Journal of Korean Medical Science
Vol. 11, No. 1, 84-87 February 1996

Ovarian Serous Cystadenoma Associated with Sertoli-Leydig Cell Tumor

—A Case Report—

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We Describe a case of ovarian serous cystadenoma having Sertoli-Leydig cell tumor, well differentiated, in the cystic septum. Well differentiated Sertoli-Leydig cell tumor coexisting with other tumor, including serous tumor, has not yet been described. In all cases of Sertoli-Leydig cell tumor with heterologous components or other tumors, the androblastomatous components are intermediately or poorly differentiated. The present case revealed a well differentiated Sertoli-Leyding cell tumor arising in a septum of serous cystadenoma, as a circumscribed nodule. With these finidngs, we discuss the possibility of this Sertoli-Leydig cell tumor, considered a mural nodule, which is well established in cystic common epithelial tumors of the ovary.

Key Words : *Sertoli-Leydig cell tumor, Serous cystadenoma, Ovary*

INTRODUCTION

Sertoli-Leydig cell tumors(SLCTs) of the ovary, by themselves, are rare and account for less than 0.2 % of all ovarian neoplasms(Young et al., 1994). Heterologous components are noted in about 20~25 % of SLCTs of intermediate and poor differentiation(Zaloudek, 1994). SLCTs have rarely been reported in association with other ovarian neoplasms, including mature cystic teratoma(Seidman et al., 1989), and mucinous neoplasms as a composite form(Waxman et al., 1981). Well differentiated SLCTs have not been described in combination with other components or tumors.

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This work was supported by a Research Grant from the Catholic Medical Center.

We describe a SLCT, well differentiated, arising within a septum of ovarian serous cystadenoma. On the basis of a single and well circumscribed mass, within a septum, we suggest a mural nodule as an possible name for this SLCT, although the pathogenetic mechanism might be different from the usual spindle cell mural nodule of common epithelial tumors.

CASE REPORT

A 46-year-old woman, P2G4, was admitted to St. Paul's Hospital, Catholic University Medical College, Seoul, Korea, due to intermittent lower abdominal pain and urinary frequency for 20 days. She had had irregular menses but usually normal flow lasting 5~7 days. She had had high blood pressure for several years. On physical examination, a large mass was palpated in the lower abdomen. A CT of the pelvis showed a huge multiseptated cystic mass occupying the lower abdomen and pelvic cavity. On pelvic exploration, a large cystic mass, 25 cm at maximum

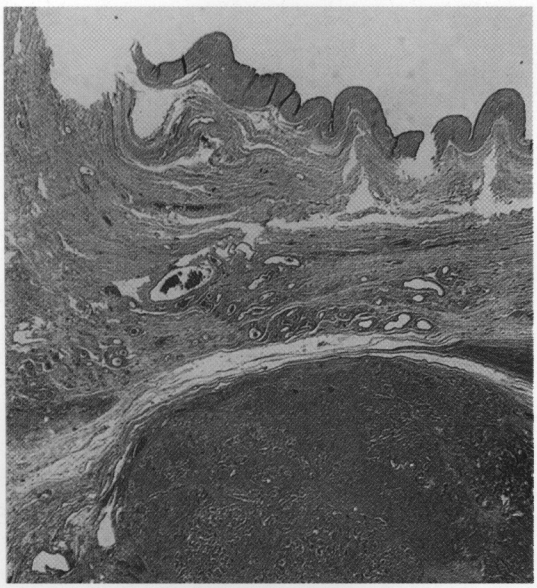

Fig. 1. A lobulated nodule of Sertoli-Leydig cell tumor within one thick septum, andoverlying simple serous epithelium.

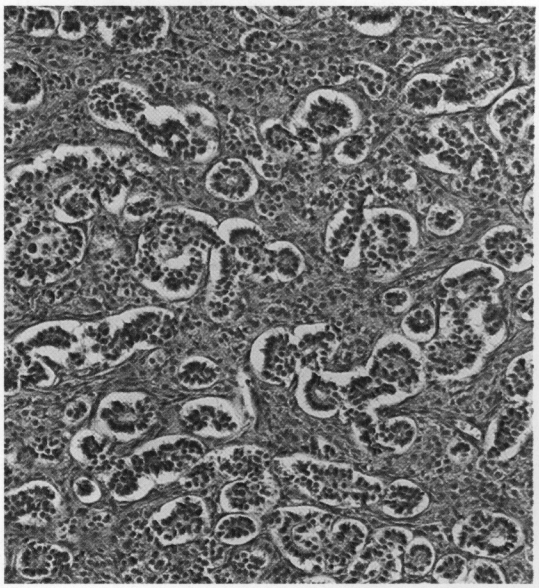

Fig. 2. Well differentiated Sertoli-Leydig cell tumor. Hollow or closed tubules of Sertoli cells and Leydig cells lying among the tubules.

diameter, arising in the left ovary, was noted. Serum CA125 was 51.66, and alpha-fetoprotein and CEA were within normal limits.

Gross Findings

The left ovarian mass, 25 cm in diameter, showed multiple locules, the majority of which were large, up to 8 cm in diameter, on section. The septae were fibrotic and thin, ranged from 0.3 to 0.5 cm in thickness, except in one area which had an ovoid yellow well demarcated nodule, 1.5 × 1 cm in size. The cystic locules showed smooth inner surfaces, and contained serous or serosanguinous fluid. The uterus, right ovary and both fallopian tubes were unremarkable.

Microscopic Findings

A lobulated ovoid nodule of SLCT was noted in one cystic septum, which was lined by simple ciliated cuboidal epithelium(Fig. 1 and 3). Occasionally, old hemorrhage or hemosiderin-laden macrophages were noted in subepithelial stroma. The nodule in the locular septum was a typical form of well differentiated SLCT, which disclosed hollow or closed tubules, and solid areas of Sertoli cells in lobular pattern. Leydig cells were lying among the tubules(Fig. 2).

Immunohistochemically, the Sertoli and Leydig cells were stained for alpha-fetoprotein and neuron-specific anolase. The vimentin was only positive for interstitial stromal tissue including Leydig cells(Fig. 4). The cytokeratin, EMA, S-100 protein and chromogranin were negative.

DISCUSSION

SLCTs are rare and comprise less than 0.2 % of all ovarian neoplasms. Approximately 20~25 % of SLCTs contain heterologous components and these are otherwise intermediate or poorly differentiated. Among the heterologous elements, gastrointestinal type mucinous epithelium is most frequently revealed, but other tissues including carcinoid, skeletal muscle and cartilage also occur(Young and Scully, 1985). Endodermal heterologous elements tend to be associated with SLCT of intermediate differentiation, whereas mesodermal heterologous elements are associated with tumors of poorly differentiated androblastomatous components(Prat et al., 1982b ; Young et al., 1982). In this case, the SLCT was a single nodule with a sharp demarcation from the surrounding stroma and the overlying serous epithelium, and was well differenti-

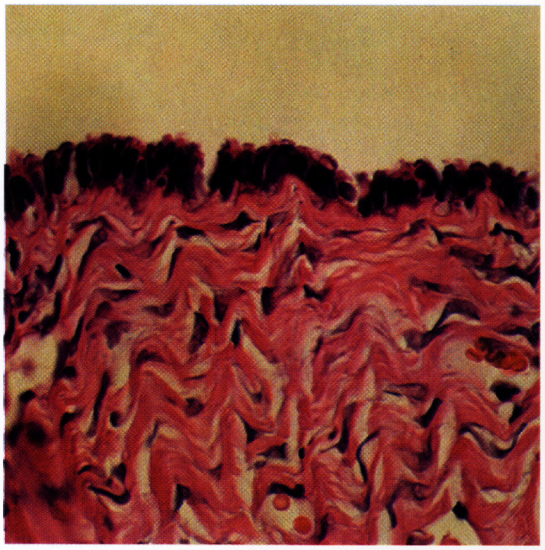

Fig. 3. Simple ciliated serous epithelium and fibrous wall of cystic septum.

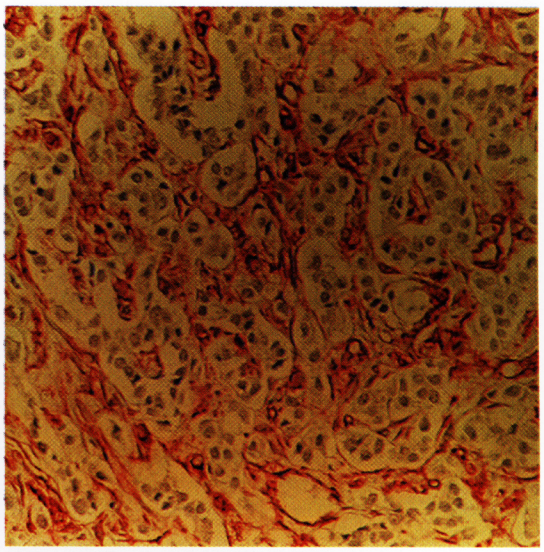

Fig. 4. Vimentin positivity is noted only in the stroma between the tubules.

ated histologically. On the basis of these findings, SLCT with heterologous component is not a compatible name for the present case.

Considering the origin of SLCT, Sertoli cells are assumed to arise from undifferentiated cells of sex cord origin, and at this time, Leydig like component represents a non-neoplastic stromal response to a Sertoli cell neoplasm(Sternberg and Dhurandhar, 1977). Alternatively, those who consider that both components of SLCTs are neoplastic, regard primitive medullary mesenchyma which is differentiating along the two pathways, as the origin of this tumor(Langley and Fox, 1987). Therefore, it is very unlikely to that the SLCT mural nodule may have originated from the differentiation of stoma in the cystic septum, in present case.

More than 50 cases of mural nodules in common epithelial tumors of the ovary have been reported(Prat et al., 1982a ; Czenobilsky et al., 1983 ; Tsujimura and Kawano, 1992) under various confusing names, since it was first described by Prat and Scully(Prat and Scully, 1979). To clarify the lesions, Baergen and Rutgers defined mural nodules as follows ; They are associated with a common epithelial tumor of the ovary. The nodules are well cirumscribed within the cystic epithelial tumor grossly. There should be no or only focal admixture of the elements of the nodule and the epithelial tumor microscopically. Finally, the mural nodules may consist of reactive, benign neoplastic, or malignant cells(Baergen and Rutgers, 1994). Benign tumors in mural nodules are extremely rare, only one leiomyoma has been reported(Lifschitz-Mercer et al., 1990).

The pathogenesis of mural nodule in common epithelial tumor is not obvious. These lesions are heterogeneous, so a single explanation may not be possible. In the case of sarcoma-like epulis-type nodule, the stromal reaction to the hemorrhage or extracellular mucin is thought to be a pathogenetic mechanism(Prat and Scully, 1979). The undifferentiated mesenchymal tissue of the stroma which has multipotential ability for differentiation, is the most likely candidate for the origin of sarcomatous nodules(Tsujimura and Kawano, 1992), and under certain condition, of carcinomatous nodule, too(Fujii et al., 1985). The unique case of benign neoplastic mural nodule in the literature is leiomyoma(Lifschits-Mercer et al., 1990). They explain the pathogenetic mechanism of this myomatous mural nodule in mucinous cystadenoma as a collision tumor of two different neoplasms.

This case arose in a septum of serous cystadenoma with a good demarcation. By the definition of Baergen and Rutgers(Baergen and Rutgers, 1994), and as an example of leiomyomatous nodule, the

SCLT of the present case might be another benign mural nodule and the first sex cord stromal nodule of common epitheial tumor in a wide sense, although the most probable pathogenetic explanation for this case seems to be that of coincidental development of SLCT in a septum of a serous cystadenoma.

In conclusion, this case describes a very unusual occurrence of SLCT in a septum of a serous cystadenoma, and we discuss the possibility of considering this SLCT as a mural nodule.

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