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Case Report

Florid genital tuberculosis co-existing with adenomyosis and evading diagnosis

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ABSTRACT

When tuberculosis affects genital organs of young females, the disease often remains silent or may present with symptoms which are common to other Gynaecological conditions as well. The diagnosis of genital tuberculosis is challenging and the diagnosis can be missed even with extensive investigations including molecular studies. A case of florid tuberculosis affecting the uterus, cervix, ovaries and tubes, co-existing with adenomyosis of the uterus and the cold abscess masquerading as bilateral ovarian endometriomas is reported.

Keywords: Genital tuberculosis, Diagnosis, MRI features

INTRODUCTION

Tuberculosis is a chronic infectious disease with predilection for respiratory system. However, in recent years, tuberculous infection is being reported more frequently in extra-pulmonary locations. It has been shown that genital tuberculosis (GTB) account for nearly 9% of extrapulmonary tuberculosis.[1] In countries where there is high prevalence of tuberculosis, genital tuberculosis is a significant cause of damage to the pelvic structures leading to infertility. When tuberculosis affects genital organs of young females, the disease often remains silent or may present with symptoms which are common to other Gynaecological conditions as well. The most common initial symptom for which a woman seeks medical advice is infertility, followed by pelvic pain and menstrual disturbances. Routine laboratory tests for tuberculosis are of little value in the diagnosis of GTB and laparoscopy may not be helpful in the early stages of disease.[2]

Accurate identification of mycobacterium tuberculosis (MTB) through culture remains the gold standard technique. However, in extra-pulmonary sites such as pelvic organs, because of the pauci bacillary nature, the pick-up rate by culture and histopathological examination (HPE) is very low. Therefore, the diagnosis of GTB is challenging and the diagnosis can be missed even with extensive investigations including molecular studies. Here we report a case of florid tuberculosis affecting the uterus, cervix, ovaries and tubes, co-existing with adenomyosis of the uterus and the cold abscess masquerading as bilateral ovarian endometriomas.

CASE REPORT

40 years old, Mrs. S was admitted with irregular heavy periods, severe congestive dysmenorrhoea and deep dyspareunia, right sided lower abdominal pain and suprapubic pain of 2 years duration and she was requesting hysterectomy. She was married for 14 years and there were no conceptions. She attained menarche at the age of 13, and she has always had irregular periods once in 2-3 months, lasting for 3 days. For the past 2 years, the periods have become very heavy, lasting for 7-10 days which was associated with passage of clots. She also suffered from severe congestive dysmenorrhoea, which was gradually getting worse, intolerable and was not relieved by any medication. In the previous 1 year she had consulted more

than 10 clinicians with the same problem and there was no relief. She had continuous lower abdominal pain, more on the right side and there was deep dyspareunia. During periods, she also had painful micturition and defecation. There was no history of fever or discharge per vaginum. She is a known hypothyroid on 100 µgm of levothyroxine. In her personal history, there was history of loss of appetite and loss of weight of 1-year duration. There was no past history of tuberculosis or any other medical illness. However, in her family history, her father in law and a neighbor were suffering from pulmonary TB.

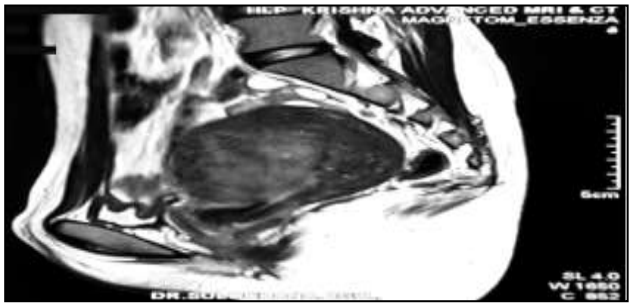

Figure 1: MRI saggital section showing a large lesion with mixed intensity seen within the endometrium, extending predominantly into the anterior myometrium with loss of endometrial interface.

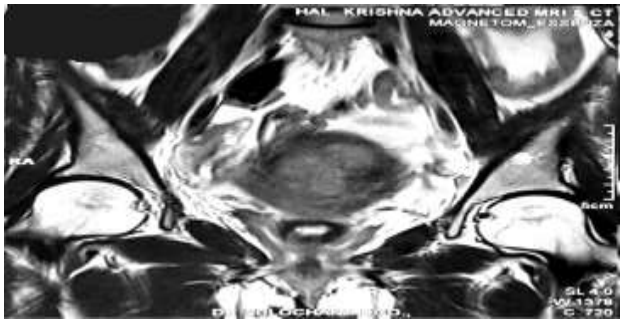

Figure 2: MRI - coronal view showing a large lesion within the endometrium.

Figure 3: Cut section of the uterus with thick material in the endometrial cavity.

In her past investigation and treatment history, she was extensively investigated and treated for infertility by many clinicians. On reviewing her records, there were no male related causes for infertility. Hysterosalpingogram (HSG) done 6 years ago was reported normal. There had been many attempts of induction of ovulation. Two years ago, she was evaluated for Infertility with diagnostic hystero-laparoscopy at a tertiary care hospital and the findings were minimal peritoneal adhesions, minimal intrauterine adhesions, and bilateral cornual block. At this point of time, GTB was suspected and the endometrium was sent for HPE, culture for mycobacterium tuberculosis (MTB) and Polymerase chain reaction (PCR) studies. The endometrium was proliferative and the culture and PCR for MTB were negative. Following these investigations, there were two attempts of In vitro fertilization and both failed.

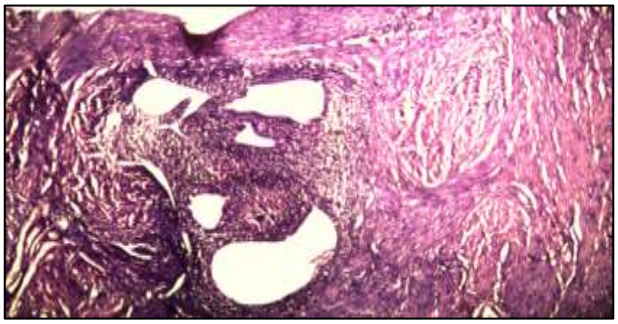

Figure 4: HPE of the myometrium with adenomyotic areas showing granuloma H and E (10X).

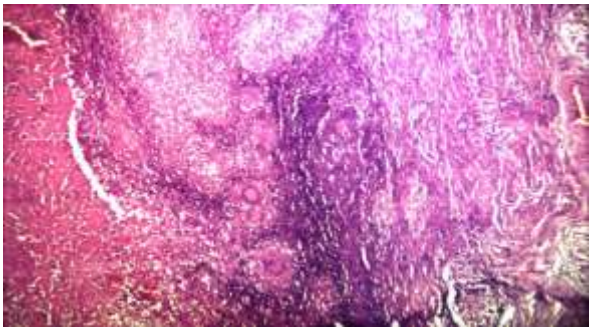

Figure 5: Granulomas in the endometrium H and E (10X).

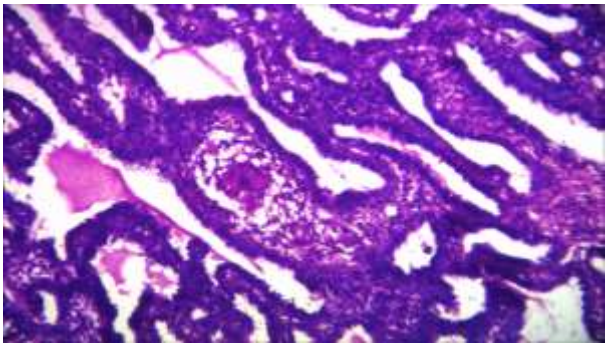

Figure 6: Fallopian tube with granulomas H and E (10X).

On examination, there was no pallor, no peripheral lymphadenopathy, chest, breasts and thyroid examination were normal. Her body mass index (BMI) was 20. On abdominal examination there was tenderness over the supra pubic region and right iliac fossa, there was no free fluid or mass in the abdomen. On bimanual palpation, the cervix and vagina were healthy, the uterus was enlarged to 12 weeks size, and was extremely tender. Through the right fornix tender vague mass was felt. On rectal examination, there was tenderness in the pouch of Douglas and in the fornices.

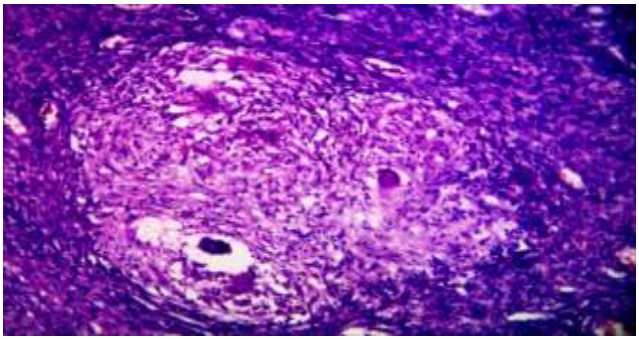

Figure 7: HPE of ovary with granulomas H and E (10X).

The investigation results were as follows: haemoglobin 10.8 gm with normocytic, normo chromic anemia, total count was 6,000, and the differential count was P64, L28, B4, and M4. Erythrocyte sedimentation rate (ESR) was 18 and 34 and the Mantoux test measured 5 mm and HIV was non-reactive. Random blood sugar was 94 mgs and the thyroid function tests (TFT) was normal. X- Ray chest was reported normal. Urine culture and high vaginal swab did not show growth of any organisms. Imaging studies were carried out by ultrasound sonography test (USG) and magnetic resonance imagining (MRI) abdomen and pelvis. The USG findings were, uterus was enlarged to 11.6×8.8×7.6 cm with multiple isoechoic lesions in anterior and posterior myometrium, largest measuring 4.8×4.4×4 cm, and the endometrial thickness was 7 mm. The right adnexa measured 9×7 cm, the left adnexa measured 6.5×6 cm, both showing cystic lesions with internal echoes with septations. The USG impression was adenomyosis with complex bilateral ovarian masses, probably endometriosis. On MRI the uterus was enlarged to 10.2×7.5 cm, with heterogenous hyperintense lesions, measuring 5.5×4.5 cm in T1w and T2w sequence. Both ovaries were enlarged in size with multiple small cystic areas (Figure 1, 2). The impression was large adenomyotic lesion in the anterior myometrial wall and diffuse adenomyosis in the posterior myometrium.

Based on the clinical evaluation and investigation findings a provisional diagnosis of adenomyosis with endometriosis was made. In view of long standing abnormal uterine bleeding and to rule out GTB (recent history of loss of weight and loss of appetite and contact with tuberculosis in the family) endometrial sampling was done with pipelle sampler. The endometrial samples were sent for culture by BACTEC method, and for HPE. The culture was negative and HPE showed endometrial hyperplasia.

There was a strong request from the patient and her family for hysterectomy as the patient suffered from intolerable pain. The patient was given extensive counselling and alternate options were discussed for the possible conservation of the uterus and ovaries. The patient did not desire a child anymore and she wanted only relief from pain. Based on the patient's request and a provisional diagnosis of large adenomyosis and bilateral endometriomas, it was decided to proceed with hysterectomy. After necessary precautions abdomen was opened by a transverse incision. There was no free fluid in the abdomen. The uterus was enlarged and there were bilateral tubo-ovarian masses all covered by adhesions. Separate ovarian tissue could not be made. While dissecting, there was discharge of pus like material from both adnexal masses which was sent for culture and sensitivity. With careful dissection, total hysterectomy with removal of both adnexal masses was carried out and the specimen was sent for HPE. The cut section of the uterus showed hypertrophied myometrium with thick material in the endometrial cavity (Figure 3). The culture did not grow any organisms. The HPE of the myometrium showed adenomyosis with endometrial glands and stroma and granulomas were seen within the adenomyotic areas. (Figure 4). The HPE of the endometrium showed granulomas with langhans giant cell, caseating necrosis and hemorrhage (Figure 5). The endocervix also showed evidence of granulomas. The HPE of the fallopian tube showed tubal plicae with granuloma showing langhans giant cells (Figure 6). The HPE of the ovary showed epitheloid cell clusters with a collar of lymphocytes) and langerhans giant cell (Figure 7).

Post-operatively, the patient was treated with anti-tuberculous treatment (ATT) for 6 months with category I drugs. The patient did not develop post-operative complications and was started on hormone therapy for menopausal symptoms. At one-year follow-up, the patient was healthy and was on hormone replacement therapy.

DISCUSSION

This case is being presented for its difficulty in diagnosing GTB, as well as for its rare presentation at unusual sites. In spite of being investigated twice, GTB was missed in this case. At the initial evaluation two years earlier, laparoscopy did not show significant findings except minimal adhesions. In early and latent cases, there may not be evidence of pelvic tuberculosis at laparoscopy.[2,3] Probably in this patient, the infection was either latent or very early and did not produce any changes externally, therefore the diagnosis could not be made laparoscopicaly. Both culture and HPE were also negative on both the occasions. It is difficult to diagnose GTB either by histopathology or by culture. In the extra-pulmonary sites,

the organisms are sparse in number, therefore, the genital tract lesions are bacteriologically mute.[4] There may also be bacteriostatic substance in the tissue that inhibits the growth of bacilli.[5] It is also difficult to diagnose tuberculosis of the endometrium histologically, because, due to the cyclical sloughing and shedding of the endometrium every month granulomas do not have enough time to form, so the endometrium may not show evidence of tuberculosis in all cycles.[6] Nucleic acid amplification tests (NAAT) enable the clinician to make a rapid and accurate diagnosis. However, false negative results can occur due to paucibacillary nature of the specimen and the presence of PCR inhibitors which are common in blood containing samples. As endometrial samples are always mixed with blood, this could possibly explain the negative PCR result in this patient.[7] The fallopian tubes are believed to be the initial and most frequently affected genital organ in pelvic mycobacterial infection. From the fallopian tubes secondary spread can occur to the peritoneum in 45% or to the ovaries in10 to 30% of cases and the endometrium is involved in 50 to 80% cases.[8] Myometrium and cervix are rarely involved. In this patient, granuloma was seen within the adenomyomatic foci in the myometrium and the endocervix was also involved.[9]

It is possible that a latent disease got activated following investigations and treatment two years ago. When the ovaries are involved, the infection is usually limited to peri-oophoritis, due to spread from adjacent peritoneum or fallopian tube. Rarely, tubercles can be formed within the ovarian parenchyma which can lead to the formation of a cold abscess.[10] A tubo-ovarian abscess is seen as a heterogeneous hypoechoic collection with moving internal echoes and echogenic debris in the adnexa, with the ovary not being recognized separately. In this patient the ovarian abscess was diagnosed as endometriomas because of the similar imaging features. The uterus and ovaries could not be conserved in this case, as the uterus was enlarged with large adenomyosis and the ovaries were replaced by large T-O masses. In spite of the florid nature of genital tuberculosis, the patient responded well to ATT, and did not develop complications such as abdominal tuberculosis, military tuberculosis or fistula formation.

CONCLUSION

Genital tuberculosis is a challenging disease and is difficult to diagnose in spite of specific investigations. Whenever, there is a strong clinical suspicion, one may have to perform the investigations repeatedly.

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