

Infantile Hemangioma in the Gluteal Region

¹Mary Sthelly Da Mata Costa Rodrigues, ¹Maysa Alves Rodrigues Brandão Rangel

1 Anhembi Morumbi University, Av. Dep. Benedito Matarazzo, 6070 - Jardim Aquarius, São José dos Campos - SP, 12230-002

Corresponding Author: Maysa A. R. B. Rangel, Profa. Dra.

Anhembi Morumbi University, School of Medicine- Av. Dep. Benedito Matarazzo, 6070 - Jardim Aquarius, São José dos Campos - SP, 12230-002

Abstract

This case report describes the management of an infantile hemangioma (IH) in a 5-year-old child, diagnosed prenatally as an amorphous mass located in the right gluteal region. Over the child's first five years, the vascular lesion progressively infiltrated adjacent soft tissues, compromising the functionality of the affected area. Despite initial treatment with propranolol, the patient showed a limited response, with lesion progression and associated symptoms. Due to the therapeutic ineffectiveness of the medication, surgical resection was chosen as the primary approach. The macrocystic lesion, with multiple loculations measuring approximately 10x4x7 cm, was removed without adverse complications. This case underscores the importance of early diagnosis and appropriate management of infantile hemangiomas, highlighting the surgical approach over the first-line treatment with propranolol in cases where initial treatment does not yield satisfactory results, especially in situations posing functional risks. The patient's favorable outcome adds to the existing literature on the therapeutic outcomes of this approach.

Keywords: infantile hemangioma, surgery, propranolol, treatment, case report.

Introduction

Infantile hemangiomas (IH) are elevated, red or purplish vascular hyperplastic lesions and are among the most common benign tumors in childhood, affecting 5% to 10% of all infants¹. These lesions can be classified based on general appearance (superficial, deep, or cavernous) or by descriptive terms (e.g., strawberry hemangioma). However, as all these lesions share the same pathophysiology and natural history, the term infantile hemangioma is preferred¹. IHs are generally benign vascular tumors and do not pose serious risks for most children. Nonetheless, around 10% to 15% of IH cases lead to significant issues, including skin expansion, ulceration, and necrosis, particularly in facial areas. The tumor follows a life cycle of rapid proliferation, growing between 4 and 18 months of age, followed by a slow involution phase over 3 to 9 years, leading to a reduction in size and vascularity². Most IHs have a favorable course, ultimately regressing without complications.

Standard treatment for problematic hemangiomas includes propranolol, which has demonstrated efficacy in the majority of cases. However, in situations where drug therapy proves ineffective or when there is a high risk of functional impairment, surgical intervention may be warranted³. This case report describes a child with an extensive gluteal region hemangioma, in which surgical management was selected as the primary treatment strategy.

The justification for this report lies in the need to discuss surgical management as a viable alternative in cases where pharmacological treatment fails or is unsuitable. Current literature remains limited regarding specific indications for surgery in infantile hemangiomas, especially in high-risk lesions. This study aims to underscore the importance of individualized evaluation and to highlight surgery as a safe and effective method in certain IH contexts.

Materials And Methods

Case Description: This study described a clinical case of an infantile hemangioma located in the gluteal region. The patient was selected from the medical records of a healthcare institution, with informed consent obtained from the parents or legal guardians.

Data Collection: Data were collected retrospectively from the patient's medical records, including demographic information, medical history, clinical characteristics of the hemangioma, diagnostic methods used (such as imaging exams), and treatments administered.

Data Analysis: The collected data were analyzed qualitatively, focusing on a detailed description of the case, including clinical progression and treatment response. The analysis was comparative with the existing literature on infantile hemangiomas, highlighting specific aspects of the studied case.

Data Collection Instruments: Medical records, imaging exams, and consultation records were used. All data were handled confidentially, in accordance with ethical and privacy standards.

Literature Review: A literature review was initially conducted to contextualize the case within the current knowledge of infantile hemangiomas, using databases such as PubMed, Scielo, and Google Scholar.

Ethical Considerations: The project was submitted to the Research Ethics Committee for approval before data collection began. Informed Consent (IC) was obtained from the patient's parents or guardians, and Assent was obtained from the child when applicable, following the terms defined in the Helsinki Declaration, with the ethics committee approval protocol number: CAAE: 83637324.3.0000.5492.

Case Report

M.G.C, a 5-year-old male, mixed-race, from Bolivia, with a regular prenatal history, vaginal delivery without complications at 40 weeks, negative neonatal screening tests, good vitality at birth, and no syndromic stigmata. Prenatal diagnosis indicated an amorphous mass in the right gluteal region.

At birth (2019), the patient underwent a biopsy of the gluteal mass (approximately 3 cm), which showed no content or risk signs in imaging exams, so an outpatient follow-up was chosen. However, the patient lost follow-up during the pandemic. Over his five years of life, the lesion grew in size but remained relatively stable in the months leading up to surgery. On physical examination, the lesion was non-tender to palpation, with no skin changes or systemic symptoms/signs. The caregiver denied any changes in shape, size, or consistency of the gluteal mass. The patient had no comorbidities, allergies, or continuous use medications.

In April 2024, the patient underwent an ultrasound of the right gluteal soft tissue, revealing the following features: a large, complex mass with thin walls, macrocystic appearance, multiple loculations, hypo/anechoic, infiltrating the adjacent soft tissue, with distinct margins, and no color flow on Doppler study. The mass measured approximately 10x4x7 cm, with a minimum distance from the skin of 2 mm. (Figure 1 and 2)



Based on the clinical diagnosis and ultrasound, surgery to remove the tumor in the gluteal region was scheduled electively for June 2024. During surgery, the patient was placed in the prone position, the aseptic technique was performed, and subsequent an arciform incision was made in the right gluteal. During surgery, a mass was identified that was difficult to limit the division of the gluteal muscle, but without being adhered to deep planes, with apparent vessels nourishing the mass, tumor with a multicystic aspect with the presence of blood inside large cysts, openings and the presence of vessels nourishing the adjacent mass (figure 3 and 4). Mass dissection of the adjacent planes and mass excision en bloc were performed, and finally, hemostasis was reviewed. After removing the mass (figure 4 and 5), which was apparently surrounded by the gluteus, it was necessary to approach it with vycril 3.0 with the positioning of the portovac drain.

After surgery, the patient evolved with a good surgical wound, coapt edges, no signs of secondary infection, portovac drain with serous content and low output, and was discharged from the hospital without any complications.

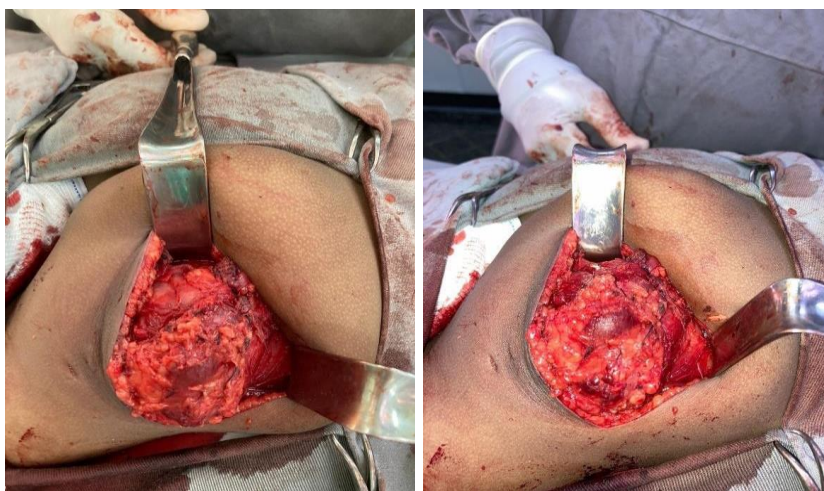
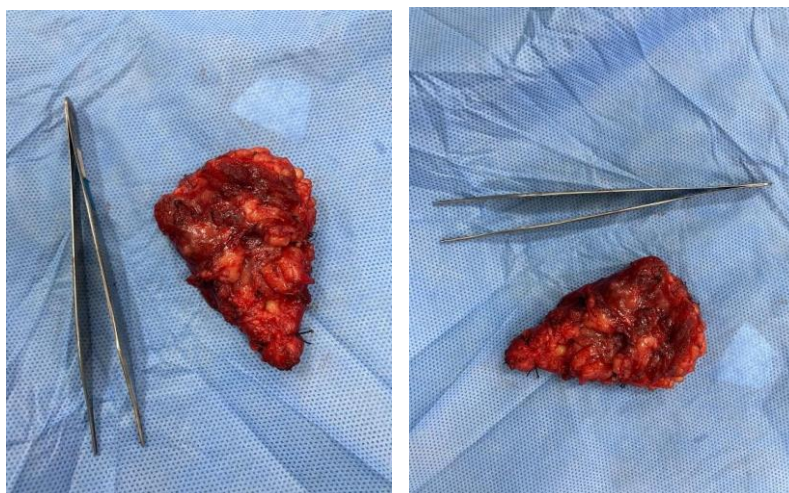


Figure 3 and 4: Tumor with multicystic aspect



Figures 4 and 5: Hemangioma removed.

Discussion

Infantile hemangioma (IH) is the most common pediatric tumor. It is a benign vascular tumor and, for most children, does not pose any serious risk. The tumor appears in early childhood (2–7 weeks of age), proliferates by 4–18 months of age, followed by slow involution over 3–9 years², leaving a fibrofatty residue in 50%–70% of cases, as well as telangiectasia and redundant skin³. In the case reported here, the lesion progressed from intrauterine life to 5 years of age, with no involution of the lesion being observed.

HI is phenotypically classified according to depth and pattern of involvement. The superficial cutaneous HI is located in the papillary and reticular dermis, presenting as a red, finely lobed plaque. Deep cutaneous HI infiltrates the reticular dermis and subcutaneous tissue and may present as a protruding flesh-colored or bluish mass⁴. Patterns of engagement are focal, multifocal, or regional. In the case described, the tumor had a multicystic aspect, not adhered to deep planes with several loculations, hypo/anechoic that infiltrated the adjacent soft tissue, from different margins.

HI with minimal or stopped growth (HI-MAG) is a subtype that does not follow the typical life cycle of HI, but can be confused with capillary malformations due to the distinctive flat telangiectatic appearance. It is present at birth, resolves spontaneously and has a predilection for the lower part of the body. The reason for the lower proliferative potential of HI-MAG is unclear; The distinct superficial anatomical distribution is suggested as an explanation^{5,6}.

Complications, although infrequent in the general population, may be present in up to 25% of patients referred to tertiary services¹. Size and location are the main risk factors for its occurrence. HI can cause complications such as bleeding, ulceration, deformation, and obstruction with functional impairment. In the case in question, the tumor was promoting deformation of the region, although it was not compromising the functionality of the organ.

The diagnosis of HI, for the most part, is clinical. In early stages, superficial HI, including HI-MAG, can be difficult to differentiate from a capillary malformation. Doppler ultrasound is particularly useful in diagnosing deep IH, revealing fast-flowing characteristics, partly with arteriovenous shunts. This can lead to misdiagnosis of arteriovenous malformations⁶. The Doppler study of the case did not show color, however, in the surgical procedure, a multicystic tumor was seen nourished by blood vessels, concluding that it was a hemangioma.

Regarding medical management, given its potential for regression, most cases of IH are conducted expectantly, with adequate guidance to the parents about the natural history and potential complications. For patients with complicated HI who require treatment, medication is the choice for most patients. The main therapeutic options are propranolol, glucocorticoids, and interferon alfa. Propranolol is currently the first drug treatment option, due to its efficacy and safety, demonstrated in recent studies⁹. Treatment of problematic HIs with corticosteroids has largely been replaced by treatment with propranolol. Prednisone or methylprednisolone are still prescribed for patients with IH with contraindications or inadequate response to propranolol¹⁰. However, acute and long-term adverse reactions of corticosteroids in infants should be considered.

HI usually resolves within 6 months of age when propranolol is given early during the proliferative phase; HI regrowth after discontinuation of propranolol occurred in 10%¹¹. Propranolol is currently the only drug approved by the Food and Drug Administration (FDA) for HI. Despite its success, propranolol can cause adverse events in infants: hypotension, bradycardia, peripheral vasospasm, diarrhea, hypoglycemia and seizures, bronchospasm, growth retardation, agitation, and sleep disturbances^{11,12}. Alternatives to propranolol, such as oral atenolol and nadolol, have been used to treat IH, as a selective β 1-adrenergic receptor blocker, atenolol has a lower risk of bronchospasm and hypoglycemia¹³. Up to 25% of HIs with an initially good response to propranolol grow back after discontinuation — an observation that is called the rebound phenomenon. Predictive factors for IH regrowth include discontinuation of propranolol treatment before 9 months of age, profound component of IH, and emergence in a female child¹⁴.

Surgical treatment is usually reserved for patients with extensive scarring fibrosis after lesion regression, pedunculated cutaneous hemangiomas (due to the risk of fibrosis) and lesions with slow regression in aesthetically delicate areas. Surgical treatment may also be considered in cases of ulcerated hemangiomas refractory to systemic treatment, periorbital lesions, and those located at the tip of the nose. It is important to highlight that surgical scarring can have more deleterious aesthetic consequences than spontaneous regression fibrosis, and the risk/benefit ratio should always be adequately evaluated¹⁵.

The treatment time should be just enough to regress the lesions to the point that they are no longer life-threatening or have functional or aesthetic complications. As soon as this goal is achieved, discontinuation of treatment should be considered to minimize the possibility of adverse effects occurring. Since the response to drugs tends to occur early in most cases, a patient should be considered refractory to treatment when there is no regression of more than 25% of the lesion after 90 days of the start of treatment. Once treatment is completed, patients should remain under semiannual medical follow-up for the first 2-3 years. Patients whose lesions cannot be completely evaluated by physical examination should undergo imaging tests in a complementary way at follow-up. After 2-3 years, the risk of disease relapse is minimal, and patients can remain under the usual pediatric follow-up¹⁶. In conclusion, Infantile hemangioma (IH) is the most common benign tumor in childhood, the diagnosis is clinical and eventually associated with imaging tests to classify the depth and involvement of the tissues. The initial approach is expectant and in the face of complicated HI, drug therapy is initiated, with propranolol being the most recommended drug for treatment. Surgical resection is reserved for specific cases that are complicated and refractory to drug treatment. In view of the case described, and the non-response to drug therapy, the surgical approach was shown to be more effective to resolve the condition through tumor resection.

Conclusion

Infantile hemangiomas are common pediatric vascular tumors, often managed conservatively. However, cases unresponsive to propranolol may require surgical intervention. This report highlights the necessity of personalized treatment approaches for IHs, ensuring optimal patient outcomes.

Acknowledgements

We would like to express our sincere gratitude to the dedicated medical professionals who provided valuable insights and expertise on infantile hemangiomas, helping to enhance the clinical interpretation and treatment strategies discussed in this report. Special thanks are extended to the radiology and pediatric teams for their support in diagnostic imaging and patient care, as well as to the surgical staff for their skilled assistance in the reported case's treatment. We are also grateful to the families of the patients for their trust and collaboration throughout the clinical management process.

Declaration Of Conflict Of Interests

The authors declare no conflicts of interest.

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