MÆDICA - a Journal of Clinical Medicine

CASE REPORT

MAEDICA – a Journal of Clinical Medicine
2021; 16(3): 538-541
https://doi.org/10.26574/maedica.2021.16.3.538

Bone Formation in a Mucinous Cystadenoma of the Ovary – Report of a Rare Case

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ABSTRACT

Bone formation in the ovary is exceedingly rare, except in the setting of dermoid cysts. Here, the author reports a case of incidental finding of heterotopic bone formation in a mucinous cystadenoma of the ovary in a 45-year-old woman who had underwent a total abdominal hysterectomy and right salpingo-oopherectomy because of hypermenorrhea during last one year with an ultrasonography report of right ovarian cyst and simultaneous multiple uterine leiomyomatas. Microscopic examination of the ovarian cyst revealed a mucinous cystadenoma with the striking finding of several thin plates of lamellar bone identified in fibrous tissue in the cyst wall. Although it is a benign finding and does not seem to have prognostic significance, it may lead to sonographic findings of concern during the evaluation of ovarian cysts.

Keywords: bone formation, ovary, mucinous cystadenoma.

INTRODUCTION

Ossification in an ovarian cyst is exceedingly rare, except for developing in the setting of mature cystic teratoma, which also has other additional features like hair, cartilage and muscle (1). Ovarian mucinous cystadenoma is a benign tumor that arises from the surface epithelium of the ovary and comprises 80% of mucinous ovarian tumors and 20-25% of all ovarian tumors (2). The morphologic characteristics of their growth patterns seem to have been exhaustively described. It was unexpected, therefore, to change upon bone in the wall of a mucinous cystadenoma; we herein describe this rare case. ❑

MATERIALS AND METHODS

A 45-year-old female (p4 and younger child 18 years old) presented with irregular menstruation and hypermenorrhea for the last year. In past medical history, she had hypertension during the past two years. In drug history, she was using Captopril daily for blood pressure and Advil

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Article received on the 10th of June 2021 and accepted for publication on the 23rd of September 2021

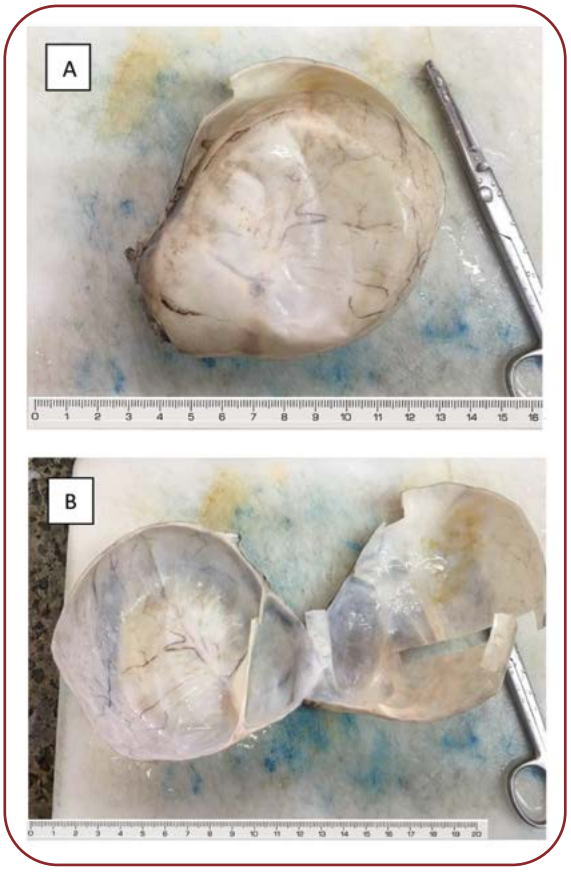

FIGURE 1 (A and B). Gross specimen shows a cream-yellowish ovarian cyst with smooth external and internal surfaces, and wall thickness measuring 0.1 to 0.2 cm.

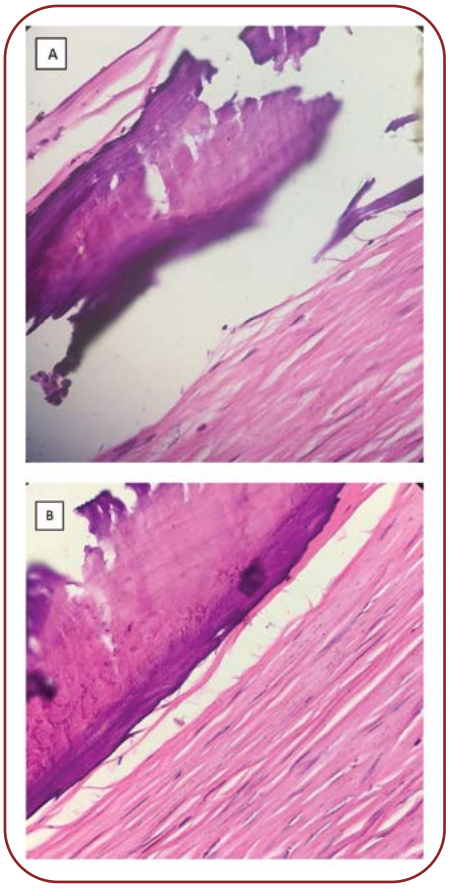

FIGURE 2 (A and B). Sections of ovarian mucinous cystadenoma revealing unexpected plates of lamellar bone formation within fibrous tissue of wall. Hematoxylin and eosin(×40)

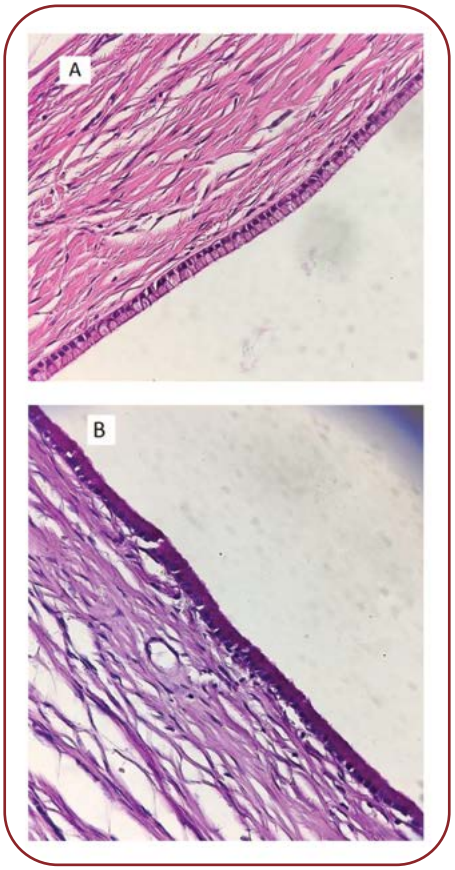

FIGURE 2. A) The wall of the mucinous cystadenoma consisting of loosely textured fibrous tissue and a layer of columnar cells with abundant cytoplasm and small, round to oval, hyperchromatic, basal nuclei constitutes the inner lining of the cystadenoma. Hematoxylin and eosin(×40). **B)** Lining of cyst showing PAS-reactive mucus containing columnar cells

on an occasional basis. Clinical examination was unremarkable. A pelvic ultrasound was done and reported a multiloculated right ovarian cyst with simultaneous multiple intramural leiomyomas in the uterus.

Following this, a total abdominal hysterectomy and right salpingo-oophorectomy were done. Gross examination of the specimen reveals an unopened uterus with no specific pathologic changes, except multiple submucosal leiomyomas measuring 1.5×1.5×1 cm to 0.6×0.6×0.5 cm with a white/tan whorled appearance without hemorrhage or necrosis. The right fallopian tube was grossly unremarkable. A cream-yellowish cystic structure measuring 12.2×9.5×5 cm was attached with a fallopian tube. On opening, it was filled with mucinous fluid, and both external and internal surfaces were smooth; the firm-elastic wall of the cyst measured 0.1 to 0.2 cm in thickness (Figure 1).

Microscopic examination showed the uterus lined by hormone-affected endometrium with multiple leiomyomas in the myometrium. Cervix examination was insignificant. The right fallopian tube had normal histology, with no specific pathologic changes. Histologically, the ovarian cyst wall consisted of loosely-textured fibrous tissue, lined by a single layer of columnar cells with small, round to oval, bland, monotonous nuclei. The cytoplasm of these cells contained abundant intracellular PAS-reactive mucus (Figure 2). While the lining of the cyst was typical for an ovarian mucinous cystadenoma, several thin plates of lamellar bone identified in fibrous tissue in the wall of the cyst were striking (Figure 3).

With the help of gross features and microscopic findings, a diagnosis of ossification in mucinous cystadenoma of the ovary was made. ❑

DISCUSSION

Calcification has been reported in a wide range of both primary neoplasms and metastatic diseases of the ovary. Psammomatous calcification is likely to account for the majority of calcified diseases in the ovaries, particularly in the case of serous cyst adenocarcinomas (3). However, bone formation in the ovary, with the exception of developing in the setting of a mature cystic teratoma or a heterologous mixed mesodermal tumor, is exceedingly rare (4).

From the review of the literature we found only 10 cases of other ovarian neoplasm associated with osseous metaplasia. Two cases of osseous metaplasia in ovarian serous cystadenocarcinoma (5, 6), one case in a benign ovarian cyst associated with cloacal anomaly (7), one case in endoimetrioma (8), one in luteinized thecoma (9), one in the endometrioid carcinoma of ovary (3), one in sertoli-leydig cell tumor of ovary (10), another one in chocolate cyst of ovary (4) and regardless of our case, there were two other cases of ossification in ovarian mucinous cystadenoma (11, 12), with one of them being reported in 2000 in a 75-year-old patient, and the other one in 2001 in a 19-year-old girl; both cases were presented with an asymptomatic pelvic mass. Although mucinous tumors comprise 15% of ovarian tumors (2), only these two documented cases of mucinous tumors with bone formation have been reported in the literature till now, more suggestive of its apparent rarity.

Benign osseous metaplasia in ovarian tumors is rare, and its histogenesis remains unclear (3). However, the cause of bone formation may be hyalinization, dystrophic calcification and subsequent osseous metaplasia.

Another theory postulated by some authors that the tumor may produce bone-forming factor-like transforming growth factor b or bone morphogenetic protein (BMP) causing metaplastic transformation of the undifferentiated mesenchymal stromal stem cell in osteoblasts (13). Bone morphogenetic protein is a family of growth factors regulating various biological processes such as bone formation and psammoma body formation in ovarian tumors (4).

As far as we know, this process does not appear to have any prognostic or pathological significances (14). Still, it is essential to know about it because it may lead to sonographic findings of concern during evaluating patients with a pelvic mass (11). ❑

CONCLUSION

Though only a few cases of osseous formation in mucinous cystadenoma of the ovary have been published in the literature and, to our knowledge, no cases from Iran have been reported until now, pathologists and radiologists need to be aware of the existence of heterotopic bone in the ovary associated with a mucinous cystadenoma. ❑

Conflicts of interest: none declared.
Financial support: none declared.

References

1. **Shaco LR, Lazer T, Piura B, et al.** Ovarian ossification associated with endometriosis. *Clin Exp Obstel Gynecol* 2007;34:113-114.
2. **Remah M Kamel.** A massive ovarian mucinous cystadenoma: a case report. *Reproductive Biology and Endocrinology* 2010;8:24-27.
3. **Mardi K, Gupta N, Sood S, Rao M.** Granulosa cell tumor-like variant of endometrioid carcinoma of ovary with osseous metaplasia: report of a rare case. *Middle East Journal of Cancer* 2015;6:107-110.
4. **Singh AK, Singh MK, Ghavghve S, et al.** Ossification of ovarian cyst: a rare case report. *Journal of Evolution of Medical and Dental Sciences* 2013;2:4523-4527.
5. **Bosscher J, Barnhill D, O'Connor D, et al.** Osseous metaplasia in ovarian papillary serous cystadenocarcinoma. *Gynecol Oncol* 1990;39:228-231.
6. **Barua R, Cox LW.** Occurrence of bone in serous cystadenocarcinoma of the ovary. *Obstet Gynaecol* 1982;22:183-186.
7. **Godbole P, Outram A, Sebire N.** Osseous metaplasia in a benign ovarian cyst in association with cloacal anomaly. *J Clin Pathol* 2005;58:334-335.
8. **Badawy SZ, Kasello DJ, Powers C, et al.** Supernumerary ovary with an endometrioma and osseous metaplasia: a case report. *Am J Obstet Gynecol* 1995;173:1623-1624.
9. **Morizane M, Ohara N, Mori T, et al.** Ossifying luteinized thecoma of the ovary. *Arch Gynecol Obstet* 2003;267:167-169.
10. **Mooney EE, Vaidya KP, Tavassoli FA.** Ossifying well-differentiated Sertoli-Leydig cell tumor of the ovary. *Ann Diagn Pathol* 2000;4:34-38.
11. **Zahn CM, Kendall BS.** Heterotopic bone in the ovary associated with a mucinous cystadenoma. *Mil Med* 2001;166:915-917.
12. **Misselevich I, Boss JH.** Metaplastic bone in a mucinous cystadenoma of the ovary. *Pathol Res Pract* 2000;196:847-848.
13. **Pervatikar SK, Rao R, Dinesh US.** Ossifying lutinized thecoma of the ovary with endometrial adenocarcinoma. *Indian j Pathol Microbiol* 2009;52:222-224.
14. **Mukonoweshuro P, Oriowolo A.** Stromal osseous metaplasia in a low- grade ovarian adenocarcinoma. *Gyneocol Onco* 2005;99:222-224.