

Case Report

Multicentric Infarcted Leiomyoadenomatoid Tumor: A Case Report

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Abstract: Adenomatoid tumor is a benign, usually small lesion that may be found within the wall of fallopian tubes or beneath the uterine serosa near the uterine cornu. It is often accompanied by smooth muscle hypertrophy that may obscure the adenomatoid tumor. We herein report a very unusual case of infarcted leiomyoadenomatoid tumor of the uterus and ovary in a 24-year-old woman who presented with severe lower abdominal pain and masses in the uterus and right ovary. Pelvic ultrasonography and computed tomography revealed a 5 cm mass in the myometrium and a 4 cm mass in the right ovary. Laparoscopy-assisted transvaginal mass removal was performed under the clinical impression of a uterine leiomyoma and benign ovarian teratoma. On a microscopic examination, prominent fascicles of smooth muscle separated or infiltrated by cuboidal or signet ring-like vacuolated cells, as well as tubular formations lined by flattened mesothelial cells and extensive necrosis were observed in both masses. The microscopic appearance often suggested the possibility of a malignant neoplasm due to irregular pseudoinfiltration with atypical cuboidal cells and the paucity of a typical adenomatoid tumor due to infarction, and the presence of epithelial-appearing cells in the hypertrophic smooth muscle bundles that mimicked an infiltrating carcinoma for a leiomyoma or myometrium. These unemphasized features of leiomyoadenomatoid tumors may potentially lead to more aggressive therapy than warranted if not correctly interpreted, especially for infarcted cases.

Key Words: Leiomyoadenomatoid tumor, uterus, ovary, infarction

Introduction

Adenomatoid tumors are benign neoplasms of mesothelial origin that occur most frequently in the myometrium or fallopian tubes in females, and the epididymis in males [1-3]. However, rare cases have been reported where the tumor is in or adjacent to the hilum of the ovary [4]. Uterine adenomatoid tumors, in particular the intramural type, are often accompanied by smooth muscle hypertrophy, which usually is represented by an entrapped myometrium permeated by the adjacent tumor [5]. Uncommonly, adenomatoid tumors accompany findings of infarction, namely, extensive or focal necrosis with associated reactive changes that impart an appearance that suggests a malignant process [6]. When the smooth muscle component is as prominent as the tumor component within

the adenomatoid tumor, the lesion is denoted as a 'leiomyoadenomatoid tumor' [7]. We experienced a case of leiomyoadenomatoid tumor in a 24-year-old woman. The patient had a mass in the uterus and a mass in the right ovary. Both masses had the morphological appearance of adenomatoid tumors with excessive smooth muscle hypertrophy and coagulation necrosis. These findings were also suggestive of an infiltrating adenocarcinoma, which made the diagnosis difficult. We herein report this rare case of leiomyoadenomatoid tumor involving both uterus and right ovary, and discuss its differential diagnosis.

Case report

A 24-year-old woman presented with severe lower abdominal pain relieved by analgesics. The patient usually had menorrhagia and



Figure 1 Radiological findings. A pelvic CT scan demonstrated a 5 cm uterine mass (upper panel, asterisk) pushing on the uterine cavity and a 4 cm right ovarian mass (lower panel, triangle) which showed a similar appearance of a round and low-density masses with mild heterogeneity within the lesions. In this level, the right ovarian mass (triangle), uterus (open asterisk) and left ovary (arrows) were observed simultaneously.

dysmenorrhea. Pelvic ultrasonography and computed tomography revealed a 5 cm mass in the myometrium and a 4 cm right ovarian mass (**Figure 1**). The two masses showed low-density with mild heterogeneity. Under the impression that the lesions were a uterine leiomyoma and ovarian teratoma, laparoscopy-assisted transvaginal mass removal was performed. On the operative findings, the ovarian mass was clearly separated from the uterine mass and it was attached to the discernible right ovary with broad base. Ovarian mass removal with preservation of the right ovary was performed.

Grossly, the uterine mass measured 4.5 × 3.5 cm in size, and the mass was well circumscribed. The tumor had a smooth surface and hard consistency. Microscopically, the two specimens from the uterus and ovary showed very similar

histopathological features and immuno-histochemical characteristics. Prominent fascicles of smooth muscle separated or infiltrated by cuboidal or signet ring-like vacuolated cells, as well as tubular or cystic formations lined by flattened mesothelial cells were observed in both masses. There was little nuclear atypia or mitotic activity and no stromal desmoplastic response (**Figure 2**). A wide area of extensive necrosis was present. Some areas showed ghost patterns of smooth muscle bundles with cystic lesions or single cells, while other areas showed nondescriptive necrosis with no recognizable underlying pattern and hemorrhage (**Figures 3A, B and C**). In an adjacent area of necrosis, reactive changes including plump infiltration of fibroblasts and acute and chronic inflammatory cells were observed (**Figure 3D**). Based on immunohistochemical analysis, epithelial looking tumor cells demonstrated a positive reaction for cytokeratin (CK), CK 7, and calretinin (**Figures 2 and 3**), and a negative reaction for carcinoembryonic antigen (CEA). Fascicles of the spindle cells showed a positive reaction for desmin and smooth muscle actin (**Figure 2**). As we regarded the two components, i.e. the mesothelial-originated cells with cystic formation and the plump smooth muscle hypertrophy, as a balanced existence of tumor components, we diagnosed the lesions as leiomyoadenomatoid tumors accompanied by infarction arising in the uterus and ovary.

Discussion

Golden and Ash [8] first proposed the term adenomatoid tumor in 1945. The incidence of adenomatoid tumors in the uterus that have been surgically removed has been reported to be uncommon at about 1% [9]. Ovarian adenomatoid tumors are rare, although they have been occasionally described [3, 10]. Most ovarian pure adenomatoid tumors have a similar histological appearance to uterine adenomatoid tumors, although the smooth muscle component seen in the uterine examples is not a feature of the ovarian lesions [11]. The histogenesis of the adenomatoid tumor was subject to debate, but it was ultimately proven by ultrastructural and immunohistochemical studies to be a

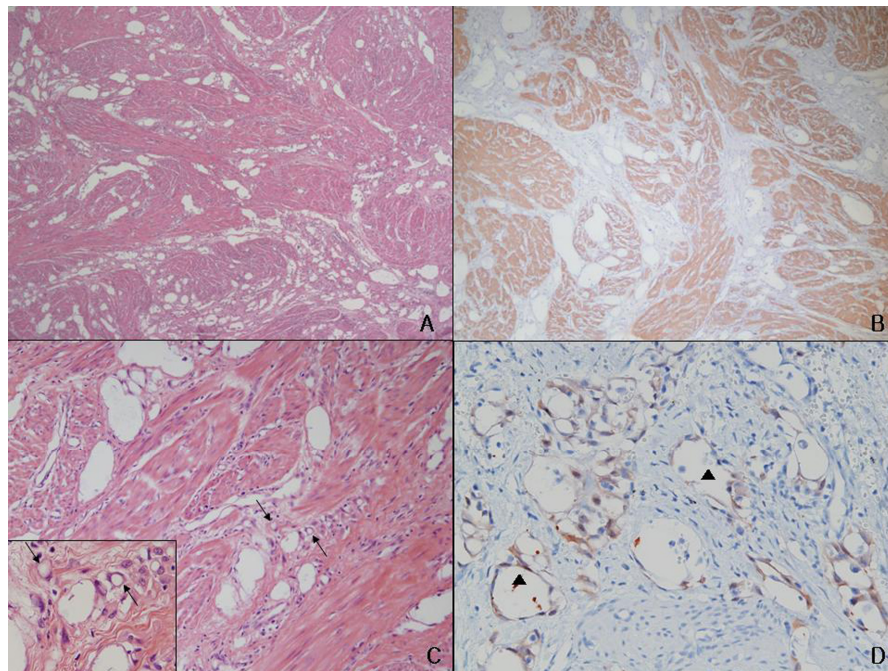


Figure 2 Histopathological findings of the non-infarcted areas. Bundles of smooth muscle with small and inconspicuous cystic spaces infiltrating stroma and smooth muscle bundles (A, H and E stain; B, Immunostain for α -smooth muscle actin). Scattered single vacuolated cells (arrows) and mesothelial cell-lined cyst (triangle) formation (C, H and E stain; D, immunostain for calretinin).

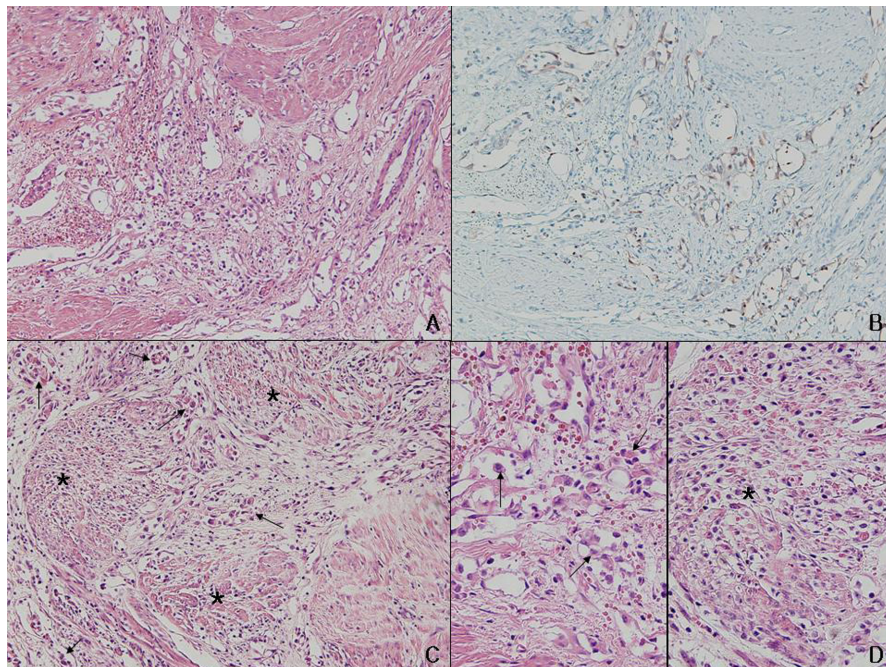


Figure 3 Histopathological findings adjacent to the infarcted areas. Degenerative changes such as mummified nuclei, ballooning, denuded mesothelial cell lining (arrows), myxoid change of stroma, residual mesothelial cell-lined cysts, and coagulative necrosis of smooth muscle bundle (asterisk) were observed (A, C and D, H and E stain; B, Immunostain for calretinin at the identical site to A. D represents higher magnification of A and C).

tumor of mesothelial origin. The tumor cells positively express CK, calretinin, anti-human mesothelioma antibody (HBME-1), and vimentin, and do not express epithelial membranous antigen (EMA) and CEA [12, 13]. In the present case, based on immunohistochemical analysis, expression of CK and calretinin was strongly positive, but vimentin was not expressed.

The majority of adenomatoid tumors are readily diagnosed based on location and characteristic microscopic features [1-3]. However, various issues may cause diagnostic difficulty. For example, as in the present case, an adenomatoid tumor with a prominent smooth muscle component may be mistaken for a leiomyoma. More important is the possibility of tumors with a solid growth pattern or cords of cells being mistaken for an infiltrating malignant epithelial or mesothelial neoplasm, or those with small vacuoles being confused with a signet ring cell adenocarcinoma [6].

The term 'leiomyoadenomatoid tumor' was first described by Epstein [7], which describes a variant of adenomatoid tumor with a prominent smooth muscle component. Sometimes, the smooth muscle overgrowth is so extensive that it obscures the adenomatoid tumor [5], and results in the tumor being misdiagnosed as a leiomyoma or malignant tumor infiltrating smooth muscle bundles. Smooth muscle bundles usually represent an entrapped myometrium permeated by the adenomatoid tumors, or reactive hyperplasia of the indigenous myometrial smooth muscle [5]. However, some investigators have postulated that smooth muscle bundles represent a neoplastic component [3], which claim may explain the presence of a leiomyoadenomatoid tumor at the sites, and not at an anatomic site associated with prominent smooth muscle, such as an ovarian mass.

In addition, another diagnostic problem, as described in the present case, is extensive coagulation necrosis due to tumor infarction. The cause of the infarction remains speculative. One could speculate that larger tumors may be more susceptible to infarction [6], but the size of the tumors in the present case was similar to that of typical adenomatoid tumors. Such infarcted

leiomyoadenomatoid tumors have a microscopic appearance that often suggests the possibility of a malignant neoplasm, such as irregular pseudo-infiltration of smooth muscle by an adenomatoid tumor that mimics an infiltrating carcinoma in a leiomyoma or the myometrium and the paucity of a typical adenomatoid tumor due to the infarction [6]. The present case also described the infarcted type of leiomyoadenomatoid tumor that is suggestive of a malignancy, resulting in confusion in the diagnosis.

In summary, we have described adenomatoid tumors showing neoplastic hypertrophied smooth muscle bundles and extensive necrosis consistent with infarction, with characteristic features that may cause diagnostic difficulty. Awareness of this disease entity (infarcted leiomyoadenomatoid tumors) will result in a correct diagnosis.

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