

## CASE REPORT

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## Prolapse of the fallopian tube after hysterectomy associated with exuberant angiomyo fibroblastic stroma response: a diagnostic pitfall

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**Abstract** We report two cases of prolapse of the fallopian tube associated with an exuberant angiomyo fibroblastic stroma response, which occurred after hysterectomy and which is a hitherto unreported feature of this lesion. The tumors were composed of richly vascularized stroma arranged in a retiform pattern and mildly atypical glandular inclusions, which had the morphology of tubal epithelium. The stroma of the lesion was composed of either thin bipolar cells with tapered nuclei and stellate-shaped cells with minimal amount of cytoplasm or small epithelioid-looking cells with eosinophilic cytoplasm. The tubal glandular inclusions displayed mildly atypical nuclear features. If the tubal glandular component was overlooked, these tumors might be erroneously diagnosed as mesenchymal lesions of the vagina, such as vaginal fibroepithelial polyp, angiomyo fibroblastoma, aggressive angio myxoma, or superficial myo fibroblastoma.

**Keywords** Tubal prolapse · Fallopian tube · Exuberant angiomyo fibroblastic stroma formation · Angiomyo fibroblastoma

### Introduction

Prolapse of the fallopian tube following vaginal or abdominal hysterectomy was first described in 1902 by Pozzi [19] and, until now, less than 100 cases have been described [1, 2, 3, 4, 5, 11, 21, 24, 25, 26, 27, 28]. The morphology of the infiltrating tubal epithelium can often be atypical, and it can closely resemble adenocarcinoma [3, 21, 25]. We describe two cases of patients with tubal prolapse, in which the tubal epithelium caused exuberant stromal response and resembled a malignant tumor. We are not aware that such reactive stromal proliferation in association with tubal prolapse has ever been published.

### Materials and methods

Tissues were fixed in formaldehyde, paraffin-embedded, and routinely stained. Sections from both cases were also examined immunohistochemically using the avidin-biotin complex or streptavidin-biotin complex immunoperoxidase techniques, antibodies to smooth muscle actin (1A4, Dako Corp., Glostrup, Denmark), muscle-specific actin (HHF-35, Dako Corp.), S-100 protein (polyclonal, Dako Corp.), cytokeratins (AE1/AE3, Boehringer Mannheim and CAM 5.2, Becton Dickinson), desmin (D-33, Dako Corp.), and CD 34 (monoclonal, Dako Corp).

### Results

#### Case 1

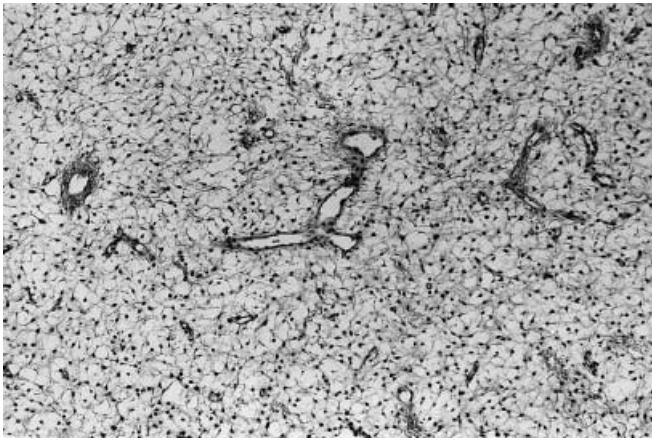
A 47-year-old woman underwent an abdominal hysterectomy and unilateral, left ovariectomy for multiple uterine leiomyomas. Four months after the hysterectomy, the patient presented with a right-sided vaginal vault tumor, which was 3 cm in diameter. No signs of pregnancy were observed in the patient. Two years after the excision of the tumor, the patient is well and without signs of recurrence.

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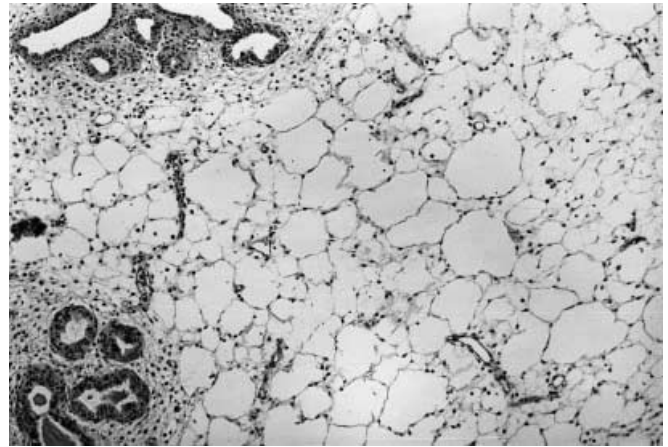
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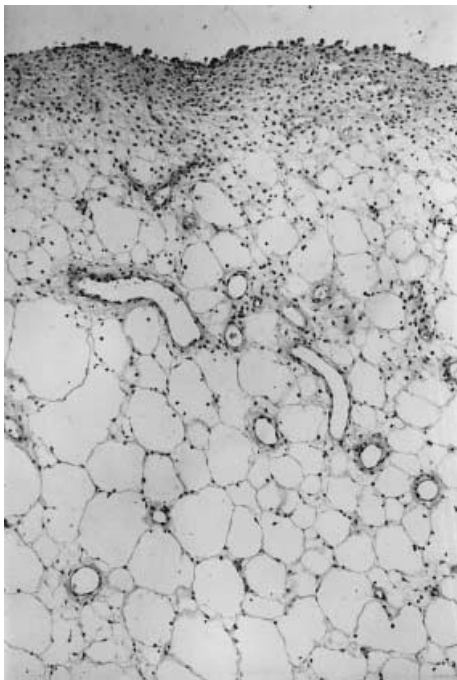
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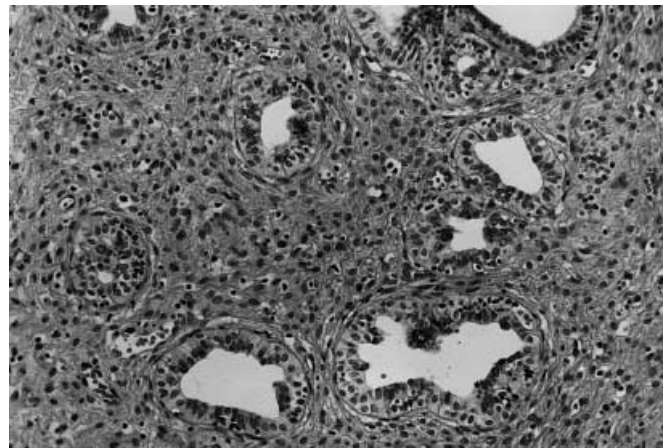
**Fig. 1** The main bulks of the tumors were composed of large areas consisting of richly vascularized stroma arranged in a retiform pattern. Hematoxylin and eosin stain;  $\times 125$



**Fig. 3** In some areas, the tumors contained tubal glandular inclusions. Hematoxylin and eosin stain;  $\times 125$



**Fig. 2** In one case, the retiform stroma was nearly microcystic. Hematoxylin and eosin stain;  $\times 125$



**Fig. 4** The stroma around the glandular tubal inclusions was composed of small epithelioid-looking cells with eosinophilic cytoplasm and contained a small amount of fibrous tissue lacking the myxoid quality of the purely stromal regions as seen in previous figures. Hematoxylin and eosin stain;  $\times 200$

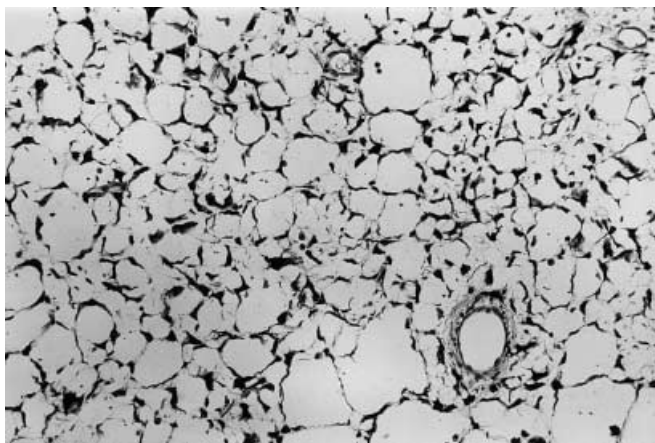
## Case 2

A 36-year-old woman, gravida 2, without any significant previous history of genital tract pathology, underwent a vaginal hysterectomy with bilateral conservation of the adnexa for multiple uterine leiomyomas. The postoperative period was uneventful. Twenty months later, at the age of 38 years, the patient returned to the hospital complaining of stress incontinence of urine. Upon vaginal exploration, there was a tender exophytic lesion 2 cm in diameter with bloody discharge at the left angle of the vaginal vault, which was surgically excised. No signs of

pregnancy were observed in the patient. Six years after the excision, the patient is well and without signs of recurrence.

Grossly, both botryoid-looking excised tumors had a red color and myxoid–gelatinous consistency. Histologically, both lesions looked similar. The main bulk of both tumors was composed of large areas consisting of richly vascularized stroma arranged in a retiform pattern (Fig. 1). In case 1, the retiform stroma was nearly microcystic (Fig. 2). The stroma in these areas was composed of thin bipolar cells with tapered nuclei and stellate-shaped cells with a minimal amount of cytoplasm. Both tumors also contained some glandular tubal inclusions (Fig. 3). These glandular tubal inclusions had focally mildly atypical nuclear features. The stroma around the glandular inclusions displayed a different appearance from that of the predominantly stromal areas, being composed of small epithelioid-





**Fig. 5** The stroma of the tumors was strongly desmin positive. Hematoxylin counterstain;  $\times 200$

looking cells with eosinophilic cytoplasm. The stroma in these areas contained a small amount of fibrous tissue and lacked the myxoid quality of the purely stromal regions (Fig. 4). A scant amount of round inflammatory cells was seen throughout in both lesions. Immunohistochemically, the tumor stroma was actin, cytokeratin, and S-100 protein negative and CD 34 and desmin strongly positive (Fig. 5).

## Discussion

Prolapse of the fallopian tube is a long known and histologically misleading phenomenon. Owing to the atypical nuclear features of the tubal glandular epithelium in some of the cases, prolapse of the fallopian tube can easily be confused with adenocarcinoma [3, 7, 9, 10, 18, 20, 21, 25]. Protean features of this lesion have been illustrated in two recently reported cases. A case of tubal prolapse was reported, where the tubal glandular epithelium contained signs of the herpes virus infection, adding to the pleomorphism of this lesion [15]. The other case of prolapse of the fallopian tube presented as a urinary bladder mass caused by proliferation of the tubal epithelium, which extended to the urinary bladder through the vesicovaginal fistula. This prolapse was difficult to differentiate from urinary bladder adenocarcinoma [2]. The exuberant stromal formation in tubal prolapse is a hitherto unknown additional feature, making it difficult to diagnose this lesion. One of the authors (MM) has reviewed, mostly in consultation, eleven cases of tubal prolapse in the past, and this exuberant stromal response was found in two of these eleven patients. Morphological and immunohistochemical profiles of the stroma are similar, if not identical, to that of cervical and vaginal angiomyofibroblastoma [8, 13, 16, 29]. The stromal proliferation, which is morphologically similar and focally identical to the vaginal angiomyofibroblastoma in our two cases, seems to be a reactive phenomenon to the presence of tubal glandular epithelium. Indirect support for this hypothesis is the fact that the stroma grew asso-

ciated with the tubal glands in our cases and nowhere have we observed autonomous growth of the stroma entirely devoid of this epithelium. It suggests that at least a part of the pure angiomyofibroblastomas of the vagina, and possibly vulva as well, might be of reactive rather than neoplastic nature, being caused by various mechanical or other injuries to the region.

The differential diagnosis of the fallopian tube prolapse associated with exuberant stromal formation is easy if one does not overlook the presence of glandular inclusions. The lesions we describe have tubal-type glandular epithelium. This differs from all other benign and malignant mesenchymal lesions of the vagina, such as vaginal fibroepithelial polyp [17], angiomyofibroblastoma, aggressive angiomyxoma [22, 23], superficial myofibroblastoma [14], and botryoid rhabdomyosarcoma [6]. A tuboendometrial variant of vaginal adenosis [12] might be confused with tubal prolapse, even if the former condition is associated with prenatal exposure to diethylstilbestrol and grossly appears as a flat, nonpolypoid lesion. However, no such angiomyofibroblastic stroma, as seen in our two lesions, has ever been described in the vaginal adenosis. Conclusively, it must be underlined that the most important characteristic feature of the tubal prolapse, which facilitates the right diagnosis, is the previous history of hysterectomy dating several weeks to a few years prior to the occurrence of the tumor mass of tubal prolapse.

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