Case Report

Secondary Vaginal Involvement Following Radical Surgical Treatment for a Stage I Ovarian Adenocarcinoma Arising in Mature Cystic Teratoma

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Background: *Vaginal carcinoma represents 1-2% of all gynecologic malignancies. Most cases reported secondary involvement from adjacent organs including cervix, uterus, and colorectum. Vaginal involvement from adenocarcinoma arising in mature cystic teratoma (MCT) has never been reported.*
Case: *A 29-year-old female presented with postcoital vaginal bleeding. She had had a history of right ovarian adenocarcinoma arising in MCT, FIGO stage IC, for 18 months' duration. Incisional biopsy of the vaginal lesion revealed adenocarcinoma, morphologically and immunohistologically identical to the right oophorectomized specimen. She received three courses of paclitaxel and carboplatin chemotherapy; however, she developed massive right pleural effusion with superior vena cava syndrome and finally succumbed to the disease, three months later.*
Conclusion: *Adenocarcinoma is rarely found in MCT. This is the first case of ovarian adenocarcinoma arising in MCT with secondary vaginal involvement, presenting as postcoital vaginal bleeding.*

Keywords: *Adenocarcinoma, Malignant transformation, Mature cystic teratoma, Vaginal involvement, Ovary*

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Vaginal carcinoma represents 1-2% of all gynecologic malignancies and only 16% of carcinoma involving the vagina are primary[1]. Therefore, extravaginal origin needs to be considered when evaluating vaginal carcinomas. The most frequent primary site is the uterus (32%), followed by colorectum (26%) and ovary (17%)[1]. Vaginal involvement from carcinomatous change arising in ovarian mature cystic teratoma (MCT) is relatively uncommon. Its diagnosis often is incidental and associated with advanced stage of cancer. It can be mistaken clinically and pathologically for primary vaginal tumor. It is important to distinguish from the primary vaginal neoplasm because the treatment is quite different. The secondary vaginal involvement from the other neoplasms almost always tends to yield a dismal outcome. Identification and confirmation of the primary tumor is important to facilitate oncological treatment. In the present study, the authors describe an early stage adenocarcinoma arising in MCT in a young woman who subsequently developed vaginal involvement.

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Case Report

A 29-year-old Thai thalassemic woman, gravida 1, para 1, presented with a three day history of right lower abdominal pain. Physical examination revealed a large cystic tumor occupying the lower abdomen. Upon exploration, a 13-centimeter right ovarian cyst was found and removed intact as the right salpingo-oophorectomy. The pathology turned out to be MCT with adenocarcinomatous change involving the ovarian surface. Optimal complete surgical staging

was achieved about six months later with total hysterectomy, left salpingo-oophorectomy, pelvic node dissection, and omentectomy. FIGO stage IC was concluded. She received no adjuvant treatment.

Eighteen months later, she developed postcoital vaginal bleeding. Vaginal examination revealed a contact-bleeding lesion in the upper vagina. Pelvic examination revealed nodular, solid tumor, measuring 8 x 9 cm in size, occupying the pelvis extending to the left pelvic wall. Incisional biopsy of the vaginal lesion was performed and showed adenocarcinoma. Morphologically and immunohistochemically, these were almost identical to the tumor seen in the previous right oophorectomized specimen. One week later, she developed intestinal obstruction. Exploratory laparotomy demonstrated a frozen pelvis with the tumor encasing the rectum and carcinomatosis peritonei with minimal ascites. The tumor could not be debulked thus, transverse loop colostomy was performed.

She refused a combination of concomitant chemoradiation as adjunctive treatment. Therefore, only chemotherapy was given as paclitaxel (175 mg/m^2) and carboplatin (5 AUC) on the seventh day after surgery. Massive right pleural effusion developed afterward and the pleural cytology was positive for malignancy. After the third course of chemotherapy, a metastatic tumor was discovered at the right axillary area measuring 2 x 2 cm. She developed superior vena cava syndrome. Computed topography of the chest revealed multiple paratracheal lymph nodes, the largest one measured 4.2 x 2.9 x 2.5 cm encasing the superior vena cava and causing obstruction. After 900 cGy of palliative radiation to the chest, the patient refused further treatment. Finally, she succumbed to the disease, three months after the diagnosis of recurrence.

Pathologic findings

The first operative specimen consisted of the right ovary measuring 13 x 11 x 8 cm in size and weighing 420 grams. Cut surfaces of the ovary revealed a multilocular cyst containing yellow turbid sebaceous material with numerous matted lanugo. The cystic wall was hard and measured 1 to 6 mm in thickness. The sections of the right ovary revealed cystic spaces lining by columnar epithelium and skin tissue equipped with hair follicles, sebaceous glands, and sweat glands. The subepithelial areas of the cyst showed nonciliated atypical dysplastic columnar epithelial cells proliferated with a tubular configuration, which was regarded as well-differentiated adenocarcinoma (Fig. 1). The tumor involved the ovarian surface. Immunohistochemically,

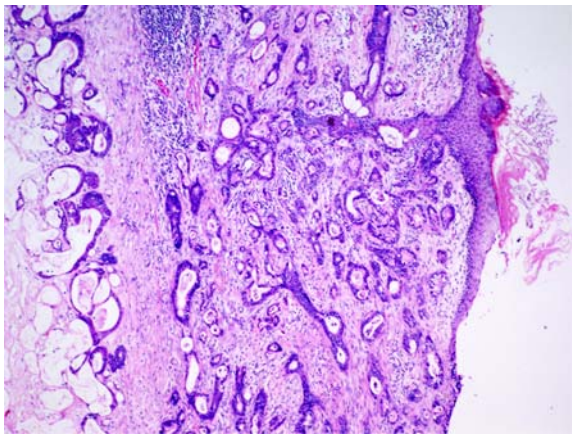

Fig. 1 Section of the right ovarian tumor shows the junction between the well-differentiated adenocarcinoma and the mature squamous epithelium, the subepithelial ovarian stromal area shows nonciliated atypical columnar epithelial cells proliferated with a tubular configuration; H&E, X40

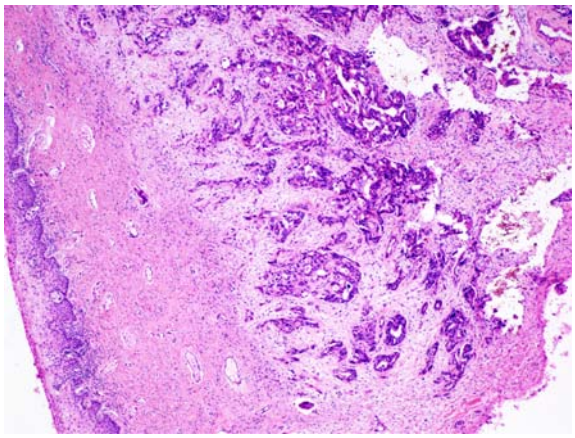

Fig. 2 Section of the vaginal tumor shows well-differentiated adenocarcinoma in the subepithelial vaginal connective tissue and lymphovascular channels; H&E, X40

the adenocarcinomatous cells were positive for cytokeratin 7 as well as 20 and negative for caudal homeobox 2 (CDX2). There were no immature teratomatous elements or other germ cell components, including embryonal carcinoma, yolk sac tumor, choriocarcinoma, or dysgerminoma. The pathological diagnosis of the right ovarian tumor was adenocarcinoma arising in MCT.

The following incisional biopsy of the vaginal mass revealed well-differentiated adenocarcinoma in the subepithelial vaginal connective tissue and

lymphovascular channels (Fig. 2). Immunohistochemically, the adenocarcinomatous cells were positive for cytokeratin 7 as well as 20 and negative for CDX2. Morphologically and immunohistochemically, these were almost identical to the tumor seen in the right ovary.

Discussion

MCT of the ovary is one of the common tumors in women during reproductive life. MCT composes of a variety of tissues usually representing three embryonic layers[2,3]. The totipotent MCT may give rise to any type of malignant transformation that is relatively uncommon and occurs in 2% of cases[3]. Squamous cell carcinoma is the most common type of malignancy found, accounting for 75-85% of all malignancies arising in MCT. Adenocarcinoma arising from MCT is extremely rare and occurs in 6.8% of the malignant cases[4].

Clinically, malignant transformation in a component of a MCT cannot be readily differentiated from an uncomplicated MCT or other ovarian tumor, although evidence of its rapid growth, pain, weight loss, and other systemic symptoms suggest the presence of a malignant tumor. The routine initial laboratory investigations are noncontributory. Diagnosis is often delayed because of protean nonspecific and diverse clinical manifestations. The imaging procedures such as Doppler ultrasonography, and magnetic resonance image (MRI) may allow early recognition of malignant transformation in MCTs. Doppler ultrasound shows malignant transformation in MCT having a higher intratumoral blood flow and lower mean resistance index and pulsatility index than the benign MCT[5]. MRI facilitates the diagnosis of MCT due to characteristic signal strength, and enhanced images of solid parts in tumors detected possible malignancy[6].

The malignant transformational part in MCT usually spreads by the direct invasion, followed by peritoneal implantation and metastasis. The previously reported secondary sites include the opposite ovary, uterus, liver, ileum, colon, urinary bladder, and lymph nodes[7, 8]. Vaginal involvement from adenocarcinoma arising in MCT has never been reported. The definite diagnosis is primarily based on the combination of the histopathology, immunohistochemistry, and clinical feature. Identification and confirmation of the primary tumor is important to facilitate oncologic treatment.

The stage of the disease is the most important prognostic factor. Five-year survival rates for patients with malignant transformation in MCT have been 77% for stage I, and 11% for stage II to IV[9]. Poor prognostic factors include cyst wall invasion, rupture, tumor dissemination, ascites, adhesions, and tumor type other than squamous cell carcinoma. Because of its rarity, the surgical and postoperative management of this uncommon germ cell malignancy are not established in the literature. Aggressive surgical staging is advocated in all cases of malignant transformation in MCT because of the high incidence of spread. Various treatments for stage I tumors have consisted of chemotherapy, radiotherapy or the combination. Recently, many authors have documented the use of a combination of chemotherapy and/or radiotherapy[10]. Therefore, the optimal treatment for a malignant transformation in MCT should be individualized based upon accurate stage and experience of the care providers.

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เนื้อร้ายชนิดอะดีโนคาร์ซิโนมาที่เกิดในเทอราโทมาชนิดสมบูรณ์ของรังไข่แล้ว ลุกลามไปยังช่องคลอดภายหลังการผ่าตัดรักษา

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มะเร็งช่องคลอด พบร้อยละ 1-2 ของเนื้อร้ายทางระบบนรีเวช โดยส่วนใหญ่เกิดจากการลุกลามของเนื้อร้ายอวัยวะข้างเคียง เช่น ปากมดลูก, มดลูก และลำไส้ใหญ่ เนื้อร้ายชนิดอะดีโนคาร์ซิโนมาที่เกิดในเทอราโทมาชนิดสมบูรณ์แล้วลุกลามไปยังช่องคลอดยังไม่พบการรายงาน

คณะผู้รายงานเสนอผู้ป่วยหญิงไทยอายุ 29 ปี มาพบแพทย์ด้วยอาการมีเลือดออกทางช่องคลอดหลังร่วมประเวณี โดยเมื่อ 18 เดือนก่อน ผู้ป่วยได้รับการวินิจฉัยว่าเป็นเนื้องอกชนิดอะดีโนคาร์ซิโนมาที่เกิดในเทอราโทมาชนิดสมบูรณ์ของรังไข่ข้างขวา ระยะ FIGO IC ในครั้งนี้ได้รับการตรวจชิ้นเนื้อที่ช่องคลอด พบว่าเป็นเนื้อร้ายชนิดเดิม ผู้ป่วยได้รับยาเคมีบำบัด 3 ครั้ง ต่อมาผู้ป่วยมีน้ำในช่องปอดขวา เกิดอาการของการอุดกั้นหลอดเลือดดำใหญ่ส่วนบน และหลังจากนั้น 3 เดือน ผู้ป่วยได้เสียชีวิต