

Ultrasonography Findings of Rare Form Deep Vulvar Hemangioma: A Case Report

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ABSTRACT

Background: Hemangiomas are the most common vascular tumors in infants and children, they typically appear after birth, grow rapidly, and then gradually involute. Early diagnosis is crucial, but the clinical presentation of hemangiomas can resemble other vascular anomalies, necessitating the use of imaging studies like ultrasonography (US) in order to establish accurate diagnosis. This study aims to present the ultrasonographic characteristics of a rare case of vulvar hemangioma, with the objective of aiding in its differentiation from other vulvar vascular malformations.

Case Presentation: A 9-year-old girl presented with a painless vulvar mass that had been enlarging over 3 months. Physical examination revealed a 3 x 2.5 x 1.5 cm, well-defined, skin-colored, non-tender, and immobile mass. Greyscale ultrasonography showed a septated cystic lesion with internal echoes. Doppler ultrasonography showed low flow vascular patterns, suggestive of a hemangioma. Surgical excision was performed, and histopathology confirmed the diagnosis of a hemangioma

Results: Although vulvar hemangioma is rare, it can mimic other vascular anomalies, making imaging essential for accurate diagnosis and management. Ultrasonography, as a inexpensive and non-invasive imaging modality, plays a crucial role in differentiating hemangiomas from other vascular malformations.

Conclusion: Hemangioma is a form of vascular mass and the most common one, at early phase the clinical presentation might resemble other vascular anomalies. Thereby diagnostic modalities such as ultrasonography might be able to help to establish the diagnosis

Keywords: ultrasonography, vulvar hemangioma, vascular tumors

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BACKGROUND

Hemangiomas are benign vascular neoplasms that can develop in cutaneous or

internal locations. According to the International Society for the Study of Vascular Anomalies (ISSVA), in 1992, vascular

anomalies were classified into true vascular tumors or hemangiomas and vascular malformations. While vascular tumors arise and regress in infancy and childhood, vascular malformations usually arise and develop until adulthood (Itinteang et al., 2014). Hemangiomas or true vascular tumors are further classified based on depth (superficial, deep, or a mixed hemangioma), growth pattern (reticular, abortive, or minimal), distribution on the body (focal, multifocal, or segmental), and microscopically (Braun et al., 2020). It is the most common neoplasm in infants and children, affecting about 1-12% of younger patients, they typically appear after birth, grow rapidly, and then gradually involute. Clinically, it may mimic other types of vascular anomalies or non-vascular benign and malignant tumors (Abraham et al., 2016).

Hemangiomas originate from stem cells, a theory posits that hemangiomas may originate from an abundance of endothelial progenitor cells, resulting in the formation of aberrant blood vessels (Abraham et al., 2016). Several factors can stimulate these stem cells to proliferate and differentiate into various cell types (Merlino et al., 2024). These cells subsequently differentiate into endothelial cells, which subsequently become uncontrolled and proliferate into vascular tumors (Ding et al., 2019). The identified risk factors encompass first-trimester bleeding, preeclampsia, prematurity, advanced maternal age, placental abnormalities, female sex, hormonal therapy, and low birth weight (Dehart et al., 2019).

Hemangioma is usually diagnosed based on clinical history, physical examination, and, noninvasive modalities (Dehart et al., 2019). Clinical manifestation of hemangioma may vary according to depth and distribution (Venkatraman et al.,

2014). Cutaneous/superficial hemangioma, commonly seen as a red macule, localized petechia, and raised in its early form, while deep hemangioma consistently appears bluish and less prominent (Clemente et al., 2023). Focal lesions are solitary and well-circumscribed, multifocal types involve multiple discrete lesions, while segmental hemangiomas are large, plaque-like, follow dermatomal distributions, and carry a higher risk of ulceration and complications (Dehart et al., 2019). Hemangiomas are typically self-limiting but may require early intervention when complicated by severe bleeding, functional impairment, or psychological impact. While corticosteroids are the first-line therapy, stepwise embolization followed by surgical excision can be effective for intractable or large lesions, offering both functional and cosmetic benefits. Additionally, treatment is more effective at earlier stages, hence underscoring the significance of a timely and accurate diagnosis (Clemente et al., 2023). Morbidity and mortality are varied, depending on location and phase. (Abraham et al., 2016).

Vulvar hemangioma is a rare form of hemangioma of female genital tract (Cebesoy et al., 2008). When present cutaneously, vulvar hemangiomas often appear as soft, compressible blue-purple lesions. It may also appears as raised lump beneath the skin similar to other vascular anomalies (Merlino et al., 2024). Although they frequently occur in pediatric and adolescent groups, postmenopausal women may occasionally experience them. (Verma et al., 2023) It can be life threatening because they may lead to uncontrollable bleeding, deformities, and functional impairment. (Bava et al., 2002; Mundeli et al., 2024).

In cases where the diagnosis is uncertain, particularly in deep type hema-

ngiomas, noninvasive radiological studies can be instrumental in establishing the diagnosis (Cheung, 2018; Merlino et al., 2024). Noninvasive modalities such as Ultrasonography, CT-scan and MRI are useful in this case, however ultrasonography is a preferred modalities to diagnose hemangioma.

Ultrasonography is one of the most common, conventional, readily available, inexpensive modalities that is important to help differentiate hemangioma with other vascular anomalies (Dehart et al., 2019). Ultrasound has the ability to distinguish skin layers, making it a valuable tool for evaluating many cutaneous pathologies (McNab et al., 2021). Both gray-scale ultrasound and doppler ultrasound can be used in order to monitor hemangioma. Gray-scale ultrasound evaluates the lesion's size, shape, borders, echogenicity, and internal texture. Color Doppler ultrasound assesses the vascularity and blood flow patterns within the lesion (Ding et al., 2019).

CASE REPORT

A 9-year-old young girl came to the pediatric surgery department with her parents, with the main complaint of a painless mass in her left vulva. The mass had appeared 3 months ago and was getting bigger in size gradually during the 3-month course. There was no history of a similar lump before. The girl was born healthy without a history of birth anomalies or prematurity. There was no significant family history related to this case. No medication was applied to the mass. No history of allergy was notable. During the physical examination, a mass was seen in the left vulva and it was about 3x2.5x1.5 centimeters in size, with a well-defined border, smooth surfaces, and circumscribed boundaries. There was no discoloration observed; it was covered with skin. The mass was non-tender when palpated, immobile, and soft in texture (Figure 1). A transillumination test was negative.

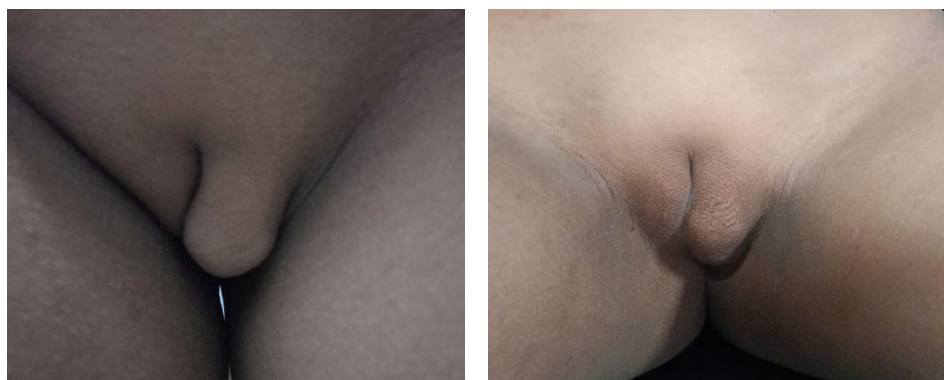


Figure 1. Clinical presentation of the vulvar mass

Doppler ultrasonography was later performed in the following days. It showed a septated cystic lesion with internal echoes and septal flow measuring 2.3 x 2.7 x 1.6 cm in the left labia majora. A low-flow lesion was noted. The right and left canals of Nuck appeared obliterated. There are no

abnormalities seen in the bladder, uterus, adnexa, and no free fluid in the abdominal cavity was observed. The ultrasonography findings suggest hemangioma with a differential diagnosis of venous malformation or lymphatic malformation (Figure 2).

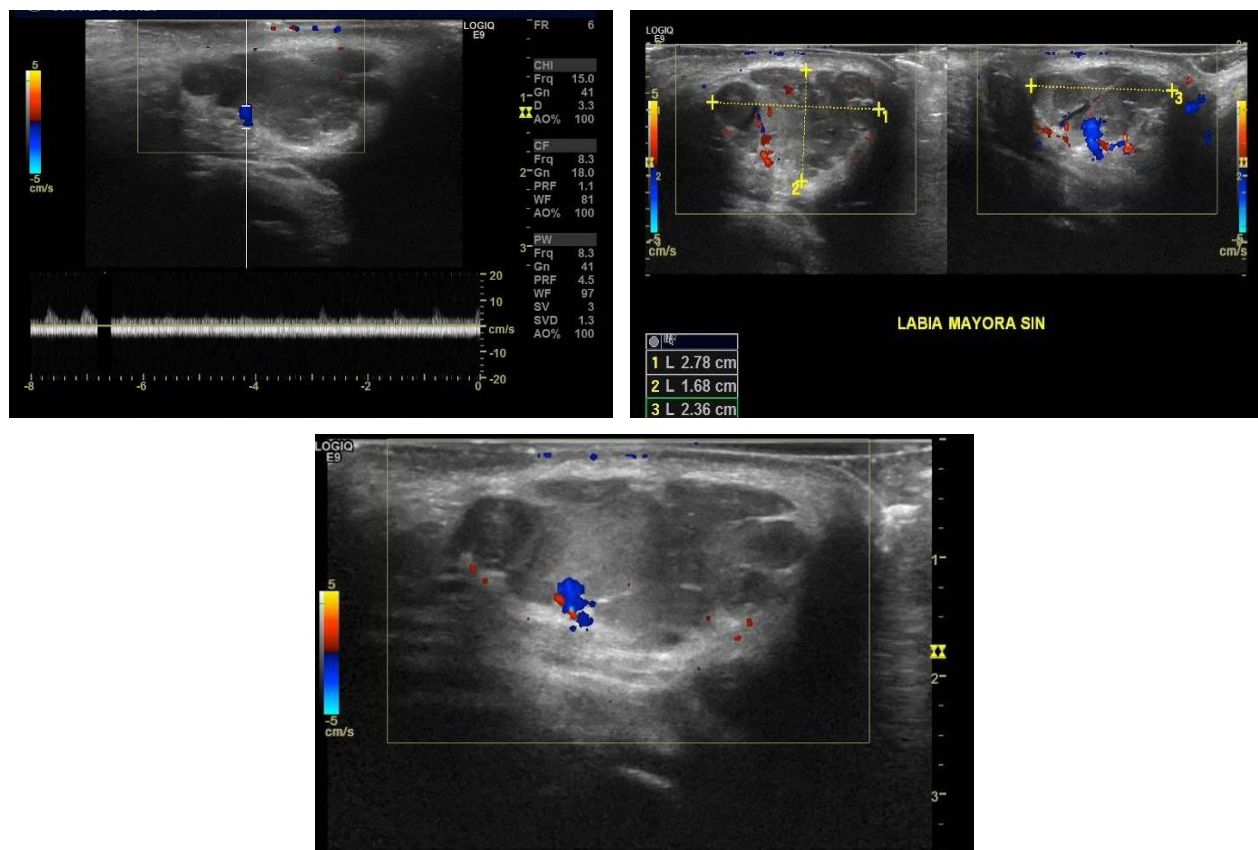


Figure 2 Doppler ultrasonography of the vulvar mass : well defined septated cystic lesion with internal echoes and septal flow measuring 2.3 x 2.7 x 1.6 cm in the left labia majora. A low flow lesion was noted.

Surgery was then scheduled for the next week, and preoperative requirements were then tested. An en bloc excision of the tumor from the surrounding tissue was performed during surgery, with bleeding controlled. The tissue sample was later sent

to the Pathology Department to be examined microscopically (Fig. 3). A post-surgical medication regimen included IVFD D5 1/2 NS fluid, Cefuroxime, Ranitidine, and Metamizole.



Figure 3 Tissue sample of the vascular mass

There were no complications during surgery. Because of the stable condition, the patient was later discharged from the hospital the following day. Follow-up in the following week showed no complications, there was no infection observed from the wound, and the patient also did not have any complaints regarding the excision wound.

DISCUSSION

Hemangiomas are the most common vascular anomaly that occurs in infants and children. The tumors usually appear after birth, grow rapidly, and involute over the years (Verma et al., 2023). Mainly around 60% of hemangiomas are located in the cervicofacial region ((Kayar et al., 2013)). On rare occasions, it has been reported in both female and male genitalia, as documented in the case reports by Gangkak et al. and Kim et al (Gangkak et al., 2015; Kim et al., 2018).

In regard of its location and risk of ulceration, surgical excision is preferable in this case, however most of hemangioma don't always need intervention, conservative therapy should be first option in order to treat hemangioma (Abraham et al., 2016; da Silva et al., 2018). Although some cases reported vascular embolization as an option (Celemente et al., 2023). Hemangioma is usually diagnosed on the basis of history and clinical appearance.

However, in the early phase of the hemangioma, the clinical presentation might be the same as other vascular anomalies such as venous malformation and lymphatic malformation; thus, imaging might be needed to differentiate it. Ultrasonography is the primary diagnostic tool for evaluating soft tissue vascular anomalies and can analyze the vascular flow (Clemente et al., 2023; Merrow et al., 2016).

The pathology results of tissue sections showed a proliferation of large, dilated blood vessels, lined by a single layer of thin-walled endothelial cells. The lumens contained erythrocytes (red blood cells). No signs of malignancy were seen. This description is consistent with a hemangioma, a benign (non-cancerous) tumor of blood vessels.

Ultrasonography is considered fast and inexpensive modality to diagnose hemangioma (Esposito et al., 2018). The sonographic appearance of vulvar hemangioma is the same as the rest of hemangiomas, it can vary depending on their stage and morphologic type. During the proliferative phase, hemangiomas appear as solid masses or plaques (focal type) or mixed but variable echogenic areas (segmental type). They have internal echogenic septa and marked, diffuse, increased vascularity on Doppler ultrasonography. Most hemangiomas, especially those of the focal type, are well-defined with lobulated borders. However, indistinct and infiltrative borders can also be present. As the lesion shrinks during the involuting phase, hemangiomas become less circumscribed and indistinct. They have less vascularity on Doppler ultrasonography images and more echogenic areas indicating fibrofatty tissue (Park et al., 2021).

Hypoechoic or hyperechoic lesions are insubcutaneous and have variably increased vascularity. The flow either high or low depending on the phase (Clemente et al., 2023). In the early phase of hemangioma, ultrasonography findings include well-defined hypoechoic, heterogeneous texture with cystic sinusoidal spaces, and fast flow pattern (Venkatraman et al., 2014). In case of subcutaneous lesions or mass, high frequency radiology is preferable in order to help to distinguish the differential diagnosis (Rodríguez Bandera

et al., 2021). Hemangioma phases could be distinguished using Colour Doppler because of its different vascularity and echogenicity patterns (Wortsman, 2018). Sometimes Colour Doppler might shows tortuous blood vessel with multidirectional turbulent flow (Cheung et al, 2018).

If surgical removal is a potential treatment, colour Doppler ultrasound can help plan the procedure by showing how far the lesion extends (Cheung, 2018; McNab et al., 2021). It's worth noting that during the involution phase, a slow-flow pattern may be observed, which could potentially lead to a misdiagnosis of venous malformation. However, in the case of venous malformation, phleboliths are always present. In this case report, it's likely that the hemangioma was in the proliferative phase because the diffuse imaging and internal echogenic features were noticeable. These findings were corroborated by the histopathological examination. (Ding et al., 2019).

In conclusion, although vulvar hemangioma is rare, it can mimic other vascular anomalies, making imaging essential for accurate diagnosis and management. Ultrasonography, as a inexpensive and non-invasive imaging modality, plays a crucial role in differentiating hemangiomas from other vascular malformations.

AUTHORS CONTRIBUTION

All authors contributed equally to the conception, design, writing, and revision of this manuscript. All authors have read and approved the final version of the article.

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CONFLICT OF INTEREST

The authors declare that the study was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest

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