Malignant struma ovarii: a case report of laparoscopic management

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Abstract

Background. Struma ovarii is a rare disease. Malignant transformation is even rarer. Data about its management are lacking. We describe the first reported case of a malignant struma ovarii treated and staged by laparoscopy.

Case. A 49-year-old patient was operated by laparoscopy for a right ovarian teratoma. The patient did not show symptoms of hyperthyroidism. The ovarian teratoma was removed in a plastic bag and definitive histology showed foci of papillary adenocarcinoma in a struma ovarii. The patient was then staged by laparoscopic surgery undergoing left adnexectomy, multiple peritoneal and omental biopsies, and common iliac and paracaval lymph node sampling. Hysterectomy was not performed. The postoperative course was uneventful and the patient was released on the second day. Thyroglobulin level was monitored and the patient is free of disease after more than 1 year.

Conclusion. The preoperative diagnosis of malignant struma ovarii is difficult. Even with cautious evaluation of the patient, some risk of wrong diagnosis is possible. This is why a meticulous technique of laparoscopic surgery in removing the ovary is important. Laparoscopic staging may also intervene in very limited cases; the expertise to perform open staging of the patient is necessary but the postoperative course is fast.

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Introduction

Struma ovarii was first described by Von Kalden in 1895 and Gottschalk in 1899 [1] and is diagnosed when thyroid tissue is predominant in a teratoma [2]. Even if malignant transformation is rarely reported, metastatic disease is possible [3–6].

Due to the rarity of malignant transformation which occurs in about 5% of all struma ovarii [7], data about the management of the tumor are lacking and based on the experience of ovarian tumor and of thyroid carcinoma.

We report on a case of thyroid papillary adenocarcinoma in a struma ovarii which was treated and staged by laparoscopic surgery.

Case report

M.C., 49 years old, Caucasian, was scheduled for laparoscopic salpingooophorectomy because of a right ovarian mass. She was still menstruating, was 1.67 m tall, and weighted 85 kg for a body mass index of 30.47.

The patient did not show any sign of hyperthyroidism and had already had an appendectomy.

Preoperatively the patient was evaluated with transvaginal ultrasound repeated twice. Sonography revealed a dishomogeneous mass diagnosed as an ovarian teratoma 7 × 6 × 6 cm. In the cyst a solid area was seen. Ca-125 was within the limit range. The patient refused the first scheduled procedure in July 2001 and underwent surgery in late October 2001. In the meanwhile the ultrasound scan showed no modification. The patient gave consent only to monolateral salpingo-oophorectomy, refusing bilateral adnexectomy.

At laparoscopy, right salpingo-oophorectomy was performed. At entry, the right ovary was white and smooth. The left ovary and all the abdomen appeared free of disease. Even if peritoneal washing was obtained it was not delivered for cytological examination. Laparoscopic surgery was performed isolating the infundibulopelvic ligament about 4 cm cranial to the ovary and visualizing the ureter. Bipolar coagulation was used for hemostasis. After coagulation of
the utero-ovarian ligament and of the tube, the mass was extracted by means of an Endo-catch (Auto Suture). A 3-cm minilaparotomy was performed and morcellement of the mass was accomplished in the plastic bag. Sebum and hair were seen and so a frozen section was not asked for.

Pathologic findings were consistent with a diagnosis of foci of thyroid papillary carcinoma in a struma ovarii. The right ovarian mass, 6 cm in size, showed a smooth outer surface. Microscopic examination revealed a mature cystic teratoma containing predominantly thyroid tissue. Arising within the thyroid tissue a multifocal papillary adenocarcinoma of the thyroid was found. The fallopian tube was unremarkable (Fig. 1).

We proposed to the patient a staging procedure and a laparoscopic approach was planned.

Before staging the patient had a complete thyroid function evaluation and thyroid sonography, which were all in the normal range. Thyroglobulin level was 1.7 ng/ml (normal value between 1.0 and 43.0).

The patient underwent a second laparoscopic procedure with peritoneal washing, left adnexectomy, multiple peritoneal and omental biopsies, and common iliac and paracaval lymph node sampling. Hysterectomy was discussed but we decided not to perform it. Four trocars were used for laparoscopy; they were positioned in the usual diamond position. No left upper quadrant trocar was used. The right ancillary trocar was passed in a different site than the previous extraction port. The latter was excised and sent for histology. The duration of the surgery was about 2.5 h (Fig. 2). Definitive histology was negative. Eight lymph nodes were retrieved.

The postoperative course was uneventful and the patient was released after 2 days. No postoperative treatments were performed.

Serum thyroglobulin level evaluation and clinical examination were performed every 4 months for the first year. Thyroglobulin levels were in the normal range. The patient is free of disease after more than 1 year. Thyroid function is at present normal.

Discussion

To the best of our knowledge, this is the first report of complete management of a papillary adenocarcinoma in a struma ovarii by means of laparoscopy.
Even if ultrasound can frequently diagnose dermoid cysts, it is not useful in showing struma ovarii. However, struma ovarii should be remembered, mostly in “solid-looking” teratomas. Anyhow, malignancy in the struma ovarii is so rare that by no means can it be ruled out. The preoperative diagnosis of struma ovarii is possible in patients having hyperthyroidism by thyroglobulin measurement [8] or by scanning [9], but our patient was totally asymptomatic. Furthermore, the reported rate of hyperthyroidism in patients having struma ovarii is only 7–8% [3,5].

It is our policy to ask for frozen section analysis in the diagnosis of suspicious masses during laparoscopic surgery. In this case, it was not asked for because the ultrasound of the patient had remained unchanged for about 4 months and part of the content of the cyst presented as sebum and hair. This may also demonstrate a slow pattern of growth of this papillary tumor in this patient.

We usually perform bilateral salpingo-oophorectomy in a 49-year-old patient. In this case the patient wanted to have the healthy ovary left.

Some authors [10,11] claim that immediate visual examination of the sample by means of the laparoscope can rule out malignancy. We are used, in suspicious cases, to remove the ovary with a long pedicle and to avoid spillage as much as possible. Removal through a plastic bag in these cases is necessary. Since we wanted to be completely sure of the port site of extraction, even if morcellement was performed in the bag, at the second laparoscopy we changed access, performing a biopsy of the peritoneum underlying the port and also of the scar and subcutaneous tissue. Histology showed no disease in the port site.

Difficulties with clinical diagnostic criteria combined with the rarity of this tumor make evaluation of various treatments controversial [4].

After the pathologic findings of a malignant struma ovarii, we decided to stage our patients according to ovarian cancer and thyroid cancer guidelines. Thyroid sonography was performed, thyroid function was evaluated, and thyroglobulin level was measured as a future marker of disease. Even if it has been reported in few cases [12–14], we believe that laparoscopic ovarian staging can be performed safely in some particular patients. Hysterectomy was an important source of discussion, since complete staging would require also this procedure which some authors claim to be necessary in these cases [3]. However, we eventually decided to remove the other ovary, to evaluate the peritoneum and the lymph nodes, avoiding hysterectomy. We feel that laparoscopic assisted vaginal hysterectomy would only increase the operative time, but not the complexity of surgery.

In the staging procedure proposed for stage I papillary thyroid carcinoma recognized nodal involvement should be removed, but selective node removal can be performed and radical neck dissection is not required [15]. According to this staging procedure and after consultation of our hospital’s endocrine surgeon, we decided to perform a sampling
of lymph nodes. In this case, common iliac and paracaval lymphadenectomy would be difficult since the patient had a BMI of over 30. The lymph node sampling was performed up to about the origin of the inferior mesenteric artery.

In these cases, laparoscopic surgery has shown great advantages with respect to open staging since the patient was released within 2 days.

After evaluation of definitive histology and according to the advice of the endocrine surgeon, the patient did not receive I\textsuperscript{131} therapy.

Thyroglobulin levels have been shown to be more sensitive for recurrent thyroid carcinoma than whole-body diagnostic I\textsuperscript{131} scanning and thyroglobulin levels have replaced periodic scanning for monitoring disease status [16]. Therefore we decided to follow patients with serum thyroglobulin level evaluation.

In conclusion, we believe that laparoscopic surgery can be important in removing suspicious ovarian masses but the technique must be careful and meticulous and, even if laparoscopic staging is not the standard procedure for ovarian cancer, it can be accomplished safely andatraumatically in particular cases of malignant tumors of the ovary.

References