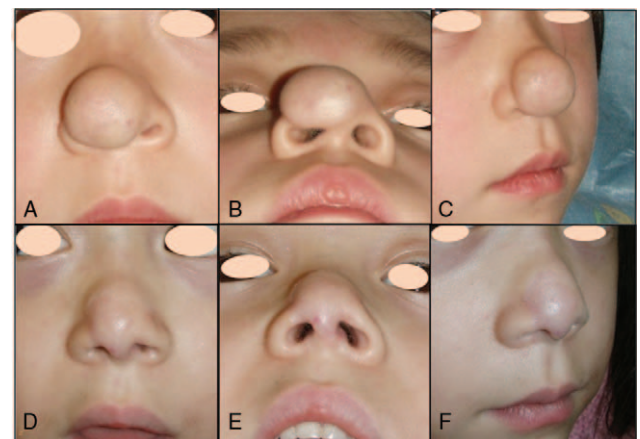


FIGURE 2. Patient 2. Before (A, B, C) and after (D, E, F) surgery image of the IH managed with direct excision plus open rhinoplasty. Final Strasser score 1 (good). Medical treatment was the choice until age 6 without response. Skin indeminity may be seen in spite of fibro fatty degeneration over the years. Direct excision was performed following aesthetic subunit delimitations. Final Strasser score 2 (good).

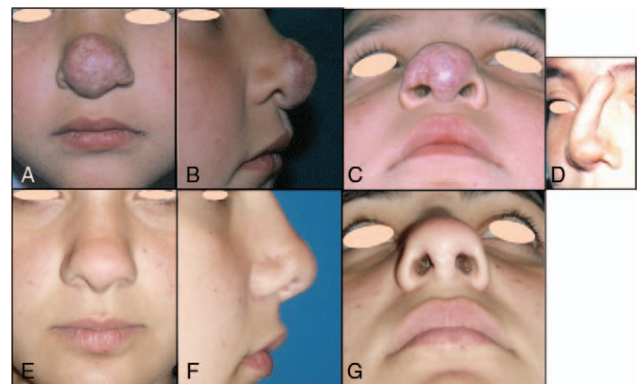


FIGURE 3. Patient 3. Before (A, B, C) and after (E, F, G) of persistent Nasal IH with defective skin in nasal tip subunit repaired with a forehead flap. The reconstruction involved 3 sequential surgical timings in a 9-week period. Above there are the pictures before surgery, inferior after surgery. At the right (D) the image the forehead flap in an intermediate surgical time can be seen. Final Strasser score 1 (good).

TABLE 1. Surgical Techniques Applied and All Cases Summary

Nasal Surgical Technique	No of Patients
Open rhinoplasty	9
Forehead flaps	5
Open resection	4
Open rhinoplasty + direct resection	2
Cheek flap	1
Lipoaspiration	1
Close rhinoplasty	1
Total	23

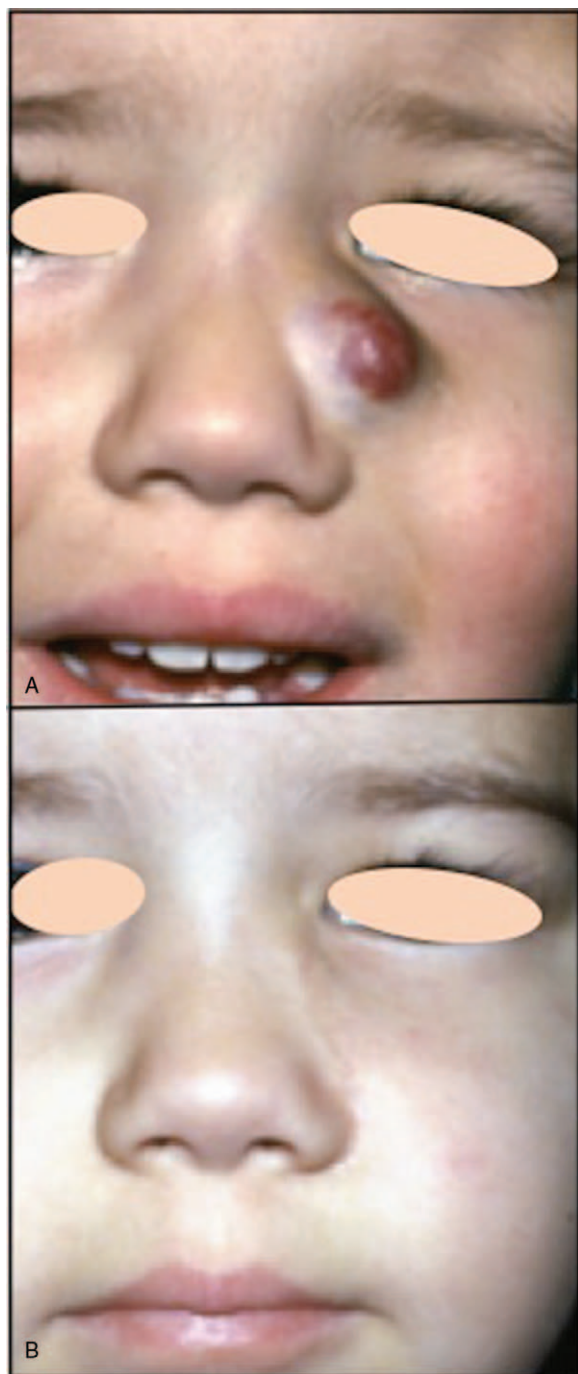


FIGURE 4. Patient 4. Before (A) and after (B) surgery image of persistent nasal IH with defective skin in lateral sidewall subunit repaired with a cheek flap, with excellent results. Final Strasser score 0 (excellent).

example of persistent nasal IH of the nasal sidewall and its surgical resolution with cheek flap is shown as patient 4 (Fig. 4).

A follow-up was done for an average of 26.6 months (range 3–168). Aesthetic results according to the Strasser method were: 3 excellent scores (13%), 15 good score (65%), 5 fair (22%), and none poor results (0%). Based on these results a surgical algorithm is proposed (Fig. 5).

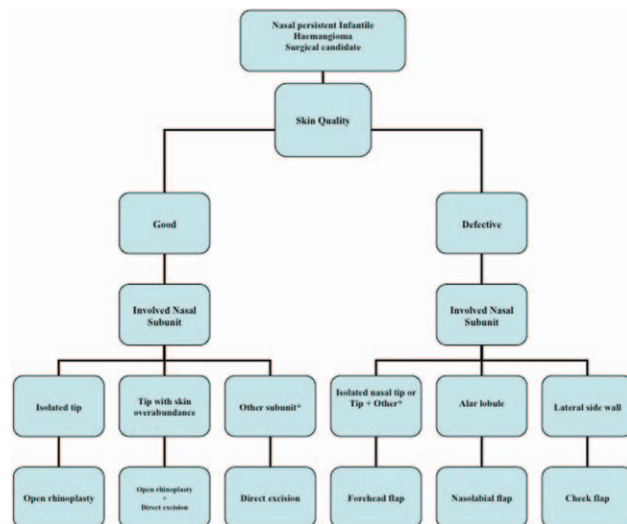


FIGURE 5. Surgical treatment algorithm proposed for persistent infantile nasal hemangioma.

DISCUSSION

IH are the most common benign tumors of infancy characterized by a rapidly proliferative initial phase followed by a slow involution over the first years of life, and a last involute phase.⁸ This unique growth pattern is what differentiates IH from other vascular anomalies.^{9,10} After the involution phase which is complete approximately after 6 to 9 years, cutaneous sequel may be seen as scarring with irreversible skin damage or changes in skin color and texture. In deeper layers of the skin the normal vascular pattern is substituted by the presence of fibro-fatty tissue mixed with collagen and reticular fibers.^{11,12}

Over the past years, the potential of permanent disfigurement and psychosocial impact of IH sequel has been increasingly addressed.¹³ The risk of disfigurement depends on its location, morphological subtype, size, shape, and growth phase of the IH. Some studies reveal that over 50% of affected patients will develop persistent telangiectasia, scars, pigmentation disorders, and a fibro-fatty remnant in the area of the original lesion requiring dermatologic and surgical care to regain an adequate aesthetic appearance.^{14–16}

IH treatment is usually medical and surgery is reserved for a minority refractory or life-threatening cases. B-blockers are now considered first-line treatment for high-risk lesions. It is very important to point that medical treatment of IH has evolved during history. Propranolol has largely replaced oral corticosteroids for those IH requiring systemic therapy, due to its high effectiveness and infimal adverse effects, providing a safe alternative, especially during the high proliferative phase.^{11,17–19}

According to our experience the oral corticosteroids were used approximately until 2008, being replaced by propranolol. Also, in our case series there was conservative treatment in 26% mainly due to delayed first evaluation at involution phase (late diagnosis).

Surgical indication in IH depends on the each case characteristic. Excision is indicated in patients with propranolol nonresponding obstructive IH, especially in the head and neck peri-orificial regions, or those causing visual, hearing, or breathing impairment.²⁰

Visible deforming tumors caused significant psychological distress. Some facial IH involute incompletely leaves fibro-fatty redundant tissue, which alters the aesthetic lines of the face. In our experience, in these cases liposuction can be used as an alternative in order to restore normal aesthetic appearance.²¹

In the presence of a lesion with recurrent bleeding or ulceration surgery may be considered before the proliferative phase has ended, which was not the case in our series.

In our experience, a multidisciplinary team consisting in dermatologists, radiologists, vascular surgeons, and plastic surgeons who analyzed all the different therapeutically options, better decides the type of therapy. Usually, surgical treatment, in these series, was decided once all chance of medical therapy had failed. It was also considered an option for the treatment of sequel.

Nasal IH is considered a “high risk hemangioma,” mainly due to its location, which can cause functional compromise secondary to respiratory obstruction and risk of permanent disfigurement,¹³ for which early referral for surgical excision is a reasonable option.

The nose is an extremely important aesthetic structure and because of its permanent exposure the possibility of concealing any lesion in this area is virtually impossible. This fact may cause severe psychological and social impact on any patient and family; therefore, watchful waiting for spontaneous involution of nasal hemangiomas may not be the best option.²² Maintaining the child’s self-esteem by offering an adequate treatment at the right age (which in the majority of patients was nearly 5 years of age before first grade of school) was the best solution in many patients.^{23,24} Some authors suggest even an earlier age for surgery in order to avoid possible cartilage growth impairment.²⁵ This issue is controversial, since at least in our series, there was no growth alteration observed in the cartilage at the moment of surgery which was performed at an average age of 6 years. Nonetheless, separation of different structural elements of the nasal cartilages was evidenced due to compression by tumor growth requiring suturing procedures in order to obtain adequate repositioning of the alar complex. This fact concurs with the experience reported by Waner who reports 44 patients that underwent surgery for nasal hemangioma, between 9 months and 12 years of age and only 1 cartilage destruction in a patient with an ulcerated hemangioma and in most patients described separation only with indemnity of cartilage growth.²⁴ Hochman and Mascarenno²⁶ supports the same observation by sustaining that cartilage destruction is unusual except for massive lesions or extensive ulceration.

Nasal reconstruction in young children is extremely challenging. Therefore, we consider preferable to perform surgery later in childhood (4 or 5 years old) if clinical conditions allow it. Around this age anatomic structures are better defined and the original lesion may have reduced its original size making reconstruction technically easier with a consequently better outcome.^{10,27}

Ultimately, a key concept is preservation of skin indemnity, and because of this reason topical or systemic corticosteroids and cryotherapy have been left behind due to their high side effects and lack of effectiveness, making first therapy election in medical treatment oral propranolol due to its significantly high efficacy, efficiency, and security. These concepts correlate to the final skin quality prior to surgery.

Grounded on our experience and an extensive revision of the literature, we propose a surgical management algorithm for persistent nasal IH patients with surgical indication based on the skin quality and affected—and largely known—nasal subunits.²³ This proposal was applied after considering the medical algorithm suggested by Frieden et al.¹³

Once the patient has been analyzed by the multidisciplinary team and referred for surgical treatment the following step is to determine the quality of skin.

Nasal tip involvement alone is indication for the open rhinoplasty approach as suggested by McCarthy et al.²⁷ Concordantly, in our case series if skin was considered appropriate and the nasal tip alone was affected an open rhinoplasty approach was made (Fig. 1).

Open rhinoplasty associated with direct skin resection is needed when an overabundance of relying nasal skin is detected (Fig. 2). A key feature is to consider that the compensating skin excisions must be exactly localized on the nasal subunits limits. Thus, a combination of both techniques respecting local aesthetic subunits is indicated.²² Therefore if the persistent IH affects other subunits direct excision is preferred since it allows skin compensation along subunit margins.²⁸

When other subunits are affected a direct excision can be made. In our algorithm if skin is defective in nasal tip subunit or nasal tip subunit plus other subunits a forehead flap is indicated (Fig. 3). In this series of patients 5 forehead flaps were performed with no complications and good aesthetic outcome, according to the previous experience.²³

In other patients with skin damage and IH in the alar lobe subunit a local flap—such as the nasolabial—is indicated. Otherwise, when the lateral sidewall is compromised a cheek flap is recommended in order to provide an optimal reconstruction (Fig. 4).

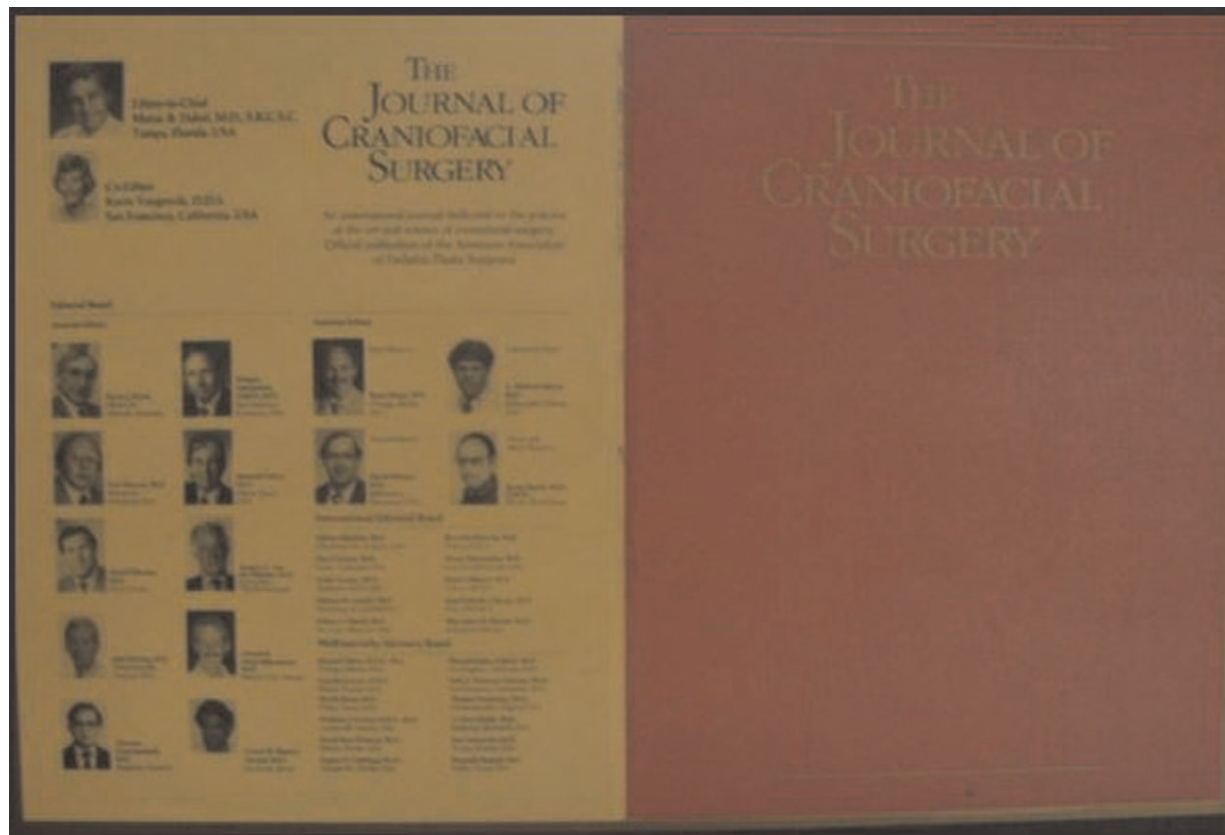
CONCLUSION

According to our aesthetic results, the perception of preserved child self-esteem, and the low rate of complications, we propose that the therapeutic options summarized in the algorithm are adequate alternatives and hopefully can be a base step in the treatment of patients with persistent nasal IH in other centers. This algorithm for treatment of nasal persistent IH was developed after a systematic analysis of demographic and clinical data complemented with the personal experience. The results obtained in this series and recommendations of different authors after an extensive revision of the literature could be beneficial for future interventions, although further studies must be made.

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