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Case Report

An asymptomatic malignant Struma ovarii at saint pierre clinic of Ottignies, Belgium

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Abstract

Malignant struma ovarii is a rare tumor. This is a report of case of 45 year old female patient who was coming for a routine control at gynecology service of Saint Pierre clinic of Ottignies. During the check-visit, a left –attached pelvic mass was observed. The mass was confirmed at the abdominal ultrasound scan and magnetic resonance imaging scan. It was found localized at the left ovary. Follicular variant of papillary carcinoma type of malignant struma ovarii was diagnosed after a total radical hysterectomy and an omentectomy were done. Among two critical treatments that are classically proposed in such cases, a thyroidectomy followed by a 131 iodine radiotherapeutic ablation was found to be effective. Thyroxin suppressive treatments followed by biological and clinical assessments were applied. They were found to be effective since the patient had a positive evolution.

Keywords: Struma ovarii, ovarian teratoma, iodine radio-isotope, thyroidectomy, ovariectomy, saint-pierre ottignies clinic, Belgium.

INTRODUCTION

The malignant struma ovarii is an extremely rare tumor. So far, few cases have been reported in the literature about this issue (Devaney et al., 1993).The malignant struma ovarii represents 0.01 % of all ovarian tumors cases and 5 to 10 % of overall struma ovarii.

This struma ovarii is defined as a mature teratoma exclusively or partially composed of thyroid tissues. It may be subjected to histological changes identical to those of thyroid tissues (Devaney et al., 1993). Despite the diversity of biological markers and imagery techniques, the best diagnosis remains the pathological exam.

Currently, there is no agreement among practitioners on the surgical and post-surgical treatments of patients suffering from malignant struma ovarii.

This short communication is reporting the case of a forty-year-old patient affected by a malignant struma ovarii in its follicular variant of papillary.

Case Report

A patient of 45 years with no desire of conception came

at the clinic for a routine gynecological consultation. Reading from the disease history of the patient, there was no evidence or suspicion for the patient to have ever suffered from tumor or thyroidal pathologies. In addition, no single member of her family has ever suffered from such pathologies. However, the patient seems to have been an active smoker .The patient reported using “Gestofène-30” as a unique contraceptive method. The clinical examination revealed a left ovarian attached mass. This was confirmed by an endovaginal ultrasound scan that highlighted the existence of a heterogeneous mass (4.5 x 3.8cm) developing parasitically on the left side of the left ovary. When the magnetic resonance imaging scan was applied, an important solid component inside the tumor was identified. Characteristics damaged signals indicated an aggressive but continuous slow progress. The contralateral ovary is of normal dimension and structure during nuclear magnetic resonance scan and ultrasound scan observations.

There was no evidence of the presence of pelvic nodes. This led to conduct a laparotomy by making a pfannenstiel incision. An entry peritoneal cytology was realized. A peritoneal cavity exploration conducted

further revealed the existence of a cyst (around 1.5cm²) of papillary aspects of mixture components on the left side of the ovary.

A classical left adnexectomy was after conducted. Various biopsies were also realized on the peritoneum, on the right sacro-uterine ligament and on the right ovary (which 40% of its surface presents papillary aspect). An infracolic omentectomy was also realized. The pathological results revealed the existence of an malignant struma ovarii. The malignant struma ovarii found was a thyroid carcinoma into his follicular variant of papillary type. (Figure 1 and 2)

The right ovarian biopsy revealed the existence of a superficial confined serous papilloma with absence of suspected neoplastic damages. The other biopsies were with no particular characteristics. The patient was later referred to an endocrinologist for further treatments (hormonotherapy). Three months later, the physical examination was normal. The thyroid was no longer palpable. The biological thyroid check-up revealed the existence of an euthyroid balance (see table 1).

Two treatments options were presented to the patient. The first include theurapeutic abstention followed by a monitoring and the second consisted to a thyroidectomy followed by iodine radioisotope treatment. The patient adopted the second type of treatment which permits to optimize the surgery treatment and simultaneously to exclude the presence in situ of thyroid carcinoma and an ectopic implantation. Later, the thyroidectomy was realized.

The administration of the first ablative dose of 100mCi of iodine-131 was realized 4 weeks after thyroidectomy. The patient was in the hormonal weaning period. (see Table 1).

Scintigraphy indicated an excellent capture of iodine radioisotope at the level of the two residual thyroidal islands and at the level of pyramid of Lalouette in the cervical region. There was no distant iodine capture channel at the abdominal level. An alternative treatment with Thyroxine was administered followed by a monthly monitoring.

DISCUSSION

Epidemiology

Embryonic tumors represent 15 to 20% in total ovarian tumors. The majority of these tumors consist of dermoid cysts those may contain in 5 to 15% of cases, thyroid tissues. When the thyroid tissues represent more than 50% of total contents of dermoid cysts, then, this is a case of struma ovarii (Bolat et al., 2005). This one may involve into a form of malignancy in 5% of cases (Bolat, et al., 2005).

The most frequent forms are, like primitive tumors of thyroid gland, papillar carcinoma, secondary follicular carcinoma. A mixture of the two histological types is also described. To our knowledge, neither a medullar carcinoma of thyroid nor an anaplastic form have ever been described and reported from studies related to struma ovarii worldwide.

The transformation of a struma ovarii to a malignancy is always done during the fifth decade and is most frequent on the left ovary. In 6 % of cases, malignant forms of struma ovarii are automatically bilateral. About 5 à 6 % of malignant struma ovarii are automatically metastatic at the time of diagnosis (Savelli et al., 2008).

Previous studies reporting about malignant struma ovarii, indicate that in 50 % of cases, a dermoid cyst or another teratoma in the contralateral ovary is observed (Robboy et al., 2009).

Diagnosis

Physical abdominal examination gives a pelvic mass or an adnexal mass. This adnexal mass can be easily observed at a pelvic ultrasound scan. The patient can also present other gynecological symptoms such as metrorrhagia. Rarely, the first clinic sign of the disease is an ascites or a pleural effusion; and this is called a « pseudo-Meigs syndrom » (Devaney et al., 1993).

When an adnexal mass is suspected, a complementary investigation is classically conducted. this is consist of an abdominal and an endovaginal ultrasound scan and CT scan. A RMI(resonance magnetic imaging) of the pelvis is also proposed in case of doubt during the diagnosis process.

Practionners suggest that during a diagnosis process, if one has to observe on a abdominal ultrasound scan or abdominal CT scan that shows a solid multilocular ovarian mass which contains one or many round hyperechogenics structures with smooth borders in cystic spaces or « struma pearls »;then struma ovarii may be suspected (Savelli et al., 2008).

In almost all the cases, the diagnosis of struma ovarii is post operative and secondary to the histological analysis of the surgical piece. This diagnosis can be suspected in certain cases in pre-operative period with the presence of hyperthyroidism. Some scientists indicate that in 8 % of laboratory examination,if the hyperthyroidism is diagnosed then one has to suspect struma ovarii presence (Ayhan et al., 1993).

The measure of the tumor marker CA-125 has little diagnosis value for the patients who present an struma ovarii. The tumor marker CA-125 test is less specific for detecting malignant ovarian tumors. This marker gives constant values indicating a normal situation in more than two third of cases of malignant struma ovarii (Ayhan et al., 1993; Yoo et al., 2008). From the fact that struma ovarii are rare, in it malignant form, criteria of histological diagnosis have been controversial over longtime. The increase of cellularity compared to connective tissu and the presence of atypic cellular, may guide to a diagnosis of malignancy. Nuclear overlapping gives an aspect of frosted glass and increased mitosis correlated to characteristics of a papillar carcinoma.

Others signs like vascular or capsular invasion represent, as for all the malignant tumors, signs of bad pronostic, but which are usually complex and difficult to establish with the anatomo-pathologic test. In



Figure 1. Left ovary with contain a cyst. Macroscopic (view after ovariectomy)

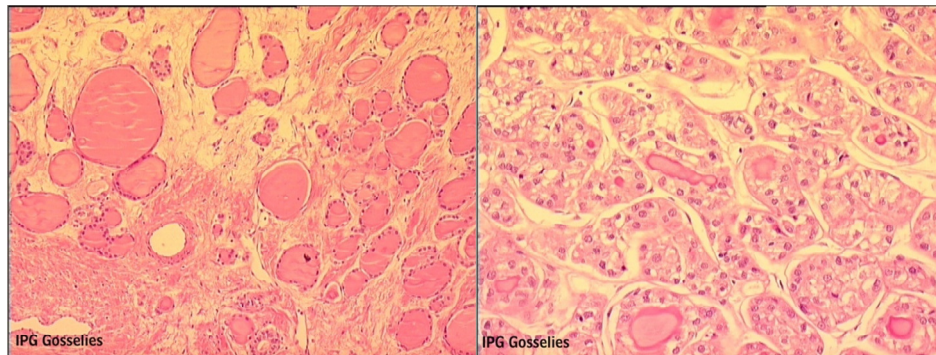


Figure 2. Anatomical Microscopical view. (a) The struma resembles a thyroid microfollicular. (b) follicular variant of papillary thyroid carcinoma.

Table 1. Biological thyroid check-up results

Examen	Thyroid check-up before radioisotope treatment	Thyroid, check-up after radioisotope treatment
TSH	1.60 microUI/ml (laboratory standard : 0.2 To 4.2)	>65.10 microUI/ml (laboratory standard : 0.4 to 4)
Free T4	(2,4pg/ml (Laboratory standard : 12 to 22)	< 3.0 pg/ml (laboratory standard : 8.9-17.6)
Thyroglobulin	27.28 ng/ml (Laboratory standard : <84)	2.1ng/ml (laboratory standard : 0-60)
Antibody antithyroglobulin	<222UI/ml (Laboratory standard : <280)	3.5 UI/ml (laboratory standard : 0-50)

immunohistochemistry, the findings of the thyroglobulin or TTF-1 (thyroid transcription factor 1) may help to confront the diagnosis ;these are classically present in well differentiated tumors (Bolat,et al., 2005).

There is an histological entity called follicular carcinoma highly differentiated from the ovary (HDFCO: highly differentiated follicular carcinoma of ovarian origin) because of that character of differentiation of the histological form and rarity of neoplastic signs during pathological exams. The diagnosis of malignancy is confirmed after apparition of metastasis. The HDFCO type is the malignant struma ovarii form with the best prognosis (Roth et al., 2008).

Macroscopically, the size of the tumor can also guide towards a malignant or benign lesion. It has been reported that a neoplastic tumor can observed in 75 %

of cases of struma ovarii when the size of the tumor is above 16cm;on the opposite, it is rare in case of a benign struma ovarii when the size of the tumor is above 5cm(Young 1993).

It's important to differentiate a malignant struma ovarii with an ovarian metastasis of a thyroid tumor.In exclusive presence of thyroid tissues in the ovarian lesion or an absence of a component of teratoma ;and if the test shows a primitive neoplastic lesion in the thyroid gland, a metastasis is suspected (Bolat et al., 2005).

Treatment

Occurrence of rare cases and the challenge to

diagnose pre-operatively, makes treatment not clearly defined across literatures (Hinshaw et al., 2012).

Previously, the treatment of a malignant struma ovarii had 2 different parts.

On one side, some authors suggested to treat a malignant struma ovarii like a germinative ovarian tumor which is becoming malignant. This kind of treatment is radical: radical total hysterectomy, omentectomy, biopsies of pelvic and lombo-aortics nodes and a cytologic test of the peritoneal fluid (Hatami et al., 2008). For a young patient, with desire of conception, an unilateral salpingo-oophorectomy can be realized with a complete examination of the abdominal cavity and biopsies of the contralateral ovary (Hatami et al., 2008). This treatment can be only applied naturally to tumors which are confined to the ovary. During the surgery, a scintigraphic exam with ¹³¹-iodine is necessary to exclude metastasis and in case of lack of secondary lesions, no adjuvant treatment is indicated.

Other side, others authors suggest to treat a malignant struma ovarii like a primitive tumor of the thyroid gland. We must start by a surgical resection of the lesion. The surface of resection depends on the stage of the lesion, from a simple cystectomy or ovariectomy for lesions limited to the ovary to a wide resection in case of massive capsular extension. A thyroidectomy must be realized even in case of lack of tumor lesions on the thyroid gland. A scintigraphy with ¹³¹-iodine is also realized to exclude the presence of secondary lesions. Thereafter, a treatment by radioisotope must be realized. The follow-up must be realized by testing the thyroglobulin and the realization of the Scintigraphy ¹³¹-iodine.

The prognosis of papillar carcinoma which is developed in a struma ovarii is better than a follicular type, like in primary thyroid carcinoma (Roth et al., 2008).

CONCLUSION

In brief, thyroidectomy followed by a radiotherapy ablation allows optimizing the surgery treatment and

simultaneously to exclude the presence in situ of thyroid carcinoma and an ectopic implantation is suggested.

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