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Bilateral massive osseous metaplasia in ovaries: “ovarian stones”

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Abstract We report a case of bilateral ovarian ossifications with images that mimic stone formations. A 65-year-old woman presented with a 2-year history of pelvic pain. Computed tomographic scan and pelvic ultrasound identified an enlarged uterus with two calcified lesions interpreted as leiomyomas. Surgical exploration revealed two ovaries with a cystic appearance and stony hard areas. The cyst contents consisted of chocolate-colored material. The pathologic findings were compatible with benign bilateral ovarian endometriotic cysts with extensive ossification. The pelvic pain resolved completely after the surgery. Though the cause of this rare case remains unknown, recognition of cysts with a content of chocolate-colored material and pigment-laden histiocytes allowed us to make the diagnosis of bilateral ovarian endometriotic cysts with extensive ossification. Complete excision was the treatment of choice.

Keywords Ovary · Ossification · Endometriotic cyst

Bilateral massive osseous metaplasia in ovaries: “ovarian stones”

In the absence of an ovarian neoplasm, extensive ossification and calcification involving the whole ovary is an unusual occurrence. It may develop within periovarian adhesions or the walls of endometriotic cysts and rarely within otherwise normal ovaries [1]. We describe a rare case of bilateral massive ossification in ovaries with a cystic appearance.

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Case report

A 65-year-old woman, with a previous history of cholecystectomy, presented with a 2-year history of pelvic pain leading to the preoperative diagnosis of two highly calcified uterine masses interpreted as leiomyomas on computed tomographic (CT) scan (Fig. 1). Also pelvic ultrasound confirmed an enlarged uterus with two calcified lesions. Laboratory investigations, including serum calcium levels, were in the range of normality. Unexpectedly, surgical exploration revealed bilateral enlarged, ovoid, pale brown ovaries, closely connected with the posterior surface of the uterus and looking like ovaries made of stones. Therefore, the patient underwent a hysterectomy and bilateral salpingo-oophorectomy. On gross examination the left ovary measured 5.5×3.5×2 cm and the right ovary measured 2.5×2×1.3 cm. The cut surface revealed in both ovaries a cyst with stony hard areas, a diffusely gritty texture, and a yellow surface (Fig. 2). The cyst contents consisted of chocolate-colored material. Both ovaries, which required 2 days of decalcification, were entirely embedded for histologic examination. Contrary to the preoperative CT scan and pelvic ultrasound diagnosis, both on gross and histological examination, the uterus and the fallopian tubes were unremarkable. Instead, histological examination revealed in both ovaries a cyst without epithelial lining cells, but surrounded by dense fibrous tissue, a diffuse infiltration of pigment-laden histiocytes (Fig. 3), and extensive calcification with areas of metaplastic ossification (Fig. 4). Osteoblasts and osteoclasts surrounded the surface of the heterotopic bone. Haversian canals were occasionally identified in the bony trabeculae. Many corpora albicantia were also evident in the ovarian stroma (Fig. 5). The ovarian surface was characterized by fibrous tissue and inflammatory cells that justified the close connection with the posterior surface of the uterus. In conclusion the morphological aspect was highly suggestive, but not conclusive, for the diagnosis of bilateral ovarian endometriotic cysts with extensive ossification. The pelvic pain resolved completely after the surgery.



Fig. 1 Two highly calcified masses interpreted as uterine leiomyomas on computed tomographic scan

Discussion

Focal calcifications are quite common in neoplastic and non-neoplastic diseases of the ovary. They are usually associated with serous tumor, mucinous tumor, dermoid tumor, fibroma, thecoma, and gonadoblastoma. Other ovarian non-neoplastic diseases such as torsion, infarction, and hypoplasia may be associated with ovarian calcification [2]. On the other hand, massive ovarian calcification, discernible at gross examination, is rare. From the review of the literature we found only one case of extensive, bilateral, multifocal calcification of the ovarian stroma with no apparent cause [3]. Microscopic examination showed, in fact, numerous spherical, laminated, calcific foci without accompanying cells. In cases like this one, occasional neoplastic cells must be sought in order to exclude tumors such as a serous borderline or malignant tumor with confluent psammoma bodies [4] and a gonadoblastoma replaced by laminated calcified masses, but with evidence of abnormal gonadal development. Also ossification of an ovary is extremely rare and usually associated with ovarian cystic teratoma [5], mucinous cystadenoma [5], papillary serous cystadenocarcinoma [6], endometrioid adenocarcinoma [7], thecoma [8], and endometriosis [9]. In fact, a common manifestation of ovarian endometriosis is cysti-

Fig. 2 On gross examination both ovaries revealed a cyst with stony hard areas, a diffusely gritty texture, and a yellow surface

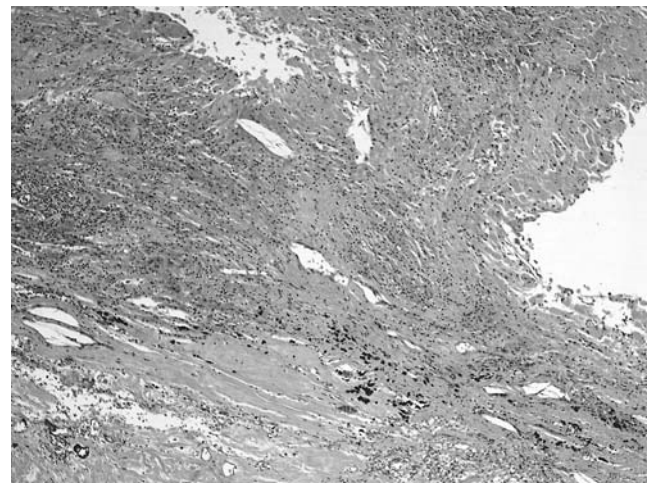
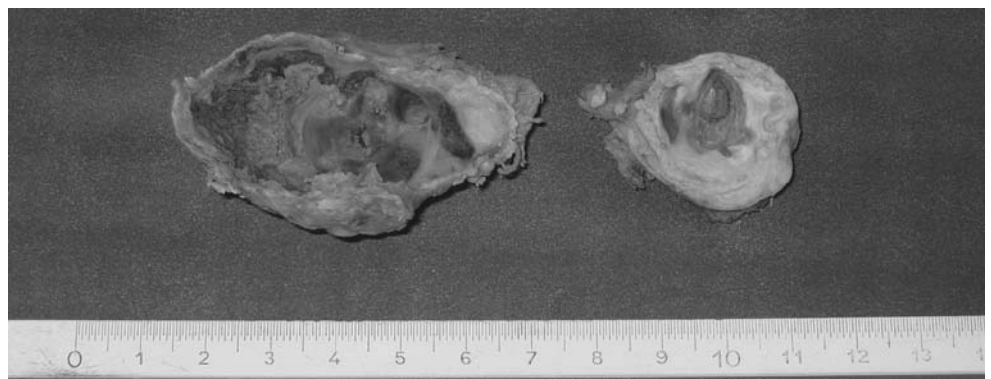


Fig. 3 Diffuse infiltration of pigment-laden histiocytes (E.E., ×10)

fication resulting in an endometriotic cyst. In many circumstances the epithelial lining becomes attenuated and recognition of an endometrial cyst may only be possible if a rim of subjacent endometrial stroma persists. Commonly in old endometrial cysts, the lining of the endometrial epithelium and stroma is totally lost and replaced by granulation tissue, dense fibrous tissue, and variable amounts of pigment-laden histiocytes. In those cases, ossification and calcification may be observed. In our case, recognition of bilateral cysts without epithelial lining cells, but with a content of chocolate-colored material, surrounded by dense fibrous tissue, a diffuse infiltration of pigment-laden histiocytes, and extensive calcification with areas of metaplastic bone, suggested the diagnosis of bilateral ovarian endometriotic cysts with extensive ossification. In the literature there is also a case of osseous metaplasia in a benign ovarian cyst in association with a complex urogenital malformation. In this case, histological examination revealed a tubal cyst with chronic salpingitis and a simple follicular ovarian cyst, in the wall of which osseous metaplasia was noted [10]. All those cases raise the question about the pathogenesis of osseous metaplasia in neoplastic and non-neoplastic diseases of the ovary. Various hypotheses have been suggested for this phenomenon; in some circumstances, such as torsion and infarction of the ovary, it appears to be an unusual reaction

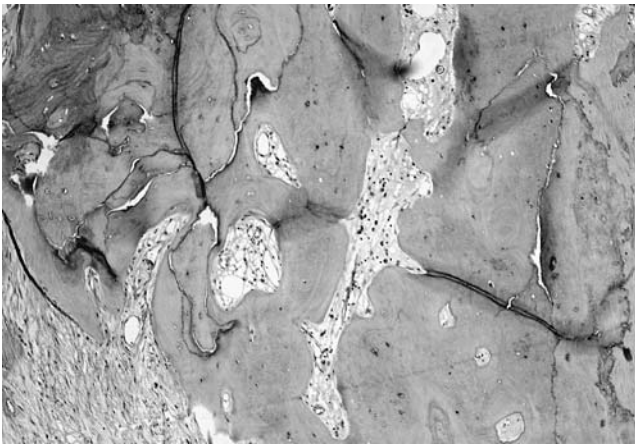


Fig. 4 Extensive calcification with areas of metaplastic ossification (E.E., ×10)

to tissue damage and repair. The overgrowth of coexisting coalesced psammomatous calcifications is another plausible explanation, but not all diseases of the ovary have this type of calcification [11]. The histogenesis of psammoma bodies is also not very clear. Some authors believe they may arise from spontaneous or induced necrosis of the tissue; for others they are related to secretion of a family of growth factors, called bone morphogenetic proteins. These factors may induce osseous metaplasia of multipotential stromal stem cells with psammoma body and bone formation in ovarian cancer [7].

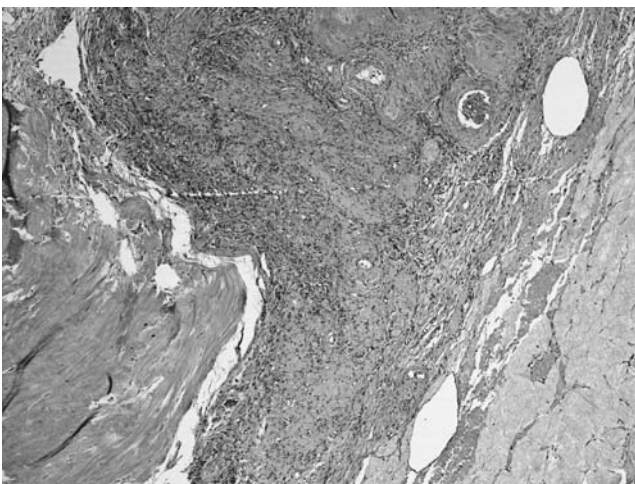


Fig. 5 Coexistence of corpus albicans (*right*) and calcification (*left*) (E.E., ×10)

In conclusion, our case of bilateral ovarian ossifications is interesting because even if a correlation with neoplastic diseases does not exist, also non-neoplastic diseases of the ovary are difficult to identify because we could not find any epithelial lining cells in the luminal cysts. Nevertheless, recognition of cysts with a content of chocolate-colored material and pigment-laden histiocytes allowed us to make the diagnosis of bilateral ovarian endometriotic cysts with extensive ossification. The calcification was not related to hypercalcemia because serum calcium levels were in the range of normality. Also a diagnosis of uterine leiomyomas was made based on the computed tomographic scan and pelvic ultrasound, and only surgical intervention with gross and histological examination supplied the right diagnosis. Surgical intervention is the treatment of choice in cases of adnexal masses presenting with extensive calcification and/or ossification.

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