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Uterus-like mass in the uterine cervix: superficial cervical endometriosis with florid smooth muscle metaplasia?

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Abstract This study describes a 47-year-old woman, gravida 2, para 2, with a uterine polypoid mass. The 25-mm mass, which was growing into the endocervical canal, had a central cavity lined with endometrium and surrounded by smooth muscle layers. It strongly resembled a miniature uterus. It is concluded that the mass represented superficial cervical endometriosis with florid smooth muscle metaplasia of endometrial stromal cells.

Keywords Uterus-like mass · Endometriosis · Smooth muscle metaplasia · Uterine cervix

Introduction

A uterus-like mass, which is characterized by a central cavity lined by endometrium surrounded by a thick wall composed of smooth muscle, is a very rare condition. Such a mass in the ovary was first described by Cozzutto [3] in 1981. The entire ovary was replaced by a mass that had a striking macroscopic and microscopic resemblance to the uterus [2,3,12,13]. Uterus-like masses arising from extraovarian sites such as the broad ligament [1], small intestine [13], and scrotum [18] have also been reported. The histogenesis of such masses has been controversial [1,3,4,11, 12, 13, 14, 15, 17, 18]. To my knowledge, there have been no reported cases of these masses arising in a uterus in the English language literature. The present study describes my observations of a uterine polypoid mass resembling a uterus in a 47-year old woman. The differential diagnosis and its histogenesis are also discussed.

Case report

Clinical findings

A 47-year-old Japanese woman, gravida 2, para 2, presented with a 2 week history of abnormal vaginal bleeding. Her past history was unremarkable except for an endometrial adenomyomatous polyp 5 years previously. The patient had no history of hormonal therapy. Physical examination revealed a polypoid mass measuring 25 mm growing into the endocervical canal. The uterus was not enlarged and there were no palpable adnexal masses. Clinically, there were no anatomic abnormalities. Laboratory data were unremarkable. Under the clinical diagnosis of myoma delivery, a polypectomy was performed. The patient was well with no evidence of disease 10 months after surgery.

Pathological findings

Macroscopically, the polypectomy specimen measured 25×18×17 mm and was a well-demarcated, rubbery mass with a smooth surface. Microscopically, horizontal sections showed a cavity measuring 10 mm surrounded by endometrial mucosa and smooth muscle layers that resembled myometrium (Fig. 1). The outer surface of the

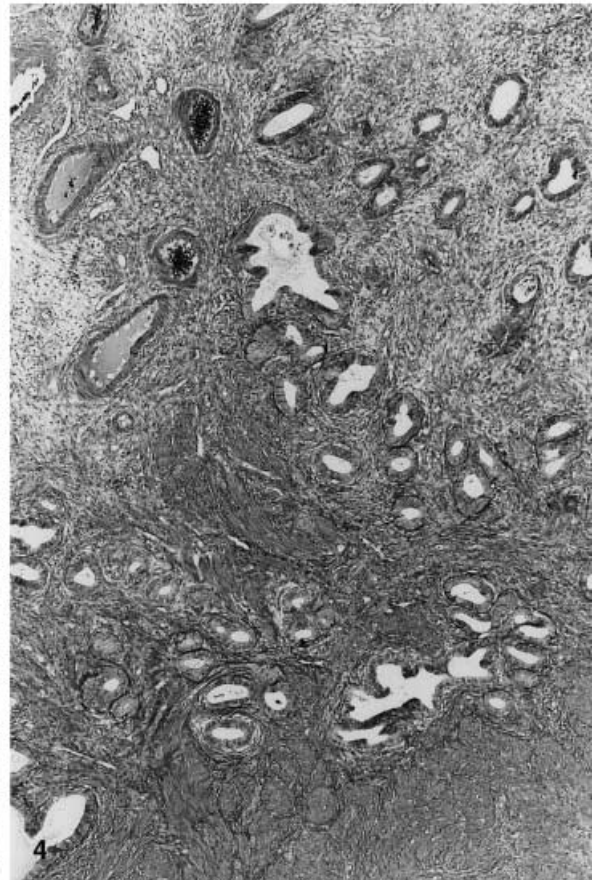
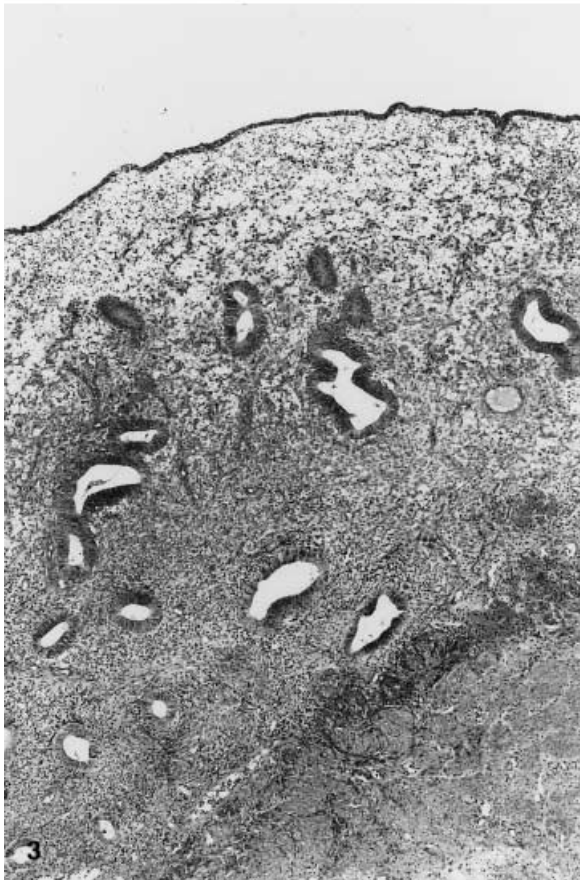
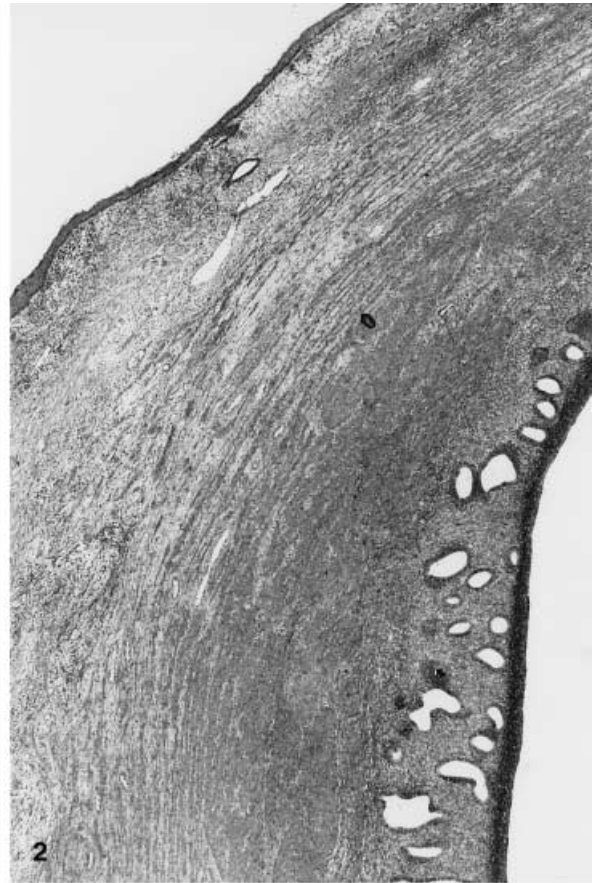
Fig. 1 Horizontal sections (*top* is an upper part of the mass and *bottom* is a lower part) show endometrial tissue, containing a cavity measuring 10 mm, surrounded by smooth muscle layers (H&E, ×3.7)

Fig. 2 The outer surface of the lower part of the mass is covered by squamous epithelium (*top*) and the inner surface is covered by endometrial epithelium and stroma (*lower right*) surrounded by smooth muscle layers (H&E, ×20)

Fig. 3 The outer surface of the upper part is covered by proliferative endometrium (H&E, ×40)

Fig. 4 Histology of the previous polypectomy specimen shows an intimate proliferation of endometrial glands and stroma and smooth muscle tissue (H&E, ×400)

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mass was covered by squamous epithelium in the lower part (Fig. 2) and by proliferative endometrial epithelium in the upper part (Fig. 3). The stroma was composed predominantly of layers of smooth muscle surrounding the inner endometrial tissues (Fig. 2). The inner endometrial tissues surrounding the cavity consisted of proliferative endometrial glands and endometrial stroma. The endometrial glands were lined by pseudostratified ciliated cells, non-ciliated cells, and intercalated cells resembling tubal epithelium. A few endometrial glands with a small number of endometrial stroma were observed in the muscle layers. Thick-walled blood vessels were present in the stroma. There were neither cellular atypia nor mitotic figures in the smooth muscle tissue.

On review of the polypectomy specimen diagnosed as adenomyomatous polyp 5 years before, the lesion was described as a 35×20×20 mm, fragile, hemorrhagic, tan mass. Histologically, it consisted of a proliferation of endometrial glands with tubular metaplasia, endometrial stroma, and smooth muscle tissue (Fig. 4). Some glands were cystically dilated. These three elements were virtually identical to those of the current polypoid lesion.

Discussion

This polypoid lesion resembling a uterus may represent superficial cervical endometriosis with florid smooth muscle and tubular metaplasia [2]. Both the current mass and the previously excised polypoid lesion could be explained by this theory. Cystically dilated endometrial glands are often present in endometriosis. Smooth muscle and tubular metaplasia in ovarian endometriosis is not an uncommon phenomenon [7,8]. A rim of smooth muscle metaplasia surrounding endometriotic cysts has been occasionally observed in ovarian endometriosis [5,7,10].

The differential diagnosis included Mullerian anomaly, mesonephric hyperplasia, adenomyoma, atypical polypoid adenomyoma, and adenosarcoma. None of the Mullerian anomalies in the American Fertility Society classification [19] could be applied to the current lesion although laparotomy and hysterectomy were needed to clarify it. The glandular proliferation in this case was different from mesonephric hyperplasia, which is characterized by ducts lined by bland cuboidal cells containing luminal periodic acid-Schiff-positive eosinophilic secretion. Mesonephric ducts are devoid of ciliated cells [6].

The designation of adenomyoma should be reserved for those polypoid lesions in which the stromal component is largely or exclusively composed of smooth muscle [9]. It is a very rare lesion and may be located within the myometrium or it may involve or originate in the endometrium and grow as a polyp. Approximately 2% of endometrial polyps are adenomyomas [20]. The uterus-like structure and the substantial element of endometrial stroma in the present lesion are unusual for adenomyoma. This polypoid lesion was different from atypical polypoid adenomyoma because it lacked glandular hyper-

plasia with atypia and loss of polarity, squamous differentiation and short interlacing fascicles of smooth muscle that are characteristic to the latter. Since this lesion lacked periglandular stromal condensation, stromal atypia and active mitotic activity of stroma cells, adenosarcoma was also ruled out.

The histogenesis of uterus-like masses has been extensively controversial. There are two basic theories of the cause: (1) metaplastic changes [3,11,12,15], (2) congenital anomalies [13,14,17]. With regard to the present lesion, the metaplastic theory, possibly originating from superficial cervical endometriosis with smooth muscle metaplasia of the endometriotic stroma, is more compelling than the congenital anomaly hypothesis. The previously excised polypoid lesion would not contradict this theory. It is hard to completely exclude the possibility of a uterine anomaly unless the uterus is removed, but clinical urogenital and gynecological examinations failed to find any anatomic anomalies.

In response to an earlier case described by Cozzutto [3], who proposed an origin from smooth muscle metaplasia in a focus of ovarian endometriosis, Rosai [17] mentioned that the uterus-like mass represented a congenital anomaly and was an example of Mullerian duct fusion defect or, less likely of a true partial duplication of the Mullerian system. The mass is often associated with a deformed urinary system [3,12, 13, 14]. However, Rahilly and Al-Nafussi [15] and Noel et al. [11] reported such masses in the ovary without any congenital anomalies of the uterus or the renal system and they supported the metaplastic theory.

Endometriosis has been described in cases of leiomyomatosis peritonealis disseminata, in which some of the nodules have resembled microuteri [16]. Scully [18] reported the occurrence of a uterus-like mass in the scrotum of men who received estrogenic therapy for carcinoma of the prostate. What specific event led to the metaplasia is not clear. Peterson et al. [13] and Pai et al. [12] suggested a vital initiating role of estrogen in the pathogenesis of a uterus-like mass.

It remains possible that both mechanisms, metaplastic changes in endometriosis and congenital anomaly, such as Mullerian duct fusion defect, may give rise to morphologically similar lesions.

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