



Villoglandular papillary adenocarcinoma co-existing with high-grade squamous intraepithelial lesion; arising from an endocervical polyp

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ABSTRACT

Adenocarcinomas account for 10-20% of invasive cervical carcinomas. The villoglandular papillary adenocarcinoma has been recognized as a subtype of mucinous adenocarcinoma and identified as a distinctive histological entity from the usual invasive adenocarcinomas of the uterine cervix. We report a new case of villoglandular papillary adenocarcinoma, which was peculiar because of its association with a co-existing high grade squamous intraepithelial lesion and arising from an endocervical polyp. Excellent prognosis of isolated villoglandular papillary adenocarcinoma is important in terms of making differential diagnosis from other adenocarcinomas and eliminating unnecessary aggressive treatments. It is also important to keep in mind for pathologists since it allows them to examine endocervical polyps that we often encounter in daily life and shows that malignant tumoral lesions can develop from polyps.

Keywords: villoglandular papillary adenocarcinoma, high grade squamous intraepithelial lesion, endocervical polyp, rare

INTRODUCTION

Adenocarcinomas account for 10-20% of invasive cervical carcinomas (1). The villoglandular papillary adenocarcinoma (ADK VG) has been recognized as a subtype of mucinous adenocarcinoma and identified as a distinctive histological entity from the usual invasive adenocarcinomas of the uterine cervix. Villoglandular papillary adenocarcinoma of the uterine cervix was (1989) described by three main histological features: Exophytic proliferation, papillary architecture and mild to moderate cellular atypicality. The authors report a case of villoglandular papillary adenocarcinoma with clinical stage IB. Five main clinicopathological features of the villoglandular papillary adenocarcinoma could be emphasized: Rare histological variant, young age of patients (25-45 years old), superficial stromal invasion, usual association with other tumoral patterns and excellent prognosis (2). We report a new case of ADK VG, which was peculiar because of its association with a co-existing high grade squamous intraepithelial lesion (HSIL) arising from an endocervical polyp.

CASE REPORT

A 41-year-old woman, G3P2, presented with a 3-month history of abnormal vaginal bleeding. The patient had no history of oral contraceptive use. Cervical examination revealed an endocervical polypoid lesion. The patient underwent polypectomy. Macroscopically, the polyp was approximately 1 cm in diameter and no macroscopic feature was seen. In the microscopy, a tumoral structure composed of fine papillary structures with fibrovascular cores toward the surface in the polypoid lesion covered with a single layered mucinous epithelium was observed. The papillary structures lined by stratified columnar epithelium with hyperchromatic nucleus and narrow pink cytoplasm showing mild to moderate atypia, containing some atypical mitosis. Tumor cells were

immunohistochemically diffuse with pancyokeratin and focal positive with CEA. PHH3 was positive in mitosis in the tumor cells. No invasive areas were seen in many serial sections. HSIL was detected on the tumor-adjacent surface epithelium which is free of tumor (Figure a-f). Based on these findings, the case was diagnosed with villoglandular papillary adenocarcinoma developed from endocervical polyps. Laparoscopic hysterectomy and salpingectomy was applied on the patient. No residual tumor was seen in the surgical material.

DISCUSSION

The ADK VG is a rare histological variant. Since it was first described by Young and Scully in 1989, few case series have been reported worldwide. Clinically, VGPA is most prevalent in younger women compared to common types of cervical adenocarcinoma. Young and Scully described 13 cases with a mean age of 33 years (3). In our case, the patient was 41 years old. The clinical manifestations of ADK VG are non-specific and the majority of cases show abnormal vaginal bleeding or postcoital bleeding (3). Our patient is presented with abnormal vaginal bleeding.

In the literature, there are studies suggesting that oral contraceptives either have a role (4) or don't (3) in the etiology of ADK VG. In our case, there was no history of oral contraceptive use. The role of oral contraceptives in the etiology is still controversial and there is a need for further studies.

The three main histological features of ADK VG were exophytic proliferation, papillary architecture and mild to moderate cellular atypicality (3). The villous component of the tumors was similar in appearance to villous adenoma of the colon (5). Microscopically, the papillae were tall, thin and had a fibrous central core. The invasive portions of the tumor were composed of elongated branching glands, separated by a fibromatous stroma. Occasionally, a desmoplastic stromal

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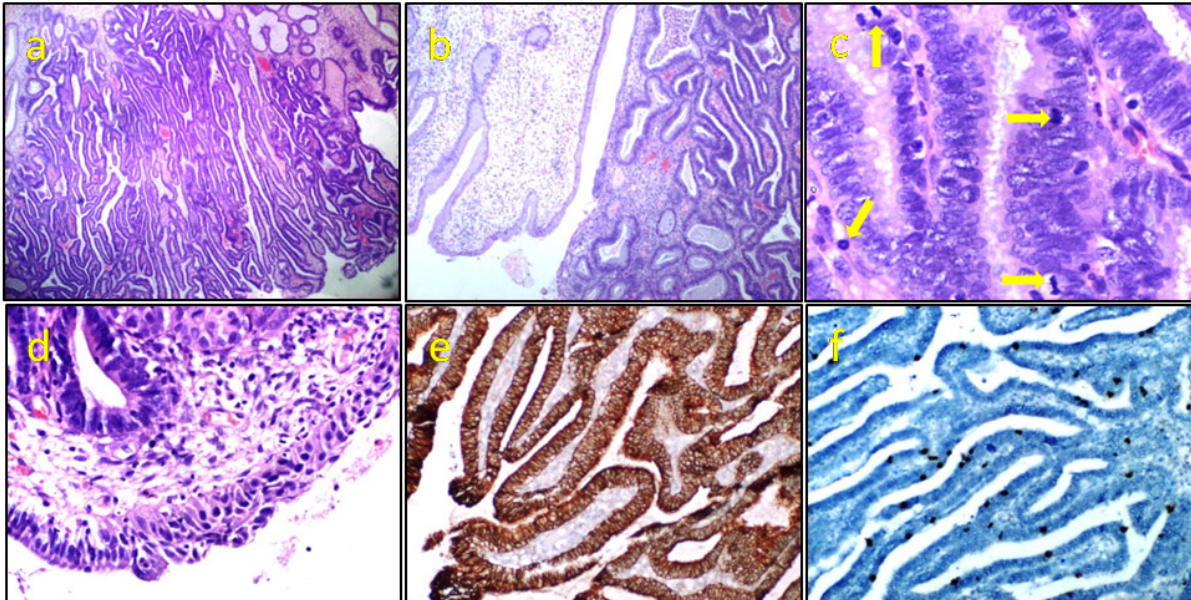


Figure 1: Representative pathological images of the tumoral lesion. a. Tumoral lesion (H&E x 20). b. Transition zone from the endocervical epithelium to tumor (H&E x 40). c. The arrows indicate an atypical mitosis (H&E x 400). d. The area of HSIL (H&E x 200). e. Positive immunostaining for pancytokeratin (x 100). f. Positive immunostaining for PHH3 in atypical mitosis (x 100).

response was noted. The epithelial lining of the papillae was covered by cells exhibiting mild to moderate nuclear atypicity (3). In our case, the mid-severe level nuclear atypia was seen in the epithelial of tumoral structure.

According to Jones et al., ADK VG was commonly associated with other patterns, of which the most frequent were: Adenocarcinoma in situ (40%), cervical intraepithelial neoplasia (30%) (4). Both squamous cell carcinoma in situ and ADK VG were described in three cases (3) and both invasive squamous cell carcinoma and ADK VG in two cases (5, 6). In our case, HSIL was co-existed with ADK VG in an endocervical polyp.

In the study of Young and Scully (3) and Jones *et al* (4), 9 tumors with no stromal invasion, 24 tumors with superficial invasion confined to the inner third of the cervical wall and 3 tumors with deep invasion of the cervical wall were described. In our case, unlike those in the literature, the tumor was limited in the endocervical polyps and there was no invasion to the stroma of polyps.

Generally, the outcome of cervical adenocarcinoma is considered to be worse than that of squamous cell carcinoma (7). However, ADK VG has been reported to have a limited metastatic potential and an excellent prognosis (3, 4, 8). The excellent prognosis of this tumor raised the question of a conservative surgical approach, especially if the reproductive

function needed to be preserved (3, 4). In the literature, cases with long follow-up periods and underwent cervical conization have been reported only (3, 4, 9). Most of the authors had a radical surgical approach and in some cases, para-aortic lymph node dissection (5, 6, 10). The advantage of hysterectomy compared to conservative treatment is not requiring long-term clinical follow-up (3). According to Jones et al., the most important feature for the choice was the stromal invasion (4). On the other hand, according to Young and Scully, a cone biopsy was the initial choice of therapy in selected cases: Margins of the cone biopsy clear of disease no more than a 3-mm depth of invasion and no vascular or lymphatic space invasion (3). In our case, no stromal, vascular or lymphatic invasion was observed. Hysterectomy and salpingectomy was applied on the case without needing lymph node dissection and protecting ovaries since our case was not expecting to have any children.

Excellent prognosis of isolated ADK VG is important in terms of making differential diagnosis from other adenocarcinomas and eliminating unnecessary aggressive treatments. It is also important for pathologists since it allows them to examine endocervical polyps that we often encounter in daily life and shows that malignant tumoral lesions can develop from polyps.

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