

## Cervical Adenosarcoma: 12 Years of Follow Up After Conservative Therapy

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### Abstract

Cervical adenosarcoma is a rare gynaecological malignancy defined as a biphasic tumor with a benign epithelial component and a malignant stromal component. Cervical localization is more common in younger patients, who frequently desire fertility preservation. Conservative strategy, in the absence of heterologous elements or lymphovascular space invasion, is becoming a preferred approach. Only a few cases have been reported, with no recurrence. Therefore, long-term follow up is essential.

We present a 15-year-old patient, with a polypoid mass of the cervix. She was submitted to excision of the polypoid lesion followed by conization.

There was no recurrence in the 12 years period of follow up.

**Keywords:** Adenosarcoma; Cervix; Fertility Preservation; Surveillance

### Introduction

Uterine adenosarcoma is a rare gynecological malignancy, representing about 5% of uterine sarcomas [1]. It is a biphasic tumor, composed of a benign glandular epithelium and a malignant stromal component [1,2].

It typically arises within the uterine corpus in post-menopausal women (median age of presentation 58 years) [2,3]. Less frequently, it's located in the endocervix (2% to 9% of all pelvic adenosarcomas) [4], where it tends to occur in younger women (median age 24,5) [5], especially in an adolescent age (the youngest patient described in the literature was 10 years old) [5,6] and appears to be associated with a better prognosis [7].

In cervical adenosarcomas, the presence of pedicle seems to be a protective factor [7]. Heterologous elements (especially rhabdomyosarcoma/sarcomatous overgrowth) or lymphovascular space invasion are considered independent risk factor for disease progression [6-8].

Historically, standard treatment for cervical adenosarcomas has been hysterectomy with bilateral salpingo-oophorectomy. However, patients who desire to preserve their fertility constitute a therapeutic dilemma. The apparent poor malignant potential and limited long-term follow-up experience, open the possibility for conservative treatment.

### Case Report

A 15-year-old girl, previously healthy, non-sexually active, menarche at 10, presented with vaginal discharge and a polypoid lesion emerging from vaginal introitus. A partial resection was performed by lesion torsion. The pathology exam revealed rhabdomyosarcoma and she was sent to our institution. Physical examination demonstrated a dark red gelatinous tumor visible at the vaginal introitus during Valsalva maneuver. Uni-digital observation detected a wide inspection in the endocervix. Pathology exam was reviewed and revealed cervical adenosarcoma. The abdominal and pelvic magnetic resonance (MRI) indicated a vegetative tumor with 23 x 12 x 28 mm (longitudinal x anterior-posterior x transversal), of the posterior endocervix, filling the upper two-thirds of vagina. Thorax TC and PET TC were negative.

The patient subsequently underwent excision of the lesion followed by cervical conization. Endometrial cavity and endocervical evaluation by hysteroscopy revealed no lesions. Histologically, evaluation of the polypoid mass confirmed adenosarcoma with free margins.

Conservative management was decided.

Over the following 12 years, she underwent continued surveillance visits, with annual pelvic magnetic resonance imaging for the first five years after surgery.

She is now 27 years old, recurrence free.

### Discussion

Cervical adenosarcoma continues to pose a challenge, because of its rarity and few long-term follow up reports.

They rarely present an endophytic growth pattern. The most common presentation is a protruding mass/polyp from the external cervical os/vagina (usually simulating a benign polyp) with vaginal bleeding and/or vaginal watery discharge [9].

In most case reports, young women were treated with hysterectomy. Optimal conservative management of cervical adenosarcoma is not consensual. An accurate pathologic review is essential. The absence of poor prognostic factors, such as heterologous elements or lymphovascular space invasion [3,5,7,8], is associated with low malignant potential and very low recurrence rate. Thus, conservative management of cervical adenosarcoma may be feasible in selected patients: narrow peduncle, free margins and when the site of excision can be monitored. Local excision, lesion torsion, cold knife conization or trachelectomy have all been described with similar results [5].

Few fertility preservation cases are reported, therefore there are no formal recommendations regarding surveillance. Although cervical sampling is questionable, annual pelvic magnetic resonance could be considered, but does not appear crucial after a few years [5].

In the literature, four cases of pregnancy after cervical adenosarcoma local treatment are reported [10,11], followed by hysterectomy.

### Conclusion

This case report adds to the limited data available, where a minimally invasive approach can be considered therapy, in women who desire to preserve fertility. However, long-term follow up is recommended and definitive hysterectomy should be considered after childbearing.

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