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Case Report

Pseudomyxoma peritonei with endometrial mucinous carcinoma and appendicular mucinous tumor: An unusual association

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ABSTRACT

The association between pseudomyxoma peritonei and appendicular or ovarian mucinous tumors is usually reported in the literature, while the association with endometrial carcinoma is exceptional. Although there has been always a continuous debate regarding its primary origin, tumors of the appendix and ovary remain the most common primary sites for this disease. The association of pseudomyxoma peritonei with two primaries from endometrial mucinous adenocarcinoma and appendicular mucinous tumor is very rare. So, we report this case to raise awareness among clinicians about this rare tumor association.

1. Introduction

Pseudomyxoma peritonei (PMP) is an uncommon disease entity accounting for one to two per million per year [1]. It affects females three to four times more than males [2].

PMP is characterized by diffusely spread collection of gelatinous material into the intra-abdominal cavity along with scattered mucinous implants over peritoneal surfaces and omentum with variable cellularity [3]. Ronnett et al. [4] classified PMP into low-grade disseminated peritoneal adenomucinosis associating, high-grade peritoneal mucinous carcinomatosis, and peritoneal mucinous carcinomatosis with intermediate or discordant features. The origin of PMP is still controversial; mucinous tumors of the appendix, ovary, and gastrointestinal tract are the most common association [4–6].

PMP is not commonly encountered in our clinical practice, and to our knowledge, primary appendiceal mucinous tumors associated with primary endometrial mucinous carcinoma have been reported rarely in the medical literature. We are reporting a case of a female patient presented with PMP associated with mucinous borderline tumor of the appendix and an infiltrating mucinous endometrial carcinoma, to raise awareness among clinicians about this rare tumor association.

2. Case report

A 56-year-old postmenopausal woman presented with a history of recent uterine bleeding. Her medical history was significant for morbid obesity and hypertension. On admission, her vital signs were stable. A physical examination revealed fullness in the lower abdomen. The computed tomography of the abdomen revealed a mildly

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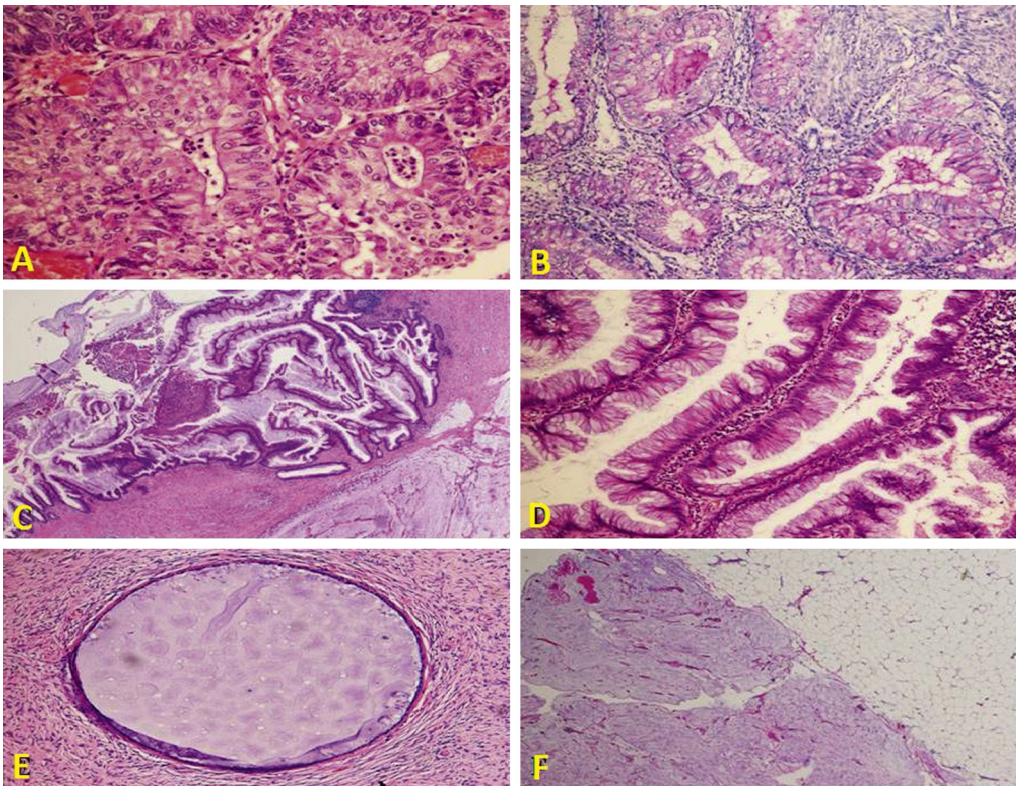

Fig. 1. (A) Endometrial mucinous carcinoma showing complex glandular structures with back-to-back arrangement [hematoxylin and eosin (H&E), 200×]; (B) endometrial mucinous carcinoma showing positive staining to periodic acid–Schiff before and after digestion (periodic acid–Schiff–diastase, 200×); (C) appendicular tumor composed of confluent papillary structures displaying a circumferential growth pattern all over the appendiceal lining mucosa (H&E, 100×); (D) appendicular tumor, high-power view showing bland-looking tumor cells (H&E, 200×); (E) small benign-looking mucin-filled cyst within the ovarian stroma (H&E, 200×); and (F) omentum revealed noninvasive acellular mucin pools bisecting fat lobules (H&E, 200×).

enlarged uterus; otherwise, there were no specific changes. No ascites or pelvic lymphadenopathy was noticed. An endometrial biopsy was obtained that was diagnosed as complex endometrial hyperplasia with atypia highly suspicious of malignancy.

An explorative laparotomy was performed; the uterine cervix was found to be distended with a small tumor. An appendicular mass was identified, measured 4 cm in length. Lobular infiltrate on the omental surface was identified, as well as mild peritoneal collection. An aggressive surgical debulking of the appendix and radical hysterectomy with bilateral salpingo-oophorectomy were performed. Due to suspected malignancy, surgical staging was completed by omentectomy, while no enlarged lymph nodes were detected. Peritoneal fluid was collected.

The specimen received in the pathology laboratory consisted of transabdominal hysterectomy with bilateral salpingo-oophorectomy, omentectomy, and appendectomy. The uterus with attached cervix weighed 250 g and measured 13 cm × 6 cm × 5 cm. The serosal surfaces are unremarkable. The endometrial cavity measured 6 cm × 3 cm × 2 cm, and contained a friable, papillary, fungating, tan-colored tumor mass. The tumor was originating from the fundus. The tumor measured 4 cm × 2.5 cm × 0.5 cm,and invaded the inner half of the myometrium (1.5/3.5 cm), reaching 0.5 cm close to the superior serosal surface. The right and left ovaries, and the attached Fallopian tubes showed unremarkable outer and cut surfaces, except fora 1-cm cyst in the left ovary. A piece of thickened omentum with mucoid outer surface was sent (weighing 600 g and measuring 25 cm × 18 cm × 2 cm). The appendix and attached mesoappendix measured 4 cm × 3.5 cm × 2.5 cm. A mucoid irregular mass measured 4 cm × 3 cm × 1 cm, and was attached to the serosal surface of the appendicular tip. The appendix lumen is filled with mucoid materials, and a perforation area is present near the appendiceal tip measuring 0.5 cm.

A microscopic examination of the uterine specimen revealed a malignant tumor composed of a complex glandular structure showing focal cystically dilated spaces with branching intraluminal papillae. The tumor cells are mucin-secreting tall columnar cells having round nuclei showing a mild degree of atypia. The glandular lumina are loaded with mucin and heavy neutrophilic infiltrate (Fig. 1A). The intracytoplasmic and intraluminal secretions were proved by periodic acid–Schiff and periodic acid–Schiff–diastase stains (Fig. 1B). The tumor was superficially invading the myometrium and lower uterine segment. There was no lymph–vascular space involvement. The lower uterine segment, cervix, and omentum were free