

Case Report

Laparoscopic Cystectomy as Exclusive Treatment for an Immature Teratoma: A Follow-Up Case Report

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Keywords

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- Laparoscopic cystectomy
- Conservative treatment

Abstract

Malignant germ cell tumors, while relatively rare, raise concerns about fertility when occurring in young women. We described the case of a 23 year old nulligravida, with a history of previous right adnexectomy for a mucinous cystadenoma, who presented with an adnexal mass compatible with a dermoid cyst at a routine pelvic ultrasound. A laparoscopic cystectomy revealed a final pathologic diagnosis of a grade 1 immature teratoma. The patient's age and history suggested conservative clinical management, without adjuvant therapy. Eight years later, the patient remains free of recurrence.

ABBREVIATIONS

CT Scan: Computed Tomography; CEA: Carcino Embriologic Antigen; CA 125: Carcinogenic Antigen 125; AFP: Alphafeto Protein; NMR: Nuclear Magnetic Resonance

INTRODUCTION

Malignant germ cell tumors are rare, and account for about 5% of all ovarian neoplasms [1]. They are divided into two groups: dysgerminomas and non-dysgerminomas. The latter include mixed germ cell tumors, endodermal sinus tumors, immature teratomas and strumaovarii. Immature teratomas comprise about 1% of ovarian neoplasms. Microscopic examination reveals a disorderly mixture of tissues derived from all three germ cell layers, with at least one component, usually the neuroectoderm, having an immature, embryonic appearance. The tumors are graded from 1 to 3 based on the amount of immature neural tissue. The tumor grade correlates with prognosis, and guides recommendations regarding chemotherapy, along with other factors. These tumors occur mainly in young women, 16 to 25 years old, raising concerns about future fertility [2-4]. Fortunately, they are bilateral in less than 5% of cases, although the contra lateral ovary may contain a dermoid cyst. In addition, in contrast to epithelial ovarian neoplasms, immature teratomas are generally diagnosed at an early stage, which permits removal of the involved adnexa with preservation of the contra lateral ovary and uterus [5-7]. Difficulties in pre-operative diagnosis have led some patients to have only the cyst removed, even when proper diagnosis might have led to a less conservative treatment,

with satisfactory outcomes [7]. Moreover, the favorable response of these tumors to the newest chemotherapy regimens allows for adequate treatment of eventual post-operative recurrences [7].

CASE PRESENTATION

N.S.E, 23 year old Asymptomatic nulligravida, with a previous right oophorectomy for a mucinous cystadenoma (pyloric, intestinal and müllerian morphology) presented on a routine evaluation with left adnexal enlargement. Transvaginal sonography revealed a complex ovarian cyst, 3.8 cm in diameter. Serum CEA was 2.6ng/ml and CA 125 was 8.9 ng/ml, both within the normal range. A CT scan did not reveal any other abnormal findings in the abdomen. To preserve the patient's fertility, in September 2004, she had a laparoscopic cystectomy, without tumor capsule rupture. No other abdominal implants or masses were seen upon thorough examination of the abdominal cavity. The final pathologic diagnosis was a grade 1 immature teratoma, composed mainly of mature tissues and sparse foci of immature neural tissues. As the neoplasm was a low grade, confined to the ovary and presented itself in a young nulligravida woman with a previous adnexectomy, the follow up has been conservative, consisting mainly of regular transvaginal sonograms and monitoring of serum tumor markers, including AFP. After five years, the patient remains asymptomatic and free of recurrence, as evidenced by her most recent pelvic NMR.

DISCUSSION

Despite their rarity, germ cell tumors deserve special attention among ovarian neoplasms as they occur mainly in young women,

for whom fertility is of special concern. About 80% of malignant germ cell tumors are improperly staged, mainly because they present themselves as misleadingly innocent-looking masses upon pre-operative evaluation and are primarily treated in non-oncologic settings, often with cystectomies that later reveal a malignant diagnosis. The oncologist must then choose between: careful postoperative follow up, including imaging techniques and serum tumor markers; re-exploration, with or without oophorectomy, for adequate staging; or adjuvant chemotherapy [8]. As immature teratomas are generally unilateral, it is currently accepted practice, when the contra lateral ovary and abdominal cavity are macroscopically normal, to proceed with a unilateral adnexectomy, preserving the normal ovary and uterus, in addition to cytologic analysis of the peritoneal fluid, careful inspection of the entire peritoneal cavity, biopsy of the main structures at risk, as well as careful palpation of pelvic and para-aortic lymph node-bearing areas [6,9]. Stage Ia, grade 1 immature teratomas do not require post-operative chemotherapy, as conservative surgical treatment is highly curative [2,6,10,11]. A study of eight patients with immature teratomas removed by cystectomy, without adequate surgical staging due to no evidence of malignancy before and/or during surgery. Only four of these patients, those with grade 2 and 3 tumors, had chemotherapy. During an average of 56.5 months of follow-up, no recurrences were observed [8]. Similarly, in six patients with dermoid cysts removed by cystectomy that were diagnosed as immature teratomas post-operatively, there were no reports of recurrence after an 18 to 84 month follow up period [12]. In another study of 32 patients with immature teratomas, of whom 30 had unilateral adnexectomy, and only 10 received adjuvant chemotherapy, there were no reports of recurrence after 10 years of follow-up [13]. Others have studied conservative surgery as the exclusive treatment of immature teratomas in children, with low recurrence rates. However, there are no references to cystectomy [10,14]. Most data suggest that, even in patients with grade 2 or 3 immature teratomas, conservative surgery may suffice. In cases of recurrence, rescue treatment regimens, consisting mainly of radical surgery and chemotherapy, are highly curative. In fact, a retrospective analysis of 35 patients treated for immature teratomas from 1975 to 1995 revealed that conservative surgery was performed in 26 patients (25 salpingo-oophorectomies and one cystectomy). Only two patients did not receive adjuvant chemotherapy, of which one had a cystectomy. The other was diagnosed with a stage Ia grade 3 immature teratoma, and presented with recurrence seven months after surgery. Treatment was completed with complete hysterectomy, salpingo-oophorectomy, and debulking and adjuvant chemotherapy (15 cycles of VAC therapy). After a 156-months follow-up period, she showed no further signs of recurrence. A significant retrospective study of 760 young patients treated between 1988 and 2001 for malignant ovarian germ cell tumors also revealed no statistically significant difference in survival among patients treated with fertility-preserving surgeries. Poor prognosis was associated with older age, advanced stage and yolk sac tumor histology [15]. Chan and colleagues concur with the fact that younger age and early-stage disease are important predictors for improved survival in patients with ovarian sex cord-stromal tumors. Conservative surgical treatment for early-stage patients wishing to retain fertility appears to be a safe

alternative in this population [11,16,17]. Yet, as most patients in the studies described have a healthy second ovary, cystectomy is seldom described as the approach to fertility-sparing surgery. In fact, most procedures include oophorectomy, staging and sparing of the uterus. In the case at hand, a grade 1 immature teratoma was diagnosed in a young nulligravida with previous contra lateral ovariectomy. Although no adequate staging was performed as there was no evidence of malignancy pre- or intra-operatively, active imaging and serum marker follow-up made conservative management possible. In addition, in case of an eventual recurrence, rescue surgery and chemotherapy produce highly satisfactory outcomes. We suggest that cystectomy might be an alternative for selected young patients with immature teratomas, coincident with accurate staging. Unfortunately, there has not been a multi centric prospective study to determine if cystectomy leads to similar cure rates as oophorectomy in stage Ia, grade I disease.

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