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Sertoli – Leydig Tumor and Meigs' Syndrome, an Infrequent Association-A Case Report

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Abstract

The Meigs' syndrome, of infrequent presentation in approximately 4% of benign ovarian tumors, is characterized by the association of a benign tumor of gynecological origin with ascites and pleural effusion, which, in the majority of cases, induces the Clinician to suspect a malignant etiology. However, these conditions are resolved after tumor excision. Fewer than 60 cases of Meigs' syndrome have been reported in the literature to date that have a rise in the CA 125 antigen. The first case was documented by Jones and Survit in 1989 in a 70-year-old patient with a fibrothecoma and with an elevation of CA 125 of 226 IU/ml. The association of a Meigs' syndrome with Sertoli–Leydig tumors is extremely rare, we present the second case reported in the literature. The fact of being able to find the association of these two entities renders it evident that the Clinician should bear this in mind at the time of carrying out the evaluation of a patient with a pelvic injury that is bears a resemblance to advanced-stage ovarian cancer, and even more so if it presents with elevations of CA 125, in that Meigs syndrome will present complete resolution after the tumor excision.

Keywords: Sertoli-Leydig; Meigs' syndrome; Ca 125

Introduction

Sertoli-Leydig tumors are extremely rare neoplasms that represent 0.5% of ovarian neoplasms [1]. One third of patients with one of these tumors present clinical and biochemical data of virilization or defeminization [2,3], although they generally have a good prognosis [4]. These tumors were initially called arrenoblastomas, and in 1958, they were designated Sertoli–Leydig tumors [5]. On the other hand, Meigs' syndrome is characterized by the presence of ascitis and pleural effusion, associated with a benign tumor of gynecological origin [6,7]. The majority of Meigs' syndromes present as associated with fibromas, thecomas, and granulosa cells, or Brenner tumor; the association of a Sertoli-Leydig tumor with Meigs' syndrome is an extremely infrequent event; thus, the importance of reporting a case in which this association is present.

Clinical Case

Patient feminine gender 48 years of age, without antecedents of importance, who initiated with a clinical condition with seven months of evolution, characterized by abdominal distension and edema of the lower extremities without other accompanying symptoms (Figure 1). The patient made an appointment with the physician, who requested abdominal Ultrasound (US), which detected a pelvic tumor, multicystic, heterogeneous, 24.4 × 23.5 × 18.4 cm in size, 5.605 cc in volume, uterus 7 × 7 × 10.8 cm in size, with intramural myomas of 6 × 4 cm, endometrium of 8 mm, ascitic fluid, which the requirement of multiple hospitalizations due to respiratory difficulty, with the performance of three evacuatory paracenteses prior to admission at our institution (Figure 2).

On her admission, the patient exhibited anasarca with paleness and respiratory difficulty, integrating bilateral pleural syndrome, globus abdominalis at the expense of fluid with tense ascites without achieving delimitation of visceromegaly or intra-abdominal tumors, weight = 87 kg, Karnofsky scale = 50.

Paracenteses was carried out, draining 6 L of cetin fluid and with placement of a pleural catheter for lung drainage. Tumoral markers on the patient's admission to the institution were the following: CA 125, 352 IU/ml; Alpha Feto Protein (AFP), 17 ng/ml; Carcinoembryonic antigen (CEA) 0.62 ng/ml; CA 19-9, 2.9 IU/ml, and CA 15.3, 12.9 IU/ml, and hormones were estradiol, 370 pg/ml, progesterone, 0.58 ng/ml, and testosterone, 42.4 ng/dl. Cytology of the ascitic fluid and pleural fluid reported reactive mesothelium without neoplastic cells.

Computer Axial Tomography (CAT) of the thorax, abdomen, and pelvis were performed, considering that the patient was not an initial candidate for cytoreduction; thus, the decision was made to conduct a guided biopsy for diagnosis and to establish norms for treatment behavior. The biopsy was conducted with a cutting needle of the adnexal lesion without complications, reporting an ovarian stromal tumor, complementing the study with the following immunohistochemistry: AFP, negative; CD99, positive; CD10, negative; C7 EMA, negative; inhibin, positive; SALL4, negative; WTQ, negative, and IRE, positive.

Exploratory laparotomy was carried out, finding a multilobulated tumor, septated, multicystic, right ovary-dependent adhered to pelvic cavity, and right parietocolic gutter with myomatic uterus and left ovary of normal appearance. The transoperative study of the lesion reported a tumor of 29 cm with integral capsule with tumor of the sex cords.

The patient was in intensive therapy for 7 days due to important bleeding during the surgical procedure, presenting resolution of the pleural effusion and of the ascitic fluid.

The definitive pathological report demonstrated right ovary with Sertoli–Leydig-cell tumor of intermediate differentiation. Tumor size

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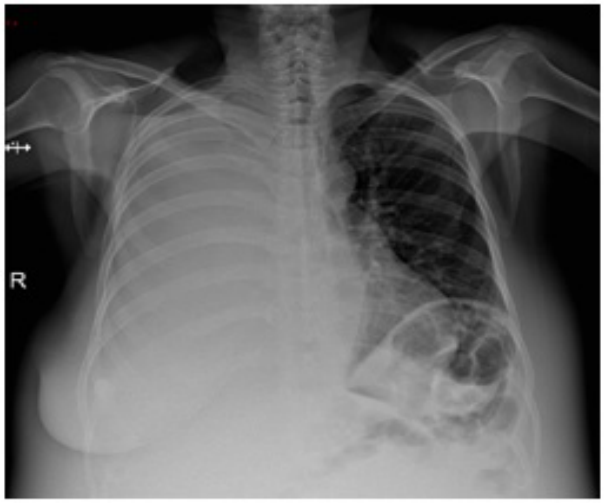

Figure 1: Teleradiography of thorax showing total collapse of the right lung at the expense of massive pleural effusion, which conditions displacement to the left of the cardiomediastinum, left lung with compensatory expansion.

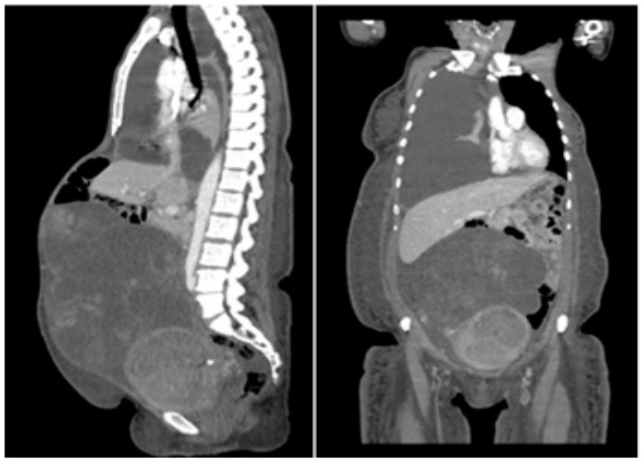

Figure 2: Coronal and sagittal reconstruction of phase-contrast Computer Tomography (CT), in which right pulmonary collapse can be observed at the expense of massive pleural effusion with displacement of the cardiomediastinum to the left. At the level of abdominal cavity and pelvis, occupation of the heterogenous neoformation dependent on right ovary, as well as implants in left mesenterinum due to carcinomatosis and uterus with increase in dimensions due to intramural myomatosis. Edema of soft tissues.

of 29 × 28 × 10 cm with integral capsule (Figure 3). Contralateral ovary with fibrous cystoadenoma and white tissues, fallopian tube with vascular congestion, slight, chronic cervicitis, basal endometrium, and conventional-type, intramural leiomyoma's 1-6 cm in diameter. The patient is found in follow-up without evidence of disease recurrence and a CA 125 marker of 4.75 IU/ml.

Discussion

Sertoli–Leydig tumors represent a very low percentage of ovarian neoplasms at the National Institute of Cancerology (INCan) in Mexico: during the 2000–2004 periods, only five cases occurred among 754 ovarian neoplasms registered [8], corresponding to 0.66%, similar to that reported in the literature [2-5,9,10]. It affects all ages, but mainly women 25 years of age [11]. Are hormonally active with estrogen secretion, which is associated with early puberty, endometrial hyperplasia, and metrorrhagia, while androgen secretion gives rise to amenorrhea, hirsurtism, mammary gland atrophy, and clitoral hypertrophy [3,12,13]. Some cases are intensely virilizing, related with elevations in testosterone of >200 ng/dl. These tumors are solid in consistency and generally unilateral, up to 98%. Only 4% present with ascitis [14] and the majority of patients have pain and abdominal distension. Elevations have been reported of the marker AFP, which have been related with the existence of an intestinal-type component, obliging the performance of differential diagnosis with germinal cell tumor and, although less frequently, with hepatic carcinoma. Immunohistochemistry allowed confirmation of the diagnosis, because it was positive for alpha-inhibin in Sertoli as well as in Leydig cells. Prognosis is general is favorable, due to that 95% of these present confined to ovary [14], with 90% survival; generally, it has a malignancy rate of 13% with intrapelvic and abdominal distension, and recurrence can present in the first 10 years, although its percentage is extremely rare, with only one case of recurrence present in the literature [15]. Poor prognostic factors associated with this neoplasm comprise the heterologous component, the presence of gastric or intestinal epithelium, carcinoid foci, or stromal elements and retiform pattern, which determine to offer of adjuvancy in the treatment [5].

On the other hand, the Meigs' syndrome, also with infrequent presentation in approximately 4% of benign ovarian tumors [16], affects middle-aged women and is characterized by the association of a benign tumor of gynecological origin with ascitis and pleural effusion [17], which, in the majority of cases, induces the Clinician to suspect a malignant etiology. However, these conditions are resolved after tumor excision. Other tumors distinct from fibroma, such as teratomas or uterine leiomyomas are associated with the Meigs' criterion, and the denomination of Pseudo-Meigs' syndrome is preferred. The distinction between Meigs' and Pseudo-Meigs' syndrome is merely academic, because therapeutic treatment is the same [18].

Multiple theories have attempted to explain the pathophysiology of ascitis and of pleural effusion from the publication of Meigs in 1960, in which the author suggests that the ascitis could be associated with the pressure exerted by the tumor itself on the lymphatic vessels, thus the accumulation in the peritoneal cavity, or associated with the exudate of the tumor itself to the peritoneum6, and up to the most recent theories, which have related the production of inflammatory cytosine's or of Vascular Endothelial Growth Factors (VEGF) and Fibroblast Growth Factors (FGF), whose values have been documented as elevated pre-operatively, in pleural fluid as well as in ascitic fluid, with a decline in these values after tumor extirpation [19-22].

Fewer than 60 cases of Meigs' syndrome have been reported in the literature to date that have a rise in the CA 125 antigen ranging between 42 and up to 7,000 IU/ml (Table I) [23-63]. The first case was documented by Jones and Survit in 1989 in a 70-year-old patient with

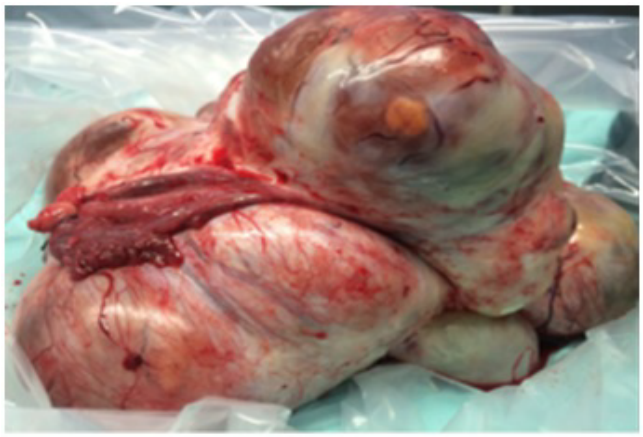

Figure 3: Right ovary with Sertoli–Leydig-cell tumor 29 × 28 × 10 cm on size with integral capsule.

Table I: Summary of reported cases of Meigs´ syndrome with elevated CA levels since 1989.

Author	Year	Age	Histopathology	Tumor size (cm/vol)	CA 125 (IU/ml)
Jones and Survit [21]	1989	70	Fibroma/thecoma	11x9x8	226
Hoffman [23]	1989	32	Thecoma	11x11x7	498
Martin et al. [24]	1990	NR	Granulosa cell tumor	NR	307
Walker et al. [25]	1990	52	Cellular fibroma	16x4x8	>5,000
	1990	67	Cellular fibroma	18x15x10	104
Le Bouëdec et al. [26]	1992	66	Fibroma/thecoma	15	645
	1992	76	Fibroma/thecoma	12	286
Williams et al. [27]	1992	74	Luteinized thecoma	15x10x9	329
Lin et al. [28]	1992	74	Fibroma	20x12x12	2,120
	1992	72	Fibroma	14x8x7	7,000
Turan et al. [29]	1993	63	Thecoma	18x9x5	744
Timmerman et al. [30]	1995	71	Fibroma	30x20.5x10	485
	1995	73	Fibroma	19x17x9	42
Aoshima et al. [31]	1995	33	Brenner tumor	NR	71
Siddiqui and Toub [32]	1995	73	Cellular fibroma	15x13x10	1,780
Abad et al. [33]	1999	51	Cellular fibroma	6x5	577
Migishima et al. [34]	2000	51	Uterine leiomyoma	2.3x24.3x 20.5	820
Chan et al. [35]	2000	13	Fibroma	20x19x10	970
Patsner [36]	2000	62	Fibroma	10	185
	2000	57	Fibroma	14	850
	2000	52	Fibroma	16	520
	2000	60	Fibroma	14	64
	2000	72	Fibroma	18	1,200
	2000	58	Fibroma	18	80
Bretelle et al. [15]	2000	71	Fibrothecoma	7x6.6	2610
Abramov et al. [19]	2001	62	Fibroma	12x13	354
Buttin et al. [37]	2001	67	Brenier tumor	11x9x6	759
Massoni et al. [38]	2001	33	Fibrothecoma	17.5x11.5	752
López et al. [39]	2002	78	Fibroma	22x8.5x20	498
	2002	68	Fibroma	18x14x10	265
Huang et al. [40]	2003	31	Sclerosing stromal tumor	7x6x6	396
Vieira et al. [41]	2003	65	Thecoma	14x12x8	319
Bildirici et al. [42]	2003	17	Sclerosing stromal tumor	25x18x15	193
M. Morillo et al. [43]	2003	59	Fibroma	22 x 17 x17	825
Cissé et al. [44]	2004	25	Fibroma	15x11x9.8	482
Salman et al. [45]	2005	56	Fibroma + Takayasu arteritis	15	509
Choi et al. [46]	2005	69	Granulosa cell tumor	6x10x12	82.49
Kurai et al. [47]	2005	79	Ovarian leiomyioma	9x7x7	163
Morán et al. [48]	2006	46	Fibroma	25x23x19	1,808
Boldorini et al. [12]	2006	26	Pure Sertoli	20	1720
Jung et al. [49]	2006	50	Sclerosing stromal tumor	19x13x10	1476.8
Benjapibal et al. [50]	2009	56	Fibroma	13x10x10	1064
Lanitis et al. [51]	2009	56	Cellular fibroma + Breast ductal carcinoma	13.5x10x8	59
Kaur et al. [52]	2009	12	Juvenile granulosa cell tumor	10x10	708
Amorim et al. [53]	2010	63	Right: Sclerosing stromal tumor Left: serous cystadenoma	4.2 x 3.7	2,168
Boufettal et al. [54]	2011	51	Fibrothecoma	7	412
Liou et al. [55]	2011	18	Sclerosing stromal tumor	14.5x13x9.5	4208.3
Monteiro et al. [56]	2012	13	Fibroma (MACF)	19x15x12	453
Su et al. [57]	2013	53	Fibrothecoma	12.2x10.7	222
Riker et al. [58]	2013	54	Fibrothecoma	15 cm	1191
Gomes et al. [59]	2013	17	Pregnancy + Sclerosing stromal tumour	volume 221.3 cm^3	625
Yazdani et al. [60]	2014	50	Fibrothecoma	12 x 10	>600
Cha et el [61]	2014	52	Fibrothecoma	NR	319.2
Park et al. [62]	2015	61	Thecoma	12x11	347

*NR = Not Reported

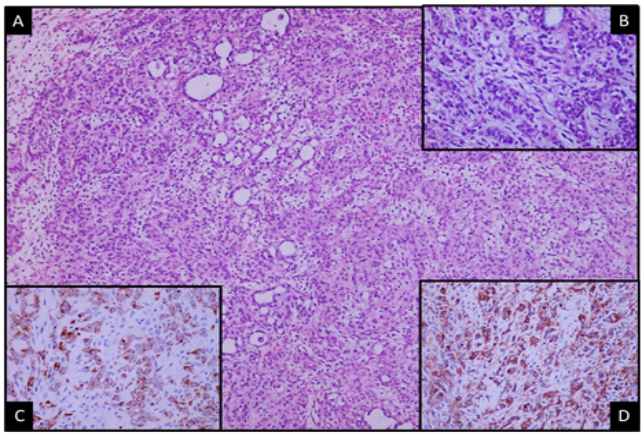

Figure 4: A) HE 4X .Low power figure shows a biphasic neoplasia with nest of Leydig cells and Sertoli cells with microcystic, nest and poorly formed tubules. B) HE20X. Cellular detail of Sertoli and Leydig components. C) IHQ Inhibin positive predominantly in Leydig component. D) IHQ CD99 positive in both components.

a fibrothecoma 11 × 9 × 8 cm in size and with an elevation of CA 125 of 226 IU/ml [21]. In our case, the patient also presented with a rise in CA 125 on her admission to our institution of 352 IU/ml. Elevation of CA 125 is considered the responsibility of the mesothelial cells, which produce the rise in response to the mechanical irritation generated by the tumor itself or by the ascitis [22].

The majority of Meigs' syndromes present as tumors, such as fibromas, thecomas, and granulosa cell tumors. Tumor size, to a greater extent than the histology itself, is found in relation to the amount of ascitis, as well as that of the pleural effusion, above all in lesions measuring >13 cm.

The presentation of the Meigs' syndrome associated with a Sertoli–Leydig-cell tumor is extremely infrequent and, on our intentionally searching in the published literature, we found only one case that had occurred prior to our own. This case was reported in 2006 by Boldorini et al., in which the authors presented the case of a young, 26-year-old patient with a pure, 20 cm Sertoli-cell tumor associated Meigs' syndrome with elevated CA 125 levels of 1.720 IU/ml [7].

Sertoli-Leydig cell tumors usually appear as solid masses, however, they may also present as heterogeneous lobulated lesions with both cystic and solid components [64], diagnostic image studies should include pelvic ultrasound and abdominopelvic computed tomography scan in selected cases [65]. Ovarian sex cord-stromal tumors may exhibit characteristic radiologic features with which radiologists should become familiar [66]. Sertoli-Leydig cell tumor are characteristic virilizing neoplasms and can easily be detected using color Doppler US rather than trasnvaginal US alone, but this presentation is small, no more than 30-50% of cases, the rest of these tumors have nonspecific appearance. On US usually present either as a distinct hypoechoic mass or a heterogeneous mass that is primarily solid with multiple cystic spaces. On CT images a soft-tissue attenuating adnexal mass is usually seen (Figure 4A-D). The solid portions characteristically exhibit avid contrast uptake [67-69]. On MRI strong hypo-intensity on T2-weighted images is not characteristic [70,71], but most of these tumors show a predominantly low signal intensity of the solid components, relating to the fibrous stroma, with some scattered cystic areas of high signal intensity [64].

Conclusion

The association of a Meigs' syndrome with Sertoli–Leydig tumors is extremely rare: this is, to our knowledge, the second case reported in the literature. Tumor size in Meigs' syndrome is related to a greater degree with the severity of the presentation of ascitis and pleural effusion than with the histology. The diagnosis should include color Doppler US and abdominopelvic computed tomography and is confirmed with immunohistochemistry positive for alpha-inhibin. Meigs syndrome will present complete resolution after the tumor excision.

Conflict of Interest

The authors declared no potential conflicts of interest with respect to the research, authorship, and publication of this article.

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