Case Report

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Osseous metaplasia of ovarian cyst: a rare case report

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ABSTRACT

Osseous metaplasia has been described at many sites, however previous reports of osseous metaplasia in ovarian lesions are rare. This is most commonly associated with dermoid cyst, osseous metaplasia in stroma rich in serous or mucinous neoplasm and ossification of endometriotic cyst (chocolate cyst). Here The authors report a case of incidental finding of osseous metaplasia in the endometriotic cyst (chocolate cyst) of resected ovary in 42-year-old female who had gone for total abdominal hysterectomy with bilateral salpingo-oophorectomy for provisional diagnosis of AUB.

Keywords: Endometriotic cyst, Osseous metaplasia, Ovarian cyst

INTRODUCTION

Osseous metaplasia has been described in many places, but previous reports of osseous metaplasia in ovarian lesions are rare. It is most commonly associated with dermoid cysts, or ovarian teratomas with additional features such as hair, cartilage and muscle, etc.¹ Other causes of this ossification include osseous metaplasia in serous or stromal-rich mucosal neoplasms or mucus. In ultrasound, a hardened endometriotic cyst (brown cyst) may be misdiagnosed as a calcified ovarian teratoma or tumor. The hardened endometriotic cyst (brown cyst) is not an indication of the operation itself, but other lesions are treated primarily by surgery. This type of ambiguity, which occurs after ultrasound, disappears only with the histopathology of the sample resected with clinical correlation.

Osseous metaplasia has been described at many sites, however previous reports of osseous metaplasia in ovarian lesions are rare. This is most commonly associated with dermoid cyst, or teratoma of the ovary with additional features like hair, cartilage and muscle

etc.¹ Other causes of such ossification include osseous metaplasia in stroma rich serous or mucinous neoplasm. On ultrasound ossifying endometriotic cyst (chocolate cyst) is may be misdiagnosed as teratoma or calcified ovarian tumor. The ossified endometriotic cyst (chocolate cyst) is not an indication of surgery itself, but other lesion treating mainly by surgical intervention. This type of ambiguity which is came after ultrasound remove only by histopathology of resected specimen along with clinical correlation.

CASE REPORT

A 46 years aged female (nulligravida) presented with heaviness and pain in lower abdomen with dysmenorrhea and heavy menstrual bleeding(hypermenorrhea) for last 1 year. On Ultrasonography: right ovary showed multiple hyperechoic masses, left ovary showed large cyst with hyperechoic with homogenous texture. Following this, provisional diagnosis of endometrioma and benign ovarian neoplasm was made based on clinical history and ultrasonography and total abdominal hysterectomy with bilateral salpingo-oophorectomy was done.

Gross examination reveals resected uterus and cervix measuring 9.5x6.5x4.5 cm in size. Cut surface of uterus and cervix was unremarkable and shows grey white, globular, submucosal mass measuring size 1.5x1x1 cm, endomyometrial thickness was 2.5 cm. Right ovary was measuring 3.5x1.7x1.7 cm with cut surface was showing a fragmented bony cyst, filled with hemorrhagic material, of 2 cm in diameter. Wall thickness of cyst wall ranging from 0.2 to 0.3 cm. Right fallopian tube was measuring 4.5 cm in length and was grossly unremarkable. Left ovary was measuring 4.5x3.5x1.5 cm in size and cut surface showed hemorrhagic cyst measuring 2.5 cm in size Left fallopian tube was measuring 11.5 cm and was grossly dilated. Microscopic examination showed submucosal leiomyoma uterus, uterine cavity lined by proliferative endometrium with histologically unremarkable underlying myometrium. Cervix examination revealed chronic cervicitis in the form of diffuse inflammatory infiltrate in to the cervical stroma. Multiple sections examined from right ovary showed a cystic structure endometriotic cyst (chocolate cyst), totally replaced by areas of ossification and calcification. Focal areas resembling normal ovary was identified peripherally. Sections examined from left ovary shows endometriosis and endometriotic cyst (chocolate cyst), while of both fallopian tubes showed normal histology.

Following above mentioned microscopic findings, different etiologies for such ossification in ovary were find out, which includes dermoid cyst, osseous metaplasia in stroma rich neoplasm of ovary and chocolate cyst of ovary. With the help of gross features and excluding other etiologies, a diagnosis of ossification in endometriotic cyst (chocolate cyst) was made.

DISCUSSION

The pathogenesis of osseous metaplasia is uncertain, but in some cases, it appears to be an unusual reaction to tissue damage and repair. It is interesting that the previously reported phenomenon associated with an endometriotic ovarian cyst occurs in an anatomically abnormal pelvis in a supernumerary ovary.² Osseous metaplasia in a benign cyst of the ovaries is an unusual meaning and an unusual manifestation of etiology, which is more common in anatomically abnormal ovarian tissue in women during reproductive age.

Osteoporosis is rare, with exception, in the development of a mature cystic teratoma or heterologous mixed mesodermal tumor. The most reliable description of bone formation in the ovarian neoplasm is a metaplastic process of the multipotent stromal cell. This extraordinary phenomenon has not been attributed prognostic significance. Pathological calcification is classified as either metastatic (associated with hypercalcemia) or dystrophic (associated with hormonal calcemia). Traditionally the calcification in neoplasm has been considered to be dystrophic-forming to secondary to degeneration of either in areas of necrosis.

The most widely accepted theory of the pathogenesis of calcification postulate that calcification is due to calcium deposition in areas of cellular degeneration associated with an infection process, such as malakoplakia or ischemic changes. Calcification has been described in various neoplasm associated with hormone production including duodenal somatostatinoma, carcinoid tumor, prolactinoma, calcifying sartoli cell tumor gonadoblastoma.⁵ In benign, the combined endometrium of combined encephalitis (combined contraceptive pills) and clomiphene citrate for contraception are the possible causes of calcification. In non-neoplastic lesion of ovarylike endometrioma, previous torsion with subsequent infarction of ovary can explain bone formation.^{6,7} In benign and malignant tumors, it is possible that multiple pathways are involved in the pathogenesis of calcification because calcification is seen in areas of necrosis and tumors with degenerative changes.

In a study of ossification, the endothelial adenocarcinoma of the ovary is reported to be the result of massive ossicular ossification in the bone. True bone formation in ovary tumors is rare. Osseous metaplasia could be cause in this setting.8 Osseous metaplasia has been documented in well differentiated Sertoli, Leydig cell tumors, mucinous cystadenoma, fibromas and serous papillary cystadenocarcinoma.9 The cause of bone formation may be hyalinization, dystrophic calcification and subsequent osseous metaplasia. Another theory postulated by some author that the production of bone morphogenetic protein (BMP) causes a metastogenic transformation of the mesenchymal stromal stem cell in osteoblasts.⁸ BMP are a family of growth factors regulating a wide variety of biological processes and formation of bodybuilding in ovarian tumors.

The pathogenesis of osseous metaplasia is unclear, but in some circumstances appears to be an unusual reaction to tissue damage and repair. It is interesting that the previously reported case associated with an endometriotic ovarian cyst occurred in an anatomically abnormal pelvis, in a supernumerary ovary.2 Osseous metaplasia in a benign ovarian cyst appears to be an unusual incidental finding of unknown importance and aetiology, which is more common in anatomically abnormal ovarian tissue in women of reproductive age. Bone formation in the ovary, with the exception of developing in the setting of a mature cystic teratoma or a heterologous mixed mesodermal tumour, is exceedingly uncommon.1 The most plausible explanation for bone formation in an ovarian neoplasm is a metaplastic process of the multipotential stromal cell.³ No prognostic significance has been attributed to this unusual phenomenon.

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The most widely accepted theory regarding the pathogenesis of calcification postulate that calcification is due to calcium deposition in areas of cellular degeneration associated with either an infection process, such as malakoplakia or with ischemic changes. Calcification have been described in various neoplasm associated with hormone production including duodenal somatostatinoma, carcinoid tumour, prolactinoma, calcifying sartoli cell tumour and gonadoblastoma.⁵ In benign, the endometrium of patient receiving enovid (combined contraceptive pills) and clomiphene citrate for contraception are the possible cause of calcification. In non-neoplastic lesion of ovary like endometrioma, previous torsion with subsequent infarction of ovary can explain the bone formation.^{6,7} In benign and malignant tumour, it is likely that multiple mechanisms are involved in the pathogenesis of calcification because calcification are seen in areas of necrosis and in tumours with degenerative changes.

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Another theory postulated by some author that the tumour 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. BMP are a family of growth factors regulating a wide variety of biological processes like bone formation and psammoma body formation in ovarian tumours.

CONCLUSION

Osseous metaplasia (bone formation) in ovarian tumours or benign ovarian cyst is probably due to interaction of multiple factor that they are not fully understood. In present case the cause of osseous metaplasia probably due to old endometriosis with subsequent infarction.

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REFERENCES

- 1. Shaco LR, Lazer T, Piura B, Wiznitzer A. Ovarian ossification associated with endometriosis. Clin Exp Obstel Gynecol. 2007;34(2):113-4.
- 2. Badawy SZ, Kasello DJ, Powers C, Elia G, Wojtowycz AR. Supernumerary ovary with an endometrioma and osseous metaplasia: a case report. Am J Obstet Gynecol. 1995;173:1623-4.
- 3. Zahn CM, Kendall BS. Heterotopic bone in the ovary associated with a mucinous cystadenoma. Mil Med. 2001;166:915-7.
- 4. Clement PB, Cooney TP. Idiopathic multifocal calcification of the ovarian stroma. Arch Pathol Lab Med. 1992;116(2):204-5.
- 5. Silva EG, Deavers MT, Parlow AF, Gershenson DM, Malpica A. Calfications in ovary and endometrium and their neoplasms. Mod Pathol. 2003;16(3):219-22.
- 6. Su WH, Wang PH, Chang SP. Ovarian stone: a case report. J Reprod Med. 2002;47(4):329-31.
- 7. Kennedy LA, Pinckney LE, Curranino G, Votteler TP. Amputated calcified ovaries in children. Radiol. 1981;141(1):83-6.
- 8. Pervatikar SK, Rao R, Dinesh US. Ossifying lutinized thecoma of the ovary with endometrial adenocarcinoma. Indian J Pathol Microbiol. 2009;52(2):222-4.
- 9. Zahn CM, Kendall BS. Heterotopic bone in the ovary associated with a mucinous cystadenoma. Mil Med. 2001;166(10):915-7.

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