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CASE REPORT

Luteinized fibrothecomas of the ovary associated with sclerosing peritonitis in a patient with systemic lupus erithematosus

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Abstract Sclerosing peritonitis is a peritoneal subserosal fibrosis that has been associated with luteinized thecomas of the ovary. Ascites, peritoneal thickening and adnexal masses were found in a 38-year-old woman. She had been an intravenous drug abuser and had systemic lupus erytematosus. At laparotomy the association of peritoneal diffuse fibrosis and bilateral luteinized fibrothecomas was diagnosed. The postoperative course was fatal due to small bowel obstruction. The concomitant finding of ascites, peritoneal thickening and adnexal masses suggested the diagnosis of ovarian cancer. Although the patogenesis is unknown, predisposing causes of sclerosing peritonitis have been described such as intravenous drug abuse and systemic lupus erithematosus.

Keywords Ovarian luteinized fibrothecomas · Ovarian fibrothecomas · Luteinized fibrothecomas · Sclerosing peritonitis · Systemic lupus erithematosus

The authors dedicate this article to the memory of Prof. J. Font who died during the preparation of this article.

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Introduction

Sclerosing peritonitis (SP) is a peritoneal subserosal fibrosis that has been associated with luteinized thecomas of the ovary (LTO) [1, 2]. It has also been described associated with certain drugs and illnesses, and as a complication of chronic ambulatory peritoneal dialysis [1]. Some authors have suggested the association of SP and autoimmune diseases such as systemic lupus erythematosus (SLE) [3]. Most of the cases described were, at first, misdiagnosed as disseminated peritoneal malignancy from an ovarian carcinoma [1, 2]. In this paper we report the first case of this rare association in a patient with SLE.

Case

A 38-year-old non-gravida woman was admitted to the hospital complaining of acute abdominal pain, nausea and abdominal swelling. She also complained of weight loss of 7 kg over the past year. Ten years ago she had been an intravenous drug abuser, and she also had SLE.

On physical examination abdominal swelling was found. She underwent abdominal and transvaginal ultrasound exploration which showed marked ascites, walled up in the upper hemiabdomen, and bilateral solid, multinodular and pooly vascularized adnexal masses of 70 and 80 millimetres (mm) each. Laboratory findings showed CA 125 levels of 112 U/mL (normal range 0–40 U/mL). Carcinoembryonic antigen and CA 19.9 levels were in the

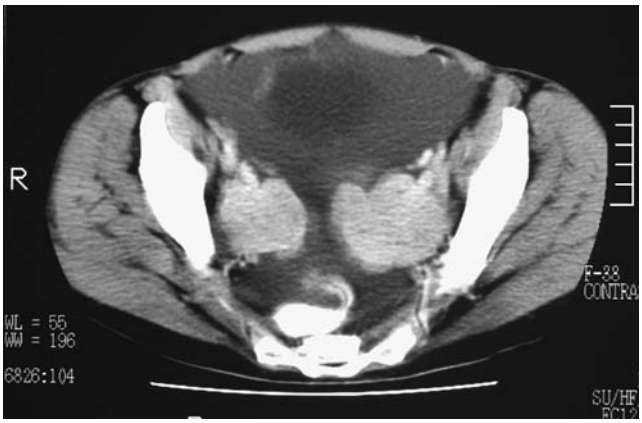

Fig. 1 CT scan of tumors

normal range. Paracentesis revealed an exudate without evidence of malignancy or infection. A CT scan showed both tumors (Fig. 1), the walled up ascites (Fig. 2), and also showed peritoneal and small bowel thickening, more evident in the greater omentum. All these findings suggested the diagnosis of ovarian cancer. A laparotomy was performed.

At laparotomy, 900 millilitres of yellowish watery ascitic fluid was found in the peritoneal cavity. Examination of the ovaries revealed multinodular and firm ovarian tumors of 75×35×30 mm in the right ovary, and 110×55×45 mm in the left ovary (Fig. 3). Diffuse fibrosis of the mesentery and the small bowel serosa and omental thickening were also found. The walled up peritoneal and omental fibrosis in the upper hemiabdomen did not let the surgeon explore the liver nor the stomach. Bilateral oophorectomy and an omental biopsy were performed. The sectioned surfaces of the tumors were whitish, solid and homogeneous (Fig. 4). Microscopically, the ovarian lesions were a mixture of cellular and edematous zones. The cellular zones consisted of densely packed fascicles of fibroblasts with nests of luteinized cells (Fig. 5). Mitoses were abundant between

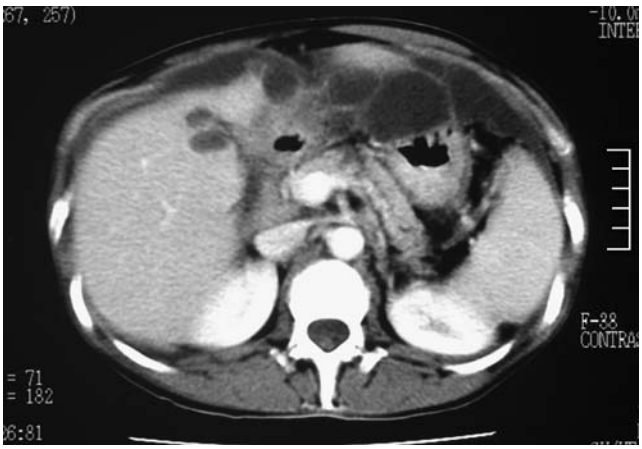

Fig. 2 CT scan showing walled up ascites

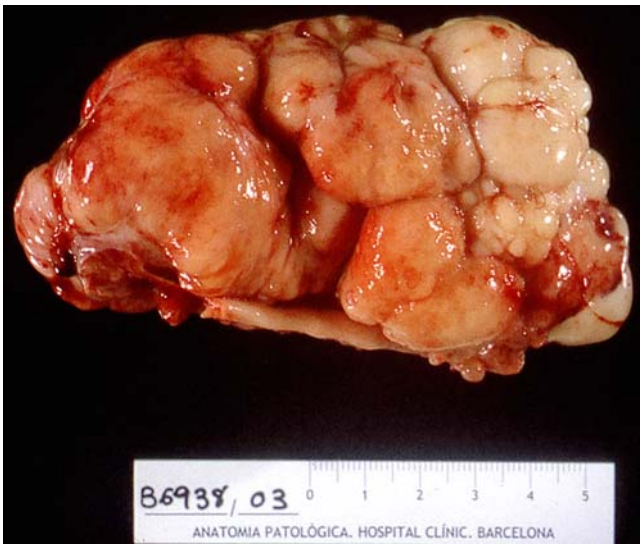

Fig. 3 Ovarian tumors

luteinized cells but no atypical mitotic figures were seen. The final diagnosis was of bilateral luteinized fibrothecomas. The omental biopsy showed fibrous tissue containing collagen with very rare mitotic figures being catalogued as sclerosing peritonitis.

The postoperative course was complicated by small bowel obstruction that improved with conservative management. Three months after hospital discharge the patient died due to small bowel obstruction and extensive peritoneal adhesions although glucocorticoid treatment and new surgical interventions had been performed.

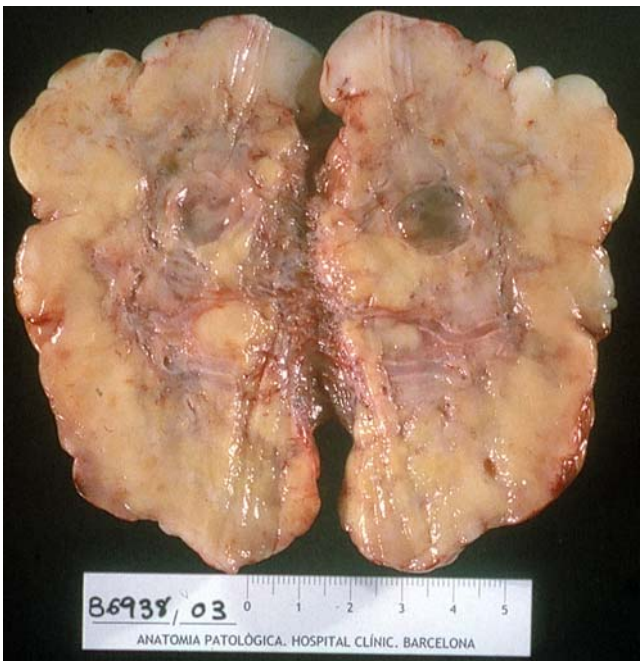

Fig. 4 Sectioned surfaces of the tumors

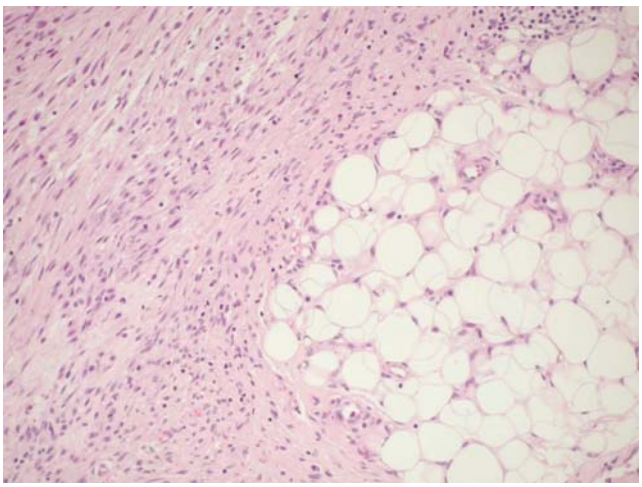

Fig. 5 The cellular zones consisted of densely packed fascicles of fibroblasts with nests of luteinized cells

Discussion

SP is a rare inflammatory process that leads to the deposition of a thick fibrous membrane on the peritoneum. The serosa of the small bowel is the most frequently affected site and clinical manifestations are usually abdominal mass and/or small bowel obstruction [1].

In our case the concomitant finding of ascites, peritoneal thickening and adnexal masses strongly suggested the diagnosis of ovarian cancer. Although the pathogenesis is unknown, predisposing causes of SP have been described [1–3]. The patient's case report had two predisposing causes of SP: she had been an intravenous drug abuser and she had SLE.

SP treatment should be conservative surgical removal of the thickened peritoneum, lysis of adhesions, and partial resection of the small bowel. When associated with LTO the treatment must include the removal of the tumor/s. Glucocorticoid treatment has been tried in another case associated with LTO with favourable results avoiding the surgery [2]. Most patients require more than one surgical intervention as the postoperative course is usually complicated with the formation of new fibrosis and more adhesions [1, 2].

In summary, we present a new case of bilateral LTO with SP that differs from those previously reported by having several possible predisposing causes of SP (SLE and being a past drug abuser). The possible diagnostic confusion with malignancy, the way the illness can degrade the quality of life, and the potentially lethal surgical complications emphasize the importance of increasing awareness of this unusually recognized entity.

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