Abstract A 51-year-old woman presented with lumbar backache leading to the preoperative diagnosis of a right solid adnexal mass with calcification on computed tomographic scan. Histological examination revealed a right ovarian luteinized thecoma characterized by extensive calcification and metaplastic ossification. Osteoblasts and osteoclasts surrounded the surface of the heterotopic bone. Haversian canals were occasionally identified in the bony trabeculae. Ossifying ovarian neoplasms are extremely rare and this case is the first to demonstrate the osseous metaplasia in ovarian luteinized thecoma.

Keywords Bone · Calcification · Luteinized thecoma · Osseous metaplasia · Ovary

Introduction

The heterotopic bone formation is extremely rare in ovarian neoplasms. To our best knowledge, only four cases of ossifying ovarian neoplasms have been reported in the literature, including serous cystadenocarcinoma [1], mucinous cystadenoma [2] and well-differentiated Sertoli-Leydig cell tumor [3]. However, no ovarian thecomas have been documented to show evidence of the osseous metaplasia. We describe an unusual case of ossifying luteinized thecoma with extensive stromal calcification.

Case report

A 51-year-old woman presented with lumbar backache in October 2000. Pelvic examination revealed a right firm adnexal mass. Cytology of the cervix and endometrium showed no atypical cells, and serum tumor markers, including serum CA724, SCC and CA125, were within normal limits. Ultrasound examination demonstrated a solid hypoechoic mass with an acoustic shadow. Computed tomographic (CT) scan displayed a right adnexal mass with calcification (Fig. 1). She underwent the right oophorectomy.

Macroscopically, the cut surface was solid and yellow in color without apparent hemorrhage and necrosis. Histological examination revealed ovarian thecoma composed predominantly of luteinized thecoma cells and partly of typical thecoma cells. Typical thecoma cells were spindle-shaped with ill-defined, pale scant cytoplasm and round nuclei, whereas luteinized thecoma cells had ill-defined, eosinophilic or vacuolated cytoplasm with central oval or round nuclei and prominent nucleoli (Fig. 2). Sudan III stain revealed abundant intracellular lipid in these cells. The mitotic figures were 2–3 per 10 high power-fields identified in the cytoplasm of tumor cells. Foci of calcification were scattered in the abundant stroma, but no psammoma bodies were noted. The meta-
plastic bone was mostly observed adjacent to typical thecoma cells at the periphery of the tumor. The heterotopic bone was rimmed by osteoblasts and osteoclasts at the surface, and intense fibrous proliferation surrounded bony spicules (Fig. 3). Haversian canals were occasionally identified in the mature bony trabeculae (Fig. 4). No teratomatous elements were present in the tumor.

Discussion

The present case appears to be the first description of ossifying ovarian luteinized thecoma with extensive stromal calcification. Although only four cases of typical thecomas have been reported to show extensive plaques of stromal calcification and/or psammoma bodies \[8\], ossifying ovarian thecomas have not been described previously.

Takemori et al. \[6\] reported the characteristic magnetic resonance imaging findings of ovarian thecoma that demonstrated homogeneous low signal intensities on T1-weighted images and predominantly high signal intensities on T2-weighted images. However, the differentiation of thecoma from other ovarian tumors by ultrasound and CT scan is not possible in many cases. In particular, the present case demonstrated an unusual CT appearances mimicking teratoma. Two cases of coexisting ovarian tumors composed of thecoma and mature teratoma were reported \[4\], but a coexistence of teratoma was definitely excluded in our case because no teratomatous elements were observed.

Although the osseous metaplasia was documented in serous cystadenocarcinoma \[1\], mucinous cystadenoma \[2\] and Sertoli-Leydig cell tumor \[3\], the mechanism underlying the heterotopic bone formation remains unknown. Takeda et al. \[5\] observed differentiation of mesenchymal cells into osteoblasts with formation of new bone trabeculae in the stroma of ameloblastoma. Furthermore, transforming growth factor ß, paracrine osteoinductive growth factor, has been reported to stimulate the osteoblastic differentiation of mesenchymal progenitor cells and metaplastic bone formation in giant cell tumor of bone \[7\]. In the present case, therefore, transforming growth factor ß or unknown osteoinductive factors may play an important role in promoting the osseous metaplasia of undifferentiated mesenchymal stromal cells with pluripotent ability for differentiation.

In conclusion, we illustrated an unusual ovarian luteinized thecoma with prominent stromal ossification and calcification. Further investigations of the effects of cytokines on the osteoblastic differentiation of undifferentiated mesenchymal stromal cells may elucidate the pathogenesis of the osseous metaplasia in ovarian tumors.

References