Surgical management of large scalp infantile hemangioma in 30-month-old infant

Cosimo Bleve,[1] Maria Luisa Conighi,[1] Enrico Valerio,[2] Mario Cutrone,[3] Giuseppe Iannucci,[4] Alessandro Segna,[5] Salvatore Fabio Chiarenza[1]

[1]Department of Pediatric Surgery and Pediatric Minimally Invasive Surgery and New Technologies, San Bortolo Hospital, Vicenza; [2]Neonatal Intensive Care Unit, Department of Woman and Child's Health, University of Padova; [3]Women and Child's Health Department, Pediatric and Neonatal Intensive Care Unit, San Bortolo Hospital, Vicenza; [4]Department of Neuroradiology, San Bortolo Hospital, Vicenza; [5]Department of Neuroscience and Neurosurgery, San Bortolo Hospital, Vicenza, Italy

Abstract

Infantile Hemangiomas (IH) are the most common benign tumor of infancy, occurring in over 10% of newborns. The head and neck is the most frequently affected area (60%), and the scalp is a typical site for such large lesions. Scalp-IHs are usually focal lesions that can be both disfiguring and may lead to complications such as ulceration and bleeding. We describe a case of a 30-months old female who presented a large scalp-IH at birth that rapidly grew in the first year of life. Topical and systemic treatments (with timolol ointment and oral propranolol, respectively) were not effective in reducing dimensions of the hemangioma. After vascular imaging study, the patient underwent surgical resection of the IH and primary closure with excellent cosmetic outcome. When medical therapy is ineffective or cosmetic and functional integrity is threatened, early surgery allows to completely removing large scalp-IHs, with good cosmetic results.

Correspondence: Cosimo Bleve, Department of Pediatric Surgery and Pediatric Minimally Invasive Surgery and New Technologies, San Bortolo Hospital, Vicenza, Italy,
Tel.: +39.0444.752642; Fax: +39.0444.752643
E-mail: cosimobleve1@gmail.com

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Introduction

Infantile Hemangiomas (IHs) occur most often in the craniofacial region (60%), followed by the trunk (25%) and extremities (15%). The more common are focal hemangiomas (85%) which usually grow as solitary or, occasionally, multifocal lesions. Segmental hemangiomas are less common (15%) and their growth follows a segmental dermatomes. They can be more aggressive and locally destructive. Their proliferation phase extends for longer periods (up to 24 months) and frequently they can ulcerate. Between focal and segmental hemangiomas exists a large variability in proliferation. Focal hemangiomas typically reach 80% of their full size by 3.2 months whereas segmental hemangiomas may proliferate for up to 24 months. All hemangiomas begin to involute, involuting phase, after a period of quiescence. The result of involution may vary from almost normal tissue to a fibrofatty mass virtually of the same size of the original IH.[1] Scalp hemangiomas could remain small, but if large are disfiguring and commonly destroy hair follicles leaving when they involute, an area of alopecia. They could often be associated with serious sequelae such as ulceration, bleeding, and cardiac failure. After regression, children may be left with a residual deformity, such as anetoderma (focal round to oval areas of flaccid skin with destroyed anatomical structures with a surrounding border of normal skin), fibrofatty residuum, redundant skin, scarring, and/or telangiectasias.[2] There are various options of treatment such as systemic propranolol that are suitable for most cases, pulsed dye laser, oral steroids. Surgery must be considered as a valid option and performed if feasible, when significant functional impairment, ulceration, bleeding, are encountered or cosmetic deformity may represent possible consequences.[3]

Case Report

We evaluated an otherwise healthy 30-month-old female for an IH of 3.5x2.5cm in size (figure 1A) located on the right frontal region of the scalp near the midline. Topical and systemic treatment (with timolol ointment and oral propranolol, respectively) were not effective in reducing dimensions of the hemangioma. Systemic propranolol administration was started late, around 6 months of life, due to delayed specialist evaluation and when the hemangioma presented a considerable size. Therapy was continued at a dosage of 2mg/kg until one year of age. Adherence to therapy was discontinuous during these months due to girl's current pathologies which required a temporary suspension and leading to minimal variation in size. After 12 months of age topical treatment was adopted which also resulted not effective. We decided for surgical resection. Before surgery an accurate imaging study with combined MRI and CT was performed demonstrating a pedunculated and vascularized lesion receiving arterial supply from distal branches of the right middle meningeal artery and from both superficial temporal arteries bilaterally with early drainage in the Superior Sagittal Sinus (SSS). No other vascular abnormalities or intracranial communications were described (Figure 1B). Preoperative blood examinations were in the normal range. The electrocardiography and the echocardiographs were normal. Surgical resection of Hemangioma was performed under general anesthesia (Figure 2A). Surgical incision was marked along the borders of the tumor. Careful and meticulous ligation of the major vessel (vein) and bipolar coagulation of the small vessels were essential for bleeding control as well as prevention of post-surgical hematoma. The hemangioma was completely removed resulting in a 4x2cm soft tissue scalp defect, which was treated with primary closure with interrupted not-absorbable sutures (Figure 2B). There was no ischemia or necrosis of the wound edges. At 24-month follow up, wound healing was complete. Histology described the specimen as capillary hemangioma, which resulted positive for

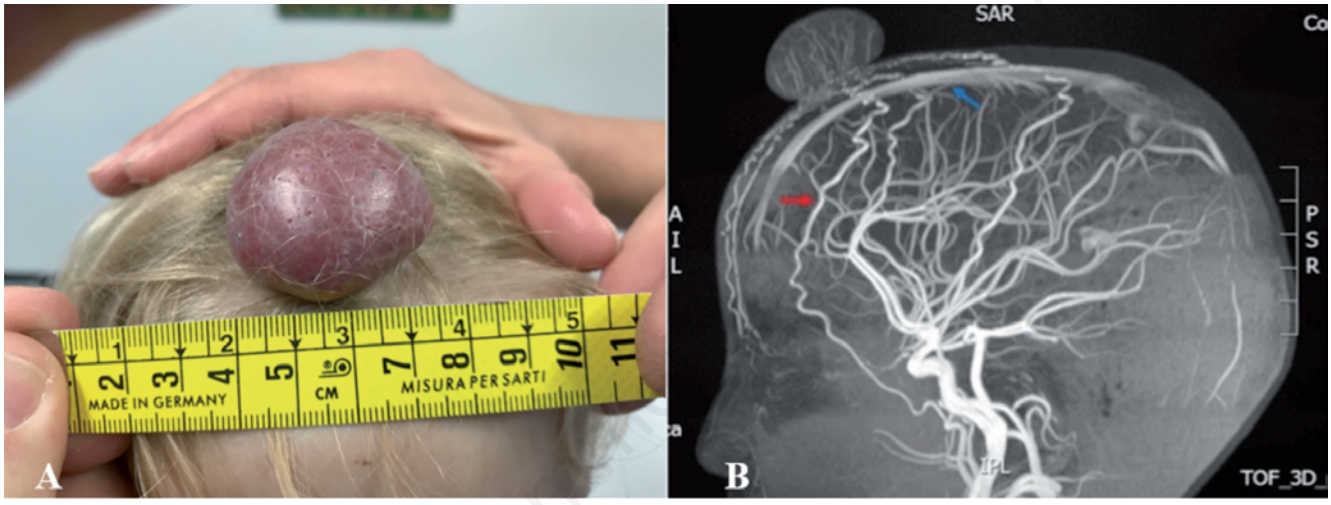

Figure 1. A) Congenital hemangioma (3.5 x 2.5 cm) located on the right frontal region of the scalp near the midline, as appeared at first evaluation. B) MRI sagittal view showing mass vascular connections: arterial supply (red arrow) was from distal branches of right middle meningeal artery and from superficial temporal arteries bilaterally with early drainage in the superior sagittal sinus (blue arrow).

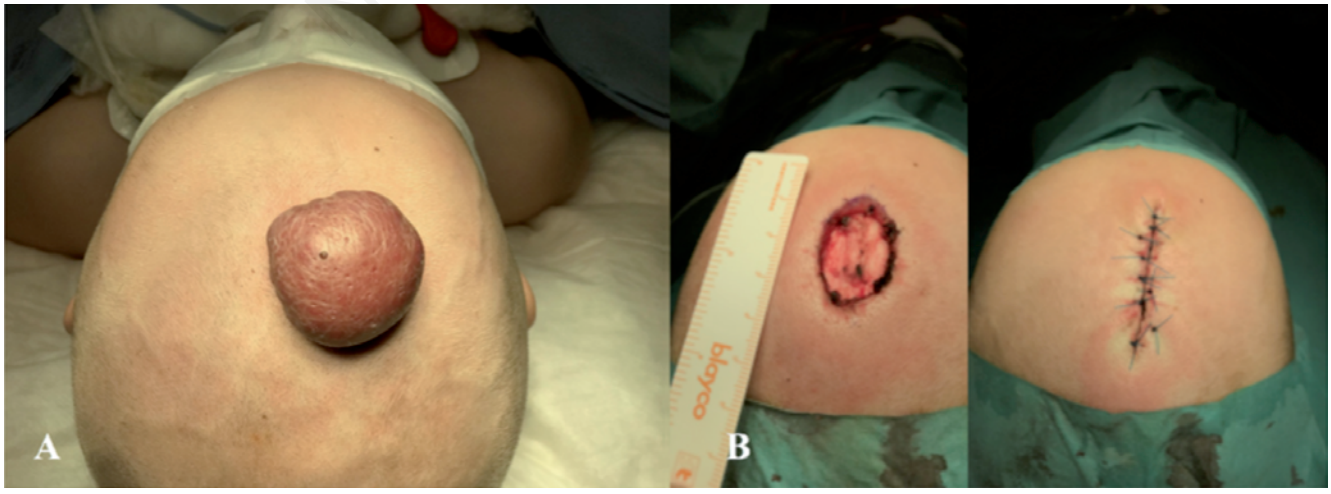

Figure 2. A) Mass view before surgical excision. B) Post-excisional soft tissue scalp defect (left), treated with primary closure with not absorbable sutures (right).

glucose transporter 1 protein (GLUT1) confirming the diagnosis of an IH. The entire surgical procedure lasted for about 2 hours, achieving immediate primary wound closure with estimated blood loss being <15cc and the patient was hemodynamically stable throughout the entire procedure.

Discussion

HIs are the most common benign neoplasms of vascular origin in paediatric age, made by proliferating endothelial-like cells and affect up to 4% to 5% of Caucasians. They are usually first noticed within 1 to 4 weeks after birth and immediately undergo a proliferative phase of variable duration and severity. Some IH grow very slowly, while others grow quickly and at an unpredictable rate. IHs often proliferate rapidly during the first few weeks of life and approximately 80% of the superficial hemangioma growth is achieved by 3 months of age.[4] In focal infantile hemangioma, this phase usually lasts 3 to 9 months. [1] Hemangiomas then begin to involute or regress. This phase may take 6 to 9 years.[5] Unlike other neoplasms, IHs tend to regress over time.[6] The vast majority of these lesions are not worrisome, but up to 12% of IHs need referral to specialist due to their complexity and/or place (e.g., tongue IH causing respiratory obstruction, or eyelid IH causing impaired eye opening).[7] In life or function-threatening IH, systemic therapies are needed to minimize massive bleeding, hypovolemic shock, wide ulceration, or serious infections. The mainstay of pharmacological treatment is based on beta-blockers, which can be administered both topically (timolol ointments) and systemically, oral, (propranolol).[8] Adverse effects of beta-blockers (hypoglycemia, hypotension, bradycardia, and bronchospasm) are well-known and rather infrequent; nevertheless, patients should be monitored for these issues during the first 48-72 hour after starting treatment.[9] IH of the scalp may be worth a try with pharmacological management when they are small; however, when they are very large, they may be of concern for possible complications such as ulceration, bleeding, or high-output cardiac failure. More, they may disrupt the anterior hairline or, after involution, result in large areas of alopecia. Moreover midline scalp as lumbar vascular malformations, are associated with underlying central nervous system malformations. MRI is mandatory to help uncover some of these congenital abnormalities. A connection with SSS needs to be evaluated to plan appropriate surgery. Infantile scalp IHs are more challenging to manage and require careful surgical planning than those in other regions because of their location and the potential risk of functional morbidity and aesthetic outcomes. The association of heart failure with hemangioma is common in visceral and multiple hemangiomas, but rare in the case of scalp congenital IHs. The size of hemangioma represents the main factor associated with development of heart failure because this complication has not been reported in case of tumours <7cm in diameter.[10] Furthermore, this possible complication requires further analysis.[11,12] By a revision of Literature few studies have reported sporadic cases of scalp hemangiomas that showed symptoms of cardiac failure.[12] In patients with mild symptoms of scalp IH who are controlled by medical therapy, waiting is beneficial to allow body mass to increase and minimize the interventional risks. In those cases where medical therapy is ineffective or cosmetic and functional integrity is threatened, meticulous surgical resection and primary wound closure is feasible and can be carried out safely with good cosmetic outcome.[2] Trans-arterial embolization of feeding arteries is reported in IH-management as a safe and effective associated to surgery for excision. The main aim of preoperative embolization is to obliterate the arterial feeders, decreasing the blood supply, and minimizing arteriovenous shunting at the lesion. However, embolization alone is not a definitive treatment as collateralization and recanalization can occur. Additionally, atrophic tissue and alopecia may persist resulting in poor cosmetic outcomes. Earlier surgical excision, in these circumstances, is resolutive to prevent potentially life-threatening or unaesthetic consequences. The reason for an early surgical approach is the great elasticity because of a relative "tissue excess" and pliability of the newborn scalp because of the thin galeal layer. Closing a scalp defect by primary closure or with rotation/transposition flaps is easier in newborns, without the need for tissue expansion. As the infant ages, this considerable redundancy of scalp tissue dissipates, which may limit the possibility of primary closure.

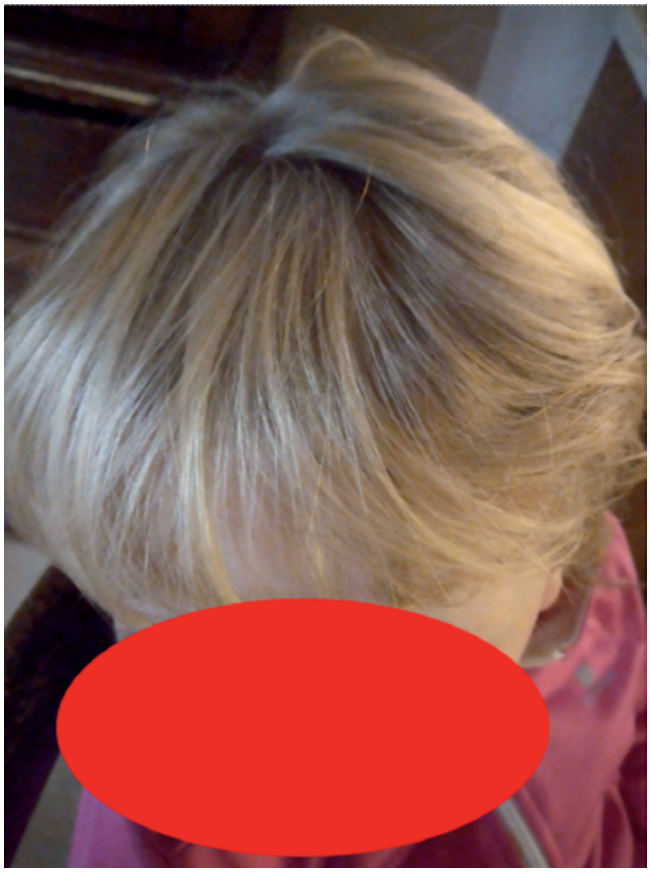

Figure 3. One year follow-up after surgery, the acceptable aesthetic result.

Conclusions

Hemangiomas usually will involute and be benign in evolution. Some scalp lesions can be large, deforming, ulcerative, and predisposed to hairless zones. Although there is no consensus on the optimal age for surgical resection of large scalp hemangioma, an early excision (under 4 months of age) is recommended.[2] Early intervention allow to correct all negative sequelae with total removal. In our case, despite the age of the patient (30 months-old), was possible to carry out a complete excision of IH with primary closure and excellent aesthetic results.

References

1. Chang LC, Haggstrom AN, Drolet BA, et al. Growth characteristics of infantile hemangiomas: implications for management. Pediatrics 2008;122:360–367
2. Spector JA, Blei F, Zide BM. Early surgical intervention for proliferating hemangiomas of the scalp: indications and outcomes. Plast Reconstr Surg 2008;122:457–62.
3. Zhu Z, Yang X, Zhao Y, et al. Early surgical management of large scalp infantile hemangioma using the TopClosure® tension-relief system. Medicine 2015;94:e2128.
4. Maguiness SM, Frieden IJ. Management of difficult infantile haemangiomas. Arch Dis Child 2012;97:266–271.
5. Cho YK, Ryu DW, Chung HY, et al. Surgical management of scalp infantile hemangiomas. J Craniofac Surg 2015;26:1169–72
6. Drolet BA, Frommelt PC, Chamlin SL, et al. Initiation and use of propranolol for infantile hemangioma: report of a consensus conference. Pediatrics 2013;131:128-40.
7. Haggstrom AN, Drolet BA, Baselga E, et al. Prospective study of infantile hemangiomas: clinical characteristics predicting complications and treatment. Pediatrics 2006;118:882–7.
8. Marey HM, Elmazar HF, Mandour SS, Khairy HA. Combined oral and topical beta blockers for the treatment of early proliferative superficial periocular infantile capillary hemangioma. J Pediatr Ophthalmol Strabismus 2018;55:37-42.
9. Léauté-Labrèze C, Hoeger P, Mazereeuw-Hautier J, et al. A randomized, controlled trial of oral propranolol in infantile hemangioma. N Engl J Med 2015;372:735-46.
10. Santecchia L, Valassina MFB, Maggiulli F, et al. Early surgical excision of giant congenital hemangiomas of the scalp in newborns: Clinical indications and reconstructive aspects. J Cutan Med Surg 2013;17:106–13.
11. Hayashi T, Ishibashi A, Hashimoto T, et al. Huge congenital angioma of the scalp. A case report. Kurume Med J 1981;28:91–4.
12. Alluhaybi AA, Abdulqader SB, Altuhayni K, et al. Preoperative trans-arterial embolization of a giant scalp congenital hemangioma associated with cardiac failure in a premature newborn. J Int Med Res 2020;12:1-7.