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Benign Ovarian Goiters: A Case Report

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Abstract **Case Report**

***Objective of Study*:** Ovarian goiter is a single-tissue teratoma made up of thyroid tissue representing 0.85 to 1.3% of ovarian tumors. The objective of our study is to report on a case of an ovarian goiter accompanied by a complete analysis of the literature in relation to the case. ***Conclusions*:** An ovarian goiter is a rare pathology, often asymptomatic, and difficult to diagnose. Few series are described in the literature. In our case, we propose a model for the management of a benign ovarian goiter. Surgical revision and adjuvant treatment is indicated in cases of malignancy of the ovarian goiter.

Keywords: Ovarian goiter, thyroid tissue, asymptomatic.

INTRODUCTION

Teratomas are germ cell tumors made up of tissues derived from the embryonic layers (ectoderm, endoderm and mesoderm). Some are composed exclusively or mainly of a single tissue defining the group of "monodermal teratomas", among which ovarian goiter is described. The presence of a thyroid contingent within an ovarian teratoma represents 5 to 15%; it is the majority, or even exclusive, in 0.85 to 1.3% of cases [1, 2]. Described for the first time by Von Kalden in 1895 and by Gottschalk in 1899, ovarian goiter or "struma ovarii" is the most common monodermal teratoma of the ovary [2–4]. We report a case of ovarian goiter treated in our institution.

CASE REPORT

This is the case of a 61-year-old patient, without any notable pathological history, multiparous and postmenopausal, who was admitted as part of the followup of an asymptomatic ovarian cyst that had been present for 3 years. She consulted our institution following minimal intensity left pelvic pain without other associated signs.

The clinical examination revealed a normal-appearing cervix, tilted on visualization with a speculum; soft to the vaginal touch; without perception of a lateral uterine mass or lateral uterine tenderness.

On ultrasound we have a solido-cystic image in the left lateral uterine, taking up the entire screen measuring 9x7 cm classified IOTA B4. Note the non-visualization of the ovaries.

A pelvic MRI carried out reveals a left ovarian cystic formation measuring 90x74x70mm classified ORADS 4 which may be related to a borderline serous cystadenoma.

A thoraco-abdominopelvic CT scan also revealed the same left ovarian mass associated with bilateral inguinal and external iliac lymph node formations, the largest of which measures 7mm, a plunging thyroid goiter identified as multi hetero nodular goiter TIRADS 3 on cervical ultrasound. Biologically, she has normal thyroid function with thyroid levels: TSH at 0.5, T4L at 18 and T3L at 9.8 respectively. Note that she did not benefit from tumor marker testing. The decision was to perform a non-conservative total hysterectomy with bilateral adnexectomy, omentectomy and peritoneal cytology. The definitive anatomy-pathology resulted in a benign ovarian goiter. The decision is therefore to continue normal follow-up with a control ultrasound.

DISCUSSION

The presence of thyroid tissue in cystic teratomas in adults represents 2 to 8.9% of mature ovarian teratomas and 0.85 to 1.3% of all solid ovarian

tumors [1]. In the analysis of the literature, the age of onset is before menopause in 85% of cases with an average age of 42 years, the age range in which our patient falls [5].

The discovery of an ovarian goiter is most often fortuitous in its localized form, as was the case for our patient, sometimes causing a diagnostic delay. Ovarian goiter is most often diagnosed during a systematic examination or in the presence of pelvic pain, an abdominal mass or dysmenorrhea [6]. This clinical picture is also compatible with a malignant ovarian tumor, leading to additional standardized examinations. Meigs pseudosyndrome, combining ascites, pleurisy and a benign ovarian tumor, is found in only 5% of cases, with ascites possibly suggesting a malignant ovarian pathology [1-7]. Our patient did not present with ascites. Ectopic thyroid tissue can have the morphological and functional aspects of eutopic thyroid tissue. Generally benign, in 5 to 10% of cases it can present histological criteria of malignancy, most often of the papillary type [8]. None of these criteria for malignancy were present in the case of our patient.

Benign ovarian goiter can also be responsible for peripheral hyperthyroidism (5 to 8% of cases) with a drop in TSHus and an increase in T3l and T4l levels. Our patient's thyroid assessment was without abnormalities. It can also be non-functional if discovered incidentally during a gynecological examination.

Ovarian goiter should be considered in the presence of persistent peripheral hyperthyroidism despite a possible total thyroidectomy or in the absence of fixation of the thyroid gland on cervical scintigraphy accompanied by a normal or high thyroglobulin level.

Our patient had known thyroid damage, but did not have symptomatic hyperthyroidism or malignant proliferation. Hyperthyroidism is more common when there is an associated cervical goiter, which is the case in 16.2% of cases [5]. Surgical excision of the ovarian tumor allows euthyroidism to be restored, but sometimes a secreting "struma ovarii" can put the hypothalamic-pituitary thyroid axis at rest with a risk of transient postoperative hypothyroidism [9].

The exceptional complication of this procedure is the thyrotoxic crisis. It can occur after surgical excision of the secreting goiter, by sudden and massive release of thyroid hormones. Medical preparation is therefore necessary with the achievement of euthyroidism before the intervention at the risk of serious complications appearing [10]. This was done with our patient who was initially euthyroid before the surgical procedure.

Finally, a few cases of "struma ovarii" with manifestation of autoimmunity – Hashimoto's disease type have been reported, which was not the case for our patient.

We know that it is most often unilateral, but the predominance of the side varies according to the studies. The left ovary is affected in 63% of cases, but the involvement can be bilateral in 6% of cases [11, 12]. Condition fulfilled by our patient who had her ovarian goiter on the left side.

On a biological level, the determination of the CA-125 marker does not help in the diagnosis of ovarian goiter, nor of the malignant component. Not requested in our patient.

Approximately 5 to 8% of ovarian goiters are accompanied by hyperthyroidism with ectopic secretion of thyroid tissue [4-9]. Radiologically, preoperative diagnosis remains difficult due to sometimes similar characteristics of different types of teratomas.

Ultrasound features of ovarian goiter are nonspecific; they can present mixed solid and cystic components or associated with other components of a mature teratoma in almost half of the cases [13]. In our case we also had this non-specific aspect. Given this nonspecific appearance, a malignant origin can be suspected by the presence of septations and vegetations on ultrasound [13]. Malignancy in our case was very unlikely due to the predominantly benign features of our mass. Doppler flow seems interesting due to the richly vascularized nature of thyroid tissue compared to other components of non-vascularized teratomas (fatty or skin appendages) but has never been evaluated [13].

The characteristics of ovarian goiters on MRI correspond to a complex unilateral adnexal mass with a multilobulated surface, a thickened septa, composed of multiple cysts of varying signal intensity [11-14]. A marked hyposignal on T1 and T2 is suggestive of colloid content [15]. A marked T2 hyperintensity of the solid portion is often noted, greater than that of malignant lesions. The solid portions which are strongly enhanced after gadolinium injection correspond to the thyroid tissue and the richly vascularized stroma. The enhancement of the solid portion of ovarian goiters appears to be greater than that of the solid portions of malignant tumors [11].

Whatever imaging tests are performed, they are not discriminating enough to distinguish the benign or malignant nature of an ovarian goiter.

Pathologically, the criteria for malignancy are listed in the study by Devaney *et al*., presence of irregular, overlapping and ground-glass cell nuclei, intense mitotic activity or signs of vascular invasion [16].

Concerning therapeutic management, it is first necessary to carry out an anatomopathological diagnosis

[3]. The anatomopathological analysis, extemporaneous or remote, should make the diagnosis of a benign or malignant ovarian goiter. In the case of benign ovarian goiter, surgical treatment should be as conservative as possible, appropriate to the age of the patient. In a young woman who wishes to become pregnant, a cystectomy is recommended [17]. In a postmenopausal woman, a bilateral adnexectomy is justified. In the case of benign ovarian goiter, no additional treatment to unilateral oophorectomy is recommended, apart from remote clinical monitoring due to very rare recurrences in the literature [9]. Our patient immediately benefited from radical treatment

In case of malignancy, an extension assessment (abdomino-pelvic CT, pelvic MRI, CA 125, CEA), then surgical revision will be necessary with bilateral salpingo-oophorectomy, total hysterectomy, total omentectomy and pelvic lymphadenectomy [5-9].

It is also necessary to consider a possible intra-ovarian metastasis of thyroid cancer, although this situation has only rarely been described in the literature [18, 19]. In this context, it is necessary to look for a possible primary thyroid carcinoma using a thyroid ultrasound and discuss a total thyroidectomy. This thyroidectomy is also necessary before considering additional treatment with iodine 131, which must be discussed – in the event of histological signs of aggressiveness – [20]. The risk of recurrence, which is difficult to assess, given the small number of series reported, is estimated at 15% but warrants clinical monitoring with thyroglobulin testing [21].

CONCLUSION

Cases of Ovarian goiter are currently little described in literature. It is a rare pathology and difficult to diagnose because it is exclusively histological. Its endocrine function can lead to hyperthyroidism and recurrence remains exceptional. Its sometimes-malignant evolution close to thyroid carcinoma conditions its future and its therapeutic approach. We propose here a support model. The recommended treatment is conservative if patients wish to preserve their fertility. In the event of malignancy, treatment then becomes multidisciplinary between gynecologist, endocrinologist and oncologist with surgical revision, and discussion of thyroidectomy and treatment with iodine 131. Systematic monitoring of thyroglobulin is carried out remotely. Other multicenter studies will be necessary to improve diagnosis and decision-making.

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