UNIVERSIDADE DA BEIRA INTERIOR
Ciências da Saúde

# Adenomatoid Tumour of Myometrium
## A Case Report and review of literature

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*Aos meus pais*

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# Resumo

Um caso de tumor adenomatoide é apresentado. A paciente, uma mulher de 49 anos, foi submetida à uma histerectomia vaginal por dor pélvica severa renitente à terapia médica. O diagnóstico de tumor adenomatoide foi feito com base na histologia e imunohistoquímica. O tumor adenomatoide deve ser considerado como diagnóstico diferencial de leiomiomas em casos de pacientes com dores pélvicas severas e útero volumoso à palpação.

## Palavras-chave

Tumor adenomatoide, Tumor uterino, Leiomioma, Tumor benigno.

# Introduction

The adenomatoid tumour (AT) is a rare benign mesothelial proliferation (1). In the genital tract it occurs predominantly in the myometrium or fallopian tubes, rarely in the broad ligament, the ovary, and the extra genital peritoneum of females and the epididymis, spermatic cord, tunica vaginalis and tunica albuginea of males (2). Most AT of the uterus present as solitary asymptomatic lesions diagnosed as incidental findings in hysterectomy specimens, and multiple AT (mAT) are rare (3). We describe a clinical case of a patient with symptomatic mAT.

## Goals

The main goals of this project were:

1. To do a theoretic review on the theme and all its trends; aetiology, diagnosis, treatment and also to report a case.
2. To describe a rare case of multiple AT, diagnosed and treated at Covilhã Hospital Centre, Portugal

## Methods

After consulting the clinical records of the patient, our first step on approaching the theme was a broad search on the internet using the sites Pubmed, Medscape with the following keywords; Adenomatoid tumour, Benign uterine tumours, Leiomyoma mimicker, Adenomatoid diagnosis, Histoimmunochemical diagnosis of adenomatoid, Treatment of adenomatoid tumours. About 30 articles were selected, most of which written in English and Spanish. A search in each paper´s references was also carefully done using the keyword “Adenomatoid Tumour”.

# Abstract

A case of multiple adenomatoid tumours (mAT) of myometrium is presented. The patient, a 49-year-old woman underwent vaginal hysterectomy for severe pelvic pain renitent to medical therapy. The diagnosis of mAT was made based on histology and in immunohistochemistry. AT should be considered in the differential diagnosis with leiomyoma in patients with severe pelvic pain and an enlarged uterus due to multiple tumours.

## Keywords

Adenomatoid tumours, Uterine tumours, Leiomyoma, Benign tumours.

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# Acronyms List

| | |
|---|---|
| UBI | Universidade da Beira Interior |
| FCS | Faculdade de Ciências da Saúde |
| mAT | Multiple Adenomatoid tumours |
| AT | Adenomatoid tumours |

# Results

## Aetiology

Although the mesothelial origin have been the most accepted nowadays (4-6), histogenesis have been heavily discussed and Endothelial, Müllerian, and Mesonephric origin have been also proposed (7). The term adenomatoid reflects its histologic appearance rather than its histogenesis. Golden and Ash suggested an hypothesis that the origin of the tumour, or at least, gland-like element of the tumour is composed of epithelial cells due to the tendency of these cells to develop vacuoles (8). Allying to Golden and Ash, many authors have also considered the AT to be endothelial in origin, accepting the flattened cuboidal appearance of the tumour where large spaces are formed as the cell type (8).

It may well be valid that the common origin of genital mesothelium and Müllerian epithelium makes it difficult for us to reach an agreement. In addition, Marcus and Lynn have concluded that, microscopically, the evidence to distinguish between the two is inconclusive (9, 10). While a Müllerian origin is parallel with the clinical location in both sexes, histologically the tumour cells are unlike endometrial or tubal epithelium and have not demonstrated evidence of cyclic changes (9, 10).

The histologic similarity between mesonephric remnants and tumor cells, the stereocilia of normal epididymis and the clinical occurrence in males are all consistent with a mesonephric hamartomas. However, adenomatoid tumors have never been reported in or around the cervix or vagina where mesonephric rests frequently occur. Furthermore, the tumors occur in places along the tube and uterus where the embryonic Wolffian duct does not pass, and where remnants, therefore, would be unexpected (11).

The mesothelial aetiology of the AT has also been a source of controversy. Although many investigators agree on this hypothesis, recent ultra-structural evidence suggests that some adenomatoid tumours may be a vascular neoplasm. A study pursued in 1982 using an immunoperoxide method of detecting factor VIII related antigen (a tissue specific marker for vascular endothelium) have shown that ATs can be subdivided into tumour of either mesothelial or vascular endothelial origin, and both of these groups could be distinguished on light microscopy (12).

## Pathophysiology

To this date, we could not find any theories regarding the pathophysiology of this lesion.

## Clinical Presentation and Differential Diagnosis

Adenomatoid tumours are usually asymptomatic and frequently are diagnosed after the surgery.

According to a study made by the department of obstetrics and gynaecology of National Taiwan University (13) the most frequent preoperative diagnosis of the AT is a Leiomyoma, followed by an Adnexal cyst. This coincides with the study created by the Pathology and OBGYN department of Clinica Las Condes in Chile which also places Leiomyoma first, succeeded by Adenomyosis (2).

Despite of the fact the AT are benign, and frequently solitary, it has been described cases of coexistence with an endometrial adenocarcinoma, and so it may pose a differential diagnostic problem in the pathologic staging of the endometrial carcinomas, because of their gland-like lumina and infiltration of the myometrium.

Typically AT are subserosal or located in the outermost zone of myometrium. This is in contrast to adenomyosis, where the endometrial glands and stroma infiltrate the myometrium. Interestingly, in 1992, was described a case of a diffuse adenomatoid tumour of the uterus with a serosal papillary cystic component. The presented woman was undergoing immunosuppressive therapy following a kidney transplantation for SLE when a adenomatoid tumour diffusely infiltrating the entire myometrium was found containing a serosal papillary cystic component that resembled a cystic mesothelioma. This was the first reported case of an ATs showing both of these features. Although ATs are considered to be benign, this woman would have a 50% risk of recurrence due to the papillary cystic component (14).
A most recent publication, from 2000, reports a case of a 34-year-old women also undergoing immunosuppressive therapy.

Another interesting case, published in 1986, reports a case of a 25-year-old female with an unusual initial presentation of the AT. She underwent dilation and curettage during investigation for infertility. The endometrial curettings revealed infiltration of the stroma by epithelioid and signet-ring-type tumour cells. Subsequent hysterectomy revealed a large, somewhat ill-defined posterior myometrial tumour that on the basis of histological, histochemical, and ultra-structural investigation proved to be an adenamatoid tumour with infiltration into the endometrium (15).

Table 1 - Clinical findings/ Perioperative diagnosis of genital female adenomatoid tumours. NA Not available

| Author | Clinical findings/ Perioperative diagnosis | Main complain |
|---|---|---|
| (16) | Incidental | NA |
| (17) | Incidental, 32% presumed leiomyoma, adenomyosis, endometriosis and unspecified mass | NA |
| (13) | Incidental | Infertility (3 cases), NA |
| (1) | Leiomyomas, adenomyosis, endometrial polyp, ovarian tumour, uterine prolapse | NA |
| (18) | Leiomyoma, endometriosis | Infertility, dysmenorrhea/ menorrhagia |
| (3) | Leiomyoma | Infertility |
| (19) | Leiomyoma | Menorrhagia |
| (20) | Uterine mass | Menorrhagia, pelvic pain |
| (21) | Uterine prolapse, incidental | NA |
| (2) | Leiomyoma and ovarian teratoma | Pelvic pain, menorrhagia and dysmenorrhea |
| (22) | Uterine mass | Infertility |
| (23) | Leiomyomas | Menorrhagia |
| (24) | Leiomyomas | Menorrhagia |
| (7) | Incidental, leiomyoma, ovarian cysts | Pelvic pain, menorrhagia |
| (25) | Leiomyoma, ovarian mass | Menorrhagia |

## Complementary Methods of Diagnosis

As said it before, the adenomatoid tumour are typically asymptomatic. At microscopy, the leasion usually has an ill-defined margin with the surrounding myometrium, which helps distinguish it from leiomyoma with its distinct margin. However, at MR imaging, the lesion may appear as an ill-defined or well-circumscribed mass of low signal intensity on T2-weighted images, an appearance that can be undistinguishable from that of leiomyoma or adenomyoma (26).

Uncommonly, an adenomatoid tumour has small cystic spaces representing dilated mesothelial tubules (Fig. 1) or appears as a large cystic mass (1).

With that said, to this date, is only possible to diagnose the adenomatoid tumour post operation with the help of anatomical pathologic tests.

## Anatomical Pathological Diagnosis

In order to diagnose lesions of mesothelial origin we have to confirm the presence of at least 2 or 3 of the following specific antibodies: AE1/AE3 Cytokeratin, 5/6 Cytokeratin, Calretinin, D2/40 and Vimentin (1). Furthermore, we would have to exclude the presence of CEA and CD31 (27) and also take into account the proliferative activity of the lesion with help of the proliferation antigen Ki-67 (1).

Microscopically, AT can be classified into adenomatoid (most common type), angiomatoid, solid, or cystic, and combinations of more than one type may occur. The adenomatoid type, includes irregular gland-like spaces that are either slit-like or round or cystically dilated (24) with absence of outstanding atipia or mitotic activity (28). Occasionally, they are filled out by basophyllic material, and can present cytoplasmic vacuolization giving the cells a Signet-ring like appearance (28).

## Treatment

Simple excision with uterine conservation is the treatment of choice especially in women who desire to be pregnant in the future (29). Successful pregnancy has been reported in several cases following surgical excision, which is reassuring to women fertility concerns (3). The efficacy of hormonal therapy, which is generally used for treatment of leiomyoma, is unclear for AT as patients with this kind of lesions receive this hormonal therapy for presumed leiomyomas based on radiological imaging (18). Although two cases of a failed pharmacological therapy has been reported (30).

In 2009, Kalidindi and Odejinmi related two cases in which the tumours were removed laparoscopically. They used the harmonic scalpel (Ethicon Endosurgery) to remove as much of the tumour as possible including parts of healthy myometrium, because of the lack of lines of demarcation of the tumour. This differs from uncomplicated laparoscopic myomectomy where the lines of demarcation are clearly identifiable. They also reported some difficulties with the myomectomy screws because of the friable nature of the ATs (18).

## Prognosis / Post Treatment Vigilance

The Adenomatoid tumours have overall a good prognosis, surgery is curative, but recurrence has been reported, especially in a case of incomplete excision. In 2005, was reported a case of a 33-year-old nulliparous woman with severe menorrhagia and dysmenorrhea, thought to be due to a submucosal fibroid on ultrasound. It was later discovered to be an adenomatoid tumor, and she underwent surgery, although it was ineffective, as the tumour kept recurring. After one year of continuous attempts to remove the tumour she underwent a Strassman procedure, a procedure that consisted in dissection of ureters and pelvic vasculature, selective temporary ligation of uterine arteries, hemi section of the uterus, and excision of the tumour with frozen sections to ensure clear tumour margins and resuturing of the uterine halves. This Temporary vascular occlusion of the uterine arteries and ovarian vessels allowed a Strassman procedure, which resulted in successful resection of a recurrent giant adenomatoid tumour of the uterus, with fertility preservation in a young nulliparous woman. After that, there was no recurrence, 2 years passed and the patient is still tumour free (30). To our knowledge this was the only reported case of adenomatoid tumour recurrence.

# Case Report

A 49-year-old multiparous woman presented in our hospital with a severe pelvic pain and dyspareunia, with no complains of abnormal uterine bleeding.

The patient had an uneventful past medical history except for a Phyllodes tumour (Borderline) excision from the left breast. Gynaecologic data: Menarche at the age of fourteen, two previous (uneventful) gestations and a regular menstrual cycle (3-4/28 days). At that time the patient was taking Lorazepam and pantoprazole daily, but no oral contraception.

Pelvic examination was normal, except for a slightly enlarged uterus. Pelvic ultrasound revealed multiple tumours consistent with leiomyomas.

Vaginal hysterectomy was scheduled for treatment of patient symptomatology.

On gross examination the uterus measures were: 5. 2 x 5.3 x 4.2cm; weight 87gr. Three tumours were identified as yellowish poorly circumscribed intramural nodules, with fasciculate appearance and elastic consistency, all located on anterior uterine wall. The larger tumour measured 2.4 cm in the largest dimension and the smaller tumour measured 0.9 cm (Fig 2).

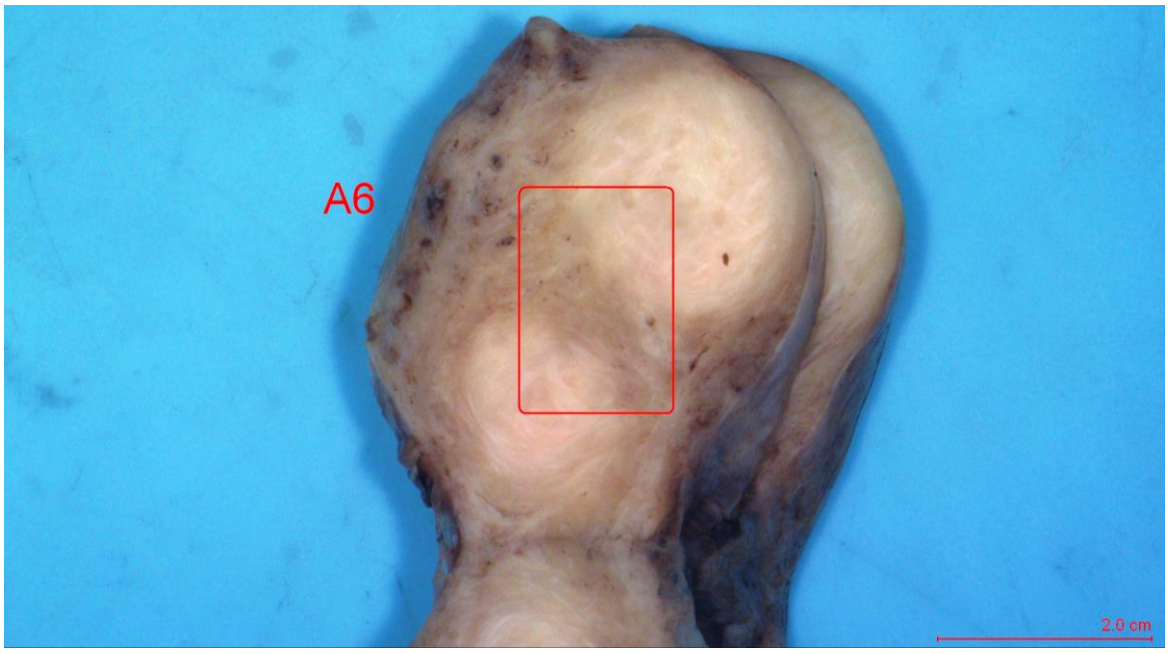


Figure 1A

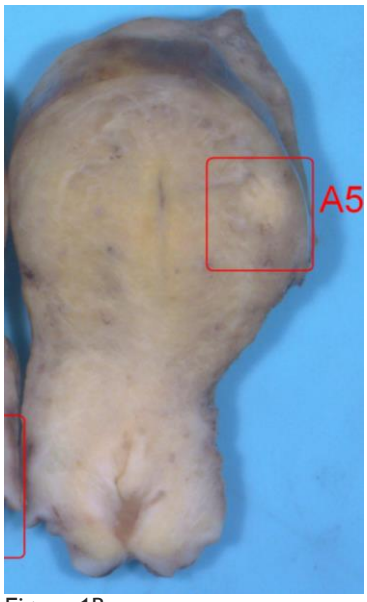


Figure 1B

Figure 1 Uterus cut surface disclosing three discrete miometrial white nodules (two contiguous-A, and another by the serosa-B)

By light microscope all the tumours disclosed tubular structures that dissociate myometrium, comprising cuboidal cell without significant atypia and a low mitotic index (Fig 3).

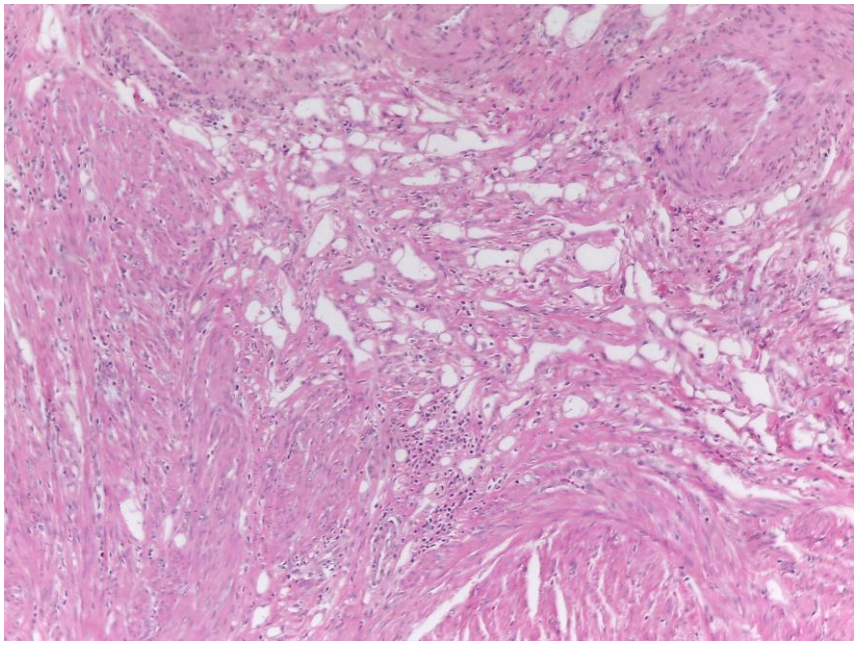

Figure 2A

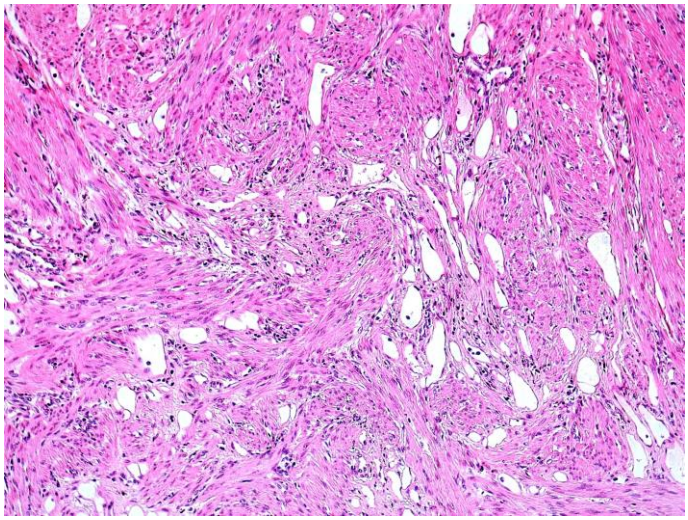

Figure 2B

Figure 2: Uterine adenomatoid tumor: tubular structures dissociating the miometrial smooth muscle (A- H&E, x 20); the cells lining the tubules are flat and without atypia (B- H&E, x 100)

By immunohistochemistry the tumour cells express calretinin and keratins (CK8/18) in the absence of oestrogen and progesterone receptors expression (Fig 4).

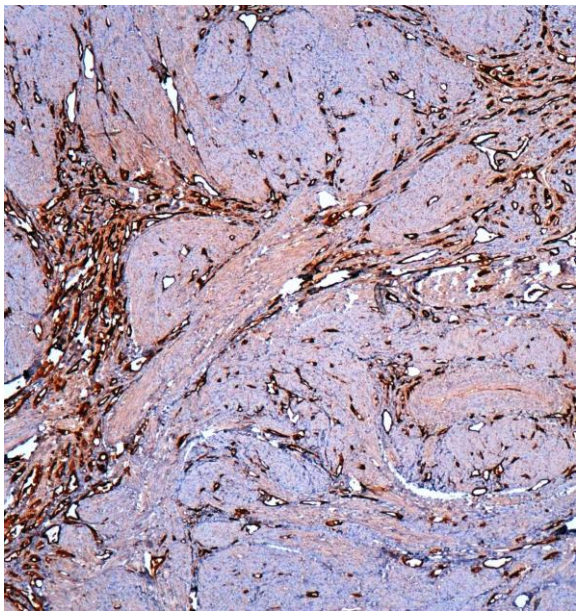

Figure 3- Immunohistochemistry expression of calretinin in the tubular structures of the adenomatoid tumor (ABC, x 40)

No further pathological alterations were found in the specimen. The pathological diagnosis was multiple uterine adenomatoid tumours.

# Discussion

The term Adenomatoid tumours was first proposed by Golden and Ash in 1945 (8) to described a benign tumour that morphologically resembles adenomas (18). It's a rare, non-recurring, benign mesothelial neoplasms of the genital tract that have been observed in women between 30-72 years of age with the median of 42 years (31). The risk of malignant transformation is low; only a case of endometrial carcinoma was reported associated with adenomatoid tumour of the fallopian tube (32). Our patient is 49 years old, in the range reported by previous studies (31).

The incidence of adenomatoid tumours in specimens of hysterectomy have been estimate to be about 1% (33). Although the true incidence may be greater as these tumours frequently are neglected and probably unreported because their small size and similar pathological appearance to leiomyomas (14, 34).

The histogenesis of adenomatoid tumours remains controversial; it is not uniformly agreed whether an adenomatoid tumour is a benign neoplasm or a form of localized mesothelial proliferation; mesonephric, mullerian, endothelial, and mesothelial origins have been suggested (19). Studies based on transmission electron microscopy, scanning electron microscopy, immunohistochemistry, and the typical location of these lesions in genital areas/adrenal glands (that have in common their origin from steroidal crest) has supported their mesothelial origin. In our case, the immunochemistry expression of keratins (CK8/18) and calretinin in the tumour cells supported the mesothelial nature of the mAT.

Our patient had complains of severe persistent pelvic pain and dyspareunia, that was the reason for vaginal hysterectomy, an unusual presentation described in literature for AT (3). Clinically the uterus was discretely enlarged; the pathology report confirmed the small volume of the uterus and allowed the diagnosis of AT. Indeed, the diagnosis of adenomatoid tumour is usually made as an accidental finding on hysterectomy specimen as in the present case (18, 35).

According to previous reports, most of AT are solitary tumours. As reported by others, we could not distinguish AT from (the more frequent) leiomyoma of the uterus by clinical examination or at ultrasound and so, initially, we suspected of leiomyoma (s). In our case, we found three independent AT, all intramural and in the same uterine wall of the surgical specimen. Interestingly, the patient had no complains of abnormal uterine haemorrhages. The three adenomatoid tumours were separated by normal myometrial tissue.

Thus, we speculate that the location of the multiple adenomatoid tumours may explain the painful symptomatology of the patient by interfering with the uterine vascularization.

Table 2- Reports of genital female adenomatoid tumours. NA- Not available.

| Authors/References | Nº Cases | Age (range) | Location | Size of the AT (cm) | Immunohistochemical Test |
|---|---|---|---|---|---|
| **(16)** | 3 | 40-46 | Myometrium (1) and Fallopian tube (2) | 0.6 - 5 .0 | ++ Pancytokeratin calretinin and HBME-1 + Vimentin |
| **(17)** | 32 (Female) | 38-79 | Myometrium (26), Fallopian tube (4), Ovary (2) | 0.1 - 1.6 | ++ CKAE1/CAM 5.2, Calretinin, D2-40 in 100% of cases and for CK5/6 in 16% and Caldesmon in 3% |
| **(13)** | 25 | 26-55 | Myometrium (23), Fallopian tube (2) | 1.0 - 7.0 | ++ CK and alcian blue staining |
| **(1)** | 32 | 29-57 | Myometrium | 0.8 - 4.5 | 100% = + CK AE1/AE3, Calretinin, Vimentin and D2-40. 6% = + CK 5/6. |
| **(18)** | 2 | 36-39 | Myometrium | 6.0 - 7.0 | ++ CAM5.2 CK |
| **(3)** | 1 | 39 | Myometrium | 0.6 - 3.0 | ++ CK, CD34, HBME1 and Vimentin. |
| **(19)** | 1 | 40 | Myometrium | 5.2 | ++ CK |
| **(20)** | 1 | 39 | Fallopian tube | 11.0 | ++ CK, HBME1, Ca-125(M11) + Ca-125 (OC 125) |
| **(21)** | 2 | 36-60 | Myometrium and fallopian tube | 2.0 and unknown | ++ Calretinin and + CK |
| **(22)** | 1 | 33 | Myometrium | 5.0 | ++ CK and vimentin |
| **(23)** | 1 | 40 | Myometrium | 6.0 | ++ CK AE1-AE3 and calretinin |
| **(24)** | 1 | 43 | Myometrium | 2.0 | ++ CK and calretinin |
| **(32)** | 1 | 60 | Fallopian tube | 2.0 | ++ CK and vimentin |
| **(7)** | 2 | 33 and 36 | Myometrium | 0.5 - 3.5 | NA |
| **(25)** | 2 | 26 and 39 | Myometrium and Ovary | 2.5 - 11.5 | ++ Calretinin and + for CK 5 |
| **(36)** | 1 | 47 | Myometrium | 2.5 | NA |
| **(37)** | 1 | 38 | Myometrium | 8.0 | ++ CK |
| **(34)** | 2 | unknown | Myometrium | 12.0 - 13.0 | NA |
| **(38)** | 9 | 28-54 | Myometrium (7), Fallopian tube (1), Ovary (1) | 0.4 - 5.8 | ++ Calretinin and CK AE1/AE3 |
| **(39)** | 24 | unknown | Myometrium (21), Ovaries (2) and both myometrium and ovary (1) | 0.2 - 5.5 | ++ Calretinin, Vimentin and CK AE1/AE3 |
| **(40)** | 60 | unknown | Myometrium | 0.2 - 10 | ++ CK, calretinin, vimentin, and HMBE-1 |

# Conclusions

We report a rare case of a patient with multiple adenomatoid tumours of the myometrium, diagnosed after a hysterectomy performed to treat severe and persistent pelvic pain and enlarged uterus, interpreted as leiomyoma(s). Definitive diagnosis was made in the pathologic study of the hysterectomy specimen.

In a patient with severe pelvic pain and an enlarged uterus by multiple tumours, suggesting leiomyomas, a differential diagnosis of AT should be considered.

# Recommendations

While performing this review I came across a few flaws that could be improved: Because of the rarity of these tumours and its resemblance to leiomyomas many cases go undiagnosed, and so it is hard to calculate the true incidence of these tumours. Hence, I would recommend a creation of a data base relating these cases. The data base could be lodged in the FCS, in other that health science students would have access, and could pursue further studies on the matter, for example, a case study regarding the connection between immunosuppressant drugs and the appearance of Adenomatoid tumours, or regarding the pathophysiology of these lesions.

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# Anexo (s)

*O seguinte artigo foi submetido para publicação na revista JCOG (Journal of Cases in Obstetrics and Gynecology) onde aguarda a aprovação.*

**MULTIFOCAL ADENOMATOID TUMOR OF MYOMETRIUM: A CASE REPORT AND REVIEW OF LITERATURE.**

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**Abstract:** A case of multifocal adenomatoid tumour (mAT) of the myometrium is presented. The patient, a 49-year-old woman underwent vaginal hysterectomy for severe pelvic pain renitent to medical therapy. The diagnosis of mAT was made based on histology and immunohistochemistry. AT should be considered in the differential diagnosis with leiomyoma in patients with severe pelvic pain and an enlarged uterus due to multiple nodules.

**Keywords:** Uterine tumour, Adenomatoid, Benign, dyspareunia.

**Introduction**

The adenomatoid tumour (AT) is a rare benign mesothelial proliferation (1). In the genital tract it occurs predominantly in the myometrium or fallopian tubes, rarely in the broad ligament, the ovary, and the extra genital peritoneum of females and the epididymis, spermatic cord, tunica vaginalis and tunica albuginea of males (2). Most AT of the uterus present as solitary asymptomatic lesions diagnosed as incidental findings in hysterectomy specimens, and multifocal AT (mAT) are rare (3). We describe a clinical case of a patient with symptomatic multifocal adenomatoid tumour.

**Case Presentation**

A 49-year-old multiparous woman presented in our hospital with a severe pelvic pain and dyspareunia, with no complains of abnormal uterine bleeding. Pelvic examination was normal, except for a lightly enlarged uterus.

Pelvic ultrasound revealed multiple nodules consistent with leiomyoma. Vaginal hysterectomy was scheduled for treatment of patient symptomatology. On gross examination the uterus weighted 87 gr and measured 5. 2 x 5.3 x 4.2cm. Sections of the uterus disclosed three yellowish intramural nodules, with fasciculate appearance and elastic consistency. The larger nodule measured 2.4 cm in the largest dimension and the smaller nodule measured 0.9cm (Fig. 1 A and B).

By light microscope all the nodules disclosed tubular structures that dissociate the myometrium, comprising flat/cuboidal cells without significant atypia and a low mitotic index (Fig 2). By immunohistochemistry the tubular lining cells expressed calretinin and keratins (CK8/18) in the absence of oestrogen and progesterone receptors expression (Fig 3).

No further pathological alterations were found in the specimen. The pathological diagnosis was multifocal uterine adenomatoid tumor.

**Discussion**

The term adenomatoid tumour was first proposed by Golden and Ash in 1945 (8) to described a benign tumor that morphologically resembles adenomas (18). It is a rare, non-recurring, benign mesothelial proliferation that has been reported in the genital tract of women between 30-72 years of age, with the median of 42 years(31). The risk of malignant transformation is low; only a case of endometrial carcinoma was reported associated with adenomatoid tumour of the fallopian tube (32). Our patient is 49 year-old, in the range reported in previous studies (31) (Table 1).

The incidence of adenomatoid tumours in specimens of hysterectomy have been estimate to be ~1% (33). Although the true incidence may be greater as these tumours are frequently neglected and probably unreported because of their small size and similar pathological appearance to leiomyomas (14, 34).

The histogenesis of adenomatoid tumours remains controversial; it is not uniformly agreed whether an adenomatoid tumor is a benign neoplasm or a form of localized mesothelial proliferation; mesonephric, mullerian, endothelial, and mesothelial origins have been suggested (19).

Studies based on transmission electron microscopy, scanning electron microscopy, immunohistochemistry, and the typical location of these lesions in genital areas/adrenal glands (that have in common their origin from steroidal crest) supported their mesothelial origin (14, 26, 34, 37, 41).

In our case, the immunochemistry expression of keratins (CK8/18) and calretinin in the tumour cells supported the mesothelial nature of the mAT.

Our patient had complains of severe persistent pelvic pain and dyspareunia, that were the reasons for vaginal hysterectomy, an unusual presentation described in literature for AT (3). Clinically the uterus was discretely enlarged; the pathology study confirmed the small volume of the uterus and allowed the diagnosis of mAT.

Indeed, the diagnosis of adenomatoid tumor is usually made as an incidental finding on hysterectomy specimens, as in the present case (Table 2).

According to previous reports, most of AT are solitary tumours. As described by others, we could not distinguish mAT from (the more frequent) multiple leiomyoma of the uterus by clinical examination or at ultrasound and so, initially, we suspected of multiple leiomyoma. In our case, we found a multifocal AT. Interestingly, the patient had no complains of abnormal uterine haemorrhages.

Since the adenomatoid tumour was multifocal in the present case, we speculate that the dimension and location of the adenomatoid tumour nodules may explain the painful symptomatology of the patient by interfering with the uterine vascularization.

Table 1 - Reports of genital female adenomatoid tumours. NA- Not available.

| Authors/References | Nº Cases | Age (range) | Location | Size of the AT (cm) | Immunohistochemical Test |
|---|---|---|---|---|---|
| **(16)** | 3 | 40-46 | Myometrium (1) and Fallopian tube (2) | 0.6 - 5 .0 | ++ Pancytokeratin calretinin and HBME-1 + Vimentin |
| **(17)** | 32 (Female) | 38-79 | Myometrium (26), Fallopian tube (4), Ovary (2) | 0.1 - 1.6 | ++ CKAE1/CAM 5.2, Calretinin, D2-40 in 100% of cases and for CK5/6 in 16% and Caldesmon in 3% |
| **(13)** | 25 | 26-55 | Myometrium (23), Fallopian tube (2) | 1.0 - 7.0 | ++ CK and alcian blue staining |
| **(1)** | 32 | 29-57 | Myometrium | 0.8 - 4.5 | 100% = + CK AE1/AE3, Calretinin, Vimentin and D2-40. 6% = + CK 5/6. |
| **(18)** | 2 | 36-39 | Myometrium | 6.0 - 7.0 | ++ CAM5.2 CK |
| **(3)** | 1 | 39 | Myometrium | 0.6 - 3.0 | ++ CK, CD34, HBME1 and Vimentin. |
| **(19)** | 1 | 40 | Myometrium | 5.2 | ++ CK |
| **(20)** | 1 | 39 | Fallopian tube | 11.0 | ++ CK, HBME1, Ca-125(M11) + Ca-125 (OC 125) |
| **(21)** | 2 | 36-60 | Myometrium and fallopian tube | 2.0 and unknown | ++ Calretinin and + CK |
| **(22)** | 1 | 33 | Myometrium | 5.0 | ++ CK and vimentin |
| **(23)** | 1 | 40 | Myometrium | 6.0 | ++ CK AE1-AE3 and calretinin |
| **(24)** | 1 | 43 | Myometrium | 2.0 | ++ CK and calretinin |
| **(32)** | 1 | 60 | Fallopian tube | 2.0 | ++ CK and vimentin |
| **(7)** | 2 | 33 and 36 | Myometrium | 0.5 - 3.5 | NA |
| **(25)** | 2 | 26 and 39 | Myometrium and Ovary | 2.5 - 11.5 | ++ Calretinin and + for CK 5 |
| **(36)** | 1 | 47 | Myometrium | 2.5 | NA |
| **(37)** | 1 | 38 | Myometrium | 8.0 | ++ CK |
| **(34)** | 2 | unknown | Myometrium | 12.0 - 13.0 | NA |
| **(38)** | 9 | 28-54 | Myometrium (7), Fallopian tube (1), Ovary (1) | 0.4 - 5.8 | ++ Calretinin and CK AE1/AE3 |
| **(39)** | 24 | unknown | Myometrium (21), Ovaries (2) and both myometrium and ovary (1) | 0.2 - 5.5 | ++ Calretinin, Vimentin and CK AE1/AE3 |
| **(40)** | 60 | unknown | Myometrium | 0.2 - 10 | ++ CK, calretinin, vimentin, and HMBE-1 |

Table 2 - Clinical findings/ Perioperative diagnosis of genital female adenomatoid tumours. NA – Not available

| Author | Clinical findings/ Perioperative diagnosis | Main complain |
|---|---|---|
| **(16)** | Incidental | NA |
| **(17)** | Incidental, 32% presumed leiomyoma, adenomyosis, endometriosis and unspecified mass | NA |
| **(13)** | Incidental | Infertility (3 cases), NA |
| **(1)** | Leiomyomas, adenomyosis, endometrial polyp, ovarian tumour, uterine prolapse | NA |
| **(18)** | Leiomyoma, endometriosis | Infertility, dysmenorrhea/ menorrhagia |
| **(3)** | Leiomyoma | Infertility |
| **(19)** | Leiomyoma | Menorrhagia |
| **(20)** | Uterine mass | Menorrhagia, pelvic pain |
| **(21)** | Uterine prolapse, incidental | NA |
| **(2)** | Leiomyoma and ovarian teratoma | Pelvic pain, menorrhagia and dysmenorrhea |
| **(22)** | Uterine mass | Infertility |
| **(23)** | Leiomyomas | Menorrhagia |
| **(24)** | Leiomyomas | Menorrhagia |
| **(7)** | Incidental, leiomyoma, ovarian cysts | Pelvic pain, menorrhagia |
| **(25)** | Leiomyoma, ovarian mass | Menorrhagia |

**Conclusion**

We report a rare case of a patient with a multifocal adenomatoid tumour of the myometrium, diagnosed after a hysterectomy performed to treat severe and persistent pelvic pain and enlarged uterus, interpreted as multiple leiomyoma. Definitive diagnosis was made in the pathologic study of the hysterectomy specimen.

In a patient with severe pelvic pain and an enlarged uterus by multiple nodules, suggesting multiple leiomyoma, a differential diagnosis with adenomatoid tumour should be considered.

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**Figures**

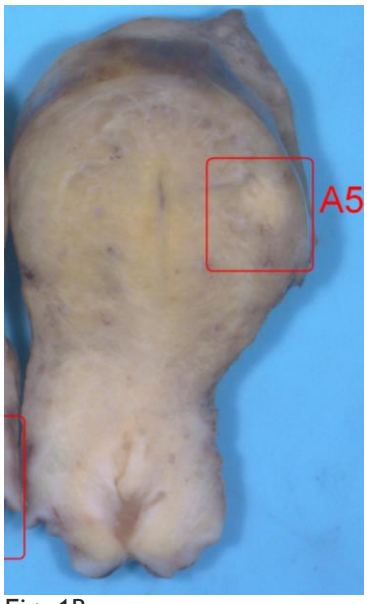


Fig. 1B

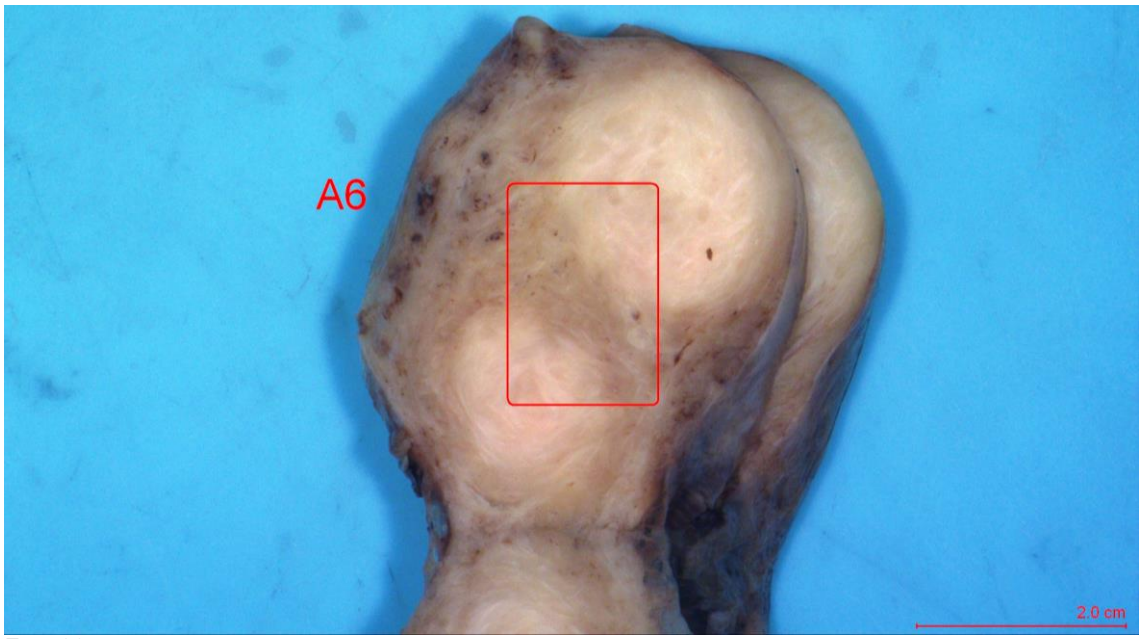


Fig. 1A

Figure 1 Uterus cut surface disclosing three discrete miometrial white nodules (two contiguous-A, and another by the serosa-B)

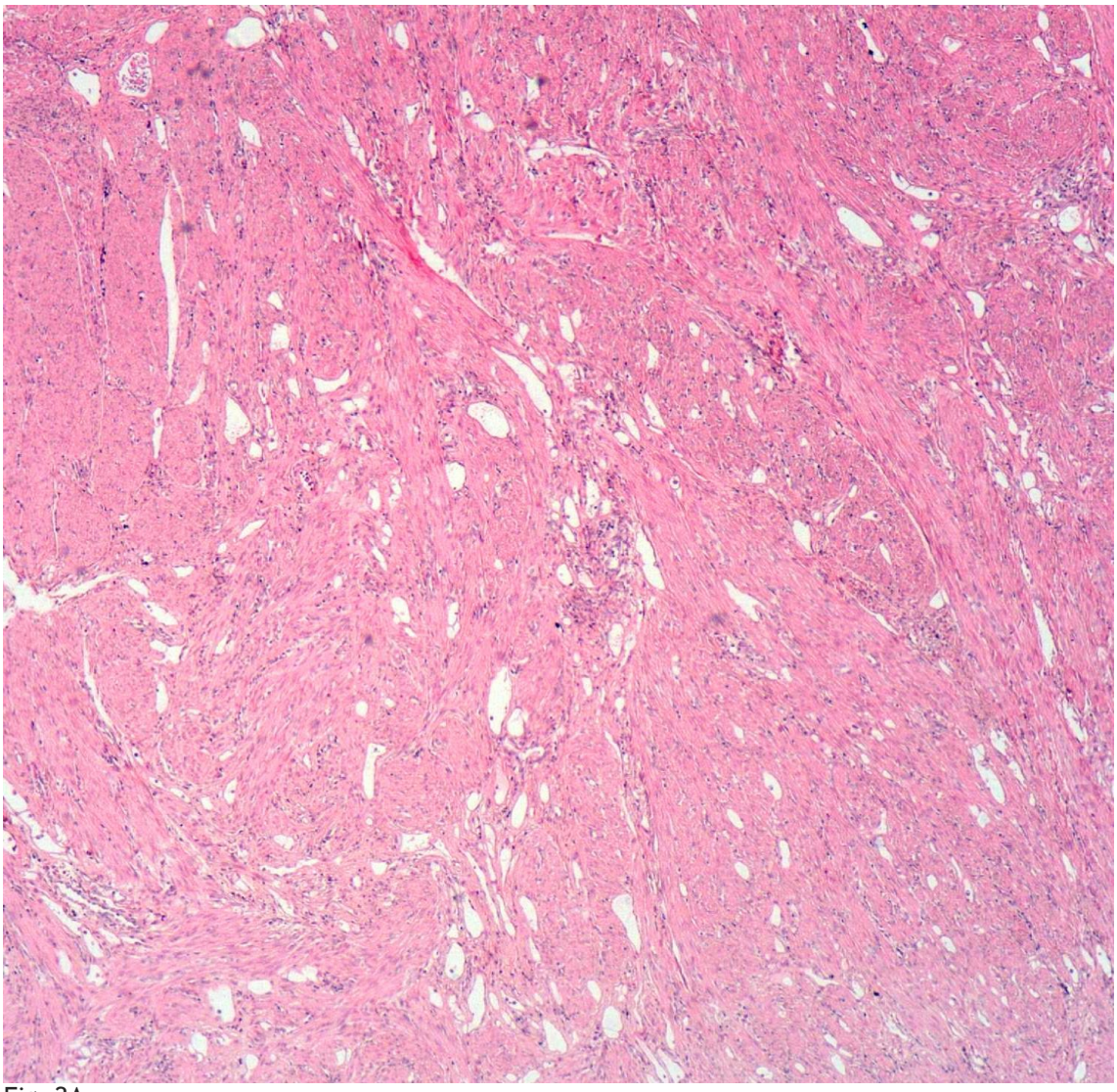

Fig. 2A

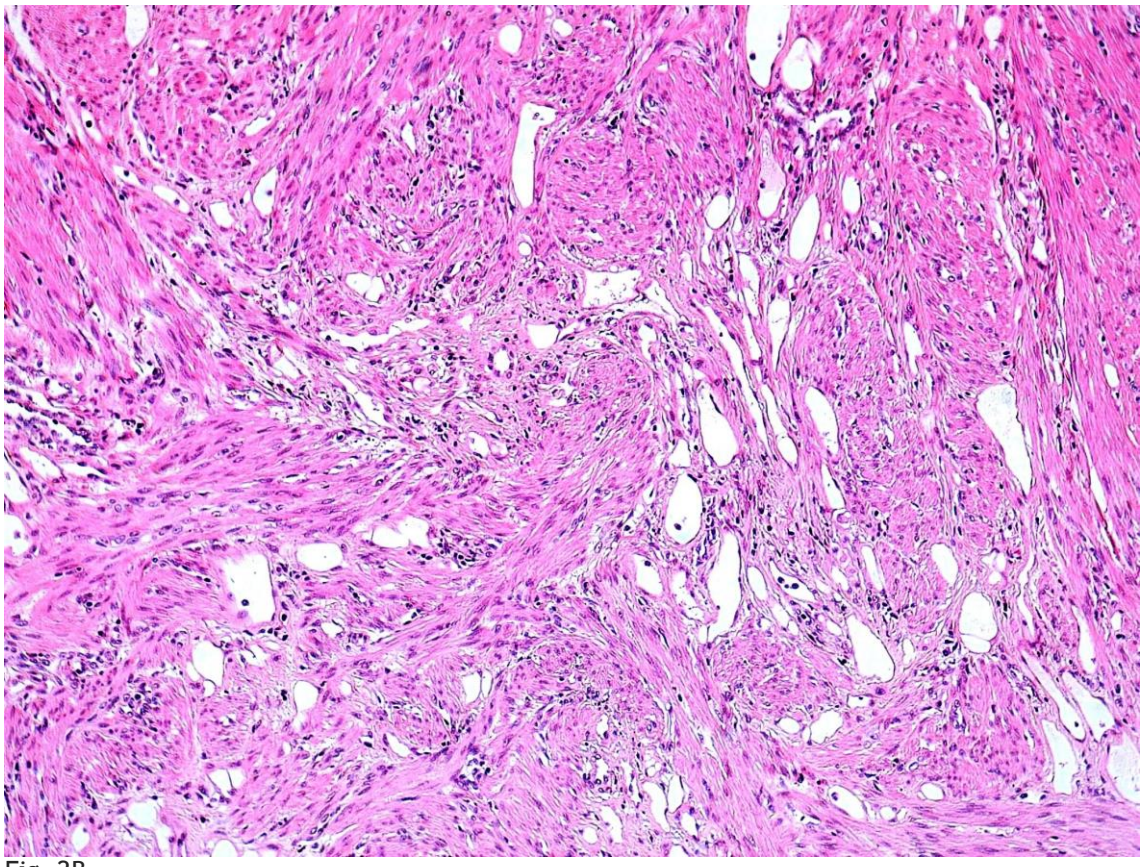

Fig. 2B

Figure 2: Uterine adenomatoid tumor: tubular structures dissociating the miometrial smooth muscle (A- H&E, x 20); the cells lining the tubules are flat and without atypia (B- H&E, x 100)

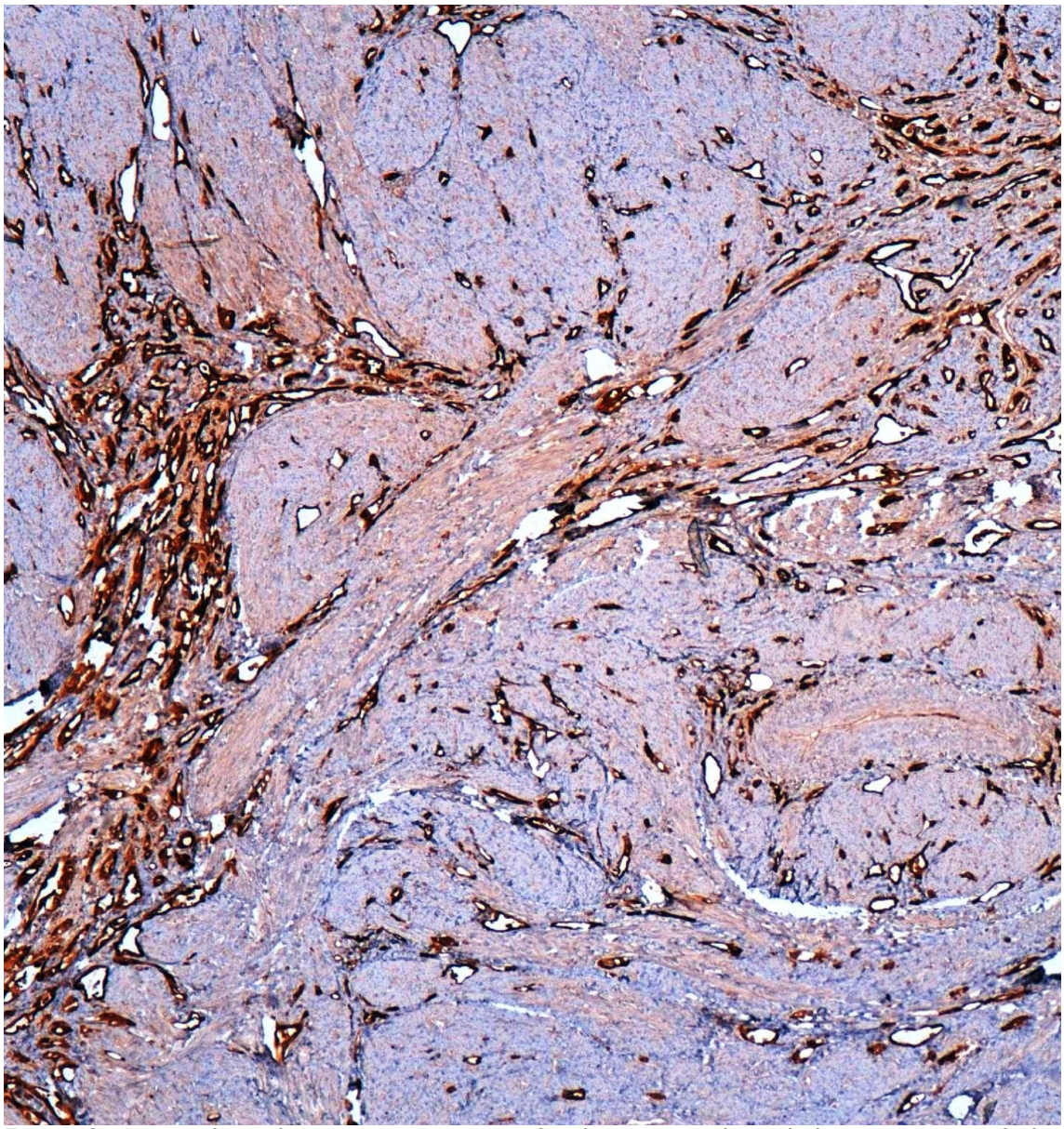

Figure 3: Immunohistochemistry expression of calretinin in the tubular structures of the adenomatoid tumor (ABC, x 40)