



Conservative surgical staging as a means to preserve fertility in patients with dysgerminoma: a case report

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Introduction and importance: Ovarian tumor is a rare condition in pediatrics. Due to the improvement in surgical techniques and chemotherapy in recent years, it is currently possible to preserve fertility in selected cases of patients who desire motherhood.

Case presentation: We present a case of pregnancy following conservative surgery and complete chemotherapy for ovarian dysgerminoma stage IIA. A 16-year-old female presented with complaints of abdominal mass and discomfort. Histopathological examination displayed dysgerminoma arising from the right ovary. Conservative treatment with right salpingo-oophorectomy and six cycles of chemotherapy was performed. Within 2 years following the surgery, the patient conceived and did not indicate tumor recurrence.

Clinical discussion: Dysgerminoma is the most common ovarian malignant germ cell tumor and usually affects women at a young age. Conservative surgery followed by chemotherapy is the treatment of choice, particularly in young patients with a strong desire to have a family. Conservative fertility-sparing surgery does not have inferior outcomes in terms of survival or recurrence. It is possible that in certain cases, patients previously treated for ovarian cancer may retain their fertility, thereby improving their quality of life.

Conclusion: Most patients with dysgerminoma can be treated with the maintenance of normal reproductive function. Reassurance regarding the high probability of conceiving and having normal children after conservative surgery and chemotherapy should be informed to the patient and family.

Keywords: case report, chemotherapy, conservative surgery, dysgerminoma, pregnancy

Introduction

Surgical management of gynecological malignancies often involves removing ovaries and the uterus. This will affect women of reproductive age due to the loss of fertility. New developments in assisted reproductive technologies, including ovarian tissue, oocytes, or embryo cryopreservation, have been developed to preserve fertility in a woman with cancer. Fertility preservation prior to gonadotoxic treatments is an imperative option. Unfortunately, it is still experimental in Indonesia, and the

HIGHLIGHTS

- Ovarian tumor is a rare condition among children.
- Many young patients with gynecologic cancer are not aware of fertility preservation.
- Ovarian cryopreservation has not been established in Indonesia.
- Conservative surgical staging may be an alternative to preserve fertility.

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technique has not yet been established. Radiation should be evaded to minimize the pelvic adhesions that might compromise fertility^[1].

Many young patients with gynecologic cancer are not attentive to the opportunity to preserve fertility. Fear of cancer prognosis and treatment can become overwhelming^[2]. Moreover, they are not offered such an option by a surgeon who is uninformed, skeptical, or not proficient at accomplishing fertility-preserving surgery^[1]. Conservative surgical staging is defined as surgery with complete staging and preservation of at least the uterine corpus and a part of one ovary. This can be an alternative when ovarian tissue cryopreservation or other means of fertility preservation are not conceivable^[3].

We present a case of ovarian dysgerminoma, which was successfully treated with conservative surgery and chemotherapy. This work has been reported in line with the Surgical CAse REport (SCARE) criteria^[4]. This study is expected to provide an overview of the successful preservation of fertility function after treatment in a woman with ovarian dysgerminoma.

Presentation of the case

A 16-year-old Indonesian female patient was referred to our hospital with a solid ovarian neoplasm. This patient had complained of an abdominal palpable mass and discomfort for 6 months before admission. She felt that the mass progressively increased in size and reached the size of 5 months of gestation on the day of admission. There was a history of nonspecific gastrointestinal complaints such as nausea, vomiting, and dyspepsia. Menarche was at the age of 13, with a regular menstrual cycle and an average flow and duration. The voiding and defecation were normal. There was no history of unexplained weight loss or fatigue and no family history of either breast, ovarian, or colon cancer.

On physical examination, we palpated a firm, smooth, round-shaped mass. The mass upper limit was in the middle of the xiphoid process and the navel. A pelvic examination confirmed the presence of a large, immobile pelvic abdominal mass. Ultrasonography showed a large solid mass measuring $20 \times 11 \times 17$ cm adjacent to the lower part of the uterus and posterior to the urinary bladder. The volume was ~ 2070 ml. The mass had a clear border and neovascularization with a resistance index (RI) of 0.38. The left ovary was normal, and no evidence of ascites or hydronephrosis. A prominent level of lactate dehydrogenase (LDH) [2203 IU/l (normal 135–225 U/l)] with an elevated serum human chorionic gonadotropin (hCG) [122 mIU/ml (normal < 10 mIU/ml)] and CA-125 [117.2 U/ml (normal < 21 U/ml)] were detected. Whilst serum α -1-fetoprotein (AFP) was within the normal range [1.9 ng/ml (normal 40–129 U/l)].

A team of experienced gynecologic oncologist carried out laparotomy salpingo-oophorectomy procedure on the patient. A lobulated mass sized $24 \times 16 \times 8$ cm was found arising from the right ovary (Fig. 1). Frozen section discovered a suspicious germ cell tumor originating from the right ovary. Conservative treatment with right salpingo-oophorectomy was performed. No ascites, adhesions, or metastatic lesions were found. The left ovary, appendix, and uterus all appeared normal. Peritoneal cytology was also done. Surgical procedures continued with omentectomy, appendectomy, and biopsies of the bilateral paracolic and para vesical lymph nodes. Pathological examination revealed a pure dysgerminoma that had reached the right



Figure 1. Laparotomy showed a $24 \times 16 \times 8$ cm lobulated mass arising from the right ovary.

fallopian tube corresponded to stage IIA. The patient's post-operative course was uneventful and without complications. She then received six cycles of chemotherapy with bleomycin, platinum, and vinblastine every 3 weeks.

After six courses of chemotherapy, AFP and CA-125 were reduced to levels of 2.1 ng/ml and 12 IU/ml, respectively. Ultrasound examination showed no residual mass. The patient was in good health during the follow-up period. She continued to have a regular menstrual cycle, lasting 4–5 days and occurring every 30 days. No recurrence was observed within 2 years after the surgery.

The patient then came in 29 weeks pregnant. The estimated fetal weight was 1250 g, with a normal amniotic index and placenta attached to the front wall of the uterus. No adnexal mass or other abnormalities of the genital tract were perceived. She delivered the baby 3 months later; the pregnancy was without complications, and the child was healthy.

Discussion

Dysgerminoma is the most common ovarian malignant germ cell tumor and usually affects women at a young age^[5]. This tumor is generally symptomatic and mostly present at stage I. However, patients may misinterpret the early symptoms as pregnancy, which can lead to diagnostic delay. Approximately 90% of cases are unilateral; thus, conservative surgery followed by chemotherapy is the treatment of choice, particularly in young patients with a strong desire to have a family^[6–9]. Most ovarian germ cell malignancies, including dysgerminoma are chemosensitive and have brought about a dramatic amendment in the prognosis for malignant ovarian germ cell tumors^[3,9]. Fertility tends to be lost in association with ovarian failure, although dysgerminoma is highly radiosensitive with a 75–90% survival probability^[5]. Considering the development of surgical performance and chemotherapy regimens, it is possible that in certain cases patients previously treated for ovarian cancer may retain their fertility, thereby improving quality of life. Nonetheless, the dispute between conservative versus nonconservative primary surgery in patients with dysgerminoma has been raised for decades.

Proponents of nonconservative therapy have argued that 3–25% of patients have bilateral ovarian involvement at the time of surgery; 15.9–52.4% of patients had tumor recurrence, with up to 35% occurring in the well-preserved opposite ovary. Likewise, the survival after relapse has been limited to 45–65%^[10]. The exclusively early-stage disease presentation underpinned our decision to conserve fertility in addition to the patient's desire to establish a family. Women of childbearing age who survived cancers and want to have children in the future may face significant regret because of potential infertility^[11]. All factors involved in tumor characteristics and patient compliance should be considered in making the decision. Dysgerminoma may coincide with menstrual irregularities. However, these were not present in this case. Appropriate surgical treatment for patients whose fertility wishes to be preserved consists of laparotomy with unilateral salpingo-oophorectomy and resection of all visible tumor masses. Based on NCCN (National Comprehensive Cancer Network) Guidelines 2020, management of the ovarian germ cell tumors for patients with fertility desires is fertility-sparing surgery and comprehensive surgical staging. Comprehensive surgical staging should still be implemented to rule

out occult, higher-stage disease^[12]. As in this case, the frozen section of the mass revealed a suspicious germ cell tumor originating from the right ovary. Besides conservative treatment with right salpingo-oophorectomy, peritoneal cytology was also performed. No ascites, adhesions, or metastatic lesions were observed. The left ovary, appendix, and uterus all appeared normal. Surgical procedures continued by omentectomy, appendectomy, and biopsy of bilateral paracolic and perivesical lymph nodes.

Pathologic examinations indicated a pure dysgerminoma that had reached the right fallopian tube and corresponded to stage IIA. Chemotherapeutic agents suggested by NCCN are bleomycin, etoposide daily for 1–5 days and cisplatin for 1–5 days for 3–4 cycles. The patient displayed a complete clinical response. We decided to examine several tumor markers every 2–4 months for 2 years. It is worth noting that conservative fertility-sparing surgery does not have inferior outcomes in terms of survival or recurrence. A study involving 182 patients with malignant germ cell tumors confined to one ovary showed that fertility-sparing surgery has no poorer prognosis. Fertility-sparing surgery is favored when a woman wishes to preserve reproductive function since there is no robust evidence to suggest the tumor spreading to the contralateral ovary, even in the case of extra ovarian disease^[13].

Of the several classes of chemotherapeutic agents, those most related to ovarian toxicity are alkylating agents. Furthermore, when combination therapy is applied, the effect of any single agent is more difficult to measure^[14,15]. The accurate mechanism by which a given chemotherapeutic agent provokes ovarian injury is inadequately understood. Chemotherapy and radiation have been linked to depletion of primordial follicles. During the treatments, ovaries are subjected to DNA damage and apoptosis leading to accelerated follicles loss and infertility^[16,17]. The influence of chemotherapy on follicular health in term of the first cell types affected by chemotherapy are varied among studies^[18,19]. Cisplatin has been proven to trigger the apoptosis events in granulosa cells within primordial follicles, resulting in increased follicle loss^[20]. The magnitude of follicular loss is interconnected with the accumulative dose and duration of chemotherapy^[14].

Conclusion

In summary, most patients with dysgerminoma can be treated with the maintenance of normal reproductive function. Fertility function is unremarkable after fertility-sparing surgery and chemotherapy. Patients and family should be reassured about the high probability of retaining the ability to conceive and have normal children after conservative surgery and chemotherapy.

Ethical approval

This study is exempt from ethical approval.

Patient consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Author contribution

M.M. was responsible for the conception, study design, literature search, data acquisition and analysis, drafted, revised, and critically reviewed the manuscript. P.D.G. contributed to literature search, data acquisition and analysis, and drafted the manuscript. T.D.A. contributed to literature search, data acquisition and analysis, and drafted the manuscript. All authors read and approved the final manuscript.

Conflicts of interest disclosure

The authors declare that they have no conflicts of interest.

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