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CASE REPORT

INVERTED PAPILLARY ADENOFIBROMA OF THE UTERINE CERVIX

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Abstract

A case of benign "inverted" papillary adenofibroma of the uterine cervix in a 48 year-old woman was presented. It was considered to be a benign counterpart of malignant mixed müllerian mesodermal tumor like other papillary adenofibromas of this region. Our case was unique for its inward papillary growth and review of the literature disclosed no other case reports of identical lesions.

INTRODUCTION

A variety of polypoid lesions arise from the uterine cervix, and the commonest is the mucosal polyp[1]. Benign tumors in the uterine cervix is relatively infrequent. Squamous papillomas and leiomyomas are common among these[1]. Lately, another type of polypoid cervical tumor with distinct histologic characteristics has been reported[2]. Because of its lobulated and papillary configuration with endocervical epithelium covering a solid fibroblastic growth, and its resemblance to the neoplasm in the ovary and female mammary gland, it has been termed papillary adenofibroma. Recently, we have encountered a polypoid lesion in which papillary adenofibroma grew within the polypoid mass without the evidence of external proliferation. In analogy to the inverted papilloma of the nose and urinary bladder, we named it inverted papillary adenofibroma. So far, ten typical[2,3,4] and two possible[1] papillary adenofibroma of the cervix have been reported, however, no reports of lesions identical to ours have been found in the literature. Herein, the first case of inverted papillary adenofibroma of the uterine cervix is presented.

CASE REPORT

A 48 year-old female (Gravida 1, Para 1) was pointed out to have anemia a few months ago. Her menstrual cycle was regular (every 35 to 40 days)

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with eight days' duration. Originally, she visited the Out-patient clinic of the Hospital of the Kawasaki Medical School and was transferred to OB-GYN service for further evaluation. A large polypoid mass was found protruding through cervical os. It was apparently arising from the anterior wall of the endocervix (12 o'clock), 1.5 cm inner from the verge. This polypoid mass was entirely removed and submitted for histological examination. There has been no recurrence during observation period of two and half years after polypectomy.

PATHOLOGICAL FINDINGS

An excised polypoid mass measured 8 × 15 mm in size. It was bisected and examined histologically. The outer surface was lined mostly by squamous mucosa with focal chronic inflammatory infiltrates but partially by endocervical

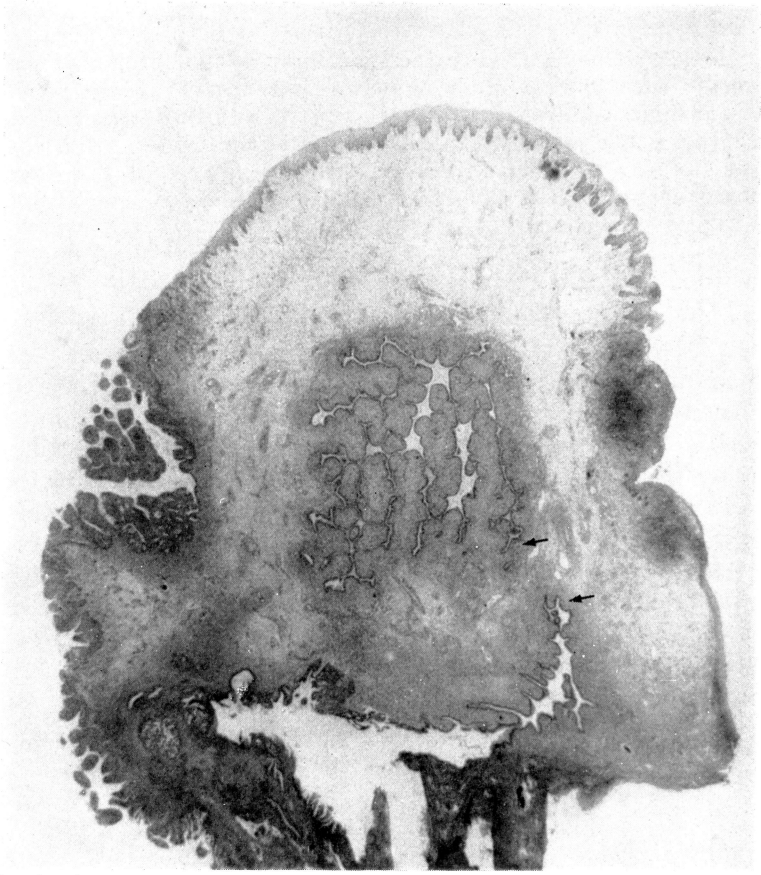

Fig. 1. An area with papillary projections of adenofibroma in the midst of the specimen. Arrows indicate a possible communication between space in the tumor and endocervical canal. Note that the outer surface of this polyp is of normal endo-and exo-cervical mucosa. (H-E, ×8)

mucosa with focal squamous metaplasia, which showed somewhat papillary structures. A nodular area with papillary projections was present in the midst of this polypoid mass (Fig. 1). This lesion consisted of papillary and nodular excrescences of cellular fibrous tissue. Fibroblastic cells ran in haphazard fashion. Their nuclei were rather small and uniform with absence of nucleoli. No mitotic figures were identified. Stroma contained fairly large numbers of capillaries and arterioles. The periphery of the lesion was more myxomatous. The covering epithelium was usually columnar and composed of tall columnar cells with picket-fence arrangement of nuclei, ciliated cells and secretory cells (Fig. 2). It gave some resemblance to that of normal endocervix or fallopian tube. Squamous metaplasia was not present in any areas. Luminal space contained a small amount of faintly eosinophilic mucoid material. Multiple serial sections suggested the continuation of these lining cells with the outer surface epithelium (Fig. 1 arrows). Therefore, it was felt that papillary projections present in the midst of this polyp was in fact an inward growth of papillary tumor. Both epithelial and stromal element showed no cytological atypism.

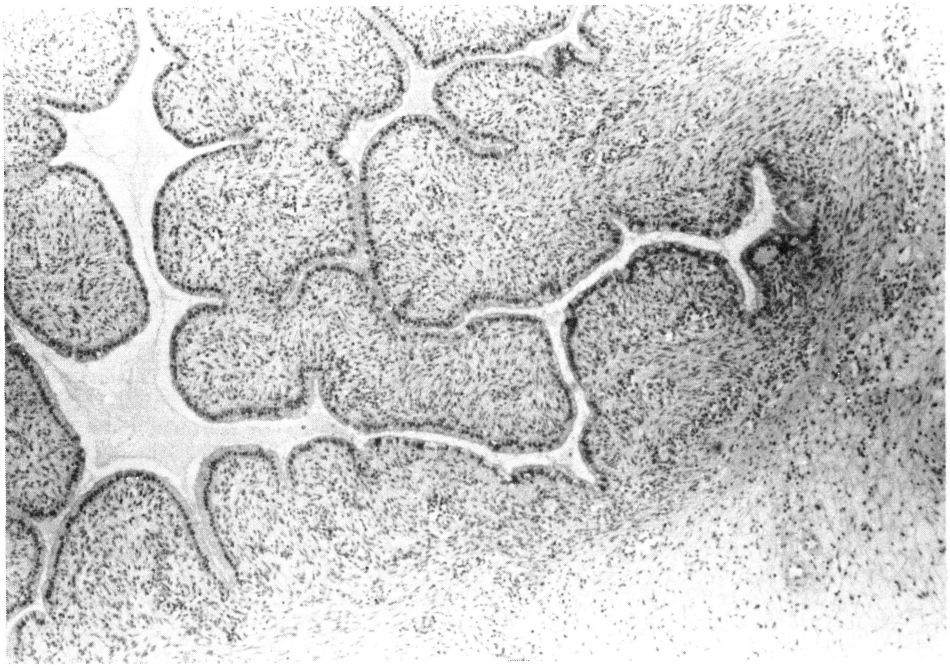

Fig. 2. Papillary and nodular excrescences of fibroblasts. Various types of epithelium are seen here. (H-E, ×100)

DISCUSSION

Papillary adenofibromas may arise from any portion of the uterus. It is

a benign papillary tumor of fibroepithelial nature and gives some resemblance to similar growths seen in the ovary and female mammary gland. Such tumors have been observed in the endocervix[2,3,4], in the fallopian tube[5], and in the endometrium[3,4,5].

So far, ten typical[2,3,4] and two possible[1] cases of papillary adenofibroma arising in the uterine cervix have been reported. Such tumors usually grew outward but some microcysts with papillary excrescence were almost always present within the stroma. Unlike our case, none reported an inward growth of such tumor producing a polypoid mass, the outer surface of which was lined by normal cervical epithelium. For this reason, we designated our tumor more descriptively as "inverted papillary adenofibroma."

The origin of such tumors has been still in debate. Vellios et al.[3] felt that both epithelial and stromal elements were neoplastic and regarded it as the benign counterpart of the malignant mixed müllerian tumor. Furthermore, citing Christopherson's statement, Vellios[4] proposed a term of uterine cystosarcoma phyllodes to cover an entire spectrum of such tumors with both benign and malignant potentials. In contrast, Grimalt et al.[6] considered that the epithelial component of the tumor was rather passive, and that it was a pure stromal tumor with surrounding pre-existing epithelial lining. Because of inward growth of our tumor, it is difficult to believe that stromal element has evoked the proliferation of epithelial element. We agree with the idea of Vellios et al. and feel the mixed müllerian origin to be most likely in our case.

A lack of stromal cell pleomorphism, necrosis and mitotic figures as well as the absence of the recurrence during 2 1/2 years of observation makes us consider strongly that the tumor under discussion is benign.

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