



Leiomyoadenomatoid Tumor of the Uterus: A Case Report of a Rare Entity

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Abstract

Adenomatoid tumor is a benign neoplasm that occurs frequently in the wall of fallopian tubes in females. Leiomyoadenomatoid tumor of the uterus is a variant of adenomatoid tumor which is extremely rare and difficult to recognize on microscopic examination. We report here a case of a 37-year-old woman who presented with vaginal bleeding during the past 6 months. Pelvic ultrasonography revealed three masses in the myometrium. Laparoscopy transvaginal masses removal was performed under the clinical impression of uterine leiomyomas. Histological examination followed by immunohistochemistry straightened the diagnosis of leiomyoma in one mass and confirmed the presence of a biphasic proliferation: an adenomatoid component, which was immunoreactive with calretinin, intermingled with interlacing fascicles of neoplastic smooth muscle cells. Here in we report the fourteenth case of leiomyoadenomatoid tumor of the uterus. Ignorance of this rare entity may lead to false diagnoses, such as a malignant epithelial or mesothelial neoplasm, resulting in a more aggressive therapy.

Keywords: Adenomatoid tumor; Leiomyoadenomatoid tumor; Uterus

Introduction

Adenomatoid tumors are benign mesothelial neoplasms that are usually present in the fallopian tube in females and epididymis in males [1]. Rarely they are seen as incidental finding in uterine myometrium, in this site they are often associated with smooth muscle hypertrophy and if this component is prominent, the lesion is denoted as a leiomyoadenomatoid tumor [1]. There are only 13 cases reported in the English literature [2-11]. We report here a new case of this rare entity presenting as incidental finding in conjunction with leiomyomas and we discuss its histological differential diagnosis.

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Received Date: 14 Oct 2019

Accepted Date: 30 Oct 2019

Published Date: 06 Nov 2019

Citation:

Makni S, Moussa A, Njima M, Njim L, Zakhama A. Leiomyoadenomatoid Tumor of the Uterus: A Case Report of a Rare Entity. *Ann Gynecol Obstetr Res.* 2019; 2(2): 1012.

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Case Presentation

A 37-year-old woman, without a significant past medical, presented with a 6 months history of irregular bleeding per vagina (menorrhagia and dysmenorrhea). Ultrasound revealed three fibroids in the myometrium. Laparoscopic transvaginal masses removal was performed. Grossly, the uterine masses were well circumscribed white with hard consistency, and measured 3, 4 and 5 cm. Sections from the 3 and 5 cm fibroids showed histological picture consistent with trivial leiomyomas. However, microscopic examination of the 4 cm mass showed prominent fascicles of smooth muscle, separated by cuboidal, flattened or signet ring-like cells, lining tubular or cystic formations. Few cells had vacuolated or amphophilic cytoplasm, but there was neither nuclear atypia nor mitosis (Figure 1 and 2). Immunohistochemistry study, showed these cells to be positive for cytokeratin (AE1/AE3) and calretinin and negative for CD31 (Figure 3). Overall features were diagnostic of leiomyoadenomatoid tumor. The outcome was favorable without any complications at the 2 years follow up evaluation.

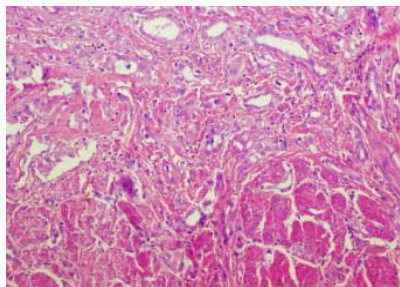
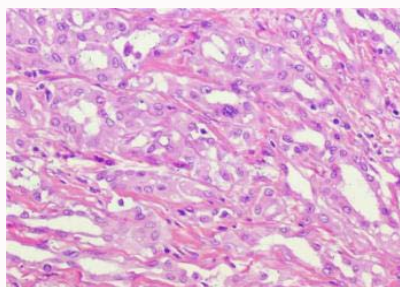
Discussion

The term leiomyoadenomatoid tumor was first described by Epstein in 1992 [1] as a variant of adenomatoid tumor with a prominent smooth muscle component. These tumors were incidental findings in hysterectomy or fibroids specimens [2]. By reviewing the literature, the mean age of diagnosis was 45 years old with extremes of 24 and 65 years old (Table 1).

The gross appearance consisted on grayish or white, well circumscribed nodules with whorled cut surface [3]. Case with gray fibrous and polycystic area containing gelatinous substance was described [4]. These nodules had an average size of 2 cm, with extremes of 0, 5 and 4 cm.

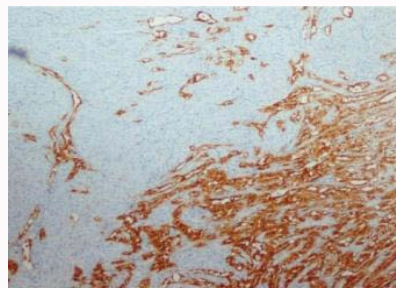
Table 1: Age and tumor size of uterine leiomyoadenomatoid tumors reported in the literature.

Author	Age	Tumor size (cm)
Amre et al. [2]	52	2.5
Hong et al. [5]	24	4
Erra et al. [7]	44	3
Mathew et al. [8]	51	3
Amérigo et al. [9]	55	2,5
Dobrosz et al. [4]	57	3,5
Pransgaard et al. [10]	24	3
Sarma et al. [3]	45	2
Bahuguna et al. [6]	65	0.5 and 1.5
	36	0.5 and 3
Ranjan et al. [11]	43	1.2
	37	4
Present case 2016	37	4

**Figure 1:** Small cystic and tubular spaces infiltrating or intermingled the Bundles of smooth muscle (H&E x100).**Figure 2:** These spaces are lined by flattened, cuboidal or signet ring-like vacuolated cells (H&E x400).

Microscopically, tumors showed a well-defined neoplastic process with a biphasic pattern: prominent fascicles of smooth muscle separated or infiltrated by an adenomatoid neoplastic component consisting on multiple anastomosing gland-like or pseudo-vascular spaces, tubules, cysts and solid growth, the spaces are lined by single layer of flattened to cuboidal cells with scant pale to eosinophilic cytoplasm that may contain vacuoles and round nuclei with small nucleoli. There is minimal cytological atypia and mitotic activity [3,5]. An extensive coagulative necrosis due to tumor infraction has been reported in one case by Hong et al. [5].

The histogenesis of this adenomatoid neoplastic component was debated, but it was ultimately proven by immunohistochemical studies to be a tumor of mesothelial origin. The cells express calretinin, HBME-1 (Hector Battifora mesothelial antigen-1 or anti human mesothelial antibody), cytokeratin (CK) 5/6, CK7, CK18, CK19,

**Figure 3:** Cell lining the cystic spaces are calretinin positive (IHC x200).

WT1, D2-40 and did not express EMA (Epithelial Membranous Antigen), CEA (Carcino-Embryonic Antigen) and vascular antigens (CD31, CD34). In our case report, tumor cells were strongly and diffusely positive for CK and calretinin, but they were negative for CD31. Fascicles of spindle cells are, as expected, positive for smooth muscle actin and desmin [3,5-7].

A possible explanation for this phenomenon is that adenomatoid component arises from inclusion of mesothelium incorporated into subserosal connective tissue or myometrium. Another theory proposes that tumor cells arise directly from uterine mesenchymal cells, which retained a potential to differentiate into mesothelial cells [3].

Diagnosis may be difficult; an adenomatoid tumor with a prominent smooth muscle component may be mistaken for leiomyoma. The cords of cells may be diagnosed as an infiltrating malignant epithelial neoplasm or an hemangioendothelioma; also the small vacuolated cells may be confused with a signet ring cell adenocarcinoma, the contribution of immunohistochemistry in these situations is important [5,6].

In conclusion, we have reported a new case of leiomyoadenomatoid tumor of the uterus, ignorance of this rare entity may lead to false diagnosis so more aggressive therapy.

References

- Epstein JI. Differential diagnosis in pathology: Urologic disorders. New York: Igasku-Shoin; 1992;173-4.
- Amre R, Constantino J, Lu S, Charney D. Pathologic quiz case: A 52-year-old woman with a uterine mass. Leiomyo-adenomatoid tumor of the uterus. Arch Pathol Lab Med. 2005;129(3):e77-8.
- Sarma NH, Srinivasulu M, Surchitra MJ. Leiomyoadenomatoid tumor of the uterus: Report of a rare entity and review of the literature. Indian J Pathol Microbiol. 2014;57(3):450-2.
- Dobrosz Z, Palen P, Wlasczuk P, Stojko R. An atypical leiomyoadenomatoid tumor of the uterus: A case report and literature review. Ginekol Pol. 2013;84(8):730-2.
- Hong R, Choi DY, Choi SJ, Lim SC. Multicentric infarcted leiomyoadenomatoid tumor: A case report. Int J Clin Exp Pathol. 2009;2(1):99-103.
- Bahuguna G, Misra D, Malhotra V, Sinha A. Multicentric leiomyoadenomatoid tumor of the uterus: A rare and distinct morphological entity. J Obstet Gynaecol India. 2014;64(suppl 1):152-3.
- Erra S, Pastormerlo M, Gregori G, Costamagna D, Pavesi M. A case of leiomyoadenomatoid tumor of uterine seosa: Speculations about differential diagnosis. BMJ Case Rep. 2009;2009:1586.
- Mathew M, Goel G. Leiomyoadenomatoid tumor of the uterus. Turk J Pathol. 2010;26:168-9.

9. Amérigo J, Amérigo-Góngora M, Giménez-Pizarro A, Velasco FJ, Gallardo SP, González-Cámpora R. Leiomyo-adenomatoid tumor of the uterus: A distinct morphological entity? *Arch Gynecol Surg.* 2010;282(4):451-4.
10. Pransgaard T, Lykke R, Hansen ES. Leiomyoadenomatoid tumor of the uterus: Report of a rare entity. *J Gynecol Surg.* 2013;29(4):219-21.
11. Ranjan R, Singh L, Nath D, Sable MN, Malhotra N, Bhatla N, et al. Uterine adenomatoid tumors: A study of five cases including three cases of the rare leiomyoadenomatoid variant. *J Obstet Gynaecol India.* 2015;65(4):255-8.