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Treatment and follow-up in an asymptomatic malignant struma ovarii: A case report



Antoni Lluca^{a,b,*}, Yasmine Maazouzi^a, Jose Luis Herraiz^{a,b}, Mari Carmen Medina^a, Dolores Piquer^a, Blanca Segarra^a, Raquel Del Moral^a, Anna Serra^{a,b}, Guillermo Bassols^b

^a Multidisciplinary Unit for Abdominal Pelvic Oncology Surgery (MUAPOS), University General Hospital of Castellon, Spain

^b Dept. of Medicine, University Jaume I (UJI), Castellon de la Plana, Spain

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ABSTRACT

INTRODUCTION: Struma ovarii is a rare ovarian tumor, representing 0.5–1% of all ovarian tumors and 2–5% of ovarian teratomas. It is defined as an ovarian teratoma composed mostly of thyroid tissue. The symptoms are nonspecific, and the imaging studies can help in characterize the mass; however, the definitive diagnosis is usually given by the Pathologist. Classically, the treatment is the surgical resection of the ovarian mass, however there is no consensus regarding the follow-up.

PRESENTATION OF CASE: An asymptomatic malignant struma ovarii in a 43 year-old patient is presented. The diagnosis was postoperatively following a laparoscopic adnexectomy due to an apparently benign ovarian teratoma. The histopathology results revealed a mature ovarian cystic teratoma with papillary carcinoma with immunohistochemical characteristics suggesting a thyroid origin. Seeing that there was no thyroid affectation or metastatic disease, we decided a conservative management. A yearly follow-up with CT scan and tumor markers was performed. The endocrinologist also performed annual controls with thyroid ultrasound and serum tests. The patient has remained asymptomatic during these last four years.

DISCUSSION: There is little evidence in literature on the conservative management in cases with evidence of malignancy. If fertility preservation is desired, an unilateral oophorectomy could be performed, along with levels of serum thyroglobulin as a marker of relapse. Other authors claim for aggressive ovarian cancer surgery followed by a total thyroidectomy. There is still no established management for struma ovarii patients and the choice for a conservative or radical approach depends only on the professional decision.

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1. Introduction

The Struma ovarii is a rare ovarian tumor, representing 0.5–1% of all ovarian tumors and 2–5% of ovarian teratomas [1], that has been first described by Von Klden in 1895, followed by Gottschalk in 1899 [2]. It can occur in females of all ages, although with a predilection for women in their fifth and sixth decade of life [3]. It is defined as an ovarian teratoma composed mostly of thyroid tissue, occupying more than 50% of the ovarian mass. They are usually benign, although 5–10% of all cases are malignant, with the most common histological type being the papillary thyroid carcinoma (70%), and does not usually lead to metastatic disease (5–6%). [3,4]. When metastasis does occur, the struma ovarii behaves in a way similar to other primary ovarian cancer. In other words, it tends to cause peritoneal carcinomatosis, also affecting the contralateral adnexa

and/or epiplon. It can either present a lymphatic or haematogenous dissemination, spreading to pelvic/paraortic lymph nodes or bone, liver and brain respectively [5].

The symptoms of the Struma ovarii are similar to those described in other ovarian tumors, and also tend to be nonspecific (abdominal pain, abnormal menstrual cycles and vaginal bleeding) [6]. Thyroid dysregulation symptoms, such as hyperthyroidism, are rare, occurring only 5–8% of the cases. [7,8]. The imaging studies can help characterize the mass; however, the definitive diagnosis is given by the Pathologist.

Classically, the definitive treatment is the surgical resection of the ovarian cyst, however there is no consensus among practitioners regarding the follow-up. The adjuvant use of radioactive Iodine-131 in malignant struma ovarii remains controversial due to the scarcity of information and the lack of significant prospective studies, and is usually employed to treat recurrences and metastasized disease. According to the various studies, the management of the struma ovarii, following the removal of the pelvic mass, is individualized, depending on the patient and the medical team. [9,10].

* Corresponding author at: Av Benicasim, 12004, Castellon, Spain.
E-mail address: antonilluoca@gmail.com (A. Lluca).



Fig. 1. Macroscopic view of left ovarian mass.

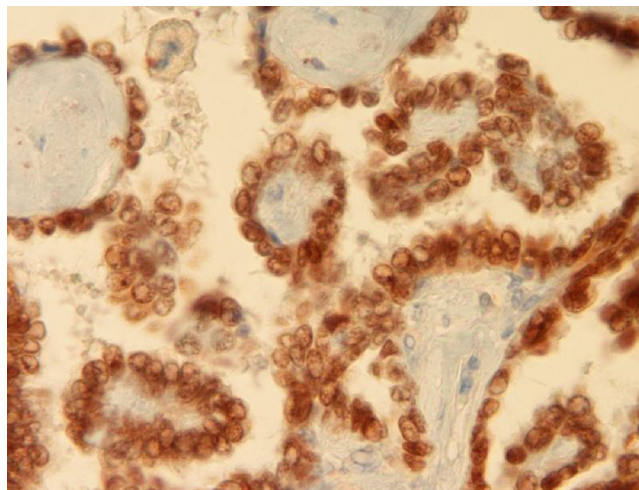


Fig. 3. Microscopic view of papillary thyroid carcinoma in struma ovarii: Thyroid Transcription Factor-1 (TTF-1) positive staining.

We report a case of an asymptomatic malignant struma ovarii in a 43 year-old patient diagnosed postoperatively following a laparoscopic adnexectomy due to an apparently benign ovarian teratoma and its management. This work has been done in line with the SCARE criteria [11].

2. Clinical case description

An ovarian mass was detected in an asymptomatic 43 year-old women, TPAL 2002, with no relevant familial or personal antecedents, in a routine gynecological check up. The preoperative ultrasound revealed no apparent signs of malignancy, and was described as a 7 cm solid solid-cystic mass compatible with cystic teratoma. The basal tumor markers were as follows: CEA 3.0 ng/ml; AFP 1.5 ng/ml; CA 125 17.9 UI/ml; CA 153 11.9 UI/ml; CA 199 91.8 UI/ml. The patient underwent a scheduled laparoscopic left adnexectomy with no incidents, and the mass was sent to pathology for analysis. There were no other pathological findings during laparoscopic exploration. The histopathology results revealed a « mature ovarian cystic teratoma with papillary carcinoma with immunohistochemical characteristics suggesting a thyroid origin (positive TTF-1, uncertain positive for thyroglobulin stain) (Figs. 1–4). Following these results, the patient was referred simultaneously to the gynecological-oncology and endocrinology consults for further studying. The diagnostic ¹³¹I scintigraphy revealed a normal pattern of thyroid uptake and the thyroid ultrasound was normal. The stimulated serum thyroglobulin, and antimicrobial antibodies levels were also normal.

Upon seeing that the rest of the results were negative, we decided on maintaining a conservative management with a gynecological, with high resolution gynecological ultrasounds, and endocrinology controls every 6 months during the first year fol-

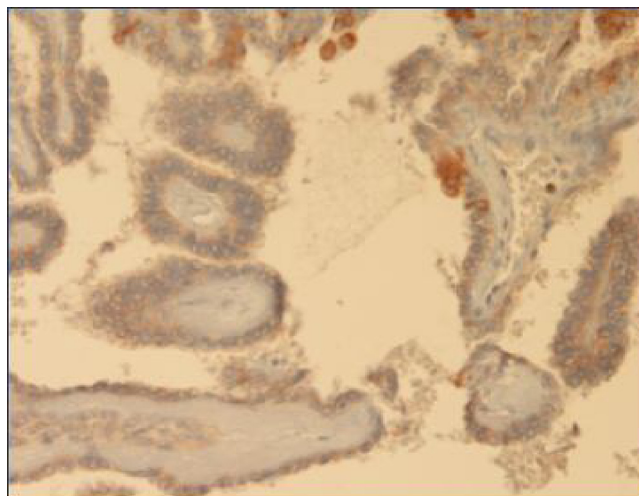


Fig. 4. Uncertain positive stain for Thyroglobulin.

lowing the diagnosis. A yearly follow-up with an annual Thoracic abdominopelvic CT scan and tumor markers (CEA, CA125, CA153, BHCG) were performed. The endocrinology department also performed annual checkups with thyroid ultrasound and serum panels (thyroglobulin, free T4, T3, TSH). The patient is currently 47 years-old, and has remained asymptomatic during these last four years, with no signs of relapse or thyroid affectation.

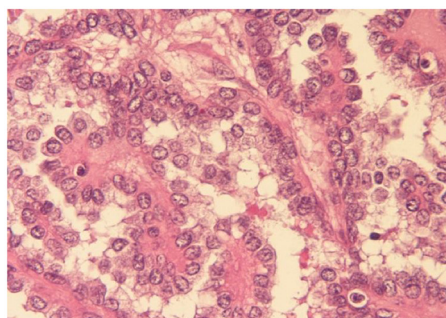
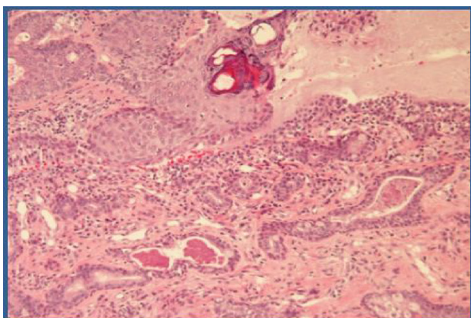


Fig. 2. Microscopic view of papillary thyroid carcinoma in struma ovarii.

3. Discussion

There is little evidence in literature on the conservative management in cases with evidence of malignancy. In cases of fertility preservation, a unilateral salpingo-oophorectomy could be performed, along with levels of serum thyroglobulin as a marker of relapse. In our case, a unilateral salpingo-oophorectomy was performed, not for fertility preservation, but because the mass was considered to be benign, nevertheless an endobag was used for the retrieval of the specimen as we usually do in suspicious adnexal masses and the patient consented only to a unilateral procedure. Some authors, especially in patients with no need for fertility preservation, claim that the cases of malignant struma ovarii requires an ovarian cancer surgical staging with pelvic washings, total hysterectomy with double adnexectomy and lymphadenectomy, as well as a total thyroidectomy followed by ¹³¹I radioablation, which would allow us to obtain the histological confirmation that the struma is of ovarian origin and prevent recurrences [12].

On the other hand, other authors consider that pelvic surgery is sufficient in patients with thyroid cancer confined to the ovary, and do not recommend prophylactic thyroidectomy and/or ¹³¹I radioablation. [13]. The management of thyroid nodules in these patients varies as well, including regular checkups with ultrasound and thyroglobulin levels or a total thyroidectomy with or without ¹³¹I radioablation therapy postoperatively [14,15].

In conclusion, there is still no established treatment algorithm for struma ovarii patients and the choice for a conservative or radical approach depends solely on the Professional in charge of the patient. There is a need for multicentric studies and further research in order to resolve the treatment and management dilemma.

Conflicts of interest

The authors declare no conflict of interest.

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Ethics approval and consent to participate

All procedures performed in studies involving human participants were in accordance with the ethical standards of the local ethics and research committee and followed the Declaration of Helsinki guidelines. 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.

All procedures performed in studies involving human participants were in accordance with the ethical standards of the local ethics and research committee and followed the Declaration of Helsinki guidelines. Written informed consent was required for collecting data.

Consent

Written informed consent was required for collecting data.

Author contribution

Study concepts: Antoni Lluca.

Study design: Antoni Lluca.

Data acquisition: Anna Serra, Blanca Segarra.

Quality control of data and algorithms: Raquel del Moral.

Data analysis and interpretation: Yasmin Maazouzi.

Manuscript preparation: Antoni Lluca.

Manuscript editing: Antoni Lluca, Dolors Piquer.

Manuscript review: Anna Serra, Jose Luis Herraiz.

Guarantor

Dr. Lluca is the guarantor of the paper.

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