CASE REPORT

Multicentric Leiomyoadenomatoid Tumor of the Uterus: A Rare and Distinct Morphological Entity

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Introduction

Adenomatoid tumors are benign mesothelial tumors that are usually present in fallopian tubes in females and epididymis in males. Rarely they are seen as incidental finding in uterine myometrium, when uterus is removed for various other indications. These have also been described in extragenital sites such as liver, heart, adrenal gland, mediastinal lymph node, pleura, and pancreas. We describe a rare case of multicentric leiomyoadenomatoid tumors of uterus presenting as incidental finding in conjunction with leiomyomas.

Case History

A female 65 year old, menopausal for last 15 years, visited hospital with history of bleeding per vagina since 3 months. Ultrasound revealed multiple fibroids. Total hysterectomy and bilateral salpingo-oophorectomy was done. Uterus with cervix measured 11 × 6 × 5 cm. Cut surface of these nodular masses showed whorled

appearance. The gross appearance was consistent with multiple fibroids measuring 3.5, 3, 1.5, and 0.5 cm.

Sections from the two larger nodular masses showed histological picture, consistent with the diagnosis of leiomyoma(s). However, sections from the other smaller nodular lesions revealed cuboidal to flattened cells lining and surrounding tubular formations. Few cells had vacuolated or ample amphophilic cytoplasm resulting in an epithelioid appearance. Nuclear pleomorphism and mitosis were not seen in these cells. These cells were closely intermixed with smooth muscle bundles and fibers (Fig. 1a).

Immunohistochemistry studies showed positive staining for calretinin in these cells (Fig. 1b). Staining for CEA was negative. CD34 highlighted the rich vascularity but tumor cells were negative. Staining by Ki67 did not show proliferation in these cells. Overall features were diagnostic of leiomyoadenomatoid tumor.

Discussion

The term leiomyoadenomatoid tumor was first described by Epstein in 1992 as a variant of adenomatoid tumor with a prominent smooth muscle component [1]. Cystic adenomatoid tumors and multicentric tumors have also been reported in uterus.

In our case, positive staining with calretinin was consistent with an adenomatoid tumor and ruled out an epithelioid leiomyoma. Negative staining with CD34 and CEA and lack of proliferation ruled out epithelioid

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Fig. 1 **a** Cuboidal to flattened cells lining tubular spaces along with cells with abundant amphophilic cytoplasm surrounding these tubular spaces. Myometrial fibers are present in between (H&E, $\times 200$). **b** Tumor cells show positive staining with calretinin (IHC, $\times 200$)

hemangioendothelioma and metastatic carcinoma. The presence of adenomatoid component intermixed with muscle fibers was diagnostic of leiomyoadenomatoid tumors of myometrium in this case.

Histologically these tumors have been classified into four types: adenoid or tubular, angiomatoid or canalicular, solid or plexiform, and cystic. Our case had mixed tubular and angiomatoid morphology.

These tumors can be mistaken for leiomyoma(s) with degenerative changes, epithelioid hemangioendothelioma, and a metastatic adenocarcinoma. Immunohistochemistry studies help in differentiating these lesions and reaching a precise diagnosis.

The histogenesis of these tumors is debated. Immunoprofile of these lesions in view of calretinin and HMBE-I (anti-human mesothelial antibody) positivity is consistent with a mesothelial origin. Endothelial origin, and mullerian origin with single elements and hamartomatous etiology have also been considered but not supported. As

adenomatoid component is intermixed with smooth muscle in leiomyoadenomatoid tumor, a suggestion has been made that this tumor is a variant of adenomatoid tumor that originated in precursor cells with dual differentiation, i.e., mesothelial and muscle cells [2]. However, most of the reports favor hyperplasia of entrapped smooth muscle by adenomatoid tumor rather than dual differentiation in this tumor.

References

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