

## A Rare Case of Adenoma Malignum: Preparing for the Unforeseen

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### About the Author



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Adenoma malignum is a rare cancer of the cervix. It is usually an incidental finding following a hysterectomy that is generally performed for other indications. We present a case that had all the radiological features of adenoma malignum, but the diagnosis could not be confirmed pathologically prior to definitive surgery.

### Case Report

Mrs. S, 35 years old, married for 8 years presented to us with copious mucoid discharge from the vagina for 6 months. This discharge was not associated with a cyclical pattern, and she had to use panty liners to contain the same. There was no history of post coital bleed. She had an 8-year old son and had undergone a caesarean section in view of foetal distress. She was not interested in any more children and was using barriers for contraception. Her menstrual cycles remained regular and were not associated with any unusual symptoms. Per speculum examination revealed profuse mucoid discharge with a ballooned appearance of the cervix. No obvious mass or ulceration was seen and the cervical surface looked healthy. Per vaginal examination revealed a ballooned cervix with a normal sized uterus and no adnexal masses. Liquid-based Pap smear yielded no abnormality.

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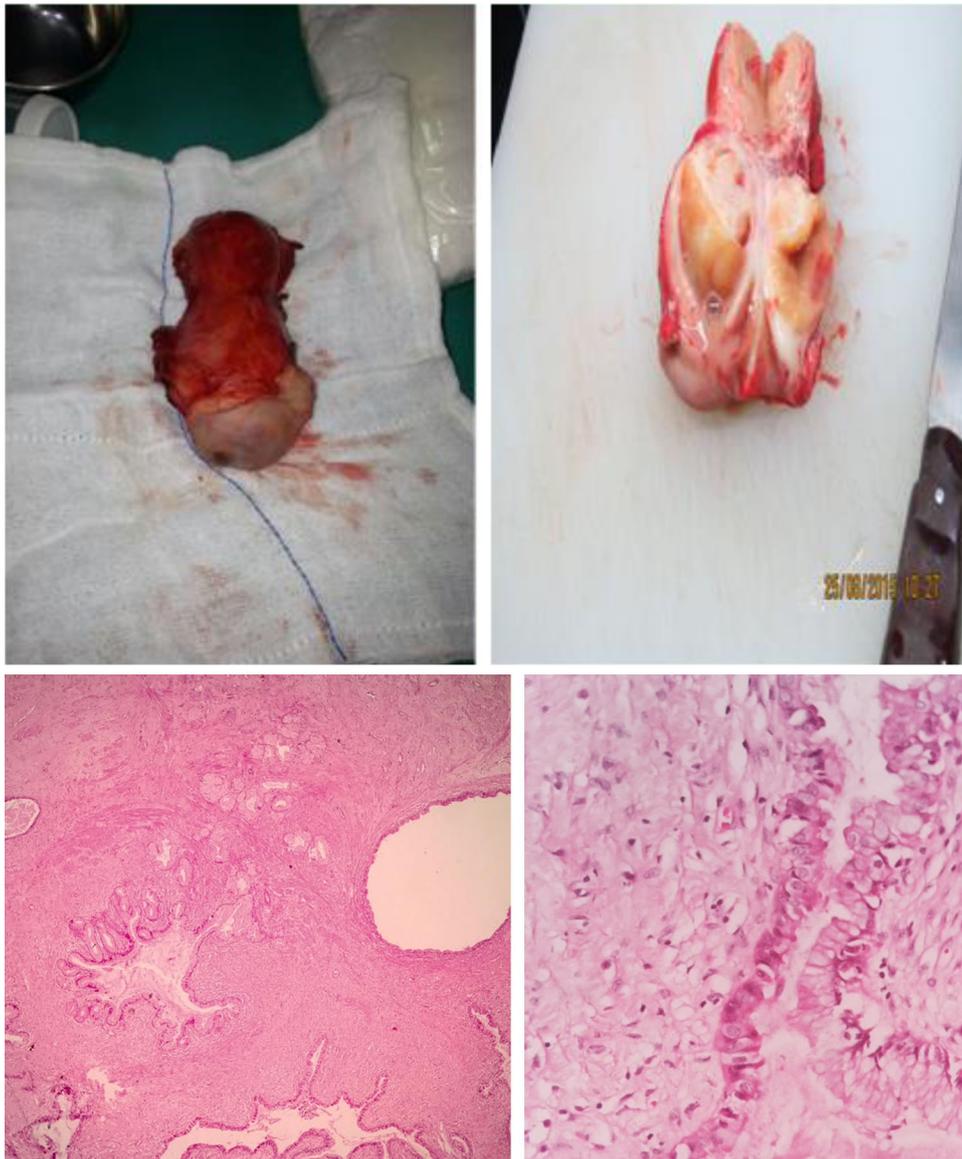
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Transvaginal ultrasound revealed a well-defined complex cystic lesion measuring  $5.9 \times 5.7$  cm within the stroma of the cervix with internal solid areas measuring about  $4.8 \times 2.7$  cm and inner vascularity. MRI pelvis revealed a well-defined space occupying lesion arising from the anterior wall of the cervix measuring  $6.1 \times 6.1 \times 4.2$  cm extending exophytically to right side from the endo to the ecto cervix. The cervical canal was deviated to the left side. The lesion showed multiple loculated cysts. Radiological diagnosis was suggestive of adenoma malignum (AM) with a differential diagnosis of nabothian cyst. Ultrasound guided FNAC revealed benign endo cervical cells and no definite malignant cells. However, since the FNAC was done targeting the solid areas under ultrasound and showed gland formation, malignancy could not be ruled out.

A multidisciplinary meet was organised comprising of a surgical oncologist, pathologist, radiologist and gynaecologist and the patient and her husband were also involved in the meeting. The radiological findings and the lesion size were less in favour of a nabothian cyst or tunnel clusters and pathologically malignancy could not be ruled out. Even if the lesion was benign and if local excision had to be considered, the cervix would have to be completely amputated and future pregnancies could be a challenge in terms of an incompetent cervix. The couple was counselled regarding the pros and cons of local excision versus hysterectomy. It was also clarified to the family that if there was a plan of childbearing cervical

amputation and an abdominal cerclage could be performed before embarking on pregnancy. The couple was clear in having a definitive surgery at one sitting if possible and clearly expressed no desire for future child-bearing. She was subjected to a type two radical hysterectomy in view of the size of the lesion, and the specimen was sent for a frozen section. The frozen section was not conclusive and hence bilateral pelvic lymph node sampling was performed, and ovaries on both sides were retained. Without a histopathologic diagnosis going ahead with a radical surgery was debatable as if the lesion were benign, the morbidity due to radical surgery would be undesirable. In view of the strong radiologic suspicion and the patient profile and desire, a radical hysterectomy was, however, performed.

The histopathology report revealed adenoma malignum with deep stromal invasion, no lymphovascular space involvement and lymph nodes were all negative for malignancy. The classical features of adenoma malignum which were observed were barrel-shaped cervix on gross examination with an endophytic growth pattern and deeply invasive glands. The glands were claw-shaped, tubular and crowded. Desmoplastic stromal reaction and cytologic atypia were also observed. Parametrial margins were free. Based on the pathological finding of lesion being more than 4 cm with deep stromal involvement, a pathological stage of Carcinoma Cervix 1B1 was given and she was subjected to adjuvant radiotherapy. Currently, it is 3 months after radiotherapy and she is symptom free.



## Discussion

Adenoma malignum was first described by Gusserow [1]. It is also known as minimal deviation carcinoma (MDA). It is pathologically a rare subtype of mucinous adenocarcinoma. It accounts for 1–3 % of uterine cervical adenocarcinoma and 0.15–0.45 % of all cervical carcinomas of the uterus [2]. Essentially, it is a tumour which looks benign but has a malignant behaviour.

The origin of adenoma malignum is uncertain and studies have shown that it is not related to HPV infection like the other cervical cancers. It is seen in 10 % of the cases with Peutz Jeghers syndrome.

The major clinical symptoms of AM are profuse mucoid vaginal discharge and irregular/contact vaginal bleeding [3].

Adenoma malignum has characteristic radiological features. It is seen on MRI as a multicystic mass, with high-signal intensity on T2-weighted images and isointensity or moderate hyperintensity on T1-weighted images. The lesion arises in the endocervical glands and extends deep into the cervical stroma.

The differential diagnoses are adenomyoma, deep nabothian cysts, tunnel cluster or endo cervical glandular hyperplasia as in OC pill users. Tunnel cluster is a specific type of Nabothian cyst characterised by complex multicystic dilatation of endo cervical glands [4].

Literature search has suggested that this variant of mucinous adenocarcinoma of the cervix is difficult to diagnose pre-operatively [5]. Cytology is insufficient to make a diagnosis of adenoma malignum. A deep incision biopsy or conization is needed to make a pre operative

diagnosis. Histologic adenoma malignum is characterised by well-spaced, deeply invasive branching glands lined with uniform columnar mucin-distended cells and infrequent foci of less well-differentiated neoplastic cells. It is essential to make an appropriate diagnosis of adenoma malignum before posting for definitive surgery as suboptimal surgery could increase the chance of recurrence and residual disease; however performing radical surgery on an innocent cervix can increase morbidity. Adenoma malignum has been for long considered to be a benign looking tumour with an aggressive clinical course. However, recent reports have suggested that if diagnosed early, the long-term outcome is not different from conventional cervical cancer. Surgery is considered to be the standard treatment when possible. Though there are no guidelines for what surgery is appropriate, it is treated as other adenocarcinomas of the cervix [6]. Similarly, after surgery, the adjuvant treatment is based on the pathological stage as with other cervical carcinomas.

## Conclusion

Adenoma malignum is a rare variant and awareness can help to diagnose more cases. In women with profuse mucoid discharge with typical MRI pictures revealing cystic lesion with solid areas in the substance of the cervix, one may have to consider adenoma malignum as a possibility. We need to be aware that there are several benign lesions which may have similar features and obtaining a diagnosis of adenoma malignum by fine needle aspiration and cytology may be inconclusive. Current recommendations are to treat adenoma malignum surgically like

cervical adenocarcinoma and adjuvant therapy to be considered in a similar manner as per the pathological staging.

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This case presentation has not been funded or sponsored.

## Compliance with Ethical Standards

**Conflict of interest** Dr. Gayathri Dinesh Kamath, Dr. Aditi Bhatt and Dr. Veena Ramaswamy declare that there has been no conflict of interest.

**Ethical Statements** All procedures performed on our patient were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

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