

Minimal deviation adenocarcinoma ("adenoma malignum") of the cervix: A diagnostic dilemma

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Abstract: Adenoma malignum is a rare variant of cervical adenocarcinoma. A woman of 26 years with complains of profuse mucoid discharge per vaginum on speculum examination per vaginum showed multiple cauliflower like growth hanging within the introitus. Punch biopsy gave a report of endocervical glandular polyp with squamous metaplasia. On excision, the mass was found to arise from the cervix and external Os separately identified. On histopathological examination the final diagnosis was adenoma malignum. Adenoma malignum of the uterine cervix is difficult to diagnose because of the deceptively benign appearance. To make a correct diagnosis, it is necessary to find a characteristic pathological feature such as multiple irregular lobulations of distorted glands demonstrating a "hair-pin" shape. Despite the presence of well-differentiated histopathological features, the prognosis of adenoma malignum is known to be poor because of early dissemination into the peritoneal cavity and early distant metastasis.

Key word: Adenoma malignum (AM)

I. Introduction

Adenoma malignum or minimal deviation adenocarcinoma of the cervix is a rare variant of cervical adenocarcinoma with poor prognosis¹. Most of the cases are diagnosed not by the attending gynecologist, rather by the pathologist while examining the pathology specimen. Preoperative diagnosis is difficult as cervical cytology, colposcopy guided biopsy and serum markers such as carcino embryonic antigen (CEA) are not very sensitive for detecting this variety of cervical cancer^{2,3}. A preoperative punch biopsy fails to confirm a diagnosis of MDA in most cases⁴. Imaging study such as pulsed power Doppler and Magnetic Resonance Imaging are upcoming investigation modalities used for differentiating adenoma malignum from other cystic lesions of cervix⁵⁻⁷. Currently genetic markers linked to chromosome 19p13.3 are found to be associated with the Sporadic Form of adenoma malignum of the uterine cervix⁷. Here we present a case of adenoma malignum for its peculiarity of presentation.

II. Case report

The present case is a woman of 26 years age married for 9 years P0+1+1+0 who came to the gynecology out patient department with complains of profuse mucoid discharge per vaginum with occasional spotting. She had a regular menstrual history. General examination findings were normal. On speculum examination per vaginum we saw multiple cauliflower like growth hanging within the introitus. Per vaginal palpation showed a growth occupying the vagina. Lateral vaginal walls were free while the cervical margins couldn't be felt.

Multiple possibilities came into our mind such as cervical cancer, a degenerated polyp or sub mucous fibroid, Sarcoma botryoides, uterine inversion, viral warts, vaginal growth and Hydatidiform mole. Smear from the growth was sent for bacteriological culture, Human papilloma virus antigen assay and a punch biopsy was sent for histopathological examination. Bacterial and virological studies were negative while punch biopsy gave a report of endocervical glandular polyp with squamous metaplasia.

We then performed an examination under anesthesia where we couldn't identify the external Os, uterus was normal size, anteverted and fornices were free. On Per rectal examination pouch of Douglas and parametrium were free. Then after inserting a Foley's catheter we attempted to excise the mass after applying successive haemostatic clamps (picture 1). On excision, the mass was found to arise from the cervix and external Os was separately identified (picture 2). On introduction of uterine sound uterine cavity was found to be of normal size.

After excision and histopathological examination the final diagnosis was adenoma malignum. The patient subsequently underwent Wertheim's hysterectomy followed by radiotherapy. She is disease free for two years after treatment completion.

III. Discussion

As early as in 1896, Hellier reports a case of Adenoma Malignum Cysticum Cervicis Uteri as “a growth which sprang from the cervix and filled the vagina without involving it. Vagina was found to be filled with cauliflower-like growth, necrotic and foul. When this was scraped away it was found that the vaginal walls were not involved and that the uterus was quite mobile” a description very similar to the present case⁹.

Subsequently Mckelvey and Goodlin in a case series described various presentations of Adenoma Malignum, red, granular, nodular cervix with the lesion invading the vagina; friable, cauliflower- shaped mass; cervical erosion; recurrence in cervical conisation stump or vault recurrence subsequent to hysterectomy¹⁰.

Adenoma malignum of the uterine cervix also referred to as the mucinous type of minimal deviation adenocarcinoma (mucinous MDA) of the uterine cervix, is a unique neoplasm that is difficult to diagnose because of the deceptively benign appearance of the tumor cells¹. It is a subtype of mucinous adenocarcinoma of the uterine cervix, which represents about 3% of adenocarcinoma cases³. In its well-developed form, the malignancy of the tumor, particularly as judged from biopsy, is being missed by competent pathologists, leading to a possibility of being under treated. Some reports suggest cytological examination as a better aid to detect this tumor with an 83% success rate¹¹. The common cytological features of mucinous MDA are (1) the frequent appearance of large sheets of cells with a honeycomb pattern on end view and a palisading arrangement at the periphery, (2) abnormal glandular cells with abundant cytoplasm showing slight to overt atypia in addition to benign-appearing glandular cells, and (3) slightly enlarged mucous cells with ovoid nuclei¹²⁻¹⁵. To make a correct diagnosis, it is necessary to find a characteristic pathological feature such as multiple irregular lobulations of distorted glands demonstrating a “hair-pin” shape⁴. However, the histopathological features of adenoma malignum are occasionally similar to pseudoneoplastic glandular lesions^{16, 17}. The MR findings of pseudoneoplastic lesions were almost identical to those of adenoma malignum, which suggested that the presence of cellular atypia or structural dysplasia in the pseudoneoplastic lesions did not significantly change MR findings^{5, 15}.

Mucinous MDA characteristically expresses gastric phenotypes. Immunostaining using the monoclonal antibody HIK1083, which is specific for gland mucous cell mucin of the stomach, facilitates the identification of tumor cells in tissue sections¹⁸. Despite the presence of well-differentiated histopathological features, the prognosis of adenoma malignum is known to be poor because of early dissemination into the peritoneal cavity and early distant metastasis. This is true of even the early lesions so that it has a bad prognostic outlook²⁻⁴. This is in contradistinction to the classical adenocarcinoma of the cervix which has shown a 50% 5-year cure rate and is quite comparable in this respect to the squamous cell carcinomas.

Figure Legend

AM Legend 1: mass seen during EUA

AM Legend 2: excised mass



Picture 1



Picture 2

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