Case Report

A rare case of postmenopausal immature teratoma with a recurrent grave course

Ali Babacan¹, Yaşam Kemal Akpak², Cem Kizilaslan³, Ismet Gun¹, Ercument Mungen¹, Vedat Atay¹

¹Department of Obstetrics and Gynecology, GATA Haydarpasa Training Hospital, Istanbul, Turkey; ²Department of Obstetrics and Gynecology, Ankara Mevki Military Hospital, Ankara, Turkey; ³Department of Obstetrics and Gynecology, Beytepe Military Hospital, Ankara, Turkey

Received March 19, 2014; Accepted April 11, 2014; Epub April 15, 2014; Published April 30, 2014

Abstract: Immature teratoma is quite uncommon older women, particular in postmenopausal period. Only a few cases of postmenopausal immature teratoma of the genital tract have been reported. This report describes a postmenopausal aggressive pelvic immature teratoma case with a grave course despite treatment. A 67-year-old woman being in menopause for the past 16 years was diagnosed with immature teratoma in the pelvic cavity. The mass was removed completely and hysterectomy plus bilateral salpingo-oophorectomy was done. Histopathological diagnosis was grade 2 immature teratoma. The patient had a grave course with recurrences despite therapy and died within one year after initial diagnosis. In teratomas diagnosed over 45 years of age, the possibility of malignant teratoma should also be borne in mind.

Keywords: Immature teratoma, menopause, recurrence

Introduction

Less than 1% of all ovarian cancers are germ cell tumors and immature teratoma of the ovary is the third most common germ cell tumor after dysgerminoma and endodermal sinus tumor [1]. Immature teratoma is mostly seen in women younger than 30 years of age. It is quite uncommon in older ages, particular in postmenopausal women. Only a few cases of postmenopausal immature teratoma of the genital tract have been reported so far [2, 3].

This case report describes a postmenopausal aggressive pelvic immature teratoma case with a recurrent and fatal course despite surgery and chemotherapy.

Case report

A 67-year-old woman (gravida 13, para 2) being in menopause for the past 16 years, presented with abdominal pain and distention for the last two weeks. Medical and family history was unremarkable except for aspirin use since a deep vein thrombosis attack 11 years ago. An abdominal mass was palpable on physical exa-

mination. Abdominal ultrasonography revealed an 18 x 16 x 8 cm solid mass with irregular contours filling right adnexal region of the right lower abdominal quadrant. The lesion had heterogeneous hypoechoic cystic regions. Computerized tomography showed an 18 x 15 x 12 cm lobulated mass with distinct borders at the same localization. The lesion had septations and solid regions. Since the lesion was in continuity with the right ovary, a malign neoplasm was suspected. Abdominal exploration revealed a retroperitoneal mass with a diameter of approximately 20 cm, originating from paratubal area, extending to pelvic floor and completely filling pelvic cavity The lesion was adjacent to the right ovary, but without continuity. It consisted of a 6 x 5 cm solid region at its lower part and had adhesions to surrounding organs. Both ovaries and tubes were normal. The histological examination findings of frozen section material were suggestive of a chondroid tumor, possibly chondrosarcoma. The mass was removed completely and hysterectomy plus bilateral salpingo-oophorectomy was done. Postoperative histopathology report revealed a grade 2 immature teratoma. Chemotherapy was offered to the

patient but she refused. Follow-up abdominal MRI examination after 3 months showed an 8-cm mass. The recurrent lesion located at close proximity with the sigmoid colon was removed completely and omentectomy was done. However, the mass was necrotic, irregular and with muddy consistency. Histological diagnosis was the same. The patient received two courses of BEP (bleomycin, cisplatin, etoposide) chemotherapy. Follow-up MRI examination after 5 months revealed two masses, each with 3 to 4-cm size. These masses were surgically removed and histopathological diagnosis was again recurrent immature teratoma. Two courses of EP (etoposide, cisplatin) protocol was planned but during the first course the patient developed pancytopenia and associated severe adverse effects, thus chemotherapy was terminated and abdominal radiotherapy was administered. The patient re-admitted 2 months after radiotherapy with massive ascites, abdominal mass and poor general condition, and died at the seventh day of hospitalization. Tumor markers had been ordered at each visit and each time they were unremarkable.

Discussion

In contrast with the malignant tumors of the ovary mostly occurring in the 5th and 6th decades of life, ovarian immature teratomas primarily affect young women of reproductive age [4]. In a study involving a total of 244 patients with immature teratomas, only 4 (1.6%) were over 40 years of age [5], while reports of postmenopausal cases are even more scarce [2, 3]. In addition, two large series reported upper age ranges in sixties but without mentioning menopausal status [6, 7]. Due to the scarcity of elderly with immature teratomas in large patient series, the association between age and survival has not been clearly established. However, grade and stage correlate with the survival in immature teratomas [8]. The case presented herein has been postmenopausal for 16 years with only recent emergence of the symptoms. The period between menopause and disease manifestation was rather prolonged in our subject.

Although most patients describe some nonspecific symptoms, the usual presenting complaint is abdominopelvic pain [8]. In a study examining mature cystic teratomas, those lesions with a diameter of at least 10 cm were more likely be malignant [9]. Similarly, imaging studies performed in our patient showed the presence of a mass, the dimensions of which were above this limit. Of all immature teratomas, only one third exhibit elevated AFP [10]. Our patient also had normal tumor markers preoperatively.

Management of immature teratomas involves laparotomic surgery and chemotherapy. Although fertility-sparing surgery is planned for younger patients, a more extensive surgery is considered appropriate in the absence of pregnancy expectation. In this type of cancer surgery, the resection of the suspicious mass lesion should be extensive enough to reduce the size of the lesion below 2 cm [11, 12]. The role of adjuvant chemotherapy remains controversial [7] and it is recommended in the presence of a suspicion for remaining malignant tissue [13]. BEP (Bleomycin/Etoposide/Cisplatin) is the most frequently used regimen, offering the significant advantage of not interfering hormone levels. In our patient, despite surgical treatment combined with chemotherapy, the grave prognosis could not be prevented. Recurrences despite optimal surgery are associated with the high proliferation rate of immature teratoma cells. In stage 1 germ cell tumors the 6 year survival rates may reach up to 93% [14], while the reported recurrence and mortality rates in stage 2 and 3 disease are 67% and 33%, respectively [15]. Unfortunately, our patient had rapid recurrences in spite of optimal surgery and combined chemotherapy, leading to death within the first year following initial diagnosis. The unfavorable prognosis in our patient can be primarily explained by advanced age, similar to previously reported rare cases of immature teratomas occurring in the postmenopausal elderly women [2, 3].

Postmenopausal immature teratoma is a rare tumor with an unfavorable prognosis. Despite this, in teratomas diagnosed over 45 years of age, the possibility of malignant teratoma should also be borne in mind.

Disclosure of conflict of interest

None.

Address correspondence to: Ali Babacan, GATA Haydarpasa Egitim Hastanesi, Kadin Hastaliklari ve Dogum Servisi, Uskudar 34668 Istanbul, Turkey. Tel:

Postmenopausal immature teratoma

+90 532 3458254; Fax: +90 216 3487880; E-mail: ababacan_@hotmail.com

References

- [1] Quirk JT and Natarajan N. Ovarian cancer incidence in the United States, 1992-1999. Gynecol Oncol 2005; 97: 519-523.
- [2] Ornvold K, Detlefsen GU, Horn T and Rorth M. Immature ovarian teratoma in a postmenopausal woman. Acta Obstet Gynecol Scand 1987; 66: 473-476.
- [3] Doss BJ, Jacques SM, Qureshi F, Chang CH, Christensen CW, Morris RT and Lawrence WD. Immature teratomas of the genital tract in older women. Gynecol Oncol 1999; 73: 433-438.
- [4] Zanetta G, Bonazzi C, Cantu M, Binidagger S, Locatelli A, Bratina G and Mangioni C. Survival and reproductive function after treatment of malignant germ cell ovarian tumors. J Clin Oncol 2001; 19: 1015-1020.
- [5] O'Connor DM and Norris HJ. The influence of grade on the outcome of stage I ovarian immature (malignant) teratomas and the reproducibility of grading. Int J Gynecol Pathol 1994; 13: 283-289.
- [6] Lee KH, Lee IH, Kim BG, Nam JH, Kim WK, Kang SB, Ryu SY, Cho CH, Choi HS and Kim KT. Clinicopathologic characteristics of malignant germ cell tumors in the ovaries of Korean women: a Korean Gynecologic Oncology Group Study. Int J Gynecol Cancer 2009; 19: 84-87.
- [7] Vicus D, Beiner ME, Clarke B, Klachook S, Le LW, Laframboise S and Mackay H. Ovarian immature teratoma: treatment and outcome in a single institutional cohort. Gynecol Oncol 2011; 123: 50-53.

- [8] Norris HJ, Zirkin HJ and Benson WL. Immature (malignant) teratoma of the ovary: a clinical and pathologic study of 58 cases. Cancer 1976; 37: 2359-2372.
- [9] Yamanaka Y, Tateiwa Y, Miyamoto H, Umemoto Y, Takeuchi Y, Katayama K and Hashimoto K. Preoperative diagnosis of malignant transformation in mature cystic teratoma of the ovary. Eur J Gynaecol Oncol 2005; 26: 391-392.
- [10] Pectasides D, Pectasides E and Kassanos D. Germ cell tumors of the ovary. Cancer Treat Rev 2008; 34: 427-441.
- [11] Munkarah A, Gershenson DM, Levenback C, Silva EG, Messing MJ, Morris M and Burke TW. Salvage surgery for chemorefractory ovarian germ cell tumors. Gynecol Oncol 1994; 55: 217-223.
- [12] Wu X, Han LY, Xu X and Li Z. Recurrent immature teratoma of the ovary: a case report of radical secondary cytoreduction with replacement of the aortic bifurcation. Gynecol Oncol 2004; 95: 746-749.
- [13] Tanaka T, Toujima S, Utsunomiya T, Yukawa K and Umesaki N. Experimental characterization of recurrent ovarian immature teratoma cells after optimal surgery. Oncol Rep 2008; 20: 13-23.
- [14] Patterson DM, Murugaesu N, Holden L, Seckl MJ and Rustin GJ. A review of the close surveillance policy for stage I female germ cell tumors of the ovary and other sites. Int J Gynecol Cancer 2008; 18: 43-50.
- [15] Patterson DM and Rustin GJ. Controversies in the management of germ cell tumours of the ovary. Curr Opin Oncol 2006; 18: 500-506.