Case Report:
Xanthogranulomatous Degeneration in a Leiomyoma – First Documentation

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Citation

Submitted: Jun 21, 2021; Accepted: Aug 4, 2021; Published: Aug 25, 2021

Abstract: Xanthogranulomatous inflammation of the female genital tract is rare with documented cases confined to the endometrium, fallopian tube, and ovary. A careful search of English literature yielded only two reports pf myometrial involvement by xanthogranulomatous inflammation. Herein, we present a case of 37-year-old diabetic female with a previously diagnosed anterior wall fibroid who presented with lower abdominal pain. A repeat transvaginal sonography confirmed the presence of a fundal subserosal fibroid with degenerative changes. The patient underwent myomectomy and there were no adhesion or involvement of adnexa in intraoperative examination. Pathological examination of the same revealed leiomyoma with degenerative changes and xanthogranulomatous inflammation. The present case of xanthogranulomatous degeneration occurring de-novo in a leiomyoma, is to the best of our knowledge the first documentation of this entity.

Key Words: Xanthogranulomatous inflammation, Leiomyoma, Foam cells, Granuloma, Necrosis

Introduction:
Xanthogranulomatous inflammation is an uncommon form of chronic inflammation that causes destruction of the affected organ tissue. It is characterized by the presence of numerous lipid-laden macrophages in a background of lymphocytes, plasma cells along with occasional presence of multinucleated giant cells, parenchymal destruction, and fibrosis.[1,2] Xanthogranulomatous inflammation is most widely reported in kidneys and gall bladder[3] and is a rare find in the female genital tract. Within the female genital tract it is documented affecting the endometrium[4,5], fallopian tubes[6,7] and ovaries.[8] Leiomyoma is the most common tumor affecting the uterine corpus and is known for its varied histopathology. As these tumors grow in the size, the blood supply gets limited and they undergo degeneration. Diverse types of degeneration reported in a leiomyoma include hyaline, myxoid, cystic, and red degeneration[9]. Involvement of myometrium by xanthogranulomatous inflammation is limited to two case reports in the English literature[2,10] and has not been reported occurring de novo in a leiomyoma. The present report is first of a kind case of xanthogranulomatous inflammation causing degeneration in leiomyoma in a 37-year-old female.

Case Report:
A 37-year-old diabetic female presented with complaints of lower abdominal pain for last 4 days. She gave the history of similar episodes of pain for the last 3 years. She had no complaints of abdominal distension, abnormal uterine bleeding, loss of weight and loss of appetite. Laboratory investigation showed a poor glycemic control. The patient had obstetric history of one past pregnancy with gestational diabetes mellitus managed on diet with a normal vaginal delivery and no post-natal complications 6 years back. During the last pregnancy, the patient was diagnosed with an anterior wall subserosal fibroid, measuring 5.5cm in greatest dimension. The present gynecological examination revealed an enlarged uterus, right and anterior fornical fullness, minimal erosions over cervix with no cervical motion tenderness. On transvaginal sonography, a subserosal fibroid with degeneration measuring 8.8cm was seen over the anterior wall of uterus with no other significant findings. After relevant pre-operative workup, patient was taken up for myomectomy. Intraoperatively, the uterus was found to be enlarged with a subserosal fibroid with areas of degeneration. Bilateral adnexa appeared normal. The fibroid was resected and submitted for pathological examination.

The excised myometomy specimen weighed 150 grams and measured 8.5x6x5cm. The cut section showed circumscribed tumour with grey, white whorled areas along with central region of tan, orange colored degeneration (Figure 1). Histopathological examination of representative areas showed peripheral region with intersecting bundles of smooth muscle fibers consistent with a leiomyoma. The central region showed...
extensive coagulative necrosis with cholesterol clefts and foci of dystrophic calcification. The periphery of degenerated region showed foci of hyalinization along with aggregates of histiocytes, numerous foam cells, lymphocytes, and plasma cells (Figure 2). There was no mitotic activity and cellular atypia. All these features were coherent with the diagnosis of a leiomyoma with xanthogranulomatous degeneration.

Figure 1: a) Transvaginal ultrasonogram showing a subserosal hypoechoic fibroid b) Cut section of the fibroid with extensive tan orange necrosis in the center

Figure 2: Peripheral intersecting bundles of benign smooth muscles (a) with central necrosis and sheets of xanthoma cells and histiocytes (b) The central region shows extensive coagulative necrosis with dystrophic calcification (c) and cholesterol clefts (d) (H&E, x100)

Discussion:
Xanthogranulomatous inflammation of the female genital tract is rare with most documented cases in fallopian tubes and ovary, followed by endometrium and rarely in cervix and vagina[7]. In 1976, Kunakemakorn et al reported the first case of xanthogranulomatous inflammation involving the fallopian tube and ovary presenting as pelvic inflammatory disease and termed it as the inflammatory pseudotumor[11]. Liao et al first described involvement of myometrium by xanthogranulomatous inflammation in 2013 in a 66-year-old female who presented with lower abdominal pain. They localized the pathology to the myometrium, which caused perforation of uterine corpus, and lead to tubo-ovarian abscess, pelvic adhesions and pyometra, however, could not elucidate the etiology in their case[10]. Inoue et al reported the second case of xanthogranulomatous inflammation arising as an abscess in the posterior perimetrium and extending into the surrounding myometrium in a 68-year-old female. The patient presented with vaginal bleeding and developed endometritis following a curettage while being investigated. They postulated the etiology of xanthogranulomatous inflammation to the instrumentation of the genital tract[2]. Du et al reported a case of xanthogranulomatous endometritis causing perforation and attributed the etiology to uncontrolled diabetes[5]. The proposed theories for etiopathogenesis of xanthogranulomatous inflammation in female genital tract include pelvic inflammatory disease, occlusion of cervical canal, and abnormal lipid metabolism. Localized abscess, necrosis and hemorrhage can also show replacement of damaged tissue by lipid containing foam cells eliciting xanthogranulomatous reaction[8].

The present case of a 37-year-old female has a considerable lower affected age group from the earlier reports. The patient had no complaints of heavy menstrual bleeding, no history of instrumentation, intrauterine contraceptive device, and history of pelvic inflammatory disease. There was no involvement of the myometrium outside the leiomyoma. The endometrium and bilateral adnexa were also not involved. Uncontrolled diabetes can be a contributing factor in the present case. Malacoplakia is an important differential diagnosis for xanthogranulomatous inflammation in patients with diabetes. It shows special histiocytes known as von Hansenn cells admixed with intracellular and extracellular basophilic laminated inclusions known as Michaelis–Gutmann bodies. The absence of these characteristic findings in the presented case, excludes this diagnosis[12].

Conclusion:
Xanthogranulomatous inflammation involving the myometrium is rare and had not been reported in a leiomyoma. To the best of our knowledge, this is the first reported case of xanthogranulomatous inflammation occurring in a leiomyoma with sparing of endometrial and adnexa. The present report adds information to the sparse documentation of xanthogranulomatous inflammation affecting the female genital tract.

References:
