

Bilateral asymptomatic incidental vulvar/labial hemangioma: Kissing Hemangiomas

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Abstract

We present a rare case of genital hemangiomas. Generally, hemangiomas are present at birth or in the first year of life and completely resolve by age 10. Most are asymptomatic. We present an 11 year old girl who presented with incidentally diagnosed asymptomatic bilateral hemangiomas on her labia majora. The treatment for genital hemangiomas is not well defined in the literature. We elected to choose watchful waiting as our treatment approach at this time. Long term follow-up is necessary with intention to intervene with multispeciality approach involving gynaecology and plastic surgery to avoid any disfigurement of the local anatomy.

Introduction

Hemangiomas are the most common benign tumor of infancy with 8-12% occurrence in full-term Caucasian infants, and 22% in preterm infants (weight <1000g). The incidence is higher in females (3-5:1). However, they are not often found in the genitalia (1% of all hemangiomas) and only 5 cases of genital hemangiomas have been reported worldwide.¹⁻⁶ The natural history of genital hemangiomas is not well defined in the literature.

Vulvar hemangiomas are often present at birth or are frequently found during early childhood. The median age for presentation is 2 weeks; however, 30-50% are present at birth.⁷ Most hemangiomas are expected to involute by age 10.

We present a case of asymptomatic bilateral labial hemangiomas in an 11 year old female. Since they are two distinct masses that are in direct opposition to each other we refer to them as Kissing Hemangiomas.

Case Report

An 11 year old girl presented to our department to discuss her persistent Grade 2 Vesicoureteral Reflux (VUR). On physical exam the patient was found to have bilateral hemangiomas on the labia majora (1x1.5 cm) (Figure 1). The hemangiomas are two distinct masses separated by normal tissue in the midline at the level of the clitoris. On clinical examination they appeared blue-purple, compressible, non-expansile, and non-pulsatile. While the patient was being treated for her VUR, vaginoscopy and examination under anesthesia were performed to rule out any anatomical or congenital anomalies. This examination revealed a single cervix and a deep vestibule. On further questioning, the mother confirmed that the lesion was not present at birth, and had not been noticed prior to physical exam in our office. The patient also denied any trauma and was not sexually active.

Physical examination revealed significant obesity and no other hemangiomas/cutaneous lesions on any other part of the body.

Discussion

A very few number of genital hemangiomas have been reported worldwide in females. In a majority of these cases the presentation was clitoromegaly or an enlarging mass.¹⁻⁶ Terminologies such as vascular malformations and hemangioma are used interchangeably with significant overlap between the definitions. Here in we elect to discuss both entities with regards to presentation, cutaneous findings, and management.

Based on the literature review it is difficult to delineate which entity was actually presented in each individual case report due to the wide definitions being used to characterize them. We reviewed the literature for age of presentation, site, diagnosis, treatment and investigations performed prior to treatment (Table 1).

It is important to differentiate a true hemangioma from a venous malformation. Strawberry hemangiomas, also known as capillary hemangiomas, result from the proliferation of immature capillaries.⁹ Morphologically they are bright red to blue and can vary in size from a few millimeters to a few centimeters. They are usually single, but 20% of the time they are present in multiple cutaneous sites and other organ systems. The systems most commonly involved are the liver, gastrointestinal tract, and the brain.⁷ While the majority are benign, 1% may be life threatening.¹

While hemangiomas are common, they are rarely found in the genitalia and these are only

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responsible for 1% of all hemangiomas. Generally these lesions grow for the first 6-8 months of life, but 60-90% gradually involute and often require no treatment. Complications are rare, and generally include ulceration and bleeding.^{1,7,10,11}

A vascular malformation, on the other hand, is a structural anomaly and can be differentiated from a capillary hemangioma by clinical exam. They are generally present at birth and are progressive. They may get progressively larger under a few circumstances including infection, trauma, or hormonal influence.³ Morphologically, they are flat, soft, and range in color from brown to red or purple.¹⁰

Diagnosis of a vulvar hemangioma can be established by clinical inspection of the lesion. A capillary hemangioma changes in size with compression, and can be level with the surface of the skin or slightly elevated.¹¹

The natural history of a hemangioma is as follows: 50% of the time they are present at birth as a faint cutaneous manifestation. From 3-9 months of age rapid postnatal growth is present. This can last up to 18 months. Involution occurs from age 2-9, and by age 10 the hemangioma is resolved. This regression has not been found to be dependent on original size, location, or appearance.¹

If the hemangioma is asymptomatic, it rarely requires treatment. However complications may require further intervention. Ulceration occurs in 5% of lesions, and is more often found in those lesions that are located on the lips and anogenital areas.¹ Treatment option includes a short course of oral steroids.^{1,10,11} Another recommends daily cleansing and application of topical antibiotics, and even oral antibiotics if topical fails. If eschar formation is present, then dressing is desired. An alternative therapy is use of a pulse dye laser, cryosurgery, or use of an argon laser. However the best results for laser appear in cutaneous lesions that are less than 3 mm

Table 1. Literature review.

Article	Patient's age	Diagnosis	Presenting symptom	Complications	Treatment	Diagnosis reaching
Bruni et al.	20	Cavernous hemangioma	Clitoral enlargement at age 17	N/A	Surgical excision	Vulvar u/s
Kempinaire et al.	12	Vascular malformation	Increasing size of mass in pubic region	N/A	Local excision by Plastics	Doppler u/s, MRI
Ishizu et al.	4	Arteriovenous hemangioma	Clitoral enlargement at age 1	N/A	Excision by Urology	N/A
Kaufman-Friedman	18	Cavernous hemangioma	Clitoromegaly, diagnosed with 8 adrenogenital syndrome at age	N/A	Excision by plastics	Clinical
Strayer et al.	30	Epithelioid hemangioendothelioma	Nodule of the clitoris, progress to painless ulcer, +LAD	N/A	Modified radical vulvectomy and bilateral inguinal LN dissection	Pathological staining
Wang et al.	15-85, 8 cases	Vascular lesions: 2 cervix and vagina, 1 vagina, 5 vulvar. All were venous malformations	Patients complained of bleedings, gradually enlarging vulvar masses, or incidentally			Routine blood-CEA, AFP, CA-125, Beta hCG, pap tests, Doppler u/s, MRI



Figure 1. Bilateral hemangiomas on the labia majora.

thick. Surgical indications for excision include blockage of airway or vision, painful or intractable ulceration, or continued bleeding.^{1,7}

When the diagnosis is in doubt excisional biopsy is indicated for a definitive pathologic diagnosis.¹⁰ This case is unique in nature because the patient has a hemangioma that is bilateral, and presented at a late age. Since there is vulvar involvement it is necessary to be as conservative as possible.³ Due to lack of current guidelines on how one should manage patients with asymptomatic bilateral labial hemangiomas, we currently elected to watch them closely with regular follow-up. The patient's family has been notified to follow-up to the emergency department if they notice any signs of bleeding or ulceration. On review of literature some questions regarding treatment and follow-up are unanswered such as is any intervention needed immediately? What is the treatment protocol that needs to be followed? At what age should this patient be treated? Should she be followed by Gynecology, Urology, Plastics, or a multidisciplinary team? There

are several aspects that need to be considered including lack of symptoms and cosmesis.

Conclusions

Bilateral labial hemangiomas are rare and have not been reported in the literature. These may present later in life in the adolescent age group as asymptomatic lesions on clinical examination. Close follow-up is critical and well defined treatment guidelines are necessary.

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