A Rare Case of Invasive Mature Teratoma of the Uterus

Mehrangiz Zamani*, Alireza Rastgoo and Maryam Jamali

Department of Oncogynecology, Hamadan University of Medical Sciences-UMSHA, Asadabad School of Medical Sciences, Iran

Abstract

Teratoma is a germ cell tumor usually occurs in young people, it is derived from germinal layers and main sites of this tumor are ovary. Few cases have been reported with teratoma of uterus in English literature till now. We want to describe a mature teratoma of uterus with aggressive behavior in 24-year-old women.

Keywords: Invasive mature teratoma; Uterus; Germ cell tumor

Introduction

Teratoma is a germ cell tumor, derived from two or more germinal layers, like, ectoderm, mesoderm and endoderm [1]. Teratomas may be classified as mature or immature base on the presence of immature / embryonic elements [2-4]. Malignant teratomas demonstrate aggressive growth of one or more of the histological components [2]. Teratomas usually arise in the gonads, extragonadal type of teratomas are rare. They mainly develop in midline structures such as the retroperitoneum, mediastinum, sacrococcegeal area, pineal gland and the head and neck region [2-4]. Teratomas presentation peak age is between 20 and 40 years. Mann for the first time described a case of primary mature uterus teratoma [3]. To the best of our knowledge, since then only 24 cases of mature and immature teratomas of the uterus and cervix have been reported. Here we want to report a mature teratoma of uterus with aggressive behavior in a 24-year-old woman which is very rare.

Case Report

A 24-year-old woman presented with vaginal bleeding and abdominal pain. Her menarche was 12 years of age and the menstrual periods were regular with a cycle of 26 days. The patient also had no history of sexual activity and was fit, well, with no past medical history.

On physical examination, she had a bulky uterus with no adnexal mass. Trans abdominal ultrasonography demonstrated a solid heterogenous echopattern mass in uterus. The size of mass in sonography was about 100 × 70 mm in uterus with some cystic endometrial sites and 6 mm endometrial thickness. She underwent a laparotomy when en bloc endometrial mass with musinous tenacious materials, which filled the uterus cavity, was performed. Some hours after surgery, the patient had the signs of lung micro-embolization with pulse rate: 190/min, and respiratory rate: 60/min. There were findings of emboli in radiologic management. Therefore, we prescribed Heparin and until 12 days later she hospitalized.

She discharged with Rivaroxaban, 10 mg twice a day. Microscopically, the tumor was a mature teratoma. Three months later, the patient presented sever vaginal bleeding. On examination, there was a friable polypoid mass protruded from the cervical canal in to the vagina perforated the hymen. The infected mass filled her vagina with sever bleeding. patient was counseled emergency laparotomy. Intra operatively, the uterus was found to be bulky with discharging sinus on the fundal site. Both ovaries were healthy – shape and there was no free fluid in the pouch of Douglas. The tumor perforated the posterior site of uterus and invaded to recrosigmoid and bladder. There was no free margin of tumor to en bloc and adhesion bands existed between uterus and marginal organs.

The decision to perform a total hysterectomy was reached. After surgery, we transfused one unit of blood and intravenous antibiotics,

ceftriaxon and metronidazol, were given with an addition of heparin. The patient had a recovery. The samples were sent for histological examination. At the pathology department, diagnosis of necrotized mature teratoma was made (Figures 1-4).

Figure 1: 1) Mature teratoma perforated the fundal of uterus, 2) Active mature teratoma with aggressive manner.

***Corresponding author:** Mehrangiz Zamani, Department of Oncogynecology, Hamadan University of Medical Sciences-UMSHA, Asadabad School of Medical Sciences, Iran, Tel: +98-918-811-2098; E-mail: maryamdr_59@yahoo.com

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Figure 2: 1). Fundal of uterus, 2) Teratoma of uterus.

Figure 3: 1) Mature teratoma.

Figure 4: The pathologic findings.

The patient received 3 courses of combination therapy with BEP. The treatment strategy included three courses of chemotherapy (BEP regimen) based on Bleomycin (30 mg/day, days 1-8 and 15) Etoposide (100 mg/m²/day, days 1-5) and Cisplatin (20 mg/m²/day, days 1-5) every 3 weeks. She had no sign of disease recurrence for 6 months following hospital discharge.

Discussion

A primary invasive mature teratoma was described in 24-year-old woman. this tumor is completely foreign to this site. In pathologic study, we found significant amount of mature teratoma elements. The origin of uterus teratoma is not exactly clear. We discussed an unusual case because only few cases of uterus teratoma were reported, and infected teratoma is also rare event [5]. Because of the extremely rapid growth of tumor in our case, it perforated the hymen and protruded in vagina. This malodor polypoid mass is the piece of a huge mass in uterine that perforated the fondal with invasion to rectosigmoid and bladder.

First report of teratoma was described by Mann in 1929. The majority of case reports are mature and to date only 7 immature teratomas were reported [4,6,7] (Table 1). The prevalence of extragonadal teratoma is about 1-2% of all teratomas [8]. Whenever a uterus teratoma presents after pathologic study, it is necessary to exclude spreading from another current site of this tumor frequently ovaries, based on the case was reported by Galko et al. [9]. Most of the cases were in reproductive age although two cases were post-menopausal women [10]. The age range is 15 to 82. There are no specific symptoms, patients presented vaginal bleeding, pelvic pain, lower abdominal distention, urinary symptoms and in two cases were complicated with uterus inversion [6,11]. In all cases, diagnosis was failed based on the radiological studies. The cause of this failure is related to the lack of characteristic features, therefore, most of them were described as polyp, lipoleiomyoma or mixed mullerian tumor (MMT) [5]. The accurate diagnosis in all cases was made after pathologic survey of surgical specimen [7,12].

Table 1: Clinical characteristics and treatment of patients with teratoma of uterus.

Treatment of relapse	Relapse (months)	Treatment	Histology	Site of tumor	Symptoms	Age	Author
BEP 1 ×	Yes (3)	Hysterectomy	Imature Teratoma	Corpus	Urinary symptoms Lower abdominal distention	65	Ben Ameer El-Youbi et al., [5]
EP × 1, ET × 1, TP × 1 + surgery	Yes (6)	Hysterectomy and bilateral Salpingoophor ectomy	Imature Teratoma	Corpus	Vaginal bleeding	28	Newsom-Davis et al., [11]
—	ns	Lesion excision + chemotherapy	Imature Teratoma	ns	Vaginal bleeding Pelvic pain	16	Gomez-Lobo et al., [7]
—	No	Hysterectomy + chemotherapy (VAC ×)8	Imature Teratoma	Fundus	Pelvic pain Lower abdominal distention	65	Iwanaga et al., [4]
—	No	Hysterectomy + radiotherapy	Imature Teratoma	ns	Vaginal bleeding	67	Ansah- Boateng et al., [5]
-	no (12)	Hysterectomy + chemotherapy	mature teratoma with aggressive manner	Fundus	Vaginal bleeding	82	Our study

Conclusion

Our patient presented vaginal bleeding and in radiological study, there was no significant findings to make decisions. In addition, our case was mature teratoma with aggressive behavior and a relapse after primary surgery. There is no standard guideline to treat extragonadal teratoma. Our knowledge about the management of this tumor depends on few reports were published. The best approach is complete tumor resection by total abdominal hysterectomy or conization in cervical mass [5,8,10]. The role of chemotherapy is not clear. In some cases, the courses of chemotherapy with BEP regimen were recommended. Ansah-Boateng et al. reported a patient with uterus teratoma who managed by pelvic radiotherapy after hysterectomy [5,6]. Chemotherapy after surgical treatment was prescribed by Newsom-Davis et al. and Ben Ameer et al. [5,10].

The only similar malignant mature teratoma was reported in an 82-year-old woman who underwent surgery and chemotherapy and finally died after last surgery was performed for retroperitoneal lymph node dissection [10]. Although, there is lack of experience about the management of this tumor, this appears to be reasonable to have a close contact clinical surveillance, regular cross-sectional imaging and monitoring serum tumor markers. Because of our restricted knowledge the prognosis of the extragonadal teratoma is uncertain. In general, uterus teratoma is extremely rare, we should consider this tumor as a reason of uterine mass even in the absence of specific radiological findings, because it would be possible to be surprised doing the surgery or after pathological results.

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