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Original Article

Mitotically active cellular luteinized thecoma of the ovary and luteinized thecomatosis associated with sclerosing peritonitis: Case studies, comparison, and review of the literature

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ABSTRACT

In this study, we distinguish two clinical and pathological entities that are similarly named: luteinized thecoma and luteinized thecoma associated with sclerosing peritonitis. Ovarian luteinized thecoma lacks definitive criteria for malignancy. Based on our case study of a mitotically active neoplasm without nuclear atypia in which the patient was living and well 19 years after operation and comparison with prior studies of luteinized thecoma and the closely related entity of cellular fibroma, we propose presumptive criteria for malignancy for this rare neoplasm. Increased mitotic activity in luteinized thecoma without significant nuclear atypia is not an indication of malignant behavior, and such cases should therefore be referred to as mitotically active cellular luteinized thecoma. We also contrast neoplasms in the luteinized thecoma category with the entity originally reported as luteinized thecoma associated with sclerosing peritonitis. In the latter, the ovarian stromal proliferations are typically bilateral, can have an exceedingly high mitotic rate as was seen in our illustrative case, often incorporate non-neoplastic ovarian structures at their periphery, and are responsive to medical therapy. In our patient with sclerosing peritonitis, both the ovarian masses and peritoneal sclerosis underwent complete regression following treatment with gonadotropin-releasing hormone agonist and high doses of steroids, and an ovarian biopsy taken 2 months after therapy showed a histologically normal ovary. The patient subsequently became pregnant and delivered a normal infant. This is, to our knowledge, the first case of successful medically conservative treatment of a young patient with this entity that led to complete relief of symptoms and allowed preservation of fertility. Because recent observations support the non-neoplastic nature of the ovarian stromal proliferations, we advocate use of the previously proposed term *luteinized thecomatosis associated with sclerosing peritonitis* for this entity.

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Introduction

Ovarian luteinized thecomas are rare stromal neoplasms for which exact criteria for malignancy have not been established. In the largest reported series reported in 1982, Zhang et al. considered an elevated mitotic rate to be an important criterion for malignancy, but emphasized that their experience was limited [1]. They stated that in the absence of well-established criteria for separating benign from malignant luteinized thecomas, they were using the criteria of Prat and Scully, which had been previously applied to distinguish cellular fibromas and fibrosarcomas [2]. In 2006, Irving et al. studied 40 mitotically active cellular fibromas defined as having four or more mitotic figures per 10 high power fields (MFs/10 HPFs) with bland histology and determined that their behavior was similar to that of cellular fibromas having a lower mitotic rate [3]. They developed criteria for malignancy that superseded earlier studies.

Two large series have been reported as luteinized thecoma associated with sclerosing peritonitis [4,5]. Although the sclerosing peritonitis component is accepted as not being neoplastic, controversy exists regarding the nature of the ovarian component. In the original study it was considered neoplastic, but the more recent article expressed uncertainty regarding this point. Recent reports document the clinical usefulness of gonadotropin-releasing

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hormone (GnRH) agonist to achieve ovarian suppression and high doses of corticosteroids or tamoxifen in the management of these cases [6,7].

In our study, we report representative cases of these two entities, summarize evidence favoring the non-neoplastic nature of the ovarian proliferations associated with sclerosing peritonitis, and advocate use of the previously proposed term *luteinized thecomatosis associated with sclerosing peritonitis* for the latter entity to clearly distinguish it from classical and mitotically active luteinized thecoma. Our case 2 is, to our knowledge, the first case of this entity in which the ovaries were not totally excised and is thus instructive from both a biological and therapeutic standpoint.

Case reports

Case 1: mitotically active cellular luteinized thecoma

A Caucasian woman, gravida 1, para 1, had irregular periods and extreme fatigue for 6 months before developing amenorrhea at 41 years of age. On admission, the serum testosterone level was 9.5 nmol/l (3 times higher than the upper reference value for women), and computerized tomography (CT) scan of the abdomen revealed a 4 cm × 4 cm left adrenal tumor that proved to be a cyst at the time of her first laparotomy. CT scan of the pituitary gland was negative. Later, after no improvement of her condition, vaginal ultrasound showed a 6 cm × 4 cm left ovarian mass with normal echogenic density and no cystic lesions. The right ovary and uterus appeared normal. The clitoris was enlarged to 8 mm × 13 mm (the upper limit of normal size is 5 mm × 5 mm). The blood pressure was 130/85 mm Hg, weight 65 kg, height 163 cm, and body mass index (BMI) 24.5. Some terminal hairs were evident on the upper lip, chin, and back, but there was no temporal balding, acne, deep voice, or striae. She had central obesity with thin limbs and complained of muscle weakness. The patient underwent a suppression-stimulation test of the adrenal glands that showed normal adrenal function followed by a fractionized curettage and a second laparotomy with total abdominal hysterectomy and bilateral salpingo-oophorectomy at the age of 43 years. At the time of laparotomy, a left ovarian tumor was identified. The right ovary, fallopian tubes, and uterus were unremarkable. There was no evidence of ascites, adhesions, or tumor rupture. After operation, she became less tired and stated that her sex drive was less than half of what she experienced preoperatively. She received hormone replacement therapy from the 5th to 7th years postoperatively. The patient was healthy with no clinical or endocrinological evidence of neoplasm 19 years after operation.

Case 2: luteinized thecomatosis associated with sclerosing peritonitis

A 25-year-old Caucasian woman presented with severe lower abdominal pain that began a week prior to hospitalization. On admission, she had abdominal pain, nausea, and vomiting with no urinary tract symptoms or irregular bleeding. On physical examination, her abdomen was very tender with signs of peritoneal irritation. Transvaginal ultrasound showed bilateral enlarged, edematous ovaries measuring 6–8 cm in greatest dimension, as well as free pelvic and peritoneal fluid. The human chorionic gonadotropin, beta subunit (β-hCG) test was negative. Due to the clinical presentation of an acute abdomen, a diagnostic laparoscopy was performed and showed two large edematous ovaries. The omentum had an irregular edematous surface that densely adhered to the ovarian surfaces bilaterally. The laparoscopy was later converted to a laparotomy with wedge resection of the left ovarian bleeding point and partial omentecomy of the segment that adhered to the ovary. She was treated with GnRH agonist (intramuscular leuprolide 3.75 mg) to achieve ovarian suppression and high doses of intravenous corticosteroids to relieve the clinical peritonitis, followed by 5 weeks of oral steroids, resulting in complete remission of the peritoneal-related symptoms.

At the time of the second laparoscopy performed 2 months after the first one, dense fibrous adhesions involved the small and large intestines, uterus, and omentum. The ovaries were of normal size without obvious tumor. Adhesiolysis, bilateral wedge resection of the ovaries, and omentectomy were performed. She subsequently resumed normal menses, and an ultrasound examination showed normal ovaries. She conceived spontaneously and delivered a normal boy 18 months after the initial operation. She is currently living and well with normal menses 40 months after initial operation.

The past history indicated that she had been in good health prior to her present illness. There was no history of seizure disorder, anticonvulsant therapy, autoimmune disease, practolol therapy, or peritoneal dialysis.

Materials and methods

Case 1 was retrieved from the pathology files of the Department of Pathology, Rikshospitalet, University of Oslo, Oslo, Norway, and case 2 was from the files of the Department of Pathology, Meir Medical Center, Kfar-Saba, Israel. The latter case was previously reported [7]; however, additional follow up, histological illustrations, interpretations, and conclusions are provided herein. The tissue specimens were fixed in formalin, routinely processed, and embedded in paraffin. For the lipid stain in case 1, frozen sections were cut on a cryostat and stained with Oil Red O. All slides were retrospectively reviewed and diagnosed according to well-established criteria. Mitotic figures were counted in five sets of 10 HPFs, and the highest count was recorded according to the method of Irving et al. [3]. Four-micron sections were cut for immunohistochemical staining that was performed on an automated immunostainer (Dako, Carpinteria, CA). Slides were analyzed for immunoreactivity to the following antibodies: estrogen receptor (Dako), α-inhibin (Serotec, Düsseldorf, Germany), vimentin (Dako), muscle-specific actin (Dako), desmin (Dako), proliferating cell nuclear antigen (PCNA) (Dako), and MIB-1 (Ki-67) (Invitrogen, Carlsbad, CA). Appropriate positive controls for each antibody were run concurrently and showed adequate immunostaining. The clinical information was obtained from the medical records.

Results

Case 1: mitotically active cellular luteinized thecoma

The excised left ovarian tumor measured 6 cm × 5 cm × 2.5 cm, was well circumscribed with a smooth surface, and was yellow on sectioning without hemorrhage or necrosis. There was no capsular invasion macroscopically, and no sign of spread beyond the ovary. Microscopically, the tumor was composed of two cell types; i.e., spindle-shaped cells and luteinized (steroid type) cells (Fig. 1A). The spindle cell component was predominant and consisted of well-defined interlacing fascicles of plump spindle cells with round to oval nuclei and vesicular chromatin, some with small nucleoli. Most areas were densely cellular with scant intercellular substance. The luteinized cells consisted of uniform polygonal cells with eosinophilic to clear cytoplasm and a central round nucleus that sometimes contained a prominent central nucleolus (Fig. 1B). These cells were dispersed between the spindle cells in numerous small clusters, most pronounced at the periphery and usually were composed of 3–50 cells in a single plane of section. No crystals of Reinke were identified. No nuclear atypia was present in

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