Case Report

Coexistent squamous cell carcinoma and adenoid basal carcinoma in the uterine cervix and infection with human papillomavirus (HPV 31)

Yu-Chieh Lin a,b, Cherng-Lih Perng c,d, Yi-Ming Chang a, Yao-Feng Li a, Yuan-Ming Tsai e, Gwo-Jang Wu f, Chih-Kung Lin a,*

a Department of Pathology, National Defense Medical Center and Tri-Service General Hospital, Taipei, Taiwan
b Department of Pathology, Taoyuan Armed Forces General Hospital, Taoyuan, Taiwan
c Division of Clinical Pathology, Department of Pathology, Tri-Service General Hospital, Taipei, Taiwan
d Graduate Institute of Pathology, National Defense Medical Center, Taipei, Taiwan
e Department of Surgery, National Defense Medical Center and Tri-Service General Hospital, Taipei, Taiwan
f Department of Obstetrics and Gynecology, National Defense Medical Center and Tri-Service General Hospital, Taipei, Taiwan

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Abstract

Objective: Adenoid basal carcinoma (ABC) is an uncommon neoplasm of the uterine cervix. ABC can be accompanied by carcinoma in situ or invasive carcinoma. Most cases are discovered accidentally during radical hysterectomy. ABC is associated with a high risk of human papillomavirus infection (HPV), most often HPV 16 infection.

Case report: We present a rare case of an 86-year-old Taiwanese married woman who suffered from bloody vaginal discharge and occasional lower abdominal pain and received cervical biopsy. The pathological report revealed squamous cell carcinoma (SCC) of the uterine cervix. After radical hysterectomy, bilateral salpingo-oophorectomy, and bilateral pelvic and para-aortic lymph node dissection, the final pathological report revealed SCC coexisting with ABC, and both of the components were infected by HPV 31. After receiving radiotherapy, she maintained outpatient department follow-up.

Conclusion: A literature review revealed that this was a rare case of combined ABC–SCC associated with HPV 31 infection. In this case, the ABC component did not affect the tumor stage because it was confined to the cervix. However, we must avoid overestimating the clinical stage because the ABC component is thought to be a benign lesion.

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Introduction

Adenoid basal carcinoma (ABC) of the uterine cervix is a rare neoplasm usually occurring in postmenopausal woman. ABC combined with other carcinomas in situ or invasive carcinomas has been previously reported [1–6]. The coexistence of squamous cell carcinoma (SCC) with ABC is very rare, and is usually associated with human papillomavirus (HPV) 16 and HPV 33 infection. Our case is believed to be the first of combined ABC–SCC associated with HPV 31 infection. In our case, both the SCC and ABC components were positive for HPV 31 infection. HPV DNA was detected in both components by polymerase chain reaction (PCR).

Case report

We report the case of an 86-year-old Taiwanese married woman, who had never received a Papanicolaou smear and
had a past history of hypertension with regular follow-up. She suffered from bloody vaginal discharge and occasional lower abdominal pain in the past 2–3 months prior to when she visited our gynecologist. After a physical examination, cervical erosion was found, and cervical biopsy was performed. The pathology report revealed SCC of a large cell, nonkeratinizing type. Abdominal computer tomography showed an ill-defined, poorly enhanced lesion measuring 4.2 cm × 3.6 cm × 2.6 cm (Fig. 1A). Serum levels of tumor markers including SCC and carcinoembryonic antigen were <1 ng/mL. Initially, the International Federation of Gynecology and Obstetrics (FIGO) stage IB, American Joint Cancer Committee (AJCC) staging: cT1bN0M0, stage IB was diagnosed by a clinician, and the patient underwent radical hysterectomy (Fig. 1B), bilateral salpingo-oophorectomy, and bilateral pelvic and para-aortic lymph node dissection.

Microscopically, the tumor had two components (Fig. 2A): (1) a nonkeratinizing large cell type of SCC characterized by nested tumor cells with pleomorphic nuclei and frequent mitotic figures, and an absence of keratin pearl formation, with stromal invasion; and (2) an ABC characterized by small, uniform basaloid cells arranged in small nests or glandular patterns. The tumor cells were smaller with fewer pleomorphic nuclei and infrequent mitotic figures. Therefore, the final pathological diagnosis was SCC, large cell, nonkeratinizing type coexistent with ABC. Both components were immunoreactive for P16 (Fig. 2B). A biopsy of both the SCC and ABC components was positive for HPV DNA using PCR (Fig. 3). The DNA of the SCC and ABC components was directly sequenced, and a search of the sequences using the National Center for Biotechnology Information (NCBI) Basic Local Alignment Search Tool (BLAST) for similarity to HPV[7], and it revealed a HPV 31 infection in the SCC and ABC components (Fig. 4). Finally, according to the pathological T stage, excluding the size of ABC, the pathological AJCC stage of our case was pT1b2N0MX, stage IB2.

Discussion

ABC was first described in 1966[8]. It is a rare tumor, usually asymptomatic clinically, and usually discovered accidentally during hysterectomy and after an abnormal Papanicolaou smear. Most patients are postmenopausal, except for one case of a 20-year-old woman[9]. Adenoid cystic carcinoma and ABC have some common features and the latter may be distinguished from adenoid cystic carcinoma by its aggressive behavior. However, immunohistochemical stains for epithelial membrane antigen, collagen IV, and laminin can

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Fig. 1. (A) Computed tomography showed one ill-defined and poorly enhanced lesion measuring 4.2 cm × 3.6 cm × 2.6 cm situated in the uterine cervix (white arrow). (B) Gross appearance of the tumor, squamous cell carcinoma (black arrow), and adenoid basal carcinoma (white arrow).

Fig. 2. (A) Section of hematoxylin and eosin stained (10× objective lens) showed squamous cell carcinoma, large cell, nonkeratinizing type (#) characterized by nested tumor cells with stromal invasion, and adenoid basal carcinoma (*) characterized by solid and glandular tumor cells beneath the component of squamous cell carcinoma (B). The two components were immunoreactive for P16 (10× objective lens).
Microscopically, ABC is composed of small, round, uniform basaloid cells in small nests or cord growth patterns. Some pathologists think ABC should be redesignated adenoid basal epithelioma because of its benign nature[11]. Therefore, in the AJCC staging of the invasive carcinoma in the present case, taking into account the size of the ABC, would have led to overstaging.

Most of the carcinomas that coexist with ABC are in situ lesions[4,8]. Few cases of SCC accompanied by ABC have been reported.

According to the literature review, immunohistochemical staining is less sensitive than PCR analysis for high-risk HPV detection[12,13]. We looked for the presence of HPV infection in the SCC and ABC by immunohistochemical staining for P16 and detection of HPV DNA by PCR analysis, respectively. The results showed that both components were immunoreactive for P16, and the PCR analysis revealed that both the SCC and ABC components were infected with HPV 31. Our results support the findings of linking infection with...
HPV to invasive carcinoma and ABC, and our case is believed to be the first to show HPV 31 infection [3,14,15].

In conclusion, coexistent SCC and ABC in the uterine cervix are rare, and this is believed to be the first case with infection with HPV 31. Although coexistent SCC and ABC are noted, only the SCC component should be estimated for clinical staging. In our case, the ABC component did not affect the staging because the tumor was confined to the cervix. However, we must keep in mind that we might overestimate the clinical stage, resulting in overtreatment.

References