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Case Report

Huge follicular carcinoma originated from struma ovarii in young woman; Extremely rare case

Chul Min Park, Bo Ram Kim, Hye Sim Kang *

Department of Obstetrics and Gynecology, Medical School of Jeju National University, Jeju National University Hospital, Aran 13gil 15(Ara-1Dong), Jeju City, Jeju Self-Governing Province, 63241, South Korea

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ABSTRACT

Introduction: Struma ovarii (SO) is a rare ovarian mature teratoma which is composed of 50% or more thyroid tissues. Malignant transformation is reported to occur in less than 5% of all cases, and it leads to 5–6% metastatic disease. The most common type is a papillary carcinoma. However, follicular carcinoma of arising from SO is extremely rare.

Case presentation: A 23-year-old nullipara complained of right lower abdominal pain. CT scan revealed a large complex cystic and solid mass in pelvic cavity with moderate amount ascites with peritoneal thickening and omental infiltration. We performed a right salpingo-oophorectomy and surgical staging.

Clinical discussion: It was diagnosed with follicular carcinoma, stage IA. After the multidisciplinary team meeting, we decided not to conduct additional treatment, including total thyroidectomy followed by radioiodine treatment and T4 therapy because thyroid tests were normal findings.

Conclusion: There is still no established treatment consensus for follicular carcinoma of SO patients. The choice for a conservative or radical approach depends on the doctor. However, conservative surgery could be considered for low risk, early stage patients who have childbearing potential.

1. Introduction

Struma ovarii (SO) is a rare tumor, originated from an ovarian mature teratoma which is composed of 50% or more thyroid tissues [1]. It has been reported to represent 0.5–1% of all ovarian tumors and 2–5% of all ovarian teratomas [2]. Depending on the histologic features, SO can be classified as benign or malignant. While most often benign, malignant transformation is reported to occur in less than 5% of all cases, and it leads to 5–6% metastatic diseases [1]. The most common histological type of malignancy is a papillary thyroid carcinoma, which occur in about 70–85% of all cases [3,4]. Because follicular carcinoma arising from SO is extremely rare, the treatments and follow-up procedures are not clearly established. We report a very rare case of follicular carcinoma arising from SO diagnosed postoperatively following oophorectomy. This work has been reported in line with the SCARE 2020 criteria [5].

2. Case report

A 23-year-old nullipara complained of right lower abdominal pain for one week. She consulted at the local hospital and had abdominal computed tomography (CT) scan. The CT revealed a large, complex cystic and solid mass in the pelvic cavity with moderate amount ascites with peritoneal thickening and omental infiltration. Differential diagnosis for mature teratoma with complex cystic tumor such as dysgerminoma, endodermal sinus tumor, thecoma, brenner tumor, immature teratoma and combined peritoneal seeding were required (Fig. 1). She was referred to our hospital cancer center with a diagnosis of ovarian cancer.

There was nothing remarkable in her prior medical and surgical history. She had no family history of cancer. There were no specific findings on physical examination other than mild tenderness in the right lower abdomen. Her menstrual cycle was regular. The serum levels of CA-125 and CA19-9 were elevated, respectively 412.40 and 54.04. But Beta-HCG < 0.100, and AFP 1.70 were within normal range. The Positron Emission Tomography-Computed Tomography (PET-CT) scan showed a heterogeneous hypermetabolic mass (SUVmax = 5.4), which was suspected of the possibility of malignant mass in the pelvic cavity (Fig. 2). On October 11th, 2019, the patient underwent an exploratory laparotomy with suspicion of ovarian malignancy, specifically germ cell

* Corresponding author.
E-mail addresses: obgymd@gmail.com (C.M. Park), ukino327@gmail.com (B.R. Kim), kanghs@jejunu.ac.kr (H.S. Kang).

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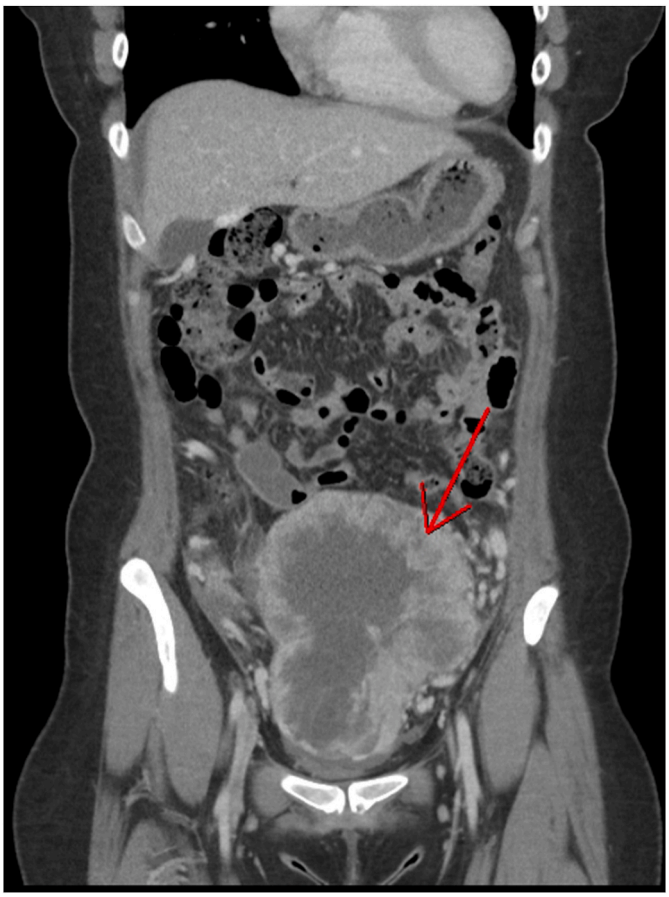

Fig. 1. Abdominal computed tomography (CT) revealed a large complex cystic and solid mass in the pelvic cavity with moderate amount ascites with peritoneal thickening and omental infiltration.

tumor. During the operation, an approximately 15cm sized solid mass was identified on the right ovary, which was severely adhered to omentum with small amount of ascites. We performed a right salpingo-oophorectomy and a partial omentectomy. The outer surface of mass was smooth and glistening. On frozen section, Histology suggested granulosa cell tumor. The uterus and left adnexa were unremarkable, grossly. After the whole abdominal exploration, we could not find any suspected metastatic lesion. Therefore, we decided to leave the uterus and left adnexa, and then after carrying out multiple peritoneal biopsies, the surgery was completed.

On final histologic diagnosis, microscopic examination revealed scattered, non-tumorous thyroid tissue around the tumor. In addition, cartilage tissue and respiratory type epithelium was observed, suggesting that this tumor developed from the pre-existing mature teratoma in the ovary (Fig. 3A and B). Tumor cells exhibited trabecular and solid pattern of follicles with minimal nuclear atypia (Fig. 3C). Upon immunohistochemistry, the tumor cells were strongly positive for thyroid transcription factor-1 and thyroglobulin (Fig. 3D and E). Based on the morphologic and immunohistochemical findings, it was diagnosed with follicular carcinoma arising from mature teratoma in the ovary. The surgical stage was IA, according to the International Federation of Gynecology and Obstetrics (FIGO).

After multidisciplinary team meeting, we decided not to conduct additional treatment including total thyroidectomy followed by radioiodine treatment and T4 therapy because thyroid ultrasonography and the thyroid function test (TFT) with thyroglobulin and thyroglobulin antibody were normal findings. The patient's CA-125 and CA 19-9 concentrations decreased gradually and normalized after 3 months. We planned to do thyroglobulin, TFT, CA-125 and CA19-9 regularly every 3 months, and pelvis Magnetic resonance imaging (MRI) every 6 months for two years, and 6–12 months thereafter. Two years later, no recurrence or metastasis has been found.

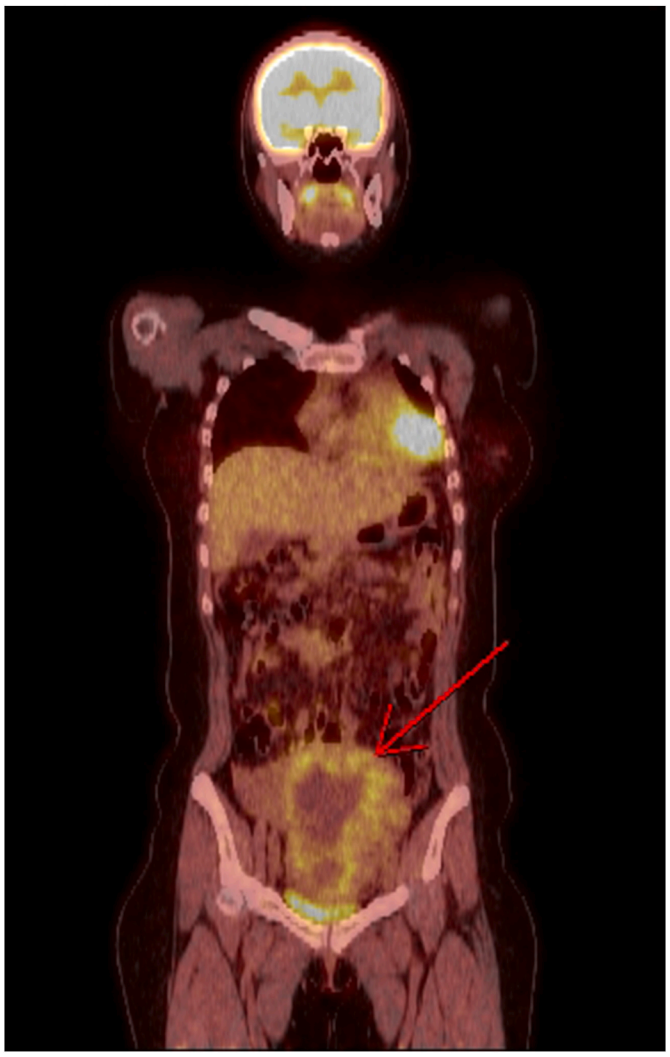

Fig. 2. Positron Emission Tomography-Computed Tomography (PET-CT) showed a heterogeneous hypermetabolic mass (SUVmax = 5.4), which was suspected of the possibility of malignant mass in the pelvic cavity.

3. Discussion

It is well-known that follicular carcinoma of the thyroid gland has a relatively worse prognosis than papillary carcinoma. It is also expected that follicular carcinoma arising from SO will have a similar prognosis. Roth et al. reported that follicular carcinoma originating from SO had a 45% dissemination frequency, which was 19% higher than the cases of papillary carcinoma [6].

There is no consensus on the optimal treatment of women with malignant SO. Ayhan et al. recommended fertility-conserving management for patients with benign SO, and radical staging surgery for those who have malignant SO without future fertility desire [7]. Robby et al. also reported that cases of malignant SO require an ovarian cancer surgical staging with pelvic washing cytology, total hysterectomy with bilateral salpingo-oophorectomy and lymphadenectomy, as well as a total thyroidectomy followed by radioactive Iodine(^{131}I) ablation [8]. There is little evidence in literature on the conservative management in cases with evidence of malignancy. But in cases of fertility preservation, a unilateral salpingo-oophorectomy could be performed, along with

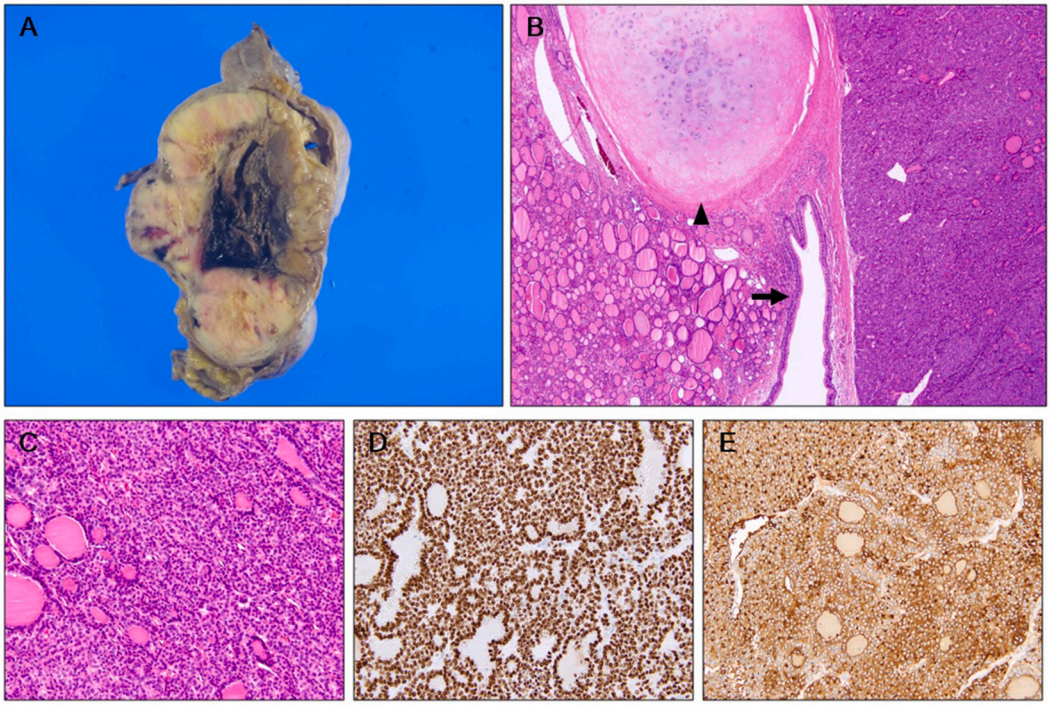

Fig. 3. Gross and microscopic findings of the tumor. (A) A huge yellowish solid tumor with central hemorrhagic necrosis. (B) Hematoxylin and eosin stain of tumor sections showed thyroid follicular cells accompanied by cartilage tissue (indicated by an arrow head) and respiratory epithelium (indicated by an arrow) (× 40 magnification). Tumor cells show diffuse and follicular growth patterns (C) with thyroid transcription factor-1 (D) and thyroglobulin (E) positivity (× 100 magnification).

levels of serum thyroglobulin as a marker of relapse [9]. DeSimone et al. also supported the conservative surgery for women with malignant SO who had childbearing potential [10]. Adjuvant treatment to perform a thyroidectomy and administer radioiodine after oophorectomy should be based upon the presence of metastatic disease and the risk of recurrence. Further, the use of iodine-131 as adjuvant therapy in malignant SO remains controversial due to the lack of information and large prospective trials. In our case, we could not find any suspected metastatic lesion except right ovary malignant SO in the young woman, therefore the uterus and what remained of the adnexa were left in place for fertility preservation. Additionally, we chose not to do adjuvant treatment, including total thyroidectomy, radioiodine treatment, and T4 therapy.

CA-125 is well-known as a specific tumor marker of ovarian malignancy, especially for non-mucinous epithelial ovarian carcinomas. And it may also increase in several benign conditions such as endometriosis, uterine fibroids, pelvic inflammatory disease, or as a secondary effect due to the presence of ascites [11]. In our case, we presumed that CA-125 was increased with moderate amount ascites and normalized after operation.

The prognosis of malignant SO is not well-known because of the rarity of this disease. In the literature, recurrence occurs in approximately 15–35% of malignant SO [10,12]. The average time to recurrence is approximately 4–6 years, mostly in patients without adjuvant treatment [10]. DeSimone et al. recommended the regular monitoring of all benign and malignant SO for at least 20 years due to late recurrence of some cases [10]. Yoo et al. also supported extended regular follow-up for a median of 65 months in malignant SO [2].

In conclusion, there is still no established treatment consensus for follicular carcinoma of SO patients. The choice for a conservative or radical approach depends on the doctor in charge of the patient. However, conservative surgery could be considered for low risk, early stage patients who have childbearing potential.

Declaration of competing interest

All authors declare no conflict of interest.

Acknowledgments

None.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2021.103018.

Ethical approval

This is a case report, therefore Ethics committee/IRB approval is not required.

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Author contribution

Chul Min Park: physician of patient, written the paper, correction of the paper.
Bo Ram Kim: written the paper, correction of the paper.
Hye Sim Kang: corresponding author and correcting the paper.

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.

Research registration

Not applicable.

Guarantor

Hye Sim Kang.

Provenance and peer review

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