

CASE REPORT

A Rare Case of Cartilaginous Heterotopia in Broad Ligament of Uterus

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ABSTRACT

Heterotopia is the occurrence of mature tissue at abnormal location. A very rare case of cartilaginous heterotopias in the broad ligament of a 47-year-old female is described. Literature contains very few references related to it. In this patient, there was no evidence of any malignancy in the abdomen or in any other part of the body, except cervical intraepithelial neoplasia changes in cervix. The peritoneal lesion was an incidental finding in this female who underwent total abdominal hysterectomy in view of low-grade squamous intraepithelial lesion of cervix on cervical biopsy. A firm to hard, white-colored, tubular, branched structure, embedded in left broad ligament reaching till serosa of left fallopian tube and undersurface of left ovary was present. Histopathology showed mature cartilage of hyaline type with well-formed chondrocytes and lacunae with surrounding fibrosis with no evidence of cytological atypia, reactive inflammatory changes, or foreign body reaction. This may represent metaplastic lesions of secondary Müllerian system or benign neoplastic lesions (chondroma) of submesothelium.

Keywords: Broad ligament, Cartilaginous heterotopia, Chondroma, Metaplasia.

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INTRODUCTION

Cartilaginous differentiation of the human peritoneal tissues is rare, although a single case is cited in surgical pathology text as a probable metaplastic lesion of submesothelium.¹ Heterotopia is the occurrence of mature tissue at an abnormal location. Cartilaginous heterotopias have been reported in the uterus and cervix.^{2,3} These lesions may represent metaplasia or

organized products of conception showing cartilaginous differentiation. Fadare et al⁴ have reported two cases of cartilaginous differentiation in peritoneal tissue, which may represent metaplastic lesions of the secondary Müllerian system or a unique peritoneal response to previous surgical manipulation. In the fallopian tube, Spanta and Lawrence⁵ have suggested subcelomic mesenchyme of the tubal serosa as the origin of chondroid metaplasia.

The peritoneal lesion was discovered accidentally while clamping the broad ligament and fallopian tube during total abdominal hysterectomy of the patient.

Histopathology showed mature cartilage of hyaline type with well-formed chondrocytes and lacunae with surrounding fibrosis, with no evidence of cytological atypia, reactive inflammatory changes, or foreign body reaction. The fallopian tube and ovary were unremarkable grossly as well as microscopically, with no evidence of any ectopic tissue or cytological atypia or dysplasia. This may represent metaplastic lesions of secondary Müllerian system or benign neoplastic lesions (chondroma) of submesothelium.

CASE REPORT

A 47-year-old female P₄L₄A₂ was admitted in the Gynae ward, Rajindra Hospital, Patiala, India, with complaint of multiple episodes of postcoital bleeding for past 2 months.

She also complained of irregular menses since past 2 months with cycle length of 10 to 15 days, duration of menses 2 to 3 days, average flow, and no dysmenorrhea. It was preceded by 3 months amenorrhea; there was no history of pain abdomen. History of discharge per vaginam was yellowish, thick consistency, nonfoul smelling fluid, not associated with itching. No urinary or bowel disturbance, no history of any lump in abdomen, no history of weight loss, or loss of appetite were found. Patient did not have any medical disorder or surgical illness and was not taking any chronic medication.

Patient has previous four vaginal deliveries with last childbirth 15 years back. Patient had two induced surgical abortions at 2 and 3 months amenorrhea 16 years back in a private nursing home.

Per abdomen examination was unremarkable.

On per speculum examination, anterior and posterior lips of cervix were hypertrophied with acetowhite areas at 5 o'clock and 11 o'clock position.

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On per vaginum examination, cervix was backward, firm, irregular in contour, uterus was anteverted, normal size, nontender, mobile, bilateral fornices were free.

Pap smear showed atypical squamous cells – high-grade squamous intraepithelial lesion cannot be ruled out.

Ultrasound showed no significant findings.

Patient underwent cervical biopsy under short general anesthesia, histopathology report of which showed low-grade squamous intraepithelial lesion.

Patient underwent total abdominal hysterectomy. There was a small healed scar at fundus of uterus on right side, suggestive of old perforation. Right fallopian tube and ovary were normal. There was a hard tubular and branched white-colored structure, 3 cm in maximum dimension, embedded in the left broad ligament reaching till left ovary's undersurface. Left ovary, left ovarian ligament, and broad ligament containing the tubular structure were resected.

Two provisional diagnosis were made:

- Displaced and retained limb of Copper T.
- Fetal tissue (bone or cartilage) as a result of previous dilatation and curettage, followed by perforation of uterus.

Histopathology report showed left tube and ovary unremarkable and normal and the hard structure as mature skeletal tissue surrounded by fibrous stroma.

On microscopy, mature chondrocytes could be seen with well-defined lacunae and also the finding of isolated mature skeletal tissue in broad ligament in the absence of any cytological atypia, reactive inflammatory changes, or foreign body reaction. It was suggestive of cartilaginous cell rest or heterotopias in paratubal region.

DISCUSSION

The histogenetic explanation for cartilaginous differentiation in peritoneal tissues has been the source of some controversy. This differentiation has been explained by the theory that there exists a population of submesothelial multipotent cells with ability to differentiate along mesenchymal and mesothelial lines.⁶

In the human peritoneum, at least seven well-documented cases of mesenteric heterotopic ossification (or osseous metaplasia) have been reported.⁷ These lesions may appear on imaging as soft tissue calcification, but in our case it was not visible on ultrasound.

In the uterus and cervix, cartilaginous heterotopia is a rare but well-recognized phenomenon. Remmele et al⁸ reported two cases and extensively reviewed the literature on the subject. Nodules of cartilage are typically identified in the endometrium, myometrium, and endocervix, and in two cases,² a cartilaginous nodule was present in the anterior uterine subserosa. In the first patient, a bar

of cartilage was noted beneath the uterine serosa during a salpingectomy for hydrosalpinx. A similarly located cartilaginous nodule in the anterior uterine subserosa of a second patient was reported. The possibility of a benign cartilaginous neoplasm (chondroma) also warrants some consideration in this case. One-third cases of extraskeletal chondromas can show immature chondroblasts with tendency of recurrence. The treatment, however, is wide local excision. In our case, only mature chondrocytes were seen.

Another possibility is that cartilaginous tissue might be implanted into the peritoneum from capsular defects in ovarian teratomas, which might explain the cartilaginous lesion in our case. However, in our case, no teratomatous lesion was identified in the ovary.

Given the obstetrical history of self-induced abortion in our case (approximately 15 years before presentation), these lesions may also represent organized products of conception showing cartilaginous differentiation. We share these cases for the benefit of surgeons who might encounter these lesions and for pathologists who would be making their intraoperative evaluation.

The differential diagnosis for the presence of cartilage in this case includes extra skeletal chondroma, mature cystic teratoma as well as fetal tissue remnants. The first two conditions would present as a tumor nodule grossly as well as microscopically with no normal fallopian tubal structure intermingled with the islands of chondrocytes in a chondroma. Teratoma, in addition, would show evidence of other germ cell elements.⁵

CONCLUSION

Cartilaginous heterotopias are cell rests of mature cartilaginous tissue which can be found in many parts of the body. Uterus and peritoneum are few rare locations. It is useful for the surgeons to know that these are usually incidental findings, detected on imaging in the form of dystrophic calcification or intraoperatively. This may represent metaplastic lesions of secondary Müllerian system or benign neoplastic lesions (chondroma) of submesothelium. In case of lesions of peritoneum in paratubal region, possibility of teratoma of ovary should be ruled out. Resection of the lesion is the definitive management. Moreover, 2 to 3% of chondromas may show immature chondrocytes with atypia with tendency for recurrence.

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