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Recurrent cervical adenofibroma progressing to adenosarcoma: a rare case report

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ABSTRACT

Objectives: Cervical adenofibroma is a rare form of mixed mesodermal tumor that can present as cervical polyps with a tendency for local recurrence and progression. Few cases progressing to adenosarcoma have previously been reported. We report a case of cervical adenofibroma progressing to adenosarcoma, and we seek to remind clinicians of the method and importance of the differential diagnosis of this disease.

Material and methods: A fertile woman was admitted in our department for the eighth recurrence of a cervical polypoidal mass which for the past 10 years. Recurrence of cervical adenofibroma was confirmed by ultrasound and MRI. A wide local excision under hysteroscopy was performed due to her strong desire to preserve the uterus.

Results: Surgical pathology and immunohistochemical interpretation revealed cervical adenosarcoma. A hysterectomy with conservation of the ovaries was recommended, with regular follow-ups for evidence of disease recurrence.

Conclusions: Differential diagnoses of cervical adenofibroma are hard to prove. Adenosarcoma should be ruled out, especially in women presenting with recurrent cervical polypoidal masses. A combined histological/immunohistochemical investigation is mandatory.

Keywords: cervical adenofibroma; adenosarcoma; recurrence; histological/immunohistochemical investigation

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INTRODUCTION

Uterine cervical adenofibroma was first reported by Abell in 1971 as a form of mixed mesodermal tumour which is composed of benign stromal and epithelial components [1]. Benign cervical adenofibroma is rare and has a strong proclivity for local recurrence and potential for developing to adenosarcoma, which is characterized by benign epithelial glands and malignant stromal elements [2, 3]. Adenosarcoma is a low-grade malignancy which is often described as a midway point between benign adenofibroma and malignant carcinosarcoma [4]. Cervical adenosarcoma is extremely unusual and presents as cervical polyps which can be confused with benign cervical polyps both clinically and pathologically [5]. In this report, we describe the case of a young woman with recurrent cervical adenofibroma presenting as cervical polyps and progressing to adenosarcoma.

MATERIAL AND METHODS

A 40-year-old woman was referred to Women and Children's Hospital of Chongqing Medical University in July 2021 with a history of a recurrent cervical polypoid mass over the last 10 years. She had her first cervical polypoid mass, which presented with no symptoms, excised in July 2011. A cervical polyp was confirmed by postoperative pathological examination. Over the next 4 years, she had recurrent cervical polypoid masses roughly every year and underwent local hysteroscopic removal of the mass. Routine histological examination of the polypoidal tissue showed cervical polyps, but no immunohistochemical tests were performed. Although fertile, she underwent an embryo transfer in September 2016 and successfully conceived twins. She delivered two healthy boys by cesarean section at 34 weeks of gestation in May 2017. During her pregnancy and lactation, the cervical polypoid mass did not recurrence

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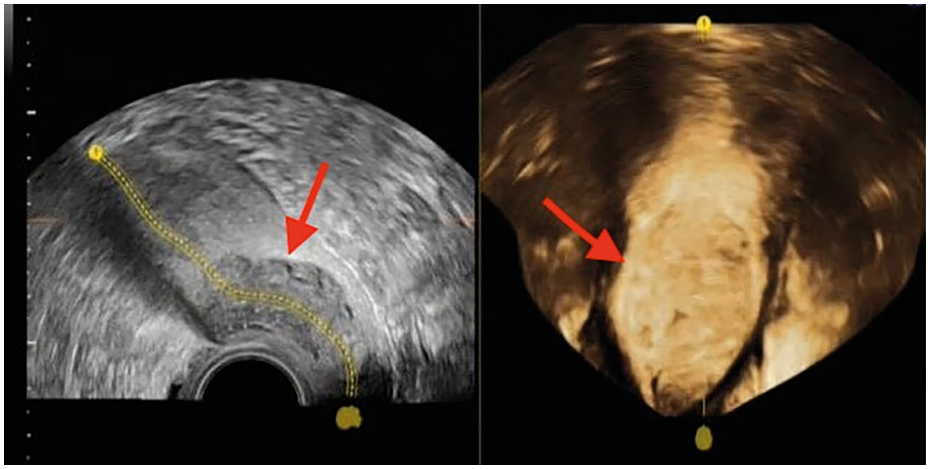

Figure 1. The polypoidal mass of the cervical canal under 3D Ultrasound (measuring 3.73 × 1.71 cm, arrow)

and there was no history of discomfort during this period. In July 2020, however, the cervical polypoid mass recurred again and was treated by hysteroscopic surgery. Postoperative pathological examination and immunohistochemistry indicated cervical adenofibroma. She had recurrence again in 2021, but she refused the hysterectomy due to a strong desire to retain her uterus, and she underwent hysteroscopic surgery again for recurrence of the disease. During this last hospital visit, her ultrasound examination showed a normal sized uterine body but a 3.73 × 1.71 cm polypoidal mass in the region of the cervical canal (Fig. 1). On examination, she was found to have a normal appearing cervix and her cervical cytology and human papilloma virus (HPV) tests were both negative. Magnetic resonance imaging (MRI) showed a heterogeneously enhancing lesion of 2.8 × 3.9 × 2.6 cm in the region of the cervix canal, and the uterine body and ovaries appeared normal (Fig. 2). There was no lymphadenopathy. Sex hormones and tumor markers were all within the normal range. Histology of the polyp confirmed adenofibroma. Hysterectomy was recommended as the primary treatment option considering the number of recurrences, however she again refused. To excise the lesion completely, we performed hysteroscopy under laparoscopic supervision. The polypoidal mass was seen to be arising from the cervix canal and decomposed clearly into the muscular layer, but the endometrial cavity was normal (Fig. 3).

RESULTS

The lesion was completely resected by hysteroscopy and was sent for pathological examination. Unfortunately, the histology showed a tumor having benign glandular epithelium with fusiform stromal tumor components. The interstitial ingredients were dense with "sleeve collar hair belt" locally. The mitotic figures were > 4/10 high-power field. Immunohistochemistry showed CK (epithelial +), CD10 (+),

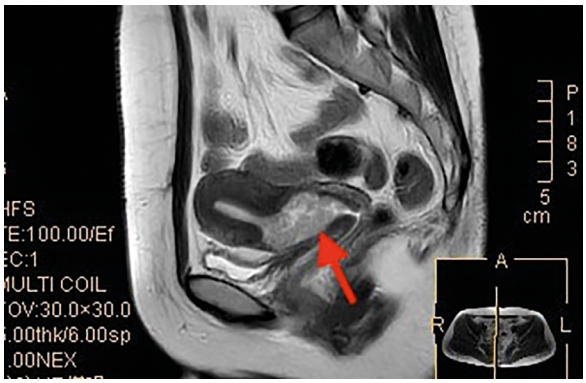

Figure 2. The polypoidal mass under magnetic resonance imaging (MRI) scan (measuring 2.8 × 3.9 × 2.6 cm, arrow)

SMA (–), CD34 (vascular +), Ki67 (about 5-10%), WT-1 (+), ER (about 70% medium intensity +), PR (about 70% strong +), Caldesmon (–), S-100 (–), p53 (–), Des (focal +) (Fig. 4). Given these results, a diagnosis of cervical adenosarcoma was decided. Hysterectomy with conservation of ovaries was again recommended along with regular follow-ups for evidence of recurrent disease.

DISCUSSION

Cervical adenofibroma is rare and usually presents as cervical polyps, and can be confused with benign cervical polyps and malignant adenosarcoma both clinically and pathologically [6, 7]. The possibility of more serious lesions such as adenosarcoma must be considered in patients with recurrent cervical polyps. Histopathological examination combined with immunohistochemistry can be more effective in differential diagnosis.

The patient in this case was only 30 years old when she was first diagnosed with cervical polyps. Her cervical lesions took 10 years to progress from benign polyps to

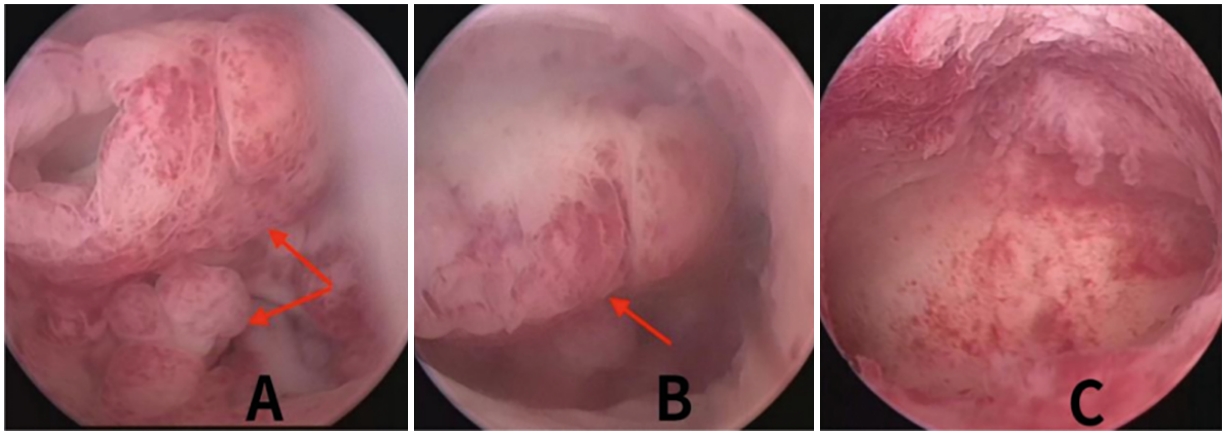

Figure 3. The polypoidal mass under hysteroscopy

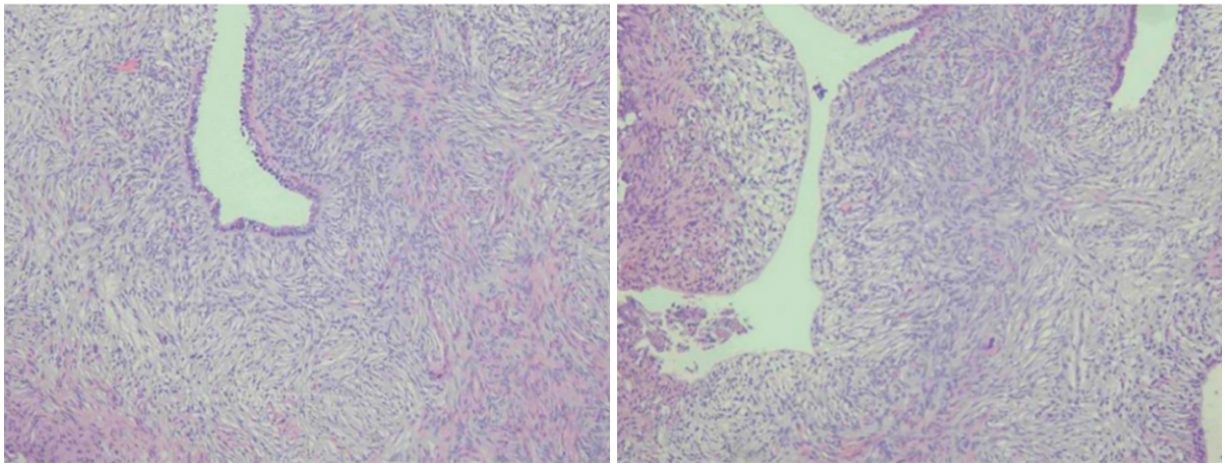

Figure 4. Histologic findings of the polypoidal mass (20× magnification)

adenosarcoma with 8 instances of recurrence, and the average recurrence time was roughly one year. After 10 years, she was diagnosed with adenosarcoma at 40 years old, which is consistent with a previous case report [8]. Benign recurrent endocervical polyps are unusual in women of reproductive age, hence the possibility of adenosarcoma in patients with a history of recurrent polyps must be considered. Some scholars often describe Mullerian adenosarcoma as a midway point between benign adenofibroma and malignant carcinosarcomas [4]. These lesions appear to represent a continuum of these diseases. Our patient's disease progression seems to confirm these views.

It is clinically difficult to distinguish adenofibroma from benign polyps and adenosarcomas, and histopathological examination seems to be a useful method. Polyps generally lack papillary processes, have more glands, and are less cellular than adenofibromas [9]. Adenofibromas contain a mixture of histologically bland epithelium and mesenchyme. They have no peripheral coagulation and no mitotic activity in the stromal compartment. Mitotic figures (MFs) are rare or invariably less than 4 MF/10 high power field (4MF/10HPF) in adenofibroma, which is a criterion to distinguish adenofibroma from adenosarcoma. Due to an insufficient understanding of adenofibroma by our clinicians and pathologists, the patient only received a general pathological examination without immunohistochemical examination during the previous pathology review. Her cervical polypoid mass was treated as benign cervical polyps many times and she continued to experience obvious recurrence. After the fifth conservative operation, the pathological and immunohistochemical examinations confirmed that the cervical polypoid mass was a cervical adenofibroma. Therefore, a careful evaluation of mitotic activity and nuclear atypia under histopathological examination combined with immunohistochemistry should always be performed in reproductive aged women with a history of recurrent endocervical polyps.

Adenofibroma is a benign lesion, but typically recurs. Recurrent tumors are prone to infiltrate the muscle layer and

blood vessels and adequate sampling is needed to exclude adenosarcoma [10, 11]. The main reasons for recurrence could be due to incomplete tumor resection or missed diagnosis of borderline tumors with recurrent adenofibroma [10, 12]. Our patient underwent cervical lesion resection 8 times for local recurrence. The fact is that the signs of cervical adenofibroma are similar to polyps and are often misdiagnosed as common benign cervical polyps for simple polypectomy. The incomplete excision of lesions from previous conservative operations was a potential reason for recurrence in this case. Another possibility is that her recurring adenofibroma itself may have potentially been malignant.

The main treatment method of cervical adenofibroma is surgical treatment including local tumor resection and total hysterectomy. Hysterectomy seems to be the preferred treatment method because the neoplasm may recur if incompletely curetted [10]. Therefore, for postmenopausal women, the standard treatment has been hysterectomy, but for young and fertile patients with limited early lesions, hysteroscopy may be considered for extensive lesion resection. Our patient was 30 at the time that her adenofibroma was diagnosed and she had a strong desire to preserve her uterus to remain fertile, so the option of conservative treatment by hysteroscopy was considered. As predicted, she experienced multiple relapses and repeated operations. At the eighth instance of recurrence, a total hysterectomy was recommended as the first treatment option, which she again refused due to her strong desire to preserve her uterus. Because of this, extensive lesion resection under hysteroscopy was the chosen method of treatment. On the one hand, there is no consensus on the optimal therapy for adenofibroma of the uterine cervix, and in rare cases, local excision has been curative. On the other hand, both MRI and 3D ultrasound indicated that the lesions were confined to the cervical canal and did not invade the muscle layer or have distant metastases. Unfortunately, postoperative pathological examination revealed adenosarcoma. For adenosarcoma, most doctors recommend total hysterectomy, usually accompanied by bilateral salpingo-oopherectomy [9]. In the latest research on adenosarcoma, investigators found that patients undergoing radical surgery have a higher overall survival rate, and early surgical resection can extend the survival time of patients [13]. Therefore, hysterectomy accompanied with salpingo-oopherectomy is generally recommended for older patients with no fertility requirements, but there is insufficient evidence to support or discourage ovarian conservation in young women [14]. Our patient is now 40 years old, has completed childbirth, and has no fertility requirements. We again recommended radical surgery, and her prognosis will be followed up on.

CONCLUSIONS

Cervical adenofibroma is highly recurrent and may have a potential malignant tendency. It is difficult to differentiate between benign polyps and malignant adenosarcoma.

Article information and declarations

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Conflict of interest

All authors declare no conflict of interest.

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