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A rare case of Pyomyoma with spontaneous rupture in a nulliparous woman

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Abstract
Pyomyoma is a very rare complication of fibroids. Rupture of Pyomyoma is much rarer condition with very few reported cases till date. Most reported cases are in multiparous women. Diagnosis is quite difficult due to non specific presentation with sometimes features mimicking malignancy. In this report we present a nulliparous woman who presented with fever, pain abdomen, lump abdomen, anemia and weight loss. CE-CT abdomen showed a Pyomyoma with uterine rupture and retroperitoneal fistula. Hysterectomy was done with coverage of broad spectrum antibiotics. Confirmation of diagnosis was done by histopathology.

Keywords: Pyomyoma, nulliparous, uterine rupture, hysterectomy

1. Introduction
Leiomyomas or fibroids are common benign smooth muscle tumors of the uterus occurring in 20-30% premenopausal women [1]. Pyomyoma or suppurative leiomyoma is a rare, life threatening complication in which there is infection of the leiomyoma, which generally shows suppurative inflammation, containing pus and necrotic exudate [2]. Since its first description in 1871, around 100 cases have been documented in the literature [3], with much less numbers in the last few years due to antibiotics use. Most cases are documented in relation to pregnancy, uterine instrumentation or in post menopausal women with co-morbidities. Pathogenesis is probably vascular compromise of the fibroid with infarction, due to some pathology, followed by infection [4]. Most infections are polymicrobial. The possible routes of infection of the leiomyomas may be contiguous spread from the endometrial cavity which may be infected from ascending infections of vagina or cervix, direct extension from the adjacent bowel or adnexa, or haematogenous or lymphatic spread from infection elsewhere in the body [5, 6, 7]. These infections are most common in submucous leiomyomas as these lie in uterine cavity and have poor blood supply [5, 6]. The most dreaded complication of pyomyoma is uterine rupture, which can lead to pyoperitonitis, sepsis and death [8]. Mortality rate is around 6-30% [7, 9, 10, 11]
Diagnosis is very difficult due to the rarity of this condition, an indolent course and non specific symptoms. Accurate diagnosis is mostly possible at time of surgery [9], though some studies have shown nearly accurate diagnosis with contrast enhanced (CE) - CT scan [12].

2. Case Report
A 32 year old nulliparous women came to the Emergency department with complaints of pain in lower abdomen for 5months, on and off high grade fever with chill and rigor for 1month and feeling of a lump in lower abdomen for 1month. She had anorexia, generalized weakness and gave history of weight loss. She was referred from a secondary health care facility where she was diagnosed with severe anemia and was given 7 units of PRBC transfusion along with injectable antibiotics for fever. Though she had history of sexual exposure, there was no history of abortions or pelvic infections. She had a history of frequent menses with heavy bleeding for last 1 year; however she was amenorrhoiec for 2 months at time of admission. CE-CT abdomen done in the previous centre gave an impression of pyometra with uterine myoma. On examination, she was febrile with pulse rate of 110/min and BP of 100/70mm Hg. On per abdominal examination, a midline, irregular, solid-cystic lump corresponding to 24weeks uterine size was palpable. There was tenderness in in fraumbilical region, extending upto right hypochondrium. On admission her total leucocyte count was normal, with hemoglobin of

6.9gm/dl, CRP of 76.4mg/L, serum albumin of 1.8gm/dl, serum magnesium of 1.3mmol/L and serum potassium of 2.3mmol/L with normal LFT & RFT. All tumor markers were normal. Blood and urine cultures were negative. Medicine consultation was taken to correct deranged electrolytes and albumin and also to investigate the causes of fever. Inj. Piperacillin-tazobactum and Metronidazole infusion were started. CE-CT abdomen showed an endometrial collection of 124×87mm. A 25mm rent was seen in right anterolateral aspect of fundus with extension of collection along right paracolic gutter upto level of hepatic flexure. Inferiorly the collection extended upto right inguinal region and anteriorly upto parietal wall. Significant post contrast enhancement with multiple pockets of air foci were seen [Figure1]. Patient was planned for laparotomy. Preoperatively electrolytes were corrected and PRBC transfused.

Abdomen was opened with a paramedian longitudinal incision. Uterus was around 20weeks size and had adhesions all around. Dense adhesions were seen between uterus and right lateral pelvic wall. On adhesiolysis, a fistula was seen between uterine cavity and retroperitoneal space. This was the site of uterine perforation [Figure2]. Thick pus was seen coming out of the tract in right lateral wall. The track was explored, pus drained and lavage given. Around 500ml of pus was removed from the uterine cavity which was thick, organized, fibrous tissue-like and extremely foul smelling [Figure3]. The uterine wall was extremely friable and completely infected. Thus total hysterectomy was done. Cut section of uterus showed thick irregular areas with blackish areas [Figure4]. Help from general surgeon was also taken to explore the retroperitoneal tract and to look for bowel injuries. Thorough peritoneal lavage was given. Patient was managed in ICU post-operatively. She developed shock which was managed with IV fluids, ionotropes and blood transfusion. Inj. Meropenem was given along with Linezolid. Patient recovered significantly in the post-operative period. Gross examination of the pus material showed necrosis and whorled appearance suggesting that it was the necrosed leiomyoma. Its histopathology showed extensive bland necrosis with a foci of viable spindle cells suggesting smooth muscle, but with no mitosis [Figure5]. Biopsy from the area of uterine rupture showed extensive areas of hemorrhage, necrosis with acute and chronic inflammatory cells in the myometrium. The area of rupture had features of leiomyoma, suggesting that it was the area of the tumor [Figure6]. Pus culture from the endometrial cavity showed no growth.

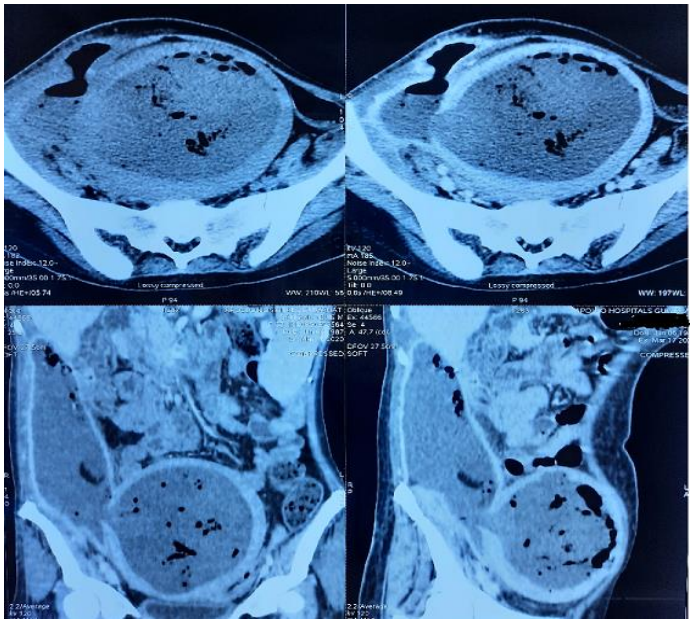

Fig 1: Different views of CT scan showing large endometrial collection with air foci with rent in lateral fundal wall with extension of the collection to right lateral pelvic wall upto hepatic flexure

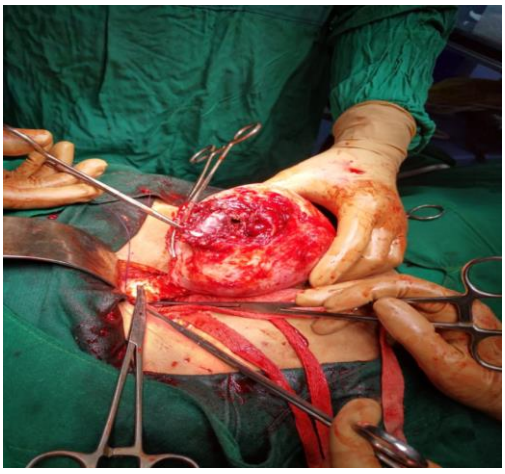

Fig 2: Site of uterine rupture which formed a fistula with lateral retroperitoneal pelvic wall

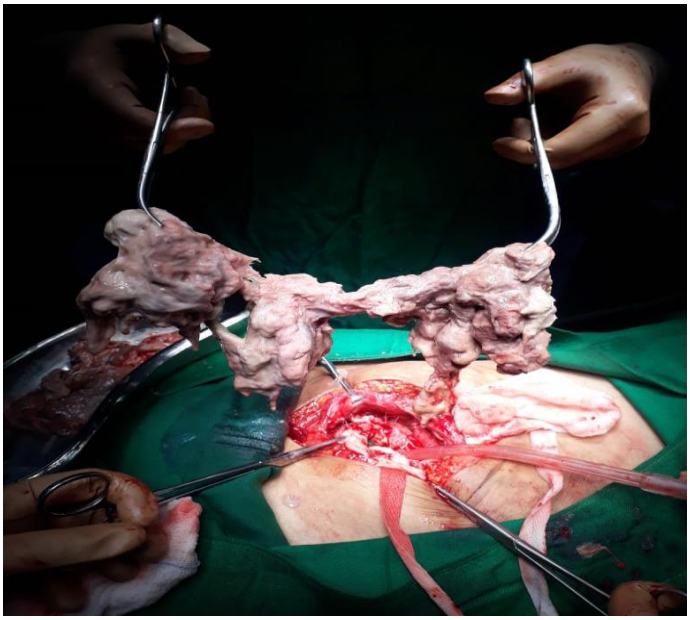

Fig 3: The organized and necrosed foul smelling pus taken out through the ruptured area of uterus

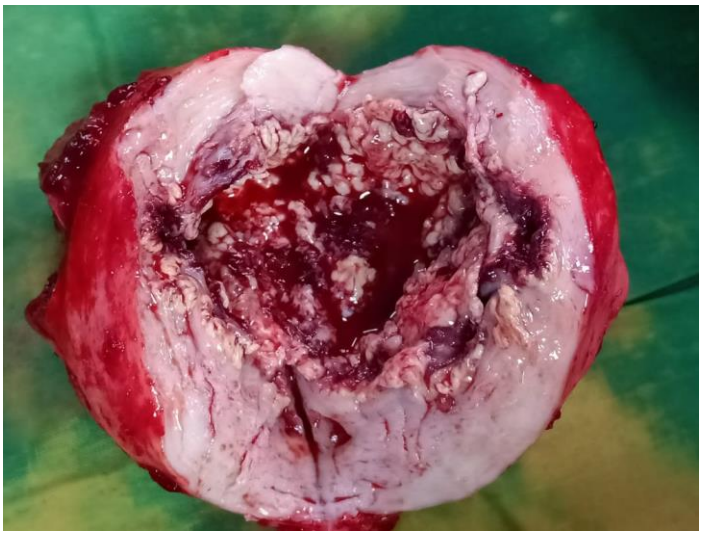

Fig 4: Cut section of uterus showing infected and necrosed endometrium and myometrium

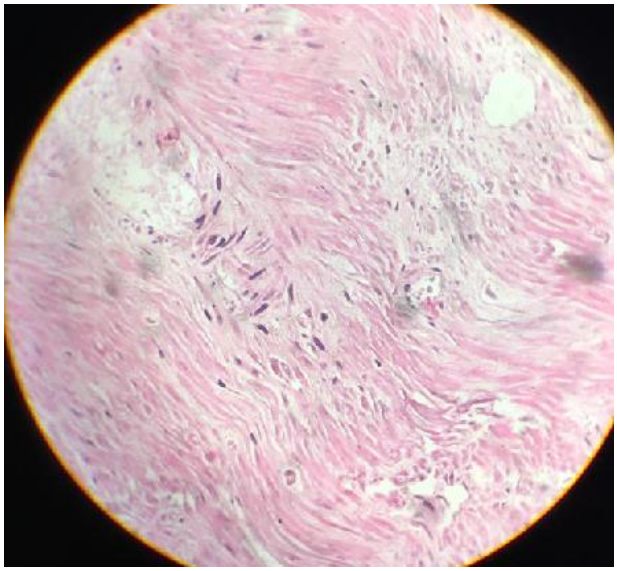

Fig 5: Low power view of pus showing few viable cells with normochromic spindle shaped nuclei having blunt ends and few inflammatory cells. No mitotic figure, no nuclear atypia, no coagulative necrosis seen

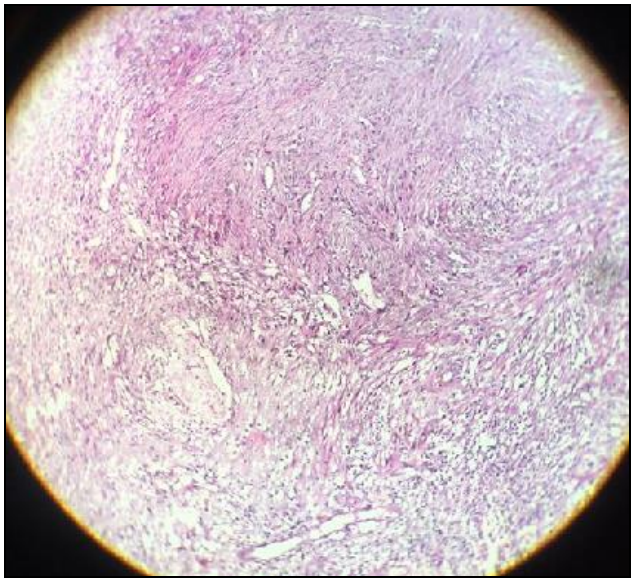

Fig 6: Low power view from myometrium showing interlacing fascicles of smooth muscle cells suggesting leiomyoma in the upper and right side along with inflammatory cells in lower field

3. Discussion

Cases of pyomyoma are quite rare and are seen in relation to pregnancy, puerperium, uterine instrumentation [2] like D & C, caesarean section [13], balloon tamponade for PPH [10] and also seen in cervical stenosis [12]. In post menopausal women, this condition have been seen in hypertensives or diabetics or in presence of atherosclerosis, probably due to immune or vascular compromise [2, 5]. There are reports of suppuration after uterine artery embolization (UAE) also, again because of infarction, necrosis, followed by infection of the fibroid [14, 15]. However our case is a nulliparous woman with no other predisposing factor and no prior history of fibroids, though she complained of menorrhagia. One explanation is that she might have had rapid growth of the fibroid which lead to ischemia, degeneration and necrosis and subsequent infection from the vagina, as she was sexually active.

Diagnosis of this condition is often difficult. Sometimes it may present with abrupt onset or may have symptoms extending upto a year [16]. It may remain silent or have nonspecific symptoms with an unidentified origin of bacteremia [5]. Symptoms include fever, pelvic pain, abdominal mass, vaginal discharge, vaginal bleeding, nausea, fatigue, changes in bowel habits, weight loss etc. [3]. However the triad proposed by Greenspoon *et al.* [5] of sepsis, fibroids and no other source of infection should raise suspicion [1, 14]. Symptoms mimic malignancy too, like leimyosarcoma or ovarian cancer [17], which was also suspected in our case due to irregular lump, tenderness, weight loss and severe anemia. In some cases CA-125 is also raised and imaging is non-specific. The differential diagnosis includes pyometra, tuboovarian abscess, infected ectopic pregnancy, malignancy, perforated viscus, or degenerating fibroids [10, 18]. Ultrasonography of a pyomyoma may show an enlarged heterogeneously echogenic pelvic mass with solid and cystic components [19]. Characteristic CT findings of a ruptured pyomyoma with peritonitis include air and debris in the leiomyoma, discontinuity of the myoma wall, and intraperitoneal free air and ascites [12]. Complications include uterine rupture, peritonitis, sepsis, endocarditis and acute respiratory distress syndrome (ARDS) [16].

Definitive treatment is often delayed due to non-specific presentations [11]. Perioperative broad spectrum antibiotics, followed by surgical intervention, either myomectomy or hysterectomy is the treatment of choice [1]. In more than half of the reported cases, hysterectomy was done. Conservative management is usually ineffective [20], but may be done with antibiotics and surgically or radiologically guided drainage [3, 21]. Histopathology may show cystic degeneration, hyaline change, hemorrhage, necrosis and acute inflammatory changes with smooth muscle cells [2, 8]. Pus culture of pyomyoma usually demonstrate a polymicrobial infection, with Staphylococcus species and Escherichia coli being the most commonly isolated organisms [1, 2, 3, 17]. Like our case, there are also few reported cases where no risk factors or etiology were found or no organism isolated from pus [3].

4. Conclusion

Pyomyomas are rare and difficult to diagnose. It has non specific presentations and mimics pelvic malignancies. A known prior history of fibroid may not be there in most of the cases. Gynecologists, general surgeons, radiologists and pathologists must be aware of this entity for early diagnosis and intervention. Delay in treatment can cause mortality. Surgery in combination with antibiotics usually gives a good outcome.

5. Conflict of Interest: None

6. Funding: None

7. References

1. Bagga R, Rai R, Kalra J, Saha PK, Singh T. “An unusual cause of postabortal fever requiring prompt surgical intervention: A pyomyoma and its imaging features,” Oman Medical Journal. 2017; 32(1):73-76.
2. Ono H, Kanematsu M, Kato H *et al*. “MR imaging findings of uterine pyomyoma: radiologic pathologic correlation,” Abdominal Imaging. 2014; 39(4):797-801.
3. Azimi-Ghomi O, Gradon J. Pyomyoma: Case Report and Comprehensive Literature Review of 75 Cases Since 1945. SM J Case Rep. 2017; 3(4):1054
4. Read S, Mullins J. Spontaneous Ruptured Pyomyoma in a Nulligravid Female: A Case Report and Review of the Literature. Case Rep Obstet Gynaecol. 2018; 2018:1026287.
5. Greenspoon JS, Ault M, James BA, Kaplan L. Pyomyoma associated with polymicrobial bacteremia and septic shock:

case report and review of literature. Obstet Gynecol Surv. 1990; 45:563-9.
6. Sah SP, Rayamajhi AK, Bhadani PP. Pyomyoma in a postmenopausal woman: a case report. South east Asia J Trop Med Public Health. 2005; 36:979-981
7. Genta Pedro R, Dias ML, Janiszewski TA, Carvalho JP, Arai MH, Meireles LP. Streptococcus agalactiae endocarditis and giant pyomyoma simulating ovarian cancer. South Med J. 2001; 94:508-11.
8. Goyal S, Mohan H, Punia RS, Tandon R. "Subserosal pyomyoma and tubo-ovarian abscess in a diabetic patient," Journal of Obstetrics & Gynaecology. 2015; 35(1):101-102.
9. Iwahashi N, Mabuchi Y *et al*. Large uterine pyomyoma in a perimenopausal female: A case report and review of 50 reported cases in the literature. Molecular And Clinical Oncology. 2016; 5:527-531.
10. Kaler M, Gailer R, Iskaros J, David AL. "Postpartum Pyomyoma, a Rare Complication of Sepsis Associated with Chorioamnionitis and Massive Postpartum Haemorrhage Treatedwith an Intrauterine Balloon," Case Reports inObstetrics and Gynecology. 2015; Article ID 609205:6.
11. Pinton A, Aubry G, Thoma V, Nisand I, Akladios CY. "Pyomyoma after abortion: Uterus conserving surgery is possible to maintain fertility. Case report," International Journal of Surgery Case Reports. 2016; 24:179-181.
12. Karcaaltincaba M, Sudakoff GS. "CT of a ruptured pyomyoma," American Journal of Roentgenology. 2003; 181(5): 1375–1377.
13. Shiota M, Kotani Y, Ami K, Mizuno Y, Ekawa Y, Umemoto M. "Uterus-sparing myomectomy for uterine pyomyoma following cesarean section," Taiwanese Journal ofObstetrics and Gynecology. 2013; 52(1):140-141.
14. Obele CC, Dunham S, Bennett G, Pagan J, Sung LY, Charles HW. "A Case of Pyomyoma following Uterine Fibroid Embolization and a Review of the Literature," Case Reports in Obstetrics and Gynecology. 2016; Article ID 9835412:5.
15. Rosen ML, Anderson ML, Hawkins SM. "Pyomyoma after uterine artery embolization," Obstetrics & Gynecology. 2013; 121(2):431-433.
16. Yeat SK, Chong KM, Pan HS *et al*. Ruptured Pyomyoma With Purulent Peritonitis: A Case Report And Literature Review. Taiwanese J Obstet Gynecol. 2005; 44(1):75-79.
17. Chen JR, Yang TL, Lan FH, Lin TW. "Pyomyoma mimicking advanced ovarian cancer: A rare manifestation in a postmenopausal virgin," Taiwanese Journal of Obstetrics and Gynecology. 2014; 53(1):101-103.
18. DeMaio A, Doyle M. "Pyomyoma as a Rare Source of Postpartum Sepsis," Case Reports in Obstetrics and Gynecology. 2015; 2015(Article ID 263518):2.
19. Tobias DH, Koenigsberg M, Kogan M, Edelman M, LevGur M. Pyomyoma after uterine instrumentation: a case report. J Reprod Med. 1996; 41:375-8.
20. Chen ZHY, Tsai HD, Sun MJ. "Pyomyoma: a rare and life-threatening complication of uterine leiomyoma," Taiwanese Journal of Obstetrics and Gynecology. 2010; 49(3):351-356.
21. Laubach M, Breugelmans M *et al*. Nonsurgical Treatment of Pyomyoma in the Postpartum period. Surgical Infections. 2011; 12(1):65-8.