Struma ovarii: Clinical presentations of an uncommon tumor

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ABSTRACT

Introduction: Struma ovarii is a rare benign tumor which represents less than 3% of ovarian teratomas. Case Series: We report three cases of different clinical presentations of struma ovarii, to attempt to define the clinical features and characteristics of this tumor with respect to ultrasonographic findings, histological characteristics and way of optimal management. Conclusion: We recommend that, without any confirmation of malignancy, conservative management should be chosen and laparoscopic way should be preferred if possible.

Keywords: Struma ovarii, Thyroid, Ovarian neoplasm

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doi:10.5348/ijcari-2012-04-106-CS-3

INTRODUCTION

Thyroid tissue is observed not uncommonly in 5–15% of dermoid tumors. Struma ovarii is a teratoma defined by the presence of thyroid tissue in more than 50% of the tumor [1]. Struma ovarii comprises 1–4% of benign ovarian teratoma [2–4].

It is a benign condition, but occasionally malignant transformation is observed in about 5% of cases. However, due to rarity of this type of tumor there has been a paucity of data in the past literature pertaining to diagnosis and treatment of this tumor.

We report three cases of different clinical presentations of struma ovarii, to attempt to define the clinical features and characteristics of this tumor with respect to ultrasonographic findings, histological characteristics and way of optimal management.

CASE SERIES

Case 1: A 17-year-old, female, gravida 0, menarche at 13, consulted for irregular menstruations. Her past medical history was not significant. Physical examination revealed a palpable mobile mass in the right lower abdomen. The patient was admitted for further investigations. Ultrasonography (USG) of the pelvis and abdomen revealed a large pelvic cyst, arising from the right ovary, sized 16.3 cm in largest dimension, with heterogeneous echogenicity and septa. The uterus and left ovary were normal. The patient had no ascites. (Figure 1). The patient’s serum CA-125 level was 11.85 IU/ml (normal level <35 IU/ml), AFP was 1.41 µg/l (normal level <10 µg/L), TSH was within normal range (3.02 IU/ml, normal level: 0.15–5 IU/ml). Routine biochemistry was unremarkable. The patient underwent a laparoscopic surgery. The uterus was in normal size

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and shape, as well as the left ovary. A 20x16 cm cyst, with regular limits had replaced the right ovary. There was no evidence of intra-peritoneal spread of disease or retroperitoneal adenopathy. Laparoscopic conversion was necessary and trans-parietal cystectomy was performed. The final pathology revealed right struma ovarii containing exclusively benign thyroid tissue confined to the ovary. Immunohistochemistry was not performed. The patient recovered uneventfully and was discharged home on the fifth postoperative day. Following up three months after her surgery, she had normal USG control and normal thyroid function tests and CA 125 levels.

Case 2: A 31-year-old woman was admitted to our department for evaluation and management of a persistent pelvic mass detected on a routine ultrasound examination. The patient had undergone laparoscopic exploration in 1998 for a two year infertility investigation. She was gravida 0. The patient had no other notable past medical history. Clinical examination was normal, and no pelvic mass was palpable. Pelvic ultrasound showed normal uterus and right ovary in addition to a heterogeneous echogenic mass of five cm in the left adnexa. Magnetic resonance imaging found a tissue mass of 5.4x5 cm in the left adnexa, well delineated with a hyper-signal in T1 and T2 and marked enhancement of the mass in Gadolinium enhanced T1 weighted images (Figure 2). Laboratory data were normal with a normal level of CA-125 (5.38 IU/ml). The patient underwent laparoscopic intraperitoneal cystectomy. Peroperative exploration showed a left ovarian mass, well delineated, with no septa or vegetations. Histopathologic examination showed a struma ovarii with benign thyroid cells. Immunohistochemistry was performed and showed the presence of thyroglobulin (+) cells. Postoperative period was uneventful and patient was discharged home on the second postoperative day. Patient was well for three months with normal CA-125 level. The patient was pregnant within a year.

Case 3: A 45-year-old, female was admitted for persistent metrorrhagia, refractory to medical treatment (nomégestrol, 5 mg/day, for three months). The patient was diabetic under oral treatment, and had hypertension under monotherapy. She was gravida 4 para 4, and had never had surgery. She were using IUD for contraception. Clinical examination as well as routine laboratory exams was normal. Cervix was macroscopically normal. Exploratory hysteroscopy after

Figure 1: Struma ovarii in ultrasonography. A) Large pelvic cyst, replacing the right ovary, sized 16.3 cm in largest dimension, with heterogeneous echogenicity and septa, B) Absence of doppler signal.

Figure 2: Struma ovarii in MRI. Tissue mass of 5.4x5 cm in in left adnexa, well delineated with a hyper-signal in T1 and T2, and marked enhancement of the mass in Gadolinium enhanced T1 weighted images. (A-T1 weighted image, B-T2 weighted image).
IUD removal was also normal. Pelvic ultrasound found a normal uterus with normal-sized ovaries. No cystic mass was detected. The patient underwent vaginal hysterectomy with bilateral oophorectomy. On peroperative macroscopic both ovaries as well of the uterus were normal. Gross histopathologic examination showed a normal endometrial aspect, as well as a normal left ovary. However, surprisingly, the right ovarian cut surface was multicystic with cysts varying from 1–2 cm in diameter filled with a brown jelly-like gelatinous material. Microscopy showed benign colloid filled thyroid follicles with no cytological feature of malignancy and histology confirmed the diagnosis of struma ovarii of the right ovary (Figure 3). Immunohistochemistry was not performed. Post-operative follow-up was uneventful.

![Figure 3: Pathologic Aspect of struma ovarii, A) Thyroid tissue in direct contact with the ovarian parenchyma, B) Thyroid follicles at high magnification.](image)

**DISCUSSION**

Struma ovarii comprises 1–4% of all dermoid tumors of the ovary, and very rarely presents in a malignant form, occurring in 0.3%–5% of all struma ovarii tumors [2–4].

The search of our database in the Hospital of Ben Arous starting from 2005 to date has found only three cases of struma ovarii among a total of 35 dermoid tumors. Therefore, the three struma ovarii patients represented 8.57% of all dermoid patients, among whom none were diagnosed as malignant struma ovarii. These figures are quite different from previously reported data [2–4], particularly our high incidence of struma ovarii among all teratomas, which may be attributed to the rarity of struma ovarii and hence the very low number of cases in previous literature.

Struma ovarii usually presents after age of 40 years and the peak age of incidence is in the fifth decade [5]. This tumor is present in only 17.6% of cases in patients under 30 years [6]. In our study, we have noticed a very early struma ovarii, discovered at the age of 17 years, and only one patient out of the three cases was above 40 years of age. This seems in contradiction with the past data.

Clinical symptoms previously reported due to the presence of a struma ovarii are very diverse, such as lower abdominal pain, palpable lower abdominal mass, abnormal vaginal bleeding, ascites, hydrothorax, elevated thyroid function and rarely thyroid tumors [7, 8]. Previous reports have shown that up to 47.1% of patients with struma ovarii are without symptoms, or are accompanied by non-specific symptoms that are similar to other ovarian tumors [6]. The results of this group seem to be in agreement with the above observation. In fact, in our cases, one patient out of the three was symptomatic and consulted because of a growing palpable mass of the lower abdomen. The two other patients had no symptoms and the tumor was discovered in one case by a routine USG and in the other case, the struma ovarii was a histological diagnosis. It has been recommended in a previous study that thyroid function tests have to be conducted in the presence of symptoms and signs related to thyroid dysfunction [9]. The incidence of thyroid hyperfunction has been reported to be 5–8% of patients with struma ovarii [10]. In our three cases, there were no patients with hyperthyroid symptoms and therefore, in only one patient thyroid function tests were done. It cannot be fully explained why there were no cases with thyroid hyperfunction within our group of patients especially with the two large-sized tumors. CA-125 is the most important clinical marker for the diagnosis, treatment and follow-up of epithelial ovarian cancer. Infrequently, benign struma ovarii is associated with elevated CA-125 levels [11]. In our cases, the two patients carrying large ovarian tumors have had pre- and post-operative CA-125 dosage, which have shown normal levels in both cases. The third patient who has been diagnosed only after histological exam, did not have any marker studies done, since the final pathology concluded to a benign tumor.

Ultrasoundography permits the diagnosis of the ovarian masses, but orient to the diagnosis of struma ovarii in about 11.8% of cases only [6]. In our cases, we clearly saw the difference in USG between patients: In one patient, USG was compatible with a benign ovarian cyst, in the other the USG was suspicious with heterogeneous echogenicity and presence of septa, and in the third case the two ovaries were normal on USG. In struma ovarii MRI typically shows a multilocular cystic mass with variable signal intensity within loculi. Some loculi show low intensity on T1 weighted images and very low intensity on T2 weighted images, corresponding pathologically to gelatinous colloid material [12]. Only one of our patients had MRI. Imaging findings in her case were not typical. Thus, struma ovarii does not have definite clinical or imaging characteristics that differentiate it from other ovarian tumors, with the exception of hyperthyroid symptoms if present.

The final diagnosis of struma ovarii is based on pathology examination of the resected cyst/ovary, which permits at the same time, to confirm or exclude malignancy. Extensive grossing is required to rule out any other component before labeling it as monodermal teratoma. Struma ovarii typically consists of normal-appearing thyroidal tissue composed of thyroid follicles of various sizes and often is associated with mature
cystic teratoma. Histologically, struma ovari can also resemble thyroid adenoma of follicular, fetal, or embryonal type or thyroid carcinoma [13]. About 5% of struma ovari are malignant [14]. Clinical features are quite similar, and malignancy should always be suspected, especially when the ovarian tumor is associated to ascites, elevated CA-125 levels, or sometimes a “pseudo-Meigs” syndrome [2]. None of our patients had malignant struma ovari.

Therapy for benign struma ovari is surgical resection. The optimal way of management is, however, very controversial [15–16]. The very suspicious clinical features and peroperative findings of the tumor add to this controversy. For women desiring further pregnancies, conservative management which consists of a simple cystectomy or a unilateral oophorectomy, seems to be the optimal treatment [15, 16]. Although infrequent, there have been reports of cases where women have had successful pregnancies after such conservative procedures in malignant struma ovarii [17]. In our cases, because of their young age, the two patients with diagnosed ovarian tumors have had simple cystectomy, one by laparoscopy and the other by laparotomy. One of the two patients has been pregnant in less than one year after surgery. We recommend that, if malignang is not confirmed, conservative management should be chosen and if possible, laparoscopic method should be preferred.

CONCLUSION

Larger studies are needed to determine an optimal diagnosis, management and follow-up protocols for this rare ovarian tumor.

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Author Contributions

Zied Khediri – Substantial contributions to conception and design, acquisition of data, Drafting the article, revising it critically for important intellectual content, Final approval of the version to be published

Chaouki Mbarki – Substantial contributions to conception and design, acquisition of data, Drafting the article, revising it critically for important intellectual content, Final approval of the version to be published

Anis Ben Abdelaziz – Substantial contributions to conception and design, acquisition of data, Drafting the article, revising it critically for important intellectual content, Final approval of the version to be published

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Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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