Gynecologic Oncology Reports 17 (2016) 56–59

Contents lists available at ScienceDirect

Gynecologic Oncology Reports

journal homepage: www.elsevier.com/locate/gynor

Case report

Treatment and follow-up of malignant struma ovarii: Regarding two cases

Aurore Oudoux [a,*], Eric Leblanc [b], Juliette Beaujot [c], Hélène Gauthier-Kolesnikov [a]

[a] Department of Nuclear Medicine, Oscar Lambret Center, 3 rue Combemale, 59020 Lille Cedex, France
[b] Department of Surgery, Oscar Lambret Center, 3 rue Combemale, 59020 Lille Cedex, France
[c] Department of Biology, Unit of Morphological and Molecular Pathology, Oscar Lambret Center, 3 rue Combemale, 59020 Lille Cedex, France

ARTICLE INFO

Article history:
Received 19 April 2016
Received in revised form 19 May 2016
Accepted 31 May 2016
Available online 8 June 2016

Keywords:
Malignant struma ovarii
Radioactive iodine therapy
Thyroid carcinoma

ABSTRACT

Malignant struma ovarii (SO) is a rare tumor, and as a consequence, treatments and follow-up procedures are not clearly established. Presented in this study are two cases of suspicious ovarian masses, resected and corresponding to malignant SO on histopathology. Similar to thyroid cancer, we proposed complementary radioiodine therapy (^{131}I) after total thyroidectomy (no malignancy was observed at this level in our two patients). Patients underwent treatment with 3.7 GBq ^{131}I followed by post-therapy whole-body scintigraphy, which can detect residual disease or occult metastases. Thyroid remnant ablation increases the sensitivity and specificity of follow-up testing using serum thyroglobulin levels as a tumor marker. Our two patients remained disease-free for 3 and 5 years, respectively, after treatment.

1. Introduction

Struma ovarii (SO) is a rare tumor defined as a mature ovarian teratoma containing 50% or more thyroid tissue, and it accounts for approximately 5% of all ovarian teratomas that are mostly benign. Malignant transformation of SO is reported to occur in less than 5% of all cases, and it is even less likely to lead to metastatic disease (5–6%) (Dardik et al., 1999). Due to the rarity of this type of tumor, there has been a paucity of data in the literature pertaining to treatments and follow-up procedures for this tumor. After local resection of the ovarian tumor, no consensus exists between surveillance alone and adjuvant treatment (De Simone et al., 2003). Similar to thyroid cancers, we proposed thyroidectomy, required before radioactive iodine ablation (^{131}I) and thyroxine suppressive therapy. We report two cases of malignant SO treated in our institution: one patient with localized disease and the other with lymph node and peritoneal extension.

2. Clinical cases

2.1. Case 1

A 49-year-old woman presented for evaluation of an asymptomatic left ovarian mass, which was incidentally discovered during a routine check of an intrauterine device (2012, October). Ultrasonography and pelvic MRI exhibited an 8-cm-diameter left ovarian mass, mixed tumor with gadolinium enhancement of the posterior solid component and low abundance intraperitoneal effusion. The appearance was consistent with an ovarian mucinous adenocarcinoma.

The CA-125 level was elevated to 111 U/ml (normal < 35), while CEA and CA19-9 levels were normal. In December 2012, this patient underwent laparoscopy with a total hysterectomy, bilateral salpingo-oophorectomy with omentectomy, and pelvic-aortic lymphadenectomy. On pathological examination, the ovarian mass was a malignant SO, consisting of follicular variants of papillary thyroid carcinoma (7 × 5.5 × 3 cm) and no other histological features of malignancy. For adjuvant therapy, it was proposed to treat this patient with procedures similar to thyroid cancer, rather than other malignant ovarian cancers. In February 2013, she underwent total thyroidectomy with the intention of administering ^{131}I therapy as an adjuvant setting. On histological examination, the thyroid consisted of normal tissue. Two months later, the patient received a treatment dose of iodine 131 (3.7 GBq) after thyroxine withdrawal for four weeks. The thyroglobulin level was increased to 19 ng/mL (TSH = 27 mIU/l), and serum thyroglobulin antibodies were negative. Two days after treatment, the whole-body ^{131} I scintigraphy showed high uptake of cervical remnants (Fig. 1A and B). The patient was maintained on thyroid hormone suppression with thyroxine. Six months later, a diagnostic ^{131}I scintigraphy (185 MBq) was conducted after four weeks of thyroxine withdrawal, which demonstrated complete remnant thyroid ablation (Fig. 1C and D). Clinical examination, neck ultrasonography and thyroglobulin level measurements were also negative (thyroglobulin <1 ng/ml, TSH = 43 mIU/l), confirming remission. Clinical and biological (TSH and thyroglobulin) examination was planned every 6 months for two years and annually thereafter. This patient has completed 3 years of follow-up with no evidence of recurrence.

* Corresponding author at: Department of Nuclear Medicine, Oscar Lambret Center, 3 rue Frederic Combemale, 59020 Lille Cedex, France.
E-mail address: a-oudoux@o-lambret.fr (A. Oudoux).

http://dx.doi.org/10.1016/j.gore.2016.05.014

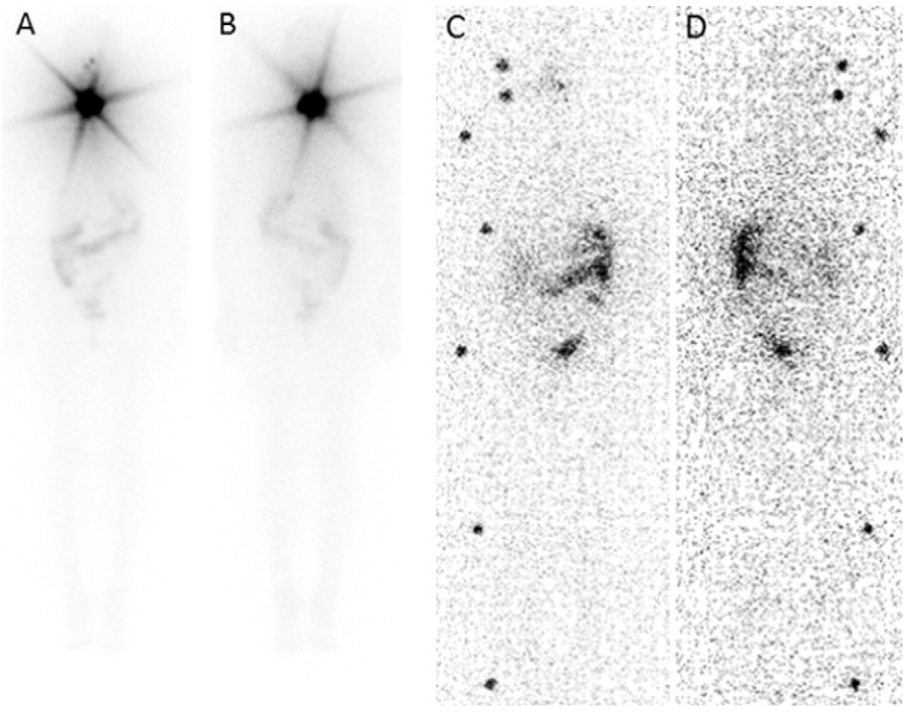

Fig. 1. A 49-year-old woman presented a left ovarian malignant struma ovarii resected followed two months later by a total thyroidectomy. (A and B) Whole-body scan (anterior and posterior views) performed two days after ^{131}I therapy (3.7 GBq) showed remnant thyroid tissue but no evidence of distant functioning metastasis. (C and D) Diagnostic whole body scan (anterior and posterior views) of ^{131}I (185 MBq) performed six months later, showing complete ablation of remnant tissues.

2.2. Case 2

A 67-year-old woman with a history of right nephrectomy for clear cell carcinoma in 1986 presented with post menopausal bleeding in December 2010. Abdomino-pelvic ultrasonography showed uterine fibroma, a 4-cm left ovarian mass associated with a right retrocaval adenopathy, without ascites. Pelvic MRI showed a left latero-uterine lesion (3.5 × 4 × 2.5 cm), hypo signal T1 and T2, without gadolinium enhancement. Appearance was consistent with a fibroma or fibrothecoma. Further exploration by ^{18}FDG PET showed a retrocaval lymph node and left ovarian mass hypermetabolisms (Fig. 2). Bone scintigraphy and thoracic computed tomography were negative. Tumor markers were normal (CEA = 2.4 μg/l, CA125 = 28.2 U/ml, CA15–3 = 13 U/ml and CA19-9 = 23 U/ml). In February 2011, she underwent a laparotomy with total hysterectomy, bilateral salpingo-oophorectomy with omentectomy, and paraaortic, retrocave and pelvic lymphadenectomy. The left ovarian tumor measured 5.5 × 4 cm. Histological examination showed malignant SO (papillary carcinoma type), associated with a metastatic retrocaval lymph node (5 × 2.5 × 4 cm) and omental nodules (Fig. 3).

After the diagnosis of thyroid-type malignancy was established, she had a total thyroidectomy (May 2011) with negative histology, to optimize uptake of radioactive iodine 131 (3.7 GBq) six weeks later. The ^{131} I post-therapeutic scintigraphy only showed thyroid remnant uptake without abnormal abdominal uptake. The stimulated serum thyroglobulin was lower than 1 ng/mL (TSH > 100 mIU/ml).

Six months later, stimulated serum thyroglobulin, ^{18}FDG PET, diagnostic ^{131}I scintigraphy and neck ultrasonography were normal. The subsequent follow-up consisted of a clinical examination and thyroglobulin measurements performed every six months for two years and annually thereafter. Pelvic MRIs performed 1 and 2 years after treatment showed no evidence of recurrent disease. She remains free of disease 5 years later.

3. Discussion

Most cases of malignant SO are incidentally discovered. The typical presenting symptoms are pelvic pain, abnormal uterine bleeding and abdominal or pelvic masses (De Simone et al., 2003; Yoo et al., 2008 Jun). It is rare for patients to present signs of hyperthyroidism (approximately 7% of SO) (Matsuda et al., 2001 Sep).

DeSimone et al. (De Simone et al., 2003) reviewed approximately 24 cases of malignant SO: lesion sizes ranged from 5 to 20 cm, more commonly on the left side (63%), and most frequently with follicular variants of papillary thyroid carcinoma (54%) and papillary thyroid carcinoma (21%). Our two clinical cases presented the same primary characteristics.

CA125 is widely accepted as tumor marker of ovarian cancers and is also elevated in other tumor lesions. Moreover, this marker can increase in non-malignant related gynecologic conditions (endometriosis, pregnancy) or as a secondary effect due to the presence of ascites (Leung & Hammond, 1993; Jiang et al., 2010 Jul 29). In our two reported malignant SO cases, the first with ascites had increased CA125, while the second patient (with peritoneal involvement but without ascites) had a normal CA125 rate at diagnosis.

Because ovarian malignancies were suspected from the images, our patients were first treated with surgery, enabling histological diagnosis and locoregional extent.

The cytological diagnostic criteria for papillary carcinoma are similar to those described for the cervical thyroid gland, which include mitotic activity, appearance of the cores (irregular, ground-glass) or vascular invasion (Devaney et al., 1993 Oct). SO with metastasis is also classified as a malignant disease.

Therapy for benign SO is surgical resection (Kunstmann & Fénichel, 2007 Jan). In malignant SO, surgery is also the first phase of treatment. A conservative surgery (unilateral salpingo-oophorectomy) may be acceptable in fertility-desiring patients if they have a localized tumor (Salomon et al., 2003). A total hysterectomy with bilateral salpingo-