

CASE REPORT

A 22nd case report of extrauterine adenomyoma of the abdominal wall

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ABSTRACT

An adenomyoma of the uterus is a nodular tumor-like mass of benign endometrial glands, endometrioid stroma and smooth muscle tissue. Extra-uterine adenomyoma is an extremely rare tumor. The majority of the cases described was from ovary and located in the pelvis. We present a case of a 70 years old woman with clinical and radiological suspicious of disseminated malignancy. In the abdominal wall, a 5-cm nodule of larger dimension detected by computed tomography-scan was biopsied for accessibility reasons. The morphological and immunohistochemical features of the biopsy led us to propose the diagnosis of an extrauterine adenomyoma. This article reports the 22nd case of extrauterine adenomyoma, a rare and poorly understood tumor that could be imagiologically and clinically indistinguishable from a metastasis.

Key Words: Adenomyoma, Extrauterine, Uterus like mass, Abdominal wall

1. INTRODUCTION

An adenomyoma of the uterus is a nodular tumor-like mass composed of benign endometrium and smooth muscle. Histologically is a nodular aggregate of benign endometrial glands surrounded by endometrial stroma, with smooth muscle bundles bordering the endometrial stroma. Extra-uterine adenomyoma is an extremely rare tumor. Cozzutto^[1] published the first case in 1981, from then until now and to the best of our knowledge, 21 cases were reported in the literature, the search was carried out using the words “extrauterine adenomyoma” in the PubMed. The pathogenesis of this tumor remains unknown. There are some possible theories to explain the histogenesis of the adenomyoma. One possible explanation is a defect in the Mullerian duct fusion, an abnormality during the formation of the genital tract that

is supported by the presence of concurrent congenital defects in the uterine and renal system reported in several cases. Other possible explanations are metaplasia or heterotopia. The metaplastic theory is based on the fact that uterus-like masses can arise from subperitoneal mesenchymal cells, retaining the ability to duplicate the Mullerian duct structures.

Of the 21 cases of extra-uterine adenomyoma/uterine-like masses reported, all were women and 11 were located in the ovaries or ovarian ligaments. The other cases involved multiple sites like medullary cone, liver, bowel, vaginal cuff and pelvic region (see Table 1).^[1-19]

We report a case of a 70 years old woman with an extra-uterine adenomyoma of the abdominal wall in the context of disseminated malignant disease.

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Table 1. Published extrauterine adenomyoma cases since 1981 to our case

Cases No. (reference)	Age	Location
1 (1)	31	Left ovary
2 (2)	18	Right ovary
3 (3)	12	Small bowel
4 (4)	38	Right ovary
5 (5)	18	Medullary cone
6 (6)	49	Right ovary
7 (7)	46	Right broad ligament
8 (8)	38	Left ovary
9 (8)	43	Ovary (side unknown)
10 (8)	39	Left ovary
11 (9)	59	Small bowel mesentery
12 (10)	50	Vaginal cuff
13 (11)	11	Right ovary
14 (12)	42	Periadnexa
15 (13)	57	Left ovarian ligament
16 (14)	47	Abdomino-pelvic region
17 (15)	39	Adjacent to the descending colon
18 (16)	41	Pelvic mass
19 (17)	56	Ovarian ligament
20 (18)	29	Liver
21 (19)	47	Liver
22 (present case)	70	Abdominal wall

2. CASE REPORT

We present a case of a 70 years old caucasian woman that came to the emergency room with back pain complaints. The CT scan revealed a well-delimited 5-cm greatest dimension nodule of the abdominal wall (see Figure 1), lymphadenopathies, liver nodules and a pathologic vertebral body fracture in the dorsal spine. The CT scan also detected another 14-cm uterine related mass located in anterior sacral region, suggesting leiomyoma. Due to clinical suspicious of malignancy and accessibility, abdominal wall tumor was biopsied for diagnostic purposes. An ultrasound core biopsy of the tumor was performed and three elongated fragments between 0.5-cm and 1.5-cm were obtained.

Pathological findings

The histologic analysis revealed three different components of the lesion, all of them without atypia, mitotic figures or necrosis. The main component was smooth muscle type with scattered stroma around glands with endometrial features (see Figure 2). Smooth muscle type component was positive to desmin and smooth muscle actin (SMA) antibodies. Positivity to SMA and CD10 antibodies in endometrial like stromal component was observed too but desmin was negative (see Figure 3). Nuclear positivity to estrogen receptors (ER) and progesterone receptors (PR) was observed in all components (see Figure 4). The morphological and immuno-

histochemical features led us to propose the diagnosis of extrauterine adenomyoma.

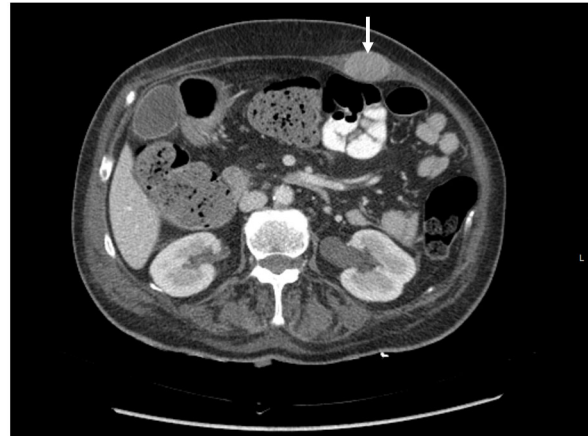


Figure 1. Contrast-enhanced Multidetector Computer Tomography image shows a solid mass within the left rectus abdominis muscle, with smooth outline, and homogenous internal enhancement

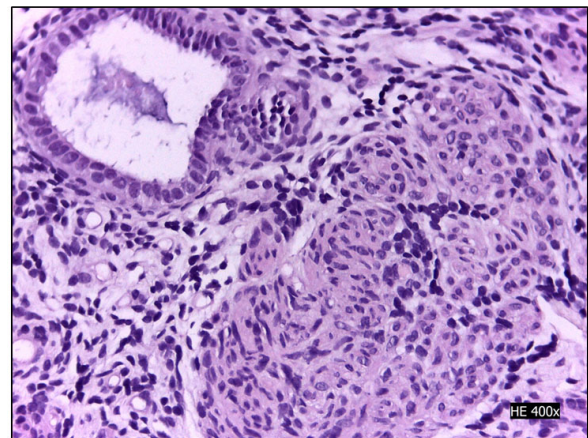


Figure 2. Hematoxylin and eosin image shows glandular proliferation admixed with endometrial stroma and smooth muscle

Immunohistochemical analysis was made in BenchMarch Ultra Ventana (Roche) platform with OptiView DAB IHC Detection Kit ABP. Primary antibodies used in the analysis included the following: ER (Novocastra 6F11), PR (Novocastra PGR), CD10 (Cell Marque 56C6), SMA (Novocastra NCL) and desmin (Dako D33).

3. DISCUSSION

We classified the lesion of our patient as extrauterine adenomyoma because it was a well-defined mass consisting of endometrioid gland, endometrioid stroma and smooth muscle tissue. These structures were well distinguished in immunohistochemical study. The present finding is the first to our

knowledge that shows extrauterine adenomyoma located in the abdominal wall.

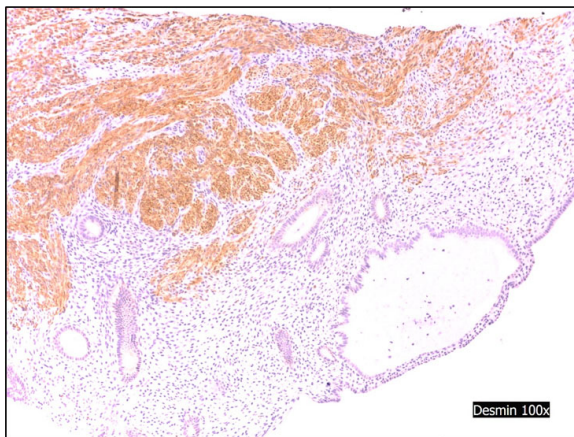


Figure 3. Desmin immunoreactivity in smooth muscle

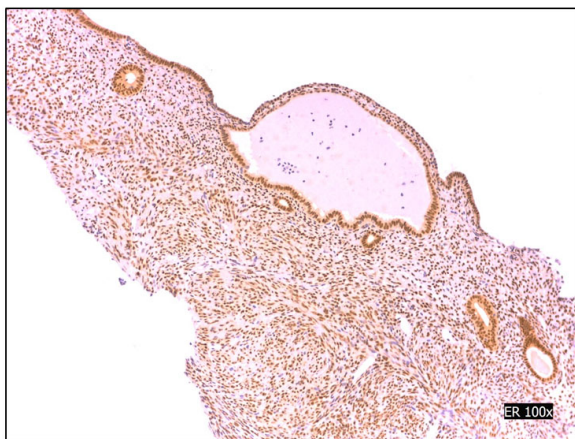


Figure 4. Nuclear positivity for estrogen receptors in the glandular epithelium and stroma

On the basis of what was published in the literature the differential diagnosis of this disease was consistent with endometriosis with smooth muscle component, or leiomyoma associated endometriosis or uterus like masses or extrauterine benign metastatic leiomyoma.

Endometriosis consists of endometrium tissue outside the uterus, in some cases admixed with smooth muscle component. The smooth muscle component could be due to adjacent smooth muscle hyperplasia/hypertrophy in continuity with endometriosis lesion or due to metaplastic changes in the endometrial stroma.^[3] The lesion reported was located in skeletal striated muscle and had a dominant smooth muscle component, which is also against a metaplastic change that is usually focal and constitutes the minor component.

Uterus-like mass lesions typically exhibit an organoid arrangement consisting of a single central cavity lined by en-

dometrial type mucosa, surrounded by a wall of smooth muscle, which resemble a uterus morphology and represent a specific form of extrauterine adenomyoma.^[3] The lesion described in this report showed multifocal glandular and stromal elements within disorganized smooth muscle. According to immunohistochemical staining results (ER/PR positive) and the clinical history of a uterus mass that imagiologically resembled a leiomyoma, this lesion could be a benign metastasizing leiomyoma, however it most frequently involves the lungs and our case has endometrioid tissue.

Regarding to pathogenesis, in this case the patient had no structural uterine abnormality consistent with Mullerian fusion defect, she had a normal anatomic reproductive system and no renal abnormality. It is most likely that this adenomyoma of the abdominal wall arose from the tissues of the secondary Mullerian system, which was derived from the subcoelomic mesenchyme.

The persistence of clinical suspicious of malignancy led the clinicians to perform a vertebral mass biopsy. The diagnosis rendered in histopathology report was melanoma. Despite this abdominal lesion didn't correspond to melanoma metastasis, the patient died after two months of melanoma progression.

Extrauterine adenomyoma is described in the literature as a benign tumor, the most of clinical symptoms were related with the mass effect of the tumor. The management of the majority of the cases was surgery with no complications reported.

4. CONCLUSION

In this article, we report the 22nd case of extrauterine adenomyoma, a very rare and poorly understood tumor that could be imagiologically and clinically indistinguishable of a metastasis.

DISCLOSURE

A part of this manuscript was accepted as a poster presentation to the 25th European Congress of Pathology, held in Lisbon, Portugal, on 31 August – 4 September 2013.^[20]

ETHICAL APPROVAL

This work was approved by the appropriate ethics committee from Centro Hospitalar do Porto. Therefore, it has been performed in accordance with the ethical standards laid of Helsinki Declaration.

COMPLIANCE WITH ETHICAL STANDARDS

This research had no financial support. The authors declare having no conflicts of interest.

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