ORIGINAL RESEARCH PAPER
Volume-8 | Issue-9 | September - 2019 | PRINT ISSN No. 2277 - 8179 | DOI : 10.36106/ijsr
INTERNATIONAL JOURNAL OF SCIENTIFIC RESEARCH

PAPILLARY CYSTADENOFIBROMA OF ENDOMETRIUM: A RARE CASE REPORT AND REVIEW OF THE LITERATURE

Pathology

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ABSTRACT

In female genital tract, an adenofibroma is commonly reported in ovary and rarely in cervix while it is extremely rare in endometrium. Here, we report a case of papillary cystadenofibroma of uterus in a postmenopausal woman with history of breast carcinoma. In our review of published case reports, very few cases have been reported so far arising from endometrium, presenting as polypoidal lesions in uterine cavity. In all reported cases, six patients had history of breast carcinoma. There has been an attempt to establish relationship between adenofibroma of uterus and tamoxifen intake in patients with breast carcinoma. However, our patient did not receive tamoxifen. In a view of reported incidence of these tumours in breast cancer patients we propose a theory that uterine mixed Müllerian tumour may be secondary to yet unknown genetic risk factor independent of tamoxifen.

KEYWORDS

cystadenofibroma, breast carcinoma, mullerian mixed tumour, tamoxifen

INTRODUCTION:

Endometrial adenofibroma is a rare mixed Müllerian tumour, which is essentially benign. Adenofibroma of uterus was first described by Ober in 1959 as a mixed mesodermal tumour(1). In 1971, Abell described a similar cervical lesion as papillary adenofibroma owing to its papillary configuration(2). Adenofibroma is composed of both active epithelial and mesenchymal elements. Presenting most often as a polypoidal mass in uterine cavity it is important to distinguish it from endometrial polyp or hyperplasia, also from its malignant counterpart adenosarcoma histologically.

There are few case reports of adenofibromas of endometrium in breast cancer patients with history of tamoxifen usage in most of them(3)(4)(5)(6). The relationship between the latter and development of adenofibroma has not been clearly established. Adenofibroma may occur in case of breast carcinoma independent of hormonal therapy as in our case.

CASE REPORT:

A 66-year-old woman (gravida 2, para 2; both LSCS), postmenopausal; known case of diabetes mellitus, hypertension and psoriasis, underwent routine pelvic ultrasound examination which showed endometrial hyperplasia. On magnetic resonance imaging (MRI) of pelvis, thickened endometrium showed a polypoidal growth with cystic changes within.

Patient gave history of left mastectomy for breast carcinoma 9 years back and was given chemotherapy thereafter. Tamoxifen was not included in the treatment. Patient had one episode of postmenopausal bleeding 4 years after mastectomy.

In view of present imaging findings, a diagnostic hysteroscopy was performed followed by endometrial biopsy. On hysteroscopy, a polypoidal growth at cervico-isthmus junction within endometrial cavity was detected; seemed to be arising from lateral wall of uterus.

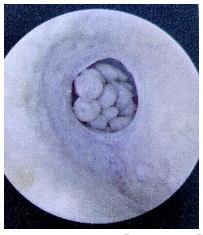

Figure 1: Diagnostic hysteroscopy shows polypoidal lesions in the endometrial cavity

The histopathological examination of the biopsy specimen on light microscopy showed a polypoidal lesion composed of fibrous tissue thrown into papillary fronds covered by cuboidal type epithelium.

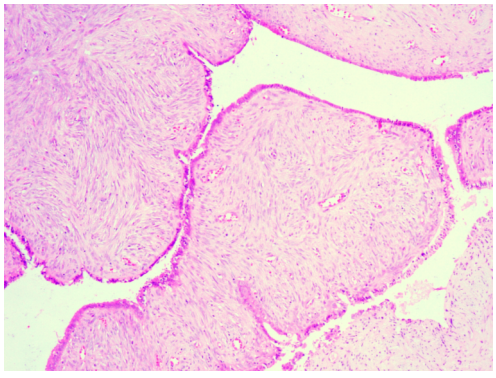

Figure 2: The micrograph shows polypoidal tissue lined by cuboidal epithelium with subepithelial spindle celled stroma(hematoxylin—eosin, original magnification x10)

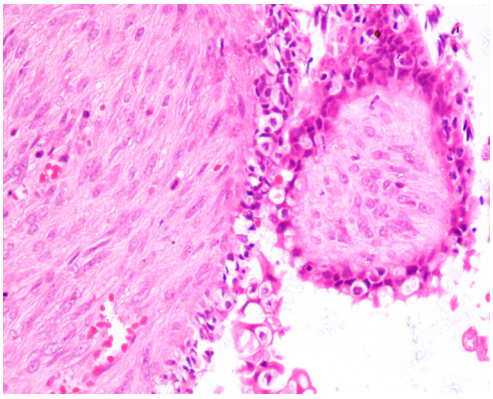

Figure 3: The micrograph shows absence of atypia or mitotic activity in the surface epithelium as well as in the subepithelial stroma(hematoxylin-eosin, original magnification x40)

Subepithelial stroma was spindle celled with a rare tubular gland. The stroma was hypocellular. Atypia and mitotic figures were not identified on the sections studied. Thus, histopathological diagnosis was that of a papillary cystadenofibroma.

DISCUSSION:

After Abell described papillary adenofibroma of uterine cervix, Grimalt, in 1975, coined the term papillary cyst adenofibroma due to presence of papillary fronds of fibrous stroma projecting into cystic spaces(7). Papillary cyst adenofibroma belongs to a group of a mixed epithelial and mesenchymal tumours(8). This tumour is most common in postmenopausal age group(8). However, there are few reports of young women presenting with the tumour(9)(10)(11). The most frequent presenting complaint is abnormal vaginal bleeding. Macroscopically, these are broad based polypoidal neoplasms with lobular or papillary surfaces. Their size ranges from 2 to 20 cm with a median diameter of 7 cm(10). Cut surface has spongy or mucoid appearance due to presence of multiple small cysts. Histologically, it is composed of a benign Müllerian epithelium and benign stroma. Gross and histologic findings in our case are similar to those described in previous cases in the published literature. It is important to distinguish adenofibroma from other endometrial lesions such as endometrial polyp and endometrial hyperplasia which have similar appearance on MRI as a polypoidal cystic lesion. However, polyps have rounded, smooth mucosa. They lack papillary processes, have more glands with central vasculature and are less cellular. Also, histologically an important differential diagnosis is a low grade adenosarcoma. It shows features of stromal mitotic count >1 mitosis per 10 high power field, marked stromal hypercellularity with periglandular cuffing and/or more than mild stromal atypia(10). All these features were not seen in our case.

In our review of literature, 6 patients with adenofibroma had a history of breast carcinoma in the past(3)(4)(5)(6). There has been an attempt to associate adenofibroma with the use of tamoxifen in breast cancer patients. Our patient did not receive tamoxifen. Wilson identified two women with malignant mixed Müllerian tumour who have had breast cancers and neither of them was given hormonal therapy(12). Causal relationship between tamoxifen usage and adenofibroma is not definitive(3). There may be yet unknown, possibly genetic component or other stochastic factor which might be responsible for development of malignant mixed tumour as well as its benign counterpart i.e. adenofibroma.

All cases reported in the literature have been treated by hysterectomy. It is the preferred treatment for this neoplasm because neoplasm may recur if it is incompletely removed(13)(1)(9). Also there is a rare possibility of invasion of underlying myometrium(14) or involvement by an adenocarcinoma(15). Hysterectomy allows thorough sampling of uterus to exclude adenosarcoma. Wide local excision via operative laparoscopy can be considered with long term follow up in women who wish to preserve their fertility(9). The patient we reported underwent total laparoscopic hysterectomy with bilateral salpingo-oophorectomy and is asymptomatic till date.

CONCLUSIONS:

Adenofibromas are clinically and histologically benign neoplasms(2). Importance of adenofibroma as a distinct clinical and structural entity lies in its resemblance with other lesions of endometrium which present with polypoidal morphology. Hence, it is essential to diagnose this lesion for proper treatment and to predict prognosis. Considerable incidence of adenofibroma in women with breast cancer with and without hormonal therapy as in our patient demonstrates the need to evaluate further a possible etiology. It may allow preventive hysterectomy in these patients.

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