

SHORT COMMUNICATIONS AND CASE REPORTS

Late struma ovarii diagnosis many years after total thyroidectomy. A rare entity

C. Iavazzo¹, G. Vorgias¹, A. Psarrou¹, I. Lekka², M. Katsoulis¹

¹Department of Gynecology and ²Department of Pathology, "Metaxa" Cancer Hospital, Piraeus, Greece

Summary

We present the case of a 53-year-old lady with incidental diagnosis of struma ovarii 10 years after total thyroidectomy due to papillary thyroid carcinoma. The disease was diagnosed owing to high levels of serum thyroglobulin.

The patient underwent exploratory laparotomy with total hysterectomy and bilateral salpingo-oophorectomy. Close follow-up was proposed and her prognosis is excellent.

Key words: diagnosis, prognosis, struma ovarii, thyroidectomy, treatment

Introduction

Struma ovarii is a rare germ-cell tumor which is defined as an ovarian mass containing thyroid tissue as the predominant cell type (over 50% thyroid tissue in the resected ovarian specimen). 5-12% of such tumors are functional and 95% are benign [1-3]. Struma ovarii usually occurs as a part of teratoma or cystadenoma. The diagnosis is often incidental. Herein we present a case of struma ovarii that was diagnosed due to high levels of serum thyroglobulin.

Case presentation

A 53-year-old lady with a history of total thyroidectomy for papillary thyroid carcinoma 10 years ago presented at the Endocrinology department of our hospital for follow up. Endocrinological examinations showed normal levels of fT3 (3.2 pg/ml, range 1.8-4.5), fT4 (14.9 pg/ml, range 7-20), TSH (2.7 µIU/ml, range 0.3-5) and elevated levels of thyroglobulin (173 U/L, normal up to 40). She underwent further examinations according to the endocrinologist's suggestions in order to identify the source of the ectopic production of thyroglobulin. Plain chest radiography

revealed no particular findings. CT scan of the neck and chest were normal except the picture of the total thyroidectomy. An enlarged right ovary was found during clinical pelvic examination. No complaints were mentioned from the patient. Abdominal ultrasound revealed a 49×30 mm right cystic ovarian mass with vascularization. Abdominal CT scan showed a complex, cystic mass of the right ovary with normal contour measuring 5 cm in its largest diameter. The tumor markers CA125 and CA19-9 were within normal range. The patient underwent exploratory laparotomy with total hysterectomy and bilateral salpingo-oophorectomy. An intraoperative frozen section of the right ovary was negative for malignancy. The histological examination of the specimens revealed a benign thyroid parenchyma with enlarged follicles filled with colloid, a picture compatible with struma ovarii of the right ovary. The left ovary, fallopian tube and uterus were without significant pathology. The thyroglobulin staining by immunoperoxidase method confirmed the diagnosis. Ten days postoperatively, fT3, fT4, TSH and thyroglobulin were within normal limits. No further treatment was necessary for our patient. We proposed to her close follow up every 3 months for the first year and then every 6 months for the following 2 years.

Discussion

Struma ovarii is a highly specialized form of mature ovarian teratoma. It was first described by von Kalden and Gottschalk in 1895 and 1899, respectively [4]. The peak incidence of struma ovarii tumors is in the 5th decade [5], as it happened in our case. Such tumors could present as an abdominal or pelvic mass with or without CA125 elevation. However, even in the malignant forms of the disease, struma ovarii is rarely associated with elevated CA125 or ascites. Only 6% of struma ovarii cases are bilateral with the tumor arising more commonly in the left rather than the right ovary [6]. In the majority of cases, the tumor's diameter does not usually exceed 10 cm [7]. The diagnosis could be suspected only by the functional changes of the ectopic thyroid tissue, like in our case. Our patient had no symptoms or clinical signs of thyroid hormone imbalance apart from the increased levels of serum thyroglobulin.

The vast majority of struma ovarii tumors are benign; malignancy is rarely seen and has an incidence of approximately 0.1-0.3% of all ovarian teratomas [8]. According to Ayhan et al. [9], pure struma ovarii consists only of thyroid tissue; mixed struma of other teratomatous elements with predominantly thyroid tissue and malignant struma are characterized by cellular atypia, mitotic activity, vascular or capsular invasion or metastases [9].

Although many could argue that laparoscopic excision of this tumor might be an option, we preferred laparotomy due to the age of our patient. We believe that total abdominal hysterectomy with bilateral salpingo-oophorectomy is a reasonable option in post- or perimenopausal patients, whereas conservative treatment with a unilateral laparoscopic or laparotomic oophorectomy could be used as a fertility-sparing option.

Histologically, the tumor is usually characterized by a dominant growth of thyroid tissue within the ovary. The gross characteristics (color and consistency) imitate those of thyroid tissue but often cystic lesions are found. The ectopic thyroid tissue may resemble any pathological change of the thyroid such as diffuse or nodular hyperplasia (which is characterized by hyperthyroidism), thyroiditis, carcinoma and malignant lymphoma [10]. Usually, like in our case, immunohistochemical staining for thyroglobulin could confirm the diagnosis [11]. Furthermore, it should be mentioned that in our case the diagnosis was also

confirmed by the postoperative reduction of the plasma thyroglobulin levels.

Many could argue that such a tumor might be a metastasis of the previous papillary thyroid carcinoma, however there are very few cases of such metastasis in the literature [12,13]. In our case, such a hypothesis was rejected as the histological examination revealed no sites of papillary formations, nuclear abnormalities, mitotic activity or vascular invasion. Usually, such tumors are benign and the prognosis is excellent. Follow up should be close in the first 3 years and could be carried out with the use of serum thyroglobulin as a "tumor marker" in both benign or even malignant forms [8].

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