#### 639

Expression of relaxin-like factor (RLF), a novel member of the insulin-IGF-relaxin family, in the human ovary

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The relaxin-like factor (RLF), also known as the Leydig cell insulin-like factor (Ley-I-L), is a novel member of the insulin-IGF-relaxin family of hormones and growth factors which has recently been shown to be expressed in very large quantities in the Leydig cells of the testis. Expression of the RLF peptide in the human ovary has not been studied so far.

In the present study, we investigated the expression of the RLF peptide in the human ovary by immunohistochemistry using a specific antibody raised against human RLF. By this method, RLF was found to be expressed in theca interna cells, while it was absent from both theca externa and granulosa cells, as well as from the ovarian stroma and surface epithelium. RLF expression could also be observed in the corpus luteum, although at a lower level than in theca interna cells.

We are currently investigating the expression of RLF in ovarian tumors. So far, a number of 19 sex cord-stroma tumors (6 granulosa cell and 17 theca cell tumors) and 4 cases of hilus cell hyperplasia have been investigated. RLF was found to be expressed in Leydig/hilus cell hyperplasia and sex cord-stroma tumors with a component of Leydig or luteinized cells. Of the analysed theca cell tumors, two displayed a diffuse staining pattern. As expected, RLF was not expressed in granulosa cell tumors.

In conclusion, RLF appears to be a useful marker for theca interna and Leydig cells in the human ovary and may be a diagnostic supplement in hyperplasias and tumors derived thereof.

### 640

## MALIGNANT DIFFUSE PERITONEAL LEIOMYOMATOSIS A CASE REPORT

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Diffuse peritoneal leiomyomatosis (DPL) is a rare condition characterized by the presence of multiple abdominal smooth muscle nodules. Malignant transformation appears to be extremely rare. This report presents the eighth case with proven malignancy.

The patient is a 43 year old, Caucasian, non-pregnant female, with no history of hormonal therapy, presented with a few months old history of intermittent and increasing right lower abdominal pain. In October 1996, explorative laparatomy was performed. Innumerable subperitoneal gray-white masses with firm to rubbery consistency, varying in size from 0.2 to 11.5 cm, were found scattered over the parietal peritoneum, omentum and mesentery. Several of these nodules were removed. Their microscopic appearance was variable. Most of the lesions appeared to consist of subperitonel nodules of benign-appearing smooth muscle cells. Nevertheless, some of them showed malignant characteristics, marked cellularity, and numerous mitoses. Immunohistochemical and ultrastuctural studies proved the smooth muscle origin of the tumors. During the next few months the abdominal tumors increased in size and the patient's condition progressively deteriorated. A second laparatomy was performed in March 1997. More than 40 nodules were removed together with a small bowel segment where inflammatory fistula was found. After receiving three courses of chemotherapy, the patient refused further treatment. In December 1997, the patient was admitted to the hospital with acute abdominal pain due to ileus, and the third laparotomy was performed. This additional case indicates that DPL has a low, but definite malignant potential, and suggests that meticulous examination of histologic material and careful follow up are required.

#### 641

# PRIMARY YOLK SAC TUMOR OF THE ENDOMETRIUM

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A 49-year-old woman was admitted with abnormal vaginal bleeding of 5-months duration. Hysteroscopy showed polypoid formations in the uterine cavity. Biopsy of a polyp revealed an anaplastic malignant tumor. A total hysterectomy with bilateral salpingo-oophorectomy and iliac lymphadenectomy were performed. A polypoid soft white-yellowish tumor originated from the endometrium. The biopsies plus sections of the operative specimen were histologically similar and displayed typical features of yolk sac tumor (YST) with tubulo-papillary pattern. Schiller-Duval bodies and PAS-positive, diastase resistant, hyaline droplets were present. There was no neoplastic involvement of the cervix, ovaries, and iliac lymph nodes. Tumor cells and hyaline globules were strongly immunoreactive with alpha-foetoprotein antibody. The patient refused chemotherapy and was subsequently treated by external radiotherapy. The patient remained free of disease 28 months after surgery. There are only four additional cases of primary YST of the endometrium reported in literature. On the basis of the small number of cases reported, such tumors appear to have clinical and pathological features similar to their ovarian counterparts.

#### 642

Uterus-like masses: four cases and a review of twelve cases in the literature.

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Malformations of the Mullerian duct system are rare, few more so than the uterine-like mass, which typically presents as an adnexal or pelvic cystic structure containing an 'endometrial' cavity surrounded by smooth muscle.

AFIP files back to 1970 were culled for this entity: 4 cases were identified. An additional 12 cases were found in the literature; reported sites included conus medullaris (2), involving spina bifida (1), small bowel wall (1), 'intra-ovarian' (3), and five cases reported as 'uterus solidaris' involving unicornuate uteri involved atretic fallopian tubes. All masses were grossly separate from the uterus.

Age range was 12 to 59 years; 3 of 4 AFIP cases arose in the right broad ligament and one, in an ovarian remnant. The four AFIP cases showed cystic cavities lined by disorganized endometrial glands and stroma, surrounded by myometrial-type smooth muscle. Immunohistochemical evaluation of 2 cases showed the glands to react with cytokeratin, estrogen, and progesterone receptor antibodies; smooth muscle was ER/PR and smooth muscle actin positive. All patients clinically are well at last follow-up.

Uterus-like masses are rare, benign entities. Origin as a metaplastic versus dysgenic phenomenon is still debated. Arguments for a metaplastic origin are based upon previously reported, intra-ovarian masses. Our results suggest a dysgenic origin, possibly from entrapped Mullerian rests, is more likely.