Bilateral struma ovarii: a case mimicking an ovarian neoplasm

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Summary

We present a case of bilateral struma ovarii which developed postoperatively and was histopathologically diagnosed after the patient was hospitalized for investigation and treatment of tumoral anexal bilateral formations. There was no evidence of clinical malignancy or metastases. Data from the literature, together with histopathologic, diagnostic and therapeutic aspects of the disease were checked again taking into account the scarcity of this lesion, especially bilaterally.

Key words: Struma ovarii; Germ cell tumors; Bilateral.

Introduction

Struma ovarii was first described in 1895 by Von Kalden and is extremely rare with only 150 cases reported in literature [1]. It is an ovarian teratoma made up entirely or predominantly by thyroid tissue (representing > 50% of tumor) with follicles of different sizes with a greenish or brownish yellow gelatinous content. It represents 0.85%-1.3% of all ovarian tumors. Most patients are asymptomatic, which is why the diagnosis is hard to establish preoperatively and is usually randomly discovered when magnetic resonance imaging (MRI) is performed [2, 3]. Struma ovarii represents an unusual tumor with extremely rare malignent changes occurring in less than 1% of cases [5] up to 5% [9], and in 90% of cases when it was situated unilaterally [10]. Most often the diagnosis has been made postoperatively by using macroscopic criteria with histopathologic confirmation [4-6].

Case Report

We present a case of bilateral struma ovarii histopathologically diagnosed from the surgical material. Based on data from the literature we tried to find the optimal therapy for this disease which was associated with uterine fibroleiomyoma and metrorragia. A 41-year-old woman, was hospitalized at the Department of Gynecology of the Clinical Hospital "Filantropia" of Craiova, with a diagnosis of bilateral anexal tumoral formations, uterine fibroma, and metrorragia. Concerning the history, the patient presented with metrorragia and for about two months she had had diffused abdominal pain, abdominal distension, constipation and irregular menses. There was no objective or subjective data to confirm thyroid hyperfunction. A local examination revealed a slightly increased abdomen, especially in the lower part, slightly diffused when superficially palpated, with maximum intensity in the same area. A gynecologic exam found a moderately increased uterus and bilateral adnexal tumoral formations. Abdominal pelvic echography revealed two ovarian cystic formations, one was pluriseptate, hypoechoic and/or hyperechogic with small nodular fibroma and endometrial thickness of 0.8 cm. Tumor markers were normal: CA-125 measurement presented a value of 19 UI/ml, while hCG, βhCG, and AFP revealed normal values. Laparotomy was performed and intraoperatively a slightly increased uterus was established with a deformed surface due to the presence of some small fibromatous nodules; the left ovary was increased to 6.5 x 5.5 x 4 cm, with variable consistency to which the left salpinx was closely joined. The right ovary was increased 7.5 x 5.7 x 4.2 cm and also had variable consistency, a globulous appearance, with the right salpinx joined to the right ovary. The two ovaries were sent for frozen section and the result was nonmalignant.

We decided to perform total hysterectomy with bilateral salpingo-oophorectomy. The material was processed and sent to the Laboratory of Pathological Anatomy of the same hospital. After the macroscopic exam, the next histopathologic technique was to parafin embed the microscopic pieces which were stained by H&E and Van Gieson. Macroscopic examination of the surgically removed piece helped us to reveal an almost normal sized uterus 10.55 x 8.4 x 6.3 cm, diminished consistency, a brown-grayish color, with the external surface slightly deformed due to the presence of some small nodular formations. On the section surface formations of variable sizes and white-necreous color were present with a slightly thickened and bleeding endometrium (0.8 cm). The left ovary was adherent to the salpinx and ovary, without any remarkable macroscopic changes. The left ovary 6.5 x 5.5 x 4.5 cm in size with a globulous appearance, yellow-greyish color, and variable consistency from fluctuant to a hard. On a section of the surface of a polymorphous aspect, with many cystic cavities ranging in diameter from 0.8 to 0.3 cm, some with serous or gelatinous contents, as the white of an egg, yellowish-orange, brownish or pistachio color. The right ovoid-shaped ovary had an irregular external surface, variable consistency presenting multiple cystic formations with a serous but also gelatinous content of variable color from pistachio to red-brownish on a section presenting a white-necrous nodular formation in the center. Microscopic examination of the uterus revealed a simple hyperplasia aspect without atypia at the level of the endometrium, leiomyofibroma at the level of the uterine body and an aspect of bilateral struma ovarii. Microscopic examination of the seriate section from both ovaries presented ovarian histologic structures containing thyroid glandular tissue with variable fibrotic areas, inflammatory infiltrates, erythrocytes here and there, and cystic spaces lined by cuboidal or flat unstratified epithelium, with an eosinophilic content (Figures 1-6).

Revised manuscript accepted for publication August 18, 2008

Fig. 1

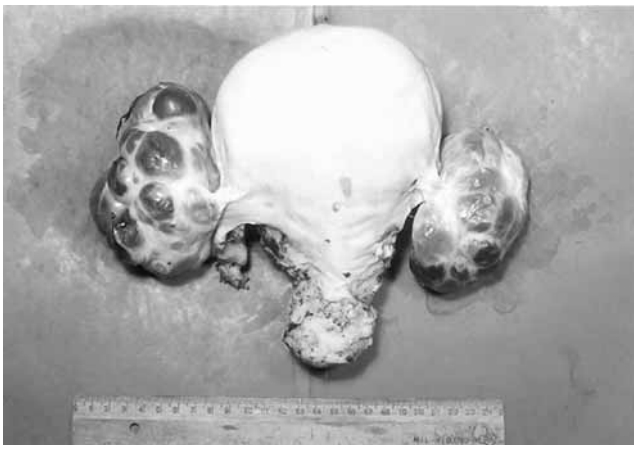

Fig. 2

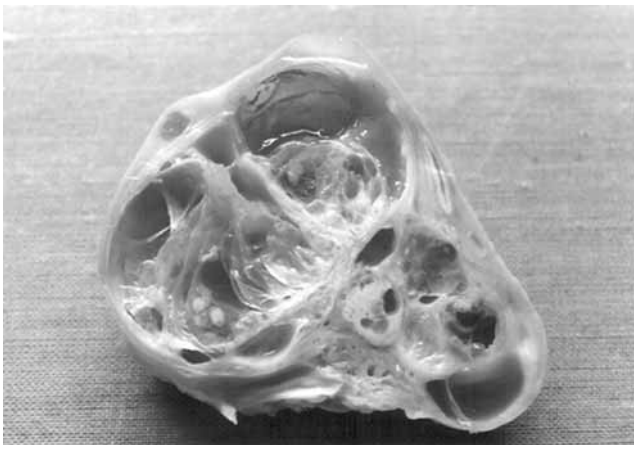

Fig. 3

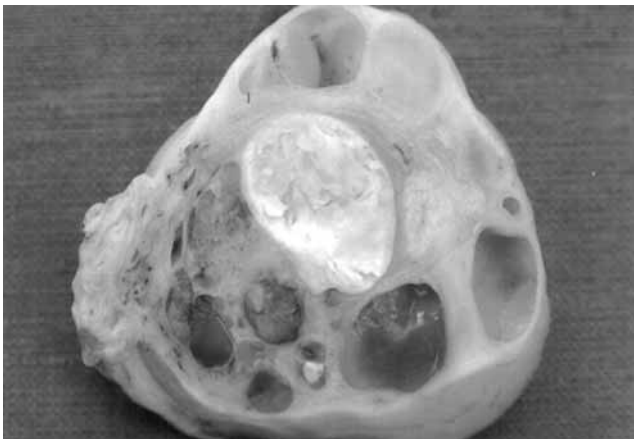

Fig. 4

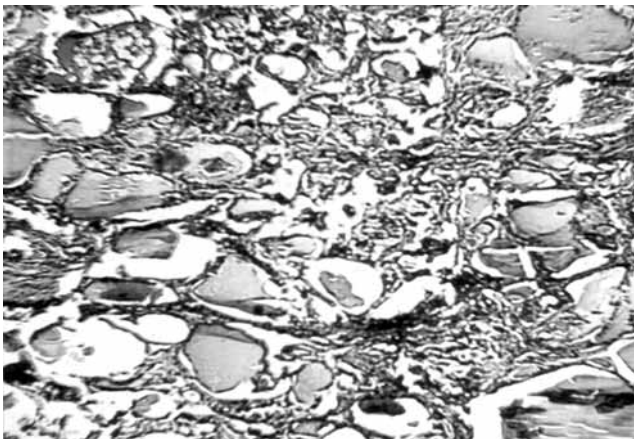

Fig. 5

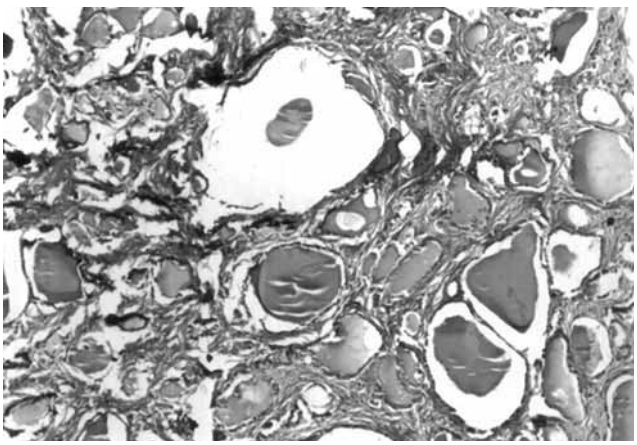

Fig. 6

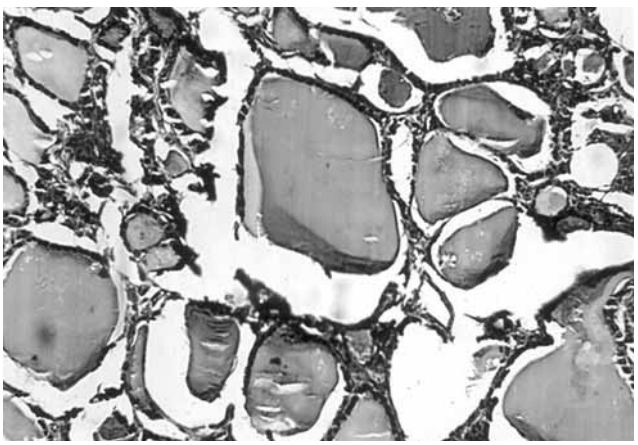

Figure 1. — Aspects of macroscopic piece of total hysterectomy with bilateral salpingo-oophorectomy.
Figure 2. — Longitudinal sections of left ovary - macroscopic image.
Figure 3. — Longitudinal sections of right ovary - macroscopic image.
Figure 4. — Colloid-filled follicles varying in size with fibrous stroma representing struma ovarii (Van Gieson 10 x 10).
Figure 5. — Aggregates of dilated "thyroid follicles" (H&E 10 x 10).
Figure 6. — Struma ovarii with predominantly follicles lined by cuboidal or flat epithelial cells (Van Gieson 10 x 20).

Discussion

Struma ovarii is an unusual tumor which rarely becomes malignant. It is dominanted by increased thyroid tissue in an ovarian teratoma. Teratomas are considered as tumors made up of tissues originating from all three germ cell layers [7, 8]. Ovarian teratomas are imature and mature. Imature teratomas are most often malignant representing about 1% of all ovarian cancers [9, 10]. Mature teratomas are made up of adult type tissues derived from the three embryo sheets. Most are cystic representing about 25% of ovarian tumors. Mature teratomas are generally benign, and rarely do they

become malignant but they can suggest a malignant appearance by their macroscopic aspect [11, 12]. Struma ovarii is the most frequent monodermal teratoma, a highly specialized form of a mature ovarian teratoma with maximum incidence in the fifth decade of life. Some cases present hyperthyroidism signs and often Meigs syndrome [13]. Macroscopically, struma ovarii is of a thyroid consistency and thyroid tissue aspect, predominantely solid and gelatinous [14]. Rarely does it appear associated with a dermoid cyst, or as a component of a stromal carcinoid [15]. Microscopically, stroma look like thyroid tissue to which any thyroid change, nodular or diffuse hyperplasia, but rarely carcinomas can be associated. Malignant transformation of the thyroid tissue may be papillary, follicular or mixed and it can include elements of mucinous cystadenocarcinoma [16]. In difficult cases, immunohistochemical stainings for thyroglobulin, triiodothyronin (T3) and tyroxine (T4) can confirm the diagnosis.

Conclusions

Because struma ovarii is extremely uncommon, each case must be individually treated. The diagnosis of struma ovarii should be suspected when a multicystic ovarian tumoral formation with a brownish colored gelatinous content – associated or not to clinical thyroidtoxicosis – is found. Surgical treatment is suggested. If it is associated with a thyroidtoxicosis, treatment of thyroid substitution should be performed but, in our case, after the surgical intervention the patient had a thyroid scan that was normal with normal thyroid function.

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