Villoglandular papillary adenocarcinoma of the uterine cervix diagnosed during pregnancy

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Summary

Villoglandular papillary adenocarcinoma (VPA) is a very rare subtype of adenocarcinoma of the uterine cervix but a well recognized variant of cervical adenocarcinoma with a favorable prognosis generally occurring in women of child-bearing age. Only five cases of VPA and pregnancy have been reported. Herein, we report a case of VPA diagnosed during pregnancy and this patient delivered a healthy baby. A successful pregnancy can be completed in patients with VPA without lymph-vascular invasion, when treated conservatively. This management is particularly desirable in young women to preserve reproductive capability.

Key words: Villoglandular papillary adenocarcinoma; Pregnancy; Conization.

Introduction

The incidence of cervical adenocarcinoma has been on the rise over the last decades. Villoglandular papillary adenocarcinoma (VPA) is a very rare subtype of adenocarcinoma of the uterine cervix. The true incidence of this form of adenocarcinoma is unknown. The classical histologic appearance of this entity is a surface papillary component of variable thickness with papillae that are usually tall and thin, but occasionally short and broad, with a fibrous stromal core. The tumor cells should have no more than mild-to-moderate nuclear atypicality and scattered mitotic figures. VPA affects a younger age group and has an excellent prognosis as compared to other endocervical adenocarcinomas [1, 2]. To our knowledge, only five cases of VPA associated with pregnancies have been reported in the literature [3-7]. We report here a successful pregnancy with Stage IB2 VPA of the cervix.

Case Report

A 32-year-old woman, gravida 0, para 0, was referred to our hospital at 33 weeks' gestation due to the positive cytological finding of adenocarcinoma. Physical examination revealed a 5-cm bleeding exophytic cervical tumor with no evidence of vaginal fornices or parametrial involvement (FIGO Stage IB2). We performed a biopsy of the uterine cervix, and verified cervical VPA (Figure 1). The patient underwent cesarean section (CS) at 34 weeks, and delivered a healthy 2,282 g newborn. Semi-radical hysterectomy with pelvic lymphadenectomy was performed two weeks after CS. Although VPA rarely has lymph node metastasis, this case had metastasis to the obturator lymph nodes. Following five courses of adjuvant chemotherapy (paclitaxel 180 mg/m^2, carboplatin AUC 6), the patient has shown no evidence of disease recurrence for 30 months.

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Discussion

We report an extremely rare case that was diagnosed with VPA during pregnancy with successful results both for the mother and baby. VPA of the uterine cervix is a rare form of cervical adenocarcinoma first described by Young and Scully in 1989 [1]. They found that this tumor has an excellent prognosis and suggested conization as a potential treatment for patients of childbearing age [1]. Conservative management of cervical VPA is considered to be a significant challenge; however, the English literature concerning treatment of VPA diagnosed during pregnancy is sparse. So far, over 115 cases of cervical VPAs have been reported worldwide; of these only nine metastases and two deaths were reported [7-11]. These few cases show an apparent discrepancy from the excellent prognosis of VPA described originally by Young, Scully and others [1, 12]. In 30% of cases, VPA is associated with other forms of invasive cancer [1-4, 7] which may have an important impact on the prognosis. Young and Scully therefore reserve the term VPA for tumors in which the villoglandular pattern is the exclusive or almost exclusive one. It has been suggested that in cases of superficial VPA diagnosed in young patients, unassociated with another type of cervical tumor and without lymph vascular invasion, less radical treatment may be suitable since these cases present a favorable outcome [12]. However, since the knowledge of the biologic spectrum of VPA appears to be evolving, a close follow-up should be pursued in VPA patients managed conservatively [13].

Pregnancy associated with VPA of the cervix has been reported in only five cases. In three cases, successful pregnancies were achieved following conservative treatment for VPA [3-5]. Two additional cases were diagnosed during pregnancy; one case ended with an early induced abortion (8 weeks of gestation) followed by a radical hysterectomy [7], and the second case, which was diagnosed during the 20th week of gestation, was conservatively

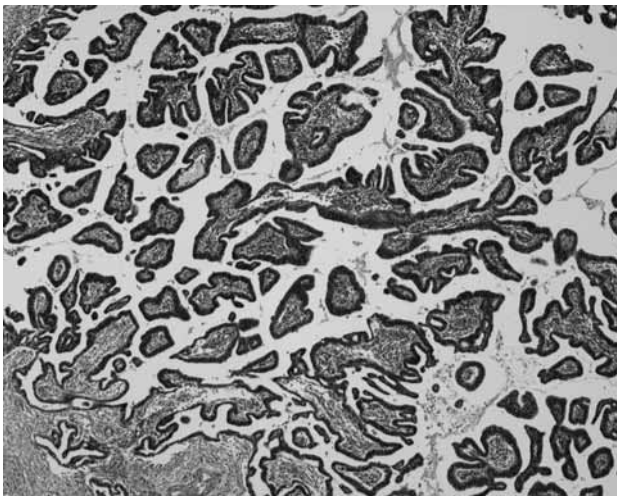

Figure 1. — Typical histological patterns for villoglandular papillary adenocarcinoma of the cervix. Tumor displaying thin and tall, well-formed papillary structures. Large glandular and papillary structures with stroma resembling those of the normal cervix (hematoxylin and eosin, original magnification, × 40).

followed until the 32nd week of gestation, when a cesarean radical hysterectomy was performed [6]. In the case whose pregnancy was terminated (8 weeks of gestation) followed by a radical hysterectomy and pelvic lymphadenectomy, the patient died of a tumor recurrence [7], suggesting that some VPAs are malicious, especially when other histological features are present. These authors recommend the attitude, "Beware of a wolf in sheep's clothing", in relation to VPA [14].

In conclusion, despite the limited experience of cervical VPA diagnosed during pregnancy, conservative treatment can be successfully achieved in selected patients after a thorough evaluation of the depth of invasion, the lymph vascular involvement and the association of other carcinoma histologies in conjunction with the VPA (i.e., adenocarcinoma or squamous cell carcinoma).

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