

Adenomyomas of the Uterine Cervix in the First-Trimester of Pregnancy: A Case Report

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Abstract

Cervical adenomyomas of endocervical type (endocervical adenomyomas) are very rare benign lesions. Here we report the case of a 33-year-old woman who referred to the Perinatology Clinic of Ommolbanin Hospital (Mashhad, Iran) in September 2017. The patient was 8 weeks pregnant and complained of spotting and feeling a mass protruding from her vagina for 2 months. Physical examination revealed the presence of three masses of approximately 10 cm in the vagina, which were treated surgically. Histopathological examination of the excised specimen showed the presence of glands lined by a single layer of endocervical-type mucinous epithelium with smooth muscle fibers. Clinicians should be aware of such lesions in order to differentiate them from other malignancies and to individualize treatment.

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What's Known

- Adenomyoma of the uterine cervix displaying simultaneous proliferation of the gland and stroma is very rare. During a pelvic examination, adenomyoma appears as an endometrial polyp projecting into the cervix. Uterine adenomyomas are uncommon lesions that can be misdiagnosed as other lesions of the uterine. Hence it is important to distinguish it from a malignant lesion.
- There are only a few case reports on adenomyomas of the uterine cervix.

What's New

- The first case report on an adenomyoma of endocervical type in an Iranian patient.
- Uterine adenomyoma should be considered in differential diagnosis with other cervical lesions.

Introduction

Uterine adenomyoma is usually found in the uterine corpus, but rarely in the uterine cervix.¹ Adenomyomas of the uterine cervix are benign and uncommon lesions that involve benign endocervical-type glands and stroma. This type of tumor is classified as mixed epithelial and mesenchymal tumors of the uterine cervix.² The majority of middle-aged women have some sort of medical problem associated with menstruation such as abnormal bleeding, vaginal discharge, and a polyp or fibroid protruding through the external cervical os as well as concerns about malignancies. A less common clinical feature of cervical adenomyoma is cervical enlargement by a mural mass without mucosal involvement. Adenomyomas may be asymptomatic and could incidentally be revealed during a regular gynecologic examination.² The biological behavior of polypoid endocervical adenomyoma is benign. There is no evidence that adenomyomas are premalignant or associated with an increased risk of subsequent development of malignancy.^{1,2} The differential diagnosis includes lobular endocervical glandular hyperplasia, adenofibroma, endocervicosis, tunnel clusters, adenoma malignum, and adenosarcoma.^{2,3}

Some pathologists are not familiar with this type of lesion and thus there exists a risk of misdiagnosis. To date, there have been two case series, each including ten cases, on adenomyomas of endocervical type. However, only a few case reports are available in the literature.³⁻⁷ To the best of our knowledge, we describe the first case report on adenomyoma of endocervical type in an Iranian patient.

Case Report

A 33-year-old woman (gravida2, para 2, abortion 1) was referred to the Perinatology Clinic of Ommolbanin Hospital (Mashhad, Iran) in September 2017. The patient was at a gestational age of 8 weeks and presented with a history of spotting and feeling a mass protruding from her vagina through the introitus for 2 months. She had no gynecologic examination during the previous 2 years and did not have a family history or genetic profile of a specific illness. The patient underwent an abdominal ultrasound as she was concerned about having a miscarriage. The results confirmed the presence of a live embryo within the uterus and no additional issues were reported.

Upon admittance, the gynecologic examination revealed the presence of three masses of approximately 10 cm in the vagina with superficial ulceration (figure 1). Subsequently, she was treated by polypectomy as an outpatient without anesthesia. Histopathological examination of the excised specimen showed that the tumor was composed of glands lined by a layer of endocervical-type epithelium surrounded by stroma with smooth muscle. No nuclear atypia or cytological atypia was noted. Mitotic activity was absent in both epithelial and smooth muscle components and no desmoplastic response was evident (figure 2, 3). The symptoms improved after the polypectomy and did not recur during the following 6 months. The rest of the pregnancy was uneventful. Written informed consent was obtained from the patient for the publication of this article and accompanying images.

Discussion

Cervical adenomyoma (endocervical adenomyoma) is an uncommon benign lesion.¹ The usual complaints due to cervical adenomyoma are spotting, hemorrhage, or copious discharge which might be asymptomatic. The tumor is usually polypoid, but can be intramural or a mass projecting into the pelvis from the cervix.² The pathological features are unencapsulated and well-circumscribed lesions 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. Mitosis is not seen in the epithelial nor in the smooth muscle components, and desmoplasia is absent.^{2,3}

To date, there have been two case series, each including ten cases, on adenomyomas of endocervical type.^{3,4} The first case series,

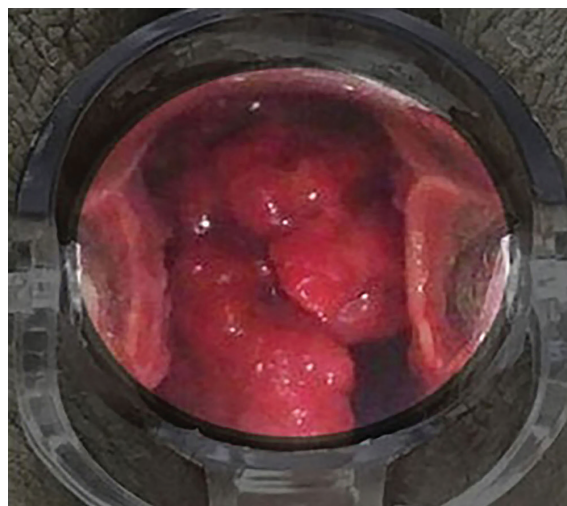


Figure 1: The cervical mass following physical examination.

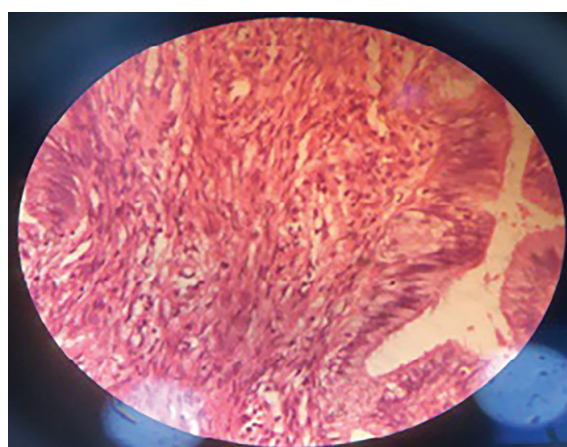


Figure 2: Endocervical type glands surrounded by stroma rich in smooth muscle with no atypia in the glands and stroma (H&E; $\times 400$ original magnification).

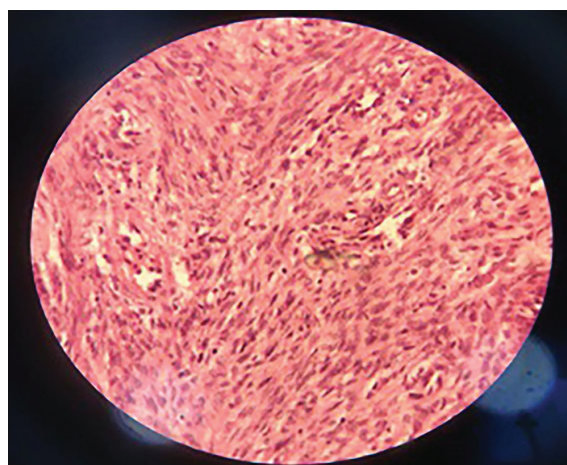


Figure 3: The proliferation of endocervical type glands surrounded by stroma rich in smooth muscle with no atypia in the glands and stroma (H&E; $\times 100$ original magnification).

by Gilks and colleagues, reported that most patients did not have any symptoms except for two patients who had abnormal vaginal bleeding.³ The differential diagnosis included

lobular endocervical glandular hyperplasia, adenofibroma, endocervicosis, tunnel clusters, adenoma malignum, and adenosarcoma.^{3, 4} The most common differential diagnosis was adenoma malignum. Polypoid endocervical adenomyoma had benign biological behavior, but adenoma malignum presents as an infiltrative mass, diffusely expanding into the cervical wall, mild nuclear atypia, and periglandular stromal desmoplasia.²⁻⁴ Gross pathology helps to differentiate between endocervical adenomyoma (polypoid with sharp margins) and adenoma malignum (diffuse expansion of lesion without polyp appearance). A desmoplastic reaction without atypia indicates adenomyoma.^{4, 5}

The second case series, by Casey and McCluggage, reported a rare case of cervical mesonephric adenomyoma; another differential diagnosis. This lesion had lobular dilated glands without mucin-secreting appearance and cuboidal epithelium.⁴ Protopapas and colleagues reported the case of a 41-year-old nulliparous patient. Hysteroscopy evaluation revealed an endocervical atypical polypoid adenomyoma (APAM). The patient was treated with hysteroscopic resection of the APAM and polyps. The patient's symptoms regressed after polypectomy.⁸

In the case of our patient, the histopathology report indicated the proliferation of endocervical type glands surrounded by stroma rich in smooth muscle with no atypia in the glands and stroma. These morphological features were largely similar to those reported in other studies.³⁻⁷ On the last note, we believe that our patient incorrectly associated vaginal bleeding with complications such as miscarriage, ectopic pregnancy, and molar pregnancy. Therefore, it is required that physicians are aware of cervical lesions and perform a speculum examination on every pregnant woman with vaginal bleeding. The main limitation of the present study was the lack of investigation of the immunohistochemistry of surgical specimens.

Conclusion

Adenomyomas of the uterine cervix should be considered upon detection of any cervical mass and differentiation should be made between benign and malignant lesions for correct treatment.

Conflict of Interest: None declared.

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