Abdominal Radiology (2020) 45:2263–2267
https://doi.org/10.1007/s00261-020-02439-0

SPECIAL SECTION: DISTINGUISHED PAPERS FROM JSAR

Uterine adenomatoid tumor associated with lymph node lesions: a case report

Yuya Wakita[1] · Junko Takahama[2] · Satoshi Yamauchi[3] · Hiroshi Okada[3] · Nagaaki Marugami[3] · Takahiro Itoh[3] · Toshiko Hirai[3] · Kimihiko Kichikawa[3] · Tomoko Uchiyama[4] · Chiho Obayashi[4] · Sho Matsubara[5] · Hiroshi Kobayashi[5]

Published online: 14 February 2020

Abstract

We report a case of uterine adenomatoid tumor (AT) with regional lymph node involvement in a 49-year-old woman. Magnetic resonance imaging revealed an aggregated cystic mass in the posterior uterine wall with partial protrusion of the tumor outside the uterus, and cystic masses of same characteristics in the bilateral obturator and right common iliac lymph nodes. FDG PET/CT revealed no significant FDG uptake in the uterine and lymph node lesions. Taking possible lymph node metastasis into consideration, hysterectomy and lymph node biopsy were performed and it revealed AT of the uterus and the lymph nodes histopathologically.

Keywords Adenomatoid tumor · Uterus · Lymph node

Introduction

Adenomatoid tumors (ATs) are relatively rare benign lesions that are reported most commonly in the genital tracts of both sexes, including the paratesticular structures in males, and the uterus, fallopian tubes, and ovaries in females [1]. These tumors occur much less frequently in extragenital locations but have been described in various organs and anatomic sites, including the mesentery [2], mediastinum [3], pleural cavity [4], adrenal glands [5], pancreas [6], heart [7], and mediastinal lymph nodes [8]. In a few reported cases, ATs involved several anatomic sites simultaneously; e.g., the uterus, ovaries, and appendix [9]. Here we report a case of AT involving the uterus and its regional lymph nodes. To our knowledge, this is the first report of uterine AT simultaneous involvement of regional lymph nodes.

Case report

A 49-year-old nonparous female presented to her primary care physician complaining of menorrhagia. She had a previous history of secondary hydrocephalus and left ovarian cystectomy but the details were unknown. Computed tomography (CT) revealed an enlarged uterus with a lobulated low attenuation mass of maximum diameter 10 cm embedded in the right posterior myometrium. On magnetic resonance imaging (MRI), T2-weighted images (T2WI) showed a high intensity septated multicystic mass protruding from the myometrium (Fig. 1a,d). The cystic components showed low intensity on T1-weighted images (T1WI) (Fig. 1b). The septa showed isointensity on T1WI and T2WI to that of myometrium, and slight enhancement on post-contrast fat-saturated T1WI (Fig. 1c). No area of significant high intensity was observed on diffusion-weighted imaging (DWI) (Fig. 1e). The bilateral obturator and right common iliac lymph nodes were enlarged (diameter 1–1.5 cm)

✉ Yuya Wakita
yuya.wakita@gmail.com

1 Department of Radiology, Nara Prefecture General Medical Center, 2-897-5, Shichijonishi-town, Nara-City, Nara 630-8581, Japan

2 Department of Radiology, Higashiosaka City Medical Center, 3-4-5, Nishiiwata, Higashiosaka-City, Osaka 578-8588, Japan

3 Department of Radiology, Nara Medical University, 850, Shijo-town, Kashihara-City, Nara 634-8522, Japan

4 Department of Diagnostic Pathology, Nara Medical University, 850, Shijo-town, Kashihara-City, Nara 634-8522, Japan

5 Department of Obstetrics and Gynecology, Nara Medical University, 850, Shijo-town, Kashihara-City, Nara 634-8522, Japan

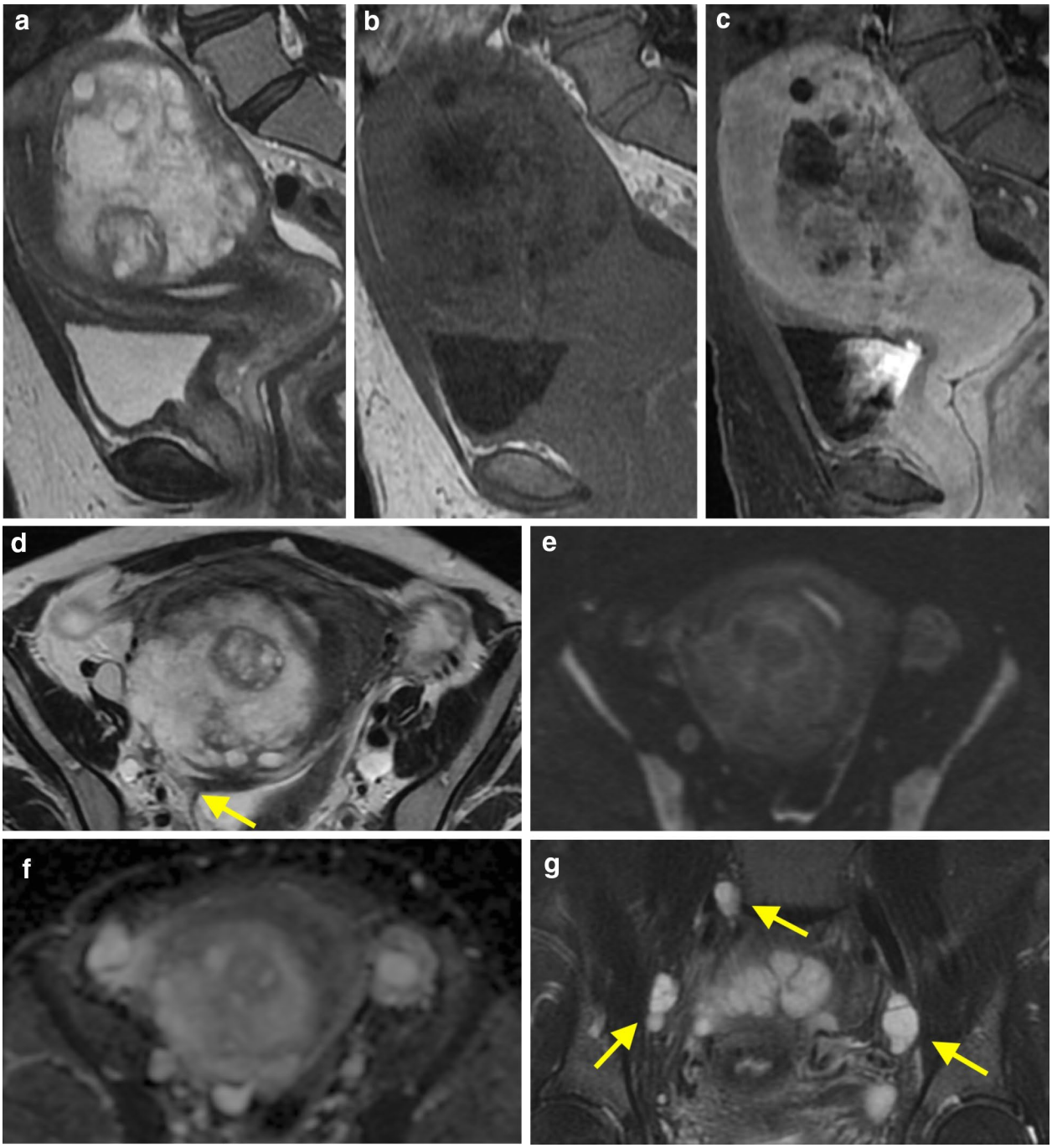

Fig. 1 Contrast-enhanced MRI. **a** Sagittal T2WI, **b** sagittal T1WI, **c** sagittal post-contrast fat-saturated T1WI, **d** axial T2WI, **e** axial DWI ($b = 1000$), **f** axial ADC map, **g** coronal fat-saturated T2WI. MRI showed aggregated cystic mass located in the uterine posterior wall. The mass showed high intensity on T2WI and mix intensity of low and intermediate on T1WI. The septa showed isointensity on T1WI and T2WI to that of myometrium, and slightly enhancement on post-contrast fat-saturated T1WI. A part of the tumor protruded outside the uterus (arrow in **d**). Signal intensity of the tumor on DWI was intermediate with its ADC map high. Bilateral obturator and right common iliac lymph nodes lesions had same characteristic as uterine lesion (arrows in **g**)

and contained solid and multicystic components, the same as the uterine mass (Fig. 1g). FDG PET/CT was performed to investigate lymph node involvement and showed minimal FDG uptake in the uterus and lymph node lesions (Fig. 2). Based on the radiologic findings, the differential diagnoses included leiomyoma, endometrial stromal sarcoma, myxoid leiomyosarcoma, and adenomatoid tumor. The patient underwent surgical resection for preoperative diagnosis of

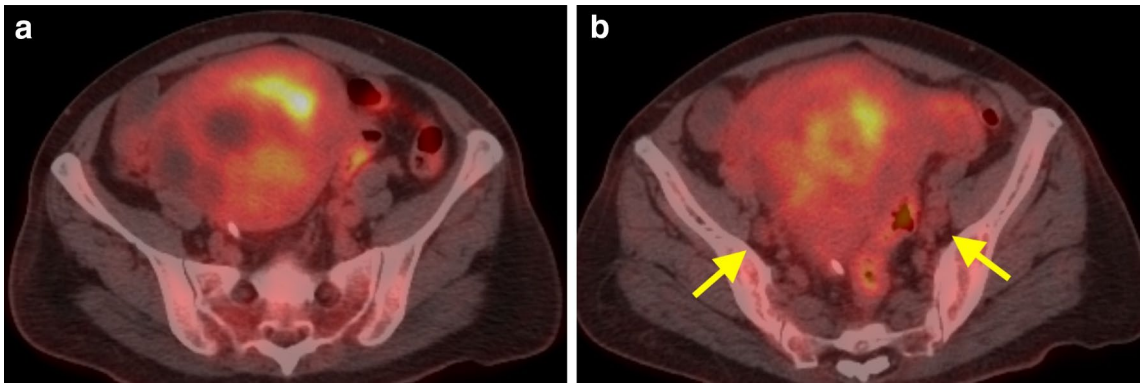

Fig. 2 **a** and **b** FDG PET/CT revealed minimal FDG uptake in the uterus and lymph node lesions (arrow)

malignancy involving regional lymphadenopathy, and total hysterectomy, bilateral salpingo-oophorectomy, and pelvic lymph node biopsy were performed. Examination of hematoxylin-eosin-stained permanent sections of the uterine lesion revealed a neoplasm characterized by pseudoacini and anastomosing channels lined by both epithelioid and flattened cells (Fig. 3a,b). The migration of the mucin into the interstitium was observed (Fig. 3a). The epithelioid and

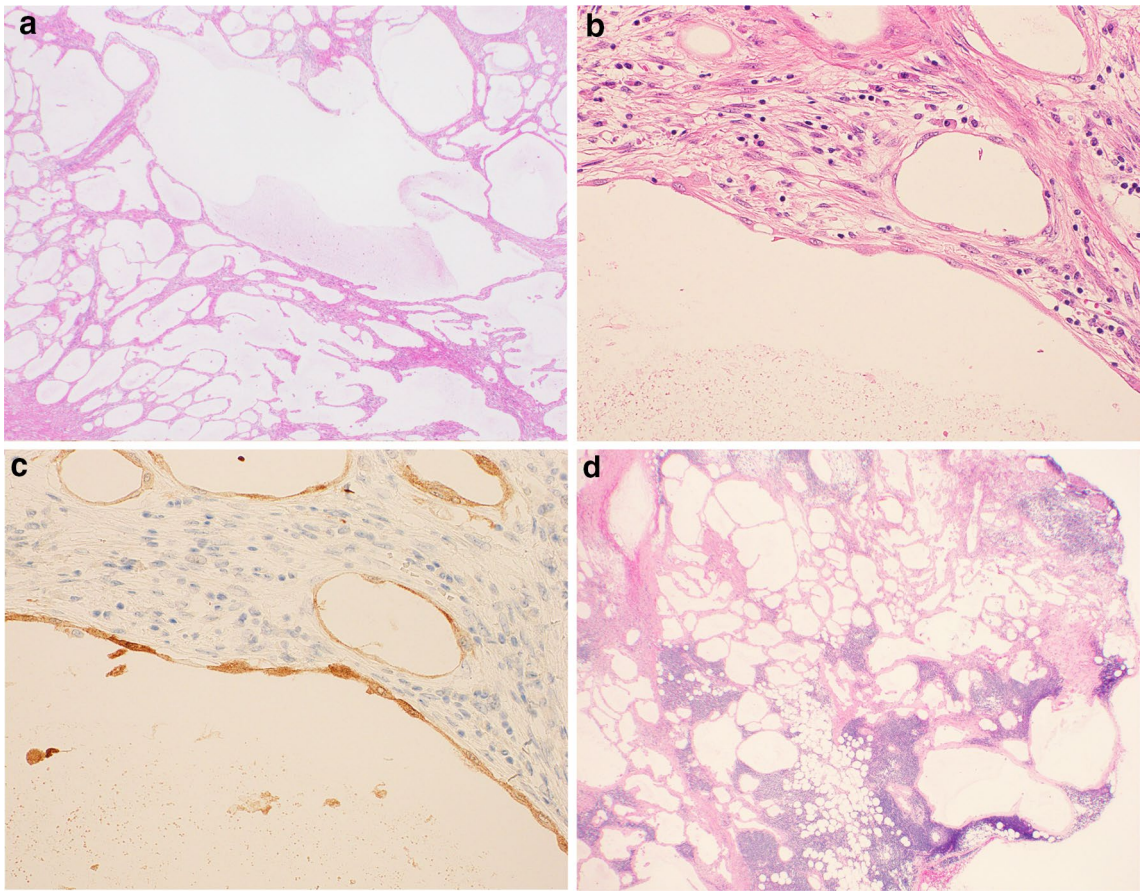

Fig. 3 **a**–**d** Histopathological images of AT (**a**, **b**, HE staining of uterine lesion, **c** calretinin staining of uterine lesion, **d** HE staining of lymph node lesion). The uterine tumor showed pseudoacini and anastomosing channels lined by both epithelioid cells and flattened cells with emigration of mucus to the interstitium. The cells lining the cystically dilated spaces showed positive immunostaining for calretinin. The lymph node tumor showed honeycomb arrangement of cystic spaces lined by a single layer of flattened cells, which showed positive immunostainig for calretinin

flattened cells exhibited strong immunoreactivity for calretinin (Fig. 3c), CAM 5.2, and D2-40, but were estrogen-receptor- and progesterone-receptor-negative, and were also negative for desmin, αSMA, and CD34. Examination of hematoxylin-eosin-stained permanent sections of the bilateral obturator lymph nodes revealed variably dilated acinar structures (Fig. 3d). Immunostaining of the lymph node lesions showed strong immunoreactivity of calretinin and D2-40, the same as the uterine lesion. Hematoxylin-eosin histology and immunophenotyping confirmed the mesothelial differentiation of the neoplasm. AT involving the uterus and lymph nodes was diagnosed.

Discussion

The term "adenomatoid tumor" was first introduced in 1945 by Golden and Ash [10] to describe benign lesions that display a strikingly adenomatous appearance. ATs are commonly asymptomatic and found incidentally [10–12]. The most common sites of involvement are the epididymis in men, and the fallopian tubes and uterus in women [10, 12, 13]. Adenomatoid tumors of epididymis shows low intensity relative to testicular parenchyma on T2WI. These tumors demonstrate slow or decreased contrast enhancement relative to the testicular parenchyma or contralateral testis. However, the enhancement can be variable because, less commonly, adenomatoid tumors show hyperenhancement relative to the testis [14]. Rarely, these tumors may originate in extragenital sites, such as the mesentery [2], mediastinum [3], pleural cavity [4], adrenal glands [5], pancreas [6], heart [7], and mediastinal lymph nodes [8].

The histogenesis of ATs is controversial, and mesonephric, Müllerian, endothelial, and mesothelial origins have all been suggested. Mesothelial cells have been proposed as the progenitor cells of these tumors [11, 12].

Histologically, ATs can be classified as adenoid, angiomatoid, solid, or cystic type [13, 15–17]. One pattern may predominate, but an admixture of two patterns is more common, with the most frequent combination being adenoid and angiomatoid. The cystic type of AT is the least common. It has been the subject of mostly single case reports [1, 15–18] and is the histological pattern found in the uterine and lymph node lesions of the present patient.

Uterine AT can be found in 0.12–1.2% of all women [13, 19]. The actual incidence is probably higher; these tumors frequently go undetected because of their small size and gross appearance, which is similar to that of leiomyoma [20, 21]. They occur most often in women of reproductive age. A previous study has reported a mean age of 41 years (range 26–55 years) at initial diagnosis [22]. The clinical manifestations of uterine AT are nonspecific and variable, and include vaginal bleeding, menorrhagia, and abdominal masses [23]. Most uterine ATs present as a subserosal mass located near the uterine horn [16]. They are often solitary and small, with a mean diameter of 2.1 cm (range 2–10 cm) [17]. On gross examination, ATs are soft and moist, and may have mucus degeneration. They can show an invasive growth pattern due to the lack of a capsule [24]. The radiologic findings of the present uterine lesion were typical of a cystic adenomatoid mass. Based on the radiologic findings, as a cystic mass forming in the uterine corpus, we differentiated leiomyoma, endometrial stromal sarcoma, myxoid leiomyosarcoma, and AT. Leiomyomas are by far the most common uterine tumors and the most common gynecologic tumors. 4% of leiomyomas undergo cystic degeneration, with extensive edema forming cystic, fluid-filled spaces [25]. Endometrial stromal sarcomas account for 10% of primary uterine sarcomas. Although they commonly appear as a polypoid mass, Park et al. reported that low-grade endometrial stromal sarcomas showed multiseptated cystic appearance (20%) or a multiple clustered small cystic appearance (30%) [26]. Rarely, myxoid leiomyosarcoma can be showed same radiologic findings [27], it is difficult to differentiate these from AT, signal intensity of diffusion-weighted image might be the key to distinguish AT from other tumors especially malignancies as endometrial stromal sarcomas and myxoid leiomyosarcomas.

Although multiple tumors in the genital tract have previously been reported as rare (2.8%) [28], a recent report stated that their occurrence is not uncommon (20%) [22]. Most previous reports describe multiple ATs restricted to the uterine corpus. Srigley reported a rare case of multifocal and diffuse ATs involving the uterus and a fallopian tube [29], and Harada reported a rare case of ATs in three different anatomical sites: the uterus, right ovary, and appendix [9].

Isotalo et al. reported AT in a mediastinal lymph node [8], which to the best of our knowledge is the first and only previous description of AT involving lymph nodes. However, there are several reports of hyperplastic mesothelial cells in lymph nodes [30, 31]. It has been suggested that ATs that develop in nonmesothelial-lined sites such as the lymph nodes may have originated from mesothelial inclusions [5] or even from embolized mesothelial cells [30, 31]. Our patient had ATs involving the uterus and its regional lymph nodes. This case supports the hypothesis of AT development from embolized mesothelial cells that originated in the uterus.

The treatment for uterine AT is surgical resection, by hysterectomy or laparoscopic tumor excision. The prognosis is good, and no cases of recurrence or metastasis have been reported in the literature. Careful follow-up and management is recommended in the present case because of the lymph node involvement.

In summary, we report a case of uterine AT simultaneous involvement of regional lymph nodes. Although the

radiologic findings of uterine AT of our case quite match the character of cystic type uterine AT, differential diagnosis must be done to distinguish AT from malignancy, in terms of involvement of regional lymph nodes.

Compliance with ethical standards

Ethical approval This article does not contain any studies with human participants or animals performed by any of the authors.

References

1. Sangoi AR, McKenney JK, Schwartz EJ, et al. (2009) Adenomatoid tumors of the female and male genital tracts: a clinicopathological and immunohistochemical study of 44 cases. Mod Pathol 22: 1228-1235
2. Craig JR, Hart WR (1979) Extragenital adenomatoid tumor: evidence for mesothelial theory of origin. Cancer 43: 1678-1681
3. Plaza JA, Dominguez F, Suster S (2004) Cystic adenomatoid tumor of the mediastinum. Am J Surg Pathol 28: 132-138
4. Kaplan MA, Tazelaar HD, Hayashi T, et al. (1996) Adenomatoid tumors of the pleura. Am J Surg Pathol 20: 1219-1223
5. Travis WD, Lack EE, Azumi N, et al. (1990) Adenomatoid tumor of the adrenal gland with ultrastructural and immunohistochemical demonstration of a mesothelial origin. Arch Pathol Lab Med 114: 722-724
6. Overstreet K, Wixom C, Shabaik A, et al. (2003) Adenomatoid tumor of the pancreas: a case report with comparison of histology and aspiration cytology. Mod Pathol 16: 613-617
7. Natarajan S, Luthringer DJ, Fishbein MC (1997) Adenomatoid tumor of the heart: report of a case. Am J Surg Pathol 21: 1378-1380
8. Isotalo PA, Nascimento AG, Trastek VF, et al. (2003) Extragenital adenomatoid tumor of a mediastinal lymph node. Mayo Clin Proc 78: 350-354
9. Harada S, Okumura Y, Kaida K (2003) Multicentric adenomatoid tumors involving uterus, ovary and appendix. J Obstet Gynacol Res 29(4): 234-238
10. Golden A, Ash J (1945) Adenomatoid tumors of the genital tract. Am J Pathol 25: 63-80
11. Mackay B, Bennington JL, Skoglund RW (1971) The adenomatoid tumor: fine structural evidence for a mesothelial origin. Cancer 27: 109-115
12. Taxy JB, Battifora H, Oyasu R (1974) Adenomatoid tumors: a light microscopic, histochemical, and ultrastructural study. Cancer 34: 306-316
13. Quigley JC, Hart WR (1981) Adenomatoid tumors of the uterus. Am J Clin Pathol 76: 627-635
14. Mittal PK, Abdalla AS, Chatterjee A, et al. (2018) Spectrum of Extratesticular and Tsticular Pathologic Conditions at Scrotal MR Imaging. RadioGraphics 38: 806-830
15. Palacios J, Manrique AS, Villaespesa AR, et al. (1991) Cystic adenomatoid tumor of the uterus. Int J Gynecol Pathol 10: 296-301
16. Kim JY, Jung K, Sung NK, et al. (2002) Cystic adenomatoid tumor of the uterus. AJR Am J Roentgenol 179:1068-1070
17. Nogales FF, Isaac MA, Hardisson D, et al. (2002) Adenomatoid tumors of the uterus: an analysis of 60 cases. Int J Gynecol Pathol 21: 34-40
18. Bisset DL, Morris JA, Fox H (1988) Giant cystic adenomatoid tumour (mesothelioma) of the uterus. Histopathology 12: 555-558
19. Christensen C (1990) Adenomatoid tumours of the uterus. Eur J Gynecol Oncol 11: 85-89
20. Livingston EG, Guis MS, Pearl ML, et al. (1992) Diffuse adenomatoid tumor of the uterus with a serosal papillary cystic component. Int J Gynecol Pathol 11: 288-292
21. Rosa GD, Boscaino A, Terracciano LM, et al. (1992) Giant adenomatoid tumors of the uterus. Int J Gynecol Pathol 11: 156-160
22. Huang CC, Chang DY, Chen CK, et al. (1995) Adenomatoid tumor of the female genital tract. Int J Gynecol Obstet 50: 275-280
23. Schwartz EJ, Longacre TA (2004) Adenomatoid tumors of the female and male genital tract. J Gynecol Pathol 23: 123-128
24. Kalidindi M, Odejinmi F (2010) Laparoscopic excision of uterine adenomatoid tumor: two cases and literature review. Arch Gynecol Obstet 281: 311-315
25. Moyle PL, Kataoka MY, Nakai A, et al. (2010) Nonovarian Cystic Lesions of the Pelvis. RadioGraphics 30: 921-938
26. Park GE, Rha SE, Oh SN, et al. (2016) Ultrasonographic findings of low-grade endometrial stromal sarcoma of the uterus with a focus on cystic degeneration. Ultrasonography 35: 124-130
27. Holzmann C, Saager C, Mechtersheimer G, et al. (2018) Malignant transformation of uterine leiomyoma to myxoid leiomyosarcoma after morcellation associated with ALK rearrangement and loss of 14q. Oncotarget 9(45); 27595-27604
28. Youngs LA, Taylor HB (1967) Adenomatoid tumors of the uterus and fallopian tube. Am J Clin Pathol 48: 536-545
29. Srigley JR, Colgan TJ (1988) Multifocal and diffuse adenomatoid tumor involving uterus and fallopian tube. Ultrastruct Pathol 12: 351-355
30. Clement PB, Young RH, Oliva E, et al. (1996) Hyperplastic mesothelial cells within abdominal lymph nodes: mimic of metastatic ovarian carcinoma and serous borderline tumor – a report of two cases associated with ovarian neoplasms. Mod Pathol 9: 879-886
31. Isotalo PA, Veinot JP, Jabi M (2000) Hyperplastic mesothelial cells in mediastinal lymph node sinuses with extranodal lymphatic involvement. Arch Pathol Lab Med 124: 609-613