

Adenomyoma of the Cervix: Clues to Make the Radiological Diagnosis

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AUTHORS' CONTRIBUTIONS

Margarida Lima – Responsible for the data acquisition and writing of the manuscript.

Ana Quintas – Responsible for performing the clinical data acquisition, medical examination and referral of the patient to an oncological specialized hospital.

Teresa Margarida Cunha – Responsible for the original conception of the article, data acquisition, supervision of the drafting and revision of the manuscript.

DISCLOSURES

The authors declare no conflicts of interest or relevant financial associations related to this study.

ETHICAL STATEMENT/HUMAN AND ANIMAL RIGHTS

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

CONSENT

Yes.

ABSTRACT

Focal adenomyosis may present as a tumor-forming variant containing smooth-muscle cells, known as adenomyoma, which usually occurs in the uterine corpus. Cervical adenomyoma is a rare benign tumor, described for the first time in 1996, with 32 cases being reported since.

Typically affecting women of reproductive age, particularly between 21 and 56 years old, cervical adenomyomas present a diagnostic challenge due to distinct radiological features compared to the adenomyomas of the uterine corpus.

This report presents a case of cervical adenomyoma, emphasizing the role of radiological imaging in the diagnosis and clinical management.

CASE REPORT

BACKGROUND

This case adds to the limited body of knowledge by illustrating the imaging features of cervical adenomyoma and reinforcing the importance of considering it in the differential diagnosis of cervical masses. By emphasizing the diagnostic value of radiologic imaging, especially MRI, this report contributes to improved recognition and appropriate clinical management of this rare entity.

CASE REPORT

A 28-year-old Portuguese woman (gravida 0, para 0) presented to a private practice complaining of increasing vaginal watery discharge over the past 5 days. She denied abnormal bleeding, postcoital bleeding and dyspareunia. The patient had an ongoing Chlamydia infection. She had never had a cervical cytology and had not received Human Papilloma Virus (HPV) vaccination. There wasn't any history of travelling

to a tropical country and familial background was unremarkable.

During the medical examination, an enlarged uterine cervix was observed, with bulging of the posterior lip that showed intact mucosa with normal vascularization, raising suspicion of a cervical malignant tumor, and prompting referral to an oncological specialized hospital.

Imaging Findings

Magnetic resonance imaging (MRI) findings disclosed an exophytic polypoid multicystic tumor with regular borders predominantly located in the posterior lip of the cervix, measuring 43 x 29 x 20 mm, with a smaller component in the anterior lip, measuring 16 x 15 x 11 mm. The cysts varied in size from 2 to 7 mm, exhibiting high signal intensity on T2-weighted images (Figure 1) and low signal intensity in T1-weighted images (Figure 2), with fine regular septa showing hypointensity in both T2 and T1-weighted images. The mass didn't show any

areas of restricted diffusion (Figure 3) and the tumor wall and septa showed slight enhancement on gadolinium-enhanced T1 weighted images (Figure 4).

The list of differential diagnoses included cervical adenomyoma (CA), gastric-type adenocarcinoma (GAS), adenosarcoma (ADS), lobular endocervical glandular hyperplasia (LEGH), mesonephric remnant hyperplasia (MH), and tunnel clusters (TC).

Management

Serum tumor markers such as carbohydrate antigen-125 and cancer antigen 19-9 were within normal limits. A cervical cytology, a punch biopsy from the surface of the tumor and a core needle biopsy of the mass were performed with inconclusive results. A conization was performed and the diagnosis of CA was made. The patient wished to preserve her fertility, so a polypectomy was performed.

Follow-up

At the 6-month follow-up appointment the patient had no signs of recurrence.

DISCUSSION

Etiology & demographics

Adenomyosis is a benign condition that affects premenopausal women, characterized by the presence of ectopic endometrium within the myometrium. It can be classified as diffuse or focal [1]. Adenomyomas are specific regions of focal adenomyosis surrounded by hypertrophic myometrium [2]. CA are rare benign lesions with unknown etiology, with only 32 cases reported worldwide, and have been observed in women ranging in age from 21 to 56 years, with a mean age of 40 years old [3].

Clinical & imaging findings

Most of the patients are asymptomatic, but symptoms like vaginal mucinous or watery discharge, abnormal bleeding and dyspareunia have been described [3-7].

CA usually presents as a polypoid mass with intact mucosa but may also be intramural. Although pre-operative diagnosis is difficult, common radiological features have been described in this tumor. Previous case reports comprising radiological evaluation of these tumors with MRI consistently displayed a large exophytic polypoid multicystic tumor with regular borders with cystic component showing high signal intensity in T2-weighted images and thin septa presenting hypointensity in both T2 and T1-weighted images. CA doesn't show any areas of restricted diffusion. The tumor wall and septa slightly enhance on gadolinium-enhanced T1-weighted MRI [3,4,6,7].

The pathological features are of a well-circumscribed lesion composed of irregularly shaped benign endocervical glands,

admixed with smooth muscle. Basal nucleus and pale cytoplasm are features of endocervical cells with tubal or tubo-endometrioid metaplasia. Desmoplasia and mitosis aren't present [5].

No reports of malignant transformation were found.

Treatment & prognosis

Appropriate treatment is still unclear [3,5,6]. Most of the published cases have undergone a hysterectomy with only five being treated by adenomyectomy. *Matsuzaki et al.* described a case of a CA resected by vaginal polypectomy that didn't show signs of recurrence at the 9-month follow-up. Despite that, recurrence is suspected to be higher in patients treated with polypectomy [3].

Differential Diagnoses

Gastric- type adenocarcinoma

GAS, formerly known as minimal deviation adenocarcinoma, represents a rare subtype of mucinous endocervical adenocarcinoma that stands as the primary differential diagnosis of CA. [9,10]. Unlike typical cervical adenocarcinomas, GAS is not causally linked to HPV infection. [10,11]. Mean age of presentation is 50 years old [10]. On MRI it manifests as a non-circumscribed gadolinium-enhancing polypoid mass containing multiple microcysts, with solid components exhibiting restricted diffusion. Histologically, this tumor presents with gastric foveolar or pyloric type epithelium infiltrating the underlying stroma. Due to its rarity and often late presentation, diagnosis commonly occurs at an advanced stage with parametrial invasion and lymphadenopathy frequently observed [10-12]. GAS is associated with a worse prognosis and lower survival rate than squamous cell carcinoma [11].

Adenosarcoma

ADS are uncommon tumors characterized by a combination of benign glandular tissue and malignant stromal components, occurring more frequently in the uterus of post-menopausal women [13]. While primarily found in the uterus, they can also manifest in the cervix and other extra-uterine sites [13,14]. Cervical ADS occurs in younger women than the uterine counterpart, with a median age at the time of presentation ranging between 37 and 39 years [14]. Notably, in a case series by *Yuan et al.*, it was observed that women with cervical ADS had a median age of 34 years, in contrast to those with uterine ADS, who had a median age of 50 years. Clinically and radiologically, lesions are often mistaken for endometrial or endocervical polyps [15]. On MRI, cervical ADS typically presents as a large, irregularly shaped polypoid mass with regions of heightened T2 signal intensity, possibly indicating internal tissue necrosis. Apparent diffusion coefficient (ADC) values are usually higher compared to other uterine sarcomas. Contrast-enhancement on MRI is typically moderate, resulting in a tumor appearance that resembles the surrounding myometrium. However, suspicion of sarcomatous overgrowth arises if signs such as invasion into the

myometrium, growth within blood vessels or outside the uterus, and distant metastases are observed [13,15].

CA differential diagnosis includes also benign glandular lesions that can be potentially misdiagnosed as malignant entities like LEGH, MH, and TC [10].

Lobular endocervical glandular hyperplasia

LEGH is characterized by discrete lobules of small glands displaying gastric-type differentiation, resembling pyloric glands, mainly situated in the upper segment of the endocervical canal. It lacks an association with persistent high-risk HPV infection, and GAS constitutes its primary differential diagnosis [16]. Both LEGH and GAS of the uterine cervix have been associated with Peutz-Jeghers syndrome [16,11].

MRI findings may reveal either a cluster of microcysts with high signal intensity on T2-weighted images or a central accumulation of numerous microcysts surrounded by larger cysts, described as a "Cosmos" pattern [17]. Conversely, CA typically presents as a well-circumscribed mass surrounded by a myomatous stromal component [3].

Tunnel clusters

TC denotes a specific type of mucinous retention cysts in the uterine cervix, predominantly occurring in pregnant women and characterized by multilocular cystic lesions without a solid component [11]. Cysts predominantly show low signal intensity in T1-weighted images, with occasional rare occurrences exhibiting heightened signal intensity attributed to mucin, and hyperintensity in T2. Additionally, they typically reveal an absence of enhancement [11].

Mesonephric remnant hyperplasia

MH presents as a proliferation of small glands resembling prostatic carcinoma, predominantly observed in the female genital tract, particularly in women with an average age of 60 years old [18,19]. In the uterine cervix, MH typically exhibits a lobular morphology, characterized by clustered, small to medium-sized, round mesonephric tubules, that didn't regress during development. MRI evaluation shows multiple cystic lesions, measuring less than 3 mm in diameter showing a high signal intensity on T2-weighted MRI, in the bilateral aspects of the variably enlarged uterine cervix. Occasionally, diffuse manifestation may occur, characterized predominantly by a non-clustered, extensive and diffuse proliferation of mesonephric tubules separated by varying amounts of cervical stroma [3].

TEACHING POINT

This article highlights the diagnostic challenges posed by cervical adenomyoma, a rare benign tumor, often mistaken for malignant neoplasms due to its distinct radiological features that present as an exophytic polypoid multicystic tumor with thin septa that don't show restricted diffusion and present only slight homogenous enhancement on contrast-enhanced

MRI sequences. Understanding these characteristic imaging features is crucial for accurate diagnosis and appropriate clinical management, particularly in women of reproductive age presenting with increased watery discharge.

QUESTIONS

Question 1: Which is a clinical manifestation of cervical adenomyoma?

- a) Hematochezia.
- b) Urinary frequency.
- c) Increased appetite.
- d) Increased vaginal watery discharge. (applies)
- e) Respiratory distress.

Explanation:

a) Not a symptom of cervical adenomyoma. [Most of the patients are asymptomatic, but symptoms like abnormal bleeding, vaginal mucinous or watery discharge and dyspareunia have been described [3-7].]

b) Not a symptom of cervical adenomyoma. [Most of the patients are asymptomatic, but symptoms like abnormal bleeding, vaginal mucinous or watery discharge and dyspareunia have been described [3-7].]

c) Not a symptom of cervical adenomyoma. [Most of the patients are asymptomatic, but symptoms like abnormal bleeding, vaginal mucinous or watery discharge and dyspareunia have been described [3-7].]

d) Increased vaginal watery discharge is one of the most common symptoms of cervical adenomyoma. [Most of the patients are asymptomatic, but symptoms like abnormal bleeding, vaginal mucinous or watery discharge and dyspareunia have been described [3-7].]

e) Not a symptom of cervical adenomyoma. [Most of the patients are asymptomatic, but symptoms like abnormal bleeding, vaginal mucinous or watery discharge and dyspareunia have been described [3-7].]

Question 2: Which of the following cervical lesions is associated with Peutz-Jeghers syndrome?

- a) Cervical adenomyoma (CA).
- b) Squamous-cell carcinoma of the uterine cervix (SCC).
- c) Tunnel clusters (TC).
- d) Lobular endocervical glandular hyperplasia (LEGH). (applies)
- e) Adenosarcoma (ADS).

Explanation:

a) Unknown etiology, association with Peutz-Jeghers syndrome hasn't been described. [Both LEGH and gastric-type adenocarcinoma of the uterine cervix have been associated with Peutz-Jeghers syndrome [16,11].]

b) Association with Peutz-Jeghers syndrome hasn't been described. [Both LEGH and gastric-type adenocarcinoma of the uterine cervix have been associated with Peutz-Jeghers syndrome [16,11].]

c) Association with Peutz-Jeghers syndrome hasn't been described. [Both LEGH and gastric-type adenocarcinoma of

the uterine cervix have been associated with Peutz-Jeghers syndrome [16,11].]

d) Association with Peutz-Jeghers syndrome is known. [Both LEGH and gastric-type adenocarcinoma of the uterine cervix have been associated with Peutz-Jeghers syndrome [16,11].]

e) Association with Peutz-Jeghers syndrome hasn't been described. [Both LEGH and gastric-type adenocarcinoma of the uterine cervix have been associated with Peutz-Jeghers syndrome [16,11].]

Question 3: What is the primary differential diagnosis of cervical adenomyoma that is not causally linked to human papillomavirus (HPV) infection and often diagnosed at an advanced stage?

- a) Tunnel clusters (TC).
- b) Adenosarcoma (ADS).
- c) Gastric-type adenocarcinoma (GAS). (applies)
- d) Mesonephric remnant hyperplasia (MH).
- e) Lobular endocervical glandular hyperplasia (LEGH).

Explanation:

a) Not the primary diagnosis of cervical adenomyoma. [GAS, formerly known as minimal deviation adenocarcinoma, represents a rare subtype of mucinous endocervical adenocarcinoma that stands as the primary differential diagnosis of cervical adenomyoma [9,10]. Unlike typical cervical adenocarcinomas, GAS is not causally linked to human papillomavirus (HPV) infection.]

b) Not the primary diagnosis of cervical adenomyoma. [GAS, formerly known as minimal deviation adenocarcinoma, represents a rare subtype of mucinous endocervical adenocarcinoma that stands as the primary differential diagnosis of cervical adenomyoma [9,10]. Unlike typical cervical adenocarcinomas, GAS is not causally linked to human papillomavirus (HPV) infection.]

c) Gastric-type adenocarcinoma is an HPV independent lesion of the cervix and it's the primary differential diagnosis of cervical adenomyoma. [GAS, formerly known as minimal deviation adenocarcinoma, represents a rare subtype of mucinous endocervical adenocarcinoma that stands as the primary differential diagnosis of cervical adenomyoma [9,10]. Unlike typical cervical adenocarcinomas, GAS is not causally linked to human papillomavirus (HPV) infection.]

d) Not the primary diagnosis of cervical adenomyoma. [GAS, formerly known as minimal deviation adenocarcinoma, represents a rare subtype of mucinous endocervical adenocarcinoma that stands as the primary differential diagnosis of cervical adenomyoma [9,10]. Unlike typical cervical adenocarcinomas, GAS is not causally linked to human papillomavirus (HPV) infection.]

e) It's associated with Peutz-Jeghers syndrome and its HPV-independent, but it isn't the primary differential diagnosis of cervical adenomyoma. [GAS, formerly known as minimal deviation adenocarcinoma, represents a rare subtype of mucinous endocervical adenocarcinoma that stands as the primary differential diagnosis of cervical adenomyoma [9,10].]

Unlike typical cervical adenocarcinomas, GAS is not causally linked to human papillomavirus (HPV) infection.]

Question 4: What is a characteristic feature of lobular endocervical glandular hyperplasia (LEGH) on MRI?

- a) "Cosmos" pattern of cysts. (applies)
- b) Low signal intensity on T2-weighted images.
- c) Being located in the exocervix.
- d) Solid components with restricted diffusion.
- e) Well-circumscribed mass surrounded by myomatous stromal component.

Explanation:

a) 'Cosmos pattern' has been described as a finding on MRI of lobular endocervical glandular hyperplasia. [MRI findings may reveal either a cluster of microcysts or a central accumulation of numerous microcysts surrounded by larger cysts, described as a 'cosmos' pattern.]

b) Cysts typically show high signal intensity on T2-weighted images. [MRI findings may reveal either a cluster of microcysts with high signal intensity on T2-weighted images.]

c) The upper segment of the endocervical canal is the most common location. [LEGH is characterized by discrete lobules of small glands displaying gastric-type differentiation, resembling pyloric glands, mainly situated in the upper segment of the endocervical canal.]

d) Solid components with restricted diffusion are seen on cervical malignancies like Gastric-type adenocarcinoma. [On MRI it manifests as a non-circumscribed gadolinium-enhancing polypoid mass containing multiple microcysts, with solid components exhibiting restricted diffusion.]

e) A myomatous stromal component is usually seen on cervical adenomyoma. [Cervical adenomyoma typically presents as a well-circumscribed mass surrounded by a myomatous stromal component.]

Question 5: Which imaging finding is typically observed on MRI evaluation of cervical adenomyoma?

- a) Solid mass with heightened T1 signal intensity.
- b) Irregular borders and central necrosis.
- c) Exophytic polypoid multicystic tumor with regular borders. (applies)
- d) High signal intensity in T1-weighted images.
- e) Diffuse infiltration into surrounding tissues.

Explanation:

a) Cervical adenomyoma shows thin solid septa that show low signal intensity on T2 and T1-weighted images. [Large multicystic exophytic mass in the cervix with multi-sized cysts and regular borders, with high signal intensity in T2-weighted images and with fine regular septa showing hypointensity in both T2 and T1-weighted images.]

b) Cervical adenomyoma shows regular borders. [Large multicystic exophytic mass in the cervix with multi-sized cysts and regular borders, with high signal intensity in T2-weighted images and with fine regular septa showing hypointensity in

both T2 and T1-weighted images.]

c) Cervical adenomyoma most typically presents as an exophytic polypoid multicystic tumor with regular borders. [Large multicystic exophytic mass in the cervix with multi-sized cysts and regular borders, with high signal intensity in T2-weighted images and with fine regular septa showing hypointensity in both T2 and T1-weighted images.]

d) The multicystic component shows low signal intensity on T1 weighted images. [Large multicystic exophytic mass in the cervix with multi-sized cysts and regular borders, with high signal intensity in T2-weighted images and with fine regular septa showing hypointensity in both T2 and T1-weighted images.]

e) Cervical adenomyoma is a well-circumscribed lesion. [Conversely, cervical adenomyoma typically presents as a well-circumscribed mass surrounded by a myomatous stromal component [3].]

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FIGURES

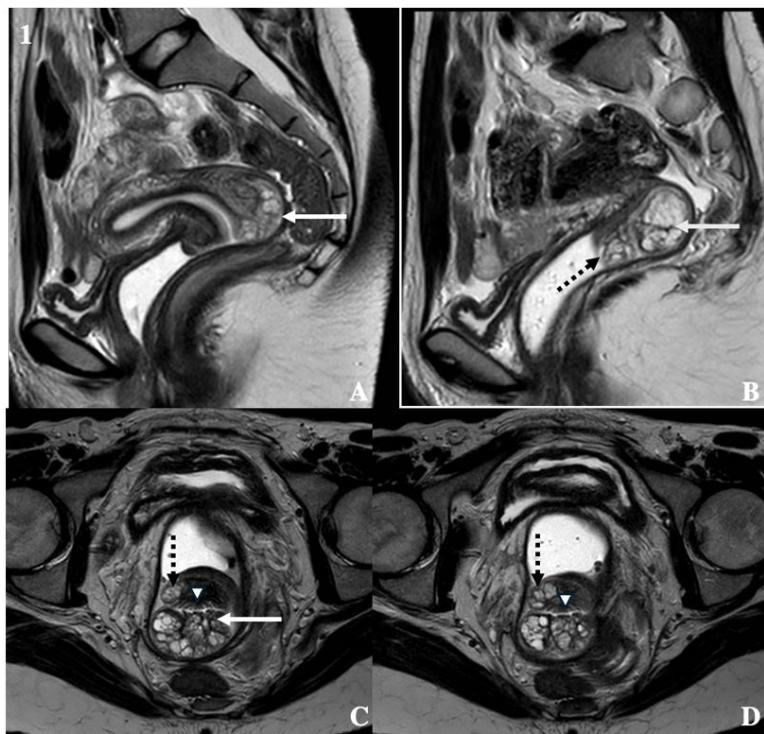


Figure 1: 28-year-old female diagnosed with a cervical adenomyoma.

Findings: A and B) Sagittal T2-weighted MRI of the pelvis showing an exophytic polypoid multicystic cervical tumor with regular borders, predominantly located on the posterior lip (white arrow), measuring 43 x 29 x 20 mm, with a smaller component on the anterior lip (dashed arrow), measuring 16 x 15 x 11 mm. No parametrial invasion is seen. C and D) Oblique axial T2-weighted MRI of the pelvis showing a well-defined multicystic tumor. Multicystic tumor on the anterior lip (dashed arrow) and cervical plicae palmatae are seen (arrowhead). Fine regular hypointense septa (white arrow) between the cysts is observed.

Technique: MRI 1.5 Tesla. Endovaginal filling with ultrasound gel is observed. A and B) Sagittal T2-weighted MRI of the pelvis . C and D) Axial of the cervix T2-weighted MRI of the pelvis.

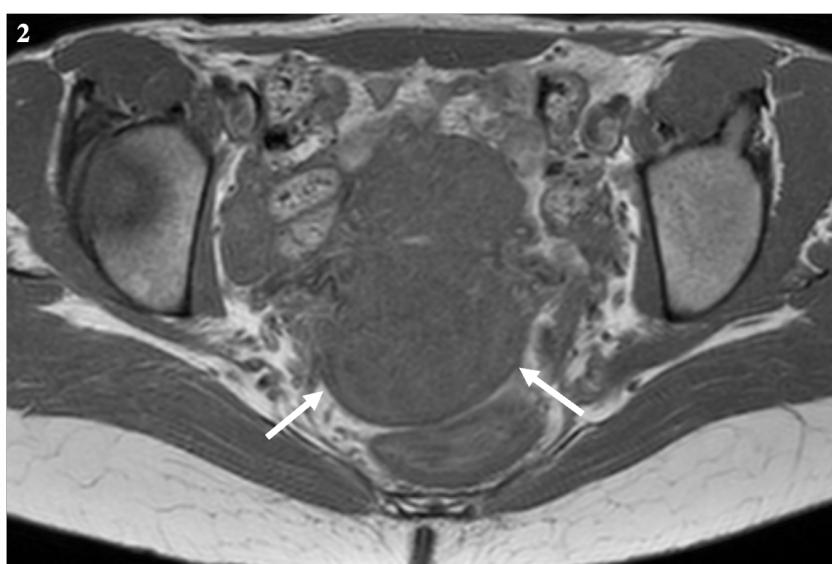


Figure 2: 28-year-old female diagnosed with a cervical adenomyoma.

Findings: Axial T1-weighted MRI of the pelvis showing an hypointense enlargement of the posterior lip of the cervix (white arrow).

Technique: MRI 1.5 Tesla. Axial T1-weighted MRI.

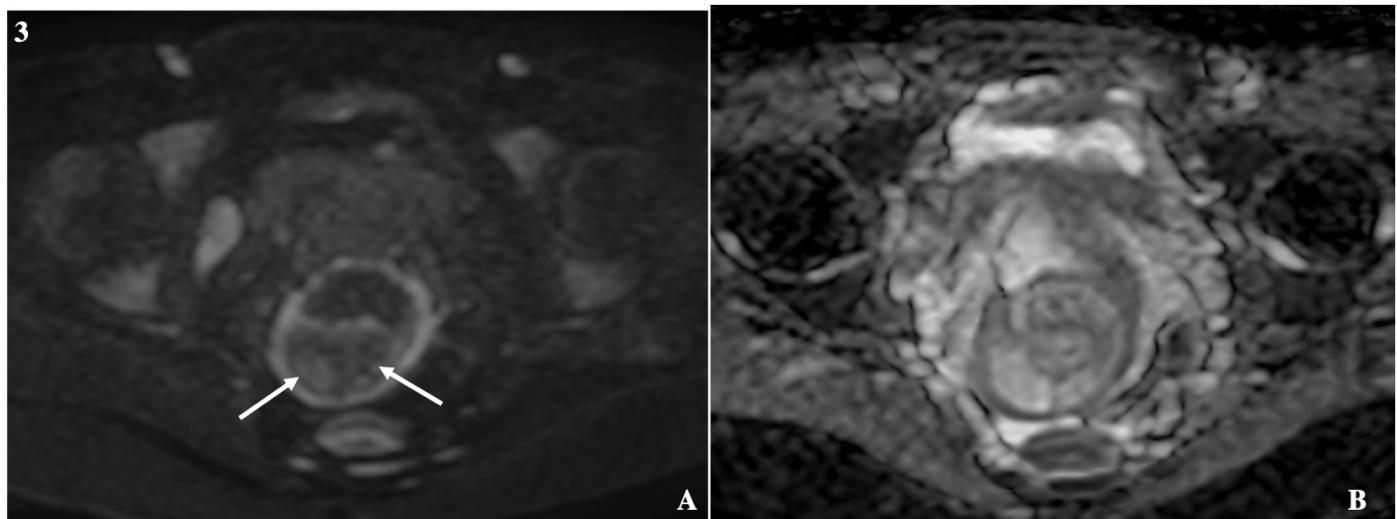


Figure 3: 28-year-old female diagnosed with a cervical adenomyoma.

Findings: A and B) Axial diffusion-weighted image ($b = 1000\text{s/mm}^2$) and ADC map images: The tumor doesn't show restricted diffusion (white arrows).

Technique: MRI 1.5 Tesla. A) Axial DWI ($b = 1000\text{s/mm}^2$) MRI B) ADC map.

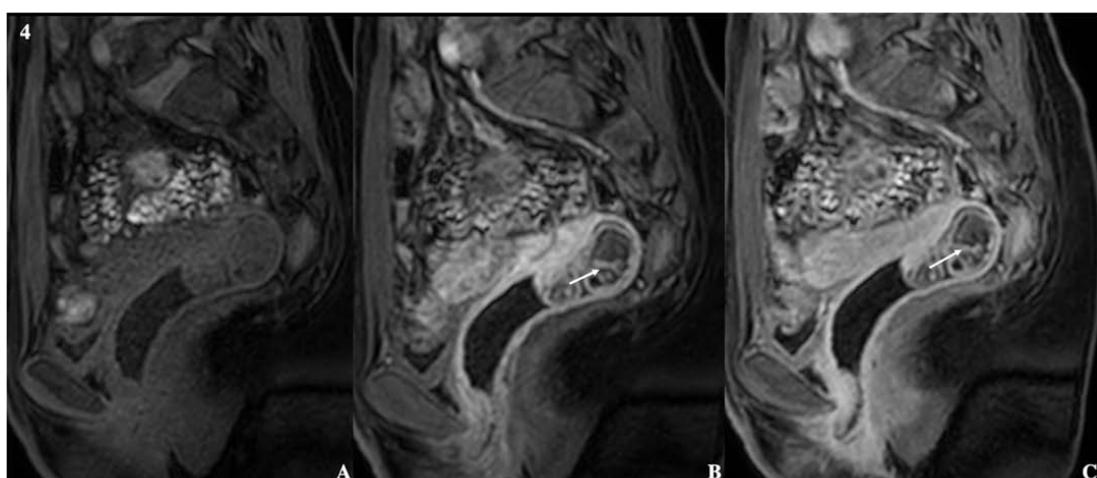


Figure 4: 28-year-old female diagnosed with a cervical adenomyoma.

Findings: A) Sagittal T1-weighted image with fat suppression showing an hypointense polypoid cervical tumor. B) Sagittal contrast-enhanced T1-weighted image with fat suppression showing a slight homogenous enhancement of the tumor wall and thin septa (white arrow) that persisted on the late-phase (C).

Technique: MRI 1.5 Tesla. A) Sagittal T1-weighted image with fat suppression. B) Sagittal contrast-enhanced T1-weighted image with fat suppression. C) Sagittal late phase contrast-enhanced T1-weighted image with fat suppression.

Table 1: Summary table of Cervical Adenomyoma.

Etiology	Unknown.
Incidence	Unknown. 32 cases have been described.
Gender ratio	Non applicable.
Age predilection	Has been described in women within an age range from 21 to 56 years old with a mean age of 40 years old.
Risk factors	Unknown.
Treatment	Polypectomy; Hysterectomy.
Prognosis	Recurrence has been described when polypectomy is performed.
Findings on imaging	Ultrasound: Large isoechoic multicystic soft mass. MRI: Large exophytic polypoid multicystic tumor in the uterine cervix with regular borders.

Table 2: Differential diagnosis table for Cervical Adenomyoma.

Differential Diagnosis	MRI
Cervical adenomyoma	Large multicystic exophytic mass in the cervix with multi-sized cysts and regular borders, with high signal intensity in T2-weighted images and with fine regular septa showing hypointensity in both T2 and T1-weighted images. Doesn't show any areas of restricted diffusion. Pattern of contrast enhancement – The tumor wall and septa slightly enhance on gadolinium-enhanced T1-weighted MRI.
Gastric-type adenocarcinoma	Cervical irregular mass with intermediate signal on T2-weighted images, that commonly invades the cervical stroma. May be purely solid or show clusters of microcysts that are hyperintense on T2-weighted images and hypointense in T1-weighted images. Solid components show restricted diffusion. Pattern of contrast enhancement – Solid parts of the tumor show heterogenous enhancement while the microcysts wall is slightly enhancing.
Adenosarcoma	Large, irregularly shaped polypoid cervical mass. Some portions of the mass may exhibit regions of heightened T2 signal intensity, indicative of internal tissue necrosis. ADC map values are usually higher than in other sarcomas. Pattern of contrast enhancement – Heterogeneous and moderate, resulting in a tumor appearance similar to that of the surrounding myometrium.
Lobular endocervical glandular hyperplasia	Aggregation of small cysts that are hyperintense on T2-weighted images and hypointense in T1-weighted images and may be displaced in a 'Cosmos pattern': central microcystic components surrounded by medium-sized to large cysts. Pattern of contrast enhancement – May show homogeneous slight enhancement of the cysts wall.
Mesonephric remnant hyperplasia	Multiple cystic lesions measuring less than 3 mm in diameter showing a high signal intensity on T2-weighted MRI, in the bilateral aspects of the variably enlarged uterine cervix. Pattern of contrast enhancement – May show homogeneous slight enhancement of the cysts wall.
Tunnel clusters	Cluster of mucinous retention cysts that are hyperintense on T2-weighted images and usually show hypointensity on T1-weighted images. May show high signal intensity on T1 due to the presence of mucin. Pattern of contrast enhancement – There is no associated enhancement.

KEY WORDS

Cervical adenomyoma, MRI, Differential diagnosis, Gastric-type adenocarcinoma, watery discharge.

ABBREVIATIONS

MRI = MAGNETIC RESONANCE IMAGING
HPV = HUMAN PAPILLOMA VIRUS
CA = CERVICAL ADENOMYOMA
ADC = APPARENT DIFFUSION COEFFICIENT
DWI = DIFFUSION-WEIGHTED IMAGE
GAS = GASTRIC-TYPE ADENOCARCINOMA
ADS = ADENOSARCOMA
LEGH = LOBULAR ENDOCERVICAL GLANDULAR HYPERPLASIA
MH = MESONEPHRIC REMNANT HYPERPLASIA
TC = TUNNEL CLUSTERS

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