International Journal of Research in Medical Sciences
Mahmood S et al. Int J Res Med Sci. 2023 Apr;11(4):1343-1345
www.msjonline.org
pISSN 2320-6071 | eISSN 2320-6012
DOI: https://dx.doi.org/10.18203/2320-6012.ijrms20230887

Case Report

An interesting case of genital infantile hemangioma

Shafqat Mahmood*, Muhammad Naeem

Department of Family Medicine, Primary Health Care Corporation, Doha, Qatar

Received: 06 February 2023
Revised: 04 March 2023
Accepted: 06 March 2023

***Correspondence:**
Dr. Shafqat Mahmood,
E-mail: shafqat_mahmood@hotmail.co.uk

ABSTRACT

Vascular lesions are commonly seen in newborns. They may be present at birth or appear in the days, weeks or months that follow. Most are benign and self-limiting but rarely they may be part of complex syndromes or systemic disorders, or they may be associated with complications requiring treatment. This is a case of a 2-month-old baby presenting with an Infantile hemangioma in the genital region.

Keywords: Infantile hemangioma, Genital hemangioma, Genital infantile hemangioma, High risk hemangioma

INTRODUCTION

The terminology used for vascular lesions has historically been inconsistent, and hemangiomas are often described using outdated terms such as angioma, capillary hemangioma, cavernous hemangioma, juvenile hemangioma, and strawberry hemangioma.[1]

The International society for the study of vascular anomalies (ISSVA) classifies vascular lesions into vascular tumors, simple vascular malformations, combined vascular malformations (two or more vascular malformations found in a single lesion), anomalies of major named vessels, and malformations associated with other anomalies (e.g., Sturge-Weber syndrome).[2]

The most common vascular lesions seen at, or soon after birth, are vascular malformations followed by benign vascular tumors such as infantile hemangiomas (IH).

IH are the most common benign tumors of infancy with a prevalence of 4.5% (3.9% in term infants and 14.4% in very premature infants) whereas congenital hemangiomas (CH) are much rarer with a prevalence of 0.3%.[3] IH and CH are clinically, genetically, and histopathologically distinct from each other.

IH usually arise within days or weeks of birth and almost always before 3 months of age whereas CH are fully developed at birth.[1,3]

IH undergo an active proliferative phase and most reach 80% of their maximum size by 5 months.[4] Proliferation after the first year is uncommon although deep lesions may proliferate until age 2 years. Involution occurs between 12 to 18 months and can last up to 5 to 6 years.[5]

Rapidly involuting congenital hemangioma (RICH) typically involutes by 12-14 months.[5] Non-involuting congenital hemangioma (NICH) and partially involuting congenital hemangioma (PICH) subtypes continue to grow at a rate proportional to the child's growth.

Risk factors for IH include female sex (female to male ratio 2.4:1), prematurity (gestation <37 weeks is seen in 20% of patients), low birthweight, fair skin, high maternal age, pre-eclampsia, placenta previa and multiple gestation pregnancy.[6]

IH have a predilection for the head and neck (>50%), although they can occur anywhere in the skin, and they less commonly involve the mucous membranes or internal organs.[7] Less than 1% are in the genital region, and so male

genital IH are rare.[8] They range from a few millimeters to many centimeters in diameter. They may be superficial, deep, or combined. Superficial lesions are bright red and well demarcated. They may be protuberant, bosselated, or smooth.

Most IH are uncomplicated and require no intervention. However, 10-15% result in complications and may ulcerate, leading to pain, bleeding, scarring, and/or infection.[9] Others may cause functional impairment and/or disfigurement during or after the proliferative phase (e.g., airway or periorbital hemangiomas). Early intervention before the completion of the rapid growth phase is recommended for infants with high-risk hemangiomas.

CASE REPORT

An eight-week-old healthy baby boy was seen at his registered primary healthcare center for his routine eight-week baby clinic assessment and vaccinations. He was born at full term following a normal vaginal delivery, to a 36-year-old mother.

On examination, he was noted to have multiple vascular lesions at the base and shaft of his penis and on the left side of his scrotum. This was not recorded at the time of birth and according to his mother, the skin condition appeared 3 weeks after birth. The remainder of the physical assessment was unremarkable.

He was diagnosed as having a genital hemangioma and referred to a pediatric specialist given the site and apparent extent of vascular lesions.

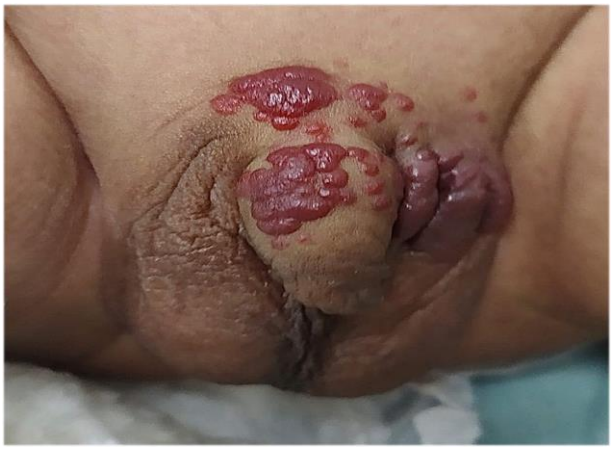

Figure 1: Genital infantile hemangioma.

Review of case

The child was diagnosed as having a genital hemangioma but the precise diagnosis is genital IH given the lesion was not apparent at birth but appeared weeks after birth. The decision to refer was correct given that genital IH are considered high risk for ulceration and possible functional impairment.

He was seen by pediatric specialist services and commenced on oral propranolol, starting at a dose of 1.2 mg twice daily, and currently on a dose of 2.5 mg twice daily.

DISCUSSION

High-risk hemangiomas have evidence or potential of the following.[10] It is recognized that anogenital IH are at high risk of ulceration.[11]

Table 1: Life threatening complications.

Life threatening complications	Hemangiomas that compromise vital organ function of the airway, liver, or gastrointestinal tract
Functional impairment	Periorbital hemangiomas that compromise vision, or ulceration
Associated structural anomalies	Posterior fossa anomalies, hemangioma, arterial anomalies, cardiac anomalies, and eye anomalies (PHACE) syndrome
	Lower-body hemangioma and other cutaneous defects; urogenital anomalies, ulceration; myelopathy; b ony deformities; anorectal malformations, arterial anomalies; and renal anomalies (LUMBAR) syndrome
Risk of permanent disfigurement	Hemangiomas located in a cosmetically sensitive area
Large size	≥5 cm, rapidly growing cutaneous hemangiomas

Extracutaneous involvement

Although extracutaneous involvement is uncommon, when present, spinal cord anomalies are the most frequent finding, seen in 70% to 80% of cases. Tethered cord, lipomyelocele or lipomyelomeningocele are the most common associated myelopathies. Anorectal anomalies (e.g., imperforate anus, fistulae) are seen in 29% to 38%, and renal anomalies (e.g., single kidney) in 25% to 31% of extracutaneous cases. Vessel abnormalities described in LUMBAR syndrome (dysplasia, narrowing, aberrant course or origin, persistence of embryonic anastomoses) present in 8% to 14% of extracutaneous cases and show striking parallels with PHACE syndrome.[12]

Management

High-risk IH should be referred for specialist treatment. Treatment aims to prevent or reverse life-threatening complications and functional impairment, and to prevent or minimize disfigurement/scarring, bleeding, pain and infection, and to minimize psychosocial distress for the patient and family.

Medical therapies for IH are most effective when initiated as early in the growth phase as possible, ideally within the first three months after birth.

Uncomplicated, low risk IH require no active intervention although superficial IH in cosmetically sensitive areas can be treated with topical timolol.

High risk or complicated IH can be treated with oral propranolol starting with a dose of 1 mg/kg per day and gradually increasing to a target dose of 3 mg/kg per day for 6 months.[13] A significant side effect of oral propranolol is hypoglycemia and so it should be administered after adequate feeding or withheld when oral intake is poor or absent.

Systemic corticosteroids are alternative agents for patients in whom beta blockers are contraindicated or may be used as combined therapy in severe, select cases.

The use of vascular laser for telangiectasia after regression of IH is well established; however, there is also weak evidence to suggest that early treatment of superficial IH with laser may arrest growth and help treat ulcerated IH. When laser therapy is used in combination with systemic or topical beta-blockade, efficacy is increased without any appreciable increase in side effects.[14]

Surgical excision is generally reserved for involuted lesions with residual scars or loose skin; hemangiomas with inadequate involution in cosmetically sensitive areas; and pedunculated, cutaneous hemangiomas.

Children with periorbital IH should be evaluated and monitored by an ophthalmologist who is experienced in the treatment of hemangiomas.

CONCLUSION

IH are very common benign tumors of infancy seen by a diverse range of providers. Most IH do not require treatment as they regress spontaneously and do not leave significant residua. Ulceration, impairment of vital structures, and potential disfigurement are considered indications for treatment. Genital IH are rare but at high risk of ulceration. Propranolol is effective at a dose of 3 mg per kilogram per day for 6 months in the treatment of IH, however, it should be used cautiously, and patients must be closely monitored for adverse effects.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Mahmood S, Naeem M. An interesting case of genital infantile hemangioma. Int J Res Med Sci 2023;11:1343-5.