

OSSEOUS METAPLASIA IN BORDERLINE PAPILLARY SEROUS CYSTADENOFIBROMA OF THE OVARY: A Case Report

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SUMMARY

We report a case of osseous metaplasia in borderline serous papillary cystadenofibroma of the ovary. Serous tumors of the ovary are common cystic neoplasms. In some serous neoplasms, the fibroblastic stromal component is unduly prominent, appearing grossly as solid, white nodular foci in a cystic neoplasm. These serous neoplasms is classified as cystadenofibroma and can be separated into benign, borderline and malignant types. The borderline category of this neoplasm extremely rare. The occurrence of osseous metaplasia within an ovarian papillary serous cystadenofibroma is very rare. So far only two cases of osseous metaplasia associated with a serous cystadenocarcinoma of the ovary¹ have been reported. The present report is, therefore, a first case of osseous metaplasia in an ovarian borderline serous papillary cystadenofibroma.

KEY WORDS: Osseous metaplasia, ovarian borderline serous papillary cystadenofibroma

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INTRODUCTION

Serous ovarian tumors are common cystic neoplasms and made up about one fourth of all ovarian tumors. In some serous neoplasms, the fibroblastic stromal component is unduly prominent, appearing grossly as solid, white nodular foci in an cytic neoplasm. Such tumors

can be separated into benign (adenofibroma and cystadenofibroma), borderline and malignant (adenofibrocarcinoma and cystadenofibrocarcinoma) types^{2,3}.

Borderline serous cystadenoma exhibit increased architectural complexity and epithelial cell stratification. Benign serous tumors that have cysts and the papillae are lined by a single layer of cells, without atypia, architectural complexity or invasion. The borderline category of this neoplasm are extremely rare and has been documented by Kao and Norris⁴.

Mature bone can be found in neoplasm of the ovary⁵. The most frequent ovarian tumor containing osseous elements is the teratoma^{5,6}. Heterologous mixing with mesodermal tumors of the ovary may also be associated with bone formation⁵. Although primary ovarian osteomas have rarely been reported and an occasional ovarian fibroma has been noted to have areas of ossification⁵.

The present report introduces an additional patient with an ovarian borderline serous papillary cystadenofibroma containing mature bone associated with bone marrow.

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CASE HISTORY

The patient was a 72 year old woman who complained of mild lower abdominal pain and discomfort for two months prior to evaluation. She had ascites and weakness. Routine biochemical blood parameters and tumor markers were all normal. Ultrasound and CT evaluation revealed a right sided multilocular cystic mass in 26 x 23 x 12 cm in size, sharply delineated and lobular in appearance. The mass was seen to be extending from the right ovary to upper part of the abdomen. The left ovary was in a size of 5.5 x 3.5 x 3 cm and also cystic.

The patient subsequently underwent a total abdominal hysterectomy with bilateral salpingoopherectomy and resection of appendix. In general, the patient postoperatively did well.

Pathologic examination: Macroscopic evaluation of the right ovarian cystic mass showed multilocular appearance and filled with serous

fluid. The firm elastic wall was 0.2 to 0.6 cm in thickness. The inner layer of the cyst wall was shiny, whitish and had papillary formation extending into the lumen. Left ovary also had a cystic mass with similar structure and appearance. The tumor demonstrated a histological structure of nested epithelial cells with a moderate amount of cytoplasm and enlarged-hyperchromatic-disorganized nuclei (Fig1). Papillary formation can be identified in many areas of the tumor mass (Fig 2). The cyst wall consisted of loosely textured fibrous tissue containing psammoma bodies and had no invasion. Foci of osseous metaplasia were seen in the ovarian stroma adjacent to areas of epithelial lining (Fig 3, 4). A transformation from psammoma body clusters into bone matrix was not observed.

DISCUSSION

Osseous tissue can be found in neoplasm of

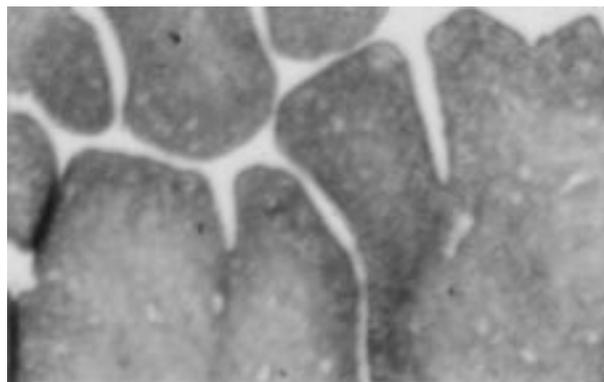


Figure-1: The stalk of papillary formation consist of fibrous tissue with thin walled blood vessels (Hematoxylin-eosin, x 48).

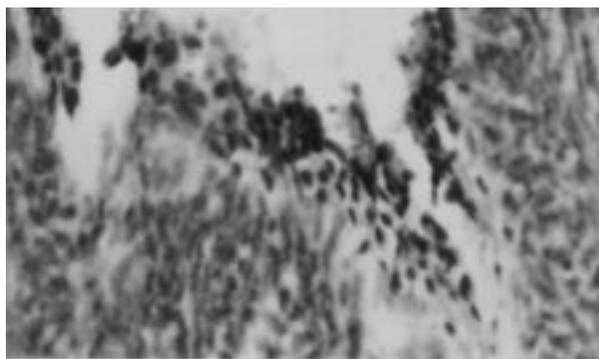


Figure-2: The lining epithelia showed nested epithelial cells with moderate amount of cytoplasm and enlarged hyperchromatic nuclei that arranged irregularly (Hematoxylin- eosin, x 240).

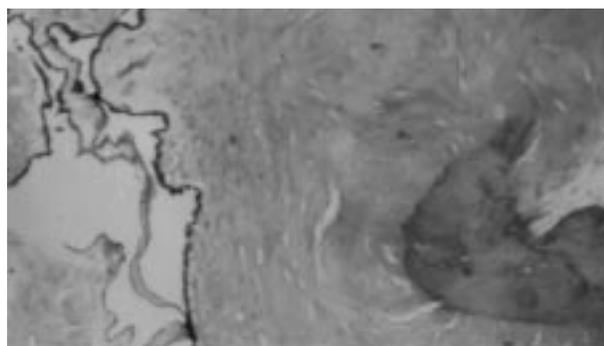


Figure-3: Foci of osseous metaplasia was seen adjacent to areas of epithelial lining (Hematoxylin-eosin, x 120).

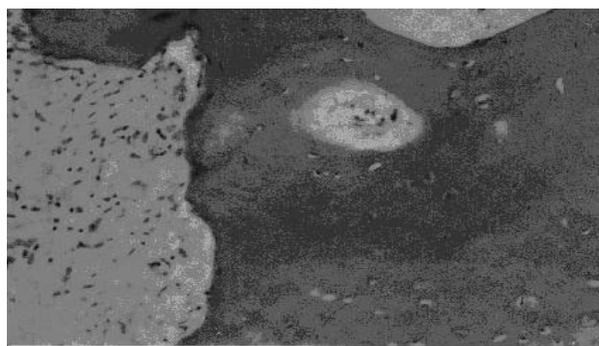


Figure-4: The lamellar bone surrounded rim of osteoblast & included bone marrow (Hematoxylin- eosin, x 240).

the ovary⁵. It is unusual in the absence of an ovarian neoplasm, and typically occurs within periovarian adhesion or the walls of endometriotic cysts, but rarely within normal ovaries⁵. The most frequent ovarian tumor containing osseous elements is the teratoma^{5,6}, but exceptionally osseous metaplasia develops in these stroma-rich tumors⁷. There have also been reported several cases of ovarian mucinous cystadenomas with bone formation^{8,9}. Only two cases of osseous metaplasia associated with a serous cystadenocarcinoma of the ovary are reported to have been published to date¹.

Because the osseous metaplasia in an ovarian papillary serous tumors which is seen to be rare the histogenetic evaluation of osseous metaplasia can be questionable. An osteoinductive agent synthesized by the tumor cells, such as transforming growth factor- β , has been implicated in osteogenesis within extraskletal neoplasms¹⁰.

In the present case, the benign osseous elements are found in small isolated foci located in the stroma. The proposed explanation for this tumor involves in the initiation of bone formation by extension or growth of already existent psammoma bodies in the serous tumor¹. Psammoma bodies are made up of microcrystals very similar to the calcium phosphate crystals of bone¹¹.

Through joining together with adjacent psammoma bodies, an area of calcification similar to bone matrix could be formed. An argument against such a theory is that no transition from psammoma bodies to bone elements has ever been found. Psammoma bodies are very common findings in ovarian papillary serous tumors, but evidence of bone formation is extremely rare. This suggests that such a relationship between psammoma bodies and bone formation probably does not exist⁵.

The most plausible explanation for bone formation in an ovarian papillary serous cystadenocarcinoma is a metaplastic process of the multipotential stromal cell. Osteoma and chondromas of the ovary are rare and most likely to occur by a metaplastic process of the

ovarian connective tissue⁷.

In the present case, the presence of prominent fibroblastic stromal component implies to support this theory. Possible explanations of the stimulus causing such metaplasia are beyond the scope of this paper, but we speculated that this may be related to the presence of desmoplastic stroma in serous papillary tumors which is also undergoing to osseous metaplasia due to exuberant fibrous tissue proliferation.

According to the previous reports, this case was unique since it is a first reported case of borderline serous papillary cystadenofibroma with osseous metaplasia of the ovary. The unusual metaplastic bone in the ovarian tumors described herein is in accordance with the current concepts of heterotopic bone formation.

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