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Spindle-cell epithelioma of the vagina diagnosed during pregnancy - a case report

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Summary

Spindle-cell epithelioma or "mixed tumor" of the vagina is an unusual and intriguing vaginal tumor consisting of both epithelial and mesenchymal components. A case of spindle-cell epithelioma of the vagina diagnosed at delivery of a 31-year-old primiparous woman is described. The excision of the mass was performed immediately after the delivery, which was uneventful. The patient was regularly followed up and no evidence of local recurrence or dissemination was found 40 months after surgery. The presentation and the diagnosis of this kind of tumor in pregnancy, and its effect on the pregnancy and delivery are still largely unknown. Since it is unlikely that any institution will have a large number of patients with this rare disease, case reports add further information to this entity. As the number of cases studied is small, close follow-up is recommended although there has been no report in the literature of metastasis so far.

Key words: Spindle-cell epithelioma; Mixed tumor; Vagina; Pregnancy.

Introduction

Spindle-cell epithelioma or intriguing mixed tumor of the vagina refers to a rare neoplasm composed of a proliferation of the spindle cells admixed with epithelial cell islands within.

Almost all of the reported tumors were small and well circumscribed, cited in or above the hymen in adult women. Many theories have been given on the origin of these tumors, ranging from possible embryonic remnants being the source, to its origin from the Müllerian ducts or urogenital sinus [1]. Its histogenesis, however, is still unclear.

This condition is most often asymptomatic and discovered accidentally on routine pelvic examination. It usually presents as a nodular mass located near the hymenal ring and may present with vaginal discharge or bleeding [2].

Case Report

A 31-year-old nullipara was referred to the obstetrics department at 41 weeks of gestation for delivery. The antepartum course was unremarkable. Her personal history contained no significant diseases or tumors. On admission, the cervix was three-cm dilated and 0.5 cm long. Vaginal examination also revealed a three-cm, mobile, painless, non-tender pedunculated mass arising from a short (0.2 cm) stalk situated on the left side of the posterior vaginal wall. The tumor was located about two cm behind the hymenal ring, and appeared neither necrotic nor infected. A rectal examination did not reveal any abnormalities, and the patient had no complaints regarding the tumor.

Since the vaginal outlet was not obstructed with the tumor, the authors decided to perform vaginal delivery, which was uneventful and performed through a right mediolateral episiotomy. A male baby of 3,500 grams with an Apgar score of 9 was born. Upon delivery, due to the unknown nature of the tumor, the authors performed the excision. Due to the unknown nature of the tumor, deep dissection was performed and both the placenta and the tumor were submitted for histopathological examination, which did not reveal any abnormalities. Grossly, the excised tumor measured 30×30×25 mm. On section, the cut surface exhibited a pale yellowish submucosal nodular mass of firm consistency.

On microscopic examination, the mass was well circumscribed but unencapsulated, with expansive growth. The tumor was covered with normal vaginal squamous epithelium and consisted of two components: stromal-type spindle cells, which were predominant, and epithelial cells with hyaline globules between them. Neither hemorrhage nor necrosis in the tumor was seen. The mitotic figures were absent (Figures 1A and B).

Immunohistochemistry was performed on paraffin sections, with a broad spectrum of immunohistochemical stains. A positive reaction for pan CK and CD 10 was observed in both components (Figures 2A and B). Vimentin activity was noted only in stromal-type cells. They were uniformly positive for CK7 (cytokeratin) and negative for CK20, smooth muscle actin, desmin, chromogranin, synaptophysin, and S-100 protein. The epithelial component was positive for CK5/6, monoclonal carcinoembryonic antigen and tumor protein 63, while the Ki67 immunopositivity was low (5%). Based on light microscopic appearances along with the immunohistochemical staining, the final diagnosis of spindle-cell epithelioma of the vagina was made.

The postoperative course was uneventful and the patient was discharged on the third postpartum day. At the four-week postpartum visit, the vaginal epithelium was completely healed and free of any lesions or scarring. The patient was regularly followed

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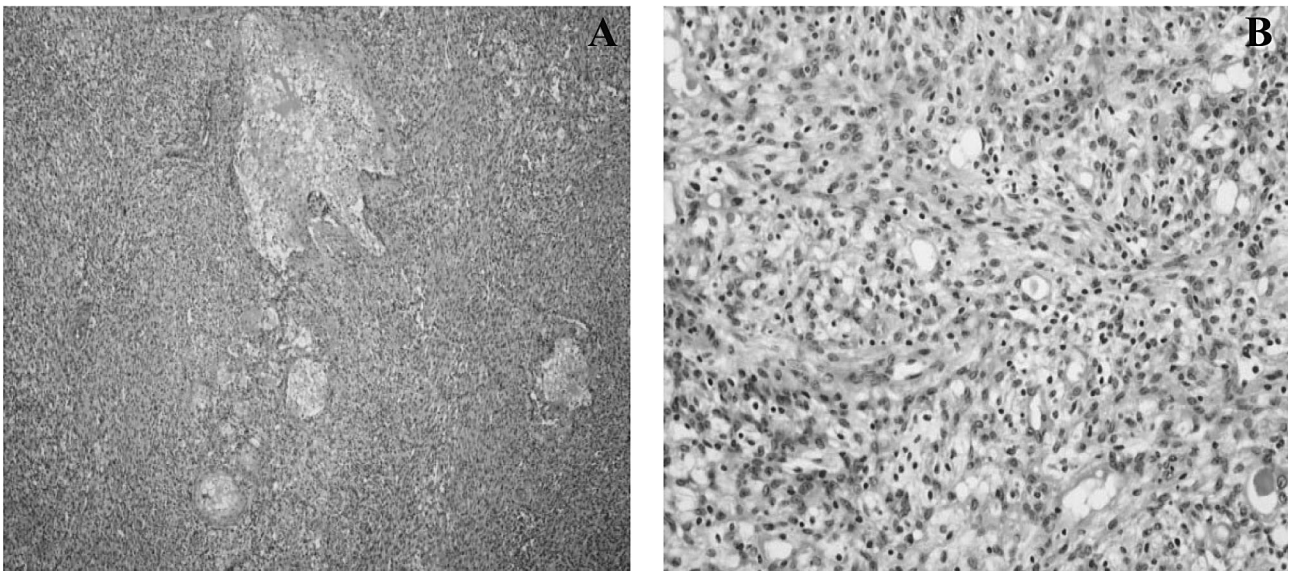

Figure 1. — A) Tumor is composed of a spindle cell population admixed with clusters of cells with epitheloid features or squamous differentiation (hematoxylin and eosin, x100). B) There are hyaline globules scattered throughout between predominantly stromal-type spindle cells and epithelial cells (hematoxylin and eosin, x200).

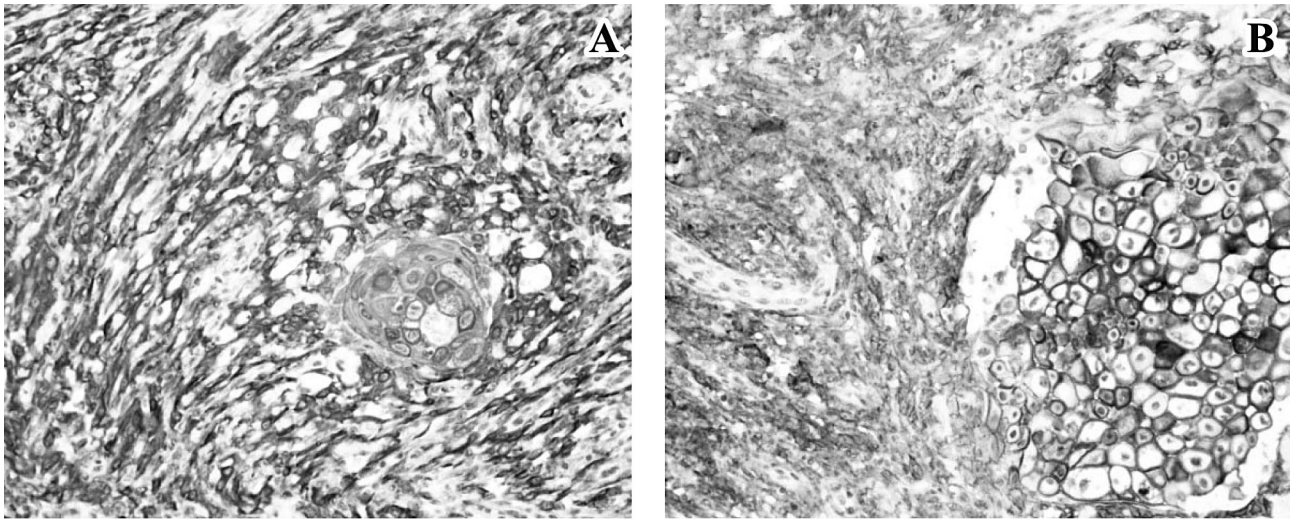

Figure 2. — On immunohistochemistry both epithelial and spindle cell tumor population are diffusely and strongly staining positive for pan CK 10 (A, x200) and for CD 10 (B, x200).

up and there was no evidence of local recurrence or dissemination 40 months after the surgery.

Discussion

Clinical experience with cell epitheliomas of the vagina is limited. The presentation and the diagnosis of this kind of tumor in pregnancy, and its effect on the pregnancy and delivery are still largely unknown. The first reported case of this kind of tumor in the English literature was published in 1953, followed by other case reports, which indicated the average age of occurrence being 40.5 years, with a reported range from 20 to 80 years [2]. In 1993 Branton *et al.* published a detailed study of 28 cases, still the largest series of its kind [3].

One would consider that a vaginal mass in pregnancy should be excised if obstructing the vaginal outlet. In the present case, the reason for surgical intervention after the delivery was unknown nature of the tumor. Because of the increased vascularisation of the vagina in a term pregnancy, special care is required to minimize the blood loss during surgery in such cases.

Based on the available reports, local excision is considered curative and the prognosis is generally good [2]. Recurrence occurs when the tumor is incompletely excised [3, 4]. In the recurrent cases, no unique features other than apparent incomplete excision were noted [2]. As the number of cases studied is small, close follow-up is recommended, although there has been no report in the literature of metastasis so far [2].

The clinicopathological features of the present case were similar to previously reported cases [5, 6]. A pathological diagnosis of spindle-cell epithelioma of the vagina should always be kept in mind whenever a polypoid mass near the hymenal ring is excised. Spindle-cell epitheliomas are distinct neoplasms and should not be confused with "mixed tumors" occurring at other sites such as the salivary and lacrimal glands, breast, mediastinum, trachea, skin, and vulva [5, 6]. These tumors should be differentiated from other tumor lesions, such as aggressive angiomyxoma, solitary fibrous tumor, malignant mixed tumor, and malignant tumor of the vagina resembling synovial sarcoma [2].

In conclusion, familiarity with these rare tumors among pathologists and gynecologists would perhaps lead to the identification of more of cases and limit misdiagnosis and surgical complications.

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