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Histopathology

ADENOMATOID TUMOR OF THE UTERUS: A RARE CASE REPORT

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ABSTRACT Adenomatoid tumors are rare benign tumors of mesothelial origin having a unique histology. These tumors have a very low incidence. They can be accidentally found in the uterus and fallopian tube and may resemble leiomyomas on gross and histological examination. They may also be present in the peritoneal cavity and other extragenital sites. Adenomatoid tumors exhibit different patterns on histological examination that can sometimes lead to an inaccurate diagnosis. An accurate diagnosis may be made by understanding the tumor's diverse histomorphological patterns and unusual appearances.

KEYWORDS : Adenomatoid tumor, cornual mass

INTRODUCTION

Adenomatoid tumors are unusual neoplasms that are benign and have a mesothelial origin. These tumors affect both male and female sexes, generally involving the genital tracts, and can be seldom seen at the extragonadal sites.[1,8] They are often discovered incidentally in pathology specimens operated on for a different cause and may resemble leiomyomas on both gross and histological examination.[8] It has been documented that uterine leiomyomas coexist with as many as 60% of these tumors.[7]

"Sakaguchi first documented it as an adenomyomata in 1916.[5] It was later in 1942 when the mesothelial origin of these tumors was confirmed, and they were first described as 'benign mesothelioma of the genital tract,' by Masson et al.[5] but it was only after 1945 that the term 'adenomatoid tumor' was used by Golden and Ash for these benign, typically well-circumscribed, and often incidental tumors that were mesothelial in origin."[3]

These tumors can occasionally have peculiar histomorphological appearances, necessitating a distinction from cancerous tumors.[7]

Case Presentation

A 60-year-old woman came to the gynecology department with the chief complaint of postmenopausal bleeding for 2 years. After a thorough workup, two fibroids were seen in the ultrasonography. The patient opted for surgical treatment, so a hysterectomy was done. The specimen was received for histological examination in our department.

On gross examination, the hysterectomy specimen measured 5x3x1 cm. The endometrial cavity was patent. Myometrium revealed two fibroids; one was intramural, measuring 1x0.5 cm, and the other one was located at the cornua, measuring 1x1 cm. The cut surface of both the fibroids was homogeneous white. Cervix and bilateral adnexa were grossly unremarkable (Figure 1).

Hematoxylin and eosin-stained sections from the endometrium showed atrophic changes, and sections from the intramural fibroid showed classical features of leiomyoma. Hematoxylin and eosin-stained sections from cornual mass revealed a variable combination of slit-like tubular branching spaces interdigitating between smooth muscle bundles (Figure 2 and 3). The cells lining the tubules were eosinophilic and cuboidal to flattened, mild lymphocytic infiltrate was also seen. Pleomorphism, nuclear hyperchromasia, mitosis, or necrosis were absent. The surrounding myometrium did not exhibit any evidence of invasion. The cervix showed features of chronic cervicitis, and the histology of the bilateral adnexa was unremarkable. A provisional diagnosis of adenomatoid tumor for the cornual mass was given, and the patient was advised IHC for confirmation.

IHC staining for calretinin was positive, thus confirming our diagnosis (Figure 4).

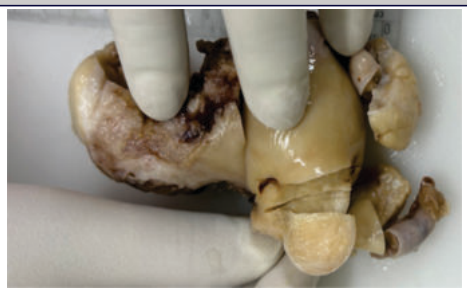

Figure 1: Gross Photograph of a Case of Adenomatoid Tumor

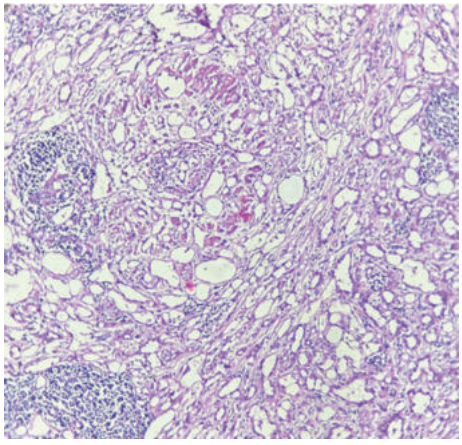

Figure 2: Photomicrograph(10x) Showing Slit-like Spaces

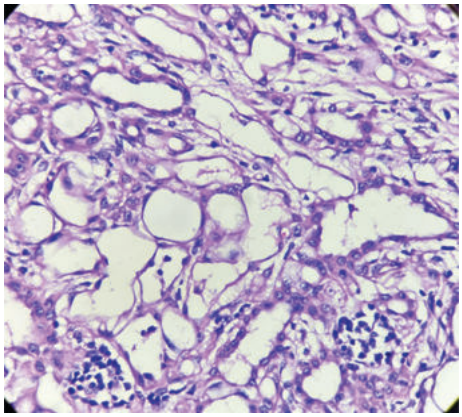

Figure 3: Photomicrograph(40x) Showing Slit-like Spaces with Mild Lymphocytic Infiltrate.

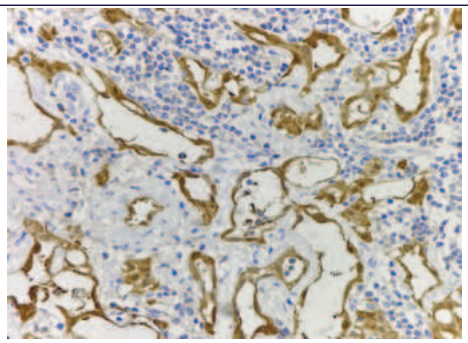

Figure 4: Photomicrograph(40x) Showing Calretinin Positivity

DISCUSSION

These mesothelial-derived adenomatoid tumors are uncommon neoplasms with an incidence of about 1% in hysterectomy specimens.[5] Due to their mesothelial origin, they occur in organs that are close to surfaces that are lined by mesothelium, the most common being the genital tracts, followed by extra genital sites like the adrenal gland, the liver, the pleura, the peritoneum, and the mediastinum.[5,6]

In males, these tumors may involve the spermatic cord, epididymis, ejaculatory duct, prostate, and tunica albuginea. In females, these may affect the fallopian tubes, uterus, and the hilum of ovaries.[1] In the case of the adenomatoid tumor of the uterus and fallopian tubes, the median age reported is 44 years and 56 years, respectively.[1] These tumors have an average diameter of 2.1cm, and their size often varies from 2 to 10 cm.[7]

There are no explicit signs and symptoms ascribed to these tumors.[7] These tumors are coincidentally found in specimens removed due to another cause.[8] There are no instances of recurrence, conversion to malignancy, or metastases that have been recorded. These tumors can either be straightforwardly excised or a hysterectomy can be done, which can pose both diagnostic and therapeutic purposes. The negative hormonal markers in these tumors make hormone therapy unfit as an appropriate treatment.[4] Similarly, in our patient, the neoplasm was found in the cornua of the uterus that was surgically removed due to the complaint of post-menopausal bleeding.

This entity is often overlooked because it has a morphological overlap with leiomyoma, and there is generally a lack of awareness among histopathologists.[5] They can have various histological patterns, like adenoid, angiomatoid, glandular, solid, and tubular.[3,8] They are often solitary,[7] but various authors have also reported a few cases of large, multicystic, papillary, and multiple tumors.[6] These tumors have an extensive differential diagnosis, which can pose a diagnostic challenge, like angiosarcoma, malignant mesothelioma, lymphangioma, primary adenocarcinoma, hemangioma, metastatic adenocarcinoma, and yolk sac tumor.[5]

The etiology of the adenomatoid tumor is unknown, and the pathogenesis is presumed to be due to TRAF7 gene somatic missense mutations akin to well-differentiated papillary mesothelial tumors.[2] Adenomatoid tumors demonstrate intact/retained nuclear expression of BAP1 and uniformly lack CDKN2A, BAP1, and NF2 deletions or mutations that characterize the majority of mesotheliomas.[2]

Although immunohistochemistry is not compulsory for the diagnosis of the adenomatoid tumor, it supports their mesothelial origin and also facilitates differentiation from adenocarcinomas. The tumor cells express WT1, calretinin, D2-40, cytokeratin AE1/AE3, and CAM5.2.[1]

In summary, this case report sheds light on the histomorphological features of an extremely infrequent tumor. These tumors are frequently neglected due to a relative dearth of understanding, familiar differential diagnoses, and low incidence. Awareness of this entity is vital for precise diagnosis, along with recognizing typical histological features backed up by specific immunohistochemical stains

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