

CASE REPORT

Adenomatoid Tumor of Myometrium: Report of Three Cases

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INTRODUCTION

The adenomatoids tumors are quite uncommon benign neoplasm, whose more frequent location is the genital tract of both genders. In women, the myometrium is the most frequent topography, usually bringing diagnostic doubts since they share macroscopic characteristics with uterine leiomyomas, and susceptible to be confused with malignant neoplasms at the microscopy.^{1,2} In this article, we present the morphological aspects of three cases whose histopathological diagnoses were performed at our healthcare service.

REPORT OF CASES

CASE 1

Forty-five year-old patient, white, female submitted, in other healthcare unit, to total hysterectomy with clinical diagnosis of uterine myomas and metrorrhage. Macroscopically, it had presented four intramural nodules, the largest measuring 2.5cm.

Under the microscopy, three of the nodules possessed compatible histology with leiomyomas. One of the nodules was composed by cystic structures with size variation, around smooth muscle bundles, without evident capsule. The cystic structures were coated by predominantly flat, occasionally cuboidal, epithelium, with discrete and focal nuclear atypias. Sometimes it was observed amorphous basophilic content inside the cysts, as well as cytoplasmic vacuolization. There were no stromal reaction or inflammatory process.

Immunohistochemical study was positive for pancytokeratin AE1/AE3, cytokeratin 7 and calretinin in the cystic covering epithelium, and negative for vascular markers (CD31 and CD 34), Ber-EP4, EMU and other cytokeratins. The smooth muscle alfa actin was positive in smooth muscle fibers around the cystic structures. Starting from the morphological and immunohistochemical characteristics, the diagnosis of adenomatoid tumor of the myometrium was made.

CASE 2

Eighty-two year-old patient, white, female was submitted, at our service, to total hysterectomy due to endometrial thickening and intermittent vaginal bleeding. Macroscopically, there were several intramural and submucous nodular formations measuring up to 1.7cm, besides endometrial thickening and polyps.

The diagnosis for leiomyomas was made, however the microscopic aspect of one of the nodules was similar to the described in the CASE 1, although with smaller variation in the size of the cystic formations. It was still made the diagnosis atypical endometrial hyperplasia. Based on the morphological discoveries of the above mentioned nodule, it was made the diagnosis of adenomatoid tumor of the myometrium.

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CASE 3

Thirty-five year-old patient, white, female, with clinical history of metrorrhage, irregular menstrual cycles and anemia; image exams suggestive of uterine myomas. Submitted in our service to hysterectomy. The surgical specimen showed 2 intramural well-delimited nodules, measuring 0.3 and 1.0 cm. Microscopically, the aspect of the two nodules was similar to the other two cases, exhibiting cystic formations of varied sizes intermixed by smooth muscle bundles, being covered by flat or cuboidal cells and occasionally filled with amorphous basophilic material. The immunohistochemistry was intensely positive for calretinin and pancytokeratin in the cystic formations, and negative for CD34. It was made the diagnosis of adenomatoid tumor of the myometrium.

DISCUSSION

Adenomatoids tumors are benign neoplasms of mesothelial origin, usually found in both female and male genital tracts, and less frequent in other locations (peritoneal and pleural surface, adrenal glands). In the female genital tract, the most frequent location is the uterus (myometrium), followed by uterine tubes and ovarian hilum. In male, they have more frequently occurred in the epididymium, spermatic string, tunic albuginea and prostate.¹

The adenomatoid tumors of the myometrium frequently make differential diagnosis with leiomyomas, by presenting similar macroscopic characteristics. In general, they are found in surgical pieces of patient submitted to hysterectomy by myomas, and can be incidental findings of hysterectomies performed by other indications.¹⁻³

They are usually located in the subserosa and serosa layers in the myometrium, however they can be found in its superficial portions, and occasionally represented histologically in uterine curettage specimens.¹ In hysterectomy specimens, they are usually isolated nodular, intramural, non-encapsulated formations, with partially defined limits with the adjacent myometrium, measuring from 0.2 to 3.5 cm, rarely multiple.^{1,3,4} There is description of tumors reaching great dimensions and multicentric tumors, usually associated to immunosuppression conditions, and more

frequently causing dubious diagnosis with malignant neoplasms.⁵⁻⁷

Histologically, they are constituted by cystic or tubular formations and rifts of varied sizes (Figure 1), with flat or cuboidal covering epithelium and absence of outstanding atypia or mitotic activity (Figure 2). In agreement with the size and proportion of these structures, they can acquire solid-like aspect.^{1,2,4,8,9} Occasionally, they are filled out by basophilic material, and can present cytoplasmic vacuolization giving the cells aspect of "signet ring".^{2,3,9} There is proliferation of smooth muscle fibers around the formations, probably of reactional origin, and focuses of inflammatory infiltrate can be observed in varied degrees.¹ Rare variations of larger diameter (7cm to 11cm) and multicystic aspect are described with possibilities of papilla and vesicle surface (in "grape bunch"), however maintaining the benign character.¹ It can coexist with malignant lesions of the genital tract, and in cases of endometrium adenocarcinoma, the adenomatoid tumor can be confused with myometrial invasion.¹⁰

Ultrastructural, histochemical, and immunohistochemical studies have demonstrated the mesothelial origin of the neoplasm. At the electron microscopy, microvilli similar to the ones found in the surface of mesothelial cells were visualized, as well as characteristic desmosomes.^{3,4,8,9} The presence of mucin containing hyaluronic acid can also be demonstrated in adenomatoid tumors through

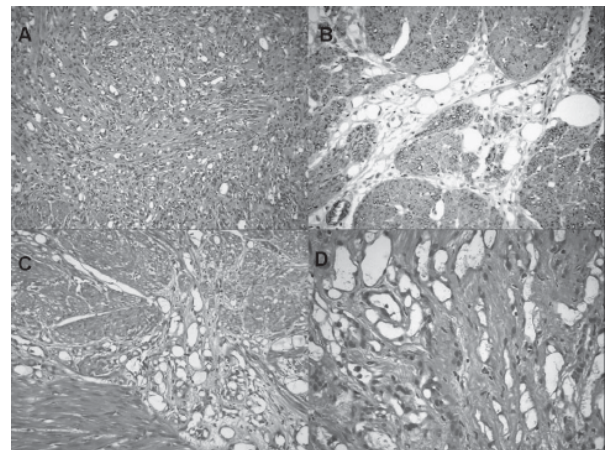


Figure 1 - A and B: Formations of cystic aspect among smooth muscle fibers (A - staining HE, 100X; muscle fibers marked by alpha actin of smooth muscle. In B, immunohistochemistry, 100X). C and D: Size variation among cystic formations, absence of infiltrative aspect or estromal reaction (HE 200X)

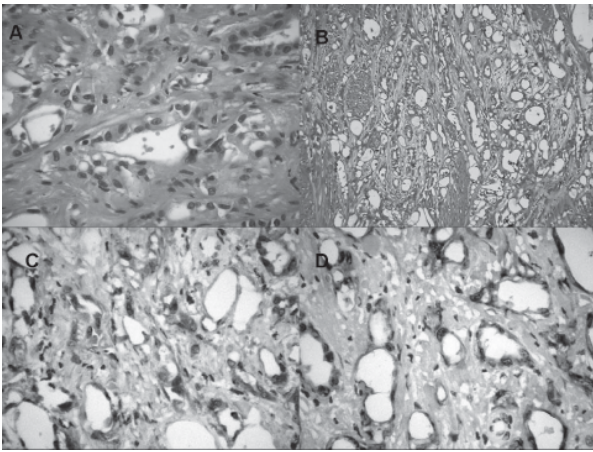


Figure 2 - A and B: cystic formations covered by plane or cuboidal cells, without cytological atypia, with occasional vacuolization (aspect of cells in signet ring) and basophilic material under the light microscope (HE, 200X and 100X, respectively). C: Immunohistochemical positive for calretinin (200X). D: Immunohistochemical positive for pan-cytokeratin (200X)

specific staining (Alcian Blue), as well in mesothelial normal cells ^{3,9}.

The adenomatoid tumors are usually positive for calretinin, HMBE-1, WT-1 and low molecular weight cytokeratins, and negative for Ber-EP4, EMU and vascular markers. Such immunohistochemical profile helps to clarify doubtful diagnosis in differential diagnosis with lymphangioma and metastatic carcinoma.^{1,2,11,12}

In conclusion, the adenomatoids tumors of the myometrium represent a less frequent entity, however benign, with no report of metastasis. It is a neoplasm of mesothelial origin (probably originated by the invagination of the coelomic epithelium during the development of the mullerian ducts), and its recognition is important in the differential diagnosis with other lesions of the uterine body, including malignant neoplasms.

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