Case Report

Primary strumal carcinoid of the ovary: a case report and literature review

Ling Wu¹, Zhou Wang², Shuang Dai¹, Jingyu Zheng¹, Jianmin Li¹

¹Department of Pathology, The First Affiliated Hospital of Wenzhou Medical University, Wenzhou, Zhejiang, P. R. China; ²Department of Urology, Tongde Hospital of Zhejiang Province, Hangzhou, Zhejiang, P. R. China

Received October 29, 2015; Accepted December 25, 2015; Epub February 1, 2016; Published February 15, 2016

Abstract: Thyroid tissue is a relatively frequent component of mature teratoma and can occur in 5-20% of cases. Primary strumal carcinoid tumor of the ovary (SCTO) is an extremely rare entity, though the survival rate is excellent if the disease is confined to one ovary. A 51-year-old, gravida 2, para 2, woman presented to our hospital with lower abdominal distension for 2 days. The patient had no signs or symptoms of carcinoid syndrome. B-ultrasound tip: the right accessories area cystic mass (considering ovarian cyst torsion). Patient had a right attachment surgery. Because of the slow development of the tumor, the patient is still alive after surgery two years.

Keywords: Ovarian, goiter carcinoid

Introduction

Primary ovarian carcinoids are very rare tumors. Only 5% of all carcinoids are ovarian and account for less than 0.1% of all ovarian malignancies [1]. Primary ovarian carcinoids were divided into four subtypes: island, trabecular, goiter and mucinous type. Carcinoid goiter is thyroid tissue composition and intimately mixed with carcinoid. It's reported that patients had primary ovarian carcinoid limited to one ovary got a good prognosis [2, 3]. However, some literature suggests that these tumors may be transferred, even fatal [4, 5]. In this study, we present a rare case of primary ovarian carcinoid goiter merger mature teratoma and review of the literature. Microscopically, the tumor contained definite a carcinoid tumors and thyroid organizations.

Case report

A 51-year-old, gravida 2, para 2, woman presented to our hospital with lower abdominal distension for 2 days on August 10, 2013. She had no other medical history and without any symptoms of carcinoid syndrome. On physical examination, her body temperature was 37.5

degrees. Superficial lymph nodes were not palpable.

The results of relevant laboratory studies were: WBC 16,000/mm³, RBC 359 × 10^4 /mm³, Hb 10.2 g/dL, platelet 148,000/mm³, aspartate aminotransferase 15 U/L, alkaline phosphatase 27 U/L, total bilirubin 0.4 mg/dL, total protein 6.4 g/dL, α -fetoprotein (AFP) 1.1 ng/ml (normal range, <9 ng/ml), carcinoembryonic antigen (CEA) 4.9 ng/ml (normal range, <5 ng/ml), carbohydrate antigen (CA) 199, 13.2 U/ml (normal range, <35 U/ml). HBtsAg and anti-HCV antibody were both negative. There were no abnormal findings on chest X-ray or gastroscopy, and the patient's medical, surgical, and family histories were unremarkable.

B ultrasound showed

Pelvic cystic tumors cystadenoma? Teratoma be ranked pelvic fluid, see a cystic mass about the size of $132 \times 68 \times 125$ MM of pelvic, intracapsular see many separate samples and floculent echo, intracapsular see a hyperechoic size of about 32×25 MM nodules; CDFI: no abnormal blood flow in the uterus color display (**Figure 1**).

Ovarian carcinoid goiter

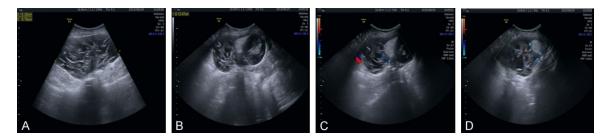


Figure 1. A and B. B ultrasound showed: Pelvic cystic tumors cystadenoma? Teratoma be ranked pelvic fluid, see a cystic mass about the size of $132 \times 68 \times 125$ MM of pelvic, intracapsular see many separate samples and floculent echo, intracapsular see a hyperechoic size of about 32×25 MM nodules; C and D. CDFI: no abnormal blood flow in the uterus color display.

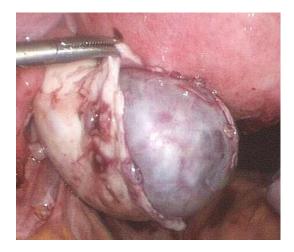


Figure 2. A capsule, the size of $132 \times 68 \times 125$ MM, capsule with sebum, hair, bladder wall thickness of $0.1 \sim 0.6$ cm, local thickening, thickening the range of about 0.5 cm in diameter.

Gross examination

A capsule, the size of $132 \times 68 \times 125$ MM cm, capsule with sebum, hair, bladder wall thickness of $0.1 \sim 0.6$ cm, local thickening, thickening the range of about 0.5 cm in diameter (**Figure 2**).

Microscopic features

Strumal carcinoid is an unusual form of ovarian teratoma composed of an intimate admixture of thyroid and carcinoid tissues that vary in their relative proportions. Carcinoid region intimately mixed with different sizes of follicles. The tumor cells have relatively consistent eosinophilic cytoplasm, flat core, "salt and pepper" chromatin kind, full of eosinophilic follicular thyroid kind of gum. There is a transition

between two components, but they have a closely mixed (Figure 3).

Immunohistochemistry

This case of carcinoid tumor cell areas were expressed synaptophysin, chro-mogranin A, CD56 and CK19; follicular colloid contains thyroglobulin expression. All cells did not express calcitoni (Figure 4).

The postoperative course was uneventful. B ultrasonic examination showed normal image after the operation for three months.

Discussion

Goiter carcinoid belongs to monodermal teratoma, which is mixed with thyroid tissue and carcinoid. The patient had no signs or symptoms of carcinoid syndrome. However, since the matrix luteinizing endocrine changes, such as masculine, hirsutism, endocrine hyperplasia.

It has been reported that goiter carcinoid may be associated with C cell [6]. According to the site where the tumor growth and the pattern, it had been divided into three types [7, 8]:

(1) The aneurysm wall nodules, which located within the cyst wall and protruded into the cavity has 1-8 cm diameter. Occasionally tumor tissue showed diffuse distribution, wall thickening; (2) Simple type. Tumor has diameter of 0.2-20 cm and homogeneous. Carcinoid is pale yellow or gray, often accompanied by hemorrhage or necrosis, and thyroid follicular lumen area is filling with glue-like substance; (3) Hybrid, this cancer is usually mixed with teratoma and cystic teratoma, which capsule has

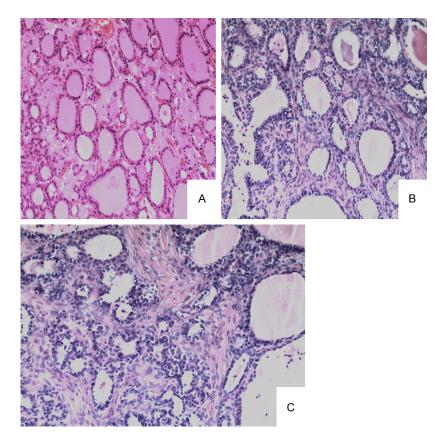


Figure 3. A. Tumor contains the carcinoid and a mixture of thyroid tissue, thyroid tissue show normal thyroid tissue morphology (H&E, \times 100); B and C. Carcinoid region intimately mixed with different sizes of follicles, tumor cells have relatively consistent eosinophilic cytoplasm, flat core, "salt and pepper". chromatin kind, full of eosinophilic follicular thyroid kind of gum (H&E, \times 100).

visible hair and sebum-like substance. It is often constituted by the carcinoid tumor, thyroid follicular and other ingredients, but pure island type are rare. The cells of Beam-like type of cancer showed short spindle, consistent size, single or several layers arranged elongated or curved strip, the tumor cells arranged in a vertical way. Abundant cytoplasm, eosinophilic, granular, nuclear is small and uniform fine chromatin, mitotic rare. The tumor cells arranged in solid nests, the nest mostly palisade. Normal follicular thyroid follicular size mixed, stratified squamous lining, cubic or columnar epithelium, eosinophilic intraluminal is filling with glue-like substance. Tumor or the number of different areas of the same tumor thyroid follicular has different sizes. The patients immunohistochemical staining showed: syn and CgA are positive support which is derived from thyroid C cells, is a tumor APUD [9].

Due to small number of cases, the treatment remains controversial. This matter underwent

bilateral tubal cancer. ovarian and uterine resection or unilateral oophorectomy, the prognosis is similar. Therefore, if the young patients, the contralateral ovary without exception, can unilateral oophorectomy [10, 11]. If the tumor follicular papillary carcinoma or follicular cancer, the patients should be combined with chemotherapy or radiotherapy. Tumor metastasis and recurrence of adjuvant treatment including radiation therapy, chemotherapy and isotope therapy [12, 13]. Goiter carcinoid classified as borderline, the prognosis is relatively good. A clinicopathological study of ovarian carcinoid tumors has found that the merger of teratoma's ovarian carcinoid tumors has less metastasis and carcinoid syndrome than non-consolidated teratoma, and 5-

year survival rate is higher [14]. Benign ovarian goiter has a good prognosis; If the tumor confined to one side of the ovary, surgery can achieve the purpose of complete cure, the prognosis is good [15, 16]. Kurabayashi et al. [4] reported the case of patients with clinical stage Ia and breast bone metastasis. Two cases have reported that goiter carcinoid had killed the patient [5]. Davis et al. [1] reported that patients with stage I of goiter carcinoid, they can survive 5-10-year of 100%.

In addition, carcinoid can concomitant heart disease (tricuspid insufficiency, pulmonary stenosis, etc.). It may persist after tumor resection, and even continue to progress, to become the main factors affecting the quality of life and survival. It is worth the long-term follow-up.

Disclosure of conflict of interest

None.

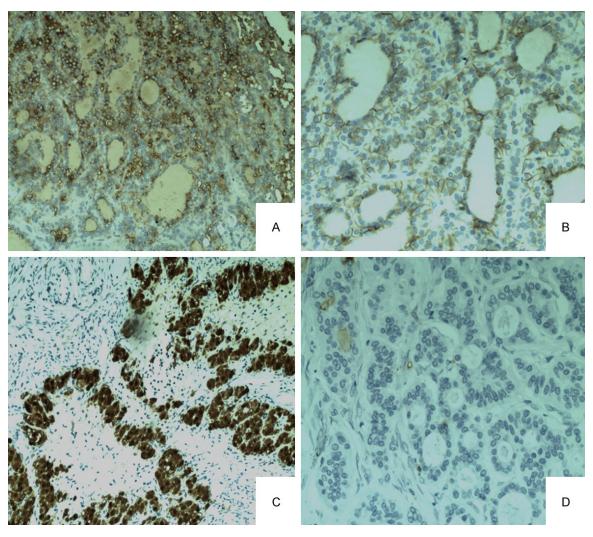


Figure 4. A-C. CD56, chro-mogranin A (CgA) and synaptophysin (Syn) were expressed in the case of carcinoid tumor cell areas (H&E, × 100); D. All cells did not express calcitoni (H&E, × 100).

Address correspondence to: Dr. Jianmin Li, Department of Pathology, The First Affiliated Hospital of Wenzhou Medical University, Wenzhou 325000, Zhejiang, P. R. China. Tel: +86-15088928123; Fax: +86-577-86689900; E-mail: wuxiao50@126.com

References

- [1] Davis KP, Hartmann LK, Keeney GL, Shapiro H. Primary ovarian carcinoid tumors. Gynecol Oncol 1996; 61: 259-265.
- [2] Gorin I, Sastre-Garau X. Strumal cacinoid tumor of the ovary. J Clin Oncol 2008; 26: 2780-2781.
- [3] Somak R, Shramana M, Vijay S, Nita K. Primary carcinoid tumor of the ovary: A case report. Arch Gynecol Obstet 2008; 277: 79-82.
- [4] Kurabayashi T, Minamikawa T, Nishijima S, Tsuneki I, Tamura M, Yanase T, Hashidate H,

- Shibuya H, Motoyama T. Primary strumal carcinoid tumor of the ovary with multiple bone and breast metastases. J Obstet Gynaecol Res 2010; 36: 567-571.
- [5] Armes JE, Ostor AG. A case of malignant strumal carcinoid. Gynecol Oncol 1993; 51: 419-423.
- [6] Scvlltr E. Recent, progress in ovarian cancer. Hum Pathol 1970; 1: 73-98.
- [7] Davis KP, Hartmann LK, Keeney GL, Shapiro H. Primary ovarian Carcinoid tumors. Gynecol Oncol 1996; 61: 259-265.
- [8] Chou YY, Shun CT, Huang SC, Chuang SM. Primary ovarian carcinoid tumor. J Formos Med Assoc 1996: 95: 148-152.
- [9] Ayhan A, Yanik F, Tuncer R, Tuncer ZS, Ruacan S. Struma ovarii. Int J Gynecol Obstet 1993; 42: 143-146.
- [10] Dardik RB, Dardik M, Westra W, Montz FJ. Malignant strumal ovarii: two cases reports

Ovarian carcinoid goiter

- and a review of the literature. Gynecol Oncol 1999; 73: 447-451.
- [11] O'Connell ME, Fisher C, Harmer CL. Malignant strumal ovarii: presentation and management. Br J Radiol 1990; 63: 360-363.
- [12] Khadilkar UN, Pai RR, Lahiri R, Kumar P. Ovarian strumal carcinoid report of a case that matastasized. Indian J Pathol Microbiol 2000; 43: 459-461.
- [13] Matsuda K, Maehama T, Kanazawa K. Strumal carcinoid tumor of the ovary: a case exhibiting severe constipaion associated with PYY. Gynecol Oncol 2002; 87: 143-145.
- [14] Soga J, Osaka M, Yakuwa Y. Carcinoids of the ovary: An analysis of 329 reported cases. J Exp Clin Cancer Res 2000; 19: 271-280.
- [15] Ranchod M. Strumal carcinoid of the ovary. Cancer 1976; 37: 1913-1922.
- [16] Takemori M, Nishimura R, Sugimura K, Obayashi C, Yasuda D. Ovarian struma carcinoid with markedly high serum levels of tumor markers. Gynecol Oncol 1995; 58: 266-269.