Case Report

Mucinous cystadenoma with ossification: An uncommon finding in a common neoplasm

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ABSTRACT

Lamellar bone formation (osseous metaplasia/heterotopic bone formation) is an unusual occurrence in the ovary, with the exception of mature cystic teratoma. Herein, we report a case of incidental finding of heterotopic bone formation in a mucinous cystadenoma of the ovary in a 59-year-old female who presented with abdominal pain. The subsequent evaluation led to the detection of the right hydrosalpinx, for which she underwent total abdominal hysterectomy with bilateral salpingo-oophorectomy. Gross examination of the specimen received showed an enlarged cystic right ovary and atrophic uterus. The microscopic examination revealed a right ovarian mucinous cystadenoma with foci of ossification. Ossification in an ovarian cyst is a benign finding of minimal prognostic significance. However, awareness of this entity is necessary to understand the sonographic findings of apprehension during radiological examination.

Key words: Mucinous cystadenoma, Ossification, Ovary

varian ossification or heterotopic bone formation is a common occurrence in mature cystic teratoma. Other conditions exhibiting ossification include endometriotic cysts, ovarian stones, benign, and malignant tumors of the ovary [1]. The pathogenesis of ossification remains unknown. It is hypothesized that the condition may occur as an unusual reaction to tissue damage and repair [1]. Mucinous cystadenoma is a benign ovarian neoplasm, comprising around 80% of all the cases of surface epithelial ovarian tumors [2]. It commonly presents as a unilateral lesion, between 3rd and 6th decades of life. Multilocular cyst with numerous thin septations containing low-level internal echogenicity due to increased mucin content is the characteristic ultrasonographic finding [3]. However, ossification in the cyst wall is exceedingly rare. As of 2020, only 33 cases of ossification in cystic ovarian lesions have been reported in the age group of 30-70 years with only five cases of mucinous cystadenoma showing ossification [1].

Here, we report an unusual case of mucinous cystadenoma with ossification in a 59-year-old female.

CASE REPORT

A 59-year-old female presented to the gynecology department of our hospital with a complaint of abdominal pain for 1 month.

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The patient attained menopause 15 years ago. She was a known diabetic since 5 years and hypertensive since 1 month, on regular medications. The patient had no significant past history.

Clinical examination and blood investigations were within normal limits. An abdominopelvic ultrasound done showed atrophic uterus and ovaries along with the right hydrosalpinx measuring 10.1×3.9 cm with no evidence of calcification. Following this, a total abdominal hysterectomy and bilateral salpingo-oophorectomy was done.

A gross examination of the specimen received in the histopathology section showed an atrophic uterus with a cervix together measuring $7 \times 4 \times 3.8$ cm. Bilateral tubes and the left ovary appeared unremarkable. The right ovary was cystically enlarged and measured $7 \times 7 \times 4$ cm. The external surface was smooth without capsular rupture. On sectioning, the cyst exuded mucinous material and showed a multiloculated cyst, with locules ranging in size from 1.5 to 5 cm with multiple firm to gritty yellow areas in the cyst wall. Average cyst wall thickness measured 0.2 cm (Fig. 1).

On microscopic examination, the right ovary showed a cystic neoplasm, lined by a single layer of columnar epithelium with apical mucin. The underlying fibrous stroma showed foci of ossification in the form of mature bony trabeculae with interspersed osteocytes (Fig. 2). The endometrium showed an

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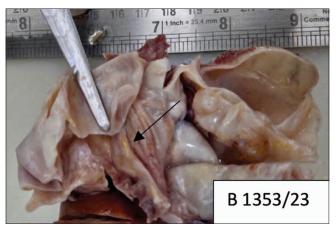


Figure 1: Gross examination of the specimen showed an atrophic uterus with a cervix together measuring $7 \times 4 \times 3.8$ cm. On sectioning, the cyst exuded mucinous material and showed a multiloculated cyst, with locules ranging in size from 1.5 to 5 cm with multiple firm to gritty yellow areas in the cyst wall

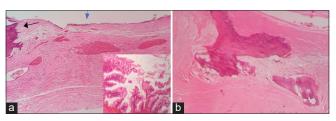


Figure 2: Microscopic examination of the right ovary showed a cystic neoplasm, lined by a single layer of columnar epithelium with apical mucin (a). Also seen are foci of bony trabeculae (b)

atrophic pattern with Monckeberg's medial sclerosis. Features of chronic cervicitis with Nabothian cyst were noted. Bilateral fallopian tubes and the left ovary showed no significant pathology.

DISCUSSION

Calcification in the ovary has been reported in neoplastic as well as non-neoplastic lesions. The most common is psammomatous calcification seen in serous cystadenocarcinomas. Bone formation in the ovary is rare, except in mature ovarian cystic teratoma [4]. Among the rare known causes, ovarian endometriosis, fibromas, mucinous, and serous cysts exhibiting ossification have been reported [4]. From the review of the literature, we found two very rare incidences at a young age, 11 and 19 years old with mucinous cystadenoma showing ossification [5,6].

Pathological calcification is classified as either metastatic (associated with hypercalcemia) or dystrophic (associated with hormonal calcemia). Conventionally, the calcification in neoplasm has been considered to be dystrophic - secondary to degeneration at areas of necrosis [7,8]. The pathogenesis of ossification in ovaries is not very well understood but is usually attributed to unusual reactions to tissue damage and repair. Another hypothesis postulated that bone-forming factors like transforming growth factor beta (TGF-β) and bone morphogenetic proteins (BMP) secreted by tumor cells stimulate metaplastic processes of multipotent stromal cells in neoplastic lesions, leading to bone formation [1] The tumor cells may produce bone-forming factors such as TGF-β and BMPs [1].

Although the reported cases of ectopic bone in the ovary have been associated with both benign and malignant conditions the finding of bone in the cyst walls likely leads to thickening of the cyst and therefore a complex appearance of the masses on sonographic evaluation as in our patient [5]. In the present case, the ovarian cyst with ossification was mistaken as hydrosalpinx by radiology. As far as we understand, this process does not have any prognostic or pathological significance; however, it is essential to know about it because it may lead to sonographic findings of concern during evaluating patients with a pelvic mass [4].

CONCLUSION

Ossification in an ovarian cyst is an infrequently reported entity; hence, the awareness of this entity is pertinent for pathologists and radiologists. Extensive gross sampling of yellow gritty or firm areas in an ovarian cyst is important to identify this condition. The bone formation associated with cystic neoplasms of the ovary possibly represents a metaplastic process occurring in the stroma of the cyst walls.

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