Adenoid Cystic Carcinoma of Cervix: A Case Report
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Abstract: Adenoid cystic carcinoma of the cervix is a relatively uncommon tumor accounting for < 1% of all cervical adenocarcinomas. The tumor is most often seen in 6th-7th decade. A 50-year-old postmenopausal woman presented to Department of gynecology with vaginal bleeding and lower abdominal pain. Per speculum and vaginal examination showed a friable, irregular cervical growth measuring 5 × 4 cm which bled on touch. Biopsy of cervical growth was done and sent for histopathological examination. Sections examined showed nests and sheets of neoplastic cells arranged in cribriform and tubular pattern with areas of necrosis and ulceration. Diagnosis of adenoid cystic carcinoma was made on the basis of these findings. Adenoid cystic carcinoma of the cervix is a rare and aggressive tumor with frequent local recurrence. Its clinical course is influenced principally by the clinical staging and the histological pattern of growth. Radical surgery with adjuvant radiotherapy and/or chemotherapy is the treatment of choice.

Keywords: Cervical growth, vaginal bleeding, adenoid cystic carcinoma.

INTRODUCTION
Adenoid cystic carcinoma (ACC) is a malignant epithelial neoplasm derived from the salivary glands and can occur in a variety of other sites. It has been found in minor and major glands, lacrimal glands, and mucous glands of the aero-digestive tract, skin, breast and lung. In the female reproductive tract, adenoid cystic carcinoma occurs most commonly in the Bartholin gland, however can also be found in the cervix [1].

It accounts for around 1% of all cervical carcinomas. The tumour is aggressive with high rate of local recurrence [2]. The tumor is most often seen in 6th-7th decade. Because of the rarity of the disease, standard treatment could not have been established. We present a rare case of a post-menopausal woman who presented with a friable growth on cervix which turned out to be an adenoid cystic carcinoma of cervix.

CASE REPORT
A 50-year-old postmenopausal multiparous woman presented to the Department of gynecology with history of irregular vaginal bleeding for the last 5 months, and lower abdominal pain for the last two months. General physical examination was unremarkable. On per abdominal examination no abnormality could be detected. Per speculum and vaginal examination showed a friable, irregular cervical growth measuring 5 × 4 cm which bled on touch. On bimanual examination vagina, uterus and bilateral adnexal structures were found to be normal. There was no parametrial involvement.

Biopsy of cervical growth was done and sent for histopathological examination. Histopathological sections were examined which showed nests and sheets of neoplastic cells arranged in cribriform and tubular pattern (Fig 1 and 2). Surrounding stroma shows acute on chronic inflammatory infiltrate with areas of necrosis and ulceration. Diagnosis of adenoid cystic carcinoma was made on the basis of these findings.

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DISCUSSIONS

ACC of the cervix is a rare malignancy in general. Primary ACC of the cervix accounts for around 1% of all cervical carcinomas [2]. The first case was described in 1949 by Paalman [3]. Origin of this tumor is still unknown. Most accepted view is that it arises from reserve cells of endocervix [4]. Most common carcinoma of cervix is squamous cell carcinoma of cervix, cause of which is Human Papilloma virus. Role of HPV in the pathogenesis of ACC is not well defined. The tumor generally occurs in post-menopausal woman; however it has been reported in young woman too. Most common symptom of ACC is vaginal bleeding which may be associated with uterine enlargement in some cases. The tumour is generally reported to be non-friable although can be a friable growth in rare cases. Adenoid cystic carcinoma is believed to originate from endocervical glands [5].

The most characteristic pattern in ACC is cribriform pattern; the other two are tubular and solid pattern. Cribriform pattern is composed of polygonal to spindled cells forming numerous duct-like structures that contain extracellular matrices filled with homogenous eosinophilic periodic acid-Schiff (PAS) positive material or granular basophilic material [6]. A grading system was described by Perzin et al for ACC based on distinctive histologic patterns i.e tubular, cribriform and solid [7]. It was believed that the three patterns reflect a progression of cellular proliferation and aggressiveness of biologic behavior. The tumor is assigned to three histologic grades: grade I, a well-differentiated tumor composed of tubular and cribriform patterns without solid components; grade II: a tumor with a pure cribriform pattern or mixed with less than 30% of solid areas and grade III, a tumor with marked predominance of the solid pattern.

Immunohistochemically, the tumor cells located in recognizable duct stain positive for keratin, S-100 protein and CD117 (C-kit) and those around pseudocysts stain positive for S-100 protein and actin and variable for keratin [8]. Immunohistochemical analysis can be used whenever there is doubt in diagnosis and histopathology failed to establish the diagnosis.

The differential diagnosis includes small cell carcinoma, adenoid basal carcinoma and non-keratinizing squamous cell carcinoma. In our case, the patient was 50-year-old postmenopausal female with history of vaginal bleeding and lower abdominal pain. On per speculum examination, it usually presents as a non-friable mass in contrast to friable growth usually seen in squamous cell carcinoma (SCC) of cervix. In our case, per speculum examination revealed a friable growth which bled on touch, making it difficult to diagnose on clinical examination. ACC is mostly invasive and often associated with perineural invasion, mesenchyme-like areas and foci of squamous metaplasia are consistently absent. No standard treatment has yet been proposed because of its rarity and absence of prospective studies. However in most of cases, surgery was performed followed by radiotherapy as in SCC [5, 9]. Chemotherapy has little if any role in the adenoid cystic carcinoma of cervix.
CONCLUSION

Although, a rare tumor, possibility of ACC should be kept in mind when we encounter a tumor with glandular differentiation in cervix in an elderly woman. It is an aggressive tumor with fatal outcome. Prognosis is modified by its size and status of resection margins. Early diagnosis and full resection followed by radiotherapy can prolong life of patient.

REFERENCES