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Case Report

Strumal Carcinoid Focus in Mature Cystic Teratoma in a Patient with Breast Cancer and Desire for Fertility Preservation

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ABSTRACT

Ovarian primary carcinoid tumors are very rare, accounting for 0.5-1.7% of all carcinoid tumors and 3% of ovarian teratomas. This type of tumor frequently arises from a mature cystic teratoma. The primary ovarian carcinoid tumors are divided into 4 major types, based on their histological characteristics: insular, trabecular or mucinous pattern. Strumal carcinoid refers to insular or trabecular pattern associated with struma ovarii. The strumal type is the most common histologic form, accounting for 40% of primary ovarian carcinoid tumors. Approximately 5% of carcinoid tumors of the non-mucinous type can be malignant. However, in the case of ovarian strumal carcinoid, the occurrence of metastasis is unusual. This article is based on a case of a 30-year-old nulliparous woman diagnosed with strumal carcinoid, who presented a right ovarian tumor suggestive of teratoma synchronous to a breast cancer. She was referred to our center for fertility preservation.

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Introduction

Ovarian strumal carcinoid (OSC) is an ovarian germ cell tumor composed of thyroid tissue mixed with a carcinoid tumor [1, 2]. The incidence of this type of tumor is around 0.3-1% of all ovarian tumors and 3% of all mature teratomas. The OSC frequently arises from mature cystic teratomas, which are the most common benign germ cell tumor in women, accounting for 58%, and between 27-44% of all ovarian tumors, including benign and malignant [3]. We present a case of an OSC diagnosed in a woman who had a right ovarian mass suggestive of teratoma.

Case Report

A 30-year-old nulliparous woman visited her gynecologist complaining of pelvic pain. Her relevant medical history included a left breast cyst excision at age 17, menarche at age 15, had regular menstrual cycle, and

no previous pregnancies. The gynecological examination revealed a suspicious right breast tumor, that was biopsied; and in the transvaginal ultrasound a heterogeneous and polylobed right ovarian mass of 54x60mm, suggestive of teratoma was found. The breast biopsy informed of an infiltrating ductal carcinoma, so a complete breast and axillary ultrasound was performed, in which suspicious axillary lymph nodes were detected and biopsied resulting positive for malignancy. Neoadjuvant chemotherapy was indicated prior to breast surgery.

The patient was referred to our hospital for fertility preservation after the diagnosis of breast cancer, and before starting neoadjuvant treatment. Prior to surgery, a hormonal analysis was performed, the result of which was: TSH 1,82 mU/L (normal), prolactin: 300 mU/L (normal). LH: 2, 6 U/L, FSH: 9,8 U/L, estradiol: 130 pmol/L and antimüllerian hormone: 1,11 ug/L.

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The patient underwent laparoscopic surgical treatment. During the procedure, two ovarian cystic tumors were visualized in the right ovary and another cystic tumor was detected in the left ovary. Bilateral excision of cystic lesions in both ovaries and cryopreservation of ovarian tissue was performed. According to the histological report, the cystic tumor located in the left ovary was found to be a mature cystic teratoma. As for the tumors in the right ovary, one of the cysts was a simple cyst and the other was a mature cystic teratoma with a focus of a strumal carcinoid.

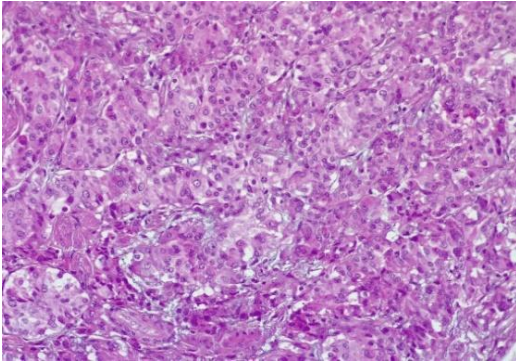


Figure 1: Carcinoid component forming solid nests and trabeculae.

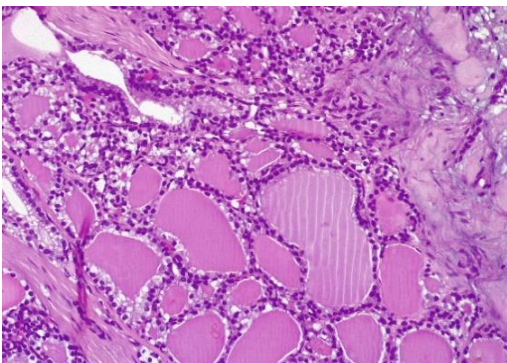


Figure 2: Thyroid tissue composed of follicles filled with colloid.

The strumal carcinoid lesion (Figure 1) was composed of thyroid tissue (Figure 2) intimately mixed with a solid component. The thyroid tissue showed immunohistochemical expression of cytokeratin 19 (Figure 3), thyroid transcription factor 1 (TTF1), and thyroglobulin. To rule out the suspicion of a solid papillary carcinoma, a BRAF mutational study was performed, the result of which was negative, excluding the diagnosis of thyroidal malignancy. The solid component showed trabecular pattern and expressed neuroendocrine markers like synaptophysin.

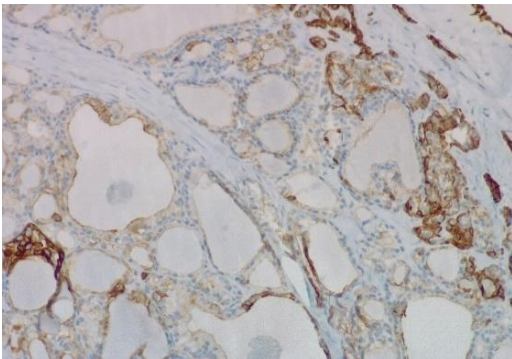


Figure 3: Immunohistochemical expression of CK 19 in thyroid follicle cells.

The patient received neoadjuvant chemotherapy treatment and underwent a right mastectomy with immediate repair, with dorsal width and right axillary lymphadenectomy. 18 months following the surgery, the patient is free of the disease.

Discussion

Carcinoid tumors are endocrine tumors that generally appear in the gastrointestinal or digestive system. Primary carcinoid tumors of the ovary are very rare, accounting for 0.5-1.7% of all carcinoid tumors and 3% of ovarian teratomas [4, 5]. The primary ovarian carcinoids are divided into different types, based on the histological characteristics of the tumor: insular, trabecular, mucinous and strumal carcinoid, when there is a mixed of thyroid tissue and carcinoid tumor [6]. Ovarian strumal carcinoids account for 40% of primary ovary carcinoids [7]. The OSC are tumors that originate from the endoderm and always contain thyroid tissue mixed with a carcinoid tumor. These ovarian tumors appear more frequently in peri and postmenopausal women [1]. The age of presentation varies between 21 and 77 years old. These tumors have a variable size that can reach up to 26 cm. The OSC are usually unilateral as in our case, although, there are bilateral cases described [8, 9]. In 10% of cases, the contralateral ovary may present a tumor, which is usually a cystic teratoma, as occurred in our case [8]. 60% of strumal carcinoids appear within dermoid cysts or solid mature teratomas [8].

Generally, patients who present these ovarian tumors do not have carcinoid syndrome, which typically presents symptoms such as flushing or diarrhea, or symptoms of thyroid hyperfunction [8, 10-12]. These patients may have an enlarged mass or pelvic pain or not present symptoms. In our case, thyroid function was normal, and the patient only reported pelvic pain. Robboy and Scully, in a review of 50 cases of ovarian strumal carcinoid found that 8% of women had elevated levels of steroid hormones and impaired thyroid function [8]. It is difficult to diagnose these tumors prior to surgery. Ca 125 may be elevated in these women, although it is also elevated in women who have ovarian cancer [13].

The histological study is essential for the diagnosis since thyroid tissue (adenomatous changes) and a component of a carcinoid tumor will always be detected. Immunohistochemistry is essential to complete the diagnosis. The carcinoid component is positive for synaptophysin and the thyroidal tissue expresses cytokeratin 19, TTF1 and thyroglobulin, as occurred in our case [14]. The main differential diagnosis must be made with the existence of a papillary thyroid carcinoma on the struma component of the teratoma [2]. In our case, it was ruled out by studying the BRAF mutation, which proved negative.

In young women the treatment is surgical and is based on the excision of the ovary and the ipsilateral fallopian tube; although in women of non-reproductive age, hysterectomy with double adnexectomy may be considered [5, 15]. Approximately 5% of carcinoid tumors of the non-mucinous type can demonstrate malignant behavior [5]. However, in the case of ovarian strumal carcinoid, the appearance of metastasis is unusual [1, 9]. In our case, the excision of the ovarian cystic lesions was performed, as the patient was young and wanted to preserve her fertility.

Conflicts of Interest

None.

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Declaration

The views expressed in the submitted article do not reflect an official position of the institution or funder.

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