International Journal of Academic Health and Medical Research (IJAHMR)
ISSN: 2643-9824
Vol. 5 Issue 12, December - 2021, Pages:9-12

Ovarian Angioleiomyoma: About A Case and Review of the Literature

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Abstract : *Angioleiomyoma (ALM) is a rare, painful benign tumor that refers to a rare type of leiomyoma that arises from the smooth muscle cells of the arterial and venous walls. ALM is very rarely found in female genitals such as the uterus or ovary. We report the case of a huge primary ovarian ALM in a 40-year-old nulligestous woman admitted for the management of a progressive increase in abdominal volume with chronic pelvic pain. We believe that angioleiomyoma should be recognized and classified as a benign variant of leiomyoma. Although we did not have follow-up data, angioleiomyomas almost certainly presented as benign, as the morphology clearly had indications of a benign lesion.*

Keywords: Angioleiomyoma, ovary, degeneration

INTRODUCTION

Angioleiomyoma (ALM) is a rare benign tumor that arises from smooth muscle cells with a vascular component.

The genital location in women at the uterine or ovarian level is extremely rare, there is a limited number of cases reported in the literature [1,2], which makes preoperative diagnosis difficult.

We will describe the case of a 40-year-old woman who presented with a huge ovarian mass with an ultrasounds appearance of a degenerating cystic fibroid.

OBSERVATION :

A 40-year-old, married, nulliparous patient presented with progressive pelvic pain associated with an increase in abdominal volume that had progressed for 1 year, without other associated signs.

The patient does not report taking hormonal treatment.

Pelvic ultrasound objectified the presence of a predominantly solid double-component latero-uterine and retro-uterine image taking up the entire dopplerized screen, suggesting either a cystic degenerative fibroid or uterine sarcoma or another ovarian pathology benign. (Figure1).

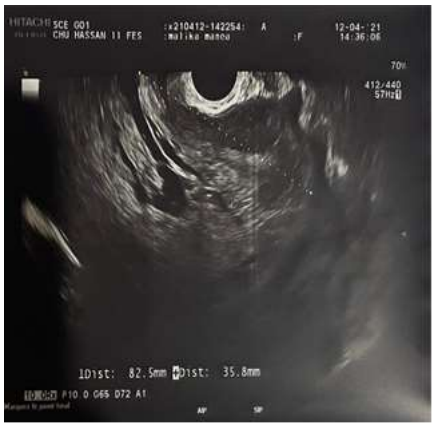

Figure 1: Double-component, predominantly solid latero-uterine and retro-uterine image taking the entire screen

The scan result showed a large intra and subperitoneal abdomino-pelvic mass of encapsulated mid-site.

Tumor markers were not requested in this case except for the negative return BHCG.

Surgical intervention is indicated on clinical and radiological criteria.

The Pfannestiel-type approach, on exploration we found no effusion; discovery of 2 polylobed masses with smooth wall without exophytic vegetations, measuring 22x13 cm long axis filling the pelvis and the abdomen presenting multiple intimate adhesions with the uterus and the right appendix and the right and left parieto-colic gutters, the left annex was unremarkable. The decision was to perform a right adnexectomy removing the 2 masses (Figure 2).

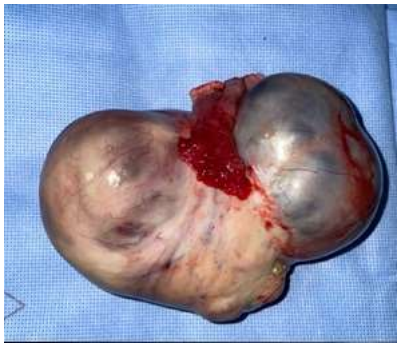

Figure 2: 2 polylobed masses with smooth wall without exophytic vegetations, measuring 22x13 cm long axis

The microscopic examination showed a double contingent tumor proliferation, a vascular contingent made up of dilated vessels with thickened walls with turgid endothelial cells and a sometimes loose mesenchymal contingent, sometimes dense in perivascular appearance, it is made of cells. Spindle-shaped with minimal cytonuclear atypia without having seen any mitosis. In addition, the ovarian parenchyma and the proboscis are free from any tumor proliferation.

An immunohistochemical study was carried out: the cells located between the vascular structures express smooth muscle actin quite markedly. It is also expressed on the cells that line the vessels. No expression of CD34. No expression of the PS100

These cells also express estrogen receptors and desmin.

No expression of HMB45, no expression of cytokeratin, no expression of inhibin, KI 67 is weakly expressed by about 2% of cells.

The histological and immunohistochemical appearance in favor of ovarian angio leiomyoma.

DISCUSSION :

Angio leiomyoma is a rare smooth muscle tumor with an endothelial vascular component [3], It usually presents as a solitary and painful subcutaneous lesion, localized in the extremities, most often in the lower limbs The review of the literature showed that Angio leiomyoma is rarely localized in the genitals in women, only a few cases of ovarian localization have been reported [3–6].

Characteristic features are the presence of spindle-shaped smooth muscle cells, numerous thick-walled arteriole-like vessels, and a swirl of smooth muscle cells around the vessels [7,8].

In our case, we were able to find some of these characteristics such as the presence of dilated vessels with a thickened wall with turgid endothelial cells. The presence of spindle cells at the level of the mesenchymal component.

The main differential diagnoses are other benign uterine leiomyomas with prominent blood vessels; the term angioleiomyoma should be reserved for lesions exhibiting the typical characteristics that we have previously described.

Our results were similar to those described in cases in the literature concerning the histological examination.Many thick-walled and proliferated vessels were found as well as special stains for smooth muscle cells, such as actin and desmin, clearly represented

the bundles of smooth muscles. CD31 and CD34, on the other hand, demonstrated endothelial cells on the vessel walls. These special stains are useful in differentiating ALM from other spindle cell neoplasms, such as fibro-thecomas, cell fibroids, sclerosing stromal tumors, angiofibromas, fibroids, and angiomyofibroblastomas. The absence of CD31-positive stromal cells within the tumor is also associated with the characteristic morphology [9]

The pathogenesis of ALM is not yet conclusive. Additional karyotypic changes have been reported [10]. Some studies suggest that ALM is hormone-dependent because it occurs more often in women, and the results show that ALM is immunoreactive with progesterone receptors but not estrogen receptors [11]. These findings are not, however, consistent with those in the present case. Our case was positive for estrogen receptors only in the immunohistochemical study.

Although it is very rare, a diagnosis of ALM should be considered in a patient with a hypervascular tumor. In particular, the preoperative diagnosis of angioleiomyoma is rarely possible, and it usually cannot be differentiated from malignant tumors [2]. Differential diagnosis on the basis of radiological features is difficult and histopathological examination is essential [2]. In one case by Hsu et al., Ultrasound was probably the most convenient and non-invasive diagnostic tool [1]. Strong blood flow observed on color Doppler indicated the presence of abundant vessels among the tumor]. Magnetic resonance imaging and computed tomography can also be useful for differential diagnoses.

Management must be surgical, and careful to avoid the risk of intraoperative bleeding

CONCLUSION :

We have emphasized the importance of histological and immunological examinations in the diagnosis of ALM of ovarian origin and we believe that angioleiomyoma should be recognized and classified as a benign variant of leiomyoma.

Origin of the aid: This work is the fruit of my own work without outside help in the form of a grant, material, medicine or other support.

Declaration of absence of conflict of interest: I declare on my honor, as well as all the participants in this study, that we have no affiliation (financial or otherwise) to disclose, with a for-profit or non-profit organization that can influence the results and analysis of this study.

Authors' contribution:

-Mochtari Houda (main author): Planning of the study, exploitation of the archives, analysis of the results and drafting of the manuscript.
-Alami Meryem: Use of archives and analysis of results
-K. Saoud, N. Mamouni, S. Errarhay, C. Bouchikhi, A. Banani: Critical review and final approval.

Number of figures: 2 figures

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Figure 2: 2 polylobed masses with smooth wall without exophytic vegetations, measuring 22x13 cm long axis

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