Intramuscular Hemangioma Mimicking an Adnexal Malignancy

Adneksiyel Maligniteyi Taklit Eden İntramüsküler Hemanjiyom

Nadir Bir Pelvik Kitle / An Uncommon Pelvic Mass

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Özet

İntramüsküler hemanjiyomlar, uluslararası vasküler anomaliler sınıflama çalışmasına göre vasküler malformasyonlar olarak tanımlanmaktadır. Radyolojik görüntülemedeki agresif görünüme rağmen agresif olmayan klinik seyir, özellikle bu lezyonlara alışık olmayan klinisyenler için tanıda zorluk yaratmaktadır. Biz burada 49 yaşındaki bir kadın hastada, pelvik taban kaslarından kaynaklanan ve adneksiyel malign bir kitleyi taklit eden mikst tip intramüsküler hemanjiom olgusunu sunmaktayız.

Anahtar Kelimeler

Hemanjiyom; Pelvik Kitle; Pelvik Kaslar; Pelvik Cerrahi; Vasküler Malformasyon

Abstract

Intramuscular hemangiomas were defined as vascular malformations according to the International Society for the Study of Vascular Anomalies (ISSVA) classification. Aggressive appearance on radiologic images but nonaggressive course of these lesions results in diagnostic challenge for the clinicians that are unfamiliar with these lesions. Here, we present a 49-year-old woman as a rare case of mixed type intramuscular hemangioma originating from pelvic floor muscles, mimicking a malignant adnexal mass.

Keywords

Hemangioma; Pelvic Mass; Pelvic Muscles; Pelvic Surgery; Vascular Malformation

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Introduction

Intramuscular hemangiomas (IMH) were defined as vascular malformations according to the International Society for the Study of Vascular Anomalies (ISSVA) classification [1] . They comprise 0.8% of all hemangiomas [2]. Since it was reported as a first case by Liston at 1843 [3], orthopedicians are familiar with these benign lesions due to their localization predominantly in the upper and lower limb [4]. However, unusual localizations of IMH was reported in the literature, including chest wall, orbicularis oculi muscle, and levator anguli oris muscle [5-7]. Aggressive appearance on radiologic images but nonaggressive course of these lesions is a diagnostic challenge for the clinicians unfamiliar with these lesions, especially when they arise in unexpected locations. Here we present probably the first case report of an IMH originating from pelvic floor muscles, mimicking a malignant adnexal mass.

Case Report

A 49-year-old woman was admitted to our clinic with a diagnosis of pelvic mass detected accidentally on abdominal sonography for the evaluation of dyspeptic complaints. Abdominal and transvaginal ultrasonography showed a 98x74 mm heterogeneous mass with cystic-solid, cavitary appearance with a mixed echo pattern. Color Doppler imaging did not exhibit increased color flow or alterations in vascular resistance parameters on the cavitations (Figure 1a). Vaginal examination revealed stiff dullness on the left vaginal wall and bulging on left upper vagina. Serum tumor markers and hormonal parameters including CA 125, CA 15-3, CA 19-9, CA 72-4, CEA, Alpha-feto protein, estradiol and inhibin were all within normal limits. Pelvic magnetic resonance imaging (MRI) showed a marked hyperintense mass measuring 85x83 mm with lobulated contours, filling the left adnexal region on sagittal T2-weighted images (Figure 1b). Cervical cytology was normal.

The woman had a history of myomectomy 15 years ago and reported no other significant health problem. We preferred laparoscopy for adnexal evaluation. Laparoscopic examination revealed a well circumscribed, mostly solid, sphere-shaped retroperitoneal mass bulging from bottom at left anterior peritoneum. Because of the deep retroperitoneal location of the lesion, we shifted from laparoscopy to laparotomy. A soft, compressible, cystic mass with a fibrous dense capsule growing through vesicovaginal space was detected. The mass was completely removed with adequate margins. Large prominent vascular connections were not observed, and intraoperative bleeding was minimal during dissection. Frozen section analysis was not diagnostic and reported as “nonspecific mesenchymal tumor”.

On final pathologic examination, a 7x8x4 cm mass with lobulated surface and solid-cystic, hemorrhagic appearance on cross-sectional analysis was observed macroscopically. Microscopically, hematoxylin and eosin staining showed multiple small and large vascular spaces scattered between the bundles of skeletal muscle. Immunohistochemically, endotelial cells were markedly positive for CD 34 staining (Figure 2) . Therefore, the tumor was diagnosed as mixed type intramuscular hemangioma of deep pelvic muscles. Patient is well with normal MRI findings postoperatively, and follow up during 5 months revealed no recurrence.

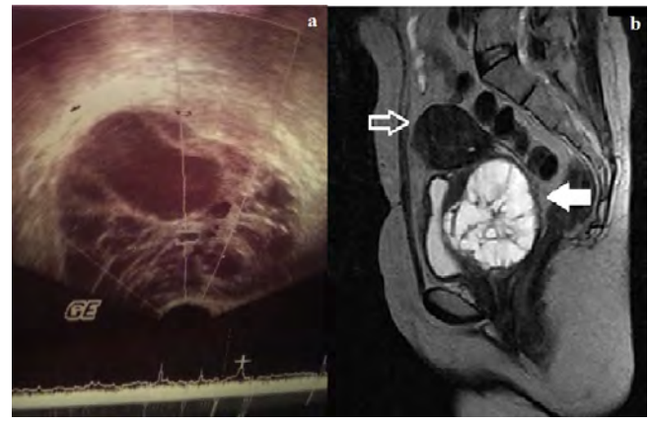


Figure 1. No vascular filling on color Doppler ultrasonography(A). White arrow shows hyperintense mass, blank arrow points uterus pushed upward due to mass effect (MRI sagittal plane, T2 weighted image)(B).

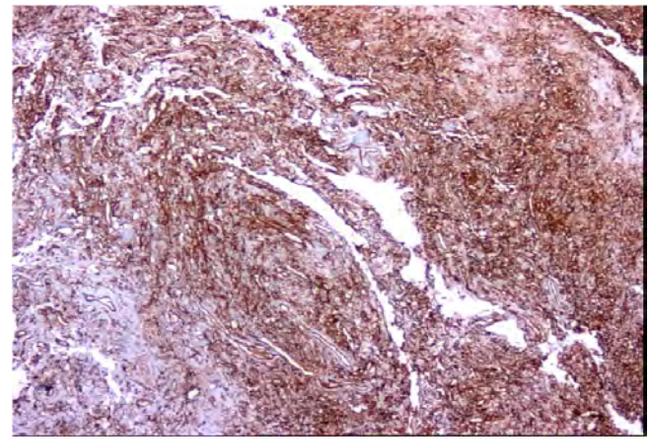


Figure 2. Endothelial cells shows marked positivity for CD34 stain (magnification ×50)

Discussion

IMH is a disease of young-adults and is usually diagnosed before age 30 [4]. Pain is the major symptom in reported series [8]; however, it may remain asymptomatic despite its large size as in our case. Thrills and bruits are uncommon. There are several classifications for IMH. While Allen and Elzinger classified their 89 IMH cases according to vessel size as “small vessel”, “large vessel” and “mixed” types [8], Beham and Fletcher used a histological classification as capillary, venous, cavernous, lymphatic, and complex lesions. However, they reported that vessel type and size has no influence on symptoms and risk of recurrence [4].

Regarding pathophysiology, it is suggested that nerve fibers and related neuropeptides such as calcitonin gene related peptide (CGRP) have some stimulating role in the development of these lesions [9]. Congenital causes and/or acquired stimulating processes such as trauma and pregnancy are other theories considered responsible for the development of these lesions [5-10].

Calcified bodies on plain radiography (phlebolitis) may aid the diagnosis of IMH located at upper and lower limbs [10]. MRI is a potential diagnostic modality for IMH [11]. A characteristic appearance reflecting vascular and nonvascular components of tumor such as adipose and fibrous tissue facilitates the diagnosis of these lesions. Marked hyperintensity on T2 images secondary to the blood in large vessels and low signal structures

reflecting fibrofatty septa between vessels can be observed on MRI [11-12]. Angiography may expose the vascular structure and its relationships with the neuromuscular bundle; however, MRI is usually proves to be satisfactory.

For treatment, complete excision is important due to the high rate of recurrence in case of incomplete surgery [4-13]. Hemorrhagic complications are not common during surgery [8]. Allen and Elzinger reported 18% recurrence rate; 7% recurred more than once but all of these recurrences were local with no distant metastases observed [8]. Wild reported 11 cases and one recurrence treated with adjuvant interferon alpha in his series [11]. Regarding predictors of recurrence, size of tumors and margin status are the only identified risk factors [13]. Mixed type was reported to have the highest rate for recurrence [8]. If surgical excision will result in functional impairment in adjacent tissue radiotherapy, sclerotherapy, and angiographic embolization are alternative treatment modalities [14-15]. On differential diagnosis, angiosarcomas are the most important lesions to be considered due to altered treatment and prognosis [8].

In conclusion, IMH originating from pelvic muscles was not reported previously to our knowledge. Pelvic location and complex appearance on radiologic images should raise suspicion of gynecologic malignancy.

Competing interests

The authors declare that they have no competing interests.

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