

# INTRAMURALLY LOCATED ADENOMATOID TUMOR OF THE UTERUS: A CASE REPORT

<sup>1</sup>Nüket Üzüm , <sup>2</sup>Fırat Ortaç , <sup>3</sup>Ömür Ataoğlu

## ABSTRACT:

Adenomatoid tumors are benign tumors originating in the mesothelium. They are mostly underdiagnosed as they simulate leiomyoma in the uterus. We present a 43-year-old woman admitted to hospital due to menorrhagia. Her examination revealed an intramurally located, ill-defined uterine mass diagnosed as an adenomatoid tumor.

**Key words:** Adenomatoid Tumor, Uterus, Mesothelium, İmmunohisto Chemistry

## UTERUSTA İNTRAMURAL YERLEŞİMLİ ADENOMATOİD TUMÖR: BİR OLGU SUNUMU

### ÖZ:

Adenomatoid tümörler benign karakterde mezotelden köken alan tümörlerdir. Çoğunlukla uterusda leiomyom olarak yanlış tanı alırlar. Burada, 43 yaşında kadın hasta menoraji nedeniyle hastaneye başvurmuştur. İnceleme sonrası; uterusda intramural yerleşimli, çevre dokudan düzensiz sınırla ayrılan, adenomatoid tümör olarak tanı alan kitle gözlenmiştir.

**Anahtar Kelimeler** Adenomatoid Tümör, Uterus, Mezotel, İmmünhistokimya

## INTRODUCTION

Adenomatoid tumor is a rare benign proliferation of gland-like structures arising from the genital tract of both females and males, the fallopian tubes and uterus being the most common sites in females<sup>1</sup>. We investigated a uterine tumor in a 43-year-old woman.

## CASE REPORT

A 43-year-old, previously healthy woman was referred to hospital because of recent onset of menorrhagia. Physically, the uterus was firm and slightly enlarged. Hysterectomy with conservation of the adnexa was performed after laparotomy.

The hysterectomy specimen revealed a uterus without macroscopically obvious leiomyomas, but one nodule 2 cm in diameter was observed with ill-defined borders, merging within the myometrium, a picture resembling leiomyoma. It was grayish-brown (Figure 1). Histological examination of this intramural lesion showed gland-like structures in clusters like ectatic vascular spaces, with hyperplastic smooth muscles around these structures (Figure 2). They were lined with a single layer of cuboidal or flattened cells without atypia. There were mild to moderate chronic inflammation and focal hemorrhagic areas within this tumor. Immunohistochemical study revealed cytokeratin (NeoMarkers) (Figure 3) and calretinin (NeoMarkers) positivity in glandular structures, and CD34 (NeoMarkers) and CD31 (NeoMarkers) were negative in the tumor. The endometrium was in the proliferative phase and minimal inflammation was observed in the cervix.

One year after surgery, she is well and without evidence of recurrence.

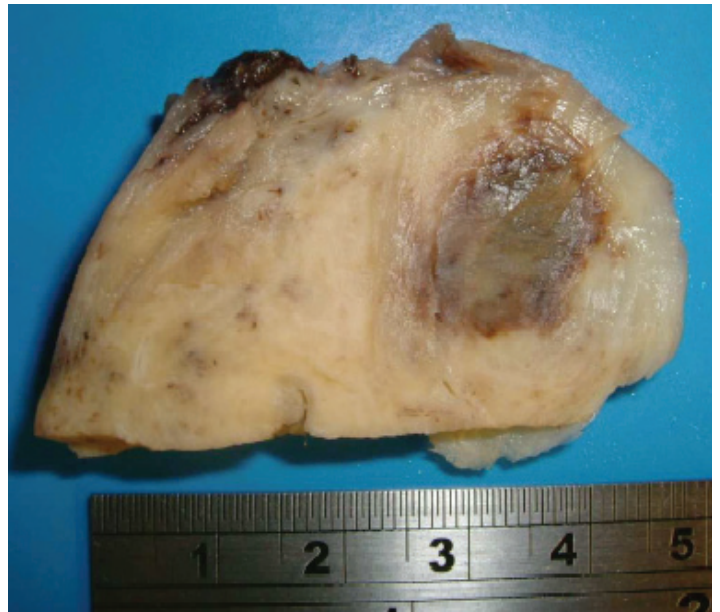


Figure 1: The cut surface of the macroscopic appearance of the adenomatoid tumor

<sup>1</sup> Mikro-Pat Pathology Laboratory, Patoloji, Ankara, Türkiye

<sup>2</sup> Ankara University School of Medicine, Obstetric and Gynecology Department, Ankara, Türkiye

<sup>3</sup> Gazi University School of Medicine, Pathology Department, Ankara, Türkiye

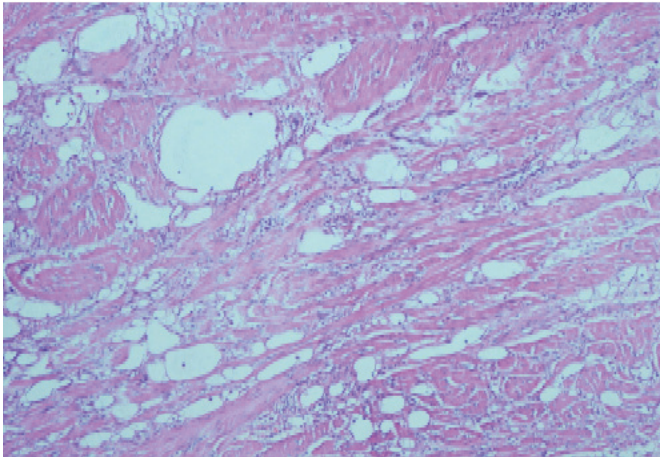


Figure 2: Typical pattern of adenomatoid tumor with a mixture of flattened and cuboidal cells enclosing spaces within hyperplastic muscle layers. Note the moderate chronic inflammation (Hematoxylin and eosin x100)

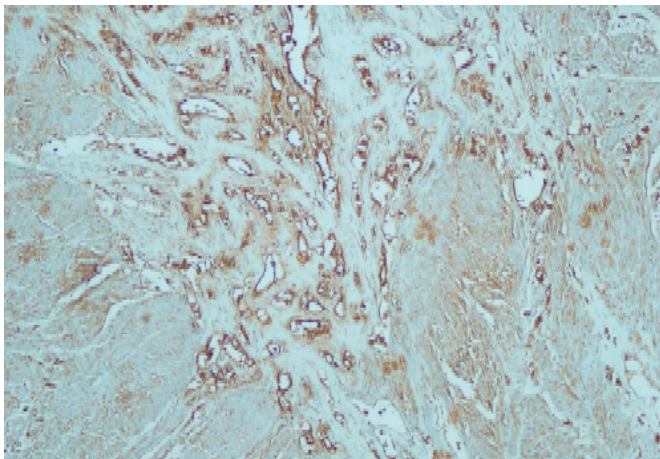


Figure 3: Positive staining of tumor cells with cytokeratin (DAB x40)

## DISCUSSION

Adenomatoid tumors are presumed to be of mesothelial origin (benign mesotheliomas). They occur in areas or organs close to mesothelium-lined surfaces<sup>2</sup>. The true incidence of these tumors is unknown, since they are often detected by chance or are removed because of a preoperative diagnosis of leiomyomas<sup>3</sup>. In males, they are found in the epididymis, spermatic cord, tunica albuginea, prostate, and ejaculatory duct. In the female genital tract, they occur, in order of frequency, in the uterus, tube, and ovarian hilus<sup>4</sup>. Although they are mostly located subserosally, they may be seated deeper in the myometrium as in the present case<sup>5</sup>.

Most cases are under 3 cm in diameter, but giant variants up to 15 cm in diameter are also described<sup>6</sup>. Microscopically, it can have an adenomatoid (most common type), angioma-toid, solid, or cystic architecture, and combinations of more than one type may occur<sup>7</sup>. The adenomatoid type, as described in our case, includes irregular gland-like spaces that are either slit-like or round or cystically dilated<sup>3</sup>. It would seem that the mesothelial cells induce a reactive proliferation of muscle<sup>8</sup>.

Uterine location, in particular the intramural types, is often accompanied by smooth muscle, which usually represents entrapped myometrium permeated by the adenomatoid tumor. In some instances, this type simulates a leiomyoma<sup>9</sup>. The presence of inflammation, as in our case, was observed in up to 80% of cases in the literature<sup>10</sup>. Identification of these tumors needs immunohistochemistry like cytokeratins, or mesothelial markers, since they appear only in empty spaces within the myometrium<sup>3</sup>. Typical circumscribed gross appearance, the bland cytologic features, and lack of mitosis allow distinction from malignant tumors<sup>1</sup>. Multiplicity and multifocality are unusual<sup>3</sup>. Titman found 12 uteri containing adenomatoid tumors in a series of 1000 hysterectomy specimens (1.2%) in contrast to only 1 tubal adenomatoid tumor in the same series<sup>11</sup>. We found only one adenomatoid tumor in 100 uteri retrospectively, a frequency of 1% in our series. This was similar to a previous prospective study, which reported a frequency of 1% in 100 consecutive hysterectomy specimens<sup>1</sup>.

Here, we reported a very rare case of adenomatoid tumor located intramurally, which is a rare benign histologic process.

Correspondence Address: Nüket ÜZÜM

Mikro-Pat Pathology Laboratory,  
Patoloji,  
Ankara, Türkiye  
Tel: 0312 431 32 70  
E-mail: nuketuzum@yahoo.com

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