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COMMUNICATION

A rare case of endometrioma and metastases of previously operated struma ovarii

Vincenzo Campo · Sebastiano Campo

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Abstract We present the first case of association between an ovarian endometrioma and peritoneal metastases of struma ovarii. In a 20-year-old woman, previously subjected to right adnexectomy at 12 years old for struma ovarii, during operative videolaparoscopy, we observed on left ovary the presence of endometrioma of 6 cm in diameter and on pelvic peritoneum and right lateral abdominal wall, red colored nodules of multiple sizes; one of these was visible on the diaphragm and one on the right anterior lobe of the liver. Definitive histological examination revealed endometriotic cyst, and the immunohistochemical analysis of peritoneal nodules revealed the presence of thyroid tissue.

Keywords Struma ovarii · Metastases · Endometrioma · Laparoscopy

Introduction

Mature cystic teratomas account approximately 20% of all ovarian tumors [1]. These tumors are composed by epithelial tissue and can include hair, skin, teeth, bone [2], and other tissues, and approximately 15% contain thyroid tissue [3]. To classify a teratoma as struma ovarii, it must contain thyroid tissue greater than 50% [4]. Struma ovarii account for about 2.7% of all ovarian teratomas [5]; it occurs at all ages, but most commonly affects women in the fifth and sixth decades of life [6]. It is often diagnosed during an exploratory laparotomy for pelvic mass, which is the most frequent presenting symptom. It is usually a benign condition even if occasionally a malignant transformation is observed in less than 5% of cases [6].

We present the first case of association between an ovarian endometrioma and peritoneal metastases of struma ovarii (Figs. 1 and 2) previously operated.

Case report

We report the case of a 22-year-old Caucasian woman who, at the age of 12 years, was admitted to her local hospital complaining of right lower quadrant pain. She underwent exploratory laparotomy and right salpingo-oophorectomy for an ovarian cyst. Uterus and left ovary seemed to be normal. The pathologic examination showed “struma ovarii” associated with mature cystic teratoma. A review of the pathology revealed no cellular atypia or malignant features. The postoperative course was free of complications, and the patient did not receive any further investigation during the following years. After 8 years, the patient was admitted with signs and symptoms of a left pelvic tumor. A pelvic ultrasound scan revealed a left ovarian lesion approximately 6 cm in diameter with a cystic component, suggestive of endometrioma. The levels of alfa-fetoprotein was 3.56 ng/ml, CA-125 was 6.5 UI/ml, CEA 0.1 ng/ml, Ca19–9 was 5 UI/ml, and she had normal thyroid parameters. The patient was taken to the operating room for an operative laparoscopy. Surgical exploration could find neither pelvic nor Douglas implants suggestive of peritoneal endometriosis or ascites. However, multiple pink nodules were observed (diameter between 1.0 and

V. Campo (✉) · S. Campo
Department of Obstetrics and Gynecology,
Catholic University of Sacred Heart,
L.go Gemelli, 8,
00168 Rome, Italy
e-mail: vincecampo76@gmail.com

Fig. 1 Peritoneal metastases of struma ovarii and endometrioma on left ovary

3.0 cm) on the pelvic peritoneum, right lateral abdominal wall, diaphragm, vesical plica, and liver. Cystectomy with spillage of like chocolate liquid during ovarian mobilization and multiple biopsies of the nodules on the anterior abdominal wall and vesical plica were performed. We removed cystic capsule by endobag. The patient was discharged the day after surgery. On histopathological examination, the ovarian cyst was found to be an endometriotic lesion, and the nodules were made up of fibrous tissue with follicular spaces lined by thyroid cells with grooves and clear nuclei. Immunohistochemical study of the abdominal nodules showed strong expression of thyroglobulin (Tg), suggesting a diagnosis of recurrent struma ovarii with features consistent with the follicular variant of papillary thyroid carcinoma.

Discussion

Only three cases of simultaneous presence of endometriosis, and mature cystic teratoma of the ovary are described in literature. Ferrario [7], in a 23-year-old woman submitted to laparotomy for bilateral salpingo-oophorectomy because of pelvic mass, described the simultaneous presence in the right ovary of an endometriotic cyst and a dermoid cyst. Caruso and Pirrelli [8], in a 28-year-old women with bilateral ovarian dermoid cysts, show in the left ovary an endometrioma. Frederick et al. [9] reported a case of a young woman with bilateral dermoid cysts and endometriotic deposits in the pelvis.

To our knowledge, we report the first case of association between ovarian endometrioma and peritoneal metastasis of a recurrent struma ovarii 8 years after annessectomy and classified as benign at histopathology and with no sensible explanation to justify the malignant transformation of the ovarian tissue and the resultant metastatic spread.

Our case is quite similar to Vadmal's [10], who described a case of recurrent struma ovarii with malignant transformation 6 years after abdominal hysterectomy and bilateral salpingo-oophorectomy in a 51-year-old woman operated for a left ovarian mass diagnosed at histopathology as a pure struma ovarii without cellular atypia or malignant features.

The interest of our case is due to the occasional finding, during operative laparoscopy for other disease, of a recurrent metastatic struma ovarii after 8 years.

The real incidence of malignancy in struma ovarii is difficult to assess due to the rare nature of this condition. An incidence of 0.1–0.3% has been quoted in the literature [11]. Even if many authors [12] consider as benign the presence of peritoneal metastasis of thyroid tissue, others suggest that any struma ovarii exhibiting metastatic behavior should be regarded as malignant [4]. Metastasis of malignant struma ovarii is seen in approximately 5% of cases [13], even if a recent review [14] demonstrated a higher metastasis rate of 23%, which was mainly intra-abdominal, although blood-borne metastasis can occur in the liver, brain, lung, bone, and the contralateral ovary. In our patient, we found metastatic implants on pelvic peritoneum, right lateral abdomen wall, diaphragm, and on the right anterior lobe of the liver. It is doubtful if these lesions are due to spillage during the first operation, even if a laparotomic adnexectomy, which presents a lower risk of spillage, was performed.

Most of the cases of malignant struma ovarii are subclinical. Approximately 5–8% of patients have clinical hyperthyroidism [6]. In our experience, after surgery, the patient had normal thyroid parameters (TSH 1.65 mcg/ml, FT3 2.1 pg/ml, and FT4 2.8 pg/ml) but presented higher levels of thyroglobulin (305 ng/ml); at ultrasound examination, no thyroid nodules were found.

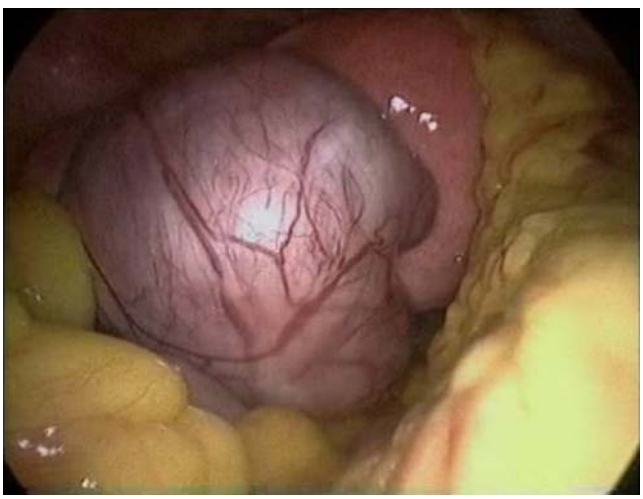

Fig. 2 Left ovarian endometrioma

The treatment of malignant struma ovarii remains controversial. Since metastatic struma ovarii is similar to metastatic thyroid carcinoma, 131I therapy represents an ideal way to treat both diseases [14]. However, as in patients with differentiated thyroid carcinoma, before 131I treatment, it is necessary to remove the thyroid gland completely to exclude a primary thyroid carcinoma with subsequent ovarian metastasis.

According to others [10], and considering the presence of peritoneal metastasis, to increase the uptake of 131I by the metastatic nodules, we suggest to the patient to have a total thyroidectomy followed by radioiodine ablation. Furthermore, we propose to the patient an ovarian tissue cryopreservation and GnRHa administration before 131I treatment in order to preserve fertility. We believe that these means should be offered to all young women diagnosed with cancer.

In conclusion, our case shows that even if malignant transformation is uncommon, close follow-up is advised after surgery, to identify precociously the progression of the disease and its potential transformation to a higher grade neoplasm; at least a follow-up period of 10 years is recommended [15], consisting of sequential thyroglobulin measurements and total-body scintiscanning with 131I if recurrence is suspected.

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