Case Report

A Case of Villoglandular Papillary Adenocarcinoma of the Uterine Cervix Diagnosed during Early Pregnancy Followed by Successful Term Delivery

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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 and 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 early pregnancy and managed successfully with conservative measures; our patient delivered a healthy baby in full term. 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.

1. Introduction

The incidence of cervical adenocarcinoma is 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. It 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]. In two cases, the patients were diagnosed VPA during pregnancy followed by conservative treatment and delivered healthy children [5, 7]. We report here a successful term pregnancy with stage IB VPA of the cervix diagnosed during early pregnancy.

2. Case Report

A 28-year-old Japanese woman, gravida 1, para 1, was admitted with atypical genital bleeding and underwent polypectomy of the uterine cervix at 9 weeks’ gestation. The 1-cm resected polyp was pathologically diagnosed as VPA (Figure 1). The tumor is purely exophytic without invasion of the underlying stroma and lymphvascular involvement. Hence, she was referred to our hospital. As the polyp had no lymph capillary space invasion, we performed conization at 16 weeks’ gestation when the patient decided to continue her pregnancy. The depth of surgical specimen was 1 cm and width of that was 3 cm diameter. No cancer cells were identified in the resected specimens. The final diagnosis was FIGO stage IB1. She delivered a healthy 2,946 g newborn vaginally at term 38 weeks, and she has been free of the disease for 44 months.

3. Discussion

We report an extremely rare case who was diagnosed VPA in the 1st trimester and was managed conservatively
throughout the pregnancy with successful results for both
the mother and the 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, 6], 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].

Young and Scully recommended careful inspection of
the histological specimen and if the villoglandular com-
ponent is the exclusive or almost exclusive pattern then
a diagnosis of VPA can be ascribed [1]. Other papillary
adenocarcinomas can present a difficulty in diagnosis. Serous
papillary adenocarcinomas of the cervix have finer, more
irregular and more cellular papillae than VPA. The clear cell
papillary adenocarcinomas of the cervix are characterized
by marked cytological atypia, high mitotic activity and
occasionally the presence of psammoma bodies. VPA should
be distinguished from endocervical adenocarcinoma with a
minor villoglandular component. The rare adenosarcoma
and adenoma malignum should also be considered in the
differential diagnosis of VPA [8].

Pregnancy associated with VPA of the cervix has been
reported in only five cases [3–7]. In two cases, success-
ful pregnancies were achieved following a conservative
treatment for VPA [3, 4]. Three additional cases were
diagnosed during pregnancy (Table 1); the first case, which
was diagnosed during the 20th week of gestation, was
conservatively followed until the 32nd week of gestation,
when a caesarean radical hysterectomy was performed [5],
the second case ended with an early induced abortion (8
weeks of gestation) followed by a radical hysterectomy [6],
and the third case, which was diagnosed during the 13th
week of gestation, was conservatively followed until the 37th
week of gestation, when a caesarean radical hysterectomy
was performed [7]. In the case whose pregnancy was
terminated (8 weeks of gestation) followed by a radical
hysterectomy, the patient underwent second, third and
fourth laparotomies because of recurrent pelvic masses. At
the end of five years follow-up period, she died because
of the complication of recurrent tumor [6]. Bouman et al.
reported three cases of VPA, and two of these are malicious
because they have other histological features (the first case
showed well-differentiated adenocarcinoma with abundant
squamous differentiation, and the second case has well to
moderately differentiated papillary adenocarcinoma) [14].
These authors recommend the attitude, “Beware of a wolf
in sheep’s clothing”, in relation to VPA [14]. However, we
can not discuss the clinical outcome in pregnant patients,
because there is no report of VPA accompanied with other
histological features or extended tumor invasion in pregnant
patients.

Figure 1: Typical histological patterns for villoglandular papillary
adenocarcinoma of the cervix. (a) Tumor displaying thin and tall,
well-formed papillary structures (hematoxylin and eosin, original
magnification, x4), (b) Higher magnification of (a). Large glandular
and papillary structures with broad stroma (hematoxylin and
eosin, original magnification, x40), (c) Higher magnification of (b)
(hematoxylin and eosin, original magnification, x200).
Table 1: Profile of patients with villoglandular papillary adenocarcinoma diagnosed in pregnancy.

| Reference | Present case | [5] | [6] | [7] |
|-----------|--------------|-----|-----|-----|
| Age       | 28           | 22  | 28  | 31  |
| Gravida   | 1            | 3   | 3   | 2   |
| Para      | 1            | 2   | 2   | 1   |
| Gestational age at diagnosis (weeks) | 9 | 20  | 8   | 13  |
| Gestational age at delivery (weeks) | 38 | 32  | 8   | 37  |
| Mode of delivery | VD | C/S | Termination | C/S |
| Macroscopic feature | Polypoid | Polypoid | Polypoid | Polypoid |
| FIGO stage | IB1 | IB2 | IB1 | IB1 |
| Treatment | Conization | RH | RH | RH |
| LCS1 | — | — | — | — |
| Lymph node metastasis | Not examined | — | Not examined | — |
| Follow-up (months) | 38 | 14 | 60 | 18 |
| Outcome | NED | NED | DOD | NED |

VD: vaginal delivery; C/S: cesarean section; FIGO: Fédération Internationale de Gynécologie et d’Obstétrique; RH: radical hysterectomy; LCS1: lymph capillary space invasion; NED: no evidence of disease; DOD: dead of disease.

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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