CASE REPORT / OLGU SUNUMU

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MEDICAL SCIENCES / DAHİLİ TIP BİLİMLERİ

Ultrasonography and MR Imaging Findings in a Patient with Tubal Adenofibroma

Tubal Adenofibromlu Bir Olguda Ultrasonografi ve MR Görüntüleme Bulguları

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Abstract

Adenofibroma is an uncommon tumor containing both epithelial and stromal components. In this report, we aimed to present a case of fallopian tube adenofibroma with an emphasis on imaging findings. On ultrasonography, a spongy semisolid mass, and on T2-weighted MR sequences, a multilocular hyperintense mass adjacent to normal left ovary were seen. Tiny hyperintense locules were noted in the hypointense fibrous component, which resembles the "black-sponge" appearance previously described for ovarian adenofibromas. Although there is no current evidence that the diagnosis of tubal adenofibroma can solely be made by imaging, the presence of "black sponge"-like appearance may support this diagnosis.

Key Words: Fallopian Tubes, Adenofibroma, Ultrasonography, Magnetic Resonance Imaging

Öz

Adenofibrom, hem epitelyal hem de stromal bileşenler içeren nadir bir tümördür. Bu yazıda fallop tüpü kaynaklı adenofibrom olgusunun görüntüleme bulgularını sunmayı amaçladık. Ultrasonografide süngerimsi semisolid kitle, T2 ağırlıklı MR sekanslarında normal sol overe komşu multiloküler hiperintens kitle görüldü. Daha önce over adenofibromları için tanımlanan "siyah sünger" görünümüne benzeyen hipointens fibröz komponentte küçük hiperintens loküller kaydedildi. Tubal adenofibrom tanısının yalnızca görüntüleme ile konulabileceğine dair güncel bir kanıt bulunmamakla birlikte, "siyah sünger" benzeri görünümün varlığı bu tanıyı destekleyebilir.

Anahtar Kelimeler: Fallop Tüpleri, Adenofibrom, Ultrasonografi, Manyetik Rezonans Görüntüleme

Introduction

Adenofibroma is an uncommon benign tumor composed of an admixture of glandular and stromal tissues. The fallopian tube adenofibromas are considered to be analogous to those of the ovary however, they develop much less frequently than those in the ovary (1-3).

Most tubal adenofibromas are discovered as incidental findings at the time of histological examination for other gynecological disorders. Since the fallopian tubes are not routinely completely sampled and examined histologically, the incidence of tubal adenofibromas is not known. Several studies revealed that these tumors probably develop much more frequently than was formerly believed. In a study published in 2008, the frequency of adenofibromas arising from tubal fimbriae was found to be higher than expected. However, in this study most of these adenofibromas, limited to fimbriae, were smaller than 1 cm and were considered to be incipient. It was emphasized that they were very likely to be overlooked by methods other than histological examination (4). In a more recent study published in 2020, incidental adenofibromas were found histologically at a rate of 2.2% in the tubes of patients who underwent surgery for ovarian tumor (5).

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Large clinically significant tubal adenofibromas are very uncommon. The tumor is usually a round, solitary mass that has developed either intraluminally or has attached to the serosal surface or fimbriated part of the fallopian tube (2).

Our purpose here is to present the ultrasonography (US) and magnetic resonance imaging (MRI) findings of an adenofibroma of the distal portion of the fallopian tube. To our knowledge, MRI features of adenofibroma along with US findings in this location has not been previously described.

Case Report

A 42-year-old, gravida 5, para 3 woman with clinical symptoms of menstrual disturbance for 4 months was referred to the MRI department because of a pelvic mass detected by US. Because there was a suspicion of intestinal origin of the mass on US examination, MRI was planned to determine the site of origin and lesion characterization. She had one cesarean delivery and there was no significant past history. The serum levels for CA-125, CA-19-9, and CEA were all within normal limits, as well as blood cell counts and blood biochemistry. Transabdominal US and HD-flow™ power Doppler US performed on the same day of MRI revealed a 57x50 mm hypovascular, spongy mobile, and semisolid mass adjacent to normal left ovary (Figures 1a and b). Its US features were not found to be typical for a degenerated exophytic myoma.

Pelvic MRI was performed on a 3.0-Tesla scanner with phased array body coil. It revealed a well-circumscribed nodular mass with dimensions of 63x59x54 mm (LxTrxAP) in the left anterolateral side of the uterine corpus and in the inferior aspect of the left ovary. On T1-weighted MR images, the mass appeared mostly homogeneous and was isointense with skeletal muscle. On T2- weighted and short tau inversion recovery (STIR) images, it was predominantly hyperintense mass containing linear hypointense bands (Figures 2a and b). There was a hypointense solid component on its superior aspect (Figure 2a). On diffusion weighted images, diffusion restriction was not detected. After IV gadolinium chelate injection, mild enhancement, more prominent in the upper solid portion and linear bands of the mass was observed. The MRI appearance of the lesion suggested a degenerated leiomyoma. The lesion was considered to be originated from serosal surface of the uterus or from the broad ligament.

In laparoscopic exploration, the uterus and left ovary were normal. There was a solid mass of 6 cm in diameter that involved the distal ampulla, infundibulum and fimbriae of the left fallopian tube. The left ovary was seen clearly separate from the lesion. The left ovarian fossa was normal. Right ovary, right fallopian tube, pouch of Douglas, and the bladder peritoneum were normal. Preserving the left ovary, a left salpingectomy was performed and the mass was excised.

The laparoscopic specimen sent to pathology department was a cream-colored solid tissue of 9x6x5 cm in size. It was well circumscribed with the cut surface exhibiting a cream colored fibrillary and multicystic-spongy appearance. On microscopic examination, cystic and cleft like glands were lined by bland tubal epithelium. The stroma was highly fibrotic. No mitosis or periglandular cuffing was identified (Figure 3a). Immunohistochemically, the stroma was negative with desmin (Figure 3b); positive with steroidogenic factor 1. No fibrous framework was seen with reticulin staining. In the light of this findings tubal adenofibroma diagnosis was made.

Discussion

Adenofibroma of the fallopian tube is an uncommon benign tumor of the female genital tract containing both epithelial and fibrous stromal components. It locates mostly within the fimbriated end of the fallopian tubes (1-4). There are evidences

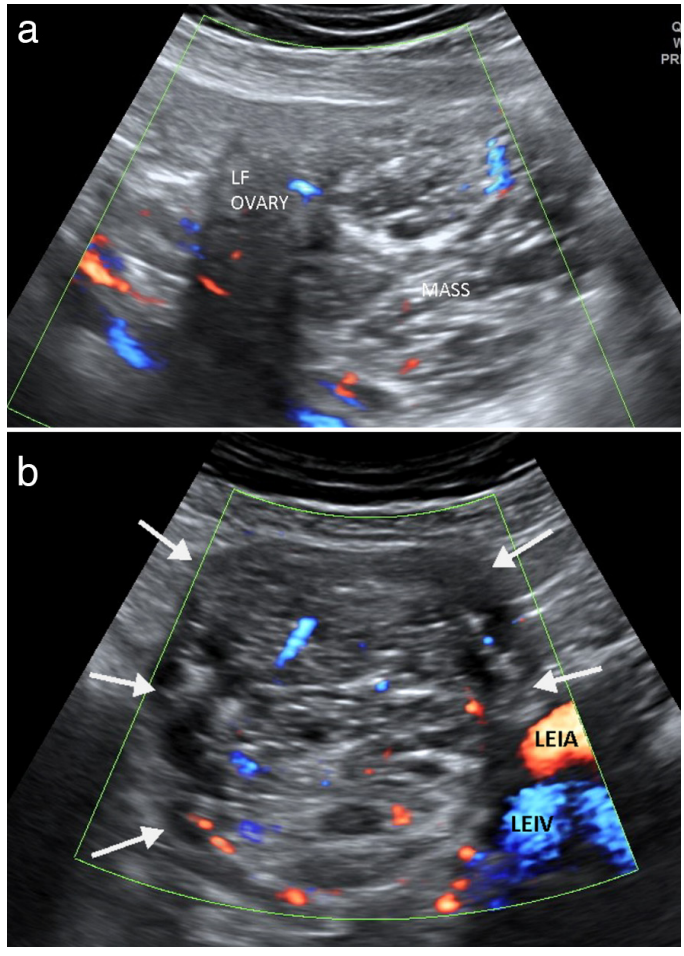


Figure 1a, b: Transabdominal HD-flowTM power Doppler US images of the adnexal mass showing its close relation to the left ovary **(a)**, and its hypovascular, spongy and semisolid texture **(b)**. The mass (arrows) was separate from the uterus, and its US features were not typical for a degenerated exophytic myoma

LEIA: Left external iliac artery, LEIV: Left external iliac vein, US: Ultrasonography

that the tumor is an embryologic remnant originating from the Müllerian duct rather than a proliferating neoplastic process (6).

Tubal adenofibroma may present diagnostic difficulties on clinical and radiological basis due to its rarity and nonspecific presentation. Most common symptoms are menstrual disturbances, pelvic pain and reproductive dysfunction (2,3).

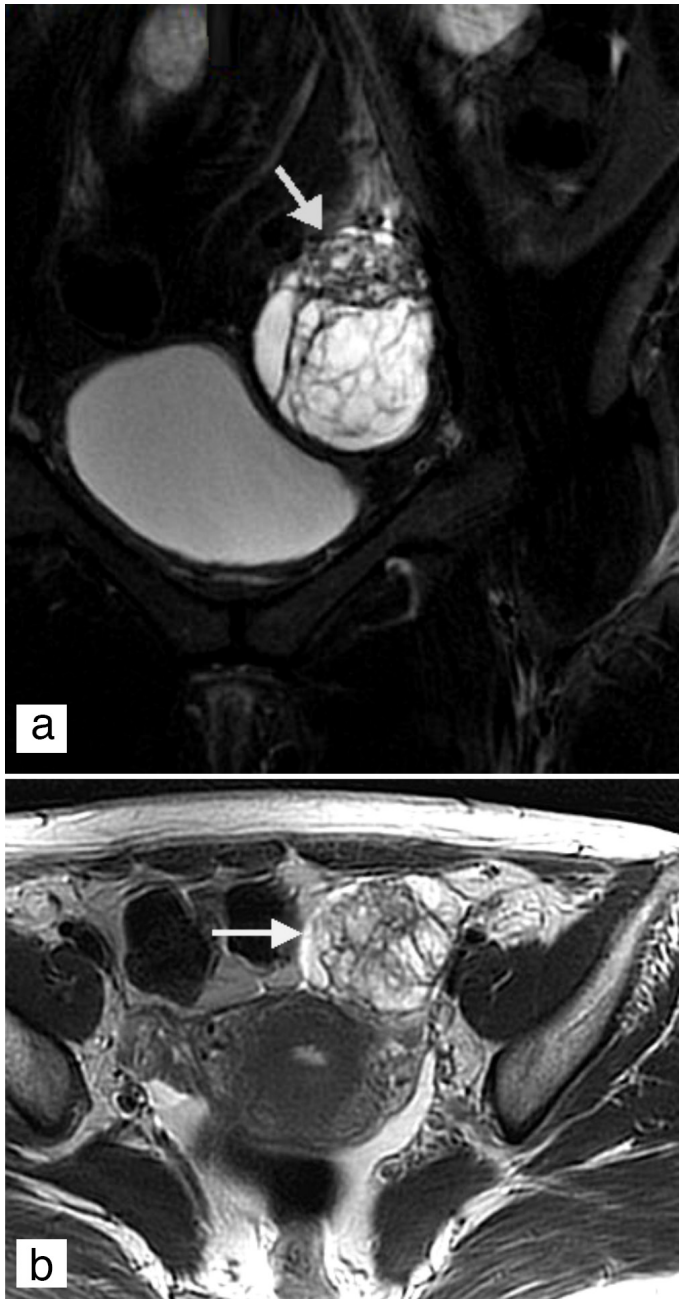


Figure 2a, b: STIR coronal MR image **a)** shows fibrous component (arrow) containing multiple, small hyperintense cystic locules. This corresponds to "black sponge-like appearance". T2-weighted axial MR image **b)** shows well-defined multilocular hyperintense mass in left adnexal region (arrow). Linear hypointense bands within the lesion representing fibrous strands are well visualized

STIR: Short tau inversion recovery, MR: Magnetic resonance

On US, tubal adenofibroma may be seen as a complex structure with regular wall and fine papillae on its internal surface. It may attain a considerable size between the uterus and ovary. On Doppler examination, the tumor is hypovascular (2). The fallopian tube adenofibroma may be misdiagnosed as ectopic pregnancy during US if it presents as a cystic tumor containing a subcentimetric solid mass on its wall (3). The tumor, however, more frequently mimics malignancy on US and CT due to its complex cystic and solid appearance (7,8).

A pattern-based approach, combining tumor morphology and signal patterns on MRI, may allow substantial narrowing of the differential diagnosis of gynecological masses (9). In our case, however, both US and MRI did not enable a specific diagnosis of tubal adenofibroma. Although the surgical specimen was described as solid spongy mass and the ultrasonographic

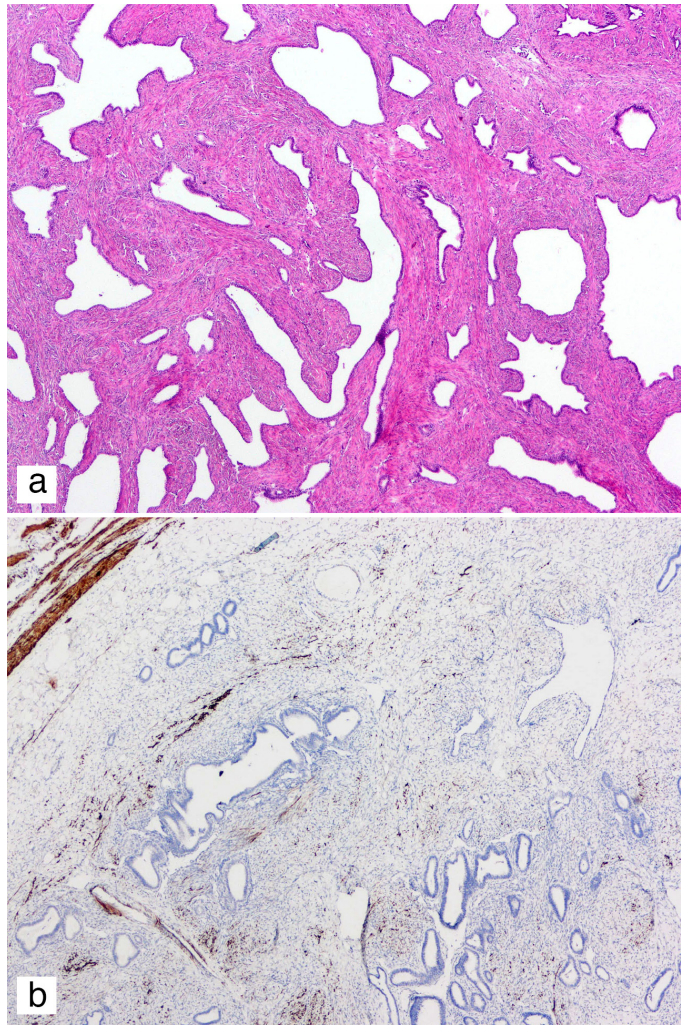


Figure 3a, b: Microscopic examination. **a)** Cystic and cleft glands are embedded in highly fibrotic stroma. This appearance is responsible for sponge-like macroscopy (H&E, 40x). **b)** Stroma is negative with desmin except internal positive controls such as tunica muscularis of the tuba uterina (upper left corner). This indicates the neoplasm does not show any smooth muscle differentiation

echo texture was semisolid with tiny anechoic cysts, the hyperintensity of the mass on T2-weighted images mostly favored the cystic content. Because the lesion was solid in consistency both macroscopically and microscopically, T2 hyperintensity on MRI was thought to be due to the spongy structure formed by cystic glands.

There is limited data about the imaging features of tubal adenofibroma. Therefore, US and MRI characteristics described for its ovarian counterpart may be helpful to recognize the tubal form. Ovarian cystadenofibroma may present as a purely cystic lesion, almost indistinguishable at MRI from a cystadenoma. However, it more often produces a complex cystic appearance with solid components and thick septations. In case series, it is shown that the fibrous components of ovarian adenofibromas tend to have very low signal intensity on T2-weighted MR images (10). Takeuchi et al. (11) noted that the dense fibrous stromal proliferation with scattered small cystic glandular structures create a "black sponge"-like appearance on T2-weighted images. The very hyperintense tiny cysts within the hypointense solid component are found to be characteristic findings of ovarian cystadenofibromas (11). Jung et al. (12) presented clues for the MRI differential diagnosis of ovarian cystadenofibroma and cystadenocarcinofibromas. Diffuse or partial thickening of the cyst wall with dark-signal-intensity in multilocular cystic masses are found to be suggestive findings of ovarian cystadenofibroma. A mild enhancement in the solid components of the mass can be seen (12). These previous observations are also mentioned in a more recent study involving a larger series of patients (13). After retrospective evaluation of images in our case, we realized that the tiny hyperintense cystic structures in the solid component of the lesion which have been described by Takeuchi et al. (11) for ovarian cystadenofibromas were apparent on both T2-weighted and STIR images. We also retrospectively noted that tumor morphology and T2-weighted signal patterns of mucinous type of ovarian cystadenofibromas presented in literature were very similar that in our case (14,15).

Conclusion

In conclusion, currently, there is no substantial evidence that, the diagnosis of tubal adenofibroma can be made by imaging findings. Normal uterus and ovary separate from the mass can suggest the tubal origin of the lesion. Although it may exhibit variable patterns, adenofibroma may be included in the differential diagnosis list when complex mass containing cystic components and septa, as well as solid tissue with low T2 signal and minimal enhancement are seen. The presence of a "black sponge"-like appearance on T2-weighted sequences may support diagnosis, with a pattern-based approach.

Ethics

Informed Consent: Informed consent was obtained.

Peer-reviewed: Externally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: M.E., M.C.S., Concept: E.Ö., Design: E.Ö., Data Collection or Processing: E.Ö., M.E., D.K.Ö., D.A.Ö., M.C.S., Analysis or Interpretation: E.Ö., M.E., D.K.Ö., D.A.Ö., Writing: E.Ö., D.K.Ö.

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