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Case Report

Solid Variant of Adenoid Cystic Carcinoma Coexisting with Invasive Squamous Cell Carcinoma in Uterine Cervix – A Case Report with Immunoprofile

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Abstract

Primary adenoid cystic carcinoma of cervix is a rare entity. Co-existence of adenoid cystic carcinoma with squamous cell carcinoma is even more rare. Here we present the case of an 85 year old postmenopausal lady who presented with on and off vaginal bleeding and was found to have a hard cervical mass. Hysterectomy with bilateral salpingo-oophorectomy was done and histopathological evaluation revealed coexisting solid variant of adenoid cystic carcinoma and squamous cell carcinoma. High grade squamous intraepithelial lesion was seen adjacent to invasive squamous cell carcinoma. Adenoid cystic carcinoma was positive for CD117 and squamous cell carcinoma was positive for p63. Both the components were also immunoreactive for p16.

Keywords: Adenoid cystic carcinoma, Squamous cell carcinoma, p16.

INTRODUCTION

Adenoid cystic carcinoma is a malignant epithelial neoplasm commonly derived from the salivary glands. It can also occur in the lacrimal glands and the mucous glands of the respiratory tract, breasts and skin [1]. The occurrence of primary adenoid cystic carcinoma in cervix is very rare and accounts for less than 1 % of cases [2]. Morphologically, it can have cribriform, tubular or solid pattern. It carries worse prognosis in cervix when compared to their usual salivary gland counterpart. It is usually seen in postmenopausal women [3]. Rarely it has been seen in combination with other neoplasms including intraepithelial squamous neoplasia, invasive squamous cell carcinoma, adenoid basal cell carcinoma, and sarcoma in the uterine cervix [4]. Here we report a case of solid variant of adenoid cystic carcinoma coexisting with invasive squamous cell carcinoma

CASE REPORT

85 year old postmenopausal female presented with on and off vaginal bleeding of 4 months duration. Per vaginal examination showed a hard mass in the anterior fornix. Total abdominal hysterectomy and bilateral salpingo-oophorectomy was done and the specimen was sent for histopathological examination.

Gross examination showed a cervical mass measuring 6x5x4 cm. Cut section of the mass was grey white with areas of necrosis. Microscopy showed neoplasm with two distinct components. 70 % of the tumor showed diffuse sheets and nests of basaloid cells (Figure 1 & 2) Focal area (5%) showed similar cells arranged as tubules and in cribriform pattern with pseudo-cystic spaces containing basophilic material. Cells were small to medium sized with scanty cytoplasm and hyperchromatic nucleus. Some cells showed vesicular nucleus, dispersed chromatin and distinct nucleoli. Areas of necrosis, brisk mitosis and lympho-vascular invasion seen. 25% of the tumor showed invasive nests of nonkeratinizing squamous cell carcinoma arising from dysplastic squamous epithelium (Figure 3). Tumor showed extension into bilateral parametrium. On Immunohistochemistry, solid area of adenoid cystic carcinoma was diffusely positive for CD117 and negative for p63 (Figure 4 & 5). Invasive squamous component was positive for p63 and negative for CD117. Both the components were in turn positive for cytokeratin and p16 (Figure 6). Tumor cells were negative for LCA, synaptophysin and chromogranin.

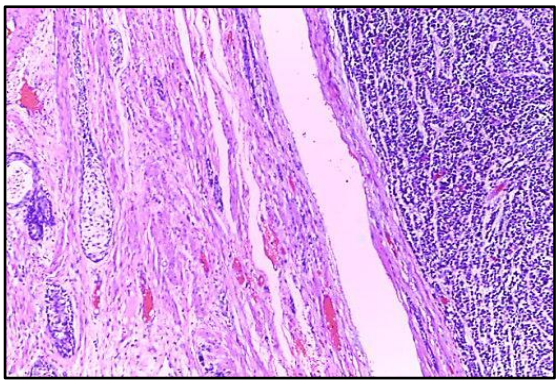

Figure 1: Solid area of adenoid cystic carcinoma(right) and invasive squamous component (left) H&E x 100.

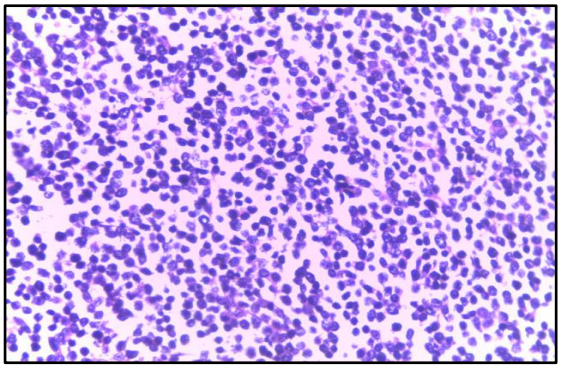

Figure 2: Solid area of adenoid cystic carcinoma (H&E x400).

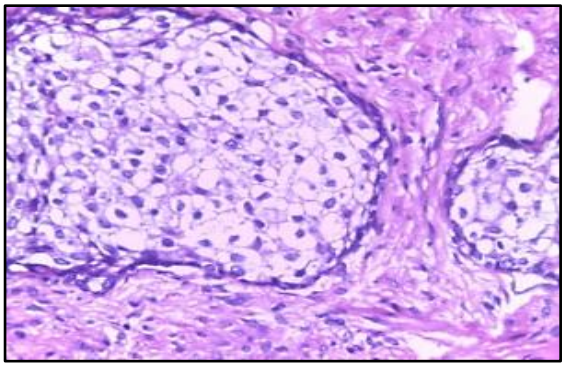

Figure 3: Invasive squamous cell carcinoma (H &E x 400).

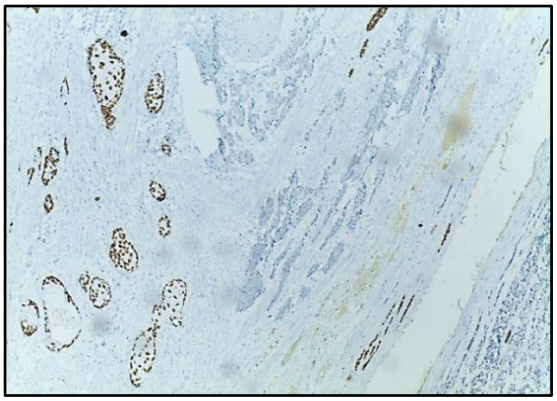

Figure 4: p63 showing positivity in squamous cell carcinoma and negativity in adenoid cystic carcinoma (x100).

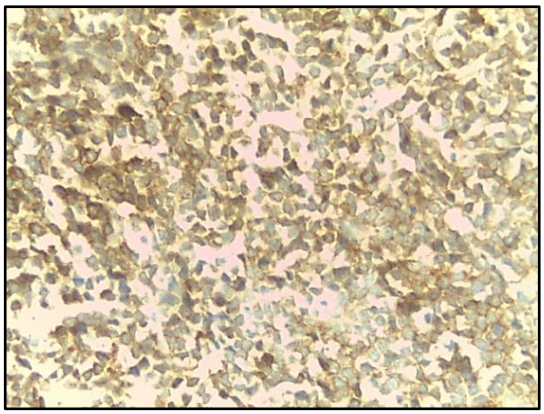

Figure 5: CD117 showing diffuse positivity in solid area of adenoid cystic carcinoma(x 400).

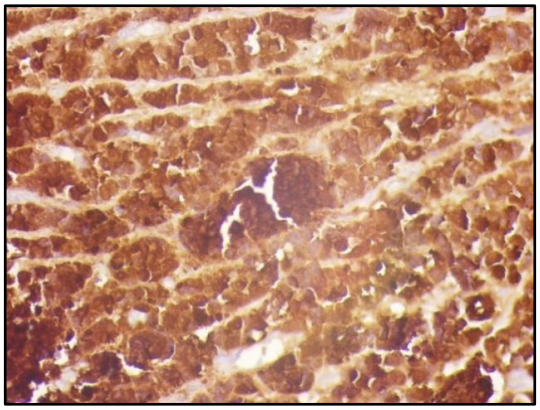

Figure 6: Diffuse cytoplasmic and nuclear staining for p16 in adenoid cystic carcinoma (x 400).

DISCUSSION

Adenoid cystic carcinoma of the cervix is a rare malignant tumour. In 1949 Paalman and Conseller first reported adenoid cystic carcinoma that appeared in cervix as Cylindroma [5]. It usually occurs in postmenopausal women, usually between the sixth and seventh decade of life [6]. Adenoid cystic carcinoma coexisting with squamous cell carcinoma is even more rare. In a study by Shi *et al.*, he had quoted that only 27 such cases were reported in literature. Adenoid cystic carcinoma is thought to arise from the reserve cells of endocervix [7]. Role of HPV infection in the pathogenesis of adenoid cystic carcinoma is not well defined [8]. However, in a case series by Shi et al, he had demonstrated p16, along with HPV mRNA ISH positivity in both components of coexistent adenoid cystic carcinoma and squamous cell carcinoma. Our case also showed diffuse cytoplasmic and nuclear staining for p16 in both components.

Differential diagnosis of adenoid cystic carcinoma includes adenoid basal cell carcinoma, small cell carcinoma and basaloid squamous cell carcinoma. Both adenoid cystic carcinoma and adenoid basal cell carcinoma are common in postmenopausal women, but the latter disease is usually asymptomatic and has an indolent course. Adenoid basal cell carcinoma has less

pleomorphic nucleus, low mitotic activity and show absent CD117 immunostaining [9]. Small cell carcinomas are positive for neuroendocrine markers like NSE, synaptophysin and chromogranin. Basaloid squamous cell carcinomas are positive for p63 and CK5/6. Both the entities are negative for CD 117. Adenoid cystic carcinoma is generally negative for p63 [4].

Prognostically, adenoid cystic carcinoma is more aggressive when compared to squamous cell carcinoma. Solid variant in turn has a greater potential for distant metastasis. Factors contributing to prognosis include location of tumor, invasion to adjacent anatomic structures, deep stromal invasion and lympho-vascular invasion. There are no specific treatment guidelines for adenoid cystic carcinoma cervix. It is rather treated just as any other squamous cell carcinoma cervix [1]. Most patients with adenoid cystic carcinoma are thus treated with surgery and radiotherapy. Chemotherapy is said to have less role in the management of this entity [7]. Dixit *et al*., recommends surgery with adjuvant chemoradiotherapy for early stage and chemoradiotherapy in advanced stage [10]. In our case, the patient was referred to a cancer centre and was started on radiotherapy.

CONCLUSION

Adenoid cystic carcinoma can be rarely encountered in uterine cervix, particularly in postmenopausal women. It can be seen sometimes in combination with squamous cell carcinoma. Adenoid cystic carcinoma can have different morphological patterns, of which solid variant can be confused with other neoplasms including basaloid squamous cell carcinoma, small cell carcinoma and even lymphoma. In such cases, immunohistochemistry can be useful to come to a definite diagnosis. Though the exact pathogenesis is not clear, positivity for p16 was detected in both adenoid cystic carcinoma and squamous cell carcinoma. As adenoid cystic carcinoma is an aggressive disease, with greater chances for local recurrence and distant metastasis, exact recognition of the disease with prompt treatment is important.

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