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CASE REPORT

HETEROTOPIC CARTILAGE IN THE MYOMETRIUM

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ABSTRACT

We describe a rare case of chondroid heterotopia in the myometrium with associated foci of adenomyosis, in a 45 years old female, presenting with abnormal uterine bleeding and abdominal pain. Designated as metaplastic and benign, it needs to be differentiated from other benign conditions as impacted fetus, teratoma, chondroma, and certain malignant conditions like malignant mixed mullerian tumor, adenosarcoma and leiomyosarcoma.

KEYWORDS: Mullerian mixed tumor; Heterotopic cartilage; Myometrium; Uterus.

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INTRODUCTION

Heterotopia means normal tissue at an abnormal site. Cartilaginous heterotopia is presence of cartilage rests and has been described throughout the female genital tract. Some sites include endometrium, endocervix, subserosa and serosa. Few cases describing cartilaginous heterotopia have been described in paratubal and tubal locations. Heterotopic cartilage within the myometrium is an extremely rare phenomenon. Neumann in 1925, first described the presence of cartilage in the endometrium and myometrium.[1]

Heterotopic cartilage in the myometrium is usually asymptomatic and an incidental finding, but may present with abnormal bleeding, infertility, subfertility and abortion. It is detected on radiology as foci of calcification. It represents metaplasia of Mullerian system or uterine endometrial or myometrial stroma in differentiation towards cartilage. There are several postulated mechanisms described for the metaplastic transformation of multipotent cells of normal endometrial stroma. Inflammation and endometriosis are thought to play a role in initiating the process of metaplasia. Dystrophic calcification and cartilage formation may be secondary to chronic inflammation as in pyometra. Some reports considered hypercalcemia and hyperestrinism as the causative factors.[2]

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Differential diagnosis of heterotopic cartilage includes benign conditions like impacted or organized fetal parts in the uterus, chondroma, teratoma, and malignant conditions like malignant mixed mullerian tumor (MMMT).[3]

The aim of this case report is to enhance knowledge of this benign condition amongst the practicing pathologist and surgeons and to emphasize the importance of pre surgical radiological investigations and a conservative approach to treatment whenever possible.

CASE REPORT

A 45 years old female, para 4, presented with the complaint of abdominal pain and menorrhagia for four months with no history of abortion, which was unresponsive to medical treatment. With the clinical impression of fibroid uterus, total abdominal hysterectomy and bilateral salpingo-oophorectomy was done. Pre-surgical pelvic ultrasonographic examination was not available.

On gross examination, the uterus was large and measured 15x8x5 cm in superior-inferior, lateral and antero-posterior dimensions respectively. Endometrium was intact and 0.2 cm in thickness. Myometrium was thick 3-4 cm, trabeculated and showed multiple well-circumscribed translucent nodules near the serosal surface of posterior wall with largest focus of 0.7x0.5 cm. Microscopic examination demonstrated the presence of variable sized lobules of benign hyaline cartilage. These lobules of cartilage were deeply penetrated with foci of adenomyosis within the posterior myometrium. Endometrium was weak secretory type and suggestive of progesterone administration for control of abnormal bleeding. (Fig-1) Cervix, bilateral ovaries and fallopian tubes showed normal morphology.

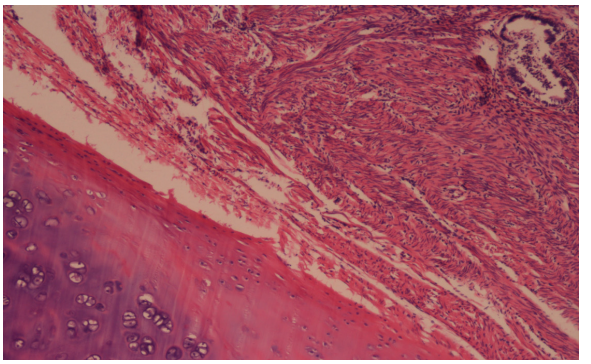

Figure 1: High power view of cartilage in myometrium with foci of adenomyosis.

DISCUSSION

Heterotopia is occurrence of normal tissue at an abnormal location. Heterotopic cartilage in the myometrium is an extremely rare event. The histiogenesis of cartilage within the female genital tract has remained a matter of academic interest. In 1925 Neumann for the first time described the presence of hyaline cartilage in the fundus of uterus between endometrium and myometrium; the reason of hysterectomy was pelvic inflammatory disease.[1] After that Taylor and Roth published a detailed case series and considered possible causes of heterotopic cartilage within the myometrium.[2] They witnessed that each of their cases of heterotopia was associated with either endometriosis or endometritis, suggesting metaplasia as the prime cause of heterotopia.[2] Metaplasia is replacement of one type of tissue with another and is a normal response to chronic inflammation, tissue damage, repair and regeneration.[1] Endometrium is the commonest site of cartilaginous heterotopia. Multipotential endometrial stromal cells have the capacity to transform into various types of cells and these cells contain high amount of acid mucopolysacharide in the adjacent stroma, as also occurs in both normal and experimental cartilage.[3] Bhatia and Hoshiko (1982) suggested that calcification and cartilage are due to prolonged retention of fetal parts, chronic inflammation and tissue destruction following repeated spontaneous or therapeutic abortions and that in some reported cases infertility was secondary to calcification of the endometrium.[4-6] In our case however, no history of previous abortion was present. Third possibility suggested in literature is that of heteroplasia, which is displacement of mesodermal cells during embryonic life already destined to differentiate into cartilage.[7]

MMMT also termed as carcinosarcoma is a highly malignant neoplasm, exhibiting malignant epithelial and stromal components. It may exhibit the terologous elements which most commonly are cartilage and skeletal muscle. The presence of cartilage in MMMT is considered a favorable prognostic factor.[8] Uterine sarcoma accounts for 3–5% of all corpus uteri malignancies; undifferentiated uterine sarcomas arise from the endometrium or myometrium, lacking any resemblance to normal endometrium and may show heterologous stromal elements in the form of cartilage, bone or rhabdomyoblasts.[9] In our case the hyaline cartilage nodules were mostly located deep in the myometrium, close to the serosal surface and adjacent tissue exhibited foci of adenomyosis; the closest differential diagnosis ruminated was adenosarcoma. Adenosarcomas have a biphasic morphology comprising of benign epithelial elements and malignant stromal elements.[10] Stromal malignant sarcomatous component may be either homologous (composed of tissues normally found in the uterus) or heterologous (containing tissues not normally found in the uterus, most commonly malignant cartilage or skeletal muscle).[9] The poor prognostic sign is marked pleomorphism and atypia in sarcomatous element with deep myometrial invasion.[3] We excluded the possibility of adenosarcoma by performing extensive sampling of the uterus and submitting the chondroid areas completely for microscopic examination.

In our case hysterectomy was performed for this benign condition, current treatment strategy is hysteroscopic removal of these foci.[9] However, these foci were deeply located and may not have been amenable to hysteroscopic removal. Pakistan is a developing country where patients have diminished access to health services, and has a considerably high rate of hysterectomy especially in patients with benign diseases, presenting with abnormal uterine bleeding.[11]

CONCLUSION

Cartilage in the myometrium is an interesting challenge, as the differential diagnoses are wide. Based on available research it appears that the pathogenesis of cartilage in myometrium in our case is possibly due to metaplasia. We recommend complete radiological workup and a conservative approach in the treatment of this benign condition.

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CONFLICT OF INTEREST
Authors declare no conflict of interest.
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All the authors agree to be accountable for all aspects of the work in ensuring that questions related to