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Primary signet ring cell carcinoma of the cervix: A case report and review of the literature

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ABSTRACT

INTRODUCTION: Primary signet cell carcinoma of the cervix has been reported only in 18 cases to date.
PRESENTATION OF CASE: A 48-year-old woman was seen at our Gynecologic Oncology Unit, because she complained postcoital bleeding during the last three months. She had 1–2 cm cervical mass, originating from the endocervical canal. A biopsy revealed a signet ring cell-type adenocarcinoma. Suspected primary sites were excluded after gastroscopy, colonoscopy and mammography. The patient underwent a laparoscopic type-3 radical hysterectomy with bilateral salpingo–oophorectomy, pelvic lymph node dissection and paraaortic lymph node dissection with a presumed diagnosis of primary signet ring cell carcinoma of the cervix. Microscopically, the tumour consisted of 70% signet ring cell type and 30% endocervical adenocarcinoma. She did not receive any adjuvant treatment. Follow-up at 18 months after surgery showed no evidence of recurrence.
DISCUSSION: Nineteenth case of a primary signet ring cell carcinoma of the cervix was presented. Immunohistochemical studies and HPV DNA positivity may help in diagnosis.
CONCLUSION: It is crucial to differentiate primary tumour from metastatic signet cell carcinoma, while treatment and prognosis differ significantly.

1. Introduction

Pure or predominantly signet-ring cell carcinoma of the cervix is extremely rare in the literature. In total, 18 cases of primary cervical adenocarcinoma containing signet-ring cells have been reported to date [1]. The infrequency with which it is encountered makes primary signet-ring cell carcinoma of the cervix a diagnostic challenge. Possible metastasis from any site should be excluded, as management and prognosis vary between metastatic and primary signet ring cell carcinomas of the cervix.

Herein, we report a case of primary predominantly signet ring cell carcinoma of the cervix with immunohistochemical findings and review the literature.

2. Case report

A 48-year-old, gravida 5, para 3 woman with post-coital vaginal bleeding during the last 3 months was seen in our hospital. Her body mass index was 24 and she had no significant medical or family history. A pelvic examination revealed a 1–2 cm cervical mass, which appeared to originate from endocervical canal. A biopsy revealed a signet ring cell-type adenocarcinoma. Laboratory studies, including cancer antigen 125 (CA-125), carcinogenic antigen (CA 19-9), cancer antigen 15–3 (CA 15-3), carcinoembriogenic antigen (CEA), and alpha-fetoprotein (AFP), were within normal limits. Magnetic resonance imaging (MRI) showed a 1.7 × 1.5-cm cervical mass with a homogeneous intensity on T1-weighted images and a heterogeneous intensity on T2-weighted images (Fig. 1). Increased FDG uptake on the positron emission tomography (PET)/computed tomography (CT) images were found for the cervical mass (SUVmax: 13.5). A gastroscopy and colonoscopy were performed to reveal the possible primary site of the tumour, however, both did not the site. In addition, her preoperative mammogram was negative. The patient underwent a laparoscopic type-3 radical hysterectomy with bilateral salpingo–oophorectomy, pelvic lymph node dissection and paraaortic lymph node dissection with a presumed diagnosis of primary signet ring cell carcinoma of the cervix. There was no pathologic finding in the pelvic cavity or abdomen.

Macroscopically, tumour measured 25 × 18 × 13 mm in size and it was located in ecto- and endocervix. Microscopically, the tumour consisted of 70% signet ring cell type and 30% endocervical adenocarcinoma. Signet ring cells were within pools of extracellular

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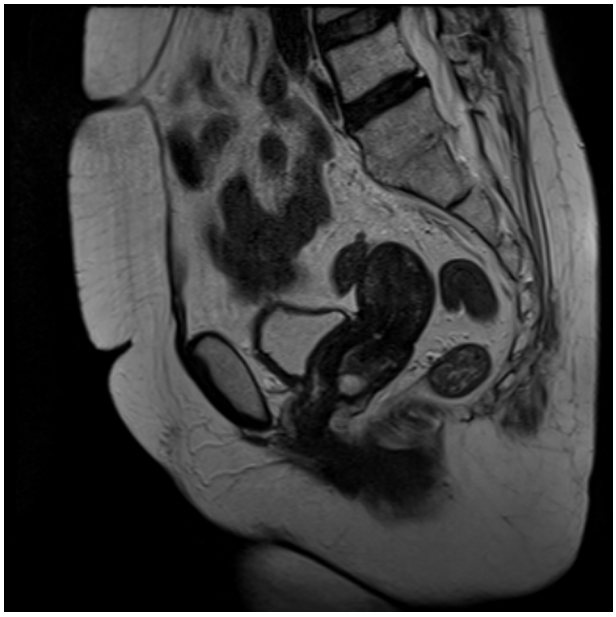

Fig. 1. (A) 1.7 × 1.5-cm cervical mass with a homogeneous intensity on T1-weighted images and a heterogeneous intensity on T2-weighted images on posterior cervix.

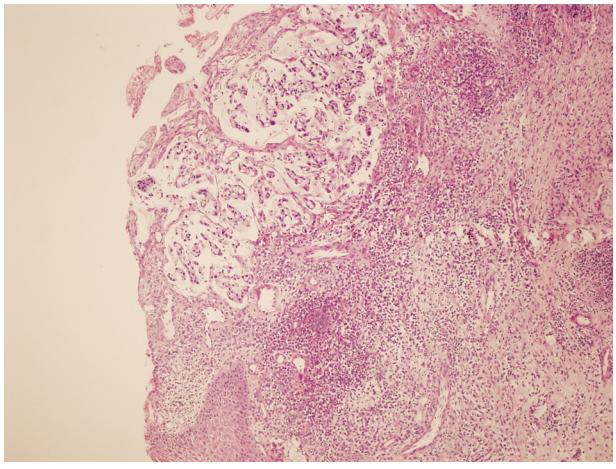

Fig. 2. The tumor was located in ecto-endocervix. HE X50.

mucin (Figs. 2 and 3). The tumour cells had hyperchromatic, eccentrically located nuclei and large mucin filled cytoplasmic vacuoles. An immunohistochemical study of the tumour showed positivity for p16 (Fig. 4), CDX-2, MUC1, MUC2 and MUC5AC and negativity for synaptophysin, chromogranin A and CK–20. The parametrium, pelvic and paraaortic lymph nodes were negative and no lymphovascular space invasion was observed.

The patient did not receive postoperative chemotherapy. Follow-up at 18 months after surgery showed no evidence of recurrence.

Written informed consent was obtained from the patient to publish these data.

3. Discussion

Previous cases of primary signet-ring cell carcinoma of the cervix are presented in Table 1. It is essantial to distinguish a primary tumour from metastasis when there are signet ring cells in a carcinoma within cervix. The stomach, colon, breast, appendix, bladder are possible primary sites for metastasis [1]. Therefore,

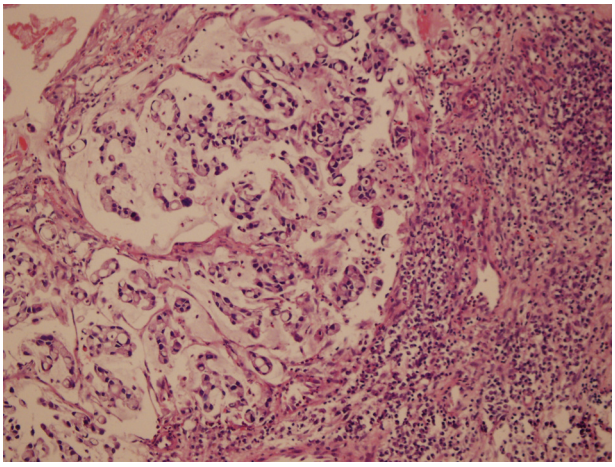

Fig. 3. The tumor was composed of signet ring cells within pools of extracellular mucin. HE ×200.

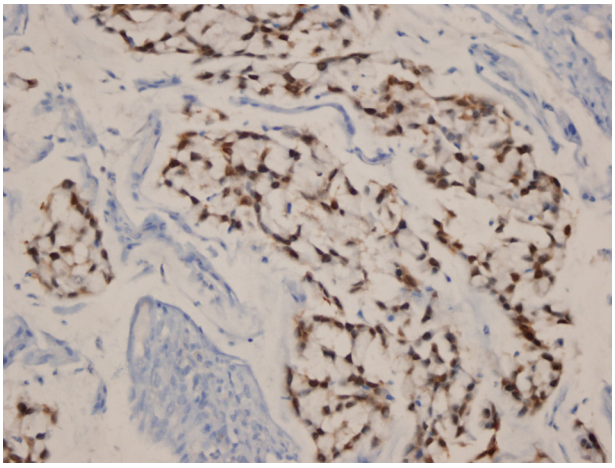

Fig. 4. The nuclei of tumor cells were diffusely positive for p16. p16 ×400.

further evaluations for other primary sites are mandatory to exclude metastasis. In addition, an earlier report described some features in favour of a primary cervical tumour such as a history of HPV infection, the coexistence of high-grade squamous intraepithelial lesion and adenocarcinoma in situ with an invasive disease and HPV type 18 in tumour tissue [4]. In our case, a gastroscopy and colonoscopy were performed and the patient underwent MRI and PET/CT. However, no other tumour lesion was found. Our patient had a history of HPV infection, and HPV type 18 was found in her tumour tissue. HPV type 18 is a well-known risk factor for cervical adenocarcinomas. Hence, almost all of the reports searching for HPV-DNA in cases with primary signet ring cell adenocarcinoma of the cervix including ours showed HPV type 18 positivity, an association with HPV type 18 and primary signet ring cell adenocarcinoma of the cervix may be easily suggested.

Immunohistochemical studies were performed in most of the previous studies. However, conflicting results have been obtained (Table 1). Three cases were negative for mammoglobin and no positive case was reported. Similarly, two cases reported positive reaction with p16, which may show an HPV effect on the tumour. No negative case was reported with p16 immunohistochemical staining. To date, positivity for cytokeratin 7 was shown in five cases, and it seems to be the most prominent immunohistochemical marker. In a recent study, cervical cytokeratin 7 positivity was found to be associated with progression of low grade cervical lesions to high grade cervical lesions [15]. The literature regarding