A case of advanced stage and poorly differentiated Sertoli-leydig cell tumor of the ovary

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Abstract

Sertoli-leydig cell tumor (SCLT) is a rare ovarian tumor that usually occurs unilaterally. These tumors constitute less than 0.5% of ovarian tumors and are further categorized based on the degree of mitotic activity. We present the case report of a young woman with an SLCT along with different considerations of treatment.

Introduction

Sertoli-leydig cell tumor (SCLT) is a rare ovarian tumor that belongs to the group of sex-cord stromal (1). Most tumors are unilateral and constitute less than 0.5% of ovarian tumors. These tumors are characterized by the presence of testicular structures that produce androgens. The second characteristic feature of these tumors is the degree of differentiation of structures in them. Women of all ages may be afflicted, but the average age of onset is 25.

Case Report(s)

A 27 years young nulliparous women, with no history of disease consults for appearance of the following clinical symptoms: voluminous abdomino-pelvic mass, secondary amenorrhea, and a mild hirsutism. Her general physical examination was normal except for the presence of hirsutism and clitoromegaly. Vaginal examination revealed a firm and mobile cystic mass in the right adnexa.

An abdominal and chest computed tomography scan was performed which confirm the presence of a 13 centimeter mass in the right ovary. The left ovary and the uterus were normal. No other lesion was individualized.

The level of CA125 was elevated to 336UI/ml.

Operative findings showed replacement of the right ovary by a 13cm solid, grey-white, smooth-surface mass. The omentum and peritoneal surface were covered with numerous tumor implants.

The radical surgery was not possible and a non-optimal cytoreduction was performed.

Only a biopsy of the ovarian mass was performed and a histopathological analysis showed that it is an ovarian sertol-leydig tumor with a positif Anticorps anti-cytokératine, a positif anticorps anti-calretinine, a positif anticorps anti-CD117 and a positif anticorps anti-E cadherine after an immunohistochemistry analysis. The tumor was poorly differentiated.

Based on the above findings, a final diagnosis of ovarian sex-chord tumor (Sertoli-Leydig cell), stage IIIIC, poorly differentiated, was made.

After the histopathology report was received, we had a detailed discussion of various treatment options with the patient. After her consent, 3 cycles of chemotherapy with a bleomycin, etoposide, and cisplatin regimen were administered 3 times a week before a debulking surgery.

After these three cycles radiological assessment showed a stabilization of the ovarian mass. The CA125 had decreased to 135 Ui/ml.

The decision was then to administrate a second line chemotherapy consisting on VIP protocol combining Etoposide, Ifosfamide and cisplatine.

Unfortunately, the patient died after two courses of chemotherapy from a severe sepsis.

Discussion

We here present an unusual case of SLCT poorly differentiected with advanced stage where the radical surgery was difficult to perform.

the most prognostic factors in these tumors other than stage appear to be histologic differentiation, mitotic index, the presence of heterologous elements, and tumor rupture. In a review of 207 cases by Young and Scully in 1985,(1) all well-differentiated tumors were benign, whereas 11% of tumors with intermediate differentiation, 59% of tumors with poor differentiation, and 19% of those with heterologous elements were malignant. In another study of 64 patients who had intermediate or poorly differentiated SLCT, a survival rate of 92% was noted at both 5 and 10 years. (2)

The majority of these patients are seen during the second and third decades of life, with the average age at diagnosis 25 years. Around 50% of cases come to
clinical attention because of progressive defeminization, as was seen in this patient who had a mild hirsutism(1). Other symptoms can be observed like menorrhagia, metrorrhagia, amenorrhea, abdominal pain.

Most of these tumors are unilateral and diagnosed in stage I, so conservative surgery in a young patient is an appropriate treatment. Adjuvant chemotherapy is considered for patients who have poor prognostic factors.(3)

The use of platinum-based chemotherapy in the form of BEP has emerged as the most common treatment for many patients with advanced or recurrent SLCT of the ovary. The following chemotherapy regimens were also tested: Cisplatin, doxorubicin, cyclophosphamide (PAC)(4); Vincristine, actinomycin-D, cyclophosphamide (VAC)(5); with poor response rates.

The patient in this report received chemotherapy for SLCT in stage IIIIC. The factors responsible for this decision were advanced stage, incomplete surgery and poorly differentiation in histopathologic study of the tumor.

There was no good good response demonstrating that other regimen of chemotherapy should be tested for this histological type of tumor.

**Conclusion**

SLCT is a rare ovarian sex-cord tumor that usually occurs unilaterally. SLCT should always be considered in a young female patient who has symptoms of virilization and an ovarian mass on examination or investigation.

Poorly differentiated tumors require aggressive management because the chances of them being malignant are high.

Because of their extreme rarity, the limited published studies the optimal treatment of this group of tumors remains somewhat enigmatic.

Advances in this area will be made only through cooperative group trials or through information gathered from case reports and small series.

**References**

Illustrations

Illustration 1

CT scan showing the ovary tumor

Illustration 2

CT scan showing the peritoneal implants