Immature uterine teratoma associated with uterine inversion

Karla Teixeira Souza,1 Marcelo Vailati Negrão,1 Lucila Soares da Silva Rocha,1 Giovanni Di Favero,2 Samantha Cabral Severino da Costa1 Maria Del Pilar Estevze Díz1

1Department of Radiology and Oncology, Instituto do Câncer do Estado de São Paulo, Faculdade de Medicina da Universidade de São Paulo, Avenida Doutor Arnaldo, 251, Cerqueira César, São Paulo SP, 01246-900, Brazil. E-mail: maria.pilardiz@gmail.com

Key words: immature teratoma, uterine teratoma, uterine inversion.

Abstract

Teratomas are the most commonly diagnosed germ cell tumors and occur primarily in testes and ovaries. Platinum-based therapy followed by surgical resection of the residual lesion is generally the recommended treatment. In contrast, immature uterine teratomas are rare, with few cases reported in the literature. Moreover, there is no standard treatment for these tumors. Non-puerperal uterine inversion is also rare in women younger than 45 years of age, and neoplastic lesions are responsible for this condition. Here, we report a case of an immature uterine teratoma associated with uterine inversion. The patient underwent surgery followed by adjuvant chemotherapy and continues to be monitored.

Introduction

Teratomas are the most common type of germ cell tumor diagnosed and they primarily develop in testes and ovaries.1,2 Extragonadal teratomas represent 2-5% of adult germ cell tumors, and they mainly localize in mediastinum. Teratomas rarely originate in the uterus. To date, only four cases of immature teratoma of the uterus have been reported.3-5

Case Report

A 23-year-old woman presented with a three-month history of vaginal bleeding and malodorous discharge. She had no known comorbidities and her gynecological history included normal menstrual cycles that started at age 11. The patient also had no history of previous sexual activity. During a gynecological consultation in January 2013, a bulky tumor was visualized at the vaginal opening and a biopsy of the lesion was performed. No adnexal masses were detected. Histopathological examination was consistent with adenocarcinoma of the cervix. Since the material was not available for confirmation of this finding, the patient was referred to a gynecological oncologist who performed a second biopsy of the tumor. The patient developed a sudden episode of intense vaginal bleeding, fatigue, and dyspnea, resulting in hospital admittance. Her initial hemoglobin value was 5.4 g/dL (normal range, 12-16 g/dL) and blood transfusion was required. Concomitantly, the results of the second biopsy indicated necrotic tissue with inflammatory findings and an absence of neoplastic cells.

According to guidelines of the International

Correspondence: Maria Del Pilar Estevez Diz, Department of Radiology and Oncology, Instituto do Câncer do Estado de São Paulo, Faculdade de Medicina da Universidade de São Paulo, Avenida Doutor Arnaldo, 251, Cerqueira César, São Paulo SP, 01246-900, Brazil.

E-mail: maria.pilardiz@gmail.com

Key words: immature teratoma, uterine teratoma, uterine inversion.

Acknowledgments: we appreciate the contribution of Esther Oliva, MD, gynecological pathologist of Massachusetts General Hospital, Boston, MA, USA.

Contributions: KTS, conception, acquisition of data, analysis and interpretation of data, drafting, revising, and final approval of the article; MAVN, acquisition of data, analysis and interpretation of data, revising and final approval of the article; LSSR, acquisition of data, revising and final approval of the article; GDF, acquisition of data, analysis and interpretation of data, revising and final approval of the article; MDPED, conception, analysis and interpretation.

Conflict of interests: the authors declare no potential conflict of interests.

Received for publication: 26 June 2014. Accepted for publication: 28 July 2014.

This work is licensed under a Creative Commons Attribution NonCommercial 3.0 License (CC BY-NC 3.0).

©Copyright K.T. Souza et al., 2014
License PAGEPRESS, Italy
Rare Tumors 2014; 6:5530
doi:10.4081/rt.2014.5530

[Rare Tumors 2014; 6:5530]
Federation of Gynecology and Obstetrics, the current patient had a stage IA immature uterine teratoma. However, the patient had poor prognostic characteristics due to the carcinoid component detected. For this reason, adjuvant chemotherapy was performed with three cycles of bleomycin, etoposide, and cisplatin (BEP). Since August 2013, the patient has continued to be monitored and is disease-free twelve months later.

Discussion and Conclusions

Non-puerperal uterine inversion is rare in women younger than 45 years of age, and neoplastic lesions are often its cause. Few carcinomas and sarcomas of the endometrium have been found to be associated with uterine inversion, and there is only one report of an immature teratoma of the uterus causing this complication.

Since immature uterine teratomas are rare, there is no standard treatment for this condition. Therefore, most of the evidence for stage I disease after surgical treatment is obtained from case reports. In these case reports, clinical observations were used to determine various treatment methods, and these included two cycles of chemotherapy vincristine, actinomycin-D, and cyclophosphamide (VAC), an undescribed treatment regimen, and radiotherapy (Table 1). In addition, not all of the reports described patient follow-up and outcomes, thereby preventing a comparison of treatment decisions. Iwanaga et al. reported a patient that received adjuvant treatment with VAC, and subsequently presented no evidence of tumor recurrence five years post-treatment.

However, Newsom-Davis et al. described a para-aortic lymph node recurrence that occurred six months after surgery in a patient that was submitted to clinical follow-up alone. Therefore, rescue treatment was needed and involved an initial dose of cisplatin (20 mg/m²) and etoposide (100 mg/m²) in week 1, followed by a two-week alternating regimen of paclitaxel (135 mg/m²) and etoposide (150 mg/m²), and paclitaxel (135 mg/m²) and cisplatin (60 mg/m²). This rescue regimen led to a partial lymphadenopathy reduction and an increase in the cystic component of the lesion. Therefore, the patient underwent retroperitoneal lymph

Table 1. Previous uterine teratoma case reports with their management and outcomes.

| Study                        | Management                                      | Recurrence                |
|------------------------------|-------------------------------------------------|---------------------------|
| Newsom-Davis et al.          | Surgery + clinical observation                   | Para-aortic lymphadenopathy |
| Gomez-Lobo et al.            | Surgery + adjuvant chemotherapy (not described) | Not described             |
| Iwanaga et al.               | Surgery + adjuvant VAC ×2                        | No                        |
| Ansah-Boateng et al.         | Surgery + adjuvant radiotherapy                  | No                        |

VAC, vincristine, actinomycin-D, and cyclophosphamide.

Figure 1. Radiological (A, B) and surgical images of uterine inversion (C) associated with an immature uterine teratoma. Arrows point to uterine fundus.

Figure 2. Microscopy images (200× magnification) of immature glandular tissue (A); squamous epithelium (B); glandular, muscular, and cartilaginous components (C), and smooth muscle and glandular tissue (D).
node dissection, which confirmed a metastatic teratoma recurrence by histopathological analysis. The patient died one month after surgery due to postoperative complications.

In the current report, both previous publications regarding the treatment of ovarian germ cell tumors and the presence of a carcinoid tumor as a factor for poor prognosis were considered in the decision to select an adjuvant BEP treatment.11-15

In summary, this case report describes a rare puerperal uterine inversion due to an immature uterine teratoma, for which there is no established therapeutic management. According to the available case reports, surgical treatment is the only standardized treatment previously described, while indications for adjuvant therapy remain uncertain. For the current patient, a treatment of a germ cell tumor and a neuroendocrine tumor included three cycles of BEP in order to reduce the potential for relapse.

References

1. Albany C, Einhorn LH. Extragonadal germ cell tumors: clinical presentation and management. Curr Opin Oncol 2013;25:261-5.
2. Newsom-Davis T, Poulter D, Gray R, Ameen M, et al. Case report: malignant teratoma of the uterine corpus. BMC Cancer 2009;9:195.
3. Gomez-Lobo V, Burch W, Khanna PC. Nonpuerperal uterine inversion associated with an immature teratoma of the uterus in an adolescent. Obstet Gynecol 2007;110:491-3.
4. Iwanaga S, Shimada A, Hasuo Y, et al. Immature teratoma of the uterine fundus. Kurume Med J 1993;40:153-8.
5. Ansah-Boateng Y, Wells M, Poole DR. Coexistent immature teratoma of the uterus and endometrial adenocarcinoma complicated by gliomatosi peritonei. Gynecol Oncol 1985;21:106-10.
6. Moulding F, Hawnaur JM. MRI of nonpuerperal uterine inversion due to endometrial carcinoma. Clin Radiol 2004;59:534-7.
7. Oguri H, Maeda N, Yamamoto Y, et al. Nonpuerperal uterine inversion associated with endometrial carcinoma—an case report. Gynecol Oncol 2005;97:973-5.
8. Lupovitch A, England ER, Chen R. Nonpuerperal uterine inversion in association with uterine sarcoma: case report in a 26-year-old and review of the literature. Gynecol Oncol 2005;97:938-41.
9. Tucket JD, Yeung A, Timmons G, Hughes T. Non-puerperal uterine inversion secondary to uterine sarcoma and ascites demonstrated on CT and MRI. Eur J Radiol Extra 2010;75.
10. Case AS, Kirby TO, Conner MG, Huh WK. A case report of rhabdomyosarcoma of the uterus associated with uterine inversion. Gynecol Oncol 2005;96:850-3.
11. Pectasides D, Pectasides E, Kassanos D. Germ cell tumors of the ovary. Cancer Treat Rev 2008;34:427-41.
12. Gershenson DM. Management of ovarian germ cell tumors. J Clin Oncol 2007;25:2938-43.
13. Parkinson CA, Hatcher HM, Earl HM, Ajithkumar TV. Multidisciplinary management of malignant ovarian germ cell tumours. Gynecol Oncol 2011;121:625-36.
14. Vicus D, Beiner ME, Clarke B, et al. Ovarian immature teratoma: treatment and outcome in a single institutional cohort. Gynecol Oncol 2011;123:50-3.
15. Patterson DM, Murguasen N, Holden L, et al. 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.