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Uterine Adenofibroma Arising From Uterıne Adenomyosis: An Extremely Rare Localisation

Adenomyozis Odağından Kaynaklanan Uterusun Adenofibromu: Oldukça Nadir Bir Lokalizasyon

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ÖZET

Uterusun Adenofibromları nadir mikst epitelyal ve mezenşimal tümörlerdir. Endometriumdan kaynaklanırlar ve benign epitelyal ve mezenkimal komponenetler içerirler. Adenomyozis; myometrium tabakası içinde endometrial gland ve stromanın bulunduğu lezyondur. Adenomyozisteki endometrial odaklardan nadiren malign tümöral transformasyon gelişebilir. Bir yıldır vaginal kanaması ve pelvik ağrısı olan 51 yaşındaki hastada myometrial adenomyozis odakaları içinde kistik uterin adenofibrom tespit edildi. Literatür taramamızda, Uterine adenomyozisten kaynaklanan adenofibrom olgusuna rastlanmadı ve olgu patolojik ayırıcı tanıları eşliğinde sunuldu.

Anahtar Kelimeler: Endometrium, Adenofibrom, Adenomyozis

ABSTRACT

Uterine Adenofibromas (UAFs) is a rare mixed epithelial and mesenchymal neoplasm composed of benign epithelial and mesenchymal components and usually arise from the endometrium. Adenomyosis is a common gynaecological disorder characterized by presence of endometrial focus consist of endometrial gland and stroma within the myometrium. The endometrial neoplasia originating from adenomyosis uteri or malignant transformation of adenomyosis are rare. A cystic UAF and adenomyosis were detected in the myometrial layer of the uterus in a 51-year-old woman presenting with an abnormal vaginal bleeding and pelvic pain for a year. In this report, we presented a case of cystic uterine adenofibroma case arising from uterine adenomyosis. To best of our knowledge it has not been reported in the literature yet.

Key words: Endometrium, Adenofibroma, Adenomyosis

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Introduction:

Uterine Adenofibromas (UAFs) is a rare mixed epithelial and mesenchymal neoplasm composed of benign epithelial and mesenchymal components (1). UAFs usually arise from the endometrium (90%) and less often occur in the other anatomical locations such as the cervix (2). This benign mixed mesodermal tumor is often seen in the postmenopausal women, while it seems rare in the younger population (1). In this article, we report a case of cystic UAF arising from uterine adenomyosis. To best of our knowledge it has not been reported in the literature yet.

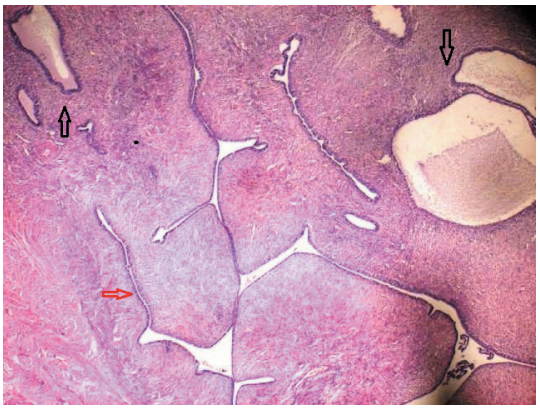
Case presentation:

A 51-year-old woman presented with an abnormal vaginal bleeding and pelvic pain for a year. Pelvic examination and routine haematological parameters of the patient were normal. Pelvic sonography revealed myometrial thickening. Total abdominal histerectomy and bilateral salpingoophorectomy were performed due to uncontrolled bleeding. Macroscopically, the size of the uterus was 10x6x5,5cm and the fallopian tubes and ovaries were normal measures. The thickness of the Endometrium was about 0,5 cm. On Sectioning of the uterus spaces, some foci of hemorrhage and a large cystic foci measured approximatelly 2.3 cm in size were present within the myometrium the thickness of the measured was 2.5-3.0 cm. Polypoid projections were detected into the cystic cavity from the inner surface of the cysts. On histopathological examination, a cystic lesion which contain papillary projections lined by endometrial epithelium and composed of cellular mesenchymal tissue without endometrial gland was presented in the sections of the myometrium (figure 1,2). In addition to there were a large number of adenomyosis focus composed of benign endometrial glandular and stromal elements (figure 1). Neither the mesenchymal component composed of spindle-shaped fibroblasts nor epithelial cells covering the papillae and cysts did not show cytologic atypia and mitotic figures (figure 3). İmmunohistochemically, fibrous mesenchymal tissue was negative for CD10 and ki 67 labelling index was very low (<% 1). The endometrium was in the proliferative phase. The adnexes and cervix did not contain abnormal pathological changes.

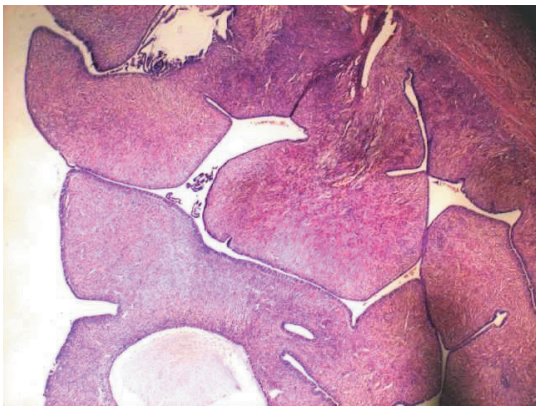
Discussion:

Adenomyosis is a common gynaecological disorder characterized by presence of endometrial focus consist of endometrial gland and stroma within the myometrium. Menorrhagia, dysmenorrhea, pelvic pain and abnormal gynaecological bleeding are the most common clinical symptoms The aetiology is unclear (3). The endometrial neoplasia originating from adenomyosis uteri or malignant transformation of adenomyosis are rare. Endometrial adenocarcinoma (especially endometrioid subtype) and müllerian adenosarcoma are usually reported in the literature as tumors arising uterine adenomyosis, (2,5).

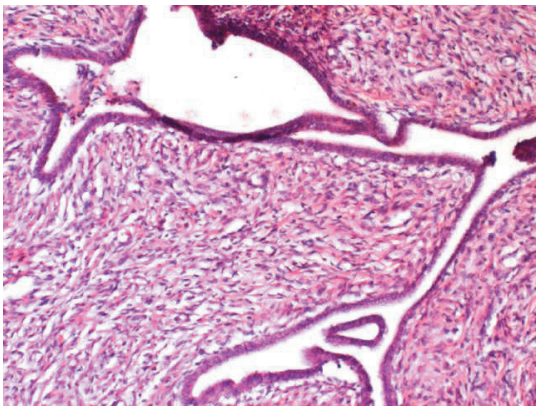
Adenofibroma of the uterus is a rare. It usually occur endometrium (90%) and less often occur in the other anatomical locations such as the cervix (2). UAFs similar to the papillary adenofibroma of the ovary (4). Histologically, these tumors have papillae projecting into clefts and cystic space. The papillae are composed of benign epithelial and mesenchymal components. The epithelial component may be endometrioid, tubal type or endocervical type. Rarelly, the metaplastic transformation of the covering epithelium into



Şekil 1. H&E, X200; Focus of adenofibroma (red arrow); Focus of adenomyosis (black arrows)



Şekil 2. H&E, X200; Benign biphasic epithelial and stromal proliferation. Papillary projections covered by endometrial epithelium and endometrioid type glands.



Şekil 3. H&E, X400; The mesenchymal component is fibroblastic; Mitotic figures and atypia are not observed in the mesenchymal component

squamous epithelium may be seen. The mesenchymal component under the epithelium is usually composed of spindle-shaped fibroblastic type cells. The epithelial and mesenchymal components are bening (1,4,6). The differential diagnoses include mainly low grade adenosarcoma of uterus. High mitotic count, marked hypercellularity and mild or markedly cellular atypia of stromal component favors adenosarcoma, while the stromal component of adenofibroma is morphologically benign(1,2,4). Hysterectomy is the preferred treatment for UAF because of the high recurrence rate after incomplete curettage or excision and the possibility of developing malign lesions(7). In present case, the differantial diagnosis was made from adenosarcoma, as the mesenchymal and epithelial component of the myometrial lesion were benign.

Endometrial tissue is required for the development of UAF. Uterine adenofibromas (UAFs) may be associate with endometriosis which is the presence of endometrial glands and stroma focus in another anatomical locations outside the endometrium (2,6). But, to our best of knowlege, UAF arising from adenomyosis focus consist of endometrial epithelium and stroma in the myometrial layer of the uterus has not ever reported in the literature. In our case, a cystic UAF and adenomyosis were detected in the myometrial layer of the uterus.

In this report, we presented a case of cystic uterine adenofibroma case arising from uterine adenomyosis. In conclusion, UAF arise within adenomyosis is extremely rare. This entity should be considered in a patient with cystic adenomyosis and it should be differentiated from the uterine adenosarcoma.

Conflicts of interest statement: "The authors declare they have no conflict of interests"

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